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Goal Management Training of Executive Functions in Patients with Spina Bifida: A Randomized Controlled Trial

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Abstract

Executive dysfunction causes significant real-life disability for patients with spina bifida (SB). However, no previous research has been directed toward the amelioration of executive functioning deficits amongst persons with SB. Goal Management Training (GMT) is a compensatory cognitive rehabilitation approach, addressing underlying deficits in sustained attention to improve executive function. GMT has received empirical support in studies of other patient groups. The purpose of the present study was to determine the efficacy of GMT in treating subjects with SB, using inpatient intervention periods. We hypothesized post-intervention changes in scores on neuropsychological measures to reflect improved attentional control, including sustained attention and inhibitory control. Thirty-eight adult subjects with SB were included in this randomized controlled trial. Inclusion was based upon the presence of executive functioning complaints. Experimental subjects ($n = 24$) received 21 hr of GMT, with efficacy of GMT being compared to results of subjects in a wait-list condition ($n = 14$). All subjects were assessed at baseline, post-intervention, and at 6-month follow-up. Findings indicated superior effects of GMT on domain-specific neuropsychological measures and on a functional "real-life" measure, all lasting at least 6 months post-treatment. These results show that deficits in executive functioning can be ameliorated in patients with congenital brain dysfunction. (*JINS*, 2013, *19*, 672–685)

Keywords: Executive functioning, Brain injury, Cognitive rehabilitation, Myelomeningocele, Goal management, Evidence-based

INTRODUCTION

Executive functioning (EF) refers to aspects of complex human behavior that are primarily involved in the control and direction of self-regulating behavior (Cicerone, Levin, Malec, Stuss, & Whyte, 2006; Levine et al., 2011; Stuss & Levine, 2002). EF encompasses not only cognitive skills, such as updating of working memory representations, planning, strategy application, and monitoring, but also more emotionally mediated aspects of control, including self-regulation, inhibition, insight, and motivation, all of which are necessary for goal-directed behavior (Duncan, Emslie, Williams, Johnson, & Freer, 1996; Miyake, Emerson, & Friedman, 2000; Stuss & Levine, 2002). The resulting breadth of the EF domain forges its tremendous impact on everyday functioning and correlative importance to human adaptation.

Impairments in EF are typically seen after frontal lobe damage (Stuss & Levine, 2002). However, EF dysfunction has been observed after a range of etiologies and lesion locations, such as traumatic brain injury (Levine et al., 2011), stroke and tumors (Zald & Andreotti, 2010), aging (Raz, 2009), the dementias (Neary et al., 1998), and spina bifida (SB) (Burmeister et al., 2005).

As executive dysfunction hampers the capacities for changing and adapting behavior in new or altered situations (Norman & Shallice, 1986), it often constitutes a significant hindrance to the acquisition of independent living skills (Stuss, 2011). Accordingly, effective interventions aimed at improving EF are needed.

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Spina bifida myelomeningocele (SBM), accounting for approximately 70% of all SB cases (Charney, 1992), is a severe birth defect resulting from a failed closure of the neural tube during fetal development and is associated with several brain abnormalities, including hydrocephalus and Arnold-Chiari malformation (Chiari II) (Barkovich, 2000). Subsequently, SBM has a pervasive multisystemic impact on physical (Fletcher et al., 2005; McDonnell & McCann, 2000) and cognitive functioning (Dennis & Barnes, 2010; Dennis, Landry, Barnes, & Fletcher, 2006; Hampton et al., 2011).

The core cognitive deficits of SBM emerge in infancy and persist throughout life (Dennis et al., 2006). One of the most consistent areas of impairment in SBM is EF (Kelly et al., 2011), interfering with day-to-day living (Burmeister et al., 2005; Mahone, Zabel, Levey, Verda, & Kinsman, 2002; Rose & Holmbeck, 2007) and representing an area of concern in this population (Stubberud & Riemer, 2012; Tuminello, Holmbeck, & Olson, 2011). Fletcher et al. (1996) suggested that the impaired performance on executive tasks in SBM patients is due to injuries in the right posterior region of the brain associated with arousal and activation (Petersen & Posner, 2012). Failure to behave in a goal-directed manner may result from reduced alertness, a foundational form of attention or processing capacity, from which more complex cognitive functions draw (Coull, 1995; Duncan et al., 1996; Greene, Bellgrove, Gill, & Robertson, 2009; Raz & Buhle, 2006; Robertson & Murre, 1999; Smith & Nutt, 1996). Despite the documented presence of executive deficits, there are no published studies that have addressed the treatment of these problems in individuals with SBM. Hence, there is a need to explore the efficacy of cognitive rehabilitation interventions for this patient group.

There have been relatively few validated rehabilitative interventions addressing executive dysfunction (Boelen, Spikman, & Fasotti, 2011; Cicerone et al., 2000, 2005, 2011; Levine, Turner, & Stuss, 2008; Rohling, Faust, & Beverly et al. 2009). However, increasing evidence supports the effectiveness of group-based compensatory interventions involving problem solving and goal management training that have incorporated "stop-and-think training" (D'Zurilla & Goldfried, 1971; Evans, 2005; Levine et al., 2011; Miotto, Evans, de Lucia, & Scaff, 2009; Rath, Simon, Langenbahn, Sherr, & Diller, 2003; Spikman, Boelen, Lamberts, Brouwer, & Fasotti, 2010; von Cramon, Matthes-von Cramon, & Mai, 1991; Wilson, Gracey, Evans, & Bateman, 2009). One of these, Goal Management Training (GMT), is a promising compensatory intervention that teaches strategies for improving attention and problem solving (Levine et al., 2000; Robertson, 1996; Stuss et al., 2007).

GMT is based on a theory of sustained attention (Levine et al., 2011; Robertson & Garavan, 2004), and thus attempts to address underlying deficits in sustained attention such as those associated with acquired brain injuries and SBM (Chen et al., 2011; Fletcher et al., 1996). Studies have demonstrated that low-level arousal deficits can contribute to high-level executive deficits (Coull, 1995; Duncan et al., 1996; Greene et al., 2009; Raz & Buhle, 2006; Robertson & Murre, 1999; Smith & Nutt, 1996). As attention and arousal have a significant role in facilitating experience-dependent plasticity underpinning neurorehabilitation (Robertson & Murre, 1999), in GMT sustained attention and alerting techniques are included in a larger metacognitive intervention to enhance EF (Levine et al., 2011).

