The effect of Functional Electrical Stimulation on abdominal muscle strength and gross motor function in children with cerebral palsy: a randomised control trial

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Declaration

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Abstract

Keywords: cerebral palsy, electrical stimulation, abdominal muscles, weakness, balance, physiotherapy, GMFM, GMFCS

Background: While spasticity has long been considered the primary impairment in children with cerebral palsy (CP), the presence of weakness is gaining increasing recognition and importance and the coexistence of the two impairments may result in greater participation restriction. Advances in neuroimaging technology have shown deficiencies in the muscle morphology of those with CP, including decreased muscle volume, cross-sectional area and muscle belly length, abnormal cocontraction and muscle imbalances. Postural dysfunction such as poor reactive balance control, delayed muscle activation, top-down muscle recruitment and a limited ability to accomplish task-specific postural adaptations by those with CP, has been well documented. Postural control is necessary for the achievement of most functional activities and the lack thereof can be severely disabling. Inconclusive evidence exists in the literature to support one therapeutic intervention over another in the management of CP and, in particular, in addressing postural control. Studies researching electrical stimulation in those with CP have shown modest, significant improvements in gait, lower limb muscle strength, hand function and anecdotal improvements in health related quality of life (HRQoL). These positive effects warrant further study to determine the efficacy of electrical stimulation in improving the strength of the abdominal muscles in children with CP and whether this translates into improvements in gross motor function and balance.

Pragmatic trials best determine the effect of treatment interventions in routine, everyday situations. As these trials compare an intervention to the accepted standard/norm of intervention rather than to a placebo, it is necessary to establish what specific regimes of treatment are currently in use, how they are implemented and for what durations. In order to determine whether FES adds value to existing regimes, it is therefore necessary to describe the baseline treatment offered by therapists at the Institution and at similar institutions in Johannesburg, in order to increase the generalisability of the results.

Objective: The primary objective of this study was to investigate the impact of functional electrical stimulation (FES) on abdominal muscle strength and gross motor function in children with CP. A secondary objective was to compare the content and intensity of physiotherapy intervention offered by the therapists at the Institution, with that offered by similar facilities in Johannesburg.

Survey of Intervention Strategies:

Methodology: A cross-sectional descriptive research design was used for this portion of the study. Participants comprised all 25 physiotherapists employed at nine government special needs schools in Johannesburg, offering physiotherapy to children with CP.

Instrumentation consisted of a checklist of intervention techniques created by the researcher and approved by a panel of paediatric specialists, as well as a description of the treatment regularity and intensity of children with CP treated by each practitioner. Individuals completed the checklist anonymously after providing consent. As the numbers included in this section of the study were small, descriptive statistics were used to describe the characteristics of the therapists and the interventions utilised. The rank ordering of the frequency with which the different interventions were used at the Institution, and in the other centres, was calculated using Spearman's rho, a non-parametric test.

Results: As three physiotherapists did not return their intervention checklist, the sample consisted of 22 therapists. The mean years of experience in the experimental school were 17.75 (SD=7.89) and in the control schools 14.88 (SD=10.66). Nineteen therapists had some degree of Neurodevelopmental Therapy (NDT) training. Post allocation varied from one to five physiotherapists at each school. The Institution, at which the intervention was based, is the only one of these schools that treats learners into their teens and offers a mainstream curriculum until grade 12. The percentage of learners at the Institution with a physical disability was 84.68% while the mean percentage at the other special schools was 54.74%. The majority of schools treated children with cerebral palsy in groups. Treatment techniques most commonly used by all participating physiotherapists were facilitation of transitions between positions and general limb strengthening. No therapists employed the

Power Plate, FES or Vojta therapy techniques in practice. The rank ordering of the frequency of use of the most commonly used techniques by physiotherapists at the Institution and the other special schools in Johannesburg combined were similar with a correlation of rho=0.68 ($p<0.001$).

Discussion: The physiotherapists at the Institution where the study took place were found to perform similar treatments to those therapists based at other special needs schools in Johannesburg. Most therapists have specialist NDT training and many years' experience but have different workloads due to very high patient to practitioner ratios. Those treatment techniques not employed were usually due to financial and/or time restraints. The Institution was the only one of the nine schools still providing therapy for children with CP in their teens.

Intervention study

Methodology: An experimental, randomised control trial with single blinding was used, with a pragmatic design, for this part of the study. A sample of convenience was taken from the Institution at which the researcher works. All children with CP between 5-18 years were eligible for inclusion, if parental consent and participant assent was gained and children did not suffer from epilepsy or have other scheduled medical interventions taking place prior to or during the trial. Ethical approval was obtained and consent/demographic data collected. Intervention was carried out by the four physiotherapists employed at the Institution (including the primary researcher), after training and piloting took place. Randomisation was stratified based on the Gross Motor Function Classification System (GMFCS). Names were placed into the five relevant envelopes and alternating names drawn were placed in the control and then the intervention group and so on by an impartial individual. A trained assessor, blinded to the group allocation of participants and with no knowledge of each child, evaluated each participant using the Gross Motor Function Measure-66 (GMFM), the Timed Up and Go test (TUG), Timed Sit-Ups, the Pediatric Reach Test (PRT), a Peak Flow Meter, and the Pediatric Quality of Life Inventory 3.0 CP Module (PedsQL) prior to and following the intervention. A participant evaluation of the intervention was also completed, by those participants in the experimental group, after the study took place.

The control group continued to receive their usual weekly/biweekly physiotherapy intervention, as did the intervention group. The experimental group received additional concurrent FES for a six-week period during their usual physiotherapy sessions, as well as an additional two passive sessions a week. The Microstim 2 (v2) devices were applied to the bilateral external oblique muscles for 20 minutes each session. Therapy sessions lasted 45 minutes.

Data analysis: The STATISTICA version 11 software programme was used for all data analysis. Descriptive statistics were used to portray demographic and medical characteristics of participants. If normally distributed, parametric tests were used. If not, non-parametric tests were employed. Ordinal data was analysed using non-parametric tests. The two groups were compared at baseline to ensure that they were analogous. To determine within group differences from baseline to post-intervention, the paired t-test or Wilcoxon signed rank test was used. To test between group differences the independent t-test or Mann-Whitney U test were used. Sub-group analysis was performed to determine if participants classified at different levels of the GMFCS responded differently to intervention. Correlations were performed to determine if changes in the different parameters were correlated with one other. A one-way ANOVA determined if participants classified at different levels of the GMFCS or of different ages responded differently to intervention.

Results: Fifty-one out of 56 possible participants returned consent forms. A further four did not meet the inclusion criteria set out for the intervention study. Twenty-two children were randomly allocated to the experimental group and 25 to the control group. All of these children completed the six-week intervention, as well as all pre- and post-intervention outcome assessments applicable to each individual. No significant difference between groups was detected on any outcome measure post intervention. Significant differences found on analysis were improvements within group for GMFM-66 scores in all participants combined ($p=0.002$), for the experimental group ($p=0.016$) and for the control group (0.048). Within group differences were found for the whole group ($p=0.009$) and for the control group ($p=0.023$) on peak flow meter readings in that expiratory flow was stronger post- intervention. Significant improvements in HRQoL were detected within groups for the combined group of participants ($p=0.001$), the experimental group ($p=0.018$) and the

control group (p=0.029). The greatest number of participants showing improvements in GMFM-66 percentage scores was between the ages of 14 and 18 years old.

Discussion: No significant improvements were found to support the effectiveness of FES of the abdominal muscles of children with CP in comparison to conventional therapeutic interventions (in terms of abdominal strength, gross motor function, balance, expiratory lung function and HRQoL). Based on this study, one cannot recommend the use of FES for abdominal strength, balance and gross motor function in children with CP. While the sample size of the study was large enough to detect a clinically important difference should it have occurred, the participants ranged from the highest to the lowest GMFCS level, standard deviations were large and subgroup analysis did not have enough power.

Both experimental and control groups did show significant improvements in GMFM-66, peak flow and PedsQL measures following the intensive six-week intervention, thus confirming that the therapeutic interventions used at the Institution, in the treatment of CP, were associated with significant improvements in function. Despite the effectiveness of an NDT-based, motor-learning approach used during routine intervention still being debatable in current literature, regular implementation of these modalities, and the other techniques employed at special needs schools in Johannesburg, does seem to be associated with significant improvements in function, in children with CP.

In contrast to the widely accepted gross motor development growth curves that predict that children with CP tend to plateau in function after seven years of age and reach a ceiling to their motor development in their early teens, those children in this study making the greatest improvements fell between the ages of 14 and 18 years old. This finding has led the researcher to question the validity of these predictive curves, as well as use of the apparently stable GMFCS for classification of functional ability, particularly for children with CP in low-income countries who have not been exposed to regular therapeutic intervention from a young age and into adulthood. Older children, particularly if they have had limited access to treatment, may continue to benefit from intervention.

Conclusions: The physiotherapists at the Institution at which the study took place are performing therapeutic interventions similar to those at the other government special needs schools in Johannesburg. These interventions were found to be associated with significant improvements in gross motor function, lung function and HRQoL over a six-week period. Based on the experimental study, FES does not appear to improve these functional outcomes and resources should not be used unnecessarily on including this modality as an adjunct in the treatment of CP. Teenage children with CP in middle to low-income countries that have not been exposed to regular therapeutic intervention from a young age and through their teenage years, appear to possess the ability to continue making functional, gross motor improvements beyond the plateau illustrated by internationally accepted gross motor development growth curves. These results provide a basis for further study as, should these results be corroborated, continued treatment of children with CP, in middle to low income countries, into their teens, should be considered, especially if early intervention was not possible or treatment was ceased early.

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Abbreviations

American Academy for Cerebral Palsy and Developmental Medicine	AACPDM
Analysis of Variance	ANOVA
Ankle Foot Orthoses	AFOs
Botulinum Toxin A	Btx-A
Caregiver Priorities and Child Health Index of Life with Disabilities	CPCHILD
Central Nervous System	CNS
Cerebral Palsy	CP
Complementary and Alternative Medicine	CAM
Conductive Education	CE
Consolidated Standards of Reporting Trials	CONSORT
Constraint-Induced Therapy	CIT
Electrical Stimulation	ES
Electromyography	EMG
External Oblique	EO
Forced Expiratory Volume in One Second	FEV_1
Forced Vital Capacity	FVC
Functional Electrical Stimulation	FES
Functional Reach Test	FRT
Gamma-Amino-Butyric-Acid	GABA
Gastro-Oesophageal Reflux	GOR
Gross Motor Ability Estimator	GMAE
Gross Motor Function Classification System	GMFCS
Gross Motor Function Measure	GMFM
Health Related Quality of Life	HRQoL
Human Immunodeficiency Virus	HIV

Internal Oblique	IO
International Classification of Functioning, Disability and Health	ICF
Intraclass Correlation Coefficients	ICC
Intrathecal Baclofen	ITB
Magnetic Resonance Imaging	MRI
Mann-Whitney U test	MWU
Minimum Clinically Important Difference	MCID
Neurodevelopmental Therapy	NDT
Neuromuscular Electrical Stimulation	NMES
Pediatric Evaluation of Disability Inventory	PEDI
Pediatrics Outcomes Data Collection Instrument	PODCI
Pediatric Quality of Life Inventory	PedsQL
Pediatric Reach Test	PRT
Professional Functional Electrical Stimulation South Africa	PROFFESSA
Proprioceptive Neuromuscular Facilitation	PNF
Quality of Life	QoL
Randomised Control Trial	RCT
Rectus Abdominis	RA
Selective Posterior Rhizotomy	SDR
Single Event Multi-Level Surgery	SEMLS
South Africa	SA
Treatment	Rx
Threshold Electrical Stimulation	TES
Timed Up and Go	TUG
Transversus Abdominis	TA
Typically Developing	TD
World Health Organization	WHO

1. Introduction

1.1 Background

Scholars and medical practitioners have debated the definition of cerebral palsy (CP) for over 150 years (1). The most recent definition describes a heterogeneous group of permanent movement and postural disorders due to disturbances occurring in the foetal or infant developing brain (2). This definition has been expanded to include several accompanying impairments and activity limitations experienced by this population, as laid out by the World Health Organization's (WHO) International Classification of Functioning, Disability and Health (ICF) system (3). This model proposes a biopsychosocial model of disability and health, providing a framework allowing for an integrated, globally understood and accepted language to be used by health professionals when discussing and documenting the assessment of and intervention provided for patients (4).

The prevalence of CP, estimated at between 2-2.5 per 1000 live births, has remained essentially unchanged over the last 40 years (5). Due to advances in medical technology and improving standards of neonatal care, more extremely preterm children now survive, with up to 50 percent of those born before 27 weeks gestation being diagnosed with CP (6).

While spasticity has long been considered the primary impairment in children with CP, the presence of weakness associated with the upper motor neuron lesion is gaining increasing recognition and importance (7) and the coexistence of the two impairments may result in greater participation restriction for the individual (8). With advances in neuroimaging technology (6), studies have been able to exhibit deficiencies in the muscle morphology of those with CP. Such changes include decreased muscle volume, cross-sectional area and muscle belly length (9) as well as abnormal cocontraction and muscle imbalance between agonist and antagonistic muscle groups (7).

A questionnaire completed by 83 adults with CP in America revealed that 55% experience poor balance and 52% listed weakness as a worrying impairment. The study further highlighted a significant association between weakness and limitations in social interaction and participation outside the home (10).

Further to this, postural dysfunction such as poor reactive balance control, delayed muscle activation (11), top-down muscle recruitment and a limited ability to accomplish task-specific postural adaptations have been well documented (12). As a whole, the muscles forming the core contract in order to increase the stability of the spine (13). The muscles are typically activated in muscle synergies best suited to the specific activity required of the individual (13)(14)(15). The decreased abdominal muscle activation (16) and lack of motor planning allowing for accurate speed and force of muscle contraction, often evident in CP (17)(18), further contribute to fallouts in balance and postural control (16). As postural control is necessary for the achievement of most functional activities, the lack thereof can be presumed to be severely disabling (19)(20)(21) and perhaps even argued to be the primary impairment in people with CP (22).

The physiological contribution of the abdominal muscles to breathing is well documented and respiratory muscle fatigue has been shown to limit exercise performance (23). Due to decreased voluntary control of muscles, those with CP have been shown to have a decreased total lung capacity and a significantly reduced functional residual capacity when compared to normal predictive values (24).

Global strategies for treatment of CP include such techniques as Neurodevelopmental Therapy (NDT) (25), conductive education (26) and constraint-induced therapy (27), all of which, on systematic review, have shown inconclusive evidence to support the use of one approach over another (28). An investigation of available literature by Harris and Roxborough (29) was conducted due to the paucity of evidence available for intervention to enhance postural control in children with CP. Their search confirmed that conclusive support for techniques such as adaptive seating, lower extremity orthoses, external perturbations or NDT, in the treatment of postural control, is still lacking.

Researchers have expressed the need to address the pathophysiological weakness present in CP therapeutically (7). Trunk stabilisation and deep muscle targeting programmes have been shown to improve abdominal strength and motor control in adults with no neurological fallout (30). Hibbs et al maintain that core strengthening programmes can avoid muscle recruitment imbalances, help re-learn motor control and thus improve neural co-ordination and function (31).

Functional Electrical Stimulation (FES) is the application of an electrical current stimulating a muscle's innervating nerve, causing a muscle contraction to obtain functionally useful movement (32). The use of electrical stimulation in the treatment of children with CP is being increasingly investigated as the potential to improve strength and motor function exists. Regrettably, any positive results reported need to be interpreted with caution due to the poor methodological design or small sample sizes employed in all of these studies (33).

Studies researching the use of electrical stimulation of upper and lower limb muscles in CP have shown modest significant improvements in gait parameters (34), lower limb muscle strength (35), hand function (36) and anecdotal functional improvements to indicate possible changes in health-related quality of life (HRQoL) (37). Only one study was found investigating its use on the abdominals in infants with CP. The authors reported improved sitting posture, trunk control and gross motor function in participating infants with CP (following a six-week intervention using concomitant electrical stimulation over the abdominals and posterior back muscles during NDT-based therapy) (38). The positive effects demonstrated warranted further study, to determine its efficacy in abdominal muscle use considering the large effect postural control has on motor skill acquisition (20)(21). The use of FES of the abdominal muscles in children with CP as an adjunct to therapy may enhance the effect of therapy and result in improved functional outcomes.

Pragmatic trials best determine the effect of treatment interventions in routine, everyday situations (39). These trials compare an intervention to the accepted standard/norm of intervention rather than to a placebo and have the distinct benefit of allowing practitioners to continue with their customary treatment (where this would not be possible in a clinical laboratory setting) (39). In order to establish whether FES adds value to existing regimes, it is necessary to establish what specific regimes of treatment are currently in use, how they

are implemented and for what durations (40). If results are to be generalisable, then a comparison between the baseline treatment offered by therapists at the Institution, with those offered by therapists working at similar facilities in Johannesburg, need be made.

1.2 Aims and objectives

Based on the reasoning above, the overall aim of the study was to investigate the impact of FES on abdominal muscle strength and gross motor function in children with cerebral palsy.

The specific objectives were:

- To determine whether the physiotherapy treatment of children with CP offered at the Institution was similar to other special needs schools, by comparing the content and intensity of physiotherapy intervention offered by the therapists at the Institution with that offered by other similar facilities in Johannesburg
- To determine whether concomitant therapeutic intervention using FES would result in a significantly different improvement in children with cerebral palsy, in comparison with a control group of children with cerebral palsy receiving only routine physiotherapy, in the following parameters over time:
    - Abdominal muscle strength as measured by the number of sit-ups completed in one minute
    - Gross motor function, with particular emphasis on balance, as determined by scores on the Gross Motor Function Measure-66 (GMFM-66), Pediatric Reach Test (PRT) and the Timed Up and Go (TUG)
    - Respiratory function, as measured by Peak Flow Meter readings
    - Self-perceived health-related quality of life (HRQoL), as assessed by the Pediatric Quality of Life Inventory 3.0 CP Module (PedsQL) and the participant evaluation of the intervention
- To determine if a relationship exists between demographic and medical characteristics and improvements in functional status made by participants

1.3 Research question

Does FES administered regularly, in conjunction with typical physiotherapeutic intervention, over a period of six weeks, improve abdominal muscle strength and gross motor function in 5-18 year old children with cerebral palsy compared to routine physiotherapy treatment?

1.4 Significance of the study

Weakness (41) and dysfunction of postural control mechanisms are central to CP, causing disturbances in activities of daily living and attainment of functional independence (18)(19)(22)(42)(43)(44)(45)(46)(47)(48). This may necessitate complete or partial dependence on the assistance of a caregiver for essential daily tasks detrimental to family dynamics, parental psychological well-being and time spent on work or other responsibilities (49)(50)(51). As CP cannot be cured, an enormous economic burden is placed on family members and the state while covering life-long therapy, adaptive equipment and medical interventions to correct secondary impairments (52)(53)(54)(55)(56)(57). This is further compounded by the fact that current medical technology has helped prolong the expected life span of those with CP (58).

Premature birth remains the single highest risk factor for the development of CP in high-income countries (59). Peri- and postnatal causes of CP, such as maternal or neonatal infection, stroke and encephalopathy (60) need to be considered in low to middle-income countries, like South Africa (SA). Poor health care standards, frequent mother-to-child transmission of Human Immunodeficiency Virus (HIV)(61) and high rates of postnatal trauma caused by violent crime or motor vehicle accidents (62) may possibly account for a greater prevalence rate of CP in such countries (61)(63).

Researchers have speculated that almost 80% of the world's people with disabilities reside in resource-poor countries but, unfortunately, accurate prevalence rates of CP in countries like SA are generally not available (63)(64).

National governing bodies require that any therapeutic intervention given by health professionals complies with current evidence-based practice (29)(65)(66)(67). To date very few studies of high methodological quality have provided evidence for the effective use of different physiotherapy techniques for the treatment of CP (68)(69). This is concerning as CP has been cited as the most commonly treated condition by paediatric physiotherapists (70). While much time is spent by therapists on enhancing postural control in those with CP, little evidence is available to support the interventions used to achieve this (29)(46). The same can be said for FES, where a paucity of available evidence and contradictions in the few studies that have been undertaken make it difficult to advocate for its use in the treatment of CP, despite generally positive findings (33)(71)(72). Early intervention is seen as crucial to maximise the potential for plastic changes to the developing brain, as well as to the outcome prognosis for children with CP (73). Consequently, the therapists at the Institution have the opportunity to make clinically and functionally relevant changes in the lives of the children they treat, as they are most often accepted to the school from a very young age. Should this study show statistically significant effects on gross motor function, strength, postural control and HRQoL, then FES could provide a cost effective and easily administered adjunct to normal physiotherapy intervention, improving the lives of the children at the Institution with CP.

If a greater sample size and more stringent methodology, than previous studies, are employed during the study, this may provide the most reliable evidence for the introduction and use of FES in paediatric clinical practice to date. Conversely, if the intervention is not found to be effective, then valuable resources will not be wasted on an ineffective treatment in the future.

1.5 Research setting

The Institution, at which the study was based, is a special needs school for children with physical disabilities in Johannesburg, SA. The school offers a mainstream curriculum from nursery school level until matric/grade 12. While approximately one third of the school admissions have a diagnosis of CP, the school also accepts children with a range of orthopaedic, neurological, congenital and medical conditions causing limitations in physical functioning. Classes are small, to cater for adapted education and physiotherapy, occupational therapy, speech therapy and nursing services are provided (during the school day) to those children requiring such intervention. These therapies are provided to children of all ages attending the school. The school is government-funded and most children attending the school hail from poor socio-economic circumstances, thus necessitating external bursaries to compensate for hostel accommodation, transport, school fees, etc.

2. Literature review

2.1 Overview of the review

The following literature review presents arguments for the feasibility of FES as an intervention strategy for improving abdominal strength and gross motor function in children with CP. The review begins with the current definition of CP, a review of classification systems, followed by the incidence and prevalence of CP in low, middle and high-income countries, based on different subtypes of CP. Diagnosis and the aetiology of CP are presented next. The following section describes the various impairments associated with CP, as well as a description of the development of postural control in both typically developing children and children with CP, leading into the activity limitations and participation restrictions that these impairments can result in.

Research explaining motor learning and the gross motor development growth curves for prognosis in CP is presented. This is followed by an overview of the available treatment techniques used by physiotherapists in the treatment of CP. A description of FES and empirical evidence advocating the use of FES and at what parameters is then presented, with an overview of different practice schedules subsequently. The next section of the review describes available standardised outcome measures for the assessment of children with CP and the selection of those used in the study. Finally, different study designs are explained and a summary of the above information concludes the literature review.

Relevant literature for use in the review was sought by entering keywords such as cerebral palsy, impairments, abdominal muscles, balance, strength, physiotherapy, electrical stimulation, GMFM and GMFCS, etc. into online databases such as CINAHL, PubMed, Science Direct, BioMed Central and Medline. While reading these articles, any applicable references cited were also sought, using the above method. Articles were limited to those freely available, full text articles, published in English.

2.2 Definition, classification and epidemiology

2.2.1 The definition of CP

CP has been alluded to in literature since as early as 1862. William Little, a British orthopaedic surgeon, was the first person to group birth defects resulting in spasticity and contractures together; referring to this disorder as "Little's Disease". By the 1960s Sigmund Freud (a Viennese neuropathologist at the time) coined the term CP to describe a group of infantile motor disorders (1)(74). As our knowledge of the workings of the human brain continues to grow and evolve so too does the definition of CP (5)(74). Despite numerous attempts at multidisciplinary conferences to determine a consensus on the definition of CP, this has never been fully realised with various definitions still employed in different parts of the world (2)(74). One aspect that throughout history has always been agreed upon is the permanent nature of CP and that it is a non-progressive disorder (2)(75).

CP should be considered not as a diagnostic term, but rather a clinical description of a group of conditions (2)(76). In 2006, an executive committee comprised of specialists in their respective fields was assembled at an international workshop in America to develop an updated definition of CP that might be acceptable for international use. After deliberation the agreed upon definition was as follows:

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).

This definition recognises that the disorder is primarily a motor defect but also includes impairments of a different nature that can equally affect activity (2)(77). While generally recognised and accepted globally, this definition does still create some ambiguity in that no

upper age limit is given for diagnostic purposes and there is no consensus regarding the age at which brain maturation ceases, and a diagnosis of CP can no longer be given. Several researchers deem cortical development complete by two years of age while other groups of professionals consider any child under the age of five, with damage to the brain, as having CP (26)(59)(74)(77).

In summary, CP can be seen as a group of primarily motor disorders, which occur due to damage taking place while the brain is still developing. These motor disorders cause difficulties with movement and posture and may be accompanied by a multitude of concomitant impairments. These impairments all lead to limitations in daily functioning as well as participation in society making CP a globally experienced disorder with extensive consequences for the individual.

2.2.2 The classification of CP

Generally speaking, classification requires the meaningful grouping of distinctive features of data in a way that can be clearly understood by anyone using the classification system (78). A global attempt to standardise the classification of CP has been made in order to provide a language all practitioners involved in the management of CP can use to communicate with one another and with those with CP and their families (77)(79)(80). Such systems also enable more accurate diagnosis, comparison between similarly classified individuals with CP, as well as provide a baseline to assist with monitoring of progress or regression and to help plot prognosis (2).

As with the definition of CP, the widespread acceptance of the classification system outlined by the Task Force on Childhood Motor Disorders, the group for the Surveillance of Cerebral Palsy in Europe and the International Workshop on Definition and Classification of Cerebral Palsy is acknowledged. This system encompasses all of the different subsystems of classification that have been previously used to try and give each individual the most specific classification possible; attempting to allow for the recreation of the same classification by any professional assessing the person with CP (81)(82). Unfortunately, little

empirical evidence exists to support the interrater reliability of these models and as CP covers a heterogeneous group of disorders different in each instance; classification by different practitioners remains inconsistent (80)(81). Nevertheless, the system outlined by the above professional bodies remains the most thorough system of classification to date and will be described below. It is suggested that any assessment of CP should include the person's age, any radiological findings and any clinical observations made (2).

2.2.2.1 Classification by type of motor disorder

The first subsection of classification is that most widely used in other systems; classification by type of motor disorder (2). In the past, this merely meant making a distinction between "pyramidal motor disorders"/upper motor neuron lesions (with damage occurring at a corticobulbar or corticospinal level resulting in weakness and increased stretch reflexes) or "extrapyramidal motor disorders" (due to damage to the non-primary motor cortical areas, cerebellum or basal ganglia with no associated weakness or changes in spinal reflexes but still causing a loss of motor control) (79). Previously, pyramidal damage was thought to be associated with spasticity and extra-pyramidal damage with dystonic or athetoid-type features. It is now widely accepted that most people with CP have elements of both; further complicating classification (79). With this in mind, systems now further divide motor disorder into spastic, ataxic or dyskinetic (subdivided into dystonic and choreoathetotic) motor types (2)(76)(77).

Tone can be explained as the relaxed muscle state when a limb is passively moved causing a stretch on the moving muscles. Hypertonia is therefore a higher than normal resistance to this externally induced movement or stretch (79). Spasticity, on the other hand, is hypertonia but with added resistance to passive stretch based on both the speed and direction of the externally imposed movement or with increased resistance above a certain threshold speed or range of movement (i.e. a "spastic catch") (77)(79).

Dystonia involves involuntary twisting movements characteristic to each individual with CP seen at rest or usually with attempts at voluntary movement. This is due to activation of

muscles in an in-coordinated sequence resulting in the sustained or intermittent, repetitive abnormal postures seen (77)(79)(83). Where dystonia's involuntary movement causes sustained postures, chorea causes distinct, fragmented involuntary movement observed in random sequences. The rapid, unpredictable movements seen in chorea appear to flow from one portion of the body to another at different speeds and in different directions and do not cease at rest (77)(83). In contrast, during athetosis, no distinct fragments of movement are seen and involuntary movement involves slower, writhing-type movements of the distal body (and may involve the head, neck and trunk). Such involuntary movements occur continuously and occur more rhythmically than athetotic movements, making maintenance of a stable posture difficult. Generally these two abnormal motor types do not occur in isolation and "choreoathetosis" is usually seen (2)(77)(83).

Finally, in ataxia, a difficulty in motor planning leads to an in-coordination of the speed, force, directionality and accuracy with which movement is executed. This may lead to a visible intention tremor and/or past pointing. Children with this classification tend to be primarily hypotonic. The incoordination of movement experienced is often associated with difficulties in maintaining the body's sense of equilibrium, leading to balance fallouts and thus an uncoordinated, unsteady gait (2)(84)(85).

It is well documented in literature that spastic CP originates from damage to the cortex of the brain, dyskinetic disorders from damage to the basal ganglia and ataxia from lesions of the cerebellum (2)(86)(87). Historically, CP has been linked to lesions of white matter in the brain (76)(88). With advances in and more regular use of MRI in neonatal brain imaging, greater understanding of the mechanism causing CP has been achieved and different injuries seem to occur based on the prenatal or postnatal period at which damage takes place (88)(89).

Due to vulnerability of the periventricular white matter in the first two trimesters of pregnancy, a lack of myelination of nerve cells occurs while the lungs are still underdeveloped resulting in poor nerve transmission and decreased oxygenation during this period (89)(90). Damage occurring at this time almost exclusively causes periventricular leukomalacia (PVL) (89)(90). This is a common finding in babies born prematurely due to foetal distress (89) and in those children later presenting with spastic diplegic CP

(2)(76)(82)(91). Insult during the third trimester more often leads to thalamic and brain stem injury (89). Asymmetrical myelination of the posterior capsule seen in term infants or asymmetrical PVL is most often associated with hemiplegia and spasticity is most often seen when damage has occurred after 37 weeks due to migrational brain malformations (such as schizencephaly, lissencephaly and heterotopia) (2)(76)(82). In addition to PVL those children presenting with quadriplegic CP tend to have damage to cortical, subcortical and other areas of the brain (92).

As most people with CP present with "mixed" motor types, numerous texts have suggested classifying individuals based on their predominant motor type. It is advised that relevant secondary motor types are still documented as this can make for more appropriate surgical, pharmacological and therapeutic intervention (2)(77)(81)(83)(93) (94). It is further suggested that the functional ability of the individual is made explicit, as well as any secondary impairments listed at this point (2).

2.2.2.2 Classification by anatomical distribution/topographical characteristics

The second subsystem of classification is based on the anatomical/topographical distribution of CP. This section is divided into hemiplegia (where upper and lower limbs on the same side of the body are most affected), diplegia (where the lower limbs are more affected than the upper limbs) and quadriplegia (where the upper limbs are equally or more affected than the lower limbs) (2)(59)(77)(86). This system can be difficult to utilise as there is often ambiguity surrounding the extent to which each body part is affected and many systems still make use of terms such as "monoplegia" or "tetraplegia" further confounding classification (2)(59)(77).

2.2.2.3 Classification by causation and timing of CP

As medical technology has improved, it has become more customary in high resourced settings that infant brain imaging takes place. The final subsystem of classification suggested by the International Workshop on Definition and Classification of Cerebral Palsy panel is to record the causation and timing of the brain insult in CP, where possible. This allows one to correlate findings on imaging with physical motor fallouts observed (89).

2.2.2.4 Classification using the Gross Motor Function Classification System (GMFCS)

Most classification systems have been supplemented with an indication of the proposed severity of the condition but this can be very subjective and thus unreliable in its allocation (95)(86). It was for this reason that the GMFCS (Appendix I) was first created, based on the ICF framework previously described (see section 1.1 Background), in order to provide a more reliable and valid standardised tool with which to classify the severity of CP (2)(96)(97)(98)(99)(100). It has since become the most internationally accepted measurement instrument for the severity of CP (2)(97)(101)(102). After much adaptation, in 1997 the GMFCS was published, providing a system of classification of children with CP between the ages of one and 12 based on self-initiated rather than passive movement and focused on overcoming environmental and societal factors (96)(97)(100)(103)(104). This system provides five broad categories into which practitioners can place a child, even after having had very little initial contact with that particular individual (101). Distinction between the levels is based on general motor functioning and mobility and the degree to which adaptive aids are required in order to be independent (96)(100). As the scale is ordinal, no inference can be made that an equal distance exists between each level and the next (96)(103)(100). The original scale gave guidelines for allocation of level to children with CP in categories below two years, 2-4 years, 4-6 years and 6-12 years of age (2)(101)(105). In 2007, a further category was added allowing the classification of children with CP from ages 12-18 as well. The descriptions for levels I-V vary depending on the relevant age category (100). A simple overview of category break down can be seen in Table 1 below (100).

Table 1: Gross Motor Function Classification System (GMFCS) overview of basic levels (adapted from Rosenbaum et al (2002) (104))

GMFCS Level	Description
I	Walks without limitations (problems encountered are with higher level gross motor activities)
II	Walks with limitations (may struggle with walking longer distances, have difficulty running and jumping and require holding rails when using stairs)
III	Walks using a hand-held mobility device indoors (and wheeled mobility outside in the community)
IV	Self-mobility with limitations; may use powered mobility (highest level of function is sitting)
V	Transported in a manual wheelchair (generally dependent on others for all mobility)

The validity of the GMFCS has been studied extensively (2)(50)(97)(104) and interrater reliability has been quoted as 0.75 (50)(96), with good content validity (95) and excellent agreement between classification performed by medical practitioners and the families of those affected with CP (97).

2.2.2.5 Conclusions regarding classification

There is unfortunately still no consensus over which classification system should best be used in describing CP, but the more thorough the recording of presentation the better the

opportunity for similar classification between medical professionals and the more accurate the prognosis for families. Comprehensive classification involves determining the most predominant type of motor disorder and listing any secondary motor types, as well as recording the anatomical distribution of symptoms, the causation and timing of the onset of CP (if known) and using the GMFCS to measure the severity of CP based on an individual's age-related functional ability. As CP is such a heterogeneous group of conditions, one might assume that studies including all classifications in their sample would have greater generalisability to the wider CP population.

2.2.3 Incidence and prevalence

In this section, the incidence and prevalence of CP, as well as the association between gestational age, subtype of CP and prevalence, are discussed. Subsequently, prevalence in high and middle to low income countries are compared, followed by statistics available for the prevalence of CP in SA.

Incidence accounts for the number of new cases of a particular disease or disorder occurring over a particular period. More relevant is prevalence, which accounts for the proportion of a specified population living with that disease/disorder at a particular point in time (63). Disappointingly, despite advances in antenatal and neonatal care, global incidence rates have remained constant over time at between 1-2.5 per 1000 live births (5)(60)(76)(82)(106)(107)(108)(109)(110)(111)(112)(113). This is due, in part, to the fact that very low birth weight and severely preterm babies now survive (5)(58)(60)(76)(106)(107)(114). Estimated prevalence rates may be inaccurately low as very severely affected infants may die prior to official diagnosis of CP and those with only minor impairments may underreport their diagnosis (76).

Prevalence can be further broken down by classification where various studies with large samples have listed that between 71-90.6% of those with CP have a predominantly spastic motor type, with between 2.6-16% being dyskinetic and 1-5.6% ataxic. These studies did not all account for those classified as having a mixed motor type of CP

(10)(57)(81)(111)(115)(114)(116). When separating prevalence rates by anatomical distribution, those with diplegia account for the majority with 49-73%, hemiplegia 26-38% and quadriplegia 8-11% of the total prevalence rate of CP (96)(114)(117).

Internationally, there is a trend for a slightly higher prevalence of CP in males, with studies citing statistics of between 58-61% (57)(59)(82)(87)(110)(111). It is reported that 15 million babies worldwide are born preterm each year (accounting for a tenth of all new-borns), with a trend toward increased incidence of preterm birth due to the current availability of infertility treatments, increasing maternal age and maternal health problems (118). All 11 countries listed as having a preterm birth rate of 15% or more in 2010, were low income countries (118). The incidence of CP is inversely proportional to gestational age (60)(91)(119). Preterm births account for the greatest proportion of those diagnosed with CP and the prevalence rate of very preterm and very low birth weight new-borns is raised from the overall prevalence of 1-3 to 19-152 per 1000 live births in these children (76)(82)(111)(119)(120). Genetic mutations associated with advanced paternal age have also been linked to a higher incidence of CP; particularly in athetoid/dystonic presentations (121).

Disability incidence and prevalence rates are universally higher in low income countries and areas, with CP cited at 3.33-4.7 per 1000 live births in these areas (82)(111)(112)(114). Low birth weight is also associated with socioeconomic status (82)(111)(112)(114). Two schools of thought surround these higher rates. Firstly, rates may only appear higher due a lack of accurate diagnosis due to diagnosis being made by unqualified professionals or without the use of standardised outcome measures or neuroimaging technology in low-income areas (63)(64). Secondly, poor socioeconomic circumstances can lead to a lack of antenatal care, home births, where avoidable birthing complications cannot be treated, and a lack of finances to seek medical assistance after such births (118). Such problems may be self-limiting for prevalence rates though as many of these children do not survive (64).

Very little evidence is available regarding the prevalence of CP in SA, as a national CP register does not exist. Other studies have cited SA incidence statistics as high as 28-64 per 1000 live births in their discussions (63), but no South African based country-wide prevalence studies could be found.

2.2.4 Diagnosis and aetiology

In recent years, neonatal Magnetic Resonance Imaging (MRI) has become more commonplace, in order to establish a diagnosis of CP based on damage to the brain evident on such scans (76)(87). Due to a lack of resources in middle and low-income countries, it can be assumed that diagnosis of CP is primarily made based on clinical observations, as funding for expensive MRI technology is not always available. The wider picture of motor or anatomical distribution of CP may be different in such countries, due to the differing causes and timing of injury seen, compared to high-income countries.

Determining the aetiology of disease is important as it gives valuable insight into how the condition should be managed, what prognosis is expected and whether future prevention is possible (59)(87)(110). Greater detail on diagnosis and the aetiology of CP (Appendix II) can be found in section 6.

Poor socioeconomic status forms a further risk for the development of CP (76)(107)(111)(122) with perinatal risk factors particularly prevalent in less resourced sections of society (113). Statistics on the breakdown of subtypes of CP are generally not available for low-income countries and neuroimaging even less available (76). As preventable perinatal causes of CP are still rife in these areas, due to lack of ante- and postnatal quality medical care and poor maternal education, one can assume that prevalence of quadriplegic and dyskinetic CP is higher than in rates given for the developed world (63)(76)(111).

Assumptions can also be made that in SA, listed as having 5.6 million persons living with HIV in 2011 (123), and almost all new paediatric cases occurring due to mother-to-child transmission (61), that postnatally acquired CP due to encephalopathy will be higher than in high-income countries (63).

2.3 Manifestations of CP

Based on an internationally accepted biopsychosocial model of medicine (3)(124), in 2001 the WHO released a system of documenting disability called the International Classification of Functioning, Disability and Health (ICF), following almost a decade of collaboration between professionals. The aim of doing so was to create a classification system using a universal language for health practitioners to describe disability (77)(125)(126)(127). In 2006, the United Nations Convention on the Rights of Persons with Disabilities defined disability as:

…a difficulty in functioning at the body, person, or societal levels, in one or more life domains, as experienced by an individual with a health condition in interaction with contextual factors (127).

Rather than concentrating on the cause of a supposed disabled minority, the ICF acknowledges that a decline in health, at one point or another in a lifespan, is a universally experienced phenomenon and the focus is rather on the impact this has on functioning (4). The ICF classifies disability based on the interaction between three different components: impairment, activity limitation and participation restriction. Impairment is defined as a loss or abnormality of anatomical structure/physiology or of psychological function, an activity limitation is a difficulty in performing daily tasks due to impairment, and participation restriction is any limitation of the disabled individual in their interaction within society (77)(98)(128)(129)(130)(131).

While historically a focus has generally been placed on treating disability at impairment level, research now encourages a move toward addressing participation and activity to achieve more meaningful outcomes (124)(132). By using an ICF framework for assessment and addressing the different components of disability, the aim is to enable the planning of interventions that will provide functional improvements, creating opportunities for greater participation of those with disabilities (127). This functional-developmental approach is

particularly relevant in a paediatric context (133). A discussion of each of the three ICF categories follows.

2.3.1 Impairments

Physical impairments such as spasticity have been cited in literature since the 1800s (1). Below, the impairments of tone and mobility, muscle strength, postural control, energy expenditure and respiratory dysfunction are explained. Further to these physical/musculoskeletal impairments are those of the sensorimotor system and cognition, along with other impairments such as epilepsy, impairments of vision, hearing and speech, and behavioural difficulties. While not directly relevant to the study at hand, these secondary impairments all influence performance and thus, need to be understood in order to comprehend the global picture of CP and how an intervention may affect these parameters. For this reason, these impairments are briefly described in Appendix III.

Studies have shown that the severity of musculoskeletal impairment experienced is a strong indicator of the likelihood of developing sensorimotor and/or other accompanying impairments (6) and the significance these other impairments may have on limiting function should not be underestimated (2). GMFCS level is associated with the severity and number of accompanying impairments; with level V persons with CP experiencing the most severe and the greatest number of impairments (134)(120).

While the condition itself it not progressive, the physical impairments discussed below commonly give rise to secondary complications (50)(68)(76)(129)(135), particularly in more severe presentations of CP (136). These, mostly orthopaedic, complications are briefly illustrated in section 6.3 in the appendices.

The following sections are of great importance to the understanding and management of CP, as numerous studies have shown that the number of people living with CP who now experience a multitude of disabling impairments is increasing (59). Approximately 60% of

children with CP experience at least one impairment over and above their primary physical impairments (120).

2.3.1.1 Musculoskeletal impairments

Abnormal muscle tone and joint mobility

As was outlined in section 2.2.2, tone can be explained as the relaxed muscle state when a limb is passively moved causing a stretch on the moving muscles. Hypertonia is therefore a higher than normal resistance to this externally induced movement or stretch (79). Spasticity, on the other hand, is hypertonia but with added resistance to passive stretch based on both the speed and direction of the externally imposed movement or with increased resistance above a certain threshold speed or range of movement (77)(79)(93)(137)(138). Spasticity is said to be a result of the upper motor neuron lesion occurring in CP (139), hyperexcitability of the tonic stretch reflex (137) and a lack of cortical inhibition of excitatory neurotransmitters acting at spinal cord level (140)(141)(142). Although primarily as a result of neural damage, hypertonia and/spasticity may have a musculoskeletal component (54). Spasticity can be exacerbated by voluntary attempts at movement, changes in psychological state and, at times, innocuous external stimuli (79). Dyskinetic and ataxic movement disorders have already been described in section 2.2.2.

Although difficult to prove, the assumption is made that the resistance to movement caused by spasticity eventually leads to muscle shortening and thus decreased active joint excursion. Over time, this leads to limited passive joint range of motion and subsequently joint contractures in those with CP (49)(93)(142)(143)(144)(145). This limitation in range is, in fact, a consequence of changes to the actual intrinsic muscle structure of those with CP. While agreement has not been met on the exact mechanism by which this occurs, studies have shown the muscle fibres of those with spasticity to be shorter than normal samples, “rounded” or to have a “moth-eaten” appearance (146)(147). Such alterations in intrinsic muscle fibre properties, and possible fibrosis of muscle fibres in these subjects, lead to

what is referred to as myoplastic hyperstiffness or passive muscle stiffness, with reports of spastic muscle fibres being up to twice as "stiff" as normal muscle fibre samples (147)(148)(149). This concept has been further validated by the fact that hypertonia has been shown to exist in the absence of electromyographic (EMG) activity as well (150).

Muscle weakness

While spasticity has historically been considered the most disabling impairment experienced by those with CP, literature now supports the idea that weakness causes greater limitation in function (7)(129)(151)(152)(153). Furthermore, it is possible for both spasticity and weakness to coexist (77). Weakness is an inability of muscle structures to exert a maximal force when contracting actively (7)(84)(154). This is often difficult to observe in children with spasticity, as the resistance exerted can be mistaken for muscle strength (84). Weakness has been expressed as being inversely proportional to gross motor function (153) and increasingly more authors now acknowledge that weakness is a major cause of functional limitation in CP (155).

Muscle weakness forms part of the so-called "negative motor signs" group of impairments linked to the upper motor neuron lesion of CP. Other negative signs include reduced selective motor control (i.e. difficulty activating the correct sequence of muscles), ataxia (uncoordinated activation of muscles during movement) and apraxia/developmental dyspraxia (uncoordinated activation of muscles needed to achieve a specific goal-directed task) (84).

Research has shown that even those mildly affected by CP have considerably decreased strength in comparison to their typically developing (TD) peers (151)(156) and particularly in those with spastic CP (9). In 2010, a systematic review by Mockford and Caulton concluded that all studies examined showed that all persons with CP experience some degree of weakness and that all children in the studies reviewed were weaker than their TD counterparts (7).

Studies have shown that, in general, the weakness experienced in CP tends to manifest more distally with the greatest weakness seen in the ankle dorsiflexors (causing drop foot during gait) (129); followed by the hip extensors. The quadriceps muscles seem to fall within normal muscle strength ranges (7)(157)(158)(159). Little evidence exists to enable comment on the strength of the upper limb in CP. Significantly decreased abdominal muscle activation has been noted in children with CP when compared to their TD peers (16) and a link has been recorded between poor abdominal muscle activation and poor functional outcomes in this population (160).

Dysfunction may be linked more to the imbalance between the strength of agonist and antagonist muscle groups in CP rather than the weakness of any single muscle group (157). As can be expected, weakness is more pronounced in those with diplegic CP than those with hemiplegia (156). Interestingly, studies testing the strength of children with hemiplegia have found that even the supposedly 'unaffected' side is markedly weaker than TD children of the same age and height (157)(161).

Muscle properties of those with CP compared to TD persons

The most fundamental part of the motor system is a motor unit, comprised of muscle fibres innervated by a motor neuron and the adjoining neuromuscular junction (8). During typical development, muscles grow by increasing muscle fibre size. This occurs for the first two decades of life after which progressive atrophy begins to occur with ageing (162)(163). The muscle force produced by an individual depends on how many and at what rate these motor units are recruited. In TD individuals, this differs depending on the specific muscle contracting (8). As a self-preserving mechanism, the smallest motor neurons will activate the weakest motor units first and then, as an increasing intensity of muscle contraction is required, motor units will fire sequentially until ultimately the largest units are engaged (145). Muscles can be distinguished as either "phasic"/fast acting (containing almost equal amounts of slow/type I and fast/type II muscle fibres) or "tonic"/postural muscles (which are made up of a greater proportion of slow-twitch/type I muscle fibres) (145).

The imbalance between agonist and antagonist muscle strength and the impaired cortical inhibition of neural pathways previously explained may account for the frequently documented co-contraction of antagonist muscle groups seen in CP (42)(163)(164)(165)(166). Another explanation frequently cited is that the intrinsic muscle properties themselves are altered in those with CP (137). It has been speculated that these muscles may be essentially normal at birth but due to inactivity, impairment and abnormal postures and attempts at movement, muscle morphology begins to adapt and change over time (90)(144)(145)(166).

Due to advances in imaging technology and biopsy histology, studies have revealed differences between the visco-elastic properties of the muscles of those with CP and TD children and adults (54). Such changes may include a greater predominance of type I/slow-twitch muscle fibres (8)(93)(145), larger variation in muscle size (8)(54)(93)(163) (with generally decreased cross-sectional area and thickness of muscles) (9)(167) and decreased sarcomeres in series present (8)(167). The less sarcomeres present in series, the shorter the length of the fascicle; impairing muscle extensibility and thus inhibiting the muscle power possible (9)(168)(146). Other morphological changes seen are an increased collagen and fat content in muscles, as well as poorer quality collagen when compared to age-matched TD peers (8)(54)(144)(163)(169). Neurological causes of weakness in CP may include an inability to increase the firing rate of motor units sufficiently during movement (8)(145) and pre-emptive muscle contraction before the necessary time, leading to inaccurate movement (8).

Over and above the intrinsic muscle changes evident in CP, immobility and disuse can lead to further muscle atrophy aggravating the weakness seen in this population (170). Further exacerbating the problem of weakness is the sheer number of treatment modalities used in the management of CP that inhibit muscle strength. Almost all surgical procedures and medications used to address spasticity and secondary impairments, as well as certain immobilising orthotic devices, have weakness as an unfortunate side effect of the intervention (151)(156)(171). This will be discussed in more detail in upcoming sections of the study.

All of the factors discussed above negatively affect the muscle force, torque and speed of contraction possible in CP (167). As is evident by the sheer volume of research on the subject, weakness is a critical impairment experienced by all of those with CP and its effect on function and independence is vast. While it is widely accepted that postural disturbances are, in part, due to a lack of abdominal muscle activation in this population, few studies have specifically concentrated on assessing this empirically. Most researchers proclaim that due to the negative impact of weakness on global functioning in CP, this parameter should be addressed during rehabilitation. The lack of convincing investigation into abdominal strength and intervention to improve this in CP warrants further study, as gains in strength may lead to gains in posture and motor function.

Posture

Posture can be defined as the vertical positioning of body segment/segments against gravity and balance is how one adapts this posture in response to internal/external perturbations in order to prevent falling (172)(173). Postural control is essential for independent functioning (46)(174). Efficient balance requires the effective operation of the visual, somatosensory and vestibular systems (43)(46)(172)(173)(175). The primary responsibility of the motor system in humans is to maintain an upright spine for function and this requires an efficient synergy between muscular structures around the trunk and sufficient postural tone (14)(176). In order for adequate posture to take place, one needs to be able to maintain the centre of mass within the body's base of support (18)(43)(48)(174).

Previously, maintenance of balance was assumed to be due to reflexive reactions to external stimuli (177)(178). Balance is however, nowadays perceived as a complicated mechanism requiring many intact processes. Studies have shown that postural strategies can be learnt with practice and exposure to certain environments and motor experiences, thus negating the idea of balance originating from pre-programmed reflexes (177)(178).

The somatosensory system is employed first in order to alert the brain to the fact that a challenge to posture has occurred (whether an internal perturbation due to movement about to take place or an external stimulus threatening the stability of the body in space) (172)(177)(176). Secondly, the brain predicts the nature of the disturbance about to take place, based on the above information received, and anticipatory postural adjustments occur in order to minimise the fallout caused by the perturbation (176). This can be seen as early as in 4-6 month old TD infants, where anticipatory neck and trunk muscle activation occurs simultaneously with upper limb muscle contraction during reaching (12)(18)(43)(164)(176). This is the start of developing postural control and balance (174). In adults, these patterns become integrated and more refined over time so that trunk adjustments to perturbation do not alter the trajectory of the reaching hand and movements in general are smaller and more graded (17)(20). In TD children, these postural adjustments continue to develop until approximately 10 years of age, by which time adult-like postural control mechanisms are possible (16)(20). In general, in standing, young children tend to stabilise themselves during tasks by limiting the degree to which the trunk is allowed to move (to provide a stable base from which movement can take place). Adults, on the other hand, tend rather to improve stability by controlling the size of all anticipatory movements for greater precision during tasks (20). In the first year of life top-down muscle recruitment is typically employed but thereafter balance reactions generally become organised from the supporting surface upwards (i.e. distal upwards) when attempting to regain control of posture (46)(179). It has also been said that specific, predictable muscle activation sequences/synergies occur in response to the direction of the perturbation taking place (18)(43)(180). For example, a perturbation displacing the body forward results in activation of dorsal muscles and backward displacement causes contraction of the ventral muscles (12)(18). These gross direction-specific muscle synergies develop first and are later refined with practice and repetitive feedback during movement (18). As well as being direction-specific, postural adjustments are also task-specific (178). Postural control mechanisms may require interplay between numerous body systems, but most essential to the attainment of adequate postural control for basic function, is the ability to activate the "core" appropriately.

Core stability

"Core" stability can be explained as the balance between passive spinal structures and active muscle control enabling appropriate trunk movement and posture against gravity and during the activities expected of the body (15)(31). The core acts as a stable base from which movement of the limbs can take place (15).

Debate surrounds exactly which structures should be included in descriptions of the core. There is consensus that the muscles around the trunk form a type of cylinder encasing the vital organs and the spine. A comprehensive list of suggestions for inclusion consists of the axial skeleton (i.e. the spine, pelvic and shoulder girdles), the diaphragm (forming a roof over the core), the paraspinals and gluteal muscles (posteriorly), the abdominal muscles (anteriorly), the pelvic floor muscles (creating the base of the cylinder) and all related ligaments, tendons, intervertebral discs, etc. (13)(15)(31)(181). Other muscles that have, on occasion, been included as part of the core are the psoas major, quadratus lumborum, latissimus dorsi and shoulder girdle muscles (31)(181).

As a whole, the muscles forming the core contract in order to increase the stability of the spine (13). The way in which these muscles are activated depends on the specific movement or activity required of the body. Each muscle plays a particular role but generally the core muscles do not act in isolation but rather form synergies to achieve the desired goal-directed movement (13)(14)(15). A balance between the so-called local muscles (which act segmentally due to their attachment to the lumbar vertebrae) and more global muscles (attached to the pelvis and/or hips) is necessary to ensure adequate mobility and stability of the spine (31). Figure 1 shows the anatomical situation of these muscles. Each relevant muscle, and its role in postural stability, is described below.

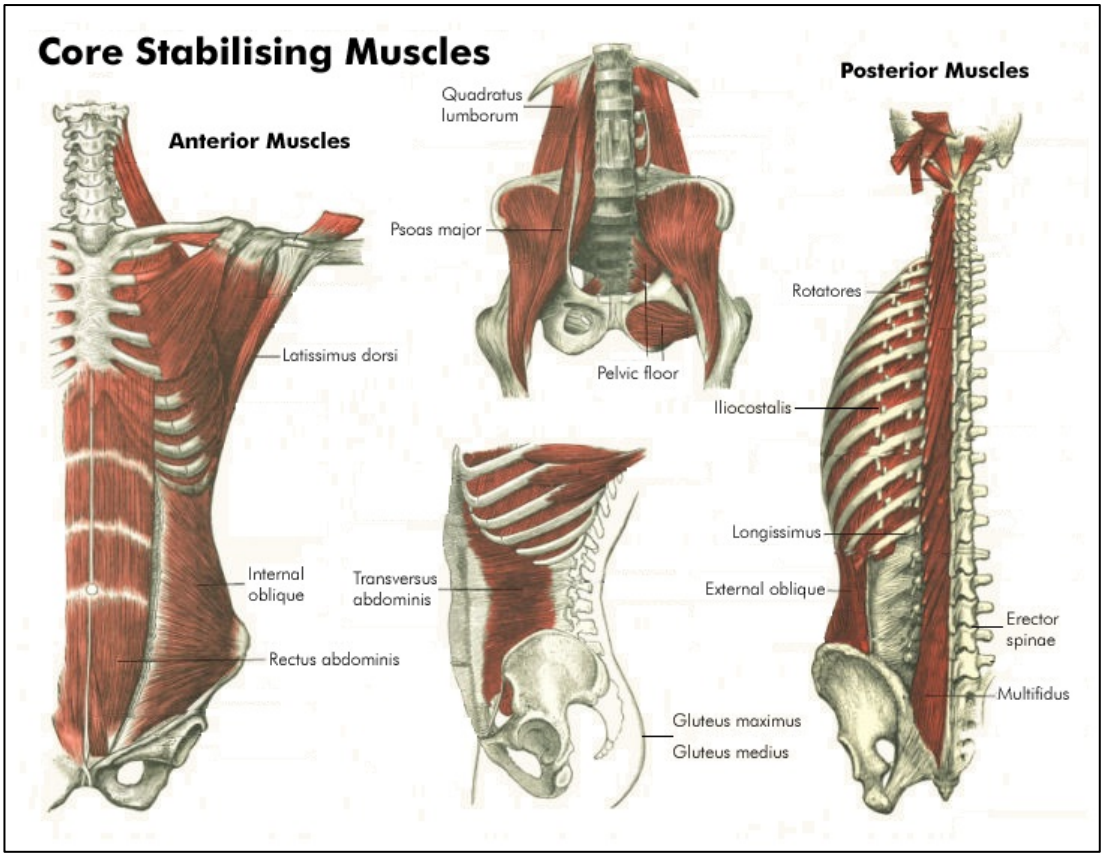


Figure 1: Core stabilising muscles

(adapted from http://www.zazzle.com/deep_core_stabilizing_muscles_anatomy_poster_1-228846587710743159) (182))

The transversus abdominis (TA) muscle is the deepest of the abdominal muscles and is widely considered central to core muscle function (14)(181)(183). During complex tasks, the TA tends to be activated before the other abdominal muscles, as well as preceding limb muscle activation. Its horizontal fibres act as a corset around the abdomen and its contraction inward stabilises the spine and can regulate intra-abdominal pressure (14)(13)(181)(183)(184). It is less active during passive or supported postures than it is when the subject is moving in upright and unstable positions (14). A study by Saunders, Rath and Hodges also showed that the TA is the only abdominal muscle that contracts tonically during gait and running (while the others act phasically during these tasks) (183).

