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Executive functions, prospective memory and metacognition in children with traumatic brain injury: assessment, characterization and rehabilitation

Agata Krasny-Pacini

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Université Pierre et Marie Curie

Ecole doctorale Cerveau Cognition Comportement

Laboratoire d'Imagerie Biomédicale UPMC / Equipe de recherche : Equipe Systèmes dynamiques anatomo fonctionnels chez l'Homme : altération et récupération fonctionnelle

Executive Functions, Prospective Memory and Metacognition in children with traumatic brain injury: assessment, characterization and rehabilitation.

*Fonctions Exécutives, Mémoire Prospective et Métacognition chez l'enfant
traumatisé crânien : évaluation, caractérisation et rééducation.*

Présentée par Agata KRASNY-PACINI

Thèse de doctorat de Science de la vie

Spécialité : Neurosciences

Dirigée par Mathilde CHEVIGNARD et Jonathan EVANS

Présentée et soutenue publiquement le 24 Mai 2016

Devant un jury composé de :

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To all children and adults experiencing a traumatic brain injury and to their families, whose lives will never be the same again after such a dramatic event.

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Preliminary Note

This PhD uses the « Paper-based» model, (« *thèse sur articles* » in French).

Because of the paper-based thesis format, some information may be repeated, or appear not chronologically. Chapters corresponding to published or submitted papers are reproduced faithfully to the journal it has been published in (therefore abstract and paper length, use of UK/US language corresponds to the journal requirements and is not uniform across chapters). The results are discussed in each chapter separately and the final discussion is only a global discussion of themes not developed in the papers.

List of abbreviations (in bold: most often used abbreviations in this PhD)

ABI: acquired brain injury

ADHD: Attention Deficit – Hyperactivity Disorder

BADS-C: Behavioural Assessment of the Dysexecutive Syndrome for Children

BRI: Behavioural Regulation Index

BRIEF: Behaviour Rating Inventory of Executive Function

CBCL: Child Behaviour Checklist

CCT: Children’s Cooking Task

CMS: Children’s Memory Scale

DEX-C: Dysexecutive questionnaire for children.

EF: Executive functions

FSIQ: Full Scale Intellectual Quotient

GAS: Goal Attainment Scaling

GCS: Glasgow Coma Scale

GEC: Global Executive Composite Score

GMT: Goal Management Training

GN: Goal Neglect

ICF: International Classification of Functioning

IRR: inter-rater reliability

MK: metacognitive knowledge

MVA: Motor Vehicle Accident

pGMT: Pediatric Goal Management Training

PIQ: Performance intelligence quotient

PM: prospective memory

PMR: Physical Medicine and Rehabilitation

PSI: Processing Speed Index

RCT: Randomized Controlled Trial

RoM: Range of motion

SA: School Assistant

SMART criteria: Specific, Measurable, Attainable, Relevant, Time-specific

TBI: Traumatic Brain Injury

ToM: Theory of Mind

VIQ: Verbal intelligence quotient

WCST: Wisconsin Card Sorting Test

WISC: Wechsler Intelligence Scale for Children

WM: Working Memory

INTRODUCTION

CHILDHOOD TRAUMATIC BRAIN INJURY

Traumatic brain injury (TBI) is the leading cause of death and acquired disability in children^{1,2}. Annual incidence of TBI in children, adolescents and young adults (up to 24 years) has been recently estimated in a systematic review¹ at 691 per 100.000 attending the hospital emergency room, 74 per 100.000 being hospitalized, and 9 per 100.000 deaths. Boys are more represented than girls (sex ratio 1.4/1 before the age of 10 and 2.2/1 after the age of 10)¹. Childhood *severe* TBI is usually defined as a TBI with initial or lowest Glasgow Coma Scale (GCS) score ≤ 8 . The incidence of TBI is 75 % per year³. Most TBIs are mild and the prevalence of severe TBI is unknown. Of hospitalized TBIs 17% are estimated to have severe TBI¹. In McKinlay et al.'s cohort³, moderate and severe TBI accounted for 10% of all TBI.

The leading cause of TBI in infants and very young children aged less than 2 years are falls followed by assaults or abusive head trauma and motor-vehicle crashes⁴. In Thurman's review¹, among children aged less than 5 years, falls accounted for about two-thirds of injuries. In this age group, motor vehicle-related incidents accounted for comparatively small proportions of injuries but most were fatal. Among children aged 5 to 14 years, falls remained the leading cause, followed by sports or recreation-related injuries, followed by motor vehicle injuries. Among youths aged 15 years or older, motor vehicle-related incidents are the leading cause of brain injury, followed by assault, then sports-related incidents and firearms.

Childhood TBI often results in impairments in children's sensory-motor and cognitive functioning including deficits in executive functioning, language, memory, attention, processing speed, working memory⁵⁻⁷. Behavioural and emotional problems are also frequently reported in children with TBI^{7,8}, as well as impaired social problem-solving and moral reasoning^{9,10}. Those deficits often have a significant and longstanding impact on the child's everyday functioning, communication skills, adaptive functioning, social interactions,

academic achievement, as well as poorer participation in society, difficulty maintaining employment and social relationships ¹¹⁻¹⁶.

TBI in children takes place in the context of ongoing cognitive development, so as well as disrupting current abilities, TBI may interrupt or affect skills that are still developing or that are to develop in the future and therefore may have more unpredictable consequences than adult TBI that occurred in a mature brain (see Anderson for a review¹⁷).

Despite some improvement of cognitive function after the acute phase, it has been observed that children with severe TBI tend to show a slower rate of subsequent development so that the gap between the severe TBI and the control groups may expand over time ⁶. This has been termed the « growing into deficit » model, and was based on large meta-analysis by Babikian and Asarnow⁶, where most studies did not follow children more than two years post injury. Subsequent cohorts that followed children longer after TBI, however, noted less pessimistic outcome trajectories: by 30 months after insult, recovery seems to stabilize and children begin to make appropriate developmental gains, with age-appropriate progress documented five years¹⁸ and ten years post injury¹⁹ (but persistent impairment after severe TBI, as children make progress but do not catch up to expected level of cognitive development).

EF IN CHILDREN

Executive functions (EF) are a collection of related but distinct abilities that allow individuals to engage efficiently in intentional, goal-directed problem-solving actions^{20,21} through conscious and effortful processing²² and to adapt to new situations in the real world ²³. Different models of EF have been proposed, but Diamond's model²⁴ is developmentally most robust and practical to use.

There is general agreement that there are three core executive functions (EF) ^{25,26}:

-inhibition [inhibitory control, including self-control (behavioural inhibition) and interference control (selective attention and cognitive inhibition)]

-working memory (WM)

-and cognitive flexibility (also called set shifting, mental flexibility, or mental set shifting).

Inhibitory control of attention enables us to selectively attend, focus on what we choose and suppress attention to other stimuli, irrelevant for the current task²⁴. We need such selective attention during activities such as cooking when we want to concentrate on the recipe rather than the TV or thinking about the holidays that are about to start. We can also choose to voluntarily to inhibit attention to particular stimuli (e.g. taking the comb on the table and comb our hair) and attend to more appropriate stimuli, based on our goal or intention (e.g. take the mixer to mix the ingredients). Inhibitory control of attention (or selective or focused attention) is an endogenous, top-down, active, goal-driven, voluntary, volitional, form of attention, sometimes called «executive attention »²⁷, and is contrasted with automatic, bottom-up exogenous attention, driven by a salient stimulus such as a klaxon or a strong smell²⁸. Cognitive inhibition involves resisting extraneous or unwanted thoughts or memories, including resisting interference. Cognitive inhibition is usually in the service of aiding WM and tends to correlate more with WM measures than with other types of inhibition. Self-control is the aspect of inhibitory control that involves control over one's behaviour and control over one's emotions in the service of regulating behaviour²⁴. Inhibition is extremely hard for young children²⁴, with a very rapid progress in inhibition to 5 years of age²⁰, less rapid between 5 and 8 and then slowed but continued progress during childhood and adolescence²⁹.

Working memory (WM) involves holding information in mind and mentally working with it (or said differently, working with information no longer perceptually present)^{30,31}. Working memory supports inhibitory control²⁴. You must hold your goal in mind to know what is relevant or appropriate and what to inhibit. "By concentrating especially hard on the information you are holding in mind, you increase the likelihood that information will guide your behaviour, and you decrease the likelihood of an inhibitory error (mistakenly emitting the default, or normally prepotent, response when it should have been inhibited)"²⁴. Various models of WM have been proposed, the most often used in adults being Baddeley's³⁰: the central executive acts as supervisory system and controls the flow of information from and to its two slave systems: the phonological loop and the visuo-spatial sketchpad. The phonological loop stores verbal content, whereas the visuo-spatial sketchpad caters to visuo-spatial data. Infants of only 9 to 12 months can update the contents of simple information. However, being able to hold many things in mind or do mental manipulation is far slower to

develop and shows a prolonged developmental progression³². Other contemporary models of WM are discussed later in relation to prospective memory.

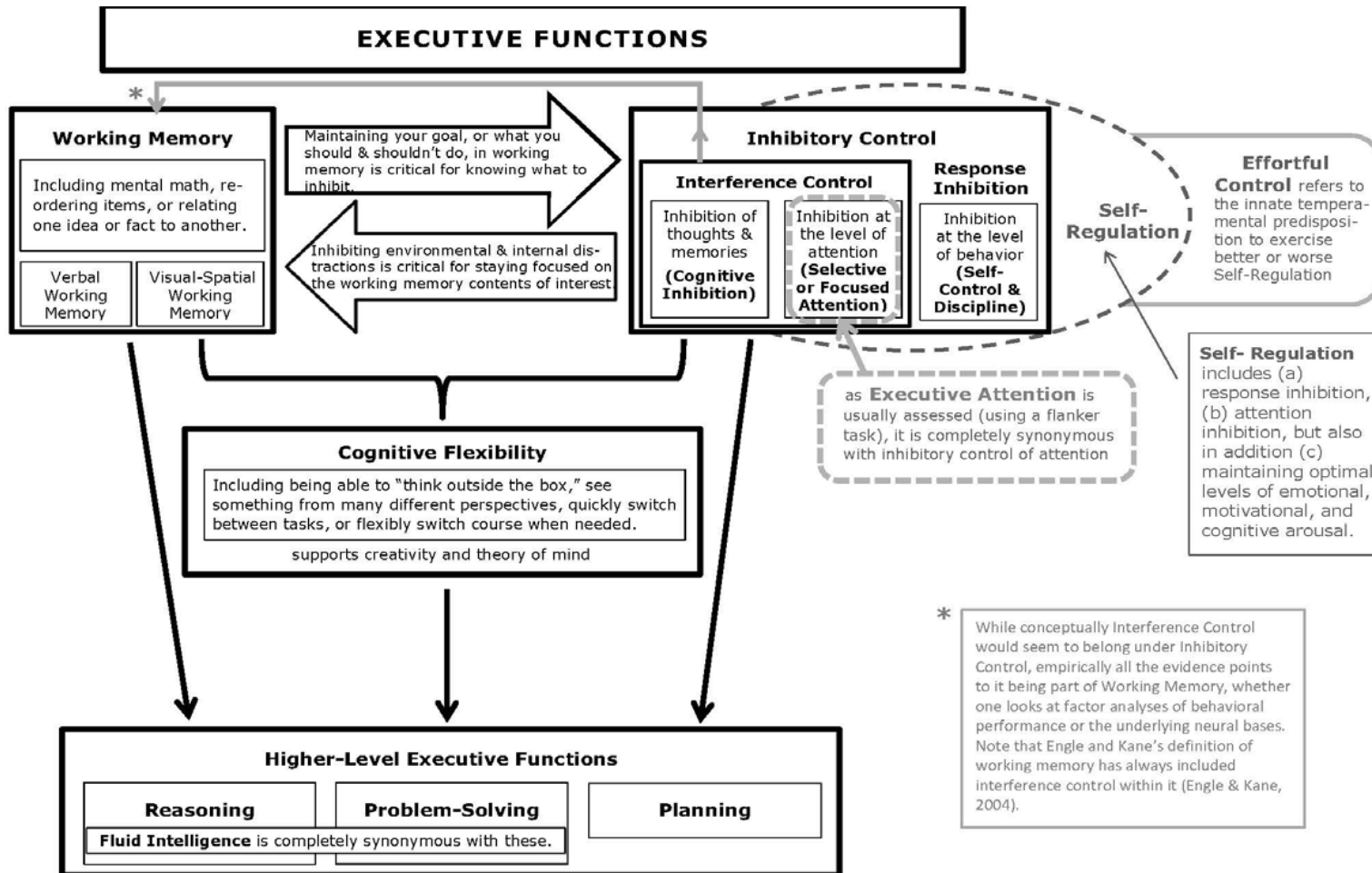
Inhibitory control supports working memory²⁴. To relate multiple ideas or facts together you must be able to resist focusing exclusively on just one thing, and to recombine ideas and facts in new, creative ways, you need to be able to resist repeating old thought patterns. Inhibitory control can also aid WM by helping to keep our mental workspace from becoming too cluttered by suppressing extraneous thoughts (i.e., gating out irrelevant information from the WM workspace), resisting proactive interference by deleting no-longer relevant information from that limited-capacity workspace.

Cognitive flexibility is being able to change perspectives and requires to inhibit our previous perspective and load into WM a different perspective²⁴. One aspect of cognitive flexibility is being able to change perspectives spatially (e.g., “What would this look like if I viewed it from a different direction?”) or interpersonally (e.g., “How does it look from your point of view”). Flexibility builds on inhibition and WM and comes in later in children’s development³³. By age 4.5-5 years, most typically developing children perform simple flexibility tasks (such as identify both figures in an ambiguous figure or switch sorting dimensions³⁴, but not until 7 to 9 years of age, can children switch flexibly on a trial-by-trial basis as usual paradigms testing flexibility require³³.

From these three core EFs, higher order EFs are built such as reasoning, problem solving and planning³⁵. Fluid intelligence is included in these higher-order EFs. Previous research has shown that intelligence is inextricably linked to EFs in childhood³⁶⁻³⁸ and the view that EFs are independent has been questioned in the literature, even in adults^{39,40}.

Diamond’s model of EF in development is summarised in the figure Intro.1 ²⁴.

Figure Intro.1. Model of Executive functions (Diamond 2013), reproduced with the permission of the author.



Based on the study of adults with brain insult, traditional localizationist views have argued that executive skills are primarily mediated by prefrontal cortex. It remains unclear whether a similar pattern of localization exists in childhood. In Jacobs' study⁴¹ that compared a range of EFs, there was little difference in executive processes between frontal and extra-frontal lesions groups. Their results provide support for contemporary models that propose a distributed, but integrated neural network for executive skills, suggesting that the integrity of the entire brain is necessary for adequate executive functions in childhood. Further, focal lesions to any brain region during development may render children vulnerable to a range of executive deficits that would not normally be expected following similar pathology in adulthood.

OTHER COGNITIVE FUNCTIONS RELYING ON EXECUTIVE FUNCTIONS

EFs are specifically involved in a series of related cognitive functions that impact everyday functioning in social and academic areas⁴², vocational outcome and participation in society. Examples of such functions include social cognition, social problem solving¹⁰, social behavioural regulation^{13,43}, metacognition/self-awareness^{44,45,46} and prospective memory^{47 48 49 50}. This PhD focuses on the latter two.

Metacognition is the conscious knowledge of cognitive processes as well as the ability to consciously monitor and regulate one's ongoing activities or processes while engaging in a task⁵¹. Metacognition refers to the awareness of one's own cognition and is used in the context of normal functioning. The concept of awareness/self-awareness (or more usually lack of awareness), on the other hand, is usually used in the context of pathology and refers to the awareness of one's deficits, including cognitive deficits. As such, self-awareness can be viewed as metacognition applied to difficulties in cognitive functioning.

Although terminology varies across the disciplines of psychology, education or neuropsychology, metacognition comprises metacognitive knowledge (MK), metacognitive skills and metacognitive beliefs.

Metacognitive *knowledge* is the knowledge of cognitive processes and products⁵² and has three components: Declarative MK (assertions about the world and the knowledge of the influencing factors of human thinking such as memory and attention⁵³), Procedural MK (knowledge of the methods for achieving goals and the knowledge of how skills work and how they are to be applied⁵⁴) and Strategic/Conditional MK (the awareness of the conditions that influence learning such as why strategies are effective, when they should be applied and when they are appropriate⁵⁵). According to Montague, procedural MK is necessary to apply declarative MK efficaciously and to coordinate problem solving; conditional MK enables a learner to select appropriate strategies and to adjust behaviour to changing task demands.

Metacognitive *skills*, also termed executive control are the voluntary control people have over their own cognitive processes and rely heavily on EF. These skills include prediction, planning, monitoring and evaluation. Metacognitive *beliefs* are the broader general ideas and theories people have about their own and other people's cognition⁵⁶ (attribution, motivation, self-esteem) although their truly metacognitive nature is debated^{57,58}.

Metacognition is known to be poorer in younger children. It has been studied mostly in relation to metamemory^{59,60,61,62,63}. From 8 years, when prompted, children begin to accurately predict that they would later recall specific details correctly and predict that the answers they provided were correct^{59,60, 61}. Not until 11-and 12- years-of age, however, can children selectively withdraw answers that would have been incorrect, although 9-and 10-year-olds already present well-developed monitoring skill^{62,63}.

Prospective memory (PM) is the ability to remember to perform intended activities⁶⁴. PM tasks require retrospective memory to remember the task, but depend on executive functions (EF)⁶⁵ for successful goal maintenance, retrieval and implementation at the right moment. At least three attributes are accepted as characterizing a PM task: (1) a delay between formation of the intention and the opportunity to carry it out; (2) absence of an explicit reminder to carry out the task at an appropriate moment; (3) the need to interrupt one's ongoing activity in order to carry out the intention. Literature on prospective memory in typically-developing children shows improvement in PM performance across the age range from 2 to 12 years^{50,66}. Small children as young as two can succeed in PM if motivation is high (remind their Mum to buy them sweets)⁶⁷. As they grow older, children become increasingly skilled at using external

reminders to cue PM and increasingly proficient at applying time-checking strategies. It has been proposed, however, that those age effects that have been documented in the literature, may be entirely attributable to factors such as: (1) unequal difficulty of the on-going task, allowing less attentional resources to PM tasks in younger children; (2) retrospective memory component; (3) motivation. When taking these into account, age effects of PM are typically small⁶⁸.

EXECUTIVE DYSFUNCTION AND RELATED COGNITIVE DYSFUNCTIONS IN CHILDREN WITH TBI

EF can be impaired after traumatic brain injury (TBI) ^{12,69–72} ^{6,69} and influence a series of outcomes including academic achievement^{42,73,74}, social cognition¹⁰, vocational outcome ¹⁵ and ability to live independently. The level of executive dysfunction is found to be related to the injury severity and younger age at injury^{14,71,75–79}. Relationship between age at injury and outcome is complex and literature begins to suggest that it is not linear^{19,80}. TBI sustained at an earlier age (when cognitive functions have not yet developed) is prone to generalized delayed adverse outcome, but outcome may be even worse when the function is at a rapidly developing stage⁷⁷ at the time of injury.

Impaired awareness/metacognition, sometimes also termed “anosognosia”, is a common phenomenon described in children who have sustained a TBI ^{81,82,83}, but few studies have explored it, partly because of a lack of underlying models and lack of adequate assessment tools. In children, self-awareness is particularly difficult to capture and assess, as it results from a combination of organically based unawareness (due to brain injury) and simple developmental immaturity ⁸³ present in typically developing children as well, which makes it harder to characterize. In metamemory, children with TBI have exaggerated overconfidence in their performance and poor estimation of memory span⁸⁴.

Prospective memory (PM) has been shown to be impaired in children with brain injuries ^{85,86}. PM problems are reported as a major concern by the parents of children with TBI ⁸⁷. The

ability to compensate for PM deficits in adults significantly predicts the ability to live independently⁸⁸. Parents of children with TBI report serious concerns for their child's safety and ability to be left unsupervised even briefly because of substantial PM impairments⁸⁷.

EXECUTIVE DYSFUNCTION AND GOAL NEGLECT

As noted earlier, working memory (WM) involves holding information in mind and mentally working with it (or said differently, working with information no longer perceptually present)^{30,31}.

Cowan's model of WM^{89,90} in adults distinguished between (1) information that is accessed at any moment for further processing (the focus of attention), and (2) the information held available in the background for later use (the activated part of long-term memory). Only the focus of attention is assumed to have limited capacity. The activation of representations in long-term memory is not capacity limited, but activation may be reduced through decay or interference. In his model, information can be processed in the focus of attention without being impaired by the demand to hold other information in the activated part of long-term memory. Cowan's model is supported by simple experiments such as Garavan's⁹¹: Garavan asked participants to count squares and triangles that appeared sequentially, in random order, on the screen, so that participants had to remember and update two running counts simultaneously. The task was self-paced, so the latency of individual counting operations could be measured. Garavan observed that a switch from one mental counter to the other (e.g., when a triangle followed a square) took 300–500 ms longer than updating the same counter again (e.g., when a square followed a square). This indicates that participants held one counter in the focus of attention while memorizing the other outside the focus. He concluded that the 300-500 extra ms reflected the time it takes to switch the focus of attention from one list (square) item to the other (triangle).

Cowan's "focus of attention" was then split further into two different regions in Oberauer's model. In a series of experiments, Oberauer⁹² showed that:

-There are items memorized for later recall but not accessed during a concurrent processing task: these items do not affect the speed of the processing task because they are in the activated long-term memory (e.g. holding in memory a number list while updating another number list with arithmetic operations).

-There are other items that are held available for direct access and which affect the processing latencies (updating another number list with arithmetic operations).

-Finally, there is a single item at any moment that is actually selected as the object of a cognitive process (the number currently undergoing the arithmetic operation).

Oberauer conceptualized working memory as a concentric structure of representations with three functionally distinct regions (see Intro.2).

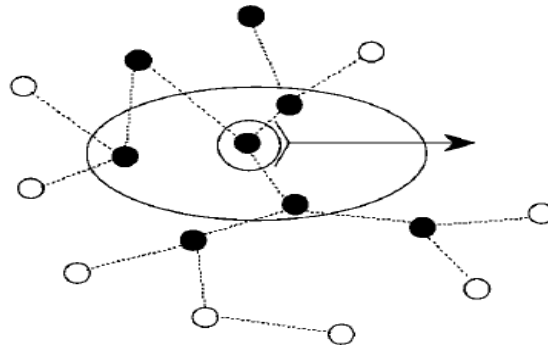
1. The activated part of long-term memory can serve, to memorize information over brief periods for later recall.
2. The region of direct access holds a limited number of chunks available to be used in ongoing cognitive processes.
3. The focus of attention holds at any time the one chunk that is actually selected as the object of the next cognitive operation.

So for Oberauer, working memory is an organized set of representations characterized by their increased state of accessibility for cognitive processes and contents of working memory can be categorized with respect to their access status.

The limits of working memory capacity reflect the limited number of independent elements that can be held in the region of direct access at the same time. This region, therefore, corresponds most closely to what Cowan^{89,90} named the *focus of attention* (i.e. Cowan's focus of attention corresponds therefore to both Oberauer's region of direct access AND focus of attention). Retrieving an item from working memory, either for recall or for manipulation, means bringing this item into the focus of attention. Only objects within the region of direct

(and not activated long-term memory) access are regarded as selection candidates by the focus of attention. “The focus of working memory therefore has a function with respect to memory that is equivalent to the function of a focus of attention in perception. (p412)”⁹²

Figure Intro.2: Oberauer’s concentric model of working memory.

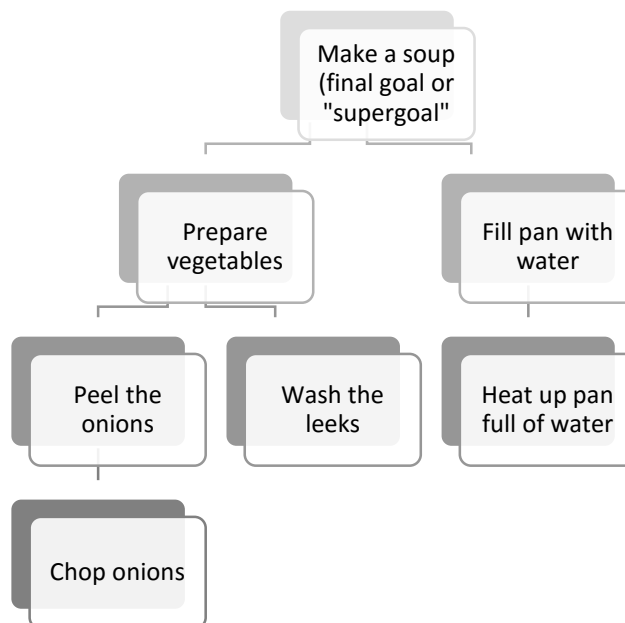


Nodes and lines represent a network of long-term memory representations, some of which are activated (black nodes). A subset of these items is held in the region of direct access (large oval). Within the region of direct access, one item is selected for processing by the focus of attention (small oval). Activated items outside the region of direct access but inside the activated part of long-term memory, are accessible only indirectly, via associative links (dotted lines) to representations in the more central regions

DUNCAN’S MODEL OF GOAL NEGLECT

In adults, Duncan’s theory of “goal neglect”^{93,39,40} suggests that much of human behaviour is controlled by goal and subgoals lists. As we proceed in a task, new candidate goals arise by working backward from a currently active supergoal (see figure Intro.3). These candidate goals are bringing the supergoal closer. For example, a final goal of making a soup (i.e. “supergoal” in Duncan’s terminology), suggests a requirement for peeled and chopped onions (subgoals serving to achieve the super goal). At a particular time during the cooking, peeling onions becomes the current goal. Current goals can be totally overturned, however, by novel input (e.g. making soup is abandoned if the baby tips over its high-chair). In this case it is the input from the environment that suggests a new goal, by working forward from current situation to new current goal. Candidate goals suggested both by working backward and by working forward compete for control of behaviour.

Figure Intro.3: Working backwards from the supergoal to subgoals, according to Duncan's model of goal neglect



Goal competition is influenced by a series of factors that Duncan et al. ³⁹ explored in successive experiments: a goal was more likely to be neglected if the task was novel, complex, if patients were not prompted, if they were stressed by time pressure, if they had frontal lobe damage or low fluid intelligence *g*, or if the delay between goal instruction and possibility to fulfil the goal was long. Finally, in the case of two concomitant goals, the goal given second was more likely to be neglected.

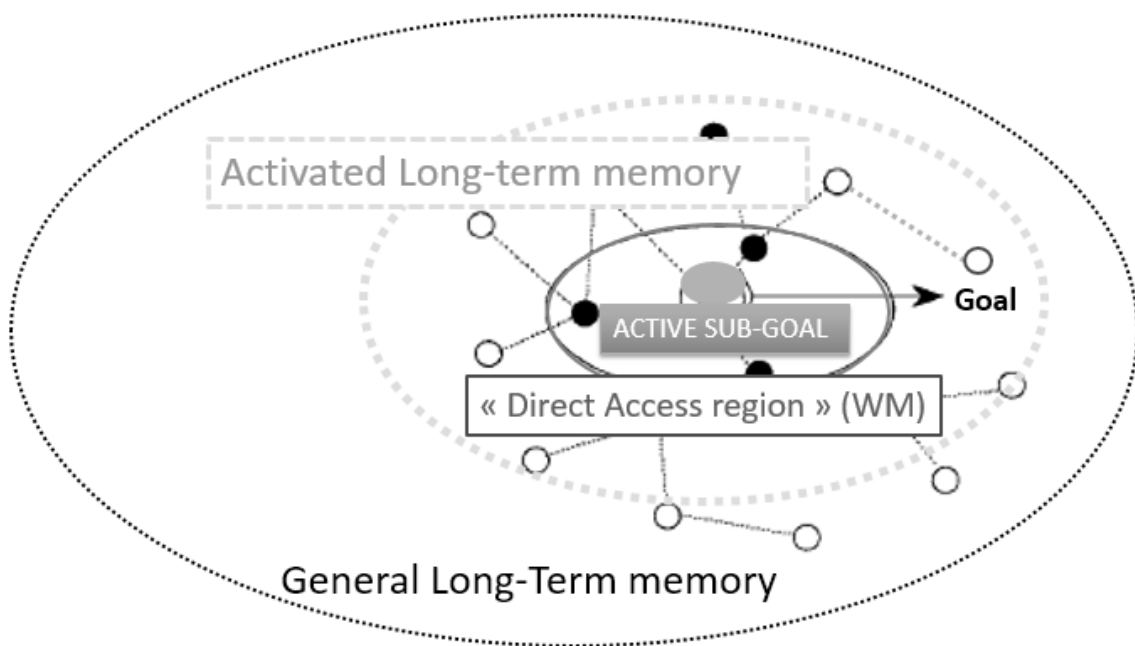
Duncan proposed that dysexecutive patients are impaired in the construction and use of goal/subgoal lists, and are more vulnerable to external input changing their planned behaviour, resulting in disorganized behaviour and neglect of the main (super-) goal.

OBERAUER'S MODEL EXPLAINING GOAL NEGLECT PHENOMENON

Oberauer's model can be very useful in explaining goal neglect in patients with executive dysfunction experience. As the focus of attention can manipulate one item only, only the

current sub-goal is held in the focus of attention, while the final goal is held in the activated long-term memory. Following Oberauer's model, the final goal is therefore not accessible directly but linked to the current sub goal through associative links (see figure Intro.4). The activation of representations of the final goal in long-term memory can, however, become lost through decay or interference. As tasks proceed, the final goal progressively drops towards less activated parts of long-term memory. Following Oberauer's view that WM is an organized set of representations characterized by their increased state of accessibility for cognitive processes, a goal whose activation levels have dropped within long-term memory is less accessible and more likely to be forgotten/neglected.

Figure Intro.4: Oberauer's model applied to goal maintenance that allows an organized behavior during a task and the retrieval and implementation of an intention in prospective memory.



Sustained attention and the right frontal-thalamic-parietal sustained attention system especially⁹⁴ allow patients with intact EF to regularly update the level of activation of the goal and maintain it at a level of activation sufficient to be accessed by the region of direct access and to come to the focus of attention when needed. Dysexecutive patients on the other hand,

have impaired sustained attention and do not maintain the activation of the goal, which eventually becomes inactivated in long-term memory (note it is not deleted - as patients can usually recall what their goal was if they are prompted, but they will not implement the goal spontaneously in their action because it has become inaccessible to the focus of attention). In such cases, external input (habits or environmental triggers) are more likely to oppose and displace higher order goals, resulting in cue-dependent or distracted behavior, relying more on environmental inputs than self-generated goals needed to achieve the goal of the task at hand (supergoal).

