Goal Management Training for rehabilitation of executive functions: a systematic review of effectiveness in patients with acquired brain injury.

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Introduction

The inability to be organized and to develop efficient strategies for completing everyday tasks is one of the most common and persistent sequelae following acquired brain injury. Such difficulties impede patients' ability to function independently in daily life. They relate to executive functioning impairment, arising from damage to the frontal lobes or to circuits that include frontal structures [1]. The term executive functioning (EF) refers to those integrative cognitive processes that support goal-directed and purposeful behavior and that are necessary to the orderly execution of daily life activities [2]. These integrative functions include the ability to formulate goals; to initiate behavior; to anticipate the consequences of actions; to plan and organize behavior; to monitor and to adapt behavior to fit a particular task or context.

The challenge for cognitive rehabilitation and particularly for EF interventions is to improve the ability to participate in meaningful activities through transfer and generalization. For example, if a stepwise strategy was trained in multitasking involving paper-and-pencil exercises at the rehabilitation center, it is hoped that the patient will use it in daily life tasks such as preparing a meal (generalization) at home (transfer).

Presently, metacognitive approaches appear to have the best level of evidence in relation to improving EF [3][4][5]. Metacognition (or "thinking about your thinking") includes self-awareness, self-monitoring and self-control of cognition while performing an activity [5]. Metacognitive approaches have negligible effect on underlying impairments compared to control interventions, but when activities and participation are the primary outcome measures, patients receiving metacognitive training improve

significantly more than controls [5]. Rehabilitation teams are therefore eager to administer metacognitive training and face the question of how and which form of metacognitive training to use.

One of the best known and most extensively studied metacognitive approaches is Goal Management Training (GMT). GMT is a theoretically derived intervention for executive dysfunction intended to promote a mindful approach to completing complex everyday activities by raising awareness of attentional lapses and reinstating cognitive control when behavior becomes incompatible with intended goals [6]. GMT rehabilitation comprises self-instruction strategies, self-monitoring exercises, cognitive techniques aimed at improving planning, prospective memory and cognitive control, mindfulness practice exercises, stories promoting discussion about executive dysfunction in daily life, and homework assignments to practice GMT ([6] for more detailed description).

Levine et al. [7] note that GMT was initially based on Duncan's theory of "goal neglect" [8][9][10] which suggests that much of human behavior is controlled by goal lists and subgoals. Dysexecutive patients are impaired in the construction and use of such goal lists, resulting in disorganized behavior. Later authors [6] developed this theory further and referred to "sustained and vigilant attention theory" [11] as the theoretical model of GMT. Ongoing activation of the right frontal-thalamic-parietal sustained attention system is required to actively, endogenously maintain higher order goal states in working memory. When the sustained attention system is compromised, habits or environmental triggers may oppose and displace higher order goals, resulting in cue-dependent or distracted behavior. In GMT, easy-to-grasp analogies are used to help patients understand these models, such as "checking main goal in the mental blackboard" to avoid "automatic pilot behavior" [12].

GMT was one of the first EF interventions tested in a randomized controlled trial (RCT) [7]. However, the intervention consisted of a single hour of instructions, which limits the translation of its findings in terms of clinical application and effectiveness [13]). GMT was then developed further, different versions were published, ranging from a single session [7] to a 14-hour GMT program ready for group rehabilitation in clinical settings [6]. Cueing became a key feature of GMT to remind patients of their goals [14][15][16] and trigger GMT strategy use [17].

It is not clear what defines GMT precisely as the GMT theoretical model has evolved and many different GMT interventions have been published, each using different outcome measures : attention

impairment [6], prospective memory performance [15], multitasking ability [14], daily life activities such as cooking [7]. GMT use has even extended to non-brain injured patients [18] [19] [20] [21]. This makes it more difficult to specify the “active ingredients”[22] of GMT programs and to summarize on what it is effective and for whom.

Nevertheless, GMT is manualized [12] and uses detailed PowerPoint slides supporting group sessions, workbooks and easy-to-find materials. It is therefore easy to use and easily reproducible for clinicians who have little experience in metacognitive training. As such rehabilitation teams may want use it to satisfy Cicerone’s practice standard of metacognitive training for EF rehabilitation[3]. However, there is limited evidence of superior effectiveness of GMT compared to other metacognitive training formats [5]. Recent literature suggest that interventions using GMT combined with other training methods are more effective than GMT-alone interventions [23][24][25]. Furthermore it remains unclear if interventions using GMT aim at improving impairments (like the diminution of attentional lapses as suggested by GMT authors [6]) or participation. Finally, not much is known about the best dose of treatment and for which patients GMT is more effective.

Kennedy [5] provided a very complete review of metacognitive rehabilitation studies through 2004, including 2 early GMT papers [7][14] but did not conclude as to how and which form of metacognitive training to use. Since the period covered by Kennedy’s review, GMT has been the subject of ongoing research and it seems timely to review that research to determine whether GMT is useful and if so, in what form and for whom. Finally, there is no review to our knowledge that focuses on participation and ecological outcome measures as criteria for intervention effectiveness.

The primary aim of this paper was therefore to review the evidence relating to the effectiveness of interventions using GMT. The question as to whether GMT improves executive function impairments and/or increases participation in everyday activities in patients with a dysexecutive syndrome following acquired brain injury was examined. Secondary aims were to determine the best treatment dose, best candidates for treatment and best delivery format, comparing evidence relating to the effectiveness of GMT delivered alone with GMT delivered in combination with other training methods/principles.

Methods

Studies published up to December 2011 were sought from : Ovid MEDLINE, PUBMED, PsycINFO, ERIC, PROQUEST by searching the following items: (1) (brain inj* OR head inj* OR traumatic head inj* OR traumatic brain inj* OR stroke OR encephalitis) (2) (intervention* OR rehabilit* OR train* OR therapy) (3) (executive OR prospective memory OR metacognitive OR cognitive) (4) (goal management training OR goal neglect OR problem solving). The symbol * denotes database operators, which include truncations or possible extra letters in the term to be included within the search. These four searches were then combined using 'AND'.