The oblique muscles are also considered primary spinal stabilisers (154). They consist of the internal oblique (IO) and the large external oblique (EO) muscles (which are the most

superficially situated abdominals) (185). Together the obliques act as trunk lateral flexors and rotators. Axial rotation occurs in the direction of the contracting side while the contralateral side contracts isometrically for stability (186)(187). In order to maintain equilibrium, core stability in all three planes of movement are necessary, making rotation a vital component of the maintenance of balance (15). This rotation is also necessary in order for dissociation between the trunk and pelvis to take place during the swing through phase for efficient gait as well as to allow reaching of the upper limbs across the midline (188). As well as rotating the trunk, these muscles are responsible for preventing an anterior tilt of the pelvis to allow the maintenance of an upright spine (181). It has been shown that an increased degree of anterior pelvic tilt in children with CP is associated with a decreased ability to complete sit-ups, possibly representing poorer abdominal strength (153). It could be postulated that the increased lumbar lordosis associated with the anterior pelvic tilt seen frequently in CP further exacerbates the "crouch" gait commonly experienced by this population (explained later in section 2.3.2 Activity limitations), further limiting balance and stability as the pelvis is considered the level of the body's centre of gravity. Assaiante has also shown that a stable pelvis is a precursor to the attainment of independent locomotion in TD children (179).

The rectus abdominis (RA) muscles run anteriorly, parallel to the spine, and are a major flexor of the trunk. They are said to brace the spine during heavy loading activities (31). In their review Akuthota, Scott and Nadler express the opinion that the RA is commonly overemphasised during strength training, thus usually neglecting the weaker EOs (181). The oblique muscles and RA only contract after the muscle responsible for limb movement has been activated (184). The abdominal muscles are also responsible for forced expiration, lung volume regulation and changes to the length of the diaphragm (184)(189)(190)(191). EMG and ultrasound imaging have shown all the abdominal muscles to be inactive during quiet breathing. These measures demonstrated strong contractions of the RA, EO and TAs in adult subjects during voluntary and forceful expiration, speech, laughter and voluntary coughing (192).

The thoracolumbar fascia encases the muscles of the back and communicates between the upper and lower limbs for coordinated movement (181). The erector spinae muscles (i.e. longissimus and iliocostalis) run superficially from the pelvis to the cervical region of the

spine (181) and act as powerful spinal extensors (186). The local muscles, deep to the erector spinae muscles, include the multifidi, rotatores and intertransversi muscles of the spine that act segmentally to assist with stability (181).

Often excluded in literature about the core is the quadratus lumborum that acts strongly in an isometric fashion to assist with stabilising the spine (181). Also neglected in research is the role the muscles around the hip (namely the gluteus maximus and medius) play in core stability (181). These muscles contract to stabilise the trunk over the body's base of support and provide strength for forward propulsion during gait (15)(172). To a lesser extent (due to its attachment in the lumbar region), psoas major may contribute to some extent toward spinal stability (181). As with the TA, the diaphragm contracts increasing intra-abdominal pressure and increasing the stability of the spine. The pelvic floor muscles contract in synergy with the TA drawing up the base of the core (181).

Posture and core stability in CP

As CP is defined as a disorder of movement and posture, it is assumed that all children with CP will experience difficulty with maintaining balance and postural control (42)(164)(180)(193)(194). As has been previously discussed, the development of postural control begins in early infancy. Any delays in attaining postural control can have detrimental effects on gross motor milestone acquisition, as postural adjustments are required for all activities (20)(21). In addition, without opportunities to experience movement and attempt new motor tasks in more severely affected children with CP, and due to the numerous impairments experienced by this population, the learning of postural adjustment may not be achieved (177)(195). Without reactive postural control, children with CP are unable to counteract unexpected threats to their balance (21).

During the exploration of new gross motor tasks, infants and children rely heavily on visual cues for postural control. The vestibular system begins to contribute more toward balance from approximately six years of age (179). As is discussed in the appendices, one of the

difficulties experienced by the CP population is that of visual impairment (section 6.3), which further contributes toward postural instability in these particular children (175).

A study on ambulant children with CP revealed that all participants showed direction-specific postural adjustments but that muscle activation in children with diplegia and hemiplegia was in a top-down fashion (43). Only children with level V GMFCS mobility limitations do not show direction-specific postural adjustments (46). The same few stereotyped muscle synergies were noted in all children with diplegia limiting their freedom of movement (43). Those participants who were ataxic employed TD bottom-up muscle synergies but the activation of these muscles was delayed. All children taking part in the study showed excess co-activation of antagonist lower limb muscles in standing. Interestingly, when TD children were made to stand in a crouched position like those with CP during platform perturbations, they too demonstrated excess co-activation of these muscles (43). Children with quadriplegic CP tend to recruit muscles more slowly and weakly than their TD and less affected CP counterparts (7). These particular postural characteristics seen in children with CP are endorsed in numerous studies (12)(16)(17)(18)(42)(164) (187).

Younger children with CP tend to activate the neck and spinal extensors en bloc in order to increase stability during reaching tasks but this tendency seems to dissipate from approximately four years old (12)(175). Motor planning in order to activate muscles with accurate speed and force is another frequently cited problem in CP (17)(18) and decreased abdominal muscle activation contributes to reduced balance and postural control (16). While numerous studies indicate deficits in abdominal muscle strength and activation in this population (16)(17)(18)(160), no studies could be found investigating the relative strength or activation of back extensor muscles in people with CP, making it difficult to comment on whether deficits in these muscles also exist. Increased postural sway during static standing has also been noted in CP populations (47). Poor core strength is strongly associated with an increased risk of soft tissue injury and pain in all populations (13).

The development of postural stability is essential for the attainment of developmental gross motor milestones in infants and later independent functioning. Postural control and balance requires a complex interaction of muscle synergies and sensory systems. Core stability requires balanced functioning of all of the core components to enable appropriate postural adaptations, depending on the activity. CP, by definition, involves disordered

posture and movement, thus severely limiting the ability to adequately attain the postural control necessary for physical tasks and the ability to counteract external threats to balance. The top-down muscle recruitment, delayed or in-coordinated muscle synergies and muscle weakness/decreased activation of the abdominal musculature experienced by people with CP, all contribute to postural deficits. Balance and posture-specific rehabilitation has the potential to improve task-directed activity as it has been shown that postural strategies can be learnt, if not naturally acquired.

Energy expenditure

All of the above listed morphological differences and physical impairments present in those with CP contribute to an increase in energy expenditure during activity. These factors coupled with slower movement (especially during gait), manifest in compromised cardiovascular fitness and general endurance (22)(70)(124)(132)(162)(196). Indicators of poor cardiorespiratory fitness such as poor oxygen uptake, increased heart rate and low blood lactate concentrations in people with CP have been described in medical literature (197). One could perhaps assert that the adapted biomechanics seen in CP are employed in order to reduce the physiological cost of walking.

Respiratory dysfunction

Respiratory dysfunction is widespread in children with CP, with a reduction of up to 35% in respiratory function compared to their TD peers (198)(199). The physiological contribution of the abdominal muscles to breathing is well documented and respiratory muscle fatigue has been shown to limit exercise performance (23). Due to decreased voluntary control of muscles, those with CP have been shown to have a decreased total lung capacity, decreased vital capacity and a significantly reduced functional residual capacity when compared to normal predictive values (24)(199)(200). Without adequate abdominal muscle strength, children with CP experience a weak cough making expectoration during illness

difficult (189). The impaired mucociliary clearance often found in this population may also contribute toward a greater frequency of chest infections (198).

These changes are of great consequence as the leading cause of mortality in this population is respiratory related complications (such as aspiration pneumonia and swallowing difficulties). The highest mortality rates are seen in more severely affected quadriplegics and severe cognitive impairment is directly associated with early death (5)(198)(200)(201). Difficulty swallowing liquids can lead to the need for the insertion of a gastrostomy for feeds but the aspiration of saliva in those severely affected cannot be completely eliminated (58)(198).

Increased incidence of respiratory disorders, postural discomfort due to physical impairments, aspiration and gastro-oesophageal reflux (GOR) (as the coordination of smooth muscle internally is also affected) can all lead to altered sleep patterns in children with CP. Difficulties with breathing during sleeping can lead to increased waking during the night and disturbance of circadian rhythms. This further negatively impacts cognition, concentration and general behaviour (5)(10)(108)(198)(200)(202).

It can be postulated that by improving abdominal muscle strength, respiratory function may be improved in this population, thus lessening the effect of secondary complications that lead to higher mortality rates.

Summary of impairments

As is evident from the sheer volume of research on the subject, the impairments seen in CP are many and their effect on functional independence considerable. The primary physical impairments of tone, posture and balance are seen in all subtypes of CP and morphological changes to muscle, when compared to TD study participants, are now evident. Almost all people with CP experience concomitant impairments as well. These include impairments to cognition, perception, proprioception, vision, hearing, speech, behaviour and respiration. Epilepsy is also commonly associated with CP. These impairments, coupled with the

primary physical impairments seen in CP, can lead to a perceivable worsening in the condition (despite the primary cortical lesion being non-progressive) due to the development of secondary complications, such as bony deformity and pain.

While each of the above impairments, when observed individually, have negative effects on functioning, all of these impairments have a negative effect on the development of postural control, vital to all activities and daily tasks. It is likely that addressing core strength and postural control during rehabilitation may prevent many of the secondary complications experienced, while improving independence and motor skill acquisition in CP.

2.3.2 Activity limitations

Activity limitations are those difficulties of an individual in fulfilling daily tasks or activities (130). Limitations in mobility are widely understood and almost one third of people with CP require assistive devices in order to mobilise (10)(57). As previously mentioned, in recent years there has been a move away from purely impairment-based treatment for CP as the outcomes of these interventions have little carry over into real life situations (79)(124). As secondary complications can further limit functionality over time, greater attention needs to be paid by practitioners to addressing the development of activity limitations (59)(79).

The GMFCS system discussed in section 2.2.2 is considered the gold standard for classifying the mobility of children with CP (77) as it is the only classification system that has been proven valid and reliable for use in this population (80). Its accessibility and ease of use enables both practitioners and family members/caregivers involved in the lives of those with CP to make classifications using the measure for greater ease of communication between stakeholders (80). Studies have shown the measure has a high level of acuity in predicting future functioning, with classifications on the scale made at 12 years of age generally representing the level that individual will remain at through adulthood (77)(80)(203). A study by Rethlefsen, Ryan and Kay went so far as to say that those

classified as levels I and II have an 88% chance of still walking as adults and those children in wheelchairs will remain wheelchair bound as adults with 96% certainty (77).

Gait is particularly difficult to achieve without adequate postural control as single leg stance is required during swing through (179). For those children with CP who are able to mobilise, patterns of gait differ in comparison to their TD peers. In CP, a lack of core strength is said to contribute to the typical upright posture seen which includes an increased lumbar lordosis and an anterior pelvic tilt; although this has not been proven empirically (204). Unger determined that a decreased ability to complete sit-ups was associated with an increased anterior pelvic tilt in children with CP able to stand (160).

A typical "crouch" gait is often adopted by older children with CP, as spasticity, the effects of gravity and increasing body weight with age tend to pull the hips and knees into a markedly flexed position while walking. This is usually observed in children with a diplegic distribution of CP (205). Due to spasticity, this may be accompanied by excess hip adduction and internal rotation resulting in a scissoring gait. This is further exacerbated by the inherent hip abductor weakness seen in CP (41). In those children that toe walk (due to a limited range of Achilles tendon and gastrocnemius muscle length) balance is also compromised as the base of support available is small and heel strike is not possible when mobilising (205).

Due to limited range of motion and active control at the ankle joint, those with CP tend to utilise greater degrees of hip protraction/retraction than is necessary during gait to compensate for the lack of contribution from the ankle (48). The abdominal muscles have been found to contract for longer periods than in TD children during mobility (22). This widespread muscle activation may be necessary to compensate for a lack of stability against gravity but limits the smaller postural adaptations that are possible when external perturbations take place (22)(44). The larger the threat to balance the more plainly the compensatory mechanisms used in CP can be seen (45). Such patterns present problems as the overcompensating muscles are no longer adequately able to complete the function they were originally intended for (22).

Gross motor function is directly associated with the ability to complete activities of daily living (132). Tasks such as dressing, independent or caregiver assisted transfers, washing and basic mobility are all hindered by the development of the secondary complications mentioned in Appendix III and can worsen HRQoL (50)(55). Such limitations in activities of daily living are more frequently experienced the more severe the level of CP (125). As would be expected, children with diplegic CP perform better during activities of daily living than those with quadriplegic CP and better during self-care tasks than those children with one side of the body more affected (125).

Abdominal activation and postural control fallouts experienced by those with CP have widespread implications on gait patterns, energy expenditure and the fulfilment of activities of daily living. This, in turn, affects the HRQoL of those with the condition.

2.3.3 Participation restrictions

As well as describing activity limitations experienced by those with disabilities, the ICF advocates that greater attention be paid to social integration and functionality within the environment appropriate to this population. More importantly, the focus of this framework is on ability rather than disability (2)(3)(56)(124). In order to achieve this, individuals with CP, family members/caretakers and the medical practitioners concerned all need to be involved in management and planning. In addition, psychosocial factors need to be taken into account (3)(70). This transition to a less impairment based model of treatment has been slow and a 2007 study by Engelen, Ketelaar and Gorter showed that within the ICF framework only 20.4% of 451 treatment goals set by practitioners included in their study were aimed at activity limitation or participation restriction (126). Treatment outcomes should also be assessed in real life situations as an improvement in a clinical setting does not necessarily equate to a functionally meaningful improvement in the individual's working or home environment (3).

Interviews with adolescents with CP and their parents have revealed that communication between them and strong family relationships, as well as social integration with their peers,

is vital for satisfactory HRQoL. Maintaining functional independence after leaving school was also highlighted as important to this population (206). Unfortunately, barriers to achieving this level of independence include those of the physical environment, transportation, access to appropriate assistive devices and therapeutic interventions required, as well as societal views and stigmas surrounding disability (206)(207).

It is understood that the level of physical independence possible in CP is directly associated with the level of social participation that takes place (208). Another source has claimed that while GMFCS level is associated with motor activity, the scale is not correlated to participation and this varies across GMFCS levels depending on the individual's circumstances (209). Socioeconomic factors, access to medical intervention and social support systems have higher correlations to levels of participation (125)(207).

A lack of social integration has a negative impact on the psychological wellbeing of children with CP and many adolescents and adults with CP find themselves socially isolated (26)(210). In order to ensure adequate participation is achieved it is essential that activities are planned that the child wants to do rather than those that the family or therapists think will be appropriate. These activities should then be adapted to ensure a level of success during these tasks to promote self-esteem and social interaction (3)(124)(135)(211).

A Canadian study revealed that adults with CP tend to be involved in meaningful relationships, attend university, move out of home and achieve gainful employment less than their TD counterparts. They are also less likely to take part in recreational activities (210) which negatively affects social integration and the development of a community identity (135). This study concluded by recommending that treatment and educational systems should include vocational training, psychological counselling and assistance with social integration and that these factors should be combined with the treatment of physical impairments and mobility restrictions in order to offer a more holistic service (210). Participation in leisure/outdoor and other physical activities has been directly linked to psychological wellbeing and physical development (135)(207).

Related studies have confirmed that less than half of those with CP are employed in either full or part time work and 65-88.7% have never married. Seventy-five percent of people

with CP were found to still live with their parents as adults (10)(212). The majority of those with CP cease receiving rehabilitative services after their eighteenth birthday. A possible reason for this, provided by Bottos et al. is that practitioners feel greater outcomes are achieved with early intervention (212).

In conclusion, as children with CP participate significantly less than their TD peers do, treatment needs to focus less on physical impairments and more on facilitating social integration and active participation in their environment (207)(213)(214). As has been illustrated above, all stakeholders involved need to be included in the management of those with CP and, more importantly, interventions need to be meaningful to the individual in order for relevant psychosocial outcomes to occur. For this reason, the inclusion of HRQoL outcome measures have been advocated during experimental studies in order to determine whether the intervention assessed has psychosocial as well as physical implications (56)(53)(67)(215). In order for resultant functionally meaningful outcomes, interventions should be assessed in everyday situations rather than in clinical/laboratory settings. It is for this reason that the outcome of pragmatic trials is more relevant in the field of rehabilitation (39). While impairment-based interventions may improve those specific impairments, in order for clinically meaningful changes to take place therapeutic interventions should be implemented in a more functional, task-oriented way. This will be elaborated on in the section below.

2.4 Motor learning and physiotherapy for the child with CP

Brain development continues after a baby is born, with maturation thought to continue until at least two years of age (26). Motor development does not cease at this point and, as has been discussed in previous sections, skill acquisition and refinement of gross motor function continues until adolescence (194). This natural phenomenon has led to much research in the field of motor learning in order to try to understand what processes occur in the brain to allow for the learning and accomplishment of new tasks.

This section begins with an explanation of the principles of motor learning and neural plasticity. It is followed by the development of gross motor growth curves for children with CP. Intervention techniques used in the treatment of children with CP in Johannesburg are many and varying. Those treatment modalities used most often by Johannesburg physiotherapists, in the management of CP, as well as those techniques aimed at addressing weakness and postural deficits, are discussed next. A description of the remaining techniques used can be found in the appendices. The section below ends with an explanation of electrical stimulation and its use in different conditions, its use in CP and the recommended parameters for use, followed by a section on practice schedules and the frequency of intervention in general.

2.4.1 Motor learning and neural plasticity

Neuroplasticity is the human brain's ability to adapt itself to the changing needs of the individual (87). Once neural maturation of the brain has been reached in infancy, the brain determines which synaptic connections are still necessary and the remainder are eliminated in order to refine neural processing (87). Plasticity is the ability of the brain to enable structural transformation to facilitate motor recovery and development (173). These changes include additional sprouting of dendrites, cortical re-organisation and the development of new synaptic connections (173)(216). Changes to the neurons themselves, as well as adaptations to pre- and postsynaptic functionality are also possible (173). After brain insult, new neurons do not develop but rather the circuitry of existing neurons is rerouted to compensate for damaged cortical areas unable to elicit particular motor actions (194).

The above can be explained by the "neuronal group selection theory" where genetics determine how neural networks develop (in terms of cell division, cell migration and dendritic sprouting or death) into interconnected neuronal groups each responsible for particular motor outputs (194). The process of neural development is then further established by somatosensory input from the environment (164). The "dynamic systems theory" takes this a step further by explaining neuroplasticity as an interaction between sensory, psychological, musculoskeletal and environmental factors (194).

As has been demonstrated in neuroimaging studies on adult stroke patients, neuro- and cortico-genesis occur following the brain insult experienced as an attempt to try to re-establish synaptic connections in the brain (87). Motor learning can be defined as "the acquisition of new skills with practice" (69).

While neural plasticity occurs due to insult/injury to the brain as described above, it may also occur due to varying sensory experiences and through repetition and learning (90)(173)(216). The brain adaptations that take place occur in response to the specific environmental circumstances of the individual (217).

Physiotherapy can enhance this process by providing the opportunity for repetition of movement and the teaching of specific skills (218). The practitioner should also allow for exposure to external sensory experiences, otherwise not encountered, to further promote motor learning (90). When a task is not yet independently achievable, therapists can assist with the motor learning process by facilitating/guiding the movement required to achieve the expected goal. With repetition the brain begins to recognise the motor demands of the task and the muscle synergies required and hands-on facilitation can progressively be withdrawn (178)(219). In order for these synergies to be retained for future application, both internal and external feedback of success is needed (69)(173). Without the physical experience of the activity, one cannot interpret the requirements of its successful completion and so practitioner demonstration of the task does not suffice (220).

As CP is a disorder of the developing brain, it occurs at a time when the potential for neuro-plastic changes are most likely (194)(216). It is for this reason that early intervention ensures the best possible functional outcomes in children with CP (22)(216). Cortically, the greatest changes are seen when skill development is goal-directed (i.e. relevant/meaningful to the particular individual with a specific and attainable goal) and when practice takes place over time (71)(75)(90)(221). Such therapy may enable new synaptic connections to develop allowing motor outcomes that the damaged areas of the brain are not able to generate (90). The more active the participation of the child in the task at hand, the more likely that compensatory neural mechanisms will become established (75)(194). The massed practice of functional, task-specific activities, by those with CP, has been shown to be effective. The active engagement of the participant and intensive repetition of such

activities leads to the development of the motor plans/programmes otherwise absent in such subjects by reinforcing neural pathways, and many professionals now advocate for quantity of movement over quality of movement (222).

In Bar-Haim et al's 2010 study, 78 Israeli children with CP were randomly allocated to two groups; one receiving Neurodevelopmental Therapy (NDT) and the other an experimental group receiving therapy based on motor learning. Both treatment groups made significant immediate improvements in gross motor function but while at six months post-intervention the NDT control group demonstrated deterioration in GMFM-66 scores, the intervention group displayed stability in their scores up to nine months following the motor learning intervention (69). This treatment included the practice of goal-directed activities that were meaningful to and selected by each individual, feedback provided by different means during and after the task for knowledge of results, intermittent cognitive tasks and changes to the environment in which the task was completed (69).

In order for tasks to be meaningful to the individual and for motor learning (and thus the development of new synaptic connections and retention of motor improvements) to occur, intervention needs to be goal-directed, task orientated, and the individual needs to be an active participant. It can be surmised that treatments aimed at improving impairments experienced by those with CP, introduced during functional, goal-directed activities, may provide greater opportunities for motor learning to take place and thus, ensure better functional outcomes of treatment.

2.4.2 Motor development growth curves

In 2002, Rosenbaum et al published an investigation that revolutionised the ability of practitioners to prognosticate about children with CP (101). This study tracked the progress of 657 children with CP, under the age of 12 on acceptance into the study, for almost four years. Participants were part of the West Sweden CP register and were recruited through the ambulatory rehabilitation centres at which they all received regular treatment in Ontario, Canada (96)(115). Treatment received by participants at these facilities included developmental interventions such as physiotherapy, occupational and speech therapies, medicinal and surgical intervention and the prescription of orthoses, when indicated (96)(105). As these centres are publicly funded, the authors concluded the sample was

representative of the greater CP population, as the majority of affected children had access to these facilities (101)(105). The children were assessed repetitively using the standardised GMFM-66 (which will be discussed in detail later in this literature review). Participants were stratified based on their GMFCS level of motor functioning as well as their age, making it possible to create a predictive "motor curve" for each level (101)(115). Each of these curves was found to be significantly different from the other (105). As the large sample was deemed representative of the tested population, the authors inferred generalisability to a much wider population (105). These curves provide a platform from which to base comparisons for all practitioners globally and the study has been widely accepted and adopted (101)(105). The curves created allow greater ease in communication with families regarding their child's prognosis and a way to monitor the success or failure of therapeutic interventions (96)(105)(216).

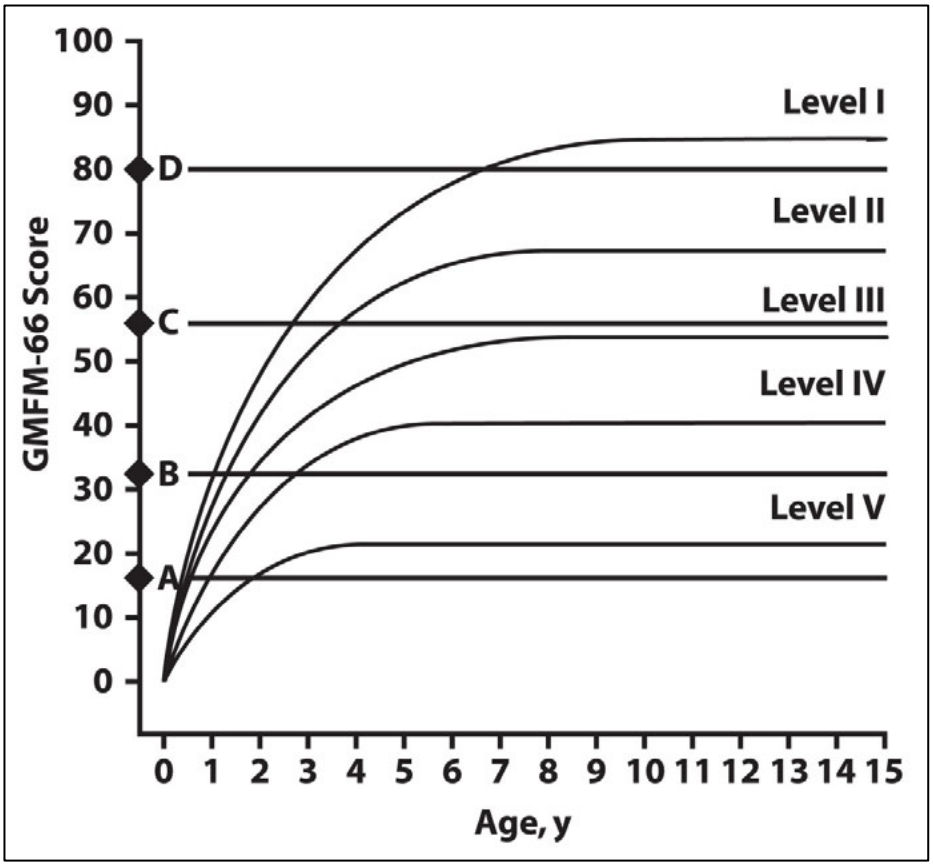


Figure 2: GMFM-66 developmental gross motor growth reference curves (adapted from Hanna et al (2008) (101))

As can be seen in Figure 2 above, basic assumptions regarding the motor abilities of a child with CP can be made based on the motor curves. Once classified using the GMFCS, one needs only find the child's age on the graph and the relevant GMFCS line graph will give an indication of the expected GMFM-66 score, as a percentage, up to a 95% confidence interval (101). This can be done visually by assessing the graph or for greater accuracy by consulting the tabulated percentiles laid out by the authors (101). As can be seen on the graph, at birth all GMFCS levels are unable to perform any of the items on the GMFM-66 outcome measure. The period of greatest gross motor development for all levels is during infancy, with development eventually levelling off as individuals reach a ceiling to their potential skill attainment in early adolescence (103)(105)(106). As would be expected, the less severely affected children have the greatest potential for improvement (223).

As no child is the same and as the presentation of CP is not stable, variability from one individual to another is inevitable. Performance may undulate and this should not automatically be associated with clinically significant effects of treatment (101). It has been proposed that up to 90% of children with CP reach their motor potential by five years of age with a generalised plateau in progress seen by age seven (96)(105). The same is seen in typically developing children (221).

Between 1981 and 2002, 317 of the 409 children born with CP listed on the West Sweden CP register took part in a study assessing the natural history of CP. The authors found that 75% of the population classified at GMFCS level I was able to accomplish 90% of the GMFM-66 items by the age of five, with a large proportion reaching 100%. Many of these children (most of who were classified as having hemiplegia) reached similar scores to their age-matched TD peers. Those at level II closely followed these scores with the majority of this category having a diplegic distribution. By age seven, in general, the remaining groups were able to complete 80%, 30% and 20% of the outcome measure for levels III, IV and V respectively, plateauing after this point (96).

The attainment of independent sitting has been strongly associated with the potential for independent gait, with 90-100% of those sitting before the age of two ambulating independently later in life and only 50% of those sitting between 2-3 years having the probability of doing the same (70). The most severe two groups also show a tendency

toward gross motor decline over time (50)(115). It could be speculated that this is due to secondary complications and an increase in body weight with growth rather than deterioration in the condition itself (115). Accelerated decline in walking ability and in physical impairments occurs from adolescence into adulthood (212).

A further study, by Hanna et al, tracked the progress of the original participants following the Ontario study. The curves that were developed, as these children aged, established that those with a classification of a level I or II on the GMFCS plateaued as was previously shown, and a steady decline in GMFM-66 scores for GMFCS levels III-V as children with CP enter their teenage years was noted, which continued into adulthood (115).

The developmental gross motor growth curves established by Rosenbaum et al have created a globally accepted way of communicating between therapists and predicting the motor outcomes of children with CP, of varying severities, over time. The curves have demonstrated that children make the greatest gains in motor functioning during infancy and thus one can advocate for the need for early intervention in order to achieve the greatest therapeutic outcomes possible. The fact that these curves show most children plateau in their attainment of new motor skills by the age of seven has led to debate surrounding whether or not therapy should continue into adolescence and adulthood. While further improvements may not be possible, the curves have also demonstrated that, with secondary complications due to primary impairments and environmental factors, a steady decline in function may occur over time. For this reason, one might assert that therapy should be a lifelong service offered to those with CP in order to maintain physical functioning, promote independence and social participation, and thus prevent the development of secondary complications. These curves were developed based on the repetitive assessment of children with CP receiving regular therapeutic, medical and surgical intervention of a high quality, as needed, from a young age until adulthood. For this reason, it may be proposed that such curves might not accurately represent the prognosis of children with CP in low-income countries, where early intervention is not always possible, resources and professional expertise are limited and intervention may not continue beyond a certain age.

2.4.3 Intervention strategies

The above outcome measures and the predictive curves based on them have allowed for more specific planning for therapeutic intervention for CP (101) and goal attainment and progress should be tracked based on the expected motor outcomes for each specific child's age and gross motor level (103). The World Confederation for Physical Therapy claims the role of this profession is to "develop, maintain and restore maximum movement and functional ability throughout the lifespan" (224).

No treatment exists capable of reversing the cortical damage present in CP and thus it can be categorically stated that no cure exists (90). As discussed in section 2.4.1, in order for treatment outcomes to have clinical and functional carry over into real-life situations, goals set need to be meaningful to each individual (70). Based on ICF concepts, a global move toward family-centred treatment has occurred with emphasis placed on the individual and relevant family members playing an active role in goal setting and the management options chosen (70)(87). A person is more motivated to work toward achieving a goal when they themselves have outlined the activity as something of importance to them (126). Goals set should always be specific, realistically attainable and measurable in order to monitor progress (225)(226). A general consensus has been met that early intervention leads to more promising outcomes of treatment (26).

Physiotherapists play a central role in the multi-disciplinary team involved in the management of children with CP (68) and therapy is usually ongoing throughout childhood (227). With the integration of the ICF into many physiotherapy practices there has been a move away from solely focusing on the maintenance of body functions and prevention of secondary complications at impairment level only, with a greater emphasis placed on participation and accomplishing maximal functional independence (67)(68)(69)(138). Previously, therapy strictly followed a system of trying to attain developmental milestones in the chronological order in which TD infants attain these gross motor skills. Waiting for a particular milestone to be reached before progressing to work on other proficiencies is unrealistic, as children with central nervous system (CNS) disorders may never reach certain milestones but may still be able to achieve higher functioning tasks with

adapted/compensatory patterns of movement (124). If therapeutic activities set are not age-appropriate and child friendly; goals will not be realised (228).

Any treatment modality adopted should be based on scientifically proven data (25)(143)(229). This is made difficult by the vast contradiction evident in current literature either supporting or negating the use of each intervention available for the treatment of CP (27). This is confounded by the lack of research available of sound methodological quality (48)(230) and systematic reviews have been constructed to try and bring together all of the available literature based on certain techniques to try and improve practitioner understanding (25)(48)(231). Numerous treatment options exist but as each child with CP presents differently, the choice of modalities needs to be made based on each individual's primary difficulties (232).

CP intervention can be broken up into rehabilitative techniques, medical treatments and surgical procedures (87). As pragmatic trials require that an experimental intervention be compared to 'typical/routine' intervention (see section 2.6), a checklist of possible techniques, for the rehabilitation of children with CP, was created by the researcher (Appendix IV) for use in this study. This will be discussed in detail in section 3.1. As was explained above, those popular therapeutic interventions, with an emphasis on improving gross motor function, strength and balance, included in the checklist, are described below. Other techniques employed by physiotherapists in Johannesburg, as well as a summary of medically/surgically based interventions, alternative therapies and other therapeutic modalities that have been not been included in the intervention checklist (as they do not form part of typical physiotherapy intervention) can be found in the appendices proceeding the conclusion of the study (Appendix V).

2.4.3.1 Neurodevelopmental therapy

Since its inception in the 1940s, neurodevelopmental treatment/therapy (NDT) (more commonly known as Bobath therapy after its founders Dr. Karel and Mrs. Bertha Bobath)

has become the most widely accepted and administered system approach to the management of children with CP (169)(178)(219)(233).

In her commentary on the evolution of NDT, Margaret Mayston describes this treatment approach as a way of thinking and analysing rather than as a specific treatment technique (48)(170). The NDT trained individual is expected to use their expertise in analysis of movement and critical reasoning to formulate treatment goals specific and functionally meaningful to each individual. Any intervention performed in order to achieve these goals should be based on empirical evidence (170). In 2000, it was estimated that 88% of physiotherapists working with patients with CNS disorders in the United Kingdom were NDT trained (170).

Initially the Bobaths surmised that the motor deficits seen in children with CP were purely due to their physical impairments and that if one could "inhibit" hypertonicity, spasticity and abnormal reflex activity then positive changes would carry over into functional mobility (170)(178). However, hands on techniques cannot inhibit neurotransmitter release to effect cortical changes, and facilitatory techniques used in NDT work rather on the visco-elastic properties of muscle and stretch reflexes (170). Thus the initial static "reflex inhibiting postures" children with CP were placed in to reduce spasticity were abandoned and replaced by "tone-influencing patterns" of movement that are facilitated using key points of control on the body to try to elicit more natural and effective patterns of movement (25)(178). These techniques still did not amount to functional overflow into real-life situations though and improvements seen ceased after handling was withdrawn. Consequently, current NDT based ideas now encourage greater active participation of the individual during treatment (25)(233) and practice and repetition of tasks is necessary to ensure retention of realised goals (170) with an emphasis on play in young children (178). One can consider the NDT process as being initiated by preparation of the body through stretching and facilitation, leading into more active functional tasks (169).

Handling techniques used include facilitation of transitions between functional positions, facilitation of repetitive movements with assistance, facilitation of weight shifts to co-activate trunk muscles and facilitation of trunk rotation vital for developing equilibrium and postural reactions (173)(234). Once motor learning begins to take place, the degree of

hands-on facilitation provided is withdrawn to begin to enable independent achievement of functional movement (234). The premise behind these techniques is the supposed promotion of "normal" motor and postural development and the prevention of secondary complications at impairment level (25)(27). Constraint-induced therapy (CIT) has long been incorporated into NDT treatment in order to facilitate compensatory mechanisms and forced use of the more affected side (170). For more details on this modality, please see Appendix V.

In order for studies evaluating treatments to be of a high standard, they require randomisation of trial situations as well as large sample sizes to allow for generalisability (226). As can be seen above, NDT is an approach that encompasses a multitude of treatment ideas and techniques. As CP is a heterogeneous group of disorders, with presentation varying widely, it is exceptionally difficult to conduct a study where each practitioner would use the same techniques as one another or even where a single practitioner would use the same treatment protocol for two different subjects with CP (229). It is also difficult to account for external variables such as environment, social participation and other therapies that may also affect motor performance in this population during studies (229). Further confounding study results are wide variances in teaching methods employed during NDT training throughout the world and the fact that it is a continuously changing concept so course content varies depending on when the course was undertaken (27)(173)(233). Studies assessing the effectiveness of NDT have also not been standardised in terms of treatment length and frequency, making comparison between studies difficult (25). The aspects that are globally agreed upon though, are the need for children with CP to be active participants in therapy and that there is a need for long-term management of this population in order to maintain progress made and continue to prevent secondary complications from arising (27).

In 2001, the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM) released an evidence report evaluating research available on the NDT approach. Twenty-one studies were included in their systematic review and were analysed based on the quality of each study. Small sample sizes were utilised in most studies and power calculations were generally not listed, so any positive effects should be interpreted with caution. Eighty-six of the 101 studies demonstrated no significant effect of NDT over other

modalities and a number of studies showed greater improvements in the control group. Therapy administered more frequently did not appear to improve functional outcomes. Only eight studies demonstrated statistically significant gross motor improvements and while all other outcomes showed inconclusive or contradictory results, findings for improvement in joint range of movement were generally positive (25). A 2010 review of physiotherapy interventions available for the treatment of CP had similar findings (68).

NDT is the most widely accepted and implemented approach to the rehabilitation of children with CP globally. It is a system approach encompassing many, varying treatment techniques based on the facilitation of active movement to assist motor learning and skill acquisition. Handling techniques focus on the attainment of functional, goal-directed activities through facilitating transitions between postures, abdominal muscle activation and weight shifts/postural adjustments. As the approach is constantly changing, based on changing neurological evidence, and as the approach to teaching professionals varies widely throughout the world, the parameters included in studies assessing the treatment modality are inconsistent, making comparison between studies difficult. Small sample sizes or poor methodological reporting further compounds this problem. While this intervention continues to be the most commonly used modality in the treatment of CP, a lack of convincing empirical evidence supporting its effect leaves room for the need for further, more rigorous, quality study on NDT or the need for new and alternative, effective treatments to replace its use.

2.4.3.2 General limb strengthening

As has been discussed throughout this literature review, weakness is a primary deficit in children with CP. Speculation exists that if strengthening of weak muscles were possible in this population then functional improvements may be associated (235). Previously, strengthening in CNS disorders has been strongly discouraged, as it was believed that such exercises would worsen spasticity and muscle tightness and that the requirements for strengthening could not be fulfilled due to difficulties with selective motor control experienced by this population (26)(151)(205)(236). Current literature dispels this belief as

no change in spasticity has been observed after resistance training trials in numerous studies involving people with CP (152)(158)(171)(236).

The American Academy of Pediatrics claims that strengthening in adolescents can be effectively used as long as safety precautions are adhered to and submaximal resistance is added (especially in those with disabilities) (205). Authoritative bodies have stated that strengthening can begin as early as a child is able to take part in sporting activities (i.e. as young as six years old) (159)(237). Damiano, Dodd and Taylor noted that repetition of movement is not enough to gain changes in muscle strength and individuals with CP require principles of muscle loading and resistance in order to improve muscle torque, as do those who are TD (151). A 2005 summary of systematic reviews on the topic noted strengthening programmes used in CP selected a few specific exercises (that could be performed using free weights or gym equipment)(236) and 5-10 reps of each activity were performed in sets of 3-4, at 50-65% of maximal output. These studies noted promising results of strength training programmes in this population (236)(237). Resistance should be increased progressively as strength improves for the best possible outcome (152)(238)(239). Positive changes in torque have also be seen when programmes include exercising until fatigue (236)(238)(240).

In their 2013 randomised control trial (RCT), Taylor et al recruited 48 children between 14 and 22 years old who were randomised into equal groups. Half continued conventional therapy while the experimental group took part in a biweekly specific progressive resistance-training programme for 12 weeks. The study was of high methodological quality and results can be generalised for children of GMFCS levels II and III with confidence. While participants showed statistically significant improvements in targeted lower limb muscle strength post intervention, these improvements did not translate into improvements in mobility parameters (235). Recent reviews have echoed this finding with significant improvements in strength seen post intervention with no carry over into improvements in functional performance (68)(152)(158)(171)(205)(236)(238).

Numerous studies exist portraying the positive effects of strengthening programmes in CP but most do not specify the exact programme used, making comparison between studies and prescription of strengthening parameters difficult (152). The only possible side effect of

progressive resistance training noted is transient muscle soreness following exercising (156)(152)(236) and no negative effect on growth (237) or spasticity (152)(158)(171)(236) is associated with strengthening as was previously thought. As weakness is such a vastly limiting impairment seen in most, if not all people with CP, positive gains in muscle strength observed in most experimental studies in this population are encouraging. The need exists for intervention protocols to incorporate goal-directed activities so that gains in strength might also equate to gains in mobility and general independent functionality as well.

2.4.3.3 Core/abdominal muscle strengthening and posture-specific/balance training

Core stability has been explained at length in section 2.3.1.1. As the core is seen as fundamental to the body's stability and to all limb movement, much research has surrounded ways in which to strengthen this group of muscles. Most of this research has been in the field of musculoskeletal rehabilitation and sports conditioning (181). Despite the popularity of core strengthening programmes, little evidence exists to support the use of techniques such as plyometrics, perturbations, concentric/eccentric/isometric abdominal contractions or balance-specific training (21)(29)(31).

The assumption exists that improvements in core muscle strength will translate into improved upright sitting or standing posture in children with CP (38). It has been proposed that reconditioning should begin with teaching the individual how to actively recruit inhibited muscles in the appropriate sequences before strengthening can begin (31). Voluntary contraction of the abdominal muscles needs to be at greater than 60% of the maximal possible contraction in order for hypertrophy of the muscles and neural activation to take place (31). Unfortunately, no one exercise exists that is able to activate all the muscles of the core simultaneously so training programmes need to contain varied, adapted resistance activities for the greatest strength outcomes (154)(181). Activities proven to activate each of the abdominal muscles individually include sit-ups or 'crunches' for the rectus abdominis, sit-ups with additional trunk rotation for the EO and IO muscles

and drawing-in activities for the deeper TA (154). Exercises should start in the most stable supine and prone positions with the base of support slowly decreasing to four-point kneeling and eventually standing during training. To further increase the postural demands of activities, strengthening exercises themselves may be performed on an unstable surface (13). Such surfaces include wobble boards, balls, rollers etc., adding the stimulation of different proprioceptors during balance rehabilitation (181).

Another way of training balance is to impose an unexpected external perturbation to provide the recipient with the opportunity to learn the necessary postural reactions to maintain the body within its base of support (172)(177). This can be done in a number of ways, either with a manual hands-on approach or by the machinery often used in balance studies. This includes a handle held that pushes and pulls on the upper limbs or a moving platform that rotates or tips in different unanticipated directions (172)(177). Studies exposing TD infants to up to 100 perturbations daily have shown significant improvements in direction-specific postural adaptations and the use of accurate muscle synergies to counteract the specific balance perturbations applied (18). Similar results were found for studies including participants with CP (18). Unger found that a four-week trunk targeting intervention, using whole body vibration for prepubescent children with CP, resulted in significantly improved abdominal muscle strength (as measured by timed sit-ups) (160). No studies could be found investigating resistance training for the abdominal/core muscles specifically in children with CP.

It is important to note that when a more functional, meaningful outcome/target was added to balance training, postural adaptations and reaching distance were greater (47)(220). Abdominal strengthening can also improve functional forced vital capacity in children with CP; positively influencing expiratory function (191).

Poor abdominal muscle activation has been closely linked to postural deficits in children with CP. While no studies exist investigating resistance training for the abdominal muscles specifically, in this population, significant gains in limb muscle strength with resistance training, and improvements in strength of the core using vibration therapy and external perturbations, provide evidence that abdominal muscle strength gains may be possible in these children with resistance training programmes. If one can improve what is considered

to be the most significant impairment in children with CP, then functional gains may be possible in this way and secondary complications may be avoided if such intervention is regularly provided.

2.4.3.4 Summary of treatments

Numerous approaches to management and techniques for treatment of CP exist but very little concrete evidence is available to recommend one over another. Regardless the treatment modality, the individual should always be considered when selecting a means of management and possible positive functional outcomes/patient comfort, etc. should always outweigh possible side effects (93). While NDT is the most widely employed treatment system for CP globally, studies on its effect are inconsistent as its implementation varies widely from professional to professional and the concept is constantly evolving, making comparison between studies difficult. Many studies have indicated no significant improvement in gross motor function with intensive NDT intervention compared to traditional treatments. The use of passive stretching, CIT, orthoses, Vojta therapy and conductive education have different facilitative, educational and supportive aims but all have in common the fact that little, if any, scientific evidence exists to support their use (see section 6.5.1 Rehabilitative treatment modalities). The same can be said for complementary and alternative therapies used in the rehabilitation of children with CP (see section 6.5.4)

Medical and surgical interventions (discussed in section 6.5.2 and 6.5.3) for the treatment of CP can lead to significant improvements in spasticity and the prevention of secondary impairments, yet all carry severe side effects, one of which being excessive weakness. The only convincing positive outcomes found, on a review of available literature, were for those rehabilitative interventions aimed at improving muscle strength. Resistance training programmes have seen statistically significant improvements in limb muscle strength and preliminary studies on external perturbations and vibration therapy have shown encouraging improvements in abdominal strength with treatment in CP. As well as improving cardiovascular function and psychosocial factors, gait training, exercise therapy

and hydrotherapy have also demonstrated improvements in strength in those with CP. As has been reiterated throughout this review, weakness is a prevalent and severely disabling impairment experienced by those with CP. It is not surprising, then, that so many currently used treatment modalities have improving this impairment as their aim. Many of these techniques require expensive equipment (e.g. treadmills, vibration plates, hydrotherapy pools) and thus a need for a more practical, cost-effective and easily administered intervention for the strengthening of those with CP exists.

2.4.4 Functional Electrical Stimulation as a rehabilitation tool

Surface electrical stimulation (ES) can be defined as the application of an alternating current to a nerve supplying an impaired muscle. This stimulates an electrical impulse which travels to the affected muscle mimicking an active muscle contraction (33). Neuromuscular electrical stimulation (NMES) is a term used for any application of great enough amplitude to elicit a visible muscle contraction. Muscles require intact innervation for this modality to work (37). Cyclic NMES is generally a passive treatment whereby the affected muscle is electrically stimulated at a set duty cycle, over a specific period of time, without voluntary or active movement of the stimulated muscles by the participant. This intervention is based on the assumption that the electrical stimulation causing repetition of movement stimulates the somatosensory cortex, which, in turn, causes long-term potentiation in the motor cortex allowing for motor learning to take place by changing the excitability of motor neurons (241).

In order for ES to be classified functional electrical stimulation (FES), the above muscle contraction needs to take place during a meaningful functional activity (32)(33)(242)(243). This stimulation may be timed specifically to contract when that particular muscle is meant to contract during a task, but not necessarily so (71)(244). FES has also been described as the stimulation of impaired muscles in order to augment task-specific movement to improve motor control with time (32)(245). By achieving a muscle contraction during functional activities it is believed the muscle may 'learn' how to contract actively over time and strength gains may allow for the weaning off of machine use with time (246).

The mechanism by which ES can improve muscle strength is assumed to be the overload principle which increases muscle size and selective type II muscle recruitment to improve synaptic potential of the stimulated muscle (33)(71)(242). A 2013 study investigating FES use for drop foot and improved gait parameters in 14 ambulant children with CP confirmed this by demonstrating statistically significant gains in gastrocnemius muscle size when compared to baseline measurements and measures of the untreated side following FES intervention (245). Application to the antagonist muscle groups may reduce spasticity in the agonists via reciprocal inhibition (35).

Threshold electrical stimulation (TES), on the other hand, is a low threshold current stimulating a subcontraction that can be administered over long periods and while asleep. It has been proposed that stimulation during the release of hormones whilst sleeping can aid circulation and promote muscle bulk (33)(71)(242). Merrill affirmed, in his 2009 review, that the few studies available investigating TES are inconclusive (71). In Sommerfelt et al's randomised, controlled crossover trial, 12 children with CP received nightly TES for 12 months. No changes in spasticity or muscle strength were noted when applied to the lower limb antagonist muscle groups (247). These results were mirrored by Dali et al in their double-blind RCT (248). Steinbok, Reiner and Kestle did note significant improvements in GMFM scores in an experimental group of 20 children receiving nocturnal TES but, again, no strength gains or changes in spasticity were seen (249).

Transcutaneous electrical nerve stimulation (TENS) is a modality more often used for pain relief, but can be administered passively at home to reduce spasticity and increase blood flow in distal muscles (250).

For all electrical modalities, electrical current is administered through self-adhesive electrodes of varying shapes and sizes applied via leads to the skin (251). Contraindications for use include implanted pacemakers, open wounds and cancerous tumours and ES should not be applied over the temples or neck (252). Care should be taken to monitor for worsening of spasticity or epileptic symptoms and possible skin reactions to the intervention. Therapy should cease should these side effects not subside

(244)(251)(252)(253). A slight reddening of the skin under the electrodes following stimulation is deemed normal (251).

ES has been used in the rehabilitation of numerous neurological disorders, including spinal cord injuries, stroke, multiple sclerosis, Parkinson's disease and traumatic brain injuries, in an attempt to reverse the effects of muscle atrophy and regain voluntary motor control (217)(244)(254).

FES is most commonly used during gait training, with application over the dorsiflexors to allow for heel strike and swing through, not possible due to the frequent equinus deformity or drop foot seen in CP and strokes (244)(246). This application can act as a substitute for more uncomfortable and limiting rigid orthoses aimed at achieving the same functional outcome (255). Two RCTs using the Odstock Dropped Foot Stimulator (ODFS) in stroke patients demonstrated significant improvements in walking speed and up to 25% decreases in the physiological cost of walking over the control group. Improvements seen ceased immediately once the ODFS was removed (244). A third study also showed significant improvements in dorsiflexor muscle strength and notable reductions in ankle spasticity following use of the ODFS (218).

NMES used in a RCT over the abdominals of adults with no neurological fallout demonstrated a 100% increase in abdominal isometric strength and endurance in the experimental group, a 72% greater improvement than the control group. No other changes in body composition parameters were seen following intervention (256). A case study following a C3/4 tetraplegic patient revealed that daily use of abdominal ES, timed for the end of inspiration, improved his chronic postural hypotension by increasing blood pressure and peak expiratory flow sufficient enough to stimulate a cough not otherwise possible (257). These results were confirmed by studies by Zupan et al, Gollee et al and Linder, who also noted improvements in resting tidal volume and maximum expiratory pressure with abdominal ES in patients with spinal cord injuries (189)(190)(258).

Reviews of such studies warn that caution should be exercised when interpreting these results as bias may have been introduced to many studies due to poor methodological design and quality (259).

In a 2009 RCT, a large sample of children with CP was divided into a control group and an experimental group receiving NMES to the gluteus medius muscles during gait. A further control group of TD children was added and this was one of the few studies available listing their treatment parameters in detail. After three different management programmes using the NMES were completed, results showed significant improvements in walking speed, stride length and hip adductor muscle tone. The authors explained the mechanisms behind these improvements as reciprocal inhibition and increased blood flow to the abductor muscles (41).

Other positive changes noted by studies, after the use of NMES/FES for the treatment of those with CP, include significant increases in dorsiflexion range of movement passively and during the swing-through phase of gait (32)(33)(255)(260) and wrist extensor strength and range of movement, as well as bilateral hand function (36)(261)(262)(263). Anecdotal, and not necessarily significant, improvements cited include decreased falls, increased participation in sporting activities (35) and increased awareness of the more affected limb (36) following periods of ES.

In contrast, studies have demonstrated no significant difference in quadriceps (242) or gluteus maximus strength (264), walking cadence or stride length (34) or gross motor function (264) following these interventions.

The only study found evaluating the effect of ES over the core muscles included 26 infants with CP. Both control and experimental groups continued with their usual NDT-based therapy and the intervention group received concomitant ES over the abdominals and posterior back muscles simultaneously for 30 minutes, six days a week, for the duration of the six-week study. The ramped delivery of ES was set at a 35 Hz frequency, 250 μs pulse width and intensity was adjusted based on each child's individual tolerance levels. Following the intervention, improvements in the sitting dimension of the GMFM and radiographic spinal kyphotic angle were significantly greater than those improvements seen in the control group. Only the experimental group experienced significant lessening of Cobb angle as well (i.e. the magnitude of curvature present in the spine). Park et al

hypothesised that these improvements in posture were due to improved abdominal strength and control due to the ES intervention (38).

While many studies exist investigating the effect of ES in children with CP, generally positive findings cited are difficult to accept, based on the lack of controlled trials available and the lack of information presented on the parameters or outcome measures used. The statistical power of these studies is also questionable due to small sample sizes included (32)(71). Most of these studies note that any positive treatment effects encountered are not maintained after cessation of the intervention (71)(263)(265).

Unfortunately, most studies found that investigated the use of ES in children with CP do not make mention of the particular parameters used when ES was administered (243). Those studies with statistically significant positive findings employed frequencies of between 20 and 40 Hz and a 50-350 μs pulse width (38)(41)(249)(260)(262). In contrast, no improvements were observed in studies using a 40 Hz frequency with a 300 μs pulse width (247), 35 Hz and only 1-5 μs (248) and 32 Hz with 300 μs (34). Maffiuletti has suggested that stimulation frequencies need to be between 50 and 100 Hz and output intensities as high as possible in order for muscle strength changes to take place (243), but no studies exist to illustrate whether such high parameters are either safe or tolerable in children (33).

Despite conflicting research outcomes, interest in the use of ES in the rehabilitation of various CNS disorders continues, as it provides a cost effective, non-invasive and easy to use intervention that can also be applied at home and during daily activities (27)(33)(248)(249)(266). As was mentioned in the summary of available treatment techniques for the rehabilitation of CP above (section 2.4.3.4), weakness and a lack of postural control are considered the most disabling impairments present in CP. Preliminary studies on the application of NMES/FES have shown significant improvements in dorsiflexor and wrist extensor muscle strength in children with CP and a 100% increase in abdominal isometric strength and endurance in adults without neurological fallouts. The only study available investigating the use of ES over the abdominal muscles of children with CP demonstrated significant improvements in postural outcomes and gross motor function, accounted for by the authors as improvements in abdominal strength and control.

Additional clinically significant changes seen during these studies were increased walking speed, stride length, physiological cost of walking, dorsiflexion and quadriceps range of movement, respiratory function, hand function and reductions in spasticity. As with any treatment modality, studies always exist with opposing outcomes and due to a general lack of quality methodological design, it is difficult to either advocate or discourage the use of ES in this population with confidence. With this said, the numerous impairments possibly addressed by ES makes it a plausible adjunct to current therapeutic interventions. Added to this, few, if any, contradictions and side effects of this modality exist.

While NMES elicits a muscle contraction over the areas stimulated, the added contraction during meaningful functional activity of FES may lead to motor learning with repetitive use and thus, as well as the improvements in parameters listed above, may also lead to improvements in independent function. With so many possible beneficial results and the ease of application, lack of side effects and cost-effectiveness of FES, the need exists for studies of greater methodological rigour on the intervention to take place in order to clarify whether its use should be added to routine physiotherapy intervention.

2.4.5 Practice schedules, frequency and structure of treatments

As with many of the treatment techniques available for the use in CP, studies on practice schedules and the recommended frequency of treatment vary greatly. Numerous systematic reviews exist which collate research studies on the subject and the general consensus is that there is no statistically significant difference in physical or functional outcomes based on the intensity of therapy given (25)(68)(225)(267). These results can be regarded highly, as study designs and statistical power have improved greatly in such studies in recent years (68). There is uncertainty whether any positive effects seen in certain studies offering high intensity therapy are maintained after these periods of high frequency and the practicality and cost of administration needs to be considered before implementation (225)(267)(268). Martin, Baker and Harvey proffered that the greatest confounding variable affecting all CP studies is the lack of detail provided on what “usual” therapy entails when control groups continue to receive their accustomed or “usual”

therapy during the study (68). Despite the lack of sound scientific evidence to support the use of more intensive intervention, many professionals still, feel that the neural reorganisation taking place following repetitive, task-orientated activity warrants more intensive therapeutic input (222).

Quality systematic reviews have determined that intensive physiotherapy does not lead to greater functional outcomes than more traditional weekly or biweekly physiotherapy sessions. In order for the results of any intervention study to be interpreted accurately, the baseline typical/conventional therapy received by control groups needs to be explained in detail in the study write up.

2.5 Outcome measures

In the following section, the general psychometric properties inherent in scientific research design are explained, followed by descriptions of standardised outcome measures available for the measurement of gross motor function, balance, abdominal strength, lung function and HRQoL. In each instance, the options available for the measurement of these factors in children with CP specifically are mentioned, ending with a description of the specific outcome instruments selected for inclusion in this study.

2.5.1 Psychometric properties

In order for an outcome measure to be considered worthy of use in scientific, evidence-based study, certain psychometric properties need to be inherent. These include validity, reliability and responsiveness (33)(126)(215)(269)(270). Reliability explains obtaining similar outcomes after repeated administration of a test, if the characteristic being measured remains constant (56)(128). This can be further broken down into test-retest and intra- and interrater reliability that infer stability in different testing circumstances (139).

