Intensive somatosensory stimulation to improve upper limb recovery and

reduce unilateral neglect after stroke

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Abstract

Case: 64-year old male with right anterior stroke; increased tone and typical

posturing of left upper limb (UL); hyperreflexia and resistance to passive movement.

No voluntary or functional movement in limb apart from slight shoulder girdle

elevation. Shoulder joint subluxed, painful on passive movement. Head rotated and

attention biased to the right; unilateral neglect behaviour apparent.

Method

N=1 study (A-B-A); mobilisation and tactile stimulation (MTS) delivered (B phase)

daily for 6 weeks, involving manual manipulations to mobilise, sensitise and retrain

selective movement. Action Research Arm Test (ARAT), Motricity Index arm section

(MI), and Semmes-Weinstein Monofilaments (SWM) tested daily. Data analysed

using visual analysis.

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Findings

Initial A phase: ARAT scores consistently zero, MI scores stable, SWM scores absent. Following one MTS treatment session (B phase), immediate improvement noted in MI and SWM scores, and unilateral neglect behaviour.

Conclusion

The potential for MTS to reduce unilateral neglect warrants further study.

Key words: upper limb; stroke; mobilisation and tactile stimulation; unilateral neglect; sensory stimulation

Introduction

Stroke affects more than 100,000 people each year in the UK (The Stroke Association, 2017). Conventional rehabilitation after stroke should be provided by a specialist multidisciplinary team working in partnership with stroke survivors and their families, to identify impairments and disabilities, and to inform and direct assessment and treatment (National Institute for Health and Care Excellence, 2013; Intercollegiate Stroke Working Party, 2016). A 24-hour approach to stroke rehabilitation is encouraged (Aries and Hunter, 2014) to maximise the potential for recovery and restoration of movement and function.

Unilateral neglect

Unilateral neglect (UN) is a disorder of perception that can affect between 10–82% of stroke survivors (Plummer et al., 2003), particularly following a right cerebral hemisphere lesion. UN has been reported to be a predictor of long-term activity

limitation and poor rehabilitation outcome (Jehkonen et al., 2006). Typical features of UN include an unawareness of the side of space opposite to the side of the stroke lesion in the brain, and failure to respond to a stimulus from the neglected side (Yang et al.,2013). People with UN often behave as if half of the world (the neglected side) no longer exists, for example bumping into objects on the neglected side, reading only one side of a page in a book, eating from only one side of the plate.

Treatment for unilateral neglect

The national clinical guideline for stroke recommends various strategies and interventions for people with neglect (Figure 1). These include the use of limb activation and sensory stimulation to raise awareness of the neglected side (Intercollegiate Stroke Working Party, 2016). Visual and auditory stimulation from the environment provide sensory cues, for example, by approaching and talking to the stroke survivor from the neglected side. All members of the multidisciplinary team are encouraged to do this to raise awareness of the neglected side. Somatosensory, proprioceptive and kinaesthetic stimulation provide information about the position and movements of the body, which contributes to 'body schema' and an integrated neural representation of the body (Holmes and Spence, 2004). This sensory information arises from feedback about movement which, after stroke, is usually the domain of the physiotherapist. However, other members of the multidisciplinary team, particularly nurses who are with stroke survivors throughout the 24 hours of the day, could potentially have a role in delivering aspects of this type of sensory stimulation.

Figure 1: Strategies for people with unilateral neglect (Intercollegiate Stroke Working Party, 2016: p64)

People with unilateral neglect should:

- "have the impairment explained to them, their family/carers and the multidisciplinary team
- be trained in compensatory strategies to reduce the impact on their activities
- be given cues to draw attention to the affected side during therapy and nursing activities
- be monitored to ensure that they do not eat too little through missing food on one side of the plate
- be offered interventions aimed at reducing the functional impact of the reduced awareness"

