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techniques for unilateral neglect: An ABACADAEAF single-case experimental design

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Unilateral neglect is a debilitating attentional disorder whereby patients fail to report, respond or orient to information presented on one side of space. Previous studies have demonstrated improvements in neglect symptoms using rehabilitation techniques, such as anchoring or limb activation. We investigated the effectiveness of five interventions in reducing the unilateral neglect observed in patient F.P. A single-case ABACADAEAF design was used to investigate the effectiveness of musical stimulation (B), anchoring (C), vibratory stimulation (D), limb activation (E), and anchoring and vibratory stimulation combined (F), compared to baseline (A). Severity of neglect was measured using star cancellation, line crossing and line bisection tests. Tau-U statistical analyses were used to investigate significant differences between conditions. All interventions resulted in improvements in F.P.'s neglect. Anchoring (C), vibratory stimulation (D) and the combination of these two techniques (F) led to greatest improvements on all three tests of neglect. Musical stimulation led to improvements on the line bisection task only.

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Anchoring and vibratory stimulation were the most effective techniques for reducing neglect for this patient. Further research is needed to investigate whether the observed gains can be sustained on a longer-term basis, generalised to other tasks, and replicated in larger samples.

Keywords: Neglect; Rehabilitation; Anchoring; Limb activation; Vibratory stimulation.

INTRODUCTION

Unilateral neglect is a debilitating attentional disorder whereby patients fail to report, respond or orient to information presented on one side of space opposite a brain lesion (Heilman, Watson, & Valenstein, 2011).

Numerous studies have investigated, and demonstrated, improvements in neglect symptoms using various rehabilitation techniques. One of the most commonly used techniques involves spatial cueing, training the individual to look towards a salient "anchor" positioned on the neglected side. Over three decades ago, Weinberg et al. (1977) showed that using a highly salient cue, such as a drawing a thick red line down the left side of the page could reduce neglect and improve performance. This was part of the scanning training taught to patients with neglect. Another strategy that has been used is limb activation. In 1989, Halligan and Marshall found that neglect could be reduced if tasks were carried out using the left hand. This, in turn, led Robertson, North, and Geggie (1992) to develop limb activation treatment in which the patient moves the affected limb or, if hemiplegic, moves some part of the affected limb such as the shoulder or one finger. They later found that the movement must take place on the neglected side of space in order to be effective (Robertson & North, 1994). Several studies, including a randomised control trial, have confirmed the efficacy of limb activation (Robertson, McMillan, MacLeod, Edgeworth, & Brock, 2002).

In a series of studies, Robertson and colleagues also introduced a neglect alert device (NAD), a portable box which sounds at intermittent intervals, cueing the patient to use the neglected limb to prevent or terminate the sound by pressing a button. Training with the NAD resulted in improved performance on tests of neglect across a range of tasks including spatial tests and everyday activities, with improvements being maintained for some patients following training (Robertson, North, & Geggie, 1992; Robertson, Hogg, & McMillan, 1998).

More recent studies have investigated the potential beneficial effects of using tools emitting electrical stimulation. In a study of nine stroke patients, both active and passive limb activation improved performance on visual scanning tests (Eskes, Butler, McDonald, Harrison, & Phillips, 2003). In the active

limb activation condition, patients were required to push a button using the left hand on the neglected side, and two out of three patients improved. In the passive condition, electrical stimulation was applied to the neglected forearm and six out of eight patients showed an improvement. Harding and Riddoch (2009) also reported benefits of electrical stimulation applied to the ipsilesional forearm in three out of four patients that were absent from a control right-arm condition. In their study, electrical stimulation was applied daily for 20 minutes over a period of seven weeks in total, and the benefits were sustained at six months post-treatment.