Validity, in general terms, demonstrates that an outcome measure is able to measure that which it was intended for (128) (i.e. content validity) (104). More specifically, construct validity looks at whether the instrument actually measures characteristics specific to that subgroup of people (56). Concurrent validity determines whether different measures evaluating the same health outcome are correlated to one another (271) and criterion-related validity is used to ensure a new measurement tool and the gold standard for that characteristic are in agreement with one another (104). Finally, responsiveness describes the sensitivity of a measure to detect real or clinically meaningful changes in outcome determinants (269)(272)(273).

Measurement tools can be divided into three subgroups, depending on what they are intended to measure. A discriminative tool differentiates between individuals with or without certain characteristics, predictive tools categorise individuals based on their prognosis and evaluative tools are those generally used in RCTs to determine change in functional status following an intervention or over a particular period of time (128)(269)(274).

Other factors that should be considered when selecting a standardised outcome measure are the specific sample to be investigated, funding availability and the practicality of its use for the specific study outlined (128)(228)(271). Certain measures may have floor or ceiling effects, where the most and least severely affected participants may achieve the lowest/highest possible score on the measure respectively, despite their functional abilities varying greatly (271)(275).

As so many different outcome measures for the evaluation of children with disabilities exist, one needs to take all of the above into account before making the appropriate selection. Evaluation tools should also be selected to fulfil all aspects of the ICF framework (3).

2.5.2 Measuring impairment

2.5.2.1 Measuring balance

Laboratory based studies on postural control often cite electromyography (EMG) as their primary outcome measure for balance related changes, following postural interventions (276). In their 2008 study, Zaino and McCoy determined that centre of pressure kinetic measures of postural control have superior test-retest reliability to EMG measures in a sample of children aged 8-14 (276). Such measures include computerised moveable force plates on which participants stand, as described in section 2.4.3.3 (11)(277). Measures may be calculated for the time it takes an individual to recover from an unexpected perturbation or the distance their centre of pressure travels, following this balance disturbance (11). While such measures are considered the gold standard for measurement of postural control, they are expensive, space consuming and inaccessible to most practitioners (278).

Simple measures that are more practical in their clinical application include timed single leg standing (with eyes open at first and closed as a progression), the degree of tilt possible when standing on a tilt board (interrater reliability Spearman's r =0.87-0.99 and r=0.98; test-retest reliability r=0.59-1.00 and poor respectively) and tandem walking or walking along narrow surfaces, as tested in children aged 5-12 years (277).

Very few balance measures created specifically for use in disabled children exist (277) and the above measures all require that the participant have the ability to stand independently. The Berg Balance Scale has been found to have excellent reliability when used in adult populations (ICC≥0.98) but has proved more complicated to administer in children and has shown less promising psychometric properties when used for this purpose (174)(279). It is for this reason that Franjoine, Gunther and Taylor piloted a modified paediatric version of this scale in 2003, using 40 TD children and 20 5-15 year old children with mild to moderate motor impairments. This version decreased the time expected for static positions, rearranged the order of tasks into a more logical, sequential developmental sequence and

added meaningful goals to each item (174)(279). Results from the pilot study revealed excellent test-retest reliability (ICC=0.998) and interrater reliability (ICC=0.997). The authors concluded that the Pediatric Balance Scale might provide an excellent alternative means of measuring balance in children between the ages of 5-15 with disabilities as it is quick and easy to administer and shows excellent preliminary reliability. Limitations listed were the lack of validity calculations for the scale and the absence of higher functioning balance tasks included (such as balance during gait and overhead activities) (174)(279).

The original Functional Reach Test (FRT) was created for the prevention of falls in the elderly (278). The individual is made to stand (wearing whatever footwear or orthoses are preferred) and a measurement is taken from the acromion process to the third metacarpal phalangeal joint with the upper limb outstretched in front of the person, parallel to the floor. The distance reached forward before a step is taken is then measured and the difference in lengths recorded (280). Such measurements are usually taken with the assistance of a yardstick or drawings on the wall behind the participant for greater accuracy of measurement. Three trials may be attempted before recording the final measurement (281).

While the FRT has proved reliable for use in paediatrics, it is only relevant to those children able to stand independently (278). Bartlett and Birmingham tested the original FRT and a modified version that has the participant sitting with their back upright against a backrest and their lower limbs angled at 90 degrees, with the feet placed flat on the floor. The same procedure as above took place for forward reaching and further measurements were taken with the shoulder abducted to 90 degrees for lateral reaching toward the left and right (281). Testing, using 19 TD and 10 children with CP below the age of 12, proved good construct validity of the new Pediatric Reach Test (PRT), as seen by significant associations between this new measure and computerised balance plate measurements during reaching and GMFCS levels ($r=0.65$; $p=0.003$). Reliability of the different reaching components proved moderate-to-excellent (ICC=0.50-0.97) (278). In children, this measure is a good indication of postural responses to internal rather than external perturbations (i.e. to “self-initiated movement”) (278).

While numerous studies have since supported the use of the PRT for balance testing in disabled children, it is limited in that it only tests anticipatory postural control and not balance in more dynamic situations (174). Such reviews have suggested the inclusion of measures such as the Timed Up and Go (TUG) or the Timed Up and Down Stairs to assess transitions between movements and balance reactions during more complex motor tasks, such as walking and stair climbing (174).

Originally called the Get Up and Go when used for mobility testing in frail, aged populations the TUG sees the participant seated as for the PRT, wearing their usual shoes and orthotics if indicated. On a specified verbal cue, the individual is expected to stand and walk a distance of three metres marked on the floor. At a designated point, signalling the end of the three metres, the participant turns and returns to their starting position seated comfortably. A trial may be attempted prior to timing the procedure. Assistive devices are allowed during testing if necessary (171)(280)(282).

Studies in the elderly have demonstrated excellent interrater (ICC=0.99) (280) and test-retest reliability (283), for the measure and use is simple, quick and bears a low administrative burden (283)(282)(284). Studies demonstrating the reliability of use in children are more limited (284) and Williams et al have suggested giving very simple instructions, timing starting only when the child decides to walk and adding a more meaningful task to the procedure to encourage best performance in children (284). In their study, within session (ICC=0.80-0.89) and test-retest reliability (ICC=0.83) were good and responsiveness and validity were proved when administered in children with disabilities (CP and spina bifida) aged 3-19. The TUG was also able to differentiate between the first three GMFCS levels (284).

2.5.2.2 Measuring abdominal strength

Laboratory-based research generally employs computerised dynamometers, such as Biodex units, for strength testing. Testing of isokinetic knee strength in children with CP aged 9-15 proved high reliability of the Cybex II isokinetic dynamometer (285). Due to their extreme

size and expense, such units are not practical for use in general research or clinical settings (286). On the other hand, more portable and less expensive hand-held dynamometers are growing in popularity (286)(155), although authors have cautioned against the use of this equipment due to the great variability between raters found on testing (155). One such study found intrasession and intersession ICCs of 0.84 or greater but with standard errors of measurement from 6.72 to 25.26 in children with CP aged 7-17 (287). For more specific abdominal strength readings, surface EMG is available for superficial muscles but in order to target the deeper abdominals much more invasive fine-wire EMG is necessary (30). Muscle properties, such as abdominal muscle thickness, can be assessed using ultrasound imaging but these readings give a more indirect idea of muscle strength left open to interpretation (30).

Core muscle strength is routinely tested in sports rehabilitation and conditioning settings. Isokinetic and isometric dynamometers are generally used for strength testing (as reports of excellent interrater reliability exist) and core endurance via more subjective field-side tests (13)(288). A pre-recorded cadence has been used to time curl-ups during sport testing. In supine, with the knees flexed to 90 degrees, the number of curl-ups possible (touching a predetermined point) before fatigue or before losing timing with the metronome is counted (212)(289). The same procedure can take place against the resistance of a dynamometer when curling up. To test endurance, one would time how long an individual is able to maintain this sit-up position against resistance (289). Testing performed in 39 healthy adult subjects (aged 17 and above) revealed good reliability (ICC=0.89) for measures of dynamic abdominal endurance on timed curl-ups, but inconsistent results for all other measures of abdominal strength (289).

In contrast, muscle power may be measured during one-minute speed tests whereby the individual completes as many sit-ups as is possible over a 60-second period (290). On investigation, Teyhen et al determined that out of 12 different exercises activating the abdominal muscles, an abdominal “crunch” triggered the greatest muscular challenge with the lowest lumbar load on ultrasound imaging (30). This activity also brings about the longest duration abdominal muscle activation and it was suggested that abdominal crunching be used as a safe, easily administered and effective means of measuring core muscle strength in children when used to assess 3-7 year old children (291).

2.5.2.3 Measuring lung function

Lung function testing generally involves obtaining forced vital capacity (FVC) and forced expiratory volume in one second (FEV_1). FVC is measured as the volume of air expelled when a forced maximum expiration takes place after a full inspiration. This is relevant as it gives an indication of the expiratory muscle strength available to enable an effective cough. FEV_1 is the volume expelled during such an exhalation in the first second only and indirectly allows one to determine an individual's ability to expectorate (189)(191)(292).

While spirometry has commonly been used for lung function testing in the past, it is recognised that use in children is impractical due to the sheer size and formidable appearance of many spirometers and the lack of compliance with donning a nose clip (292). Peak flow meters have been accepted as an inexpensive, portable and easy to use alternative for the measurement of FEV in children with asthma and obstructive pulmonary disorders since the early 1960s (292)(293). Compliance and accurate use has been seen in children as young as five years old (292)(293). An investigation of 12 school-going boys, aged 11-17, with asthma revealed that four different brands of mini flow meter readings were poorly correlated with those taken by a spirometer and the authors concluded that these measures may overestimate expiratory lung function (294). Despite this, peak flow meters remain the only cost-effective outcome measure for expiratory function in children with CP.

2.5.3 Measuring activity limitations and participation restrictions

2.5.3.1 Measuring gross motor function

The Bayley Scales of Infant Development is a discriminative tool used to assess the acquisition of motor milestones in developing babies (128). Also measuring the development of gross and fine motor skills is the Peabody Developmental Motor Scale, a 3-point scale administered for children between birth and seven years of age (128).

The Pediatric Outcomes Data Collection Instrument (PODCI) is a tool validated for use in children with musculoskeletal disorders. This self or parent proxy questionnaire reliably assesses motor function and health status in this population, as determined by studies of children aged 3-23 years old (273).The use of the PODCI is limited by the potential floor and ceiling effects of this measure (271).

The Bruininks-Oseretsky Test of Motor Proficiency involves 46 items testing gross and fine motor skill acquisition applied to children from 4-21 years of age (295). While the measure shows reliability and validity, its use in CP is limited as many of the test items are very difficult to achieve for those children with CNS disorders and it is best used in the assessment of children with generalised developmental delays or developmental coordination disorder (128).

The Pediatric Evaluation of Disability Inventory (PEDI) measures functioning in children with disabilities/chronic illnesses in two spheres: capability/competence in completing certain functional goals and functional performance in terms of the degree to which assistance or adaptation is required in order to achieve these tasks, established through parent interviews. This norm-referenced measure is further divided into self-care, mobility and social function scales addressing the activity limitation and participation restriction components of the ICF (126)(128)(221)(266)(269). As items on the scale more dominantly encompass lower level functioning, more severely affected children with disabilities can still be assessed with the measure (271). Studies have shown the PEDI to have higher quality psychometric properties than other functional outcome measures, such as the PODCI, and studies demonstrate the measure has excellent internal consistency ($\alpha \geq 0.98$) (271), satisfactory reliability (0.76-0.96) (75)(296), validity (128) and responsiveness to change (75). Its use is limited to children between 6 months and 7.5 years old (128)(273) (297) and children of GMFCS level I may demonstrate a ceiling effect on assessment with the scale (125).

Systematic reviews have asserted that the PEDI and the Gross Motor Function Measure (GMFM) are the most reliable and valid evaluative measures responsive to changes in the

functioning of children with CP (269)(128)(126)(273) and the two scales are strongly correlated with and complement one another (126)(128)(271). They are widely considered the gold standard in evaluating gross motor function in CP (65)(69)(226).

While all the measures examined above were created for the assessment of disabled children in general, the GMFM was specifically created to evaluate the movement disorders present in CP (269) and has since been used in countless studies appraising the effectiveness of rehabilitative, surgical and medical interventions addressing CP (273)(274). The GMFM-88 was first introduced by Russell et al in 1990. Eighty-eight gross motor tasks thought to be fully accomplished by a TD five year old were included in their developmental sequence and broken up into five motor groupings; namely: "Lying & Rolling", "Sitting", "Crawling & Kneeling", "Standing" and "Walking, Running & Jumping". These activities comply with the ICF's concept of functioning in that transitional movements are included, rather than static positions (126)(140). A generic 4-point Likert scale explains the scoring for each item, whereby a 0 is achieved if the task cannot be initiated, a 1 is gained if the movement is initiated or <10% is achieved, a 2 is scored for partial completion of the task (i.e. 10%>task< 100%) and a 3 is awarded for full completion of the task as laid out in the guidelines. The GMFM User's Manual delineates more specific scoring prerequisites for each individual item (274). A description of how the measure is administered and scored, details of the original validation study of the measure and how the GMFM-66 was developed can be found in Appendix VI.

Studies of the GMFM-66 have shown that intraclass correlation coefficients (ICC) are as high as 0.99 for test-retest and interrater reliability (65)(101), 0.76-1.00 for the stability of participant conditions (65) and 0.98 for concurrent validity (298). The measure is responsive to both positive and negative changes in function while showing longitudinal construct validity and greater specificity than the GMFM-88 (65)(223). The GMFM has also been found to be able to differentiate between the different GMFCS levels (153)(299). The measure is used to compare a child's gross motor development over time and not to make comparisons between children with CP and their TD peers (i.e. it is criterion and not norm-referenced) (105)(128). The original validation study investigated 111 children with CP, aged between five months and 16 years old (274) but no upper age limit is given for use (128).

Limitations of the measure include a possible ceiling effect for higher functioning children with CP that has not been negated completely by the introduction of the GMFM-66 (226)(273) and it has been intimated that the GMFM-66 may not be sensitive enough to detect changes in those very low functioning children with CP (300).

As the GMFM measures purely the attainment of certain gross motor tasks, the Gross Motor Performance Measure was created to allow for the assessment of quality of movement during these tasks. No upper age limit is given for use (128). The observational measure contains 20 items from the GMFM evaluated based on alignment, weight shifts, stability, coordination and dissociation seen during these tasks and is scored on a 5-point scale. Research shows that while reliability is good for therapists trained in the use of the measure, responsiveness to change does not correlate with practitioner opinion on the change seen in participants and its use outside of laboratory research has been cautioned (128)(225). It is for this reason, and the limitations of the other gross motor outcome measures discussed above, that the GMFM-66 was selected for use in this study.

2.5.3.2 Measuring health related quality of life (HRQoL)

Quality of life (QoL) can be explained as a holistic perception of one's life taking environmental, social and personal factors into account (210)(301). While most outcome measures validated for use in children with CP assess impairments or mobility, in order to comply with all three spheres of the ICF framework, studies should include measures to assess participation and HRQoL (33). This is particularly important as children with CP have been found to participate less than their TD peers (214)(302). In order to evaluate the effect of an intervention one cannot merely look at physical outcomes but one also needs to ascertain whether the intervention outcome is meaningful to the individual and alters their HRQoL (53)(56)(215). While not entirely based on activity limitations and participation restrictions, these measures generally include items based on activities of daily living, social functioning and individual's perceptions on their daily interactions and thus, HRQoL outcome measures have been included in this section.

Unfortunately, many children with CP are unable to communicate and one needs to rely on parent proxy for the completion of most HRQoL measures available (56). This has led to debate surrounding the reliability and validity of these outcomes (56)(303). In 2007, Oeffinger et al compared child and parent proxy versions of numerous HRQoL measures for 562 children with CP, between level I and III on the GMFCS, aged 4-18. Results showed that significant differences existed between child and parent reports for ¾ of PedsQL (described below) and PODCI (see 2.5.3.1) domains, with the children rating their HRQoL as better in all instances. The more severe the child, the greater the disparity between child and caregiver scores (275)(304). The results of this study were mirrored by Majnemer et al in 2008 (301). Interestingly, the experience of pain and suffering is not correlated with severity of CP (200) and studies have shown that TD school-going children involved in mainstream education rate their HRQoL as poorer than age-matched children with disabilities based at special needs schools (303).

HRQoL measures determine a person's participation in activities of daily living and social functioning whilst gaining insight into their perception of their own health and how this affects their perceived well-being (56)(301)(304). In their 2010 systematic review, Carlon et al suggested the use of condition specific HRQoL measures over more generic measurement tools and they concluded that no gold standard for the measurement of HRQoL in this population exists (302). Those CP specific HRQoL measures available are discussed below.

The Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) is a HRQoL measure developed through interviews with 52 caregivers and validity testing. The measure is completed by the primary caregiver of a child with CP and questions are scored based on the domains of Personal Care; Positioning, Transfer and Mobility; Communication and Social Interaction; Comfort, Emotions and Behaviour; Health; and Overall QoL. The comfort section is rated in terms of frequency and intensity of discomfort, while all other skills are rated based on difficulty using a 7-point scale (where zero means the child has no difficulty fulfilling the required task and six means the task is impossible). As well as scoring difficulty, the amount of assistance required during each particular task is noted on a 6-point ordinal scale (from no to total assistance required). A final section allows parents to rank which of the domains they feel are most to least significant to their child's perceived

HRQoL (56). Initial investigations of test-retest reliability demonstrated ICCs of between 0.88 and 0.96 for the various CPCHILD domains (56). The same measure is completed for all children aged 5-18 years (53), thus incurring questions regarding the validity of such a measure (302).

The Cerebral Palsy Quality of Life Questionnaire for Children (CP QOL-Child) was compiled by a multidisciplinary team from around the world and validated for use in children with CP between the ages of 4-12. The authors have suggested that both child and parent-proxy versions be used for accuracy. Sections include questions on social and emotional wellbeing, participation, physical and family health, functioning, pain, acceptance and access to services. Initial testing of the instrument revealed sound construct validity and test-retest reliability (ICC=0.76-0.89) (305).

The PedsQL 3.0 CP Module was developed based on research and practitioner experience and has been shown to have excellent validity, reliability and internal consistency (67)(301)(302). It is the only HRQoL measure allowing for self-report in children with CP from the age 5-18 (67). The measure has been adapted slightly so that children of different ages complete a version appropriate to their age group's level of understanding (i.e. ages 5-7, 8-12 and 13-18). The instructions request answers relating to experiences over the past month. Sections are divided into questions related to activities of daily living, participation at school, movement and balance, pain, fatigue, feeding and communication. For those children unable to complete the measure independently, an adapted version has been created allowing for parent proxy. The items on the measure are scored by the participant, on a 5-point ordinal scale, with zero stating that the problem is never experienced and four confirming that the particular item is always a problem. The version applicable to children aged 5-7 simplifies scoring by providing a visual analogue scale of happy, indifferent and sad faces to assist answering "not at all", "sometimes" and "a lot" to the related questions on the measure (53)(67)(130)(301)(306). This concept has been supported by Eiser, Mohay and Morse, who explained that it is vital to determine children adequately understand what is being asked of them during testing with HRQoL measures and that the use of simple pictures may assist this understanding in younger children (307).

The instrument is limited in that questioning of teenagers lists very simple activities of daily living etc. that one would expect mild-moderately affected children to be able to complete and more appropriate social interaction and participation questions are not included in the measure (53). Despite this, the PedsQL 3.0 CP Module is still the only validated HRQoL measure developed specifically for use in children with CP that can be tested using self-reports from children aged 5-18. As self-report scores differ greatly from parent-proxy scores on different measures, it is more accurate to obtain the individual child's perception of their HRQoL, wherever possible.

2.6 Study designs

While qualitative, observational and descriptive studies all provide valuable information in scientific research, they are unable to categorically state whether an intervention actually makes a statistically or clinically significant difference or not (29). Credible, quality studies require thorough methodology, adequate sample sizes, limited possibility for the introduction of bias, valid outcome measures and proof of internal validity (25)(67). In order to ascertain that improvements in outcome determinants are in fact due to the experimental intervention provided, a randomised controlled trial (RCT) needs to be implemented (230). RCTs are widely accepted as the "gold standard" for scientific enquiry (66)(308)(309)(310). Study participants are randomly allocated to different treatment groups with the expectation that both groups' demographic characteristics will be similar. The quality of such studies is further improved if one of these groups serves as a placebo-based control, if both assessors and those implementing the intervention are blinded to group allocation of participants, if said practitioners are trained and experienced (67)(309)(311) and if the relevant ethical body has approved the study prior to implementation (312). These factors serve to reduce the opportunity for human error and limit the possibility of human bias/prejudice affecting scientific results (309). Unfortunately, such rigorous trial design often leads to smaller treatment effects than seen in observational studies (309).

Experimental studies are inherently longitudinal and prospective in nature (311). While explanatory trials investigate treatment interventions in ideal, laboratory-based

circumstances, pragmatic trials determine their effect in routine, every-day situations (39). For ethical and practical reasons pragmatic trials compare an intervention to the accepted standard/norm of intervention rather than to a placebo. In order for results to be generalisable to a wider population, MacPherson has suggested that pragmatic trials incorporate broad inclusion criteria (thus a large sample is required) and that outcome measures evaluate functional performance and QoL (39). Pragmatic trials have the distinct benefit of allowing practitioners to continue with their customary treatment, where this would not be possible in a clinical laboratory setting. However, they are limited by the impracticality of blinding in certain instances (39).

Inadequate reporting has allowed the introduction of bias and limitations in the external validity of RCTs. As this makes comparison between studies difficult, an international multidisciplinary group of professionals, qualified in statistics, research and editing, came together to create the Consolidated Standards of Reporting Trials (CONSORT) statement in an attempt to generate guidelines that would assist with improved quality of reporting on RCTs (40). The external validity of a study can be negatively influenced by underreporting of treatment parameters, especially if the intended intervention differs from "usual clinical practice" or if the control group does not receive what is considered best practice (313). It is for this reason that one of the items included in the revised CONSORT checklist is the need to include the specific details of the intervention to be administered to both control and experimental groups, in terms of what this treatment will entail, how it will be implemented and for what durations it will take place (40).

Experimental studies, investigating new interventions for the treatment of children with CP, cannot ethically include a control group that receives no intervention and including a placebo into such trials may be considered equally difficult or unethical. Pragmatic trials are more practical in their implementation for this population as the additional intervention can be tested under usual treatment circumstances, in real-life situations. In order for the accuracy of analysis of results of such studies, the treatment parameters of baseline/conventional therapy provided during the study needs to be described in detail. This allows for reproduction of the study in the future, as well as prevents the introduction of bias. It is recommended that the CONSORT be used when reporting RCTs.

2.7 Summary of the literature review

CP is the most commonly experienced CNS disorder in children, as well as the most commonly treated disorder by paediatric physiotherapists worldwide. SA and other developing countries may experience even higher prevalence rates. While spasticity was always considered the primary impairment in children with CP, the presence of weakness, resulting from the original upper motor neuron lesion and specific morphological changes to muscle in this population, results in greater activity and participation limitations. This weakness also results in deficits in balance and postural control.

A multitude of treatment interventions exist for the management of CP, including modalities that fall under rehabilitative, surgical, medicinal and alternative categories. All of these available interventions have conflicting evidence supporting their effectiveness (including the globally, most frequently employed intervention, NDT) and many studies' results demonstrate inadequate statistical power due to poor methodological design and small sample sizes. Current trends in therapy include the once rejected idea of strength training, as a growing body of literature shows no change to levels of spasticity but encouraging improvements in muscle strength with loading/resistance training. The same has been found for abdominal strengthening in CP and it is postulated that core strength gains may lead to positive postural improvements and functional development. The use of ES in populations of people with disabilities is gaining increasing interest as preliminary studies show modest improvements in certain gait parameters, respiratory function and muscle strength. When used in a functional context, ES has the potential not only to improve strength but also to have carryover into the performance of every day functional activities.

Outcome measures used should be developed specifically for use in the tested population, and be validated and reliable. Pragmatic, RCTs provide the most practical and methodologically sound structure for investigating the effect of a new intervention for the treatment of children with CP. This requires that a detailed description of the therapy administered at baseline or to the control group as part of routine/conventional therapy is included in the write-up of the study. This would allow for a study on FES in children with CP to accurately determine whether this modality is an effective adjunct to physiotherapy.

A previous study showed significant strengthening of abdominal muscles following a four-week trunk targeting intervention and thus it was surmised that effects should be seen following six weeks of trunk targeted FES intervention.

3. Intervention strategies employed by physiotherapists in Johannesburg

As has been discussed in the literature review, the need for a description of what conventional/routine intervention consists of exists. This limits the potential for the introduction of bias and makes the results of pragmatic trials both replicable and generalisable. It is for this reason that this subsection of the study was included. Comparing a new intervention to conventional therapy aids the ability to conclude with conviction that any significant changes seen are in fact due to the intervention under investigation. In order to do so, it is necessary to determine whether the conventional therapy offered is similar to that offered in other settings as well. Below follows a description of the methodology of this subsection of the study, the procedure followed, how data collection took place and was later analysed, the results of this study, as well as a discussion of these results. This section's limitations are discussed before concluding the write-up of the substudy.

3.1 Methodology

3.1.1 Aims and objectives

- To determine whether the physiotherapy treatment of children with CP offered at the Institution was similar to other special needs schools, by comparing the content and intensity of physiotherapy intervention offered by the therapists at the Institution with that offered by other similar facilities in Johannesburg

3.1.2 Null hypothesis

The rank order correlation of frequency of the different interventions, between those used at the Institution and those used by physiotherapists at other schools in Johannesburg, would not be significant.

3.1.3 Research design

A cross-sectional descriptive research design was used for this segment of the study.

3.1.4 Sample/participants

The sample included the entire eligible population based on the inclusion and exclusion criteria below. All government-funded special needs schools in Johannesburg offering physiotherapy services to children with CP were included.

3.1.4.1 Eligibility criteria

Inclusion criteria met by participants were as follows:

- Qualified physiotherapist, permanently employed by the Gauteng Department of Education at one of the above schools

Exclusion criteria:

- No verbal consent to participate given

3.1.5 Research setting

There are several government-funded special needs schools in Johannesburg offering physiotherapy services, but only nine of these accept children with CP. Most other schools cater for children with visual, auditory or learning disabilities. The nine eligible schools

accept children who have both physical and intellectual impairments and most offer education from nursery school level until grade seven or nine, with both mainstream and adapted/remedial streams. Only the Institution offers a mainstream educational curriculum up until matric/grade 12. These schools are government subsidised and the majority of learners attending these schools come from poor socio-economic backgrounds. The number of physiotherapy, occupational therapy and speech therapy posts available at each school is determined by the government, based on a weighting system for the number of learners enrolled at each school and the type of disability each child has.

In SA, basic, eight-week, paediatric NDT courses are offered annually. Courses are monitored by the South African NDT Association and are run by internationally qualified and accredited NDT lecturers and facilitators. Introductory, one-week courses are held throughout the year in order to provide rehabilitation professionals with a basic overview of the NDT concept and its implementation. Once the eight-week, basic NDT course has been completed and passed, numerous advanced NDT courses of 3-4 weeks duration are available. The NDT concepts are incorporated into paediatric physiotherapy training at university undergraduate level in most cases, but in order to become an NDT practitioner, the basic eight-week course must be completed as a postgraduate (314).

3.1.6 Instrumentation

The instrumentation used consisted of a checklist of intervention techniques and a description of the treatment regularity and intensity of children with CP treated by each practitioner.

The checklist was developed by the researcher based on literature and post-graduate courses that she has attended, as well as on her own experience of working with children with CP and discussions with other therapists. This list was submitted to a panel of three physiotherapists, from three different provinces, expert in paediatric neurology, to be checked for face and content validity. Through email correspondence, this panel provided suggestions to better represent all physiotherapy interventions currently used in the

treatment of children with CP throughout the country. As well as adding missing interventions, the researcher also added a glossary of definitions of each intervention for better understanding, broke down each concept into its components and added a column for how regularly each checked intervention was utilised, based on the proposals of the expert panel. After alterations were made to the original checklist, all three professionals on the panel unanimously accepted the final checklist before use as an outcome measure for this portion of the study. These professionals confirmed the face and content validity of the intervention checklist as an outcome measure for the assessment of physiotherapeutic interventions used in daily practice at special needs schools in Johannesburg.

3.2 Procedure

3.2.1 Ethical considerations

3.2.1.1 Approval

Ethical approval for the sub-study was obtained from the University of Cape Town, Faculty of Health Sciences, Human Research Ethics Committee. Following ethical approval, permission for commencement of the study was gained from the Gauteng Department of Education. The clinical trial was registered on the Pan African Clinical Trials Registry (identification number PACTR20131000634685)(315).

3.2.1.2 Risk to participants

Time was not taken away from therapeutic intervention of children, as appointments were arranged during tea times or before the school day, when convenient for each therapist, at the respective special needs schools. Informed consent was obtained verbally from each

physiotherapist prior to completion of the intervention checklist, which was completed anonymously, with only a number coding for the particular school evident on each form. Completed checklists were kept in a secure location, separate from the list explaining which number related to which school, assuring confidentiality of all information completed.

3.2.1.3 Benefit to participants

Participants were advised they could contact the researcher should they wish to see the final results of the study, thus gaining valuable knowledge on the use of FES in CP, as well as a comparison of interventions taking place at other schools in their area.

3.2.2 Pilot study

Completion of the intervention checklist was piloted, using four physiotherapists. They found the checklist easy to understand and complete and felt it was thorough and representative of all the general techniques they use in practice. The checklist was found to take 10-15 minutes to complete and the glossary of definitions was found self-explanatory, meaning no further questions for clarity (after initial explanation of how the checklist works) were asked.

3.2.3 Data collection

The premise of the study was explained to each school's therapy Head of Department, either telephonically or via email prior to setting up visits to each respective school, at a time that suited each department. Visits to all nine eligible special needs schools were conducted by the researcher, from May to June 2013, visiting one or two schools a week. The aim of the study and the intervention checklist were explained to each participant and

verbal consent to take part was obtained. The researcher was available to assist if any queries arose, but each participant completed the checklist individually, anonymously and without observation of their answers. The completed forms were collected by the researcher before leaving each participating school. Once all school visits had been completed, this data was collated and analysed.

3.3 Data analysis

As the numbers included in this section of the study were small, descriptive statistics were used to describe the characteristics of the participating therapists and the interventions utilised. The rank ordering of the frequency with which the different interventions were employed at the Institution and in the other facilities was calculated using Spearman's rho, a non-parametric test.

3.4 Results

3.4.1 Sample

Twenty-five physiotherapists are employed by the Gauteng Department of Education, at nine special needs schools, in the Johannesburg area. Four of these therapists (including the primary researcher) work at the Institution where the study took place and their demographic data are represented in Table 2.

Table 2: Demographic characteristics of physiotherapists working at the Institution (n=4)

Qualification	Years of experience	Other qualifications (specific to the treatment of CP)
BSc (Physiotherapy)	7	NDT basic 8 week course
Bachelor degree of Physiotherapy	22	NDT basic 8 week course
BSc (Physiotherapy)	17	NDT basic 8 week course
Bachelor degree of Physiotherapy	25	NDT introductory 1 week course

Of the original total sample, three physiotherapists did not return their intervention checklist (both physiotherapists from school 2 and one from school 9) and thus their demographic data are not represented. The remaining 18 physiotherapists' demographic data (working at other special schools in Johannesburg) can be seen below (Table 3).

Table 3: Demographic characteristics of participating physiotherapists working at other special schools in Johannesburg (n=18)

School	Qualification	Years of experience	Other qualifications (specific to the treatment of CP)
1	BSc (Physiotherapy)	6	None
1	BSc (Physiotherapy)	12	NDT basic 8 week course
1	BSc (Physiotherapy)	7	None
1	MSc (Physiotherapy)	17	NDT advanced course or other specialised courses
1	BSc (Physiotherapy)	7	NDT basic 8 week course
4	Bachelor degree of Physiotherapy	29	NDT advanced course or other specialised courses

4	Bachelor degree of Physiotherapy	3	NDT introductory 1 week course
4	National Diploma in Physiotherapy	19	NDT basic 8 week course
5	BSc (Physiotherapy)	3	NDT introductory 1 week course
5	Bachelor degree of Physiotherapy	12	NDT advanced course or other specialised courses
5	BSc (physiotherapy)	15	NDT advanced course or other specialised courses
6	National Diploma in Physiotherapy	41	NDT basic 8 week course
6	BSc (Physiotherapy)	13	NDT advanced course or other specialised courses
6	Bachelor degree of Physiotherapy	11	NDT basic 8 week course
7	RNIB Diploma (UK)	35	NDT basic 8 week course
7	BSc (Physiotherapy)	15	NDT basic 8 week course
8	BSc (Physiotherapy)	17	None
9	BSc (Physiotherapy)	5	NDT introductory 1 week course

Nineteen of the 22 physiotherapists at the participating schools had undergone some degree of NDT training, with 15 of these being qualified NDT practitioners (having completed the basic eight-week or other additional advanced NDT courses). The mean years of experience in the experimental school was 17.75 (SD=7.89) and in the control schools 14.88 (SD=10.66). Fifteen of the 22 therapists in both school groups had more than ten years' experience (Table 2 and Table 3).

3.4.2 School characteristics

The number of posts allocated varied between a minimum of one and a maximum of five physiotherapists working at each school. All nine schools offered nursery schooling and accepted learners until the cut-off age of 18-22. The Institution has 222 enrolled learners and the mean number of learners attending the other special schools in Johannesburg is 378 (SD=86.95). The Institution is the only special school in Johannesburg offering a mainstream curriculum with no remedial/vocational stream also available. The percentage of those learners enrolled at the Institution with a physical disability is 84.68% while the mean percentage at the other special schools is 54.74 % (SD=25.79). The ratio of physiotherapists to physically disabled learners at the Institution is 47:1 and to those with CP 19:1. The mean ratio at other special needs schools was 89:1 (SD=48) and 56:1 (SD=32) for learners with physical disabilities to physiotherapists and those with CP to physiotherapists respectively (Table 4).

Table 4: School characteristics of participating special needs schools in Johannesburg (where school 3 is the Institution at which the study took place) (n=9)

School no.	Mainstream only or vocational/ remedial stream	No. physios at school	Total no. learners	Earliest age accepted	Oldest age accepted	Approx. no. physically disabled	Approx. no. with CP	% Total no. physically disabled	Physically disabled: Physio	CP: Physio
1	Both	5	330	3	18	120	114	36.36	24	22.8
2	Both	2	450	5	18	350	200	77.78	175	100
3 – The Institu-tion	Mainstream only	4	222	3	22	188	75	84.68	47	18.75
4	Both	3	450	3	20	275	110	61.11	91.7	36.67
5	Both	3	425	3	20	142	74	33.41	47.3	24.67
6	Both	3	450	3	20	300	120	66.67	100	40
7	Both	2	216	5	22	216	205	100	108	102.5
8	Both	1	401	3	20	120	75	29.93	120	75
9	Both	2	300	5	18	98	93	32.67	49	46.5

3.4.3 Treatment characteristics

Physiotherapy sessions last 30 minutes at seven of the special needs schools, while the Institution at which the study took place and School 2 performed 45-minute treatment sessions. Five of the special schools treat each timetabled learner once a week maximally and the remaining four schools generally treat their learners twice weekly. Four schools only rehabilitate learners in groups of from two to a maximum of 12 learners simultaneously, where the remaining schools treat in smaller groups of up to a maximum of seven children at a time as well as still performing individual treatment sessions. The Institution is the only school that treats learners in their teens and does not place an emphasis on treating children in the foundation phase only (Table 5).

Table 5: Treatment characteristics of participating special needs schools in Johannesburg (where school 3 is the Institution at which the study took place) (n=9)

School no.	Length of Rx sessions in minutes	Frequency of Rx of CP learners per week (mostly)	Individual or group Rxs	Routine cessation of Rx once learners reach their teens	No. learners per group minimum	No. learners per group maximum
1	30	2	Both	Yes	1	5
2	45	1	Both	Yes	1	7
3 – The Institution	45	2	Both	No	1	4
4	30	2	Group	Yes	8	12
5	30	1	Group	Yes	4	4
6	30	1	Both	Yes	1	5
7	30	1	Both	Yes	1	7
8	30	2	Group	Yes	2	5
9	30	1	Group	Yes	4	7

Note: Rx=treatment

Treatment techniques most commonly used by all participating physiotherapists were facilitation of transitions between positions and general limb strengthening, followed by items spread between the NDT, Orthopaedic, Strengthening and Gait Training treatment approaches. The only treatment techniques not used by any physiotherapists were the Power Plate, Functional Electrical Stimulation and Vojta Therapy techniques (see Figure 3, Figure 4 and Table 6). When comparing techniques used and frequency of use between physiotherapists at the Institution and other special schools in Johannesburg, it was noted that those techniques most commonly used were similar. Techniques not used by the Institution that were used at other schools (although not commonly) were electrical stimulation, treadmill training and constraint-induced therapy. No core or general limb strengthening at the Institution was performed until fatigue. The use of heat and ice and external perturbations during balance-specific training was more common at the Institution than at other special schools (Table 6).

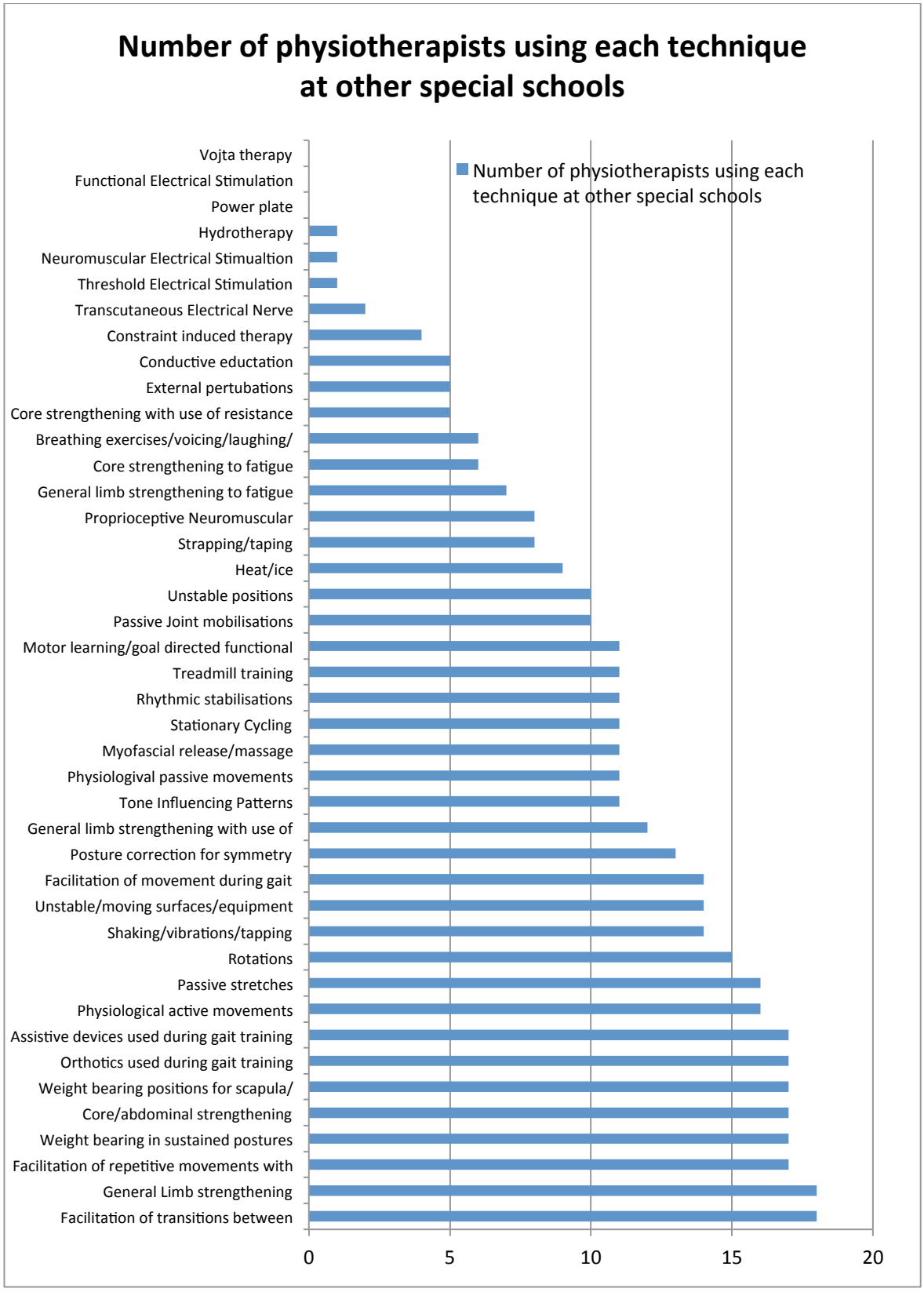


Figure 3: Number of physiotherapists using specific treatment techniques at other special schools in Johannesburg (n=18)

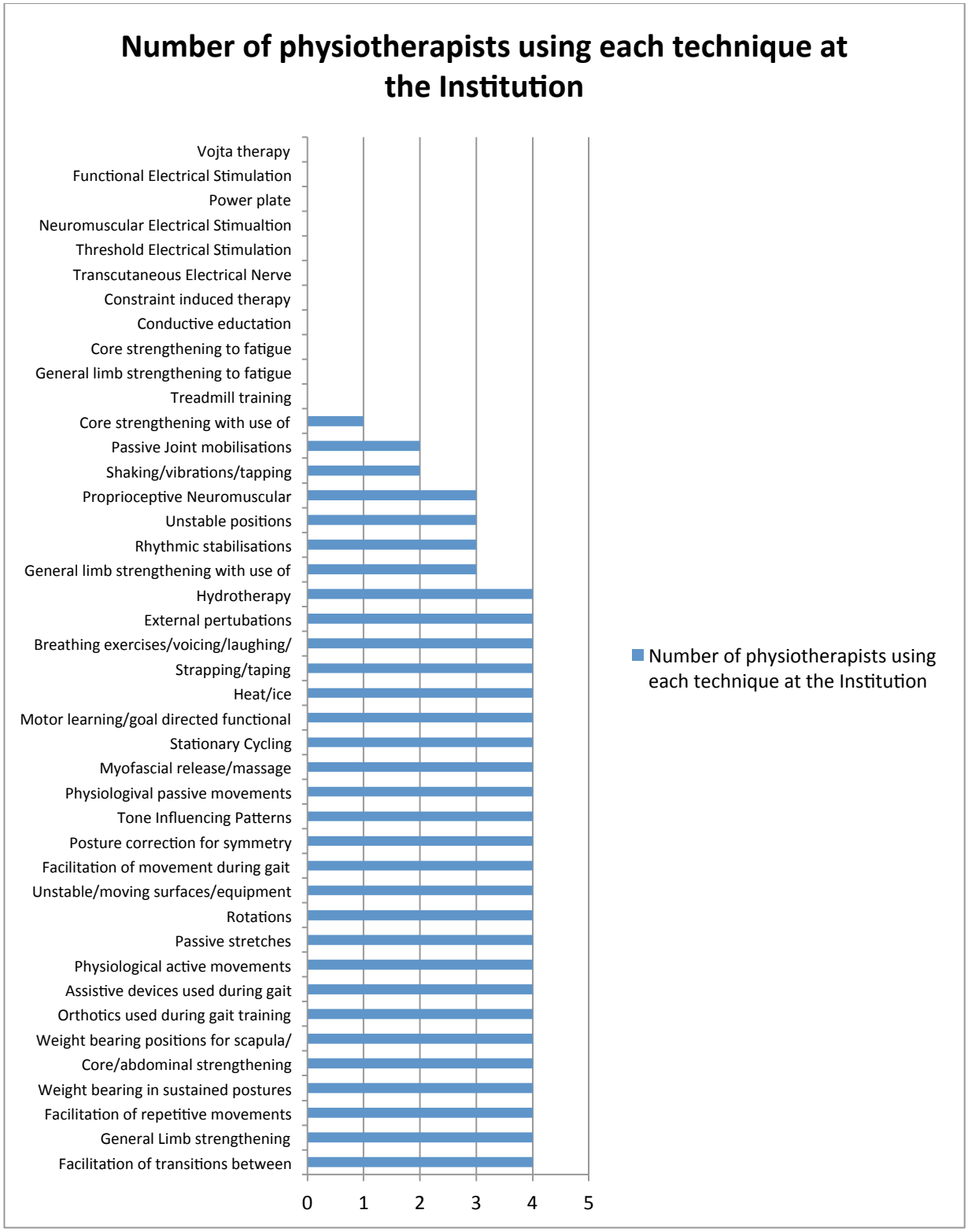


Figure 4: Number of physiotherapists using specific treatment techniques at the Institution (n=4)

Table 6: Treatment techniques used and their general frequency of use by physiotherapists at the Institution (n=4) compared to other special schools in Johannesburg (n=18)

Treatment Approach	Specific treatment technique	No. therapists using technique at other schools (n=18)	Most common frequency (no. participants)	No. therapists using technique at the Institution (n=4)	Most common frequency (no. participants)
Neuro-develop-mental Therapy	Facilitation of transitions between positions	18	Daily (15)	4	Daily (3)
	Facilitation of repetitive movements with assistance	17	Daily (13)	4	Daily (2)
	Weight bearing in sustained postures	17	Daily (12)	4	Biweekly (2)
	Rotations	15	Daily (12)	4	Daily (3)
	Shaking/vibrations/tapping	14	Daily (7)	2	Weekly (2)
	Posture correction for symmetry	13	Daily (10)	4	Daily (3)
	Tone Influencing Patterns	11	Daily (8)	4	Daily (3)
Orthopaedic Approach	Physiological active movements	16	Daily (7)	4	Daily (3)
	Passive stretches	16	Daily (9)	4	Daily (3)
	Physiological passive movements	11	Weekly (5)	4	Daily (2)
	Myofascial release/massage	11	Infrequently	4	Weekly (2)

			(4)		
	Passive joint mobilisations	10	Weekly (5)	2	Weekly (1); Monthly (1)
	Heat/ice	9	Infrequently (5)	4	Daily (2)
	Strapping/taping	8	Infrequently (5)	4	Infrequently (2)
Strengthening	General limb strengthening	18	Daily (10)	4	Daily (2)
	Core/abdominal strengthening	17	Daily (10)	4	Weekly (2)
	Weight bearing positions for scapula/pelvic stability	17	Daily (11)	4	Daily (3)
	General limb strengthening with use of resistance	12	Daily (6)	3	Biweekly (2)
	Stationary cycling	11	Biweekly (3); Infrequently (3)	4	Monthly (2)
	Proprioceptive Neuromuscular Facilitation	8	Weekly (3); Infrequently (3)	3	Monthly (2)
	General limb strengthening to fatigue	7	Daily (5)	0	
	Core strengthening to fatigue	6	Daily (4)	0	
	Breathing exercises/voicing /laughing/shouting	6	Daily (2); Biweekly (2)	4	Infrequently (2)

	Core strengthening with use of resistance	5	Daily (3)	1	Biweekly (1)
	Power plate	0		0	
Balance specific Training	Unstable/moving surfaces/equipment	14	Daily (7)	4	Weekly (3)
	Rhythmic stabilisations	11	Weekly (6)	3	Weekly (3)
	Unstable positions	10	Daily (8)	3	Weekly (2)
	External perturbations	5	Biweekly (2); Monthly (2)	4	Weekly (2); Monthly (2)
Electrical stimulation	Transcutaneous Electrical Nerve Stimulation	2	Infrequently (2)	0	
	Threshold Electrical Stimulation	1	Infrequently (1)	0	
	Neuromuscular Electrical Stimulation	1	Monthly (1)	0	
	Functional Electrical Stimulation	0		0	
Gait training	Orthotics used during gait training	17	Daily (10)	4	Daily (3)
	Assistive devices used during gait training	17	Daily (11)	4	Daily (3)
	Facilitation of movement during gait	14	Daily (10)	4	Daily (3)

	Treadmill training	11	Infrequently (6)	0	
Constraint-induced Therapy	Constraint-induced therapy	4	Infrequently (2)	0	
Hydrotherapy	Hydrotherapy	1	Infrequently (1)	4	Weekly (4)
Vojta therapy	Vojta therapy	0		0	
Conductive education	Conductive education	5	Weekly (2)	0	
Motor learning	Motor learning/goal directed functional therapy	11	Daily (9)	4	Daily (2); Weekly (2)

The correlation between the number of therapists using the above interventions at the Institution and at other special schools in Johannesburg was rho=0.68 ($p<0.001$). In other words, those techniques used the most or least often by the physiotherapists at the Institution were used most or least often by physiotherapists at other special needs schools in Johannesburg. The null hypothesis was thus rejected.

When asked if any other treatment techniques not listed on the checklist were used, two therapists at other special needs schools listed hippotherapy, another two therapists listed craniosacral therapy and one mentioned sport-specific training as interventions performed during routine practice. One physiotherapist at the Institution added active mobilisation as an intervention not shown on the checklist.

3.5 Discussion of intervention strategies employed by physiotherapists in Johannesburg

This additional sub-study was added to the original intervention study as, as discussed in section 2.6, for ethical and practical reasons pragmatic trials compare an intervention to the accepted standard/norm of intervention rather than to a placebo (39). As has been described in the literature review (section 2.6), the revised CONSORT statement of 2000 stipulates the "precise details of the interventions intended for each group and how and when they were actually administered" should always be presented in a RCT (40). This increases the external validity of a study and improves its reproducibility (313), as it has been expressed that the greatest confounding variable affecting CP studies is the lack of detail provided on what "usual" therapy entails when control groups continue to receive their accustomed or "usual" therapy for the duration of the study (68).

The objective of this section of the study was to compare the content and intensity of physiotherapy intervention offered by the physiotherapists at the Institution with that offered by other similar facilities in Johannesburg in order to ensure the baseline therapy offered during the intervention period of the experimental study was similar and, thus, results generalisable.

The sample used during this part of the study and the demographics of each school are discussed first, followed by a deliberation of the results found, using the intervention checklist created by the researcher and the frequency of treatment administered at the various schools. A discussion of this part of the study's limitations and any recommendations for future studies follow. This section is summarised and concluded, leading into a description of the intervention study.

3.5.1 Sample and school demographics

There are nine government special needs schools in the Johannesburg area where children with CP receive physiotherapy. School characteristics and treatment frequencies were gained for all of these schools. Of the 25 physiotherapists employed at these schools, one was absent on the day of assessment and two from a single school who requested completing their checklist at a later stage and sending it to the researcher never did so, leaving 22 of a possible 25 participants as part of the included sample. Results therefore represent 100% of the school population and 88% of the total possible physiotherapist population. As the characteristics of the schools from which this missing data came were similar to that of the remaining schools, it is assumed that this dropout rate did not introduce bias into the study.

Both the Institution and other schools combined, showed means of over 14 years' experience for therapists (with large standard deviations at the other schools explained by three therapists employed having qualified in the last 3-6 years). While a United Kingdom study of neurological physiotherapists determined that 88% were NDT trained (170), only 68% of Johannesburg special needs school physiotherapists had completed their full NDT training. When the one-week introductory NDT course was included, this brought NDT trained therapists up to 86% at special needs schools in Johannesburg, in line with these results.

All of the schools included in the sample are government run and children attending generally come from poor socioeconomic backgrounds. The percentage of children with physical disabilities (84.68%) attending the Institution can be assumed higher than the mean (54.74%) at others schools, as the Institution is the only one of the nine special needs schools offering a purely mainstream curriculum for physically disabled learners, while the other schools accept children with learning disabilities in addition, as they offer a vocational/remedial stream.

3.5.2 Results of the practice schedules of the different schools and the intervention checklist

As all of the participating schools have such large physically disabled learner to physiotherapist ratios, group treatments have become necessary in order to provide intervention for as many children as possible attending the schools and seven out of nine schools offer only 30-minute treatment sessions to further accommodate these numbers.

When correlations between those techniques used most or least frequently were calculated between the therapists at the Institution and at other special needs schools in Johannesburg, it was established that that these were statistically similar. All 22 therapists included in the sample stated that they facilitate transitions between positions and only one therapist does not facilitate repetitive movements with assistance or utilise sustained weight bearing positions in order to influence tone. This outcome may have been predicted by the large proportion of therapists working at these schools that are NDT trained and thus implement these NDT based techniques in daily practice.

With this said, it must be considered how this is practically possible when sometimes up to 12 children are treated in a group simultaneously at these schools. As a maximum of four children are seen at a time for 45 minute sessions at the Institution and most children with CP also receive a weekly individual session, it can be surmised that such hands-on approaches to therapy are possible and that treatment aims can be more individualised in these instances at the Institution. It is interesting to note that such a large majority of therapists still employ these techniques, as empirical evidence in their support is generally lacking (see section 2.4.3.1).

The entire sample of physiotherapists also noted that they employ general limb strengthening in regular practice (with their patients with CP). This is aligned with current trends in scientific research stating that strengthening does not influence spasticity, as previously thought (152)(158)(171)(236), and can lead to statistically significant improvements in the muscle strength of targeted muscles when implemented in this population (68)(152)(158)(171)(205)(235)(238). Twenty-one out of 22 therapists also

perform core/abdominal strengthening when treating this population of children and thus it can be concluded that they believe core strength to be essential to rehabilitation in CP.

In contrast, 75% of therapists at the Institution and 66.67% at other schools use any sort of external resistance (other than the child's own body weight) during general limb strengthening, with only 25% and 27.78% of the Institution and other school therapists respectively doing so during core strengthening for this population. Additionally, 38.89% of therapists at the other special needs schools strengthen to fatigue for the limbs and 33.33% for the core, but no therapists at the Institution do so for either.

In order to gain the best possible improvements in muscle force it has been suggested that resistance be increased progressively during training (152)(238)(239). Davidson and Hubley-Kozey have been cited as imparting that in order for core strength gains to take place, loads provided during strength training need to be 60-100% of the individual's one repetition maximum (31). Taylor, Dodd and Damiano have confirmed this by suggesting exercising to fatigue is a means to ensure programmes result in positive changes in torque (236).

As mentioned in the results section (3.4.3), the only techniques not used by any physiotherapists in the total sample were the Power Plate, Functional Electrical Stimulation and Vojta Therapy and no electrical stimulation, treadmill training or constraint-induced therapy were performed at the Institution (but, similarly, only one, 11 and four other therapists use these techniques in practice). On general discussion, no therapists at the nine schools had heard of Vojta therapy but this is not a generally accepted intervention and bares little, if any, scientific evidence to support its use (27). The reason cited by all therapists for all other techniques not used, or used the least often, was limited funding available to the school and thus not having the equipment necessary in order to employ these techniques. On further discussion, it was also established that most therapists had not considered the use of FES in children or in the treatment of CP, nor had they read any research on its potential benefits.

3.6 Limitations

While the checklist of intervention techniques was checked for face and content validity by an expert panel of paediatric physiotherapists, this is still not a standardised measurement instrument and reliability testing was not performed. While it was hoped that the anonymity of completing this checklist would mean that therapists completed the measure honestly (recording only those techniques which they do in fact employ in regular practice), researcher observation of these therapists in practice may have provided more reliable results. Due to the heterogeneity of children with CP (2) and the wide variety of treatment techniques used by these therapists (section 3.4.3) this may have taken weeks, if not months, of observation in order to guarantee all practiced treatment techniques were observed for each individual therapist working in Johannesburg special needs schools. During a pragmatic trial, and due to limited resources, this would not have been practically possible. Observation may also have provoked therapists to perform their idea of 'best practice', rather than what actually occurs in the therapy department on a daily basis.

3.7 Recommendations

- Should this sub-study be reproduced in future, it is recommended that those interventions used by therapists not present on the intervention checklist are added and reliability studies are performed before use.
- Further detail on the timing and intensity of interventions used and for which particular classifications or ages of children with CP should be established with future use of the intervention checklist.
- The outcome measures employed by therapists for assessing the impact of therapy provided should be included to further clarify whether impairment, activity limitation or participation restriction are addressed during goal setting and whether intervention provided is successfully assisting in achieving these goals.

3.8 Conclusion

This sub-study showed that all nine government, special needs schools in Johannesburg, where physiotherapeutic intervention is offered to children with CP, have experienced therapists in their employ, who are mostly NDT trained. All of these schools share the problems of limited resources, both in terms of financial support and staff allocations. The majority of these therapists follow NDT principles during daily practice and aligned with current research, use strengthening as a regular rehabilitative tool for this population of children treated. Limitations in their implementation of this modality were seen, in that few therapists use resistance or working until fatigue during strengthening programmes for children with CP. Those techniques used least frequently at these schools were also those techniques shown to have the least scientific evidence to support their use, in the earlier literature review.