A key mechanism in experience-dependent plasticity is the capacity to allocate processing resources selectively to a particular stimulus (Blake, Heiser, Caywood, & Merzenich, 2006; Recanzone, Schreiner, & Merzenich, 1993). This form of attention, however, is dependent on adequate levels of arousal (Coull, 1995; Smith & Nutt, 1996). Studies have demonstrated that arousal can be manipulated by both external and internal alerts; hence, one can improve sustained attention by voluntarily increasing arousal (Robertson, Mattingley, Rorden, & Driver, 1998; Robertson, Tegner, Tham, Lo, & Nimmo-Smith, 1995). This type of training has been addressed for patients with executive dysfunction in combination with a metacognitive strategy with which the temporary arousal could be linked and, hence, invoked periodically to produce enduring effects (Fish et al., 2007; Manly, Hawkins, Evans, Woldt, & Robertson, 2002). GMT promotes internalization of such prompts through training of a self-cueing process to aid in sustaining attentional control. In fact, a central procedure of GMT is to stop ongoing behavior periodically to monitor and adjust goals, an activity supporting the maintenance of goal-related information essential to managing the sequence of stages needed to accomplish the goal. In light of this framework, the role of attentional control, and especially sustained attention and inhibitory control, in supporting processes collectively referred to as executive, is crucial (Andres, 2003; Aron, 2007; Logan, Cowan, & Davis, 1984; O'Connor, Robertson, & Levine, 2011; Stuss & Alexander, 2007).

In everyday life, responses contiguous with features of performance contexts may oppose and displace higher order goals when the sustained attention system does not function optimally (Manly, Robertson, Galloway, & Hawkins, 1999; Reason, 1990; Robertson, Manly, Andrade, Baddeley, & Yiend, 1997). GMT attempts to prevent goal failure by raising awareness of attentional errors through the use of participants' real-life attention deficits, in-session practice on laboratory tasks of attention supplemented with periodic alertness cueing, in-session practice of complex real-life tasks, and homework assignments (Levine et al., 2011). There is also an emphasis on mindfulness-based attention strategies (Kabat-Zinn, 1990).

GMT and its modified versions have been shown beneficial in treating EF deficits across several groups, including acquired brain injury (Chen et al., 2011; Fish et al., 2007; Grant, Ponsford, & Bennett, 2012; Levine et al., 2000, 2011; McPherson, Kayes, & Weatherall, 2009; Novakovic-Agopian et al., 2011), normal aging (Levine et al., 2007; van Hooren et al., 2007), addiction (Alfonso, Caracuel, Delgado-Pastor, & Verdejo-Garcia, 2011), intensive care unit survivors (Jackson et al., 2011), and in case studies of patients with focal cerebellar damage (Schweizer et al., 2008), encephalitis (Levine et al., 2000), craniopharyngioma (Metzler-Baddeley & Jones, 2010), and schizophrenia (Levaux et al., 2012).

When GMT is effective, it is assumed to be a result of underlying alterations in brain networks supporting sustained attention (Chen et al., 2011). Reliable goal-directed behavior requires the capacity to sustain attention over time. Targeting attentional control may therefore lead to improvements in functioning that generalize to broader domains of goal-directed functioning. In fact, GMT is associated with reduced attentional lapses, increased behavioral consistency and improved performance on neuropsychological measures of attention and executive functions, including sustained attention and inhibition (Alfonso et al., 2011; Levine et al., 2011; Novakovic-Agopian et al., 2011). Significant effects in support of GMT were also found for performance on analogues of real-life tasks requiring complex attentional skills (Levine et al., 2000, 2007; Novakovic-Agopian et al., 2011) and in surveys of real-life executive deficits (Levine et al., 2007; van Hooren et al., 2007).

The present study is a randomized controlled trial (RCT) with one treatment group (GMT) and one wait-list control group (WL), and using a repeated-measures design across three time points (baseline, post-intervention, and 6-month follow-up). Experimental participants were assigned to 21 hr of GMT. The neuropsychological test measures included directly assessed attentional control processes targeted by GMT, as well as behaviors supported by sustained attention and inhibitory control and in turn affected by GMT, albeit not specifically trained. In exploring generalization (Burgess, Alderman, Evans, Emslie, & Wilson, 1998; Burgess, Veitch, de Lacy Costello, & Shallice, 2000; Manchester, Priestley, & Jackson, 2004; Wilson et al., 2009; Wilson, 2008) a functional “real-life” measure was also included. Additional data, from questionnaires of cognitive functioning, mental health, quality of life, and coping, will be reported elsewhere.

The main objectives of this study were to determine the efficacy of GMT as a group-based treatment program for patients with SBM and EF deficits. The current study addresses several areas with a lack of research knowledge. First, we believe that no studies have investigated the effect of cognitive rehabilitation of EF in patients with SBM. Our particular interest was in examining the effect of an intervention targeting processes of attentional control, specifically sustained attention and inhibition. Second, the evidence related to long-term effects of GMT is weak. In fact, only two group-based GMT studies (Levine et al., 2011; Novakovic-Agopian et al., 2011) have reported follow-up analyses more than three months post-intervention. No GMT studies other than the current one have evaluated treatment effects at 6-month follow-up. Finally, we are unaware of studies applying GMT over extended time periods. Earlier GMT studies have conducted trials with weekly sessions (e.g., Levine et al., 2011; van Hooren et al., 2007). However, as SBM is a rare disorder, participants had to be recruited from throughout Norway, necessitating coverage of the GMT modules during three 3-day inpatient intervention periods across a 3-month period. To examine the feasibility of this treatment-delivery method, we planned to monitor treatment compliance, as measured by completed GMT modules. We hypothesized post-intervention changes in scores to reflect improved sustained attention and inhibitory control: on Conners’ Continuous Performance Test II (CPT-II; Conners, 2000), reduction in omission and commission errors and increase in reaction time; on D-KEFS Tower Test (Delis, Kaplan, & Kramer, 2001), increase in mean time to first move and total achievement score, and reduction in rule violations; on D-KEFS Trail Making Test and Color-Word Interference Test (Delis et al., 2001), reduction in errors. On the task chosen to measure generalization, the Hotel Task (Manly et al., 2002), it was hypothesized that both time deviation scores would decrease following treatment and that the total number of tasks attempted would increase. Additionally, as patients were taught to use compensatory strategies autonomously, the long-term presence of treatment effects at follow-up was considered as even more important.