The MotivAider® is a small electronic device which can provide attentional cues and help individuals manage their own behaviour more effectively. It can be set to emit vibrations at intermittent intervals (ranging from seconds to minutes) and can be attached to an individual (e.g., on a belt or strapped to the arm). Previous studies conducted in educational settings have reported improved attention in children using the MotivAider®. Flaute, Peterson, Van Norman, Riffle, and Eakins (2005) outlined various ways in which the device could be used to aid both students and teachers to attend to their behaviour across different classroom scenarios, such as reminding a child to keep on task, or cueing a teacher to praise the class, reinforcing good behaviour. Legge, Debar, and Alber-Morgan (2010) also investigated the use of the MotivAider® in helping two students with autism and one with cerebral palsy difficulties to stay on-task. The students were trained to self-monitor how on-task they were each time the MotivAider® vibrated and results showed improvements in on-task behaviour following this training. Furthermore, benefits were maintained following termination of the self-monitoring procedure. To our knowledge, no studies to date have determined the feasibility of using the MotivAider® to help reduce unilateral neglect.

Attentional processes have been proposed to account for hemispatial neglect and researchers have investigated this in a variety of ways. In a recent study, Soto and colleagues (2009) investigated the effect of mood and music on neglect, and reported an improvement on neglect tests when subjects listened to preferred music and reported positive mood. They proposed that neglect could be modulated by emotional affect and result in improved visual attention.

Although large amounts of research have been conducted on possible neuropsychological rehabilitative techniques for unilateral neglect, few studies have directly compared different techniques within a single study.

The aim of the present study was to compare the effectiveness of five rehabilitation interventions in reducing neglect in patient F.P. The conditions included: musical stimulation; anchoring; vibratory stimulation (Motiv-Aider®); limb activation (tapping); and anchoring and vibratory stimulation combined.

CASE STUDY

Background and clinical history

F.P. is a 51-year-old right handed male with severe left sided neglect. He had been a successful musician (drummer) and toured the world performing until August 2010 when he experienced a sudden onset of left hemiparesis and slurred speech suggestive of a right hemisphere stroke. He was admitted to hospital and was on one-to-one supervision due to risk of falls and potential risk to self and others.

In January 2011, F.P. was transferred to a neurological rehabilitation centre in the south of England. At the centre he required one-to-one supervision in activities of daily living. Due to comorbid mental health difficulties and psychotic episodes he was transferred, in the same year, to a neurological rehabilitation centre with a psychiatric inpatient unit.

Medical records showed that F.P. had a history of mental health difficulties and had also been diagnosed with epilepsy in 2004, Type 1 diabetes in 2005 and alcoholic Korsakoff's syndrome.

Neuroimaging investigations

A computed tomography (CT) scan conducted in March 2011 revealed: "Prominence of the ventricular system with minor asymmetry (the right lateral ventricle is larger than the left) with further enlargement of the third and fourth ventricles. This raises the possibility of open or low pressure hydrocephalus. There is no true small vessel disease and no periventricular lucency." No focal change within the cerebral substance, recent infarction or haemorrhage was reported.

Neuropsychological assessments

A neuropsychological assessment was conducted in April 2011 and repeated in 2012. There was little change in the assessment results over the year and Table 1 outlines results of the assessment conducted in 2012.

As shown in Table 1, F.P.'s level of premorbid functioning was estimated to be in the low average range. On the WAIS-4 he scored below the first percentile on his full scale IQ, however this is likely to have been an underestimate due to neglect and visual attention difficulties. Although he just passed the screening test of the Visual Object and Space Perception Battery (VOSP), F.P. failed all of the remaining subtests. He also had poor naming abilities, measured using the Graded Naming Test (GNT), and impaired memory. On the Rivermead Behavioural Memory Test - 3 (RBMT-3) his highest age-scaled scores were 4 for name learning, face recognition and delayed messages, where 10 is average. The Hospital Anxiety and Depression Scale