Mobilization and tactile stimulation

Mobilization and tactile stimulation (MTS) is a physical therapy intervention that provides somatosensory, proprioceptive, and kinaesthetic stimulation and feedback (Hunter et al., 2006). Described as a discrete 'module' or unit of physical therapy, MTS for the upper limb includes various routine physical therapy techniques that are provided in combination to the hand and forearm after stroke (Hunter et al., 2006). These include manual joint manipulation and soft tissue mobilization techniques, sensory stimulation (specifically touch, pressure and proprioception), and limb reactivation. Sensory receptors (cutaneous mechanoreceptors) in the glabrous (nonhairy) skin of the hand are stimulated in response to mechanical deformation from touch, stretch and compression. This provides proprioceptive feedback to the brain, supplementing that from receptors in joint capsules, ligaments, muscles and tendons in response to the mobilization techniques. In quasi-experimental replicated single

system (n=1) experimental studies, using A-B-A design, an intensive dose of MTS has been shown to improve sensorimotor function in the upper limb after stroke (Hunter et al., 2008; Winter et al., 2013).

Single system (n=1) experimental studies

In this type of study, the first A phase acts as the baseline or control for comparison with subsequent phases. The experimental intervention is delivered during the B phase, and subsequently withdrawn in the second A phase. The key features of this design are summarised in Figure 2.

Figure 2: Key features of A-B-A design

- First A phase = period before the intervention under investigation is delivered. Considered to be the 'control' phase in which behaviour or performance should be stable, often referred to as the baseline phase
- B phase = period during which an intervention is introduced, and the behaviour or task performance during this phase can be compared with that in the baseline (A) phase. Referred to as the intervention phase
- Second A phase = period after which the experimental intervention is withdrawn. Known as the withdrawal phase
- Outcome measures are recorded at regular intervals throughout all three phases.

Data are primarily analysed using visual analysis of charted data to identify observable changes in behaviour or task performance between phases. More specifically, changes in the level, trend and slope of the plotted data, particularly at

the point of transition from one phase to the next, are of interest. Individual cases (n=1) are studied, rather than a group of participants. Evidence of causal effect can be shown if the results are replicated in at least four or more cases (Barlow and Hersen, 1984). The benefit of this design is that individual responses to an intervention can be examined in detail, often providing unique insights into responses to treatment that might otherwise be unseen in a group study. The use of n=1 studies is considered to be particularly useful in the modelling and early evaluation of therapy (Craig et al., 2008), and in evaluating complex behaviour (Barlow and Hersen, 1973), such as in stroke.

Mobilization and tactile stimulation for unilateral neglect

In addition to MTS being an effective intervention to improve UL sensorimotor function after stroke, therapists have also reported that MTS is an intervention used in rehabilitation of UN (Hunter et al., 2006). Yet, the effects of MTS on UN have not been evaluated. However, in the study of Hunter et al. (2008), one participant demonstrated significant observable UN behaviour in conjunction with severe UL dysfunction during the baseline phase. It was noteworthy that, after only one treatment of MTS, both his observable UN behaviour and UL muscle strength improved significantly. The purpose of this paper is to report this single case and his response to MTS, and to discuss multidisciplinary treatment implications and opportunities for patients presenting in this way.

The case study report

This case study report is of a 64-year old male, referred to as 'Derek', who had been admitted with dense left sided stroke (right brain lesion) as a result of a large right middle cerebral artery infarct and evidence of acute haemorrhage. He was diagnosed on admission with Total Anterior Circulation Infarct (TACI) (Bamford et al., 1991), and

had a Barthel score of 1. On recruitment to the study, at 61 days post-stroke, he presented with increased tone, hyperreflexia and resistance to passive movement in his left UL, which was held in a typical pattern of flexor synergy. He had no voluntary selective movement in his left arm apart from slight shoulder girdle shrug, and no UL functional activity. He reported pain on passive movement, and his shoulder joint was subluxed. His head was rotated to the right when he was at rest, with his attention biased to the right side of space. Clinical assessment concluded that he had UN with observable associated neglect behaviour.

Derek was one of six stroke survivors who received MTS for up to one hour per day, five days per week, for six weeks (Hunter et al., 2008). MTS was delivered by an experienced and skilled physiotherapist. Outcomes of UL function and activity capacity, motor impairment, and sensation were measured using the Action Research Arm Test (ARAT), the Motricity Index (MI) arm section, and the Semmes-Weinstein Monofilaments (SWM) respectively. The combination of both sensory and motor outcomes was selected because both are important for UL function. Unilateral neglect behaviour was assessed and monitored clinically by observation of behaviour during activities of daily living and communication throughout the day.