Inclusion criteria were (1) interventions using any version of Goal Management Training (GMT), entirely or partly, alone or in combination with another intervention OR Interventions acknowledging their link with Duncan's theoretical model of goal neglect, (2) interventions conducted with brain injury patients, irrespective of type of injury or age, (3) group studies and single-case experimental design (SCED) studies or single case reports, (4) peer-reviewed journals, (5) articles written in English, French or Polish. Exclusion criteria were (1) Theoretical articles or description of programs with no specific intervention, (2) feasibility studies not including effectiveness data on EF (3) review articles, (4) articles that did not include participants with brain lesions.

Four-hundred-and-twenty-nine papers were obtained from the computerized database searches. 359 were excluded on title alone, leaving 70 abstracts. A further 33 were excluded based on abstracts, and 18 on reading the intervention content in the full paper, leaving 19 articles referring to GMT in their description of the intervention studied. On reading the full 19 original papers, a further seven were excluded for the following reasons: four papers used GMT in non brain injury samples: drug addicts, elderly, healthy adults [21][19][26][27]. McPherson's feasibility study [28] used GMT for goal setting, without including effectiveness data on EF. Two studies of metacomponential approach [29] [30] for problem-solving in adults [31] and children [32] with ABI, referred once to Duncan's goal neglect model but as a very minor part of the program, without sufficient explanation of its use. Twelve articles published up to December 2011 remained for review.

Classifying studies: GMT proof-of-principle study versus GMT rehabilitation study

Included studies were classified according to the initial aims of the study:

-Four “Proof-of-principle studies” aimed at testing the potential effectiveness of a GMT principle or model. These studies typically used a single session of the GMT component being tested, without extensive training and without examining the long term effects or effects on participation in everyday activities.

-Eight “Rehabilitation studies” aimed at testing GMT as a full multi-session rehabilitation intervention, assessing long-term outcomes, measuring effects on participation in everyday activities, generalization and potential maintainance of effects.

Methodological appraisal of included studies

The criteria for the appraisal of articles were based primarily on the Consolidated Standards of Reporting Trials (CONSORT) guidelines [33] but extended, similarly to the methodology of Ross’s [34] systematic review on children’s cognitive rehabilitation. Items that are important in cognitive rehabilitation were added (e.g.: evidence of generalization of effects to untrained tasks, provision of data on brain injury severity, evidence of controlling for intervention duration). Furthermore, the nature of outcome measures used in each study was appraised with greater weight being given to studies that examined the impact of interventions on novel and ecologically relevant tasks.

The other six CONSORT items were applicable only for group studies (exclusion criteria, power calculation, intention-to-treat analysis) whereas the review also included single-case studies, describing interesting GMT variations [17]. Therefore, the six CONSORT items that were not applicable to single-case studies were replaced by items from the Single-Case Experimental Design (SCED) rating scale [35] (www.psycbite.com). The SCED scale is used for methodological appraisal of papers reporting interventions with single, or small N methods [35][36]. In SCED, causality is obtained by sufficient sampling at baseline (minimum 3 baseline assessments prior to intervention) and during intervention (sufficient sampling to differentiate a treatment response from fluctuations in behavior that may have occurred at baseline) and replication of the findings on another patient.

As a result, nine questions based on CONSORT and 10 cognitive and EF rehabilitation-specific questions were applied to all studies (see table 1). Additional questions were applied according to study methodology: six SCED-specific questions were applied only to single-case studies and six additional CONSORT questions were applied only to group studies. Finally, each study was assessed

on 25 questions, reported in table 1. With this rating method, well designed SCEDs could receive good ratings regardless of small number of patients.

Each item was awarded a score of 1 (if the criterion was met) and 0 if not met or if it was not possible to determine from information given in the article. Papers that met 75% of the methodological criteria were considered 'high' quality; those that were rated 50-74% were considered 'moderate' quality and those achieving less than 50% were 'lower' quality. When a single study was published in two papers[23][37], it was assessed only once, conversely when a paper included two distinct studies it was assessed twice [7]. Two reviewers rated the final 13 papers. Individual disagreements were resolved by discussion between the reviewers.

Table 6.1: Methodological Quality appraisal of papers

Score 1 if met, 0 if unmet or unable to determine

<u>CONSORT questions applied to all included studies</u>	<u>Cognitive and EF rehabilitation-specific questions applied to all included studies</u>
<ol style="list-style-type: none"> 1. Were specific hypotheses and/or objectives stated? 2. Were the settings and locations where data was collected stated? 3. Was there completely defined pre-specified primary and secondary outcome measures? 4. Were those assessing outcome blind to the treatment? 5. Was the intervention described in detail? 6. Were the characteristics of the patients clearly described? 7. Did the results relate to the initial hypotheses? 8. Was statistical analysis appropriate? 9. Were data adequately described (mean, range)? 	<ol style="list-style-type: none"> 1. Did the article specify the severity of brain injury and was the method of diagnosis appropriate? 2. Did the injury occur at least 6 month ago (to ensure that effects were not a reflection of the recovery process)? 3. Was there some kind of control for program intensity (to see if effect is specific of therapy or rather due to general cognitive stimulation)? 4. Were main outcome measures untrained tasks (ie a testing tasks not having the same structure as tasks used during training)? 5. Was generalization evaluated (to see if EF intervention effect generalize to other cognitive functions)? 6. Were outcome measures ecologically valid (i.e. ecological tests like "multiple errands" tasks rather than improved neuropsychological paper and pencil tests)? 7. Were effects observed in daily life activities (improved participation)? 8. Did at least one of the EF tasks administered post intervention bear a novelty aspect challenging patient's EF in a non routine situation (i.e. not the same task as pre-intervention as practice effect may decrease pressure on EF)? 9. Were at least some of the measures standardized assessment tools? 10. Were follow-up data collected at least 3 months after intervention (to see if effects were maintained post intervention)?