GOAL NEGLECT AND PROSPECTIVE MEMORY

In the same way that goal neglect theory attempts to explain failure to maintain a task's final goal in WM (i.e. in a time-limited duration), goal neglect can be applied to prospective memory failures, to explain failure to maintain memory for a goal over a longer duration (hours, days or month). Because the construct is the same, again Oberauer model is useful to explain how an intention can inappropriately drop to general (inactivated) long-term memory and no longer be activated and therefore become inaccessible to the focus of attention. In this case, environmental cues (e.g. seeing a letter box and remembering to post a letter) that normally act as associative links are no longer sufficient (because the goal is too low in activation and accessibility for the focus of attention to use it). When exploring PM in the laboratory, PM tasks tend to use infrequent targets: the higher the frequency of target stimuli, the more the task relies on working memory, because the frequent target acts as a constant reminder of the intention and is therefore maintains its activated state in long-term memory⁹². On the other hand, if the target is infrequent, as the on-going task proceeds, the intention drops progressively to lower attentional levels. Dysexecutive patients therefore are more likely to experience PM failures, by being impaired in upgrading the activation of their intention.

INTERVENTIONS BASED ON THE MODEL OF GOAL NEGLECT AND IMPAIRMENT IN VIGILANT ATTENTION

Goal Management Training (GMT) approaches to executive dysfunction aim to train adult patients to maintain goal activation, by compensating for vigilant attention impairment

through the use of content-free cues that can be either external, exogenous (such as auditory alerts^{95,96}, text messages with a “Stop” cue⁹⁷) or patient-generated (by training patients to regularly pause action and remind themselves of their goal ^{98,99}) - In GMT patients are encouraged to stop ongoing behaviour in order to define goal hierarchies³⁹ and monitor performance and regularly update current goals in working memory. Goal Management Training methods target executive dysfunction as well as prospective memory, because the underlying impairment (goal neglect resulting from deficient endogenous reactivation of relevant goals) is considered to be the same. While GMT is widely used in adults⁹⁸⁻¹⁰¹, the validity of interventions derived from Duncan’s model has never been explored in children. A number of questions arise: Is it possible to alleviate prospective memory impairment by using goal neglect-based interventions? Will an improvement in PM be accompanied by improvement in complex task management?

REHABILITATION OF EXECUTIVE DYSFUNCTION IN CHILDREN

Apart from visuo-spatial and verbal working memory ¹⁰²⁻¹⁰⁶, and attention¹⁰⁷⁻¹⁰⁹ where some approaches have proven effective, there is a lack of validated methods for EF rehabilitation in children ^{107,110-113}.

While in adults metacognitive strategies are recommended to alleviate EF dysfunction¹¹³⁻¹¹⁵, in children most metacognitive strategies described in the literature are theory-based and not validated. Evidence of their age-appropriateness is lacking. Many questions do not have an evidence-based answer yet: from a developmental point of view, from which age is a child able to learn to use a particular metacognitive strategy (MS), irrespective of brain injury status? Are brain injured children capable of understanding, learning and using a metacognitive technique efficiently, despite specific impairments in EF-related function including metacognition? Can metacognition (and especially metacognitive strategies use) be trained in children who have suffered a TBI? And from which age? Can metacognitive strategies be used in children who are not fully aware of their difficulties because of developmental immaturity and, in the case of TBI, additional deficits in organic anosognosia?

In adults, awareness it is an important factor mediating the effect of interventions: (1) individuals with acquired brain injury (ABI) who lack awareness of their deficits often demonstrate poorer motivation for therapy, resist treatment recommendations and develop fewer compensatory strategies¹¹⁶; (2) patients are more willing to strategies to compensate for their memory difficulties than to compensate for their dysexecutive syndrome¹¹⁷, possibly because it is easier to be aware of memory problems than of EF problems; (3) EF training programmes that has been demonstrated to be effective in adults with ABI include awareness interventions in order to obtain maximum motivation for the rehabilitation program^{118,119}. In children, very little is known about how to assess self-awareness and influence of self-awareness on intervention effects. Can self-awareness be assessed in children and how? Are children with TBI aware of the impairment caused by their executive dysfunction? What steps are needed for a child to become aware of his/her impairment? Will self-awareness influence the effectiveness of interventions aiming at compensating an EF deficit?

Given the frequency of executive dysfunction after TBI, the extensive consequences of EF impairment on academic achievement, everyday life and overall independence, and the lack of validated rehabilitation methods, there is an urgent need for research to progress in the domain of EF rehabilitation in children.

PHD OVERVIEW

Part one of this PhD aims to characterize executive dysfunction after childhood TBI and its impact on two specific related cognitive functions: prospective memory and self-awareness.

Chapter 1 examines the degree of EF deficit in children with severe TBI using a prospective longitudinal study, and explores the controversial age at injury effects. Chapter 2 examines the severity of PM impairment using an ecological test of EF, the Children's Cooking Task (CCT)^{120,121}, in a large convenience sample of children with TBI and other acquired brain injuries. Chapter 3 examines the frequency of PM impairment long-term post severe TBI using the same cohort as chapter 1. Finally, Chapter 4 is a pilot study that explores self-awareness in a small (N=5) sample of children with severe TBI and proposes a model of child anosognosia. Note that the data of this chapter 4 was gathered during a pilot EF intervention that is detailed in section 2. Together the aim of section one is to document, assess,

characterize and quantify impairment in EF, in PM and in self-awareness following childhood severe TBI.

Part 2 of this PhD aims to develop an intervention to improve executive functions after severe childhood TBI. First, chapter 5 gives an overview of current practices, promising approaches and challenges in EF rehabilitation in children (*NB: A French adaptation of this chapter can be found in the appendix*). Chapter 6 then reviews the effectiveness of approaches to executive dysfunction used in adults, based on Duncan's model of goal neglect (and especially Goal Management Training - GMT), that were considered as possible candidates for treating executive dysfunctions in children as well. Chapters 7 and 8 then present the experimental data on the GMT adapted to children, identify possible factors limiting intervention effectiveness and challenges in research on EF intervention and discuss the use of metacognitive strategies in children with TBI.

PART 1: CHARACTERISATION
AND ASSESSMENT OF
EXECUTIVE FUNCTIONS AND
TWO RELATED FUNCTIONS:
PROSPECTIVE MEMORY AND
SELF-AWARENESS

CHAPTER 1: EXECUTIVE FUNCTIONS FOLLOWING SEVERE CHILDHOOD TRAUMATIC BRAIN INJURY – THE TGE PROSPECTIVE LONGITUDINAL STUDY: AGE AT INJURY VULNERABILITY PERIODS

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INTRODUCTION

Childhood severe traumatic brain injury (TBI) is usually defined as a TBI with initial or lowest Glasgow Coma Scale (GCS) score ≤ 8 . The incidence of TBI is high (1.75 % per year)³. Most TBI are mild and the prevalence of severe TBI is not precisely known. Annual incidence of hospitalized TBI is estimated at 74/ 100 000, of which 17% are estimated to have severe TBI¹. In McKinlay et al. cohort³, moderate and severe TBI accounted for 10% of all TBI.

Executive functions (EF) are a collection of related but distinct abilities that allow individuals both to engage efficiently in intentional, goal-directed, problem-solving actions^{20,21} through conscious and effortful processing²² and to adapt to new, complex situations in the real world^{20,23}. According to Diamond's developmental model of EF²⁴, there are 3 core EF: inhibition, working memory and flexibility. (1) Inhibition enables us to selectively focus on what we choose and suppress attention to other stimuli and resist interference²⁴. One particular form of inhibition is Inhibitory control of attention which is interference control at the level of perception. Terminology and models of EF are numerous, but in this paper, we will call "attention", the function termed in literature "inhibitory control of attention", "attentional inhibition" or "focused"/"selective" attention which is an endogenous, top-down, active, and volitional, form of attention, that is part of inhibition in Diamond's model (as opposed to automatic attention, driven by a salient stimulus such as a klaxon)²⁸. Inhibition is extremely hard for young children²⁴, with a very rapid progress in inhibition up to 5 years of age²⁰, less rapid between 5 and 8 ad then slowed but continued progress during childhood and

adolescence²⁹. (2) Working memory (WM) involves holding information in mind and mentally working with information no longer perceptually present^{30,31}. Infants of only 9 to 12 months can update the contents of simple information. However, being able to hold many things in mind or perform mental manipulation is far slower to develop and shows a prolonged developmental progression³². (3) Cognitive flexibility (“thinking outside the box”) is being able to change perspectives and requires to inhibit our previous perspective and load a different perspective into WM²⁴. Flexibility builds on inhibition and WM and comes in later in children’s development³³. By age 4.5-5 years, most typically developing children perform simple flexibility tasks (such as identify both figures in an ambiguous figure or switch sorting dimensions³⁴, but not until 7 to 9 years of age, can children switch flexibly on a trial-by-trial basis as required by usual paradigms testing flexibility³³. From these three core EF, higher order EFs are built such as reasoning, problem solving and planning, which develop later, around the age of twelve¹²², although these skills exist and develop during early childhood as well²⁰.

EF are often impaired after TBI^{6,69} and influence a number of outcomes including academic achievement^{42,73,74} social cognition¹⁰, vocational outcome¹⁵ and ability to live independently. The level of executive dysfunction is found to be related to the severity of injury and younger age at injury^{12,14,71}. TBI in children takes place in the context of ongoing cognitive development, so as well as disrupting current abilities, TBI may interrupt or affect skills that are still developing or that are to develop in the future and therefore may have more unpredictable consequences than adult TBI that occurred in a mature brain (see Anderson for a review¹⁷). Relationship between age at injury and outcome is complex and literature begins to suggest that it is not linear^{19,80}. TBI sustained at an earlier age is prone to generalized delayed adverse outcome when cognitive functions have not yet developed at the time of injury but outcome may be even worse when the function is at a rapidly developing stage⁷⁷ at the time of injury. Anderson et al. compared intellectual, executive and behavioural outcomes after focal injuries across six age at injury groups (congenital to late childhood injuries) and found patterns of impairment that varied across domains, suggesting that different stages of brain development may be critical for different functions⁸⁰. For EF, Anderson et al. found that the 10-12 years at injury group had better EF compared to children injured at a younger age, but children aged 3-6 at injury performed better than those injured at 7-9⁸⁰. Crowe et al. found

that for intellectual ability (IQ), children injured in middle childhood (7-9 years) demonstrated poorer outcome¹⁸ than their older and younger peers. Because the relationship may not be linear, evidence is contradictory, some studies showing an increased risk of impairment with early age at injury^{80,123,124}, while others do not find an age at injury effect^{19,71,73,125}.

The aim of this study was to estimate EF impairment after childhood severe TBI, using a prospective, longitudinal, two-year follow-up of children with severe TBI, and to explore factors predicting EF outcome. Secondary aims were (1) to analyze differences in component EF development post TBI across assessment times; (2) to analyze differences when outcome is based on questionnaires or on performance-based tests only; (3) to compare recovery trajectories across age at injury groups, testing the hypothesis of greater vulnerability when TBI occurs during rapid cognitive development stage.

METHODS

All children consecutively admitted to the pediatric neurosurgical intensive care unit of University Necker Enfants Malades Hospital between January 2005 and December 2008, within the first 6 hours following TBI, and meeting the inclusion criteria were offered to participate in the study, and followed-up prospectively over two years. Children included in this study constitute the TGE cohort (this stands for “*Traumatisme Grave de l’Enfant*”, i.e. Severe Childhood Trauma). Inclusion criteria were: children aged 0-15 years, who had sustained a severe accidental TBI defined as Glasgow Coma Score (GCS) of 8 or lower and/or an Injury Severity Score¹²⁶ > 16. Exclusion criteria were: children with no vital signs upon admission, children with non-accidental head injury, previous history of diagnosed neurological, psychiatric or learning disability. The study was approved by the local ethics committee and parents gave their informed written consent to this observational study. At the initial acute phase, the following data was collected: lowest GCS score, coma duration. After acute care management, survivors in need of rehabilitation were referred to a specialized rehabilitation center for children with acquired brain injury in Saint-Maurice Hospitals, and followed at 3, 6, 12 and 24 months.

Demographic and environmental data was collected, such as parental education level (graduation from high school or not), and the language(s) spoken at home (French exclusively, or other language/bilingual). Children's EF were evaluated on a series of performance-based, paper-and-pencils tests of EF as well as more ecological questionnaires of EF completed by parents.

Regarding performance-based tests, there were no tests in French that could be administered across the full 0-17 years at testing age range, therefore not all children were administered the same tests (see table 1.1). Tests were selected for this study based on: large administration age range, adequate psychometric properties and previous use in children with TBI. The Wisconsin Card Sorting Tests (WCST), the Stroop and the Tower Test have been used in the same or similar form in major publications on childhood EF, both in developmental¹²² and TBI literature^{6,72}. The NEPSY-1¹²⁷⁻¹²⁹ was, at the time of inclusion, the main available assessment tool for EF. Its psychometric properties are well-established according to the Society of Pediatric Psychology Evidence-Based Assessment Task Force Workgroup¹³⁰, and it has been used in neurological populations¹³¹⁻¹³³. There are no validated N-back working memory tasks available in French. Therefore the Children's Memory scale¹³⁴⁻¹³⁶, often used in children with TBI¹³⁷⁻¹³⁹, was chosen, as its concentration-attention index includes classical working memory tests such as backwards spans and letter number sequences. Description of performance-based tests can be found in supplemental data 1.1.

Table 1.1: Executive Functions (EF) tests used to construct component EF Z-scores.

Inhibition	Flexibility	Attention	Planning	Working Memory
Stroop Test ^{140,141} Interference score (child version: 12-16; adult version: after 16)	Wisconsin Card Sorting test ¹⁴² , percentage of perseverative errors (8 – 89 years)	NEPSY-1 ¹²⁷ Visual Attention subtest (age range: 3-12)	NEPSY-1 ¹²⁷ Tower test (age range: 5-12)	Children’s Memory Scale ^{134,135} Attention/Concentration Index (age range: 5-16)
NEPSY-1 ¹²⁷ Statue subtest (age range: 3-12)	NEPSY-1 ¹²⁷ Design Fluency subtest (age range: 5-12)	NEPSY-1 ¹²⁷ Auditory Attention (age range: 5-12)	Lussier Tower of London ¹⁴³ : number of success (age range: 7-17, used from 13 in our study)	
	Total Number of trials in Lussier Tower of London ¹⁴³ (age range: 7-17 used from 13 in our study)			
BRIEF * ^{144–146} inhibition subscale (age range 5-18)	BRIEF ^{144–146} flexibility subscale (age range 5-18):	CBCL† ¹⁴⁷ Attention subscale (preschool: 1,5-5; school version 6-18)	BRIEF ^{144–146} planning subscale (age range 5-18)	BRIEF ^{144–146} working memory subscale (age range 5-18)

Note: Details and description of performance-based tests can be found in supplemental data 1. BRIEF: Behavior Rating Inventory of Executive Function, parental form; CBCL: Child Behavior Checklist

Supplemental Data 1.1: Performance-based tests of executive functions

Stroop tests requires children to name the ink color of a list of words representing colors (e.g.: If the word “blue” is written with a red color, the child must name it as “red”, thus inhibiting the prepotent reading response “blue”).

In the statue test, the child is to pretend s/he is a statue and not move, regardless of the distractions, during 75 seconds.

In the Wisconsin Card Sorting Test (WCST), the child is given cards with drawings differing in the type of geometrical form (square, triangle...), in the color of forms and in the number of forms. The child is expected to classify the forms according to different classification criteria (form, color, number) s/he must find her/himself. In order to succeed, the child must think flexibly and change classification system when s/he is cued to.

In the Tower tests (Lussier and NEPSY-1), analogs of the widely studied Tower of London and Tower of Hanoi^{148,149}, the child must replicate a tower, made of pegs and must plan the order in which s/he displaces pegs. The NEPSY-1 tower records only the number of succeed towers (measure of planning), whereas Lussier’s version allows also to screen for flexibility by reporting perseverative behaviors, indicated by the number of trials (a child with impaired flexibility may perseverate on the same error several times).

In drawing fluidity (NEPSY-1), the child must create as many different designs as possible by joining points in different ways.

The visual attention test (NEPSY-1) requires the child to cross target drawings (age-adapted complexity) among a list of non-target drawings.

In auditory attention (NEPSY-1), the child must pick colored forms from a box, following auditory commands (e.g. pick up squares when he hears “square” while listening to a list of words), increasing in complexity.

Working Memory (WM) was assessed using the Children’s Memory Scale measure of WM¹⁵⁰, referred to as the “attention/concentration index” in the manual: tests including retaining a series of numbers and of number-letters sequences as well as backwards digit span.

Regarding “ecological” questionnaires of EF, we used the Behavior Rating Inventory of Executive Functions (BRIEF) questionnaire^{144, 145} and the attention subscore of the Child Behavior Checklist (CBCL)¹⁴⁷. The BRIEF yields 8 subscales, leading to 3 composite scores: a Metacognition index (MI), a Behavioral Regulation index (BRI), and a Global Executive Composite score. Higher scores correspond to increased EF difficulties in daily life. The BRIEF has large normative data for children aged 5 to 18, high internal consistency¹⁴⁶, good validity¹⁵¹, good test-retest reliability¹⁴⁶ and is a recommended outcome measure for children with TBI¹⁵². It is the most commonly used questionnaire of EF and is sensitive to EF deficits

in children with TBI^{151,153,71,14}. The CBCL has high test–retest reliability and validity¹⁵⁴ and is the most commonly used test to assess behavior problems after TBI^{155,156}. Questionnaires were obtained only for children whose parents could read French.

All scores were transformed into Z-scores, in order to ease comparisons and to compute composite scores. This comprehensive assessment yielded a single score for each EF (inhibition, attention, WM, flexibility and planning), that incorporate performance-based tests and questionnaires. Composite EF scores were also calculated: a performance-based score (mean of all performance-based tests scores) and a global EF score (mean of all performance-based tests scores, BRIEF MI and BRI). The global and performance-based EF scores were computed only for children aged 3 or more at assessment because only one test could be administered before three and these scores would therefore not have been accurate.

STATISTICAL ANALYSIS

All analysis were computed on IBM SPSS Statistics 21. The few outliers were trimmed to three standard deviations from the mean for analysis.

Injury variables and outcome variables were analyzed for normality using Kolmogorov-Smirnov test, and quantified analysis of skewness and kurtosis divided by their standard error (1.96 criteria). Normality analysis revealed that coma duration and GCS were not normally distributed (skew towards shorter coma duration; skew towards higher GCS and significant kurtosis), therefore a dichotomized version of those variables was created, in order to meet the assumptions of the general linear model: coma duration ≤ 7 days versus ≥ 8 days and GCS ≤ 6 versus ≥ 7 . For all covariance analysis, covariates included: family characteristics (language spoken and parental education), injury severity characteristics (dichotomized coma duration and GCS). These covariates were used based on previous literature reporting their importance and on correlations with the global EF score in our sample (see supplemental data 1.2).

Scores of children with TBI were compared to norms by the one-sample t-test to detect if their mean Z-scores differed significantly from the normal population. A repeated measures ANCOVA was used to assess if scores changed over time. A Multivariate analysis of covariance (MANCOVA) was used to explore outcome on the 5 component EF, across 3 assessment times.

Age at injury effects were explored using a two-way mixed repeated measures Analysis of Covariance (ANCOVA) that examined change in global EF score and in component EF scores across assessment time and across age at injury groups. Age at injury was not used as a continuous variable, because previous research suggests its relationship with recovery is not linear, some age at injury groups being more vulnerable. Children were divided into 5 age-at-injury groups, the first 4 were based on age groups previously used in research on age at injury effects^{80,157}, based on neurosciences developmental data¹⁵⁸⁻¹⁶⁰: 0-2 years, 3-6 years, 7-9 years, and 10-12 years. A fifth group (13-15 years at injury) was necessary as our sample used a larger age range than previous research on age at injury groups. Age at injury groups' differences on covariates were tested by Chi squared test.

Recovery was assessed by the difference in Z-scores between 24 and 3 months, indicating how much progress a child had made, rather than how close to norms s/he was at 24 months post-injury. Differential recovery trajectories were explored for this progress score for the 5 age groups, using ANCOVA. In the 0-2 year's group, only two children were old enough to be tested at both 3 and 24 months, enabling calculation of an EF progress score, thus this age group was not considered in EF progress analysis.

Effect sizes for ANCOVAs were calculated using partial η squared (η^2_p) and interpreted by Cohen's guidelines as follows: small: 0.01; medium: 0.06; large: 0.14.

A hierarchical multiple regression was used to analyze factors influencing outcome by entering in step one family characteristics (language spoken and parental education), in step two injury severity characteristics (dichotomized coma duration and GCS) and in step three, five age at injury group dummy variables. Missing values were excluded listwise. For predicting progress, only children aged >2.75 years at injury were included, as this was the minimal age allowing EF progress calculation from 3 to 24 months.

RESULTS

PARTICIPANTS

Eighty-one children were included at the acute stage of TBI between January 2005 and December 2008. Sixteen children died during acute care, leaving 65 children at the 3 months

assessment. Of those 65, eleven were under the age of 3 at 3 months post-injury, which did not allow a reliable global EF score to be computed. Among the older children, two were in a minimally responsive state, precluding any cognitive assessment. At 12 months post-injury, one child was lost to follow-up, for one child parents withdrew consent and nine children were still under three. All remaining children could by then be tested. At 24 months, 2 more children were lost to follow-up, and three were still under of the age of three. Characteristics of included children are presented in table 1.2.

Table 1.2: Participants demographic and injury severity characteristics.	
<i>Demographic Variables</i>	
Gender: Boys; N (%)	43 (66%)
Age at injury (years): Mean (SD; range)	8.1 (4; 3 months - 14 years 11 months)
Number of children in each age at injury group: 0-2 / 3-6 / 7-9 / 10-12 / 13-15	13/12/16/10/14
Language spoken at home (N=64)*: French/other or bilingual; N (%)	37 (58%)/ 27 (42%)
Country of origin of parents (N=58)*: France/ at least one parent from another country; N (%)	29 (50%)/29 (50%)
Parental education (N=63)*: under/above high school graduation; N (%)	34 (54%) / 29 (46%)
<i>TBI medical and severity characteristics</i>	
Glasgow Coma Scale score [GCS; mean (SD; range)]	6.7 (1.41; 3 – 9)
GCS dichotomized (≤ 6 ; ≥ 7); N (%)	26 (40%) / 39 (60%)
Coma duration [days; mean (SD; range)]	6.6 (4.83; 1 – 23)
Coma duration dichotomized (≤ 7 days; ≥ 8 days); N (%)	46 (71%) /19 (29%)
<i>TBI immediate outcome</i>	
Admitted to a rehabilitation center after acute care: yes/no; N (%)	54 (83%) / 11(17%)
Performance IQ at 3 months post injury; mean (SD)	83.1 (16)
Verbal IQ at 3 months post injury; mean (SD)	88.0 (19)
Full Scale IQ at 3 months post injury; mean (SD)	85.2 (18)

Note: * data missing in a few cases; IQ: Intelligence Quotient

Supplemental Data 1.2: Correlations between global EF score and demographic and injury variables

Spearman's Rho	Global composite score at 3 months (Nmin= 50) ‡	Global composite score at 12 months (Nmin= 52) ‡	Global composite score at 24 months (Nmin= 57) ‡	Progress on EF global score. (Nmin=52) ‡
Parental education†	.38*	.31*	.33*	0.16
Language spoken at home†	-.28*	-0.17	-0.14	0.10
GCS (continuous)	0.20	0.12	0.24*	-0.11
Coma Duration (continuous)	-0.23	-0.24*	-0.07	0.25
Age at injury (continuous)	0.11	-0.04	0.22	0.00
Coma duration < 7 days or ≥ 8 days†	-0.27*	-.28*	0.00	.45*
GCS dichotomized (≤ 6; >7) †	-0.31	-0.13	-0.34*	0.10

† Point-biserial correlations

*p<0.05

‡ children aged <3 years at assessment time were excluded from analysis, as few test could be administered in this age range, leading to inaccurate composite score

COMPARISON WITH NORMS AND CHANGE OVER TIME

Composite EF scores and BRIEF T-scores are reported in table 1.3, and component EF scores are displayed in figure 1.1. Patients did not differ from the norms on planning at any assessment time, whereas they did for the global EF score, and the attention and working

memory scores at all time points ($p < 0.001$). Performance-based scores differed significantly from the norms only at the 3 months assessment.

Table 1.3: Composite scores and BRIEF T-scores at 3, 12 and 24 months post injury.

	3 months			12 months			24 months		
Z-scores	N	Mean	95% CI	N	Mean	95% CI	N	Mean	95% CI
Global EF score †	52	-0.47**	-0.67; -0.26	54	-0.26*	-0.45; -0.08	58	-0.34*	-0.54; -0.13
Performance-based EF score †	52	-0.36**	-0.56; -0.16	54	0.05	-0.13; 0.23	58	-0.11	-0.31; 0.08
T-scores	N	Mean	95% CI	N	Mean	95% CI	N	Mean	95% CI
BRIEF BRI	28	57*	53; 61	34	57*	53; 62	35	58*	53; 63
BRIEF MI	28	55*	50; 60	34	55*	51; 58	35	55*	51; 59
BRIEF GEC	28	56*	52; 61	34	56*	52; 60	35	57*	52; 61

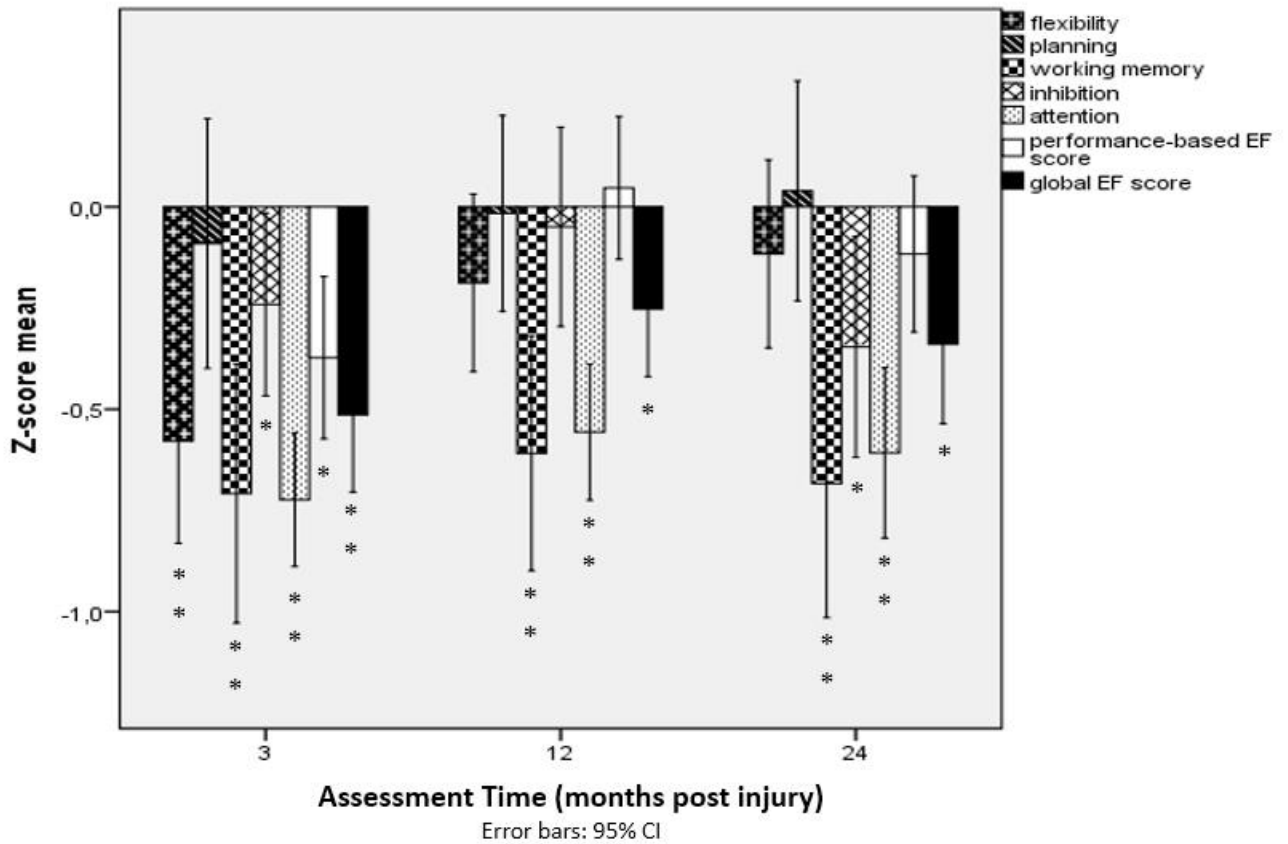
*Score statistically different from norms, $p < 0.05$

** Score statistically different from norms, $p < 0.001$

† Children aged < 3 at assessment time were excluded from analysis, as few tests could be administered in this age group, leading to inaccurate composite score.

Note: BRIEF= Behaviour Rating Inventory of Executive Functions; MI= Metacognition index; BRI= Behavioral Regulation index; GEC=Global Executive Composite Score

Figure 1.1: EF Z-scores across assessment times compared to norms



Statistically different from norms: * $p < 0.05$ ** $p < 0.0001$

Note: significance for attention at 24 months should be interpreted with caution as its distribution was bimodal and violated assumption for normality.

Performance-based score change across time was significant, [$F(2,86)=4.48$, $p=0.014$, $\eta^2_p=0.09$], with significant pairwise Sidak-adjusted comparisons showing a difference between 3 and 12 and 3 and 24 months ($p < 0.001$), but no significant difference between 12 and 24 months. Global EF score improved between 3 and 12 months [Huynh-Feldt $F(1.64, 91.2)=6.35$, $p=0.03$, $\eta^2_p=0.12$; Sidak-adjusted comparison $p=0.01$], but the change between 3 and 24 months was not significant ($p=0.09$). BRIEF indices differed from norms at all assessment times. Changes in BRI and MI across time were not significant. Detailed scores for BRIEF subscales can be found in supplemental data 1.2.