As the intervention techniques used most and least frequently at the Institution were strongly correlated with those used most and least frequently at the eight other participating special needs schools in Johannesburg, one can conclude that the baseline therapeutic intervention performed at the Institution was similar to that offered by the greater population of neurological physiotherapists treating children at government special needs schools in this area. In compliance with the CONSORT statement of 2000 (40), the typical therapy both the control and experimental groups in the intervention study below will receive has been explained in detail and is comparable to that of the greater population, thus ensuring the external validity and reproducibility of this study. No therapists at these schools employed the use of FES in their treatment of children with CP, when assessed. As preliminary evidence has shown the potential for positive improvements, in those with CP, in spasticity, gait parameters and muscle strength (as well as cost-effectiveness), the need for a study of high methodological quality, investigating this modality in children with CP in Johannesburg exists. The intervention study that took place will now be systematically described below.

4. Intervention study

As has been described at length throughout the literature review and sub-study above, the potential for strength gains which may lead to clinically important postural and functional gains in children with CP, with the use of FES, exists. The sections below describe an experimental study examining the effectiveness of this modality, used over the abdominal muscles, in a sample of children with CP in Johannesburg. The methodology, procedure and data analysis used are described in detail, followed by the results gained during the study and a discussion of these findings. Limitations experienced and recommendations for future studies are given before concluding this dissertation.

4.1 Methodology

4.1.1 Aims and objectives

The overall aim of the study was to investigate the impact of FES on abdominal muscle strength and gross motor function in children with cerebral palsy.

The specific objectives of the experimental study were:

- To determine whether concomitant therapeutic intervention using FES would result in a significantly different improvement in children with cerebral palsy, in comparison with a control group of children with cerebral palsy receiving only routine physiotherapy, in the following parameters over time:
    - Abdominal muscle strength as measured by the number of sit-ups completed in one minute
    - Gross motor function, with particular emphasis on balance, as determined by scores on the Gross Motor Function Measure-66 (GMFM-66), Pediatric Reach Test (PRT) and the Timed Up and Go (TUG)
    - Respiratory function, as measured by Peak Flow Meter readings

- Self-perceived health-related quality of life (HRQoL), as assessed by the Pediatric Quality of Life Inventory 3.0 CP Module (PedsQL) and the participant evaluation of the intervention
- To determine if a relationship exists between demographic and medical characteristics and improvements in functional status made by participants

4.1.2 Research question

Does FES administered regularly, in conjunction with typical physiotherapeutic intervention, over a period of six weeks, improve abdominal muscle strength and gross motor function in 5-18 year old children with cerebral palsy compared to routine physiotherapy treatment?

4.1.3 Null hypothesis

The addition of FES to routine physiotherapy treatment will lead to no significant difference in abdominal strength or percentage total scores of the GMFM-66 compared to routine treatment.

4.1.4 Research design

Double-blind randomised control trials (RCT) have been described as the "gold" standard for research methodology in that they are able to produce evidence that is unmarred by bias allowing results to be generalised and research methods repeated (309).

An experimental, RCT with single blinding was used for this section of the study. As the intervention was active, and apparent, double blinding was not possible as both the participants and the treating therapists were aware of which children were receiving the intervention. A pragmatic design was employed as such studies are successful in determining the effect of an intervention compared to typical conventional treatment in everyday practice. Larger samples and outcome measures of function and HRQoL are selected to ensure results have wide generalisability to different clinical circumstances (39).

4.1.5 Sample/participants

A sample of convenience was taken from the Institution at which the researcher works; a special needs school in Johannesburg. All children with CP were eligible for inclusion and randomisation was stratified based on their GMFCS level.

4.1.5.1 Eligibility criteria

Inclusion criteria met by participants were as follows:

- Enrolled full time at the Institution
- Aged 5-18 years
- Diagnosed by a medical practitioner as having CP

Please note: The upper limit of the inclusion age group was selected as the South African Children's Act defines a child as "a person under the age of 18 years" (316). The lower limit for inclusion in this study was chosen as younger children may not adequately understand HRQoL measures, and the PedsQL selected for use in this study has been validated for use in children from 5-18 years (see section 2.5.3.2).

Exclusion criteria included:

- Uncontrolled epilepsy
- Surgical or other medical intervention scheduled during the intervention period or up to three months prior to the trial
- Participant unable to give clear voluntary assent and/or no written consent given by parent or guardian (Appendix VII/VIII)

4.1.6 Research setting

Research took place at the Institution; a government-subsidised special needs school for children with physical disabilities in Johannesburg. The school offers mainstream education and therapy to children with a variety of disabilities (including CP) from nursery school level until grade 12/matric. The curriculum is taught in English. Most learners enrolled reside in the school hostel during the week. The majority of learners attending the school come from poor socioeconomic circumstances and thus receive bursaries to cover their schooling and hostel costs. A multi-disciplinary team approach to therapy is employed whereby (if indicated) children receive physiotherapy, occupational and/or speech therapy on a weekly or biweekly individual or group basis. Due to the socioeconomic status of the vast majority of school learners, the therapies received during the school term are the only interventions received by these learners and thus, during 2-6 week long school holidays (four times a year) these children receive no further intervention. As described in the results section of the earlier sub-study (section 3.4.2 School characteristics and 3.4.3 Treatment characteristics), the Institution is the only facility in the area offering a mainstream curriculum up until matric and thus many learners from neighbouring schools transfer to the Institution in grade seven or nine (when aged approximately 13-15 years old). On acceptance to the Institution, as described, many of these children ceased receiving any therapies at their previous schools after the age of 12.

4.1.7 Sample size determination

The 2010 CONSORT statement recommends that sample size calculations be included in the reporting of RCTs (310). Calculating the necessary sample size allows authors to state with certainty that the results found are able to detect a true difference in effect size (25) and that the study was powerful enough to detect statistically significant changes in outcome parameters (227)(310)(317).

While statistical significance is used to determine whether an important change occurred following a study, that may not have resulted purely by chance, determining whether an intervention resulted in a change that is clinically meaningful, or perceived to be beneficial by the participant, is more important. By ascertaining the minimum clinically important difference (MCID) following an intervention, one can determine the proportion of participants that would respond positively to the same intervention, should the study be repeated. This is important, as improvements seen on a specific outcome measure do not necessarily equate to observable improvements in the participant's disability or QoL etc (318)(319)(320).

To clarify: while minimally detectable change represents the smallest possible detectable change on an outcome measure (usually that falling outside a measurement error or standard deviation of 0.5), the MCID depicts the smallest possible clinical change which is seen as beneficial by the participant or clinician (319). This is based on the responsiveness of a measure, which is its ability to accurately measure meaningful change over time. Unfortunately, little consensus exists over exactly how much change is clinically sufficient, as measured by specific outcome determinants. This is further complicated by the widely varied rehabilitation goals set for individuals with disabilities whose populations are heterogeneous by nature and, when measures with a large number of items may not reflect clinically important changes that take place when only a few items show change/improvement (272).

The primary outcome measure of the study is the GMFM-66 and was thus used to determine the required sample size for the study. Numerous studies have evaluated the

responsiveness of the GMFM-66, with changes in scores measured over time (from 1-6 years) varying between an absolute maximum change observed of 7% (for participants classed GMFCS levels I and II only) (274) and “moderate” improvements considered as between 3.7 and 1.7 (274)(223)(320).

It was anticipated that a 10% increase in performance in the GMFM-66 would result in observable improvements in gross motor function; a large enough magnitude improvement to give rise to clinically significant functional gains positively influencing the lives of the individual participants. Based on the mean scores and standard deviations from a previous study (264) and the parameters outlined in the table below (Table 7), a sample size of 24 was deemed necessary per group for a one-sided t-test (STATISTICA version 11 power calculation).

Table 7: Sample size calculation

	Value
Population Mean Mu1	80.6
Population Mean Mu2	90.6
Population S.D. (Sigma)	13.6
Standardized Effect (Es)	-0.74
Type I Error Rate (Alpha)	0.05
Critical Value of t	1.68
Power Goal	0.80
Actual Power for Required N	0.81
Required N (per group)	24

4.1.8 Instrumentation

The study intervention is described below. The details gathered by the socio-demographic data collection form created by the researcher, to ensure all personal parameters were available for comparison during data analysis, are then presented. The standardised outcome measures used and the application of each are then described in detail.

4.1.8.1 Functional Electrical Stimulation

Microstim 2 (v2) units were purchased new from Professional Functional Electrical Stimulation South Africa (PROFFESSA), a local subsidiary of the Salisbury Healthcare National Health Service Trust. The units were then programmed by a trained PROFFESSA employee to ensure proper calibration. Those studies reporting statistically significant improvements in children with CP after using FES/NMES, over various muscle groups, used the modality for 15-60 minutes, 6-7 days a week during the study intervention period (36)(38)(41)(260). Significant improvements in the abdominal strength of adults without neurological fallout were seen with NMES use five days a week for eight weeks (256), as well as improvements in the lower limb muscle strength and gait parameters of stroke patients with FES use for only 15 minutes, three times a week (218). This frequency of use was made possible as in most of the studies the participants used the ES devices at home (36)(256)(260). The frequency settings used in these studies with positive outcomes varied from 20-40 Hz, with the majority in the lower range (36)(38)(41)(218)(260).

On discussion with the PROFFESSA representative, it was established that Odstock Medical (the manufacturer of the Microstim 2 units to be used in this study), governed by the National Health Service in the United Kingdom, advocate use of their FES unit for 20 minutes, five times a week. This is to encourage eventual integration of unit use into everyday living and not only during therapy. It was also discussed that a frequency of 20 Hz may best serve the function of stimulating tonic muscles and provide the greatest level of comfort for use in the abdominals of child participants (personal communication with Deon Buhrs, owner and representative of PROFFESSA, January 2013).

The aim of this pragmatic intervention study was to incorporate FES into routine physiotherapy treatment (in order to establish whether use during functional activities may lead to improvements in independent function, as well as the anticipated strength gains). Participants would usually receive physiotherapy intervention once or twice a week and so, in order to maximise the stimulation time, within the pragmatic constraints, additional sessions of stimulation were added. Over and above the two FES sessions a week performed for 20 minutes during therapy sessions, two additional passive, 20-minute FES sessions were added before school or during lunch breaks weekly during the intervention period. During these sessions, children were able to continue with their usual social interaction or play throughout stimulation, ensuring these more "passive" sessions still took place during functional activities.

The appropriate mode (i.e. mode 4) remained programmed on each unit throughout the study and machines were not used in the treatment of patients outside the study to ensure exact settings were used on each occasion. Children were not allowed to handle the units for safety of use and strict adherence to necessary precautions was observed.

Parameters for FES intervention were set at a stimulation frequency appropriate for postural muscles of 20 Hz (as discussed above), 300 μs pulse width and output amplitude of 0-100 mA for 20 minutes at a time, applied over the external oblique muscles of each participant (154). The external obliques were selected, as they are considered primary core stabilising muscles but are large and superficially situated, allowing for easier electrical stimulation than deeper core muscles (181). Their rotatory action is also central to the maintenance of balance and stabilising of the pelvis for upright posture (see section 2.3.1.1 Musculoskeletal impairments for more details).

The external obliques originate from the external surfaces of the fifth to twelfth ribs and insert into the linea alba, pubic tubercle and the anterior half of the iliac crest. The muscle fibres therefore run inferomedially. The muscle is innervated by the inferior six thoracic nerves and the subcostal nerves (321). This is illustrated in Figure 5 below. Two electrodes of an appropriate size for the size of the participant were applied bilaterally to the external obliques. Therapists used points superior-laterally from the umbilicus above the eleventh rib and superior to this below the fifth ribs as landmarks for application. The output stimulation intensity was modulated per participant, based on the individual's comfort

levels and ramped upward as the person became more accustomed to the sensation (251). Treatment started once the patient was comfortable and visible muscle contractions of the left and right external obliques were simultaneously evident.

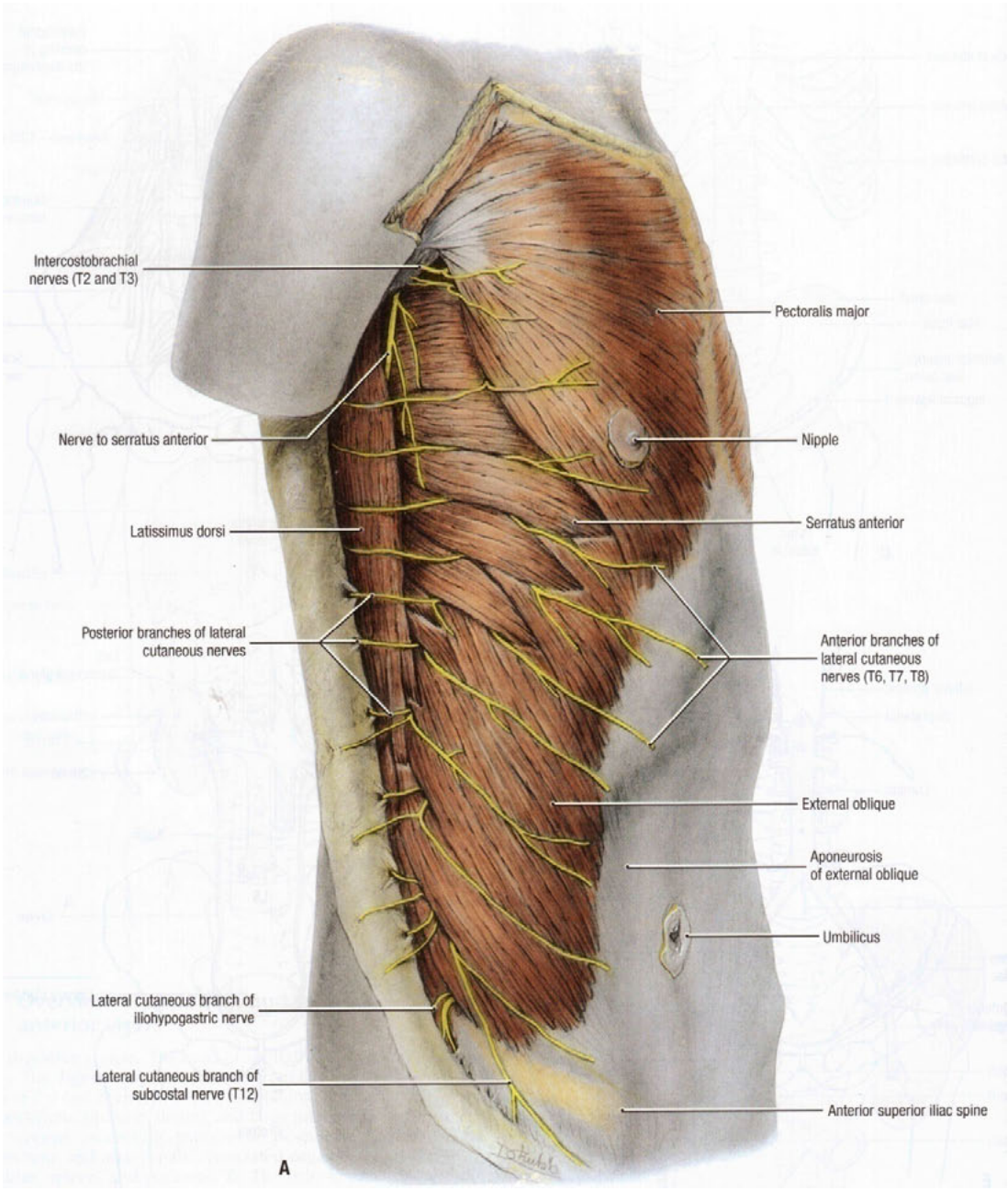


Figure 5: Anatomy of the external oblique muscle (adapted from Agur and Lee (1999) (321)**)**

4.1.8.2 Socio-demographic form (Appendix IX)

Information was drawn from the Institution's learner profiles. Basic demographic data, such as participant name, age, gender and contact numbers for ease of communication with relevant parents/guardians regarding the study, were recovered from these records. Home language was included in data collection to ensure the accurately translated version of consent forms were sent to parents. As suggested by the International Workshop on Definition and Classification of Cerebral Palsy panel (2)(89), all possible classification data was also included on the data collection form for eligible participants. This included clinical observations of secondary impairments, the primary type of motor disorder observed and the anatomical distribution of CP noted for each participant by their treating physiotherapist (1). Where possible, the causation and timing of CP were also recorded (1)(85). The treating physiotherapist of each possible participant was listed and the GMFCS level of each child determined (1)(100). Other therapies received by participants at the Institution, previous surgery and the date of surgery, if relevant, was also recorded. This information was used to determine whether possible participants met the criteria for inclusion in the study, as well as providing background information relating to participant functional level.

4.1.8.3 Gross Motor Function Classification System (GMFCS)

The GMFCS was chosen for use in stratifying the sample as it classifies the motor functioning of children with CP until 18 years of age, using five standardised levels (95). Studies of validity and reliability are well established and its use has become increasingly important in clinical decision-making and practice (97). The researcher and the three other physiotherapists working at the Institution discussed each of the study participants, coming to a consensus regarding a GMFCS level allocation for each.

4.1.8.4 Gross Motor Function Measure -66 (GMFM-66) (Appendix X)

The GMFM is a therapist-administered test able to determine the extent to which a child is able to perform certain motor functions. It has been specifically developed for use in children with CP and has been shown to be responsive to change as well as the absence of change (128). It is regarded as the "gold standard" for gross motor measurement and has been shown to have intrarater, interrater and test-retest reliability (298). Both the GMFM-88 and GMFM-66 versions have shown impressive longitudinal construct validity in a long-term follow up (65).

As outlined by the GMFM user's manual, the GMFM-66 was administered (by a trained external assessor blinded to group allocation) in a private and spacious gym with the required equipment constant for pre- and post-intervention testing. Children were made comfortable in order to move freely and were tested barefoot. A test trial was demonstrated or facilitated if the item was not understood but all scorable attempts had to be independently performed. A maximum of three trials were allowed according to manual recommendations (including any spontaneous observations of the desired task) and the best attempt was scored. Scoring was based on the guidelines outlined for each item in the manual and all reasonably attainable items were at least attempted. A "Not Tested" was recorded if the participant refused to complete that particular item. Scores were loaded onto the Gross Motor Ability Estimator (GMAE) computer software for a percentage score calculation (see 6.6 for a description for this programme (322).

4.1.8.5 Pediatric Reach Test (PRT) (Appendix XI)

Many measures of balance exist but are mostly time consuming or utilise expensive computerised equipment (278). The Functional Reach Test (FRT) was originally designed to measure balance for the prevention of falls in the elderly and has since been modified to allow for use in sitting or standing as well as for forward and sideways reach. The Modified FRT has been adapted for use in children with CP by creating the PRT (323). Preliminary studies (278) have shown this simple tool to be valid and reliable with moderate-to-

excellent test-retest reliability and construct validity in both typically developing and disabled children.

Participants were all able to sit independently. They were placed in a comfortable seated position, with their feet supported and told to keep their backs upright by a trained external assessor blinded to group allocation. This assessor requested they hold their right upper limb ahead of them in 90 degrees shoulder flexion with elbow extension and a neutral wrist. The length from their acromioclavicular joint to the third metacarpal phalangeal joint was measured with a measuring tape by the research assistant standing behind the participant. They were then asked to reach forward in this position as far as they could and maintain this position for three seconds whilst a new measurement from their starting shoulder position to their final end point was taken. This same process was repeated laterally to the right and left sides. A trial attempt was allowed before the final measurement was recorded. All three of these measurements were also tested in a static standing position wearing the participant's usual footwear and orthoses (if indicated). The distance reached was determined by the difference between initial and final measurements in each position. If the participant was unable to attain the desired upper limb posture, maintain a static sitting or standing posture independently for 15 seconds, or maintain the reaching position without fallout or taking a step then measures for this particular item were not recorded.

4.1.8.6 Timed Up and Go (TUG) (Appendix XII)

As all levels of the GMFCS were represented by the sample, it was necessary to include a measure for higher functioning children who may encounter a ceiling effect on the GMFM, in order to prevent inadequate representation of possible improvements made (324).

The TUG is a simple performance-based test with a low respondent and administrative burden that assesses basic mobility and balance (originally designed for the purpose of falls prevention in the elderly). It has been shown to have high intrasession stability and interrater reliability (282). It is also reliable and valid for use in those with physical

disabilities from the age of 3-19 years and is able to differentiate between the highest levels of the GMFCS, showing responsiveness to change over time (284).

Participants were seated in a wooden desk chair with their back flush to the seat backrest (by a trained external assessor blinded to group allocation) and wearing their usual footwear and orthoses (if indicated). They were requested on the command "go" to stand from the chair, mobilise forwards three metres to a pre-marked line at a comfortable pace, turn and then return the three metres and be seated again. Participants that could not walk independently could use their usual assistive device during testing. A trial was possible prior to time keeping if the directions were not understood and measurement was made in seconds by the use of a stopwatch used by the research assistant. (Note that the lower the score in seconds, the faster the walking time).

4.1.8.7 Timed Sit-Ups

Hand-held dynamometers and other laboratory-based isokinetic devices have been used in the measurement of abdominal strength in previous studies but are not financially viable options for use at a government-funded school. The total number of sit-ups an individual is able to complete in one minute can serve as an alternative measure of abdominal muscle strength and endurance in this instance (160).

A trained external assessor blinded to group allocation assisted with implementing the timed sit-ups. A position in supine, comfortable to the individual participant, was used as the starting position (either with extended lower limbs or in crook lying). The participant was then instructed to complete as many 'crunches' as was possible in 60 seconds, as timed by a stopwatch used by the research assistant. For more capable participants, this procedure was completed independently with the upper limbs extended at their sides reaching up toward their flexed knees, crossed behind their heads or across their chests, at their own discretion. For those participants with higher GMFCS levels, the hands were allowed to pull up on their pants to assist and/or their feet could be supported passively by the research assistant (if necessary). 'Crunches' were only counted if the head, humeral

heads and scapulae left the ground. On post-intervention evaluation, the preferred starting position at pre-intervention evaluation was noted for the assistant on the assessment form (specifying both upper and lower limb position), to ensure this exact position was used again post-intervention, for consistency.

4.1.8.8 Peak Flow Meter

The major morbidity and mortality seen in CP has been related to respiratory compromise (198). As the abdominals play such an important role in breathing and respiratory function, it was necessary to add an outcome measure to determine whether FES over the abdominal muscles has any effect on respiratory function. The Peak Flow Meter has been widely accepted as a simple, easily administered, portable tool for the assessment of obstructive respiratory disorders in children (292).

Children were instructed, by a trained external assessor blinded to group allocation, to sit upright, close their lips over the mouthpiece and take a large, deep breath in. They were then asked to exhale as hard and as fast as possible keeping lip closure over the meter. As suggested by the American Lung Association, the highest reading of three attempts on the Peak Flow Meter was recorded for each participant and the outcome tool was sterilised before use by the next participant (293).

4.1.8.9 Pediatric Quality of Life Inventory 3.0 CP Module (PedsQL) (Appendix XIII)

While physical function is most often evaluated in children with CP, due to its difficulty to quantify and the varying effect of social environment and experience, HRQoL is often neglected in study. In their research, Kerr, McDowell & McDonough (208) found a significant correlation between gross motor function and restriction in participation. It is

with this and the WHO's emphasis on participation and inclusion in mind that the PedsQL was included in the study.

While other HRQoL measures are only validated between the ages of 8 and 12 (305), the PedsQL 3.0 CP Module has been validated for self-report between the ages of 5-18. This measure shows reliability, construct validity and sensitivity among children with different diagnostic categories of CP (67).

The appropriate version of the PedsQL (i.e. ages 5-7, ages 8-12 or ages 13-18) was allocated to each participant. While this measure has not been validated for use in the South African context, cognitive debriefing was provided by the research assistant (a trained external assessor blinded to group allocation) and the Institution's physiotherapy assistant (who was not otherwise involved in the study), in a language each participant could understand, in order to ensure children were able to comprehend exactly what was being asked of them (325). As was done in a study by Varni et al, if unable to write, the measure was read to the participant and their verbal response or pointing at the appropriate answer was recorded by the research assistant (67). If the participant was unable to communicate their response in any way or if they did not understand the measure, scores were not recorded for that particular child.

4.1.8.10 Participant evaluation (Appendix XIV)

By introducing a biopsychosocial model of disability, the ICF recognises the importance of participation in and interaction with one's environment, thus introducing the need for research and clinical practice to be more relevant to the lives of each individual (3). It is for this reason that a participant evaluation was included so that any self-perceived benefit or negative impact of FES intervention could be duly noted. The evaluation is the only measure that was only completed at the end of the intervention period and was explained to and completed by participants as above for the PedsQL. A simple visual analogue scale was created by the researcher to determine what those participants in the intervention

group thought of the sensation of the FES, whether they would like to use it again and whether they thought it helped them in any way. This scale was adapted from that used for younger children on the PedsQL where happy, indifferent and sad faces were used to assist answering “not at all”, “sometimes” and “a lot” to the related questions on the measure. Eiser, Mohay and Morse suggested the use of simple pictures to assist young children in understanding what is being asked of them in subjective outcome measures (307). A space was provided for any further comments participants wished to make following the intervention period.

4.2 Procedure

4.2.1 Ethical considerations

4.2.1.1 Approval

As was explained earlier, in section 3.2.1.1, ethical approval was obtained from the necessary statutory bodies as well as the principal of the Institution and the study was registered with the Pan African Clinical Trials Registry. All ethical principles delineated by the World Medical Association Declaration of Helsinki were adhered to throughout the study (326).

4.2.1.2 Risk to participants

The principal of beneficence was observed in that both the control and intervention groups continued to receive their usual weekly or biweekly physiotherapeutic intervention, thus ensuring no harm in loss of treatment time or in a greater time spent outside of the classroom compared to normal.

The only possible side effects of treatment with FES (as long as contra-indications mentioned in the exclusion criteria are observed) are minor skin reactions. This adverse effect was not encountered by any of the participants. Individuals decided on their own comfort level in terms of FES intensity and informed their treating physiotherapist if they would like to either increase or decrease this intensity during use, thus respecting the principal of non-maleficence.

4.2.1.3 Benefit to participants

This study was undertaken due to preliminary evidence to show that FES intervention in children with CP may have positive effects on function. Were this to be observed in the results of the study, children in the intervention group would immediately benefit as well as providing empirical evidence to either substantiate or negate the use of FES, thus further expanding evidence bases currently available.

4.2.2 Training of investigators

Intervention was carried out by the four physiotherapists employed at the Institution (including the primary researcher), none of whom had worked with FES before. The therapists had a mean of 17.75 years (SD=7.89) practical experience between them ( ). In order to ensure regulated use, prior to study commencement, a training session with the owner of PROFFESSA, the manufacturer of the FES device used, was undertaken. All four treating therapists attended.

The treatment protocol was discussed and use of the FES device was practiced on one another preceding use on study participants.

4.2.3 Data collection

Data collection commenced on the 7 January 2013, when children returned to the Institution after a 5-week vacation. Socio-demographic data was collected and eligibility criteria was assessed, based on medical records for each child held at the Institution, as well as the inclusion/exclusion criteria listed previously (see 3.1.3.1). As the researcher is employed at the Institution, this did not breach confidentiality. Children were classified based on the nature and typology of the motor disorder, anatomical distribution, and causation and timing (where possible) as prescribed by Rosenbaum et al (2).

Information leaflets and consent forms containing relevant information about the study, any risk factors involved and their right to either not consent to the study or to withdraw their child at a later stage, were sent home to the relevant children's parents/guardians in English (Appendix VIII). Non-English first language speaking parents also received a translation of the information sheet and consent form in Zulu, if speaking an Nguni language (Zulu or Xhosa) (Appendix XV) or in Tswana if speaking Tswana, North or South Sotho or Venda as these languages are all understood if speaking one of them (Appendix XVI). The remaining South African languages were not spoken by the sample. Only children who returned signed consent forms were considered for inclusion in the study. There was no fiscal compensation for participants.

Participant autonomy was respected as children were required to give voluntary written (or verbal, if this was all that was possible) assent to both assessment and intervention (Appendix VII) and it was made known that they were able to withdraw from the study at any given time.

All demographic information and outcome measure results were kept in individual folders for each participant, numbered on the exterior of the file for confidentiality purposes and stored in a safe location. Participant names were not included in the socio-demographic or assessment data files. Only the principal investigator and supervisor had access to this information and no names were mentioned during any discussions regarding this dissertation.

4.2.4 Pilot study

As only standardised outcome measures were used, their validity and reliability did not need to be tested. However, the reliability of use within the research context did need to be established. An experienced paediatric physiotherapist, with no knowledge of the participants, was employed to administer all assessments in order not to introduce confounding variables to the study. Interrater reliability of the measurement instruments was determined by comparing outcomes obtained by the simultaneous rating of the performance of children not participating in the final study, by both the researcher and an assessor who did not participate in the socio-demographic data collection or group allocation. Rating of two children resulted in 90% agreement on all items in both children. Therefore, a priori criterion for reliability was met and the ability of the external assessor to reliably assess children was deemed satisfactory. This also helped establish that the intended assessments would take between 30-45 minutes to complete for each participant.

Prior to commencement of the study, each of the four physiotherapists involved piloted the use of the FES devices on one child who was not involved in the final study as they did not meet the inclusion criteria or had undergone recent surgery. This was done to ensure familiarity of use, practice for explanation to the child and implementation concurrent to normal weekly physiotherapy intervention. No problems were encountered prior to study commencement.

4.2.5 Randomisation

The researcher and the three other physiotherapists working at the Institution discussed each of the study participants, coming to a consensus regarding a GMFCS level allocation for each. Participants were then stratified into their relevant GMFCS levels and names were placed into five envelopes representing each level on the score. It was not possible to see the names of participants once inside the envelope. The first name drawn from each envelope, by a therapist not involved in the study, was allocated to the control group and the second to the intervention group and so on until each participant was allocated.

The trained assessor evaluated each child with the above-mentioned outcome measures from the 9-16 January 2013. Assessment took place in a quiet, private physiotherapy gym with all necessary equipment for the outcome measurements used readily available. To ensure outcome determinants were not confounded, the external assessor performing standardised measures was blinded to group allocation and treating physiotherapists were not involved in pre- or post-intervention assessments.

4.2.6 Intervention

The control group continued to receive their usual physiotherapy weekly or biweekly intervention with their usual physiotherapist, as did the intervention group (see results section 4.4.1 Sample for specifics regarding treatment frequency). The experimental group received concurrent FES for a 6-week period during their usual weekly or biweekly physiotherapy sessions, as well as an additional two passive sessions a week during lunch breaks or before school (from 21 January to 8 March 2013), as has been discussed in section 4.1.6.1 previously.

As stipulated, the area was cleaned and free of skin care products prior to the application of self-adhesive electrodes (the appropriate size for the specific child) (251) to the bilateral external oblique muscles. The Microstim units were set at the parameters outlined previously and leads and the unit placed in a sealed plastic bag and attached to the child's pants so as to prevent children playing with the controls and to ensure ease of movement. Please see 4.6.1.2 for a detailed account of the FES application and parameters used in the study.

The same assessor then re-evaluated children from both the control and intervention groups six weeks later, using the same measures as performed pre-intervention and, in addition, the participant evaluation, from 11-14 March 2013.

4.3 Data analysis

In order to discuss the outcome of a study statistical analysis needs to take place. Effect size signifies the size of change taking place as determined by standardised outcome measures (65). Power calculations allow researchers to determine whether the sample included and the study design chosen are able to detect a "true difference" in effect size indicating that two intervention strategies are statistically significantly different from one another (25). Such calculations are also essential for preventing type II errors where too small a sample size equates to an intervention that has caused a significant change but the study design is not powerful enough to demonstrate this (227)(310)(317). For this reason the 2010 CONSORT statement for RCTs requires the inclusion of sample size calculations in such studies (310). An alpha threshold or p value of less than 0.05 signifies that two sample groups are inherently different and allows one to reject a null hypothesis with more than 95% certainty. When a result has a p value of < 0.05 it is said to be statistically significant (311)(327). A type I error occurs when a significant result is found when in actuality none exists. Without power analysis, it is difficult to accept reported significance at face value.

Variance can be described as a measure of the degree to which sample participants deviate from the mean. To calculate standard deviation one obtains the square root of the variance. The lower the variance the smaller the sample need be as the sample is more likely to represent a wider population (327).

Graphs are often used to condense statistics into more interpretable formats. The frequency distribution of categorical variables can be demonstrated in bar graphs or pie charts and that of numerical data in a histogram. The appearance of either graph will demonstrate whether data is symmetrical or 'normally distributed' (i.e. parametric). This, in turn, dictates the statistical tests that will be used to analyse data (311). Other deciding factors for the selection of specific statistical tests include the nature of the variable (categorical or numerical), whether comparison is being made between two or more groups and whether the groups are independent from or related to one another (311).

When two categorical, independent variables are compared, Chi-squared or Fishers' Exact tests are used, while paired variables make use of McNemar's test for analysis. Ordinal or numerical variables, on the other hand, make use of a t-test for comparing two independent variables and a paired t-test for paired variables. If numerical/ordinal data is skewed (i.e. non-parametric) two independent variables can be compared using the Wilcoxon rank sum or Mann-Whitney U (MWU) tests and when paired, the Wilcoxon signed rank test. Should more than two numerical/ordinal variables be compared, then parametric data will employ a one-way analysis of variance (ANOVA) or a repeated measures ANOVA and non-parametric data the Kruskal-Wallis test or Friedman two-way ANOVA for independent and related variables, respectively. More specifically, an ANOVA determines the significance of between group and within group differences between two or more means, when data is normally distributed (328). The one-way ANOVA is reported to be robust against violations to the normality assumption (329)(330) and the test can tolerate analysis of non-parametric data with minimal potential for a Type I error (328).

In order to determine linear relationships between parametric variables, a scatter plot of all data is drawn and the Pearson correlation coefficient or 'r' is calculated. If r is found to be zero then no linear relationship exists between the variables. An r of -1 or 1 denotes a perfect linear relationship in one direction or the other but this is rarely the case as certain outliers tend to exist for any set of data. Relationships between ordinal or non-parametric variables are calculated using the Spearman correlation coefficient (which also ranges from -1 to 1)(311).

During this study, demographic characteristics and data from all pre- and post-intervention assessments were entered into Excel spreadsheets and the STATISTICA version 11 software.

Descriptive statistics were used to portray the demographic and medical characteristics of the participants. The data was tested for normality and, if normally distributed, parametric tests were used. If not, non-parametric tests were employed. Ordinal data was analysed using non-parametric tests. The two groups were compared at baseline to ensure that they were equivalent. Within group differences (paired t-test or Wilcoxon signed rank) and between group differences (independent t-test or MWU), from baseline to post-intervention, was determined. As the data were not normally distributed and the Kruskall

Wallis test does not allow for a repeated measures ANOVA with two factors, the Wilcoxon signed rank test and the MWU tests were used to determine within and between group differences. The difference in the amount of change, exhibited by the two groups, will be included in future publications.

The ANOVA was used to determine if participants classified at different levels of the GMFCS or of different ages responded differently to intervention. Correlations (Spearman's and Pearson's) were performed to determine if changes in the different parameters were correlated with one other (e.g. did a gain in the number of sit-ups possible correlate with a gain in GMFM-66 score?)

There are several different methods of calculating effect sizes for non-parametric statistics such as Cliff's delta (by enumerating the number of occurrences of an observation from one group having a higher response value than an observation of the reverse), quantile absolute deviation (dividing the z value by the square root of N) (331) or the U value by the product of the two sample sizes (332). As there appears to be little consensus on which test to use and how to interpret the different values calculated using the different methods, effect sizes were not included in this analysis.

Table 8 below shows the specific statistical tests used to analyse study data.

Table 8: Summary of statistical tests used

Question	Outcome measure	Test	Type of data
Were the demographic characteristics and outcome measures normally distributed?	All	Shapiro-Wilk test	Various
Were the two	Gender, language, CP cause, all	Chi-squared test	Categorical

groups equivalent?	secondary impairments, CP motor disorder, anatomical distribution, previous surgery and other therapies received		
	Age, gestational age, GMFCS level	Mann Whitney U test	Numeric non-normal, ordinal
Was there a within group difference from baseline to post-intervention?	Peak Flow	Paired t-test	Numerical - normal
	Timed sit-ups, GMFM-66 and TUG	Wilcoxon signed rank test	Non-parametric
	PedsQL and participant evaluation	Wilcoxon signed rank test	Ordinal
	PRT	Note: Within group differences were not calculated for this outcome measure as there were too many variables, with small subgroup sizes, to perform this analysis on in this particular test.	
Was there a between group difference from baseline to post-intervention?	PRT	t-test for equal variances	Numerical - normal
	Peak Flow	Independent t-test	Numerical
	Timed sit-ups, GMFM-66 and TUG	Mann-Whitney U test	Non-parametric

	PedsQL and participant evaluation	Mann-Whitney U test	Ordinal
Was there a difference in the amount of change in GMFM-66% for children of different ages and different GMFCS levels?	GMFCS level, age, motor disorder and anatomical distribution of CP	ANOVA	Numerical and categorical
Were changes in GMFM-66% scores correlated with other outcome measure scores?	GMFM-66, TUG, peak flow	Pearson's correlation	Numerical
Was there a difference in the amount of change in HRQoL for children of different ages and different GMFCS levels?	Age, GMFCS level	ANOVA	Numerical and ordinal

4.4 Results of the intervention study

Results for the sample included in the study are presented first, followed by their demographic data and medical characteristics for both control and intervention groups. The impact of the intervention, as measured by each of the outcome measures employed, is discussed next. Finally, results are presented for how participant demographic characteristics influenced changes in functional status.

4.4.1 Sample

Information leaflets and consent forms were sent out to the parents or guardians of 56 children enrolled at the Institution who met the outlined inclusion criteria. Of these, 51 returned consent forms. Four of these were then excluded from the study as they were scheduled for surgery within the intervention period. Forty-seven children were stratified, according to their GMFCS level, and then randomised into control and intervention groups (as described in section 4.2.5). Ten children in each group did not begin their schooling at the Institution, but were transfers to the school in their teens, having not received physiotherapeutic intervention since the age of 10-12. All of these children, aged between 14 and 18, began physiotherapy immediately on admission to the school. Six of these 20 participants only joined the Institution in the year the intervention study took place. Three, aged 14, 15 and 16, were randomly allocated to the intervention group and three, two aged 16 and one 17, to the control group. It is assumed that these six children had not received intervention for the last 2-5 years as all of the other special needs schools in the area cease treatment when children reach their teens.

All 47 participating children completed the six-week intervention, as well as all pre- and post-intervention outcome assessments. However, as not all measures were applicable to each child and some participants were unable to complete the PedsQL measure, there was some missing data (refer to Figure 6 below). A summary of the frequency of physiotherapy received by participants during the six-week intervention period can be found in Table 9. There was no difference in the mean number of treatments for each group, $t=1.46$, $p=0.925$.

Table 9: Summary of frequency of physiotherapy treatment of control and intervention groups over the six-week intervention period

	Sample size	Total Treatments	Mean Treatments	Standard Deviation
Intervention	22	224	10.18	2.20
Control	25	230	9.2	2.29

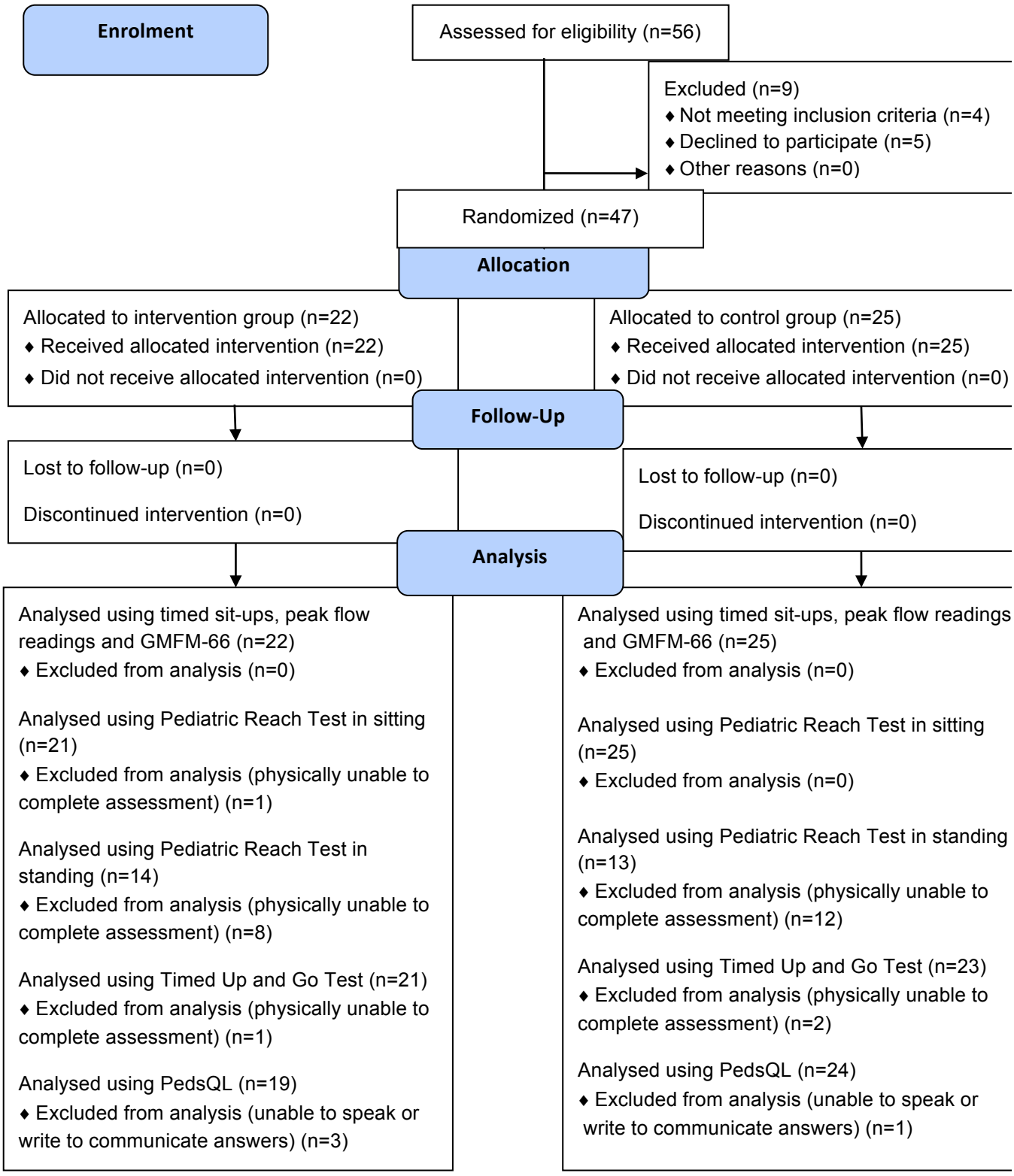


Figure 6: Flow diagram of recruitment and retention of participants (adapted from Schulz, Altman and Moher (2010) (308)**)**

The figures included in the above flow diagram for analysis include those participants able to complete the various outcome measures post-intervention. Those excluded from analysis were not able to complete the particular measure at all (i.e. either at pre- or post-intervention assessment).

It should be noted that five of the nine participants classified as a level IV on the GMFCS were able to complete the TUG on pre-intervention assessment. This classification was established through discussions between the physiotherapists at the Institution based on their experienced observation of the children over time. While this may appear counter-intuitive (as level IV children are generally unable to mobilise in standing), the GMFCS guidelines laid out by Palisano et al, for children in the age 6-12 and 12-18 year old categories, explain that to be classified as a level III, children need to walk with an assistive device in "most indoor settings" and are generally able to climb stairs using a rail for support. None of these children were able to complete these tasks but were able to walk short distances indoors, "with physical assistance" (in this case the assistance of a wheeled walking frame) in order to mobilise independently for six metres to complete the measure (100). It was for this reason that a classification of level IV was selected for these participants.

4.4.2 Demographic characteristics

Twenty-two participants were allocated to the experimental and 25 to the control group (Figure 6). The 22 females and 25 males who participated were equally distributed between these two groups (p=0.862) (Table 10).

Table 10: Demographic characteristics compared between the two groups (experimental n=22, control n=25)

		Experimental	Control	Count		
Gender					**Chi-Square**	**p-value**
	Female	10	12	22	0.031	0.862
	%	45.5	48.0			
	Male	12	13	25		
	%	54.5	52.0			
	Totals	22	25	47		
Age					**MWU z- value**	**p-value**
	Median	12.5	14.0		-0.827	0.408
	Range	5-18	6-18			
Language					**Chi-Square**	**p-value**
	Other	18	14	32	3.590	0.058
	%	81.82	56.00			
	English	4	11	15		
	%	18.18	44.00			

The ages of the two groups are depicted by histograms in Figure 7. The ages were not normally distributed (experimental group Shapiro-Wilk W=0.858, p=0.005; control group Shapiro-Wilk W=0.865, p=0.004) and a Mann-Whitney U test determined that there was no significant difference in the ranking of the ages of the two groups (adjusted z= -0.827; p=0.412).

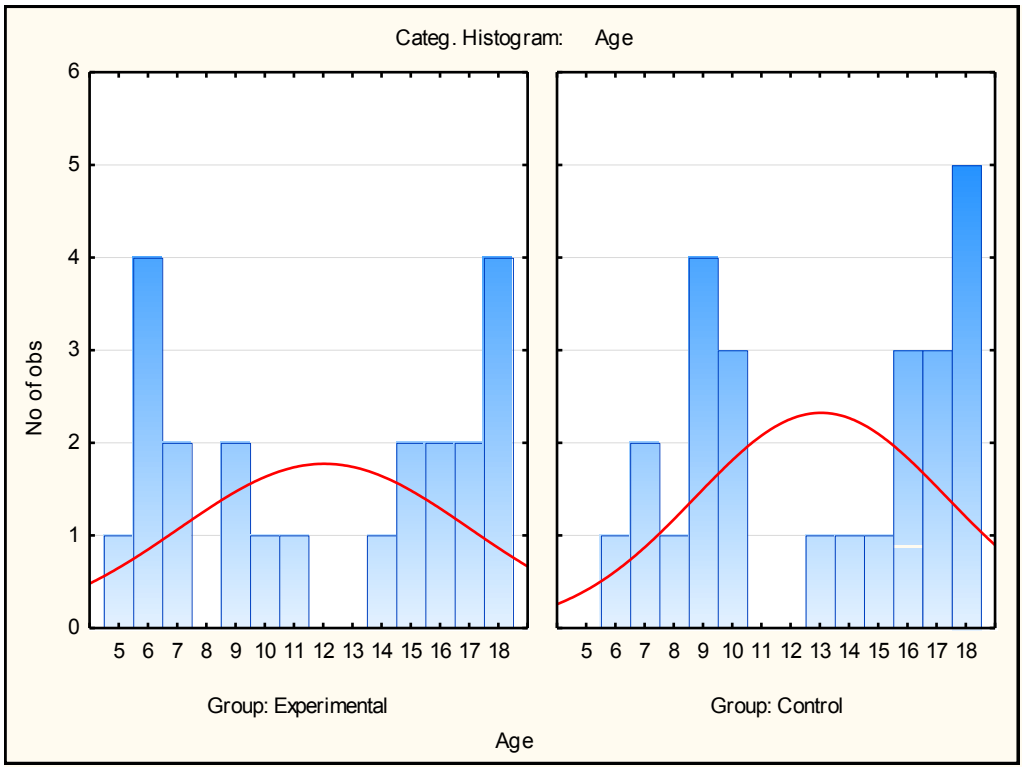


Figure 7: Histogram of the ages of participants in the two groups (Experimental n=22, Control n=25)

There were more home language English speaking participants in the control group and this approached significance (p=0.058) (Table 9).

4.4.3 Medical characteristics

The gestational age of participants was not normally distributed (Shapiro Wilk=0.816; p<0.001) (Figure 8) and showed a bimodal distribution with more participants falling in the 25 to 30 week and 35 to 40 week categories.

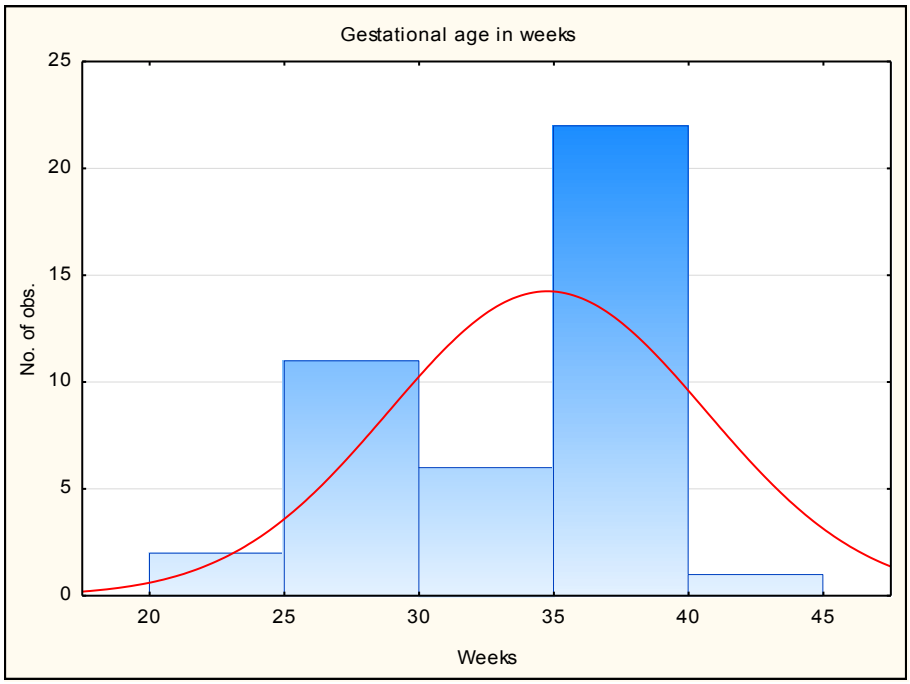


Figure 8: Gestational age in weeks (n=42, 5 missing as information not available/unknown)

The rank ordering of gestational age of the two groups approached being significantly different, with the control group having an earlier gestational age (p=0.071) (Table 11).

Table 11: Medical characteristics and CP classification of participants between the two groups (Experimental n=22, Control n=25)

		Experimental	Control	Count		
Gestational Age					**MWU z- value**	**p- value**
	Median	40	32		1.808	0.071
	Range	26-42	24-40			
Cause					**Chi-Square**	**p- value**
	Prenatal	5	7	12	1.584	0.663
	%	22.7	28.0			

	Perinatal	5	8	13		
	%	22.7	32.0			
	Postnatal	9	6	15		
	%	40.9	24.0			
	Unknown	3	4	7		
	%	13.6	16.0			
	Totals	22	25	47		
Cognitive impairment					**Chi-Square**	**p-value**
	No	9	17	26	3.475	0.062
	%	40.9	68.0			
	Yes	13	8	21		
	%	59.1	32.0			
	Totals	22	25	47		
CP motor disorder (grouped)					**Chi-Square**	**p-value**
	Dyskinetic	3	8	11	2.201	0.138
	%	13.64	32.00			
	Spastic	19	17	36		
	%	86.36	68.00			
	Totals	22	25	47		
CP anatomical distribution					**Chi-Square**	**p-value**
	Diplegia	12	14	26	1.185	0.553

	%	55	56			
	Quadriplegia	4	7	11		
	%	18	28			
	Hemiplegia	6	4	10		
	%	27	16			
	Totals	22	25	47		

The most common causes of CP in participants were hypoxia and maternal bleeding during pregnancy (Table 12). The cause of CP was not associated with group allocation (p=0.663) (Table 11) or secondary visual (p=0.348), speech (p=0,230), behavioural (p=0.747), auditory (p=0.476) or orthopaedic (p=0.138) impairments. There was a trend toward association of cognitive impairment with group, with more participants in the experimental group having a mild cognitive impairment and/or a learning disorder (p=0.062) (Table 11).

Table 12: Specific causes of CP (n=47)

	Count	**Percent**
Hypoxia	9	19.1
Maternal bleeding during pregnancy	6	12.8
HIV encephalopathy	5	10.6
Post-natal infection (other than HIV)	4	8.5
Maternal hypertension	3	6.4
Cerebral haemorrhage	2	4.3
Premature labour	2	4.3
Twin	2	4.3
Delay in receiving medical care	1	2.1

TB meningitis	1	2.1
Congenital brain defect	1	2.1
Prolonged labour	1	2.1
Hydrocephalus	1	2.1
Substance abuse during pregnancy	1	2.1
Postnatal seizures	1	2.1
Unknown aetiology	7	14.9

Most children had associated secondary impairments, with visual impairments and/or visual perceptual problems being the most common, followed by mild cognitive impairments and/or learning disorders (Table 13).

Table 13: Associated secondary impairments (n=47; some had multiple impairments; others had none)

	Count	**Percent**
Visual	29	61.7
Cognitive	21	44.7
Orthopaedic	11	23.4
Speech	10	21.3
Behavioural	5	10.6
Auditory	3	6.4

There were more children who displayed the spastic motor form of CP than any other and the majority of these were diagnosed with a diplegic anatomical distribution (Table 14), however specific motor disorder and anatomical distribution of CP was not associated with group allocation (p=0.138 and p=0.553 respectively) (Table 11).

Table 14: Motor disorder type and anatomical distribution of cerebral palsy (n=47)

CP anatomical distribution	Spasticity	Dystonia	Athetosis	Ataxia	Hypotonia	Totals
Diplegia	24	2	0	0	0	26
%	51.06	4.26	0.00	0.00	0.00	55.32
Quadriplegia	2	0	3	4	2	11
%	4.26	0.00	6.38	8.51	4.26	23.40
Hemiplegia	10	0	0	0	0	10
%	21.28	0.00	0.00	0.00	0.00	21.28
All Groups	36	2	3	4	2	47
%	76.60	4.26	6.38	8.51	4.26	100

Three quarters of the participants were at a GMFCS level of III or lower (Table 15), with the highest number of participants classified as a Level II on the scale. As the levels were used as a basis for the stratification of participant allocation, the different levels were evenly distributed across the two groups (Chi-square =0.456, df=4, p=0.978).

Table 15: GMFCS level (n=47)

	Count	Percent	Cumulative - Count	Cumulative - Percent
I	7	14.9	7	15
II	17	36.2	24	51
III	12	25.5	36	77
IV	9	19.1	45	96
V	2	4.3	47	100

Eighteen participants (38%) had undergone previous surgery and this was not associated with group allocation (p=0.798). Most children receive regular occupational and/or speech

therapy at the Institution, with only 15 (32%) participants receiving neither. There was no association between the receipt of other therapies and group allocation (p=0.604).

In summary, the control and intervention groups were equivalent in every respect apart from there being more English home language speakers in the control group and this group having an earlier gestational age, and the experimental group having slightly more participants with mild cognitive impairments (all of which approached but did not reach significance).

4.4.4 Impact of intervention

The primary outcome measures selected were timed sit-ups and the GMFM-66. The impact of these measures will therefore be presented first, followed by the PRT, the TUG, peak flow readings and the PedsQL. Finally, the results of the participant evaluation form will be presented.

All of the numerical scales were tested for normality. The outcomes that were not normally distributed (Table 16) were then tested using non-parametric statistics. The PedsQL yields ordinal data and was analysed using non-parametric statistics as well. All other outcome measures were tested using parametric statistics (as discussed in section 4.3).

Table 16: Outcome measures which were not normally distributed on analysis

	Shapiro-Wilk	p- value
Sit-ups pre-intervention	0.934	0.010
GMFM % post-intervention	0.937	0.013
TUG pre-intervention	0.685	<0.001

4.4.4.1 Timed sit-ups

The results of the timed sit-ups are depicted in Table 17. There was no significant difference in the rank ordering of the number of sit-ups between the two groups either before or after the intervention (p=0.701). There was a slight decrease in the number of sit-ups in each group post-intervention but this was not significant either for the entire group (p=0.123), the experimental group (p=0.498) or the control group (p=0.119).