<u>CONSORT question applied only to group studies</u>	<u>Single-case experimental design (SCED) ³⁴³ questions applied only to single case studies</u>
<ol style="list-style-type: none"> 1. Was a power or sample size justified? 2. Were the inclusion/exclusion criteria clearly stated? 3. Was there a control or comparison group used? 4. Were patients randomly allocated to groups? (1 also for well designed pseudo-random allocations) 5. Were all patients included in the analysis? 6. Was intention-to-treat analysis used if randomized (0 for non randomized)? 	<ol style="list-style-type: none"> 1. Was there a clearly defined target behavior that the reflected cognitive function the intervention aimed at improving? 2. Were sufficient baseline assessments conducted to ensure stability prior to intervention? 3. Was there sufficient sampling during intervention to differentiate a treatment response from fluctuations in behavior that may have occurred at baseline? 4. Was replication performed: (study on 2 patients at least)? 5. Was Inter-rater reliability of the target behavior used in baseline and intervention assessed? 6. Did the design allow examination of cause and effect?

Calculation of effect size (ES)

ES were calculated as a standard difference between means, using Hedge's g [38] adapted by Morris and Deshon [39]. Their approach has been used in important cognitive intervention reviews [4][34] and was used here as the preferred procedure for ES calculation. Where scores were reported as Z-scores, without raw data from pre to post intervention [7][23], effect size was calculated by subtracting control Z from intervention group Z, assuming pretest standard deviation (SD) were used for Z-scores.

Equation 1 is the formula used for calculating ES in single group pretest-posttest designs:

$$ES = (M_{\text{post,exp}} - M_{\text{pre,exp}}) / SD_{\text{pre,exp}}$$

Equation 2 is the formula used for calculating ES in independent group pretest-posttest designs:

$$ES = [(M_{\text{post,exp}} - M_{\text{pre,exp}}) / SD_{\text{pre,exp}}] - [(M_{\text{post,com}} - M_{\text{pre,com}}) / SD_{\text{pre,com}}]$$

Where M is the mean, exp is the experimental group, com is the comparison group, $post$ is the posttests, pre is the pretest and SD is the standard deviation.

ES was interpreted subjectively by Cohen's rating [40]: $g < 0,20$ is a small ES; $g = 0,20-0,50$ is a medium ES; $g > 0,80$ is a large ES.

Results

The review included twelve studies. Four studies were “Proof-of-principle” studies, testing the potential effectiveness of a GMT principle or model and nine were Rehabilitation studies. One proof-of-principle study [7] tested GMT’s “STOP-STATE your goal-SPLIT goal into subtasks-CHECK” cycle. Two proof-of-principle studies [14][16] tested the effect of content-free cueing on goal management and one study [15] examined both approaches (content-free cueing triggering GMT use in a daily life context). Eight studies were rehabilitation studies: one group study [6] and three case studies [7][17][41] tested GMT alone, whereas the other four studies [24][25][23][37][42] tested comprehensive EF interventions that relied heavily on GMT but that also included other approaches described in the literature for EF rehabilitation, which are not addressed by GMT. These include Problem Solving Therapy, Mindfulness practice, personal goal training and ecological activities. Table 2 gives a description of the interventions, of the methodology used and the methodological quality rating. Most samples included mixed injuries, mainly traumatic brain injuries, frontal tumors and strokes. Severity of injury was reported for most TBI patients and ranged from mild to extremely severe, but using different severity classifications. No classification could be used for severity classification of other types of ABI.

Insert Table 2 about here

How are GMT interventions administered?

Interventions testing GMT component principles were either as short as a single sentence before starting the test [14][16] (“you might find these periodic beeps useful in reminding you to think about what you are currently doing and your overall aims during the session”) or of short durations such as 1 hour[7] or 30 min[15].

Interventions testing rehabilitation ranged from 5 hours [7] to 43 hours training [23] and were administered between once [6][24] and two-three times a week [23][25]. Most studies were administered as individual sessions [7][15][17]. Apart from three studies [6][24][41], all interventions included some individual sessions [37][23] or some adjustment of the program to the personal needs of each patient [25].

Personal between-sessions homework is a key component of GMT rehabilitation and was used in most rehabilitation studies using GMT. Patients were instructed to apply at home the goal

management strategies they had learned during the session [25][6][23][17]. Homework included monitoring EF failures in daily life called “Slips”[6], identifying factors that helped or not in everyday goal management performance [25][7], listing occasions on which participants used GMT strategies during the week [6], mindfulness practice assisted by an audio CD [37][23].

Are GMT principles effective?

The “STOP-STATE your goal-SPLIT task into sub-goals-CHECK” cycle was tested after a one-hour training in Levine et al.’s RCT [7]. The “everyday tasks” used as outcome measures were paper-and-pencil tests : for example, proofreading consisted of underlining, circling and crossing out words that met certain criteria (e.g.: circle all numbers). The intervention group made less errors in two out of the three “everyday tasks” after intervention, but needed more time to finish the tasks. The outcome was examined just one hour after the session, so that effectiveness and generalizability to daily life activities of this GMT principle cannot be affirmed. Furthermore, patients practiced two of these tasks during the intervention session.

The Content-free cueing principle has been tested in three studies. In two studies [14][16], auditory alerts during a complex multi tasking activity reminded patients about their current goal. In Manly et al.’s Hotel task [14] patients had to do some of each of five “hotel employee” tasks (sorting conference badges, compiling bills, looking up telephone numbers...) as well as a time-related prospective memory task (opening and closing garage door). Sweeney used a virtual reality “Removal task” [16] where the patient had to move around a storage unit selecting furniture as well as performing prospective memory tasks (visit the front door every five minutes, shut a particular door each time, label some furniture as “fragile”). In both studies, periodic alerts were introduced to the patients as a possibly useful way to remind them their overall goals but there was no training to ensure that patients linked the auditory cue with reviewing their goal. Conversely, Fish et al. [15] combined content-free cueing with a 30-minutes GMT to help patients associate the word “STOP” with reviewing goals. Text messages reading “STOP” were sent to participants eight times throughout the day to remind them of their prospective memory goal (to make a phone call at a certain time). In all three studies the cues were not specific cues to trigger task switch (i.e. changing to another task or making the phone call immediately) as cues never occurred at a time when task switch would produce an optimal performance. Content-free cueing was effective on reminding patients of their goal and increasing task performance in Manly et al. [14], but not in Sweeney et al. [16]. Fish et al. [15], who combined text

alerts with brief GMT found evidence that the intervention was effective with a moderate to large ES. Overall, evidence is contradictory (no effect to large effect).