Supplemental Data 1.2: BRIEF subscores at 3, 12 and 24 months post injury

	3 months N=28		12 months N=34		24 months N=35	
	Mean (SD)	95%CI†	Mean (SD)	95%CI†	Mean (SD)	95%CI†
BRI	57 (12)*	53 ; 61	57 (13)*	53 ; 62	58 (14)*	53 ; 63
MI	55 (12)*	50 ; 60	55 (12)*	51 ; 58	55 (12)*	51 ; 59
GEC	56 (12)*	52 ; 61	56 (13)*	52 ; 60	57 (13)*	52 ; 61
Inhibition	57 (12)*	52 ; 61	56 (13)*	51 ; 60	56 (12)*	52 ; 61
Shift	57 (10)*	54 ; 61	58 (12)*	54 ; 62	57 (13)*	52 ; 61
Emotional Control	56 (12)*	52 ; 61	56 (12)*	52 ; 60	58 (15)*	53 ; 63
Initiation	53 (12)	48 ; 58	54 (13)	50 ; 58	53 (13)	49 ; 58
Working Memory	57 (11)*	52 ; 62	58 (13)*	54 ; 62	60 (15)*	55 ; 65
Plan/Organize	56 (13)*	50 ; 60	54 (12)*	51 ; 58	53 (11)	50 ; 58
Organization of Materials	52 (10)	47 ; 56	49 (9)	46 ; 52	51 (10)	48 ; 55
Monitor	55 (12)*	51 ; 60	55 (12)*	51 ; 58	54 (12)*	50 ; 59

*Score statistically different from norms, $p < 0.05$

†BCa 95% Confidence Interval, based on 1000 bootstrap samples

Note: Standard error for all means = 2

BRIEF= Behavior Rating Inventory of Executive Functions; MI= Metacognition index; BRI= Behavioral Regulation index; GEC=Global Executive Composite Score

INFLUENCE OF DEMOGRAPHIC AND INJURY CHARACTERISTICS ON COMPONENT EF

Using Roy's largest root, MANCOVA revealed a significant effect of parental education [$\Theta = 0.15$; $F(4,111)=4.07$, $p=0.004$] and assessment time [$\Theta=0.11$; $F(4,112)=2.98$, $p=0.022$] on component EF. Note attention was removed from the MANCOVA as its distribution was binomial at 24 months and violated the assumption of normality. Follow-up ANOVA revealed children whose parents had a lower education showed significantly worse scores on all EF components than those with higher education level, [$F(1,114)=5.62$ for flexibility, 5.53 for planning, 15.06 for WM, 6.44 for inhibition (all $ps < 0.05$, with WM $p < 0.001$)]. Planned simple contrast revealed that flexibility scores improved significantly between 3 and 12 months ($p=0.034$) and 3 and 24 months (0.007), while repeated contrast revealed that the change from 12 to 24 month was not significant. For other EF components, change in score was not significant.

AGE AT INJURY EFFECTS

Age at injury groups did not differ significantly in terms of demographic and severity factors, except for parental education, which was significantly lower in the 7-9 years age at injury group ($p=0.03$).

AGE AT INJURY EFFECTS ON REAPETED MEASURES OF EF

A two-way mixed repeated measures ANCOVA was used to examine change in EF composite score across assessment times and across age at injury groups, Mauchly's test of sphericity was significant ($p = 0.040$), therefore Huynh-Feldt estimate was used for significance. All covariates made a significant contribution, as denoted by time x coma duration interaction [$F(2,90)=11.01$, $p < 0.001$, $\eta^2_p = 0.20$]; main effect of GCS [$F(1,45)=6.85$, $p=0.012$, $\eta^2_p = 0.13$] and of parental education, [$F(1,45)=5.61$, $p=0.022$, $\eta^2_p = 0.11$].

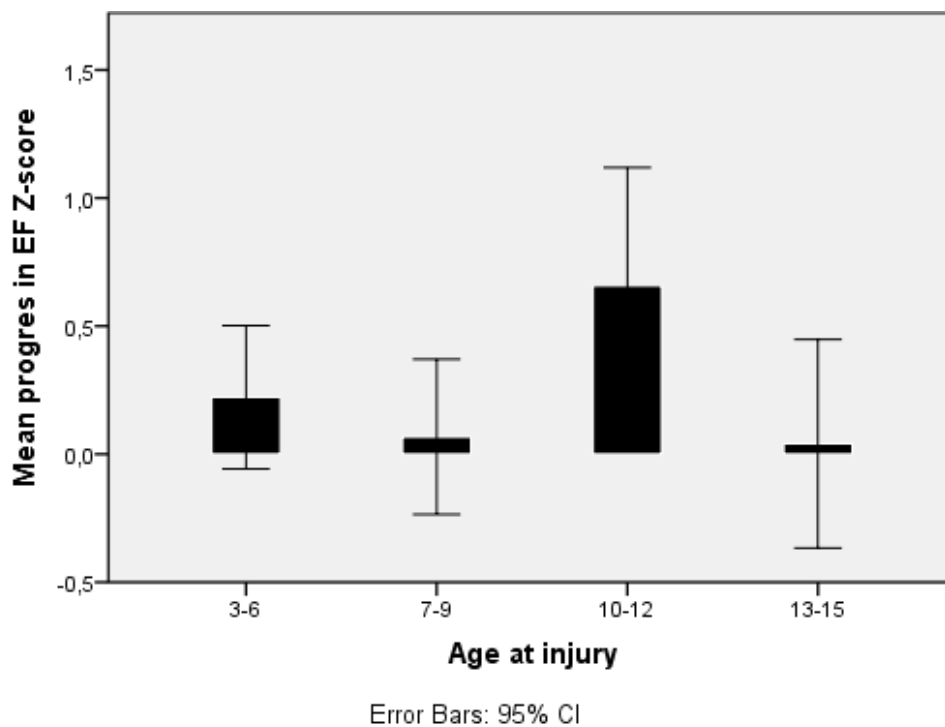
There was a significant time x age at injury group interaction [$F(8,96)=3.65$, $p=0.001$, $\eta^2_p=0.23$]; however, none of the planned contrasts was significant. Main effects of age at injury group were not significant. There was a significant and large effect of assessment time, [$F(2,90)= 8.40$, $p < 0.001$, $\eta^2_p = 0.16$]. Post hoc comparisons adjusted with Bonferroni method showed a significant progress in the global EF score only between the 3-and 12-months

assessments: mean difference in Z-score=0.30 [0.14-0.45], SE=0.06, $p<0.001$ (with Bonferroni adjustment for multiple comparisons). The difference in scores between 12 and 24 months was not significant, nor was the difference between 3 and 24 months, although there was a trend towards better scores at 24 months, with a Z-score mean difference =0.20 [0.15-0.46], SE = 0.09, $p=0.086$.

AGE AT INJURY EFFECTS ON OVERALL EF PROGRESS

There was a significant large effect of age at injury group on the amount of EF progress [$F(3,38)=4.10$, $p=0.01$, $\eta^2_p =0.25$]. Planned repeated contrasts revealed that children injured 3-6 recovered similarly to those injured 7-9 (see figure 1.2), but children injured 10-12 recovered better than those injured 7-9, and than those injured 13-15 (difference in Z score progress =0.71, $p=0.003$ and 0.65, $p=0.006$ respectively). The 13-15 age at injury group seemed to show nearly no recovery.

Figure 1.2: Progress in EF global score between 3 and 24 months according to age at injury group



AGE AT INJURY EFFECTS ON COMPONENT EF

A repeated ANCOVA was used for each component EF across 3 assessment times. There was no time x age at injury group interaction. Age at injury effect was not significant.

MODEL PREDICTING EF SCORES AT 3, 12, 24 MONTHS AND AMOUNT OF EF RECOVERY OVER TIME

Family characteristics alone did not significantly predict EF outcome, however when combined with injury severity, the model accounted for 34% of the variance in EF at 3 months, and for 23% of the variance at 24 months.

Regarding improvement over time, the regression model showed that better EF progress between 3 and 24 months was predicted by longer coma duration and age at injury group 10-12. Detailed results of the regression can be found in table 1.4

Table 1.4: Regression Models

	F	Sig.	ΔR²	Sig	Standardized Beta	t	Sig.
EF at 3 months model							
Step 1	3.91*	0.0026	0.13*	0.026			
Step 2	6.48*	0.000	0.21*	0.001			
					Parental Education*	0.26	2.14 .037
					GCS*	0.34	-2.89 .006
					Coma duration*	-0.29	-2.39 .021
Step 3	3.24*	0.05	0.19	0.845			
EF at 12 months model : all steps non-significant							
EF at 24 months model							
Step 1	2.29	0.111	0.076	0.11			
Step 2	3.95*	0.007	0.15*	0.008			
					Parental Education*	0.28	2.18 .034
					GCS*	0.40	-3.24 .002
Step 3	2.82*	0.011	0.085	0.21			
					GCS*	0.35	-2.87 .006
					Age at injury 10-12*	0.35	2.36 .022
Amount of EF recovery over time model							
Step 1	0.27	0.767	0.12	0.767			
Step 2	2.34	0.071	0.17*	0.019			
					Coma duration*	0.45	2.96 .005
Step 3	3.40*	0.06	0.20*	0.013			
					Coma duration*	0.46	3.33 .002
					Age at injury 10-12*	0.50	3.15 .003

Note: Step 1 = family characteristics (language spoken and parental education); step 2 = injury severity characteristics (dichotomized coma duration and GCS); step 3 = five age at injury group dummy variables.

**Significant at 0.05*

DISCUSSION

Children had significant impairment on WM, inhibition, attention and global EF following severe TBI. For flexibility and performance-based tests, children were impaired at 3 months only and improved to reach scores in the normal range by 12 months. No impairment was found on planning at any time. Global EF was predicted at 3 and 24 months by GCS score and parental education. Global EF outcome at 24 months and progress on EF were also predicted by age at injury but the relationship was not linear, children aged 10-12 at injury showing better outcome than older and younger children. Coma duration predicted global EF at 3 months but was no longer a significant predictor at 24 months.

STRENGTH OF THE STUDY

This was a prospective longitudinal study including a large group of children with severe TBI. Larger cohorts of childhood TBI published so far usually include heterogeneous samples of children with a range of TBI severity, where children with severe TBI are usually relatively few compared to the rest of the group (42 severe out of 330¹⁶¹, 50 out of 132⁴², 18 out of 54⁷¹, 13 out of 40¹⁶², 33 out of 122¹²³, 14 out of 59⁷⁹, 13 out of 40¹⁹, 33 out of 63¹⁴, 23 out of 87⁷²). Further, most samples focus on a restricted age range. Larger samples of severe TBI (N=118)¹⁶³ did not assess EF and were not prospective studies, with an important selection bias (towards very severe injuries and inclusion of children hospitalized in an intensive rehabilitation department). Children in this sample were aged 0-15 years, and were included prospectively immediately after uniformly severe TBI. The prospective longitudinal nature of this study allowed capturing positive outcomes compared to retrospective studies based on the inclusion of patients in rehabilitation^{73 163}. This is to our knowledge the largest study assessing EF prospectively in children with severe TBI. The disadvantage was the lack of uniform EF measures across the sample, which is the main reason for age restriction in other studies. Nonetheless, the use of a global EF score allowed comparing children who had different tests. Using both performance-based tests of EF and ecologically-valid questionnaires to assess EF, the global EF score was likely to be representative of true EF impairment.

Finally, this sample was representative of the TBI population in the Paris region, as children whose parents did not speak French were not excluded, contrary to other studies¹⁶¹. This factor did not allow obtaining questionnaire data for the whole group, however all children could be tested at least on standardized tests. As part of non-French speaking and low education parents are likely to have more difficulty to apply for help and understand counselling regarding their child because of language restrictions, it is important that those children are not excluded from trials.

EF TESTS

PERFORMANCE-BASED TESTS VERSUS QUESTIONNAIRES, THE ISSUE OF NOVELTY

The performance-based EF score showed a significant recovery at 12 and 24 months compared to the 3 month assessment, with no residual difference when compared to norms. This is consistent with previous reports that children's scores on performance-based tests of EF improve over time^{79,164}. It is difficult, however, to interpret reported improvements on performance-based measures of executive functioning, because better scores may be related either to recovery, or increased practice with the tests. As the whole purpose of EF tests is to prevent lapses into automaticity and promote conscious, novel and effortful processing^{22,165}, the scores at 12 and 24 months may not be reliable because requirement of novelty of EF tests to make significant demands on EF^{166,165,93,167,22} was not met. Further, in Vriezen and Pigott's¹⁵³ study, performance-based tests underestimated the child's level of executive functioning when compared to questionnaires of daily EF, perhaps due to the highly structured nature of standardized assessment in performance-based tests. Another explanation to consider is the comparison to norms instead of to a matched control group, which would have allowed controlling for the practice effect.

Contrary to performance-based tests of EF, no recovery was found on BRIEF questionnaires, which is consistent with studies reporting no improvement in EF (measured by BRIEF) during the first year after TBI¹⁶¹. The major limitation of questionnaires, which was obvious in our study, is the need for parents to speak the language of questionnaire.

When using the global EF scores that integrated both questionnaires and performance-based scores, our findings were somehow in-between, as there was a trend towards improvement between 3 and 24 months, although it did not reach significance. These results confirm previous concerns about EF performance-based tests' validity and promote their use combined with questionnaires. The significant improvement found between 3 and 12 months but not between 3 and 24 months is not surprising: it confirms Babikian and Asarnov meta-analysis that showed initial good recovery followed by an absence of age expected progress, resulting in lower Z-scores⁶ at longer term follow-up.

RELATIONSHIP BETWEEN EF AND IQ

Most EF tasks involve complex, overlapping and multi-faceted functions that tap both executive and non-executive processes¹⁶². Isolating the influence of these processes is difficult^{20,122} and purely "executive" tasks do not exist¹⁶². The current findings of EF impairment may be impacted by the patients' impaired general cognition (general intellectual skills, visual-spatial processing, processing speed, memory...), which may have exaggerated the observed deficits. Previous research has shown that intelligence is inextricably linked to EF in childhood³⁶⁻³⁸ and the view that EF are independent has been questioned in the literature, even in adults^{39,40}. Further, a number of studies have used IQ subtests to assess EF^{22,34,58}. For these reasons IQ was not used as a covariate in the analyses.

COMPONENT EF EVOLUTION OVER TIME

Ewing-Cobbs et al.⁷⁷ found that children tested 1-2 years post-injury performed worse on measures of inhibitory control and working memory, but not on measures of cognitive flexibility, when compared to controls. Our findings are similar, with important recovery for flexibility across time but persistent inhibition and WM deficit at 24 months post-injury. Similarly to Ewing-Cobbs's⁶⁹ and Anderson's³³ studies, flexibility was found to be quite robust to TBI effects, with rapid significant recovery across assessments and no significant deficit by 12 months post-injury. Because inhibitory control and working memory undergo rapid development during early childhood, but flexibility has a relatively flatter developmental trajectory, Ewing-Cobbs and colleagues interpreted this finding as supporting the idea that skills in a rapid stage of development are more vulnerable to the effects of TBI. Our study may support this view. A large meta-analysis also found largest effects on WM⁶.

Similarly to other studies, we did not find planning impairment⁷⁹. This skill is thought to emerge later, around 12 years of age¹⁶⁸ and therefore deficits may not be clearly apparent before that age. A meta-analysis has found large effects on planning, becoming apparent only after 24 months⁶ post-injury, but overall the children in the included studies were older than in our study. As 60% of the children in our study were under 12 by the 24 months testing, a longer follow-up may have allowed detecting significant impairment as children grew older. Another explanation could be the lack of sensitivity of the tests used, especially for the NEPSY-1 Tower test that has failed to produce lower scores for the clinical groups in Schmitt et al. study¹⁶⁹ compared to controls. However, this is unlikely as our planning score also included other tests than the NEPSY-1 tower.

In this study, attention was impaired at all assessment times, compared to norms. Our results are consistent with Babikian and Asarnov meta-analysis who reported attention impairments following severe childhood TBI⁶, although other studies reported opposite results^{71,123,162}. As attention is a complex construct, including different types of attention, the limited number of performance-based tests used in past studies may not have captured attention deficits. Indeed, some of the tests used in previous studies can be argued to offer a poor reflection of attentional skills (e.g. forward digit span only^{79,162}). In our study, performance-based tests of attention were combined with a more ecological measure of everyday attention, which probably led to a more sensitive reflection of attentional deficits experienced following severe TBI, as already suggested in previous research¹²³. To support this interpretation, we reran the analysis without including the CBCL attention subscale and found no attention deficit compared to norms. The lack of sensibility of performance-based tests is an on-going concern in attention and EF research^{161,170,171}.

AGE AT INJURY

A number of studies did not report effects of age at injury. This is probably due to the restricted ranges of age at injury used (1-7 years^{19,71}, 3-6⁷², 0-6 years⁷³, 12-17¹²⁵), as studies using larger age at injury ranges found an effect both for general intellectual ability^{80,123,157,172} and EF⁸⁰. Previous research has often assumed that age effects are linear, that is, younger age at injury leads to poorer outcomes. However, literature has now shown the child's brain step-

wise maturation, with critical maturational periods¹⁷³⁻¹⁷⁵, separated by less rapid development periods, which makes a linear relation between age at injury and cognition unlikely.

Crowe et al.¹⁵⁷ found that the 7-9 years at injury period is the most vulnerable to adverse outcome after brain injury compared to younger and older at injury periods, but also found a better recovery for 10-12 years at injury (better processing speed and performance IQ), which is consistent with our results of best EF recovery for children injured 10-12. Visual inspection of progress (figure 1.2) seemed indeed to show very little recovery in the 7-9 years at injury group. In Crowe's study there wasn't a 13-15 age at injury group to compare our results to: in our study this group's recovery seemed very close to that of 7-9 years at injury. Nonetheless their progress was significantly worse than the 10-12 group. After the age of 12, higher order EF develop, such as planning, higher order attention; frontal lobes show important myelination during adolescence. Therefore, the 13-15 group may have been more vulnerable to brain injury because of rapidly developing EF skills at the time of injury. Unfortunately, most studies do not include large age ranges and adolescents have rarely been compared to younger age groups. Overall, our results seem to support age at injury as an important predictor of executive functioning after childhood severe TBI, but with a nonlinear pattern of vulnerability. These results support the theory of children's brain increased vulnerability during rapid EF development, or more precisely, that when a cognitive function is not in a rapidly developing stage (10-12 group), EF are less vulnerable to adverse effects of severe TBI. Adolescence may represent another higher risk group, comparable to the 7-9 age group.

We did not find an age at injury effect for component EF. This may be due to a lack of power as a number of tests could not be administered before the age of 5, resulting in smaller sample size than for the global EF score. Another explanation is that the age at injury groups, defined by overall developmental stages^{80,157}, did not represent meaningful periods for each EF component, which develop at different speeds and different critical maturation periods. Literature does not have a clear agreement on developmental stages of each EF, as results are very dependent on how an EF is assessed: not all tests make the same demands on a same EF, not all tests have the same developmental discriminant validity, and most of tests rely on other cognitive processes and other EF.

PREDICTORS OF EF

Our study used a simplified dichotomous measure of socio-economic status (SES) using parental education. Effect of SES confirms earlier literature^{72,176} that found that more educated home environment is associated with better cognitive performance. Parental education influenced all components of EF, especially WM, and was a significant predictor of EF at 3 and 24 months.

Coma duration has been found to significantly predict EF in past research in children^{163,177} and adults¹⁷⁸ with brain injuries, even if different results have been found using the BRIEF questionnaire exclusively in a mixed sample of children with mild and severe TBI¹²⁴. In our study, longer coma duration predicted worse EF at first (3 months) but no longer at last (24 months) assessment, suggesting that after initial increased EF difficulty, in the long term, longer coma duration is not predictive of poorer outcome following severe TBI. Further, progress on EF was significantly larger when coma duration was longer, probably because of lower EF at 3 months yielding mathematically larger difference between the 2 assessments. As such coma duration effect on progress was somehow artificial. Babikian and Asarnov meta-analysis also found largest recovery for most severe TBI, who had the most impaired scores at initial assessments⁶.

Most studies have associated GCS<9 (severe TBI) with poorer outcome^{179,71,73}, but most of them compared children with mild and moderate injuries to those with severe TBI. GCS has been shown to predict outcome in TBI on a range of dependent variables¹⁷⁹, including general intellectual skills^{73,157}, EF^{71,79} and mathematics skills⁷³. Our study shows further that among children with severe injury, GCS ≤6 is a significant predictor of worse EF immediately after injury (3 months) and at 2 years, compared to those with a GCS of 7-9.

Overall in our sample, higher GCS, higher parental education and age at injury 10-12 (i.e. probably during less rapid brain maturation) predicted better EF at 24 months. Only age at injury (10-12 group) predicted amount of EF progress between 3 and 24 months post injury, when coma duration's artificial effect was removed.

RECOMENDATIONS

Literature published so far shows a lack of consistency in the way component EF are conceptualized and assessed^{6,71,72,124,162}. Future studies should aim at selecting tests that are validated measures of EF, and not IQ subtests thought to reflect EF. Ecologically valid tests, such as the BRIEF questionnaire should be included, as performance-based tests alone tend to underestimate EF impairment. Because of language barriers with questionnaires, the combined use of performance-based ecological tests with strong psychometric properties, would make a great contribution to the field^{120,180,181}. The issue of retest effect in EF tests remains an important concern for results interpretability, and future research should encourage development of parallel forms of ecological EF tests.

STUDY LIMITATIONS

Relying on norm-referenced tests of EF and comparing the TBI group scores to norms instead of using a prospective control group may have underestimated EF difficulties. Massagli et al.¹⁷⁹ who compared the magnitude of impairment in a cohort of children with TBI using both methods, found that using population norms underestimated impairment compared to use of controls. Similar findings have been reported in other causes of acquired brain injury, with a difference magnitude of up to 1-2 SD.³⁸

We did not have a measure of pre-injury functioning and therefore could not include pre-injury factors in our analysis, while pre-injury factors such as cognitive reserve have shown to mediate effects of TBI^{79,172}. Also the measure of family environment variables was rough as only parental education was included, rather than measures of family functioning, which has been shown to be crucial in TBI outcome^{72,182}. Family functioning questionnaires were not available in French at the time of the study planning (and the same language issue would have limited the findings to a subgroup).

Since the study has been performed, guidelines have been published regarding valid outcome measures recommended in childhood TBI¹⁸³, and could be used in future studies. At the time of the study, few tests of EF were available in French. Results based on NEPSY-1 should be interpreted with caution, as the hypothesized five-factor structure outlined in NEPSY-1

manual (EF being one of the factors) was never evaluated by the authors and Stinnett et al.¹⁸⁴ provide evidence for a single factor for the NEPSY-1. Further some of the discriminant validity findings of NEPSY-1 are weak (e.g.: statue test discriminates children with dyslexia from controls but not children with ADHD from controls); and NEPSY-1 validity in TBI is based on small samples (n=9 for attention and tower test). EF impairment may have been underestimated due to NEPSY-1's insufficient discriminant validity. Finally, a number of children were assessed on performance-based tests only, either because their parents were not sufficiently fluent in French, or because the BRIEF questionnaire can only be used from age 5. As performance-based tests of EF are known to underestimate EF impairment, the overall EF impairment in our study was probably underestimated. Children whose parents do not sufficiently master the French language are, however, an important and challenging population clinicians need to take care of and future studies should make all possible efforts to integrate them in longitudinal studies, in spite of assessment difficulties.

CONCLUSIONS

EF are impaired following childhood severe TBI. While flexibility and planning seem quite robust to TBI effects, impairment on WM, inhibition, attention and global EF is persistent at 24 months post injury. Longer coma duration is a pejorative factor shortly after injury, but does not seem to influence EF on the longer term (24 months). Parental education is an important predictor of EF recovery. Ewing-Cobbs's hypothesis of greater vulnerability of the developing brain during phases of rapid development is supported by our results, with children aged 10-12 (corresponding to a relatively stable phase of cognitive development) at injury showing better progress from 3 to 24 months, irrespective to how impaired they were at 3 months, and better EF at 24 months. Relying only on performance-based EF tests can underestimate EF impairment and our study confirms concerns about ecological validity of usual EF tests.

CHAPTER 2: ECOLOGICAL PROSPECTIVE MEMORY ASSESSMENT IN CHILDREN WITH ACQUIRED BRAIN INJURY USING THE CHILDREN'S COOKING TASK

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BRIEF REPORT

Ecological prospective memory assessment in children with acquired brain injury using the Children's Cooking Task

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Abstract

Prospective memory (PM) has been shown to be impaired in children with acquired brain injuries (ABI) and is a major concern for parents. Few studies have addressed this issue and most used tasks that are not ecologically valid. The aims of this study were (1) to explore if children who have sustained an ABI suffer PM impairment, measured both by the Children's Cooking task (CCT) PM score and using the 2 PM subtests of the Rivermead Behavioral Memory Test (RBMT), and (2) to explore if the CCT PM score is sensitive to developmental changes in PM in typically developing children and in children with ABI. Fifty-four children with ABI and 33 typically developing controls participated in the study. Children with ABI had significantly lower PM scores and poorer performance in the CCT than their typically developing peers. PM scores increased significantly with age, indicating developmental progress of PM performance.

Keywords

Acquired brain injury, children, cooking, ecological, executive functions, prospective memory

History

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INTRODUCTION

Prospective memory (PM) is the ability to remember to perform intended activities⁶⁴. Children with PM impairment can fail to deliver important messages to parents, forget appointments, or fail to bring necessary items for planned activities. PM tasks require retrospective memory to remember the task, but depend on executive functions (EF)⁶⁵ for successful goal maintenance, retrieval and implementation at the right moment. PM depends upon frontal lobe integrity¹⁸⁵, with a key role for rostral prefrontal cortex (BA10)¹⁸⁶.

Literature on prospective memory in typically-developing children shows improvement in PM performance across the age range from 2 to 12 years^{50,66}: children become increasingly skilled at using external reminders to cue PM and increasingly proficient at applying time-checking strategies. In typically-developing children aged 6-12, performance on EF tasks such as planning and switching⁴⁷, working memory⁴⁸, and inhibition⁴⁹ is correlated with PM⁵⁰. PM and retrospective memory are not strongly associated in typically developing children¹⁸⁷.

PM has been shown to be impaired in children with brain injuries^{85,86}, even after cues are given¹⁸⁸, and even under strong incentive conditions¹⁸⁹. PM problems are reported as a major concern by the parents of children with traumatic brain injury (TBI)⁸⁷. Adult patients with ABI generally obtain lower results than control participants on experimental PM tasks¹⁹⁰, detecting fewer prospective cues and retrieving fewer actions¹⁹¹. The majority of memory failures they report are prospective in nature¹⁹². The capability to compensate for PM deficits in adults significantly predicts the ability to live independently⁸⁸. Parents of children with TBI report serious concerns for their child's safety and ability to be left unsupervised even briefly because of substantial PM impairments⁸⁷. Children with ABI may display normal performance on standard neuropsychological assessment, and still be impaired on functional memory¹⁹³ and especially PM.-In spite of these reports, very few standardized tests address PM issues, especially in naturalistic settings¹⁸⁰.

McCauley et al.^{85,189,194} used a monetary incentive to increase children's performance on an event-based PM task consisting of asking the examiner for points (exchangeable for dollars or for pennies) before each new neuropsychological test. Although the incentive was naturalistic, the testing used an office-based setting and a verbal non-ecological task. A promising

approach for studying PM is the use of virtual reality and computer games to assess PM (e.g.: TEMP task¹⁹⁵ in adults, simulating driving around shops to prepare a dinner or a holiday; the CyberCruiser^{66,50} in children simulating driving a vehicle across a variety of scenes to explore, and requiring to remember to refuel the vehicle after a cue), but unfortunately this approach is not yet used in clinical practice. A functional memory test that has been used in children with acquired brain injuries (ABI)^{196,197} is the Rivermead Behavioural Memory Test (RMBT) for Children^{198,199}: it has shown to be sensitive to everyday memory problems^{196,193}, and it includes two subtests tapping prospective memory. The RMBT however is an office-based test, performed in a very structured environment, and it may not fully capture children's PM in real, motivating, complex and playful activities. Recently it has been proposed to use a real cooking task (Children's Cooking Task – CCT) to explore EF in children with ABI^{120,121}, as children find it fun²⁰⁰ and because cooking is a novel open ended task, requiring adaptability and innovative strategies that challenge the child's EF.

The aims of this study were (1) to explore if children who have sustained an ABI suffer PM impairment, measured both in a complex ecological real life EF task, the Children's Cooking task and using the two PM subtests of the RBMT ; (2) to explore if the CCT PM score is sensitive to developmental changes in PM in healthy controls and in patients with ABI; (3) to replicate the findings that the CCT discriminates children with various types of ABI from typically developing controls^{121,120} on a larger number of patients.

METHODS

Children were recruited from 3 different regions of France. Inclusion criteria for children with ABI were: age 8-20, a brain injury sustained after birth and prior to the age of 16, at least one year prior to the study, necessitating on-going rehabilitation or follow-up. Exclusion criteria were: severe reading or vision impairment that would impeded the child to read/see the recipe, severe behavior problems, and homogenous intellectual disability (full scale IQ < 70). Exclusion criteria for typically developing controls were diagnosed neurological or psychiatric condition or history, intellectual disability, inability to attend a mainstream school. All children were divided in 3 age groups (8-10; 11-13 and 14-20 groups).

Eighty-seven children participated in the study, separated in three age-groups: 54 children with ABI [8-10 years (n=10), 11-13 years (n=16) and 14-20 years (n=28)] and 33 typically developing matched controls (n=7, 17, 19 children in each group respectively). There was no significant difference between groups regarding age or gender (see table 1). Causes of ABI were TBI (N =43), brain tumours (N=7), ischemic or haemorrhagic stroke (N=3), drug intoxication (N=1). Mean age at injury was 8.98 years \pm 3.35 (range 7 months – 14.4 years). For TBI, mean Glasgow Coma Scale Score was 7.48 \pm 3.14 (range 3-15), length of coma was 7.13 days \pm 8.5 (range 0-45).

All children with ABI had been recently (< 6 months) assessed for general intellectual ability, using the age-appropriate Wechsler Scales (WISC III, WISC IV or WAIS III) prior to inclusion in the study, and the assessment was recorded for each patient. WISC III was used for children who were part of a cohort study that used WISC III as a follow-up measure. When the neuropsychological assessment had included the 2 PM sub-tests of the Rivermead Battery Memory Test (RBMT) ^{198 199}, results were also recorded. In the “remembering a hidden belonging” sub-test, an object is hidden, and the subject is required to ask for it at the end of the test session and to remember where it was hidden. In the “remembering an appointment” sub-test, an alarm is set to ring after 20 minutes, and the child is required to ask a specific question when the alarm goes off. The total of both PM tasks raw scores was used to screen for a possible correlation with the CCT PM score.