Test	Subtest	Raw score	Statistic scores and comments	
TOPF		25	Predicted pre-morbid $IQ = 88$	
WAIS-IV	Full scale IQ	59	Below 1 st percentile	
	Verbal Comprehension Index	83	13 th percentile	
	Perceptual Reasoning Index	50	Below 1 st percentile	
	Working Memory Index	74	4th percentile	
	Processing Speed	50	Below 1 st percentile	
	Information (general knowledge)		Age Scaled Score $= 9$	
	Vocabulary		Age Scaled Score $= 7$	
	Block design/symbol search/visual puzzles and coding		Age Scaled Score $= 1$	
GNT		10/30	5 th percentile	
VOSP	Screening Test	15/20	Bare pass	
	Incomplete Letters	12/20	Fail	
	Dot Counting	0/10	Fail	
	Position Discrimination	11/20	Fail	
	Cube Analysis	0/10	Fail	
RBMT - 3	General Memory Index	53	Below 1 st percentile	
HADS	Depressive symptoms	5/21	Normal range	
	Anxiety symptoms	11/21	Abnormal range	
BIT	Line crossing	19/36 omissions	Severely impaired	
	Line bisection	4/9	Impaired	
	Figure and shape copying	0/9	Severely impaired	
	Letter cancellation	14/36	Severely impaired	
	Star cancellation	40 omissions	Severely impaired	

 TABLE 1

 Neuropsychological assessments conducted with F.P. in April 2012

TOPF = Test of Premorbid Functioning; WAIS-IV = Weschler Adult Intelligence Scale 4th edition; GNT = Graded Naming Test; VOSP = Visual Object and Space Perception Battery; RBMT-3 = Rivermead Behavioural Memory Test - 3; HADS = Hospital Anxiety and Depression Scale; BIT = Behavioural Inattention Test. (HADS) revealed no evidence of depression but anxiety scores were in the abnormal range.

In April 2012, F.P. was severely impaired on all of the conventional subtests of the Behavioural Inattention Test (BIT; Wilson, Cockburn, & Halligan, 1987), except for the line bisection task on which he was slightly less impaired but still scored outside the normal range.

Measures of neglect

In the present study, severity of neglect was measured by F.P.'s performance on the star cancellation, line crossing and line bisection subtests of the BIT. In the line bisection test, F.P. was asked to mark the midpoint of three horizontal lines, positioned towards the right, centre and left hand side of the page. The stimulus paper was aligned to the midsagittal plane of F.P.'s body and deviations from the midpoint of each of the three lines were measured to the closest millimeter. Positive values denote rightward deviation from the midpoint, while negative values denote a leftward bias.

On the star cancellation and line crossing tests, F.P. was instructed to search for, and cross out all of the stars or lines (respectively) presented on the page. The number of stars/lines omitted was recorded.

F.P. was impaired on way finding and needed reminding about the route to take when walking around the hospital. He frequently collided with doorways, walked towards the left side of corridors and had difficulty with balance (e.g., falling backwards) when walking upstairs. The Catherine Bergego Scale (Azouvi, 1996) was completed by F.P. and also by a member of staff who worked with F.P. pre- and post-intervention in order to obtain a rating of F.P.'s functional difficulties in activities of daily living (e.g., grooming and shaving the left part of the face) due to neglect. Neglect behaviour is rated on a 4-point scale: absent (0), mild (1), moderate (2), or severe (3). This scale was completed both prior to the treatment interventions and 4 months following the end of the study.

Design

F.P. participated in the present study over a period of five months and completed each of the tests on five occasions across consecutive days. An ABA-CADAEAF design was used to test performance across five experimental conditions. A stable baseline (condition A) of F.P.'s test performance was initially obtained, and repeated between each of the test conditions. In the musical stimulation condition (B), F.P. selected a track of his own music and listened to this while completing the tests. Anchoring (condition C), involved the experimenter drawing a thick red line down the left hand side of the test page. F.P. was instructed to look for the red line before beginning

388 TUNNARD AND WILSON

the tests. In the passive limb activation condition (D), vibratory stimulation was applied to the lower part of F.P.'s left arm using a MotivAider®. This device was set to vibrate at 10 second intervals, and provided a cue towards the left side. In the active limb activation condition (E), F.P. again wore the MotivAider® as outlined above, but was also instructed to tap the table-top every time that he felt a vibration. In the final treatment condition (F), both the anchoring (as described previously) and active limb activation techniques were used in combination.