Is GMT rehabilitation alone effective?

Levine et al. [6] report some positive results after a seven-week GMT training on the Sustained Attention to Response Task (SART) test (a go/no go test consisting of withholding of key press to one of nine targets) [43]. However this test is very close in format to one of the GMT training exercises that involves clapping to all targets (fruit) apart from one (pears) and thus reflects a near transfer of a learned skill rather than an effect on EF in daily life. There was a positive effect of GMT on the “Tower Test” which measures visual spatial planning: disks must be placed on dowels to match models of increasing complexity, with « rules » constraining the movement of the disk. The GMT group made significantly less rule violations on the tower test compared to the control group. However, there was no effect on self-rated Dysexecutive (DEX) Questionnaires relating to executive complaints in daily life in the GMT group and a statistically significant *decrease* in performance for the GMT group on the Hotel Task immediately after intervention when compared to controls. Proxy-rated DEX questionnaires were not obtained in this study and the authors suggested that stability on the DEX may reflect an increase of patients’ insight.

Two case-studies of low quality [7] [41] make it difficult to examine cause-effect of GMT, as patients started the GMT rehabilitation five and four and a half month after their brain injury (encephalitis and cerebellum hemorrhage) and there was no multiple baseline collected to ensure that the results observed were not due to spontaneous recovery.

What are the ingredients of effective comprehensive EF interventions using GMT?

Comprehensive EF interventions appeared to use one or more of the following elements in addition to GMT: (1) Problem Solving Therapy; (2) Ecological activities to promote transfer (3) Goal Setting; (4) Initiation facilitation.

Problem-Solving Therapy (PST) was developed by Von Cramon for enabling patients to be more effective in breaking down problems, adopting a slowed down, controlled and stepwise processing approach in contrast to more impulsive approaches. The therapy was effective in office-set problem solving but there was no evidence on generalization to everyday functioning [44][45]. Nevertheless,

PST principles are easy to integrate to the GMT “STOP-STATE the goal-SPLIT into subgoals cycle”. Before planning steps, patients are instructed to engage in the problem solving process of brainstorming for potential other solutions or means to achieve a current goal. PST was used in three studies [24][46][23][37] with good results.

Ecological and daily life activities using GMT to promote transfer

The necessity to include ecological and meaningful activities when training EF has been outlined and tested by many authors [47][48][49][50]. In the articles reviewed, cooking activities were used by Levine [7] and Pachalska [42] as cooking is a complex, open-ended, ill-structured multi-tasking activity requiring EF performance [51][52][53]. Levine’s patient reported less difficulties in cooking and examiners observed less problematic behavior during meal preparation but generalization to other tasks was not reliably assessed. Other ecological activities included bringing refreshments for the rest of the group following a budget and practicing stepwise goal management strategies to functional tasks of their choice [23][37]. GMT principles [7] [14] were combined ecologically and applied with success in a rehabilitation case-study by Metzler- Baddley [17]: a female with a craniopharyngioma was trained to use naturally occurring distractions at work (instead of the auditory alerts she was first trained on) as triggers to use GMT and refocus on present goal through the GMT “STOP-STATE-SPLIT” cycle. The patient successfully returned to work using this technique. Similarly in Pachalska’s study [42], applying the learned strategies to one’s own professional activity was the final stage of a holistic program involving GMT.

Goal-setting is a well known approach to increasing goal achievement in rehabilitation and other settings [54]. Although causality cannot be determined, in the interventions showing the best results on participation assessment tools, participants were asked to choose personal goals to be achieved by the end of the intervention. Patients were trained to set goals realistically by practicing short-term goal setting in individual projects [25][7] (planning a meal, learning to use an organizer) and group projects [37][23]. In Spikman’s study [25], control patients also had to set realistic goals for themselves even though the control intervention provided no specific training to achieve them (repeated computer exercises from CogPack). Controls progressed significantly towards their goals so that no significant effect was detected between intervention and control groups on Goal Attainment. This shows perhaps how powerful goal setting can be for enhancing motivation in rehabilitation and boosting effectiveness

of an intervention on personal goals, irrespective of the intervention. It is worth mentioning McPherson's study, although it did not meet the inclusion criteria of this review: McPherson [28] used GMT as a tool of informed goal setting and goal attainment in patients with TBI. GMT appeared "particularly helpful in providing a structured framework for error prevention in attempting goal performance"[28], and all patients progressed towards their goals; however no effectiveness data relating to EF was collected as the aim of the study was to assess acceptability of GMT.

Initiation facilitation was used in Miotto [24] and Spikman [25] to help patients who have difficulty in translating intention into action or initiative problems. They were instructed to link plans to an external device (mobile phone, alarm) or to a routine activity (lunch, morning hygiene) to prompt the first step of GMT strategy.

Are comprehensive EF interventions incorporating GMT effective?

Four papers report that complex interventions including GMT are effective, with medium to large effect sizes. These interventions were tested in randomized [25] or pseudo-randomized controlled trials [24][37][23].

Miotto et al. investigated an Attention and Problem Solving (APS) Program [24], focusing on problem awareness and monitoring of actions, that teaches the patients to develop, initiate and implement a plan. The authors report significant effects of the intervention on the carer-rated DEX questionnaire and on the "Modified Multiple Errands Task" in which patients were asked to carry out a series of activities requiring planning, strategy, sequencing, monitoring in a shopping center with a given amount of money.