Prospective memory (PM) was assessed in all children using the Children’s Cooking Task (CCT)^{120,121}. In the CCT, children have to prepare a real chocolate cake and a fruit cocktail following a structured, photo-cued, child-friendly recipe contained in a cookbook including distracters. Instructions for all the key steps are clearly stated, with a picture for each step, although some actions embedded in a step are not necessarily explicitly detailed, in order to assess children’s adaptability to the context (e.g.: using straw to open and empty an apple juice pack into a glass). The CCT can be performed from the age of eight. Scoring is based on the number of errors and qualitative data (goal achievement, occurrence of dangerous behaviours, spontaneous initiation of both recipes and necessity of an adult intervention to prevent task failure or consequences of a dangerous behaviour). Details of CCT scoring can be found in the CCT scoring manual (unpublished, available from the last author) and previous publications about the test^{120,121}. Normative data is not available yet. The CCT has

good inter-rater and test–retest reliability, high internal consistency, as well as good discriminant and concurrent validity¹²⁰.

Four PM tasks are embedded in the CCT instructions, although the original CCT scoring had not used these tasks for a specific PM subscore: (1) making a fruit cocktail after finishing the chocolate cake; (2) putting the dirty dishes in the sink; (3) putting the rubbish in the bin; (4) telling the examiner that the task is finished. These 4 PM tasks are explicitly mentioned to the child before the start of the task, and they are written in the instruction sheet that is given to the child and placed in front of him/her during the whole duration of the test (“Today, you will prepare a chocolate cake and a fruit cocktail please place everything that needs to be washed in the sink and the rubbish in the bin... Tell me when you have finished”- refer to Chevignard et al.¹²¹ and CCT manual²⁰¹ for complete detailed instructions). Furthermore, before beginning the task the child is asked to summarise the instructions; thus ensuring the child has understood and remembered them.

Scoring for PM tasks was developed as follows: 0: failure to achieve: 1: achieved after a **specific** cue is given (“look at the instructions, you have forgotten something”); 2: achieved after a **non-specific** cue (“are you sure you have finished?”); 3: achieved spontaneously (without any cue). The CCT manual provides examiners with a progressive range of cues to be given to the child if s/he forgets one of the PM tasks after finishing the chocolate cake. The final score for all four PM tasks developed for this study ranged from 0 to 12. With this scoring system, a PM score < 8 showed inability to react to non-specific cues.

Observation and scoring were performed by three different examiners (AKP, VS, CA), who received a specific training on the use of the CCT and regularly consulted the author of CCT (MC). Ambiguous behaviour scoring was discussed with the author of CCT (MC).

Data were analyzed by IBM SPSS statistic 19, using non parametric statistics, as the number of errors in the CCT was not normally distributed. In order to compare children with ABI to controls, a Mann-Whitney U test for independent samples was used for quantitative data (number of errors on CCT, task duration, PM score) with reported standardized test statistic (Z). For CCT qualitative data, a Fisher exact test was used with reported Cramer’s V as measure of effect size (range 0 and 1). Differences between age groups were explored by Kruskal-Wallis test and Jonckheere test to screen for trend. A Bonferroni adjustment was used

for multiple comparison of PM scores between controls and children with ABI for 3 age groups with a significance level of $0.05/3 = 0.017$. Correlations between CCT PM score, number of errors in the CCT, RMBT PM subtests scores and age were analyzed by Spearman correlation coefficient.

RESULTS AND DISCUSSION

Total number of errors – replication of earlier findings: ABI and controls significantly differed on the total number of errors in the CCT (see table 2.1) ($p < 0.001$), replicating earlier findings that children with ABI are severely impaired on ecological EF assessment using real cooking^{121,120}. The number of errors in the CCT showed a developmental trend in typically developing children: as children grew older, they made less errors (Jonckheere test $J = -3.16$; $p = 0.002$), with a significant difference in the number of errors across age groups (Kruskal Wallis test, $H(2) = 11.6$; $p = 0.003$). Children with ABI also showed a developmental effect on number of errors in the CCT (Jonckheere test $J = -3.39$; $p = 0.001$), with a significant difference in the number of errors across age groups (Kruskal Wallis test, $H(2) = 11.4$, $p = 0.003$). Further there was a significant medium size negative correlation between the total number of errors and age, both in controls ($r = -0.561$; $p = 0.001$) and children with ABI ($r = -0.502$; $p < 0.001$), indicating that as children grow older, they make less errors in the CCT. This is consistent with literature on progressive maturation of EF^{20,122}.

Table 2.1: Comparison of demographic characteristics and of test scores in children with acquired brain injury and typically developing controls.

	ABI [mean (SD)]	Controls [mean (SD)]	Effect size and significance †
N	54	33	
Age (years)	13.3 (3.05)	12.5 (2.40)	p=0.13
Sex (% boys)	64	61	p=0.5
CCT Prospective Memory Score (/12)	8.6 (2.6)	11.1 (1.7)	z=-4.7; p=0.000*
Cocktail	2.4 (0.9)	2.9 (0.4)	z=-2.8; p=0.006*
Rubbish in the bin	2.2 (1.1)	2.7 (0.6)	z=-2.0; p=0.039*
Dirty dishes in the sink	2.2 (1.1)	2.77 (0.6)	z=-2.4; p=0.016*
Say “I finished”	1.8 (1.5)	2.8 (0.75)	z=-3.4; p=0.001*
Total number of errors in the CCT	75.4 (52)	28 (36.6)	z=5.6; p=0.000*
Qualitative analysis of CCT			
Task duration [min]	47 (17.3)	31.9 (9.5)	z= 4.5; p= 0.000*
Goal achievement [no]	55%	9 %	V = 0.46†; p=0.000*
Dangerous behaviours [yes]	63%	9 %	V = 0.3† ; p=0.003*
Spontaneous initiation of both recipes [no]	33 %	8%	V= 0.28†; p=0.013*
Intervention of an adult necessary [yes]	60%	11%	V = 0.45†; p=0.000*
RMBT PM subtest Standard score (0-2)			
Appointment	1.22 (0.6)		
Hidden belonging	1.41 (0.6)		
Wechsler Standard scores# (WISC III n= 18, WISC IV n= 27, WAIS IV n=8)			
Processing Speed Index	80.7 (18.3)	–	
Working Memory Index	82.8 (19.9)	–	
Perceptual Organization/Reasoning Index#	84.0 (13.4)	-	
Verbal Comprehension Index	89.2 (18.3)	-	

Note: apart from **qualitative** data denoted by † which is reported by Cramer’s V from Fisher’s exact test, the statistics reported are standardized Z statistic obtained by Mann-Whitney U test for independent samples;

* denotes significant results (p<0.05);

Perceptual Organization index from WISC III and WAIS III and Perceptual Reasoning Index from WISC IV were pooled to obtain a unique mean score of perceptual abilities;

ABI: Acquired brain injury; **CCT:** Children’s Cooking Task; **RMBT:** Rivermead Behavioural Memory Test; **PM:** Prospective Memory.

As expected, the total number of errors in the CCT, which is a measure of ecological global executive functioning, was correlated with the CCT PM score ($r=-.336$, $p=0.039$) in the control group; i.e. as children made more errors, they had a lower PM score. This is consistent with literature showing that in typically-developing children aged 6-12, performance on EF tasks such as planning and switching⁴⁷, working memory⁴⁸, and inhibition⁴⁹ is correlated with PM. In children with ABI however, the CCT PM score and the total number of errors were not correlated, similar to Ward et al. study, who found that most EF tests were not predictive of PM⁸⁶.

Prospective Memory: PM scores for typically developing controls were significantly higher (Mean rank=59.77) than those of children with ABI (Mean Rank= 34.36; $p<0.001$; see table 1). The performance of older children (14-20) with ABI did not differ significantly from the performance of young (8-10) controls, while 89% of their typically developing peers aged 14-18 obtained the maximum possible PM score (see figure 1). The difference was still significant ($p<0.001$) when subjects with impaired working memory index (<65) were excluded from analysis, suggesting that poor PM was not due to poor short-term or working memory. Examples of behaviour of children with ABI in relation to PM tasks and before the task ended included: leaving the kitchen after having prepared the cake to go play in the next room, licking various utensils and objects on the table instead of picking up the utensils and rubbish on the table, drinking the fruit cocktail, beginning to play with a smartphone halfway through tidying and leaving the cake burn in the oven. Children with ABI were not helped by non-specific cues: 44% of them did not manage the PM tasks even after non-specific cues were given (PM score <8), versus only 6% of the controls, all of whom were in the youngest age group (8-10). In the two PM sub-tests of the Rivermead Behavioural Memory Test (RBMT), only 11% of children with ABI obtained the maximum score on the two sub-tests used, whereas according to the normative data, 80% of children aged 8 to 14 are expected to do so.

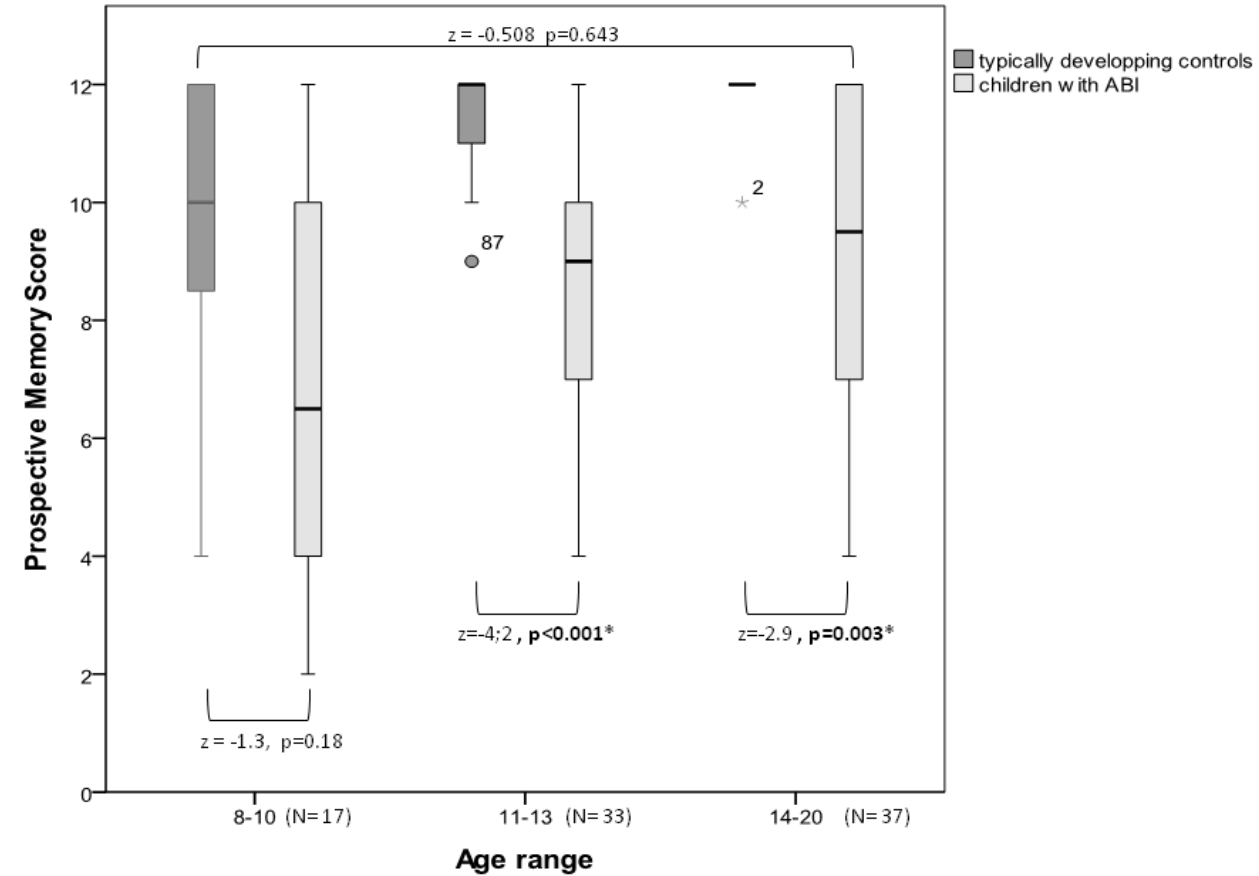
In the youngest age group only, we found a significant correlation between age at injury and PM score ($r= 0.7$; $p=0.025$): the earlier the injury, the lower the PM score was. There was no overall effect of age at injury in the whole ABI group.

No significant correlation was found between the PM score on CCT and sum of PM raw scores of RMBT in the ABI group.

Developmental changes in PM scores: PM scores of the youngest typically developing controls (8-10) and of children with ABI of all ages showed important variability (see figure 1), whereas older controls (11-13 and especially 14-20) showed very homogeneous high scores, suggesting important PM development from age 8 to 11 in typically developing children, with mature PM by the age of 14 on the CCT PM tasks. In typically developing controls only, PM showed a developmental trend: as children grew older, they had higher (better) PM scores (Jonckheere test $J=2.701$; $p = 0.07$), with differences between age groups that were statistically significant (Kruskal Wallis test, $H(2)= 6.99$; $p=0.03$) and a significant correlation ($r=0.373$, $p=0.032$) between age and the CCT PM scores. Difference between age groups and trend in children with ABI were not significant, but there was a correlation between PM score and age in this group ($r=0.275$, $p=0.044$), suggesting improvement of PM with age. Given the variability in patient's CCT PM scores in the 11-13 and to a lesser extent the 14-20 group, we expect that a larger sample size would have enabled us to find an age effect with the statistical tests used. An alternative explanation could be that children with ABI precisely do not improve their PM at the critical time of PM improvement found in typically developing controls of our sample, potentially due to lack of normal EF maturation found in typically developing children. However, ABI severity could not be considered in multivariate analyses along with age and age at injury given the heterogeneous sample. One cannot exclude that effects of age at injury or severity also contributed to this finding.

Our study adds objective data to the parental reports⁸⁷ and to the previous studies⁸⁵⁻⁸⁶⁻¹⁸⁸⁻¹⁸⁹ reporting that PM is impaired after brain injury in children. In this study, however we used an ecological assessment consisting of simple and natural PM tasks, very close to what is expected of a child (e.g.: put the rubbish in the bin after finishing an activity, follow instructions at school, similarly to the CCT where two recipes need to be prepared). Results showed severe impairment on these simple PM tasks after childhood ABI. Similarly to McCauley et al. study¹⁸⁸, children with ABI were not helped by non-specific cues.

Figure 2.1: Boxplot of Prospective Memory Scores according to age group and injury status



Note: The tinted box represents the interquartile range (i.e. the middle 50% of scores), the thick horizontal line represents the median, the whiskers represent the range of the top 25% and 25% bottom, excluding outliers (represented by a circle) and extreme cases (represented by a star).

It might be argued that the success or failure on these tasks was not related to PM ability only but that it may have been influenced by other factors and especially, social cognition (children are expected to clean after finishing an activity) and theory of mind (deducing that “I have finished” is to be said if examiner is not reacting at the end of the test). If, however, implicit,

socially-driven cues were the reason children with ABI performed poorer on PM tasks, performance should have been corrected after being given non-specific cues explicitly, which was not the case. The lack of correlation between the two assessments of PM (RBMT subscores and CCT MP score) speaks to the problem we face in studying PM in non-natural settings: do the ecologically valid tests really measure the same thing as the laboratory tests in children? Although meant to be “ecological”, the RBMT is still a paper-and-pencil task that comprises only two PM items and is therefore probably insufficient to adequately screen for PM difficulties. The same discrepancies have been found in other ecological assessments of executive functioning and remain an important issue that needs further research^{170,180}. The main limitation of this study is the absence of retrospective memory data that could account for PM failure. Also, mechanisms of injury may have influenced PM but the small numbers of children in each condition did not allow a differential analysis in each diagnostic group. Another drawback is that PM sub tests from the RBMT were administered only to children with ABI, impeding a direct comparison of performance with typically developing peers on these subtests. As suggested previously^{189,86}, future research should include an evaluation of both retrospective and prospective memory, as well as an assessment of EF, in order to better understand memory functional impairment in children with ABI. Another limitation of the study is the small sample size in the typically developing healthy controls. To examine age effects and typical development of PM skills, a much larger sample size would be needed. This study does, however, support previous literature on typical PM development but future research should examine the age-effects in a much larger sample.

Our study shows that PM is significantly impaired in children with ABI performing an ecological, close to real life cooking task including four simple PM tasks. Unfortunately, usual neuropsychological assessments of memory often do not explore PM, and focus mostly on episodic memory. Ecologically valid and sensitive tests of PM should be included in assessments of children with ABI, in order to better reflect actual impairment in everyday life and at school.

CHAPTER 3: PROSPECTIVE MEMORY SEVEN YEARS AFTER SEVERE CHILDHOOD TRAUMATIC BRAIN INJURY - THE TGE 2 PROSPECTIVE LONGITUDINAL STUDY

To be submitted

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INTRODUCTION

Prospective memory (PM) is the ability to remember to perform intended activities⁶⁴. The majority of memory failures reported by adults are prospective in nature¹⁹². Children with PM impairment can fail to deliver important messages to parents, forget appointments, or fail to bring necessary items for planned activities. PM tasks require retrospective memory to remember the task, but depend on executive functions (EF)⁶⁵ for successful goal maintenance, retrieval and implementation at the right moment. At least three attributes are accepted as characterizing a PM task²⁰²: (1) a delay between formation of the intention and the opportunity to carry it out; (2) absence of an explicit reminder to carry out the task at an appropriate moment; (3) the need to interrupt one's ongoing activity in order to carry out the intention.

Although PM problems are reported as a major concern by the parents of children with traumatic brain injury (TBI)⁸⁷, there are surprisingly few studies that have evaluated PM in children with TBI (as opposed to adults with TBI, on which there is an extensive body of existing research^{65,203,204}). Ward et al.²⁰⁵ found that children with TBI had poorer PM than their uninjured peers, and that this may be attributable to worse executive functioning, especially if the PM task is cognitively demanding. Recently, a real cooking task (Children's Cooking Task^{120,121}) has been used to explore PM in children with acquired brain injury¹⁸¹: it showed a striking impairment in PM, with older (14-20) children with brain injuries performing close to young (8-10) healthy controls. Further, children with acquired brain injury were not helped by non specific cues. McCauley et al.^{85,189,194} used a monetary incentive to increase children's performance on an event-based PM task consisting of asking the examiner for points (exchangeable for dollars or for pennies) before each new neuropsychological test. Children with severe TBI were impaired on PM. Motivation (dollars versus pennies) influenced PM performance of controls and of children with chronic severe TBI⁸⁵, while it had no effect on children with subacute severe TBI^{189,194}. However even the chronic severe TBI group performance remained significantly below the low-motivation condition performance of controls.

Limitations of these studies were that they used either a biased sample of children with TBI (recruited from patients needing rehabilitation) or, for the one prospective study¹⁹⁴, children early after TBI. Therefore the frequency and degree of PM impairment after severe TBI in the long term is unknown. This is particularly needed information because PM can impair functional memory and daily life even more than retrospective memory⁸⁷, but most patients in clinical practice (and even in research) are assessed only on the latter while PM is not included in expertise or rehabilitation assessments.

The aim of this study was to investigate long term PM outcome, after childhood severe TBI, using short novel ecological tasks of PM in a prospective longitudinal cohort.

METHODS

PARTICIPANTS

Participants were a cohort of children with severe accidental TBI (defined as Glasgow Coma Scale (GCS) score of 8 or lower and/or an Injury severity score¹²⁶ > 16), aged 0-15 years at the time of inclusion and recruited between January 2005 and December 2008 at the pediatric neurosurgical intensive care unit of Paris 5 University “Necker Enfants Malades” Hospital, and assessed at seven years post inclusion for PM prospective memory (PM). By the seven year testing the cohort contained therefore both children and young adults. Exclusion criteria at the time of cohort recruitment were: children with no vital signs upon admission, children with non-accidental head injury, previous history of diagnosed neurological, psychiatric or learning disorders. A group of controls was recruited at the seven year follow up point, matched individually (in terms of age, gender and parental education) with the TBI group. This study is part of a larger study, which was approved by the Paris ethic committee CPP VI.

PM TASKS

Three ecological tasks of PM were developed for the study. Because PM performance is influenced by motivation⁸⁵, by the ecological features of the task (meaningful task versus exercise type task, home versus laboratory context)²⁰⁶ and by delay⁶⁶ between formation of an intention (PM instructions in this case) and the target situation, the tasks were designed to

vary on these three factors. Further, because standard tests administered to the cohort were numerous and time-consuming, the PM tasks had to be short and not disturb the other tests. For these reasons, time-based PM tasks were not used because children would have invested attentional resources into time monitoring rather than the on-going testing.

Low-motivation, ecological context, ecological task, long delay: the letter TASK

Individuals received an envelope with the address of the hospital and were asked to send to the hospital a short note on the next day it rained in the place they live. The letter had to contain four pieces of information: name of the participant, date, place and a mention of rain. The participant was also told that even if s/he forgot to send the letter the first time it rained, s/he could still do it later on, the important thing being to send the letter at some point. Instructions were given twice to the child during the testing and the examiner checked if the individual had understood and encoded the instructions. The task was not explained to parents, but parents were warned the child had a task to do when s/he was at home and that is why s/he had an envelope for the hospital and that they were not to help him/her. Data from the letter task was treated as categorical (success or failure) in relation to two components: (1) sending the letter with all adequate information (which comprised a retrospective memory component - the letter content - as with most PM tasks of daily life) and (2) sending the letter irrespective of content (which assessed the prospective component of PM, as the child had only to remember the intention to send a letter).

High-motivation, ecological (meaningful) task, non ecological context, medium delay task: the amusement park prize-draw competition.

At the beginning of the testing, individuals were told they could enter a prize-draw competition to thank them for participating in the study. The examiner showed them a colored entry sheet that mentioned the draw. They were told that they could enter their name for the draw only at the end of all tests. The prize was two entries for an amusement park of their choice. At the end of the testing (6-7 hours after the instructions for the children with TBI and 2.5-3 hours after the instructions for the controls who had less tests to complete), the examiner said “ok, we have finished all the tests, well done”. If the participant did not ask spontaneously to enter the draw, the examiner made discreetly visible the colored competition entry-sheet so that it came into the participant visual space but without giving it to him/her. If

the participant did not react to this visual cue, the examiner used a verbal nonspecific cue: “did you want do to something else before leaving?”. If this was not enough, the participant was reminded explicitly s/he could enter his/her name for the draw competition. Data from the draw competition PM task was treated as ordinal: 3 points were awarded for individuals succeeding without cues (individuals asking spontaneously to enter the competition at the end of testing); 2 points for individuals needing the visual cue only; 1 point for individuals needing the verbal cue and zero points for individuals needing an explicit reminder to enter their name for the draw.

On the competition entry-sheet the participant was instructed to add his/her name and telephone number. In addition, there was an instruction to read and to tell the examiner which amusement park s/he would like to go to if s/he won, and to fold the completed sheet in two for the draw box. These tasks did not constitute PM tasks as they could be carried out immediately, but tested the child’s ability to follow through a series of task instructions.

Low-motivation, exercise-type, non-ecological, short delay (retrieve-execute): post-it/faces task

In the last task, the delay was short. For this task, we used two existing tests as the on-going task: the NEPSY-2 affect recognition subtest and the Bordeaux Faces Test, which were included in the social cognition evaluation protocol (PECS-B)²⁰⁷. In the adult version and most items of the child version, the individual had to name an emotion by looking at a face. Colored post-its were placed on some pages of these tests. The individuals were told to remove all the post-its apart from the pink ones throughout the task, but only after they had stated the emotion, not before. There were 9 post-its to remove (and 4 pink post-its to leave), placed in pre-determined position on nine of the 39 (children version) or 40 (adult version) pages of the test. The instruction to take off only post-its of certain colors was meant to increase task difficulty and prevent the individuals from automatizing post-it removal without effortful processing.

STATISTICAL ANALYSIS

Individuals with severe TBI participating in the study were compared to individuals lost to follow-up, by Mann-Whitney tests on GCS Score, coma length, age at injury, and one-year post injury intellectual quotients and executive functioning. Further, Chi squared was used to

compare participants for language spoken at home, gender and parental education (defined as high if above high school or low if high school and below). The same tests were used to compare the participating severe TBI group to the controls on age at testing, parental education, gender and language spoken at home. Controls and individuals with TBI were compared for categorical data (fail/success in sending the letter with all adequate information, fail/success in sending the letter irrespective of content) using chi squared analysis. Effects size was calculated by odds ratios. The draw competition PM ordinal data was analyzed using Mann-Whitney test. The Post-it task yielded a score of number of post-its taken out which was analyzed between groups using Mann-Whitney tests because score distribution was not normal, and effect size was calculated using $r = \frac{z}{\sqrt{N}}$ and interpreted according to Cohen's guidelines²⁰⁸. Additionally, among each group (TBI and controls), adults' performance was compared to children's' performance using Mann-Whitney test or Fisher's exact test.

RESULTS

PARTICIPANTS

Eighty-one children were included at the acute stage of TBI between January 2005 and December 2008. Sixteen children died during acute care, leaving 65 children entering the follow-up. By seven years post-injury, 26 were lost to follow-up, leaving 39 patients (aged 7-22, mean: 15 years; 64% boys) tested for PM.

Individuals remaining at 7 years follow-up had an initial coma length of 6.4(SD: 5) days; initial GCS of 6.5 (SD=1.4, range 3-8) and were aged 8.9 years (SD=4.5, range 0-15) at injury. 56% of them used French exclusively at home, and 54% were from low parental education background. Individuals remaining at 7 years follow-up did not differ significantly from those lost to follow-up on these parameters, nor on their one year post-injury intellectual quotients and executive functioning (see chapter 1 for details on cognitive abilities at one and two years post injury).

Most individuals (26) were still children at the time of the study but 13 had become young adults. 37 controls were recruited (two controls could not be recruited within the time frame

of the study). There was no significant difference between the TBI and the control groups on gender, age at testing, parental education, and language spoken at home.

PM TASKS

Letter task – Overall, 56 % of participants sent a letter. Individuals with TBI failed significantly more on both the prospective and the retrospective component of the task. One letter contained weather and date information but no name and therefore it could not be identified to which individual it belonged. If an individual had sustained a TBI he was 2.6 times more likely to forget to send the letter and 3.7 times less likely to send a letter with all required information. Results are presented in Table 1.

Table 3.1: Results of PM tasks			
	Individuals with severe TBI	Controls	
Letter Task			
% individuals who succeeded to send the letter with all information required	20%	49%	$\chi^2(1) = 6.68$, p=0.01*
% individuals who succeeded to send a letter, irrespective of adequate content	44%	68%	$\chi^2(1) = 3.96$, p=0.047*
Prize Draw Competition			
% individuals who spontaneously entered their name for the draw	28%	27%	U=660, z=-0.07, p= 0.94
% individuals who needed <i>visual</i> cues to enter their name for the draw	19%	14%	
% individuals who needed <i>verbal</i> cues to enter their name for the draw	31%	46%	
% individuals who needed an explicit reminder to enter their name for the draw	22%	13%	
Post-it Task			
% individuals who totally forgot to take out the post-its	36%	13%	$\chi^2(2) = 8.43$, p=0.015*
% individuals who remembered to take off all required post-its	28%	60%	
% individuals who remembered partially to take off post-its	36%	27%	

Amusement park prize-draw task - There was no significant difference between controls (Mean Rank =37.16) and individuals with TBI (Mean Rank = 36.8) in the prize-draw competition ($U=660$, $z=-0.07$, $p= 0.94$).

Post-it Task- Individuals with TBI remembered to take out significantly less post-its (Mean = 5.06, SD = 4.07) than their matched controls [Mean = 7.35, SD = 3.07, ($U=420.5$, $z=-2.87$, $p= 0.004$, $r=0.34$)]. There was no difference between groups in taking post its too early. Apart from one control who took out all four pink post its, individuals respected the rule of leaving the pink post its equally well in the TBI and the control group ($p = 0.16$).

CAPACITY TO FOLLOW THE 5 INSTRUCTIONS TO ENTER THE DRAW

When entering the amusement park draw competition, individuals with TBI managed to follow less instructions (fill in name, read instructions, fill in table with phone number, state the amusement park they would like to go to, fold the participation sheet for the draw) than controls (mean in controls = 4.2, mean in TBI group = 3.7; Mann-Whitney $U=452.5$, $z= -2.20$, $p= 0.028$, $r=0.26$).

DEMOGRAPHIC AND INJURY EFFECTS

None of the PM tasks was influenced by parental education, initial GCS or coma length. In the whole sample, individuals whose parents had higher education (Mann-Whitney $U=789$, $z=2.28$, $p=0.023$, $r=0.27$) and who spoke exclusively French at home (Mann-Whitney $U=377$, $z=-2.34$, $p= 0.019$, $r=0.28$) followed more instructions.

AGE AT TESTING EFFECTS

Children performed poorer than young adults on the Post-it Task, irrespective of injury status (in the TBI group: Mann-Whitney $U=210$, $z=2.61$, $p=0.009$, $r=0.38$; in the typically developing controls: Mann-Whitney $U=237$, $z=2.69$, $p=0.007$, $r=0.44$).

On the prospective component of the letter task (sending a letter irrespective of content), there was a trend for adults to perform better than children in both groups (Fisher's exact test = 0.068 in the TBI group and 0.084 in the typically developing controls group).

There was no difference between younger and older participants for the prize-draw competition and the overall letter task.

DISCUSSION

Across the large age span of 7-22, individuals with severe TBI systematically recruited for a longitudinal prospective follow-up showed significantly poorer PM seven years' post-injury than matched controls in two PM tasks. Their performance on a high motivation PM task did not differ from controls. Younger children performed generally poorer on PM tasks, except for the high motivation task (amusement park draw). However, although individuals with TBI managed to retrieve and implement their intention of entering the draw (PM success), they were significantly poorer than controls in following detailed instructions. The ecological task consisting of sending a letter on a rainy day, showed significant differences both in its PM component (sending the letter irrespective of adequate content), and the overall task (sending the letter containing all adequate information).