Ongoing treatment

Throughout the study F.P. continued his regular timetable of activities including daily physiotherapy (mainly weight lifting and cycling) and occupational therapy (OT). In OT sessions, F.P. carried out various table-top activities (e.g., a pegboard task), reading, computing and also participated in various group sessions (e.g., orientation). His therapist reported that despite his neglect he could attend to the left side with verbal cues and prompts and was actively encouraged to use his left hand as much as possible during sessions. During the reading sessions he would ignore the left side completely but this could be improved within sessions when verbal prompts were given. F.P enjoyed computing sessions to keep in contact with his family but required full assistance in reading and writing e-mails due to his neglect. Despite therapy sessions aiming to improve F.P.'s neglect, it has remained stable with little improvement when assessed in April 2011 and April 2012.

Statistical analysis

Tau-U analyses were conducted to investigate whether significant reductions in neglect were found between the different conditions. Tau-U is a method for measuring data non-overlap between two conditions and it offers the possibility to control for baseline trend, that is, it allows quantifying the amount of improvement in the intervention phase beyond a potential improvement starting before the intervention. Moreover, this effect size index has been reported to be suitable in single case research where data often do not conform to parametric assumptions. It has a statistical power of 91–95% of (ordinary least squares) linear regression when data conform to parametric assumptions, or 115% when data do not. This method can control for baseline trend and, where necessary, analyses controlled for baseline trend. All calculations were performed via the website: http://singlecaseresearch.org/ (Vannest, Parker, & Gonen, 2011). Further information on the Tau-U method has been detailed by Parker, Vannest, Davis, and Sauber (2011).

Results

All interventions resulted in improvements in F.P.'s left-sided neglect, with anchoring (C), vibratory stimulation (D) and the combination of these two techniques (F) leading to greatest improvements on all three tests of neglect compared to baseline. Musical stimulation was the least effective intervention on the cancellation tasks but led to improvements on the line bisection task.

As shown in Figure 1, on the star cancellation task, a reduction in mean omissions went from a high of 67% in the first baseline phase to a low of 45% in the anchoring condition (C) and 36% in the final treatment stage (F). Tau-U analyses revealed a significant reduction in omissions between the second baseline and the anchoring condition (-1, p < .01). A significant reduction was also observed in the fifth baseline and the anchoring and vibratory stimulation combined condition after correcting for the improvement already present in the baseline data (-0.76, p < .05).

On the line crossing task, the mean number of omissions was highest in the music condition (50% omitted) and the first and second baseline phases (48% and 49%, respectively), and lowest in the anchoring (18%), vibratory stimulation (18%) and anchoring and vibratory stimulation combined conditions (9%) (see Figure 2). A significant difference in line crossing

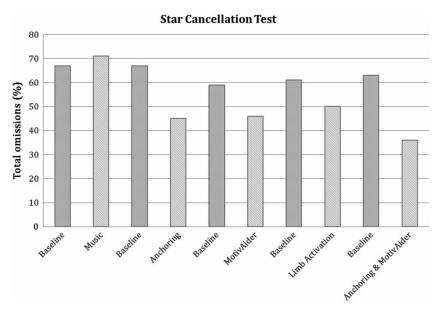


Figure 1. Mean percent of omissions across all trials for each condition on the Star Cancellation Test.

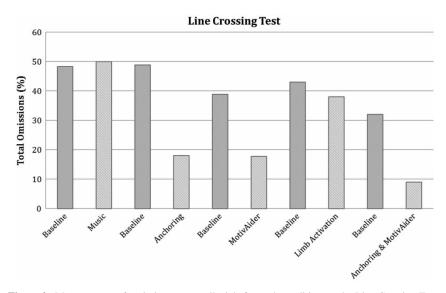


Figure 2. Mean percent of omissions across all trials for each condition on the Line Crossing Test.

performance was found between the second baseline and anchoring condition (-1, p < .01), and the fifth baseline and the combined anchoring and vibratory stimulation condition (-1, p < .01).