Spikman et al.'s "multifaceted treatment of executive dysfunction" [25] used GMT and Problem Solving Therapy (PST), tailoring the intervention to patients' specific goals. It was tested in a high quality multicentre single-blind RCT. The multifaceted treatment of executive dysfunction comprises three phases. First, improving awareness of executive deficits is enhanced by continually monitoring and evaluating performance during training, predicting performance and analyzing factors that did or did not help. Then goal setting and planning are trained, based on Ylvisaker's work [47] and Sirigu's scripts [55]. Intended activities are formulated in terms of goals and steps, explicitly and concretely verbalized, put in the right order on worksheets, eventually leading the patients to apply the same

strategy to real life goals, relying on GMT for execution and monitoring. Finally PST is introduced to address problems that might arise during task execution. The primary outcome measure was the Role Resumption List, a validated Dutch interview, which assesses changes in amount and quality of activities compared with premorbid levels in four daily life domains: vocational functioning, social interaction with proxies, leisure activity and mobility. The experimental group showed larger improvement than the control group, who received computerized cognitive training [56].

“Goals training” is a 43 hour training in goal-oriented attentional self-regulation. It relies on GMT, Problem Solving therapies, “Mindfulness” and attention interventions for accomplishing individually-salient, self-generated and complex goals. Novakovic [23] reports that the intervention group improved more on EF and attention tests than a control group receiving two hours of education. A transfer to memory and learning tests was present. However, the difference between the two groups on an ecological assessment (the “Multiple Errands Test”) was not significant. Chen [37] reports the fMRI results of this intervention on a cognitive task presenting images relevant or irrelevant to a goal: modulation of neural processing in extrastriate cortex was significantly enhanced in “goals training” whereas training effects within pre-frontal cortex depended on an individual’s baseline state.

Overall, GMT combined programs were effective on participation with medium-large effect sizes on participation questionnaires ($g = 0,49 - 2,14$) when these were included, whereas GMT alone did not have a significant effect on questionnaires.

Does GMT rehabilitation improve EF at the level of impairments?

All studies that found an intervention effect on measures of participation failed to detect statistically significant intervention effect on impairments assessed by classical neuropsychological tests. In Miotto’s [24] and Spikman’s [25] studies, patients showed some progress on some neuropsychological impairments, but not significantly more after GMT than after the control intervention. This suggests that improvement on tests measuring impairment is not specifically due to the GMT intervention. This is consistent with Kennedy’s conclusions [5] regarding metacognitive strategy training. Conversely, in four articles [23][37][41][6] relating to two studies, patients receiving the intervention improved on paper-and-pencil neuropsychological tests, but not on more ecologically relevant tests of EF (Hotel Task [6] and Multiple Errands Task [37][23]) and the effects on measures of participation were either

not detected [6] or not assessed objectively [37][23]. However, patients generally reported using GMT strategies more than before the intervention [23].

Do GMT effects generalize to untrained tasks and does GMT rehabilitation improve participation?

Novakovic-Agopian et al. [23] reported an improvement on EF that generalized to tests of memory and learning, but this result must be interpreted with caution as the intervention groups was highly stimulated by a 43 hour program whereas the control group had two hours education on brain injury. Carers [24] [41] and patients [25] reported improvement on questionnaires of EF, on interview of role resumption [25] or reported subjective improvement in daily life [7][41][17] in most studies (except [6][37][23]). Improvement on questionnaires[24][25][41] can be considered as a generalization effect and transfer to home-context activities but questionnaires are known to be influenced by many confounding factors other than intervention effectiveness [57]. This was addressed in the studies of Miotto [24] and Spikman [25] as questionnaires of active group patients were compared to questionnaires of active control patients who received a time-matched control intervention with participants being blind to which treatment was active. In both of these studies significant effects of the intervention were reported.

GMT had a positive effect on EF “ecological” real-life tasks like the Multiple errands Task [24], the Executive Secretarial Task [25] or test that are supposed to be close to daily life complex tasks, like the Hotel Task [14] and the Everyday tasks [7][17] although the truly ecological value of these tests is uncertain.

Who can benefit? EF dysfunction severity and self-awareness

GMT is often said to be more suited to less severely impaired patients. Heterogeneity of interventions, and of types of brain injuries in the reviewed studies does not allow any conclusion on which population is the best target for GMT. Most studies did not report on the severity of the dysexecutive syndrome and some did not report on TBI severity [23][37]. There is some, albeit limited, evidence that more severely impaired patients would benefit less [16].

Awareness of impairments is thought to be essential for GMT success. Problem awareness was specifically trained in most interventions, often prior to GMT, as a key step before engaging the patient in the metacognitive training [24][25]. The first few GMT modules emphasize awareness of “slips”

during daily life and encourage patients to monitor them throughout the day. Monitoring EF performance “on task” and recognizing errors or even predicting them could trigger metacognitive strategies use [51][58].

Discussion

GMT training effectiveness and validity of goal-neglect theory in interventions

At the present time there is insufficient evidence to support the application of GMT as a stand-alone intervention in patients with brain injury. Studies reporting effectiveness of GMT alone were either GMT proof-of-principle-studies [7][14][16] assessing the immediate effect of a cognitive strategy but not the effectiveness of use of that strategy in everyday contexts, or low quality case studies [7][41][17]. Levine’s RCT [6] is not sufficient to affirm that GMT administered alone is effective as a rehabilitation program because no effects were observed on EF daily life complaints and because two out of three outcome measures that reached statistical significance were similar to tasks used during GMT training.

GMT’s theoretical model of goal-neglect was supported by Manly’s periodic alerts[14] study but not confirmed in Levine ‘s [6] that used the same outcome measure nor in Sweeney’s study [16] that used a novel virtual task. In the Fish et al study[15], on days where patients received content-free cues on their mobile phones, prospective memory performance was better, emphasizing the important role of alerting in GMT effectiveness. No published studies have yet proven the generalizability of such cueing on patient-set goals and daily life tasks.