METHODS

Participants and Procedures

Figure 1 depicts the flowchart of study participants. All patients diagnosed with SBM (19–45 years) and registered in 2010 at TRS national resource center for rare disorders, Sunnaas Rehabilitation Hospital (Norway), were requested to participate ($n = 201$). The information letter specifically solicited participants with subjective complaints of executive dysfunction such as impaired planning, attention, multitasking, decision-making, and organization. Accompanying the letter was a self-report questionnaire, the Behavior Rating Inventory of Executive Function-Adult Version (BRIEF-A; Gioia, Isquith, Guy, & Kenworthy, 2000), which respondents were asked to complete and return. In addition to the reported difficulties from the information letter, inclusion of patients was also based upon an elevated score ($T > 60$) on at least one of the subscales constituting BRIEF-A. Criteria for exclusion, based on review of medical records and baseline measures, included impaired essential linguistic, perceptual, or motor function that would interfere with the capacity to participate in training. Additionally, patients with Axis I psychiatric disorders or IQ below 70 were excluded. Six subjects met exclusion criteria; four had IQ below 70, and two had Axis I disorders. Eight subjects who met inclusion criteria could not participate due to somatic illness and/or hospitalization, or educational requirements. A final sample of 38 subjects (58% female) ages 19–45 ($M = 32$; $SD = 8.3$) were included in the study (see Figure 1 for Consort diagram) (Schulz, Altman, & Moher, 2010).

Table 1 displays the sample demographic and medical characteristics. The study was approved by the Regional Ethic Committee for Medical Research Ethics (2009/2188b), South-Eastern Norway. All patients gave informed consent for participation. The research was completed in accordance with the Helsinki Declaration.

Figure 2 illustrates the randomization, assessment, and intervention/waiting list procedure. The randomization method was block design with block size 2, with stratification

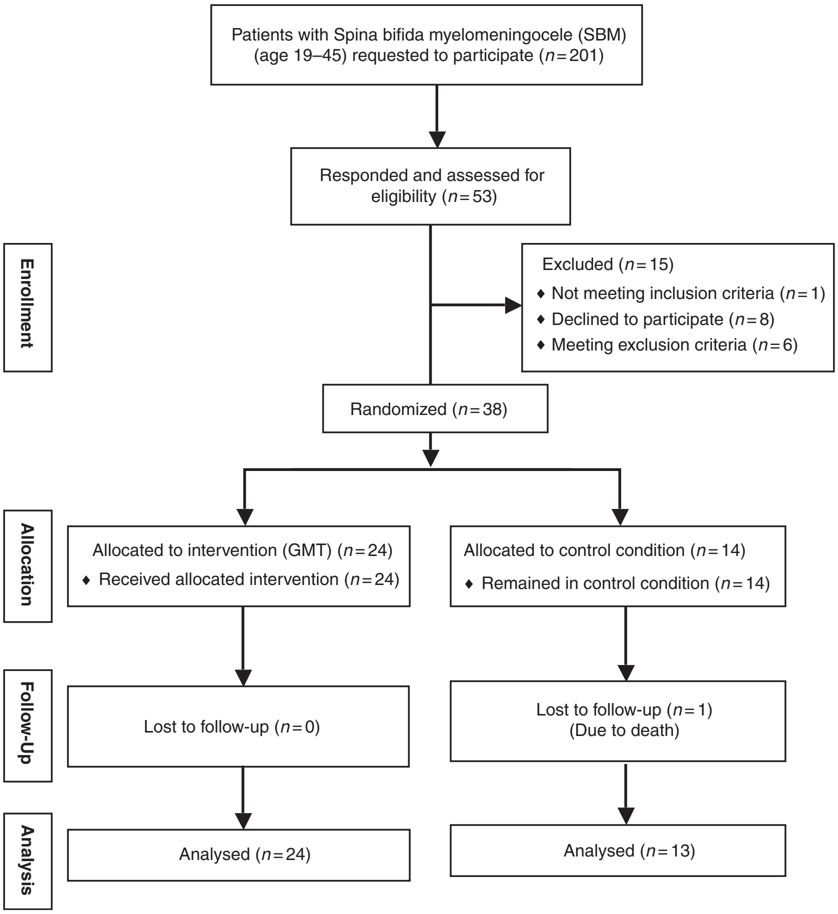

Fig. 1. Consort diagram.

for age (above/below 33 years) and education (above/below 12 years). An unequal randomization ratio of 2:1 was used to ensure maximum use of the available intervention and to gain experience of GMT (Dumville, Hahn, Miles, & Torgerson, 2006). The investigator responsible for randomization was not involved in the training procedures. The participants were informed about randomization outcome, and, if assigned to the WL group, told that they would receive GMT one year later. GMT consists of seven modules, with a minimum of 3 hr being necessary to complete each module (see Figure 2). A clinical neuropsychologist and a nurse/social worker conducted the training. None of the participants received any other intervention during the study period.

GMT was administered following a manualized protocol, also used by Levine and colleagues (2011), consisting of PowerPoint slides and participant workbooks. The GMT materials were translated into Norwegian, and back-translated to English by an independent translator, whose mother tongue was English and who had no previous knowledge of the materials.

Intervention

Training involved discussions and exercises intended to increase awareness of different features of goal management. Specifically, participants were trained to use strategies such as stopping and orienting to relevant information, partitioning goals into subgoals, encoding and retaining goals, monitoring performance (Levine et al., 2011), and mindfulness training (Kabat-Zinn, 1990). Complex training tasks used involved multitasking exercises (e.g., do five different tasks within a 4-min allotment; sorting cards, put dates of birth in order, connect the dots, word search in a grid, and spot differences between two pictures). Throughout the intervention, discussion of patients' real-life executive problems was encouraged, and application of GMT strategies to these difficulties and to the complex training tasks was reviewed. Assignments between sessions included monitoring, recording of absentminded slips and activities that went well, along with present-mindedness practice (see Table 2).