Results on the line bisection task are shown in Figures 3 and 4. Figure 3 shows F.P.'s overall ability to bisect the centre of all three lines on the page. A significant reduction in this bias towards the right hand side was found between the first baseline condition and the music condition, the second baseline and the anchoring condition and the fifth baseline and the anchoring and vibratory stimulation combined condition (-0.76, p < .05 for all three comparisons). The improving trends in the first and second baselines were controlled for and, thus, the effect quantified by Tau-U is the decrease observed in the B and C phases beyond the initial improvements.

In addition to measuring F.P.'s overall performance at bisecting all three lines, we found that his performance at bisecting the right hand line appeared to be greatly improved in the anchoring and vibratory stimulation combined condition, whereby he bisected to the left of the midpoint (see Figure 3). However, this difference was not found to be significant due to overlapping data points between the two conditions.

However, interestingly, F.P. showed a significant improvement on bisecting the right hand line when he listened to the music, compared to baseline (-0.84, p < .05) See appendices for raw data.

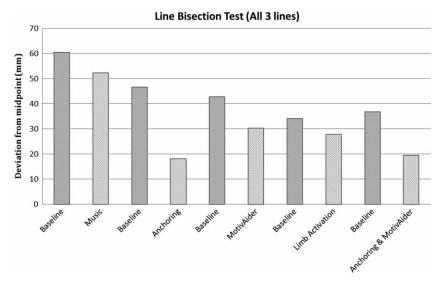


Figure 3. Performance on the Line Bisection Test (for left, centre and right positioned lines).

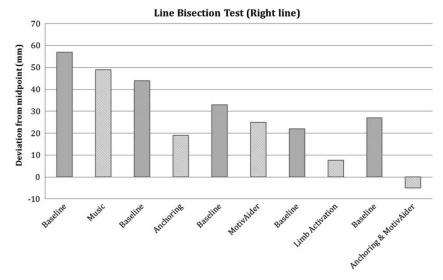


Figure 4. Performance on the Line Bisection Test for the right line only.

Functional assessment of neglect

Ratings on the Catherine Bergego Scale (CBS) showed reductions in both self-rated and carer-rated neglect on everyday activities following the

training. At baseline, F.P.'s self-rated and carer-rated scores were both 26 indicating F.P. had good insight into his difficulties. Following the interventions, reductions were seen in both self-rated and carer-rated scores, rated as 15 and 19, respectively.

DISCUSSION

A number of treatment procedures have previously been demonstrated as helpful for people with unilateral neglect including music therapy, anchoring, limb activation and alerting systems. However, to our knowledge no previous study has compared several different treatment strategies in a well-controlled single case experimental design (SCED).

The patient described here, F.P., had left-sided neglect that had persisted for three years and, despite regular inpatient treatment, his neglect remained severe, causing him to require help in his everyday functioning. He seemed to show reasonable insight into his difficulties as his score on a rating scale agreed with that of a carer who knew F.P. well.

We compared five different treatment methods using an ABACADAEAF design, thus returning to baseline after each intervention strategy. One finding was that musical stimulation improved F.P.'s neglect on the line bisection task but not on the cancellation tasks, thereby partially supporting findings by Soto and colleagues (2009). This highlights the importance of measuring performance across different tests of neglect.

Another finding was that anchoring consistently and significantly reduced F.P.'s neglect across all three tests compared to baseline. Furthermore, cueing using vibratory stimulation also reduced the neglect, but the combination of anchoring and the vibratory stimulation was the most effective treatment strategy on the line crossing and line bisection tests. Thus we have shown that anchoring and vibratory stimulation, and the combination of the two appeared to be the most consistently effective techniques in reducing symptoms of neglect for this patient.