Action Research Arm Test (ARAT)

The ARAT is a test of functional capacity in the UL. Divided into four sections, it tests grasp, grip, pinch and gross movement, with each section scored separately (Lyle, 1981). It is performed from seated position at a dining table. The maximum total score is 57, and the minimum clinically important difference (MCID) is 12 points for the contralesional dominant UL, and 17 points for the contralesional non-dominant UL (Lang et al., 2008). The ARAT tasks are summarised in Figure 3.

Figure 3: Summary of the Action Research Arm Test

Section 1: Grasp (maximum score = 18)

Reach to pick up and place on a shelf (37cm height above the table and 37 cm away from the table edge) a range of different sized objects: wooden cubes, cricket ball, sharpening stone

Section 2: Grip (maximum score = 9)

Pour water from glass to glass, pick up two different sixed metal tubes, place a washer over a bolt

Section 3: Pinch (maximum score = 18)

Pick up and place on the shelf marbles and ball bearings using just thumb and individual fingers

Section 4: Gross movement (maximum score = 12)

Take the hand up to the mouth, or place it on the top of the head, and behind the head

Motricity Index (MI) - arm section

The MI (arm section) is a valid and reliable measure of muscle strength in the hand (pinch grip), elbow (biceps muscle) and shoulder (deltoid muscle) with a maximum score of 33 for each (Wade 1992). The scoring system for the MI is summarised in Table 1.

Table 1: Summary of the Motricity Index (upper limb section) scoring

Criteria	Score
Pinch	
No movement	0
Palpable contraction of muscle but no movement	11
Movement seen but not full range or unable to hold against gravity	19
Movement full range against gravity but not against additional	22
resistance (force applied by the rater in the opposite direction)	
Movement against resistance but weaker than the other side	26
Normal power	33
Elbow and shoulder	
No movement	0
Palpable contraction of muscle but no movement	9
Movement seen but not full range or unable to hold against gravity	14
Movement full range against gravity but not against additional	19
resistance (force applied by the rater in the opposite direction)	
Movement against resistance but weaker than the other side	25
Normal power	33

Semmes-Weinstein monofilaments (SWM)

The SWM are a set of 20 nylon filaments or rods calibrated to exert a pre-set force when applied perpendicular to the skin surface (Bell-Kotroski & Tomancik, 1987). Forces range from 0.008g to 300g, and for this study each filament was numbered according to the order in which they appeared in the full set, from 1–20. Filament number 1 represents the smallest force (0.008g), and filament number 20 represents the highest force (300g). The test involves determining the lowest pressure threshold at which the sensation is accurately perceived. Consequently, the larger the filament

number, the less able is the participant to register the sensation of touch-pressure.

Where there is no sensation at all, even to a force of 300g, there is no score.

Key events noted during the study

Table 2 summarises Derek's key feelings and events during the study.

Table 2: Key events occurring during the study according to day of occurrence

Day of study	Event
1	Start of A phase.
	Feeling tired and lethargic; unable to attend to the sensory tests
2	More alert but still very tired
7	Feeling unwell
8	Feeling unwell
9	Feeling better
18	Start of B phase – MTS intervention introduced
21	Reported pain in contralesional forearm
22	No report of pain
28	Reported pain in contralesional forearm
29	No report of pain
30 & 31	Christmas Day and Boxing Day Bank Holidays – no therapy
36	Overnight stay at home
37 & 38	New Year's Eve and New Year's Day Bank Holiday – no therapy
44	Feeling very distracted by removal of urinary catheter
45	Feeling very distracted by removal of urinary catheter
46	Re-catheterised and feeling more relaxed and able to attend to tests
60	End of B phase; MTS treatment withdrawn.
73	End of withdrawal phase and end of study