This is to our knowledge the largest cohort of severe childhood TBI. Most cohorts usually include heterogeneous samples of children with a range of TBI severity, where severe TBI are usually relatively few^{19,71,162}. It is also to our knowledge the first cohort study that assessed PM. The prospective longitudinal nature of this study was expected to capture more positive outcomes compared to retrospective studies based on the inclusion of patients in rehabilitation^{73,163}; nonetheless PM impairment appeared significant and should raise awareness about frequent PM deficits, that are not explored by usual memory tests used in children¹³⁶ and that should be given more attention.

Our results are consistent with previous publications on both developmental⁶⁷ and clinical samples⁸⁵ that showed that deficits in PM performance may be reduced under high motivation conditions. The lack of age effects in the high motivation condition may have been due to an unequal degree of motivation, as the experimenter noted that younger children seemed more enthusiastic about the prize-draw, while some young adults and adolescents appeared less interested (one did not even want to enter the draw). An unequal motivation effect was probably also present for the letter task, which was designed to be a low motivation condition: children aged 7-10 seemed very proud to have a letter to write and post and many parents

reported the enthusiasm of their child for the task. Also, we cannot exclude that parents helped their children with the letter task, because young children are not expected to send a letter of their own and it may have seemed natural for their parents to help them despite the examiner's explanations.

Whether PM improves with age is still a matter of debate. Small children as young as two can succeed in PM if motivation is high (remind their Mum to buy them sweets)⁶⁷. Age effects that have been documented in the literature may be entirely attributable to factors such as: (1) unequal difficulty of the on-going task, allowing less attentional resources to PM tasks in younger children; (2) retrospective memory component; (3) motivation. When taking these into account, age effects of PM are typically small⁶⁸. This is in line with our findings, as the prize-draw competition showed no difference between age groups.

Implications: There is now more evidence that PM is a common sequelae of childhood severe TBI^{85,87,181}, also at long-term. When assessing sequelae post TBI, the evaluation should include an assessment of PM in addition to classical episodic memory assessment. When PM is found impaired, interventions should aim at using high motivation tasks/incentives for most essential PM tasks of daily life, in order to help individuals with TBI with most important goals. However other methods should also be considered (e.g. pager, alarms), given the frequent overall PM impairment.

Limitations: The tasks used are not validated, their relative difficulty was not explored and therefore the differences observed between the high and low motivation tasks may be due to the task difficulty and not the motivation factor per se. Most tasks generated categorical or ordinal data that did not allow assessment of PM impairment severity. In the prize-draw competition, delay between instructions and opportunity to carry out the intention (entering the draw) was unequal between control and TBI groups, which may have disadvantaged the individuals with TBI who, further, were more tired by a longer assessment than controls. However, on this task there was no difference between groups. For the other tasks the delay was the same.

The post-it task could be argued to reflect rather a dual task (dependent more on working memory ability than prospective memory), as the target (non-pink post-its) were relatively frequent (9 targets out of 40 pages), while other experimental laboratory PM tasks tend to use

less frequent targets (e.g.: 2 out of 13⁴⁸, 1 out of 20¹⁸⁷) but not always (e.g.: 3 out of 10⁶⁴). Probably, the higher the number of target stimuli, the more the task relies on working memory, because the frequent target acts as a constant reminder of the intention and is therefore kept in the attentional focus of working memory⁹². On the other hand, if the target is infrequent, as the on-going task proceeds, the intention drops progressively to lower attentional levels. Further, dual task performance is more about switching between two activities while PM more about delaying an intention until a favorable moment to execute it is encountered. Our post-it task may have better assessed PM if the target stimuli were less frequent. Further, because children had to switch between facial recognition, post-it removal and pink post-it inhibition of removal, the task evaluated probably both working memory and PM.

Conclusions: PM is impaired in individuals at long-term after severe childhood TBI. High motivation conditions may enhance PM.

CHAPTER 4: SELF-AWARENESS ASSESSMENT DURING COGNITIVE REHABILITATION IN CHILDREN WITH ACQUIRED BRAIN INJURY: A FEASIBILITY STUDY AND PROPOSED MODEL OF CHILD ANOSOGNOSIA

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RESEARCH PAPER

Self-awareness assessment during cognitive rehabilitation in children with acquired brain injury: a feasibility study and proposed model of child anosognosia

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Abstract

Purpose: To compare three ways of assessing self-awareness in children with traumatic brain injury (TBI) and to propose a model of child anosognosia. **Method:** Five single cases of children with severe TBI, aged 8–14, undergoing metacognitive training. Awareness was assessed using three different measures: two measures of metacognitive knowledge/intellectual awareness (a questionnaire and illustrated stories where child characters have everyday problems related to their executive dysfunction) and one measure of on-line/emergent awareness (post-task appraisal of task difficulty). **Results:** All three measures showed good feasibility. Analysis of awareness deficit scores indicated large variability (1–100%). Three children showed dissociated scores. **Conclusions:** Based on these results, we propose a model of child self-awareness and anosognosia and a framework for awareness assessment for rehabilitation purposes. The model emphasizes (1) the role of on-line error detection in the construction of autobiographical memories that allow a child to build a self-knowledge of his/her strengths and difficulties; (2) the multiple components of awareness that need to be assessed separately; (3) the implications for rehabilitation: errorless versus error-based learning, rehabilitation approaches based on metacognition, rationale for rehabilitation intervention based on child's age and impaired awareness component, ethical and developmental consideration of confrontational methods.

Keywords

Brain injury, children, cognitive rehabilitation, executive functions, model, self-awareness

History

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INTRODUCTION

Metacognition is the conscious knowledge of one's own cognitive processes as well as the processes involved in consciously monitoring and regulating one's ongoing actions⁵¹. Thus metacognition refers to the awareness of one's own cognition and is used in the context of normal functioning. The concept of awareness/self-awareness (or more usually lack of awareness), on the other hand, is usually used in the context of pathology and refers to the awareness of one's deficits, including cognitive deficits. As such, awareness can be viewed as metacognition applied to difficulties in cognitive functioning.

Awareness and metacognition have been defined in many ways and there is little agreement among neuropsychologists, psychologists, developmentalists and education researchers as to the exact nature and limit of the two concepts. In developmental psychology, metacognition is thought to comprise metacognitive knowledge and metacognitive skills. Metacognitive knowledge is defined by Flavell as knowledge about one's own cognitive strengths and limitations, including factors that may interact to affect cognition⁵². The concept of metacognitive skills, refers to the voluntary control of cognitive processes including prediction, planning, monitoring and evaluation of behaviors.

Many models of awareness and metacognition have been proposed for adults²⁰⁹. Two models of awareness/metacognition are particularly useful: (1) Crosson's model²¹⁰ that offers a pragmatic classification of awareness levels linked with efficient compensation which can be proposed in each level and (2) Toglia and Kirk's comprehensive model of awareness after brain injury²¹¹.

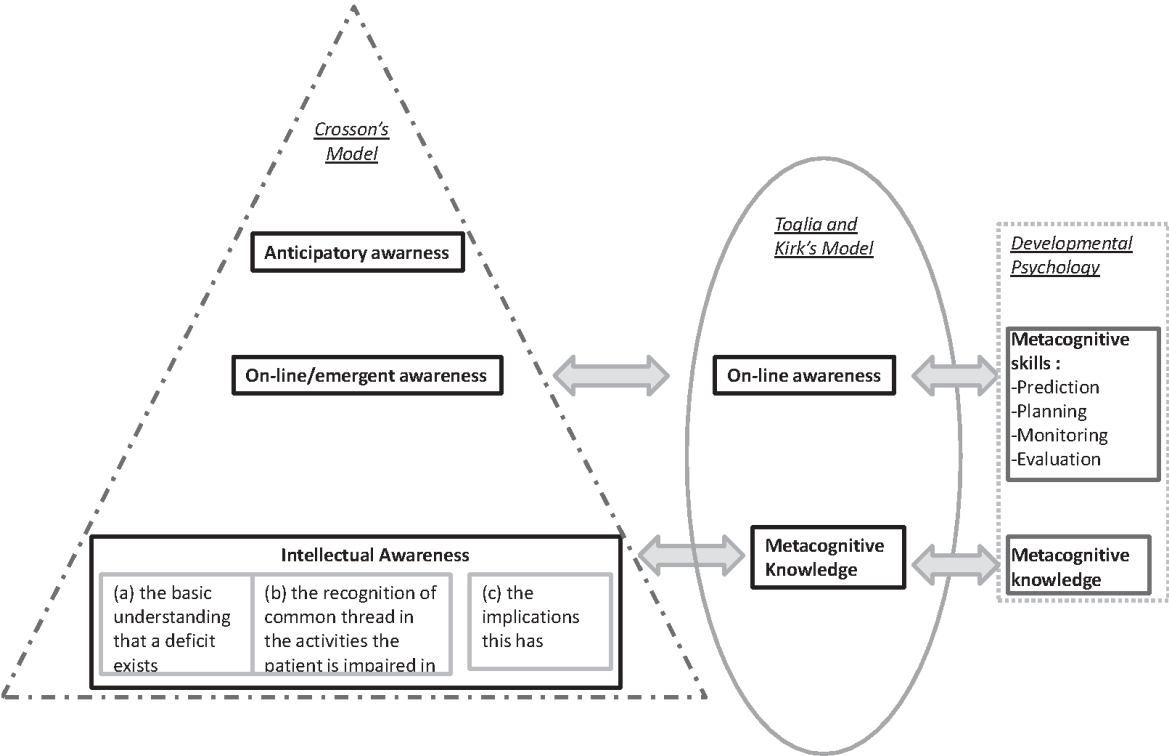
Crosson's model has an hierarchical structure, although this hierarchy has never been confirmed empirically²¹². At the base of awareness is intellectual awareness. Intellectual awareness comprises three sublevels: (a) the basic understanding that a deficit exists, (b) the recognition of a common thread in the activities the patient is impaired in, (c) the implications this has. On-line Awareness (also called emergent awareness) refers to the ability to recognize a problem while performing an activity. Emergent awareness is crucial to rehabilitation as patients who do not realize that a problem is occurring will not recognize the need to correct it and /or to initiate compensation. Anticipatory awareness is the highest level

of awareness in Crosson's model, and is defined as the ability to anticipate that a problem will occur as a result of some deficit, and take some action to prevent that problem occurring.

Toglia and Kirk's comprehensive model of awareness after brain injury views the relationship between different aspects of metacognition and awareness as a dynamic process rather than as a series of hierarchical levels. It clearly differentiates between knowledge and beliefs related to one's self (i.e. *metacognitive knowledge* that pre-exists and is stored within long term memory) and knowledge and awareness that is activated during a task (i.e. *on-line awareness* which Toglia and Kirk define as "the ability to monitor performance "on-line", within the stream of action")²¹¹. Metacognitive knowledge is what one brings to a task, whereas on-line awareness involves ongoing evaluation of performance within the context of a task²¹³²¹¹. Metacognitive knowledge and on-line awareness are distinct functions and have been found not to be correlated in adults with brain injury²¹⁴.

Although Crosson's and Toglia and Kirk's models offer two distinct frameworks to study awareness, we argue they are complementary and that their combined use allows a better understanding of patients' difficulties: (1) Crosson's Intellectual awareness corresponds to the metacognitive knowledge of Toglia and Kirk's model; (2) Crosson's emergent awareness corresponds to on-line awareness of Toglia and Kirk's model (and comprises metacognitive "skills" from the field of developmental psychology); (3) anticipatory awareness is the behavioral manifestation of good metacognitive knowledge and good on-line awareness. Hereafter, the terms of awareness and metacognition will be used interchangeably, postulating that they have the same underlying construct, irrespective of its application to pathology or to normal functioning. Correspondence between the models and vocabulary used in developmental psychology are summarized in figure 4.1.

Figure 4.1: Correspondence between Crosson’s and Tolia and Kirk’s model and Developmental psychology terminology



Impaired awareness/metacognition, sometimes also termed “anosognosia”, is a common phenomenon described in children who have sustained a traumatic brain injury (TBI) ^{81,82,83}. However it results from a combination of organically based unawareness (due to brain injury) and simple developmental immaturity⁸³ present in typically developing children as well. Metacognition is known to be poorer in younger children. Even typically developing children are not “fully aware”: they may have some basic intellectual awareness about things they cannot do that their parents can, however they are not able to fully understand the consequences of their cognitive limitations in recognizing a problem when it is actually happening, or predicting a problem will occur as a result of some developmental immaturity. Children only gradually come to gain awareness over the entire span of developmental

years⁸³. This has been mainly studied in relation to memory (termed meta-memory) in typically developing children who show gradual development of metamemory throughout childhood^{59,60,61,62,63} and in the field of learning disabilities^{54,215,55,216}. Nelson and Narens proposed a comprehensive model of metacognition in metamemory²¹⁷ where metacognition is described as the interplay between two levels of information processing (an object-level processing and a meta-level processing) that interact with feedforward and feedback control and monitoring loops. A neural description of the model has been proposed by Shimamura²¹⁸.

To the best of our knowledge metacognition in relation to other cognitive functions, and especially executive function (EF)/complex task management in daily life, has not yet been explored. Here we will focus on metacognition for executive functions and complex task management in children with TBI because EF deficits are a frequent and disabling consequence of TBI^{6,69} and because TBI outcome is strongly predicted by executive functioning level¹²².

Although metacognition is poor in young children, it has been shown to be even poorer in children who have sustained a brain injury^{81,82,83,219,220,221}. However, to date, objective measurement of awareness in children with TBI is scarce (see Wales et al.²²² for a review) and most studies evaluate single metacognitive skills such as prediction, evaluation and confidence of performance^{223,224,84,225,226}. Conversely, Beardmore et al. reported the use of the “Knowledge Interview for Children” (KIC)⁸¹, a semi-structured interview related to twelve areas of knowledge about TBI (coma, story of the accident, brain functioning...) and ten potential areas of difficulty (attention, fatigue, memory, behavior...). Interview of the child and the parents yields an Awareness Discrepancy Index, by summing the number of items endorsed by the child’s parents but rejected by the child. Children reported significantly less problems than their parents and demonstrated extremely limited knowledge about TBI. The SAND-C (Subjective Awareness of Neuropsychological Deficits Questionnaire for Children) is a self-report instrument in which children are required to estimate their neuropsychological functions²²⁷, however self-report is not compared to parental judgment and as such is not a measure of anosognosia if used alone.

Josman et al. evaluated children with TBI in relation to metamemory²²⁴ and categorizations skills²²⁸ with three types of self-awareness measures (1) intellectual awareness –termed general awareness by the authors (e.g.: “have you noticed any changes in memory?”), (2) self-

prediction (of task difficulty²²⁸; “how many pictures will you remember ?”²²⁴) and (3) self-estimation of performance after the task. General awareness questions were not fully understood by the children and therefore not recommended. Prediction was difficult in both healthy children and children with TBI, self-estimation was significantly less correlated with actual performance in brain-injured children than in typically developing ones²²⁸, and children with brain injury overestimated their memory performance²²⁴. Similarly, in Hanten et al.^{84,225} and Crowther et al.²²⁶ studies, children who had sustained a TBI had poor estimation of their memory span and overconfidence in performance when compared to healthy children and children with mild TBI⁸⁴, suggesting impaired metacognition. The scarce literature assessing awareness in children with brain injury explores metacognitive skills such as prediction and evaluation, using assessments performed in structured, unecological environments. Questionnaires of intellectual awareness (KIC, SAND-C) proposed to date are not domain specific and do not specifically explore awareness of executive functioning.

The use of metacognitive training is a practice standard in adults with deficits in executive functioning¹¹⁴. Adult metacognitive training programmes, such as Goal Management Training (GMT)²²⁹ have begun to be used in children²⁰⁰. GMT is mostly known for its algorithm “STOP! - Define the main task – List the steps –Learn the steps – Do it - Check” that can be used to train specific tasks²³⁰. However, the full GMT version⁹⁹ is a truly “metacognitive” training in that GMT encourages patients to think about their cognitive failures, to identify factors promoting or preventing these failures and to reflect and monitor how their thoughts may drift away from the main goal and switch to “automatic pilot”. When used alone, GMT group training^{231,99} does not offer a repetitive intensive practice of EF and monitoring skills, but rather teaches and prompts the patients to monitor their actions, to detect their cognitive slips and gain control over their actions, through a conscious and attention-demanding monitoring of their flow of thoughts and actions. The relatively short duration of the programme (7 x 2 hours of group training in adults, including GMT theory presentation and group discussions) does not allow automatization of monitoring and checking. Rather, it relies on patient’s awareness of difficulties and ability to actively implement the GMT algorithm in daily life, under conscious and ‘top-down’ control. Therefore, GMT in adults relies heavily on a patient’s awareness. A patient who considers that his/her cognition is efficient, will not easily engage in such an attention-demanding

programme aiming at improving cognition efficiency in an effortful way. Awareness (before treatment or acquired throughout the GMT programme) is a core factor for programme success. GMT programmes usually enroll patients with mild or moderate brain injuries with relatively preserved awareness or integrate an awareness intervention component before the GMT ^{118,119}.

As young children with brain injuries have impaired awareness because of developmental immaturity ⁸³ added to the organically based awareness deficits due to their injury, metacognitive training programmes such as GMT may not be effective because the core factor for programme success – awareness – is missing. It is therefore crucial to evaluate children's awareness when conducting a metacognitive training such as GMT. To our knowledge there is a lack of self-awareness measures for children. Tools used in metacognition developmental research ^{232,54,233,217} are not easily accessible or transferable to the context of rehabilitation (e.g.: use of event-related potentials)²³². Furthermore, as cognitive rehabilitation of EF aims at understanding and improving daily life executive functioning in the natural context of the child, classically used measures of metacognition (Judgments of Learning - JoL; Ease of Learning judgments – EoL; Feeling of Knowing – FoK; during a word list learning, in an office-based un-ecological environment ^{217,59,60,61,62,63}) are not clinically useful.

The primary aim of this pilot study was to examine the feasibility of three ways of assessing awareness of executive dysfunction in children with a TBI during a rehabilitation programme based on GMT.

METHODS

This awareness study was part of a pilot study that tested an intervention based on a context-sensitive pediatric Goal Management Training (GMT) combined with ecological activity practice. Details and effectiveness of the intervention are reported elsewhere ²⁰⁰. Children were taught metacognitive strategy use through discussion of stories in which story characters experience cognitive failures. Children were introduced to ideas of how those may be prevented, and were given practice at applying metacognitive strategies on paper-and-pencil exercises, then on ecological activities in the rehabilitation centre, and finally on real life

activities at home and school. The programme used a range of functional, meaningful activities including cooking. The training was administered weekly, for 15-20 hours over 4-6 months. Because of the availability and time required by the intervention for the children included in this pilot study, it was not judged ethically possible to include typically developing children. Children included in the study had sustained a severe TBI at least two years earlier, had a documented dysexecutive syndrome, including executive functioning difficulties in daily life as reported by parents and school staff.

Awareness was assessed using three different measures: two measures of metacognitive knowledge (intellectual awareness) and one measure of on-line/emergent awareness.

The two measures of metacognitive knowledge (MK) corresponded to two different levels of Crosson's intellectual awareness (see figure 4.1). The first measure assessed level (a) of Crosson's intellectual awareness i.e. the basic understanding that a deficit exists. The second measure assessed levels (b) and (c) of Crosson's intellectual awareness i.e. (b) the recognition of a common thread in the activities the patient is impaired in and (c) the implications this has.

The first measure of MK (the basic understanding that a deficit exists) consisted of a discrepancy score using the "goal management training questionnaire"⁹⁹ translated into French, simplified and adapted for children (see appendix 1). This questionnaire, based on the adult GMT questionnaire, is specific to goal management difficulties and 'goal neglect' (failure to take actions to achieve goals despite the intention to do so). It is however not validated and has no norms. Because poor reading skills and vocabulary might have influenced children's responses, the questions were read to the child who answered orally. The questionnaire presents common executive failures that can happen at home, at school or during leisure activities (e.g. "Forgetting something that needed to be done at a certain time, running out of time because you got too caught up in something that you were doing, starting an exercise and realising once you've started that are not doing what was asked..."). For each of the 30 items of the questionnaire, the child had to report if the item was a problem for him/her. The questionnaire was answered during an interview with the child without his/her parents, before the intervention. The trainer provided age-appropriate examples for the items the child did not understand spontaneously. At the end of the intervention the same person who trained the child throughout the rehabilitation programme, answered the questionnaire as

well, based on what she witnessed of the child's behavior during the intervention and based on contacts with parents and school. As the trainer had spent 15-20 hours with the child, it allowed her to observe carefully the child's functioning. The questionnaire score of MK awareness was obtained similarly to the KIC⁸¹, from the number of discrepant items between the child's rating of the questionnaire and the rating of the trainer. An item was judged as discrepant if the child responded it was not a problem, or a minor problem but the investigator thought it was a significant problem, obtaining a binary response for each item (aware/unaware). Potentially inversed discrepancies (the child thinking an item was problematic while the trainer responded it wasn't) did not occur. Items not relevant (e.g. forgetting books in the school bag, when school bag is not prepared by the child) were excluded, as well as items the examiner could not judge reliably at the end of the intervention. The final score was a percentage of "unaware items" divided by the number of relevant and reliable items.

The second measure of MK (the recognition of a common thread in the activities in which the patient is impaired and the implications this has) used stories contained in the pediatric GMT intervention²⁰⁰, and was assessed throughout the intervention. In these stories, characters have a series of daily life problems related to their executive dysfunction (e.g. putting an essay to hand in at school into a sports bag instead of a school bag). Stories are age-appropriate and consequences of the problems are emphasized (e.g. stress while looking in the school bag for the essay, feeling upset when finding the essay in the evening while getting dressed for football, getting a lower mark because the essay was handed in late...), as well as factors that contributed to the problem (e.g.: going to bed late because writing the essay at the last minute, being in a rush when preparing the school bag and the sports bag the next day). Use of PowerPoint slides with child friendly drawings, allowed children to follow the story without too much pressure on their working memory. The intervention contained a total of six stories, one every two weeks. At the end of each story, the child was asked "Do you think this could happen to you?" and it was followed by a discussion with the child about personal examples of cognitive failures and slips (called "Oops errors" in the intervention), implications and factors that contributed to these "Oops errors", and similarities with the stories. The awareness deficit score was the percentage of stories the child thought would never happen to him/her, while the trainer saw similar events regularly happening to the child, divided by the total number of stories. A child who acknowledged that this kind of story could

have happened to him/her, but could not provide any personal examples of similar cognitive failures, was still considered as being aware on that story (i.e. providing personal examples was not mandatory to score as aware on a story).

Emergent (i.e. on-line) awareness was assessed throughout the intervention by asking the child at the end of each session if s/he thought the exercise/activity had been difficult for him/her. Exercises could be office-set paper-and-pencil school type exercises or complex daily life tasks like real cooking. An awareness deficit was taken as a percentage of activities judged by the child as easy while s/he completely failed or required a lot of help to achieve the goal, divided by the total number of intervention sessions. On-line awareness assessment was conducted throughout the training, at the end of each session, with a total of 15 sessions. Children were also asked to identify “Oops errors” (cognitive slips) s/he had made during the exercise/activity, to qualitatively assess their on-line awareness.

Because awareness measures (except the questionnaire) were embodied in the intervention program, throughout the 15-20 hours of training, it was not possible to obtain data from healthy controls who did not follow the intervention on those specific awareness measures.

Although this was not intended to be part of the awareness assessment, we also report here the behavior of children during a pre-intervention test using cooking, the Children’s Cooking Task^{121,120} (CCT). In the CCT, children have to make a chocolate cake following a child-friendly photo-cued recipe. Children repeated the test twice before the intervention (to obtain two baselines). Children were not asked about their performance on the CCT because it would have acted as a cue for subsequent CCT assessments. However as behavior gave the trainer some insight into children’s on-line awareness through error detection on task, and possibly anticipatory awareness on the second attempt to make a chocolate cake (for second baseline), trainer qualitative observations are reported.

RESULTS

Five children were initially included in the study. One child (YR) dropped out after four sessions. Children’s detailed medical history, neuropsychological assessments and overall intervention effect can be found in table 1. All children suffered a severe dysexecutive syndrome, especially on ecological measures of executive functions (Children’s Cooking

Task and questionnaires). Neuropsychological assessment showed relatively preserved or even normal episodic memory (see table 1- except for RK, in story recall which was poor probably due to attention rather than memory problems) therefore high anosognosia scores in the stories and questionnaire did not reflect a memory problem.

Table 4.1 : Demographic, medical and neuropsychological characteristics of the participants

	YR	PB	CS	RK	IP
Sex	Boy	Girl	Girl	Boy	Boy
Age at inclusion (years)	14	11	11	13	8
Medical history					
Age at injury (years)	2.5	2.5	6.5	7	5.5
Initial GCS	<7	6	4	3	6
Brain imaging	Unknown	Large right hemisphere hemorrhage and edema, right parietal depression fracture	Cerebellar and right parieto-occipital lesion with depression fracture	Subdural hematoma with diffuse edema and pneumocephalus	Brain stem hemorrhage, Diffuse subarachnoid hemorrhage
Duration of coma (days)	Unknown	Unknown	1	10	6
Associated impairments, reported in medical records and previous assessments	Severe behavioral disorders	Epilepsy absences treated by carbamazepine	FSIQ 69. Impaired ToM and language pragmatics Spastic equinus foot	Attention problems Left arm weakness	ADHD
Glasgow Outcome Scale	3 (severe)	2 (moderate)	3 (severe)	3 (severe)	2 (moderate)
Neuropsychological assessment					
WISC IV matrices	11	9	7	16	11
WISC IV vocabulary	6	9	5	7	12
BADS-C 6 part test	7	10	7	8	6
CMS stories - immediate	9	8	8	4	8
CMS stories- delayed	8	5	7	4	9
CMS backward span	Missing data	8	8	10	18
CMS words list - immediate	Missing data	9	12	12	14
CMS words list - delayed	Missing data	13	7	16	16
Parents BRIEF T-score: BRI	63	71	87	95	57
Parents BRIEF T-score: MI	68	82	76	79	56

Table 4.1 (continued): Demographic, medical and neuropsychological characteristics of the participants						
Parents BRIEF T-score: GEC	68	80	82	88	57	
Parents DEX-C : Z score	3,53	4,7	3,7	5,1	0,7	
CCT: Z-score	Scoring impossible due to complete failure on task		4,8	2,2	7,1	18,0
Effect of Intervention (Context-sensitive pediatric Goal Management Training)						
Dropped out	Improved on EF questionnaires. No effect on cooking task.	Improved on EF questionnaires. No effect on cooking task.	Improved on EF questionnaires. No effect on cooking task.	Improved on EF and strategies on complex tasks, improved on cooking task, no effect on EF questionnaires		

Note: **GCS**: Glasgow Coma Scale score; **ToM**: Theory of Mind; **FSIQ**: Full Scale Intellectual Quotient; **ADHD**: Attention Deficit – Hyperactivity Disorder; **WISC**²³⁴: Wechsler Intelligence Scale for Children; **BADS-C**: Behavioural Assessment of the Dysexecutive Syndrome for Children; **CMS**¹⁵⁰: Children’s Memory Scale; **BRIEF**^{144, 145}: Behavior Rating Inventory of Executive Functions; **BRI**: Behavioral Regulation Index; **MI**: Metacognition Index; **GEC**: Global Executive Composite Score. **DEX-C**²³⁵: Dysexecutive questionnaire for children. **CCT**^{120,121}: Children’s Cooking Task.

Neuropsychological test results are reported as standard scores, unless otherwise stated. For the CCT and for executive functions questionnaires, BRIEF and DEX-C, a higher score indicates greater impairment. The clinical cut-off score for the BRIEF is set at a T-score of 65.

Paper and pencil tests of **executive functions (EF)**²³⁶ and detailed effect of intervention are reported elsewhere²⁰⁰; all children had at least two out of three EF tests indicating impairment relative to controls (< 2SD below controls’ scores)²⁰⁰.

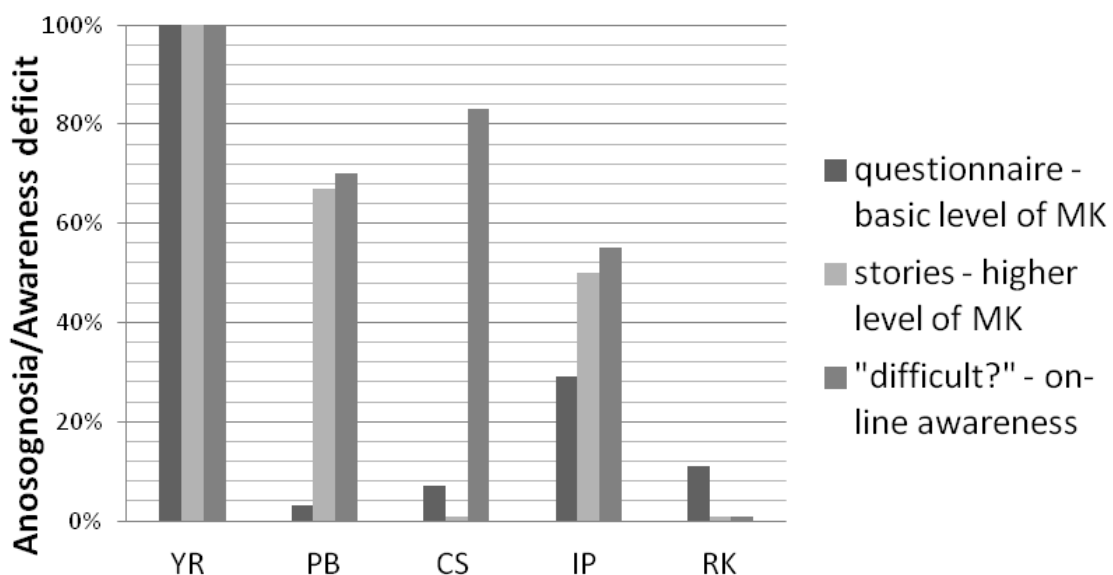
FEASABILITY

Our method of awareness assessment showed good feasibility. For the first measure of MK, all children were able to understand the thirty-one items of the questionnaire when provided with examples. The maximum number of items per child the trainer could not reliably assess at the end of the intervention was two. For the second measure of MK (stories), children enjoyed GMT stories and could understand them easily. All stories could have happened to the children and were relevant. Children could identify no “Oops errors” in activities they had judged as easy, even when they had failed the task.