Table 17: Pre- and post- intervention scores for timed sit-ups completed in 60 seconds (Experimental n=22, Control n=25)

		Median	Minimum	Maximum
All groups	**Sit-ups pre-intervention**	21	1.0	54
	Sit-ups post-intervention	19	5.0	52
Experimental	**Sit-ups pre-intervention**	20.5	6.00	54.0
	Sit-ups post-intervention	19.0	8.00	52.0
Control	**Sit-ups pre-intervention**	21	1.0	49
	Sit-ups post-intervention	19	5.0	37

	Rank Sum - Experimental	Rank Sum – Control	U-adjusted	z –adjusted	p-value
Sit-ups pre-intervention	509.5	618.5	256.5	-0.384	0.701
Sit-ups post-intervention	527.0	601.0	274.0	-0.011	0.991

4.4.4.2 GMFM-66

As can be seen in Table 18, the median scores of both groups improved over time (as measured by the GMAE). This within group improvement was significantly different in all

participants combined (p=0.002), for the experimental group (p=0.016) and for the control group (0.048) (Table 19).

Table 18: Pre- and post- intervention scores for the GMFM-66 (Experimental n=22, Control n=25)

		Median	Minimum	Maximum
All groups	**GMFM-66 pre-intervention**	65.0	33.9	100
	GMFM-66 post-intervention	65.6	33.9	100
Experimental	**GMFM-66 pre-intervention**	68.0	41.1	100
	GMFM-66 post-intervention	72.5	40.0	100
Control	**GMFM-66 pre-intervention**	61.8	33.9	100
	GMFM-66 post-intervention	64.0	33.9	100

The median and range of the GMFM-66 remained the same for the entire group, but the median of the individual groups improved by about three points. (Note that the median post-intervention of the control group remained lower than the median of the combined groups and this would account for the lack of change in the group median score).

Table 19: Comparison of GMFM-66 scores within groups (Wilcoxon Matched Pairs test) (n=47 adjusted for ties)

	Non-ties	T	z- value	p-value
All groups	40	180.50	3.08	0.002
Experimental	19	35.00	2.41	0.016
Control	21	58.50	1.98	0.048

As can be seen in Table 20, there was no between group difference in GMFM-66 total scores either before or after the intervention.

Table 20: Comparison of GMFM-66 pre- and post-intervention scores between the two groups (Mann-Whitney U test) (Experimental n=22, Control n=25)

	Rank Sum - Experimental	Rank Sum – Control	U-adjusted	z-adjusted	p-value
GMFM-66 % score pre-intervention	551.0	577.0	252.0	0.480	0.631
GMFM-66 % score post-intervention	569.0	559.0	234.0	0.864	0.388

4.4.4.3 PRT

As the results for the PRT were normally distributed, the t-test was used to compare PRT scores. One participant with a GMFCS level I classification was unable to reach while sitting independently to complete the measure. Two hemiplegic participants were unable to reach in the direction of the affected side in sitting. Not all participants were able to stand independently and thus the number of participants completing the standing portion of the PRT was limited, with 20 participants unable to complete the standing test at all and a

further two hemiplegic participants unable to reach in the direction of their affected side in standing. These participants were thus excluded from analysis. No between group differences were detected on any of the items, apart from the pre-intervention sitting reach to the left, in which the experimental group demonstrated a significantly larger reach than the control group (p=0.034) (Table 21).

Table 21: PRT scores for the difference between start and end position (in different test positions) and the comparison of pre- and post- intervention PRT scores between the two groups (n=based on the number of participants able to execute each component of the PRT adequately)

	Valid N - Exp.*	Valid N - Control	Mean - Exp.	Mean - Control	Std. Dev - Exp.	Std. Dev - Control	t-value	df	p- value
PRT pre-int.* sitting forward reach	21	25	11.31	10.73	3.620	4.135	0.499	44	0.620
PRT post-int. sitting forward reach	21	25	12.97	10.74	3.472	4.209	1.934	44	0.060
PRT pre-int. sitting reach right	20	25	6.45	6.43	2.634	3.195	0.020	43	0.984
PRT post-int. sitting reach right	20	25	6.56	5.56	2.220	2.419	1.423	43	0.162
PRT pre-int. sitting reach left	20	25	7.26	5.49	2.787	2.613	2.190	43	0.034
PRT post-int. sitting reach left	20	25	6.72	6.19	2.501	2.621	0.685	43	0.497
PRT pre-int. standing forward reach	13	13	8.38	7.92	3.375	4.109	0.308	24	0.761
PRT post-int. standing forward reach	14	13	9.93	10.67	3.136	3.369	-0.592	25	0.559
PRT pre-int. standing reach right	12	13	6.79	6.91	3.922	2.500	-0.089	23	0.930
PRT post-int.	13	13	7.86	6.95	2.408	2.978	0.855	24	0.401

standing reach right									
PRT pre-int. standing reach left	12	13	7.17	7.81	2.807	3.497	-0.503	23	0.620
PRT post-int. standing reach left	13	13	7.38	7.89	1.842	1.516	-0.779	24	0.444

* int. = intervention, Exp. = Experimental

Note that the t-test for equal variances was used, as the variances were equal in the two groups.

4.4.4.4 TUG

Not all of the participants were able to mobilise independently or with a walking aid and consequently the number of participants, completing the TUG test was limited. The correlation between the pre- and post-intervention scores was high, (Spearman's rho=0.88, $p<0.001$). While seven participants were unable to complete the TUG pre-intervention, during the course of the intervention, three children in the experimental and one in the control group began to walk and their scores were included in the post-intervention analysis (Table 22). One participant gaining the ability to walk was classified as a level III on the GMFCS and the remaining three participants were all a level IV.

Table 22: Pre- and post-intervention scores for the TUG (note that the higher the TUG score in seconds the longer it takes the participant to complete the measure) (n = no. of participants able to complete the measure)

		Valid N	Median	Minimum	Maximum
All groups	**TUG pre-intervention**	40.0	14.0	7.00	196.0
	TUG post-intervention	44.0	14.5	7.00	177.0
Experimental	**TUG pre-intervention**	18.0	13.5	7.00	196.0
	TUG post-intervention	21.0	15.0	8.00	128.0
Control	**TUG pre-intervention**	22.0	14.5	7.00	102.0
	TUG post-intervention	23.0	14.0	7.00	177.0

There was no significant improvement in TUG scores detected within groups (Table 23) or between groups (Table 24) even when the participants who began to walk during the intervention period were excluded from analysis.

Table 23: Comparison of TUG scores within groups (Wilcoxon Matched Pairs test, adjusted for ties)

	Valid N	T	z- value	p-value
All groups	37	284.5	1.011	0.312
Experimental	17	40.50	1.704	0.088
Control	20	96.00	0.336	0.737

(Note that n has decreased as the results have been adjusted for ties)

Table 24: Comparison of pre- and post-intervention scores for the TUG between the two groups (Mann-Whitney U test) (n=based on those participants able to execute the TUG appropriately).

	Valid N - Experimental	Valid N – Control	Rank Sum - Experimental	Rank Sum - Control	z-adjusted	p-value
TUG pre-Intervention	18	22	365.0	455.0	-0.095	0.924
TUG post-intervention	21	23	466.5	523.5	-0.130	0.897

4.4.4.5 Peak flow

As the values for peak flow were normally distributed, the t-test was used to compare peak flow meter scores. No between group differences were found for either pre- or post-intervention scores (Table 25).

Table 25: Comparison of pre- and post- intervention peak flow meter readings between the two groups

	Valid N - Experimental	Valid N - Control	Mean - Experimental	Mean - Control	Std. Dev- Experimental	Std. Dev- Control	F - ratio - Variances	p - Variances	t-value	df	p-value
Peak flow pre-intervention	22	25	219.3	217.4	118.2	118.1	1.001	0.991	0.056	45	0.956
Peak flow post-intervention	22	25	228.9	244.4	116.0	119.0	1.051	0.915	-0.452	45	0.654

Within group differences were found for the whole group (p=0.009) and for the control group (p=0.023), in that expiratory flow was stronger on the post- intervention outcome measure when compared to pre-intervention measure scores. No within group difference was seen for the experimental group (p=0.196) (Table 26).

Table 26: Comparison of peak flow meter readings within groups (dependent t-test)

		N	Mean	Std. Dev.	Diff.	Std. Dev. Dif	t-value	df	p-value
All groups	**Peak flow pre-intervention**		218.3	116.8					
	Peak flow post-intervention	47	237.1	116.6	-18.8	47.07	-2.74	46	0.009
Experimental	**Peak flow pre-intervention**		219.3	118.2					
	Peak flow post-intervention	22	228.9	116.0	-9.55	33.56	-1.33	21	0.196
Control	**Peak flow pre-intervention**		217.4	118.1					
	Peak flow post-intervention	25	244.4	119.0	-27.0	55.79	-2.42	24	0.023

4.4.4.6 PedsQL

As the PedsQL uses ordinal data, non-parametric tests were used for analysis (Table 27).

Table 27: Pre- and post- intervention scores for the PedsQL (note the higher the score on the PedsQL the worse the perceived HRQoL) (Experimental n=22, Control n=25)

		Median	Minimum	Maximum
All groups	**PedsQL pre- intervention**	28.0	4.00	73.0
	PedsQL post- intervention	21.0	1.00	76.0
Intervention	**PedsQL pre- intervention**	26.0	4.00	70.0
	PedsQL post- intervention	16.0	2.00	61.0
Control	**PedsQL pre- intervention**	29.5	7.00	73.0
	PedsQL post- intervention	23.0	1.00	76.0

Significant improvements in HRQoL were detected within groups for the combined group of participants (p=0.001), the experimental group (p=0.018) and the control group (p=0.029) with the experimental group showing a greater change than the control group (Table 28).

Table 28: Comparison of PedsQL scores within groups (Wilcoxon Matched Pairs test, adjusted for ties)

	Valid N	T	z - value	p - value
All groups	40	172.0	3.199	0.001
Experimental	17	26.50	2.367	0.018
Control	23	66.00	2.190	0.029

(Note that n has decreased as the results have been adjusted for ties)

No significant difference was noted between the two groups for either pre- (p=0.242) or post- intervention PedsQL scores (p=0.138) (Table 29).

Table 29: Comparison of pre- and post- intervention scores for the PedsQL, between the two groups (Mann-Whitney U test) (n=based on those participants able to complete the PedsQL independently).

	Valid N – Experimental	Valid N - Control	Rank Sum – Experimental	Rank Sum - Control	z - adjusted	p - value
PedsQL pre-intervention	18	24	340.5	562.5	-1.17	0.242
PedsQL post-intervention	19	24	357.0	589.0	-1.48	0.138

4.4.4.7 Participant evaluation of intervention

The 22 participants in the experimental group were assisted in completing the participant evaluation following the 6-week intervention (as described in section 4.1.8.10). Their answers to three questions regarding use of the FES are represented in Table 30. The vast majority of participants responded “A lot” to liking how the FES felt, how much they wanted to use the FES again and their perception of the amount the FES helped them make functional improvements.

Table 30: Scores for the participant evaluation questions (n=22)

	I liked the feeling of the FES	I would like to use the FES again	I found that the FES helped me
Not at all	3	0	1
A little	3	5	5
A lot	16	17	16

Table 31 shows those intervention group participants who chose to comment's responses to use of the FES. Blank spaces represent those participants who chose not to comment.

Table 31: Participant comments on use of the FES (n=17)

Participant no.	Comment
1	
4	It tickleishing
5	It felt hard
6	
7	
9	
11	It felt fine
12	It was tickling
14	The machine did work for me a little because now can stand for a long time
20	It was shocking me a little bit
21	It felt quite weird. It helped me improve in balance and strength
25	It was nice but didn't like when I had to put on the machine because every time the machine was cold
26	It felt like my whole body was vibrating. I enjoyed it and I think I would like to use it sometimes
27	
28	The feeling nice and good gentle helps
32	It helped my muscle strength a lot
35	I miss the machine a lot because it was helping me about my excising my stomach I miss it's a lots

37	I loved the machine. Can I use it again please I'm begging you. Please!
39	The machine has helped work muscles I didn't know were there
42	I found that this machine help me to be able at home and I lost weight
46	It is relaxing
47	It helped my stomach muscles

4.4.5 Demographic characteristics influencing changes in functional status

4.4.5.1 Interaction between GMFCS, GMFM-66 and age

The plot of GMFM-66 percentage scores across GMFCS levels (Figure 9) indicates that there was a decrease in score commensurate with an increase in level for both the pre-and post-intervention measurements.

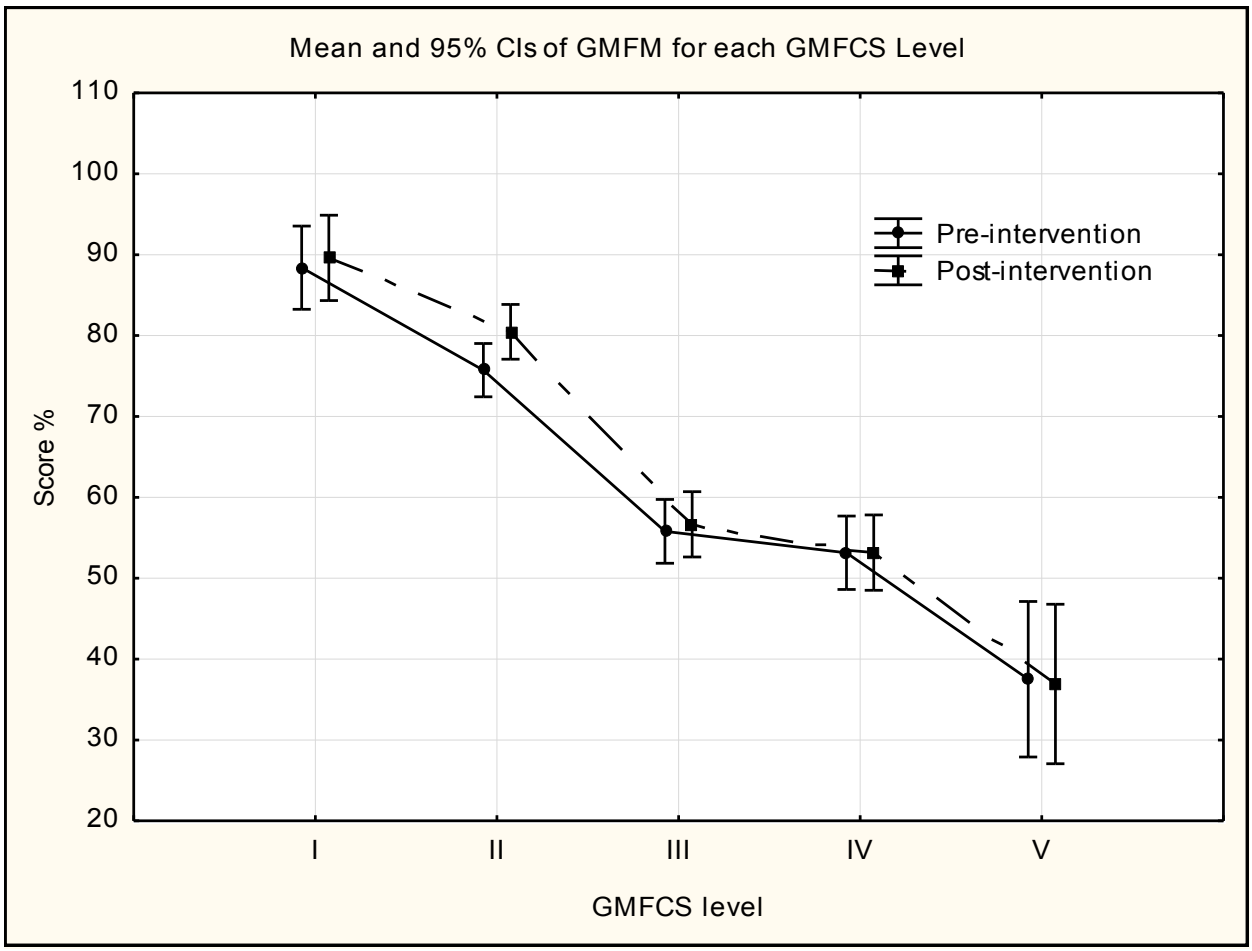


Figure 9: Comparison of GMFM-66% scores across GMFCS level pre- and post-intervention (n=47)

The largest difference in GMFM-66 % scores was seen between the pre-and post-intervention scores for children performing at level II and, to a lesser extent, level I on the GMFCS scale. The change in GMFM-66 scores across the different GMFCS levels were significantly different (ANOVA F (4, 42) =2.879, p=0.034) with participants with a level II functioning, showing the greatest improvement over time (Figure 10).

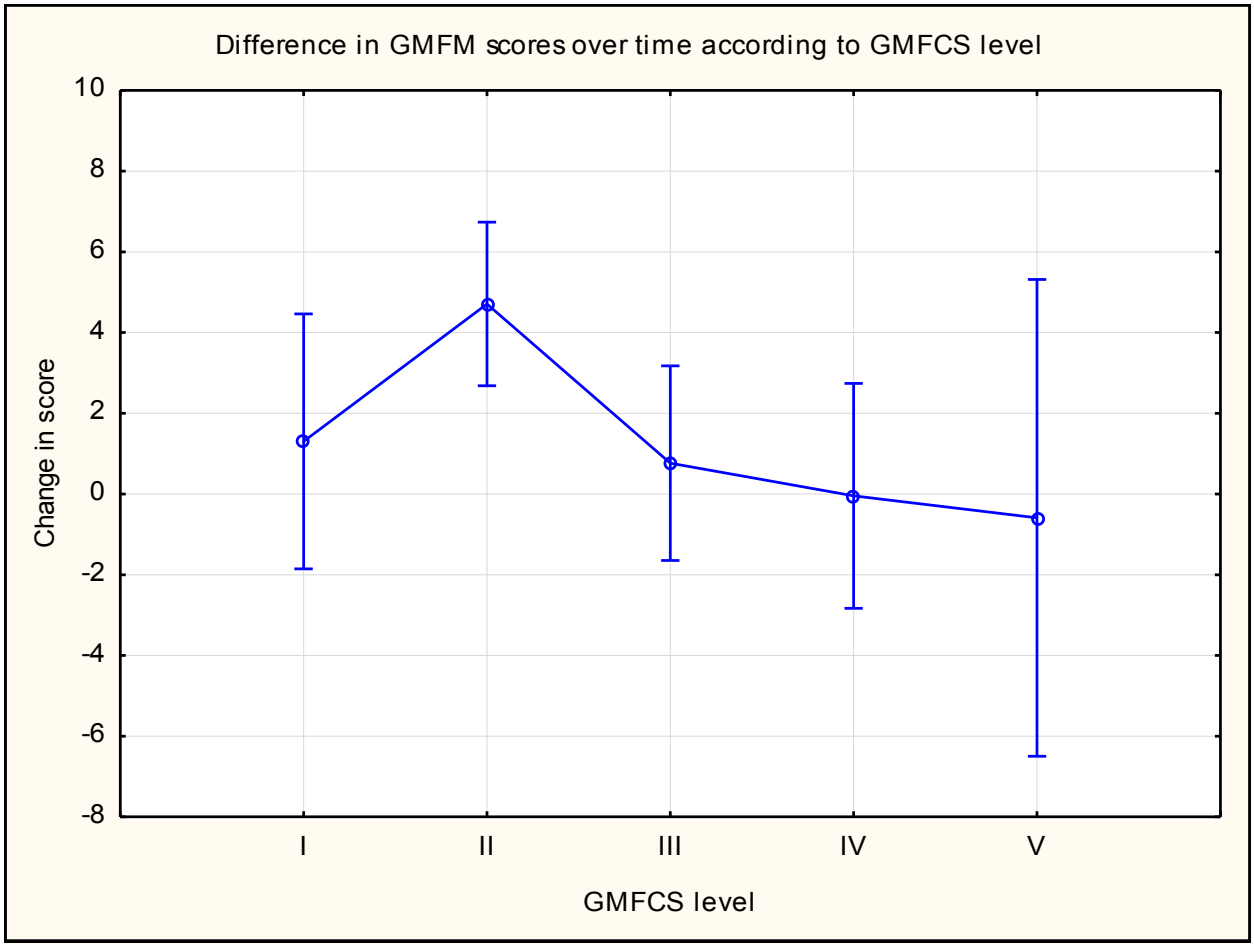


Figure 10: Comparison of change in GMFM-66 scores over time by GMFCS level (n=47)

As can be seen in the scatter plot of age against change in GMFM-66 % score (Figure 11), improvements noted spanned all ages with the greatest number of participants improving between the ages of 14 and 18 years old. There was no significant correlation between age and improvement in GMFM-66 (r=0.27, p=0.067) but a Mann-Whitney U test comparing the rank ordering of children 11 years and below with children 12 years and above indicated that the older children showed a significantly higher ranked improvement (Z= -2.042, p=0.41). See table Table 32.

Table 32: Comparison of rank ordering of children 11 and below and 12 years of age and above (n=47)

Ranks

	VAR00003	N	Mean Rank	Sum of Ranks
VAR00002	1.00	20	18.18	363.50
	3.00	24	26.10	626.50
	Total	44		

Participants classified as level IV and V fell mostly on or below the zero line of the scatter plot (Figure 11) indicating no change or deterioration in GMFM-66 percentage score after the 6-week intervention. Again, it can be seen that the top five improvements in GMFM-66% score were all classified as level II on the GMFCS.

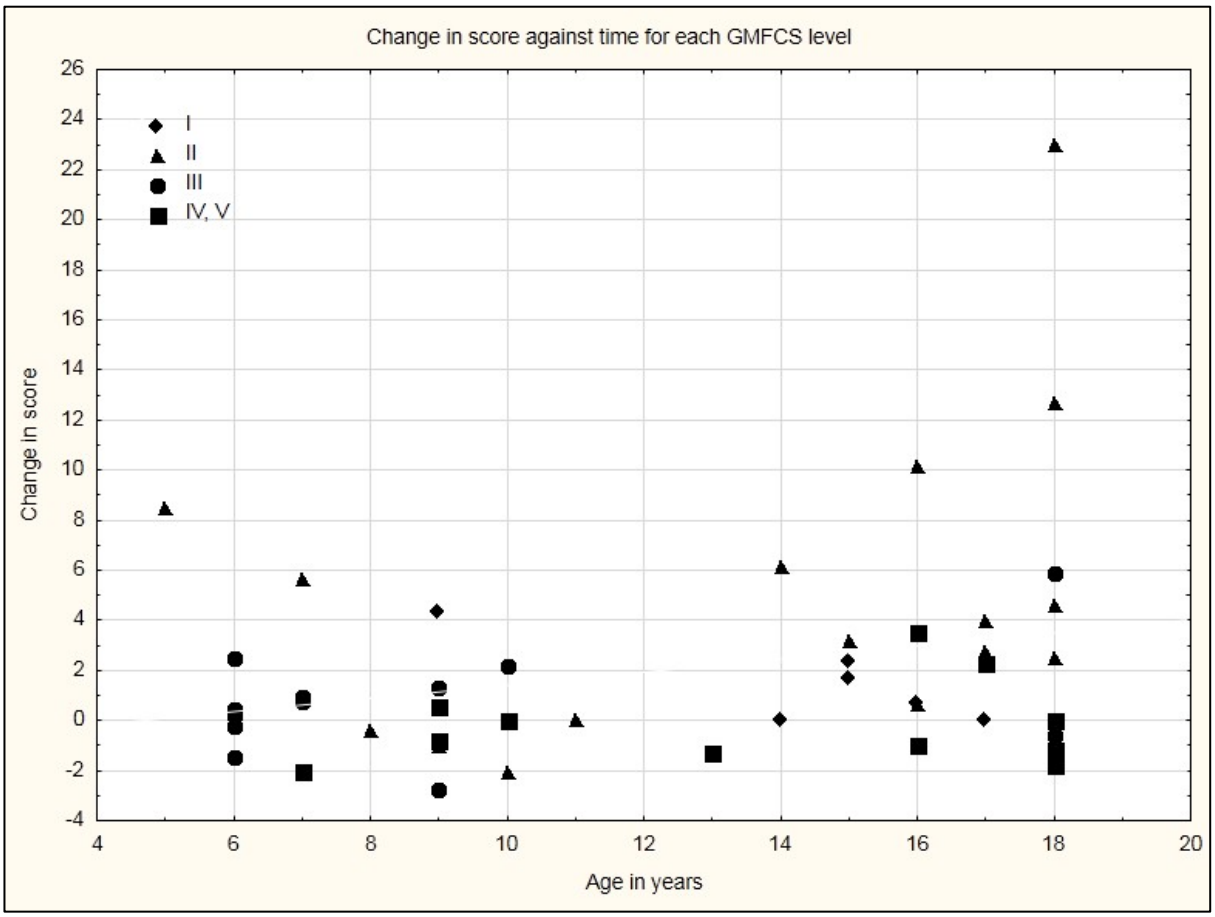


Figure 11: Change in GMFM-66% score over six weeks against age in years (n=47)

No significant difference was detected in the amount of improvement shown by the different motor types of CP ($F(4, 42) = 1.174$, $p=0.336$) or by the anatomical distribution of CP ($F(2, 44) = 0.652$, $p=0.526$).

4.4.5.2 Interaction between TUG and GMFCS

At baseline, Figure 12 shows that GMFCS level I and II TUG scores (in seconds) were quite similar, with level III scores much slower, as these participants used walking aids and/or orthotics in order to complete the measure.

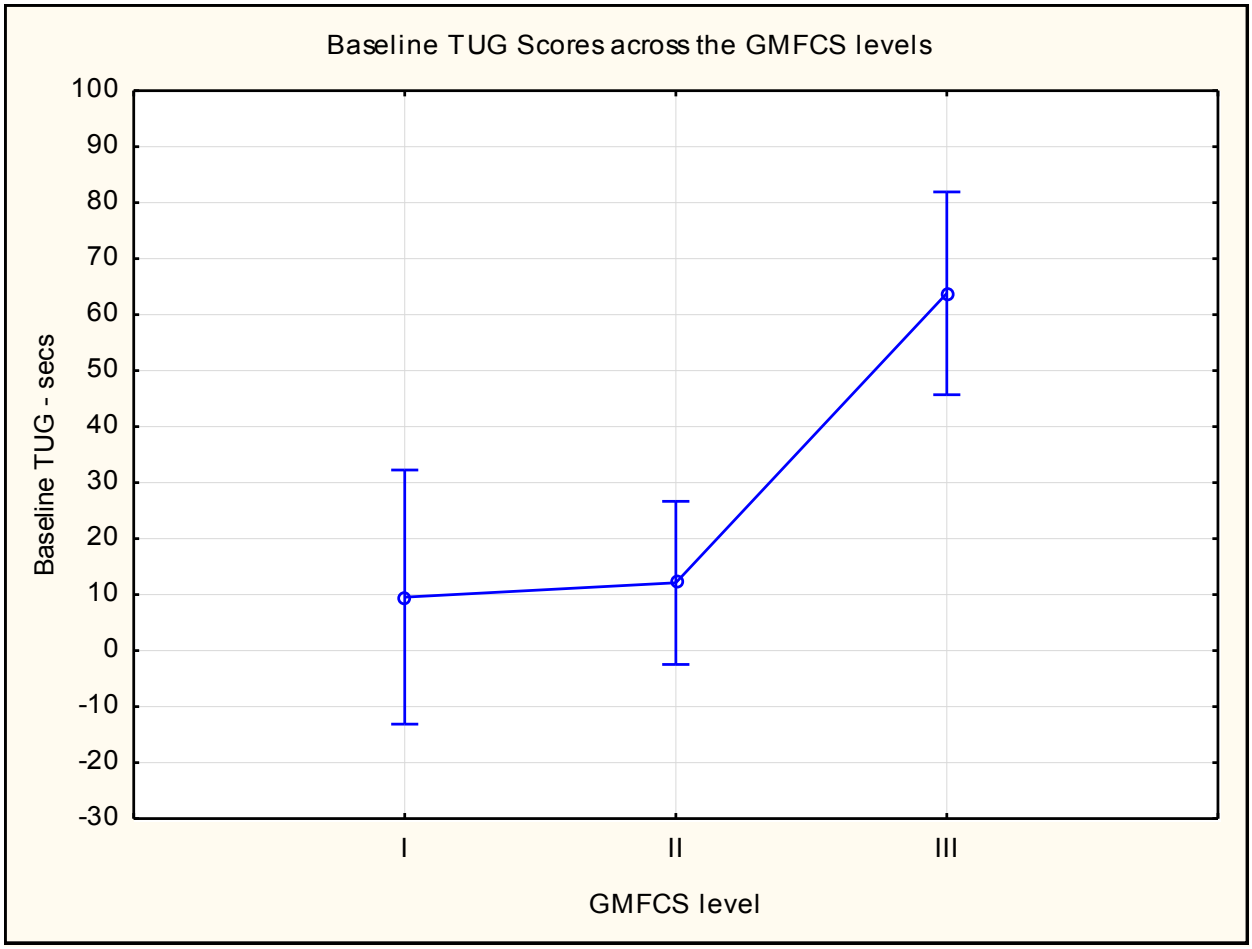


Figure 12: Pre- intervention TUG scores for GMFCS levels I-III (n=36)

An ANOVA indicated that there was no difference in improvement in the TUG between participants classified at GMFCS level I, II or III (Current effect: F (2, 32) = 0.774, p= 0.470). However, participants at level III did show considerable improvement in TUG scores in comparison to level I (who deteriorated) and II (who showed 0 change post- intervention) (Figure 13).

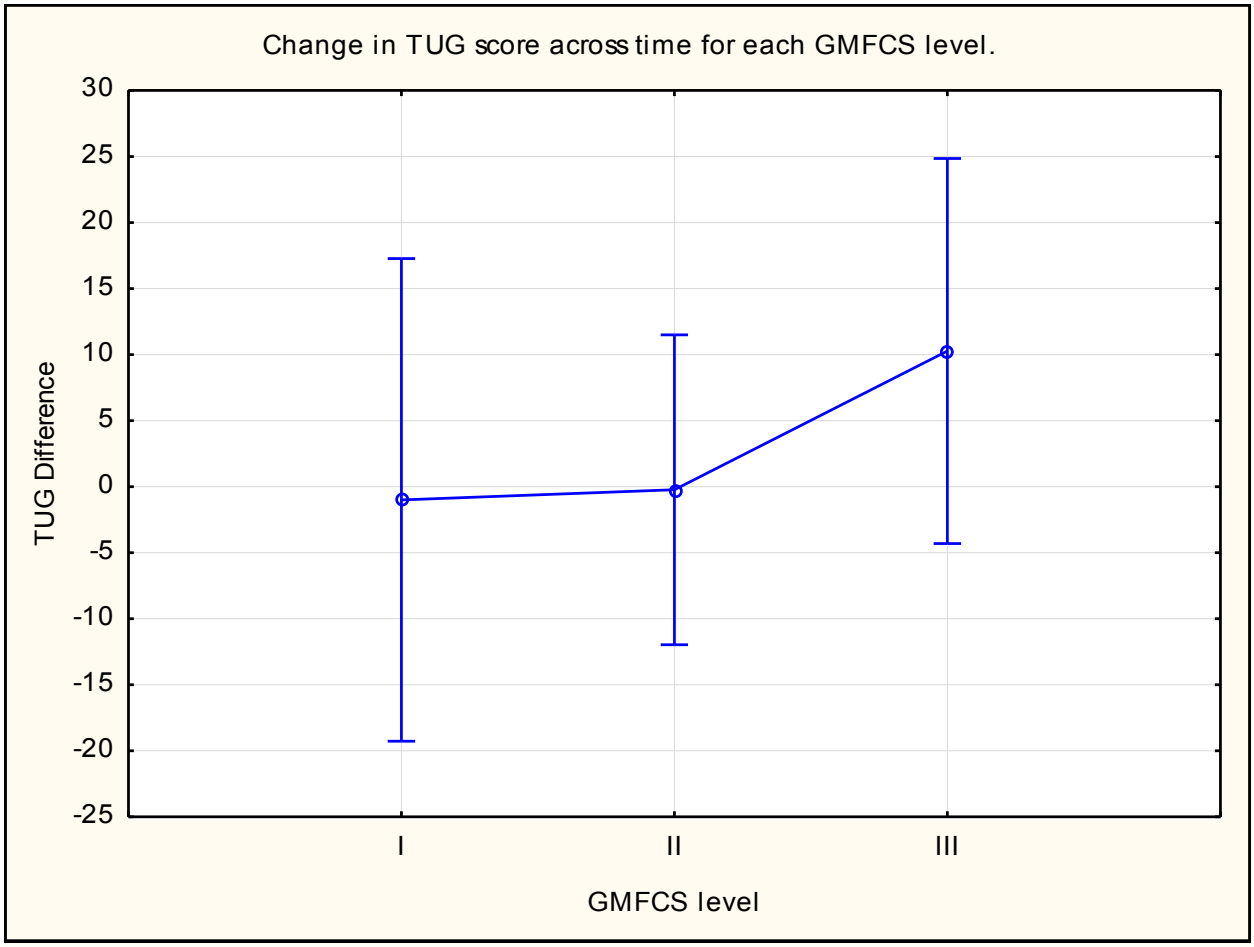


Figure 13: Comparison of change in TUG scores in seconds after 6-week intervention, by GMFCS level (n=36) (difference=pre-intervention scores-post-intervention scores)

Note that the larger the score in seconds the slower the participant is able to mobilise and the greater the change, the greater the improvement in times post-intervention.

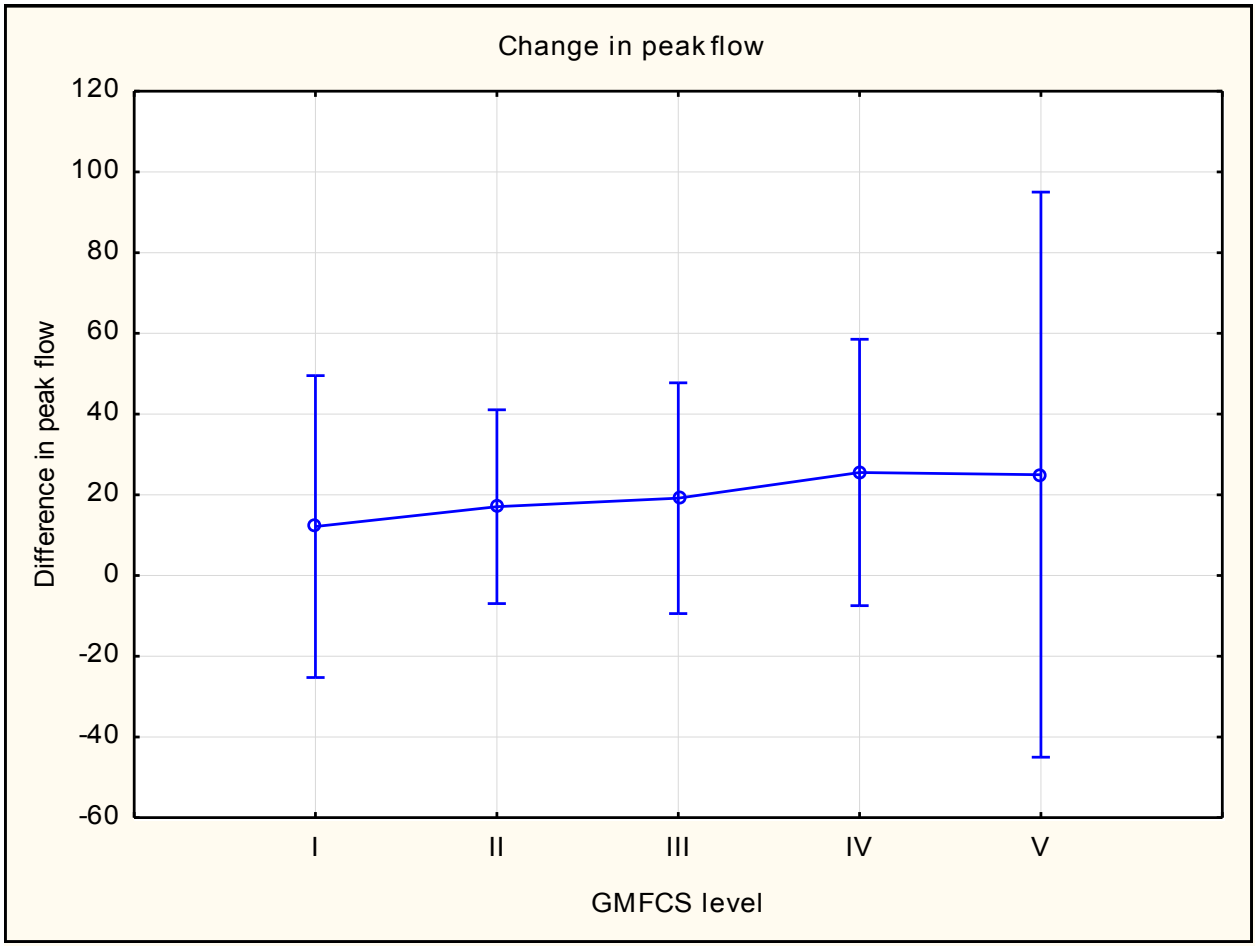
Table 33: Comparison of participant GMFCS level pre- and post- intervention

	After					
Before	**I**	**II**	**III**	**IV**	**V**	**Row (Totals)**
I	7	0	0	0	0	7
II	0	17	0	0	0	17
III	0	0	12	0	0	12
IV	0	0	***3***	***6***	0	***9***
V	0	0	0	0	2	2
Totals	7	17	15	6	2	47

Note: Chi-sq=166.07, df=16, p<0.001

As can be seen in Table 33, three participants progressed from a GMFCS Level IV (prior to the six-week intervention period) to a level III following intervention. This was a significant change (p<0.001). Two of these three participants were older than 17 years of age.

4.4.5.3 Interaction between change in peak flow readings and GMFCS



(Difference=post-intervention scores-pre-intervention scores. Note that the larger the score the greater the peak flow).

Figure 14: Comparison of change in peak flow scores after 6-week intervention, by GMFCS level (n=47)

ANOVA analysis revealed that there was no difference in improvement in peak flow meter readings between participants of different GMFCS levels (Current effect: F (4, 42) =0.088, p=0.986). Participants of all levels did improve on their baseline scores following the 6-week intervention, with level IV and then V making the greatest improvements (Figure 14).

4.4.5.4 Interaction between HRQoL and age and GMFCS level

As can be seen in Figure 15 participant perception of HRQoL, as measured by the PedsQL, varied greatly and age was not significantly associated with a higher or lower self-perceived HRQoL (Spearman-Rho=0.022, t=0.137, p=0.891).

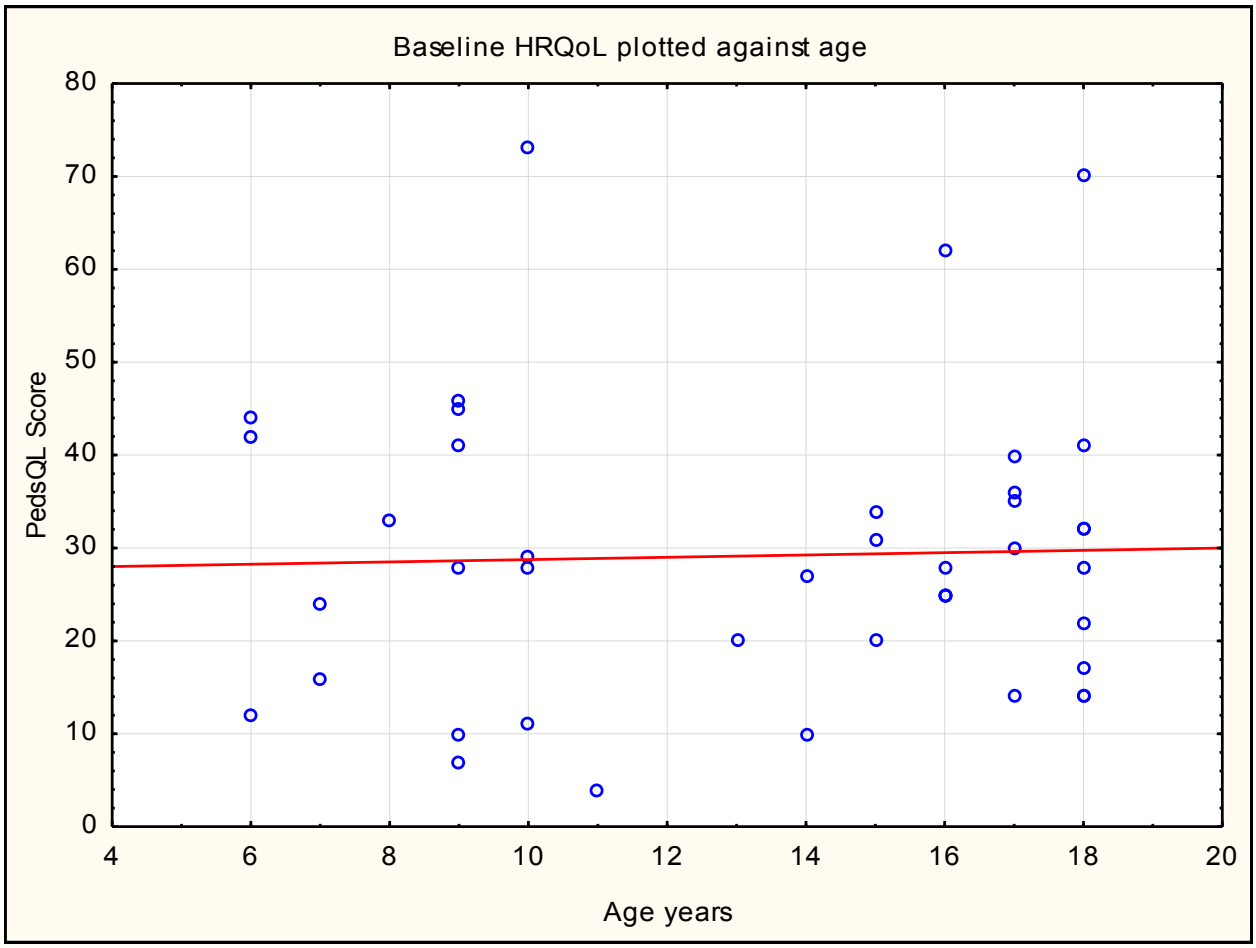


Figure 15: Change in PedsQL score over six weeks against age in years (n=42) (note that the lower the PedsQL score the higher the perceived level of HRQoL)

While ANOVA analysis showed no statistically significant difference in improvement in HRQoL, measured with the PedsQL, between participants of different GMFCS levels (Current effect: F (4, 37)=1.360, p=0.266), Figure 16 shows a general trend towards a decreasing improvement in HRQoL from Level I (who perceived the greatest improvement

in HRQoL over time), downwards one level at a time to level IV and V who perceived a deterioration in HRQoL after the 6-week intervention period.

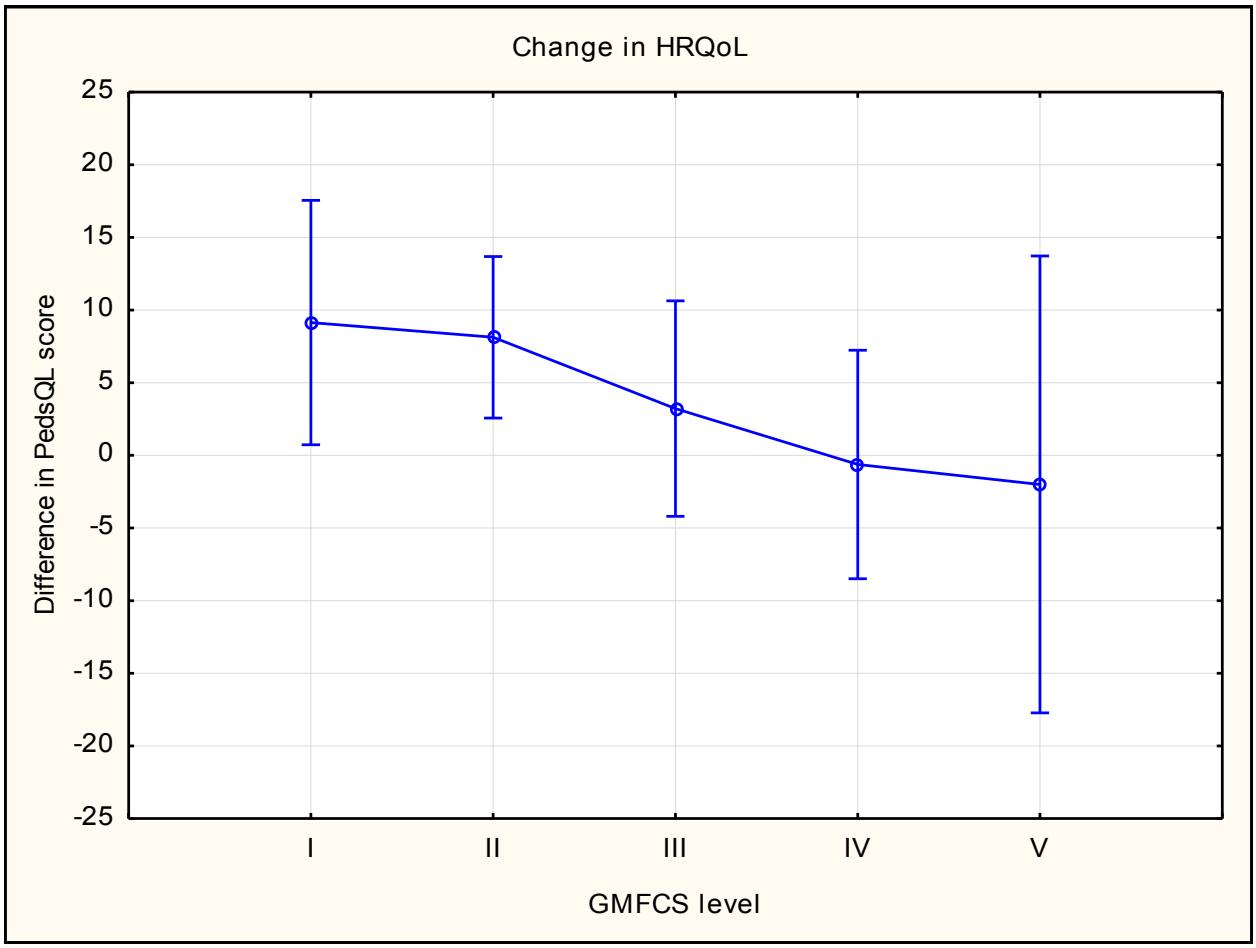


Figure 16: Comparison of change in HRQoL after 6-week intervention, by GMFCS level (n=42) (difference=pre-intervention scores-post-intervention scores. Note that the lower the PedsQL score the greater the self-perceived HRQoL)

4.4.6 Summary of results

Forty-seven children met the study inclusion criteria and were stratified, based on their GMFCS level. Randomisation placed 22 participants in the intervention group and 25 into the control group. There was a zero dropout rate and all participants completed all outcome measures they were physically able to complete. The most common causes of CP

were hypoxia and maternal bleeding. The control and intervention groups were statistically equivalent in all respects (with a trend toward more English home language speakers and an earlier gestational age in the control group and more mild cognitive impairments seen in the intervention group, but none of these factors reached significance).

No between group differences were found for any of the outcome measures used. Statistically significant within group improvements were seen for both groups and the total group for GMFM-66% scores, for the control and total group peak flow meter readings and both groups and the whole group for PedsQL scores after the 6-week intervention. The majority of those children in the experimental group stated they liked the feeling of the FES; they wanted to use it again and thought it helped “a lot”.

Participants classified as GMFCS level II and then I made the greatest improvements in GMFM-66% scores over time and the greatest number of participants showing the largest improvements on this outcome measure were those aged 14-18. GMFM-66% score improvements were not correlated with age, motor type or anatomical distribution of CP but were correlated with GMFCS level. GMFCS level was not associated with TUG, peak flow or PedsQL (but a trend did exist for the greatest improvements to be experienced by the highest functioning participants) and HRQoL was also not associated with age.

Although not statistically significant, there was a trend toward participants of GMFCS level III making the greatest improvements in TUG scores and levels IV and V improving the greatest amount on peak flow meter readings following the 6-week intervention. Three participants gained the ability to walk short distances with an assistive device during the study.

4.5 Discussion

This study aimed to investigate the impact of FES on abdominal muscle strength and gross motor function, in children with CP. Specifically, objectives were set to determine whether an experimental group of 5-18 year old children with CP, receiving FES administered four times a week, in conjunction with typical physiotherapeutic intervention (in comparison with a control group of children with CP receiving only routine physiotherapy) would demonstrate significant differences in abdominal muscle strength, gross motor function, balance, respiratory function and self-perceived HRQoL over a period of six weeks. A further objective looked at whether a relationship exists between the demographic and medical characteristics of participants and any improvements made in functional status.

The most notable findings revealed on statistical analysis of results were that there were no between group differences in the selected outcome measures and that the addition of FES to standard treatment did not result in a greater improvement in function in children with CP over a group of children receiving conventional therapy only. On the other hand, both groups showed significant within group differences in gross motor function and HRQoL, and the control and total groups demonstrated significant improvements in peak flow readings over the course of the six-week intervention period.

The characteristics of the study sample, in comparison to the greater population, are considered below, in order to determine the generalisability of these results. This section is followed by a discussion of the effect of FES, the effect of routine physiotherapy intervention and of the results gained for each individual outcome measure used. This section concludes with an analysis of the study's strengths and weaknesses, as well as the study's limitations. Finally, recommendations for future studies are presented and the dissertation is summarised and concluded.

4.5.1 Sample/participants

Forty-seven of a possible 56 participants met the inclusion criteria laid out by the study and consented to taking part. Pre-study sample size determination (section 4.1.7 Sample size determination) demonstrated that 24 participants were necessary in each group to give the study adequate power. This was reached by the 25 participants randomly allocated to the control group but was slightly underpowered by the 22 participants in the experimental group. Both groups still contained a greater number of participants than most studies investigating the use of ES in CP. There was a zero dropout rate between pre- and post-intervention evaluations. The fact that participants were randomised based on stratification, all participants included in the study completed the study, and all participants were receiving therapy at the Institution prior to intervention so baseline treatment did not change, all limit the potential for the introduction of bias (25)(67)(333).

4.5.1.1 Generalisability of results

As with international trends, male participants slightly outnumbered females (57)(59)(82)(87)(110)(111). The distribution of those study participants with either a primarily spastic or dyskinetic motor disorder fell within the range specified in other studies, with the 8.51% of ataxic children in the sample slightly higher than that quoted in the literature (10)(57)(81)(114)(115)(116). When looking at anatomical distribution, only the number of diplegic children in the study fell within the percentage of total cases found in other studies, with hemiplegics slightly lower than the norm and the 23.4% of study participants classified as quadriplegic higher than the 8-11% cited in international studies (96)(114)(117).

Another divergence from published literature was that while prenatal causes generally account for 70-80% of all CP cases elsewhere (26)(87), the split between pre-, peri- and postnatal causes in this sample was fairly even. While, in high-income countries, birth asphyxia/hypoxia reportedly accounts for 6% of all CP cases (57)(60), 19.15% of medical records in this sample revealed this as the cause of CP. The next most frequent aetiologies noted were maternal bleeding, HIV encephalopathy and postnatal infections, with parents/guardians of participants also citing delays in receiving medical attention,

Tuberculosis Meningitis and prolonged labour as causes of their children developing CP. All of these factors are preventable risks, supporting the speculation made in the earlier aetiology section of the literature review (2.2.4 Diagnosis and aetiology) that prevalence rates are higher in low-income countries due to a lack of ante- and postnatal, quality medical care, poor maternal education (63)(76)(111) and due to the high prevalence of HIV in SA, in particular (123).

As quadriplegic CP has been closely connected to hypoxic events (83)(109)(110), and this was the leading cause of CP found for this sample, it is understandable that a higher proportion of this distribution of CP was noted.

When looking at additional impairments, the percentage of study participants experiencing visual, auditory, cognitive and behavioural problems was similar to those recorded in the literature, with only those with concomitant speech and communication problems in the sample slightly less than that cited (see Appendix III for relevant statistics). This may be because more severely affected children with CP are more likely to experience multiple accompanying impairments (6)(120) and children enrolled at the Institution are expected to be able to take part in a mainstream curriculum. The participants are therefore likely to represent the higher end of the functional spectrum.

In summary, the participants included more children with quadriplegia and less with prenatal causes than international samples. As no association was found between aetiology, type or distribution and outcome, these differences are unlikely to limit international generalisability of the results. However, the sample was drawn from children with less cognitive impairments than their peers with CP who were not able to cope with mainstream schooling. These factors might limit the generalisability of the findings to some degree.

4.5.1.2 Equivalence between the control and intervention groups

After randomisation, there was a trend toward more participants in the control group speaking English as a first language and more individuals experiencing mild, concomitant cognitive impairments in the experimental group, but both of these factors did not reach significance. As the school offers a mainstream curriculum, all of these children would have been able to understand the necessary instructions given during completion of outcome measures and as classes are taught in English, all non-first language English speakers have a good command of the language. It can be concluded that neither of these factors would have had an influence on results. The control group also had a slightly earlier gestational age; again, not reaching significance. The most premature child in the control group was born at 24 weeks and in the experimental 26 weeks (with most children included in the study being born before full term). This corresponds with an increased incidence of CP with decreasing gestational age shown worldwide (60)(91)(119) and evidence that due to improving medical technology more very preterm children now survive (6).

Following stratification and randomisation, the distribution of gender, age, aetiology, other impairments, motor and anatomical distribution of CP, other therapies received and previous surgery undergone was similar between the control and experimental groups.

The control and intervention groups were also statistically comparable at baseline, ensuring bias was not introduced before intervention took place and no statistical difference existed between the frequency of physiotherapy intervention received by each group during the six-week intervention. The results are therefore likely to be as a result of the intervention and not due to other confounding variables.

With all of the above taken into consideration, one can conclude that the sample included in the study is representative of other children with CP capable of following a mainstream curriculum in Johannesburg, and perhaps a much wider population.

4.5.2 Effect of the FES

The study supported the null-hypothesis that the addition of FES to routine physiotherapy treatment would not lead to a significant difference in outcome parameters compared to routine treatment. Statistical evidence is therefore lacking to support the beneficial effect of FES in the treatment of CP, under the specific study parameters. It was anticipated that electrical stimulation during treatment might lead to improvements in abdominal muscle strength and that by incorporating stimulation while performing the task-specific, functional activities included in routine treatment would provide a feedback loop in that, as trunk stability improved, posture, limb control and balance would also improve. Better control of these body functions would then provide feedback to, in turn, increase abdominal activation during functional activity (235). Steinbeck et al found that electrical stimulation in children with CP resulted in the greatest changes in GMFM scores for those children unable to independently mobilise and he hypothesised that the weaker the muscle the greater the opportunity for change (264). This was not the case during this RCT, and the common finding that improvements at impairment level do not necessarily translate into functional improvements was demonstrated by this study (68)(76)(119)(152)(158)(171)(205)(236)(238).

FES, rather than NMES or more passive TES, was selected for use in this study as, with the wide acceptance of the ICF framework of disability, a move away from impairment-targeted intervention has created a focus on treatment techniques that might create functional improvements, thus allowing for improved participation (127). It was anticipated that by utilising ES currents during the functional activities provided during routine treatment at the Institution, that both impairment and activity limitation/participation would be simultaneously addressed. As seen by a lack of between-group differences, this was not the case for either.

Studies have hypothesised that strength gains with FES are due to the overload principal (28)(66)(225), whereby muscle size improves, as has been observed in a 2013 study of walking children with CP (30). With repetition of use, the learning of accurate muscle activation and timing has been proposed (228). All studies investigating the use of FES, that have shown both statistically significant improvements at impairment and activity

limitation level, have employed ES over lower limb muscles during gait, thus achieving improvements in various gait parameters. Stimulation, in these instances, was timed for specific activation of the relevant muscles during swing-through (36)(227). In this study, stimulation was consistent for 20 minutes during all activities. Due to the heterogeneity of CP, goal-directed activities were obviously different depending on the individual participant's needs, and activities used to achieve these specific goals also would have varied throughout the six-week intervention, to ensure patient interest and participation in activities. Had stimulation of the abdominals been timed for specific activation during the same task, repeated over six-weeks as in these studies, greater improvements may have been seen.

In studies where significant improvements in strength or gross motor function have taken place with the use of ES, any positive treatment effects encountered were not maintained after cessation of the intervention; either after a given period without the intervention or immediately after removing the device (217)(244). Unfortunately, this study did not include outcome measures taken while the FES device was in use, so one can only speculate that results may have been greater had this been done.