Measuring outcome in EF intervention studies

The issue of novelty

An outcome measure needs to be novel to make significant demands on EF [59][60][8][61][62]. Multi element tasks (requiring a patient to do at least some of each of n tasks included in the test in a set amount of time) are sensitive to executive dysfunction [63][64] and have a higher veridicality than classical neuropsychological EF assessments. However, their test-retest reliability tends to be low [65] [66] perhaps because a patient who was overrun by time on first testing may remember to do some of each task the next time. In GMT training, patients are explicitly trained during GMT modules to

manage multitasking in the same conditions as the tests used to evaluate their progress. When the trained task is repeated after intervention, its familiarity may make it less demanding on EF as it is likely to require the application of learned knowledge and task-specific procedures which may have become automatic, rather than more general problem solving and goal management processes [67][62]. As the whole purpose of EF tests is to prevent lapses into automaticity and promote conscious, novel and effortful processing [60][62], apparent progress after EF interventions may not necessarily reflect changes in underlying executive processes. Even parallel forms of the same test [66] may not overcome the novelty problem. The content may be new but the format is not [67].

With the exception of one study [25], all rehabilitation studies used the same task pre- and post-intervention. This limits the opportunity to draw conclusions regarding the EF component in task success as a result of the intervention. Although it is natural to use the same test before and after the intervention, this may not be the best methodological solution for EF research [62]. Spikman et al [25] overcome this issue in their trial as they administered an ecological EF task (Executive Secretarial Task) only once, *after* the intervention and compared the results of the experimental group with controls that had a time-matched control intervention. This single administration made the task genuinely novel, though of course there was no direct comparison of pre-intervention performance.

The issue of ecological validity

As the primary aim of any cognitive rehabilitation intervention is to improve functioning in everyday life, the ideal is to use outcome measures that reflect everyday functioning.

Some studies use measures that are clearly not similar to everyday life tasks but have been shown to correlate with performance on activities of daily living. For example, the SART task used by Levine et al. [6], is a computerized sustained attention task that consists of withholding key press to one of nine single-digit numbers targets. Although the SART has proven to have a good correlation with reported everyday attentional failures and performance [43], it has never been demonstrated that improvement on the SART is correlated with improvement in everyday functioning. Indeed, Levine et al. [6] found no improvement on a questionnaire on everyday cognitive failures after GMT while patients had significantly improved on the SART.

Tests designed to be more ecologically valid such as the “multiple errands” format tasks [68] [69] offer a challenging, novel, open-ended, multiple goal context, and require multiple, innovative and higher level strategies. As such, they have the strongest verisimilitude to daily EF requirements and offer a good approach to providing evidence for intervention effectiveness in daily life (for a review of useful tests assessing EF in intervention studies see Lewis et al [57]).

Another way of assessing EF in novel and challenging situations is to ask proxies how patients function in daily life with a specific focus on aspects of daily life that require EFs. This has been done through questionnaires or structured interviews such as Dysexecutive Questionnaire [6][41][24], Cognitive Failures Questionnaire [6][41] or Role Resumption List [25]. A disadvantage is subjectivity in responses, especially when a patient and his/her family engage in a long, demanding rehabilitation protocol, something which might account for the progress seen in control patients’ questionnaires [25] when the control intervention is active and duration-matched.

Measuring impact on neural processes

Another approach to examining the impact of GMT is to investigate impact on brain processes associated with goal management. To date few paradigms are available to do this, but one example of an attempt to do this was that of Chen et al [37] who used a visual selective attention task in which only goal-relevant stimuli had to be selected for further processing (1-back matches within the relevant category in a series of images composed of faces or scenes categories, with varying instructions as to which category is relevant). However, what is also needed is evidence that measures of EF or specific measures of processes related to goal management that can be used in the scanner environment also have validity in terms of measuring processes that impact on everyday functioning.

Limitations and future research

One limitation of this review is that the quality appraisal method used has not been independently validated. The appraisal system was developed to capture important aspects of research investigating interventions for EF, with a clear focus on impact on improving EF in everyday life. As the same quality rating was applied to all studies, inevitably studies that were designed as “proof-of-principle” studies scored lower than more substantial rehabilitation studies. Thus studies that might be considered to be equally strong as far as the individual aims of each study were concerned scored differently.

Nevertheless, the primary focus of this review was to address the question of where the evidence is at just now in terms of whether GMT is useful for improving functioning in the everyday lives of people with brain injury. In this case, using consistent appraisal system for all studies seems appropriate.

A second limitation was the lack of objective criteria to decide which outcome measures are truly ecological and not only called as such by authors. This issue was been emphasized in a recent review [70] and our review followed the same approach

For as many studies as possible effect sizes were calculated. The possibility of a meta-analysis was considered, but given the heterogeneity of specific interventions and outcome measures, and the level of variability in effect sizes, it was considered that an average effect size (even a weighted effect size) had the potential to be misleading. However this does mean that at this stage in the development of GMT interventions it is not feasible to estimate an overall average effect size.

Finally, the main difficulty in analyzing the results comes from the fact that in several studies, especially those who were most effective, GMT was only a part of a larger therapeutic program and so it is impossible to state to what extent GMT contributed to the observed effect." Multiple baseline single case experimental designs, in which GMT and the other components of comprehensive programs are introduced sequentially might give some insight into the respective effectiveness of each compound. A controlled trial with cross-over with three groups similar to Miotto's methodology for example would give valuable knowledge about the respective effectiveness of different compounds of GMT combined interventions by comparing a group receiving general cognitive stimulation, a GMT group and a group receiving Spikman's multifaceted treatment of executive dysfunction. In any case, research on GMT should use ecologically valid outcome measures targeting participation rather than impairments, and where possible use objective, ecologically valid tests with parallel versions that ensure task novelty. More reflection is needed as to what constitutes a parallel version of an EF task: tasks that have the same format but differ in content may not be sufficient, but designing tasks that draw on the same EF processes but use quite different presentation formats is challenging.

