Effects of Specifically Sequenced Massage on Spastic Muscle Properties and Motor Skills in Adolescents with Diplegic Cerebral Palsy

A Thesis Submitted for the Degree of Doctor of Philosophy in the Faculty of Biomedical and Life Sciences

by

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April 2009

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AUTHOR'S DECLARATION

I declare that this thesis embodies the results of my own work, and that it does not include work forming part of a thesis presented successfully for a degree at this or any other University

Signed:…………………………

Date:…………………………...

ACKNOWLEDGEMENT

My unending thanks goes to God, for guiding me to the right people at Glasgow University, and for giving me the perseverance to complete this PhD.

My thanks and appreciation go to my supervisors Dr. M. H. Gladden and Dr.R. H. Baxendale for their support and encouragement. Special thanks go to Dr. Gladden for her continued enthusiasm and input even in her retirement and to Dr Baxendale for his patience and attention to detail during the final write-up. Thanks also to go Dr Linda Ross and her consultant colleagues at Yorkhill Sick Children's Hospital, for embracing the work from the outset.

Thanks also goes to Kristen Hefner, for her enthusiasm for the project and help with formatting, to Ian Watt, Paul Paterson, Robert Auld for being willing control participants, and to numerous staff members at Glasgow University for their encouragement, even when the project was spawned at undergraduate level.

Lastly, thanks to family and friends for their continued interest, especially to my wife for the patience and encouragement that was always on hand.

ABSTRACT

Cerebral Palsy (CP) is the most common childhood disability, with an incidence around 2-2.5 in every 1,000 live births in Europe. It results from damage to the developing brain and adversely affects motor control. The limitations in motor control range from an inability to even hold the head erect and an inability to self-feed, to cases where for example walking is hampered by spasticity in one limb.

The cornerstone of current treatment is physiotherapy in which the aims are to maintain and improve mobility and to prevent limitation of the range of joint movement. Specific forms of physical therapy include Conductive Education and Bobath treatment. Other interventions include botulinum toxin injections, intrathecal baclofen, selective dorsal rhizotomy and multi-level orthopaedic surgery. Despite these varied and concerted inputs, improvements in motor skills are very limited – motor skills tend to plateau around the age of seven and in fact deteriorate in adolescence.

Of the four classifications of CP (Spastic, Athetoid or Dyskinetic, Ataxic and Mixed), the spastic type is the most common. Around 75% of all cases are spastic and around 60% of these are diplegic (meaning it affects both limbs, usually the legs). Spastic diplegia results from periventricular leucomalacia, where oligodendrocytes are damaged by hypo-perfusion of the periventriucular areas predominantly affecting the corticospinal tracts supplying the legs. This results in a deficit in the development of the white matter forming the insulation around those nerves and consequently compromises the signal transduction to

the legs. As spastic diplegia is the most common type of CP, and the presenting symptoms are considered less complex than the other types, patients with spastic diplegia were chosen for participation in the current studies.

The main symptoms presenting in spastic diplegia are reduced gross motor function, increased reflex response to muscle stretch, reduced range of ankle movement and shortened calf muscle/tendon units, evident in equinus.

Whilst the main cause of spasticity in conditions other than CP is considered to be the neuropathology, altered muscle properties are considered to be the main problem in CP. Masseurs contend that they bring about a healthy response in damaged muscle by altering the resting state of the muscle, although this has not been scientifically proven until now.

The initial aim of the present series of studies was to test if a specific massage sequence could increase the range of movement at the ankle joint by altering the mechanical properties of the muscle in adolescents with spastic diplegia. However the investigations indicated that instead, this type of massage changed sensory feedback from the spastic muscles, which led to significant improvements in motor skills. The physical limitations of the 12 participants with CP range from habitual wheelchair users to one participant who is able to run. Their abilities classified by the Gross Motor Function Classification System (GMFCS) ranged from level I to IV. The investigation involved the use of goniometry to measure change in the active and passive range of movement at the ankle joint and EMGs to measure incidence of stretch reflex contractions.

Motor skills were assessed by an independent physiotherapist, using the Gross Motor Function Measure-66 (GMFM-66).

In chapter 3, three passive ankle dorsiflexions at a controlled rate were carried out before and after massage which was given twice weekly for 5 weeks. The incidence of stretch reflex contractions during passive dorsiflexion was reduced from 40% in the first 5 massage sessions to 22% in the last 5 sessions, in the 5 participants tested. After massage the resistance of the calf muscle to stretch was not reduced as expected; in fact the muscles were stiffer (more force was needed to take the ankle through the same range of movement). However, the resting angles of the ankles often changed, indicating alteration of the resting length of the calf muscles. The change was not always in the one direction, although, on the whole, muscles lengthened after massage (shown by an average increase in dorsiflexion of 1.4º). It is argued that thixotropic properties of muscles were responsible and that the massage changed the mechanical properties of the calf muscles.

In chapter 4, Gross Motor Function Measure-66 scores for all 12 adolescent participants who received the specialised massage were shown to be improved by an average of 5.8. Five of the 7 participants showed improvements in their ability to descend stairs, which is recognised to be particularly difficult in spastic diplegia. The range of voluntary ankle movement improved in some participants in some tests.

Despite a lack of scientific evidence, masseurs also contend that their intervention brings about change by altering the blood flow. In the current studies, near infrared spectroscopy was used to measure oxygenation of the muscles and changes in the skin temperatures were also recorded. In chapter 5, temperature recorded from the skin over the calf muscle after massage was increased in both the CP group and the controls. Both finished with comparable temperatures although the CP group's temperatures started 1-1.5°C below those of the control group. Contralateral effects of raised skin temperature were also observed. It was confirmed that the extent of change in skin temperature over the massaged muscles could be used to determine the effectiveness of a trainee using the massage technique. Additionally, the oxygenation of the tissue was altered significantly at some stage during massage for all participants. It is proposed that spastic muscles in CP may sometimes operate in oxygen debt, particularly in cold conditions.

The improvements in GMFM-66 with massage are at least as effective as other current therapies and the massage has none of the adverse side effects of surgery and drug interventions. The mean improvement in GMFM-66 score after massage was 5.8, whereas treatments using selective dorsal rhizotomy and baclofen showed improvements of only 2.7 and 3.8 respectively.

It is proposed that the mechanical properties and the feedback from spastic muscles are altered by the massage and that the CNS is able to accommodate the change in feedback to produce improved motor function.

It is recommended that the massage used here be incorporated into the physiotherapy regime for individuals with CP.

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ABBREVIATIONS

CP	Cerebral palsy
GMFM	Gross Motor Function Measure
GMFCS	Gross Motor Function Classification System
PVL	Periventricular leucomalacia
SDR	Selective dorsal rhizotomy
CIBI	Continuous intrathecal baclofen infusion
Botox	Botulinum toxin-A
EMG	Electromyography
NIRS	Near infrared spectroscopy
cm	centimetre
s	seconds
m/s	metres per second
N/s	Newtons per second
m	metre
V	volt
H reflexes	Hoffman reflexes
Hb	Haemoglobin
HbT	Total haemoglobin
HbO_2	Oxyhaemoglobin
CtOx	Cytochrome c oxidase
IOS	Inter-optode spacing
DPF	Differential path factor
Hz	Hertz
C	Centigrade
b	Baseline
e	Effleurage
pu	Pick up
puh	Pick up and hold
r	Rolling
k	Kneading
ar	Appositional rolling
w	Wringing
eam	Effleurage end of massage
bam	Baseline after massage
CI	Confidence intervals

CHAPTER 1

Introduction and Literature Review

1. Background and Rationale

As the most common childhood motor disability, cerebral palsy (CP) can have a devastating affect on the lives of many children and their families (Molnar, 1991). There is no cure for CP, which occurs in 2-2.5 births in every 1,000. A review of the literature indicates that even the most up-to-date interventions aimed at alleviating the spasticity present in most cases have met with very limited success.

It is considered that the major cause of the spasticity presenting in CP is due to altered properties of the skeletal muscle, even though the original damage was neurological (see later sections on spasticity). Masseurs contend that they are affecting the mechanical properties of muscle during massage although this had yet to be confirmed scientifically. During a lifetime of using massage, the author has had empirical results showing improved motor function and a reduction in spasticity following massage in two family members and others who have CP. It seemed important to assess if these results could be replicated on a larger scale. Additionally, whilst working at the Scottish Centre for Children with Motor Impairments, it became clear to the author that the massage intervention needed to be scientifically investigated in order for it to be incorporated into the physiotherapy input as a matter of course for those children.

However, there is very limited published research on the effects of massage on CP. Only one controlled study has been published (Hernandez-Reif *et al*, 2005). Those authors had previously shown improvements using massage in other conditions of disability. From their results they propose that massage attenuates physical symptoms associated with CP and that their results should promote further study. Having reviewed that work (see point 1.13 later), it became clear that a series of studies could be undertaken which could further advance our understanding of the effects of massage in those with CP. This includes investigations in the current thesis intended to uncover possible underlying mechanisms by which the massage may have its effect.

The rationale for the chosen outcome measures and the size of the participant populations was as follows; a general linear statistical model indicated that an improvement of as little as 5% in gross motor function would be considered significant with as few as 5 participants. By measuring gross motor function rather than a change in spasticity, the practical use of the improvements was also self evident. For example, if a participant could walk down stairs after a series of massage sessions, having been unable to do so before the massage. Other examples of this are the participant being able to sit or stand unaided after the intervention, resulting in more independence for them.

1.1 Characteristics of Cerebral Palsy

The following definition alludes to the complexity of this condition. 'Cerebral palsy (CP) describes a group of disorders of the development of movement and posture causing activity limitation, which are attributed to non-progressive disturbances that occurred in the developing foetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, cognition, communication, perception, and/or behaviour, and/or by a seizure disorder' (Bax *et al.*, 2005). It should be noted that although reduced cognitive ability occurs in around 30% of CP cases the term 'cerebral palsy' describes only the motor component of the disability (Nelson & Ellenberg, 1978).

The term 'cerebral palsy' is a description of the presenting clinical symptoms, not a specific diagnosis (Ketelaar *et al.*, 2001). It is a complex amalgam of motor function deficits and consequently its treatment and assessment of interventions can also be complex and difficult. In his review of the efficacy of lower limb orthoses used for cerebral palsy, Morris highlighted a major difficulty in the understanding, assessment and treatment of CP when he said 'The nature of the cerebral palsies is that each child has a slightly different cerebral pathology' (Morris, 2002).

It is important to note that whilst the damage to the brain by the initial insult does not progress, the resulting musculoskeletal and movement problems can often become worse as the children move into adolescence (Mutch *et al.*, 1992). Other workers have stated that it is inappropriate to emphasise that the cerebral lesion is static without stating clearly that the musculoskeletal pathology will be progressive in many cases (Graham *et al.*, 2003). They observe that the new born child with CP usually has no deformities or musculoskeletal abnormalities at birth and that scoliosis, dislocation of the hip and fixed contractures develop during the rapid growth of childhood (Kerr Graham & Selber, 2003). Not surprisingly therefore, Boyd and colleagues argued that the basic definition of cerebral palsy should be extended to acknowledge the progressive nature of the musculoskeletal pathology (Boyd *et al.*, 2001)

For ambulatory individuals with CP the effects of the neural damage were summarised by DeLuca in 1991. He said that mass limb and postural reflexes in conjunction with spasticity are responsible for the muscle imbalance that exists between joint agonists and antagonists (flexors and extensors). In the growing child such imbalance rapidly produces the secondary problems of fixed muscle contracture and joint and skeletal deformity. An additional primary deficit of central origin is an impaired balance mechanism, which produces the adaptations of a flat-foot, crouched, wide-based gait. This muscle imbalance is also evident in the upper limbs and trunk in triplegic and quadraplegic CP (DeLuca, 1991).

The motor condition in CP may be spasticity, hypotonicity, or dyskinesia, with the added complication of the development of fixed deformity, and may change as the child develops (Badawi *et al.*, 1998).

Any assessment of interventions, therefore must take account of the type of disability, the severity of the condition and the age of the child.

1.2 Types of Cerebral Palsy

Spastic CP is characterized by a much-reduced capability of the skeletal muscles to stretch. The majority of CP cases are of the spastic diplegic type, where 'spastic' refers to the manifestation of the movement disorder and 'diplegic' refers to the distribution within the body, affecting mainly the legs (Albright *et al.*, 1993; Kuban & Leviton, 1994). Around 75% of all CP cases are spastic and around 50% of these are diplegic (Stanley *et al.*, 2000). *Athetoid (Dyskinetic) CP* is much less common (around 10%) and has been attributed to a brief period of profound asphyxia at term, which damages the basal ganglia. This results in very unsteady movements of the head, arms and legs, necessitating support from others (Nelson & Ellenberg, 1978). *Ataxic CP* is a rare form (less than 5%), with low muscle tone and poor coordination in evidence. Children with ataxic CP look very unsteady and shaky with a wide-based gait (Nelson & Ellenberg, 1978). In *mixed CP*, the children have both the taut muscle tone of spastic CP and the involuntary movements of athetoid CP. This affects about 10% of children with CP (Nelson & Ellenberg, 1978).

The type of CP is further described by reference to the number and location of the limbs involved, i.e. monoplegia, hemiplegia, diplegia, triplegia and quadraplegia.

1.3 Severity and Gross Motor Function Classification System

It has generally been considered that the degree of severity of involvement for patients has similar numbers in each of the ‘mild’, ‘moderate’, and ‘severely involved’ categories of disability, and this wide range of capabilities has made description, assessment and treatment difficult (Scherzer, 2001).

However the recently developed Gross Motor Function Classification System (GMFCS) has proved invaluable in this respect. It was developed in response to calls for a standardized system to measure the ‘severity of movement disability’ in children specifically with CP (Morris & Bartlett, 2004). This classification system consists of five levels, I – V, where I is the most capable and V is the worst affected. Throughout childhood children in all levels have physical impairments that limit voluntary control of movement but there is a differential in motor skills between the levels. For example, before the 2nd birthday, children in level I can move in and out of sitting with both hands free to manipulate objects, whereas children in level V are unable to maintain anti-gravity head and trunk postures in prone and sitting, and they require assistance to roll. As the children become older, their motor skills develop until around the age of 7 years, but the differential in motor skills between levels continues (Rosenbaum *et al.*, 2002) (fig 1.1).

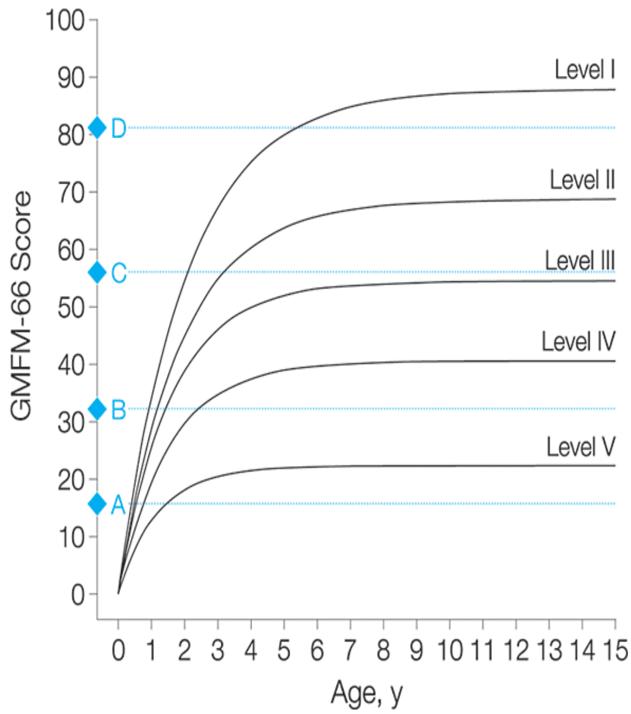


Figure 1.1
Predicted Average Development by the Gross Motor Function Classification System (GMFCS) Levels

This figure shows the predicted mean GMFM-66 scores from 4 tests of 737 children aged from birth to 15 years. The classification levels I-IV are indications of the level of ability which the child is expected to be able to achieve having achieved a particular score with GMFM-66. The diamonds on the vertical axis identify 4 Gross Motor Function Measure-66 (GMFM-66) items that predict when children are expected to have a 50% chance of completing that item successfully. The GMFM-66 item 21 (diamond A) assesses whether a child can lift and maintain his/her head in a vertical position with trunk support by a therapist while sitting; item 24 (diamond B) assesses whether when in a sitting position on a mat, a child can maintain sitting unsupported by his/her arms for 3 seconds; item 69 (diamond C) measures a child's ability to walk forward 10 steps unsupported; and item 87 (diamond D) assesses the task of walking down 4 steps alternating feet with arms free. Adapted from (Rosenbaum *et al.*, 2002).

There are different criteria for the age bands 0-2years, between 2nd and 4th birthdays, between 4th and 6th birthdays, and between 6th and 12th birthdays. The researchers who developed the GMFCS intend to develop a 5th age band from 12 to 18 years (Morris, 2002).

1.4 Age of Child

The usefulness of GMFCS is that it is a predictor of motor development for individuals with CP (Rosenbaum *et al.*, 2002). Note that fig. 1.1 shows that even in the least affected children motor development is expected to level off by age seven.

The developmental curves reveal nothing about the *quality* of motor control used to accomplish the activities and the children may improve their gross motor performance over the developing years through increased balance, stamina, energy efficiency, or quality of motor control. Thus Rosenbaum emphasizes that parents, physicians, therapists, and other decision makers do not assume further therapy is unhelpful or unnecessary when the curves appear to level off.

However, a decline in motor function is often noted as the child moves into adolescence (Bottos *et al.*, 2001; Chapple *et al.*, 2001; Campbell *et al.*, 2002). Under hormonal control their limbs become longer and stronger with greater strength in their muscles. Bone lengthening results in already tight muscles becoming under more strain, in addition to having to transport a heavier body.

1.5 Incidence of Cerebral Palsy

The prevalence of CP is static or increasing, and is generally accepted as occurring in 2-3 per 1000 live births (Graham *et al.*, 2003). There is no single national register of children with CP, although the UK has a Scottish one and 4 regional ones in England, which now form an ongoing collaborative network (UKCP). They quote 2 per 1000 for birth years 1986-1996 (Surman *et al.*, 2006). Surveillance of Cerebral Palsy in Europe (SCPE) quote 2 - 3 per 1000 live births from 6000 children in 13 geographically defined regions in the period 1980-1990 (Johnson, 2002).

The incidence is similar in the U.S.A., where more than 100,000 under 18 year olds are estimated to have some degree of neurological disability attributed to CP (Newacheck & Taylor, 1992). There are around 750,000 individuals with CP in USA.

Recent medical advances have meant that more children with CP are surviving prematurity and living longer, and multiple births following infertility treatment adds to the number of sufferers. The life expectancy of children with CP is greater than had been suggested in some previous studies (Hutton *et al.*, 1994). For subjects with no severe functional disabilities, the 20-year survival rate was 99%. However subjects severely disabled in all three functional categories (ambulation, manual dexterity and mental ability) had a 20-year survival rate of 50%.

For the highest functioning group, with full motor and feeding abilities, life expectancy is only 5 years less than that of the general population (Strauss & Shavelle, 1998). That not withstanding, Kuban concluded that the burden imposed on society by CP had not abated despite recent advances in medical care, and this is still the case more than 10 years later (Kuban & Leviton, 1994).

1.6 Causes of Cerebral Palsy

The cause of this condition has been controversial ever since 1843 when Little first described chronic encephalopathy in children (Rotta, 2002). In 1862 the link was made between the condition and abnormal delivery and until recently, it was considered that most cases of CP were the result of obstetric misadventure. However, careful epidemiological studies and brain imaging suggest that it frequently has antenatal antecedents and is often multi-factorial (Stanley *et al.*, 2001; Kerr Graham & Selber, 2003). It is now considered that developmental and genetic factors are responsible for 90% of cases, with only 10% due to intrapartum disaster (Cook *et al.*, 2002). The type of CP and severity of the symptoms depend on the size, location and timing of the lesion, however Forssberg suggests that no one of these factors is an accurate predictor of the resulting symptoms (Hadders-Algra *et al.*, 1999). Fortunately the relationship between gestational period and CP phenotype is now well established (fig 1.2; Lin, 2003).

The participants in our studies have spastic diplegic CP, and periventricular leucomalacia (PVL) has been shown to be the major cause of this. PVL denotes a failure of myelination of nerve cell axons by oligodendrocytes and accounts for about 70% of CP in babies born before 32 weeks and 30% of CP in term babies. This suggests a common antenatal origin during the period of oligodendroglial activity and resultant myelination. PVL is due to hypo-perfusion and infarction affecting the periventricular areas predominantly affecting the corticospinal tracts supplying the legs. It can also affect those supplying the arms, though much less often.

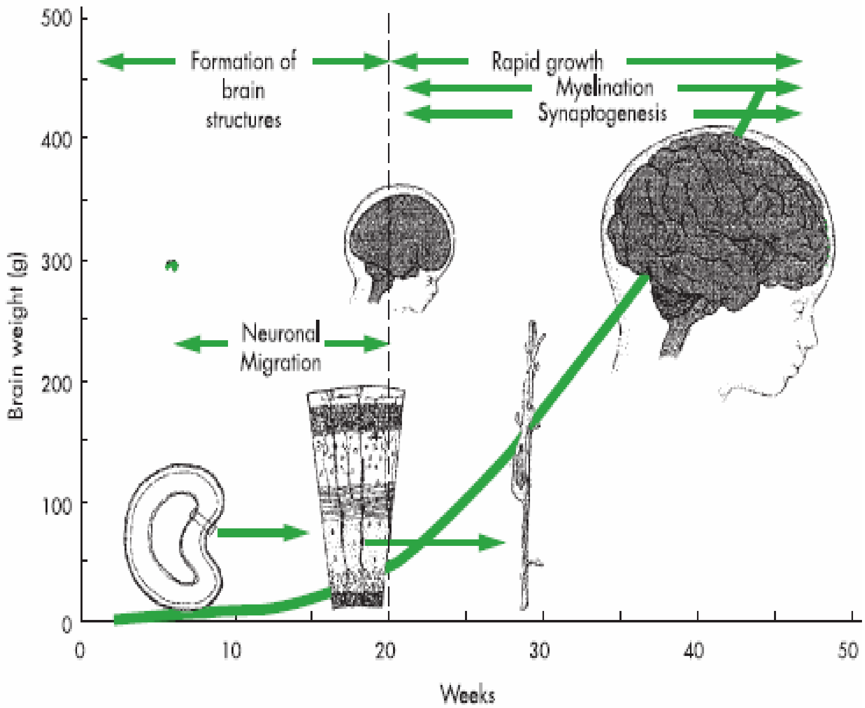


Figure 1. 2
Brain development during gestation and early postnatal life.

This figure shows the birth weight increases through gestation. It also shows the major events in brain development at each stage. Injuries between 15-22 weeks gestation result in neuronal migration defects. After about 22 weeks gestation, the oligodendrocytes are vulnerable to injury so that white matter wasting, periventricular leucomalacia, with associated expansion of the lateral ventricles is the dominant clinical pattern. Adapted from Lin, 2003.

1.7 Spasticity

Spasticity has been described as a state of increase over the normal tension of a muscle, resulting in continuous increase of resistance to stretching (Landau, 1974). Katz suggests that spasticity is more difficult to characterise than to recognise, and still more difficult to quantify (Katz 1989). However there is generalised agreement that the spastic form of CP is characterised by increased muscle tone, a positive stretch reflex, exaggerated deep tendon reflexes, and sometimes clonus (Myklebust *et al.*, 1986).

1.7.1 Spasticity - neural control of movement

Spasticity adversely affects motor co-ordination. Motor co-ordination is the process of linking the contractions of many independent muscles so that they act together and can be controlled as a single unit. Co-ordinated contraction of skeletal muscle therefore depends on neural input to and feedback from the muscle. Feedback from skeletal muscle to the CNS is provided by signals from the muscle spindles via group Ia and II afferent axons. Neural circuits in the spinal cord play an essential role in efficient motor co-ordination. Spinal reflexes provide the nervous system with a set of elementary patterns of co-ordination that can be activated either by sensory stimuli or by descending signals from the brain stem and cerebral cortex (Kandel *et al.*, 1991). Muscle tone can be seen as the continually adjusted maintenance of a muscle at its optimal length for use. The economy of the neural circuit for the stretch reflex allows muscle tone to be regulated quickly and efficiently without direct intervention by higher centres (Kandel *et al.*, 1991).

Spastic CP is considered to display heightened muscle tone, indicating that some part of the neuromuscular function is deficient. The initial brain damage affects the signals which supply the limbs via the corticospinal tracts.

Lance defined spasticity as a motor disorder characterised by velocity-dependent increase in tonic stretch reflexes ('muscle tone') where hyper-excitability of the stretch reflex is one component of the upper motor neurone syndrome (Lance, 1980). For a number of years this increase in stretch reflex excitability was considered to be the main contributor to the stiffness seen in spastic muscle (Gottlieb, 1982). However, work by Dietz and Berger on children who have CP had first suggested that muscle stiffness during locomotion in spastic patients is due more to changed mechanical properties of the muscle than to heightened stretch reflexes (Dietz & Berger, 1983).

A review of the literature since then indicates that the relative proportional contribution of stretch reflexes and that of changed muscle properties in spasticity is not clear. Additionally, current understanding does not allow us to comprehensively differentiate the spasticity displayed in individuals who have CP from those who have spasticity from other causes (Lieber, 2004). A number of studies have shown that although there is some difference between spasticity of cerebral and spinal origin, the main features such as leg muscle activation during locomotion and the physiopathology of spastic muscle tone are quite similar (Dietz, 1999).

Whilst recent advances in our understanding are being made, the way that the muscles interact with the disordered nervous system in those with CP does remain poorly understood (Lin *et al.*, 1999). Lin contends that a number of factors appear to distinguish spastic muscle from normal muscle - both the intrinsic properties of spastic muscle and its response to stretch have been shown to be abnormal.

1.7.2 Spastic muscle – neural adaptations

The myotatic stretch reflex is an unconscious neurally mediated contraction of a muscle that occurs in response to stretch of the same muscle. In this way muscle length, and thus its tension, are adjusted continuously for ease of posture, control of movement, and as a protective mechanism if the muscle is lengthened too forcefully or too quickly.

No studies appear to have been done which directly compare the stretch response of the spastic muscle in CP with that existing in other conditions. This particularly includes whether the qualities which spasticity presents from childhood are the same as that acquired in later life, e.g. following a stroke.

Many of studies of spasticity feature conditions other than CP, i.e. following a stroke, in spinal cord injured patients and in those with multiple sclerosis. Fortunately, enough studies of cerebral palsy have been done to distinguish one main difference. In the majority of cases other than CP, the main contributor to spasticity has been judged to be a reduction in the stretch reflex threshold or hyper-excitable reflex responses to stretch

(Corcos et al., 1986; Katz, 1988; Katz, 1989; Thilmann *et al.*, 1991; Sinkjaer *et al.*, 1995; Sinkjaer, 1997).

Children with CP are also considered to have exaggerated myotatic stretch reflex responses (Myklebust *et al.*, 1986). However, with CP cases, changed mechanical properties of the muscle are considered to be the *main* contributor (Deitz, 1983, 1986, 1991; Lieber, 2003; Friden, 2003; Rose, 1994; Mohagheghi, 2007). It should be noted that changed mechanical muscle properties have also been seen in cases other than CP, although they were not considered to be the main cause. (Thilmann, 1991; Sinkjaer, 1994).

In addition to altered reflex responses to stretch, other neural adaptations have been noted in those with spastic CP.

Reciprocal inhibition (RI) between muscles and their antagonists is necessary for smooth coordinated movement. However a reduction or absence of RI or, in fact, reciprocal excitation, appears to distinguish the spasticity existing in CP (Gottlieb *et al*., 1982; Leonard, 1990).

Additionally, greater reflex activity, along with electromechanical delay (EMD) has been observed in EMG recordings from 12 young people with spastic CP. However, Granata and colleagues concluded that whilst increased biomechanical stiffness was the cause of the abnormally reduced EMD, reciprocal excitation of antagonist co-contraction was also present in the group with spasticity but not in the control group (Granata, 2002)

Another adaptation was seen in 23 children with spastic CP, when Leonard and Hirschfeld noted reflex irradiation to other muscles following patellar or Achilles tendon taps. This response was greatly reduced in the control group, if participants were over the age of two years (Leonard & Hirschfeld, 1995). Reflex irradiation does not appear to have been recorded in spasticity in cases other than CP.

One confounding observation in quantifying spasticity in CP is that no correlation has yet been shown between the degree of clinical spasticity and the level of tonic stretch threshold. This was apparent in two studies of young people with spastic CP, one where cutaneomuscular reflexes were recorded from trunk and lower limb muscles in 21 subjects, the other where EMG recordings were obtained from the elbow flexor muscles of 14 subjects (Gibbs *et al*., 1999; Jobin, 2000).

Another interesting feature which appears to distinguish the spasticity in CP is the possibility of consciously reducing the effects of increased reflex activity. One group of 15 children with spastic CP managed to reduce the stretch reflex gain from the triceps surae by around 50%, using visual feedback (O'Dwyer, 1994). This observation suggests that reflex activity was achieved through voluntary relaxation in the spastic muscle. This, in turn, suggests that the stiffness is not fixed, and may therefore respond to an intervention such as massage which is aimed at reducing the mechanical stiffness in spastic muscle.

1.7.3 Spastic muscle – mechanical changes

As mentioned above, Dietz and Berger suggested that only part of the resistance of spastic muscles to stretching can be attributed to increased reflex contraction, much is due to the intrinsic stiffness of the muscle itself (Dietz & Berger, 1983).

This resistance has 3 components – passive muscle stiffness, neurally mediated reflex stiffness, and active muscle stiffness. Of these, increased passive mechanical stiffness accounts for nearly all of the increase in limb stiffness (Sinkjaer & Magnussen, 1994; Lieber *et al.*, 2004).

Changes in the structure of spastic muscle in CP are to be expected. Skeletal muscle represents a classic biological example of the relationship between structure and function, since the structural characteristics of muscle are determined by its conditions of use (Lieber, 1986).

In 2004 Lieber and co-workers acknowledged that the basic mechanisms underlying the functional deficits that occur after the development of spasticity are not well understood, and that with a few notable exceptions, the properties of skeletal muscle have largely been ignored. However, it is becoming increasingly clear that there are dramatic changes within skeletal muscle as well as in the nervous system.

Although our current understanding of spasticity is incomplete, it is now acknowledged that spasticity has both neurophysiological and musculoskeletal components (Lieber *et al.*, 2004). The authors suggest that this is the reason why therapeutic interventions involving stretching, casting, splinting, neurectomy, intrathecal baclofen, botulinum toxin A and electrical stimulation have proved to be only marginally effective.

Because the basic mechanisms underlying the deficits apparent in spasticity are not well understood, the relative proportional contribution of reflex responses and mechanical stiffness has been controversial for some time (Foran *et al.*, 2005).

The total mechanical stiffness in a contracting muscle, measured during a stretch, is the sum of the response from the properties of the muscle fibres contracting prior to the stretch, the response from the stretch reflex-mediated contraction of the muscle fibres, and the response from the passive tissues (Sinkjaer and Magnussen 1994). In that study of spastic muscles in hemiparetic patients, the passive mechanical stiffness of the muscle itself was shown to be largely responsible for the increased resistance to stretch; only part of the resistance could be attributed to heightened stretch reflexes. These findings are in accord with extensive work by Deitz and others (Dietz & Berger, 1983; Lee *et al.*, 1987; Dietz *et al.*, 1991; Ibrahim *et al.*, 1993) which suggested that the mechanical properties of spastic muscle are abnormal. This would affect mechanical behaviour in both passive and active states.

Frieden and Lieber have said that there is no clear consensus regarding whether muscle cells from patients with spasticity have normal properties. They contend that this is due to the paucity of objective data regarding the mechanical, physiological or biochemical properties of spastic muscle (Friden & Lieber, 2003). Comprehensive details of the structural changes that occur in spastic muscle, as well as the underlying mechanisms for the changes are lacking (Lieber, 2004).

Foran and colleagues assert that ‘spastic’ muscles are altered in a way that is unique among muscle plasticity models and inconsistent with simple transformation due to chronic stimulation or use. They make the case for the following alterations in spastic muscle, 1) altered muscle fibre size and fibre type, 2) proliferation of extra-cellular matrix, 3) increased spastic muscle cell stiffness, and to a lesser extent spastic muscle tissue, 4) inferior mechanical properties of extra-cellular material, compared to normal muscle (Foran *et al*., 2005) (see also Fig. 1.3, Rose *et al* 1994).

1.7.4 Spastic muscle - mechanical changes observed in CP

A number of structural differences have been observed which distinguish spastic muscle in CP from normal muscle. These include changes in muscle fibre type, length and x-sectional area, and the contribution and quality of the connective tissue component.

Changes in muscle fibre type have been observed in spastic muscle although there is no general agreement that the spasticity present in CP represents either an increased or decreased use model. Four studies involving patients with CP showed an increased percentage in the cross sectional area of type I fibres (Dietz *et al*., 1986; Rose *et al*., 1994; Ito *et* *al*., 1996; Marbini *et al*., 2002). However, three other studies of biopsies from participants with CP showed no specific change in either type (Castle *et al*., 1979; Romanini *et al*., 1989 ; Booth *et al*., 2001).

Fig 1.3 shows a good example of the change in the proportions of the fibre types. The gastrocnemius muscle is normally considered to have around 75% type 2 fibres, whereas type 1 predominates in that muscle in a 5 year-old boy with CP.

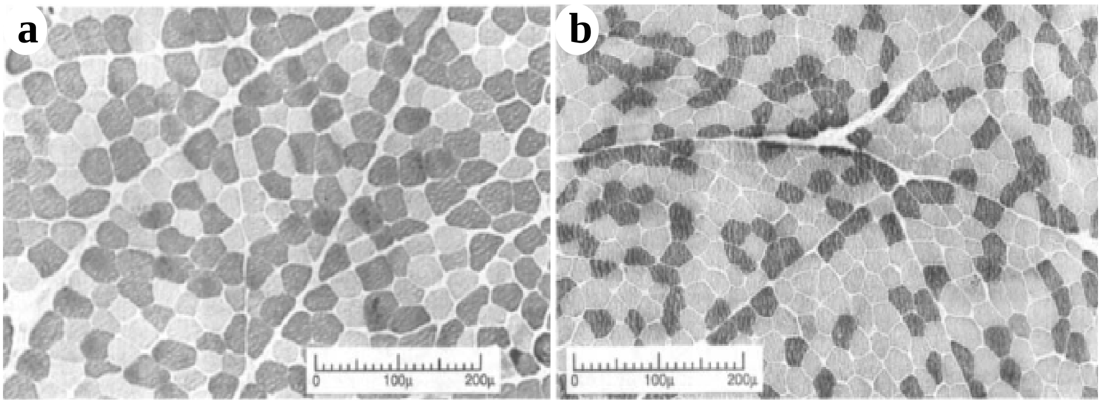


Figure 1.3 Comparison spastic muscle and control –altered proportions of muscle fibre type

Skeletal muscle stained with ATPase, pH 9.4. Type 2 fibres are dark. (a) Biopsy of the peroneus brevis muscle (x50) from a 7-year-old control subject. (b) Biopsy of lateral gastrocnemius muscle from a 5-year old boy with cerebral palsy. Type II fibres (so called fast twitch) are dark stained, showing approximately 50% of either type in a) However, type I predominates in (b) which is considered to have 75% type II fibres in healthy individuals (Adapted from Rose *et al*., 1994)

Additionally, muscle fibre bundles have been shown to differ from biopsies of spastic muscle in patients with CP who were about to undergo surgery. Only 40% of the spastic muscle bundle cross-sectional area was occupied by muscle fibres, whereas 95% of the normal muscle bundle was occupied by muscle fibre (Booth *et al*, 2001).

As well as the altered contractile component of the muscle fibre, other changes have been observed in CP muscle. The connective tissue, (mainly collagen 1) present in the muscle unit has been shown to be increased in the vastus lateralis muscle in young people with spastic CP and proportionate to the muscles' resistance to stretch (Booth *et al.*, 2001) (Fig 1.4).

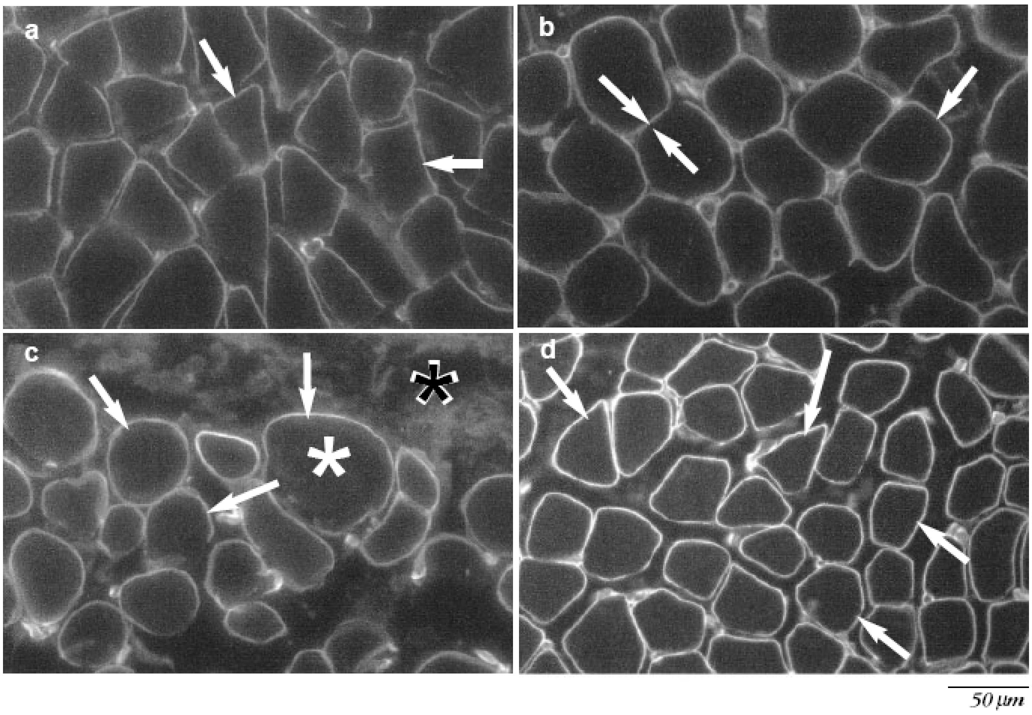


Figure 1.4
Comparison of mild to severe. Fibre diameter changes and collagen accumulation

Collagen I immunohistochemistry in spastic muscle of children with CP at different severities on Modified Ashworth Scale (MAS) and Balance (B). (a) MAS mild, B good; (b) MAS moderate, B good; (c) MAS moderate-severe, B poor; (d) MAS severe, B moderate. (Adapted from (Booth *et al.*, 2001).