The treatment modality used for intervention in this study has limitations of its own. Research has shown that the contractions initiated by NMES carry a high metabolic demand (243). In addition, where self-initiated muscle contractions recruit motor units in differing orders, FES recruits the same motor units, which may actually lead to muscle fatigue (334). Another problem is that it is difficult to accurately position electrodes when using surface electrical stimulation in order to achieve the desired muscle contraction, as the "non-selective" electrical current administered by these devices may lead to unpredictable motor responses, with unwanted, surrounding muscles also being stimulated (71).

The external obliques were selected for use in this study as they lie anatomically more superficially than most of the abdominal muscles and, as well as assisting with spinal stability (144) and providing trunk rotation (176), they also prevent an anterior pelvic tilt, prevalent in the CP population (160)(181). The transversus abdominis muscles play a more central role in core stability as they are activated before all other abdominal muscles to

precede limb activity (10) and are the only abdominal muscles to act tonically during gait (174). For this reason the TA might have been more appropriate for stimulation in this instance, to achieve the desired outcomes but, they are the deepest of the abdominal muscles (172)(174) and surface ES is unable to stimulate deep muscles (71). Invasive, intermuscular ES capable of stimulating these muscles is not appropriate for use in a paediatric population.

The higher the intensity of surface stimulation, the greater the stimulation of nociceptors, thus limiting the possible strength of muscle contraction achievable, using FES, during this study (243). While visible muscle contractions were elicited during the intervention, the intensity of said contractions may not have been sufficient to bring about significant strength gains after such a short period. This rationale was presented by Kerr et al in their RCT of 63 children with CP, investigating NMES over the quadriceps muscles, when positive strength gains were not obtained (242).

Only one other study was found in the literature investigating the use of ES over the core muscles of children with CP, in comparison to a control group receiving routine, NDT-based therapy. Participants in this study received an unspecified type of ES over unspecified "abdomen" and "posterior back muscles" simultaneously for 30 minutes, six days a week for the duration of their six-week intervention. This lack of specificity limits the interpretation of the results, as does the authors' implication that ES significantly improved the postural control and abdominal strength of intervention group participants over control group participants without including an outcome measure specifically designed to measure balance or strength. With this said, positive findings of improved radiological spinal angles and GMFM sitting dimension scores do indicate improvement in sitting posture and control (38). While analysis of the individual dimensions of the GMFM was not performed in the current study, a lack of between group differences in balance and abdominal strength encountered (as measured by the PRT and timed sit-ups) may be accounted for by the stimulation of abdominal muscles only, four as opposed to six treatments a week and stimulation at 35 rather than 20 Hz in this study. Park et al included infants as their sample and perhaps it can be surmised that ES intervention is more effective in younger participants (38).

An international trend for publishing statistically positive results exists (335) which could lead to an overemphasis of the positive effect of certain modalities. While negative results, such as those determined by this study, are discouraging, they provide valuable information for professionals basing their therapeutic intervention on empirical evidence (170). As the methodological design of this study was strong, results can be interpreted with a level of certainty and thus the use of FES under the study's circumstances cannot be supported.

It is unlikely that the results of this study are not a true reflection of the impact of ES under the specified circumstances. There were no significant differences noted between the groups at baseline and no trend was evident that might indicate that a Type II error occurred due to a small sample size. The CONSORT criteria (see below) were adhered to as far as possible.

In addition, the length of intervention was likely to have yielded significant results, if ES had been effective. A 2003 study investigating conductive education and intensive therapy programmes concluded the intervention period of five weeks was too short for developmental maturation of participants to be the explanation for improvements seen and it was concluded that these changes were due to the treatment itself (296). Similarly, this study introduced an intervention executed over a period of six weeks and thus improvements in the outcome measurements within groups can be accounted for by the baseline therapy offered by the therapists at the Institution and not by maturation.

Taylor, Dodd and Graham have maintained that notable changes in the strength of children with CP should be possible following a six week intervention of any kind, if changes are to be experienced (155). Another study by Ozer, Chesher and Scheker went so far as to say that such changes may even be detected in as little time as four weeks after NMES intervention (263). Following a four-week trunk targeting intervention, Unger also noted significant strengthening of abdominal muscles (160). Therefore, had the FES administered during this study made clinically significant differences in muscle strength, they would have been duly noted following the six-week intervention period.

With this said, while employing a sample size greater than that of most studies investigating paediatric intervention outcomes and the control group meeting the pre-determined sample size parameters (and the experimental group size nearing this calculation), a lack of power cannot be completely ruled out as this sample size was determined based on a study investigating higher functioning children (264). The standard deviation was likely much higher in this study as all five levels of the GMFCS were included in the sample and it is therefore possible that more positive results may have been found had the sample included only certain GMFCS levels.

4.5.3 Effect of routine/standard physiotherapy intervention

While the lack of significant between group differences on all outcome measures employed does not support the use of FES in this context, within group improvements prove that the baseline intervention being offered by therapists at the Institution, and in Johannesburg, is improving the gross motor function, respiratory function and HRQoL of the children with CP treated. Due to the wide variety of treatment techniques used by these therapists and the specific combination of techniques used for each child differing, one cannot say which of these techniques individually or in combination, or at which frequencies of use accounted for the improvements seen. Studies assessing intervention strategies used in the treatment of CP are always complicated by the lack of uniformity of use between therapists (75).

In order for treatment outcomes to have clinical and functional carry over into real life situations, goals set should be meaningful to the individual being treated (65) and paediatric intervention should be age-appropriate and child friendly (212). Task-orientated activities have shown a larger degree of postural adaptation and range of functional movement in children with CP, when compared to more meaningless exercises/activities (42)(205). All four physiotherapists at the Institution stated that they employ a motor learning/goal-directed therapy approach in their treatment of children with CP. Children are included in goal setting and guide the use of interventions used to attain these goals. For this reason, and due to their inclusion of facilitation of movement in daily practice allowing for the sensory experience of movement, it can be assumed that motor learning took place over the six-week intervention, thus accounting for statistically significant within

group improvements seen (64)(86)(164)(201). This approach is also followed by the majority of other special-school based therapists in Johannesburg.

The other modality most often used in the treatment of CP by all Johannesburg physiotherapists was that of strengthening. The lack of notable improvements in abdominal strength following the intervention period may be accounted for by the lack of resistance and training to fatigue employed during strengthening programmes by these medical professionals (as explained in greater detail in section 3.5.2).

As this was a pragmatic trial, physiotherapy intervention continued once or twice a week, as is usually the case at the Institution. Significant improvements in gross motor function encountered with this frequency of intervention further supports studies demonstrating that a higher intensity of therapy offered does not enable greater gains in functional outcomes (209)(248)(249).

4.5.4 Individual outcome measure findings and limitations

4.5.4.1 Timed Sit-Ups

While isokinetic and isometric dynamometers show excellent interrater reliability on evaluation (13)(288), they were too expensive for application during a study based at a government funded school. For this reason, a more practical and easy to administer measure of abdominal strength was employed in the form of timed sit-ups, as this test has shown good reliability (155)(289).

There was no difference in the rank ordering of the number of sit-ups possible, by either group, pre- or post-intervention. Surprisingly, following the six-week intervention, both the control group and the experimental group using FES were able to do slightly fewer sit-ups than prior to the intervention period. This deterioration was not significant.

In the study on the effect of improved abdominal muscle strength on respiratory function in adults without neurological fallout, Simpson also found lower than expected improvements in abdominal strength, measured by timed curl-ups, and concluded that abdominal muscle torque and FVC and FEV_1 were not correlated (191).

In contrast, a study investigating the effect of NMES for the abdominal muscles in TD adults saw a 100% improvement in abdominal isometric strength and endurance in the experimental group following intervention (256). ES over the abdominals and posterior back muscles of infants with CP during a RCT demonstrated significant improvements in the sitting dimension of the GMFM and radiographic spinal kyphotic and Cobb angles, leading the authors to conclude that improvements in posture were due to improved abdominal strength and control due to the ES intervention (38). (The differences between this and the current study have already been described in section 4.5.2 Effect of the FES).

As has been described in the sub-study on intervention techniques administered in Johannesburg special needs schools in an earlier section (3.5.2), while all four physiotherapists employ strengthening programmes in their daily intervention programmes, only three use any form of external resistance during limb strengthening and one, during core strengthening. None of these therapists works the child to fatigue during strength training for both the limbs and the core musculature. As was discussed in section 3.5.2, studies have shown that in order for strength gains to take place, loads of 60-100% of the individual's maximal voluntary contraction (31)(256) need to occur and resistance needs to be progressively increased as strength gains are made (152)(238)(239). Exercising to fatigue has also been cited as a method of ensuring strength gains are made (235). It has been claimed that repetition of movement, using one's own body weight for resistance, is not enough to cause changes in muscle strength (151). It is perhaps for this reason that no improvements in abdominal muscle strength were observed, despite the participating therapists stating that they use strengthening techniques in daily practice. The possible reason behind FES not improving abdominal strength in this sample has already been discussed (see 4.5.2).

In a study described earlier in this section, timed curl-ups were used as an outcome measure for determining abdominal strength gains following a three-week trunk targeting

intervention. Their lack of significant results was explained by testing having been performed at different times on different days, participants not performing to their maximum potential on evaluation and difficulty controlling a standardised technique being used by each participant (191).

While timed curl-ups have demonstrated good reliability as a measure of abdominal strength in other studies (289), the results established for abdominal strength in this study should be interpreted with caution. While there was an attempt at controlling variability by ensuring each individual used the same starting position pre- and post-intervention, when being evaluated during timed sit-ups, standardisation was not rigorously enforced. Other studies have ensured the exact degree of knee flexion each participant should begin lying in with a goniometer, with the feet elevated from the floor and no assistance offered in terms of stability provided by the assessor at the ankles. Each sit-up was measured once the participant was able to touch a predetermined point ahead of them during the curl-up (212)(289). While some study participants showed excellent strength in that they were able to curl completely up to a sitting position without assistance, this also meant that they were able to complete fewer of these type of sit-ups than those weaker participants performing only a small 'crunch', where only the shoulders left the ground, as full sit-ups take longer to execute.

Knudson makes the point that even with strictly standardised timed curl-up testing, a minimum of 10 achievable sit-ups is necessary in order to detect change and a standard error of measurement of 5-9 repetitions exists for this evaluative tool (154). In this sample, six children were not able to perform a maximum of 10 sit-ups either pre- or post-intervention and only nine out of the sample of 47 showed a change (whether negative or positive) in the number of sit-ups they were able to complete in one minute, of more than nine.

As has been described in the literature review (section 2.3.1.1 Musculoskeletal impairments), the muscles stimulated during this study were the EOs, whose primary mechanical function is rotation of the trunk. The "crunches" used as an outcome measure essentially measure trunk flexion (the role of the RA) and thus without having assessed sit-ups with a rotatory component little inference can be made into the improvement of

strength of the EOs of study participants. With this, and the above said, very little conclusive evidence can be drawn from the results of this particular outcome measure.

4.5.4.2 GMFM-66

While no between group differences were noted following the intervention, significant within group improvements were noted for the control, experimental and combined groups. These results may have been even greater had three participants not been experiencing pain during post-intervention testing (of a severity large enough to inhibit certain gross motor tasks), following varying musculoskeletal injuries.

As was discussed at length in the literature review (see section 2.5.3.1 Measuring gross motor function for details), the GMFM is an outcome measure specifically designed for the evaluation of change in the motor function of those with CP. It has demonstrated high test-retest and interrater reliability, concurrent and construct validity and excellent responsiveness to both positive and negative changes in gross motor function over time. As was also discussed in section 4.1.7 Sample size determination, MCID was determined in this study in order to assess whether clinically meaningful and beneficial changes could be made with use of the intervention. Due to the heterogeneous nature of CP, little consensus exists over the exact degree of change that will lead to such observable MCID changes in CP.

If compared to Bar Haim et al's 2010 RCT on motor learning, where it was concluded that a 1.5% increase in GMFM-66 scores results in clinically meaningful gross motor changes, then the median improvement of 2.2 for the control group and 4.5 for the experimental group following intervention can be described as clinically significant, as well as statistically significant improvements (69). On the other hand, Eek et al recounted that in a previous study by Wang and Yang it was reported that an improvement of 3.71 on the GMFM-66 would lead to "clinically visible" changes (158). In this case, only the experimental group would allow for observable improvements in gross motor activity. During the original GMFM-66 responsiveness study, Russell et al determined that the greatest changes were

seen in children under the age of five and that those older than five showed improvements of 2.06 overall in GMFCS levels I and II, and 1.15 and 1.6 for level III and levels IV and V combined, respectively, 12 months after their initial evaluation (274).

The greatest improvements in GMFM-66 percentage scores were achieved by those participants classified level II on the GMFCS (followed by those in level I). This may be because children classified as a level I reached a ceiling effect on the measure (see below for more information). In line with the motor development growth curves, Sorsdahl et al also noted the greatest improvements in gross motor function in children of levels I and II after an intensive, goal-directed intervention period (268).

Those children from level IV and V made no improvements or showed slight deteriorations in gross motor function over the six-week period. This may be because the GMFM may not be sensitive enough to detect changes in lower functioning children with CP (300) or because intervention for this level was aimed at maintaining current levels of function and preventing secondary impairments from occurring, rather than strengthening and focusing on functional activity attainment (230). With this said, certain individuals classified as a GMFCS level IV did make great improvements in gross motor function, as indicated by a progression to a level III as seen by their attainment of the ability to walk short distances with an aid, during the intervention period (see section 4.4.4.4 TUG for details). The lack of improvement seen on the GMFM may have been a measurement error and perhaps been a non-significant deterioration.

While the GMFM is considered the gold standard for the measurement of gross motor function in children with CP (298), a possible ceiling effect for higher functioning children with CP exists (226)(273)(336). One participant from each group had a pre-intervention maximum score of 100 and thus post intervention testing showed no change in gross motor function, although this may not actually be true if higher functioning tasks were to be taken into consideration. Both of these participants were teenagers, classified as a level I on the GMFCS, with the child in the control group hemiplegic and in the experimental group, diplegic. Two other hemiplegic children scoring in the high 80s did not make any further change in scores following the intervention period. These results are corroborated by Shumway-Cook et al, who found that two of their hemiplegic participants also encountered

a ceiling effect on testing using the GMFM-66 (21). During their large population-based study in Sweden, Beckung et al determined that 75% of children with CP classified at GMFCS level I are able to accomplish 90% of the GMFM-66 items by five years old, with a large proportion reaching 100% eventually (96).

It has been proposed that the GMFM may not be sensitive enough to detect changes in lower functioning children with CP (300). This was seen in this study, where the two children with no change between pre- and post-intervention testing (and a low score on the measure) were classified as a level IV and V on the GMFCS. Children who had scores that deteriorated post intervention were one young level III, three level IV participants and the only other level V child taking part. Had analysis of the various individual dimensions of the GMFM-66 been performed, perhaps significant improvements may have been seen in the appropriate categories, as both participants classified as a level V and one as a level IV scored between 0-6 points only for dimensions C, D and E on the GMFM which may have overshadowed any improvements made in the first two dimensions.

Testing, using the GMFM-66, is performed barefoot (274)(324) as was executed during assessments in this study. All four physiotherapists at the Institution stated that they use orthotics and assistive devices during gait training, as did 94.44% of therapists at the other special schools (the majority of whom recorded use on a daily basis). Orthotics and assistive devices are purported to improve postural support (86)(163)(232)(337) and gait parameters, compared to barefoot walking (99)(324), and aids can compensate for balance fallouts by providing a greater base of support and correcting body alignment when mobilising (170)(177)(324). One might assume that results for gross motor function seen may have been greater had these children been tested wearing their accustomed orthoses and employing the help of assistive devices when needed, allowing them to complete more of the functional tasks required, independently, in a more pragmatic sense (105). With this said, children did use these aids and devices during TUG testing, where no statistically significant improvements were noted for either control or experimental groups.

4.5.4.3 PRT and TUG

Again, no within or between group improvements were of statistical significance for these measures, following the intervention period.

Prior to the intervention, 40 of the 47 participants were able to mobilise independently, with or without an assistive device, in order to complete the TUG test to receive a recorded outcome. Following the six-week study period another four children gained this ability. While no statistically significant within group or between group improvements in TUG scores were noted following the intervention, for either control or experimental groups, these 'new walkers' further support the effectiveness of current therapeutic input provided at the Institution. Most interestingly, three of these four 'new walkers' were originally classified as a level IV on the GMFCS. One child was aged seven and this new gross motor ability may be due to natural maturation. The other two participants were aged seventeen and eighteen. This is a clinically significant finding given that, much of current literature supports Rosenbaum et al's predictive motor development growth curves for children with CP (101) which demonstrate more severely affected children have the least potential for improvement in function (223) and that 90% of children with CP reach a plateau in progress by the age of seven. These curves, however, are based on the evaluation of children having received early intervention in a first-world country (105). This concept will be discussed in greater detail in section 4.5.5 Age-related degree of improvement in gross motor function.

While children classified as a level I on the GMFCS actually showed slower times than prior to the intervention, levels II and to a lesser degree III showed considerable improvements in their TUG times (despite these results not reaching significance). This may be due to the fact that level I children are already proficient at walking and may not put in as much effort on testing of such tasks. Greater improvements in levels II and III may be due to the fact that therapeutic goals for these levels would aim at gait training (three of the four therapists facilitate gait with the use of orthoses and assistive devices at the Institution on a daily basis) in order to achieve a higher level of independent functioning.

While numerous studies support the use of the PRT for balance testing in disabled children, it is limited in that it only tests anticipatory postural control and not balance in

more dynamic situations (174). As well as being direction-specific, postural adjustments are also task-specific (178). As reaching was performed in stationary standing or sitting as the measure indicates, no functional goal is expected of reaching and this may have negatively affected the motivation of children (especially younger children) to reach with maximal effort (284).

While the TUG aims at measuring the improvement in walking speed over a three-metre distance, children appeared more concerned with safety than with increasing their speed. This was especially the case for those children completing the measure using an assistive device. Another reason a positive change in TUG scores may not have been achieved is that children with CP generally demonstrate compromised cardiovascular fitness and general endurance (22)(70)(124)(132)(196). While aerobic-specific training was not included on the intervention checklist completed by the physiotherapists included in the sub-study earlier, no participants added this intervention when additional treatment modalities used were requested of them. It can therefore be implied that this was not a treatment goal included during the intervention period at the Institution and without the cardiovascular fitness or endurance necessary, walking speed would not have improved.

4.5.4.4 Peak Flow

While no between group changes were seen for peak flow meter readings, significant improvements in FEV were seen for the control and total sample of participants post intervention. On pre-intervention testing, one child from each group was unable to gain enough lip closure to allow for a readable measure on expiration. Both of these children were classified as having dyskinetic CP (with choreoathetosis); both were under the age of seven and received speech therapy. The child randomised to the control group achieved a reading following the intervention period and this may have accounted for a lack of statistical improvement seen in the experimental group, as this child still received a zero on post-intervention testing, as the child was still unable to coordinate and maintain lip closure sufficient to gain a reading on the measure.

No particular GMFCS level made a statistically different improvement to another group but it was noted that those making the greatest gains in peak flow readings were in levels IV and V. This may be because they have the largest room for improvement or because a focus on respiratory muscle training may be a greater focus in this subgroup of children in order to prevent the secondary respiratory complications discussed in section 2.3.1.1.

A study mentioned previously cited that four different brands of mini flow meter readings were poorly correlated with those taken by a spirometer and the authors concluded that these measures may overestimate expiratory lung function (294). This may account for the within group improvements seen in respiratory function in this study.

4.5.4.5 PedsQL

During pre-intervention assessment, four children in the intervention and one in the control group were unable to complete the PedsQL measure, due to an inability to communicate their answers adequately. All of these children were under the age of eight. Other than bringing the sample number below the sample size determination of 24 participants for the intervention group this should not have greatly affected the positive improvements in PedsQL scores seen following the intervention as younger children are more likely to report satisfaction with their HRQoL than their older counterparts (271).

Eiser, Mohay and Morse have asserted that essential to the concept of HRQoL is that it is a "unique" subjective perception and thus the individual's opinion of their own HRQoL should be attained during testing (307). It was decided that a parent-proxy version of the PedsQL would not be used for those children unable to complete the measure themselves, as studies have shown that significant differences exist between child and parent reports of HRQoL, where parents tend to rate their child's HRQoL as worse than they themselves perceive it (56)(299)(303)(304).

No between group difference was seen for this outcome measure either but statistically significant within group improvements were noted for both groups and for the total group. Slightly greater improvements in self-perceived HRQoL recorded for the experimental group may be due to the Hawthorne effect, whereby the novelty of the treatment modality and participant perception that they are receiving 'extra' attention, may have accounted for these results (338).

Improvements in HRQoL, as seen for the total sample, may be due to the timing of the study. Before the intervention period, all children had been on their end of year, five-week vacation. Most children enrolled at the Institution come from very poor socioeconomic, home backgrounds and often live in child-run households. This fact is in keeping with statistics showing that in 2011, only 89.2% of Black African South Africans had access to running water inside their dwelling, and 81.5 and 89% had access to electricity and flushable toilets respectively (339). The rate of children orphaned by a single or both parents has more than doubled since 1996 (340) and many of the children at the Institution live in child-run households. Most children attending the school receive bursaries that pay for their school fees and allow them to reside in the school hostel during the week, where they receive regular meals. When at school these children also take part in sporting and cultural activities, are able to interact with their friends and receive medical attention from on-site nursing staff; all factors that may positively influence HRQoL (53)(125)(135)(207).

While age and GMFCS level were not found to be associated with HRQoL, a general trend did exist for level I participants to record the greatest improvements in PedsQL scores post intervention, followed by level II and so on until level V who reported the smallest gains in HRQoL. The more severe the presentation of CP, the higher the likelihood of developing multiple secondary complications/impairments (55), and the greater the number of impairments, the greater the participation restriction for the individual (10). It makes sense then, that those more severely affected children in the study showed the smallest gains in HRQoL.

The PedsQL is validated for use in children from 5-18 and has different versions for each particular age group to represent the improving cognitive level of children with time. However, authors have expressed the concern that as all items are equally weighted, the

PedsQL does not account for the changing views of adolescents as to what activities are more important than others (307). Only self-report was used in this study as self-report scores of HRQoL have been shown to be statistically higher than parent-proxy reports (299)(304). This did exclude those few participants who were unable to communicate and this may have introduced bias towards higher functioning children.

4.5.4.6 Participant evaluation

Three of the 22 participants in the experimental group did not like the feeling of the FES at all and only one said they felt it did not help. In contrast, 72.73% of the sample stated they liked the feeling of and thought the FES helped "a lot" and 77.27% said they would like to continue using the FES during therapy "a lot". When asked to comment, the only complaints were of the coldness of the electrodes when placed on their bare skin and one child felt a slight "shocking sensation". Some reported that the machine "tickled" and most mentioned various ways in which they felt the FES had improved their strength and balance. Again, the Hawthorne effect discussed in sections above may account for this sense of improvement with exposure to a novel intervention.

While the participation evaluation provides interesting insight into the perceptions of those participants in the experimental group of the study, this outcome tool is not standardised or validated and results speak only of each child's subjective opinion.

4.5.5 Age-related degree of improvement in gross motor function

As was seen in the results of the study on intervention techniques used by therapists at special needs schools in Johannesburg (see 3.4.3), the Institution is the only one of these nine schools treating children in their teens. The remaining eight schools focus their therapeutic intervention on the younger, foundation phases. Bottos et al explained that

this global tendency toward preferential treatment of younger people with CP occurs as practitioners feel greater outcomes are achieved with early intervention (212). This general therapist opinion accords with Rosenbaum et al's widely accepted gross motor development growth curves (96). As was discussed in detail in section 2.4.2, according to these standardised curves, up to 90% of children with CP reach their motor potential by five years of age, plateau in progress by age seven (96)(105) and reach a ceiling to their potential skill attainment by early adolescence (103)(105)(106). However, Rosenbaum et al based their predictions on the performance of children in high-income countries, where it is likely that they had received optimal physiotherapy treatment throughout their lives.

The 657 children with CP included in their predictive study were recruited from nine different rehabilitation centres, situated in Ontario, Canada (115). These centres provide intervention by experienced paediatric therapists, medical personal, surgeons and orthotists to those children with CP in the province where such treatment is indicated (96)(115). As these centres are government-funded, they are widely accessible and thus service the majority of children with CP in the geographical area (105)(115). These intensive services are made available to children with CP until early adulthood (115).

In contrast, as seen by the results mentioned above, the majority of special needs schools in Johannesburg only offer therapy services to children with CP prior to their teenage years, at which point all intervention ceases. The same has been observed by the researcher in government-funded hospitals in the area, where therapy is provided in groups, on a monthly basis, to young children with CP only. This trend can be understood, based on the wide acceptance and application of the gross motor development growth curves for prognosis and intervention planning, internationally (96)(100).

When improvements in GMFM-66 outcomes were plotted against age (section 4.4.5.1), the children making the greatest improvements in gross motor function were between the ages of 14 and 18 and a MWU test revealed children older than 12 experienced a significantly higher ranked improvement in GMFM-66 scores than those children aged 5-11.

The Institution is one of the only special needs schools in Johannesburg offering a matric/grade 12 curriculum. For this reason, every year the school accepts block transfers of physically disabled children from schools that only offer classes until grade seven or nine. These teenagers have generally not received physiotherapy for the four to five years prior to admission and immediately start with physiotherapy on acceptance into the Institution. It may be for this reason that this subpopulation of children made the greatest gains in gross motor function as, after years of being inactive, they suddenly receive intensive, goal-directed therapy.

As has been seen in neuroimaging studies of patients following a stroke, neuroplasticity is still possible in adulthood (83). Rerouting of neurons to undamaged cortical areas is possible with increased exposure to new and varying sensory experiences (86)(164)(179)(201). It has been said that physiotherapy can aid in motor learning by providing the opportunity for an individual, otherwise unable to experience movement independently, to be guided through an activity physically and thus, "learn" the motor components necessary in order to execute this particular task (169)(203)(204). As has already been established, the majority of therapists based at schools in Johannesburg use a motor learning, goal-directed or an NDT-based approach to intervention and facilitation of movement (161)(164)(217) was recorded as being used most often, on a daily basis. While not essentially a motor-learning approach to intervention, the facilitatory techniques incorporated into an NDT approach provide a child with the opportunity to experience movement not otherwise possible. This assists with motor learning in that the sensory experience of an activity is processed by the brain, later allowing the activation of muscles necessary for independent execution of these tasks. This may account for the positive changes seen in this older population of children with CP, following the six-week intervention.

The only three children in the sample not able to walk with assistance at the end of the study were two children classified as a GMFCS level V and one boy with spastic quadriplegic CP classed a level IV. Based on the definitions of these levels, citing limitations in self-mobility in a wheelchair or the need for complete dependence on a caregiver for transportation and mobility, one would expect these individuals not to be able to complete the measure (105). While one child in the control group now able to complete the TUG as a

level III is not surprising (105) and another seven year old level IV in the intervention group's walking ability perhaps accounted for by natural maturation , the final two participants gaining this ability are more surprising. Both of these children, in the intervention group, were classified as a level IV on the GMFCS, with diplegic CP. The unexpected demographic characteristic of these two participants though were their ages; 17 and 18. This occurrence may further justify the need for continued therapeutic input beyond the teen years of children with CP (as discussed above).

Improvements seen in older participants may also be the case as older children may have a greater understanding of what is expected of them and can be motivated to work harder during therapy when they understand how intervention may influence their daily functioning and participation. Whatever the reason, these results provide a basis for further study as, should these results be corroborated, continued treatment of children with CP, in middle to low income countries, into their teens, should be considered, especially if early intervention was not possible or treatment was ceased early.

These results bring into question the global acceptance of Rosenbaum et al's predictive gross motor development growth curves for children with CP, as they were developed based on a population of children receiving intensive intervention from their early years until adulthood (96)(100). The children included in this sample may have ceased therapy prior to reaching their full motor potential and, by resuming therapeutic intervention at this late stage in life, began making gainful improvements in function once again. With that said, motor curves plotted for such children may show a peak in functional activity later in life, with a plateau in performance seen at a much older age. Along this line of thinking, predictions made based on the GMFCS may also only be appropriate for children with CP who have received intervention throughout their lives. Studies of this valid and reliable measure of ability (45)(92)(93)(99) have shown a high level of acuity in predicting future functioning of children with CP, with a classification using the measure at 12 years of age generally being retained through adulthood (74)(77)(190). As was seen by a change in GMFCS level after the six-week intervention (during testing with the TUG) in this sample, this was not the case for children with CP from a low-income area, having had little intervention in previous years.

4.6 Strengths and weaknesses of the study

While most systematic reviews have cited poor methodological designs as a reason to exercise caution when interpreting study results for interventions for the treatment of CP (33)(48)(230)(259), this study has meticulously followed the 2010 CONSORT statement for RCTs (40). The CONSORT statement was drafted to assist with the transparency and quality with which reporting on RCTs are executed and to enable accurate reproducibility of studies (40)(308). Below, this study has been evaluated based on its strengths and weaknesses, as laid out by this CONSORT statement. Simpler items, such as the study title, have just been noted as the page number on which they were reported, while items that are more complex have been justified in the comments section of the table below. As the sub-study on intervention techniques used by Johannesburg therapists was not a RCT it is not included in the evaluation below.

Table 34: CONSORT 2010 checklist of information to include when reporting a randomised trial (adapted from Schulz, Altman and Moher (2010) (308)**)**

Section/topic	Item no.	Checklist item	Page no. reported on	Comment
Title and abstract				
	1a	Identification as a randomised trial in the title	i	
	1b	Structured summary of trial design, methods, results, and conclusions	v-x	Detailed overview of the relevant study sections provided
Introduction				
Background and objectives	2a	Scientific background and explanation of rationale	1-4, 5-6 and 8-75	Thorough literature review presented following in-depth reading of all available literature relevant to the study from 1980-2013
	2b	Specific objectives or hypotheses	4, 76 and 99-100	Specific aims and objectives, as well as null hypotheses presented
Methods				
Trial design	3a	Description of trial design (such as parallel, factorial) including allocation ratio	77 and 100-101	

	3b	Important changes to methods after trial commencement (such as eligibility criteria), with reasons	-	Not applicable. The methodology planned was followed
Participants	4a	Eligibility criteria for participants	101-102	See 4.7.1 below for comment
	4b	Settings and locations where the data were collected	7, 77-78 and 102	The entire population of special needs schools servicing children with CP in Johannesburg was assessed. The Institution at which the intervention study took place was comparable to all other schools in the area, apart from offering a mainstream curriculum only
Interventions	5	The interventions for each group with sufficient details to allow replication, including how and when they were actually administered	105-107 and 118	Specific dates for both assessment and intervention were provided. Step-by step procedures were explained and intervention parameters for both the control and experimental groups were described in detail allowing for reproducibility.
Outcomes	6a	Completely defined pre-specified primary and secondary outcome measures, including how and when they were assessed	108-114	The psychometric properties and application of the standardised primary and secondary outcome measures employed for assessment of participants during the intervention study are described
	6b	Any changes to trial outcomes after the trial commenced, with reasons	-	Not applicable
Sample size	7a	How sample size was determined	103-104	Sample size determination was listed and the pre-determined group size met for the control group and slightly underpowered for the intervention group
	7b	When applicable, explanation of any interim analyses and stopping	-	Not applicable

		guidelines		
Randomisation				
Sequence generation	8a	Method used to generate the random allocation sequence	117	
	8b	Type of randomisation; details of any restriction (such as blocking and block size)	117	
Allocation concealment mechanism	9	Mechanism used to implement the random allocation sequence (such as sequentially numbered containers), describing any steps taken to conceal the sequence until interventions were assigned	117	
Implementation	10	Who generated the random allocation sequence, who enrolled participants, and who assigned participants to Interventions	117	The researcher and treating physiotherapists were not involved in random selection of names from envelopes after stratification and so preferential placement of participants into groups was avoided.
Blinding	11a	If done, who was blinded after assignment to interventions (for example, participants, care providers, those assessing outcomes) and how	117-118	Bias was controlled by ensuring the research assistant executing pre- and post-intervention outcome measurements was blinded to the group allocation of participants and the therapists providing the intervention were not involved in any of these evaluations (341). Having four physiotherapists perform the intervention rather than only the researcher alone reduced the possible biasing influence one treating therapist may have had on the results (39). As no children in the sample receive any private therapy this external confounding

				factor was controlled as all participants continued with the intervention they were accustomed to throughout the trial. It was not possible to blind the treating therapists or the participants to group allocation, as it is not possible to provide placebo FES treatment.
	11b	If relevant, description of the similarity of interventions	115-117 and 118	Standard/routine physiotherapy received by both the control and intervention groups was outlined in detail in the first sub-study, preceding the intervention study, and was comparable to the intervention provided by other physiotherapists working at similar institutions in the area.
Statistical methods	12a	Statistical methods used to compare groups for primary and secondary outcomes	120-123	The purpose of each statistical test used for analysis of data is described. Despite its primary use in analysis of normally distributed data, the possible use of the one-way ANOVA for non-normally distributed data has been justified.
	12b	Methods for additional analyses, such as subgroup analyses and adjusted analyses	120-123	
Results				
Participant flow (a diagram is strongly recommended)	13a	For each group, the numbers of participants who were randomly assigned, received intended treatment, and were analysed for the primary outcome	125	The suggested CONSORT flow diagram (308) was used for this purpose
	13b	For each group, losses and exclusions after randomisation, together with reasons	125	As above

Recruitment	14a	Dates defining the periods of recruitment and follow-up	116-118	
	14b	Why the trial ended or was stopped	118	The intervention study ended at the end of the planned six-week intervention period as planned.
Baseline data	15	A table showing baseline demographic and clinical characteristics for each group	124-134	A detailed breakdown of socio-demographic and medical characteristics between groups is presented for participants of the RCT.
Numbers analysed	16	For each group, number of participants (denominator) included in each analysis and whether the analysis was by original assigned groups	124-157	n is listed in each table heading of results presented and explained when less than the total sample number
Outcomes and estimation	17a	For each primary and secondary outcome, results for each group, and the estimated effect size and its precision (such as 95% confidence interval)	135-157	Results listed are for each of the various outcome determinants employed during the study were given. As there appears to be little consensus on which statistical test to use to calculate effect sizes when data are not normally distributed, and how to interpret the different values calculated using the different methods, effect sizes were not included in this analysis.
	17b	For binary outcomes, presentation of both absolute and relative effect sizes is recommended	-	N/A
Ancillary analyses	18	Results of any other analyses performed, including subgroup analyses and adjusted analyses, distinguishing pre-specified from exploratory	135-157	
Harms	19	All important harms or unintended effects in each group	114-115	Potential harms were listed but possible side effects of the intervention were few

				and no participants experienced any adverse effects during or after the intervention period.
Discussion				
Limitations	20	Trial limitations, addressing sources of potential bias, imprecision, and, if relevant, multiplicity of analyses	181-188	Limitations of the FES intervention itself, all outcome determinants used in both studies, the sample and statistical analysis are offered
Generalisability	21	Generalisability (external validity, applicability) of the trial findings	159-161	See 4.7.1 below for comment
Interpretation	22	Interpretation consistent with results, balancing benefits and harms, and considering other relevant evidence	158-180	A thorough analysis of results and discussion surrounding pertinent findings is presented and related to scientific literature
Other information				
Registration	23	Registration number and name of trial registry	79 and 114	
Protocol	24	Where the full trial protocol can be accessed, if available	-	This item was not included in the study write-up but access to the proposal, accepted by UCT, can be obtained from the university or the primary researcher
Funding	25	Sources of funding and other support (such as supply of drugs), role of funders	iii	

4.7 Limitations

4.7.1 Study design and sample

In a sample where heterogeneity is a consideration, standard deviations may be large (226). In this sample, due to the inclusion of all five GMFCS levels, this was certainly the

case, with participant abilities ranging from those children able to obtain a perfect score on certain outcome measures and others unable to complete certain instruments at all. Although the RCT design used in this study is regarded as the gold standard in terms of proving causality, Sanson-Fisher et al suggest that the greatest limitation of RCTs is that they lack external validity and thus the ability to make generalisations about the greater population due to stringent exclusion criteria causing a sample group to be selected that does not represent the greater public (342). This may result in clinicians employing successful treatment modalities less in practice (313). For this reason, a large, representative sample was selected for inclusion in this study. While the total sample size had significant power to allow interpretation of the results of the study with confidence, when performing subgroup analysis this was not the case and these results should be accepted with caution. The main findings of the study may also have lacked sufficient power due to the large standard deviation present due to the inclusion of all GMFCS functional levels in the sample (needed to have a sample of sufficient size).

One coincidental limitation of the sample was that while participants were generally quite evenly distributed between the various age groups, only two participants in the study were aged 11-13. This can be rationalised by the fact that learners either join the Institution at nursery school level or as a transfer from another school not offering a mainstream matric curriculum in grade seven or nine (when approximately 14 or 16 years old). This has led to the school having very small intermediate phase classes (i.e. far less children enrolled at the school aged 11-13 than any other age). This may limit the results in that children of this age have generally been receiving physiotherapeutic input consistently since a young age and thus may have performed better than their teenage counterparts may.

As was described in section 4.5.1.1 of the discussion above, the results can be generalised to other 5-18 year old children with CP attending government special needs schools in the country and perhaps in other developing countries. These results cannot be generalised to those children not enrolled in schools nor can they be generalised to those attending private schools or schools with modified academic streams (112). The lack of ongoing physiotherapy since birth in many of the children who recently transferred to the Institution may limit the generalisability of the results to children in middle to low income countries where intervention is not mandatory.

4.7.2 Statistical analysis

While blinding, the use of a control group receiving equal baseline therapy to the experimental group and strict inclusion criteria all increase the internal validity of a study, these factors may also lead to low treatment effects when a greater effect exists in actuality (313).

Those outcome measures not finding within or between group differences following intervention during this study were also those outcome instruments not possible to complete by all participants. While sample size determination prior to study commencement recommended 24 participants per group (see 4.1.5), TUG viable pre- and post-intervention comparisons left only 22 participants in the control group and 18 in the experimental for statistical analysis. Likewise, the PRT saw as few as 20 paired outcomes in sitting and 12 for certain standing components of the test. These smaller useable sample sizes may have introduced a type II error, whereby a significant change may have taken place but the study was not powerful enough to demonstrate this (227)(310)(317). (In contrast, this was also the case for the PedsQL, where the necessary 24 participants were included in the control group analysis but only 18 children were able to complete the measure in the intervention group). In the same vein, GMFCS subgroup analysis should be interpreted with caution due to small numbers available for analysis; especially for level V as only two participants classified at this level met the study inclusion criteria. The limitations of the outcome measures used have already been presented during the discussion of each outcome determinant in section 4.5.4.

4.8 Conclusion and Recommendations

As pragmatic trials compare an intervention to standard/routine therapy, a full investigation was carried out to determine whether the standard of care offered at the Institution was comparable to that offered by other physiotherapists in Johannesburg. This was confirmed and a trend toward an NDT systems approach to treatment and a focus on

motor learning and strengthening was established. No therapists in Johannesburg used FES for intervention in CP prior to this study.

Despite the majority of participants enjoying the use of the FES and believing that it helped, following a six-week intervention period of concurrent FES during routine physiotherapy, no between-group differences in outcome determinants were found between experimental and control groups. Due to the rigorous study design employed, it can be concluded that FES in the treatment of CP is not effective, as used in this study. It was also confirmed that impairment-targeted intervention does not necessarily equate to functional gains.

On the other hand, statistically significant within group improvements in gross motor function and HRQoL for both groups and, improvements in lung function in the control group and the combined sample, following the intervention period, were noted. This confirms that, despite the NDT-based, motor-learning approach used during routine intervention having little scientific evidence to support it, regular implementation of these modalities, and the other techniques employed at special needs schools in Johannesburg, does seem to be associated with significant improvements in function in children with CP.

In contrast to the widely accepted gross motor development growth curves that state that children with CP tend to plateau in function after seven years of age and reach a ceiling to their motor development in their early teens, those children in this study making the greatest improvements fell between the ages of 14 and 18 years old. This finding has led the researcher to question the validity of these predictive curves, as well as use of the apparently stable GMFCS for classification of functional ability, for children with CP in low-income countries who have not been exposed to regular therapeutic intervention from a young age and into adulthood. Older children, particularly if they have had limited access to treatment, may continue to benefit from intervention and this needs to be documented and explored.

Recommendations based on the study

- The results of this study do not support the use of FES over the external oblique muscles of children with CP
- Should future studies wish to determine the effects of FES in this population, then it is suggested that EMG be used to more accurately measure muscle strength and to measure changes in the selected motor strategy used by participants. If timed-sit ups are repeated as an outcome determinant of abdominal muscle strength, then more stringent standardisation of outcome use should be employed and sit-ups should include a rotatory component in order to better target the EOs
- As younger children naturally activate core muscle flexors and extensors en bloc to increase stability, simultaneous ES of the abdominals and the spinal extensor muscles, may improve study findings. Outcome measures should be taken both with and without the FES device to determine whether greater changes are noted while the device is in use. Timed activation of the abdominals for specific contraction during more specific, repetitive activities may also demonstrate greater functional improvements
- The study should be repeated using a more homogenous group of children with CP (i.e. either GMFCS levels I-III or IV-V only) in order to narrow the standard deviations found and better clarify whether the intervention provided incurs more positive results for certain functional levels
- Further study should elucidate whether significant improvements in peak flow meter readings found are meaningful in the absence of gross motor function and how the greater improvements seen in more severely affected children might warrant further study given the mortality rates linked to poor respiratory function in these groups
- Subgroup analysis of the GMFM individual dimensions, subgroup analysis of only the higher functioning participants (i.e. levels I-III), alternative statistical tests and a higher sample size in future studies may all yield more positive results
- Calculation of effect size for the current, and future studies, may better demonstrate positive outcomes

General recommendations

- Physiotherapy treatment should continue to be made available to the subgroup of children with CP who made the greatest improvements (i.e. those higher functioning children and children in their teenage years)
- The functioning of the abdominal muscles in CP and means of targeting these muscles for improvements in strength, posture and stability needs to be explored further. The differing role of the abdominals in the varying GMFCS levels should also be investigated
- Further research is needed to clarify whether impairment-based interventions have any functional carry-over into real life situations
- More research is needed on the frequency and intensity of intervention and whether intermittent, intensive blocks of therapy are more beneficial than once/twice weekly regular intervention maintained throughout the lifespan
- A South African, population-based study is necessary to determine whether the trend for continued improvements made by children with CP in their teens, who have not had regular intervention, observed in this study are confirmed and whether new predictive gross motor curves are necessary for application in such countries.

As has been previously discussed there is an international trend toward the publication of statistically positive scientific results. This may lead to an overemphasis of the positive effect of certain modalities. While the negative results demonstrated in this study are discouraging, they provide valuable information for professionals required to base their selection of therapeutic interventions on current scientific evidence (170). This is necessary in order to prevent the overprovision of ineffective interventions to people with CP (50) and to better guide families to select appropriate, evidence-based modalities that will improve the functioning of their child (317). As the methodological design of this study was strong, based on the CONSORT statement, results can be interpreted with a level of certainty and thus practitioners might not waste unnecessary resources, especially relevant in middle to low-income countries such as SA, on a treatment modality that is not effective in this context. Should the above recommendations be implemented then more positive results may be found on future investigation.

The results of this study also provide a basis for further study as, should these results be corroborated, contrary to popular belief, continued treatment of children with CP, into their teens, in middle to low income countries, should be considered (especially if early intervention was not possible or treatment was ceased early) as they may still possess the potential for further gross motor and functional gains.

"Even minor improvements have the potential to be of major practical and psychological significance for children with cerebral palsy and their families" (230).

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6. Appendices

6.1 Appendix I

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GMFCS – E & R
Gross Motor Function Classification System
Expanded and Revised

(Reference: Dev Med Child Neurol 1997;39:214-223)

INTRODUCTION & USER INSTRUCTIONS

The Gross Motor Function Classification System (GMFCS) for cerebral palsy is based on self-initiated movement, with emphasis on sitting, transfers, and mobility. When defining a five-level classification system, our primary criterion has been that the distinctions between levels must be meaningful in daily life. Distinctions are based on functional limitations, the need for hand-held mobility devices (such as walkers, crutches, or canes) or wheeled mobility, and to a much lesser extent, quality of movement. The distinctions between Levels I and II are not as pronounced as the distinctions between the other levels, particularly for infants less than 2 years of age.

The expanded GMFCS (2007) includes an age band for youth 12 to 18 years of age and emphasizes the concepts inherent in the World Health Organization's International Classification of Functioning, Disability and Health (ICF). We encourage users to be aware of the impact that **environmental** and **personal** factors may have on what children and youth are observed or reported to do. The focus of the GMFCS is on determining which level best represents the **child's or youth's present abilities and limitations in gross motor function**. Emphasis is on usual **performance** in home, school, and community settings (i.e., what they do), rather than what they are known to be able to do at their best (capability). It is therefore important to classify current performance in gross motor function and not to include judgments about the quality of movement or prognosis for improvement.

The title for each level is the method of mobility that is most characteristic of performance after 6 years of age. The descriptions of functional abilities and limitations for each age band are broad and are not intended to describe all aspects of the function of individual children/youth. For example, an infant with hemiplegia who is unable to crawl on his or her hands and knees, but otherwise fits the description of Level I (i.e., can pull to stand and walk), would be classified in Level I. The scale is ordinal, with no intent that the distances between levels be considered equal or that children and youth with cerebral palsy are equally distributed across the five levels. A summary of the distinctions between each pair of levels is provided to assist in determining the level that most closely resembles a child's/youth's current gross motor function.

We recognize that the manifestations of gross motor function are dependent on age, especially during infancy and early childhood. For each level, separate descriptions are provided in several age bands. Children below age 2 should be considered at their corrected age if they were premature. The descriptions for the 6 to 12 year and 12 to18 year age bands reflect the potential impact of environment factors (e.g., distances in school and community) and personal factors (e.g., energy demands and social preferences) on methods of mobility.

An effort has been made to emphasize abilities rather than limitations. Thus, as a general principle, the gross motor function of children and youth who are able to perform the functions described in any particular level will probably be classified at or above that level of function; in contrast, the gross motor function of children and youth who cannot perform the functions of a particular level should be classified below that level of function.

OPERATIONAL DEFINITIONS

Body support walker – A mobility device that supports the pelvis and trunk. The child/youth is physically positioned in the walker by another person.

Hand-held mobility device – Canes, crutches, and anterior and posterior walkers that do not support the trunk during walking.

Physical assistance – Another person manually assists the child/youth to move.

Powered mobility – The child/youth actively controls the joystick or electrical switch that enables independent mobility. The mobility base may be a wheelchair, scooter or other type of powered mobility device.

Self-propels manual wheelchair – The child/youth actively uses arms and hands or feet to propel the wheels and move.

Transported – A person manually pushes a mobility device (e.g., wheelchair, stroller, or pram) to move the child/youth from one place to another.

Walks – Unless otherwise specified indicates no physical assistance from another person or any use of a hand-held mobility device. An orthosis (i.e., brace or splint) may be worn.

Wheeled mobility – Refers to any type of device with wheels that enables movement (e.g., stroller, manual wheelchair, or powered wheelchair).

GENERAL HEADINGS FOR EACH LEVEL

LEVEL I	-	Walks without Limitations
LEVEL II	-	Walks with Limitations
LEVEL III	-	Walks Using a Hand-Held Mobility Device
LEVEL IV	-	Self-Mobility with Limitations; May Use Powered Mobility
LEVEL V	-	Transported in a Manual Wheelchair

DISTINCTIONS BETWEEN LEVELS

Distinctions Between Levels I and II - Compared with children and youth in Level I, children and youth in Level II have limitations walking long distances and balancing; may need a hand-held mobility device when first learning to walk; may use wheeled mobility when traveling long distances outdoors and in the community; require the use of a railing to walk up and down stairs; and are not as capable of running and jumping.

Distinctions Between Levels II and III - Children and youth in Level II are capable of walking without a hand-held mobility device after age 4 (although they may choose to use one at times). Children and youth in Level III need a hand-held mobility device to walk indoors and use wheeled mobility outdoors and in the community.

Distinctions Between Levels III and IV - Children and youth in Level III sit on their own or require at most limited external support to sit, are more independent in standing transfers, and walk with a hand-held mobility device. Children and youth in Level IV function in sitting (usually supported) but self-mobility is limited. Children and youth in Level IV are more likely to be transported in a manual wheelchair or use powered mobility.

Distinctions Between Levels IV and V - Children and youth in Level V have severe limitations in head and trunk control and require extensive assisted technology and physical assistance. Self-mobility is achieved only if the child/youth can learn how to operate a powered wheelchair.

Gross Motor Function Classification System – Expanded and Revised (GMFCS – E & R)

BEFORE 2ND BIRTHDAY

LEVEL I: Infants move in and out of sitting and floor sit with both hands free to manipulate objects. Infants crawl on hands and knees, pull to stand and take steps holding on to furniture. Infants walk between 18 months and 2 years of age without the need for any assistive mobility device.

LEVEL II: Infants maintain floor sitting but may need to use their hands for support to maintain balance. Infants creep on their stomach or crawl on hands and knees. Infants may pull to stand and take steps holding on to furniture.

LEVEL III: Infants maintain floor sitting when the low back is supported. Infants roll and creep forward on their stomachs.

LEVEL IV: Infants have head control but trunk support is required for floor sitting. Infants can roll to supine and may roll to prone.

LEVEL V: Physical impairments limit voluntary control of movement. Infants are unable to maintain antigravity head and trunk postures in prone and sitting. Infants require adult assistance to roll.

BETWEEN 2ND AND 4TH BIRTHDAY

LEVEL I: Children floor sit with both hands free to manipulate objects. Movements in and out of floor sitting and standing are performed without adult assistance. Children walk as the preferred method of mobility without the need for any assistive mobility device.

LEVEL II: Children floor sit but may have difficulty with balance when both hands are free to manipulate objects. Movements in and out of sitting are performed without adult assistance. Children pull to stand on a stable surface. Children crawl on hands and knees with a reciprocal pattern, cruise holding onto furniture and walk using an assistive mobility device as preferred methods of mobility.

LEVEL III: Children maintain floor sitting often by "W-sitting" (sitting between flexed and internally rotated hips and knees) and may require adult assistance to assume sitting. Children creep on their stomach or crawl on hands and knees (often without reciprocal leg movements) as their primary methods of self-mobility. Children may pull to stand on a stable surface and cruise short distances. Children may walk short distances indoors using a hand-held mobility device (walker) and adult assistance for steering and turning.

LEVEL IV: Children floor sit when placed, but are unable to maintain alignment and balance without use of their hands for support. Children frequently require adaptive equipment for sitting and standing. Self-mobility for short distances (within a room) is achieved through rolling, creeping on stomach, or crawling on hands and knees without reciprocal leg movement.

LEVEL V: Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and standing are not fully compensated for through the use of adaptive equipment and assistive technology. At Level V, children have no means of independent movement and are transported. Some children achieve self-mobility using a powered wheelchair with extensive adaptations.

BETWEEN 4TH AND 6TH BIRTHDAY

LEVEL I: Children get into and out of, and sit in, a chair without the need for hand support. Children move from the floor and from chair sitting to standing without the need for objects for support. Children walk indoors and outdoors, and climb stairs. Emerging ability to run and jump.

LEVEL II: Children sit in a chair with both hands free to manipulate objects. Children move from the floor to standing and from chair sitting to standing but often require a stable surface to push or pull up on with their arms. Children walk without the need for a hand-held mobility device indoors and for short distances on level surfaces outdoors. Children climb stairs holding onto a railing but are unable to run or jump.

LEVEL III: Children sit on a regular chair but may require pelvic or trunk support to maximize hand function. Children move in and out of chair sitting using a stable surface to push on or pull up with their arms. Children walk with a hand-held mobility device on level surfaces and climb stairs with assistance from an adult. Children frequently are transported when traveling for long distances or outdoors on uneven terrain.

LEVEL IV: Children sit on a chair but need adaptive seating for trunk control and to maximize hand function. Children move in and out of chair sitting with assistance from an adult or a stable surface to push or pull up on with their arms. Children may at best walk short distances with a walker and adult supervision but have difficulty turning and maintaining balance on uneven surfaces. Children are transported in the community. Children may achieve self-mobility using a powered wheelchair.

LEVEL V: Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and standing are not fully compensated for through the use of adaptive equipment and assistive technology. At Level V, children have no means of independent movement and are transported. Some children achieve self-mobility using a powered wheelchair with extensive adaptations.

BETWEEN 6TH AND 12TH BIRTHDAY

Level I: Children walk at home, school, outdoors, and in the community. Children are able to walk up and down curbs without physical assistance and stairs without the use of a railing. Children perform gross motor skills such as running and jumping but speed, balance, and coordination are limited. Children may participate in physical activities and sports depending on personal choices and environmental factors.

Level II: Children walk in most settings. Children may experience difficulty walking long distances and balancing on uneven terrain, inclines, in crowded areas, confined spaces or when carrying objects. Children walk up and down stairs holding onto a railing or with physical assistance if there is no railing. Outdoors and in the community, children may walk with physical assistance, a hand-held mobility device, or use wheeled mobility when traveling long distances. Children have at best only minimal ability to perform gross motor skills such as running and jumping. Limitations in performance of gross motor skills may necessitate adaptations to enable participation in physical activities and sports.

Level III: Children walk using a hand-held mobility device in most indoor settings. When seated, children may require a seat belt for pelvic alignment and balance. Sit-to-stand and floor-to-stand transfers require physical assistance of a person or support surface. When traveling long distances, children use some form of wheeled mobility. Children may walk up and down stairs holding onto a railing with supervision or physical assistance. Limitations in walking may necessitate adaptations to enable participation in physical activities and sports including self-propelling a manual wheelchair or powered mobility.

Level IV: Children use methods of mobility that require physical assistance or powered mobility in most settings. Children require adaptive seating for trunk and pelvic control and physical assistance for most transfers. At home, children use floor mobility (roll, creep, or crawl), walk short distances with physical assistance, or use powered mobility. When positioned, children may use a body support walker at home or school. At school, outdoors, and in the community, children are transported in a manual wheelchair or use powered mobility. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports, including physical assistance and/or powered mobility.

Level V: Children are transported in a manual wheelchair in all settings. Children are limited in their ability to maintain antigravity head and trunk postures and control arm and leg movements. Assistive technology is used to improve head alignment, seating, standing, and and/or mobility but limitations are not fully compensated by equipment. Transfers require complete physical assistance of an adult. At home, children may move short distances on the floor or may be carried by an adult. Children may achieve self-mobility using powered mobility with extensive adaptations for seating and control access. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports including physical assistance and using powered mobility.

BETWEEN 12TH AND 18TH BIRTHDAY

Level I: Youth walk at home, school, outdoors, and in the community. Youth are able to walk up and down curbs without physical assistance and stairs without the use of a railing. Youth perform gross motor skills such as running and jumping but speed, balance, and coordination are limited. Youth may participate in physical activities and sports depending on personal choices and environmental factors.

Level II: Youth walk in most settings. Environmental factors (such as uneven terrain, inclines, long distances, time demands, weather, and peer acceptability) and personal preference influence mobility choices. At school or work, youth may walk using a hand-held mobility device for safety. Outdoors and in the community, youth may use wheeled mobility when traveling long distances. Youth walk up and down stairs holding a railing or with physical assistance if there is no railing. Limitations in performance of gross motor skills may necessitate adaptations to enable participation in physical activities and sports.

Level III: Youth are capable of walking using a hand-held mobility device. Compared to individuals in other levels, youth in Level III demonstrate more variability in methods of mobility depending on physical ability and environmental and personal factors. When seated, youth may require a seat belt for pelvic alignment and balance. Sit-to-stand and floor-to-stand transfers require physical assistance from a person or support surface. At school, youth may self-propel a manual wheelchair or use powered mobility. Outdoors and in the community, youth are transported in a wheelchair or use powered mobility. Youth may walk up and down stairs holding onto a railing with supervision or physical assistance. Limitations in walking may necessitate adaptations to enable participation in physical activities and sports including self-propelling a manual wheelchair or powered mobility.

