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NEUROPSYCHOLOGICAL PERFORMANCE IN TOURETTE SYNDROME AFTER AN EQUINE-THERAPY INTERVENTION

N. Cassimjee, R. Grobler and C. Wagner

ABSTRACT

Equine-therapy is a new form of therapeutic horse riding that purports to enhance the stimulation of the vestibular and somatosensory systems through processes of cortical sensory integration. With regard to their neuropsychological profile, Tourette syndrome children often manifest with executive functioning deficits attributable to disruptions in frontal functioning. Since equine-therapy focuses on stimulation and augmentation of frontal integrative processes, an intervention of this nature may influence the executive functioning of this group. The purpose of this study was to explore whether an equine-therapy intervention will influence neuropsychological (executive function) outcomes in a group with Tourette syndrome. The sample consisted of eight individuals with Tourette syndrome who were assessed on a neuropsychological test battery comprising the Wisconsin Card Sorting Test, Stroop Test, Rey Complex Figure Test, Trail Making Test, Raven's Matrices, and Symbol Digit Test. The statistical analysis indicated significant differences on the Wisconsin Card Sorting Test, Stroop Test and Symbol Digit Modalities Test. These changes suggest that equine-therapy may augment self-corrective strategies, selective attention, visual-associative and visuo-motor integration abilities in Tourette syndrome. The results show that the stimulation of vestibular and somatosensory systems may enhance the integrative abilities of the frontal cortex as manifested in the cognitive performance increments.

Key words: Neuropsychological assessment, Tourette syndrome, executive functions, Equine-therapy, sensory integration.

N. Cassimjee, *PhD, Psychology*
R. Grobler, *M.A. Psychology*
C. Wagner, *D. Phil Psychology*
Department of Psychology,
University of Pretoria,
0002

E-mail: ncassim@postino.up.ac.za

INTRODUCTION

Numerous studies have attested to the value of therapeutic horse riding among neurological patient groups. For example, Delius (1998) found improvement in balance, muscle strength, joint range of motion and improved co-ordination. Bertoti (1988) and Rufus (1997) indicated statistically significant improvements in posture, while subjective improvement was observed in self-confidence, muscle tone, weight bearing and sitting balance. Furthermore, positive effects on psychosocial and cognitive benefits such as self-confidence, self-concept, self-esteem, motivation, attention span, spatial awareness, concentration, listening skills, interest in learning and verbal skills were also reported (Basile, 1997; MacKinnon, Noh, Laliberte, Lariviere & Allen, 1995). Few studies, however, have focused on the influence of therapeutic horse riding on cognitive and higher order executive functioning in children with neuro-developmental disorders such as Tourette syndrome.

The first clear medical description of Tourette syndrome dates back to 1825, when J.M. Itard, chief physician at the Institution Royale des Sourdsmuets in Paris, reported a case of a French noblewoman, who developed motor tics at the age of seven, and later, began to use obscene words (Kushner, 1999).



Sixty years later, Gilles de la Tourette described a combination of involuntary motor movements and spontaneous shouting and cursing as a single disease that he aptly called 'maladie des tics' (Kushner, 1999). Tourette syndrome is now recognized as a chronic neuropsychological disorder with a suggested genetic substrate, abnormalities in the frontal cortex and sensory anomalies (Cohen & Leckman, 1992).

Researchers such as Robertson (2000) have suggested an important associative dysfunction between the limbic system and the frontal lobes in patients with Tourette syndrome. The frontal cortex and its limbic connections (including the septum and hippocampus) appear to underlie the tic and hyperactive behaviour found in Tourette syndrome patients. According to several researchers, dysfunction of the frontal-limbic structures has pronounced disinhibitory effects because of its reciprocal connections with the reticular activating system (Lezak, Howieson & Loring, 2004; Luria, 1980). Furthermore, Gedye (1991) suggested that abnormal discharges in the frontal lobe, particularly the motor area of the mesial basal region, underlie the symptoms manifested in Tourette syndrome. With sensory integration therapy it is hypothesized that dysfunctional and chaotic processing of stimuli is modulated and the optimal stimulation and functioning of the vestibular system has a cumulative effect on processing in the frontal lobes, hence executive functioning.