AWARENESS SCORES

Awareness *deficit* scores for each type of awareness are presented in figure 4.2. Higher scores indicate that children are more anosognosic/unaware, i.e. representing a deficit in awareness. Lower scores reflect better awareness.

Figure 4.2 : Anosognosia/Awareness deficit scores for each type of awareness.



Note: Higher scores indicate that children are more anosognosic/unaware, i.e. representing a deficit in awareness. Lower scores reflect better awareness.

MK: metacognitive knowledge; **YR, PB, CS, IP, RK:** children's initials

QUALITATIVE DATA

CHILD 1: YR

YR was 14. He had sustained a severe TBI at the age of 2.5 years (collision with a running child); he attended a special school but was excluded from school for half of the year for behavioral issues. YR dropped out from the intervention after 4 sessions.

YR reported none of the questionnaire items as problematic, despite having been observed making frequent errors, resulting in a 100% awareness deficit score. As for pGMT stories of the modules he completed before he dropped out of the study (4), YR thought the characters must be “stupid” and such cognitive failures would never happen to him and never gave any example of personal “Oops errors” in the story discussion, nor did he acknowledge he made mistakes or errors in the ecological intervention activities. An example is his performance on the CCT (Children’s Cooking Task) prior to the intervention: YR found the correct recipe easily, took a quick look at the ingredients needed, put all the eggs he could find on the table, all the baking powder (5 packets), added one spoon of chocolate and one spoon of flour and put it in the oven. At the end of the task, both the child and the examiner tasted the “chocolate cake” that was rather a baking powder-flavored omelet, provoking instantaneous tingling in the mouth. YR said: “I didn’t know it was so easy to make a chocolate cake. But next time I’ll put slightly more chocolate” and wanted to eat the remaining cake. After being shown that on the same page there were stepwise instructions and asked if he thought he had followed them, YR looked perplexed for a moment and said in a defensive voice “No, I didn’t but I’ve managed well anyway”. YR made it clear he was not interested in the training and that he needed no help. However he was very interested in the module that explained executive function impairment and spontaneously admitted “That’s exactly my problem”, suggesting some intellectual awareness but then returned to a contemptuous attitude for the training. Before he dropped out of the study, YR participated actively in the sessions, while affirming he participated only because his parents forced him and that he had no need for it.

CHILD 2: PB

PB was 11. She was a passenger in a motor-vehicle accident at the age of 2.5 years with severe brain lesions requiring immediate neurosurgical treatment. She attended mainstream school with a part-time school assistant. She had epilepsy absences treated by carbamazepine.

PB was intellectually aware of her impairment on nearly all items of the questionnaire. Indeed, she understood she had difficulties with some activities, (lowest level of Crosson's intellectual awareness) and answered to most items in the questionnaire as "big, big problem for me", but she seemed unable to understand the implications of her deficits (highest level of intellectual awareness). While enjoying the GMT stories very much (our second measure of MK), PB never acknowledged such things could happen to her. For example, she recognized she often forgets and loses important objects (at school, she is late half of the time because she realizes on her way to school that she didn't take her schoolbag; she doesn't give forms to be signed by parents to her mother, who then misses important school meetings). However these situations did not seem problematic to PB, and there was no emotional reaction when these failures were discussed in relation to the stories. PB regularly took examples of her highly organized mother's rare executive failures as an excuse for her own frequent failures: "My Mum sometimes forgets her bag as well". As for on-line awareness, she judged most of the activities as easy, even when she needed considerable help to manage them. She never recognized she failed an activity and it was thus impossible to convince her that the training could help her. In the CCT, on her first attempt to make a chocolate cake she forgot the baking powder. On her second attempt, she focused on not repeating the same error and managed a beautiful looking cake, containing the baking powder, but this time missing sugar. While still unable to acknowledge the need to compensate for EF dysfunction at post-intervention testing, she was reported to have made significant progress on parental and teacher post-intervention questionnaires (see ²⁰⁰) and parents were highly satisfied with the intervention, reporting that she understood her brain functioning better.

CHILD 3: CS

CS was 11. She had sustained a TBI when a soccer goal post fell on her head at the age of 6.5 years, with mainly cerebellar and right parieto-occipital lesions. She attended a special school and a mainstream school part-time. Her intellectual quotient was on the lower limit of normal and she had impaired theory of mind and language pragmatics described in her previous rehabilitation reports. CS was aware of most problematic items of the questionnaire i.e. basic intellectual awareness. In our second measure of MK (stories), CS acknowledged all of them could have happened to her. However her behaviour indicated that she often simply "guessed" that she was expected to say "yes, that story could have happened to me" and she could not

provide any personal example of a similar “Oops error” in the discussion that followed the stories. She couldn’t evaluate her performance “on-line“ during or after activities, did not express awareness of “Oops errors” after the tasks and always thought activities were easy and that she did well, although she failed or needed much help on most tasks.

CHILD 4: IP

IP was 8 at the beginning of the training. A television had fallen on his head at the age of 5.5 years. He attended mainstream school with a part-time school assistant. He was diagnosed with ADHD, with symptoms that had probably started prior to the TBI. Very protected by his carers, who confronted him as little as possible with his difficulties (e.g.: managing his schoolbag preparation), he had few opportunities to notice that he was impaired and lacked knowledge about the consequences of his TBI. All three measures of awareness showed a moderate awareness deficit, with highest unawareness for on-line awareness. Very few examples of personal “Oops errors” could be obtained from IP and most of these were not appropriate. On his first attempt to make the chocolate cake, he used a small coffee bowl instead of the required salad bowl: after pouring in the sugar, the bowl was full, however he continued adding the other ingredients until the bowl was invisible and totally covered under a mountain of flour. He showed no manifestation of having detected this error and was very surprised when he looked at the photo of the next step of the recipe, depicting a half full salad bowl of cake mixture. He also had difficulty following the recipe steps and missed the step requiring to stir the mixture until it was smooth. He was very surprised that, once cooked, the “cake” had separated into white-flour and a black-oily layer. During training sessions, he alternated from great overestimations of his abilities (“I’m the most intelligent boy of my class, this exercise is just too easy”) to deep self-depreciation (“I can’t do it because I’m just too stupid”) and presented great emotional reactions to his performance, including inconsolable crying when he was failing a cooking recipe, inappropriate laughing, and rolling on the floor when he could not find a solution to a problem.

RK was 13. He had sustained a motor-vehicle pedestrian accident at the age of 7. He attended part-time private schooling with school assistant part-time private lessons. He presented with severe attention problems. Although 13 at the time of the study, his parents could never leave him at home alone, he was forbidden to enter the kitchen as he usually forgot to switch off the gas. RK was fully aware of his impairments on all three measures. He is the only child who actually acknowledged the utility of strategies taught during the intervention and used them to compensate for EF dysfunction post-intervention. However effects did not transfer to natural contexts and no change was reported by parents on post-intervention questionnaires²⁰⁰. However he seemed unaware of his lack of cognitive flexibility. This was illustrated by a cooking episode at home reported by his parents. He decided to make finger biscuits (that he had practiced during the intervention) for his large family. He decided to multiply all ingredients by 5 (which would give over one hundred biscuits), because it would require 500 grams of butter which is the usual size of butter packs found in shops. His parents tried to persuade him that this would be too much and that multiplying recipe quantities by two would suffice. He could not accept it, even though there was no problem with mathematical skills and he refused to make the biscuits at home with any other quantity. A whole intervention session focused on this problem without success.

DISCUSSION

All three methods of awareness assessment showed good feasibility. Apart for one child who presented complete anosognosia on all awareness measures (YR), children showed relatively preserved basic intellectual awareness but more difficulty in the higher order intellectual awareness and on-line awareness. Observation of the cooking activity on the CCT allowed an interesting insight into the children's awareness and especially error detection.

MEASURING METACOGNITIVE KNOWLEDGE/ INTELLECTUAL AWARENESS

Many ways of assessing awareness in adults have been described in the literature (see Ownsworth and Clare¹¹⁶ for a review). Intellectual awareness is commonly assessed through discrepancy scores between the patient and a significant other's rating on a questionnaire (e.g.

Dysexecutive questionnaire²³⁷, Patient Competency Rating Scale^{238,239}, Awareness questionnaire²⁴⁰). Self-report versus test performance is another way of assessing intellectual awareness. Our approach was a discrepancy score approach but was close to the self-report versus test performance approach, as the therapist was the informant, and most items judgments were based on children's actual behavior and performance throughout the sessions. The questionnaire was easily understood by all children and, as the informant was the therapist (taking into account interviews of parents and school staff), we probably managed to have a more objective informant report than when parents answer questionnaires alone. However, the questionnaire was long and items related to the content of the intervention (Context-sensitive Goal Management Training²⁰⁰), that focused on improving executive functions and prospective memory. This questionnaire would need to be shortened and simplified further if applied to cognitive rehabilitation outside GMT training context.

Crosson's levels of intellectual awareness proved useful in explaining PB's dissociated intellectual awareness: excellent awareness of impairments on the questionnaire (lower level of intellectual awareness) but difficulty in recognizing common threads between activities she is impaired in (on the GMT stories) and total unawareness of the consequences of these impairments (highest level of intellectual awareness) when these were discussed during training sessions. The difference between the two levels of intellectual awareness described by Crosson: (1) knowledge of impairment and (2) implications of impairment is probably a key issue in research with children. PB could state her impairments well, but did not see the implications of them - and thus did not acknowledge the need to compensate for them.

The GMT stories offered the advantage of presenting to the child both impairment and its consequences in an accessible story, with a visual support that lowers working memory and language demands. Through these stories of daily living, we hoped the child would understand the cognitive failure and its consequences and use it to recognize how cognitive failures may impact on home, school and leisure activities. Children found the stories fun and understood them easily, as opposed to more general questions that have been used to assess intellectual awareness in children^{224,228}. However, this approach for assessment of awareness requires intact theory of mind (ToM) skills. First level ToM is needed to adequately understand how the character thinks and feels in relation to the situation. Second level ToM is needed to understand what the character thinks the surrounding characters are thinking (e.g. a

story where a boy forgets the ball he was supposed to take to a football match with his friends, who become angry with him). Unfortunately we did not assess ToM in participants, nor did we ask control questions to check if the children had sufficient ToM ability to fully understand how the character is feeling. As children who sustain a TBI can suffer impaired ToM^{11,241}, such an assessment should control for ToM. A way to control for ToM would be to ask the child how the character is feeling instead of including this information in the text of the story. For example, CS, who was described in her medical reports as having poor ToM, had difficulty understanding the stories. She could not give personal examples of similar events, and struggled to understand how the reaction of characters in the story related to cognitive failures. She also did not appear to be aware of the consequences of the cognitive slips. However, she often answered that “yes that story could have happened to me”, relying on her knowledge that she often does things wrong and she simply “guessed” what she was expected to say. Therefore her story score seemed unrealistically good and therefore unreliable, probably because of her reasoning and ToM impairment. ToM has shown to be correlated to self-awareness²⁴² in adults and must be assessed if stories are used to assess metacognition. On the other hand, some authors proposed that metacognition and awareness reflect ToM about one’s own thoughts^{243,244}, and that both rely on the same cognitive ability and common brain structures²⁴⁵. Another problem with using stories to assess awareness is that the measures can be biased by children’s incapacity to generalize the situations presented to other similar situations where the same cognitive failure can occur.

ON-LINE AWARENESS

MEASURING ON-LINE AWARENESS

On-line awareness is difficult to assess. In the literature several approaches have been described: (1) think aloud protocols (the child is told to say aloud everything he/she is thinking²⁴⁶; (2) video analysis of performance where the examiner looks for signs of error detection (verbalization, non word exclamation such as “oops!”, facial expressions, head-shaking, manual gestures) in the patients behavior²⁴⁷; (3) forced on-task verbalization of error detection (e.g: patient instructed to say “hit” whenever he/she notices that he/she made an error)²¹⁴; (4) event-related potential examination during task²⁴⁸. Think aloud and forced on task-verbalizations are problematic in that they inevitably become a dual task paradigm and/or a prospective memory task: They require additional attention allocation to the task of showing

the examiner an error has been detected. Furthermore in both these approaches, as the patient is informed of being assessed/watched on his/her error detection skills, s/he is likely to focus on error detection rather than the task itself, and as a consequence provides an unecological (and thus inaccurate) measure of error detection capacity in daily tasks. Conversely, behavioral signs of error detection probably underestimate error detection, as very early detected errors and errors with little consequences are unlikely to be expressed by overt behavior, especially if the patient wants to appear successful. Event-related potential offers an alternative way of assessing error detection²³². Following an error, an error-related negativity is registered, that has been argued to correspond to the unconscious (implicit) error detection, whereas conscious errors are followed by a positive deflexion²³². This post-error positivity is reliably associated with decreased on-line awareness of deficits²⁴⁹ and was proposed as an electrophysiological indicator of on-line awareness in adults. Event-related potential studies suggest that a child may have no experience of making an error, either because s/he truly does not *detect* errors, or because implicitly detected errors are not brought to consciousness. From a rehabilitation point of view, it has consequences as to where to focus intervention: it seems illogical to try and make children consciously aware of errors they haven't detected "electrophysiologically", at a basic, unconscious level. From a developmental point of view, it is interesting to note that the error-related negativity that follows all errors (conscious or not) has been shown to increase with age, whereas the subsequent positive deflexion (present only for conscious errors) – marker of on-line awareness –, is stable with age. Although theoretically promising, event-related potentials are impractical to use during rehabilitation sessions and will probably remain a research tool rather than being used routinely for clinical purposes.

TRULY "ON LINE" VERSUS "OFF-LINE" AWARENESS: PREDICTION AND EVALUATION OF TASK PERFORMANCE

In our study we tried to find a way of assessing on-line awareness, without disrupting the ongoing task. Our measure of on-line awareness (responses to the broad question "Was the task difficult for you?" and post-task error recollection) had the drawback of requiring preserved memory of performance, as children were asked the question only after the task. It relied on post-task evaluation, rather than "truly on-line" error detection and was therefore rather a measure of "off-line" awareness. The term "off-line" metacognition/awareness has

been proposed by Desoete⁵⁴ in her study of metacognition in math problem-solving. Off-line metacognition includes both prediction (of difficulty, of time required to complete the task) and post-task evaluation of performance. Emergent/on-line awareness can be considered to comprise truly "on-line" awareness (error detection, monitoring of performance *during* the task) and off-line awareness (immediately *before and after* a task). Off-line is included in on-line awareness because it is activated within the context of a specific situation or task and involves judgments about one's abilities and limitations in relation to the current situation. Furthermore a complex task such as cooking is a series of subtasks and therefore predictions, error detection and monitoring and evaluation are continuously needed throughout the task: as such off-line awareness is needed throughout the task.

PREDICTION OF PERFORMANCE AND ANTICIPATORY AWARENESS

The exact distinction between prediction of performance and anticipatory awareness is unclear in the literature and therefore the two terms are sometimes used interchangeably. Our view is that prediction of performance is a skill, which can be prompted and is cognitive in essence. Anticipatory awareness is the behaviour that results from *spontaneous*, correct prediction of task difficulty and of self-capacity to deal with the task. Anticipatory awareness is very difficult to capture in an assessment because it is a behaviour rather than a measurable cognitive function. Anticipatory awareness is expressed when the potential for a problematic situation/task arises in daily life. All attempts to capture anticipatory awareness in an office-based interview or assessment are unecological, because asking the patients how they perceive their ability in a hypothetical situation provides a prompt for awareness and assesses intention of behaviour. It does not reflect actual behaviour in such a situation, in daily life, and in the patient's usual environment. Most assessments termed "anticipatory awareness" assessments are really "off-line" prediction awareness measures (e.g.: asking the patients : "how do you think your performance on the task might affect your ability to live independently, work and have fun?"²¹²; or predicting memory span²¹⁴).

In our study, none of the three measures of awareness served to assess prediction or anticipatory awareness. However qualitative observation of children's behaviour gave some insight into their anticipatory awareness, especially on their second attempt on the CCT. For example PB, who had only very basic intellectual awareness, could predict she might forget

baking powder again and thus concentrated on adding the baking powder on the subsequent CCT attempt.

Anticipatory awareness probably has different levels as suggested by PB: she could anticipate she would forget the baking-powder (lower level of anticipatory awareness, in this case “baking powder-specific”), but could not generalise this anticipatory awareness to other ingredients she might forget (higher level of anticipatory awareness, “ingredient-specific”), or to the anticipation that she may forget not only ingredients but whole recipe steps, switching on the oven (highest level of anticipatory awareness), or even more broadly that she is very disorganised irrespective of the type of task (generalised anticipatory awareness, in other activities such as cooking in general, do-it-yourself, homework...).

DETECTING ERRORS ON-LINE AND ERRORLESS LEARNING

Another issue for our measure was the help given to children on the tasks. For unfamiliar tasks such as cooking, the considerable help provided by the trainer probably seemed natural to children and – with the help received – the task might be perceived as “easy”. On-line awareness can probably be experienced only if a patient is allowed to struggle on task, which was not the case during the intervention, as our first aim was to train children in novel complex task management, and only secondly to assess awareness. This issue is particularly important as some authors advocate errorless learning for patients with dysexecutive syndromes²³⁰. Classically, errorless learning has been used in patients with memory deficits: Errorless learning is based on the assumption that explicit memory for errors is impaired, whereas implicit memory is not, meaning that errors are primed, and so are more likely to be repeated. In errorless learning instructions, the aim is to try to prevent patients from making errors during the learning process. However it has also been proposed to use errorless learning in patients with a dysexecutive syndrome without memory deficits. In those patients, errorless learning is based on the assumption that the error-monitoring system is defective²⁵⁰²⁵¹, whereas implicit memory is not, yielding a memorisation of the undetected error. In errorless learning patients are not given the opportunity to detect their errors because errors are prevented by the therapist. It is possible therefore that this could impair the development of on-line awareness by ensuring success on tasks at all times and offering no opportunity for the error-monitoring system to be trained/used. Therefore error-based learning has been proposed to improve self regulation in adults with ABI²⁵². *Error-based* learning may be useful in

helping children to develop their on-line awareness through error detection, but this must be considered carefully and evaluated for children with severe episodic memory impairments, who may not remember the error they detected and therefore may profit more of an *errorless* learning approach. It would be interesting to include an assessment of awareness (and especially on-line awareness) in trials comparing errorless and error-based learning.

STEPS REQUIRED FOR CORRECT POST-TASK EVALUATION

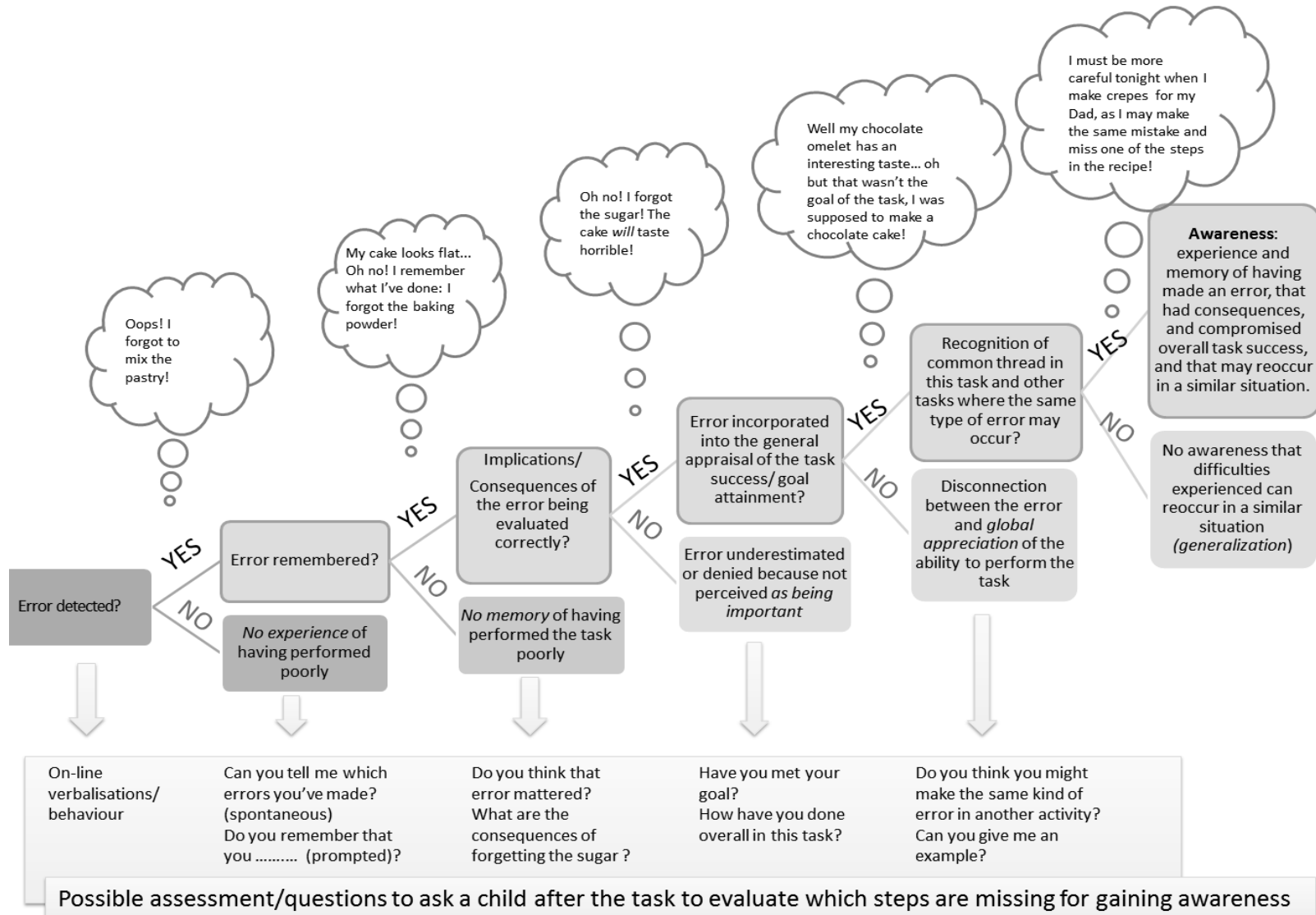
Post-task evaluation was influenced by different factors in our study, and not only error detection ability. Qualitative analysis of on-line awareness measures, based on the performance of the five children on the ecological training activities and on the Children's Cooking Task lead us to propose a series of steps necessary for adequate post-task evaluation, (where error detection is only the first step). Figure 4.3 presents examples of children's (real or hypothetical) verbalisation for each step, corresponding to a situation where the child is aware. In our view, a child must go through all the steps to truly *experience* the difficulty of having trouble with a task (i.e. on-line awareness). We suggest that to understand a child's on-line awareness deficit in rehabilitation, each required step should be assessed separately (see bottom line questions in figure 4.3).

The consecutive steps are: (1) Errors must be detected and brought to consciousness (see previous section on measuring on-line awareness); (2) Even if the error has been consciously detected on-tasks, a child may have no memory of having performed poorly on the task because of episodic memory impairment (she/he does not encode performance on the task in episodic memory or does not encode it as an error or cannot access the memory). Even if an error is detected and remembered after the task, children may (3) not appraise the consequences/implications of the errors and therefore not interpret them as errors (errors with little consequences, or unknown consequences or corrected errors not being considered as errors) resulting in a very positive self-evaluation of performance; (4) neglect the main goal of the task. These last two factors are probably extremely important in children, as they tend to focus on things that went right or that were fun, rather than on errors which are thought of as minor details. All children had so much fun trying to make a chocolate cake in the CCT, that even totally failed cakes were given a positive appreciation by the child who made them: IP did not know that not mixing the liquid and the flour would result in an inedible cake. He could not make a connection between the cake consistency and his omission of mixing the

ingredients (erroneous appraisal of consequences of the error). YR was pleased with his 'baking-powder omelette', although he acknowledged he was supposed to follow the recipe steps to make a chocolate cake (neglect of main goal); (5) For some children, in spite of adequate recognition of within-task errors, there is an inability to relate these apparently unique errors to previous similar experiences, nor to anticipate potential future situations where the same problems are likely to occur, impeding the child to generalize his/her on-line experience.

It is worth noting that a child's evaluation of his/her performance may be biased by some form of denial (i.e. the child detects, remembers, correctly appraises the consequences and places them in the context of the task goal but *denies* the error occurred, has consequences). Pure denial is rare in children⁸³ but is often the explanation for behaviour that might in fact be caused by awareness deficits⁸³. In Beardmore et al. Study⁸¹, emotionally-motivated or defensive denial (operationalised as a negative/avoidant coping style) was not supported as a contributor to the child's poor metacognitive knowledge.

Figure 4.3: On-line awareness steps required to arrive at an adequate post-task evaluation



THEORY OF AUTOBIOGRAPICAL METACOGNITIVE KNOWLEDGE ACQUISITION BASED ON ON-LINE EXPERIENCES

We hypothesize that metacognitive knowledge is constructed from on-line awareness experiences, that will progressively allow the child to construct a sense of his/her own cognitive abilities, through the construction of semantic and autobiographical memories of task performance and difficulties. In the same way that children's development of autobiographical memory²⁵³ is related to the understanding of their own mental states in the past^{254,255}, we postulate that self-awareness is related to the memory and understanding of on-line awareness experiences in the past.

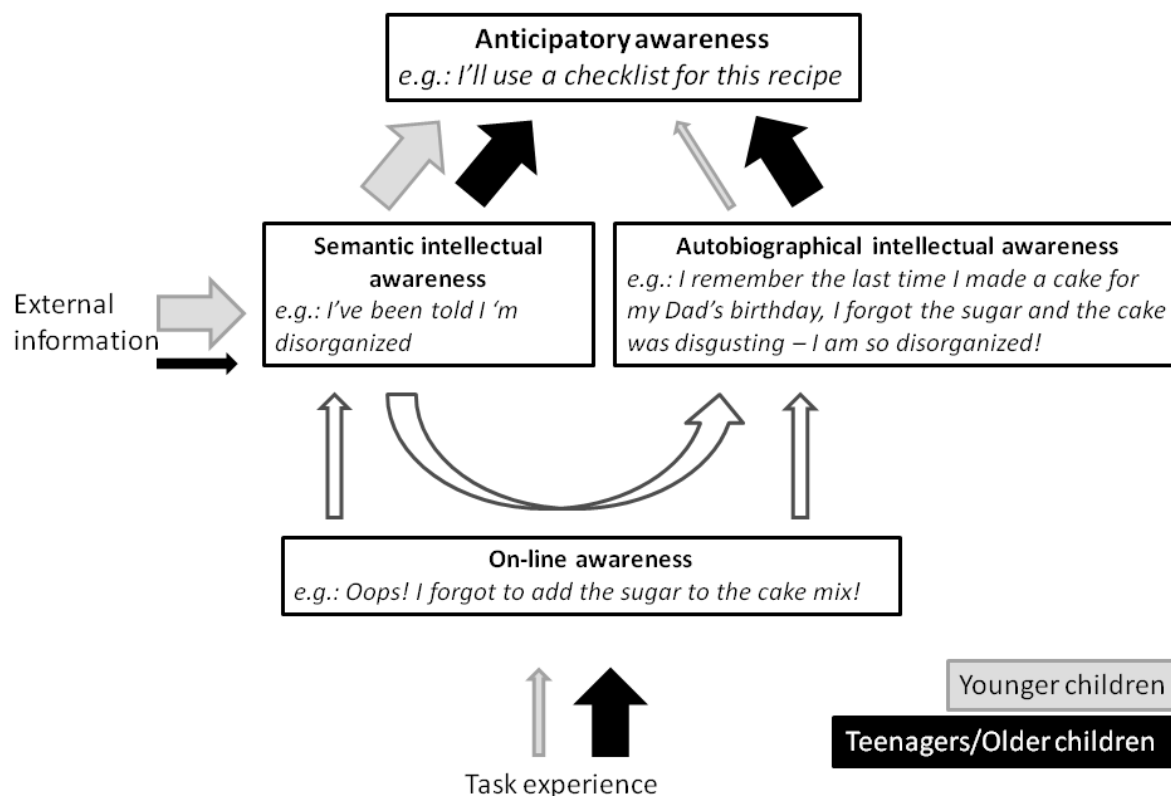
Let's first take the simpler case of an adult. Dirette¹¹⁷ suggests that awareness of cognitive deficits develops through "aha" moments, particularly in functional activities (e.g. being aware that one cannot drive from the actual experience of driving, rather than having been told you cannot drive) and familiar places (cooking at home rather than in the rehabilitation centre). When an adult is told that s/he is not able to drive, s/ he may gain some general self-knowledge about his/her driving capacity, however this will remain a very theoretical knowledge, which we will call "*semantic*" metacognitive knowledge (MK). To be truly aware of his/her driving difficulty, s/he needs his/her own experience of performing poorly in driving, with preserved on-line awareness of making errors to support self-appraisal of driving capacity. If on-line awareness is intact, it is the actual experience of being conscious of driving dangerously that will allow him/her to truly internalise knowledge of his/her driving abilities, make it his/her own, store the "trying to drive" episode in his/her autobiographical memory and acquire MK that has "*autobiographical*" characteristics, including the phenomenological details accompanying the experience of having difficulty on the task (e.g. feeling anxious, hearing the horns).

Young children acquire most MK from adult reminders and feedback ("put it in your schoolbag or you'll forget it", "you're very disorganised"). Eventually a child who has been told for years that s/he is disorganized (like PB in our study), will acknowledge s/he is, gaining some basic semantic intellectual awareness (or semantic MK), by storing the information "I am very disorganised" in his/her semantic memory. However this semantic MK will remain a fact, without a link to an episodic sense of self.

From a developmental point of view, semantic MK precedes on-line awareness⁵². With age, children become increasingly conscious of cognitive capacities, strategies for processing information, and task variables that influence performance²⁵⁶ and have more attentional resources to devote to on-line monitoring/error detection. As on-line monitoring skill becomes more efficient, children's on-line awareness increases and allows them to be aware of having difficulty with a certain task (following the steps described in the previous section). Progressively, memories of correctly appraised performance are stored in memory resulting in increased MK, that does not rely solely on what the child *knows* because s/he has been told by his/her parents (basic semantic MK) but on his/her own experience, with all the phenomenological details accompanying this experience (e.g. feeling anxious when realising school bag is missing, smelling the burnt cake). Therefore the memories of these correctly appraised experiences, which the child will truly *remember*, will allow full awareness (comprising both semantic and autobiographical components) and will hopefully be retrieved and used to anticipate future problems (anticipatory awareness) in similar situations (see figure 4.4). However, because autobiographical memory matures gradually throughout childhood²⁵³, a unique experience of failure will probably not be stored or retrieved as clearly as in adults. Children may recall for example that they enjoyed cooking, and what they managed well, rather than remembering their difficulties in planning on-task and the strategies they used to overcome difficulties.