Level IV: Youth use wheeled mobility in most settings. Youth require adaptive seating for pelvic and trunk control. Physical assistance from 1 or 2 persons is required for transfers. Youth may support weight with their legs to assist with standing transfers. Indoors, youth may walk short distances with physical assistance, use wheeled mobility, or, when positioned, use a body support walker. Youth are physically capable of operating a powered wheelchair. When a powered wheelchair is not feasible or available, youth are transported in a manual wheelchair. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports, including physical assistance and/or powered mobility.

Level V: Youth are transported in a manual wheelchair in all settings. Youth are limited in their ability to maintain antigravity head and trunk postures and control arm and leg movements. Assistive technology is used to improve head alignment, seating, standing, and mobility but limitations are not fully compensated by equipment. Physical assistance from 1 or 2 persons or a mechanical lift is required for transfers. Youth may achieve self-mobility using powered mobility with extensive adaptations for seating and control access. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports including physical assistance and using powered mobility.

6.2 Appendix II

Diagnosis

This section begins with clinical observations made in the diagnosis of CP and is followed by neuroimaging as an adjunct to diagnosis. The section is concluded with the impact learning of a diagnosis of CP can have on families and caregivers.

Coupled with the difficulty in classifying CP is the difficulty in diagnosing the condition as no specific diagnostic test exists (59)(76)(343). Consensus is generally met regarding the need for early diagnosis, as intervention should start as soon as possible to benefit from the early plastic changes possible in the brain and to allow for greater ease in communication with families regarding prognosis and other expectations (5)(59)(73)(76)(105)(140)(230). This is made difficult by the heterogeneity of the group of disorders and the fact that developmental delay is often only noted after the first 1-2 years of life (59)(73)(76)(105)(140)(343)(344) .

It is suggested that at risk infants have a thorough family and medical history taken early and that clinicians make regular observations in order to discern delays as early as possible (26)(87)(343)(345). Signs and symptoms first noted are usually delays in the development of expected motor milestones, limb hypertonia and/or hypotonia in the trunk and brisk reflexes. These symptoms may be transient in preterm infants but the persistence of such signs beyond one year of age is a strong indicator of CP (5)(26)(87)(344)(346). Parents should be asked to observe their children and report any signs of abnormal movement and posture early (96). Differential diagnoses such as metabolic disorders, muscular dystrophies, genetic anomalies and hereditary dystonias should be ruled out before an official diagnosis is made (5).

With advances in medical technology, Magnetic Resonance Imaging (MRI) in neonatology has become more commonplace (76)(87), allowing for correlations between physical presentations of CP and neurological injury to be made. In this way, professionals have been able to retrospectively determine aetiology and timing of the original brain insult as

well as the severity of the damage (2)(6)(76)(89). Studies show that MRI can accurately detect neurological abnormality in 70-92% of children with CP (5)(76)(87)(343). With this said, 25% of those infants with abnormal findings on MRI do not develop any physical symptoms and the area of brain lesion does not always directly correlate with physical fallouts (76).

CP is a lifelong condition and although non-progressive, has many secondary complications meaning continued comprehensive intervention is necessary and dependence on caregivers may be permanent. Studies have documented increased psychological, health and financial burdens placed on families of children with CP and thus ongoing support from practitioners following diagnosis is required (76)(347).

Aetiology

This section on aetiology discusses the possible risk factors for the development of CP first. Following this is a discussion of pre-, peri- and postnatal causes of CP and the aetiologies specific to the different subtypes of CP. The section is concluded by how socioeconomic circumstances influence aetiology and the effect this may have on a South African population.

Numerous risk factors for the development of CP have been reported. In the past, it was assumed CP was always caused by prenatal or birth asphyxia. MRI technology has helped demonstrate that other aetiologies are also responsible for the development of CP (59)(60)(87)(89) and that birth trauma causing asphyxia may only account for 6% of CP cases (57)(60).

Premature birth is still the highest predictor for prevalence rates of CP with gestational age and birth weight inversely proportional to the risk of developing CP (57)(59)(76)(91)(344)(346). Maternal infection (especially those affecting the reproductive tract) increases the risk of developing CP substantially and has been linked to low Apgar scores and hypoxaemia (57)(87)(106). The acronym TORCH has been used to represent the

most common infections resulting in CP; namely toxoplasmosis, others (i.e. syphilis and HIV), rubella, cytomegalovirus and herpes simplex virus. The degree, to which the central nervous system (CNS) of the developing foetus is affected, when exposed to such infections, depends on the stage of maturation at the time of infection. CP rarely occurs when infection is introduced neonatally and CNS fallouts are worse the earlier the infection takes place prenatally (60)(89).

Cerebral infarction/stroke may occur at any stage during brain development with the middle cerebral artery affected 95% of the time. Incidence is highest in the 37-40 week gestational age group with statistics decreasing steadily as gestational age decreases (60)(89). Multiple births have a higher expectancy of developing CP. This can be explained as these children are usually born with low or very low birth weights and mothers rarely carry to full term (although this is not always the case) (57)(59)(60)(87)(91)(106)(107) . CP prevalence rates have been affected due to the generalised global increase in multiple births due to improved accessibility of fertility treatments (106).

Brain malformations, such as abnormal neuronal and glial proliferation or migration and abnormal cortical organisation, are associated with the development of CP (60)(89)(107). Less frequently experienced conditions such as maternal and foetal thrombophilia (60)(87)(106), placental abnormalities (87), maternal thyroid dysfunction or hypertension (87)(91), delayed delivery from time of labour (91), meconium aspiration, meningitis (107), breech presentation at birth (107)(109) and hyperbilirubinaemia (107)(109) also increase the risk of developing CP.

Prenatal refers to the period from conception until labour and delivery. Peri- or neonatal is the period from delivery until 28 days of life (6)(107). Prenatal causes of CP can account for 70-80% of all cases; the cause of which is often unknown (26)(87). A population-based study in Sweden between 1999 and 2002 showed that children born full term with a prenatal cause of CP tended to function at a higher level (73% had a GMFCS level I or II) than term babies with perinatal origin of insult (53% of whom functioned at levels IV or V) (6).

If no pre- or perinatal aetiology is found and the infant is deemed normal prior to brain insult the cause is said to be postnatal (110). Postnatally acquired causes of CP include traumatic head injuries, child abuse, cerebrovascular insults, infection, severe dehydration, seizures, near drowning and encephalopathy (5)(60)(109)(110)(111). Only 5-18% of CP cases reported are due to postnatal causes (59).

Certain risk factors have been linked with particular presentations of CP with prematurity generally resulting in diplegic CP and cerebrovascular insults causing hemiplegia. Brain maldevelopments tend to cause ataxia and quadriplegia and dyskinetic subtypes of CP have been closely connected to hypoxic events (83)(109)(110). In the past, kernicterus was strongly associated with the development of athetosis (83)(110).

6.3 Appendix III

Sensorimotor impairments

As has been described in section 2.2.1 the comprehensive definition of CP now includes accompanying impairments to the sensory, perceptual, communicative and behavioural systems, as well as the occurrence of epilepsy and secondary complications (2)(131). The more severe the presentation of CP (i.e. the higher the GMFCS level), the more likely one is to have one or multiple accompanying impairments (6)(120).

Epilepsy is the most common comorbidity seen in CP and studies have cited its prevalence at between 33-50% of all CP cases (6)(26) (57)(82)(108)(134)(343)(347). Seventy percent of those with epilepsy will experience their first seizure within a year of their birth and it is the least prevalent in those with a spastic diplegic or ataxic distribution of CP (82)(108)(343). An epidemiological study conducted in the Netherlands showed that the risk of developing epilepsy was much higher in those with more severe CP and 94% of quadriplegics who also had severe cognitive impairment suffered from epilepsy (70)(82). Interestingly, a 2011 study by Himmelmann and Uvebrant revealed that epilepsy was more common in those children born after 32 weeks gestation (6)(343). There are numerous antiepileptic drugs available for the treatment of this additional impairment in CP (108)(343).

Visual defects occurring in children with CP can be broken up into anterior defects (of the eyes themselves or the optic nerves) and posterior defects (with damage anywhere from the optic tracts to the occipital cortex). A common posterior defect seen, mostly in quadriplegic CP, is cerebral/cortical visual impairment where the individual is said to be cortically blind with the structure of the eye itself intact. The person is not completely blind but has severe visual fallouts due to damage to the optic nerve or occipital cortex (26)(175). Another common problem in CP is visual field deficits, such as homonymous hemianopia (mostly seen in hemiplegic children), where either the right or left side of the visual field in both eyes is absent depending on the side of the cortical lesion (26)(175). Nystagmus is less commonly seen and is experienced by those where involuntary movement is a part of their classification, causing involuntary movements of the eyes as

well. This causes difficulty focusing and thus limited visual acuity (175). A strabismus or squint present may be corrected surgically (26)(175). Prematurity has also been linked to severe myopia/near sightedness (5).

Studies claim that 17-62% of children with CP experience a visual defect of some sort (6)(10)(26)(82)(120)(134)(175)(343) and, as with other impairments, the more severe the presentation of CP the higher the likelihood of having a visual impairment (5). As one could expect, those children with visual impairments have greater difficulty exploring their environment and therefore have a lower potential for gross motor skill acquisition and independent mobility (70).

Less commonly experienced is hearing loss with only 3-15% of those included in studies requiring hearing aids or auditory therapy (5)(6)(346). Causes of CP such as prematurity, kernicterus, meningitis and birth asphyxia/hypoxia are associated with the incidence of hearing loss (5)(108)(343).

Probably the most frustrating of the associated impairments in CP (as well as the most limiting in terms of participation), are communication deficits, with between 38-59% of those with CP unable to communicate appropriately and up to 25% unable to speak at all (6)(343)(347). This deficit in communication can be either in receptive understanding of language or expressive speech ability (2). The extent to which communication is hindered depends on the area of cortical damage and whether the language centres of the brain have been spared or not (343). The primary motor impairments characterising CP can cause oromotor difficulties, making articulation and the coordination of speech difficult as well (26)(343). This lack of oromotor control may also lead to excessive drooling in up to 22% of those with CP (347).

Epilepsy, visual and auditory impairments can all negatively influence the attainment of postural control, thus further limiting gross motor function.

Cognitive and perceptual impairments

Intellectual deficits are seen in as many as 40-75% of those with CP (5)(26)(57)(120)(134)(347). As with visual and auditory deficits, the development of cognitive defects in CP is associated with premature birth (87). Those with brain malformations are also more likely to experience fallouts in cognitive functioning (6). As can be expected, the more global brain insult causing quadriplegia is associated with a higher prevalence of cognitive impairment, compared to hemiplegic CP which is the least likely classification to experience cognitive short falls (5)(120)(200)(343). Epilepsy sufferers are more likely to experience cognitive impairment and a 2006 epidemiological study claimed that 97.7% of children severely affected by CP have profound intellectual disability (2)(82)(343).

While cognitive skills may be normal, many children with CP have coexisting learning disorders with 12% mildly affected and 33% with more serious specific learning difficulties (5)(6)(134). Attention may also be affected (with attention deficit hyperactivity disorder a fairly common coexisting condition) (2)(6), as may be memory retention in CP (10). Rarely, children with CP may experience concomitant autism (6). Behavioural disturbances may occur due to the primary cortical insult causing CP or due to emotions such as frustration associated with limitations in mobility and participation and occur in up to 26% of children with CP (347).

Certain perceptual deficits may also be present in children with CP. Perception can be explained as the cortical interpretation and integration of sensory information received by the brain (2). Coupled with the motor impairments experienced by the CP population, deficits in motor planning may exacerbate difficulties with task completion and the refinement of new motor skills (84). Proprioception is the body's recognition of where its limbs are in space. Errors in joint position sense were high in a study of participating children with diplegia and hemiplegia, with only the dominant upper limb measuring close to figures achieved by their TD peers. Considerable proprioceptive fallout was seen even in the least affected individuals and deficits were attributed to cortical damage, as well as dysfunction of sensory receptors in these individuals (348). Hemi-neglect may occur in

hemiplegic CP due to the difficulty of joint excursion on that side or due to complete disregard for this side due to the cortical damage present (84).

Stereognosis or the ability to recognise items by touch, based on their physical properties, may be limited in up to 50% of those with CP (82). Over and above visual acuity problems seen in children with CP, visual-perceptual problems are frequently encountered interfering with scholastic progress (349).

Cognitive, behavioural and perceptual impairments all need to be taken into consideration during intervention planning, as all of these factors have the ability to negatively influence participation in tasks or tolerance of certain treatment strategies.

Secondary complications

With advances in medical management, more children with CP live into their adult years (26). While CP as a condition is non-progressive, the perpetual abnormal postures and movement experienced, inactivity and spasticity can all lead to the development of secondary complications in CP. If not managed appropriately these then exacerbate the original impairments present causing a cycle of further disablement (117)(129)(167)(350).

In general, the more severe the presentation of CP is, the more likely the development of secondary complications (55). The resistance to stretch caused by spasticity, if strong enough, can lead to muscle shortening over time and eventually fixed muscle and joint contractures, further limiting functional mobility (86)(144). Common muscles involved are the hip and knee flexors often developing fixed contractures over time. Equinovarus deformities of the feet, patella alta (i.e. high riding patellae) and tibial or femoral torsional deformities are other frequently seen secondary complications in CP (5)(86)(117)(351).

In those that are wheelchair-bound and inactive, postural deformities are prone to occur. Due to the pull of spasticity on the body in an asymmetrical manner, deformities such as

scoliosis of the spine, obliquity of the pelvis and possible sublaxations and dislocations of the femoral heads may occur. These can lead to a windswept posture (i.e. with one hip abducted and externally rotated and the other adducted and internally rotated toward the opposing limb and thus more likely to dislocate) preventing independent mobility and further exacerbating physical impairments (5)(26)(55)(86)(82)(136). While these types of deformities do not occur in those minimally affected by CP, windswept deformities are observed in 90% of those classified as a GMFCS level V (86).

Abdominal weakness and poor external postural support given in sitting may lead to exaggerated spinal kyphosis and occasionally the development of a gibbus (i.e. rib hump). Limitations in hip flexor range of movement and poor pelvic stability can lead to a hyperlordotic spine (86). Spinal deformities may also lead to respiratory compromise in the form of obstructive airway disorders, dyspnoea, aspiration, impaired rib excursion during inspiration and GOR (10)(198).

Without an opportunity to weight bear, more severely affected individuals with CP develop bone density disorders such as osteopaenia, osteoporosis and on occasion pathological fractures. Those with a high degree of spasticity and large muscle imbalances are also at greater risk for the development of osteoarthritis due to excessive joint compression caused by these impairments (26)(117)(163).

As has been alluded to in the body of the literature review, swallowing difficulties/dysphagia can lead to several other impairments, over and above respiratory complications. Poor nutritional intake due to swallowing problems in severely affected children leads to a body mass index below predicted norms and malnourishment (5)(108). Hypertonicity may lead to constipation in 80% of those with CP and urinary incontinence or spastic bladder may occur (5)(82)(108).

Up to 82% of both children and adults with CP experience pain (5)(10)(117) which leads to higher chronic pain prevalence than in TD populations (10)(82)(117). Personal reports of pain have shown that almost a quarter of those with CP experiencing pain; list it as being over 7/10 on a visual analogue scale (10).

The secondary complications discussed above can all lead to pain or discomfort, pressure sores, respiratory complications and sleep disorders further limiting participation and HRQoL (55)(117)(136)(202)(214). Pain, fatigue and worsening physical impairments lead to deteriorations in physical functioning in adulthood; in particular, in walking ability (162)(350). Long term secondary complications may also lead to the development of psychological disorders such as depression (117). All of the above complications bring about further participation limitations of the individual (10).

Whether physical or psychological, all of the above possible secondary complications can limit the development of adequate postural control and thus further hinder functional independence if not prevented.

6.4 Appendix IV

Checklist of interventions

School Number: _____

Therapist number: _____

Date of assessment: _____

Qualifications and years obtained:

_____

NDT or other postgraduate courses relevant to treatment of learners with cerebral palsy (and years obtained):

_____

_____

PHYSIOTHERAPY INTERVENTION CHECKLIST

TREATMENT APPROACH	TREATMENT TECHNIQUE	PERFORMED ROUTINELY
Neurodevelopmental Therapy (NDT)	Facilitation of movement: Facilitation of transitions between positions Facilitation of repetitive movements with assistance Tone inhibiting techniques: Tone Influencing Patterns (TIPs) Rotations Passive stretches: Length of time held: _____ Weight bearing in sustained postures Shaking/vibrations/tapping etc. Posture correction for symmetry: Other: _____	
Orthopaedic Approach	Physiological active movements: Repetitions _____ Physiological passive movements: Repetitions _____ Passive joint mobilisations: Passive stretching: Time held _____ Myofascial release/massage: Strapping/taping: Use of heat/ice: Other: _____	
Strengthening	Core (abdominal) strengthening: Repetitions _____ Resistance used _____ To fatigue? _____ Breathing exercises, voicing, laughing, shouting etc. _____ General (limb) strengthening: Repetitions _____ Resistance used _____ To fatigue? _____ PNF Stationary cycling Weight bearing positions for scapula/pelvic stability Power Plate Other: _____	
Balance Specific Training Sitting or standing? _____	External postural perturbations: Unstable/moving surfaces/equipment : _____ Unstable positions: _____ Rhythmic Stabilisations:	

	Other: _____	
Electrical stimulation	Threshold Electrical Stimulation: Neuromuscular Electrical Stimulation: Functional Electrical Stimulation: Transcutaneous Electrical Nerve Stimulation: Other: _____	
Gait training	Facilitation of movement during gait: Orthotics used during gait training: _____ Assistive devices used during gait training: _____ Treadmill training: Other: _____	
Constraint induced therapy		
Hydrotherapy		
Vojta therapy	Goal-directed pressure given to defined zones on the body stimulating “reflex creeping” in prone or “reflex rolling” in supine/side lying	
Conductive education	Structured framework for group therapy, using a task series, rhythmical intention and specific equipment (combo of rehab and education)	
Motor learning/ goal-directed functional therapy	Goal/specific functional task selected, broken into components and full task completed repetitively, goal attainment at some level, feedback given by therapist (knowledge of performance/results)	

Other techniques used and not listed above:

_____

_____

What equipment is used during treatments?

_____

_____

DEFINITIONS:

Neurodevelopmental Therapy (NDT): The NDT/Bobath Approach is used for the assessment, management and treatment of individuals with central nervous system pathophysiology. The individual's strengths and impairments are identified and addressed in relation to functional abilities and limitations. The NDT Approach continues to evolve with the emergence of new theories, models, research, and information in the movement sciences. NDT is a hands-on, problem solving approach. Intervention involves direct handling and guidance to optimise independent function and improve the performance of everyday activities.

Facilitation of movement: The use of afferent information to effect improvements in motor performance. Facilitation is used to enable successful movement and task performance with regard to aspects such as postural orientation, components of movement, functional sequences of movement, recognition of the task and motivation to complete the task. Facilitation via handling skills is intended to provide key components of the spatial and temporal aspects of a specific movement/task to enable the individual to have an experience of movement that is not passive but one that they cannot yet do alone.

Key points of control: Parts of the body from where the therapist can most effectively control and change the patterns of posture and movement in other parts of the body) thus influencing tone.

Tone Influencing Patterns: Normal patterns of activity, which are used to modify abnormal patterns of posture and movement thereby reducing hypertonus and/or the development of hypertonus or used in low tone to facilitate stability. The whole body is controlled by the therapist in a reversal of the abnormal pattern or the abnormal pattern is partially changed while other parts of the body remain free to move.

Physiological joint movements: Pure movements of the body in different planes that can be performed actively by the patient or passively by the therapist.

Joint mobilisations: Slow, oscillatory or sustained accessory movements performed on the joints by the therapist aimed at decreasing pain and/or increasing joint mobility.

Core Strengthening: The balanced development of the deep and superficial muscles that stabilise, align, and move the trunk of the body (especially the abdominals and muscles of the back). Core strength goes beyond the surface muscles and asks us to utilise our deep internal muscles to maintain stability in motion. Generally, core strengthening is affiliated with sit-ups, crunches, plank positions and creative variations of each.

General Muscle Strengthening: The use of resistance to muscular contraction to build the strength, anaerobic endurance, and size of skeletal muscles. Different methods of strength training include the use of gravity or elastic/hydraulic forces to oppose muscle contraction. Training commonly uses the technique of progressively increasing the force output of the muscle through incremental increases of weight, elastic tension or other resistance, and uses a variety of exercises and types of equipment to target specific muscle groups. According to popular theory, sets of one to five repetitions primarily develop strength, with more impact on muscle size. Sets of six to twelve repetitions develop a balance of strength, muscle size and endurance. Sets of thirteen to twenty repetitions develop endurance, with some increases to muscle size and limited impact on strength. Sets of more than twenty repetitions are considered to be focused on aerobic exercise.

Proprioceptive Neuromuscular Facilitation (PNF): A combination of passive stretching and isometrics contractions to hasten the response of the neuromuscular mechanism through the stimulation of the proprioceptors. The most common PNF leg or arm positions encourage flexibility and coordination throughout the limb's entire range of motion. PNF is used to supplement daily stretching and is employed to make quick gains in range of motion to help improve performance.

Power Plate: A brand of vibrating platform used as exercise equipment. Traditional exercises such as squats and push-ups are done on the vibrating base. Whole-body vibration exercise may enhance muscle strength adaptations by eliciting the tonic vibration reflex, activating muscle spindles, and stimulating transient increases in growth hormones.

External postural perturbations: The use of a disturbance or upset in balance to activate specific recovery, which is not under volitional control therefore training compensatory responses (when perturbations are unexpected). A postural perturbation is a sudden change in conditions that displaces the body posture away from equilibrium. These can be applied to any body part such as a push to the trunk, head or limbs. Perturbances via a supporting surface which then displaces the base of support under the body’s centre of mass may also be used.

Threshold Electrical Stimulation: A low-level, subcontraction electrical stimulus applied at home during sleep.

Neuromuscular Electrical Stimulation: The application of an electrical current of sufficient intensity to elicit muscle contraction.

Functional Electrical Stimulation: Neuromuscular stimulation (as above) applied in a task specific manner where a muscle is stimulated when it should be contracting during a functional activity.

Transcutaneous Electrical Nerve Stimulation (TENS): A technique used to relieve pain in an injured or diseased part of the body in which electrodes applied to the skin deliver intermittent stimulation to surface nerves and block the transmission of pain signals.

Constraint induced therapy: Involves intense functionally oriented task practice of the paretic upper extremity along with restraint of the less-impaired upper extremity for most waking hours. This approach encourages use of the paretic upper extremity in daily life and is thought to help overcome "learned non-use" of the paretic upper extremity. Treatment by restraining only the less-impaired upper extremity, which is typically accomplished by placing the entire arm in a sling or placing the hand in a mitt is referred to as "forced use".

Hydrotherapy/Aqua therapy: The use of heated water to relieve discomfort and promote physical well-being. This has been used in children with disabilities as the negative influences of poor balance, postural control and excessive joint loading are reduced in water. The resistive forces of buoyancy and viscous drag permit a variety of aerobic and strengthening activities that can be easily modified to accommodate a wide range of motor abilities.

Vojta therapy: The therapeutic use of reflex locomotion to enable elementary patterns of movement in patients with impaired central nervous systems. Reflex locomotions are activated "reflexogenically" (i.e. therapeutically applied external stimuli and their predefined and always identical, "automatically" present movement responses).
The therapist administers goal-directed pressure to defined zones on the body in a patient who is in a prone, supine or side lying position. Such stimuli lead automatically and involuntarily to two movement complexes: Reflex creeping in a prone lying position and reflex rolling from a supine and side lying position. This allows a coordinated, rhythmic activation of the total skeletal musculature and a CNS response at various circuit levels. Through therapeutic use of reflex locomotion, the involuntary muscle functions necessary for spontaneous movements in everyday life are activated in the patient. Through the repeated stimulations of these "reflex-like" movements, it is postulated that a "freeing" of functionally blocked networks of nerves between the brain and spinal cord occur and after treatment, these segmental patterns should be more available spontaneously to the patient.

Conductive education (CE): Based on an educational model of intervention it integrates educational and rehabilitative goals into one programme by addressing many aspects of a child's development and personality. Outcomes therefore also include academic, communication and social skills. CE is generally provided in a group setting. A specific "conductor" is responsible for putting together the intervention based on an individual's capabilities and then intervention is given working under a highly structured framework in the group, using a task series, the use of rhythmical intention and specific equipment.

Motor learning/goal-directed functional therapy: "Motor learning" takes place through practice allowing appropriate motor programmes to be acquired and later retained. In therapy functional meaningful goals are set based on importance to the specific child and family. The child is expected to explore active solutions to functional difficulties rather than a 'hands on' approach from the therapist. Functional activities are assumed to be learned through repetitive practice of goals set, through interaction with the environment in every day functional situations.

6.5 Appendix V

6.5.1 Rehabilitative treatment modalities

Family-centred treatment

As frequency of therapy is often limited by the financial and time constraints that families experience, studies started looking at how caregivers could become more actively involved in their disabled children's management. With a shift away from a pure medical model of medicine, the ICF has advocated the inclusion of all stakeholders in the decision making process when it comes to children with special needs (352). Therapeutic intervention guided by loved ones provides a unique opportunity for children to experience activities in their own home environment and be motivated by the people they trust the most (70). As parents observe the disabled individual on a daily basis they are considered the expert on their child's needs and they are often able to create learning opportunities that the practitioner is not (70)(352). Goals set by therapists not taking into account the greatest concerns of parents or the individual are often not met (124)(299) and each family needs to be treated as unique to achieve the greatest possible functional outcome (352). The psychological wellbeing of caregivers has been listed as higher on self-reports when a family centred treatment approach has been incorporated into management (353). This concept includes training of caregivers, provision of vital information and setting up of support systems for the families of those seeking treatment (353)(354).

Constraint-induced therapy (CIT)

Constraint-induced therapy (CIT) is generally used for patients with a hemiplegic distribution of CP. The technique involves the restraint of the 'unaffected' upper limb in order to encourage the forced use of the more affected side (27)(355)(356)(357). Devices for possible use include slings, casts, resting splints and mitts (356)(357). A programme of task-specific and repetitive activities is introduced during the period of immobilisation to ensure intensive functional use of the affected side (355)(356).

In 2013, 51 children with hemiplegic CP were randomised into two groups; both receiving CIT with a rigid orthosis for an hour a day for 10 weeks and one also receiving an intensive hand function and strengthening programme concomitantly. Improvements in upper limb and grip muscle strength and single-handed tasks were seen in both groups following the intervention period. Unfortunately, no control group was included to prevent confounding variables (355).

Most studies investigating the intervention have been case studies only and the parameters regarding length and means of constraint vary widely between authors (356). In general, constraint can last from 2-15 hours per day (219)(356)(357). Speculation has been made that cortical reorganisation can be seen on neuroimaging following CIT intervention (356). Negative components of this therapy include poor compliance, decreased protective reactions during postural disturbances when splinted and patient discomfort (356)(357). Constraint-induced therapy has long been incorporated into NDT treatment in order to facilitate compensatory mechanisms and forced use of the more affected side (170).

Stretching and passive movements

After NDT, one of the most commonly used treatment techniques in CP is that of passive muscle stretching, with therapists as well as parents encouraged to stretch children on a regular basis (138). It is well known that muscle activation is strongest within mid-range of movement for that particular joint (170). The purpose of stretching is to allow joint excursion to mid-range for strength, to try and influence hyperactive stretch reflexes present in those with CP and to maintain spastic muscle length to prevent secondary contractures and joint deformities (138)(140), thus delaying the need for surgical intervention (211). Unfortunately, while in-treatment improvements in range of motion might be observed, it is now well documented that in order to exact any sort of permanent change in muscle length one would need to stretch a muscle for more than six hours a day (49)(138) which is impractical unless splinting is used.

A survey conducted amongst 46 rehabilitation centres in Canada revealed that, despite a lack of scientific evidence supporting the use of stretching in CP, practitioners still believe that stretching can maintain current joint range of motion and thus positively affect functional outcomes. Up to 33% of questioned therapists' time was spent on stretching with a further 1-25% spent on showing parents how to incorporate stretching into their home rehabilitation programmes (211).

Studies have shown that the greatest amount of pain experienced by children with CP occurs during their daily stretching routine (49)(202). Without strong empirical evidence supporting the long-term benefit of stretching (211) one must consider whether this treatment modality should still be used with such frequency.

Orthoses and assistive devices/aids

Gait training of those with CP has many forms but is most often accompanied by the use of orthoses or assistive devices. The orthotic devices most commonly used in the rehabilitation of children with CP are called ankle foot orthoses (AFOs) (99). The positive benefits of postural support provided at the ankle (allowing swing through and heel strike) in those children with limited active dorsiflexion range and muscle activation during gait may be counteracted by discomfort and the atrophy of these muscles when placed in a statically held position perpetually (32)(86)(163)(232)(337). AFOs have been shown to improve gait parameters (such as walking speed, stride length and energy expenditure) in children with CP, when compared to walking barefoot (99)(324). AFOs should be worn for a minimum of six hours a day in order to influence hypertonicity and gastrocnemius muscle length (99)(140)(337). Serial casting of the ankles also takes place fairly frequently and has its greatest effect on range and spasticity when performed in conjunction with Botulinum Toxin A (Btx-A) application (140). Upper limb orthoses are used less often and generally limit rather than improve independent function (99)(263).

Postural seating adaptations are also important considerations in this population as the correct degree and type of support/stability provided can increase functionality of the upper limb and assist with independence in activities of daily living (42)(86). Ideally, any postural stability aid should be used constantly for 24 hours a day but this is usually neither

practical nor comfortable (49)(99). Positive postural effects need to be weighed against convenience of use, affordability and ease in transportation for the caregiver (49). Options include modified wheelchair back support systems, hip abduction devices and cushions, saddle, ischial shelf and pressure relief cushions and spinal braces (42)(86)(99)(358).

While it is generally considered that the use of orthoses and postural aids in children with CP is beneficial, little evidence exists to substantiate whether they are able to prevent deformities from developing or whether they improve posture as opposed to not using a device (49)(90)(99)(337)(358).

Standing frames can be used in those unable to stand independently or even those more severely affected that are never expected to stand as weight bearing in an upright position is thought to improve bone density (49), provide a sustained stretch of lower limb muscles and improve respiratory and gastrointestinal function (99)(198). Again, little conclusive empirical evidence exists to support these claims (49). More commonly prescribed are assistive devices such as walking sticks, crutches and wheeled walking frames, compensating for balance and gait fallouts to allow for independent ambulation. This is enabled by providing the individual with a greater base of support and by correcting body alignment (170)(177)(324)(359).

Orthoses and aids cannot be used in isolation and prescription needs to be accompanied by therapeutic and/or medical input (337). Acceptance of the device by the individual is tantamount to compliance with use (359). Children with CP are generally compliant with assistive device use due to the improved independent function and the opportunities for social participation such devices can provide (359).

Orthopaedic/manual therapy

While no studies on the effect of manual mobilisations or a more orthopaedic approach to physiotherapy in the treatment of CP could be found, they are briefly described here as the researcher has experienced that these techniques are practiced in SA in the treatment of CP.

Physiological movements can be explained as pure movements of the body in different planes that can be actively achieved by the patient or performed completely passively by the therapist. In order for full range physiological movements to be possible complete range of accessory movement needs to be available. These are small joint movements not under the active control of an individual. Joint mobilisations are slow, oscillatory or sustained accessory movements performed on spinal or peripheral joints, by the therapist, aimed at decreasing pain and/or increasing joint mobility (360).

This approach to therapy may also include the use of heat, ice, massage therapy, strapping and electrotherapy modalities such as ultrasound (360).

Proprioceptive neuromuscular facilitation (PNF)

Proprioceptive neuromuscular facilitation (PNF) is a group of stretching and strengthening techniques aimed at the body's proprioceptive system often used in the rehabilitation of people with CNS disorders (361). Specific placement of the therapists' hands and guidance of movement in predetermined functional patterns (usually diagonal) allow for summation of muscle contraction in the affected limb whilst the patient resists the direction of movement by the therapist maximally. By altering the timing or degree of resistance provided, the therapist can elicit either tonic or phasic stretch reflexes or Golgi tendon reflexes to encourage flexibility and coordination throughout the limb's entire range of movement. A system of reciprocal inhibition (whereby strongly contracting the agonist group of muscles can cause reflex relaxation of the antagonist muscles) is employed through techniques such as "slow reversal", "contract-relax" and "rhythmic initiation". Together, these procedures enable quick gains in range of movement to assist with improvements in performance (361). While no studies investigating PNF-specific techniques in the strengthening of people with CP could be found, it stands to reason that this modality could successfully be included in the treatment repertoire of those treating CP.

Gait training/aerobic exercise

As has been shown in section 2.3.1.1, children with CP have compromised cardiovascular fitness levels and thus aerobic activity should be included as part of their therapeutic intervention (124)(200). Such exercises include cycling, walking, swimming, martial arts and other sporting activities (197)(362). By improving fitness, energy expenditure and the physiological cost of movement can be decreased during activity (362).

Aerobic exercise in those with CP can be administered with treadmill training. This is often performed with the assistance of a harness to enable greater ease of weight bearing and to help facilitate a more natural stepping reflex (27)(68)(363). People with CP included in a biweekly eight-week programme of cycling (for 30 minutes each session) made statistically significant gains in cardiovascular fitness (197). This is a treatment modality frequently suggested for the strength and fitness training of people with CP (130). Stationary bicycles provide a safe and stable way in which to make gains in these parameters, as the individual can be strapped in and resistance can be graded as improvements are made (130).

In a population prone to respiratory infections and/or respiratory compromise, aerobic exercise can promote less reliance on shallow breathing and an increase in ventilatory efficiency and aerobic capacity, thus reducing the risk of such infections (240)(364). Specific attention should also be paid to the particular strengthening of respiratory muscles in CP (200)(364). A trial involving 46 children with CP showed a 42% higher gain in vital capacity in children receiving a swimming and gym exercise intervention over a control group receiving conventional physiotherapy (364). In their guidelines for exercise in children, the American College of Sports Medicine has recommended that, in order for cardiovascular improvements to take place, children need to exercise for between 30-60 minutes almost every day and at an intensity where one reaches 60-80% of maximal heart rate and oxygen uptake (240).

Group training sessions at gyms or community centres also provide the opportunity for social interaction (240). As well as having positive respiratory effects, again it can be seen that current treatment modalities used in the treatment of CP are aimed at strengthening.

Hydrotherapy

Aqua/hydrotherapy involves a wide variety of therapeutic techniques performed with the patient in a heated pool. Principles of buoyancy, hydrostatic pressure and drag enable activities not possible on land to be achieved by more severely disabled children with CP (199)(209)(240). Joint loading, weight bearing and postural requirements against gravity are all lessened when submerged in the water (209).

Controlled trials have demonstrated significant improvements in respiratory function (199)(200), cardiovascular fitness (200) and muscle strength (240) in participants with CP following aqua based interventions. A review of studies with samples suffering from general CNS disorders cited improvements in pain, flexibility, resistive muscle strength, spasticity and cardiovascular fitness with hydrotherapy (209)(341)(365). The scientific rigour with which these studies were conducted varied greatly (341).

As well as the positive physical effects of aqua therapy, it provides a fun, motivating medium in which children can achieve activities they are possibly more anxious to attempt or unable to accomplish during land based therapy (209)(240).

Hippotherapy

Hippotherapy or therapeutic horse riding has become popular in recent years as it provides a means of addressing all levels of the ICF during treatment. The disabled child is placed on a horse lead by a trained instructor and generally supported by a therapist, however indicated (366). Therapy comprises different movements experienced by the individual due to different tasks the horse is taken through (366). As sessions progress, the child is expected to become more actively involved, incorporating coordinated goal-directed activities whilst on the horse (366).

Impairments of hypertonicity and postural dysfunction are addressed in that the rhythmic, repetitive gait of the horse is thought to influence tone as it mimics human pelvic movement during gait. Unexpected changes in direction and pace allow for the experience

of postural perturbations otherwise not experienced by a wheelchair bound child (27)(180)(366)(367). Movements of the horse in different directions stimulate muscle activation of either flexor/extensor or lateral trunk muscles, helping to develop muscle synergies not generally present in children with CP (367). The body warmth of the horse is thought to reduce spasticity and wide hip abduction necessary to straddle the horse can help to improve joint range of movement and muscle extensibility (171)(367). Activity limitation is addressed as children unable to independently ambulate now feel that they can. Social participation can take place during group riding classes and animal interaction assists with building motivation, self-confidence and self-esteem and provides a fun and meaningful therapeutic experience (22)(171)(366)(367).

Eleven children with spastic diplegic CP participated in a trial where they received weekly hippotherapy for 12 weeks. Statistically significant improvements in head control, reaching time and trunk coordination, post intervention, were found. No control group of children was included in the trial and thus it is difficult to conclude that improvements seen were not purely due to maturation or other external confounding variables (366). Despite studies having small and differing samples, Zadnikar and Kastrin concluded that hippotherapy results in statistically significant improvements in postural control and balance in children with CP, in their 2011 meta-analysis of eight studies on the intervention (171). Further studies have shown improvements in general gross motor function (332) and symmetrical activation of trunk muscles (66), following short periods of hippotherapy.

Vojta therapy

Vojta therapy is based on the assumption that postural control is not attained in children with CP due to the retention of primitive reflexes (27). Vojta suggested that “reflex locomotion” could be stimulated in children with CP in order to influence postural control mechanisms lacking in this population. Supposed “trigger zones” during movement are used to bring about reflexive rolling or crawling and later reflexive locomotion. It is said that repetition of these reflexive movements eventually leads to independent, active control of these activities (368). No scientific studies using Vojta therapy in the treatment of children with CP could be found to either negate or support the use of this intervention.

Conductive education (CE)

As with NDT, Conductive Education (CE) was conceptualised in the 1940s (27) for use in those with CNS disorders (296). Group intervention is carried out by trained "conductors" who facilitate participation in ADLs and motor activities by giving verbal instructions/cues. Obviously in order for these verbal instructions to be understood the participants require a certain level of cognitive functioning (27)(67). The focus is therefore on educational factors rather than a medical approach to treatment (369), as it is assumed by the founders that the primary problem in CP is one of learning/problem solving. In this sense, appropriate teaching is said to influence cortical functioning, thus improving physical functioning (369).

Peto, the creator of CE, has set up an institute for the training of CE conductors. They may be therapists, teachers or nurses (296). This conductor then takes the place of all of these roles/professions (317)(369). Working in a group may improve motivation (268). Group sessions generally take place five days a week for up to six hours a day for the duration of the school year and larger groups may require more than one conductor (296). Singing, rhyming and the use of specialised equipment for motor tasks are used to facilitate this process (296) (with a more 'hands-off' approach) , with the end goal being integration into mainstream educational systems (369). Assistive devices are discouraged to try to encourage more independent functioning (66). The day is structured and tasks are broken up to allow for a sense of achievement (317).

Very little scientific research exists to support the use of CE (26)(67)(317). A study on 19 children with CP concluded that functional improvements seen in the classroom during a 5-week CE trial generally had little or no carry over into other environments and no improvement over children receiving special education or intensive therapy were seen (296).

6.5.2 Medical treatment modalities

Medication

Medical interventions for CP are still based on trying to correct impairments and are mostly aimed at reducing spasticity. Baclofen is the most commonly used drug in the treatment of CP. Baclofen/Lioresal is a synthetic gamma-amino-butyric-acid (GABA) agonist which inhibits excitatory synaptic transmission by binding to pre- and post-synaptic GABA receptors, thus preventing neurotransmitter release (108)(138)(140)(370)(371). This oral antispasmodic does not cross the blood-brain barrier well and so cannot influence cortical causes of spasticity (138). Common side effects include drowsiness, sleep disturbances, confusion, ataxia, hypotonia and possible behavioural alterations (138)(140)(371). Dosage should be weaned down before cessation to prevent severe withdrawal symptoms (108)(140)(370).

Other medications used include Dantrolene (which reduces tone by reducing calcium release from the sarcoplasmic reticulum of muscle), Diazepam/other benzodiazepines (which inhibit pre-synaptic spinal cord and brainstem activity and act as an anti-anxielytic) and Tizanidine (which increases noradrenergic inhibition at spinal cord level) (93)(108)(138)(140)(371). These drugs therefore desensitise the nervous system to external stimuli that may increase spasticity (90). Most of these available medications are not well-tolerated due to quite severe possible side-effects of organ toxicity, bladder and bowel dysfunction, sleep disorders, sensitivity to light, impaired cognition and sedation (90) (140)(370)(371).

Phenol/alcohol nerve blocks

Phenol and alcohol nerve blocks have been used since the 1950s and offer up to six months of reduced hypertonicity in those with CP when administered by injection into the selected

muscle or nerve (140)(370). Injections work by degeneration of the axons causing denervation (93). Serious side effects of loss of sensation or pain, muscle necrosis and vascular problems have been associated with this treatment modality (93)(140) and the procedure requires a skilled practitioner able to administer anaesthetic and electrical stimulation to demarcate affected nerves (370). Dystonia is less responsive to oral medication than spastic type presentations of CP (372). Dopaminergic or anti-cholinergic drugs should be attempted in these cases (93).

Intrathecal Baclofen (ITB)

Due to the limitations oral Baclofen has in crossing the blood-brain barrier, if introduced intrathecally, toxicity can be avoided and almost 80 times the possible oral dosage can be found in the cerebrospinal fluid (138)(232)(370)(371). ITB should be considered in the management of severe spastic quadriplegia, dystonia or those individuals with CP who do not respond to other more conservative means (133)(371). If repetitive bolus doses of Baclofen have a positive effect on hypertonicity then one can be considered for the insertion of an intrathecal pump (138)(232). When inserted, the Baclofen reaches the spinal cord, substituting GABA in order to decrease spasticity by decreasing the release of neurotransmitters at spinal cord level (232)(372).

The pump is surgically inserted subcutaneously into the anterior abdominal wall with a catheter attached into the subarachnoid space intrathecally (138)(141)(370)(371). Baclofen is stored inside the pump and its release is computer-programmed based on each individual's need (86)(90), adding the benefit of possible titration of doses (141). In a 2000 review of studies based on ITB administration, 68% of trials demonstrated statistically significant improvements in spasticity, attainment of ADLs, pain and participation, as well as delays in the need for orthopaedic intervention (141). On the other hand, in their well-known review in the same year, the AACPDM found a lack of available evidence to support the use of ITB in spastic or dystonic CP (26).

The complications of ITB are common and include sedation, excess drooling, blurred vision, dizziness, seizures and more detrimental effects of lung and cardiac failure due to overdose, or infection/meningitis due to the surgical insertion procedure may occur (26)(86)(138)(141)(371). Hardware problems, such as system blockages or pump failure (in 5-10% of cases) may lead to symptoms of withdrawal (138). In 2006, Baclofen pumps were quoted as incurring a cost of $10 000, excluding the associated running costs, battery replacement (every eight years) and fees for the Baclofen itself (which needs replacing every three months) (138)(141)(371)(333).

Botulinum Toxin A (Btx-A)

Commonly referred to as Botox, this contemporary intervention for the treatment of spasticity is derived from a bacterium produced by Clostridium botulinum (106)(140)(370). Ultrasound imaging is generally used in order to guide injections into the affected muscles of those with CNS disorders, while under anaesthetic (106)(141). This neurotoxin works by permanently impeding the release of acetylcholine from the myoneural junction, thus ensuring neurotransmitters are unable to bring about muscle contraction in that particular area (in a sense 'paralysing' the injected muscle) (86)(106)(232)(370). The effects of Btx-A can be noted after approximately three days and appear to be self-limiting after a period of 3-6 months. This apparent regaining of muscle strength and tone is rather due to the body's development of new neuromuscular junctions and compensatory axonal sprouting (106)(138)(140)(232)(370).

Originally Btx-A was only administered into the gastrocnemius muscles of children with CP in order to correct toe-walking/equinus present in this population, thus attempting to modify gait and improve joint range of movement and energy expenditure (228)(232)(373). Today, Btx-A is generally administered at multiple levels in order to attain maximal functional outcomes (232)(374). It produces its greatest effect in muscles without fixed contractures (140), when used in conjunction with physiotherapy (140) and when administered to children between the ages of 1-6 years (370). In their 2009 review, Lukban,

Rosales and Dressler noted that Btx-A reduces the effects of hypertonicity more than study groups receiving placebo treatment or occupational therapy alone (373).

Unlike the oral and intrathecal medications available for the management of CP, studies have found very few side effects associated with its use (138). These include possible pain and bruising around the injection site directly after application, excessive weakness, incontinence or constipation, fatigue and nausea (141)(370)(373)(374). Coupled with the ease with which it can be administered, this lack of serious side effects has made Btx-A the widely accepted intervention that it is today (138)(232). The body does tend to build up a resistance to Btx-A with repetitive use, as antibodies attempt to fight off the bacterium. For this reason, the lowest effective dose should be administered each time and dosage should be worked out according to the individual's body weight (93)(370). On the negative side, Btx-A treatment is expensive and its beneficial effects are transient (375).

All of the above mentioned medicinal treatments list substantial weakness as a likely side effect (108)(140)(141)(333)(370)(371)(373)(374). It should be considered that children with CP may 'use' their lower limb spasticity or extensor thrust patterns in order to bear weight in an upright position (to compensate for general underlying muscle weakness). If this is the case, then caution should be exercised when contemplating such therapeutic interventions, as one should not take away independent function in order to improve impairments such as spasticity (138)(141)(370).

6.5.3 Surgical interventions

Surgical procedures used in the management of CP have typically been aimed at reducing spasticity (86)(106) and no other options really exist when associated deformities experienced by those with CP become fixed (300). Most popular has been a procedure called selective dorsal rhizotomy (SDR) which was refined in Cape Town in the 1980s (376)(377). It is a procedure where the dorsal rootlets are exposed surgically at cauda equina level, individually electrically stimulated and those producing the most abnormal electrical readings (i.e. innervating the muscles groups most disabled by the effects of spasticity) are then transected (106)(138)(376)(377)(378). Up to 50% of these rootlets can

be cut (87)(379). In this way input at the posterior roots is reduced, dampening the spinal reflex arc, in order to prevent excitation of motor neurons (138)(232). Side effects include incontinence, sexual dysfunction and sensory disturbances, with up to 24% of those undergoing the procedure experiencing transient urinary retention (26)(90)(138)(378)(379). This intervention may also increase the risk of spinal deformities in those children prone to these conditions (86)(138).

Older systematic reviews have shown inconclusive evidence to support the use of SDR as significant reductions in spasticity seen did not necessarily equate to functional improvements, and most studies were not able to convincingly establish whether positive effects observed were due to the surgical intervention or to the concomitant intensive physiotherapy that study participants received following SDR (90)(106)(232)(376). In 2009, Langerak et al demonstrated that one year following SDR significant reductions in hypertonicity achieved in 14 patients with CP were maintained (376). Similar results were found by Kondo et al (336). The selection criteria for rootlets to be cut as well as the number of rootlets to include in the procedure varies between surgeons and countries, making comparison between studies difficult (106)(377).

Another common procedure performed in this population is a tendon lengthening surgery. Muscles tightened due to spasticity or tendons already contractured are surgically released (common sites involve the Achilles tendons, hamstrings and hip flexors and adductors) (86)(133)(212). Tendon transfers may assist in stabilising a joint by compensating for the inherent weakness of distal muscle/tendon insertions (49)(138)(168). If this fails, arthrodesis of the ankle joint may be necessary (86). Major orthopaedic surgeries such as osteotomies may assist in correcting internal rotation of the femurs or tibial torsion, thus amending or preventing hip sublaxations/dislocations in more severely affected children with CP (86)(138). Spinal surgery is usually only indicated when deformities run the risk of severely exacerbating respiratory compromise (99) or impeding the ability of a child to sit (86).

Due to the changing nature of CP as a condition, any positive outcomes of the above mentioned surgeries are usually not permanent and surgeries often need redoing every few years. This can lead to a great deal of trauma, as the surgeries involved can be painful

and all carry long intensive rehabilitation periods following the procedures in order to gain the greatest benefits (232)(300). Due to this phenomenon, multilevel surgery has been popularised in recent years (86)(232). This has been termed single event multi-level surgery (SEMLS) where all (usually lower limb) impairments and/deformities are addressed at the same time (232). SEMLS is now considered the standard for orthopaedic management in CP in most developed countries (77)(351). Despite this, a 2012 systematic review of studies investigating the effect of SEMLS in children with CP found only low level quality studies and the acceptance of the positive outcomes observed was cautioned by the authors (351).

As with medicinal management, all surgical interventions have the unfortunate side effect of added weakness (86)(138)(157)(377)(378)(379), especially as the risk for 'overlengthening' of certain muscles exists (300)(82).

6.5.4 Complementary and alternative therapies

Due to the contradictory and often confusing evidence available to support more orthodox therapeutic interventions, or negative experiences when undergoing these treatments, families of children with CP often seek alternative methods of intervention (90)(131)(312).

Complementary and alternative medicine (CAM) modalities are often not held to the same rigorous level of review as more conventional techniques/treatments (66)(131)(217). No RCTs currently exist confirming the beneficial effects of any CAM interventions (90)(131). With this said, up to 50% of those with CNS disorders seek so-called fad treatments in a desperate attempt to sample all options (66)(312) and thus practitioners need to stay informed on emerging alternative treatments in order to adequately inform patients on the risks or benefits of such techniques (66)(217).

Hyperbaric oxygen

Hyperbaric oxygen chambers have been developed on the premise that exposing children with CP to 100% pressurised oxygen will stimulate areas of damage in the brain, exciting "dormant" neurons and decreasing inflammatory processes (27)(66)(312). Intervention is expensive and time consuming (with hour-long sessions, 5-6 times a week, for up to 40 treatments indicated) and severe side effects such as damage to auditory anatomical structures, seizures, pain and pneumothorax associated with exposure (27)(66)(312).

Stem cell therapy

Due to the brain's propensity for plastic changes during development, stem cell implantation has been introduced in an attempt to repair the cortical lesions present in CP (90). The mechanism by which improvements may take place is purely speculative at this point and ideas include that stem cells may differentiate into glial immune cells, may replace nerve cells and may aid regeneration of certain structures (312)(380). Possible adverse effects are seizures, ischaemic insults or infection introduced at the injection site but more rigorous investigations are needed to determine if further side effects exist (312) or whether use is indicated.

Therapeutic garments

Various individually sized and fitted garments exist (usually lycra-based) and are used based on the assumption that they can influence tone and proprioceptive input, provide postural support and reduce the risk of postural deformities (140)(169)(361). Depending on the properties of the fabric and the particular impairments of the child, the garment may provide necessary stability in certain regions and allow for mobility in others in order to compensate for asymmetries and muscle imbalances (381). Examples include the Adeli suit and Theratogs (140)(169). As these garments require full time use, compliance is usually

difficult to achieve and complaints have been made of discomfort, difficulty completing ADLs, chafe and trouble getting in and out of the suits (99)(381).

Vibration therapy

Vibration has been applied to various muscles in CP in order to simulate a stretch reflex thus allegedly correcting muscle imbalances (172). Whole body vibration during sport training has been purported to improve strength and increase circulation and the release of growth hormones following use (382)(383). A possible source of vibration is the Power Plate; a vibrating platform on which exercises can be performed (383).

Other

Other CAM treatments include the use of homeopathic or herbal medicines (90), art and music therapy (66), acupuncture (27)(66), cryotherapy (140), craniosacral therapy (66) and the Feldenkrais method (66). As mentioned in the introduction to this section, no convincing, if any, scientific evidence exists to support the use of any of the discussed CAM interventions in the treatment of CP.

6.6 Appendix VI

Application, scoring and validation of the GMFM

This therapist-administered test can take up to an hour to implement but this varies widely with familiarity with the tool, child functional ability and whether the 66 or 88 version of the measure is employed (103)(269)(274). The child should be assessed in a comfortable, spacious environment that can be kept constant between assessment intervals and should be barefoot during testing. Should the therapist wish to draw comparisons with orthotic use, these may be used on a second attempt of testing with the GMFM-88 after the completion of the entire scale barefoot (274)(324).

The original validation study for the GMFM-88 took place in Ontario, Canada and included 111 children of varying ages with CP, 25 children with traumatic brain injuries and another 34 TD children under the age of five. After reaching at least 70% agreement on criterion scores, 13 therapists agreed on the face validity of the measure and after repetitive assessing of the participants, reliability, responsiveness to change in gross motor function and validity were established (274).

As the GMFM-88 is an ordinal scale, interpretation of percentage scores can lead to much confusion and it is for this reason that Rasch analysis was applied to the measure in 2002 in an attempt to make the measure more user-friendly. As children do not linearly acquire motor milestones, later items in the Crawling dimension, for example, may actually be more difficult to complete than earlier items in the Standing dimension, etc. For this reason, the GMFM-66 was developed, streamlining the implementation of the measure and allowing for easier interpretation of results (65)(115)(274). The GMFM-66 also allows for a Not Tested (NT) score to be recorded, for those items not attempted due to time constraints or participant compliance, etc. in order to gain a more accurate overview of a child's capabilities (whereas on the GMFM-88 a zero would have been recorded for these items affecting the final measure score)(274).

While GMFM-88 scores can be summed to attain a total score, the GMFM-66 items need to be entered into the Gross Motor Ability Estimator (GMAE) programme in order to compute an interval level total score out of 100. This provides further information on the level of difficulty of the items achieved by the individual (101)(110)(125)(322).

6.7 Appendix VII

CHILD ASSENT FORM

TITLE OF THE RESEARCH PROJECT:

The effect of Therapeutic Electrical Stimulation on abdominal muscle strength and gross motor function in children with cerebral palsy: a randomised control trial

RESEARCHER'S NAME: Jessica Joffe

WHAT IS RESEARCH?

Research is something we do to find new information about how things and people work. We use research projects to increase our knowledge of illness or disabilities and how best to treat them.

WHAT IS THIS RESEARCH PROJECT ABOUT?

This project wants to see if using a machine called Therapeutic Electrical Stimulation (TES) can help to make your tummy muscles stronger. We also want to see if it helps you be able to do more things, to sit or stand more upright, and to balance better.
If you decide to take part a physio you do not know will test what you are able to do and how well you can do these things. She will ask you to try different tasks and will write down how well you do them. You will carry on with your physio at school like normal.

Half of you taking part will not change your physio at all. The other half will use the TES machine for 6 weeks during your normal physio time. Your physio will put two little stickers over your tummy muscles. When the machine is turned on it will cause a little prickly feeling. This machine is trying to make your tummy muscles contract or work better. After 6 weeks the physio from before will come to do all the same tests again to see if the machine made any difference.

WHY HAVE I BEEN CHOSEN TO TAKE PART?

You have been invited to take part because you go to the Hope School, you are between 5 and 18 years old and because you have cerebral palsy.

WHO IS DOING THE RESEARCH?

I am Jessica Joffe, one of the physiotherapists working at the Hope School. I am also studying to get my Masters degree. This research project will help me to complete my studies.

CAN ANYTHING BAD HAPPEN TO ME?

The only thing that may happen when you use the TES machine is your skin may go a little red under the stickers. This should go away after your physio session. If it does not go away or you do not like the feeling of the prickles of the machine you can decide to stop taking part. If you hurt yourself by accident your physio will help you like normal and we will call your parents to tell them about it.

CAN ANYTHING GOOD HAPPEN TO ME?

Hopefully your tummy muscles will get stronger and you will be able to do a little bit more and balance more easily. If the project has good results, other children at the school may also be able to use the TES machine during their physio sessions.

WILL ANYONE KNOW I AM IN THE PROJECT?

Only the physiotherapists at your school and my teacher at the university will know that you were in the study. I will not use your name anywhere and no one else will know you took part.

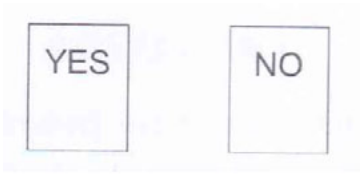
WHO CAN I ASK QUESTIONS OR TALK TO ABOUT THE STUDY?

You can ask me Jessica (083 951 7073) or any of your physiotherapists at your school any questions you may have about the project. You can also talk to my teacher Professor Jelsma if you would like to (021 406 6595).

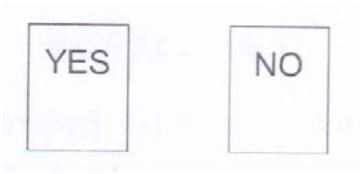
WHAT IF I DON'T WANT TO DO THIS

You do not have to take part if you don't want to. If you choose to take part, you can also change your mind later. This will not get you into any trouble and it won't affect your physio at school.