Baseline Instruments

In characterizing the cognitive functioning of the sample, the participants completed all subtests of the Wechsler Abbreviated Scale of Intelligence (Wechsler, 1999), letter-number sequencing and digit span from the Wechsler Adult Intelligence Scale III (Wechsler, 1997), the Brief Visuospatial Memory Test Revised (Benedict, 1997), and the California Verbal Learning Test II (Delis, Kaplan, Kramer, & Ober, 2000). The BRIEF-A (Gioia et al., 2000) was used as an inclusion instrument.

Table 1. Demographic and medical characteristics of both groups

	GMT (*n* = 24)	Control (*n* = 14)	Total (*n* = 38)	Significance
Age, mean ± *SD*	31.79 (8.38)	31.79 (8.50)	31.79 (8.31)	n.s.
Gender (M = men, F = female)	10 M, 14F	6 M, 8F	16 M, 22F	n.s.
Hydrocephalus *n* (%)	21 (88)	12 (86)	33 (87)	n.s.
– Shunt	20 (83)	11 (79)	31 (82)	n.s.
– 3rd ventroculostomy	1 (4)	3 (21)	4 (11)	n.s.
– >3 shunt revisions	8 (27)	7 (50)	15 (39)	n.s.
Arnold Chiari malformation *n* (%)	17 (71)	8 (57)	25 (66)	n.s.
Agenesis of the corpus callosum *n* (%)	2 (8)	2 (14)	4 (11)	n.s.
MMC level *n* (%)				
– Sacral	2 (8)	0	2 (5)	n.s.
– Lumbar	20 (83)	14 (100)	34 (89)	n.s.
– Thoracic	2 (8)	0	2 (5)	n.s.
Education, years ± *SD*	12.04 (1.71)	12.71 (1.90)	12.3 (1.78)	n.s.
– Primary *n* (%)	7 (29)	3 (21)	10 (26)	n.s.
– Upper secondary *n* (%)	15 (63)	8 (57)	23 (61)	n.s.
– Higher education *n* (%)	2 (8)	3 (21)	5 (13)	n.s.
Marital status (with partner) *n* (%)	4 (17)	3 (21)	7 (18)	n.s.
Paid work full time *n* (%)	2 (8)	1 (7)	3 (8)	n.s.
Living Situation *n* (%)				
– Living alone	12 (50)	8 (57)	20 (53)	n.s.
– With parents/siblings	7 (29)	1 (7)	8 (21)	n.s.
– Own family	3 (13)	3 (21)	6 (16)	n.s.
– Other	2 (8)	2 (14)	4 (11)	n.s.

Note. Percentage totals may not add to 100% due to rounding. Differences between groups were tested with Chi-square for dichotomous variables and *T*-tests for continuous variables.
GMT = Goal Management Training; MMC = myelomeningocele. n.s. = not significant.

Outcome Measures

The neuropsychological outcome measures included CPT-II (Conners, 2000), and D-KEFS subtests: Color-Word Interference Test (CWI), Trail Making Test (TMT), and Tower Test (Delis et al., 2001) (Table 3). Participants' performance in a cognitive domain commonly affected by SBM, but not targeted by the intervention (i.e., motor speed), was assessed as a marker of potential non-specific changes. In exploring generalization, the Hotel Task (Manly et al., 2002) was used. The Hotel Task has been demonstrated to have acceptable ecological validity, and it has proven to be sensitive in the detection of frontal dysfunction in various conditions (Roca et al., 2010, 2008; Torralva, Gleichgerrcht, Lischinsky, Roca, & Manes, 2012), even in the absence of deficits on standard cognitive tests (Gleichgerrcht, Torralva, Roca, & Manes, 2010).

Feasibility of treatment-delivery method

Participants' attendance was registered after each module.

Statistical Analyses

Data analyses were conducted using SPSS version 19.0 for Windows. Frequency distributions, means, and standard deviations (*SD*) were calculated for the demographic, medical, and neuropsychological performance variables. Differences between groups were analyzed using Chi-square for dichotomous variables and *t*-tests for continuous variables. A general linear model with repeated measures analysis of variance (RM ANOVA) was used to examine differential group treatment effects. Data were analyzed using a 2×3 mixed-design with Group (GMT, WL) as a between-subjects factor, and Session (baseline, post-intervention, follow-up) as a within-subjects factor, using a multivariate approach to avoid the more stringent univariate model assumptions. *T* tests were used to explore change scores (baseline to session 2, and baseline to session 3) within each group. Experiment-wise error was not corrected since measures were thought to reflect separate processes at different levels of ecological validity (Levine et al., 2011). The strength of experimental effects was interpreted with effect size statistics, including partial eta-squared for ANOVA results and eta-squared (η^2) for *t*-tests. According to Cohen (Cohen, 1988), thresholds for interpreting η^2 are less than .06 (small), .06 to .14 (medium), and greater than .14 (large). All tests were conducted with an alpha level of $P < .05$.

RESULTS

As seen in Figure 1, a total of 37 individuals with SBM were included in the 6-month follow-up analysis, with one person lost to follow-up due to death. Demographic and medical characteristics are summarized in Table 1, and neuropsychological data in Table 4. All subjects were Caucasian. Most of the participants had brain abnormalities (hydrocephalus, Chiari-II or agenesis of corpus callosum) and a lumbar-level