Conversely, for younger children, most MK will be gained from external information and their MK will essentially rely on the "semantic" awareness (see fig 4.4). In our view autobiographical MK/intellectual awareness develops gradually, along with the development of autobiographical memories, and young children should not be expected to have complete intellectual awareness. However it is worth noting that younger children may still exhibit anticipatory awareness, based on their semantic MK: They may not have *lived* the experience of being impaired (or developmentally immature) on a task, but may rely on what their parents taught them ("don't cross the street on your own") and on their semantic general memory (e.g.: children don't drive cars).

Figure 4.4: Autobiographical awareness mediated by on-line experience model. We hypothesise that the relative importance of different inputs varies with developmental age



Our theory could explain the dissociation in the MK scores in the discrepancy questionnaire score and GMT stories. The questionnaire assessed basic knowledge of functioning, which can be qualified as the “semantic” MK/ intellectual awareness about self and was probably mainly acquired by repetitive comments of adults (parents, teachers...). Stories on the other hand, especially when they elicited pertinent examples of personal cognitive failures in the autobiographical memory, relied more on autobiographic MK/ intellectual awareness, by cueing the children on consequences of the cognitive failures and phenomenological details developed in the story (how the character felt, what were the circumstances...).

FACTORS CONTRIBUTING TO POOR AWARENESS IN CHILDREN

It is often said that ‘It takes a whole life to know oneself’, reflecting that even for adults, being fully aware of one’s strengths and weaknesses is challenging. Children with brain injury might have specific deficits that impact on their functioning, but these are set in the context of (1) having cognitive functions that are constantly developing, (2) being supported by parents, teachers and others to carry out new tasks (so not necessarily having the experience of difficulties) and (3) having limited attentional/executive resources that make monitoring and reflecting on performance difficult ²⁵⁶. Thus it might be said that being self-aware is a challenge for everybody, but for children it is particularly difficult and hence for children with brain injury it is major problem ⁸¹.

In our study, children’s awareness was influenced by many factors. For the youngest (IP), a lack of knowledge about his impairments due to a lack of confrontation probably explained much of his unawareness. Reassurance and help from parents assured success on tasks but also contributed to his lack of awareness. However, IP’s emotional responses to failures, suggest that such a non-confrontational reassurance may not be appropriate, even for children as young as 8. Another factor is the type of activities children are confronted with: for most children, it appeared obvious that they would be given help if they attempted to cook a cake and therefore judged the task easy, because adult help was inherent to the task at their age. For YR, part of his unawareness was probably due to a denial of his impairment or at least a wish to hide any difficulty.

Awareness is probably easier to gain for some cognitive functions than others, and from our data, cognitive flexibility seemed to be the most difficult cognitive function to gain awareness of. This was illustrated by RK who showed excellent awareness on all measures, who frequently talked about his memory and planning difficulties, but who could not acknowledge his cognitive rigidity. Questionnaires and stories did not include aspects of cognitive flexibility and it would be a valuable addition for further development of awareness assessments in both children and adults. For example, it would be interesting to see whether RK would react to a story where a character shows a difficulty with cognitive flexibility (such as RK’s own relating to quantity of cooking ingredients). However being aware of one’s own cognitive rigidity and detecting it on task *is*, in itself, something that requires cognitive

flexibility; therefore awareness of cognitive flexibility deficit is probably, by definition, incompatible with such a deficit.

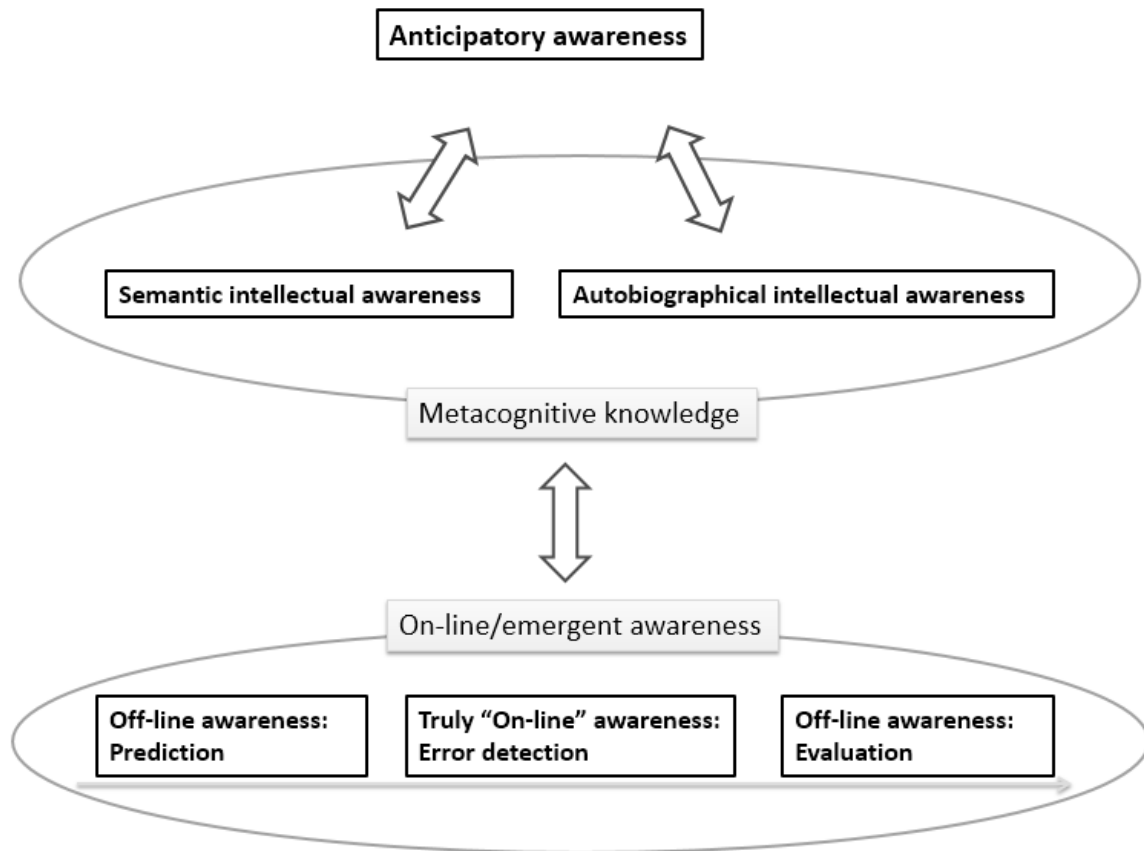
LIMITATIONS AND RECOMMENDATIONS

It is not possible to determine from our data if the children had more impaired awareness than healthy children, as we did not include healthy controls with whom to compare awareness scores. Developmental studies are needed to explore awareness in healthy children, without which literature lacks a reference to normative standards and degree of awareness impairment in our clients who suffered a TBI cannot be precisely determined.

Awareness is multicomponential, and different components have different importance depending on the child's age (e.g. autobiographic awareness is not expected before late childhood/adolescence). Each component should be assessed separately to truly understand where the child's foundation of unawareness deficit lies.

Figure 4.5 summarises the awareness components that are easily measurable separately. Real cooking seems a feasible, fun and ecologically valid way for studying on-line awareness (please refer also to figure 4.3).

Figure 4.5: Proposed framework for awareness assessment



Prediction and evaluation are important to assess but also to practice and should be included during activities at school and at home (refer to Ylvisaker⁸³ for details). For intellectual awareness, stories seem a particularly well-suited assessment because (1) children find them fun; (2) children understand them easily; (3) drawings lower working memory demands; (4) consequences and factors contributing to cognitive failures can be included in the story and elicit awareness of them. The story format however does not appear to be appropriate for children with severe reasoning or ToM deficits. Brain storming about personal examples of cognitive failures elicited by stories (or by another support including questionnaires) allows one to determine whether children rely on semantic awareness, autobiographical awareness or both for their responses. The remember/know paradigm and explicit requirement to provide phenomenological details can be used to differentiate between autobiographical and semantic awareness (see^{253,257} for details about the paradigm referring to autobiographical memory). Assessing anticipatory awareness remains a challenge but is most likely to be valid during ecological activities at home, school or during rehabilitation activities using real life settings

and activities such as cooking, without prompting and without explicit knowledge of the child of being assessed.

So far metacognition in children has been mostly explored in meta-memory research. As EF are the main problem for independent daily living, “meta-EF” studies (and especially “meta-EF” in ecological settings) are urgently required before engaging children and teams in clinical and research programmes of metacognitive strategy training that rely on awareness, which may be recommended for adults but whose appropriateness for different stages in childhood are not yet established.

DECLARATION OF INTERESTS

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PART 2: INTERVENTION

- Intervention Theory
- Experimental data

INTERVENTION THEORY

CHAPTER 5: REHABILITATION OF EXECUTIVE FUNCTIONS IN CHILDREN

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EF IN CHILDREN

There is general agreement that there are three core executive functions (EF) ^{25,26}: inhibition [inhibitory control, including self-control (behavioural inhibition) and interference control (selective attention and cognitive inhibition)], working memory (WM), and cognitive flexibility (also called set shifting, mental flexibility, or mental set shifting and closely linked to creativity). Working memory supports inhibitory control (you must hold your goal in mind to know what is relevant or appropriate and what to inhibit) but inhibitory control in turn supports working memory (by helping to keep our mental workspace from becoming too cluttered by suppressing irrelevant information from WM limited-capacity workspace. From these, higher-order EFs are built such as reasoning, problem solving, and planning ³⁵. This model of EF in child development is summarised in the figure Intro.1 (see introduction chapter) ²⁴.

DEVELOPMENTAL ISSUES IN SUPPORTING EF FOR CHILDREN

It is essential that interventions are developmentally coherent. This is of prime importance when considering executive functioning. It is known that full myelination of the frontal lobes is not reached by 20 years of age ²⁵⁸ and corresponding executive function skills are still maturing in late adolescence and early adulthood ²⁵⁹. As such adult interventions for EF are unlikely to be beneficial to children, particularly if they require cognitive functions that would not be expected to have developed yet such as metacognition and self-awareness ²⁶⁰. Strategy use develops throughout childhood and as a result specific strategies that are effective for adults may not help children ^{261,261}.

Ideally, EF interventions should be implemented after core skills that may serve as prerequisites to EFs have sufficiently recovered, are supported as well as they can be or have naturally matured. The paediatric neurocognitive interventions (PNI) model is potentially useful as a framework for deciding priorities for intervention ²⁶². In addition, a multidisciplinary approach is essential for children with moderate to severe impairment of EF

to ensure that interventions are appropriately considered within the context of the range of physical, cognitive and behavioural needs of the individual ²⁶³.

Some core principles in relation to executive function interventions are emerging. For example, EF must be continually challenged, i.e. increasing difficulty level as soon as attainment is reached, to see improvement ^{264,265}. Interventions may need to be long and intensive and this may be a major factor that impedes EF interventions in the literature so far. Increasing intensity and frequency is likely to require support in everyday settings, and Ylvisaker proposed that “targeting EF goals throughout the day in the context of everyday activities and routines is better than targeting these goals in highly specialized therapy or instructional periods.”²⁶⁶ (p243).

Ideally, parents would be involved as often as possible ^{267–269} as they are the most present “cognitive coaches”, although involving families may be difficult, especially if parents themselves lack metacognitive skills ²⁰⁰. Working on parent interactions with their child before EF, is perhaps in some cases the first necessary step. Interestingly, parenting programmes such as “I-Interact” ²⁷⁰ have shown more effectiveness for low-income families, who may need support in optimising parent-child interactions.

CURRENT PRACTICES IN EF INTERVENTIONS FOR CHILDREN

Many strategies have been proposed to help children with executive dysfunction, and useful manuals of EF interventions have been proposed ^{271–275} however most of them have never undergone rigorous evaluation and do not necessarily acknowledge the importance of the child’s age and stage of development.

There is a lack of validated methods for EF rehabilitation in children outside WM ^{110–113,276}. For most approaches there is the difficulty of a lack of generalisation and transfer to everyday contexts ²⁷⁷. In current clinical practice, four key approaches are used to help children with their EF difficulties, though each has its limitations:

(1) Providing environmental support and compensatory aids (e.g. use of electronic prompting devices ^{278,279}– though only a restricted number of situations lend themselves to such an approach;

(2) Training component EF skills (i.e. working memory, inhibitory control, cognitive flexibility). -Use of easily commercialized games (e.g.: Jenga ©, Highlights ©) that target EF ²⁶⁹ or computerized trainings are a way for training component EF skills ^{106,280,105,281}. There is some evidence in the literature to support the use of external delays to inhibit a prepotent response, an approach commonly seen in ADHD intervention manuals ²⁸², which has been shown to be effective in typically developing children as young as 3 years ²⁸³. In addition, there is an emerging evidence-base to support cognitive flexibility training through cognitive remediation therapy for adolescents with anorexia nervosa ²⁸⁴. If it is possible to train component skills in children with a brain injury, transfer and generalisation to other functional daily life situations would need to be demonstrated and the importance of integrating these components into a holistic programme considered.

(3) Training children on specific goals (e.g. preparing their schoolbag). For children with severe EF deficits, who often have a range of associated difficulties (and who are often excluded from trials) it has been proposed that such goal-focused rehabilitation, aiming at improving specific tasks (rather than improving global EF) may be a reasonable option ²⁰⁰.

(4) Providing children with metacognitive strategies applicable to a variety of everyday situations. Whilst this is effective in some adults after TBI ^{114,285}, there is little evidence that metacognitive training is effective for children with a dysexecutive syndrome post TBI ^{81,200,286}, although research on children with other primary difficulties following brain injury suggests it may be a useful approach for some ^{108,287}. Strategies are usually defined as deliberate, goal-directed mental operations that are aimed at solving a problem ²⁸⁸. Although younger children may not produce a strategy spontaneously, they can be trained to use a strategy and enhance their performance as a result ²⁸⁸. By definition strategies are effortful to use and children who use a strategy (especially a new one) may not have enough mental resources ²⁸⁸ and especially WM space left to devote to solving the problem at hand and, as a result, the strategy may not improve their performance. This phenomenon is however short-lived in typically developing children. It is still not clear if this is also the case for children

whose EF and metacognitive abilities have been compromised by a TBI and if metacognitive training can be an effective approach for them. In Krasny-Pacini et al. study ²⁰⁰, children were happy to try the metacognitive strategies on paper-and-pencil tasks but showed no consistent application of strategies in the meaningful, more complex concrete activities (e.g.: cooking, route finding). The same finding has been reported in the study by ²⁸⁶: making the intervention fun was identified as being useful, whereas the “Goal-Plan-Do- Check” strategy and promotion of good strategy use were not. Contrary to adults, Metacognitive Strategy use may not be the best rehabilitation approach for young or severely dysexecutive children. For children who may benefit from strategies, they need to be simple, concrete, and repeatedly practiced in order to be successful, unlike adults (with mild EF dysfunction) for whom providing the strategy may be sufficient.

PROMISING APPROACHES

Although evidence for effective EF interventions is scarce, a number of promising approaches have been published that emphasize (1) the necessity of a context-sensitive approach (with an important body of work by Marc Ylvisaker, Timothy Feeney and collaborators; (2) the necessity of prolonged interventions to show benefit; (3) the importance of involving children’s “everyday people” (e.g., teachers, parents, school assistants) for obtaining an intervention effect; (4) the necessity of practicing EF on ecological, meaningful tasks rather than office-based generic exercises.

Promising interventions using self-regulatory or metacognitive approaches focusing on functional task performance have been published as pilot studies in children with ABI : (1) “*Goal-Plan-Do-Review*”^{289–291}, contextualized within the routines of everyday life ^{292–294}; (2) “Goal Plan Do Check” ²⁸⁶ used in Cognitive Orientation to Daily Occupational Therapy-CO-OP ²⁹⁵; (3) “Stop, State your Goal, Split Task into subtasks, Do it, Check” ²⁰⁰ adapted from Goal Management Training ⁹⁸; (4) “Nature Planning Representing Monitoring”²⁸⁷. Feeney and Ylvisaker’s approaches offer the most robust and positive findings, using well-designed single-case experimental designs and replication studies.

In Feeney and Ylvisaker’s “Context-sensitive support”, the intervention was delivered by school staff under the supervision of a consultant. The students were given a graphic “map”

that represented the general sequence of activities from an executive function perspective: Goal (i.e., “What are you trying to accomplish?”); Identification of difficulty level (i.e., “Is this going to be hard or easy?”); Plan: (i.e., “How do you plan to get this done? What do you need? What are the steps? How long will this take?”); and Review (i.e., “What were you trying to accomplish? How did it work out? What worked for you? What didn’t work? What was easy? Difficult?”).

Case studies have also been published using similar approaches: Glang et al. ²⁹⁶ reported generalization to school functioning of an educational approach, “Direct Instruction”, that utilizes task analysis, modelling and shaping. Suzman et al. reported positive gains after a self-instruction and metacognitive strategy multi-component package ²⁹⁷.

Ylvisaker’s work remains a primary resource for clinicians ²⁹⁴ and much of the research described above is based on these fundamental concepts. For Ylvisaker, “the goals of environmental EF interventions are to help children (1) know what they are good at and what is difficult for them; (2) know that there are special things that can be done when tasks are difficult; (3) make plans for getting things done; (4) pay attention to how well they are doing; (5) try a new approach if they are not successful. [...] The overriding goal for a rehabilitation program is to create a culture within which there is developmentally appropriate focus on EF throughout the day”²⁶⁶ (p 242). The goal of EF intervention is to improve executive behaviour in everyday life. Any activity can be used to promote EF including the following aspects: (1) choice of goal; (2) a decision about how easy or difficult it will be to accomplish the goal; (3) Development of a plan; (4) Coaching in problem solving and strategic thinking as issues arise; (5) a quick review of the goal, the plan, and accomplishment at the end; (6) a summary of what worked and what didn’t work, of what was easy and what was difficult. Initially children’s participation may be minimal and adults may need to model, conversationally, most or all components of the routine.

PROBLEM-SOLVING INTERVENTIONS IN CHILDREN WITH ABI

The work of Shari Wade and colleagues ^{270,298–300} using development of web-based problem-solving interventions suggests that different interventions supporting EF may be required at

different ages. Following a pilot study in 2005 using a family problem-solving intervention³⁰¹, they further divided their interventions into a parenting programme (I-interact, ²⁷⁰ for children aged 3-9 years who were showing behavioural difficulties (i.e. poor self-regulation and inhibitory control), and a teen online problem-solving (TOPS) programme which has been demonstrated to be effective across a number of studies of adolescents with TBI ^{299,299,300,302} as well as an early intervention for adolescents in the form of counsellor-assisted problem-solving (CAPS, ^{298,303}. These problem-solving interventions involve sessions including EF related themes (Implementation, monitoring, and goals; Problem solving) as well as modules targeting behavioural issues (Self-management, Staying positive, Verbal and nonverbal communication, Controlling behaviour/handling crises...). Supplemental modules are used according to family needs (e.g.: Sleep, Pain, Social skills, After high school, Sibling issues). Following Ylvisaker's principles of ecological validity, the program empowers the families as well as training some components of EFs.

EXAMPLES OF EFFECTIVE INTERVENTIONS FOR DYSEXECUTIVE CHILDREN IN DEVELOPMENTAL PATHOLOGY

“Unstuck and On Target”³⁰⁴ is an EF intervention used in children with autism spectrum disorder without intellectual disability that targets flexibility, big picture thinking and planning, delivered at school in 28, 30-40 minutes sessions and integrating trainings for teachers and parents, that showed superiority to social skills training on EF and generalization to social skills ²⁶⁸

The positive effects of stimulant medication (e.g. methylphenidate) on inhibitory control are reported in children with ADHD ³⁰⁵ as well as for healthy individuals ³⁰⁶. However, evidence for efficacy of non-pharmaceutical interventions in ADHD is less robust, although there has been limited evidence supporting the use of peer and parent tutoring, strategy training and self-monitoring, for students with ADHD ³⁰⁷. There is also some preliminary evidence to support the use of executive function coaching for college students with ADHD: this approach “provides support for the development of skills, strategies, and beliefs needed to

manage executive function challenges. Coaches use specific types of questions that model reflective thinking and prompt students' ability to plan and carry out their goals" ³⁰⁸.

In younger children, Halperin et al. ³⁰⁹ have assessed the feasibility of an early intervention programme training executive, attention and motor skills (TEAMS) for preschool children with ADHD. The intervention aims to enhance inhibitory control, working memory, attention, visuospatial abilities, planning, and motor skills and initial results indicated significant improvements of ADHD symptoms.

INTERVENTIONS THAT HAVE PROVEN TO BE EFFECTIVE FOR CHILDREN'S EF DEVELOPMENT OUTSIDE BRAIN CLINICAL CONDITIONS

Contrary to ABI literature where few interventions on EF have been published and most have not proven to be effective because of methodological insufficiency, there is more abundant literature on interventions promoting EF in mainstream school children.

Special curricula used at school (e.g.: "Tools of the Mind" ³¹⁰, "Montessori" ³¹¹, "PATHS: Promoting Alternative Thinking Strategies"³¹², "Chicago School Readiness Project" ^{313,314}) provided by regular but trained and supported teachers have been reported to promote EF development in mainstream school children (see ³¹⁵ and ³¹⁶ for a review). However, a recent review of the literature suggests that although there is a strong correlation between EF performance and school success in typically developing children, there is little evidence to suggest a causal link ³¹⁷.

Physical activity has a body of well-designed literature that shows its effect on cognition ³¹⁸⁻³²¹ and especially EF. Physical activity ^{319,322,323}, cognitively challenging sports that develop strategy such as tennis ³²⁴, and traditional martial arts ³²⁵ are examples of physical activity that has shown to improve EF, independently of metabolic related changes. The value of aerobic

exercise in improving executive functions, and specifically inhibitory control, is also being increasingly reported in children with ADHD ³²³.

As concluded by Diamond “Stress, loneliness and not being physically fit impair prefrontal cortex function and EFs. The best approaches to improving EFs [...] will probably be those that (a) engage students’ passionate interests, bringing them joy and pride, (b) address stresses in students’ lives, attempting to resolve external causes and strengthen calmer, healthier responses, (c) have students vigorously exercise, and (d) give students a sense of belonging and social acceptance, in addition to giving students opportunities to repeatedly practice EFs at progressively more-advanced levels. The most effective way to improve EFs [...] is probably not to focus narrowly on these alone, but also address children’s emotional and social development and children’s physical development”³¹⁵.

CHALLENGES, CLINICAL PERSPECTIVES AND FUTURE RESEARCH

Clinicians working with children need to consider multiple factors, in particular the typical development of and/or damage to underlying skills that support executive functioning. In addition, it is likely to be important to assess and intervene in relation to core EFs (i.e. working memory, cognitive flexibility and inhibitory control) before trying to address higher-order and later maturing skills such as reasoning, planning and problem-solving. Ensuring that interventions are age appropriate in terms of typical development (i.e. not rehabilitating a skill that is not expected to develop until a later age) is crucial.

The evidence-base for interventions supporting executive functions is growing and spans a diverse range of fields including education, learning disabilities, cognitive psychology, neuroscience and developmental psychology. Interventions are recommended in many books and with varying degrees of theoretical and research underpinnings. Developing systematic approaches such as single-case experimental designs, to evaluate recommended approaches is important both for clinical governance and the development of the research field.

As described above behavioural approaches and environmental modifications have the most robust evidence to support their use in children with neurodevelopmental disorders and ABI. However, they require systematic implementation and support and training for staff, parents and carers. They can therefore be costly both in terms of time and money, and generalisation is not typically anticipated with these approaches.

From a clinical point of view working with executive function impairments in children is extremely challenging and needs to acknowledge the significant burden that these difficulties can have on families and schools. Interventions need to ensure that they don't further stress the systems that the child is trying to operate within, and programmes need to make sense and appeal to families and schools, so that they can accommodate them within their daily lives.

An important step forward would also be developing and using outcome measures that truly test progress in EF, and transfer and generalisation, e.g. that have good ecological validity and allow controlling for test-retest effect, as the whole purpose of EF tests is to prevent lapses and promote conscious, novel and effortful processing. Also rather than testing children on isolated components of EF, more open-ended and ecological tasks should be used ¹⁸⁰. Questionnaires should be used in association with performance-based testing to assess intervention impact at school and at home: the BRIEF ¹⁴⁴ is probably the most useful for EF.

CONCLUSION

Many EF intervention manuals/materials for neuropsychologists are commercialized, with few demonstrating scientifically validated effectiveness: neuropsychologists should be careful and alert parents to expensive, ineffective or unproven interventions.

There appears to be a need for interventions to be lengthy, repetitive, challenging, and fun. Therefore, implementation at school during a whole year (or more) could be the best format. Involving teachers and parents is a key component for success, possibly by the increased repetition it involves.

Given the research available so far, it seems unethical to diminish children's after-school leisure (sports, music...) to the profit of adult-type cognitive rehabilitation sessions as the former has shown positive effect on EF development and the latter is still at a research stage

in children. “Training may even lead to negative effects if the activities it displaces are more beneficial than the training itself”²⁶⁴.

Given the scarce literature on EF rehabilitation in children with ABI, more collaborations with specialists of other developmental disabilities (e.g. ADHD, autism) is needed with the aim of adapting existing effective EF interventions from these conditions to children with ABI rather than adapting adult ABI EF interventions to children. The latter carrying a high risk of being developmentally inappropriate, irrelevant or ineffective.

There is more evidence to encourage collaboration of neuropsychologists with school, training of school team and of parents in EF than to engage children in EF intervention decontextualized from the school and home environment ^{277,326}.

CHAPTER 6: GOAL MANAGEMENT TRAINING FOR REHABILITATION OF EXECUTIVE FUNCTIONS: A SYSTEMATIC REVIEW OF EFFECTIVENESS IN PATIENTS WITH ACQUIRED BRAIN INJURY.

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REVIEW

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

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Abstract

Purpose: To determine if Goal Management Training (GMT) is effective for the rehabilitation of executive functions following brain injury when administered alone or in combination with other interventions. **Method:** Systematic review, with quality appraisal specific to executive functions research and calculation of effect sizes. **Results:** Twelve studies were included. Four studies were “Proof-of-principle” studies, testing the potential effectiveness of GMT and eight were rehabilitation studies. Effectiveness was greater when GMT was combined with other interventions. The most effective interventions appeared to be those combining GMT with: Problem Solving Therapy; personal goal setting; external cueing or prompting apply GMT to the current task; personal homework to increase patients’ commitment and training intensity; ecological and daily life training activities rather than paper-and-pencil, office-type tasks. Level of support for GMT was higher for studies measuring outcome in terms of increases in participation in everyday activities rather than on measures of executive impairment. **Conclusion:** Comprehensive rehabilitation programs incorporating GMT, but integrating other approaches, are effective in executive function rehabilitation following brain injury in adults. There is insufficient evidence to support use of GMT as a stand-alone intervention.

Keywords

Brain injury, cognitive rehabilitation, cueing, dysexecutive syndrome, executive functions, frontal functions, generalization, goal neglect, memory, problem-solving

History

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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 ³²⁷. 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 ³²⁸. 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 ^{114,115,113}. Metacognition (or "thinking about your thinking") includes self-awareness, self-monitoring and self-control of cognition while performing an activity ¹¹³. 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 ¹¹³. 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 ⁹⁹. 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 (⁹⁹ for more detailed description).

Levine et al. ⁹⁸ note that GMT was initially based on Duncan's theory of "goal neglect"^{93,39,40} 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 ⁹⁹ developed this theory further and referred to "sustained and vigilant attention theory" ⁹⁴ 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" ²³¹.

GMT was one of the first EF interventions tested in a randomized controlled trial (RCT) ⁹⁸. However, the intervention consisted of a single hour of instructions, which limits the translation of its findings in terms of clinical application and effectiveness ³²⁹). GMT was then developed further, different versions were published, ranging from a single session ⁹⁸ to a 14-hour GMT program ready for group rehabilitation in clinical settings ⁹⁹. Cueing became a key feature of GMT to remind patients of their goals ^{95,97,96} and trigger GMT strategy use ³³⁰.

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 ⁹⁹, prospective memory performance ⁹⁷, multitasking ability ⁹⁵, daily life activities such as cooking ⁹⁸. GMT use has even extended to non-brain injured patients ^{331,332,333,334}. This makes it more difficult to specify the "active ingredients"³³⁵ of GMT programs and to summarize on what it is effective and for whom.

Nevertheless, GMT is manualized ²³¹ 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 rehabilitation¹¹⁴. However, there is limited evidence of superior effectiveness of GMT compared to other metacognitive training formats¹¹³. Recent literature suggest that interventions using GMT combined with other training methods are more effective than GMT-alone interventions^{336,119,118}. Furthermore it remains unclear if interventions using GMT aim at improving impairments (like the diminution of attentional lapses as suggested by GMT authors⁹⁹) or participation. Finally, not much is known about the best dose of treatment and for which patients GMT is more effective.

Kennedy¹¹³ provided a very complete review of metacognitive rehabilitation studies through 2004, including 2 early GMT papers⁹⁸⁹⁵ 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 adults³³⁴³³²³³⁷³³¹. McPherson's feasibility study³³⁸ used GMT for goal setting, without including effectiveness data on EF. Two studies of metacomponential approach^{339 340} for problem-solving in adults³⁴¹ and children²⁸⁷ 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) guidelines³⁴² but extended, similarly to the methodology of Ross’s¹¹⁰ 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 variations³³⁰. 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 scale³⁴³ (www.psycbite.com). The SCED scale is used for methodological appraisal of papers reporting interventions with single, or small N methods^{343,344}. 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.

Table 6.1: Methodological Quality appraisal of papers

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

<p><u>CONSORT questions applied to all included studies</u></p> <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)? 	<p><u>Cognitive and EF rehabilitation-specific questions applied to all included studies</u></p> <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 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)?
<p><u>CONSORT question applied only to group studies</u></p> <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)? 	<p><u>Single-case experimental design (SCED) ³⁴³ questions applied only to single case studies</u></p> <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?