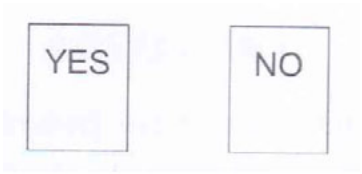
Do you understand what the project is about?



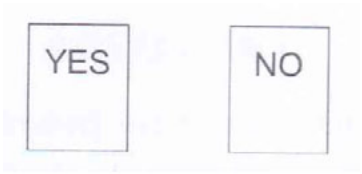
Are you willing to take part in the project?



Has the researcher answered all your questions?



Do you understand you are allowed to pull out if you want to?



_____ _____

Signature Child Date

6.8 Appendix VIII

PARENT/GUARDIAN CONSENT FORM AND INFORMATION SHEET

PROJECT TITLE:

The effect of Therapeutic Electrical Stimulation on abdominal muscle strength and gross motor function in children with cerebral palsy: a randomised control trial

PRINCIPAL INVESTIGATOR: Jessica Joffe

You and your child are being invited to take part in the research I am conducting for my Master's in Physiotherapy degree through the University of Cape Town (UCT). I am a registered physio working at the Hope School. Please take the time to read through this letter and do not hesitate to contact me with any questions you may have.

Participation in the study is voluntary. If you do not choose to take part, your child's future treatment at the school will not be affected negatively in any way. If you decide you would like your child to take part but later change your mind, you are able to withdraw from the study at any time.

This study has been approved by the University of Cape Town Faculty of Health Sciences Human Research Ethics Committee, the Gauteng Department of Education and the principal of the Hope School. The study will be carried out following strict ethical guidelines.

WHAT IS THE STUDY ABOUT?

This study will look at whether treatment using a machine called Therapeutic Electrical Stimulation (TES), together with your child's usual physiotherapy treatment; will cause improvements in tummy muscle strength, balance and physical function. Studies have shown that TES may improve balance, posture and muscle strength in some children with cerebral palsy.

WHY YOUR CHILD HAS BEEN CHOSEN

Your child has been chosen, as they are between age 5 and 18, have cerebral palsy and attend the Hope School.

WHAT YOU NEED TO DO

Your child will only be able to take part if you sign the consent form attached. If included, children will be split into two groups. Both groups will continue to receive physio at school like normal. The second group will also receive TES treatment over their tummies during their usual physio time. If chosen to be in this group, your child's physio will place small pads over their tummy for 20 minutes while they continue with their normal physio session, once a week for 6 weeks.

All assessments and treatments will take place during the normal school day and your child will not be expected to miss any more class than they usually do to attend therapy.

All assessments performed before and after the study will be based on observations made by a trained physio and will take about an hour long. All of these tests require your child to try different physical activities and none are painful or scary. They will also be asked to fill in a questionnaire about how they feel about certain aspects of their lives and what they thought of the TES treatment.

WILL YOUR CHILD BENEFIT FROM TAKING PART?

If your child is in the TES group and positive effects are found then your child will benefit straight away. These results may help us better plan your child's future therapy as well as later benefit the children who are chosen to be in the group that do not get TES treatment and other children with cerebral palsy at the school who could then also receive FES treatment after the study takes place. Taking part will not cost you anything and you will not be paid anything to take part.

ARE THERE ANY RISKS?

The only known side effect of using TES is a rare skin reaction to the pads (a mild redness after treatment is normal). If your child complains of persisting redness or discomfort, they are allowed to withdraw from the study. Any accidents that may happen during assessment or treatment will be treated by trained staff in the same way as at any other time at the school.

WILL MY CHILD REMAIN ANONYMOUS?

All information taken will be kept in a secure and private file. Only the investigator and her supervisor will have access to this information and your child's name will not be used in any discussions or publications about the research.

WHAT HAPPENS IF I HAVE ANY QUESTIONS?

If you have any questions or would like to know more about this study, please do not hesitate to call or email the researcher (Jessica Joffe) at:

Phone: 083 9517073/ (011) 646 6130
e-mail: jessicajoffe@hotmail.com

You are also welcome to contact the University of Cape Town Faculty of Health Sciences Human Research Ethics Committee representative for further information.

Marc Blockman
Research Ethics Committee
Faculty of Health Sciences
E 52-23 Old Main Building
Groote Schuur Hospital
Observatory 7925
Tel: +27- 21- 4066492
Fax: +27- 21- 4066411

DECLARATION OF CONSENT

I _____ (name) hereby agree for my child _____ (name) to take part in the study entitled "The effect of Therapeutic Electrical Stimulation on abdominal muscle strength and gross motor function in children with cerebral palsy: a randomised control trial".

I declare that:

I have read or had someone read to me the information about the study and the consent form and it is written in a language that I can understand.

I have had a chance to ask any of the questions I may have and they were answered to my satisfaction.

I, nor my child, have been pressured into taking part and understand participation is voluntary.

I am able to withdraw my child from the study at any time should I want to and this will not affect my child's future treatment at the school in any way.

Signed at _____ (place) on _____ (date).

_____ _____

Signature of parent/legal guardian Signature of a witness

6.9 Appendix IX

Participant no.	
Name	
Parent name	
Parent contact no.	
Grade	
Date of birth	
Age	
Sex	
Race	
Hostel/day scholar	
Home address	
Home language	
CP motor disorder	
CP anatomical distribution	
Other impairments	
Gestational age	
CP cause	
GMFCS level	
Treating physiotherapist	
Other therapies received	
Previous surgery	

6.10 Appendix X

GROSS MOTOR FUNCTION MEASURE (GMFM)
SCORE SHEET (GMFM-88 and GMFM-66 scoring)
Version 1.0

Child's Name: _____ ID #: _____

Assessment date: _____ year / month /day

Date of birth: _____ year / month /day

Chronological age: _____ years/months

GMFCS Level [1]

☐ I ☐ II ☐ III ☐ IV ☐ V

Testing Conditions (eg, room, clothing, time, others present)

Evaluator's Name: _____

_____

_____

_____

The GMFM is a standardized observational instrument designed and validated to measure change in gross motor function over time in children with cerebral palsy. The scoring key is meant to be a general guideline. However, most of the items have specific descriptors for each score. It is imperative that the guidelines contained in the manual be used for scoring each item.

SCORING KEY
0 = does not initiate
1 = initiates
2 = partially completes
3 = completes
NT = Not tested [used for the GMAE scoring*]

It is now important to differentiate a true score of "0" (child does not initiate) from an item which is Not Tested (NT) if you are interested in using the GMFM-66 Ability Estimator Software.

*The GMFM-66 Gross Motor Ability Estimator (GMAE) software is available with the GMFM manual (2002). The advantage of the software is the conversion of the ordinal scale into an interval scale. This will allow for a more accurate estimate of the child's ability and provide a measure that is equally responsive to change across the spectrum of ability levels. Items that are used in the calculation of the GMFM-66 score are shaded and identified with an asterisk (*). The GMFM-66 is only valid for use with children who have cerebral palsy.

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[1] GMFCS level is a rating of severity of motor function. Definitions are found in Appendix I of the GMFM manual (2002).

Check (√) the appropriate score: if an item is not tested (NT), circle the item number in the right column

	Item	A: LYING & ROLLING	SCORE				NT
	1.	SUP, HEAD IN MIDLINE: TURNS HEAD WITH EXTREMITIES SYMMETRICAL	0 ☐	1 ☐	2 ☐	3 ☐	1.
*	2.	SUP: BRINGS HANDS TO MIDLINE, FINGERS ONE WITH THE OTHER	0 ☐	1 ☐	2 ☐	3 ☐	2.
	3.	SUP: LIFTS HEAD 45°	0 ☐	1 ☐	2 ☐	3 ☐	3.
	4.	SUP: FLEXES R HIP AND KNEE THROUGH FULL RANGE	0 ☐	1 ☐	2 ☐	3 ☐	4.
	5.	SUP: FLEXES L HIP AND KNEE THROUGH FULL RANGE	0 ☐	1 ☐	2 ☐	3 ☐	5.
*	6.	SUP: REACHES OUT WITH R ARM, HAND CROSSES MIDLINE TOWARD TOY	0 ☐	1 ☐	2 ☐	3 ☐	6.
*	7.	SUP: REACHES OUT WITH L ARM, HAND CROSSES MIDLINE TOWARD TOY	0 ☐	1 ☐	2 ☐	3 ☐	7.
	8.	SUP: ROLLS TO PR OVER R SIDE	0 ☐	1 ☐	2 ☐	3 ☐	8.
	9.	SUP: ROLLS TO PR OVER L SIDE	0 ☐	1 ☐	2 ☐	3 ☐	9.
*	10.	PR: LIFTS HEAD UPRIGHT	0 ☐	1 ☐	2 ☐	3 ☐	10.
	11.	PR ON FOREARMS: LIFTS HEAD UPRIGHT, ELBOWS EXT., CHEST RAISED	0 ☐	1 ☐	2 ☐	3 ☐	11.
	12.	PR ON FOREARMS: WEIGHT ON R FOREARM, FULLY EXTENDS OPPOSITE ARM FORWARD	0 ☐	1 ☐	2 ☐	3 ☐	12.
	13.	PR ON FOREARMS: WEIGHT ON L FOREARM, FULLY EXTENDS OPPOSITE ARM FORWARD	0 ☐	1 ☐	2 ☐	3 ☐	13.
	14.	PR: ROLLS TO SUP OVER R SIDE	0 ☐	1 ☐	2 ☐	3 ☐	14.
	15.	PR: ROLLS TO SUP OVER L SIDE	0 ☐	1 ☐	2 ☐	3 ☐	15.
	16.	PR: PIVOTS TO R 90° USING EXTREMITIES	0 ☐	1 ☐	2 ☐	3 ☐	16.
	17.	PR: PIVOTS TO L 90° USING EXTREMITIES	0 ☐	1 ☐	2 ☐	3 ☐	17.
		TOTAL DIMENSION A	☐				

	Item	B: SITTING	SCORE				NT
*	18.	SUP, HANDS GRASPED BY EXAMINER: PULLS SELF TO SITTING WITH HEAD CONTROL	0 ☐	1 ☐	2 ☐	3 ☐	18.
	19.	SUP: ROLLS TO R SIDE, ATTAINS SITTING	0 ☐	1 ☐	2 ☐	3 ☐	19.
	20.	SUP: ROLLS TO L SIDE, ATTAINS SITTING	0 ☐	1 ☐	2 ☐	3 ☐	20.
*	21.	SIT ON MAT, SUPPORTED AT THORAX BY THERAPIST: LIFTS HEAD UPRIGHT, MAINTAINS 3 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	21.
*	22.	SIT ON MAT, SUPPORTED AT THORAX BY THERAPIST: LIFTS HEAD MIDLINE, MAINTAINS 10 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	22.
*	23.	SIT ON MAT, ARM(S) PROPPING: MAINTAINS, 5 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	23.
*	24.	SIT ON MAT: MAINTAINS, ARMS FREE, 3 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	24.
*	25.	SIT ON MAT WITH SMALL TOY IN FRONT: LEANS FORWARD, TOUCHES TOY, RE-ERECTS WITHOUT ARM PROPPING	0 ☐	1 ☐	2 ☐	3 ☐	25.
*	26.	SIT ON MAT: TOUCHES TOY PLACED 45° BEHIND CHILD'S R SIDE, RETURNS TO START	0 ☐	1 ☐	2 ☐	3 ☐	26.
*	27.	SIT ON MAT: TOUCHES TOY PLACED 45° BEHIND CHILD'S L SIDE, RETURNS TO START	0 ☐	1 ☐	2 ☐	3 ☐	27.
	28.	R SIDE SIT: MAINTAINS, ARMS FREE, 5 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	28.
	29.	L SIDE SIT: MAINTAINS, ARMS FREE, 5 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	29.
*	30.	SIT ON MAT: LOWERS TO PR WITH CONTROL	0 ☐	1 ☐	2 ☐	3 ☐	30.
*	31.	SIT ON MAT WITH FEET IN FRONT: ATTAINS 4 POINT OVER R SIDE	0 ☐	1 ☐	2 ☐	3 ☐	31.
*	32.	SIT ON MAT WITH FEET IN FRONT: ATTAINS 4 POINT OVER L SIDE	0 ☐	1 ☐	2 ☐	3 ☐	32.
	33.	SIT ON MAT: PIVOTS 90°, WITHOUT ARMS ASSISTING	0 ☐	1 ☐	2 ☐	3 ☐	33.
*	34.	SIT ON BENCH: MAINTAINS, ARMS AND FEET FREE, 10 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	34.
*	35.	STD: ATTAINS SIT ON SMALL BENCH	0 ☐	1 ☐	2 ☐	3 ☐	35.
*	36.	ON THE FLOOR: ATTAINS SIT ON SMALL BENCH	0 ☐	1 ☐	2 ☐	3 ☐	36.
*	37.	ON THE FLOOR: ATTAINS SIT ON LARGE BENCH	0 ☐	1 ☐	2 ☐	3 ☐	37.
		TOTAL DIMENSION B	☐				

Item	C: CRAWLING & KNEELING	SCORE	NT
38.	PR: CREEPS FORWARD 1.8m (6')	0 ☐ 1 ☐ 2 ☐ 3 ☐	38.
* 39.	4 POINT: MAINTAINS, WEIGHT ON HANDS AND KNEES, 10 SECONDS	0 ☐ 1 ☐ 2 ☐ 3 ☐	39.
* 40.	4 POINT: ATTAINS SIT ARMS FREE	0 ☐ 1 ☐ 2 ☐ 3 ☐	40.
* 41.	PR: ATTAINS 4 POINT, WEIGHT ON HANDS AND KNEES	0 ☐ 1 ☐ 2 ☐ 3 ☐	41.
* 42.	4 POINT: REACHES FORWARD WITH R ARM, HAND ABOVE SHOULDER LEVEL	0 ☐ 1 ☐ 2 ☐ 3 ☐	42.
* 43.	4 POINT: REACHES FORWARD WITH L ARM, HAND ABOVE SHOULDER LEVEL	0 ☐ 1 ☐ 2 ☐ 3 ☐	43.
* 44.	4 POINT: CRAWLS OR HITCHES FORWARD 1.8m (6')	0 ☐ 1 ☐ 2 ☐ 3 ☐	44.
* 45.	4 POINT: CRAWLS RECIPROCALLY FORWARD 1.8m (6')	0 ☐ 1 ☐ 2 ☐ 3 ☐	45.
* 46.	4 POINT: CRAWLS UP 4 STEPS ON HANDS AND KNEES/FEET	0 ☐ 1 ☐ 2 ☐ 3 ☐	46.
47.	4 POINT: CRAWLS BACKWARDS DOWN 4 STEPS ON HANDS AND KNEES/FEET	0 ☐ 1 ☐ 2 ☐ 3 ☐	47.
* 48.	SIT ON MAT: ATTAINS HIGH KN USING ARMS, MAINTAINS, ARMS FREE, 10 SECONDS	0 ☐ 1 ☐ 2 ☐ 3 ☐	48.
49.	HIGH KN: ATTAINS HALF KN ON R KNEE USING ARMS, MAINTAINS, ARMS FREE, 10 SECONDS	0 ☐ 1 ☐ 2 ☐ 3 ☐	49.
50.	HIGH KN: ATTAINS HALF KN ON L KNEE USING ARMS, MAINTAINS, ARMS FREE, 10 SECONDS	0 ☐ 1 ☐ 2 ☐ 3 ☐	50.
* 51.	HIGH KN: KN WALKS FORWARD 10 STEPS, ARMS FREE	0 ☐ 1 ☐ 2 ☐ 3 ☐	51.
	TOTAL DIMENSION C	☐	

Item	D: STANDING	SCORE	NT
* 52.	ON THE FLOOR: PULLS TO STD AT LARGE BENCH	0 ☐ 1 ☐ 2 ☐ 3 ☐	52.
* 53.	STD: MAINTAINS, ARMS FREE, 3 SECONDS	0 ☐ 1 ☐ 2 ☐ 3 ☐	53.
* 54.	STD: HOLDING ON TO LARGE BENCH WITH ONE HAND, LIFTS R FOOT, 3 SECONDS	0 ☐ 1 ☐ 2 ☐ 3 ☐	54.
* 55.	STD: HOLDING ON TO LARGE BENCH WITH ONE HAND, LIFTS L FOOT, 3 SECONDS	0 ☐ 1 ☐ 2 ☐ 3 ☐	55.
* 56.	STD: MAINTAINS, ARMS FREE, 20 SECONDS	0 ☐ 1 ☐ 2 ☐ 3 ☐	56.
* 57.	STD: LIFTS L FOOT, ARMS FREE, 10 SECONDS	0 ☐ 1 ☐ 2 ☐ 3 ☐	57.
* 58.	STD: LIFTS R FOOT, ARMS FREE, 10 SECONDS	0 ☐ 1 ☐ 2 ☐ 3 ☐	58.
* 59.	SIT ON SMALL BENCH: ATTAINS STD WITHOUT USING ARMS	0 ☐ 1 ☐ 2 ☐ 3 ☐	59.
* 60.	HIGH KN: ATTAINS STD THROUGH HALF KN ON R KNEE, WITHOUT USING ARMS	0 ☐ 1 ☐ 2 ☐ 3 ☐	60.
* 61.	HIGH KN: ATTAINS STD THROUGH HALF KN ON L KNEE, WITHOUT USING ARMS	0 ☐ 1 ☐ 2 ☐ 3 ☐	61.
* 62.	STD: LOWERS TO SIT ON FLOOR WITH CONTROL, ARMS FREE	0 ☐ 1 ☐ 2 ☐ 3 ☐	62.
* 63.	STD: ATTAINS SQUAT, ARMS FREE	0 ☐ 1 ☐ 2 ☐ 3 ☐	63.
* 64.	STD: PICKS UP OBJECT FROM FLOOR, ARMS FREE, RETURNS TO STAND	0 ☐ 1 ☐ 2 ☐ 3 ☐	64.
	TOTAL DIMENSION D	☐	

Item		E: WALKING, RUNNING & JUMPING	SCORE				NT
*	65.	STD, 2 HANDS ON LARGE BENCH: CRUISES 5 STEPS TO R	0 ☐	1 ☐	2 ☐	3 ☐	65.
*	66.	STD, 2 HANDS ON LARGE BENCH: CRUISES 5 STEPS TO L	0 ☐	1 ☐	2 ☐	3 ☐	66.
*	67.	STD, 2 HANDS HELD: WALKS FORWARD 10 STEPS	0 ☐	1 ☐	2 ☐	3 ☐	67.
*	68.	STD, 1 HAND HELD: WALKS FORWARD 10 STEPS	0 ☐	1 ☐	2 ☐	3 ☐	68.
*	69.	STD: WALKS FORWARD 10 STEPS	0 ☐	1 ☐	2 ☐	3 ☐	69.
*	70.	STD: WALKS FORWARD 10 STEPS, STOPS, TURNS 180°, RETURNS	0 ☐	1 ☐	2 ☐	3 ☐	70.
*	71.	STD: WALKS BACKWARD 10 STEPS	0 ☐	1 ☐	2 ☐	3 ☐	71.
*	72.	STD: WALKS FORWARD 10 STEPS, CARRYING A LARGE OBJECT WITH 2 HANDS	0 ☐	1 ☐	2 ☐	3 ☐	72.
*	73.	STD: WALKS FORWARD 10 CONSECUTIVE STEPS BETWEEN PARALLEL LINES 20cm (8") APART	0 ☐	1 ☐	2 ☐	3 ☐	73.
*	74.	STD: WALKS FORWARD 10 CONSECUTIVE STEPS ON A STRAIGHT LINE 2cm (3/4") WIDE	0 ☐	1 ☐	2 ☐	3 ☐	74.
*	75.	STD: STEPS OVER STICK AT KNEE LEVEL, R FOOT LEADING	0 ☐	1 ☐	2 ☐	3 ☐	75.
*	76.	STD: STEPS OVER STICK AT KNEE LEVEL, L FOOT LEADING	0 ☐	1 ☐	2 ☐	3 ☐	76.
*	77.	STD: RUNS 4.5m (15'), STOPS & RETURNS	0 ☐	1 ☐	2 ☐	3 ☐	77.
*	78.	STD: KICKS BALL WITH R FOOT	0 ☐	1 ☐	2 ☐	3 ☐	78.
*	79.	STD: KICKS BALL WITH L FOOT	0 ☐	1 ☐	2 ☐	3 ☐	79.
*	80.	STD: JUMPS 30cm (12") HIGH, BOTH FEET SIMULTANEOUSLY	0 ☐	1 ☐	2 ☐	3 ☐	80.
*	81.	STD: JUMPS FORWARD 30 cm (12"), BOTH FEET SIMULTANEOUSLY	0 ☐	1 ☐	2 ☐	3 ☐	81.
*	82.	STD ON R FOOT: HOPS ON R FOOT 10 TIMES WITHIN A 60cm (24") CIRCLE	0 ☐	1 ☐	2 ☐	3 ☐	82.
*	83.	STD ON L FOOT: HOPS ON L FOOT 10 TIMES WITHIN A 60cm (24") CIRCLE	0 ☐	1 ☐	2 ☐	3 ☐	83.
*	84.	STD, HOLDING 1 RAIL: WALKS UP 4 STEPS, HOLDING 1 RAIL, ALTERNATING FEET	0 ☐	1 ☐	2 ☐	3 ☐	84.
*	85.	STD, HOLDING 1 RAIL: WALKS DOWN 4 STEPS, HOLDING 1 RAIL, ALTERNATING FEET	0 ☐	1 ☐	2 ☐	3 ☐	85.
*	86.	STD: WALKS UP 4 STEPS, ALTERNATING FEET	0 ☐	1 ☐	2 ☐	3 ☐	86.
*	87.	STD: WALKS DOWN 4 STEPS, ALTERNATING FEET	0 ☐	1 ☐	2 ☐	3 ☐	87.
*	88.	STD ON 15cm (6") STEP: JUMPS OFF, BOTH FEET SIMULTANEOUSLY	0 ☐	1 ☐	2 ☐	3 ☐	88.

TOTAL DIMENSION E ☐

Was this assessment indicative of this child's "regular" performance? YES ☐ NO ☐

COMMENTS:

GMFM RAW SUMMARY SCORE

DIMENSION	CALCULATION OF DIMENSION % SCORES	GOAL AREA (indicated with ✓ check)
A. Lying & Rolling	$\frac{\text{Total Dimension A}}{51} = \frac{\quad}{51} \times 100 = \quad\%$	A. ☐
B. Sitting	$\frac{\text{Total Dimension B}}{60} = \frac{\quad}{60} \times 100 = \quad\%$	B. ☐
C. Crawling & Kneeling	$\frac{\text{Total Dimension C}}{42} = \frac{\quad}{42} \times 100 = \quad\%$	C. ☐
D. Standing	$\frac{\text{Total Dimension D}}{39} = \frac{\quad}{39} \times 100 = \quad\%$	D. ☐
E. Walking, Running & Jumping	$\frac{\text{Total Dimension E}}{72} = \frac{\quad}{72} \times 100 = \quad\%$	E. ☐

TOTAL SCORE = $\frac{\%A + \%B + \%C + \%D + \%E}{\text{Total \# of Dimensions}}$

$= \frac{\quad + \quad + \quad + \quad + \quad}{5} = \frac{\quad}{5} = \quad\%$

GOAL TOTAL SCORE = $\frac{\text{Sum of \% scores for each dimension identified as a goal area}}{\text{\# of Goal areas}}$

$= \quad = \quad\%$

GMFM-66 Gross Motor Ability Estimator Score [1]

GMFM-66 Score = _____ _____ to _____ 95% Confidence Intervals

previous GMFM-66 Score = _____ _____ to _____ 95% Confidence Intervals

change in GMFM-66 = _____

[1] from the Gross Motor Ability Estimator (GMAE) Software

TESTING WITH AIDS/ORTHOSES

Indicate below with a check (✓) which aid/orthosis was used and what dimension it was first applied. (There may be more than one).

AID		DIMENSION	ORTHOSIS		DIMENSION
Rollator/Pusher	☐	____	Hip Control	☐	____
Walker	☐	____	Knee Control	☐	____
H Frame Crutches	☐	____	Ankle-Foot Control	☐	____
Crutches	☐	____	Foot Control	☐	____
Quad Cane	☐	____	Shoes	☐	____
Cane	☐	____	None	☐	____
None	☐	____	Other	☐	____
Other	☐	____			

_____ (please specify)

_____ (please specify)

RAW SUMMARY SCORE USING AIDS/ORTHOSES

DIMENSION	CALCULATION OF DIMENSION % SCORES	GOAL AREA (indicated with ✓ check)
F. Lying & Rolling	$\frac{\text{Total Dimension A}}{51} = \frac{\quad}{51} \times 100 = \quad\%$	A. ☐
G. Sitting	$\frac{\text{Total Dimension B}}{60} = \frac{\quad}{60} \times 100 = \quad\%$	B. ☐
H. Crawling & Kneeling	$\frac{\text{Total Dimension C}}{42} = \frac{\quad}{42} \times 100 = \quad\%$	C. ☐
I. Standing	$\frac{\text{Total Dimension D}}{39} = \frac{\quad}{39} \times 100 = \quad\%$	D. ☐
J. Walking, Running & Jumping	$\frac{\text{Total Dimension E}}{72} = \frac{\quad}{72} \times 100 = \quad\%$	E. ☐

TOTAL SCORE = $\frac{\%A + \%B + \%C + \%D + \%E}{\text{Total \# of Dimensions}}$

$= \frac{\quad + \quad + \quad + \quad + \quad}{5} = \frac{\quad}{5} = \quad\%$

GOAL TOTAL SCORE = $\frac{\text{Sum of \% scores for each dimension identified as a goal area}}{\text{\# of Goal areas}}$

$= \quad = \quad\%$

GMFM-66 Gross Motor Ability Estimator Score [1]

GMFM-66 Score = _____ _____ to _____ 95% Confidence Intervals

previous GMFM-66 Score = _____ _____ to _____ 95% Confidence Intervals

change in GMFM-66 = _____

[1] from the Gross Motor Ability Estimator (GMAE) Software

6.11 Appendix XI

APPENDIX A: ScoreSheet Identification Number: __

Pediatric Reach Test

Child's name: _____ D.O.B.: _____

Height (supine or standing): _____ cms Foot length (heel to big toe): _____ cms

Therapist: _____ Date of Assessment: _____

Sitting on a Bench—*Can the child sit independently for 15 seconds?*

Please indicate the units of all distances.	Start	End	Difference
1. Reaching forward in sitting. Instruction: Lift arm forward to 90°. Make a fist and reach forward as far as you can.			
2. Reaching to the right in sitting. Instruction: Lift arm to the right to 90°. Make a fist and reach out as far as you can.			
3. Reaching to the left in sitting. Instruction: Lift arm to the left to 90°. Make a fist and reach out as far as you can.			

Standing—*Can the child stand independently with or without gait aides for 15 seconds?*

Please indicate the units of all distances.	Start	End	Difference
4. Reaching forward in standing. Instruction: Lift arm forward to 90°. Make a fist and reach forward as far as you can.			
5. Reaching to the right in standing. Instruction: Lift arm to the right to 90°. Make a fist and reach out as far as you can.			
6. Reaching to the left in standing. Instruction: Lift arm to the left to 90°. Make a fist and reach out as far as you can.			
		Total (sum)	

APPENDIX B

Testing Protocol: Pediatric Reach Test

Equipment:

a measuring tape (to measure height and foot length)
a flexible, retractable measuring tape to measure distance reached (with loop to secure to child's finger)
a variety of wooden benches of different heights (or an adjustable bench) (child's hips and knees should each be at 90 degrees when sitting)

Time Required: The Pediatric Reach Test can be administered to cooperative and minimally involved children within 15 minutes. More time will be required to administer the measure to children with more severe involvement and those requiring motivational prompts.

Before testing, ensure that the child is wearing his or her regular footwear (i.e., orthoses—if used, socks, and shoes) for both the sitting and standing sections and that the child's regular mobility aide is used for the standing section. The therapist will demonstrate the task to the child and then the child will have one practice trial and one test trial (as per Niznick et al[14]). Children will be asked to "sit up tall" at the beginning of the test trial for each item. The initial and final positions will be held for three seconds each. Some children might need a reminder to reach as far as they can before starting the three-second count. Repeat the trial if the child either touches a wall or the therapist or takes a step.

Sitting: Starting position: have the child sit on a surface (with no back or sides) with his/her feet flat on the floor, hips in neutral abduction/adduction, arms resting on

the lap. Keep the sitting surface constant for each testing occasion. Child should not externally stabilize otherwise using the arms and/or legs. Be sure to spot appropriately for safety. Some children with severe physical involvement might need two people present (one to administer and one to spot).

First, ask the child to sit with hands in lap for 15 seconds. If a child can sit independently for 15 seconds, administer items 1, 2, and 3. Start by putting the loop at the end of the tape around the middle finger of the child's dominant hand.

Item 1. Ask the child to "sit up tall." Then, with the tape secured on the finger, ask the child position his/her shoulder at 90 degrees of forward flexion with the elbow extended and the wrist in neutral (or as close to this position as possible). Position yourself stably behind the child and take an initial reading from the tape after the child has held the position for three seconds. Ask the child to reach as far forward as he/she can (toward a motivating object), and to hold the end position for three seconds. Measure the distance reached as designated by the difference between the initial and final positions.

Item 2. Put the loop around the middle finger of the child's distal right upper extremity. Ask the child to position his/her left shoulder at 90 degrees of abduction, with the elbow extended and the wrist in neutral (or as close to position as possible). Position yourself stably behind the child and take an initial reading from the tape after the child has held the position for three seconds. Ask the child to reach out to the right as far as he/she can, and to hold the end position for three seconds. Measure the distance reached as designated by the difference between the initial and final positions.

Item 3. Repeat item 2 on the left side.

Standing: Starting position. Have the child stand as they normally do with regular footwear and gait aide. To maintain a constant position for the reaching tests, attach a sheet of paper (or Bristol board, as necessary) to the floor with masking tape, and trace the child's foot position and also the gait aide contact points. This foot tracing should be used again for each of the interrater trials, as well as the retest trial (after the session, the foot length will be measured–perpendicular distance from the heel to the big toe). Safety concerns should be addressed as required.

First, ask the child to stand for 15 seconds. If a child can stand independently for 15 seconds, administer items 4, 5, and 6. Start by putting the loop of the tape around the middle finger on the child's dominant hand.

Item 4. Ask the child to position his/her shoulder at 90 degrees of forward flexion, with the elbow fully extended and the wrist in neutral (or as close to this position as possible). Position yourself stably behind the child and take an initial reading from the tape after the child has held the position for three seconds. Ask the child to reach as far forward as he/she can (toward a motivating object), and to hold the end position for three seconds. A child may lift a foot as long as it is replaced "approximately" over the traced foot print and the child has maintained balance throughout the reach (ie, not fallen or taken a step or relied on the therapist or a wall, etc). Loss of balance requires a retrial. Measure the distance reached as designated by the difference between the initial and final positions. For this item, it is not important whether movement is at the ankle or hip or both.

Item 5. Put the loop of the tape around the middle finger on the right hand. Ask the child to position his/her left shoulder at 90 degrees of abduction, with the elbow extended fully and the wrist in neutral (or as close to this position as possible). Position yourself stably behind the child and take an initial reading from the tape after the child has held the position for three seconds. Ask the child to reach as far to the right as he/she can (toward a motivating object), and to hold the end position for three seconds. A child may lift a foot as long as it is replaced "approximately" over the traced foot print and the child has maintained balance throughout the reach (ie, not fallen or taken a step or relied on the therapist or a wall, etc). Loss of balance requires a retrial. Measure the distance reached as designated by the difference between the initial and final positions.

Item 6. Repeat item 5 on the left side.

Note on motivation: The need for motivational prompts will vary among children, and is related to age and attention span, among other things. It might be useful to know a child's interests and/or favorite toys or activities when administering this measure to young children (for example, finger puppets might be motivating for young children).

Recent research with the Functional Reach Test in standing used on children suggests statistically controlling for the base of support (through measures of foot length and distance between the feet in stance) and height of the child[26] if making either intersubject or intrasubject inferences; therefore, these variables were added to the score sheet to provide this option.

Similar to a strategy described by Westcott et al.,[27] a total score is obtained by summing the interval data (score in centimeters).

6.12 Appendix XII

Timed "Up and Go"*

Directions

The timed "Up and Go" test measures, in seconds, the time taken by an individual to stand up from a standard arm chair (approximate seat height of 46 cm [18in], arm height 65 cm [25.6 in]), walk a distance of 3 meters (118 inches, approximately 10 feet), turn, walk back to the chair, and sit down. The subject wears their regular footwear and uses their customary walking aid (none, cane, walker). No physical assistance is given. They start with their back against the chair, their arms resting on the armrests, and their walking aid at hand. They are instructed that, on the word "go" they are to get up and walk at a comfortable and safe pace to a line on the floor 3 meters away, turn, return to the chair and sit down again. The subject walks through the test once before being timed in order to become familiar with the test. Either a stopwatch or a wristwatch with a second hand can be used to time the trial.

Instructions to the patient

"When I say 'go' I want you to stand up and walk to the line, turn and then walk back to the chair and sit down again. Walk at your normal pace."

Variations

You may have the patient walk at a fast pace to see how quickly they can ambulate. Also you could have them turn to the left and to the right to test any differences.

*Podsiadlo D, Richardson S. The timed "up and go": a test of basic functional mobility for frail elderly persons. *JAGS* 1991; 39: 142-148.

Scoring

Time for 'Up and Go' test _____sec.
Unstable on turning?
Walking aid used? Type of aid: _____

6.13 Appendix XIII

ID# _____

Date: _____

PedsQL™

Cerebral Palsy Module

Version 3.0

YOUNG CHILD REPORT (ages **5-7**)

Instructions for interviewer:

I am going to ask you some questions about things that might be a problem for some children. I want to know how much of a problem any of these things might be for you.

Show the child the template and point to the responses as you read.

If it is <u>not at all</u> a problem for you, point to the smiling face

If it is <u>sometimes</u> a problem for you, point to the middle face

If it is a problem for you <u>a lot</u>, point to the frowning face

I will read each question. Point to the pictures to show me how much of a problem it is for you. Let's try a practice one first.

	Not at all	Sometimes	A lot
Is it hard for you to snap your fingers	☺	😐	☹

Ask the child to demonstrate snapping his or her fingers to determine whether or not the question was answered correctly. Repeat the question if the child demonstrates a response that is different from his or her action.

Think about how you have been doing for the last few weeks. Please listen carefully to each sentence and tell me how much of a problem this is for you.

After reading the item, gesture to the template. If the child hesitates or does not seem to understand how to answer, read the response options while pointing at the faces.

DAILY ACTIVITIES *(problems with...)*	Not at all	Some-times	Alot
1. Is it hard for you to put your shoes on	0	2	4
2. Is it hard for you to button your shirt	0	2	4
3. Is it hard for you to pull a shirt on over your head	0	2	4
4. Is it hard for you to put on your pants when you get dressed	0	2	4
5. Is it hard for you to brush your hair	0	2	4
6. Is it hard for you to get into the bathroom to use the toilet	0	2	4
7. Is it hard for you to undress to use the toilet	0	2	4
8. Is it hard for you to get in and out of bathtub/shower	0	2	4
9. Is it hard for you to brush your teeth	0	2	4

SCHOOL ACTIVITIES *(problems with...)*	Not at all	Some-times	Alot
1. Is it hard for you to write or draw with a pen or pencil	0	2	4
2. Is it hard for you to use scissors	0	2	4
3. Is it hard for you to use a keyboard on the computer	0	2	4
4. Is it hard for you to use a mouse for the computer	0	2	4

MOVEMENT AND BALANCE *(problems with...)*	Not at all	Some-times	Alot
1. Is it hard for you to move one or both of your legs	0	2	4
2. Is it hard for you to move one or both of your arms	0	2	4
3. Is it hard for you to move parts of your body	0	2	4
4. Is it hard for you to keep your balance when you are sitting in a chair	0	2	4
5. Is it hard for you to keep your balance when you are standing	0	2	4

Think about how you have been doing for the last few weeks. Please listen carefully to each sentence and tell me how much of a problem this is for you.

PAIN AND HURT *(problems with...)*	Not at all	Some-times	Alot
1. Do you ache or hurt in your joints and/or muscles	0	2	4
2. Do you hurt a lot	0	2	4
3. Do you have trouble sleeping because of pain or aching in your joints and/or muscles	0	2	4
4. Do your muscles get stiff and/or sore	0	2	4

FATIGUE *(problems with...)*	Not at all	Some-times	Alot
1. Do you feel tired	0	2	4
2. Do you feel physically weak (not strong)	0	2	4
3. Do you rest a lot	0	2	4
4. Do you not have enough energy to do things that you like to do	0	2	4

EATING ACTIVITIES *(problems with...)*	Not at all	Some-times	Alot
1. Is it hard for you to eat with a spoon and/or fork	0	2	4
2. Is it hard for you to chew your food	0	2	4
3. Is it hard for you to hold a cup	0	2	4
4. Is it hard for you to drink on your own	0	2	4
5. Is it hard for you to cut your food	0	2	4

SPEECH AND COMMUNICATION *(problems with...)*	Not at all	Some-times	Alot
1. Is it hard for you to tell your family what you want	0	2	4
2. Is it hard for you to tell other people what you want	0	2	4
3. Is it hard for your family to understand your words	0	2	4
4. Is it hard for other people to understand your words	0	2	4

How much of a problem is this for you?

Not at all

Sometimes

A lot

ID# _____

Date: _____

PedsQL™

Cerebral Palsy Module

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CHILD REPORT (ages **8-12**)

DIRECTIONS

Children with Cerebral Palsy sometimes have special problems. Please tell us **how much of a problem** each one has been for you during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

*In the past **ONE month**, how much of a **problem** has this been for you…*

DAILY ACTIVITIES *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to put on my shoes	0	1	2	3	4
2. It is hard for me to button my shirt	0	1	2	3	4
3. It is hard for me to pull a shirt on over my head	0	1	2	3	4
4. It is hard for me to put pants on when I get dressed	0	1	2	3	4
5. It is hard for me to brush my hair	0	1	2	3	4
6. It is hard for me to get into the bathroom to use the toilet	0	1	2	3	4
7. It is hard for me to undress to use the toilet	0	1	2	3	4
8. It is hard for me to get in and out of bathtub/shower	0	1	2	3	4
9. It is hard for me to brush my teeth	0	1	2	3	4

SCHOOL ACTIVITIES *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to write or draw with a pen or pencil	0	1	2	3	4
2. It is hard for me to use scissors	0	1	2	3	4
3. It is hard for me to use a keyboard on the computer	0	1	2	3	4
4. It is hard for me to use a mouse for the computer	0	1	2	3	4

MOVEMENT AND BALANCE *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to move one or both of my legs	0	1	2	3	4
2. It is hard for me to move one or both of my arms	0	1	2	3	4
3. It is hard for me to move parts of my body	0	1	2	3	4
4. It is hard for me to keep my balance when I am sitting in a chair	0	1	2	3	4
5. It is hard for me to keep my balance when I am standing	0	1	2	3	4

PAIN AND HURT *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I ache or hurt in my joints and/or muscles	0	1	2	3	4
2. I hurt a lot	0	1	2	3	4
3. I have trouble sleeping because of pain or aching in my joints and/or muscles	0	1	2	3	4
4. My muscles get stiff and/or sore	0	1	2	3	4

*In the past **ONE month,** how much of a **problem** has this been for you…*

FATIGUE *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel tired	0	1	2	3	4
2. I feel physically weak (not strong)	0	1	2	3	4
3. I rest a lot	0	1	2	3	4
4. I don't have enough energy to do things that I like to do	0	1	2	3	4

EATING ACTIVITIES *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to eat with a spoon and/or fork	0	1	2	3	4
2. It is hard for me to chew my food	0	1	2	3	4
3. It is hard for me to hold a cup	0	1	2	3	4
4. It is hard for me to drink on my own	0	1	2	3	4
5. It is hard for me to cut my food	0	1	2	3	4

SPEECH AND COMMUNICATION *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to tell my family what I want	0	1	2	3	4
2. It is hard for me to tell other people what I want	0	1	2	3	4
3. It is hard for my family to understand my words	0	1	2	3	4
4. It is hard for other people to understand my words	0	1	2	3	4

ID# _____

Date: _____

PedsQL™

Cerebral Palsy Module

Version 3.0

TEEN REPORT (ages **13-18**)

DIRECTIONS

Teens with Cerebral Palsy sometimes have special problems. Please tell us **how much of a problem** each one has been for you during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

*In the past **ONE month,** how much of a **problem** has this been for you…*

DAILY ACTIVITIES *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to put on my shoes	0	1	2	3	4
2. It is hard for me to button my shirt	0	1	2	3	4
3. It is hard for me to pull a shirt on over my head	0	1	2	3	4
4. It is hard for me to put pants on when I get dressed	0	1	2	3	4
5. It is hard for me to brush my hair	0	1	2	3	4
6. It is hard for me to get into the bathroom to use the toilet	0	1	2	3	4
7. It is hard for me to undress to use the toilet	0	1	2	3	4
8. It is hard for me to get in and out of bathtub/shower	0	1	2	3	4
9. It is hard for me to brush my teeth	0	1	2	3	4

SCHOOL ACTIVITIES *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to write or draw with a pen or pencil	0	1	2	3	4
2. It is hard for me to use scissors	0	1	2	3	4
3. It is hard for me to use a keyboard on the computer	0	1	2	3	4
4. It is hard for me to use a mouse for the computer	0	1	2	3	4

MOVEMENT AND BALANCE *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to move one or both of my legs	0	1	2	3	4
2. It is hard for me to move one or both of my arms	0	1	2	3	4
3. It is hard for me to move parts of my body	0	1	2	3	4
4. It is hard for me to keep my balance when I am sitting in a chair	0	1	2	3	4
5. It is hard for me to keep my balance when I am standing	0	1	2	3	4

PAIN AND HURT *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I ache or hurt in my joints and/or muscles	0	1	2	3	4
2. I hurt a lot	0	1	2	3	4
3. I have trouble sleeping because of pain or aching in my joints and/or muscles	0	1	2	3	4
4. My muscles get stiff and/or sore	0	1	2	3	4

*In the past **ONE month,** how much of a **problem** has this been for you…*

FATIGUE *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel tired	0	1	2	3	4
2. I feel physically weak (not strong)	0	1	2	3	4
3. I rest a lot	0	1	2	3	4
4. I don't have enough energy to do things that I like to do	0	1	2	3	4

EATING ACTIVITIES *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to eat with a spoon and/or fork	0	1	2	3	4
2. It is hard for me to chew my food	0	1	2	3	4
3. It is hard for me to hold a cup	0	1	2	3	4
4. It is hard for me to drink on my own	0	1	2	3	4
5. It is hard for me to cut my food	0	1	2	3	4

SPEECH AND COMMUNICATION *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to tell my family what I want	0	1	2	3	4
2. It is hard for me to tell other people what I want	0	1	2	3	4
3. It is hard for my family to understand my words	0	1	2	3	4
4. It is hard for other people to understand my words	0	1	2	3	4

6.14 Appendix XIV

PARTICIPANT EVALUATION

	Not at all	A little	A lot
			
I liked the feeling of the TES machine:			
I would use the TES machine again:			
I found the TES machine helped me:			

Other comments about the machine:

_____

_____

_____

6.15 Appendix XV

Parent consent form and information sheet in Zulu

INCWADI YEMVUMELO VOMZALI/YOMLINDI

IGAMA LEPHROJEKTHI:

Umehluko okwenza yi-Therapeutic Electrical Stimulation on abdominal muscle strength and gross motor function ezinganeni ezine-Cerebral palsy: a randomised control trial

UMHLOLI OMKHULU: Jessica Joffe

Wena nengane yakho nimenyiwe ukuthi nihlangayele kwicwaningo engiyenzayo ukuze ngithole umnyezane kwi-Physiotheraphy e-Inyunivesi yaseKapa (UCT). Ngiyi-Physio ezilobile esebenze eHope School. Ngicela nithatha isikhathi nifunde lencwadi futhi ningasabi ukubuza imibuzo.

Locwaningo ivunyelwe yi-Research and Ethics committee yaseUCT noThishanhloko waseHope School. Locwaningo uzolandela imithetho.

LOCWANINGO UMAYELANA NANI?

Locwaningo uzobheka ukuthi urnuthi obizwa 'Therapeutic Electrical Stimulation' (TES) nokwelapha kwengane kuzo enza kube-ngcono amandla womsipha esiswini. Imicwaningo abonisile ukuthi i-TES inga enza kube-ngcono amandla womsipha esiswini sengane ene-Cerebral palsy.

KUNGANI INGANE YAMI IKHETHIWE?

Ingane yakho ikhethiwe ngoba inemiyaka engu-5 kuya ku-18, ine-cerebral palsy futhi ifunda eHope School.

INTO ENIDINGA UKUYENZA

Ukuze ingane yakho ihlanganyele kulocwaningo kumele usayine lencwadi. Uma ingane yakho ikhethiwe, kuzobe kunamaqhembu amabili. Lamaqhembu womabili azothola i-physio esikoleni. Iqhembu lesibili fizothola i-TES treatment phezulu kwezisu zabo ngesikhathi se-physio. Lomshini angeke ulimaze izingane.

Ukuhlola konke kuzokwenzeka ngesikhathi sesikole ngakho-ke ingane yakho angeke ishiyiwe ekilasini.

Ukuhlola konke kuzoyenzwa umuntu ufundele ukuba i-physio futhi izothatha ihora kuphela. Ingane yakho kuzomele iphendule incwadi enemibuzo ngempilo yakhe futhi nemibuzo nge- TES treatment.

INGANE YAKHO IYOSIZAKALA NA?

Uma ingane yakho ikwiqhembu le-TES futhi izinto ezihle zitholakala ngokushesha, ingane yakho iyasizakala zisasuka. Leziphetho esizitholayo zizosiza ingane yakho nabanye abantwana abane-cerebral palsy. Ukuhlanganyele kumahala kodwa angeke sikukhokhele.

ZIKHONA IZINGOZI NA?

Ingozi eyaziwayo yinkinga yesikhumba uma usebenzisa umshini (isikhumba sibabomvu). Uma kubayinkinga uvunyelwe ukuphurna kulocwaningo. Uma kubanengozi inzingane zizolapha isitafu ngendlela elungile.

INGANE YAMI IZOYAZIWA NA?

Yonke ukwaziswa kuzobekwa efayeleni engasese futhi ethembikile. Umhloli nomphathi wakhe bazoba nemvumelo kulefavela kuphela futhi angeke basebenzise igama lengane yakho.

NGENZENJANI UMA NGINEMIBUZO?

Uma unemibuzo noma ufuna ukwazi okunye ngalocwaningo, ngicela ungithinte ngocingo noma iposi likagesi (Jessica Joffe):

Ucingo: 083 951 7073 **Iposi likagesi:** jessicajoffe@hotmail.com

Uwamukekile ukuthinta i-Research Ethics Committee uma unemibuzo:

Marc Blockman

Research Ethics Committee Faculty of Health Sciences

E 52-23 Old Main Building

Groote Schuur Hospital

Observatory

7925

Ucingo: 021 406 6492

Isikhahlamezi: 021 406 6411

ISIFINYEZO SENVUMELWANO

Mina u_____ (igama) ngiyavuma ukuthi ingane yami

u _____ (igama) ihlanganyele ucwaningo obizwa "The effect of

Therapeutic Electrical Stimulation on abdominal muscle strength and gross motor function in children with cerebral palsy: a randomised control trial".

Ngiyathembisa ukuthi:

Ngifundile noma ngithole umuntu ongifundele ukwaziswa ngalocwaningo nephepha senvumelwano futhi ibhalwe ngolimi olulula.

Ngilitholile ithuba lokubuza imibuzo futhi ngithole imphendulo.

Mina nengane yami asijezisangwa ukuthi sihlanganyele futhi siyagondasisa ukuthi yithi esifuna uhlanganyela kulephenyo.

Ngiyakwazi ukukhipha ingane yami kulocwaningo noma nini uma ngifuna futhi lokhu angege kwenza abantu bajezise ingane yami esikoleni.

Sayinwe e_____(indawo) ngo _____ (usuku).

_____ _____

i-signature yomzali/umlindi i-signature yofakazi

Parent consent form and information sheet in Xhosa

KITSISO LE FOROMO YA TUMELANO YA BATSADI

SETLHOGO SA LENANE/POROJEKE:

Tiro ya Therapeutic Electrical Stimulation mo go thatafatseng mosifa wa mpa le tiro ya gross motor go baneng ba nang le cerebral palsy: e tla nna e diriwa ka tlase ga taolo.

MOTHO YO MOGOLO YO A DIRANG DIPATLISISO KE: Jessica Joffe

Wena le ngwana wa gago le lalediwa go tseya karolo mo patlisisong e e dirang ya dithuto tsa me tsa Masters in Physiotherapy degree le Unibesity ya Cape Town (UCT). Ke Physio e rejesetariweng: ke dira kwa Hope School. Ke kopa o tsee nako go bala lekwalo le, o se ke wa tshaba go ikopanya le nna ka dipotso tse o ka bang le tsona.

Tseo karolo mo dithutong tse ke boithaopo. Fa e le gore o tlhopa go se tsee karolo, tsholo ya ngwana wa gago mo sekolong ga e kitla e amega ka gope. Fe o tlhopile gore ngwana wa gago a tsee karolo, mme morago wa be o fetola tsheetso e o e tsereng, o kgona go tswa nako e nngwe le enngwe.

Thuto e amogetswe ke Unibesithi ya Cape Town, Faculty of Health Sciences Human Research Ethics Committee. Lefapa la thuto la porofenseng ya Gauteng le Mogokgo wa sekolo sa Hope Thuto e e tla tswelapele e latela melao e e beilweng ya kae lo.

THUTO E KE YA ENG?

Thuto e e tla lebelela gore a tiriso ya motshini o o bitswang Therapeutic Electrical Stimulation (TES) ga e tlhakana le tiriso ya metlheng ya physiotherapy e ngwana wa gago a emogelang, e tla dira tokafatso mo go

tieng ga mesifa ya mpa; le gore a tiriso ya mmele wa gagwe ka kakaretso e a lekana.

Dithuto tse, di bontshitse gore TES e ka tokafatsa go lekana mmele , ka mokgwa wa gonna fatshe le go tia ga mesifa mo baneng ba cerebral palsy.

GORENG NGWANA WA GAGO A TLHOPILWE

Ngwana wa gago o tlhopilwe jaaka a le magareng ga dingwaga di le 5 le 18, a na le cerebral palsy ebile a tsena sekolo sa Hope.

SE O TLHOKANG GO SE DIRA

Ngwana wa gago o tla kgona go tsaa karolo fa fela o saenele forom eya tumelano, e e leng ka morago. Ge ngwana wa gago a le mo palong, bana ba tla ntshiwa ditlhopha tse pedi. Ditlhopha tse pedi tse, di tla tswelela di amogela physio mo sekolong jaaka ba tlwaetse. Setlhopha sa, bobedi se tla amogela le tirelo ya TES mo dimpeng tsa bona ka nako e e tlwaelegileng ya physio. Fa ngwana wa gago a tlhopilwe go ba mo setlhopeng se, physio ya gagwe e tla bea mesamelonyana e me nnyane mo dimpeng tsa gagwe nako e ka nnang metsotso e 20 fa a tsweletse pele a dira physio ya gagwe gangwe ka beke mo dibekeng tse 6.

Ditshekatsheko le ditirelo tsotlhe di tla diriwa ka nako ya sekolo e e tlwalegileng, se se yaya gore ngwana wa gago, ga a tlhoke go se be teng phaposing ya borutelo jaaka ba dira ka metlha fa ba tshwanetse ba ye go dira therapy ya bone.

Ditshekatsheko tsotlhe tse di diriwang pele le morago ga dithuto tse, di tla diriwa mo go se se bonwang ka matlho; ke physio e e katisitsweng, e tla tsaya bolefele boa ura e le nngwe.

Tsotlhe tsa diteko tse, di tlhoka gore ngwana wa gago a leke go dira ditironyana tse ding tse farologanang e le lebaka la go ikatisa. Ga gona tse e

leng gore di a tshosa kgotsa di utlwisa botlhoko. Ba tla kopiwa gape gore ba tlatse pampiri ya dipotso mabapi le maikutlo a bona ka ga tse ding mo maphelong a bone le gore a ba nagana eng ka tiriso e ya TES.

A NGWANA WA GAGO O TLA THUSEGA MO GO TSEENG KAROLO?

Fa e le gore ngwana wa gago o mo setlhopeng sa TES ebile go nale ditlamorago tse ntle tse fitlhetsweng, gone o tla thusega go tloga ka yona nako eo. Ditlamorago tseo di ka rethusa ka go loga maano tebang le bokamoso boa therapy ya ngwana wa gago. Go tsaya karolo ya tiriso ya TES ke mahala, ga e duelwe.

A GO NALE KOTSI E E LENG GORE E KA TLHAGA (RISK)?

Selo se le sengwe fela se se ka tlhagelelang fa motho a dirisa TES ke phetogonyana ya letlalo le dirang ke mesamelonyana e e dirisiwang (go tlwaelegile ebile go siame gore letlalo le be le le hubidunyana morago ga tiriso ya TES).

Fa e le gore ngwana wa gago o ngongorega ka bohubidu boa letlalo bo o leng gore gabo nyelele kgotsa bo fola, ba dumelletswe gore ba tswe tirisong e. Fa go ka diragala kotsi ka nako ya tiriso e, ngwana wa gago o tla ba ka tlase ga tlhokomelo e e tlwaele gileng e a efitlhelang ka metlha fa a le mo sekolong.

A NGWANA WA ME O TLA SIRELETSEGA GORE A SE KE A ITSIWE PHATLALATSA?

Dilo tsotlhe tse re di itseng ka ngwana wa gago di tla tsholwa ka faeleng ya sephiri e e bolokegileng. Motho yo a tsamaisang lenane le, ke ena fela yo a tla fitlhelelang difaele tse. Leina la ngwana wa gago le ka se dirisiwe mo dipuisanong kgotsa go phatlalatsa dipatlisiso tse (research).

GO DIRAGALA ENG FA E LE GORE KE NA LE DIPOTSO?

Fa e le gore o na le dipotso kgotsa o rata go ka itse go le go ntsi ka dithuto tse, ke kopa gore o se ke wa etsaetsega/ wa tshaba go ikopanya le nna ka tsela e e latelang (Jessica Joffe):

Cell: 083 9517073 e-mail: jessicajoffe@hotmail.com

Le a mogetswe gape go ikopanya le moemedi wa yunibesithi ya Cape Town Faculty of Health Sciences Human Research Ethics Committee.

Marc Blockman
Research Ethics Committee
Faculty of Health Sciences
E 52-23 Old Main Building
Groote Schuur Hospital
Observatory 7925
Tel: +27- 21- 4066492
Fax: +27- 21- 4066411

KITSISO YA TUMELANO

Nna, ke le _____ (leina) ke dumela gore ngwana wa me _____ (leina) a tsee karolo mo dithutong tsa “Ditlamorago tsa tiriso ya Therapeutic Electrical Stimulation mo go tiiseng mesifa ya mpa le motor function mo baneng ba cerebral palsy: e e tla dirwang ka tlase ga taolo”.

Ke le itsise gore:

Ke badile kgotsa mongwe o mpaletse kitsiso e, mabapi le dithuto le foromo e ya tumelano le gore e kwadilwe ka loleme le ke le tlhaloganyang.

Ke bile le nako ya go ka botsa potso e nngwe le enngwe e nka bang le yona. Ke ne ka kgotsofala ka mokgwa o di neng tsa arabiwa ka teng.

Nna le ngwana wa me re ne ra se gapeletswe go tsaya karolo ya dithuto tse. Ke ya utlwisisa gore go a ithaupiwa.
Ke kgona go ka gogela morago fa ke eletsa, ka nako e nngwe le e nngwe fa ke sa tlhole ke batla go tswelela pele ka dithuto tse. Se ga se na go kgoreletsa ngwana wa me go tswelela pele ka dithuto tsa gagwe tsa sekolo mo Hope School.

Saenilwe kwa _____ (lefelo) ka _____ (letsatsi).

_____ _____

Mo go saenang motswadi/mosireletsi Mo go saenang paki