This shows that the more severe the condition, the smaller the proportion of cross sectional area that the muscle fibres form. Compare the tight apposition between muscle fibres illustrated in (a) and (b) with the larger spaces between the muscle fibres formed by connective tissue in (c) and even more so in (d).

These authors suggest that collagen may be involved in increases in the muscle stiffness observed in spasticity and that its accumulation contributes either directly or indirectly to the development of contractures and secondary bony abnormalities, thus playing a major role in mobility problems observed in CP.

Other studies have shown that although spastic muscle contains a larger amount of extracellular matrix within it, the mechanical strength of that material is poor compared with that of normal muscle (Lieber *et al.*, 2003b; Lieber *et al.*, 2004).

Direct measurement of the mechanical properties of isolated muscle fibres of spastic muscles from patients with CP showed that although they are stiffer compared to normal muscle fibres, bundles of muscle fibres are actually less stiff compared to normal muscle fibre bundles (Lieber *et al.*, 2003b). Those authors concluded that this is because the extra-cellular matrix around the muscle fibres, examined in a group of 9 year-old children with CP, was shown to have inferior mechanical strength.

Muscle stiffness in CP has also been attributed to shorter muscle fibres resulting in overall shorter muscle lengths. However, it has been observed that the diameter of muscle fibres in CP is in fact smaller (Shortland *et al.*, 2002), as much as one third normal (Lieber *et al.*, 2003b). In pennate muscles such as gastrocnemius, because the muscle fibres are angled in relation to the overall length of the muscle, the smaller diameter of the fibres explains the overall shortness of the muscle. However, in a more recent study comparing spastic muscles in hemiparetic children with CP, muscle bundles were shorter on the affected side (Mohagheghi *et al.*, 2007).

Lieber and colleagues contend that two important clinically relevant questions still remain largely unexplored – ‘Is there a difference in the muscle response to different causes of spasticity?’, and ‘Is there an effect of age at which the spasticity is acquired on muscle properties?’ (Lieber *et al.*, 2004).

Whatever the relative contribution of altered neural and muscle components in spasticity, the management of the increased muscle tone is a key factor in rehabilitating children with CP (Jobin & Levin, 2000). Flett concurs with this view, suggesting that eliminating spasticity enables the child to utilise their selective motor control more effectively and functionally (Flett, 2003). Whilst this last view seems somewhat simplistic, it was the author’s view that the massage intervention might at least reduce the heightened muscle tone of the participants with CP.

1.8 Current interventions aimed at alleviating spasticity in CP

The brain damage in CP cannot be reversed, however maturational and adaptive processes may change the clinical picture of the child over time. Treatment for CP therefore focuses on how best to help the individual to maximise his or her potential (Ketelaar *et al.*, 2001).

1.9 Invasive Therapy – Surgery

This includes muscle/tendon lengthening, muscle attachment relocation, and multi-level orthopaedic surgery, which is currently being performed more commonly.

1.9.1 Multi-level Orthopaedic surgery

Recently there has been considerable growth in new multi-level orthopaedic operations to correct soft tissue and bony deformities in the lower limbs of CP sufferers. This follows from the observation that change in the resting angle at one joint necessarily affects others, especially evident in the simple act of standing erect. Depending on the severity of the condition, a single operation to correct deformity involving the limb as a whole is seen as more likely to be beneficial than a succession of surgical interventions. The surgery itself can involve as many as 12 different procedures, taking two teams of surgeons up to 6 hours. The children may not regain their preoperative mobility for several months, and will continue to need intensive physiotherapy and occupational therapy for at least a year (Morton, 1999). Nene observed that the success of the total treatment depends on effective physiotherapy as well as the intellect and personality of the patient. Morton and others suggest that the approach must obviously be evaluated further before being universally adopted (Nene *et al.*, 1993).

Gage and others have commented that surgery which does not preserve normal muscle function could add iatrogenic injury to the already physiologically burdened child with cerebral palsy (Gage, 1991).

1.9.2 Selective Dorsal Rhyzotomy (SDR)

In this surgery, which usually takes 6 hours or more, a laminectomy L1-L5 is performed and the dura opened to expose dorsal roots L1-S2. Each one is then separated into 12 or more rootlets and divided if they appear to be associated with spasticity as determined by an abnormal electromyography response (Morton, 1999). However the underlying theory that sensory axons with particular central connections are arranged into discrete rootlets has been challenged (Landau, 1974).

The surgery itself may offer only marginal added benefit over the intensive physiotherapy, which is required following surgery and, in view of the impact on the child and continued complications, is of questionable value (Morton, 1999). SDR is still widely practised in USA for selected patients, often not necessarily the worst cases. However long term results are raising doubts about the sustained benefit (Lin, 2003).

1.10 Pharmacological Approaches

Pharmacological interventions aim to reduce the symptoms of CP by altering the effects of neurotransmission either at spinal cord level or at the neuromuscular junction.

1.10.1 Baclofen

Baclofen, which is marketed under the trade names ‘Baclospas’ and ‘Lioresal’ is a GABA agonist selective at presynaptic GABA beta receptors.

The antispastic action of baclofen is exerted mainly on the spinal cord where it inhibits both monosynaptic and polysynaptic activation of motor neurones. It is given by mouth or more commonly, because it penetrates the blood brain barrier poorly, by continuous intrathecal infusion (CIBI).

In CIBI, the pump is fixed in a subcutaneous pocket in the abdomen and is connected to a catheter tunnelled under the skin and inserted into the intrathecal space in the lumbar region, ending around T12. Every three months the pump is refilled with baclofen by needle injection.

Side effects include drowsiness, motor inco-ordination and nausea. It may also have behavioural effects (Fehlings *et al.*, 2001). The effects of baclofen are time limited and the cost has also to be considered. Steinbok and others have estimated CIBI to cost three to four times more than SDR in the first year (Steinbok *et al.*, 1995).

Morton and others have concluded that CIBI is associated with a significant number of complications in all patient groups, and that there is clearly a need for controlled trials in children (Morton, 1999).

1.10.2 Botulinum Toxin A

Botulinum Toxin A (Trade name 'Botox Dysport') is a protein produced from Clostridium botulinum. It is injected into multiple sites in the muscle and causes paralysis by blocking pre-synaptic acetylcholine release into the gap at the neuromuscular junction. It works by cleaving a specific protein (SNAP 25) involved in exocytosis and blocks synaptic function for 12 to 16 weeks. Recovery of the neuromuscular junction occurs by means of compensatory proximal axonal sprouting and takes place over 6 to 8 weeks in the experimental animal (Jefferson, 2004).

A number of reviews have attempted to evaluate the effectiveness of Botox since its initial use in the 1970s and these have produced conflicting results (Ade-Hall & Moore, 2000). An additional problem with Botox treatment is that it may be injected into the wrong muscle.

Long term use and effectiveness of Botox has yet to be validated and a cautious approach is advocated by Gough and colleagues until further evidence is available (Gough *et al.*, 2005).

1.11 Non-invasive Therapy

Physical therapies form a large part of meeting the needs of children with CP. Non-invasive therapies generally have the objectives of preventing deformity and encouraging normal growth patterns.

1.11.1 Physiotherapy

In developed countries, each child with CP has a specific record of needs drawn up by the child's consultant in conjunction with other professionals and the child's parents/carers. This includes a programme of physiotherapy tailored to their particular stage of development.

Physiotherapy is used as the primary non-invasive therapy but may also be used in conjunction with invasive therapies, for example in remobilisation following splinting after surgery.

The standard approach in physiotherapy clinics includes stretching and strengthening, balance and gait exercise, postural work (in and out of their wheelchairs), heat and ultrasonic/electrical procedures, hydrotherapy, and trampoline rebounds.

Throughout the history of physiotherapy, therapists have been challenged to provide evidence that their interventions work. In the last 30 years there have been repeated calls for research into the effectiveness of physiotherapeutic procedures specifically for the management of cerebral palsy (Mead, 1968; Taft, 1972; Pless, 1976; Pearson, 1982).

A limited number of trials have been conducted more recently. In 1992, Bower related that only three major studies had been undertaken on children with an established diagnosis of CP (Wright & Nicholson, 1973; Palmer *et al.*, 1988; Scherzer, 2001) and they showed either inconclusive or negative results (Bower & McLellan, 1992). A later study by Bower investigated the effect of different intensities of physiotherapy in 44 children with CP. They concluded that intensive physiotherapy produced a slightly greater effect than conventional physiotherapy, but physiotherapy directed to specific measurable goals resulted in increased motor skills acquisition (Bower *et al.*, 1996). A further study by Bower on 56 children with CP concluded that there was no measurable difference in the effectiveness of intensive physiotherapy against collaborative goal setting (Bower *et al.*, 2001).

In 2002 Trahan stated that over the last 15 years, reviews focusing on the effectiveness of rehabilitation programs for promoting motor development in children with CP had been inconclusive. These authors conducted a pilot study which concluded that intermittent intensive physiotherapy did indeed lead to improvements in motor function in five children with CP. A larger study is needed to consolidate those results (Trahan & Malouin, 2002).

Although there is limited scientific evidence to demonstrate the effectiveness of physiotherapy, it is also not known how much worse the child's condition might be without it.

1.11.2 Conductive Education

Conductive education (CE) is a learning system developed at the Peto Institute in Budapest, Hungary, designed to enable children and adults with disabilities to function independently. British therapists and teachers have used elements of the system for around 30 years in their work with school-age children with CP. The child's daily routine includes several series of tasks carried out in different positions, for example the sitting, lying and standing/walking positions. The emphasis here is to encourage good motor patterns and to discourage/alter poor ones.

A limited number of scientific studies have been carried out. Darrah and colleagues reviewed 15 studies carried out between 1972 and 2000 and concluded that the present literature does not provide conclusive evidence either in support of or against CE as an intervention strategy (Darrah *et al.*, 2004). Reddihough and co-workers, in their study of 66 young children, concluded that those involved in CE based programmes made similar progress to those involved in conventional programmes (Reddihough *et al.*, 1998). Massage was a core part of this treatment in Hungary. However it is not included in the approach used by conductors in the UK. (See chapter 6 for further comment on this).

1.11.3 Bobath

Also known as Neurodevelopmental Therapy (Knox & Evans, 2002), this treatment is based on the premise that the fundamental difficulty in CP is lack of inhibition of reflex patterns of posture and movement (Bobath, 1985). Here abnormal patterns are thought to be associated with abnormal tone due to over reaction of tonic reflex activity. Thus the main focus is on the treatment of tone in order to prepare for movement.

Results from the limited scientific studies are varied, with reports often showing opposing results. The American Association for Cerebral Palsy and Developmental Medicine (AACPDM) report in 2001 suggests that further, larger studies are needed with more homogeneous subject groups (Butler & Campbell, 2000). Law and colleagues found no improvement for upper limb function, using the Peabody Fine Motor Scale (Law *et al.*, 1997). Knox and Evans found gains in gross motor function using the Gross Motor Function Measure-66 and The Pediatric Evaluation of Disability Inventory (Knox & Evans, 2002). Tsorlakis found gains in motor function, with emphasis on the intensity of the intervention

(Tsorlakis *et al.*, 2004). They also used the Gross Motor Function Measure-66.

1.11.4 Electrical stimulation (ES)

This is used less often than the previously mentioned treatments and is used in two different forms. The first, called neuromuscular electrical stimulation (NMES), is the application of an electrical current of sufficient intensity to elicit muscle contraction. Contraction occurs through the stimulation of the intramuscular branches of the nerve supplying the muscle. Functional Electrical Stimulation (FES) is a type of NMES in which the stimulation is applied when the muscle should be contracting during a functional activity (Kerr Graham & Selber, 2003). The second is Threshold Electrical Stimulation (TES) where a low level, sub-contraction electrical stimulus is applied, usually at home, during sleep (Pape *et al.*, 1993).

Trials have been conducted into the efficacy of ES in the last 20 or so years. Varying results have been reported using a wide range of outcome measures both specific and empirical. However the scarcity of well-controlled trials makes it difficult to support definitively or discard the use of electrical stimulation in the paediatric CP population (Kerr Graham & Selber, 2003).

1.12. Comparison of efficacy of current treatments

Table 6.6 (in Discussion) compares the efficacy of current treatments which used the same main outcome measure (GMFM) as was used in the present study.

1.13 Massage – Background and use in Rehabilitation and in CP

Tradition defines massage as hand motions practised on the surface of the living body with a theraputic goal. The first mention of massage appears over 4,500 years ago in the Nei Ching, the oldest existing medical work (Gifford & Gifford, 1998). Hippocrates, considered the father of medicine, is quoted as saying 'The physician must become expert in all aspects, especially in the rubbing' (Wakim, 1976).

Massage fell out of favour for more than a thousand years following the social decadence of the Roman Empire. The modern era of massage began in 1863 with the publication of a treatise systematically classifying each technique according to the body system affected (Wakim, 1976).

The type of massage most widely used in rehabilitation, and the one used in this thesis is Swedish massage, also known as Western massage or Classical massage. The strokes used in Swedish massage were formalised around 1880 by a Swede called Per Ling. This employs four main strokes or variations of them – Effleurage (long slow stroking movements done with the palms of the hands and pads of the fingers moulded to the contour of the body part being worked on), Petrissage (picking up, rolling, wringing and kneading), Frictioning (small circular movements done with the tip of thumb or fingers), and Tapotement (percussion movements such as cupping and hacking (Pemberton, 1945).

In the UK, massage was first used in patient rehabilitation around 100 years ago. A number of eminent surgeons in London found that their patients were dying post operatively, although the surgery itself had been successful. This was eventually attributed to circulatory problems aggravated by extended periods of immobility due to prolonged bed rest. Consequently women masseurs were employed to give massage, and this resulted in significantly improved mortality rates.

In 1894 the British Medical Journal had raised concerns about the practices of some masseuses and masseurs who were offering massage as a euphemism for sex. The BMJ called for an institution to be formed to regulate massage, and the 'Society of Trained Masseuses' was formed by nurses and midwives keen to see their massage practices authenticated.

In 1920 the critical work of these massage practitioners was recognised when the Chartered Society of Physiotherapists was formed. Unfortunately for proponents of massage, the advent of the NHS in 1948 saw massage used less and less, as electric and later electronic equipment were considered more time efficient and physically less demanding for the therapist. Consequently, physiotherapists received less training in massage. In the UK, current trainee physiotherapists receive 10 hours tuition in their four-year degree course.

In the USA, Colby, followed by Crothers and by Phelps, all used massage in rehabilitation programmes for CP sufferers during the 1930s, with Phelps going on to develop a paediatric rehabilitation centre in Maryland, USA (Slominski, 1984).

Most of the limited published work on the effectiveness of massage is documented in relation to its effect on sports performance. However, there are few, well controlled studies that have examined the potential for massage to influence performance, recovery or injury risks (Weerapong *et al.*, 2005). Those authors concluded that further research is needed which includes an appropriate control group, where a counterbalance design is used to minimise different responses of individual participants, and appropriate outcome measures and massage techniques are used.

Although empirical results had shown the effectiveness of massage in CP, no scientific studies were recorded until the 21st century. In the first (very short) study only qualitative measures were used (Stewart, 2000). After massage it was noted that one child smiled more; another had looser limbs and displayed an open hand, which had been a tightly clenched fist prior to the massage. That author concluded that massage techniques can reduce symptoms associated with CP and help improve the quality of life for these children.

In the only controlled study to date which uses modifications of Swedish massage, the authors concluded that massage attenuates the symptoms associated with CP, enhances development and suggest that it should be considered as an early intervention for children with CP (Hernandez-Reif *et al.*, 2005). In that study, 20 participants received 30 minutes of massage or reading twice weekly for 12 weeks. However, as the mean participant age was 32 months, it is possible that some/all of the improvements could have happened spontaneously. (Rosenbaum *et al,* 2002*,* have shown that motor skills in children with CP tend to plateau around the age of seven). 90% of participants in that study had spasticity although it is not noted how many had diplegia, and so the population may not be as homogenous as it could have been. The Modified Ashworth Scale was used as a measure of reduction in spasticity. However, this relies on the therapist gauging how much resistance is felt to stretch, and consequently this is more of a qualitative measure. The Gross Motor Function Measure-66 was not used, despite being widely recognised for assessment of changes in motor function, specifically with those who have CP. Additionally, range of movement at the hip joint was measured although this is difficult to gauge as movement is possible in more than one plane. Consequently, the selection criteria for the current thesis stipulated only adolescent participants with spastic diplegic CP, changes in range of movement at the ankle joint (one plane only), and the GMFM-66 for measuring motor function.

Massage has been used for thousands of years for treating musculoskeletal problems. Despite this, there has been very little scientific research into its efficacy in any condition. However the author, who has 45 years experience in the use of massage, has had empirical results showing that massage improved motor function in two members of his family who have CP. I have also observed improvements in motor function following massage with others who have CP, although again no scientific measurements were used. Additionally, a palpable reduction in the tone of the massaged muscles was evident some times during and almost always following massage.

Consequently the investigations focussed on whether massaging spastic muscles using a specific timed sequence of massage would change their mechanical behaviour. This in turn would be likely to alter the feedback from the muscles to the CNS, allowing the possibility of better motor function.

1.14 Devising a Suitable Massage Sequence

Unfortunately there is no literature confirming the efficacy of specific sequences of massage. Consequently the sequence used in this thesis is based on the author's 40 years of practical experience in treating a wide range of musculoskeletal problems, and also from the massage used with a limited number of people with CP, already mentioned. However some general principles were followed.

Generally speaking massage has the effect of stretching the muscles mostly longitudinally and, this usually has a beneficial effect when used on damaged muscles, for sports preparation or for releasing tight muscles.

However, a pivotal point arises when trying to devise a theraputic intervention that involves manipulation of spastic muscle. A major difficulty for children with CP is that the very act of stretching the tight muscles often causes unhelpful stretch reflexes to occur. This can be felt when the patient flinches as the muscle contracts involuntarily. This results in further contractions of already shortened muscles.

Consequently, in order to avoid eliciting stretch reflexes, the devised sequence includes a number of massage strokes intended to stretch the muscles locally in a transverse direction, rather than longitudinally. Additionally, the sequence of strokes moves gradually from ones which are slow, smooth and not too deep, to ones that are faster, deeper and more vigorous. If the vigorous strokes alone were done, without preparation, the muscles would tend to contract by reflex in response to movement that is too forceful or too quick. The subsequently devised massage sequence was designed to alter the resting properties of the muscle without invoking stretch reflexes.

The massage sequence applied to the triceps surae was the same for all of the studies. It has precisely timed strokes for standardisation of the procedure and ease of replication (table1.1).

Massage stroke	Duration (Mins)	Strokes per minute
Effleurage	2.0	15
Pick up	1.5	24
Pick up and hold	1.5	12
Rolling	2.5	15
Kneading	1.5	Slow/Medium pace
Appositional Rolling	2.0	60
Wringing	1.0	Vigorously
Effleurage	2.0	15

Table 1.1 Massage sequence – calf muscles

Each stroke listed in the left hand column of table 1.1 is recognised as a 'Swedish' or classical massage stroke. The second and third columns show the specific duration and rate of application of each of the massage strokes. They are designed specifically to stretch the muscle transversely in small sections and not to invoke stretch reflexes during their application.

1.15 Hypothesis

As detailed above, current treatments that target the damaged neural component in CP have met with very limited success. The spasticity has been shown in some studies, though not all, to be reduced by some current therapies (Intrathecal Baclofen, Botox and Selective Dorsal Rhyzotomy) (Wright *et al.*, 1998; Butler & Campbell, 2000; Boyd & Hays, 2001). Even when the spasticity is reduced, the gains in gross motor function are modest. It has been proposed that the problem may not lie entirely with neural mechanisms that these interventions target, but that abnormal physiological properties of spastic muscle could contribute (Dietz *et al.*, 1986; Lin *et al.*, 1999; Lieber *et al.*, 2004).

Neural plasticity has been demonstrated following an intervention with participants who have CP. Functional MRI scans showed increased cortical activation in those who underwent weight bearing treadmill exercises (Phillips *et al,* 2007). If massage could improve the mechanical properties of the spastic muscle, the feedback to the CNS is also likely to be improved. Given the capacity for neural plasticity in this participant group, better motor function following massage then becomes a possibility.

1.15.1 Aims

The lack of documented evidence for the effectiveness of massage for individuals with CP is the very reason that this thesis was undertaken. As there is evidence that the altered mechanical properties of the spastic muscle appear to be the main contributor to stiffness in addition to altered reflex responses, the effects of massage on both of these were investigated.

The literature has shown that the properties of spastic muscle in CP and its response to stretch are altered (higher incidence of stretch reflexes), although the original damage occurred to the nervous system (Dietz *et al*, 1986, Lin *et al,* 1999, Lieber *et al* 2004). Consequently, I devised a sequence of massage strokes which were designed to change the resting state of the muscles and to avoid invoking stretch reflexes during their application.

The aims were:-

To investigate the effects of massage on the mechanical properties of the spastic calf muscles, including its response to stretch (chapter 3).

As motor skills and range of movement and have also been shown to be compromised in spastic CP (Bottos & Gericke, 2003, Shortland *et al*, 2002), these were also investigated (chapter 4).

Having observed changes in the spastic calf muscles and improvements in motor skills in the initial studies, the investigations then focussed on possible mechanisms underlying the changes.

Massage has been shown to increase the temperature of the erector spinae muscles (Longworth, 1982) and the vastus lateralis muscles (Drust, 2003). As increases in muscle temperature have been linked to improvements in sports performance (Bergh & Ekblom, 1979), the effect of massage on the temperature of spastic calf muscles was investigated (chapter 5). Additionally, it has been postulated that massage may produce its effects by altering the blood flow of the massaged tissue (Goats, 1994). Consequently, chapter 5 also included an investigation into the effects of massage on the oxygenation of the massaged muscles.

Having noted the efficacy of the massage, the final aim was to determine if the massage technique employed by the author could be learned by another therapist (chapter 5).

In this way, the ultimate aim of this thesis might be realised; that if scientific study showed that massage was beneficial for those with CP, it might be more widely accepted into the physical therapy input for young people with CP, thus reducing the debilitating effects of their condition.

CHAPTER 2

Materials and Methods

2.1 Over all design of the studies

The aims of this thesis can be summarised as investigating the effects of massage on spastic muscle in CP and determining some of the possible mechanisms underlying the changes observed after massage.

Prior to the start of this work, no other comparable studies had been documented. Consequently no overall study design was formulated prior to the completion of the first of the studies, rather each study followed on from the previous one as the results of each were analysed. Six studies were conducted to assess effects of specific massage sequences on the leg muscles in adolescents with spastic diplegic cerebral palsy. A summary of those studies follows, along with the rationale for their formulation.

Study 1 - Effects of massage on muscle properties and motor function

Study 1 assessed the effects of calf muscle massage on motor function and the response of the spastic muscle to stretch following massage.

5 adolescent participants with spastic diplegia received the calf muscle massage sequence twice per week for five weeks. The massage was tested for its effects on stretch reflex responses and muscle length changes (chapter 3). The effects of massage were also tested on active ankle

movements and on motor skills, using the Gross Motor Function Measure-66 (GMFM-66) (chapter 4).

At the time of this study, only these 5 participants were available and met the recruitment criteria. The statistician who was consulted before the studies began, confirmed that an improvement of 5% or more in GMFM-66 scores would be statistically significant with as few as 5 participants. The four ambulant participants did improve their GMFM-66 scores (mean increase 6.0). See chapter 4. As more participants became available, it was decided that additional participant numbers would add to the significance of the results.

Study 2 - Effects of calf muscle massage on motor function

Having observed improvements in motor function in four of the five participants in study 1, study 2 investigated the effects of calf massage on motor function in a further 6 adolescents with CP. They were tested with the GMFM-66 (chapter 4). All of the participants in this study showed improved motor function (range: 2.89 – 7.71, mean increase – 5.0).

Study 3 - Effects of full leg massage on motor function

It was felt that the two most disabled participants (both non-ambulant) might benefit from full leg massage as opposed to massage of only the calf muscles. One of these participants, who had no voluntary ankle movement, had already received calf muscle massage for 10 sessions over 5 weeks as participant number 5 in study 1. He had shown no appreciable improvement in GMFM-66 score in that study. Both participants were tested with the GMFM-66 (chapter 4) which showed statistically significant improvements in motor function for both participants.

Study 4 - Effects of self-administered full leg massage on motor function

Once improvements in GMFM-66 scores were noted following full leg massage sessions, options were explored to implement massage sessions at home. Parents or carers could possibly give massage if they were trained in the technique, but some of the adolescents indicated that this option was not to their liking. However, the prospect of participants giving themselves full leg massage during school hours was welcomed by three participants.

It was considered that three of the more able participants with CP might be able to administer self massage to the full leg. They had been part of the earlier study during which they received calf muscle massage, 18 months earlier. The full leg self massage was done in 33 sessions spread over 74 weeks. The assessment with the GMFM-66 and the results are discussed in chapter 4.

The focus then centred on investigating possible mechanisms underlying the changes observed following massage.

Study 5 – Effects of calf muscle massage on skin temperature

Increases in skin temperature following massage have been observed often by therapists, although no scientific measurements are taken. Increases in muscle temperature have been linked to improved athletic performance. It was of interest to assess if the massage applied here increased skin temperature, and if this might be linked to improved motor function in those participants with CP. Additionally, the change in muscle temperature was used to determine the efficacy of a trainee masseur. The results showed that temperature over the massaged muscles was consistently higher after massage than before (chapter 5).

Study 6 - Effects of calf muscle massage on skin temperature and oxygenation

It has been postulated that massage may alter the blood flow of the massaged tissue (Goats, 1995). This, coupled with the temperature increases observed in study 5, prompted an investigation into the effects of massage on oxygenation in the massaged muscles.

By this point, a control group had become available. Two groups of 6 adolescents, one with CP and an age and sex matched control group of healthy adolescents received the calf muscle massage sequence. They were tested for changes in skin temperature and oxygenation of the massaged muscles, and whether these were related. The results are discussed in chapter 5.

2.2 Conditions common to all of the studies

2.2.1 Ethics

Ethical permission was obtained from the ethics committees at Yorkhill Hospital and the Western Infirmary, Glasgow and the Faculty of Biomedical and Life Sciences, University of Glasgow.

All the adolescent participants gave informed assent to participate in accordance with the Declaration of Helsinki. Written informed assent for the spastic diplegic participants was obtained from the participants, their parents, general practitioners and their paediatric consultant at Yorkhill Hospital, Glasgow. Written informed assent was also obtained from the healthy adolescents and their parents. Informed consent was obtained from the adult participants. None of the participants subsequently withdrew.

The studies took place at Ashcraig and Richmond Park special needs schools, and Hutchesons' Grammar School, all in Glasgow, and at the University of Glasgow. Glasgow City Council Education Services and senior management of the schools gave their approval for the studies to be conducted at the schools.

2.2.2 Participants with CP

As already alluded to, there is wide diversity in the way CP manifests itself. Consequently, all of the participants with CP in the present studies were chosen because they have a diagnosis of spastic diplegia. Although each young person is affected in a specific way, this leads to as homogeneous a group as possible and thereby reduces the variables in this investigation. Although it was expected that the massage was likely to prove beneficial with all age groups (personal experience), to further reduce the variables, the participants with CP in these studies were restricted to adolescents. There were three reasons for this. 1). Gross motor ability is not expected to improve spontaneously above the age of seven years (Rosenbaum *et al.*, 2002). 2). In fact, gross motor function is likely to deteriorate as young people with CP move into adolescence (Morris, 2002). Thus any improvements in motor function are more likely to be due to the massage intervention. 3). Adolescents are approaching a stage in their lives where their disability could further handicap them e.g. in the work place. Improvements in motor function at this age are likely to better prepare them for adult life.

Whilst CP results in cognitive impairment in around 30% of all CP cases, it was considered that the participants should not be cognitively impaired in order that they might give informed consent, be able to follow instructions for active movements, and be able to give clear meaningful feedback.

2.2.3 Participant attendance

As no previous comparable work had been done, the number of sessions and the interval between the sessions had to be decided somewhat arbitrarily for these studies. Previous work (unpublished) by the author suggested that three sessions per week were likely to be most beneficial. However, in order to accommodate the school timetable, it was decided that wherever possible, a maximum of two sessions of massage per week would be given.

In study 1, participants attended for two sessions per week for five consecutive weeks. This was considered a minimum which pupils could reasonably be expected to be excused from their scheduled class work, and which was likely to be sufficient time to allow for change to happen in response to the massage. In the remaining studies participants were given the massage over a varying number of sessions, dependent on their availability, and also to afford the more disabled participants more treatment sessions. Their attendance is noted separately for each study. The reasons for the use of the particular type of massage and the chosen sequence are addressed in the individual chapters.

2.2.4 Study design - Use of controls

One possibility was that the participants could act as their own control where the effects of relaxation were tested against massage. This option was not used, as it was felt unethical to subject the participants to sessions detracting from their school lessons, when it was unlikely that the relaxation alone would produce improvement. Comparison of one leg with the other leg was also avoided as there was likely to be a contralateral effect of the massage from the treated leg to the other one. This proved to be the case when the results were analysed later. Additionally, it is very unusual for both legs to be affected by CP to the same extent. The participants with CP did, however, act as their own controls in studies 1-4 when the effects of the massage on motor function were assessed using a 'before - v - after intervention' study design. By the time study 6 was undertaken a comparable group of healthy adolescents had been recruited and acted as a control.

2.2.5 Conditions for massage and testing

A room affording quietness and privacy was set up at each venue with the treatment table and equipment suitably sited for ease of recording. The length of each session took cognisance of the participants' curricular activities, but averaged around 50 minutes. The procedure and equipment were explained to the participants at the beginning of the session and prior to each piece of equipment being attached to them. Shoes, socks and where relevant, orthotic splints were removed and trouser legs rolled to above the knee. Where full leg massage was given, the participants wore shorts. They lay prone on a treatment table, a hydraulic table for disabled participants, and a portable table for able-bodied ones. Their legs extended with the feet beyond the end of the table, supported on a pillow to allow the ankle joint to adopt its natural resting angle, and to allow unhindered movement of the joint. Only one therapist (the author) performed the massage for the adolescents. As massage often has a soporific effect, the participants were kept mentally alert throughout each session by engaging them in light conversation. A chaperone was present where the masseur might otherwise be alone with female participants.

2.2.6 Identifying muscles suited to receiving massage

Whilst CP can affect all areas of the body, a decision had to be made on which area(s) would be most suited to being massaged.

Visits were made to 10 special needs schools in Glasgow and initially around 60 pupils were considered for inclusion in our studies. It appeared that improvement in movement of badly affected upper limbs was most likely to benefit young people of school age. For example, they might be able to use wall-mounted touch screen computers, if the spasticity in the elbow flexors were reduced. However, because of the complexity of movement of the upper limb as a whole, it was considered too difficult at this stage to devise and implement a protocol that could be used and measured effectively within the confines of the schools' timetable.

Literature highlighting the deficiencies in ankle movement prompted a decision to massage the calf muscles. In a study of 7 children (mean age 10 years) with spastic dilpegia, ankle joint angles at rest were 20° more plantarflexed than normally developing children (Shortland *et al.*, 2002). Those authors concluded that some element of the calf musculotendinous unit must be short, resulting in toe walking. The equinus observed in that study is common in young people with CP. 80% of the children who exhibit equinus have problems with walking as a result of lower limb spasticity (Gage, 1991). Consequently massage of the calf muscles was investigated to ascertain if this would alter the resting angle and/or the range of movement at the ankle joint. Their ability to walk was also measured before and after a series of massage sessions.

The advantages of using the calf muscles in the initial study are, firstly, the ankle joint provides movement in one plane only. Secondly, exposure is minimal and easily carried out to avoid possible embarrassment. Third is the obvious importance of the calf muscles to gait in walking.

2.2.7 Application of massage

In diplegic CP, it is common for right and left limbs to be affected to a different extent. If massage was likely to be beneficial, it was considered important to massage both legs, in fact, it was considered unethical to do otherwise. For these reasons the option of using one leg as a control was not taken up. Throughout the series of studies both legs of the participants with CP were massaged, unless unavoidable time constraints or equipment problems made this impossible within their school timetable.

The triceps surae muscles of both legs were massaged using the sequence of strokes that emphasised gently stretching the muscles mainly in a transverse direction rather than longitudinally. Each sequence of Swedish massage lasted 14 minutes and involved seven timed changes of stroke, beginning and ending with effleurage, with five variations of petrissage in between (table 1.1). The right leg was always massaged first. Each session lasted 50 minutes, including tests.

The details for the massage specific to each study are noted in their relevant sections. The massage sequences for full leg and self administered full leg massage are listed in tables 2.2 and 2.3 respectively.

2.2.8 Assessment of effectiveness of therapy

Until recently, classifications of CP have been simply “mild,” “moderate,” or “severe” with no specific detail of what the child was expected to be capable of (Scherzer, 2001).

Hypertonia, an abnormal increase in muscle tone, is regarded as the defining feature of spasticity having both diagnostic and theraputic significance (Lance, 1980; Katz & Rymer, 1989). However quantitative objective valuation of muscle tone remains difficult since there is as yet no universally accepted means of measuring it (Mirbagheri *et al.*, 2001).

Previously devised methods of assessment often involved a degree of subjectivity from the tester. For example the Ashworth Scale, devised in 1964 and widely used for over 30 years, has the tester assess the degree of resistance felt on a scale of 0 – 4 (Ashworth, 1964). A score of 0 indicates no increase in tone, and a score of 4 indicates that the limb is rigid in flexion or extension. Good inter-rater reliability is possible (Damiano *et al.*, 2002) but the test gives no indication of whether the child’s motor skills have changed.

In proposing a new treatment for the CP condition, we were obliged to use existing recognised methods of measuring change for comparison with our work. A number of methods were at our disposal, however not all of them have been shown to be reliable (Mirbagheri *et al.*, 2001).

2.2.9 The Gross Motor Function Measure-66

Because spasticity is so difficult to quantify, it was decided that it might be prudent not to attempt measuring changes in spasticity, but to assess the ability of the subjects *to make use* of any changes following massage. The instrument used was the Gross Motor Function Measure-66 (GMFM-66) to assess motor skills. This method is now widely used and has been thoroughly validated and designed for use specifically with those who have CP (Russell *et al.*, 2000). This test was done before a block of massage sessions, after the block was complete, and for some subjects, at intervals after that.

The GMFM-66 has been designed specifically for use in assessing lower limb function in children with CP. It employs a series of 66 tests of specific motor tasks that are graded in degree of difficulty and for which each is scored 0-3. A score of 0 = does not initiate, 1 = initiates, 2 = partially completes, 3 = completes. The Gross Motor Function-66 user's manual provides a specific descriptor for the way that each task is to be performed. The tests areas are divided into 5 progressively more complex dimensions, A). Lying and Rolling - 17 items tested, B). Sitting - 20 items tested, C). Crawling & Kneeling - 14 items tested, D). Standing – 13 items tested, and E). Walking, Running and Jumping – 24 items tested.

The GMFM-66 has been designed so that not all of the 66 items have to be attempted in order to calculate an overall score (0-100). Of equal importance is the fact that the intervals between scores are of the same value no matter where a child's score is on the continuum from 1 to 100. This allows for comparison of the effects of an intervention even if the children are not necessarily affected by CP to the same extent. In order to interpret the scores for the GMFM-66 a computer programme is required. This scoring programme is called the Gross Motor Ability Estimator (GMAE), so named because it provides an estimate of the child's gross motor ability based on the child's scores on the GMFM items actually entered. The child's GMFM-66 score is calculated along with the standard error of measurement (SEM) and the 95% confidence intervals (CI) around the child's score (95% CI = ± 1.96 x SEM). The confidence intervals give an indication that the child's score is highly (95%) likely to fall between the upper and lower bounds of this confidence interval. Thus a change is considered significant where there is no overlap between the upper limit of the one assessment score and the lower limit of a subsequent score (see figs A1 and A2 in Appendix for example score sheets).

GMFM-66 is a very important measurement tool because it assesses whether the child with CP can *make practical use* of any changes following an intervention.

2.3 Material and Method for studies 1-6

2.3.1 Study 1. Calf muscle massage

The massage sequence outlined in table 1.1 was applied to both legs, right leg first. Participants were assessed for gross motor function, using the GMFM-66, before and after the completed block of massage sessions. They were assessed for response to active and passive calf muscle stretch before and after each massage session using EMGs from the soleus muscle. Range of ankle movement was measured at each massage session using electronic goniometry.

2.3.2 Participants

Five adolescents with spastic diplegia (three males, two females, age range 12-15 years, mean age 14 years) were recruited from Ashcraig Special Needs School in Glasgow. At this time, only these five met both inclusion criteria (i.e. they were adolescents with spastic diplegia, and cognitively unimpaired) and could accommodate participation in our study within their school timetable.

2.3.3 Attendance

All five participants attended two massage sessions per week for five consecutive weeks.

2.3.4 Application of Massage

Each massage stroke was applied directly to the soleus and gastrocnemius muscles in a distal to proximal direction, including the Achilles tendon and finishing over the popliteal fosa. Although it was probable, it was not possible to assess how much effect the massage had on the deeper muscles of the lower leg (tibialis posterior muscle, the toe flexor muscles, popliteus muscle and plantaris muscle, fig 2.1).