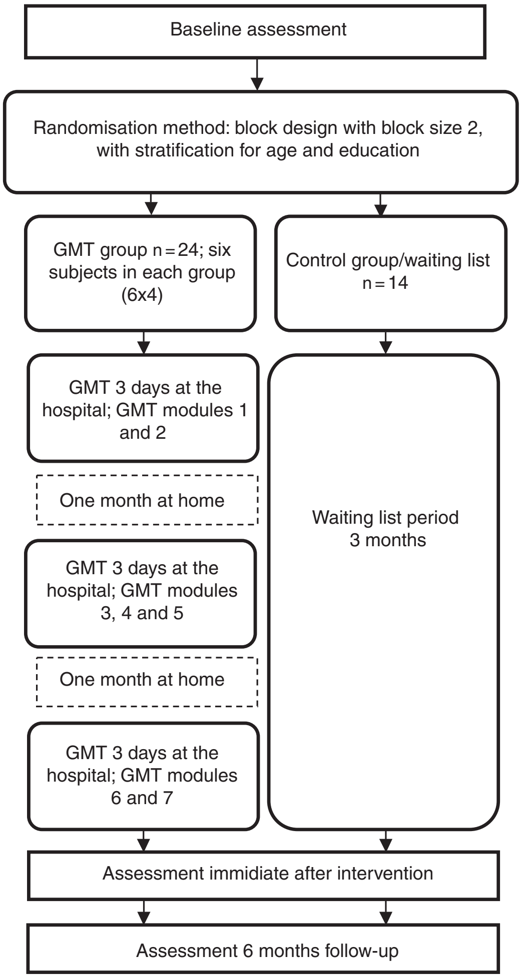

Fig. 2. Randomization, assessment, and intervention/waiting list procedure.

lesion. The majority of participants were female, had finished upper secondary school, and lived alone. A minority reported being with a partner, and very few were employed full-time. No significant differences in demographic or medical characteristics were found at baseline (Table 1).

Tables 4 and 5 demonstrate that treatment and control groups had comparable cognitive functioning (Table 4) and self-reported executive functioning (Table 5) at pre-treatment baseline assessment. Overall, both groups displayed impaired executive functioning relative to the standardization samples.

Feasibility of Treatment-Delivery Method

All study participants completed the seven GMT modules.

Effects of Treatment

Table 6 provides mean scores on cognitive outcome data by session for intervention and control groups, with session- and group-by-time effects. An examination of the pattern of scores across time and between groups revealed that the GMT group had greater gains over time than the control group. In addition, within the GMT group there were statistically significant improvements on all targeted outcome variables. Of note, effect-size estimates indicated overall large training effects ($>.14$) (Cohen, 1988). In the WL group, no significant changes were detected except for a significant retest effect on the Tower total achievement score.

Performance on CPT-II showed a significant reduction in responses given to non-target items across sessions that held at follow-up. The GMT group also demonstrated significantly less failures in responding to target letters at follow-up compared to baseline. Of interest, there was an increase in hit reaction time from baseline to 6-month follow-up that approached significance. In the Tower Test, both groups showed a significantly increased total achievement score across sessions. However, there was a significant increase in mean time used on the first move, and reduction on rule violations per item across sessions for the GMT group that held at follow-up. Furthermore, in both TMT and CWI, there was a significant reduction in errors across sessions that held at follow-up for the GMT group. These findings were evident in condition 4 in TMT and across all conditions (1–4) in CWI, in addition to conditions 3 and 4, in CWI. No significant differences in pre–post change were seen between the two groups for motor speed (TMT condition 5). In the Hotel Task (HT), there was a significant reduction in deviation from optimal time used on each subtask, an increase in number of tasks attempted, and a reduction in time deviation on the closing and opening of the garage door across sessions for the GMT that held at follow-up. In subsequent *post hoc* analyses, a linear regression analysis was used to derive a standardized residual score for each variable in CPT-II and HT, to represent change. These residuals were then correlated. Significant correlations (Pearson) were found between reduction in garage time deviation (HT) and reduction of commissions (CPT-II) ($r = .6$; $n = 23$; $P < .002$), and between reduction of total time deviation (HT) and reduction of omissions (CPT-II) ($r = .55$; $n = 23$; $P < .006$).

DISCUSSION

The main aim of this RCT was to evaluate the efficacy of GMT using inpatient intervention periods for patients with SBM and executive difficulties, with a 6-month follow-up. In terms of efficacy, participants showed significant differential improvement, compared to WL subjects, on neuropsychological measures of attentional control, including sustained attention and inhibitory control. These results were in line with the hypothesized cognitive targets of the intervention. GMT also was associated with improved performance on a desktop model of a "real-life" multitasking situation, suggesting generalization of intervention effects to functional performance in complex real-life settings. These findings suggest that strategies addressing the ability to plan activities and to structure intentions did improve after training. Participants appeared able to maintain all training gains at 6-month follow-up. Even though this sample was relatively

Table 2. Outline of the modules and objectives in GMT

GMT modules	Objectives/key concepts	Within-session exercises	Between-session assignments
1. The Absent Mind and Slip-ups	– Orientation to the GMT program – Defining absentmindedness, absentminded errors (slip-ups), and goals – Discussing and raising awareness of consequences of slip-ups	– Clapping task – Clapping task-revisited	– Record slips – Remember workbook
2. Stop the automatic pilot	– Defining the automatic pilot – Addressing automatic pilot errors. – Addressing how to stop the automatic pilot	– Card-dealing task – Clapping task with "STOP!" – Card-dealing task with "STOP!" by trainer – Card-dealing task with "STOP!" by participant	– Record Slips – 30-minute daily STOP
3. The mental blackboard and present-mindedness	– Introducing the concept of working memory as a mental blackboard – Introducing the "STOP!" technique (periodic suspension of ongoing behavior) to check the mental blackboard – Introducing a mindfulness-based meditation technique to acquire an ability of bringing one's mind to the present to monitor ongoing behavior, goal states, and the correspondence between them	– Card-dealing task with "STOP!" by participant – Card-dealing task with distraction	– Daily present-mindedness practice – Record slips/things that went well
4. State your goal	– Teaching to state goal following stopping, and present-mindedness as a way to activate goal representations "STOP!" (present-mindedness)-STATE cycle	– Complex task I – Complex task II	– Daily present-mindedness practice – Record slips/things that went well –30-min daily STOP-STATE
5. Making decisions	– Addressing competing goals – Understanding emotional reactions to conflicting goals – To-Do Lists in the "STOP"- STATE cycle – Combating indecision	– Complex task with to-do list	– Get to-do list – Daily present-mindedness practice – Catalogue task #1
6. Splitting tasks into subtasks	– Defining overwhelming tasks. – Defining tasks and subtasks (goal hierarchies) – Splitting the task up – "STOP!"- STATE - SPLIT cycle	– Wedding task	– Log STOP-STATE-SPLIT scenarios – Daily present-mindedness practice – Catalogue Tasks #2 & 3
7. Checking (STOP!)	– Recognizing "STOP!"-STATE-SPLIT errors – Using "STOP!" to monitor output/error correction – GMT review	– Clapping task with "STOP!"	