There are many striking similarities between the behaviour of patients with Tourette syndrome

and those with frontal lobe dysfunction (Denckla 1996; Gedye, 1991; Robertson, 2000). Based on the frontal dysfunction hypothesis of Gedye (1991) and the other researchers (e.g., Denckla 1996; Robertson, 2000), one can postulate that frontal integrative deficits, which are implicated in Tourette syndrome, will result in executive deficits, as this function is normally related to frontal regions of the brain. Executive functions are conceptualized as higher order cognitive processes that include volition, planning, purposive action and effective performance (Lezak *et al.*, 2004). Furthermore, executive function is often considered the basis of cognitive and psychological functioning. Functions such as attention and concentration, drive and motivation, cognitive/mental flexibility, planning and organisational skills, the ability to recognise and correct mistakes, the ability to respond to feedback cues, and the ability to understand the consequences of behaviour all fall under the ambit of higher order cognitive processes (Spreeen & Strauss, 1998).

The concept of sensory integration was introduced by Ayres (1979), who suggested that through the stimulation of the sensory network, vestibular system functioning is optimised and stimuli processing is more synchronous and organized. This optimal processing results in adaptive responses that can improve various functions such as motor planning and the execution of tasks that require reasoning and various intellectual abilities (Delius, 1998).

One of the mechanisms used to rouse sensory integration and vestibular stimulation is an alternative form of therapy known as equine-therapy. This forms part of therapeutic horse riding, in which the three-dimensional movements of the horses back is regarded as a possible mechanism to stimulate and manipulate areas of the patient's body and hence influence the sensory integration networks and vestibular system (Ayres, 1979; Bertoti, 1988). An imbalanced (aversive and defensive reactions to sensory input) vestibular system contributes to distractibility and hyperactivity because of the lack of a modulating effect, whereas a well-modulated vestibular system keeps the level of arousal balanced thereby enhancing information processing (executive functions) in the brain (Ayres, 1979; Sollier, 2003). Sensory integration of the sequential processes of praxis and its components of ideation, motor planning and execution of purposeful actions enables efficacious amalgamation of various stimuli. This results in optimal daily functioning by influencing behavioural, cognitive and emotional responses (Cassimjee, 2004). The sensory integration status of a Tourette syndrome child would be considered poor due to the associative dysfunction between cortical and subcortical brain structures. Dysfunctional sensory integration therefore implies that the brain cannot organize the flow of the sensorial impulses effectively.

In equine-therapy, a group of professionals (occupational therapist, psychologist, and physiotherapist) designs a programme suitable

for a particular patient group. The main aim of this programme is to stimulate the vestibular system, which is sensitive to movement. The horse riding activity activates the fluids in the vestibular canals, distributing sensory information to the rest of the brain (Ayres, 1979). The objective of this study therefore was to investigate changes in the neuropsychological performance of children with Tourette syndrome on selected measures of executive function after an equine-therapy intervention.

METHOD

Subjects

Several neurologists and paediatricians in the Pretoria and Johannesburg area referred children between the ages of 9 and 15 years. Participants had to meet the following criteria for inclusion in the final sample: a) definite Tourette syndrome diagnoses - (DSM-IV); b) no diagnosed related behavioural disorders such as attention deficit hyperactivity disorder or obsessive-compulsive disorder; c) live in a geographic location where they could easily access the equine-therapy, d) fluency in written and verbal English or Afrikaans; and e) free of other co-morbid medical conditions that may have an effect on performance tasks.



Furthermore, all other additional new therapies had to be delayed over the study period, although medication already in use could be continued. However, parents had to inform the researcher of medication and/or dosage changes. The study was conducted on the premises of the South African Therapeutic Riding Association (SATRA) in Pretoria. Ethical clearance for the study was obtained from the Faculty of Humanities ethical committee of the University of Pretoria. Parent indemnity forms were signed before the commencement of the intervention and testing.