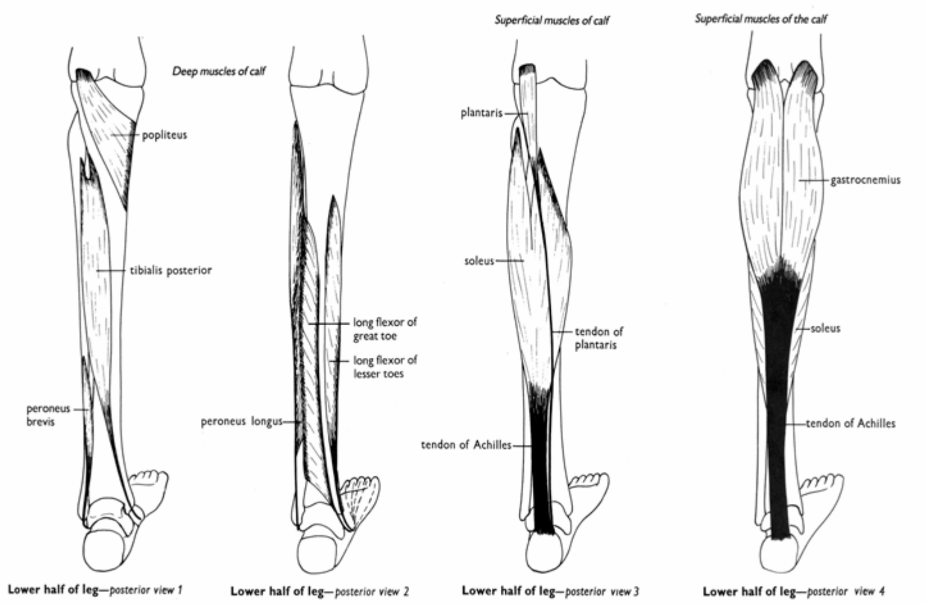


Fig 2.1 Posterior muscles of the lower leg.

This shows all of the posterior muscles of the lower leg, from the deepest muscles on the left to the most superficial on the right. (Rowatt, 1992).

2.3.5 Passive stretches

Immediately before and after massage, the ankle was passively dorsiflexed from the resting position three times with the knee at a right angle and the participant prone (Fig 2.2). A pressure pad 7cm in diameter was placed on the sole of the foot, centred over the metatarso-phalangeal joints. Force was applied at a controlled rate (around 8 N/s) by matching a force trace on a computer screen with a linearly increasing template. The pressure was applied slowly in order to reduce the possibility of invoking stretch reflexes, although nevertheless some stretch reflexes were recorded during the stretches (see below). Later statistical analysis showed that the force profile was being followed consistently (see p87).

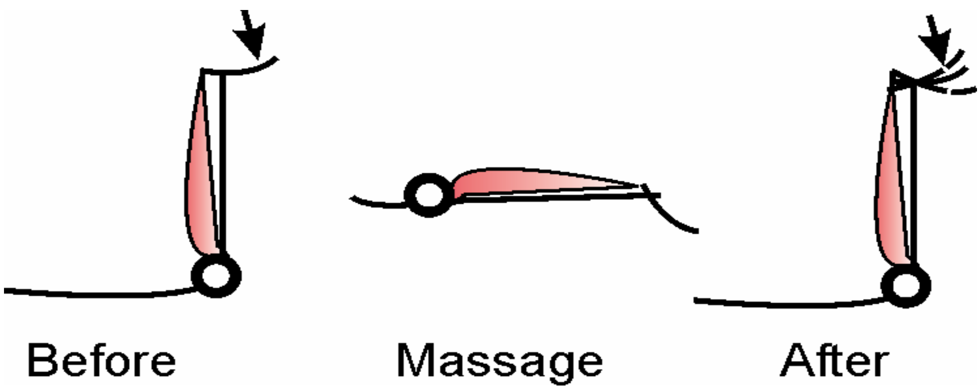


Fig 2.2 Limb positions during massage and stretches

This shows the lower limb position with the participant lying prone. 'Before' – Three stretches are done with the knee at a right angle; the arrow indicates the direction of force through the dynamometer on the sole of the foot. 'Massage' - The massage is then done with the lower limb resting with the knee extended along the treatment table. 'After' – three stretches done as 'Before.' Illustration suggests that the angle is extended further after massage.

2.3.6 Active ankle movements

With the knee flexed at 90°, immediately following three passive stretches, participants flexed and extended their ankle voluntarily in time with a metronome set at 40 beats per minute (fig 2.3). It would have been ideal to be able to compare maximal movements before and after massage. However, to avoid causing participants stress, possibly affecting supraspinal control (Leonard *et al.*, 1990), they were not specifically encouraged to make maximal movements (see discussion, ch4). Changes in ankle angle during active movements were recorded with an electronic goniometer. The change in angle during three of the voluntary movements was measured and the average change calculated.

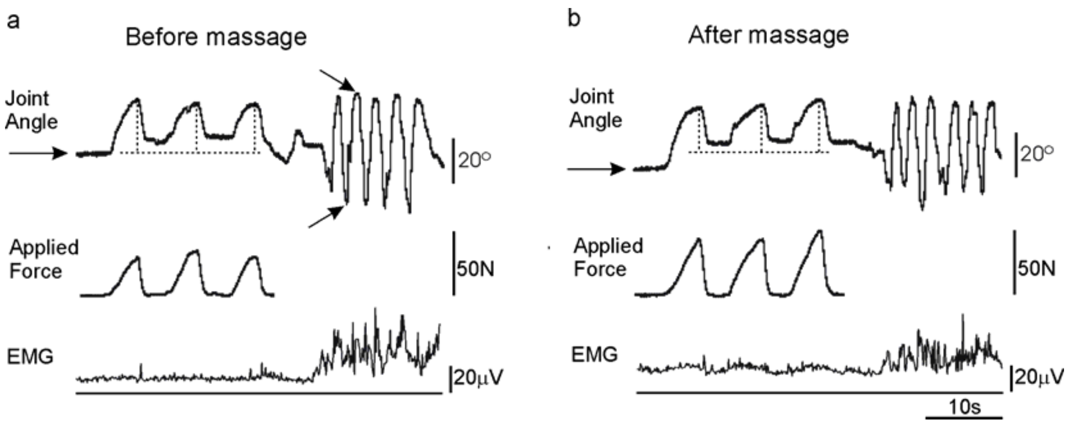


Fig 2.3 Joint angles during three passive stretches and EMG activity during active ankle movements.

This figure shows data recorded from participant 3, a) before and b) after massage. The angle of the ankle joint, the applied force and the surface EMG recorded from soleus is shown in each case. Horizontal arrows indicate rest angles. (b) shows starting resting angle has changed and greater joint angle excursion from that new position after massage. Sloping arrows in (a) indicate change in joint angle during a voluntary contraction. EMG activity during active movements is clearly reduced after massage.

2.3.7 Electromyography

Electromyographic (EMG) signals were collected with paired surface silver electrodes 6mm in diameter and 12mm apart following standard skin preparation. They were placed over the soleus muscle 2.5cm distal to the lower limit of the meeting of the gastrocnemius muscle bellies, in a line central with the Achilles tendon (fig 2.5). The pre-massage testing was carried out, then the electrodes were removed prior to massage, and replaced in the same marked position for tests following massage.

EMG signals were amplified (x1000), analogue to digital converted (sampling rate of 2kHz) using a 1401 Micro (Cambridge Electronic Design, Cambridge, UK) and stored in a personal computer, together with signals from the goniometer and pressure pad. They were band-pass filtered (50–300Hz) offline, rectified, and filtered (time constant 1ms or 100ms, to highlight detail or to show trends) with Spike2 software (version 5; Cambridge Electronic Design, Cambridge, UK). Data in Spike2 were transferred to Excel for further analysis.

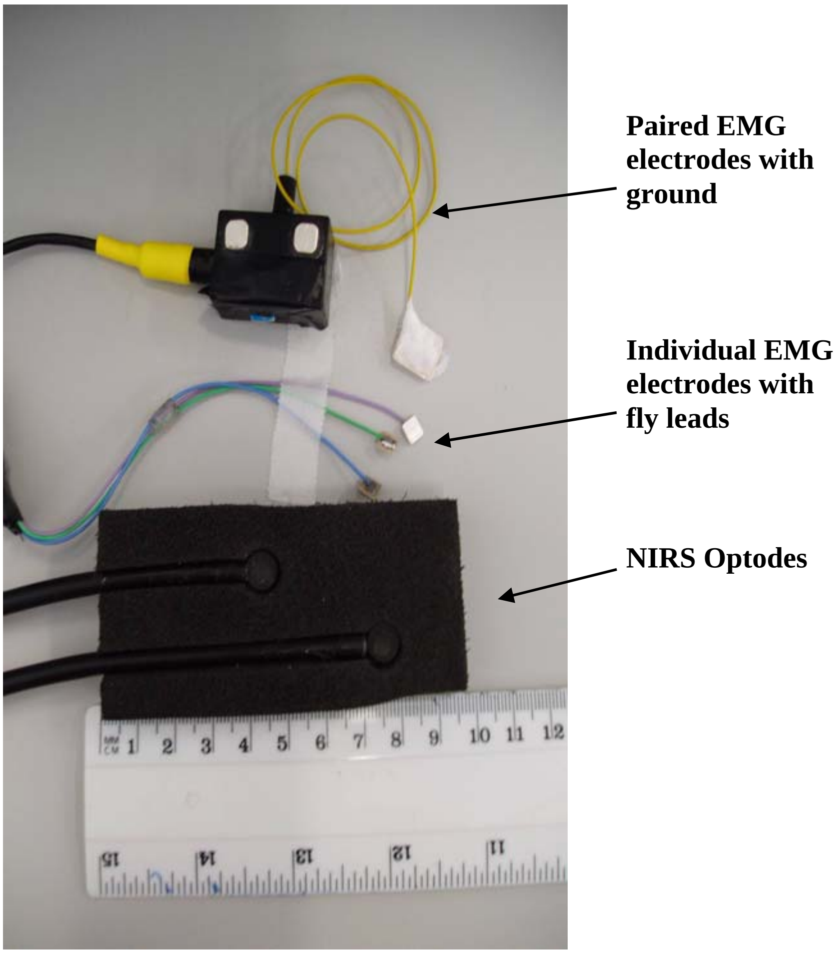


Figure 2.4 Equipment – 2 types of EMG electrodes and NIRS optodes

Photograph of items of equipment used in experiments. Paired EMG electrodes were used in preference to the individual ones as this offered a better chance of exact replication of position after massage.
The NIRS electrodes were placed in a foam holder which kept the operating distance between them exact, for replication after massage and over a number of sessions. The position EMG and NIRS electrodes is noted in the Methods.

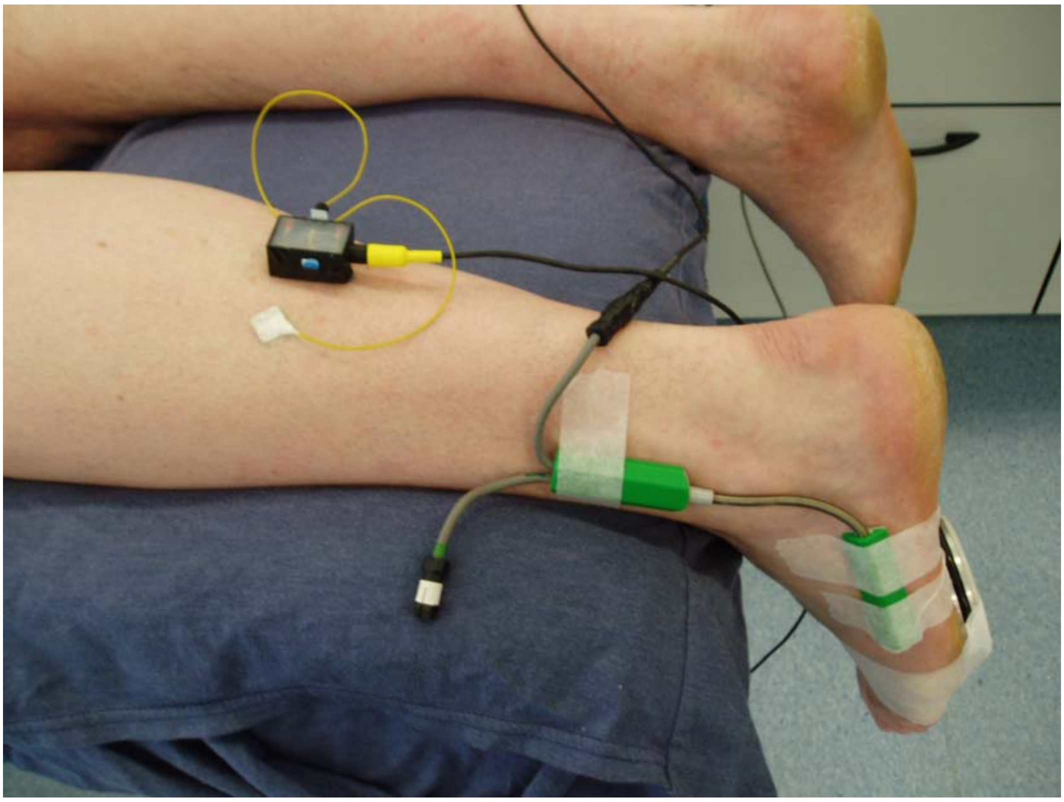


Fig. 2.5 Position of EMG electrodes over soleus muscle and goniometer position on the ankle joint.

Example from one healthy adult volunteer.

2.3.8 Goniometry

A twin axis SG110 electronic goniometer (Biometrics Ltd, Gwent, UK) assessed the changes in ankle resting angle and range of active and passive movement. This was attached with surgical tape on the outer aspect of the ankle joint. One axis was aligned along the long axis of the fibula and the other parallel to the long axis of the fifth metatarsal bone (Kilgour *et al.*, 2003) (fig 2.5). The goniometer was taped to the ankle of the first leg receiving massage and remained in position throughout the active and passive ankle movements, during massage of that leg and during the active and passive ankle movements following massage. It was then removed and the same procedure was repeated for the other leg. An indelible ink marker was used for replicating the position of the goniometer from one session to another.

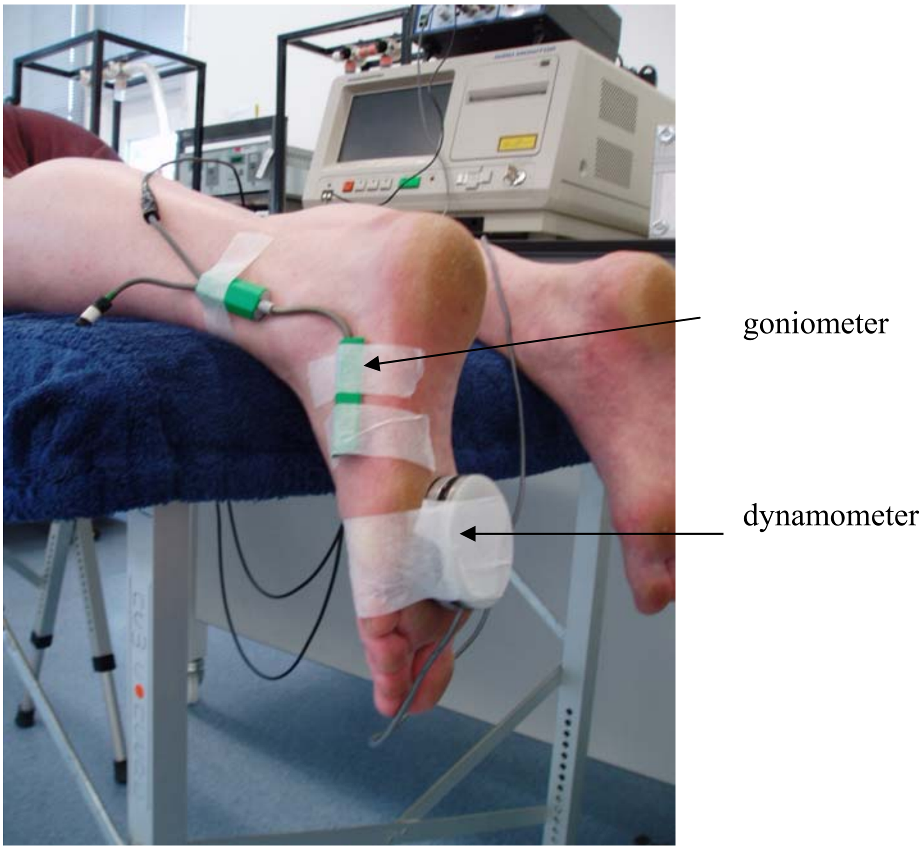


Fig 2.6 Positioning of goniometer and dynamometer

2.3.9 Dynamometer

A dynamometer was used to measure the pressure applied in passive movements of the ankle joint, using the same force each time. This was a pressure pad, 7cm in diameter designed and manufactured by the Bioelectronics Department of the University of Glasgow. Surgical tape held it in place on the sole of the foot centred along the metatarso-phalangeal joints (Fig 2.6).

2.3.10 Amplifier

An ISS Multimode Interface Unit designed and manufactured by the Bioelectronics Department of Glasgow University was used to amplify the raw signals of the EMG, goniometer and dynamometer en route to the Micro 1401 Unit.

2.3.11 Micro 1401

A Micro 1401 (Cambridge Electronic Design Ltd, Cambridge, UK) digitized the raw data and marker information, allowing traces to be displayed on a laptop.

2.3.12 Statistical analysis

The change in joint angle data (after-before, see fig 2.2) were analysed with ANOVA using a general linear model. Variables included in the model were subject, day, leg, peak force and rate of force applied. The change in EMG activity (after-before) data was also analysed using a general linear model including subject, leg and day as possible predictors. Minitab (version 13) was used for this analysis with a significance level of 5%.

2.4 Study 2. Effects of calf massage on motor function

Only motor function was assessed in this study, using the GMFM-66 because a suitable number of investigators were not available during the time that this group of participants was available.

2.4.1 Participants

6 adolescents with spastic diplegia (three males and three females, mean age 13.9 years, age range 11-18 years) had both calf muscles massaged twice per week where possible, using the standardised massage sequence listed in Table1.1.

2.4.2 Attendance

The number of sessions of massage for each individual ranged from 6 to 68 (Table 2.1) and depended on their availability and degree of disability. The more severely affected participants were offered more massage sessions. The protocol for the massage sequence and the testing conditions using GMFM-66 were identical to those used in study 1.

Participant number	GMFCS	Total number of massage sessions	Total number of weeks	Consecutive (C) Non consecutive (N/C)
1	I-II	20	47	N/C
2	II	14	7	C
3	II	27	31	N/C
4	III	6	7	C
5	III-IV	12	7	C
6	III-IV	68	126	N/C

Table 2.1 Participant attendance - study 2

This table shows details of each participants Gross Motor Function Classification System score, and the total number and the frequency of the massage sessions. ‘Consecutive’ and ‘non consecutive’ indicate whether the massage sessions happened in consecutive weeks, or whether there were breaks in attendance, as often occurred over a longer period of time due to school holidays.

2.5 Study 3. Full leg massage

The only non-ambulant participant from study 1 had shown no appreciable improvement in GMFM-66 score after 10 calf only massage sessions. It was thought that full leg massage might be more beneficial for non-ambulant participants. The sequence for this was devised using the same principles as for the calf muscle massage sequence (Table 2.2).

POSTERIOR	(Done first)		ANTERIOR		
Stroke	Area	Minutes	Stroke	Area	Minutes
Effleurage	Full leg	1.0	Effleurage	Full leg	1.0
Pick up/hold	Calf only	1.5	Rolling	Full leg	2.0
Rolling	Full leg	1.5	Kneading	Full leg	1.5
Kneading	Full leg	2.0	Appos.rolling	Quads only	1.0
Appos. Rolling	Full leg	1.0	Wringing	Quads only	1.0
Wringing	Full leg	1.0	Effleurage	Full leg	1.5
Effleurage	Full leg	1.0		Total time	8.0
	Total time	9.0			

Table 2.2 Massage sequence – full leg

This table shows the duration of each massage stroke used on the anterior and posterior aspects of the whole leg. The table also shows the sequence in which each stroke was delivered. Note: It is not possible to apply the pickup/hold stroke effectively to the anterior aspect of the leg.

The rate at which each stroke was administered is the same as that in table 1.1. Appos. Rolling = appositional rolling.

2.5.1 Participants

Two non-ambulant male participants with spastic diplegic CP (GMFCS classifications III and IV) ages 14y 8m and 15y 3m took part. Consent and ethical approval were obtained as previously described.

2.5.2 Attendance

The first participant, who had received 10 calf massage sessions 1 year prior, received 18 full leg massage sessions over 10 weeks, and then reverted back to calf only massage sessions for personal hygiene reasons. His results for the remaining calf muscle massage are included in study 2. The second participant received 60 full leg massage sessions over 114 weeks. The results for full leg massage for both participants are recorded in chapter 4.

2.5.3 Assessment

Both participants in this study were assessed with GMFM-6 only.

2.6 Study 4. Self-administered full leg massage

The author devised a specific sequence of massage strokes was which could be self-administered to the full leg by diplegic adolescents who had reasonable manual dexterity. If it could be shown to be effective, the advantages of self-massage are: firstly, the pupils would feel empowered by being directly involved and responsible for at least part of their own treatment. Second, convenience - the technique is carried out fully clothed and no oil is needed. Third, physiotherapists could supervise a group of adolescents using the routine, thus saving physiotherapists' time.

2.6.1 Participants

3 participants with spastic diplegic CP (1 male and 2 females, ages 15y 10m, 13y10m and 14y 1m respectively) were recruited from Ashcraig School, Glasgow. Consent and ethical approval were received as previously documented.

These three participants (who had previously had 10 sessions of calf muscle massage one year prior) were trained to administer their own full leg massage (table 2.3). They did this on a mat on the floor with their backs supported against a wall and the legs fully extended along the mat, i.e. in a long sitting position. This was done fully clothed, with shoes and splints, where necessary, removed.

1) Internal and external hip rotation, legs extended. (Repeat x 5. L&R)

2) Draw heel in 15cm flexing knee, then release to extend leg on floor. (x 10).

3) Alternating plantar- and dorsiflexion. (x 6)

4) Massage* hip adductor muscles. (x 4). One leg at a time

5) Massage* calf muscles. (x 4) One leg at a time

6) Seated forward stretch with legs fully extended along the floor

7) Massage* hamstring muscles. (x 4, L&R)

8) Massage** quadriceps muscles one at a time (x 4, L&R)

9) Repeat step 2

10) Repeat step 3

11) Cupping down fronts of both legs and up outsides of both legs

12) Repeat step 1

Table 2.3 List of the sequence for self massage technique

This is a list and description of the protocol followed by those administering their own massage and stretches. Brackets contain number of repetitions, L, left leg: R, right leg.

* The massage consists of two strokes, which are better demonstrated, although an attempt is made here to describe them in words. First stroke – muscles are gripped between thumb and the straightened fingers (not pincer grip) of the hand opposite the leg. Repeated gripping for approximately 5 seconds and then releasing of adductor muscles and calf muscles. Second stroke – using the same grip, hold and mobilise the muscle drawing the hand up and down. This is done in sections until eventually the full length of those muscles is massaged.

** The quadriceps are massaged with the same grip but with hands one above the other at the same time, gripping from the outside of the leg with the thumbs forward. An inward rotation of both wrists is then done to compress for 5 seconds then release the muscle. This is done along the full length of the muscles.

2.6.2 Attendance and Assessment

Participants attended for 34 sessions spread over 74 weeks. They were assessed for gross motor function (results in chapter 4) and for change in skin temperature over the massaged muscles, (results in chapter 5).

2.7 Study 5. Calf muscle massage – effects on skin temperature.

2.7.1 Participants

Group 1. 3 adolescents who have spastic diplegia (2 males and 1 female, ages 16.1y, 16.6y and 13.1y respectively). The left calf was always massaged first. The temperature of the skin over the calf muscles immediately before and after massage was recorded for this group. Ethical approval and consent were obtained as recorded earlier.

Group 2. Four healthy males (ages 23, 45, 46 and 54) were recruited from technical staff and students at Glasgow University and consented to participate, following instruction on the purpose and methods to be used. Approval was obtained from Glasgow University Ethics Committee. It was of interest to ascertain if the massage technique employed in the previous studies could be learned by another therapist. Following instruction a trainee masseur massaged the left calf first, and then the experienced masseur massaged the right calf, both using the massage sequence listed in table 1.1. This was carried out at the Laboratory, Room 121, West Medical Building, University of Glasgow. A comparison of the change in skin temperature over the massaged muscles (before massage - v - after massage) for right and left legs was used to determine the efficiency of the trainee in administering the massage.

2.7.2 Attendance

Group 1 – 3 adolescents with CP (GMFCS III, III, III-IV) received calf muscle massage twice per week for 3 consecutive weeks.

Group 2 – 4 healthy adults received calf muscle massage twice per week for 5 consecutive weeks.

2.7.3 Temperature recording

The skin temperature of the massaged limb was recorded before and after massage, using the probe of an Edale Digital Thermometer GC20 (Edale Instruments, Cambridge, UK) taped centrally on the medial belly of the gastrocnemius muscle, 10cm distal to the popliteal crease. The probe was removed during massage and replaced in the same marked position immediately following massage. A settling time of 30 seconds elapsed before the temperature was recorded each time. Results are recorded in chapter 5.

2.8 Study 6. Calf muscle massage – effects on skin temperature and oxygenation

As more participants became available, 2 groups allowed for a comparison to be made of changes in skin temperature and oxygenation following massage.

2.8.1 Participants

Groups 1 & 2. Six adolescent pupils (group1) at Ashcraig School who have spastic diplegia were age and gender matched with six healthy controls from Hutcheson's Grammar School (group 2) (14-18 years, mean 16years, three males and four females). Both groups received calf muscle massage (table 1.1).

2.8.2 Attendance

The sequenced massage previously described was administered in four sessions over five weeks at Ashcraig School, and in five sessions over four weeks at Hutcheson's School. School holidays prevented Ashcraig School pupils from attending the same number of sessions at the same time as those at Hutcheson's School. Written informed assent from all of the adolescents and consent from their parents, along with ethics approval were received as recorded earlier. Changes in oxygenation and in skin temperature were recorded for both of these groups as detailed below.

2.8.3 Procedure

The participants attended the study at their own schools during a selected period of the school timetable. A room affording privacy was set aside to accommodate the treatment table, with the equipment suitably sited for recording.

Each participant was welcomed and the protocol was explained to them before they removed their shoes and socks, and rolled their trouser legs to above the knee. Some pupils from Ashcraig School needed assistance with this. The participant lay prone on the treatment table with the lower leg extended far enough beyond the end of the treatment table to allow plantarflexion and dorsiflexion. The equipment was explained to the pupils before being attached, and they were encouraged to ask questions.

2.8.4 Temperature recording

Calf muscle temperature was recorded as noted in point 2.7.3 above.

2.8.5 Near-infrared spectroscopy

A near infrared spectroscope (NIRO 500 SRS, Hamamatsu Photonics, Japan) measured changes in oxygenated haemoglobin (HbO_2), deoxygenated haemoglobin (Hb), total haemoglobin (HbT) and cytochrome oxidase (CtOx) of the triceps surae muscles. The transmitting and receiving optodes were encased at a spacing of 3cm in an optically dense holder unit, which ensured unaltered orientation to one another. The optode unit was taped securely to the medial belly of the gastrocnemius muscle at a distance of 5cm from the popliteal crease. It was wrapped in an elastic bandage, and then covered by a black nylon sleeve to minimize the intrusion of ambient light and loss of NIRS light. The optode holder and cover stayed in place throughout the testing and massage. Once removed, a check was made to determine if the holder had moved during the process by assessing the clear indentations left in the skin by the two circular parts of the holder.

The effects of massage on oxygenation of the muscles were compared with the oxygenation changes produced by contraction of the same muscles during resisted plantar flexion. It was also possible to monitor the effects of each stroke of the massage concurrently, as well as effects before and after the massage.

2.8.6 Determining the path length for each participant

The first visit to each school was used to determine the path length for each participant. The NIRS optodes were attached as described above and shown in fig 2.7, and a series of three contractions, (plantar flexion for ten seconds, and then relaxing for ten seconds), were executed at four path lengths, 8cm, 10cm, 12cm and 14cm. The optimal path length for each participant was determined after visual inspection of the collected recordings, and that path length was then used for the rest of the study (fig 2.8). In the first session the sequenced massage was given on the right leg, with a five minute settling in and out period.

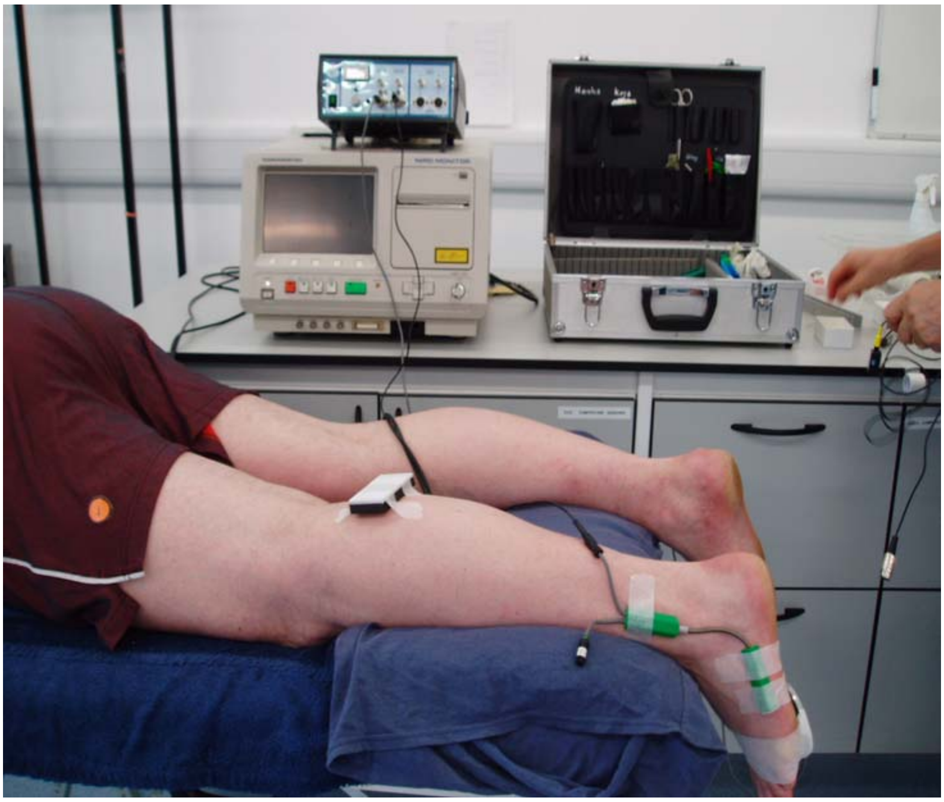


Fig 2.7 Positioning of NIRS equipment.

The black and white box is the optode holder, in position before the securing bandage and blackout material were wrapped around and taped over it. This then covered the top half of the calf. The goniometer is also visible on the ankle joint as well as the dynamometer on the ball of the foot.

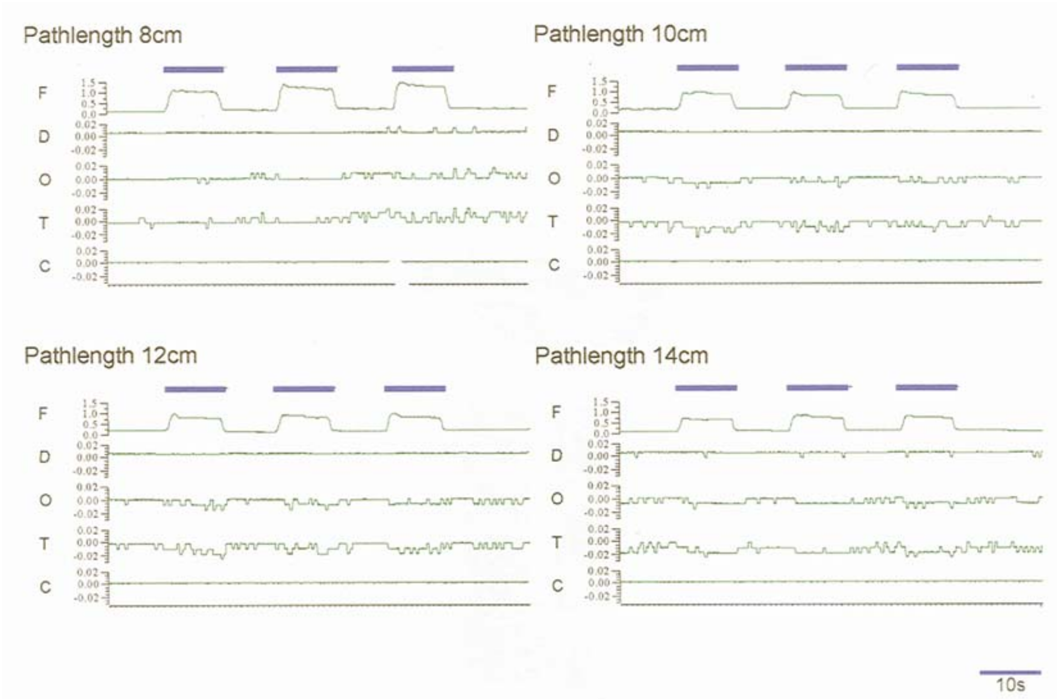


Fig 2.8 Path length data - control group.

These panels show four sets of NIRS data recorded over the gatrocnemius muscle of one participant in the control group. In each case a different path length, ranging from 8-14cm was used.
In each record a series of three plantar flexions (10s contractions with 10s rest in between), are indicated by the upward deflection of the force record (F) and the horizontal bars. Such records at each path length allowed for visual inspection of the data to determine which path length was optimal.
A clear depression of the total haemoglobin and oxyhaemoglobin during the contraction with a return afterwards for all three contractions indicated an optimal path length. In this example 14cm was the optimal path length. The raw data was used here rather than smoothed records in case the smoothing made the choice less obvious.
F: Force, D: deoxyhaemoglobin, O: oxyhaemoglobin T: Total haemoglobin, C: cytochrome c oxidase,

During subsequent sessions at Hutcheson's Grammar School, both legs were massaged (right one first) but all NIRS measurements were taken from the right leg only (table 2.4). At Ashcraig School, as the periods were shorter and the pupils needed more time for preparation, only the right leg was massaged (table 2.5).

Event sequence	
1	Participant in position prone on couch. Right leg - instruments attached and switched on, settle in period – 3 mins
2	30 secs run in data capture to establish baseline values
3	Temperature recorded from gastrocnemius
4	3 contractions (plantarflexion), each 10 secs contraction, then 10 secs rest. Total 1 min.
5	14 minute massage sequence
6	3 mins settling out
7	3 contractions as event 4 above
8	Record temperature
9	Remove instruments
	Right leg first, then sequence repeated for left leg

Table 2.4 Protocol used at Hutcheson's School – control group

Note: The same protocol was used at Ashcraig School, but as it took longer to accommodate the disabled participants within their school timetable, only the right leg was massaged.

2.8.7 Data Analysis

The sampling rate was chosen to take account of the time course of the voltage outputs on the four channels of the NIRS. The voltages change in steps of 0.008v and are updated every 0.5s, although the four channels do not update synchronously. Also, changes in voltage involve ramps each lasting 0.1s. The four voltage outputs (HbT, HbO_2, Hb and CtOx) were sampled at 5 Hz for analysis in Microsoft Excel because sampling at 5Hz provides two samples for each possible voltage step change. The mean and the SEM were estimated for data corresponding to an initial baseline period of 100s before massage began followed by each of the eight massage strokes.

Participants were asked to make 3 voluntary contractions (plantar flexion) with the relevant limb before and after massage. Changes were assessed from data collected for 8s during each contraction for comparison with voltages during a control period of 8s, 12 s before the first contraction.

The operating optical path length is the product of the interoptode spacing (IOS) and the differential pathlength factor (DPF), the depth that the laser light penetrates the tissue (Duncan *et al.*, 1995; Boushel & Piantadosi, 2000; Kowalchuk *et al.*, 2002). Previous studies have determined the DPF for the forearm, calf, and cranium using an interoptode spacing of 4cm (Duncan *et al.*, 1995; Elwell, 1995)

In this study an interoptode spacing of 3cm was used to allow secure attachment to the relatively smaller calf muscles of the adolescents with CP.

The DPFs given in Table 1.1 of Elwell's User Guide to Near Infrared Spectroscopy were generated with an IOS of 4cm (Elwell 1995). Since the IOS in our study was 3cm, we chose an appropriate DPF from Elwell's table and tested path lengths longer and shorter to determine the optimal path length for each participant. This was necessary for two reasons. Firstly, the group of participants included females, and females have a greater fat to fat-free mass ratio than males. It was anticipated that longer path lengths would be optimal in females. Secondly, A. P Shortland (personal communication) had told us that individuals with spasticity of calf muscles have a thicker overlying layer of fat.

Test contractions of calf muscles were used to detect that the infrared light reached muscle. Adult control subjects were asked to plantar flex their ankles three times. The dynamometer fastened to the ball of the foot measured the force exerted when the participant plantar flexed against a non-compliant surface with the knee fully extended and resting on the treatment table. A warning bell rang when the force reached about 80N.

Figure 2.9 shows typical traces recorded from the NIRS equipment related to the oxyhaemoglobin (HbO_2), deoxy-haemoglobin (Hb) and cytochrome oxidase (CtOx). During contractions HbO_2 fell, followed by an increase in Hb. The time course of both these changes is reflected in the signal for total haemoglobin (HbT). The signal for CtOx could increase or remain unchanged (see Discussion for interpretation). The results for male and female responses (figs 2.10 and 2.11) were used to determine the path length range to be tested for each adolescent participant in their first session.