GMT = Goal Management Training.

small, effect size estimates indicated overall large training effects. Additionally, all 24 treatment subjects successfully completed GMT, suggesting that a treatment-delivery method with extended treatment periods is feasible for patients with SBM.

Attentional Control

GMT aims to promote a mindful approach to enhance EF by strengthening sustained attention to maintain awareness of goal states and output monitoring (Levine et al., 2000). Both

Table 3. Description of outcome measures, dependent variables, and cognitive functions assessed

Test	Description	Dependent variables	Cognitive function
CPT-II	Computerized test involving speeded responses to series of letters. Participants are instructed to respond to all letters except for X′s	Omission errors, commission errors, and reaction time	Sustained attention and inhibitory control
CWI	Words are printed in dissonant ink color and participants are instructed to name the color of the ink	Total errors in condition 3 (inhibition) and condition 4 (inhibition/switching), and total errors across all conditions	Inhibitory control
TMT 4	In the Number-Letter Switching condition, participants are instructed to switch back and forth between connecting numbers and letters (i.e., 1, A, 2, B, etc., to 16, P)	Total errors in condition 4 (number-letter switching)	Attentional control
TMT 5	In the Motor Speed condition the participants are instructed to trace over a dotted line connecting circles on the page as quickly as possible	Total time condition 5	Motor speed (marker of potential non-specific changes)
Tower	Participants are asked to construct towers of discs on a set of pegs corresponding to a model, with rules limiting the movement of the disks. For the purposes of this study, the standardized test was split into two forms by alternating items	Total time to first move, rule violations and total achievement score	Inhibitory control and processes supported by sustained attention
Hotel	It mimics a real-life multitasking situation in which the participant plays a hotel manager with five different tasks. Participants must distribute time equally across five tasks within a 15-minute allotment, and remember to open and close the garage doors at two predefined times	Number of tasks attempted, time allocation (total deviation from an optimal allocation of three minutes per task), and total garage-door time deviation	Planning and organization

Note. CPT-II = Conners Continuous Performance Test II; CWI = Color-Word Interference Test; TMT 4 = Trail Making Test condition 4; TMT 5 = Trail Making Test condition 5; Tower = Tower Test; Hotel = Hotel Task.

sustained attention and inhibitory control are considered to be central elements of attentional control (Andres, 2003; Aron, 2007; Miyake, Friedman, et al., 2000; Robertson & Garavan, 2004), and both are crucial in explaining behavioral flexibility and goal-directed behavior in a dynamic environment. In the current study, there were significant effects specific to the GMT group with a reduction in errors on measures of sustained attention and inhibition (i.e., TMT, CWI, CPT-II). The robustness and specificity of these findings were consistent with the theoretical assumption that GMT targets basic aspects of attentional control. Further supporting the hypothesized targets of training, no changes were found on a control measure of basic motor speed. The GMT group showed a significant reduction in commission errors (CPT-II) across sessions, perhaps reflecting increased ability to inhibit a habitual response. Levine and colleagues (2011) also found a reduction in commission errors on the Sustained Attention to Response Task following GMT. In Reason's (1990) studies of everyday behavior, habit intrusions were especially common when people were distracted or absentminded. This type of cue-dependent behavior or failure to maintain attentional control is more prominent in various clinical groups than control groups, and is related to reports of everyday lapses in goal-directed activity (Fish et al., 2007; Manly et al., 1999; Robertson et al., 1997; Shallice & Burgess, 1991).

Furthermore, there was a significant increase in the time before first move across sessions on the Tower Test following training. The major emphasis in GMT is to "Stop and Think" before acting. This would seem to correlate with the capacity for response inhibition observed in the Tower Test, indicating that subjects in the GMT group are slowing down (i.e., stopping) for the sake of greater reflection and accuracy. Indeed, this proposition is supported by our finding that the GMT group also demonstrated a significant reduction in rule violations on Tower compared to the control group. There was also a trend for an increase in reaction time for the GMT group on the CPT-II from baseline to 6-month follow-up. On the basis of these findings and the reduction of errors in other neuropsychological measures of executive functioning, it can

Table 4. Standardized neuropsychological scores at baseline

Neuropsychological tests (*M* ± *SD*)	GMT (*n* = 24)	Control (*n* = 14)	Significance
WASI FSIQ	93 (13.41)	89.1 (15.41)	n.s.
WASI VIQ	94.9 (13.7)	86.4 (17)	n.s.
WASI PIQ	91.5 (13.6)	92.6 (14.7)	n.s.
CVLT-II Total Score	34.3 (11.9)	36.8 (16.7)	n.s.
BVMT-R Total Score	31.7 (13.9)	29.2 (9.5)	n.s.
Letter-Number Sequencing (WAIS-III)	7.4 (2.8)	8.5 (3.3)	n.s.
Digit Span Total Score (WAIS-III)	8.1 (2.8)	8.9 (2.8)	n.s.
CPT-II Omissions	70.8 (56.4)	48.9 (7.9)	n.s.
CPT-II Commissions	61.7 (11.1)	59.6 (10.3)	n.s.
CPT-II Hit RT	45.2 (12.2)	45.6 (11.2)	n.s.
Tower Test Total Achievement Score	7.8 (3.9)	6.4 (4.5)	n.s.
Trail Making Test condition 4	6 (3.3)	5.9 (3.8)	n.s.
Color-Word Interference Test condition 3	5.7 (3.7)	7.4 (3.5)	n.s.
Color-Word Interference Test condition 4	5 (3.5)	5.5 (3.4)	n.s.