Due to the exclusion criteria and low prevalence of diagnosed Tourette syndrome, 8 participants were included in the study (3 females and 5 males). This methodological shortfall has been documented in other neuropsychological studies where sample size is commensurate with low prevalence of diagnosed Tourette syndrome children (Comings, Himes & Comings, 1990).

Equine-therapy intervention

Individuals were evaluated on a neuropsychological level with regard to their level of sensory integration of the vestibular system and the somatosensory system, visuo-perceptual skills (limbic system functioning) and hemispheric integration of higher cognitive functions. These functions were assessed by an occupational therapist. The following assessment measures were used: Clinical Observations of Ayres, Southern California Sensory Integration Test (SCSIT), Test for Visual Perception (TVPS-2), Beery Test-

Developmental Test for Motor Integration (VMI) and the Brain Profile Test. Following this evaluation, a special programme was developed by the occupational therapist and a qualified equine-therapist, where certain goals were set to be achieved on the horse to suit the individual.

The final aim was to help the individual to ride the horse on his or her own. During the 3-month intervention (1-hour session per week) the sequence of therapy was as follows: a) the individual is taught to ride at a walk with helpers, b) walk without helpers, c) sitting trot, and d) a walking trot. The length of time spent on each of these phases was dependent on the performance of the participant and the assessment of the performance by the equine-therapist, who facilitated all sessions.

Study Design

A neuropsychological test battery was used to evaluate components of executive functioning 1 week before and 1 week after the completion of a 3-month (1-hour per week) therapeutic horse riding intervention. The research setting and circumstances in which this study occurred made it difficult to employ a true experimental design. Thus, for the purpose of this research a quasi-experimental design was implemented where participants were non-randomly assigned to two groups (4 in each group).



The first group (experimental) was subjected to the horse riding intervention whilst the second, over the same period, was not. However, at the completion of the 3-month intervention the second group, which initially served as a control, was also enrolled for the equine-therapy intervention. Thus, the control group was tested at baseline (pre-measure), 3-months after the pre-measure, and 6-months after the pre-measure. Allowing the control group exposure to the intervention after the initial 3-month period minimised the chance of participant mortality throughout the study and entitled all participants to experience the alternative therapy (Basile, 1997). Firstly, the sample was compared on post-measures of executive functioning, as a control ($n=4$) and experimental group ($n=4$) and secondly, on pre and post-measures, where the person serves as his/her own control ($n=8$).

Measures

A specific battery of six neuropsychological tests was administered. The choice of tests was guided by the following rationale: a) documented in other research to be sensitive to cognitive dysfunctions in Tourette syndrome; b) sensitive to dysfunctions in the modalities related to the executive functions of the brain; c) normed for use on children and adolescence; and d) practice effects are minimal and alternative versions of the tests are available, which make them suitable for retesting (Spreeen & Strauss, 1998).

A psychometrist in a private office with minimal distraction evaluated each participant individually. To standardise the testing situation across the time intervals, the same psychometrist was used and tests were administered in the same sequence. Administration took between 45 and 60 minutes. The concise neuropsychological battery comprised the following tests:

The Wisconsin Card Sorting Test

This test assesses the ability to form abstract concepts, to shift and maintain set, and to utilise feedback, thus providing information on several aspects of problem-solving behaviour beyond such basic indices as task success or failure. In general, poor performance on categories completed, perseverative errors and failure to maintain set would be indicative of frontal lobe dysfunction. Test-retest reliability coefficients are reported to be approximately 0.63 (Lezak *et al.*, 2004).

This test is often used to measure higher cognitive functions (selective attention and cognitive flexibility), and assesses the ease with which a person can shift his or her perceptual set to conform to changing demands, while suppressing a habitual response in favour of an unusual one.