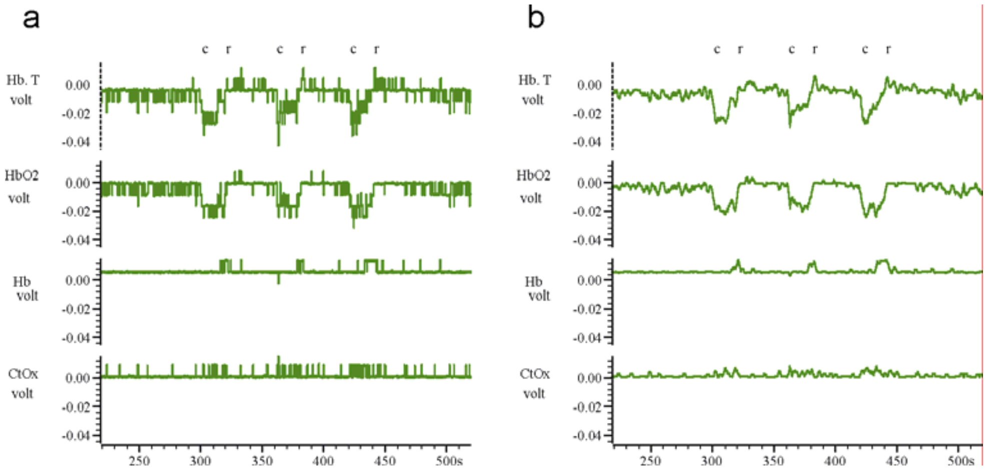


Fig 2.9 Voltage outputs from the NIRS equipment during three contractions of calf muscles

These two panels show 1 set of NIRS data recorded over the gatrocnemius muscle of one participant in the control group. b) is a smoothed record of the raw data shown at a), time constant 1s, using Spike 2 smooth function.
This records a series of three plantar flexions (10s contractions with 10s rest in between). Such a record allowed for visual inspection of the data to determine if the NIRS equipment would register clearly when a contraction was done. Note that contraction had the greatest effect in reducing HbT and HbO_2.
The smoothed record shows clear depression of the total haemoglobin and oxyhaemoglobin at the start of the contraction (point c above record) with a return towards zero on relaxation afterwards (point r), for all three contractions. This indicates suitable recording perameters.
Hb: deoxyhaemoglobin, HbO_2: oxyhaemoglobin, HbT: total haemoglobin, CtOx: cytochrome c oxidase. All voltages are displayed with the same scale (range 0.06V).

Figures 2.10 and 2.11 show the voltage outputs of the NIRS equipment for a male control and a female control participant, using a range of path lengths (4cm to 14 cm). From each of these a path length was chosen that would likely give the clearest indication of changes in oxygenation/blood flow in the muscle for that participant. Recordings from the shorter path lengths (4 and 6cm) were more variable because they were affected by changes in both skin and muscle blood flows. On the other hand, the signals relating to muscle contraction were reduced at the longest path lengths (14cm) because of bone conduction.

Figure 2.12 compares the chosen optimal path lengths from figs 2.9 and 2.10 for the male and female. It shows that the 8cm path length chosen for the male would not be so useful for the female because the deflection of HbT and HbO_2 was not maintained during contractions. Likewise, the 12 cm path length chosen for the female would not be as suitable for the male because HbT and HbO_2 are clearly changed only during the first contraction.

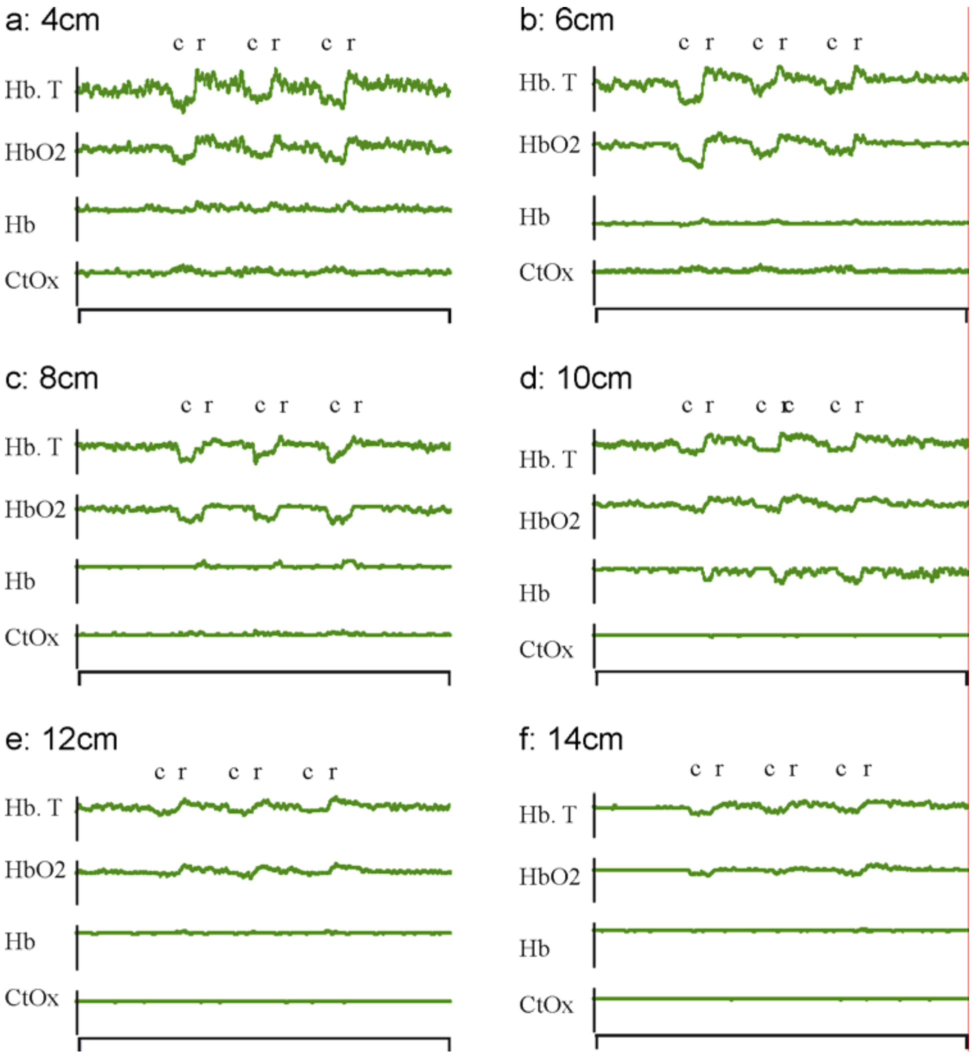


Figure 2.10 Voltage outputs of the NIRS equipment for a male control participant using a range of path lengths as indicated

These two panels show six sets of NIRS data recorded over the gatrocnemius muscle of one male participant in the control group, smoothed at time constant 1s, using Spike 2 smooth function. These were all done in the same session, in the same conditions.

Each panel records a series of three plantar flexions (10s contractions with 10s rest in between). The smoothed record shows clear depression of the total haemoglobin and oxyhaemoglobin at the start of the contraction (point c above record) with a return towards zero on relaxation afterwards (point r), for all three contractions. It was most obvious that the contraction had the greatest effect in reducing HbT and HbO_2 at the 8cm path length, which was chosen for all the subsequent recordings for this participant. All voltages are displayed with the same scale (range 0.09V).

Abbreviations as for fig 2.9.

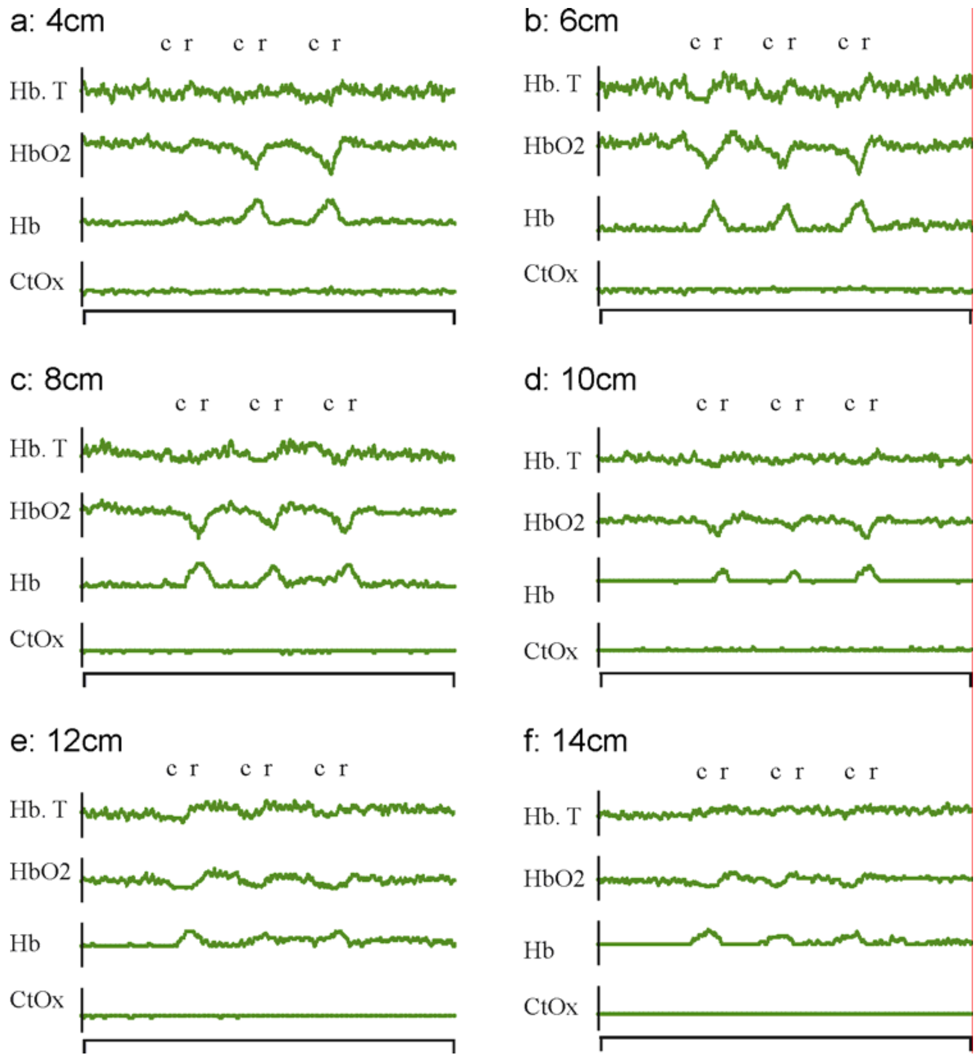


Fig 2.11 Voltage outputs of the NIRS equipment for a female control participant using a range of path lengths as indicated

These two panels show six sets of NIRS data recorded over the gatrocnemius muscle of one female participant in the control group, smoothed at time constant 1s, using Spike 2 smooth function. These were all done in the same session, in the same conditions.

Each panel records a series of three plantar flexions (10s contractions with 10s rest in between). The smoothed record shows clear depression of the total haemoglobin and oxyhaemoglobin at the start of the contraction (point c above record) with a return towards zero on relaxation afterwards (point r), for all three contractions. It was most obvious that the contraction had the greatest effect in reducing HbT and HbO_2 at the 12cm path length, which was chosen for all the subsequent recordings for this participant. All voltages are displayed with the same scale (range 0.09V).

Abbreviations as for fig 2.9.

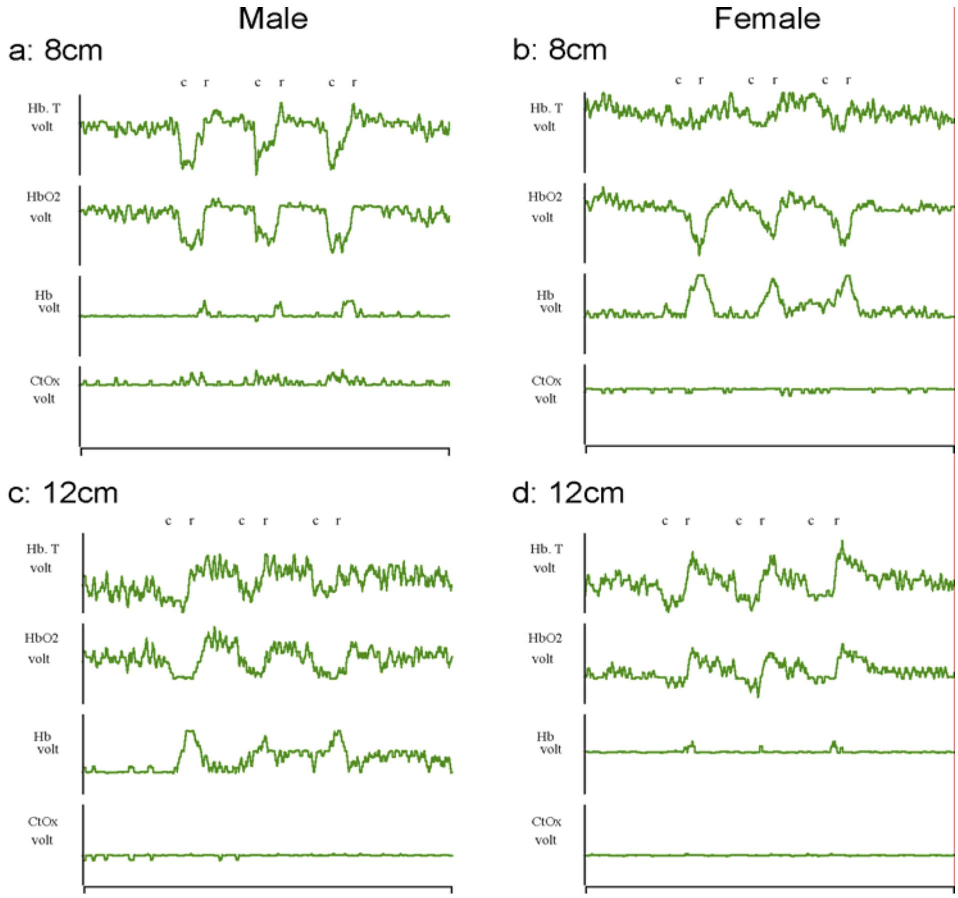


Fig 2.12 Comparison of traces at chosen path lengths for male and female from figs 2.10 and 2.11.

These four panels from figs 2.10 and 2.11 indicate differences in the path length suitability, depending on whether the participant was male of female. Top - male (a) and female (b) at 8cm path length. Bottom (c) male and (d) female at 12cm path length. Voltage ranges (a) 0.04, (b) 0.045, (c) 0.03, (d) 0.025.
In these cases, the path lengths (a) and (d) were chosen as optimal because the HbT and HbO_2 displayed the clearest drop in voltage at the start of each contraction, which was best maintained during all three contractions.
Abbreviations as for fig 2.9.

CHAPTER 3

Effects of Massage on Mechanical Properties of Spastic Muscle and Stretch Reflexes

Lieber and colleagues contend that comprehensive identification of the structures responsible for the altered stiffness in spastic muscle is not yet possible (Lieber *et al.*, 2004). Despite this, it is known that both the mechanical properties of spastic muscle and its response to stretch differ from those of non-affected muscle (Dietz & Berger, 1983).

The incidence and timing of the onset and termination of stretch reflexes is important because stretch reflexes regulate muscle stiffness (Nichols & Houk, 1976; Houk, 1979). From clinical observations, it has been concluded that the exaggerated response of stretch reflexes is responsible for muscle hypertonia, and consequently treatment is usually directed towards reducing stretch reflex activity (Dietz, 1999).

However, O'Dwyer and colleagues demonstrated that it was possible to train young people with CP to reduce their stretch reflex gain by as much as 50% by giving visual feedback. Nevertheless, there was no change in the mechanical behaviour of the muscles when they were stretched (O'Dwyer *et al.*, 1994).

Another factor that may be implicated in the changes observed in spastic CP is thixotropy. That term was first used in 1927 to describe the change from a gel to a sol in the stirred cytoplasm of sea urchin eggs. Instead of being stiff, the consistency became fluid (Walsh, 1992). Thixotropy can be described as history-dependent behaviour, and has been shown to occur in isolated muscle (Lakie & Robson, 1988). It has also been observed in normal muscle as a whole (Walsh, 1992), and Walsh found that its effects are exaggerated in spastic muscle of hemiplegic patients (Walsh *et al.*, 1994). It was felt that the very act of moving the ankle regularly during testing would be beneficial to the muscle and would be apparent in its thixotropic response.

A number of authors have concluded that the spasticity seen CP is due more to changed muscle properties than to exaggerated reflex response to stretch (see chapter 2).

Consequently the aims of the work recorded in this chapter were to investigate the effects of massage on the response of the spastic muscle to stretch. Slow stretches lasting 5 seconds were used because it was anticipated that the massage might increase the range of movement at the ankle joint. However some reflex contractions were still elicited. There were occasions when the ankle movement came to an abrupt stop before the five seconds of force time had elapsed. When this happened, almost always during the stretches before massage, the pressure was removed and the ankle rested until the time for the next of the three stretches. The frequency and nature of the reflex contractions invoked during passive stretching of the calf muscles were noted, along with the resting muscle lengths. In this study, the contribution of three factors, muscle mechanical properties, thixotropy and reflex contractions, was borne in mind.

Details of the methods are recorded in chapter 2.

3.2 Results

3.2.1 Effects of massage on the range of passive movement

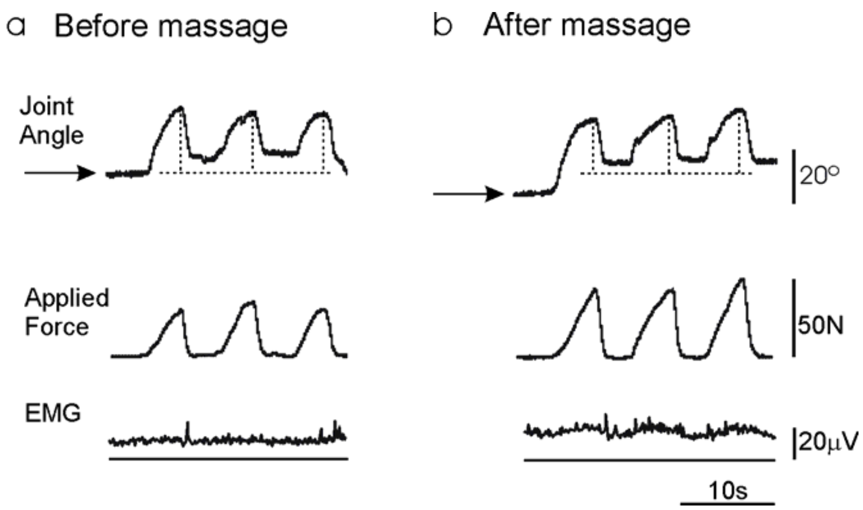


Fig 3.1 Example of data recorded during three passive stretches into ankle dorsiflexion.

Example of joint angle, applied force and accompanying EMG recordings during the 3 passive stretches done before and after each massage session for each participant. The example relates to participant 3 during the second massage session, and is typical of the recordings from her and the other four participants.
The horizontal arrows in (a) and (b) (top) show that the resting joint angle had changed after massage. Changes in joint angle when the ankles were dorsiflexed were all measured with reference to the initial resting angle (indicated by the horizontal dashed lines). Note that the changes in joint angle were approximately the same in (a) and (b). However, the applied force was greater in (b) than in (a). The inference drawn from this is that the muscle was actually stiffer after massage.

Analysis of tests of passive dorsiflexion of the ankle before and after massage (Fig 3.2) showed that only four of the 10 ankles dorsiflexed consistently further after massage. It is important to note that all changes in ankle angle were measured in relation to the initial rest angle. Surprisingly however, the force applied to all ankles after massage was greater (also compare force applied in Figs 3.1. (a) and (b). This could imply that massage had made the calf muscles stiffer, as, on average, significantly greater force was required to move the ankles through the same angles in both limbs of all 5 participants ($p < 0.001$; paired t-test). Statistical analysis also showed that there was no significant effect of peak force or the rate that the force was applied. This confirms that the force profile template was being followed faithfully in producing ankle dorsiflexion for the three stretches both before and after massage.

Any increased stiffness of the ankles after massage could have been caused by increased reflex responsiveness. Therefore the areas of the rectified EMG activity in a window lasting 2 seconds immediately before the end of the stretch were compared before and after massage. The only significant factor in the general linear model for this EMG activity was the leg ($p=0.014$), with the left leg being associated with significantly greater EMG activity (after-before) than the right; there was no significant difference in EMG activity when comparing the periods before massage and after massage.

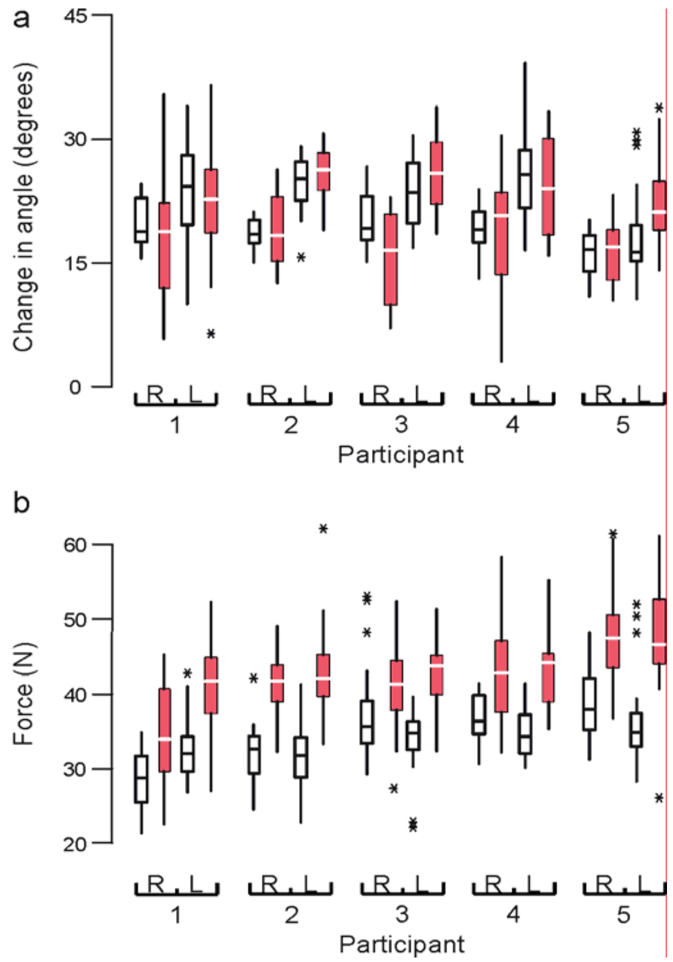


Fig 3.2 Passive dorsiflexion before and after massage – 5 participants.

Boxplots show the range and means for the amount of force (bottom) and the change in ankle angle (top) during 3 passive stretches of right and left ankles, applied before and after massage for all 5 subjects, for all 10 massage sessions.
Top and bottom horizontal lines forming each box indicate the range of change, and the horizontal line between these shows the mean for the total number of stretches for each leg for each participant. R: right leg, L: left leg. *indicates outliers.
a)**.** Average change in ankle angle during three stretches for each participant, before massage (clear box) and after massage (filled box). Note that in only four of the legs was there an increase in angle, i.e., the range of movement was not consistently increased for all 10 limbs.
b). Average of force applied to produce the stretches, before massage (clear boxes), after massage (filled boxes). After massage, in all of the stretches significantly greater force was applied in order to follow the template on the computer screen, in an attempt to replicate the same stretch each time ($p < 0.001$, paired t-test).

3.2.2 Changes in muscle length after massage

In young people with spastic CP, equinus is usually evident, indicating that an element of the calf muscle/tendon unit must be shortened. This typically presents as permanent plantarflexion of around 20º (Shortland, *et. al.*, 2002). It was therefore of interest to assess if the massage had resulted in a change of ankle angle, i.e., an expected reduction in plantarflexion, indicating that the calf muscle had lengthened following massage.

However, the results showed that how far an ankle extended during a passive stretch after massage depended on the resting ankle angle, which could have changed appreciably from the initial rest angle (compare joint angle before and after massage in Fig. 3.1 (a) and (b)). Therefore it was important to record the effects on stretch of the calf for all of the participants for each massage session.

Fig 3.3 shows the effects on the mechanical behaviour of the calf muscles during the first passive stretch of the ankle of both legs for all 5 participants before and after each massage session. The changes were measured from the natural ankle resting angle for each participant, which is signified by 0° on the horizontal axis and is arbitrary. Normally, the resting ankle angle is 90°, and any movement into plantarflexion or dorsiflexion, is measured from that point. However, unless surgery has been done to lengthen the calf musculotendinous unit, in CP the ankle usually rests in some degree of plantarflexion (Shortland *et al.*, 2002).

Repeated measures on the same subject allowed for comparison of the response of the muscle both before and after massage in any one session, and also whether there was a trend in the muscles' response over a number of sessions. This graph displays trend lines and not correlation coefficients, thus repeated measures on the same subject can be justified. The trend lines indicate that over all the muscles were longer after massage – the trend line crosses the y axis at 20.6° before massage but 22° after massage.

If the change in resting ankle angle indicated that the calf muscle had shortened, the ankle displacement during stretch was greater than before, and less if the calf muscle had lengthened. This can be seen in the reversal of the slopes of the trend lines for peak displacement in upper and lower graphs in Fig.3.3.

Surprisingly, although individual calf muscles could either shorten or lengthen when massaged, there was no consistent pattern of shortening or lengthening. Another feature was that the ankles did not spring back exactly to the resting position immediately on release from a test stretch. There was always some residual displacement. This effect was previously described as 'muscular creep' in two children with hemiplegia (Walsh *et al.*, 1994). The slopes of the trend lines for peak and residual displacements before massage (Fig.3.3) suggest that calf muscles did not extend so far, and sprang back closer to the rest position if they shortened during massage and vice-versa for those that lengthened.

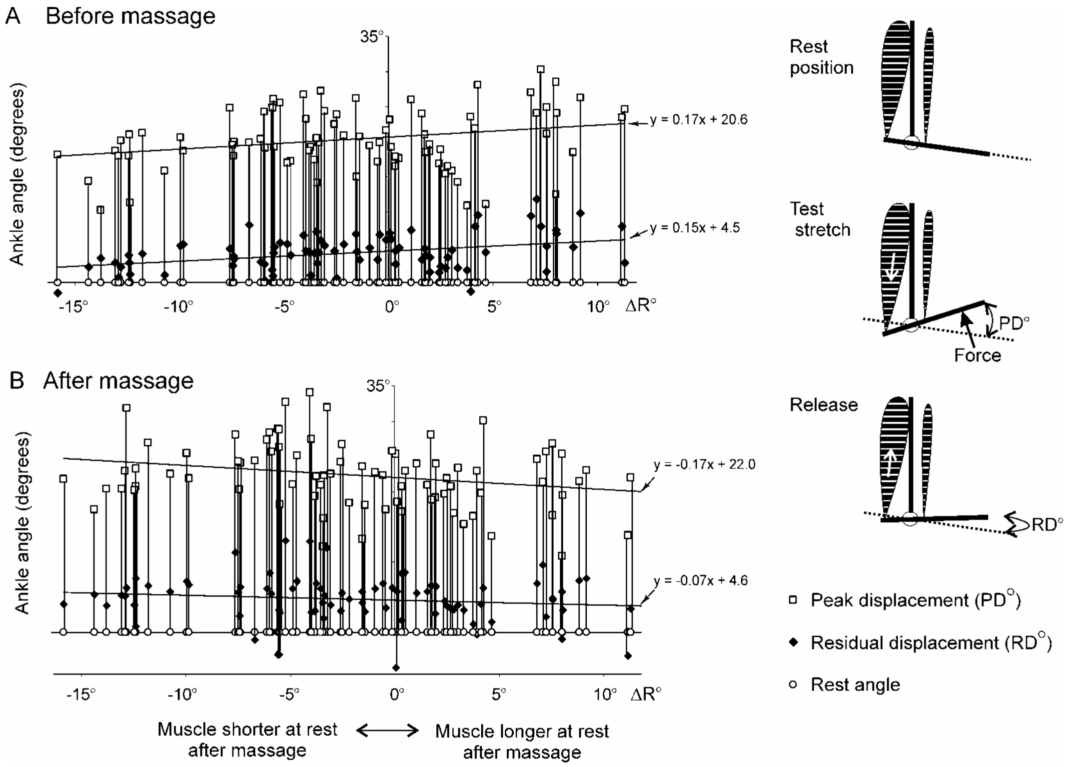


Fig 3.3 Change in passive mechanical behaviour of spastic muscle after massage.

The graph on the left of the figure records the mechanical behaviour of the calf muscles during the first stretch of both ankles, before and after each of the 10 massage sessions, for all 5 participants.
A) Ankle extension in the first test stretch before massage plotted against change in resting ankle angle after massage (ΔR°). Data are from all five participants (10 massage sessions over 5 weeks). 0° signifies zero degrees of movement into plantar flexion or dorsiflexion, i.e. no change in the rest position after massage.
The trend lines are best fit with r^2 values – top to bottom lines on graphs r^2 =0.07, r^2 =0.14, r^2 =0.06 and r^2 =0.02. These values are of a similar very low magnitude, although they slope in opposite directions before and after massage. The slope of the trend line for peak displacement suggests that ankles extended further if the muscle lengthened during massage (data to the right of 0^o). Conversely ankles did not extend as far if the muscle shortened during massage (data to the left of 0^o). The residual displacement tended to be greater if the muscle lengthened.
B) Ankle extension in the first stretch after massage plotted from new resting position. The trend line crosses the y axis at 22^o, increased from 20.6^o in A, suggesting that on the whole, muscles lengthened during massage. However muscles that lengthened the most did not extend as far as previously (right side of 0^o) and muscles that shortened the most extended further than previously (left side of 0^o).
Figures on the right side of the diagram relate to ankle dorsiflexion and illustrate a typical relationship between the ankle rest angle (top), the peak displacement during calf muscle stretch (middle), and any residual displacement after stretch (bottom). The symbols for each of these are used in the graph on the left.

3.2.3 Abnormal stretch reflexes

The incidence of abnormal stretch reflexes was assessed from soleus EMG activity during all test stretches, which were sometimes accompanied by irregularities in the joint angle record. Irregularities in the joint angle record unaccompanied by EMG activity were taken to indicate stretch reflexes in the gastrocnemius muscles (see fig 3.5). Together, these indications occurred during 40% of passive stretches during the first five massage sessions and only 22% of stretches during the last five sessions, i.e., the frequency of stretch reflexes was substantially reduced (see fig 3.4).

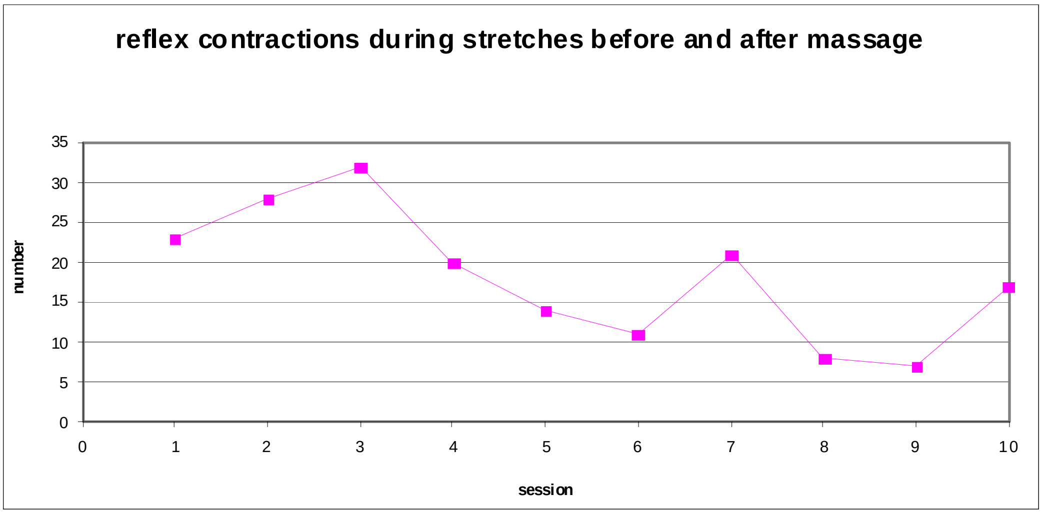


Fig 3.4 Numbers of abnormal stretch reflex contractions

This graph summarises the total numbers of abnormal stretch reflexes, recorded from all five participants during test stretches in each of the 10 massage sessions, e.g. there was a total of 23 instances of abnormal stretch reflex response in session 1. Taken together for all stretches for all participants, stretch reflexes had a 40% incidence in the first five sessions compared to only 22% in the last five sessions. This suggests a significant reduction in spasticity, which is classically known for hyper reflexive responses to stretch.

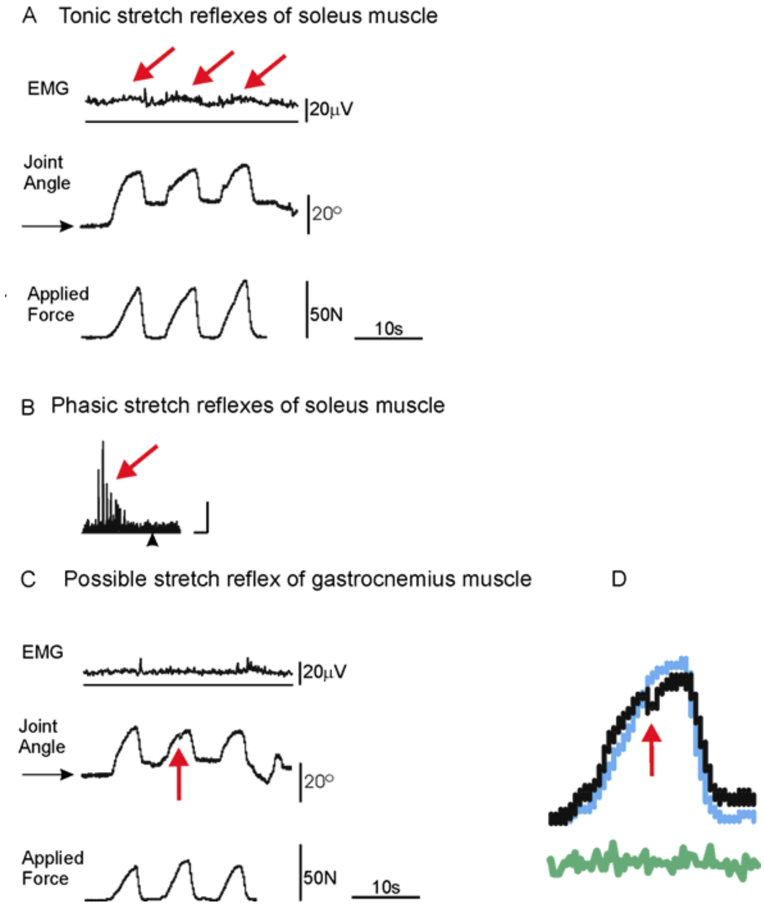


Fig 3.5 Indications of stretch reflexes

Recordings from participants with CP, showing 3 different instances taken to indicate abnormal stretch reflexes during passive stretch of the calf muscles. In each case the red arrows indicate the point of incidence of stretch reflexes.

A. Regular increases in EMG are recorded as each of the three stretches is administered during one session, indicating tonic stretches of soleus muscle (three red arrows).
B. This shows an instance of phasic stretch reflexes during one stretch of soleus muscle (single diagonal red arrow). Very large stretch reflexes are visible against the lower background EMG. The diminishing size of the spikes suggests that the reflexes were progressively inhibited as the muscle stretched. The length of the stretch is indicated from the start of the trace on the left to the black arrow head.
C. This illustrates the possibility of stretch reflex in gastrocnemius muscle. (at single up facing red arrow). Here there is no apparent increase in EMG of soleus but there is a dip in the joint angle.
D. Enlargement of C, showing that the ankle angle trace (black) has a momentary dip, but the force trace (blue) is uninterrupted.

The character of the reflexes encountered could change markedly. For example, in the first two sessions a stretch reflex in the soleus muscle of participant 2 began at a more dorsiflexed angle after massage (Fig. 3.6 a), suggesting a reduced responsiveness. However, in a subsequent massage session, decreasing phasic stretch reflexes were recorded soon after the beginning of stretch (Fig. 3.6b). Evidence of inhibition of the soleus motoneurones during stretch (see Discussion) was also seen in participant 1 in sessions 4 and 5 as a diminution of background EMG activity. Decreasing phasic stretch reflexes similar to those seen in fig. 3.6(b) were also seen in this participant.

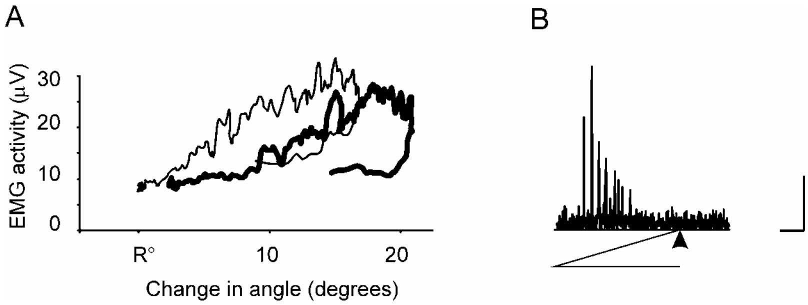


Figure 3.6: **Changing reflex responses to stretch of soleus muscle**.

(A) Averaged electromyographs (EMG) recorded from participant 2 during sessions 1 and 2 before (thin line) and after massage (thick line), plotted against the change in ankle angle during dorsiflexion. Each trace is an average of six EMG responses to stretch. Before massage, EMG activity increased almost immediately the ankle began to move. After massage, EMG activity started to increase later, and although the muscle extended further, maximum EMG activity was about the same as before massage. Calibration: vertical: 60mV; horizontal: 1s. R: rest angle.

(B) This shows an EMG plotted against time, recorded during the first stretch of soleus muscle from the same participant during session 4. Very large stretch reflexes (spikes in EMG) are visible against the lower background EMG activity. Here the EMG activity did not increase as in (A). Instead, phasic stretch reflexes were evident which decreased during the first stretch before massage. The diminishing size of the spikes suggests that the reflexes were progressively inhibited as the muscle stretched, which is a 'normal' response to stretch of the calf muscles. The period of this single stretch is indicated by a ramp below the EMG trace, with an arrow at peak of stretch.