Note. All scores reported are standardized scores. Higher neuropsychological scores represent better performance, except for scores in CPT-II where *T* scores between 40 and 60 are in the normal range. Differences between groups were tested with two-tailed results of *t* tests.
WASI FSIQ = Wechsler Abbreviated Scale of Intelligence Full Scale Intelligence Quotient (*M* = 100, *SD* = 15); WASI VIQ = Wechsler Abbreviated Scale of Intelligence Verbal Intelligence Quotient (*M* = 100, *SD* = 15); WASI PIQ = Wechsler Abbreviated Scale of Intelligence Performance Intelligence Quotient (*M* = 100, *SD* = 15); CVLT-II = California Verbal Learning Test II (*M* = 50, *SD* = 10); BVMT-R = Brief Visuospatial Memory Test Revised (*M* = 50, *SD* = 10); WAIS-III = Wechsler Adult Intelligence Scale III (*M* = 10, *SD* = 3); CPT-II = Conners Continuous Performance Test II (*M* = 50, *SD* = 10); RT = reaction time; Subtests from D-KEFS (*M* = 10, *SD* = 3); n.s., not significant.

be argued that the GMT group showed improved inhibitory control following training.

A model from experimental psychology ("horse-race model") facilitates an interpretation of the performance on tasks of inhibition in the present study. This model asserts that the stopping and reaction processes compete for the first finishing time (Logan et al., 1984). If stopping processes finish before the reaction processes, the response is inhibited. Otherwise, the response escapes from inhibitory control. Thus, the extra time taken to respond observed on our study when two dimensions are incongruent can be attributed to the disabling of the incorrect response.

Omission errors occur when the subject fails to maintain an ongoing response, possibly reflecting attentional drift and reduced top-down control leading to pre-empted responses (O'Connell et al., 2009). Further supporting the training effect on sustained attention and inhibition was the reduction of omission errors in the GMT group, a significant effect only in the baseline–follow-up comparison. The appearance of this effect only after 6 months suggests that participants may have internalized strategy use for attentional control in the months following the intervention. Similar findings have also been found in a another GMT study (Levine et al., 2011), supporting our results.

Table 5. Norm-referenced scale means for BRIEF-A self report

BRIEF-A scales	GMT (*n* = 24) *M* (*SD*)	Control (*n* = 14) *M* (*SD*)	Significance
Behavioral regulation scales			
Inhibition	53.3 (9.6)	54 (10.9)	n.s.
Self-monitor	54.5 (11.7)	54.4 (14)	n.s.
Shift	63.4 (9.8)	63.2 (15.7)	n.s.
Emotional control	58 (11.2)	57.4 (13.8)	n.s.
Behavioral regulation index	58.8 (10.6)	58.6 (15.2)	n.s.
Metacognition scales			
Initiate	67.4 (12.4)	65.1 (10.3)	n.s.
Working memory	67.8 (11.3)	70.8 (10.4)	n.s.
Plan/organize	65 (11.3)	66 (12.4)	n.s.
Organization of materials	58.3 (11.1)	57.8 (11.9)	n.s.
Task-monitor	61.4 (8.8)	61.3 (10.4)	n.s.
Metacognition index	66.3 (9.8)	66.6 (9.3)	n.s.
Global executive composite	64.1 (9.8)	64.1 (12.1)	n.s.

Note. Scores listed are *T* scores (*M* = 50, *SD* = 10), with higher scores indicating greater impairment. Differences between groups were tested with two-tailed results of *t* tests.
n.s., not significant.

Table 6. Mean scores on outcome data by session for intervention group (GMT) and wait list control (WL) with session and group by time effects

		Group		Group by session and session effects	
Test	Assessment	GMT *M* (*SD*)	Control *M* (*SD*)	*F* (df) group by session effect	*F* (df) session effect
Hotel task		(*n* = 24)	(*n* = 13)		
Deviations from optimal time (s)	Baseline	669.46 (292.94)	672.62 (303.44)	9.04*** (2, 34)	12.58*** (2, 34)
	Post-intervention	323.83 (105.74)***	653.38 (266.96)		
	Follow-up	253.13 (129.83)***	630.92 (269.23)		
No. of tasks attempted	Baseline	3.67 (1.27)	3.69 (1.18)	3.88* (2, 34)	8.64*** (2, 34)
	Post-intervention	4.92 (0.28)***	3.92 (1.19)		
	Follow-up	5 (0)***	4 (0.91)		
Time deviation garage (s)	Baseline	345 (448.39)	410.77 (442.84)	0.58 (2, 34)	4.87* (2, 34)
	Post-intervention	152.5 (317.48)**	327.69 (458.01)		
	Follow-up	92.75 (200.82)**	189.69 (292.44)		
CPT-II		(*n* = 23)	(*n* = 13)		
Commission errors	Baseline	19.57 (7.43)	18.54 (7.72)	6.98** (2, 33)	20.05*** (2, 33)
	Post-intervention	14.26 (8.76)***	16.23 (10.09)		
	Follow-up	7.09 (3.16)***	15.46 (9.77)		
Omission errors	Baseline	8.17 (15.37)	1.9 (3.07)	2.24 (2, 33)	3.43* (2, 33)
	Post-intervention	3.74 (5.22)	4.31 (7.45)		
	Follow-up	1.43 (1.67)*	2.15 (3)		
Hit RT (ms)	Baseline	372.1 (65.07)	371.9 (63.55)	2.29 (2, 33)	0.449 (2, 33)
	Post-intervention	373.11 (62.68)	369.22 (73.37)		
	Follow-up	392.06 (55.96)	362.75 (61.89)		
Tower Test		(*n* = 24)	(*n* = 13)		
Total achievement score	Baseline	13.54 (5.72)	11.92 (6.69)	0.62 (2, 34)	21.27*** (2, 34)
	Post-intervention	17.17 (4.64)	13.92 (4.27)		
	Follow-up	20.42 (4.84)***	17.23 (5.51)***		
Mean time 1st move (s)	Baseline	5.21 (4.4)	5.38 (4.96)	6.77** (2, 34)	3.92* (2, 34)
	Post-intervention	10.51 (4.93)***	4.55 (2.26)		
	Follow-up	11.10 (3.16)***	4.72 (1.97)		
Rule violation per item ratio	Baseline	.29 (.43)	.22 (.41)	2.09 (2, 34)	1.64 (2, 34)
	Post-intervention	.10 (.18)**	.24 (.31)		
	Follow-up	.05 (.14)**	.21 (.54)		
Trail Making Test		(*n* = 24)	(*n* = 13)		
Total errors condition 4	Baseline	2.58 (4.21)	.85 (1.14)	1.63 (2, 34)	4.74* (2, 34)
	Post-intervention	1.25 (2.44)**	.46 (.66)		
	Follow-up	.38 (.58)*	.77 (.73)		
Motor speed condition 5 (s)	Baseline	37.96	33.69	0.97 (2, 34)	1.2 (2, 34)
	Post-intervention	33.79	32.08		
	Follow-up	33.63	33.31		
Color-Word Test		(*n* = 24)	(*n* = 13)		
Total errors all conditions	Baseline	5.04 (5.34)	4.15 (3.89)	3.68* (2, 34)	1.33 (2, 34)
	Post-intervention	3.29 (4.44)**	3.69 (4.09)		
	Follow-up	1.67 (1.63)***	5.38 (6.08)		
Total errors in conditions 3 and 4	Baseline	4.42 (5.28)	3.46 (3.8)	3.25 (2, 34)	0.92 (2, 34)
	Post-intervention	2.83 (3.90)**	3.38 (3.52)		
	Follow-up	1.58 (1.56)**	5 (6.01)		