These findings are of interest as significant improvements were apparent over a relatively short period of training (one week), although it should be borne in mind that F.P.'s performance was measured while the interventions were in place and required prompting by the experimenter (e.g., drawing the anchoring line, etc.). This contrasts with Harding and Riddoch's (2009) study, whereby stimulation was an intensive daily treatment intervention with improvements in neglect measured following this. One advantage of the MotivAider® used in this study is that it is portable and can easily be worn while a patient carries out activities of daily living, and thus could potentially help increase independence in these tasks. At the end of the treatment phases, we had hoped to observe whether there were any longer-term benefits of the interventions, and also whether F.P. showed any behavioural improvements in functioning in daily activities following the treatment (e.g., a reduction in collisions with doorways and other objects, ability to shave, etc.). However, unfortunately he was transferred to a different rehabilitation centre before this was possible, thus we were unable to observe in detail F.P.'s behaviour in his natural environment. Nevertheless we did obtain a caregiver's rating of F.P.'s neglect following the treatment, which suggested that there may have been some generalisation of improvement to everyday life even though this was not formally measured. F.P. himself also rated his neglect as less severe.

One limitation of the study was that we did not use a different neglect task to test for any improvements post-training. Practice effects and natural recovery can be a difficulty faced by investigators conducting SCED studies. However, in this study, F.P. had longstanding unilateral neglect that had shown little change over time. We used a carefully controlled ABACA-DAEAF design and although some improvements already present in the baseline phases were observed across baselines, and we cannot be sure what led to these differences, we controlled for this using the Tau-U statistical method. It would have been useful to have included additional controls, such as applying vibration on the right arm, however we felt this was not practicable within this study considering the number of different interventions that were evaluated, and due to time constraints.

Ultimately, all clinicians want to know if their treatment is effective and this is where SCEDs are so useful as they allow us to tease out the effects of treatment from the effects of spontaneous recovery, and other non-specific factors. If we wish to find out whether a particular brain injured person is benefiting from a specific kind of procedure we need to employ a SCED. If we want to find out *how many* people are benefiting from this procedure and whether it generalises to other patients with similar difficulties, we would conduct a group study. For instance, F.P. used to be a successful musician and we do not know whether the improvements on the musical stimulation condition would also generalise to other patients with unilateral neglect. Nevertheless, conducting this study was relatively quick and feasible to do and we could accommodate F.P.'s daily timetable at the centre, which may have been difficult in a larger group study.

However, there are times when SCEDs are problematic, for example, once we have taught someone to do something we cannot then "unteach" them. Furthermore, it may be difficult to obtain stable baselines with certain patients, or unethical to postpone treatment until a stable baseline has been obtained (e.g., if treating pressure sores or severe self-injurious behaviour). Nevertheless, we should always try to determine whether our treatment, or some other factor, is responsible for any change we see and, for this, the SCED is one of the best methods we can employ.

In summary, this study shows that several treatments may impact on the manifestation of neglect. A combination of anchoring and vibratory stimulation was seen to be the most helpful for F.P., with musical stimulation also showing some promise.

Further research is required to determine whether these findings also generalise to larger samples of patients experiencing longstanding unilateral neglect. Future studies could also investigate: (1) whether the observed gains are sustained longer term following the training period, (2) whether patients would be able to implement these interventions independently following training, and (3) whether improvements are generalised to functional tasks (e.g., self-care).

CONCLUSION

Anchoring and vibratory stimulation, and the combination of the two appeared to be the most effective techniques in reducing symptoms of neglect for this patient. Musical stimulation also showed some promise, specifically on the line bisection task. Future research is required to further investigate whether the gains seen using these interventions remain following the training period and whether they generalise to other patients and across more functional tasks.