3.3 Discussion and Conclusions

3.3.1 Changes in mechanical properties

The passive mechanical properties of muscle are functionally very important as the earliest source of resistance to stretching, because stretch reflexes take longer, owing to delays for nerve conduction. Three pieces of evidence from our work suggest that the local stretches during massage may have adjusted sarcomere lengths within the muscles. Firstly, changes were observed in muscle length after massage. Second, an individual's muscles could lengthen or shorten after massage. Third, the extent of passive muscle shortening immediately after the initial stretch was related to whether there was overall lengthening or shortening after massage. Thus it is possible that the trend line slopes reverse between fig 3.3 a) and b) because massage reset the sarcomere lengths in the calf muscles. If a large proportion of sarcomeres were initially over-long (left side of graphs), muscles extended less before massage, whereas if sarcomeres were mainly over-short (right side of graphs), they extended more easily before massage.

3.3.2 Thixotropy

The initial rise in tension during passive stretch of skeletal muscle has been interpreted as being due to the presence of numbers of long-term, stable cross-bridges between the contractile filaments in resting muscle fibres (Hill, 1968). The cross-bridge explanation has recently been challenged, with the view that viscous resistance to interfilament sliding and the mechanical properties of the elastic filaments would adequately explain the observed tension changes (Mutungi & Ranatunga, 1996b, 1996a). The term ‘thixotropy’ refers to an important property of passive muscle that its response to stretch is dependent on the immediate history of its contraction and length changes. However, Proske and Morgan argue that thixotropy cannot be explained simply in terms of viscous and viscoelastic properties (Proske & Morgan, 1999). They argue that stable actin-myosin bonds are formed at the length at which a muscle relaxes, and that this influences its mechanical behaviour in the immediate future. Proske and Morgan’s views on instability of sarcomere lengths continue to be challenged (Herzog, 2004; Telley *et al.*, 2006; Lee *et al.*, 2007).

The results in this chapter could be explained using Proske and Morgan’s ideas about thixotropy. Sarcomere lengths may become more homogenous throughout the muscle after massage, reducing the proportion of over-long and over-short sarcomeres (Fig 3.7). This would increase the number of resting bonds attaching the heads of the myosin cross-bridges to the actin filaments. The formation of more cross-bridges between the contractile proteins would also explain the greater stiffness after massage. At the same time contractile strength ought to have increased. It would be interesting to test whether spastic muscles are in fact stronger after massage.

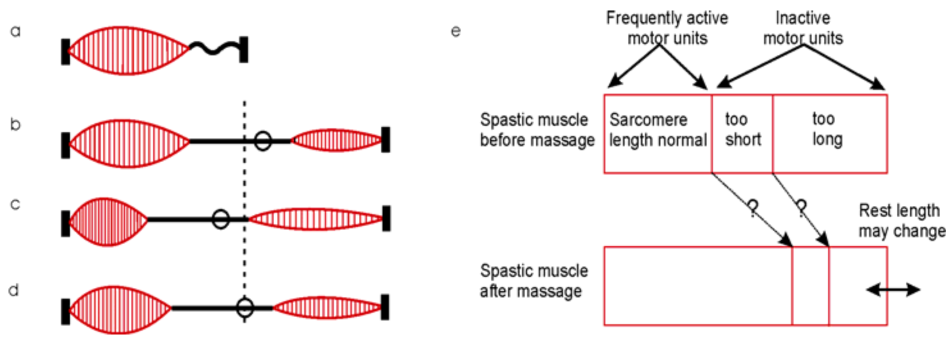


Fig 3.7 Hypothesis to explain changes to muscle rest length after massage.

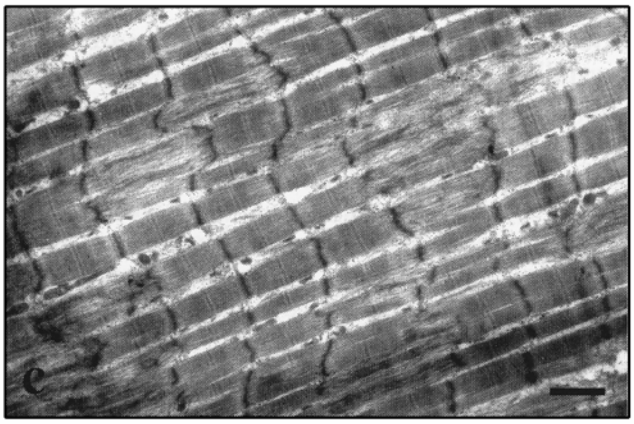
This figure explains a proposed mechanism by which improved motor function may follow massage - changes in muscle rest length. Left side shows the possible alternatives depending on the length at which the muscle as whole rests after massage (full description below). Right side shows the proposed rearrangement at sarcomere level – where more sarcomeres are of an optimal length after massage.

- **a)** According to Proske and Morgan (1999), if a contracting muscle relaxes at long length, resting actin-myosin bonds form; next, if the musculo-tendinous unit is shortened, the muscle component remains at the same length, but the tendon becomes slack.
- **b)** In the case of calf muscles, if resting bonds had formed at long length, and the muscle was shortened as in a), slack in the tendon would be taken up by shortening of the antagonist. The circle between the tendons represents the ankle joint.
- **c)** Conversely, if resting bonds had formed at short sarcomere length, the muscle would stay short, and the antagonist would be stretched.
- **d)** Massage has redistributed sarcomere lengths, resulting in a new equilibrium between the calf muscle and antagonist.
- **e)** Spastic muscles may contain a mixture of over-long or over-short sarcomeres, and massage might change only a proportion of sarcomere lengths. This would explain why muscles could be shorter or longer after massage (Fig 3.3).

3.3.3 Eccentric contractions

Spastic muscle is potentially more susceptible to eccentric contractions because of the increased risk of stretch during reflex contractions. Whitehead and colleagues have shown that when eccentric contractions were carried out at longer lengths, there was a larger shift of the optimum length for active tension in the direction of longer muscle lengths and a larger fall in peak isometric tension (Whitehead *et al.*, 2003). The optimum length for active contraction shifts to the right because there is an increase in series compliance. This is because of damage to sarcomeres (Fig 3.8 a).

a)



b)

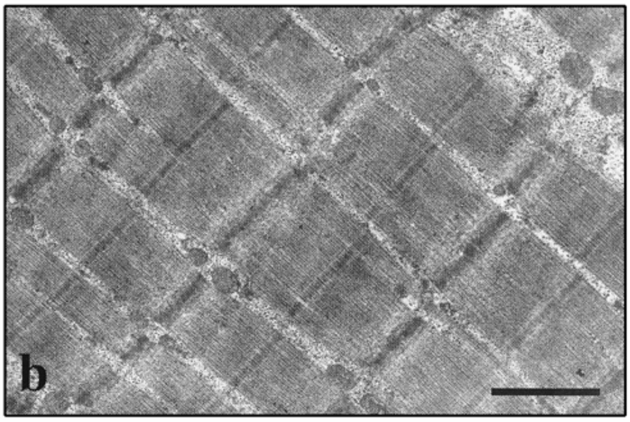


Fig. 3.8 Sarcomere damage following eccentric exercise.

Electron micrographs of longitudinal sections from the vastus lateralis muscle of one healthy human subject following eccentric exercise. Scale bars: 1μm.
a). Damaged sarcomeres one day post eccentric exercise. Note the 'wavey' line formed by the z-lines in the damaged sarcomeres.
b). Sarcomere rearrangement and repair after 14 days recovery from eccentric exercise. Z-lines restored to regular configuration, shown at double the magnification of (a). (Feasson *et al.*, 2002).

It is possible that the massage could aid reconfiguration of sarcomeres which occurs spontaneously following damage from eccentric contractions, as shown in Fig 3.8. However, it is unlikely that this was a major factor. Just as in unaffected individuals, participants in our study did not normally experience muscle soreness such as would be expected following eccentric contractions. They were only sore following unaccustomed exercise.

3.3.4 Stretch reflexes

Stretch reflexes provide the second defence against unintended lengthening of muscles as might occur for example in maintaining posture. However, in normal individuals, during passive dorsiflexion of the ankle, H reflexes recorded from the soleus muscle are inhibited indicating that passive dorsiflexion of the ankle inhibits soleus motoneurones (fig 3.9). It is therefore interesting that recordings from participant 2 during passive ankle dorsiflexion showed tonic stretch reflexes in the first two sessions, but in the fourth session phasic stretch reflexes diminished during the stretch (fig 3.6). This suggests that control of soleus motoneurones changed from abnormal stretch reflexes to the normal pattern of inhibition during passive stretch. In the other participants also the frequency of stretch reflexes approximately halved when comparing the first and last five sessions.

The reduced frequency of abnormal stretch reflexes over the 5 weeks of massage could be because regular massage sessions maintained the calf muscles in a functionally better mechanical state. Resetting sarcomere lengths to functionally more optimal values ought to change sensory feedback from muscle spindle stretch receptors since the overall length of the muscles could be reset by massage (fig 3.2). It is possible that feedback from the tendon organs could also be changed after massage. Whitehead and colleagues reported that tendon organs can monitor increases in passive tension, which they propose is caused by muscle damage following eccentric contractions (Whitehead *et al.*, 2003).

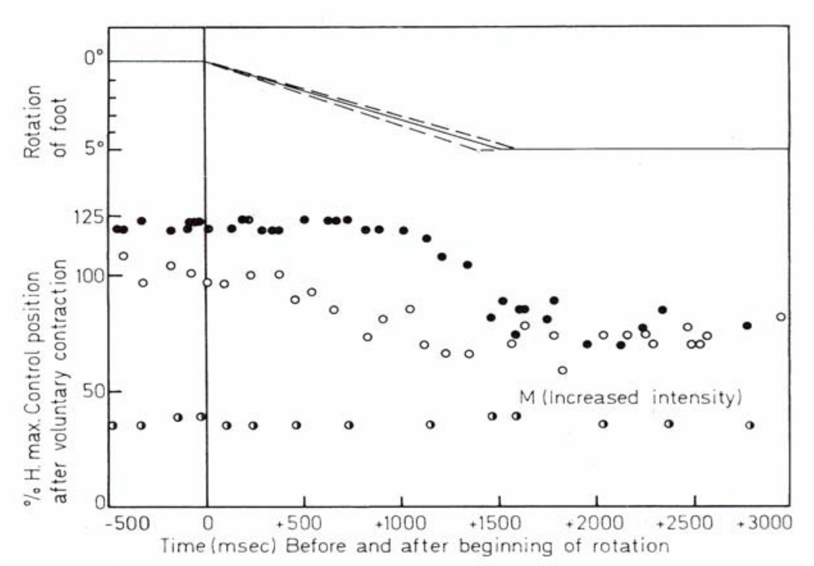


Fig 3.9 Inhibition of soleus muscle during ankle dorsiflexion

This figure from the work of Mark *et al.*, (1968) clearly illustrates reduced excitability i.e., inhibition, of the soleus muscle during stretch of the calf muscles.

The ankle of a normal subject was slowly passively dorsiflexed (top trace). H reflexes were elicited at intervals during the ankle movement (H reflexes are a measure of the excitability of alpha moroneurones). Filled circles: the reflex excitability was reduced during dorsiflexion. The authors found that after stretching and releasing the ankles, reflex excitability was increased (the initial excitability of tests indicated by open circles is 125% of control tests). After a voluntary contraction of soleus muscle prior to stretching, excitability was the same as control (open circles), but reflex excitability still fell during dorsiflexion. The direct M response of the muscle (half filled circles, tested with a stronger stimulus than the reflex tests), did not change. This indicates that movement of the ankle did not change stimulating and recording conditions.

Finally, it should be noted that much of the research into spasticity has been carried out on animals, or on humans who do not have spasticity due to cerebral palsy. For example, the work of Sinkjaer and Magnussen on the respective contribution of the component parts of stiffness in spasticity, involved patients whose spasticity was attributed to multiple sclerosis (Sinkjaer and Magnussen 1994). Mirbagheri and colleagues point out that spasticity is a syndrome and caution that their results from patients with spinal cord injuries might not be replicated in patients with spasticity from other causes (Mirbagheri and Barbeau 2001). There may also be differences if the spastic condition began in childhood or in adult life. For example, Deitz and Berger showed that adults with spasticity following a stroke could still suppress their tendon jerk reflex on heel strike, whereas children with CP could not (Dietz and Berger 1983).

CHAPTER 4

Effects of Massage on Motor Skills and Voluntary Ankle Movement

4.1 Motor skills

Motor skills were assessed in studies 1, 2, 3 & 4 using the Gross Motor Function Measure-66 (GMFM-66). As mentioned in the Introduction and Literature Review, this method has been designed and validated for use specifically with those who have CP (Russell *et al.*, 2000). It is particularly useful because the items tested and the scoring reflect situations in everyday life requiring movements of the lower limbs. For example, walking up stairs is tested with the participant holding onto one handrail only, as in normal situations; no marks are awarded for doing this if the participant has to hold onto both handrails. Russell and colleagues improved their previous measure (GMFM-88) using Rasch analysis to reflect the level of difficulty of the items. An important advantage is that any change in scoring using GMFM-66 is comparable throughout the scale of 0-100% (see Methods for a fuller explanation). Rosenbaum et al (2002) showed that motor skills as assessed by the GMFM-66 do not improve after the age of 7 in children with all types of CP (fig 1.1). Therefore, as adolescents are not expected to improve their scores spontaneously, any improvement in score is likely to be due to an intervention. For this reason the studies focused on adolescents.

4.1.2 Voluntary ankle movements

Voluntary ankle movements were assessed in 5 adolescents in study 1 as described in chapter 2, Methods. There is no precedent in the literature for testing voluntary ankle movement in CP. Surprisingly it gave an early indication of positive effects of massage in participants scoring in levels I and II.

4.1.3 Participants attendance

Twelve adolescents with spastic diplegia (six males and six females, mean age 13.9 years, age range 11-18 years) had their leg muscles massaged twice per week where possible, using the standardised massage sequences listed in Tables 1.1 & 2.2 (see methods).

Participants 1-5 (study1) had calf muscle massage twice per week for 5 weeks during April/May 2004. Participants 1-6 (study 2) had calf muscle massage twice per week where possible during the period February 2005-June 2006, although this varied (table 2.1). The more severely affected participants were offered more massage sessions. The number of weeks varied depending on availability due to curricular constraints. They were assessed for gross motor function only (GMFM-66).

Two non-ambulant participants with GMFCS classifications III and IV were given full leg massage, study 3 (table 2.2). The first of these participants had previously received massage of the calf muscles and had shown no appreciable improvement in GMFM-66 score. They were both assessed for gross motor function (GMFM-66).

Three more dextrous participants with CP administered self massage to the full leg using the sequence in table 2.3 (study 4). They were assessed with the GMFM-66.

4.1.4 Motor skills assessment – participants with CP

Motor skills were assessed by an independent physiotherapist who was accredited in using the Gross Motor Function Measure-66 (GMFM-66) (Russell *et al.*, 2002) They were assessed one week before beginning massage, and as soon as possible after their block of massage sessions finished (within one week). Some of the participants were assessed after intervals during which no massage took place. This gave an indication of how long the effects of the massage may last.

4.1.5 Statistical analysis

The GMFM-66 uses the Gross Motor Ability Estimator computer programme to calculate the individual's percentage score from their scores (0-3) for each item tested (see Methods). This includes a calculation of Confidence Intervals (CI) and Standard Error of Measurement (SEM) for scores (Russell *et al.,* 2002), indicating whether or not changes can be considered significant.

4.2 Results

4.2.1 Voluntary Movements – participants 1, 2, 3 & 4 (study1)

Trends in improved ankle joint range of movement were seen following massage (fig 4.1). The fifth participant was unable to produce active ankle movement.

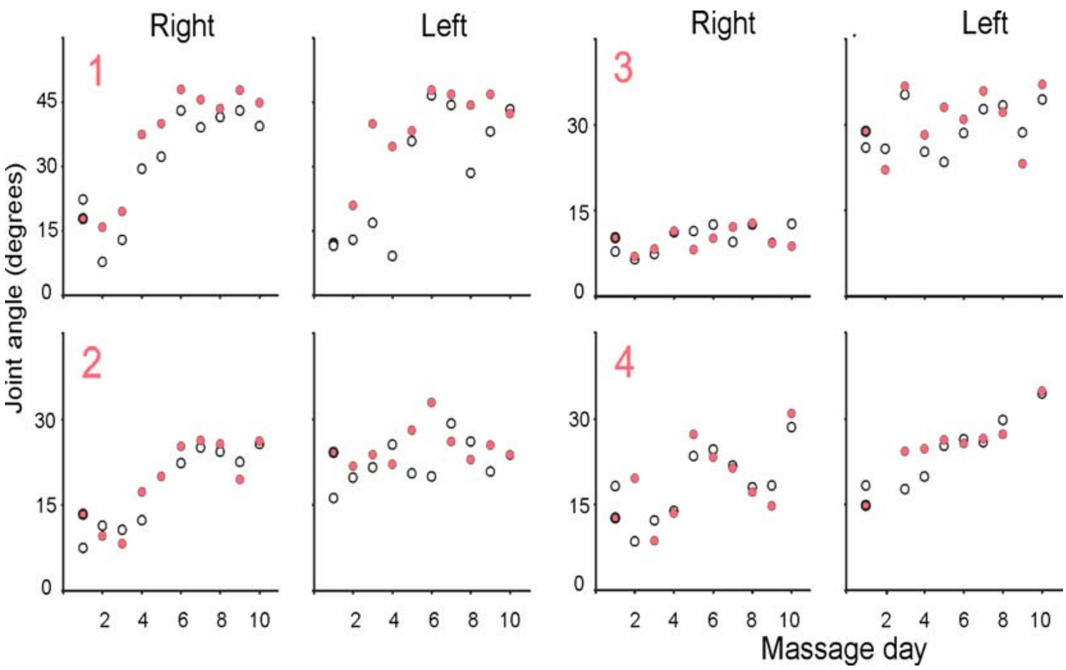


Fig 4.1 The average change in joint angle of three voluntary ankle oscillations is plotted for each individual.

Each pair of figures (1-4) plots the change in range of movement of left and right ankle joints against the session (2-10), during the first of three voluntary plantar flexions in each session before massage (open circles), then after massage (red circles).

Participant 1 (top left pair) made striking gains in the freedom of movement of both ankles as early as the third massage session. Participant 2 (bottom left) made a more modest improvement in movement in the right limb, also after the third massage session. Participant 3 (top right) made no clear gains, although there was a large disparity between legs. Participant 4 (bottom right) improved, especially in the last session when urged to contract the calf muscle, rather than inverting and everting the foot. Participant 5 in this study was not capable of active ankle movement.

4.2.2 Gross Motor Function Scores

The scores of all participants are displayed against their functional level in fig 4.2, which summarises the improvements in GMFM-66 during the periods that the participants received massage. All participants in the age range 10-18 and GMFCS range I-IV improved their scores.

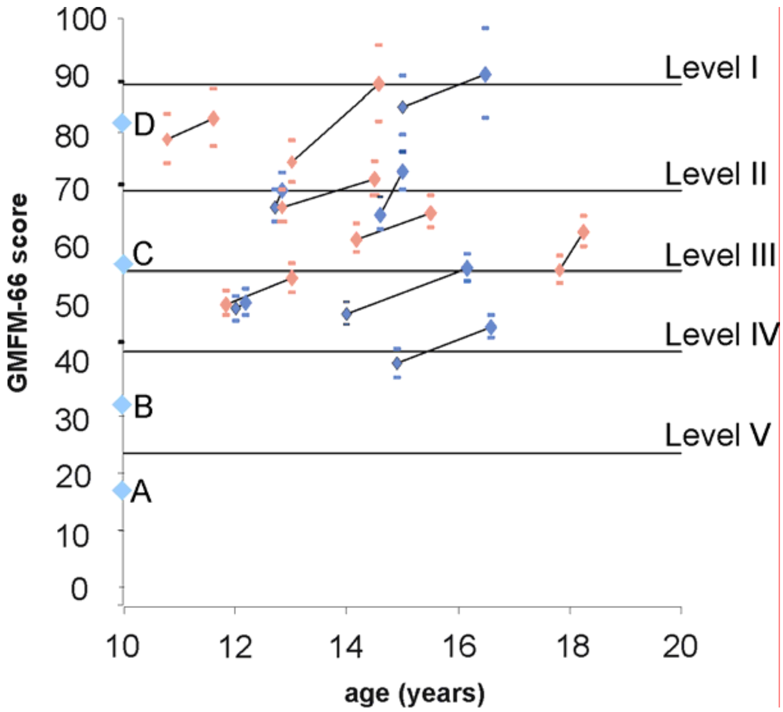


Fig 4.2 Gains in GMFM-66 scores after massage.

This figure summarises the scores and age of all participants before and after they were given massage (blue males, orange females).
I bars indicate 95% confidence intervals, calculated by the Gross Motor Ability Estimator computer programme (see p52). Five participants scoring in levels II-IV achieved significant increases.
The diamonds on the left vertical axis identify 4 GMFM-66 items that predict when children are expected to have a 50% chance of completing that item successfully. (A), is item 21 and assesses whether a child can lift and maintain their head in a vertical position with trunk support by a therapist while sitting. (B), is item 24, which assesses whether when in a sitting position on a mat, a child can maintain sitting unsupported by his/her arms for 3 seconds. (C), is item 69 and this measures the child's ability to walk forward 10 steps unsupported. (D), is item 87 which assesses the task of walking down 4 steps alternating feet with arms free.

The degree of improvement in GMFM scores did not depend on the level of disability. Figure 4.3 shows that participants with the lowest GMFM-66 scores (<50) could achieve comparable increases in score to those scoring above 60.

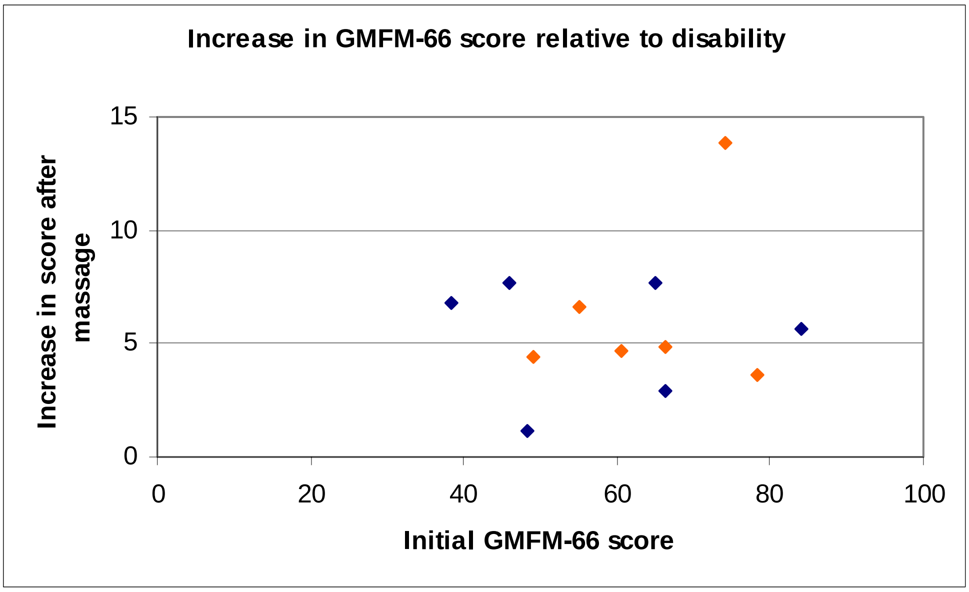


Fig 4.3 Improvements in GMFM-66 scores

The graph summarises the improvements in GMFM-66 scores for all participants with CP. No regression line is fitted here because the lengths of times for treatment vary.
Participants with lower GMFM-66 scores did respond favourably to treatment, although more massage sessions were needed in order to show changes comparable with higher scoring participants (see chapter 2 Methods for individual attendances). Blue: males, orange: females.

4.2.3 Massage of calf muscles

a). Study 1 - 5 participants received massage of their calf muscles twice weekly for 5 weeks. By the 5th week of the intervention ambulant participants numbers 1, 2, 3 and 4 had improved their GMFM-66 scores by an average of 5.11 but the score of the non-ambulant participant (participant 5) was virtually unchanged (Table 4.1).

participant number	age years/months		GMFCS level	1wk before GMFM-66	5 wks after GMFM-66	17wk after GMFM-66
1	15	0	I	84.05	89.7	89.7
2	13	1	I-II	74.16	81.93	78.28
3	12	11	II	66.33	71.22	70.81
4	14	9	II	64.98	72.16	72.63
5	14	1	III-IV	45.91	46.09	46.09

Table 4.1 GMFM-66 scores, before and after calf massage - Study 1

This table shows the changes in GMFM-66 scores before and after massage, and then after a break of 12 weeks with no massage.
Improvements were made by the more able participants (numbers 1-4) levels I-II, however the least able participant (5) did not improve measurably with only these 10 massage sessions. Later his score improved significantly after additional massage sessions - see point 4.2.4 below.

The change was statistically significant only for participant 4, although participants 1-3 also made substantial improvements. For changes to be classified as significant, participants' scores must clear the 95% confidence intervals calculated for their previous GMFM-66 scores. In general, scores improved in items testing coordination and balance (e.g. stepping over a stick at knee height) rather than muscle strength like jumping vertically and hopping. Surprisingly, gains were largely maintained 12 weeks later even though they received no massage (Table 4.1).

b). Second study - 6 further participants receiving only massage of the calf muscles all improved GMFM-66 scores (by an average of 5.0) (table 4.2).

	Age		GMFM-66	
Participant	Years/Months	Initial	Final	Increase
1	10 9	72.28	81.93	9.65
2	12 8	66.33	69.22	2.89
3	14 2	60.62	65.33	4.71
4	17 10	55.15	61.80	6.65
5	11 10	49.21	53.62	4.41
6	12 0	48.32	50.85	2.53

Table 4.2 GMFM 66 scores of participants in the second study who received massage of the calf muscles only.

Participants' GMFM-66 scores after varying numbers of massage sessions. See table 2.1 for periods during which massage was given.

Two participants (1 and 4) made significant improvements; Participants 3 and 5 made substantial improvements and participants 2 and 6 made measurable improvements. The fact that all of the participants in this study improved with the intervention is in itself remarkable. All of the improvements are at least comparable with other current interventions (see Discussion later).

4.2.4 Full leg massage – The two lowest scoring participants with CP

Increases in GMFM-66 scores for both participants were significant (Figs 4.4 and 4.5).

By the end of 65 weeks participant 1 had made a significant improvement in his GMFM-66 score (fig 4.4). He was able to achieve full marks for high kneeling (item 48) and crawling up and down 4 steps (items 46 & 47). During this period of 65 weeks, he received 21 full leg massages and 26 massages of the calf muscles. After the initial period of 65 weeks, participant 1 made further functional gains even though only his calf muscles were massaged. At 90 weeks he was able to tall kneel competently and without concentrated effort. ('Tall kneel' here means supported on the knees in the upright position with full hip extension). At 145 weeks he began to remove and put on his shoes and socks routinely. During the period between 65 weeks and 145 weeks he received 61 massages of his calf muscles. He was assessed for gross motor function at 110 weeks and again at 114 weeks. Although the total score is less in the final assessment, on this occasion he scored 2/3 for walking up four steps holding onto one rail. The effort involved may have reduced his ability to improve his score on other items (see Discussion).

Participant 2, who had the lowest initial GMFM-66 score of all the participants in any of the studies, improved his score significantly (by 6.76). Remarkably, he scored full marks (3) in item 44 (crawls or hitches forward

1.8m), item 45 (crawls reciprocally forward 1.8m), and item 46 (crawls up 4 steps) despite scoring 0 in all of these items initially.

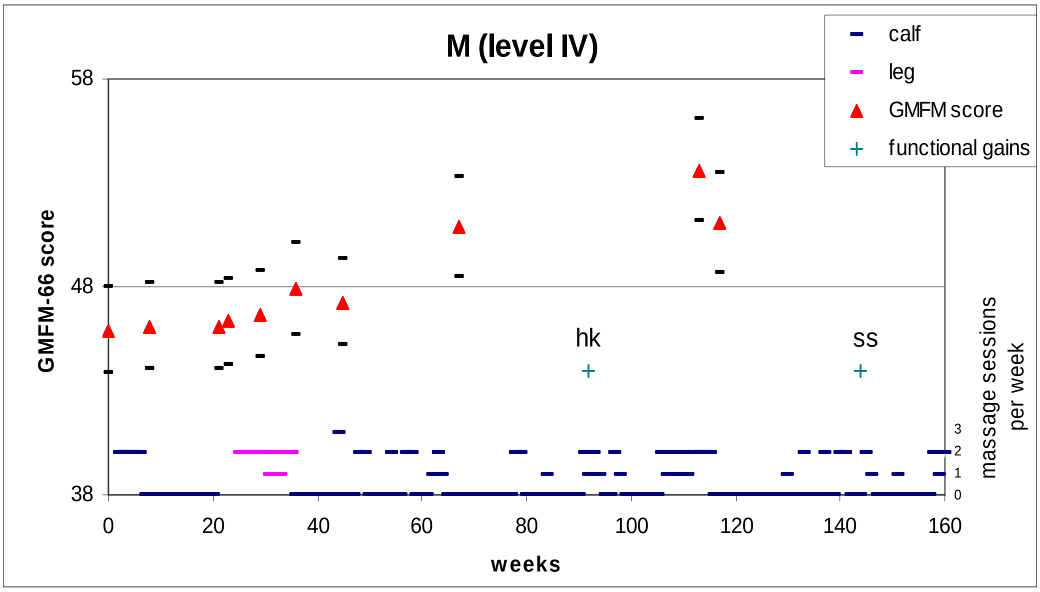


Fig. 4.4 GMFM-66 scores and massage sessions – Participant 1 (study 3)

The figure shows the progression in GMFM-66 scores made by this participant, who received both calf muscle massage (blue) and also whole leg massage (lilac) at different times (see main text for full details and reasons).
The upper part of the figure shows the participants' GMFM-66 scores (red triangles) with confidence intervals shown by short horizontal black lines above and below. The lower part of the figure shows the frequency of massage sessions for this participant over three years.

Despite being the second most disabled participant in all of the studies, he eventually made statistically significant improvements in his GMFM-66 score. Note that there were long periods during which massage was not available (bottom blue line marked 0).

Horizontal blue and lilac lines indicate the number of massage sessions per week, 0, 1, 2, or 3. Black horizontal bars indicate limits of confidence intervals, calculated by the GMAE computer programme. hk: high kneeling, ss: routinely puts on/takes off shoes and socks. See main text for a more detailed description.

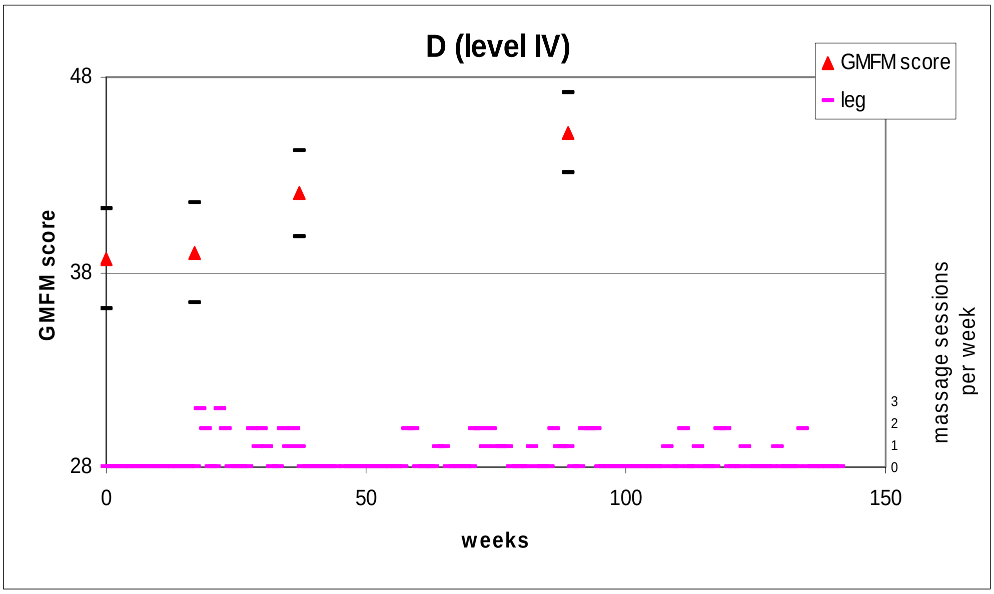


Fig. 4.5 GMFM-66 scores and massage sessions - Participant 2 (study3)

The figure shows the progression in GMFM-66 scores made by this participant, following whole leg massage sessions.
The upper part of the figure shows the participants' GMFM-66 scores (red triangles) with confidence intervals shown by short horizontal black lines above and below. The lower part of the figure shows the frequency of massage sessions for this participant over almost three years.
Horizontal lilac lines indicate the number of massage sessions per week, 0, 1, 2, or 3. Note that there were long periods during which no massage was given (horizontal lilac lines marked 0).

This figure shows that despite being the most disabled participant in all of the studies (initial GMFM-66 score 38.38), he made statistically significant improvement in GMFM-66 score (final score 45.14). This confirms that the massage can have an effect irrespective of the degree of disability, although more massage sessions may be needed.

4.2.5 Self massage – (full leg)- a pilot study

All three participants in this pilot study had participated in the initial study which involved massage of the calf muscles twice weekly for 5 weeks (see above).

Participant 1 had the highest initial GMFM score of all 12 participants. His score improved by the end of the initial study (fig 4.6) and this improvement was maintained after 32 week without any massage. He had 33 sessions of self-massage over 74 weeks but there were no further improvements in his score (fig 4.6). Although he had a score of 90 he had lost only 2 marks in the entire assessment (one mark each in items 80, jumping and 82, hopping). These items require strength more than skill.

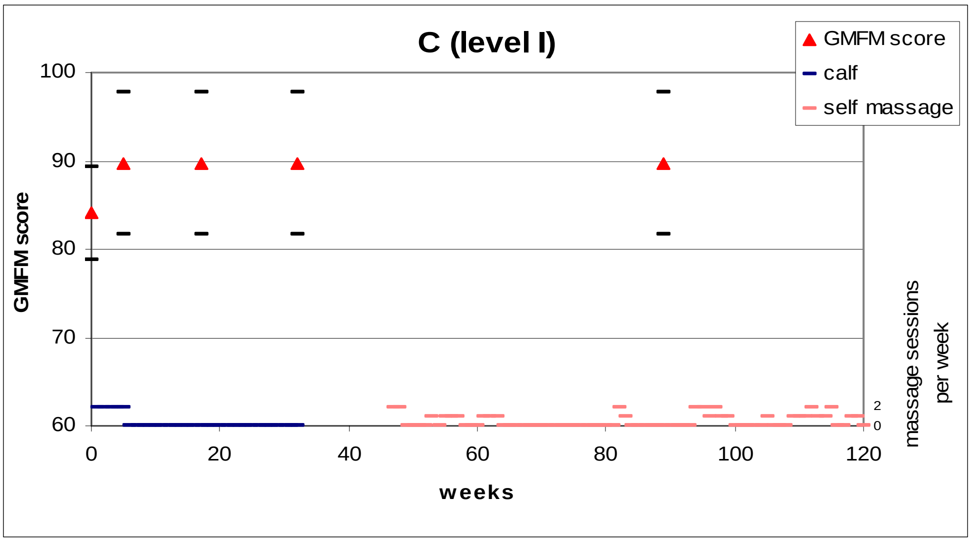


Fig. 4.6 Participant 1- GMFM-66 scores, type of massage and attendance

The figure shows the GMFM-66 scores for this participant who originally had calf muscle massage during study 1 (blue) and then self massage during this study (pink). This high scoring participant made no further improvements after his initial increase from 84.05 to 89.7. Confidence intervals (horizontal black lines above and below red triangle, score) calculated by GMAE programme.

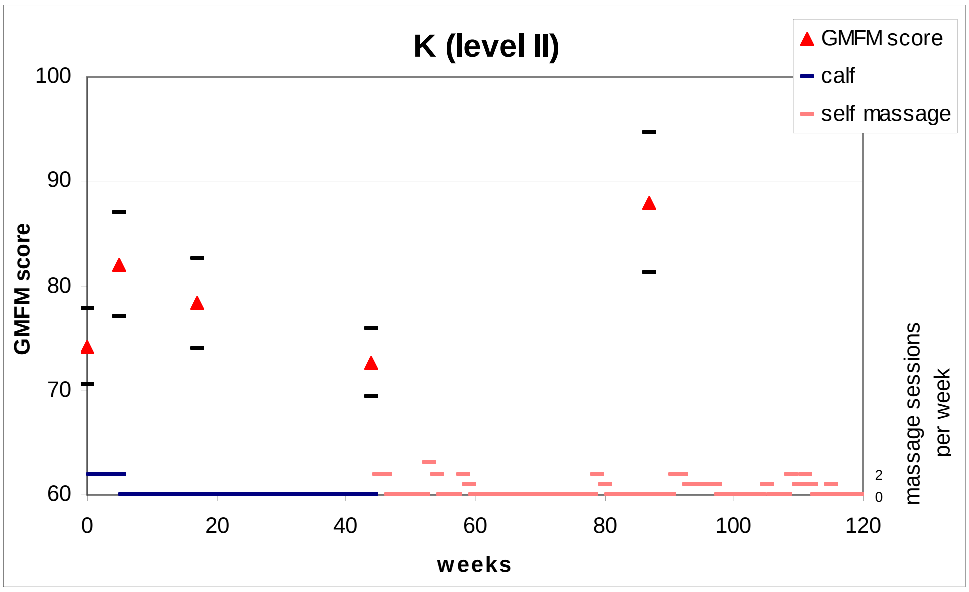


Fig. 4.7 Participant 2 GMFM-66 scores, type of massage and attendance.