Implications. Recommendations

GMT is a clearly defined, structured and manualized training which makes it attractive as an intervention for people with dysexecutive problems. To date, the evidence is insufficient to justify a

recommendation that GMT should be delivered as a stand-alone intervention. However, evidence is stronger that GMT may be a useful component of a more comprehensive intervention that includes using (1) Problem Solving Therapy, (2) focusing on patients' own personal goals as part of the GMT program including use of daily life tasks as part of the training program and with use of between session homework to encourage generalization (3) using external cueing or prompting to remind support application of GMT strategies in everyday situations (4) an intensity of training sessions that is greater than weekly and a duration of more than 15 hours . When patients are unaware of their impairments, awareness intervention seems useful before beginning a GMT-based rehabilitation.

Other approaches seem very promising but there isn't enough evidence to support their use yet: using naturally-occurring daily life distracters as cues to apply GMT to the current situation [17], using GMT for brain injured children.

The Attention & Problem Solving program [24] and the "Multifaceted Treatment of executive dysfunction" [25] programs offer the best evidence of GMT-combined effective intervention. Their comprehensively explained active ingredients, tested in well designed studies, can reasonably be recommended to rehabilitation teams in search of metacognitive strategy training effective on daily life EF for brain-injured patients.

Declaration of Interest:

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Table 2: Description, methodological ratings and main findings of included studies (by type and by chronological order of publication)							
Author	GMT alone or GMT combined with other rehabilitation methods	Sample	Description of experimental intervention and of control intervention	Outcome measures	Findings	Effect sizes according to Cohen’s rating	Maintenance of effects
Study type (group/single case)	Method						
Quality rating							
Proof-of-principle studies							
Levine et al. 2000 [7] – study 1 Group study 56%, moderate	GMT alone RCT	30 patients with mild to severe TBI 3-4 years post TBI	1 hour of individual GMT presenting goal management processes, examples of goal management failure, teaching the “STOP-STATE your goal-SPLIT task into sub-goals-Check” cycle. Practice activities including searching letters and numbers in a grid with an increasing level of difficulty (room layout grid) and proofreading tasks Control group: motor skill training (MST)	Paper-and-pencil tasks called “everyday tasks” including proofreading, and room layout grid	GMT participants made less errors in proofreading and grouping tasks but took more time for task completion that MST group	g = 0,9 for proofreading (<i>large</i>) g = 0,95 for grouping (<i>large</i>)	Not evaluated
Manly et al. 2002 [14] Group study 80%, high	GMT alone + cueing Comparison of performance in two conditions (with and without auditory alerts)	10 patients: 1 suspected ischemic stroke and 9 TBI (7 were severe to very severe on Teasdale’s system) 24 controls	A “beep” during task to remind patients about their goal was used as a content-free cueing. No specific training. Healthy controls to determine if both version of the test used are parallel.	Hotel task with parallel version of subtasks and 6 elements task from the BADS.	With auditory alerts, patients performed better (time allocation to different tasks more optimal) and close to controls.	g = 0,795 for cueing effect on patients (pretest-posttest) (<i>large</i>)	-
Fish et al. 2007 [15] Group study 64%, moderate	GMT alone + cueing Comparison intra subjects with/without GMT cue	20 patients with ABI, including 14 TBI (severe to extremely severe on Bigler’s classification).	One 30-min individual GMT session to associate the content-free cue (text message “STOP”) with reviewing goals	Number of phone calls made and timing accuracy of phone calls	Significant effect of cueing with more phone calls and better time accuracy on cued days	g = 0,508 for time accuracy (taking uncued days as controls) (<i>medium</i>)	-
Sweeney et al. 2010 [16] Group study 76%, high	GMT alone + cueing	17 patients with ABI > 1 year post-injury, unknown severity 17 healthy controls	A “beep” during task to remind patients about their goal. No specific training.	The Removals Task	No effect. A trend that less impaired patients would benefit more	No effect	-
Rehabilitation studies							
Levine et al. 2000 [7] – study 2 Single case 52%, moderate	GMT alone Single-Subject	Female, 35 years, 5 months post encephalitis	2 sessions of GMT similar to Levine 2000-study 1 + 3 sessions of GMT applied to cooking task	Observation of problematic behaviour during meal preparation Self report cooking errors dairy Paper-and-pencil tasks including proofreading, room layout grid.	Less problematic behaviour during meal preparation observation Less self-reported difficulties in meal preparation Less errors on the “everyday tasks”	-	Maintenance of effects on cooking at 6 month
Pachalska et al. 2000 [42] Group study 36%, low	GMT combined Controlled	24 patients with TBI, unknown severity	-“Executive function training” based on GMT, included in a larger holistic Polish program “MARSZ”. The program was applied to activities of increasing difficulty, including real life activities (from making a sandwich to performing one’s own professional activity) Duration: 4 weeks, intensity not reported -Control group: standard rehabilitation while waiting for inclusion in MARSZ program	WAIS-R WMS-R Polish battery of “Clinical test of Executive functions” “Cracow test of right hemisphere”	Qualitative data showing large success for MARSZ program Comparative data between groups not reported	No data for ES calculation	Not evaluated
Miotto et al. 2008 [24] Group study	GMT combined Pseudo-randomized, controlled trial with cross-over	Patients with right (n=14) or left (n=16) frontal lobe lesions (23 tumours and 7	-Group 1: “Attention and problem solving based on GMT and PST: 10 weekly 1,5 hour group sessions -Group 2: Information and education through a booklet to read that suggested cognitive exercises	Extensive executive functions paper-and-pencil assessment Modified Multiple Errands Task (MMET)	Intervention effect on MMET and carers DEX when group 1 compared to groups 2 and 3. Some improvement on paper-and-	g= 1,08 for MMET (<i>large</i>) g= 2,14 for carers DEX (<i>large</i>)	Effect maintained at 6 month follow up