Tolerance of the intervention and outcome measures

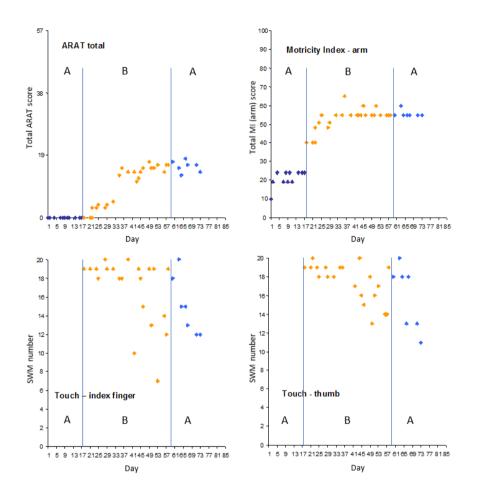
During the B phase, Derek received MTS on 24 out of a possible 30 days of intervention. Therapy was not provided on four Bank Holidays over Christmas and New Year, and on two days when he was engaged in other routine rehabilitation activities, such as home assessment visits. The average (mean and standard deviation (SD)) length of MTS treatment sessions was 48.5 (SD 11.75) minutes (range 30–60 minutes). He reported pain in his forearm on days 21 and 28 but this was not exacerbated by MTS, and on each of those days he tolerated 45 and 55 minutes of MTS, respectively, quite comfortably

Response to the MTS intervention

Derek's reports of forearm pain were not considered to be directly linked to the MTS treatment which he tolerated well. Like 60-74% of stroke survivors reported to experienced sensory loss in the arm and hand after stroke (Carey, 1995), Derek had experienced disruption to sensory pathways. This disruption can result in altered perceptions of sensation (Carey, 1995). Functional manifestations of disrupted sensation can include uncertainty of response to sensory stimulation, hyper- and hyposensitivity, and recovering sensation can be perceived as pain (Carey, 1995).

Figure 4 shows scatterplots of Derek's performance on all outcome measures through each phase.

Figure 4: Scatterplot showing scores for Action Research Arm Test, Motricity Index (arm), and Semmes-Weinstein monofilament test for the contralesional (left) arm, across all phases (A, B, A) of the study



Key:

ARAT = Action Research Arm Test; MI = Motricity Index; SWM = Semmes-Weinstein monofilaments

A = phase with no MTS intervention; B = phase with MTS intervention

MI (arm section) performance was stable with total scores of between 19–24 during the baseline A phase, achieved by activity in the shoulder and elbow subsections only. The pinch subsection remained consistently at zero throughout this phase

indicating no voluntary muscle activation. However, after only one treatment session, total MI (arm) scores increased to 40 in the intervention phase, showing an immediate change in level. Thereafter, scores continued to increase, reaching a maximum score of 65 after 11 MTS treatments. Scores remained stable, at 55–65, to the end of the intervention phase and into the withdrawal phase. Specifically, pinch scores increased from zero (no movement) in the baseline phase to 11 (representing the beginnings of prehension) in the intervention phase after one treatment session. Pinch scores continued to rise up to maximum of 24 (ability to grip cube against a pull, but weaker than the other side) during the intervention phase. Elbow and shoulder subsections remained relatively constant at 14 (movement seen but not full range / not against gravity).

ARAT scores were consistently zero during the baseline phase, indicating no UL activity function or capacity. ARAT scores increased in the intervention (B) phase after just three treatments of MTS, reaching a total ARAT score of 17 after 10 MTS treatment sessions. Thereafter, performance remained stable throughout the intervention phase and into the withdrawal phase. The difference between baseline (A) and intervention (B) phase scores (17 points for the non-dominant UL) was clinically significant.

SWM scores were absent during baseline for both index finger and thumb, indicating no perception of deep pressure sensation (300g force) in either digit. However, after just one treatment of MTS, his sensation improved, recognising a force of 180g (monofilament number 19). This represents change in level and trend at the point at which the MTS intervention was introduced. Sensory threshold continued to improve throughout the intervention and withdrawal phases. A 0.60g force (SWM number 7), representing diminished protective sensation, was the smallest threshold recorded.

Observable UN behaviour reduced substantially after just one treatment of MTS. On clinical assessment, Derek demonstrated an immediate increase in attention to and voluntary activation of the contralesional (left) upper limb. Spontaneous head turning to the left was also observed during communication.

Summary of the case study

Prior to receiving an intensive dose of MTS for the contralesional (left) upper limb,

Derek presented clinically with marked observable UN behaviour, no voluntary

movement of the left UL, and inability to attend spontaneously to the left-hand side.