The shorter Victoria version, which only has 24 items, was administered. There are three parts to the test: Part D, Part W and Part C. Part D measures response time of colour naming with the word blue, for example, written in blue. Part W measures response time of naming the colours in which common words are written. Part C is similar to parts D and W, except that here the coloured stimuli are colour names, for example, green printed in the colour red, where the print colour never corresponds to the colour name. The test-retest reliability is satisfactory with reliability coefficients of .90, .83, .91 for the three parts, respectively (Spreen & Strauss, 1998).

The Rey-Osterrieth Complex Figure Test

This instrument measures visuo-spatial constructional ability and visual memory. During administration of the test, the subject's logical construction of the figure was observed, as well as omissions and mutations to the drawing. The copy condition of this test was also analysed qualitatively, to establish if a possible 'frontal' performance profile was evident. This was assessed by observing the random order in which components of the figure were drawn (lack of planning and organisation), inclusion of components not in the original figure (disinhibition), or retracing of components once drawn (motor perseveration) (Walsh, 1987). A drawing displaying any or all of the above characteristics was classified as 'frontal'. A 'normal' performance was any copy that showed none of the above characteristics. Alpha

coefficients of .60 and .80 have been reported for this measure (Spreen & Strauss, 1998).

The Trail Making Test

This measure consists of Part A and B, which addresses selective attention and cognitive shift abilities, particularly, visual scanning and conceptual tracking. The reliability coefficients range from 0.60 to 0.90 (Lezak *et al.*, 2004).

Raven's Progressive Matrices

This test was developed as a measure of non-verbal analogical reasoning, intellectual efficiency and conceptual abilities and is useful to administer to subjects' with expressive language problems, since it does not necessarily require verbal responses. Test-retest reliability coefficients range between 0.70 and 0.90 even when retesting involves three administrations 6 and 12 months apart (Lezak *et al.*, 2004).

The Digit Symbol Modalities Test – Reversed Form

The test is administered to assess visual-associative learning ability, psychomotor/response speed, visuo-motor integration and co-ordination. Reliability coefficients range from 0.82 to 0.88 and minimal practice effects were noted when the test was administered 4 times with intervals varying between 1 week and 3 months (Lezak *et al.*, 2004).



Statistical analysis of data

Since the group (sample) sizes were small, non-parametric statistical tests (Mann-Whitney and Wilcoxon Rank Sum) were used to analyse the data and to test for a significant difference between the scores obtained on the neuropsychological tests. The Mann-Whitney compared the post-score baselines of the experimental group, which had received the intervention, and the control group, which had not. The Wilcoxon compared the pre-test performance and post-test performance on the neuropsychological measures to determine if the intervention had had any effect on performance.

RESULTS

Three significant differences were found between the scores of the experimental group and the control group. On the Wisconsin Card Sorting Test, the scores for categories completed and failure to maintain set differed significantly between the group that received the therapeutic horse riding and the group that had no exposure to the therapeutic intervention. This indicates that self-corrective behaviour was one aspect of executive functioning that improved after the therapeutic intervention. Furthermore, on the Stroop Colour Word Test, scores for uncorrected errors on part C of the test also differed significantly between the two groups. The results on the Stroop Test confirm the findings on the WCST: subjects who participated in the equine-therapy intervention appeared to engage in less perseverative actions when compared to the control group and hence the experimental group showed more self-corrective behaviour

than the control group. Table 1 indicates the means and standard deviations for the two sections of the Wisconsin Card Sorting Test and Table 2 shows the means and standard deviations for the Stroop Colour Test category.

A group mean analysis of the scores using the Wilcoxon rank sum test was obtained by comparing the entire group (after intervention was received by all participants) on a pre-measure and a post-measure. The results indicated 4 significant differences. Three were found on the Stroop Colour Test: reaction time on part W ($p = 0.47$, $p < 0.05$), uncorrected errors on part W ($p = 0.47$, $p < 0.05$), and part C ($p = 0.03$, $p < 0.05$). The fourth difference was found for correct written substitutions on the Symbol Digit Modalities Test ($p = 0.01$, $p < 0.05$). These results highlight improvements in executive functions and speed of information processing.