76%, high		mild TBI)	and presenting problem-solving frame-work -Group 3: usual care	Virtual Planning test DEX questionnaire	pencil executive tests after APS		
Schweitzer et al. 2008 [41] Case study 56%, quality rating not applicable *	GMT alone Single-Subject case report	Male, 41 years, cerebellum haemorrhage 4,5 months post injury	7 weekly 2-hour sessions of GMT	Questionnaires: DEX, CFQ SART Hotel Task R-SAT D-KEFS Tower Test and TMT CVLT-II	Reduced commission and omission errors on SART Increased awareness of difficulties Less complaints of the patient’s spouse on the DEX Patient returned to work	-	Effects maintained at 4 month follow-up
Spikman et al. 2010 [25] Group study 92% high	GMT combined Multicenter single-blinded RCT	75 patients with ABI (mainly stroke and TBI, unknown severity), living at home, >3 months post injury	-Multifaceted Treatment of Executive dysfunction, relying heavily on GMT and PST but tailored to patients ‘specific goals. -Control Group: CogPack (computerized cognitive training with repetitive exercises) Both interventions were administered in 20-24 one-hour sessions, 2/week, for 3 months	Primary outcome measure =Role Resumption List (RRL) Interview Other outcome measures: Treatment Goal Attainment (TGA) Questionnaires: DEX, QOLIBRI Executive Observational Scale (EOS) rated by therapist Neuropsychological tests: BADS, TMT, Stroop, Tower of London, 15 word test Executive Secretarial Task (EST) administered only after intervention to ensure novelty component	Both groups resumed their previous roles significantly more than pre-treatment (RRL) but experimental group to a larger and statistically significant extent Statistically more improvement in experimental group on EOS Both groups showed less complaints on DEX, better quality of life (QOLIBRI) and progress on personal goals (TGA) Significant group effect when 3 measures of daily executive functioning combined (EST, RRL and TGA)	g= 0,49 for RRL at 6 month follow-up (<i>medium</i>)	Further improvement on RRL at 6 month for experimental group, while control group non significantly worsened on RRL compared to post intervention
Metzler-Baddeley et al. 2010 [17] Case study 52%, moderate	GMT alone + cueing Single-Subject case report	Female, 40 years, Craniopharyngioma treated by surgery and radiotherapy 4 years previously. Remaining suprasellar mass.	3 x 1.5 hour of GMT with auditory cues similar to Manly et al. [22] to trigger GMT. In a second phase patient was taught to use naturally occurring distractions to trigger goal management steps	Telephone Directory and Map search from TEA	Statistically improved errors and speed on telephone directory task. The patient reported subjective improvement in her ability to complete tasks and deal with distractions at work.	-	No follow-up
Levine et al. 2011 [6] Group study 72%, moderate	GMT alone Controlled trial, partially randomized (12 patients randomized)	20 patients with ABI, mainly stroke with frontal lobe lesions	-GMT group: GMT long version presented on PowerPoint Slides -Control group: “Brain health workshop” (BHW): education on BI, matched for time Both intervention were administered 7 x 2hours, in group sessions + between-sessions homework	SART D-KEFS Tower Test Hotel Task DEX Questionnaire CFQ	Less omission errors for GMT group on SART Less rule violations for GMT group on Tower Test but no group x session interaction for achievement score Paradoxically less optimal per-task time allocation in the GMT group on Hotel Task immediately post intervention No effect on questionnaires	g = 0,43 on omissions errors on SART (<i>medium</i>) g = 0,95 for rule violation in the Tower Test (<i>large</i>)	At 4 month follow-up, effect remained on omission errors on SART and rule violation on Tower test Hotel Task time allocation did not differ from controls at follow-up.
Novakovic-Agopian et al. 2011 [23] Chen et al. 2011 [37] Group study 68%, moderate	GMT combined Pseudo-random cross-over RCT design	15 patients with ABI >6 months post-injury, unknown severity	-“Goals Training”: GMT + PST + other problem solving, mindfulness and attention interventions 10 x 2 hours of small group training + 3 x 1h individual training + 20 hours of home practice for 5 weeks. Focus on practice in daily life and self-generated complex goals (individual and group projects). -Control group : 2 hours of education about brain injury	Neuropsychological tests : -EF and attention averaged to an overall “Attention and Executive Function domain” -Memory and learning -Processing speed Multiple Errands Tasks + fMRI on a selective attention task	Significant difference between groups in the “Attention and Executive Function domain” Transfer to memory and learning No statistical difference between control and intervention group on MET fMRI: enhanced modulation of neural processing in extrastriate cortex.	g=0,81 for the Attention and Executive Function domain (group difference) (<i>large</i>) g=0,55 for memory domain (<i>medium</i>) No raw data reported to calculate MET ES	At 10 weeks, effects remained in the group that received Goals Training first when assessed after education control intervention (cross-over). Further improvement on the overall attention and executive domain and working memory.

* As Schweitzer’s patient was in the acute phase of ABI, and no controls were used, causality cannot be affirmed by this study: as such quality rating was not applicable.

Abbreviations

*Sample description: **TBI**=Traumatic Brain Injury, **ABI**=Acquired Brain Injury,*

*Methods: **RCT**=Randomized Controlled Trial, **SCED**= Single-Case Experimental Design*

*Intervention description: **EFT**= Executive function training, **GMT**= Goal Management Training, **MARSZ**= polish abbreviation for “ Active Socio-professional rehabilitation model”, **PST**= Problem Solving Therapy,*

*Outcome measures : **EF**= executive functions*

*-Paper-and-pencil tests: **CVLT-II**=California Verbal Learning Test, **D-KEFS**= Delis-Kaplan Executive Function System, **R-SAT**= Revised-strategy application test, **SART**=Sustained Attention to Response Task (**Commission** error = responding to no-go stimuli; **Omission** Error = not responding to go stimuli, **TMT**=Trail Making Test, **WAIS-R**= Wechsler Adult Intelligence Scale-Revised , **WMS-R**= Wechsler Memory Scale Revised,*

*-Paper-and-pencil tests with some ecological validity: **BADS**=Behavioural Assessment of the Dysexecutive Syndrome, **TEA**= Test of Everyday Attention*

-Truly ecological tests: **EST** = Executive Secretarial Task, **M(modified)-MET**= Multiple Errands Test
-Questionnaires : **CFQ**=Cognitive Failures Questionnaires, **DEX**=Dysexecutive Questionnaire, QOLIBRI= Quality of Life after Brain Injury, TGA=Treatment Goal Attainment