Note. All scores reported are raw scores. Time is reported in seconds (Hotel Task, Trail Making Test) and milliseconds (CPT-II). Significant effects in comparison to baseline *$P < .05$; **$P < .01$; ***$P < .001$. All *F-tests* use the Wilks' lambda statistic *$P < .05$; **$P < .01$; ***$P < .001$. *N*'s are provided as data were missing for certain measures.
S = seconds; ms = milliseconds; CPT-II = Conners' Continuous Performance Test II.

The desired goal of cognitive rehabilitation is that individuals will exhibit learned approaches to task completion and application of strategies in situations with similar task demands. Because outcome measures were chosen specifically to detect the learning of expected theory-bound behaviors, there was naturally an overlap between test demands. Therefore, although the measures used were not specifically practiced as part of GMT, it was expected, and seemed borne out, that participants were able to transfer use of strategies to these situations.

Generalization

In a similar vein, a major concern for any rehabilitation study is generalization to situations and activities not specifically addressed by the intervention, yet appropriate for application of learning strategy use (Wilson et al., 2009; Wilson, 2008). GMT is designed to promote generalization to everyday functioning. As such, the most optimal transfer of training is to the daily life of participants. However, the nature of neuropsychological tests is that they assess cognitive domains rather than the functional capabilities required in the execution of daily activities (Burgess et al., 1998, 2006; Manchester et al., 2004). Nevertheless, in the present study, GMT effects were found on basic cognitive domains necessary for complex behavior in daily life (Aron, 2007; O'Connor et al., 2011; Stuss & Alexander, 2007).

A test that is relevant to the concept of generalization is the Hotel Task (Manly et al., 2002), included as an outcome measure to examine executive functioning in a complex real-life simulation context (Shallice & Burgess, 1991). Successful performance of the Hotel Task requires individuals to develop a task-performance plan, monitor ongoing behavior, and keep track of time. In the present study, improved attentional control (specifically sustained attention and inhibitory control) was associated with improved performance on a "real-life" multitasking situation, supporting the proposal that targeting attentional control may lead to improvements in functioning that generalize to broader domains of executive functioning. Although more conjectural, the *post hoc* analyses suggest that attentional control may have a specific impact in decisions about time allocation and estimation when one is faced with multiple tasks to perform. Previous group studies using GMT techniques have demonstrated positive effects on real-life analogue tasks, consistent with our findings (Levine et al., 2000, 2007; Miotto et al., 2009; Novakovic-Agopian et al., 2011). Yet, in contrast to the positive results found in our study, Levine and colleagues (2011), using this same "real-life" simulation task, found that patients with frontal lobe dysfunction who were treated with GMT distributed their time less consistently across tasks at post-training as compared to baseline. It is possible that the longer period of intervention in our study contributed to more opportunities to exercise and practice GMT techniques in everyday life.

The findings from this study also add support to studies (e.g., Levine et al., 2000., 2011; Miotto et al., 2009; Novakovic-Agopian et al., 2011; Rath et al., 2003; Spikman et al., 2010; von Cramon et al., 1991) that have incorporated problem solving and goal management training in ameliorating executive deficits. We were able to extend this evidence by demonstrating that effects can also be achieved when using inpatient intervention periods for patients with SBM, with effects lasting at least 6 months post-treatment.

Although the content of GMT has evolved considerably since it was initially conceived (Robertson, 1996), stopping to attend to goal hierarchies is fundamental to the protocol and precedes the remaining problem solving elements. Whereas the problem solving stages are similar to components of problem solving therapy (Miotto et al., 2009; Rath et al., 2003; von Cramon et al., 1991), GMT is less focused on making decisions about how to solve a problem and more focused on suspension of ongoing behavior to determine *which* problems should be solved. Indeed, the element of sustained attention runs continuously through GMT, and is reinforced through mindfulness training.

Limitations and Future Directions

A limitation of the current study was that the persons who carried out the assessments post-treatment were not blind to group membership. It is also important to note that the inclusion criteria for this study included self-reported executive functioning deficits rather than objective test performance as these tests are of limited utility in the assessment of real-life executive deficits of interest in this study. Furthermore, non-specific effects such as professional attention or group dynamics of the intervention cannot be ruled out as contributing to the results without having an active control group. Also, the present findings need to be cross-validated in a larger and more representative SBM sample considering the relatively small sample size. Finally, additional outcome measures are recommended to explore what the nature of the improvement of executive functioning in "real-world" activities that the intervention promotes.

CONCLUSIONS

This is the first study published on cognitive rehabilitation of executive dysfunction in patients with SBM. The study reports the successful use of a focused executive-rehabilitation program in SBM patients with executive dysfunction, given that GMT led to significant treatment effects on domain-specific neuropsychological measures as well as a functional measure, with effects lasting at least 6 months post-treatment. These data show that executive deficits can be ameliorated even in patients with congenital brain dysfunction.

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