The figure shows the GMFM-66 scores for this participant who originally had calf muscle massage during study 1 (blue) and then self massage during this study (pink). Note the long periods during which no massage was given (horizontal lines pink and blue marked 0).
After the initial rise in score following 10 calf massage sessions over 5 weeks, her scores for the next two tests dropped during the period when she had no massage. Her score improved significantly following 15 sessions of self administered massage (compare the last two GMFM-66 scores).
Confidence intervals (horizontal black lines) calculated by GMAE programme.

Participant 2 improved her GMFM-66 score by 7.77 after receiving 10 calf muscle massages over 5 weeks. During the following 44 weeks she received no massage and her score dropped to 74.16. Her score then increased by 13.83 to 87.99 following 15 sessions of self administered full leg massage over 42 weeks. This improvement was significant and was the largest of all participants in any of the studies (fig 4.3). She improved her scores in 7 of the 8 most difficult items and was 3 points short of a full score (100) by losing 1 point on each of items 3 items, 74 (walking in a straight line), 80 (jumping) and 82 (hopping).

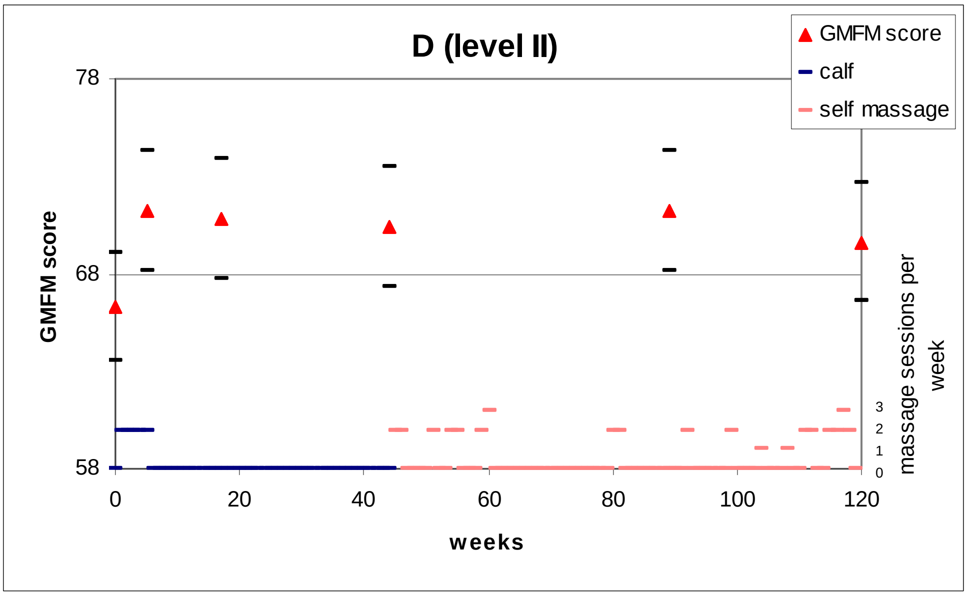


Fig. 4.8 Participant 3 - GMFM-66 scores, type of massage and attendance.

The figure shows the GMFM-66 scores for this participant who originally had calf muscle massage during study 1 (blue) and then self massage during this study (pink).
The notable point about this participants' scores is that even although there were long periods during which no massage was given (horizontal lines pink and blue marked 0), her scores remained above the original score, indicating a 'carry-over' effect of the massage.
Confidence intervals (horizontal black lines) calculated by GMAE programme.

Participant 3 initially improved her GMFM-66 score from 66.33 to 71.22, having received 10 calf massage sessions over 5 weeks. Her score fluctuated during the time she administered self-massage, but was maintained above that of her initial score. Her score for items 86 and 87 (going up and down 4 steps, hands free) increased from 2 to 3 in both cases.

4.2.6 Walking down stairs (GMFM-66, items 85 and 87).

A task that young people with spastic CP find particularly difficult is to walk down stairs with alternating steps and hands free (item 87), see discussion. Only 5 participants could attempt this test. Initially only one participant (participant 1, study 1) gained full marks for item 87. After massage three other participants (2/1, 1/2 and 2/2) also gained full marks for this test. A less exacting test is to walk down 4 steps holding on to one handrail (item 85). Two participants (3/1 and 5/2) gained full marks for item 85. Participant 3, study 2 made no improvement on item 87 (table 4.2) although he had full marks for item 85 already on his initial test. Participant 4, study 2, did not improve her scores for item 85, but her condition is not typical of spastic diplegia.

Subject/ study	GMFCS	Item 85 Before	Item 85 After	Item 87 Before	Item 87 After	Massage sessions
1/1	I	3	3	3	3	10
1/2	II	3	3	0	3	14
2/2	II	3	3	2	3	15
3/1	II	1	3	0	0	10
3/2	II	3	3	0	0	14
2/1	II	3	3	0	3	10
4/2	II	2	2	-	-	27
5/2	III	0	3	-	-	6

Table 4.3 Rating (0-3) scores for all participants tested on items 85 and/or 87

This is a record of the scores rated 0-3 for the 8 participants who were capable of attempting the tests, for each of the two items involved in walking down stairs (item 85, with hands - item 87, hands free).
Negotiating stairs involves a very complex series of motor coordination. Improvement in this shows improved motor skills and is likely to be very beneficial to those who previously could not attempt stairs.
The main text above expands on the specific details for progress in these participants' scores.

4.3 Discussion and Conclusions

4.3.1 Motor skills

This is the first study to investigate the effects of massage on motor skills, in both short and long term, in adolescents with spastic diplegia. The main findings were: firstly, improved GMFM-66 scores for all participants who received calf muscle massage. The improvements were largely maintained during spells when no massage was given. Second, massage was effective in raising scores of participants with a range of disability, from GMFCS levels I-IV. Both participants with low GMFCS scores who received full leg massage improved their GMFM scores significantly. Third, some participants classified level I and I/II responded surprisingly quickly with massage of the calf only (two improved significantly within 6 weeks). The more disabled participants required massage over months in order to make significant gains. Fourthly, one of the three participants who self-administered full leg massage improved her GMFM significantly

A disadvantage in this study was that assessments of GMFM-66 scores had to be completed within a school lesson period. This may have underestimated the abilities of some individuals, particularly in GMFCS levels III and IV owing to exhaustion. It is well known that tiredness rapidly develops in CP (Bottos & Gericke, 2003). The developers of the GMFM do not stipulate that all of the tests have to be done immediately following one another. It may be that some participants would have scored differently given rest, especially where they attempted tests which they had not be able to attempt before.

Some adolescents with CP are overweight because they are unable to take sufficient exercise owing to their disability (Bottos & Gericke, 2003). An additional factor is the necessity for fast transfer between lessons at school, which encourages the tendency to use a wheelchair rather than walk. This applies to participant 5 in study 1, who could walk when he was younger. At the beginning of the study he was adept at wheelchair hockey.

The ability to play goalkeeper in football, which he attained after receiving massage, not only improved his quality of life, but it gave him the potential to improve his cardiovascular fitness. Pimm uses the term ‘physiological burn-out syndrome’ to describe the excessive mental and physical effort that adolescents with CP experience when trying to produce movement in daily life (Pimm, 1992).

Improvement for the lower scoring participants may have been better if it was possible to massage them twice per week as had happened in the first group. Where participants had massage over a number of months, there were inevitably long periods were no massage was given, e.g. during school holidays. Despite this, with one exception (participant 3), scores did not return to or below the initial baseline. That participant’s final score did improve dramatically once massage was resumed (fig 4.7).

The largely maintained gain in GMFM scores during times when no massage was given strongly suggests that neural adaptive changes had occurred. For example, participants 1, 2, 3 and 4 study 1 were given no massage for 16 weeks due to examinations and school holidays but none of their GMFM-66 scores dropped back to their original scores (see table 4.1).

4.3.2 Alternating movements and the descent of stairs

An early indication of the effect of the massage was seen in the improved range of voluntary movements of individuals who were classified with higher GMFCS scores (fig 4.1). Alternating limb movements are particularly difficult for children with CP (Leonard, 1990, Lin, 2003). The challenge is to control Ia inhibitory interneurons which receive direct input from muscle stretch receptors (Jankowska, 1992). Those interneurones are excited by impulses in the cortico-spinal pathway as well as by other descending pathways.

There is evidence implicating Ia interneurones in the inhibition of soleus motoneurones when the calf is passively stretched (Gritti & Schieppati, 1989), (see chapter 3). Fig 4.9 shows that soleus motoneurones are similarly inhibited during active dorsiflexion of the ankle but not during plantarflexion. (Romano & Schieppati, 1987) argue that this inhibition is necessary during lengthening contractions of soleus muscles, otherwise lengthening would lead to an increased contraction owing to the mechanical properties of the muscle. Lengthening contractions come into play when descending steps (fig 4.10). According to Schieppati and co-workers soleus motoneurones are inhibited not only via Ia pathways from the antagonist, but interestingly also from gastrocnemius muscles. In fact, they suggest that the interneurones belong to a single group (Schieppati *et al.*, 1990).

Table 4.4 highlights the improvements made by the majority of participants tested in descending stairs. Three participants in GMFCS level II walked down 4 stairs with hands free; two of those would not attempt this task before. Of four others scoring in levels II to III, two gained full marks in walking down stairs holding onto one rail. There was no change in score for the remaining two. Thus 5 out of 7 participants made striking gains in their ability to walk down stairs. This could imply that these participants became more adept at inhibiting reflex contractions of their soleus muscles, which is essential in this activity.

During the voluntary movements participants were not encouraged to make maximal movements, and this could be regarded as an inconsistent means of obtaining a measure. The reason for this is stated in the methods, i.e. to avoid causing participants stress, possibly affecting supraspinal control. The test conditions could not be considered similar to those where, for example, VO_2 max is being tested in athletes. Despite this, a trend was seen where the range of ankle movement was improved after massage. This, in itself, can be considered a vindication of the methods used here, as the participants were given the same instructions before and after massage. However, the call for an absolute against which the change could be measured is accepted as a valid criticism of the method used here.

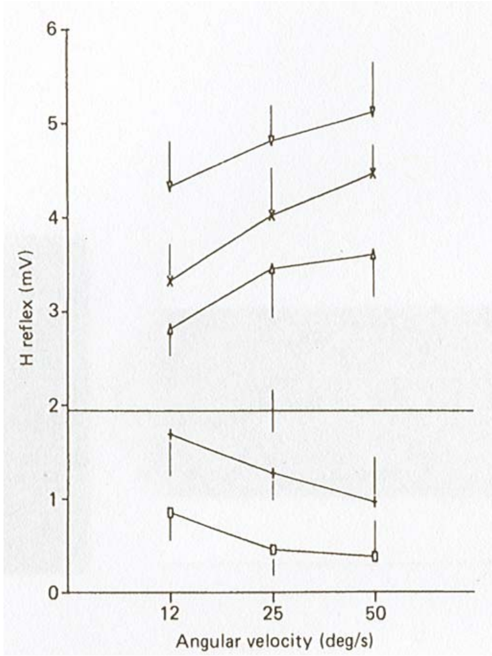


Fig 4.9 Increasing inhibition of soleus motoneurones with increased active stretch.

This figure from the work of Romano & Schieppati, (1987) illustrates that increased stretch of the calf muscles results in increasing inhibition of soleus muscle contraction.

H reflexes are a measure of the excitability of the α motoneurones. The horizontal line represents 0 degrees of plantar flexion. Below the horizontal line the soleus muscle is actively lengthening and above it the muscle is actively shortening. □ represents H reflex responses at a predetermined value for the amplitude of integrated EMG at 3 angular velocities. †and Δ indicate α motoneurone excitability at twice this predetermined value. This gives a direct comparison at the same level of contraction, showing that during active lengthening H reflexes are reduced probably as a result of soleus motoneurones receiving greater inhibition than during active shortening. X and ▼ are H reflex responses at 3 and 4 times the initial predetermined contraction during shortening of the soleus muscle.

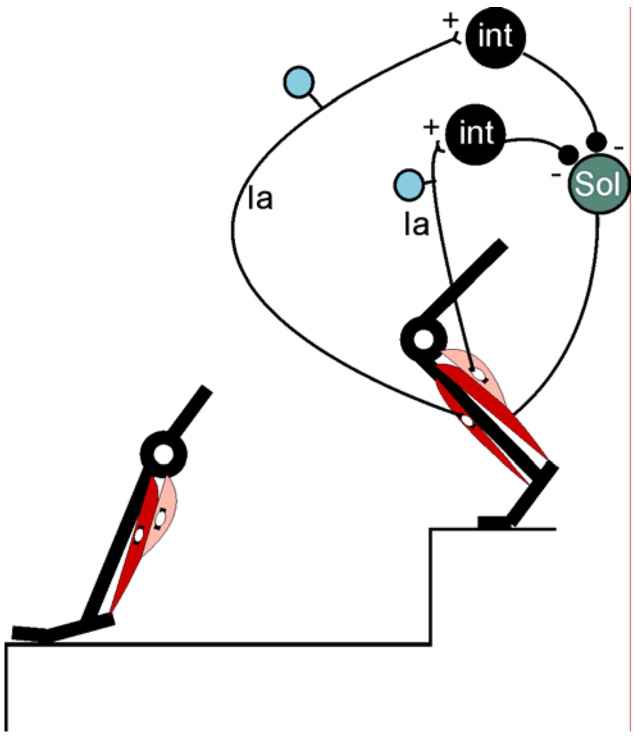


Fig 4.10 Diagram of neuronal connections highlighting the difficulties faced by an individual with cerebral palsy when walking down stairs.

The right side of the figure shows pathways involved in the inhibition of soleus motoneurones during lengthening contractions via Ia inhibitory interneurones from gastrocnemius and tibialis anterior muscles. This is consistent with the findings of Scheippati *et al*., (1990). If these motoneurones are not inhibited, lengthening increases the force of contraction owing the mechanical properties of the muscle. In spastic diplegia lengthening the soleus muscle initiates stretch reflexes rather than inhibition (see fig 3.6). Another difficulty for these individuals is the control of clonus set up when their foot reaches a step (left side of diagram) (Baird & Gordon, 1983).

CHAPTER 5

Effects of massage on skin temperature and oxygenation of spastic muscle.

5.1 Introduction and background

After massage, increased skin temperature has often been observed by massage practitioners and by their patients, although no specific measurements are taken. This is usually accompanied by an improvement in the patient's condition, shown by a reduced sensation of pain and/or a perceived greater ease of joint movement. A tenuous link is often suggested between the rise in temperature, increased blood flow and improved athletic performance. However, this effect and its proposed benefits have had very limited scientific backing.

Back massage sessions lasting six minutes have been shown to increase the temperature recorded from the skin in healthy female participants, although the temperature returned to baseline after 10 minutes (Longworth, 1982). Those results do not distinguish between increased skin temperature and the temperature of the underlying muscle. However, if the temperature change was only due to friction of the skin, it seems likely that the additional heat would be dissipated before the change in temperature could be recorded. More recently, deep effleurage massage has been shown to significantly increase the temperature of the vastus lateralis muscles in seven healthy males (Drust *et al.*, 2003). A needle thermistor was used in that study and temperature changes were detected to a depth of 35mm into the muscle. The largest temperature increases were recorded in the muscle nearer the surface of the skin (2-2.7 ^{o}C at a depth of 1.5cm, 1.5-1.7 ^{o}C at 2.5cm depth and 0.1-0.5 ^{o}C at 3.5cm depth). These results could indicate that the effects of the massage were more superficial.

In an initial pilot study conducted by the author, 3 participants with CP received the calf muscle massage sequence described in Chapter 2 twice per week for 3 weeks. The temperature recorded from the skin over the massaged muscles was always increased after massage (by around 4ºC).

It was suggested that this observation might have a practical use as a possible means of assessing the effectiveness of a trainee's massage. The increase in skin temperature during massage could be due to frictional effects on the skin, or to frictional effects in the underlying muscle - or both. The amount of friction to the skin is likely to be about the same for both the trained masseur and the trainee because they both deliver the same number of strokes per minute. In fact, oil is used to reduce skin friction. Frictional effects within the muscle are likely to depend on the pressure as well as the rate that the strokes are applied. The pressure was designed to be firm enough to move the muscle underneath without being so firm that the patient flinched (see chapter 2). If the same timed sequence is used, any differences in temperature by the end of the sequence are likely to be attributable to differences in the frictional effects on the muscle.

It has been postulated that massage may alter the blood flow of the massaged tissue (Goats, 1994). Only two of seven studies investigating blood flow, and reviewed by Weerapong and colleagues (Weerapong *et al.*, 2005), were considered to be free from technical problems (Tiidus & Shoemaker, 1995; Shoemaker *et al.*, 1997). Both of those studies showed no change in total muscle blood flow following massage. However, the Pulsed Doppler Ultrasound used in those studies detected changes in the limb large arteries and veins that supplied other tissues besides the massaged muscles but not necessarily in the microcirculation of those muscles.

The current study investigated the effects of massage on the blood supply within the muscles, using near infrared spectroscopy (NIRS). NIRS is a non-invasive method of monitoring oxygen availability and utilization by the tissues. In intact skeletal muscle, NIRS affords measurement of changes in oxyhaemoglobin (HbO_2), reduced or deoxyhaemoglobin (Hb), total haemoglobin (HbT), and assessment of the redox state of the mitochondrial cytochrome c oxidase (CtOx). It is also possible, with the use of specific tracers, to make accurate assessment of regional blood flow (Boushel & Piantadosi, 2000). However, that last option was not used in this study because it is invasive.

The aims of the work recorded in this chapter were firstly, to investigate the effects of the massage on the temperature recorded from the skin over the massaged muscles. Second, to assess if temperature changes could be used to determine the effectiveness of a trainee masseur. Third, to investigate the effects of the massage sequence on oxygenation of the massaged muscles, using NIRS equipment.

Firstly, changes in skin temperature with massage were investigated with 3 adolescents who have spastic diplegia and 4 unaffected adults (study 5). Later, changes in skin temperature and muscle blood flow/oxygenation were investigated with two gender and age matched groups of adolescents with and without spastic diplegia. The effects of massage on oxygenation of the muscles were compared with the oxygenation changes produced by contraction of the same muscles during resisted plantar flexion. It was also possible to monitor the effects of each stroke of the massage concurrently, as well as effects before and after the massage (study 6).

Material and Methods are recorded in chapter 2.

5.2 Results

5.2.1 Change of skin temperature with massage

Figure 5.1 and table 5.1 show temperature changes recorded from the skin over the calf muscles comparing before and after massage for groups 1-4.

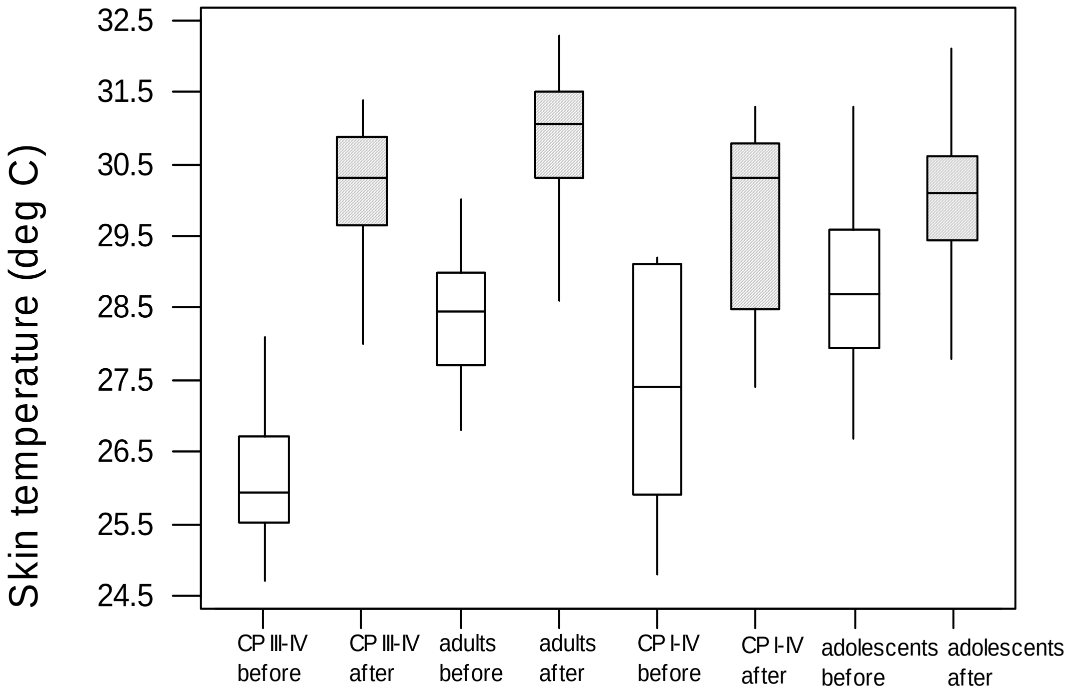


Fig 5.1 Temperatures recorded from groups 1-4 before and after massage

Box plots showing skin temperature over the calf muscle before massage (clear) and after massage (hatched) for 4 groups, over varying numbers of observations (see table 5.1 for observation numbers). Top and bottom lines of box plots indicates the range of temperatures for each group. Intermediate line in box plots signifies the mean.
Both groups of adolescents with CP had mean starting temperatures of 26.5ºC before massage. Both groups without CP had mean starting temperatures of 28ºC. However all four groups finished with temperatures statistically the same (30.5ºC) after massage. This suggests that the adolescents with CP regularly have limb temperatures lower than optimal for producing motor function.

Group	Number of participants	Number of observations	Mean before	Mean after	P value
1: CP	3	27	26.01	30.18	<0.05
2: Adults	4	32	28.37	30.92	<0.05
3: CP	7	21	27.4	29.82	<0.05
4: controls	7	21	28.8	30.05	<0.05

Table 5.1 Change of skin temperature with massage – groups 1-4

Table shows skin temperatures over the calf muscles before and after massage for all four groups, together with the number of observations. The mean temperatures for all four groups after massage were statistically the same (t-test), although the adolescents with CP had starting temperatures around 2ºC lower.

Figure 5.1 and table 5.1 show that the initial temperatures of adolescents with CP are lower compared with healthy adolescents or adults. However the skin temperatures after massage were approximately the same for all four groups (not statistically different).

It was not appropriate to compare initial skin temperatures of groups 1 and 2 (adolescents with CP and adults). One reason is that the room temperatures were significantly different (more detail of this is given later). However measurements of skin temperatures for groups 3 and 4 were made in comparable conditions. The mean room temperatures were the same. The initial skin temperatures for group 3 (adolescents with CP) were significantly lower than those of group 4 (healthy adolescents) – p=0.0014.

However, temperature after massage was not significantly different in these two groups (p=0.47).

5.2.2 Effects of room temperature on skin temperature change with massage

Throughout the study variations in room temperature were observed in both schools and in the laboratory where the adults (group 2) received massage (ranging from 17.4 – 24.9°C). Thus the skin temperature could even fall during massage if the room temperature was low. Consequently, the effects of room temperature on initial skin temperature and skin temperature following massage were examined. Although skin temperatures could increase during massage when room temperature was below 20^{o}C, there were three instances when skin temperatures were lower after massage (two in Ashcraig School and one in Hutcheson's School). There were also five instances in Hutcheson's School when skin temperatures were lower after massage despite recorded room temperatures of 20^{o}C and above. All these cases occurred in a session when there was a pronounced draught. When all the data were included, there was only a weak relationship between room temperatures and increase in skin temperature during massage (fig 5.2, R^2 0.02 and 0.29 for Hutcheson's and Ashcraig Schools respectively). Indeed, if the cases at Hutcheson's School and Ashcraig School were omitted where the room temperature fell below

20°C and when there was a draught, there was no relation between room temperature changes on skin temperature after massage (R^2 = 0.0008 and 0.16 for Hutcheson's and Ashcraig Schools respectively).

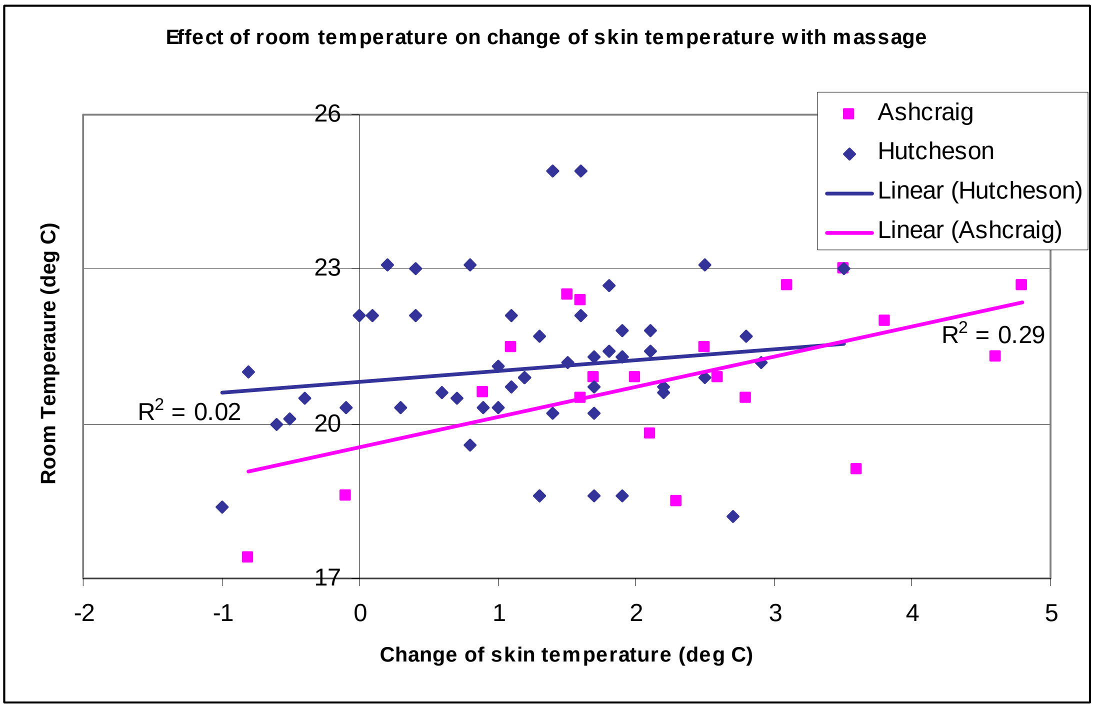


Fig 5.2 Plot of changes in skin temperature after massage against room temperature.

The graph plots the changes of skin temperature against the room temperature for the participants with CP (lilac squares) and the control group (blue diamonds) for all sessions at both schools.

Summary – the relationship between room temperature and skin temperature is weak. If room temperatures were above 20°C then there was no effect. Thus effects of ambient temperatures on change in skin temperature during massage can be discounted providing they are ≥21°C.

5.2.3 Use of skin temperature differences following massage to determine the effectiveness of a trainee

Analysis of the effect of environmental conditions on the increase in skin temperature during massage indicated that massage may not increase skin temperatures in cool or draughty conditions (fig 5.2). Therefore it is important to take this into account if trainees are to be assessed by their ability to increase skin temperature by massage. Furthermore, even when room temperatures were 21^{o}C and above, there was still a wide variation between increases in skin temperature recorded from individual participants. This suggests that a fair comparison can be made between the ability of a trainee and an experienced masseur to increase skin temperatures by massage, providing environmental conditions are the same and they massage the same subject. Fig. 5.3 shows the difference between the temperature changes achieved by a trainee and a masseur when each massaged one calf of the same individual consecutively. All individuals were healthy adults (group 4). When the trainee's skill increased with practice (as indicated by the number of sessions), the difference between their ability to increase skin temperatures decreased. This is seen clearly in fig 5.3 with a very strong trend in diminishing differences as the number of massage sessions increased.

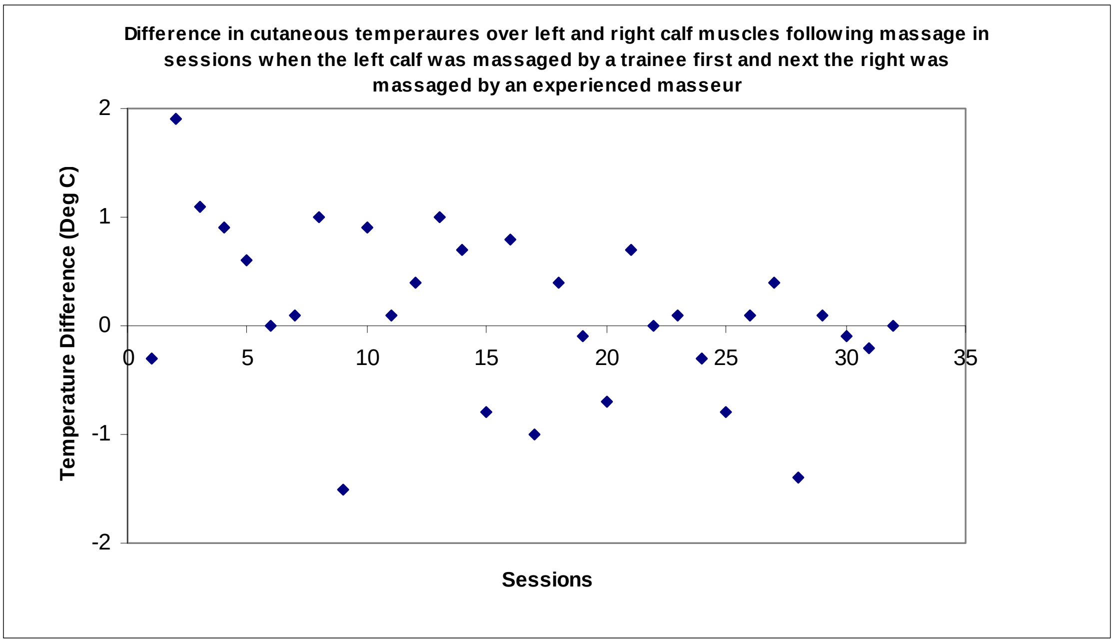


Fig 5.3 Comparison of skin temperature difference achieved by the trained masseur and a trainee in control group (4).

The graph is a record of the differences in the calf muscle skin temperature change produced by massage from the trainee masseur and that from the experienced masseur, for four healthy adult volunteers over eight sessions for each volunteer. In every session the left leg was massaged first by the trainee in the massage technique, and then the right leg was massaged by the experienced masseur. Positive changes indicate a difference of change brought about by the experienced masseur, negative ones by the trainee.

As the session numbers progressed, left to right on the graph, the difference between the change produced by the trainee and that produced by the experienced masseur became less, indicating that the trainee was becoming more proficient in the massage technique.

An additional sign of the trainee's improved effectiveness was his ability to produce a change in the temperature of the opposite leg (i.e., a contra lateral effect), as the number of sessions progressed. A contra lateral effect had already been noted in the massage by the experienced masseur. The trainee always massaged the left leg before the masseur massaged the right leg. The average temperature of the right leg before massage compared to the left before massage was +0.18°C in the first 8 sessions, whereas in the last 8 sessions it was +0.36°C. Thus the masseur was 'at a disadvantage' because the skin temperature of the right leg had already increased. Fig 5.1 shows that the massage sequence increases skin temperature to a ceiling of about 31°C, so any increase in temperature depends in the initial skin temperature.

5.2.4 Changes in oxygenation

The NIRS equipment provided an indication of changes in oxygenation of muscle during 3 resisted contractions of the calf muscles, then throughout the massage itself, and during 3 contractions following the massage (see tables 2.4 & 2.5 for the timing of those). The raw data output from the NIRS shown in fig 5.4 is from one session for one participant in the control group and is typical of those from all of the participants in the study. The figure displays changes in reduced haemoglobin (Hb), oxyhaemoglobin ($HbO_{2)}$, total haemoglobin (HbT) and cytochrome oxidase (CtOx) during each massage stroke and during three resisted contractions, both before and after massage. During tests of 3 contractions of the calf muscles (resisted plantar flexion), Hb, HbO_2 and HbT all decreased, due to compression of blood vessels within the muscle. After contractions, Hb, HbO_2 and HbT returned to baseline.

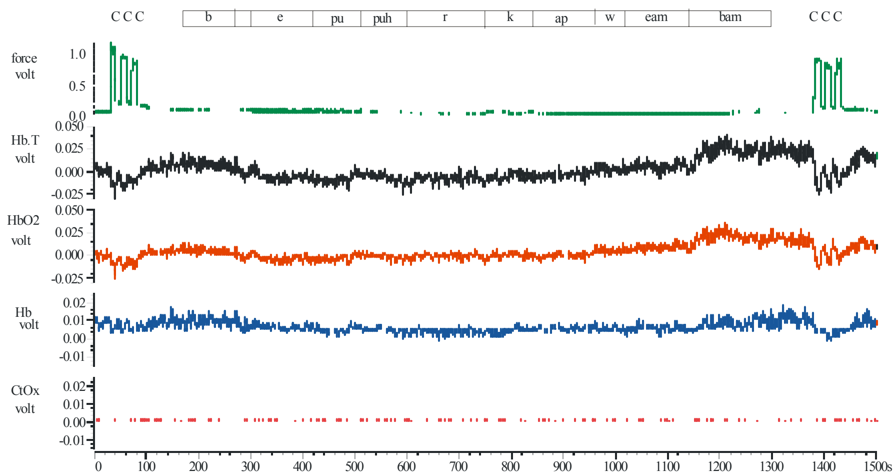


Fig. 5.4 Changes of voltage recorded from the NIRS equipment during one test sequence on a participant in the control group.

The figure illustrates results from one session for one control subject. It is typical of that from the other participants in the study. Once the massage began with effleurage, there was an initial decrease in Hb, HbO_2 and HbT. However, once the massage strokes became more vigorous (appositional rolling and wringing) a steady increase in Hb, HbO_2 and HbT occurred. This steady increase remained to the end of the massage and throughout the settling out period, up to the final test of 3 contractions, indicating an increase in oxygenation of a magnitude at least comparable with that shown during the resisted contractions.

The voltages on the vertical axis are shown at different scales for clarity of observed change. Hb: reduced haemoglobin, HbO_2: oxyhaemoglobin, HbT: total haemoglobin, CtOx: cytochrome c oxidase. ccc = three voluntary contractions. The letters in the horizontal boxes represent each of the massage strokes shown between the two sets of three contractions. b: baseline; e: effleurage; pu: picking up; puh: pick up and hold; r: rolling; k: kneading; ap: appositional rolling; w: wringing; eam: effleurage at end of massage; bam: baseline after massage.

In the example illustrated in fig 5.4, once the massage began, there was an initial decrease in Hb, HbO_2 and HbT. However, once the massage strokes became more vigorous (appositional rolling and wringing) a steady increase occurred in Hb, HbO_2 and HbT. This increase remained to the end of the massage and throughout the settling out period (bam), up to the final test of three contractions.

Changes in total haemoglobin (HbT) best reflect changes in perfusion in the massaged tissue, without using invasive methods. Fig 5.5 is an example of changes in HbT, taken from the same recording as in fig 5.4, but at a higher resolution.

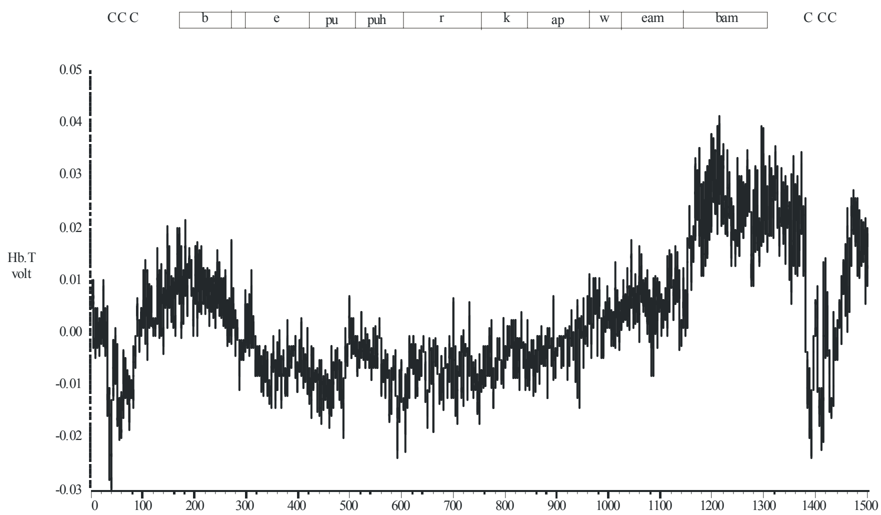


Fig. 5.5 Changes in the total Haemoglobin (HbT) at high resolution

This figure shows in higher resolution the recording of the HbT from fig 5.4. It shows a trend that was typical of the recordings from all of the participants during the massage sessions. Three active resisted contractions were done before and then after the massage sequence (ccc). This gave an indication of the significance in the size of the changes observed during and immediately after the massage.

The levels of HbT decreased during the three contractions and then returned to the initial baseline (b) before massage. From the start of massage with effleurage, the HbT decreased during the series of massage strokes until increasing with the more vigorous strokes (appositional rolling and wringing) and then remained elevated during the final effleurage. Once the massage ceased, the baseline after massage (bam) was higher than the original baseline. HbT fell again during the three active contractions after massage. The increase in HbT following massage was of the same magnitude as the reduction in HbT during the active contractions. Horizontal axis denotes seconds. Abbreviations as for fig 5.4.

The data shown in Figs 5.4 and 5.5 confirms that there are changes in oxygenation of the massaged tissue and that those changes are comparable in magnitude with the ones seen during the voluntary contractions.

The mean voltage, together with the standard error, was computed for Hb, HbO_2 and HbT (see data analysis section for detail) before and after massage, and during each massage stroke. HbT was of most interest since it can give an indication of changes of perfusion within the calf muscles.

To determine if the general trends seen in the NIRS were significant, confidence intervals (CI) were calculated for each of the means for initial and final baselines and each massage stroke. In the example shown in fig 5.6, the changes in HbT were found to be significant (95% CI). Each participant's data was examined to determine if HbT changed significantly during massage. Significant changes did occur at some stage during massage in every session for participants at both schools.