Sensorimotor performance for all outcome measures was stable during baseline (the 'control'). However, after only one treatment session of MTS, sensation and motor impairment in the neglected UL and UN behaviour all improved, with subsequent activity capacity improving after just three treatment sessions. Furthermore, these improvements were maintained once the intervention had been withdrawn.

Discussion

Of particular interest in this case was that the observable UN behaviour as well as UL sensorimotor performance improved markedly after just one MTS treatment session. It could be hypothesised that this improved UN behaviour was associated with the corresponding improved sensory perception and voluntary muscle activation, which raised intrinsic awareness of the neglected left side. However, from this single n=1 study, it is not possible to establish any relationships between these factors. Further investigation of the effects of MTS on UN needs to be undertaken with a larger group of stroke survivors with UN, and with valid and reliable measurement of UN as an outcome.

Intensity of therapy

Although there were days when therapy could not be provided by the experienced physiotherapist, the dose of MTS received in this study was intensive: 45 minutes per day, 5 days per week, for 6 weeks. Replicating this dose in routine stroke rehabilitation is limited by resources (time and cost). Experienced therapists do not have the opportunity to spend sufficient time with individual patients to deliver MTS at this intensity on a regular basis, particularly when rehabilitation goals of functional mobility are prioritised. Alternative methods of delivering MTS at this intensity should be considered to improve the quality and intensity of rehabilitation. Using the 24-hour approach to rehabilitation to optimise recovery potential (Aries and Hunter, 2014), it may be possible that other members of the multidisciplinary team, including nurses and carers / family members, could be trained to deliver some of the components of MTS.

Components of MTS that might be taught to others include massage and sensory stimulation techniques involving touch, holding and squeezing the hand. Therapeutic touch and hand-holding are often features of general nursing care, for example when providing reassurance, talking and listening to patients, and when providing comfort to patients. Consequently, there is an opportunity to integrate specific components of MTS into general care throughout the whole day. More frequent repetition of sensory stimulation might in turn improve both recovery of UL function and UN after stroke. Further studies involving nurses, carers and other members of the rehabilitation team need to be undertaken a) to explore the training needs, feasibility and acceptability of components of MTS being delivered by non-physiotherapists, and b) to evaluate the effectiveness of this transference of specific therapy intervention to non-therapists.

Multidisciplinary approach to somatosensory stimulation

Strategies to increase attention to the neglected side of space are reinforced by all members the multidisciplinary team in conventional stroke rehabilitation. Whilst these strategies may be helpful, the sensory stimulation involved in cueing (visual, auditory), for example by approaching the person and talking to them from the neglected side, is considered to be external to the body ('exteroceptive'). In contrast, somatosensory, proprioceptive and kinaesthetic sensations are considered to be internal to the body, informing about limb position and movement, and connecting body segments together to form a body schema. It may be that this internal proprioceptive stimulation, provided by intensive MTS, is more appropriate and meaningful to 'reawaken' the limbs on the neglected side of space, and to decrease UN behaviour. More frequent provision of aspects of intrinsic sensory stimulation by nurses and other members of the multidisciplinary team would supplement that provided during more formal physiotherapy. It is anticipated that this would in turn improve outcomes for all stroke survivors, but this has yet to be evaluated in robust research.

Conclusion

In addition to an immediate improvement in UL sensorimotor function following one treatment session of MTS, this intervention appeared to also have an immediate and cumulative effect on clinically observable UN behaviour in this stroke survivor. This change in behaviour and sensorimotor performance was maintained when the intervention was withdrawn. However, this has only been described in one case, and further investigation of the effects of MTS on a larger sample of stroke survivors with UN is warranted. Similarly, identifying methods to increase the regularity and dose of delivering MTS is necessary. Such a method could include non-therapists, nurses, or carers, being trained to provide some aspects of MTS during routine activities.

Delivery of more frequent and intensive MTS throughout the day and evening has

potential to improve outcomes for all stroke survivors with UL dysfunction and UN.

However, the feasibility, acceptability and effectiveness of other members of the multidisciplinary team delivering components of MTS have yet to be explored.

Acknowledgements: Thanks are extended to Research into Ageing (Bernard Isaacs Memorial Fellowship) for funding this work.

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