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396 TUNNARD AND WILSON

APPENDIX 1

Condition	Trial no.	Total omissions
Baseline (A)	1	34
	2	35
	3	35
	4	38
	5	38
Music (B)	1	39
	2	36
	3	38
	4	38
	5	41
Baseline (A)	1	38
	2	38
	3	31
	4	33
	5	41
Anchoring (C)	1	23
	2	22
	3	25
	4	27
	5	25
Baseline (A)	1	43
	2	27
	3	39
	4	31
	5	18
MotivAider (D)	1	19
	2	39
MotivAider (D)	3	28
	4	19
	5	18
Baseline (A)	1	29
	2	36
	3	31
	4	30
	5	39
Limb Activation (E)	1	23
	2	33
	3	35
	4	19
	5	26
	3	20

TABLE A1 Raw data for the Star Cancellation Test showing total omissions across each trial

(Continued)

Continued			
Condition	Trial no.	Total omissions	
Baseline (A)	1	36	
	2	37	
	3	35	
	4	28	
	5	33	
Anchoring & Limb Activation (F)	1	15	
	2	15	
	3	16	
	4	27	
	5	25	

TABLE A1

APPENDIX 2

TABLE A2 Raw data for the Line Crossing Test showing total omissions across each trial

Condition	Trial no.	Total omissions
Baseline (A)	1	18
	2	24
	3	12
	4	19
	5	14
Music (B)	1	18
	2	18
	3	18
	4	18
	5	18
Baseline (A)	1	18
	2	18
	3	18
	4	14
	5	20
Anchoring (C)	1	13
	2	7
	3	5
	4	0
	5	7

(Continued)

398 TUNNARD AND WILSON

Continued				
Condition	Trial no.	Total omissions		
Baseline (A)	1	18		
	2	18		
	3	18		
	4	6		
	5	10		
MotivAider (D)	1	13		
	2	11		
	3	1		
	4	7		
	5	0		
Baseline (A)	1	12		
	2	16		
	3	18		
	4	13		
	5	18		
Limb Activation (E)	1	5		
	2	18		
	3	18		
	4	8		
	5	19		
Baseline (A)	1	12		
	2	9		
	3	12		
	4	12		
	5	12		
Anchoring & Limb Activation (F)	1	5		
	2	0		
	3	0		
	4	3		
	5	8		

TABLE A2

APPENDIX 3

TABLE A3 Raw data for Line Bisection Test showing mean deviation (mm) from the midpoint for each line across each trial

Condition	Trial no.	Right line	Centre line	Left line
Baseline (A)	1	545	645	735
	2	525	435	685
	3	705	555	735
	4	615	555	765
	5	465	395	705

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(Continued)

	TABLE A Continue			
Condition	Trial no.	Right line	Centre line	Left line
Music (B)	1	635	505	725
	2	425	505	565
	3	505	445	575
	4	415	475	615
	5	465	485	495
Baseline (A)	1	525	565	535
	2	355	295	585
	3	455	475	575
	4	615	675	455
	5	245	225	405
Anchoring (C)	1	-245	-75	425
	2	-145	145	375
	3	-225	-5	405
	4	385	355	235
	5	325	215	535
Baseline (A)	1	245	305	355
	2	515	625	645
	3	575	695	595
	4	75	105	375
	5	235	535	535
MotivAider (D)	1	475	455	385
	2	465	355	415
	3	255	355	275
	4	-5	395	495
	5	35	275	-85
Baseline (A)	1	-25	205	475
Basellile (A)	2	445	203 465	475
	2 3	525	325	423 645
	4	25	285	575
	4 5	135		345
Limb Activation (E)	1	-245	265 315	545 275
Linio Activation (E)				485
	2 3	225 245	465	
			335	515
	4	-175	135	365
Descline (A)	5	335	305	585
Baseline (A)	1	335	495	595
	2	135	325	365
	3	195	235	485
	4	225	295	335
	5	475	465	545
Anchoring & Limb Activation (F)	1	-205	225	415
	2	-205	285	265
	3	175	275	285
	4	-195	-5	365
	5	185	405	635

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