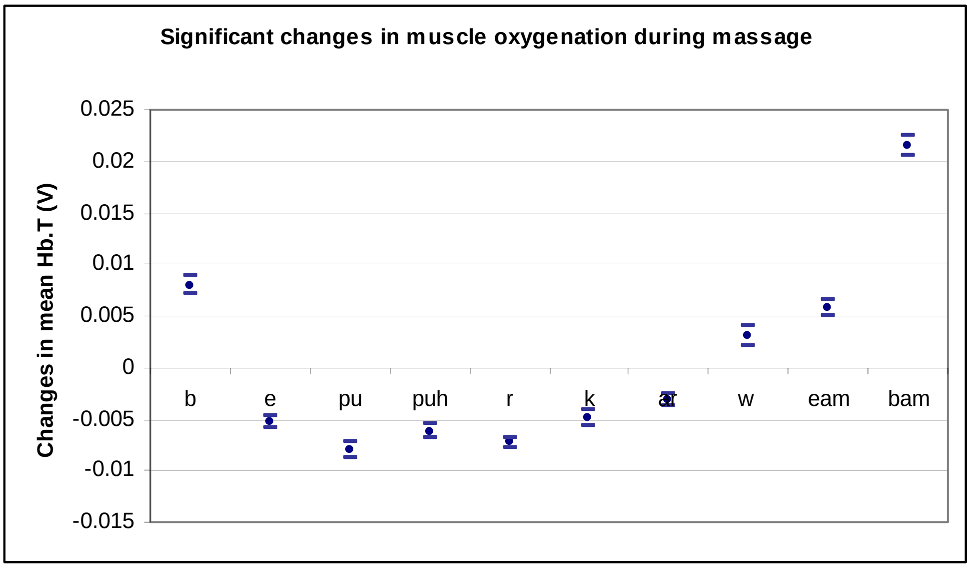


Fig 5.6 Mean values and 95% confidence intervals for total haemoglobin (HbT) during initial and final baseline periods (b and bam) and during each massage stroke (same sequence as in figs 5.4 and 5.5).

The figure records changes in HbT from the same participant as in figs 5.4 and 5.5. Short horizontal lines above and below the means indicate 95% confidence intervals. Since there is no overlap in the 95% confidence intervals for the mean HbT during initial and final baseline periods, that change is significant. Similarly, almost all confidence intervals for the means of HbT for successive massage strokes do not overlap, indicating that there were significant changes in HbT with changes in individual massage strokes. Abbreviations as in fig 5.4.

Note: the same massage sequence was used each time as this was considered likely to be most likely to bring about improvements in motor skills (see chapter 2) and the whole study was designed to assess the effectiveness of this particular sequence, not to compare the effects of different sequences.

5.2.5 Comparison of responses from participants with CP and controls

The responses to massage were compared in the control group and the participants with spastic diplegia. In the control group, two different responses were observed when the calf muscles were massaged; the first type of response was that the participants' HbT began to rise once massage started and remained elevated even after the massage finished (fig 5.7). The second type of response was a decrease in HbT during massage, but once the settling out period began, there was an increase, though not to the same extent as the first type of response (compare averages in figs 5.7 & 5.8).

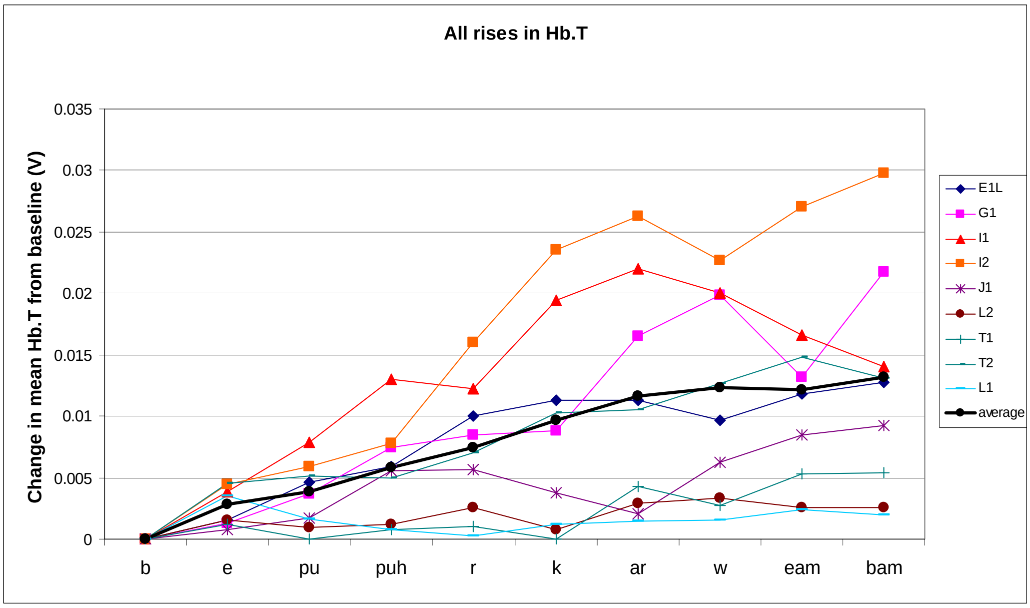


Fig 5.7 All of the rises in HbT – control group.

The figure records the first type of response to massage in the control group – elevated levels of HbT from the start and throughout the massage. This shows all of the sessions for the controls where HbT rose from the start of the massage and remained higher than at the starting baseline, even after the massage finished. Increases from the baseline were evident for every stroke for these participants. Numbered letters on the right refer to specific individuals during specific sessions. b = baseline, e = effleurage, pu = pick up, puh = pick up and hold, r = rolling , k = kneading, ar = appositional rolling, w = wringing, eam = effleurage at the end of massage, bam = baseline after massage.

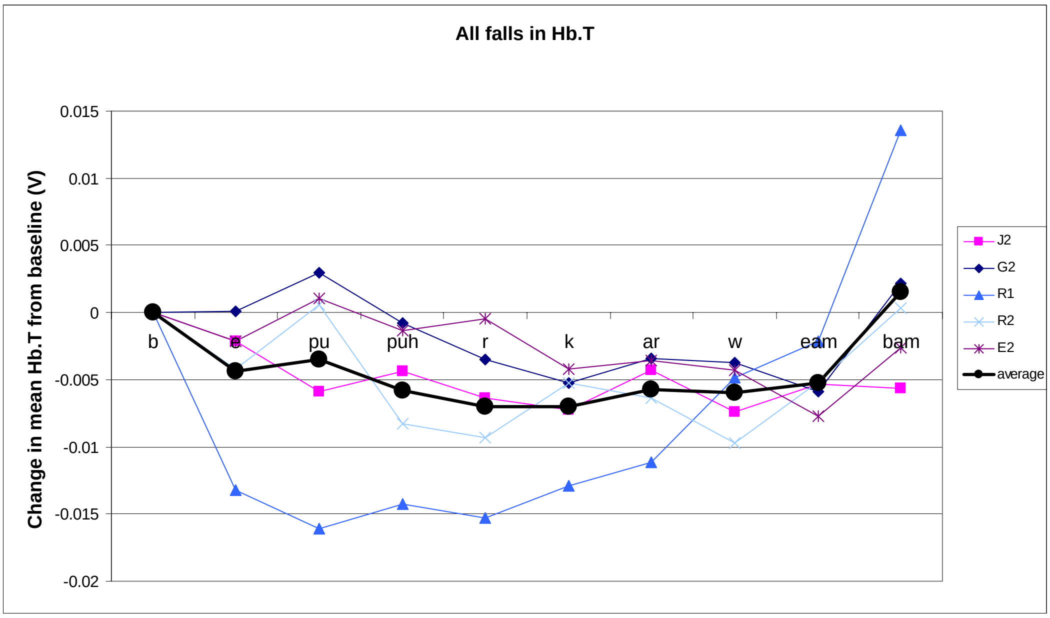


Fig 5.8 All of the falls in HbT – control group.

The figure records the second type of response to massage in the control group – sessions where HbT fell after the massage began. However, by the end of the massage the HbT had on average risen above the initial base line.

Numbered letters on the right refer to specific individuals during specific sessions. b = baseline, e = effleurage, pu = pick up, puh = pick up and hold, r = rolling, k = kneading, ar = appositional rolling, w = wringing, eam = effleurage at the end of massage, bam = baseline after massage.

Two patterns could be discerned in participants with CP, but they were not as clearly evident as in the control group. On average the HbT increased above the baseline by the end of the massage although there was a sustained reduction in HbT in two participants in two massage sessions (D1 & J1 in fig 5.9). Among the controls the HbT remained level after massage or increased in the majority of cases, even when HbT had initially fallen (fig 5.8). However, among participants with CP it usually fell. The fall was particularly striking in the participant who was an elite swimmer and has the highest score of all the participants with CP (participant C). His HbT fell significantly below the baseline. However, among the rest, despite falls in HbT after the final effleurage, HbT still remained elevated during the settling out period (bam in fig 5.9) or was unchanged from the baseline. If data from participant C were excluded, the average HbT during the settling out period remained markedly higher than baseline.

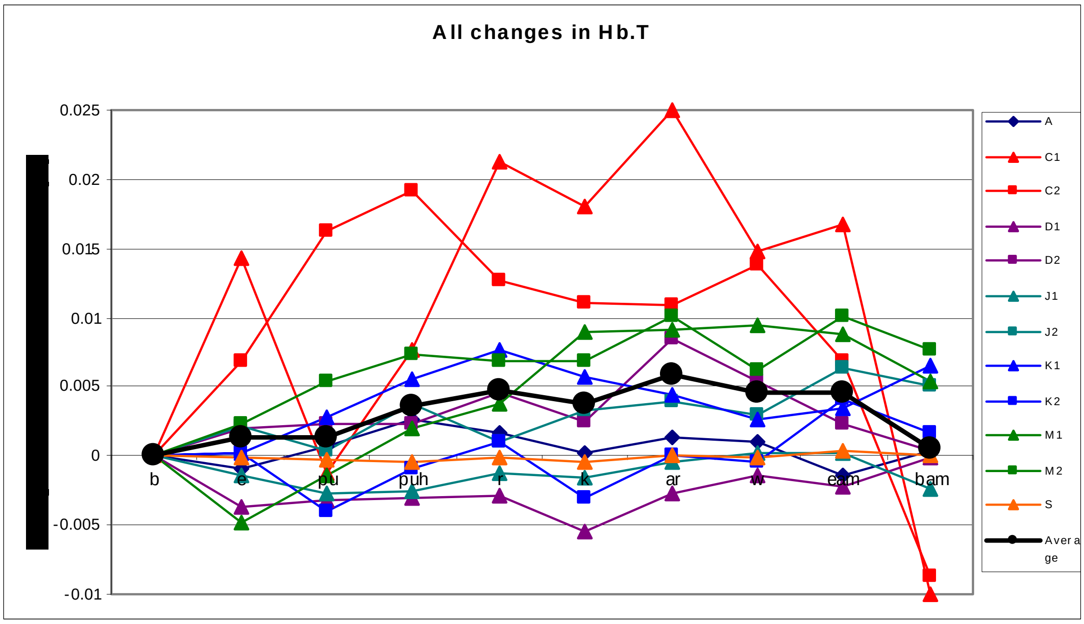


Fig 5.9 All the changes in HbT – Participants with CP

Figure shows all of the changes in HbT for the participants with CP. Two trends are discernable – one where there is an increase in HbT from the outset of massage and the other where there is a drop in HbT. There were twice as many instances of increase in HbT as there were drops in HbT. However, for both responses, the final HbT level after massage was still above initial baseline, and this is more apparent if the extreme response from participant C is excluded.
Numbered letters on the right refer to specific individuals during specific sessions.
Abbreviations as for fig 5.8.

5.2.6 Contralateral effects of massage

In all of the cases where the temperature was recorded, an increase in the temperature had been noted in the non-massaged leg, prior to it being massaged, i.e. the massage produced a contralateral effect. It was therefore of interest to assess the effects of massage on the oxygenation of the non-massaged leg. Following massage of the right leg, the NIRS optodes remained in place on the right leg while the left leg was massaged. On average the HbT increased in the right leg while the left leg received massage (fig 5.10). Unfortunately this was only possible for the control group due to the longer time necessary for the disabled participants to tae part in the massage and testing sessions.

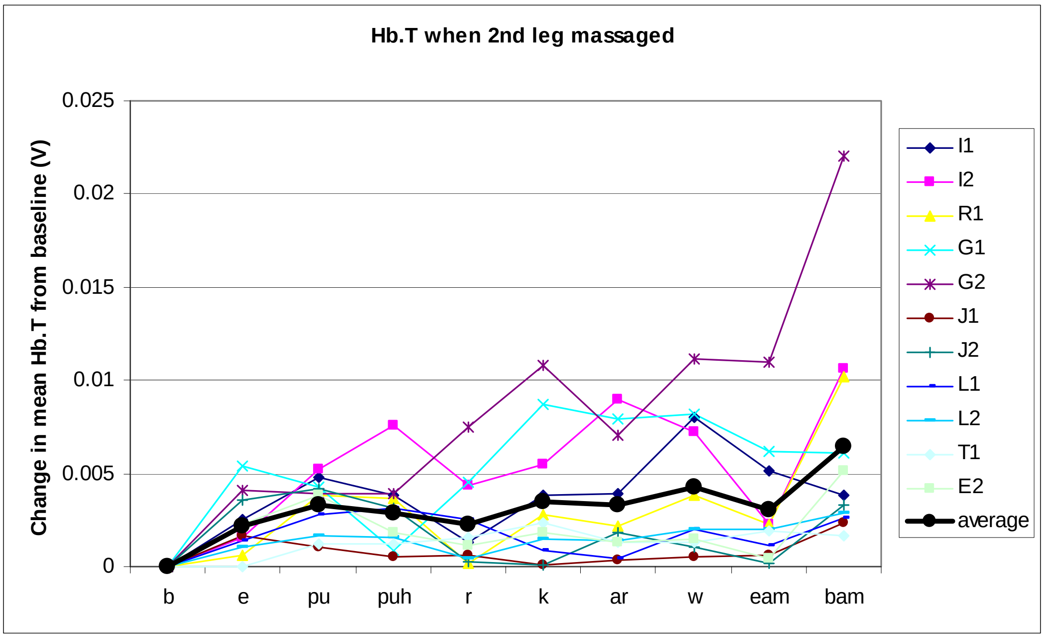


Fig. 5.10 Effects of massage on opposite leg - control group.

Data from the control group during a limited number of sessions when the NIRS equipment remained in place on the right leg whilst the left leg was massaged. This shows the changes in mean HbT from the baseline for the right leg HbT which remained elevated as the left leg was massaged. This is additional evidence of the effects of massage on the circulatory system, i.e., another contralateral effect of the massage in addition to the changes in temperature already noted. Numbered letters on the right refer to specific individuals during specific sessions. Abbreviations as for fig 5.8.

By comparing the responses of particular individuals, it was possible to draw conclusions about the effects of the massage on both the temperature change and specific aspects of the oxygenation of the massaged tissue. Suggestions could also be made on whether those changes may have a bearing on the limited motor control evident in CP.

5.2.7 Comparison of athlete and sedentary participant in control group

It was established that most of the participants in the control group exercised occasionally although not on a regular basis. Two participants were in stark contrast with one another - one was a very competitive sportsman, while the other was a very intense academic. Their HbT responses are displayed in fig 5.11, which shows that during their massage, HbT was always increased in the sportsman (squares) and always decreased in the academic (triangles). This suggests that vasodilatation could be a typical response in a trained athlete or in individuals who regularly exercised, while vasoconstriction could be a typical response from a sedentary person. It is accepted that these conclusions are based on a very limited number of observations.

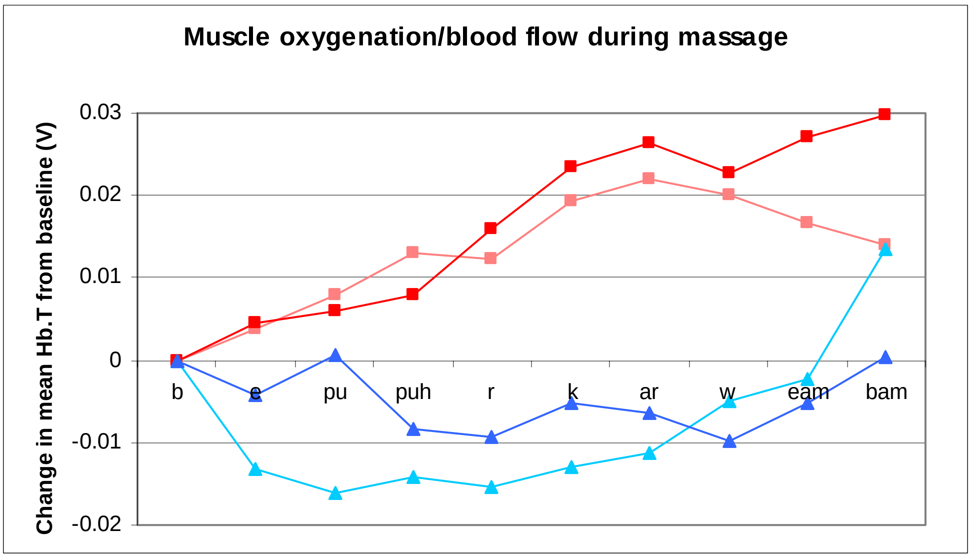


Fig 5.11 Comparison of HbT responses of an academic and a sportsman in the control group.

The figure records the HbT responses of two participants in the control group to two massage sessions each. Red squares – sportsman, blue squares – academic. Two differing responses are evident – from the outset the HbT is elevated in the sportsman, but the opposite response applies to the academic. However, in three of the four tests, the HbT was elevated after massage, suggesting improved circulation in the massaged tissue. Abbreviations as for fig 5.8.

5.2.8 Comparison of athlete and wheelchair user in CP group

Interestingly, three participants in this group showed similar disparities between an athlete and a sedentary person. Results for participant1, who swam at international level, showed HbT increasing (vasodilatation) and remaining elevated throughout the massage. By contrast, HbT decreased (vasoconstriction) in two habitual wheelchair users (participants 4 & 9) (fig 5.12) although the change was not as striking as in the academic participant in the control group.

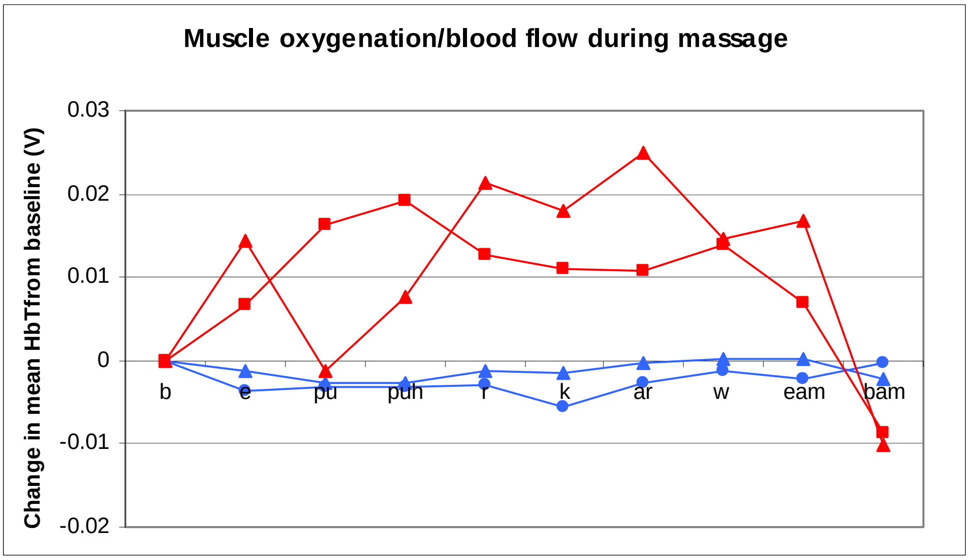


Fig 5.12 Comparison of participants with CP - 2 habitual wheelchair users and one sportsman.

The figure shows the changes in HbT, comparing two massage sessions for the sportsman with CP (red) with one session each from two habitual wheelchair users who have CP (blue). The sportsman's HbT increased from the beginning of the massage and remained elevated throughout the massage, although there was a steep drop immediately after the massage finished. The wheelchair users' HbT varied very little from the base line although it fell slightly from the outset of massage but did return to about the initial baseline after massage. These results suggest that vasodilatation is the typical response from an athlete, whereas vasoconstriction was observed in the wheelchair users. Abbreviations as for fig 5.8.

5.2.9 Comparison of participant 1 in CP group with two controls

In addition to differences in HbT between the two groups, there were also some striking differences in the recordings of HbO_2 and Hb. These were seen particularly in recordings where the optodes were orientated lengthwise along the muscle rather than transversely. Recordings with this orientation appeared to be more sensitive. The transverse orientation was preferred initially because it allowed more exposure of the muscle for massage. The optodes were orientated in the lengthwise direction for participants I and R among the control group, and participant 1 in the CP group. Fig 5.13 shows that in general massage caused a vasodilatation in participant 1. However, there was much greater variability in all outputs from the NIRS equipment, and in one recording the Hb gradually fell during the massage. These observations are referred to in the Discussion.

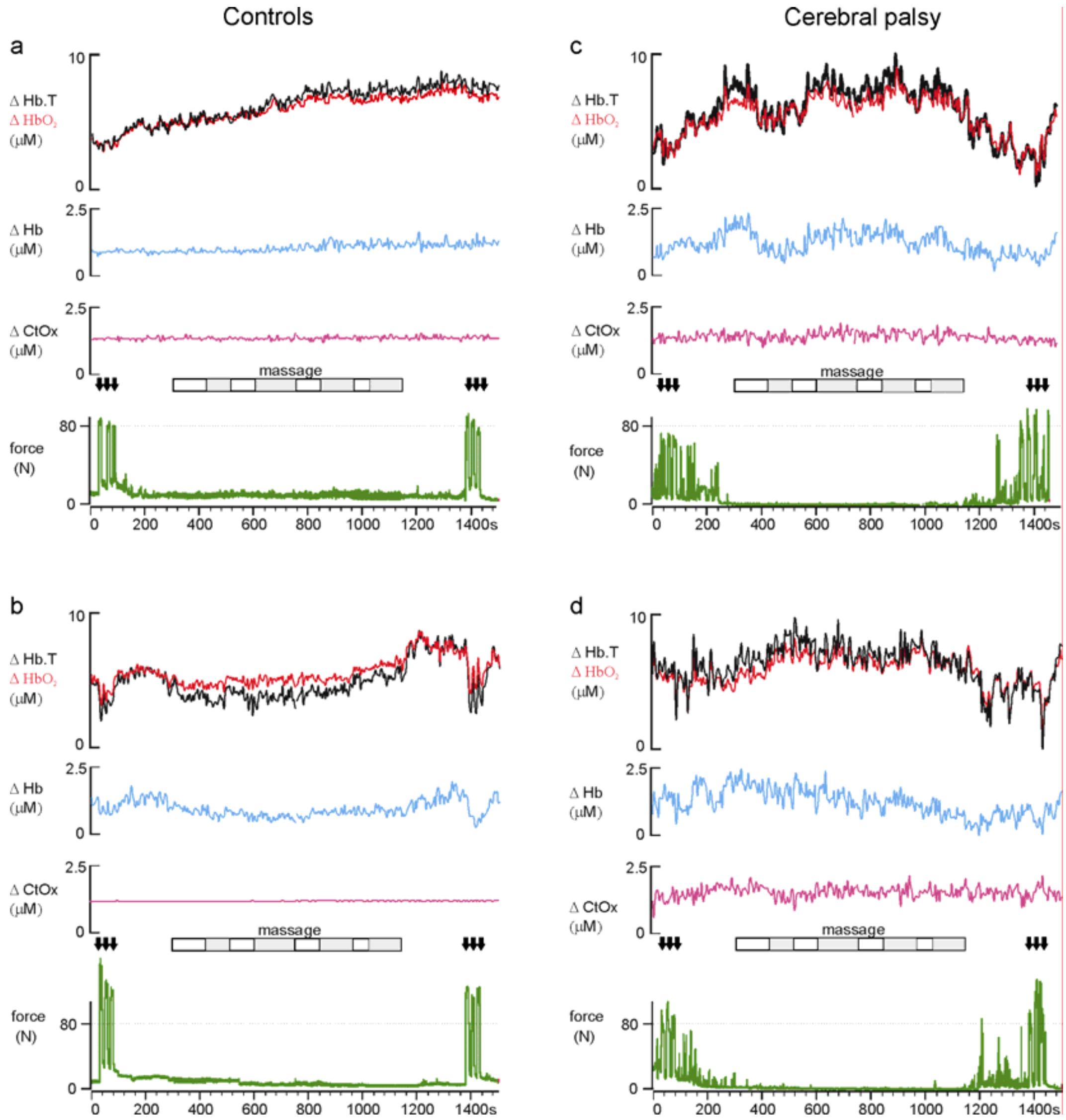


Fig 5.13 Comparison of variations in HbT, HbO_2, Hb and CtOx, participant 1 in CP group and two controls.

The figure shows four sets of data – one from a sportsman (a), and one from an academic (b) both in the control group; and two results from the sportsman who has CP (c & d).

The values for HbT, HbO_2 and CtOx are averages over periods of 5s, sampling at 5Hz. The scale for Hb and CtOx is half of that for HbT and HbO_2. Vertical arrows indicate voluntary contractions.

a) The HbT, HbO_2 and Hb all increased gradually during and after massage, suggesting vasodilatation. The HbT trace rises above the HbO_2 trace towards the end of the record because of the increase in Hb.

b) During massage HbT, HbO_2 and Hb all fell, suggesting vasoconstriction. In contrast to (a) the Hb trace lies below the HbO_2 trace because the Hb was also reduced. During voluntary contractions (arrows), HbT, HbO_2 and Hb all fell more than in (a), the force record shows that the contractions were stronger.

c) and d). The HbT trace frequently rises above the HbO_2 trace, indicating simultaneous increases in both HbO_2 and Hb as in (a). Note the much greater variability in HbT, HbO_2, Hb and CtOx traces than in (a) and (b) and also the involuntary reflex contractions in the force traces recorded during the first and last 300s when the foot contacted an unyielding surface. In (d) the Hb gradually fell during massage, and at the beginning of massage HbO_2 increased while CtOx decreased.

5.2.10 Relationship between change in skin temperature and change in HbT

The results for the groups with CP and the control group show an opposite relationship between differences in skin temperature and in HbT before and after massage. (fig 5.14).

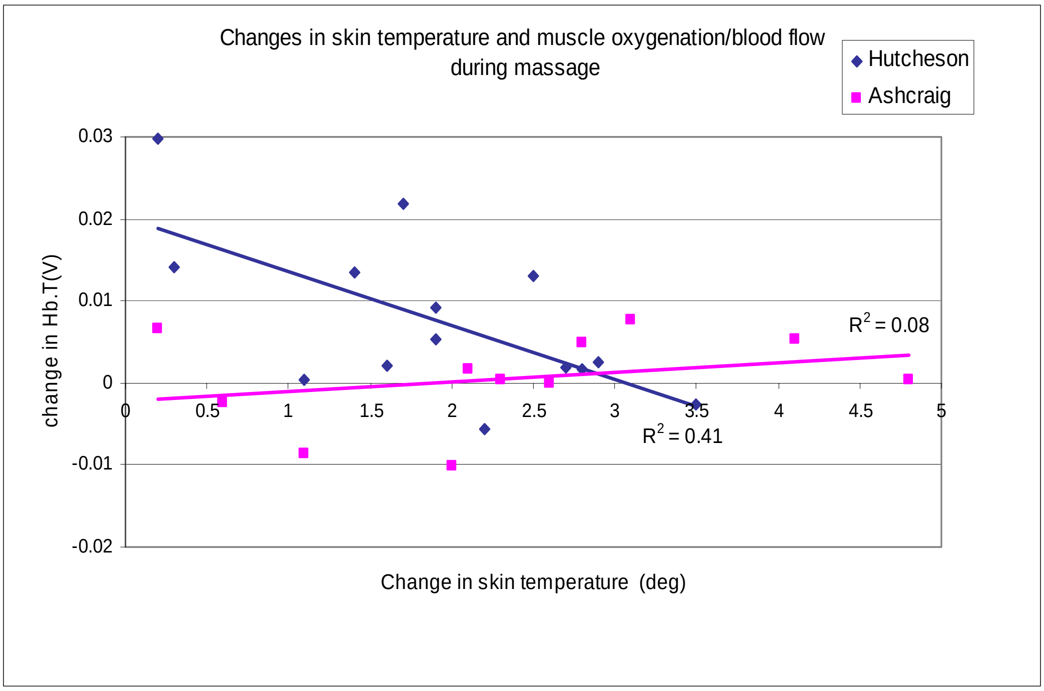


Fig 5.14 Comparison of skin and HbT changes in the CP group and the control group.

Figure compares the changes in HbT and changes in skin temperature for all of the participants during all sessions when the room temperature was above 20ºC - CP group (lilac) and controls (blue).

In the control group smaller increases in skin temperature accompanied greater changes in HbT. In those with CP, changes in skin temperature and HbT were only weakly related (R^2=0.08). When the data from participant 1 (whose HbT fell below baseline after massage, see fig 5.9) was removed, there was no relationship between the two variables (R^2 = 0.008). In the control group, there was an inverse relationship (R^2 =0.41) between change in HbT and in skin temperature with changes in HbT falling as the temperature rose. It is difficult to determine the significance of this as these were control group data, and not the CP on whom the intervention was centred.

In some initial sessions, the room temperature for the control group was below 19°C. In other sessions, skin temperature fell during the massage because of draughts very close to the participants, although the room temperature was above 20°C (see above). The data from both of those instances was not included in fig 5.14.

5.3 Discussion

This is the first study to investigate the effects of massage on oxygenation of the calf muscles in adolescents with CP. The aim of the study was to determine if massage increases muscle oxygenation, and whether this was linked with changes in temperature recorded from the skin.

5.3.1 Effects of massage on oxygenation of the muscles

The data from the NIRS equipment show distinct changes in Hb, HbO_2 and HbT, indicating that massage did have an effect on the muscle oxygenation. The magnitude of the changes was small, in the range of a few micromoles of haemoglobin (a change in haemoglobin of 2 micromoles would equate to about 10 ml of blood volume change). However, by customising the path length for each person, statistically significant changes were observed throughout the massage procedure.

The increases and decreases in HbT observed in both the CP group and the control group were likely due to the effects on the blood vessels within the massaged muscles. In normal homeostatic conditions, the blood flow in the muscles prior to massage would be constantly adjusted via vasoconstriction or vasodilatation to meet the current metabolic demands. Immediately at the onset of exercise in humans there is a reduction in muscle blood volume and oxygenation in the exercising limb, probably due to vasoconstriction (Taylor *et al.*, 1989). Vasoconstriction is likely to be the cause of the reduction in Hb, HbO_2 and HbT observed in some subjects in the current study. A reduction in muscle blood flow also occurs when blood vessels are occluded during a strong contraction. Evidence for this can be seen in fig 5.5 when the participant pressed against an unyielding surface eliciting a contraction, and decreased oxygenation was observed.

Similar decreases in oxygenation were sometimes observed during massage, suggesting a decrease in blood volume due to vasoconstriction. This vasoconstriction during massage could have been a response to direct mechanical irritation, causing contraction of smooth muscle in the vasculature. Conversely, an increase in HbT probably reflects vasodilatation of the blood vessels. When both legs were massaged consecutively, the HbT in the first leg continued to increase while the second leg was being massaged, indicating a contralateral effect. This finding is consistent with the results of Cooper and Angus who found that the blood flow in muscles of the non-exercising leg also increased when the blood flow increased in the exercising leg (Cooper & Angus, 2003). The raised starting temperature of the opposite leg to the one being massaged also suggests that the blood flow in the contralateral leg had been affected by massage. Therefore it is important to ensure that participants with CP receive massage on both legs to derive the most benefit from the massage.

In fit young adults there is not normally any measurable change in the voltage registering the levels of cytochrome c oxidase (CtOx) at rest or during exercise (verbal communication, R. Baxendale 2007). In the current study this was the case for unaffected participants. However participant 1 in the group of seven with CP did display marked changes in CtOx in two sessions (see fig 5.13 c and d). The NIRS equipment provides a voltage related to the absorption of near infrared light by the Cu_A ion of cytochrome oxidase. This ion receives electrons from cytochrome c, an electron doner, which are then distributed to the site in the enzyme where oxygen is reduced (Babcock & Wikstrom, 1992). Thus during cellular respiration Cu_A is reduced. Since Cu_A absorbs light only in its oxidised state (Ellwell, 1995), a reduction in voltage indicates an increase in cellular respiration. In fig 5.13d, CtOx decreased after massage began (between 300s and 500s from the start of the record), indicating an increase in cellular respiration. During the same period HbO_2 increased. It is also particularly striking that the Hb gradually fell during massage in this session (fig. 5.13d). This supports the idea that the muscle may have been in oxygen debt before massage began. Since the participant was resting, and had had no more exercise than any others, his frequent involuntary reflex contractions must have been responsible. In healthy individuals oxygen requirement is met by vasodilatation caused by metabolites produced during contractions. However, two observations suggest that there is more intense vasoconstriction at rest in spastic muscles in CP. Firstly, the skin over the muscles was significantly cooler. Secondly after massage in normal individuals, HbT usually remained raised, or increased even further (figs 5.7 and 5.8). In most adolescents with CP and especially participant 1, HbT usually fell (in 8 out of 12 observations), indicating a faster onset of vasoconstriction. It is possible that in CP excessive vasoconstriction may over-ride the vasodilatory effects of metabolites that accumulate during involuntary reflex contractions. This is supported by analysis of fig. 5.13 (d) as discussed above. In CP, spastic muscles could be ischaemic even at ambient temperatures comfortable for unaffected individuals. In cold conditions this vasoconstriction is likely to be even more intense

5.3.2 Comparison of skin temperature changes following massage

In the control group, when the room temperature occasionally fell below 21°C or if the participants were in a draught, skin temperatures could even decrease with massage (fig. 5.2). Those participants most likely reacted to the cold environment by peripheral vasoconstriction in order to preserve the body core temperature. Lightly clad adults lose heat to the environment when the ambient temperature falls below about 21°C (Cross, 1979). In the group with CP, skin temperatures also decreased in some individuals during massage when room temperatures were less than 20°C.

As mentioned above, the skin temperatures before massage in the CP group were approximately 1.5 - 2°C below those of the control group (fig 5.1 and table 5.1). However the temperatures after massage for both groups were essentially the same (fig 5.1 and table 5.1). This is likely to be an important observation. It is possible that in individuals with CP, vasoconstriction to maintain core body temperatures is more intense at higher environmental temperatures. This may be the reason why individuals with spastic diplegia develop thicker layers of subcutaneous adipose tissue on their limbs (Shortland A., P. personal communication, 2004). In future investigations involving the relationship between skin temperature and HbT, it would be interesting to document the effects of massage with room temperatures above 24°C. This would reduce the likelihood of the skin temperature changing predominantly in response to a cooler environment.

Experience in the pilot study to assess the effectiveness of the trainee's massage suggests that caution should be exercised in concluding that increases in temperature are necessarily a measure of the effectiveness of the massage. It is possible that a reduction or curtailed increase in temperature could be due to cool environmental conditions. In addition, even the experienced masseur did not achieve a marked increase in skin temperature with massage in some individuals. Those adolescents in the control group with the greatest increase in HbT after massage (suggesting vasodilatation) had the least increase in skin temperature (fig 5.14). This suggests that vasodilatation removes heat caused by frictional effects during massage.

In the current work, massage increased the temperature and altered the oxygenation of the massaged muscles. After massage the HbT was higher on average in the group with CP. Increased muscle temperature has been shown to have a positive effect in sports performance (Bergh &

Ekblom, 1979). Maximal isometric strength increased by 2% for every degree rise in muscle temperature. Hypoxia has a more limiting effect on muscle performance, in both static and dynamic exercise (Eiken & Tesch, 1984). Thus it is possible that increasing oxygenation with massage can improve metabolic conditions within spastic muscle in the short term.

CHAPTER 6

General Discussion

Most of the current interventions aimed at improving motor function in CP target the compromised neural system, but a review of the literature confirms that they have shown very limited success. However, it has become increasingly clear that the mechanical properties of the spastic muscle are also altered in CP (Deitz *et al*, 1986; Rose, 1994; Ito, 1996; Lin *et al*, 1999; Marbini, 2002; Lieber *et al*, 2004; Mohagheghi, 2007). The altered mechanical properties of the muscle are considered by those authors to be the main reason for muscle spasticity even though the original damage occurred to the developing nervous system. The work forming this thesis investigated the effects of massage on those mechanical properties as well as the effects on motor skills.

In devising an investigation where no directly comparable studies had previously been published, a number of problems had to be addressed. Firstly, the complexity of the way CP manifests itself in each individual. It was decided that adolescents with spastic CP would be the main participants. This made for as homogeneous a population as possible, where further spontaneous improvement in motor function is unlikely. Second, is the difficulty in quantifying spasticity. It was decided that it would be prudent to measure not the spasticity itself, but to assess if massage improved motor function. Third, was the choice of muscle group to receive the massage treatment. The calf muscles provided an ideal group as they were easily accessible, affect ankle joint movement in one plane only, are often shortened in spastic CP, and are crucial to gait during walking. Fourth, is the difficulty of quantifying the massage intervention. A specific, timed sequence of massage was devised where the strokes progressed from superficial and slow to more vigorous and deep. The aim of this was to alter the resting state of the muscle without invoking stretch reflexes, and to provide a reliable, repeatable intervention.

The main aims were to assess the effects of the massage on motor functioning and on the mechanical properties of the spastic muscle, and then to investigate possible mechanisms underlying the improvements observed in motor skills. The format for the series of six studies was devised with each study following on from the previous one, once the results of each were analysed.

Study 1 investigated the effects of calf muscle massage on motor function and on muscle properties in five adolescents with CP. The main findings were;

1). The massage improved motor function in the four ambulant participants (by an average increase of 6.1 GMFM-66 score). It was felt that neural plasticity, which has previously been demonstrated in other participants with CP, could be effective in responding to improved feedback from the changed mechanical properties of the spastic muscle following massage. This is likely to have been the case, given the improved motor function scores.