Figures 1, 2, 3, 4, are graphical representations of score differences on the test indices on which participants showed significant improvements after receiving the intervention.

DISCUSSION

The results indicated that there were significant differences between pre-test and post-test scores on specific indices of the Wisconsin Card Sorting Test, the Stroop Colour Test and the Symbols Digits Modalities Test. This suggested that participants engaged in a 3-month equine-therapy intervention showed improvements on the following aspects of executive functioning:



Table 1: Mean and standard deviations for Wisconsin Card Sorting Test indices

Category	Experimental (n=4)	Controls (n=4)	p-Value
Categories Completed	-1.75 (1.2583)	-0.25 (0.5000)	0.0885*
Failure To Maintain Set	-0.7500 (0.9574)	0.2500 (0.5000)	0.0982*

*Significant, $p < 0.1$

Table 2: Mean and standard deviations for Stroop Colour Test index

Category	Ex group (n=4)	Controls (n=4)	p-Value
Uncorrected errors Part C	2.5000 (0.5774)	4.7500 (1.2583)	0.0360 *

*Significant; $p < 0.05$

Table 3: Mean difference between the pre and post-measure on the Stroop Test

Category	Difference mean (n=8)	p-Value
Time in seconds Part W	7.2500 (7.4785)	0.0469 *
Uncorrected errors Part W	2.5000 (2.5635)	0.0469 *
Part C	1.6250 (1.3025)	0.0313 *

*Significant; $p < 0.05$

Table 4: Mean difference between the pre and post-measure on the Symbol Digits Modalities Test

Category	Difference mean (n=8)	p-Value
Correct Written Subs	-8.3750 (6.1630)	0.0078 *

*Significant; $p < 0.05$

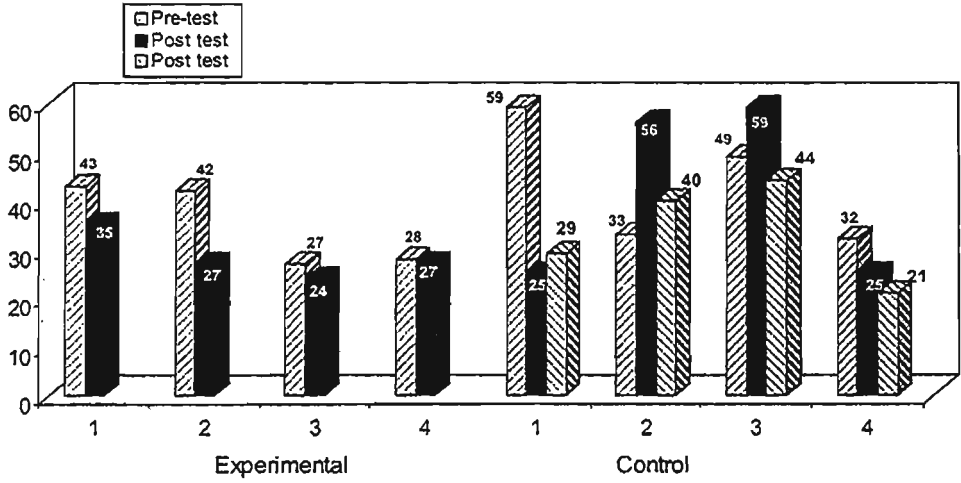


Figure 1: Pre-test and post-test scores on the Stroop Colour Test Part W (Time)