2). The incidence of stretch reflexes during passive dorsiflexion was almost halved, comparing the first five and last five massage sessions (40% in the first five sessions, 22% in the last five sessions). The specific massage sequence used in the current work had been designed to avoid invoking stretch reflexes during application by stretching the muscle locally and transversely. It was not possible to record EMGs during massage as the electrodes would have had to be placed over the muscles being massaged. However, reflex responses during massage would have been evident in the participant flinching and/or the muscle displaying spasms. The massage had been designed specifically to avoid both of those happening and, in the masseur's experience, this appeared to have been avoided during all sessions. It was also considered that massage applied in this way would, in turn, alter the response of the muscle to subsequent stretch, by altering the resting mechanical properties of the muscle.

3). The calf muscles did not always lengthen consistently with massage. However there was an increase of almost 2º in the average angle through which the ankles were able to move in dorsiflexion after massage. The normal range of movement in dorsiflexion is around 22º, and reduced range of dorsiflexion is almost universal in those with spastic diplegia. Thus, this can be seen as a considerable improvement.

Although the number of participants was small, these results can be considered significant, and compare favourably with other widely used current treatments (see later). The methods were shown to be viable, although time consuming, and were considered a sound basis on which to carry out further investigations.

Study 2 investigated the effects on motor function only on a further six adolescents with CP. Those six also improved their motor function (by an average of 5 in GMFM-66 scores) following a series of massage sessions. The more badly affected participants received more massage sessions as the effects of the massage, not surprisingly, took longer to manifest themselves. (As mentioned in the introduction, it was considered that the massage could possibly be used as a therapy for young people with CP). Constraints in school timetabling meant that the effects on muscle mechanical properties in these six participants were not investigated, although that would have added to the strength of the claim that massage is indeed beneficial for individuals with spastic CP. Given the results for the 11 participants in studies 1 and 2, (an average increase of 5.5 in GMFM-66 scores), the rationale for the use of the specific sequence, timing and application of the massage strokes appeared to be validated.

Study 3 investigated the effects of massage to the full leg of the two most disabled participants who had been recruited. One of these participants had been the only non-ambulant one in study 1, and had shown no measurable improvement in motor function after the series of calf muscle massage carried out 18 months previously. Again the motor function of both of these participants was shown to be improved, this time following a series of full leg massage sessions (5.5 and 6.7 increase in GMFM-66 scores). The same rationale was used to devise the massage sequence as had been used in the calf muscle massage, and the results validate its use.

Thus far all 12 participants had completed the series of massage sessions which resulted in improvements in motor function, by amounts which compare favourably with those of other current treatments. It is uncommon for all of the participants in a study to complete the treatment and to respond to an intervention in such a positive way. The importance of these results should not be understated, although it could be argued that the participant numbers are still small.

Study 4 tested the ability of three adolescents with CP to self administer full leg massage. Only one of these showed significantly improved motor function, (her GMFM-66 score improved by 13.8). One participant had scored very highly in the motor function test prior to participating in this study, thus the margin for his improvement was very much reduced. The third participant was unable to produce a change in the temperature over the massaged muscles, suggesting that she was unable to administer the massage effectively. The participant who did improve her score was the only one of the three who had been able to raise the temperature over the massaged muscles (by around 4ºC). Study 5 showed that effective massage consistently improved the skin temperature over the massaged muscles – see below.

These results suggest that further investigation should be considered with participants who have good manual dexterity and who score in GMFCS levels II and III. The effectiveness of the sequence devised specifically for self massage has shown promising, but not yet proven, results. An obvious benefit would be the saving of physiotherapists' time, as a group of participants could be supervised in self massage, all at once. Additionally, the participants are likely to experience a sense of self-empowerment, and may choose to massage their limbs at times out with the formal massage sessions with their therapist.

As the mechanical properties of spastic muscle are implicated in the limited motor function evident in CP, the investigations then turned to uncovering some of the possible mechanisms underlying the changes observed in the first four studies.

Massage has been shown to increase the temperature of the muscles (Longworth, 1982; Drust, 2003) and increased sports performance has been linked with increased muscle temperature (Bergh & Ekblom, 1979). Study 5 investigated if the specific massage sequence used in earlier studies did indeed change the muscle temperature. This study showed that the temperature over the massaged muscles increased after massage, by around 2ºC in non-disabled participants and by around 4ºC in disabled participants. Interestingly the temperature recorded after massage was statistically the same for both groups. This suggests that, in the participants with CP, less heat is generated by the limb muscles due, not surprisingly, to lower activity levels. As maintenance of body core temperature takes precedence, the heat is directed away from the limbs, resulting in less than optimal limb operating temperatures.

It would have been useful to have assessed for how long after the massage the temperature remained elevated and also if there was a correlation between increased muscle temperatures and improved motor function. Unfortunately neither of those options was possible within the confines of the school curriculum. However the specific massage sequence used here was proven to increase muscle temperature. This may be beneficial to those with CP as their lower limb skin temperature before the massage started appears to be consistently below normal (26.5ºC in those with CP compared to 28.5ºC in non-disabled participants).

The consistency of change in temperature suggested that this could be used to determine the effectiveness of trainee masseurs. One newly qualified physiotherapist with no prior massage training took around 10 sessions to raise the relevant temperature for control subjects to the same level as the qualified massage practitioner. Another physiotherapist already competent in massage was able to raise the temperature by around 4ºC in the very first session. This gives an indication of the length of training needed by other physiotherapists who may wish to include the massage as part of their input for those with CP.

Study 6 investigated the effects of massage on oxygenation of the massaged tissue. A group of 6 adolescents with CP and an age and gender matched control group were compared. The main observation here was that the total haemoglobin (HbT) was higher on average after massage in the group with CP. This suggests that the blood supply is regularly sub-optimal in the lower limbs of those with spastic CP. Hypoxia, not surprisingly, has been shown to limit muscle performance. It can reasonably be concluded that the massage at least temporarily improves metabolic conditions within spastic muscle in those with CP.

Additionally, significant changes in oxygenation were observed during the massage, as each stroke was applied. The massage strokes differ from one another in the way they squeeze and stretch the muscle and this was reflected in the changes in oxygenation. The last three strokes regularly produced an increase in oxygenation following on from a general slight decrease during the first four strokes. It could be argued that application of only the last three strokes would reduce the length of time the massage need be applied. However, it is the author's experience that those first four strokes are needed to gently warm and stretch the muscle in preparation for the more vigorous last three strokes. If only the last three strokes were done, it is likely that their depth and vigour would result in the muscle reflexly contracting. This can be felt as spasm of the muscle, often accompanied by flinching from the patient, which was avoided during these massage sessions. This adds to the justification for the use of the specific massage sequence used here.

Another important point is that these results were obtained with only the lower half of the calf muscles being massaged because the NIRS electrodes and blackout sleeve covered the top half of the calf. It is not unreasonable to assume that massage of the whole calf is likely to be more beneficial. This could be confirmed by massaging the whole calf and recording from the leg immediately above the popliteal fossa. This would give another indication of whether the massage affects the circulation out with the muscles being directly massaged. The massage had already shown an effect on the circulation out with the massaged muscles, as the starting temperature of the second leg to be massaged was consistently higher than that of the first leg.

It would have been useful to be able to record the length of time after massage that the oxygenation levels remained elevated. However, working within the confines of the school timetable precluded that option.

Taken together, the effects on both the temperature and oxygenation of the massaged muscles, add to our understanding of possible mechanisms underlying the effects of the massage in improving motor function for those with spastic CP in these studies.

6.1 Methods – general conclusions and observations

As mentioned in the introduction, it is common in this type of study to include a control against which the intervention is measured. However no two participants with CP are affected to the same extent by their condition. Thus the comparison of one group who have CP against another with CP was not considered to be a viable option. One option was that the participants could act as their own controls by comparing the effect of massage against the effect of relaxation alone. This option was not implemented, as the author felt that it was unethical to subject the participants to an intervention (relaxation alone) which was unlikely to prove beneficial, whilst reducing their school lesson time. A larger study where one group of adolescents with CP are compared to another group with CP is likely to be considered more ‘scientific’ than the methods used here. This should be considered in future work.

One main aim of this series of studies was to assess if the participants could *make use* of any changes following massage. Consequently a comparison was made in the participants with CP where their own motor function scores were compared before and after the massage intervention. As the mechanical properties of spastic muscle are affected in CP, and the massage appeared to alter the resting state of the muscles, the hypothesis was validated that massage might aid motor function by altering the mechanical properties of the muscle.

It is possible that the very act of stretching the muscle produced the response seen after massage. However, if that were the case, presumably physiotherapists would have had the same measure of success by simply employing a series of stretches in their regimen for young people with spastic CP. As the participants with CP all continued to have regular sessions with their physiotherapists throughout the time of the studies, it is unlikely that the stretching alone would have been responsible for the changes observed after massage.

Simple methods of testing were employed which could be accommodated within the school timetable. For example the muscles were manually stretched by using hand pressure on a dynamometer and following a template on a computer screen. It was not always possible to do this with exact accuracy. However a general linear statistical model showed that there was no significant difference over all in the rate and amount of pressure applied before and after the massage. More sophisticated methods are suggested to test the muscle mechanical properties such as those used by Sinkjaer and Magnussen (Sinkjaer & Magnussen, 1994). This could be used to confirm results from the current work, and also to assess if the muscle stretches equally over its whole length. It is suggested that the massage resulted in altered feedback to the CNS resulting in the recruitment of more motoneurones by descending command and this could be investigated further. It is possible that a shorter massage sequence could be equally beneficial, especially with younger children. Additionally, it is not yet known what the optimal frequency is for application of the massage.

The physiotherapist who conducted the GMFM tests was aware that the participants had received the massage intervention. However, because the GMFM manual is very specific about how participants should attempt the task, this was not considered to be detrimental. For example, descending stairs in a specific way is unlikely to be a false result. As mentioned previously, the adolescents did become fatigued during the testing as their cardiovascular fitness is tested as well as their motor skills. This could be avoided if the testing were not done all in one session.

It is recommended that the room temperature be above 21^{o}C. However, the chill factor could also affect the skin temperature, as was noted during the first session at Hutchesons' School when there was a considerable draught in the room. It was possible to use all of the equipment in two secondary schools, although the NIRS equipment was very heavy. This was preferable to bringing the adolescents to the University, given their curricular restraints.

The orientation of the optodes also has to be addressed as orientation along the length of the muscle proved more sensitive (3 participants only). Transverse orientation was used to expose as much of the muscles as possible for massage. In fact changes in oxygenation were significant using orientation in both directions. More recordings are needed for adolescents with CP using the orientation along the length of the muscle. This is because of the particularly interesting records from participant 1 which suggested that oxygenation might sometimes be suboptimal in spasticity.

From the results two new ideas are proposed. Firstly, sensory feedback from spastic muscles in CP is inappropriate; in effect the CNS is misled by it. This is because the mechanical properties of the spastic muscle are abnormal and thixotropic effects are more pronounced. The massage sequence that was tested acts by changing the mechanical properties. Motor control centres in the CNS respond to the changed feedback and motor control improves, notably the control of interneurones concerned with reciprocal inhibition. Gains in motor skills are potentially long term owing to the plasticity of neural function (fig 6.1). Second, there are abnormalities in the control of the blood supply to spastic muscles in CP.

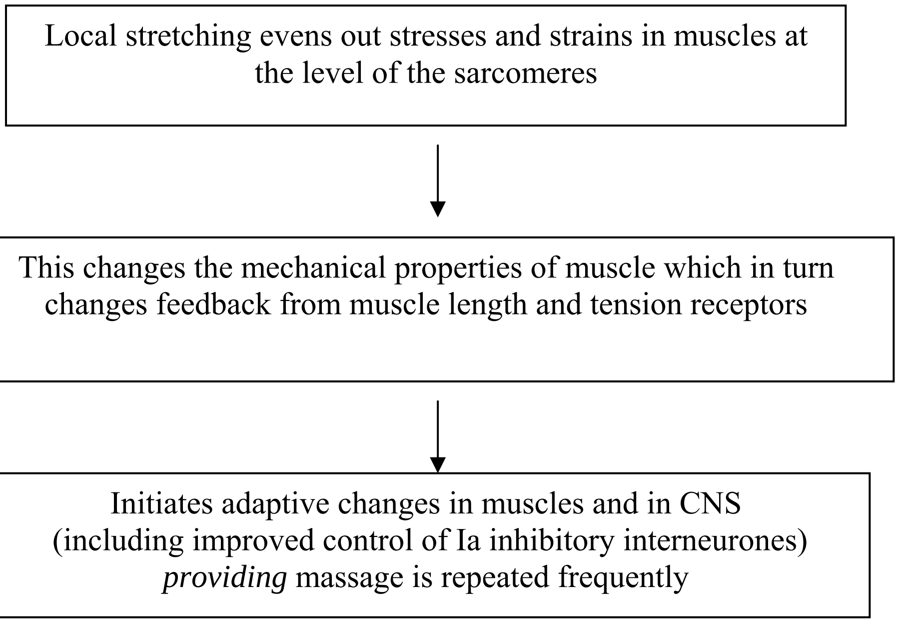


Fig 6.1 Proposed effects of massage

From the results observed in the current studies, the figure summarises the possible mechanisms by which the massage may have its effect. Massage alters the mechanical properties of the muscles at sarcomere level. This, in turn changes the resting state and the feedback from the muscle to the CNS. The plasticity, which has been demonstrated by the CNS in others with CP, accommodates the new neural signalling, which leads to improved motor function.

6.2 Supporting evidence for hypothesis concerning sensory feedback

Foran and colleagues suggest that it is reasonable to study skeletal muscle properties in disorders of neural origin because muscles respond in a fairly stereotypical manner to the amount and type of activity imposed on them (Foran *et al.*, 2005).

1). It was found that the massage affected the mechanical properties of the spastic muscles. Immediately after massage:

a) the muscle stiffness was increased (fig 3.2)

b) the length of the muscles sometimes changed (fig 3.3)

The mechanical properties of spastic muscles in CP are known to be abnormal. Although the muscle cells themselves are stiffer, spastic muscle has a higher proportion of extracellular matrix, which itself has less resistance to stretch compared with normal muscle (Lieber *et al.*, 2003a). It is argued that the changes in mechanical properties following massage can be explained by thixotropic effects, following the ideas of Proske and Morgan (see Chapter 3). 'Thixotropy' means that the mechanical behaviour of the muscle is changed by its immediate history and this is more pronounced in spastic muscle (Walsh, 1992).

2). There is no direct evidence from the present study that the massage sequence changes the sensory feedback from the muscles. On the other hand:

a). Kratch and co-workers found that children with spastic diplegia scored significantly worse than normal when tested for position and vibration sense. Muscle spindle Ia afferents make a sensory contribution to both these tests (Krach *et al.*, 2005).

b). Thixotropic effects can alter monosynaptic and H reflexes in healthy adults (Gregory *et al.*, 1990; Proske & Morgan, 1999).

3). Indirect evidence that the massage sequence changes the sensory feedback can be deduced from the notable gains in the ability to descend stairs in 5 out of 7 participants. This could imply that they had become more adept at inhibiting motoneurones during lengthening contractions of their soleus muscles, which is essential for this activity. It is argued (Chapter 4) that descending stairs entails control of reciprocal inhibition. In addition there was evidence from EMG recordings from soleus muscles that inhibition of soleus motoneurones during soleus lengthening developed in two participants over the course of a few weeks of regular massage sessions (fig 3.6). Interestingly, Leonard and colleagues found that whereas in healthy individuals, increasing levels of contraction of tibialis anterior muscle led to increasing inhibition of soleus motoneurones, this did not occur in their participants with spastic diplegic CP (Leonard *et al.*, 2006). Additionally Nardone and co-workers found evidence of disinhibition in group II afferent pathways in hemiparetic patients following a stroke (Nardone *et al.*, 2005). Consequently, it is not possible to determine whether the altered reflex responses in the current work can be attributed to changes in either group 1a or II afferents or to both.

4). Although other authors have not claimed to change the feedback from spastic muscles, there are several reports demonstrating that individuals with CP are better able to control reflexes and movements if given visual feedback:

a). When given visual feedback, children with CP reduced the gain of their stretch reflex by 50% on average after 20 weeks training; adults are also capable of this (O'Dwyer *et al.*, 1994). This demonstrates that the participants were making use of abilities that had previously not been exploited.

b). Feedback was given to a 13 year old boy with CP using a split screen to model what the movement should be like, in addition to verbal clues. This was shown to promote motor learning (Larson & Surber-Berro, 2006).

5). In the present study neuroplastic changes associated with enhanced motor skills are suggested from the largely maintained GMFM-66 scores of participants many weeks after receiving massage (see chapter 4).

a). Direct evidence of neuroplastic changes initiated by visual feedback, was reported by Young in an 8-year old boy with hemiparetic CP. Following virtual reality therapy to train elbow movements, activation of primary supplementary cortices (SMC) bilaterally together with the ipsilateral supplementary motor area was reduced to activation of the contralateral SMC during movements (Young, 1994).

b) In the lower limb, Phillips and colleagues found evidence of neural plasticity underlying improvements in motor function in individuals with CP (Phillips *et al.*, 2007). In a body-weight-bearing treadmill exercise, three children with CP showed improvements in walking. Increased cortical activation was seen in functional MRI scans, illustrating that altered feedback had been accommodated in the CNS (fig.6.2).

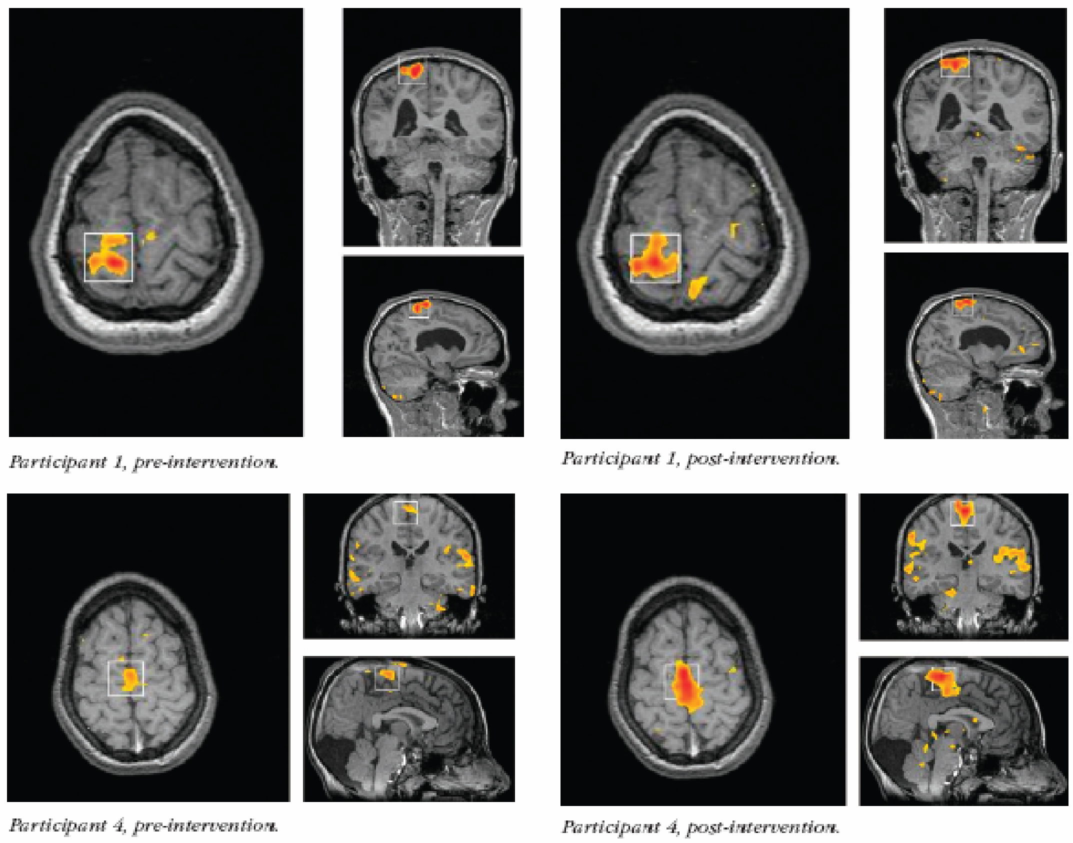


Fig 6.2 Functional MRI scans during ankle dorsiflexion.

fMRI scans from two participants (1&4) with CP who took part in weight bearing treadmill exercises. After the intervention in both cases there is increased activation in the area of cortex related to ankle dorsiflexion. This demonstrates neural plasticity in participants who have the same clinical condition as those in the current studies. Note; only these two participants had clear scans although three subjects in that work showed improvements in walking. (From Phillips *et al.*, 2007).

c). It should be noted that the area of cerebral cortex devoted to a particular muscle depends on its use in healthy individuals. When a limb is immobilised in adults with normal control, the area of motor cortex concerned with the muscles in the limb is reduced; these changes are reversible (Liepert *et al.*, 1995).

The capacity of the CNS to adapt and accommodate is pivotal to the hypothesis that improved motor control is possible if the properties of the muscle have been altered following the intervention used in the current study. In the cerebral palsies, the child has in most cases known no other motor state and has never experienced the loss of previously acquired function. However, the processes necessary for the initial development of motor skills appear to be still available long after brain damage has occurred (Lin, 2003).

Thus far only input from Ia afferents, and Ia reciprocal inhibition have been proposed as mechanisms that the massage sequence may have targeted. However, Nardone and co-workers found evidence of disinhibition in group II afferent pathways in hemiparetic patients following a stroke (Nardone *et al*., 2005). Additionally, Leonard and colleagues found no evidence of modulation of short or long latency (presynaptic) inhibition of soleus motoneurones during contraction of tibialis anterior in individuals with spastic diplegic CP (Leonard *et al*., 2006). The current work has not produced evidence as to whether massage affects these pathways, although it seems likely.

Additionally, the current work did not take account of the effects of massage on the mechano/nociceptor neurones. The majority of afferent nerve fibres in mammalian muscle are thin myelinated (A delta or group III) and unmyelinated (C or group IV) (Kniffeki *et al*., 1981). Given the vigour of the last three massage strokes and the magnitude of the changes in oxygenation and temperature observed during and after massage, it is highly likely that group III and/or IV afferents were stimulated, although again direct evidence of this was not sought during the current series of studies. Further investigation might elucidate this as another mechanism by which the massage has its effects.

6.3 Supporting evidence for hypothesis concerning abnormal control of muscle blood supply in CP

The second hypothesis is that blood supply in CP is potentially more attenuated than normal in a cool environment. Skin temperatures over the calf muscles before massage in the participants with CP were consistently below those of the unaffected individuals (by 1.5-2°C). However, after massage there was a significant difference in skin temperatures compared with healthy individuals, provided room temperatures were above 21°C, and draughts were excluded. This is because maintenance of body core temperature takes precedence, and blood is diverted away from the periphery.

Phasic stretch reflexes which are involuntary still require oxygen. The muscles of individuals with spastic diplegia may be in chronic oxygen debt in a cool environment. This may be an additional reason why muscles in those with CP do not function optimally. In the current studies vasodilatation usually occurred in participants with CP during massage, but often muscle blood flow decreased significantly after massage. The subcutaneous adipose layer in young people with CP is thicker than non-affected people (personal communication, Shortland, 2004). This may be a compensatory adaptation for excessive vasoconstriction. It is not clear at present how these ideas tie in with current thinking on the contribution of local metabolites on blood vessel diameter during exercise, for example.

Interestingly, Jahnsen and colleagues found that chronic musculo-skeletal pain is a pronounced problem reported by adults with CP in Norway (Jahnsen *et al.*, 2004). Young men in level III of the GMFCS had the most pain. The musculo-skeletal pain was worse in cold conditions, and besides analgesics the pain was relieved by massage, hot water, exercise, and living in a warm climate.

6.4 Comparison with directly related work

Previous research on the effects of massage on spastic muscles in CP has been very limited. In the only recorded controlled trial of the effects of massage on children with CP (Hernandez-Reif *et al.,* 2005), the mean age of the 20 participants was 31 months and the massage was carried out by an unspecified number of trained volunteer massage therapists. 80% of the children had spastic CP, although it is not clear how many of those have diplegia. In that study, it is possible that some of the improvement may have occurred spontaneously with age, as motor skills in children with CP have been shown to plateau around the age of seven (Rosenbaum *et al.,* 2002). The main reason our studies involve adolescent participants is that spontaneous improvement is unlikely at that stage of their lives. The variables in the current work were reduced by having only one experienced massage therapist treat spastic diplegic participants (100% of the disabled participants), using a timed, easily replicable, massage sequence. Additionally, the effectiveness of this work can be compared directly with a number of current interventions, because the same main assessment measure (GMFM-66) is used.

6.5 Comparison with other interventions

GMFM-66 has been used as the outcome measure in the current study because it has been devised specifically for use with individuals who have CP. The predecessor to this was GMFM-88 which has been used in many other studies to assess the effects of interventions in CP. GMFM-66 was developed by Russell and colleagues by applying Rasch analysis to GMFM-88, and has several advantages over GMFM-88 (Russell *et al.*, 2000). The GMFM-88 score cannot be calculated unless all 88 items are tested, whereas GMFM-66 scoring takes account of the difficulty of each item, improvements in scores are comparable at all levels, and all items need not be tested. In addition to physical therapy, selective dorsal rhizotomy (SDR) and botulinum toxin (BTX-A) are regularly used to treat CP. Table 6.1 shows that the massage sequence tested in the present study is more effective than SDR in improving motor skills.

Study	Intervention	n	Outcome interval (months)	Age (years)	GMFCS level	GMFM difference
(Steinbok *et al.* 1997)	SDR	28	9	3-6.5	II-IV	0.2*
(McLaughlin *et al.*, 2002)	SDR	38	24	3.3-18	I-IV	6.1*
(Wright *et al.*, 1998)	SDR	24	12	3.5-7.6	I-IV	7.7*
(MacGregor *et al.*, 2007)	massage	12	1.5-24	10-18	I-IV	5.8 ± 0.89 (SEM)

Table 6.1 Comparison of massage with selective dorsal rhizotomy

The table compares the effects of the massage intervention with results from three selective dorsal rhyzotomy (SDR) studies and one published by the author of this thesis. The massage intervention compares favourably using the GMFM-66. Note that the ages of those in the SDR studies ranged from a much younger age and consequently, there could have been spontaneous improvements with age in those studies.

* GMFM-88 scoring; for pooled data, change in score using GMFM-66 as outcome measure was 2.66 ± 0.82 SEM. Studies of the effects of SDR compared physiotherapy only and SDR along with physiotherapy.

It is not possible to make a direct comparison of effects of massage with BTX-A therapy as no published data used GMFM-66 as an outcome measure, and the scores for GMFM-66 and GMFM-88 are not comparable. Most recently Mall and co- workers used the Goal Attainment Score (Palisano *et al.*, 1992) rather than GMFM-88 (Palisano *et al.*, 1997) as the outcome measure for treating adductor spasticity with BTX-A, commenting that 'the GMFM failed to detect the superiority of BTX-A treatment' (Mall *et al.*, 2006).

Kratch and colleagues reported significant improvements in mean GMFM scores with continuous infusion of baclofen intrathecally, both in subjects <8 years (mean change 4.1) and in subjects 8-18 years old (mean change 3.7) (Krach *et al.*, 2005). Meythaler also reported improvements in GMFM-66 scores in 7 patients who had continuous intrathecal infusions of baclofen (Meythaler, 2007). Their mean pretreatment score was 49.9 ± 7.3; mean post treatment score was 54.5 ± 7.7, a mean increase of 4.4.

6.6 Future work

Although CP originates from neural damage, the results from the current studies underline the contention that there is an altered muscle component in spasticity, which needs to be addressed if improved motor function is to be expected. Although some form of massage is sometimes used by physiotherapists, the beneficial effects of massage have not been widely documented before, possibly because they did not use the sequence deployed in the present work. This may be the reason why massage has not yet been universally accepted for use in CP treatment.

Massage originally formed a core part of Conductive Education (CE) in its country of origin (Hungary). However it is not included in the CE practised in the UK, possibly because its efficacy had not been confirmed.

Hopefully, this practice may be reviewed in the light of the results found in the current work.

This study has demonstrated that significant improvements of motor skills can be achieved with massage in spastic diplegia that are at least comparable with outcomes of any current interventions. Next it will be important to confirm that professionals and carers can deliver the same massage sequence effectively. It was found that initially the massage of a qualified physiotherapist with no massage training was not as effective as that given by an experienced therapist. The ability to produce a temperature change in the skin over the massaged muscles could be used to measure the effectiveness of other trainees' massage. Parents could be trained to give this massage effectively. It would be very interesting to discover whether even greater improvements in motor skills could be achieved if massage were delivered more regularly and from an earlier age.

It is proposed that this massage sequence should be incorporated into the physiotherapy input for young people with spastic diplegia. It is appreciated that physiotherapists are concerned about the development of deformities in CP. As physiotherapists' time is limited, it is advocated that the existing stretch programmes be replaced with this massage sequence, followed by mobilisation of the joint(s) within the range of movement. This is more likely to avoid stretch reflexes being incurred, whilst making use of the newly altered muscle properties. Voluntary movements should also be encouraged following massage to make best use of the thixotropic changes in the muscle brought about by the massage.

6.7 Final observations and conclusions

The over all purpose of this thesis was to investigate if massage treatment might be beneficial for those with cerebral palsy. This had seemed a likely possibility following empirical results obtained by the author. The specific aims of this thesis were to investigate the effects of a particular sequence of massage strokes on motor function, on the mechanical properties of spastic muscle, and on possible mechanisms underlying changes, in those with CP. The results from the six studies suggest that those aims were achieved to a large extent. The numbers of participants and sessions could be considered small. However the changes were often statistically significant. All of the participants completed the series of massage sessions and all of those with CP improved their motor function. The author considers that these studies have made initial inroads into understanding the effects of massage on those with CP and to the possibility of massage being adopted into the physiotherapy input for that client group.

APPENDIX

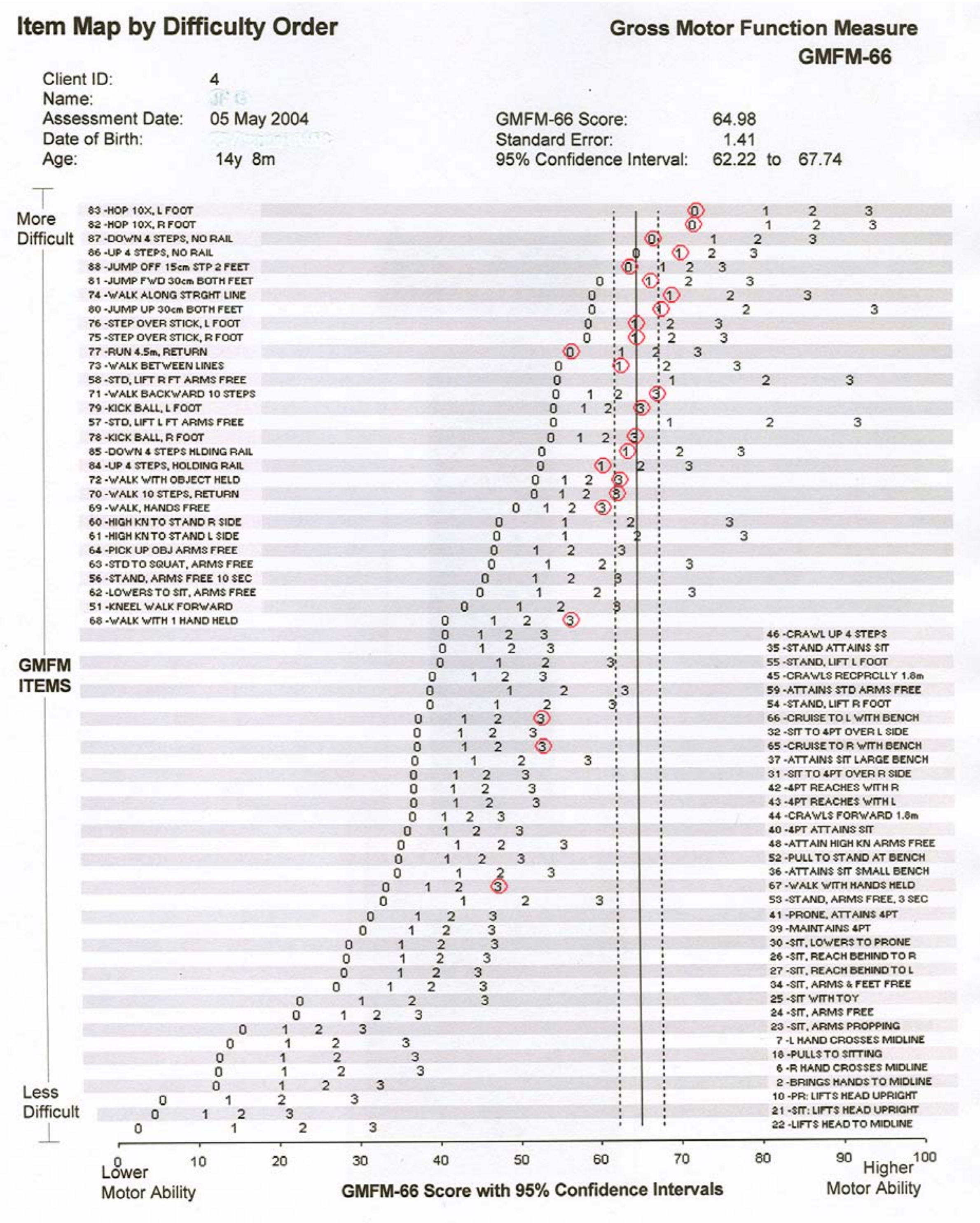


Fig A1 Example of GMFM-66 score sheet before massage sessions

See fig A2 for legend.

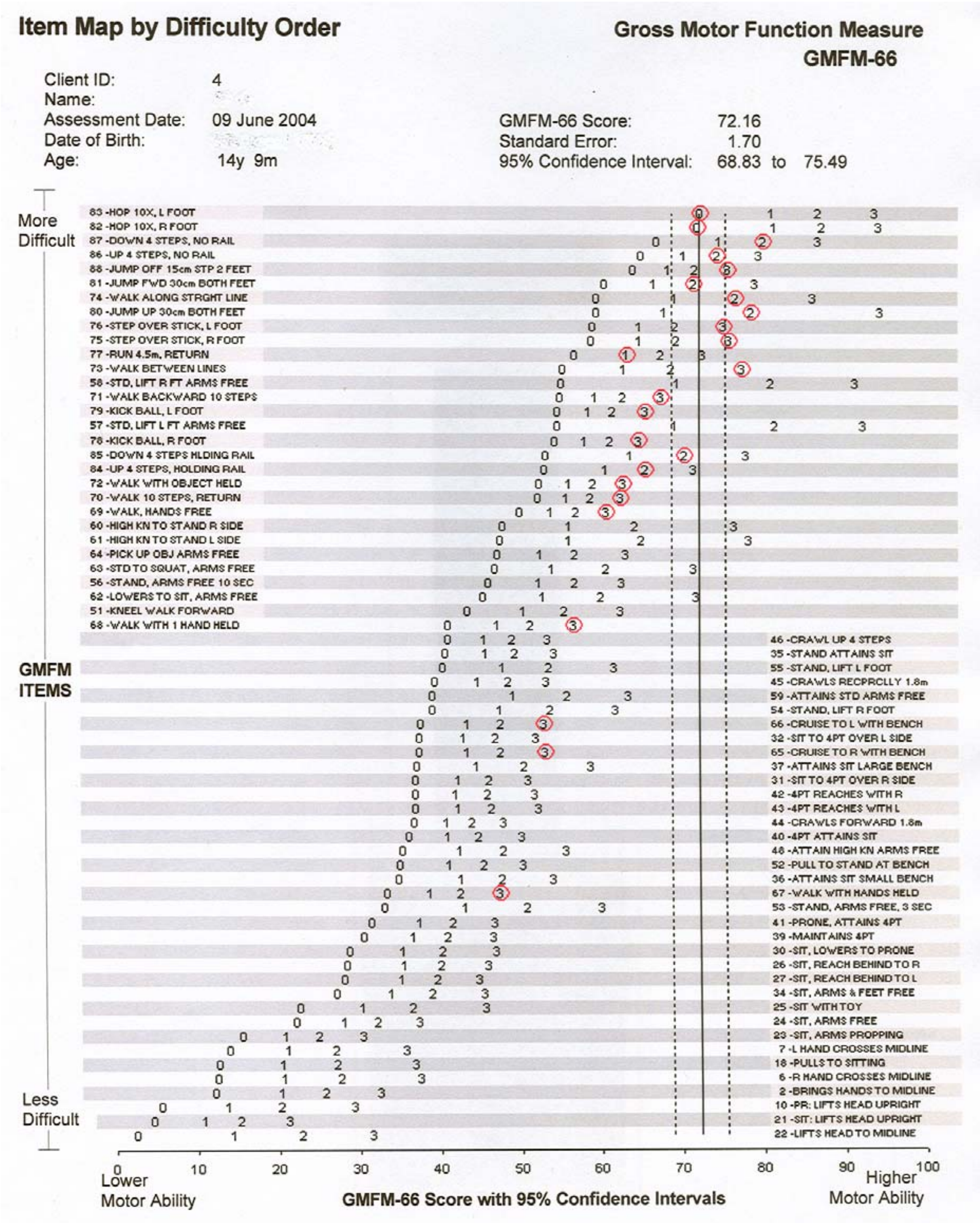


Fig A2 Example of GMFM-66 score sheet after massage sessions.

Figs A1 and A2 illustrate the GMFM-66 scores for one participant. The items tested are circled in red. The vertical lines indicate the score, with the broken vertical lines either side showing the confidence intervals (CIs). In this case the increase in score is significant as lower CI of the second test is clear of the upper CI of the first test. The GMFM-66 allows for a score to be calculated even though not all of the 66 items are tested.

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