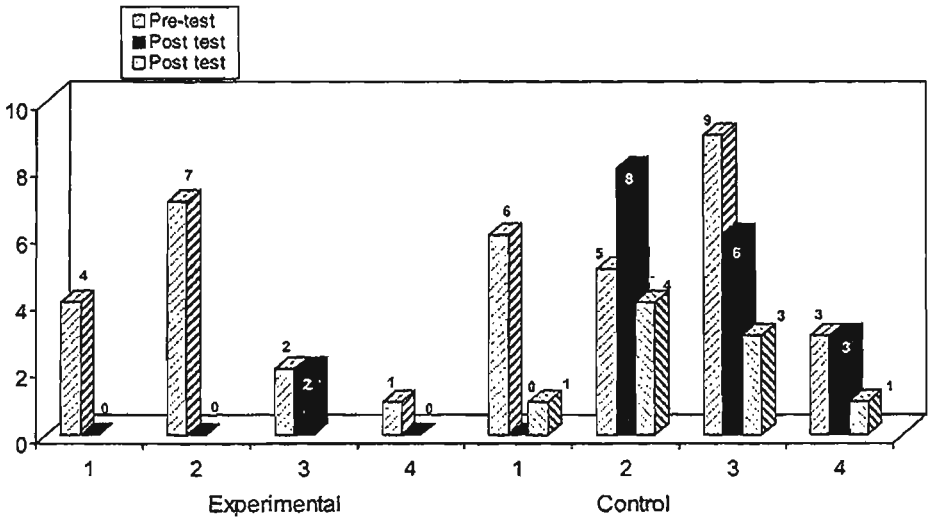


Figure 2: Pre-test and post-test scores on the Stroop Colour Test Part W (Errors)

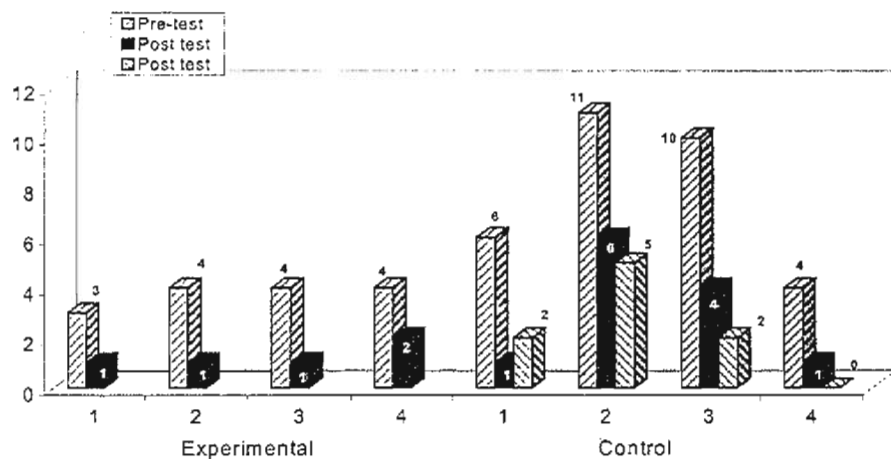


Figure 3: Pre-test and post-test scores on the Stroop Colour Test Part C (Errors)

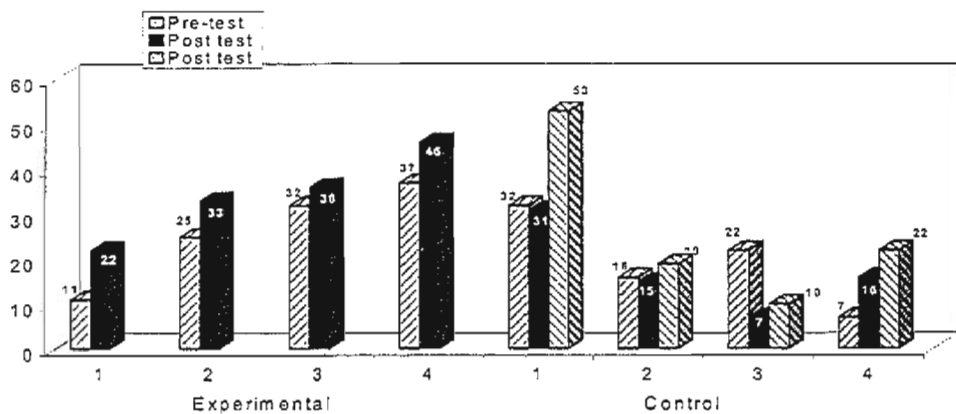


Figure 4: Pre-test and post-test scores on the Symbol Digit Modalities Test (Correct Substitutions)



self-corrective behaviour, selective attention, visual-associative learning and visuo-motor integration abilities.

In the Wisconsin Card Sorting Test, problem-solving behaviour is addressed by the person's abilities to complete card categories and to maintain the cognitive pattern according to the rules of the test. Previous research has documented that Tourette patients manifest with poor problem solving abilities because of frontal dysfunction (Denckla, 1996). Participants who completed the equine-therapy significantly improved their problem solving skills as indicated by the higher scores on the test indices. This indicated an improved ability to form abstract concepts, to shift and maintain cognitive sets and to utilise feedback. It is the latter that underpins the efficacy of processing in the frontal lobes, as the frontal lobes have numerous reciprocal feedback loops to other cortical and subcortical structures (Lezak *et al.*, 2004). Children with Tourette syndrome manifest with behavioural inhibitions that could lead to various cognitive anomalies. It seems as if most of these problems are characterised by attentional problems seemingly due to heightened activity levels and impulsivity, which are features indicative of frontal dysfunction. Robertson (2000) also confers that poor motor control and co-ordination and behavioural disinhibition indicate poor sensorial integration, and if paralleled on a neuropsychological level, are indicative of poor integrative brain functioning (Ayres, 1979; Luria, 1980). Based on this one can hypothesise that sensory integration and

vestibular stimulation enhances the flow of information passing through the neural interconnections and in the absence of chaotic stimuli processing, efferent and afferent information flow amongst brain structures is enhanced, hence frontal lobe functioning is optimised.

Aspects of selective attention and cognitive flexibility, as measured by the Stroop Colour test, significantly improved after participants received the intervention. For Tourette syndrome patients these higher scores could also parallel improved cognitive and motor adaptation. In a previous study, Moe (2000) studied changes in cognitive performance and motor adaptation and found a significant positive correlation between these performances.

In neurological terms, improved executive functions and motor elements may be associated with the regulation of dopamine levels in the brain. Sensory integration and vestibular stimulation also involve the medial-prefrontal areas that have dopamine pathways running from the ventral tegmental area via the mesolimbocortical system to regions in the frontal cortex. Altering dopamine levels in medial areas will influence feedback via the mesolimbocortical network and hence frontal dopamine metabolism (Zillmer & Spiers, 2001).



Due to the attentional processes underlying the set of executive functions measured by the Wisconsin Card Sorting Test and the Stroop Colour Test, improvement in executive performance is likely to be evidenced on the Symbol Digit Modalities Test. The latter instrument assesses the first of three elements of attention, namely the visuo-motor scanning component, an executive deficit common among Tourette patients (Robertson, 2000). Participants showed significant improvement on this executive component after receiving the intervention. The Symbol Digit Modalities Test is regarded as the most sensitive measure of cerebral integrity and has also been endorsed as the single best measure of speed of information processing (Smith, 1991; Ponsford & Kinsella, 1992). Subsequently if sensory integration is about optimising information processing then improvements on the Symbol Digit Modalities Test may imply that equine-therapy has the potential to enhance the sensory integration status and hence components of executive functioning.

Certain methodological limitations should be borne in mind when considering the results of this exploratory study. The sample size was limited and not representative (neuropsychological research is confounded by the problem of sample accessibility, especially because of co-morbid neurological profiles and low prevalence of diagnosed neurological groups). Neuropsychological test results are often susceptible to many extraneous and biographical variables (eg., age, education levels,

testing times, test battery), and future studies should control for these elements. Multi-disciplinary research in this context often provides cross-validation of data, interpretations and conclusions. For example, neuropsychological test results can be correlated with neurological scan results and clinical information from occupational therapists, etc. Finally, due to the poor accessibility of specific target groups these small samples are amenable to subjective assessment from parents, teachers, etc. The latter can enhance the robustness of more objective assessments and should be considered in studies of this nature.

Therapeutic horse riding and more specifically equine-therapy, as a new form of intervention, have received little attention in the South African context. This exploratory study provides a useful springboard for future studies on the potential benefits of equine-therapy for children with neuro-developmental disorders.

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