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^{336,345}, it was assessed only once, conversely when a paper included two distinct studies it was assessed twice⁹⁸. Two reviewers rated the final 13 papers. Individual disagreements were resolved by discussion between the reviewers

CALCULATION OF EFFECT SIZE (ES)

ES were calculated as a standard difference between means, using Hedge’s g ³⁴⁶ adapted by Morris and Deshon³⁴⁷. Their approach has been used in important cognitive intervention reviews¹¹⁵¹¹⁰ 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 intervention⁹⁸³³⁶, 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:

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

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

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

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 rating²⁰⁸: $g < 0,20$ 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 ⁹⁸ tested GMT's "STOP-STATE your goal-SPLIT goal into subtasks-CHECK" cycle. Two proof-of-principle studies ^{95,96} tested the effect of content-free cueing on goal management and one study ⁹⁷ examined both approaches (content-free cueing triggering GMT use in a daily life context). Eight studies were rehabilitation studies: one group study ⁹⁹ and three case studies ^{98,330,348} tested GMT alone, whereas the other four studies ^{119,118,336,345,349} 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 6.2 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.

Note: due to editing problems, table 6.2 (pages 142 to 144) appears here as published in Disability & Rehabilitation Journal. Page numbers and references in brackets correspond to those of the paper, not the PhD. For correct references see below:

Levine et al. 2000 ⁹⁸—study 1

Manly et al. 2002 ⁹⁵

Fish et al. 2007 ⁹⁷

Sweeney et al. 2010 ⁹⁶

Levine et al. 2000 ⁹⁸—study 2

Pachalska et al. 2000 ³⁴⁹

Miotto et al. 2008 ¹¹⁹

Spikman et al. 2010 ¹¹⁸

Metzler-Baddeley et al. 2010 ³³⁰

Levine et al. 2011 ⁹⁹

Novakovic-Agopian et al. 2011 ³³⁶

Chen et al. 2011 ³⁴⁵

Table 2. Description, methodological ratings and main findings of included studies (by type and by chronological order of publication).

Author Study type (group/single case) Quality rating	GMT alone or GMT combined with other rehabilitation methods Method	Sample	Description of experimental intervention and of control intervention	Outcome measures	Findings	ESs according to Cohen's rating	Maintenance of effects
Proof-of-principle studies Levine et al. [7] – study 1 Group study 56%, moderate	GMT alone RCT	30 patients with mild to severe TBI 3–4 years post TBI	1 h of individual GMT present- ing goal management pro- cesses, 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 train- ing (MST)	Paper-and-pencil tasks called "everyday tasks", including proofreading, and room layout grid	GMT participants made less errors in proof- reading and grouping tasks but took more time for task comple- tion than MST group	$g = 0.9$ for proofreading (<i>large</i>) $g = 0.95$ for grouping (<i>large</i>)	Not evaluated
Manly et al. [14] Group study 80%, high	GMT alone + cueing Comparison of perfor- mance in two conditions (with and without auditory alerts)	10 patients: one suspected ischemic stroke and nine TBI (seven 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 six 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. [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 con- tent-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 accu- racy (taking uncued days as controls) (<i>medium</i>)	–
Sweeney et al. [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. [7] –study 2 Single case 52%, moderate	GMT alone Single-subject	Female, 35 years, 5 months post encephalitis	Two sessions of GMT similar to Levine study 1 + 3 sessions of GMT applied to cooking task	Observation of problem- atic behaviour during meal preparation Self-report cooking errors diary, paper-and-pencil tasks including proof- reading, room layout grid.	Less problematic behav- iour during meal prep- aration observation Less self-reported diffi- culties in meal prepar- ation Less errors on the "everyday tasks"	–	Maintenance of effects on cooking at 6 month
Pachalska et al. [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 pro- gram "MARSZ". The pro- gram was applied to activities of increasing diffi- culty, 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 func- tions" "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

(continued)

Table 2. Continued

Author Study type (group/single case) Quality rating	GMT alone or GMT combined with other rehabilitation methods Method	Sample	Description of experimental intervention and of control intervention	Outcome measures	Findings	ESs according to Cohen's rating	Maintenance of effects
Miotto et al. [24] Group study 76%, high	GMT combined Pseudo-randomized, con- trolled trial with cross- over	Patients with right (<i>n</i> = 14) or left (<i>n</i> = 16) frontal lobe lesions (23 tumors and seven mild TBI)	<ul style="list-style-type: none"> Group 1: Attention and Problem Solving based on GMT and PST; 10 weekly 1.5 h group sessions Group 2: Information and education through a booklet to read that suggested cognitive exercises and presenting problem-solving frame-work Group 3: usual care 	Extensive executive func- tions paper-and-pencil assessment Modified Multiple Errands Task (MMET) Virtual Planning test DEX questionnaire	Intervention effect on MMET and carers DEX when group 1 compared to groups 2 and 3. Some improvement on paper-and-pencil executive tests after APS	$g = 1.08$ for MMET (<i>large</i>) $g = 2.14$ for carers DEX (<i>large</i>)	Effect maintained at 6 month follow up
Schweitzer et al. [41] Case study 56%, quality rating not applicable*	GMT alone Single-subject case report	Male, 41 years, cerebel- lum hemorrhage 4.5 months post injury	<ul style="list-style-type: none"> Seven weekly 2 h 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	–	Effects maintained at 4 month follow-up
Spikman et al. [25] Group study 92% high	GMT combined Multicenter single-blinded RCT	75 patients with ABI (mainly stroke and TBI, unknown sever- ity), living at home, >3 months post injury	<ul style="list-style-type: none"> 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) 	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 Telephone Directory and Map search from TEA	Both groups resumed their previous roles signifi- cantly more than pre- treatment (RRL) but experimental group to a larger and statisti- cally significant extent Statistically more improvement in experimental group on EOS Both groups showed less complaints on DEX; better quality of life (QOLIBRI) and pro- gress on personal goals (TGA) Significant group effect when three measures of daily EF combined (EST, RRL and TGA) Statistically improved errors and speed on telephone directory task. The patient reported sub- jective improvement in her ability to complete	$g = 0.49$ 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 wor- sened on RRL com- pared to post intervention
Metzler-Baddeley et al. [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.	<ul style="list-style-type: none"> 3 × 1.5 h 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		–	No follow-up

Levine et al. [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 ver- sion presented on PowerPoint Slides - Control group: "Brain health workshop" (BHW): educa- tion on BL, matched for time Both intervention were adminis- tered 7 x 2 h, in group sessions + between-sessions homework	SART D-KEFS Tower Test Hotel Task DEX Questionnaire CFQ	tasks and deal with distractions at work. 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 alloca- tion in the GMT group on Hotel Task imme- diately post interven- tion	$g = 0.43$ 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 viola- tion on Tower test Hotel Task time allocation did not differ from controls at follow-up.
Novakovic-Agopian et al. [23] Chen et al. [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 h of small group train- ing + 3 x 1 h individual training + 20 h of home practice for 5 weeks. Focus on practice in daily life and self-generated complex goals (individual and group projects). - Control group: 2 h of education about brain injury	Neuropsychological tests:- EF and attention avei- aged to an overall "Attention and Executive Function domain" - Memory and learning - Processing speed Multiple Errands Tasks + fMRI on a selective attention task	No effect on questionnaires 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 modula- tion of neural process- ing 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 educa- tion control interven- tion (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 Questionnaire, DEX = Dysexecutive Questionnaire, QOLIBRI = Quality of Life after Brain Injury, TGA = Treatment Goal Attainment.

HOW ARE GMT INTERVENTIONS ADMINISTERED?

Interventions testing GMT component principles were either as short as a single sentence before starting the test ^{95,96} (“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 hour⁹⁸ or 30 min⁹⁷.

Interventions testing rehabilitation ranged from 5 hours ⁹⁸ to 43 hours training ³³⁶ and were administered between once ^{99,119} and two-three times a week ^{336,118}. Most studies were administered as individual sessions ^{98,97,330}. Apart from three studies ^{99,119,348}, all interventions included some individual sessions ^{345,336} or some adjustment of the program to the personal needs of each patient ¹¹⁸.

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 ^{118,99,336,330}. Homework included monitoring EF failures in daily life called “Slips”⁹⁹, identifying factors that helped or not in everyday goal management performance ^{118,98}, listing occasions on which participants used GMT strategies during the week ⁹⁹, mindfulness practice assisted by an audio CD ^{345,336}.

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 ⁹⁸. 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 ^{95,96}, auditory alerts during a complex multi tasking activity reminded patients about their current goal. In Manly et al.’s Hotel task ⁹⁵ 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”⁹⁶ 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.⁹⁷ combined content-free cueing with a 30-minute 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.⁹⁵, but not in Sweeney et al.⁹⁶. Fish et al.⁹⁷, 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.⁹⁹ 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)³⁵⁰. 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 ^{98,348} 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 ^{351,352}. 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 ^{119,353,336,345} 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 ^{266,354,355,356}. In the articles reviewed, cooking activities were used by Levine ⁹⁸ and Pachalska ³⁴⁹ as cooking is a complex, open-ended, ill-structured multi-tasking activity requiring EF performance ^{247,357,358}. 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 ^{336,345}. GMT principles ^{98,95} were combined ecologically and applied with success in a rehabilitation

case-study by Metzler- Baddley ³³⁰: 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 ³⁴⁹, 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 ³⁵⁹. 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 ^{118,98} (planning a meal, learning to use an organizer) and group projects ^{345,336}. In Spikman’s study ¹¹⁸, 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 ³³⁸ 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”³³⁸, 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 ¹¹⁹ and Spikman ¹¹⁸ 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 ¹¹⁸ or pseudo-randomized controlled trials ^{119,345,336}.

Miotto et al. investigated an Attention and Problem Solving (APS) Program ¹¹⁹, 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” ¹¹⁸ 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 ²⁶⁶ and Sirigu’s scripts ³⁶⁰. 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 ³⁶¹.

“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 ³³⁶ 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 ³⁴⁵ 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 ¹¹⁹ and Spikman’s ¹¹⁸ 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 ¹¹³ regarding metacognitive strategy training. Conversely, in four articles ^{336,345,348,99} 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 ⁹⁹ and Multiple Errands Task ^{345,336}) and the effects on measures of participation were either not detected ⁹⁹ or not assessed objectively ^{345,336}. However, patients generally reported using GMT strategies more than before the intervention ³³⁶.

DO GMT EFFECTS GENERALIZE TO UNTRAINED TASKS AND DOES GMT REHABILITATION IMPROVE PARTICIPATION?

Novakovic-Agopian et al. ³³⁶ 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 ^{119,348} and patients ¹¹⁸ reported improvement on questionnaires of EF, on interview of role resumption ¹¹⁸ or reported subjective improvement in daily life ^{98,348,330} in most studies (except^{99,345,336}). Improvement on questionnaires^{119,118,348} 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 ³⁶². This was addressed in the studies of Miotto ¹¹⁹ and Spikman ¹¹⁸ 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 ¹¹⁹, the Executive Secretarial Task ¹¹⁸ or test that are supposed to be close to daily life complex tasks, like the Hotel Task ⁹⁵ and the Everyday tasks ⁹⁸³³⁰ 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 ³³⁶³⁴⁵. There is some, albeit limited, evidence that more severely impaired patients would benefit less ⁹⁶.

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 ^{119,118}. 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 ^{247,210}.

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^{98,95,96} 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^{98,348,330}. Levine's RCT⁹⁹ 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 alerts⁹⁵ study but not confirmed in Levine's⁹⁹ that used the same outcome measure nor in Sweeney's study⁹⁶ that used a novel virtual task. In the Fish et al study⁹⁷, 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^{166,165,93,167,22}. 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^{363,364} and have a higher veridicality than classical neuropsychological EF assessments. However, their test-retest reliability tends to be low^{365,366} 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 ^{367,22}. As the whole purpose of EF tests is to prevent lapses into automaticity and promote conscious, novel and effortful processing ^{165,22}, apparent progress after EF interventions may not necessarily reflect changes in underlying executive processes. Even parallel forms of the same test ³⁶⁶ may not overcome the novelty problem. The content may be new but the format is not ³⁶⁷.

With the exception of one study ¹¹⁸, 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 ²². Spikman et al ¹¹⁸ 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. ⁹⁹, 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 ³⁵⁰, it has never been demonstrated that improvement on the SART is correlated with improvement in everyday functioning. Indeed, Levine et al. ⁹⁹ 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 ³⁶⁸ ³⁶⁹ 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 ³⁶²).

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 ^{99,348,119}, Cognitive Failures Questionnaire ^{99,348} or Role Resumption List ¹¹⁸. 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 ¹¹⁸ 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 ³⁴⁵ 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 ¹⁸⁰ 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 situation³³⁰, using GMT for brain injured children.

The Attention & Problem Solving program¹¹⁹ and the “Multifaceted Treatment of executive dysfunction”¹¹⁸ 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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EXPERIMENTAL DATA

CHAPTER 7: CONTEXT-SENSITIVE GOAL MANAGEMENT TRAINING FOR EVERYDAY EXECUTIVE DYSFUNCTION IN CHILDREN AFTER SEVERE TBI.

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Context-Sensitive Goal Management Training for Everyday Executive Dysfunction in Children After Severe Traumatic Brain Injury

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Objectives: To assess the effectiveness of a metacognitive training intervention, based on an adapted Goal Management Training and Ylvisaker's principles, on 3 activity domains of executive functions: (1) prospective memory (PM) performance in ecological setting, (2) complex cooking task management, and (3) daily executive functioning (EF) at home and at school. **Participants:** Five children aged 8 to 14 years, who were 3 to 11 years post-severe traumatic brain injury, experiencing severe EF difficulties in daily life. **Design:** Single-case experimental design and assessment of EF twice prior to intervention, postintervention, and 3 and 6 months postintervention. Progress was monitored by a weekly ecological PM score. The effect on EF was assessed using the Children's Cooking Task. Transfer to the child's natural context was assessed by parental and teacher questionnaires and Goal Attainment Scaling. **Results:** All children improved both on the measure of PM and on questionnaires of daily EF. Two children improved on the Children's Cooking Task but returned to their preintervention level in a novel cooking task at follow-up. Participation of school personnel and parents in the program was low. **Conclusions:** It is feasible but challenging to use Goal Management Training in children with traumatic brain injury. Further research is needed in relation to how to promote generalization and how to increase the involvement of the child's "everyday people" in the intervention. **Key words:** *child, daily life activities, ecological, executive functions, Goal Management Training, intervention, prospective memory, traumatic/acquired brain injury*

Introduction

Executive functions (EF) are a collection of related but distinct abilities that allow individuals to engage efficiently in intentional, goal-directed problem-solving actions^{20,21} through conscious and effortful processing²² and to adapt to new situations in the real world²³. Executive functioning (EF) deficits are a frequent consequence of traumatic brain injury (TBI)⁶. TBI outcome is predicted by executive functioning level¹²².

There is a lack of validated methods for EF rehabilitation in children^{110,112,111}, although some general rehabilitation principles have proven to be useful. Ylvisaker²⁶⁶ emphasized two principles: (1) the key role of parents and of all the ‘everyday people’ surrounding the child in the “cognitive coaching” of their child (See Braga²⁶⁷ and Wade^{299,300} for examples of family-delivered interventions in TBI); (2) the necessity of a “context-sensitive” approach²⁷⁷, embedded in functional routines of everyday life and using meaningful activities rather than decontextualized exercises.

In current clinical practice, four types of approaches are used to help children with their EF difficulties, though each has its limitations: (1) Providing environmental support and compensatory aids (e.g. use of electronic prompting devices) – though only a restricted number of situations lend themselves to such an approach; (2) Training component EF skills (e.g. repeated exercises aiming at changing the brain’s working memory capacity for example) – though transfer to natural contexts and generalization to untrained activities effects of this approach have rarely been demonstrated²⁷⁷; (3) Training children on specific goals (e.g. if preparing a schoolbag is problematic, the child will be trained on this specific activity until the goal is achieved) – though generalization to similar goals in different situations (e.g. preparing a suitcase for holidays) is often not achieved; (4) Providing children with metacognitive strategies applicable to a variety of everyday situations—whilst this is effective in some adults after TBI¹¹⁴, there is little evidence that metacognitive training is effective for children with a dysexecutive syndrome post TBI¹⁶, though research on children with other forms of brain injury suggests it may be a useful approach^{108 287}.

Goal Management Training (GMT)⁹⁸ is one of the most studied metacognitive training programs, of which many variants exist²²⁹. GMT includes self-instruction strategies, self-

monitoring exercises, metacognitive strategies aimed at improving planning, prospective memory and hierarchical goal management, mindfulness practice exercises, stories promoting discussion about executive dysfunction in daily life, and homework assignments (See Levine et al.⁹⁹ for a more detailed description). GMT was developed from Duncan's theory of "goal neglect"^{39,40}, which suggests that dysexecutive patients are impaired in the construction and use of "goal lists", necessary for goal-directed behavior. They do remember the intended goal but tend to lose sight of it as they progress through a task leading to a prospective memory failure. Prospective memory (PM) (remembering to carry out intended actions) tasks require retrospective memory to remember the task, but depend on EF⁶⁵ for successful goal maintenance, retrieval and implementation at the right moment. PM depends upon frontal lobe integrity¹⁸⁵, with a key role for rostral prefrontal cortex (BA10)¹⁸⁶. In typically-developing children aged 6-12, performance on EF tasks such as planning and switching⁴⁷, working memory⁴⁸, and inhibition⁴⁹ is correlated with PM⁵⁰. PM problems are reported as a major concern for the parents of children with TBI⁸⁷. PM is impaired in children with TBI^{85,86} compared to children with orthopedic injuries, even after cues are given¹⁸⁸, and even under strong incentive conditions¹⁸⁹.

The primary aim of this study was to assess the feasibility and effectiveness of implementing a metacognitive training intervention, based on an adapted form of Goal Management Training (GMT) and on Ylvisaker's rehabilitation principles, in three domains: (1) prospective memory performance, (2) complex cooking task management (3) daily executive functioning at home and at school. Secondary aims were to determine if the effects of such a metacognitive training generalize enough to help children to (1) achieve personalized, untrained, goals and (2) manage a demanding novel task that requires EF's.

METHODS

PARTICIPANTS

The study was approved by the ethics committee of Pitié-Salpêtrière University Hospital in Paris, France. Informed parental written consent and participation assent were obtained for all participants prior to initiating any procedure.

Inclusion criteria were: (1) severe TBI (initial Glasgow Coma Score lower than 9); (2) sustained at least 2 years previously; (3) children attending one of the two participating rehabilitation departments; (4) aged 8-14; (5) evidence of a dysexecutive syndrome on neuropsychological assessment performed at least two years post injury; (6) parental report of executive functioning difficulties in daily life.

Exclusion criteria were: (1) diagnosed learning disabilities, neurologic or psychiatric condition prior to TBI; (2) severe intellectual disability; (3) insufficient French language level of the child or of his/her family.

Characterization data included classical standardized tests from the Wechsler Intelligence Scale for Children (WISC-IV)²³⁴, from the Children's Memory Scale¹⁵⁰ and a French battery of EF for children: "Fonctions Exécutives Enfant" battery (FEE)²³⁶. FEE has a much larger normative data than any other EF test available in French. Each child participating in the intervention was compared to a sample of controls matched for sex, age and socioeconomic status from the FEE database, using Crawford's method³⁷⁰ with one-tailed probability³⁷¹ taking $p < 0.05$ criteria for significance.

INTERVENTION

The intervention was inspired by Goal Management Training⁹⁸ (GMT) but extended to follow Ylvisaker's principles of involving "everyday people" in the child's social network in the cognitive coaching of the child and using a context-sensitive approach prioritizing functional ecological activities.

Prior to the intervention, parents participated in a two to three hour informal interview during which the following were discussed: 1) the child's daily life difficulties at home and at school; 2) program content and key GMT concepts; 3) the need to apply the metacognitive GMT techniques at home and at school. An emphasis was put on the key role parents played for the success of the intervention.

The intervention comprised of: (1) GMT theoretical modules and between session "missions" (promoting GMT use at home and at school); (2) practical modules in which children practiced GMT content in meaningful activities; (3) an "everyday people cognitive coaching guide". The materials included PowerPoint slides, a workbook, posters for home and school

use, mission sheets and the cognitive coaching guide. The 15 modules (theoretical modules+ meaningful activities) of the intervention required 15-20 hours to be completed, and were administered individually over a four to six-month period on a weekly basis, either in a rehabilitation centre or at home.

(1) Theoretical modules were derived from the adult GMT PowerPoint Manual⁹⁹ developed by Levine, Robertson and Manly, that has already been used (with minor changes) in children³⁷². A new, shorter, colourfull version was created for the intervention to make materials child-friendly, age-appropriate, enjoyable and simpler. For example, “slips” (referring to slips of attention) became “Oops errors”. “To Do lists” and the “mental blackboard” were combined in a unique “note book” concept in order to explain to the children how a real paper note book can help them not to overwhelm their “mental notebook”. Discussion about EF failures in daily life was triggered through illustrated stories relating to school and leisure activities. In each module we included a prospective memory task to be performed during the session (e.g. when you see a slide with X you do Y) to encourage discussion about PM failures at the end of each session. The content-free cue “Look into your mental notebook” was used as a prompt when children failed PM tasks. Throughout the training period children had to complete “mission sheets”, inspired from GMT between-session assignments. These were of three types, introduced progressively: 1) monitoring Oops errors, their consequences and factors influencing their occurrence; 2) listing occasions on which the child used a metacognitive strategy of his/her own or from the program with success; 3) identifying situations where a stepwise processing approach can be used in order to manage a goal (preparing the school bag, preparing a sandwich).

(2) Practical modules served to practice GMT content in meaningful activities (Ylvisaker’s *Content-sensitive* principle): cooking of various meals, route finding, searching for information, poster making, photo ordering. This was an explicit generalization training that was aimed to show the children that GMT metacognitive strategies are applicable to many situation in life where their EF impairment may impede success, and to promote the use of these strategies to personal (present and future) goals and untrained activities³⁵⁵. The activities involved planning, strategy generation, following steps, and monitoring of actions. Difficulty increased as children progressed in the program. Similarly to the theoretical modules, the meaningful activities contained naturally occurring prospective memory tasks to allow

discussion about PM difficulties in real activities (e.g. checking regularly if the first set of finger biscuits is cooked while preparing the next). The trainer guided the children when needed, using non-specific prompts and general cues. Explicit help was only given if these were not sufficient. At the end of each activity, the child was invited to review his/her performance using the “mission” sheets, identify Oops errors and any effective strategy that had helped in the task, and to think about situations in real life where the same kind of strategy may be useful. The child always took the ‘product’ of his/her activity (e.g. crepes) home to increase motivation from the praise s/he received at home. Moreover, the ‘product’ was expected to remind the parents about the child’s program.

(3) To encourage transfer to the child’s natural contexts, we tried to involve the child’s ‘everyday people’ as cognitive coaches for their child. Everyday people were parents, teachers, school assistants and any adult the parents identified as a potential cognitive coach (baby-sitter, student helping the child with homework). A letter presenting the program was sent to the child’s teacher and school assistant (SA), explaining briefly TBI executive problems and their implications, and asking the teacher and the SA to participate by applying GMT at school. The intervention content was not explained orally. We asked for a contact e-mail and a telephone contact to discuss the child’s difficulties and set realistic goals on goal attainment scales. The letter was sent a second time after one month as the first yielded few responses, so we had responses from at least one school staff member per child. Other potential ‘everyday people’ identified by the parents were sent a similar letter. Twice a month all ‘everyday people’ who agreed to participate received one chapter, two-pages long, of a “Cognitive Coaching Guide” that was created for the intervention. The guide was colorful, using the same drawings, diagrams and analogies as the theoretical modules, explaining the rehabilitation content and suggesting how to apply metacognitive strategies at school and at home. The intervention was organized in a way that each metacognitive strategy was (1) first introduced during a theoretical module, (2) then practiced on a meaningful activity and (3) lastly introduced to everyday people. They were sent the corresponding chapter of the coaching guide that described the activity for which the child had already practiced the strategy, and suggested other activities to which it could be applied (see supplemental digital data for examples). Through this guide, everyday people were encouraged: (1) to use non-specific prompts for PM failures (“look into your mental note book”) rather than specific instructions (“you need to feed the dog”) or negative sentences (“you’ve forgotten to feed

your dog again!"); (2) to promote strategy generation instead of giving the solution to their child, consistent with Ylvisaker’s aim of “helping the child to become a strategic thinker”²⁶⁶; (3) to prompt and help the child to fill in his/her “mission sheets” regularly; (4) to practice goal identification (“state your goal”) and stepwise processing in daily activities (preparing schoolbag, table setting). Parents were explicitly asked to go through the metacognitive strategies of the cognitive coaching guide and to sign the child’s GMT workbook every week. The ‘everyday people’ were not requested to participate in the rehabilitation session, but were advised that the therapist was available if they had any questions.

Examples of the type of exercises used may be found in supplemental data 7.1.

Supplemental data 7.1: Examples of how metacognitive strategies were applied and used in theoretical modules, meaningful activities and suggested activities for everyday people.

Metacognitive strategy	Theoretical module exercise	Meaningful activity	Cognitive coaching guide (proposal of situations the strategy can be applied to):
“Stop and State your goal”	Multi-element tasks with changing aims (do some of each sub task OR earn as many points as possible)	Cooking biscuits from a recipe containing prompts (cartoon character showing a stop sign) to stop and state what the goal of the step is before proceeding	State your goal before beginning a school exercise. Use the cartoon character whenever the child starts an exercise without understanding the aim instead of telling him/her s/he read it wrong.
“Write the steps”	Imagining you prepare a sandwich Mathematical problem Scripts about organizing a birthday party	Preparing a cake from an unordered recipe, presented without steps and without details about cooking procedure	School essay preparation Organizing homework for the week Planning a party/ an outing with the family

QUALITATIVE DATA ABOUT THE PROGRAM

Throughout the sessions, the therapist recorded (1) how the child reacted to the intervention content; (2) if the metacognitive strategies were easily understood and used; (3) if the child

seemed aware of his/her EF difficulties during discussion and performance of meaningful activities; (4) if the “mission sheets” were filled-in between the sessions.

STUDY DESIGN

Intervention effectiveness was assessed in two ways: (1) A Single-Case Experimental design (SCED)^{344,373}, with repeated ecological measurement of prospective memory was used to monitor progress throughout the intervention; (2) pre-post measurement of EF with two baseline assessments 4-8 weeks apart (B1 and B2), and three post-intervention assessments: immediately after the intervention (R1), and at three (R2) and six months (R3) to assess maintenance of effects. Baseline was expected to be stable as children had sustained their TBI at least two years earlier, minimizing chances of spontaneous recovery during the study. The pre-post measurement served to capture the key issues of EF rehabilitation, namely: (1) EF performance in ecological tasks; (2) transfer of training effects to natural contexts; (3) generalization to untrained tasks; (4) need for novelty for a task to truly test “executive” functions.

OUTCOME MEASURES

Repeated measures of prospective memory (PM) performance - SCED design: We monitored the effectiveness of the program through a weekly score on a time-based PM task. The PM task was inspired by the Fish et al. phone call task⁹⁷ that has recently been used in children³⁷⁴. Three times a week, children had to look up the day’s Saint on a calendar (e.g. 24th June is Saint John’s day), and send it to the therapist at an agreed target time, either as a text message, an e-mail or a phonecall. The child was awarded three points if correct information was given within one hour of the agreed time, two points if within the day, one point if on a different day and zero points if the child completely forgot about the task. The retrospective memory component was controlled for by checking at each session that children remembered the task and agreed times. Parents were given the timings in case the child wanted to check the target time. To encourage use of mental strategies, children were asked not to use cues such as alarms, or pre-programmed text messages. Parents were instructed not to give any cues or help to complete the task. The three target days and times were chosen individually for each child with the parents before the first assessment to ensure: (1) that the child was easily available for the task (i.e. not during school time or leisure activity); (2) that it didn’t

disturb family routines (e.g. bedtime); (3) that the time didn't correspond to a regular activity that could act as a cue (e.g. TV show); (4) that timings respected the following common rules for all the children: non-consecutive days, one week-end day and two school days, except the day of the intervention and different target time on each of the three days. As in Fish et al⁹⁷, days when the tasks had not been performed for another reason than PM failure, were not used in the analysis (medical appointment at target time, no internet connection during a week-end outing); therefore the total score was expressed as a percentage of total possible points that week.

“Children’s Cooking Task” (CCT) – pre-post ecological measurement of EF: As the aim of this study was to improve EF in daily life, assessment included an ecological¹⁸⁰ test of EF, called the Children’s Cooking Task (CCT)^{120,121}. In the CCT, children have to prepare a real chocolate cake and a fruit cocktail following a structured, photo-cued, child-friendly recipe contained in a cookbook including distractors. The task has been shown to be highly sensitive to executive dysfunction in TBI as it is novel, challenging, open-ended, and requires multiple goal management and innovative higher level strategies to succeed. The Children’s Cooking Task (CCT) has good inter-rater and test–retest reliability, high internal consistency, as well as good discriminant and concurrent validity¹²⁰. It can be performed from the age of 8. Scoring is based on the number of errors, including omissions, additions, commentaries, substitution and estimation errors. Normative data is not yet available. Therefore raw scores were used to track individual child changes. The number of errors in CCT made by each child was compared to the number of errors made by age matched healthy controls, extracted from unpublished data³⁷⁵.

Questionnaires – pre-post measurement of EF in natural contexts: Ultimately, the aim of any cognitive rehabilitation intervention is to allow a transfer of learned skills to the natural context of the child²⁶⁶, and this was particularly important to assess because metacognitive training aims at providing children with strategies they can apply to many tasks in their natural context. This was assessed through two questionnaires of EF: (1) the Behavior Rating Inventory of Executive Functions (BRIEF) questionnaire^{144, 145}, completed by both parents (transfer to home-context) and teachers (transfer to school-context); (2) a cognition subscore derived from the Dysexecutive Questionnaire for Children (DEX-C)²³⁵ that was completed by parents only. The BRIEF assesses eight domains of executive functioning in the real world,

which give together a Global Executive Composite (GEC) score. Higher scores correspond to increased EF difficulties in daily life. A T-score superior to 65 is defined as the clinical range. The BRIEF has large normative data for children aged 5 to 18, high internal consistency¹⁴⁶, good validity¹⁵¹ and good test-retest reliability¹⁴⁶ although parent-teacher agreement is only moderate¹⁴⁶. It is the most commonly used questionnaire of executive functions and seems to be sensitive to deficits in executive functioning in children with TBI^{151,153,71,14}. Its relationship with common EF cognitive tests is however inconsistent. DEX-C is a 20 item questionnaire, probing four broad areas of EF difficulties (emotional/personality, motivational, behavioural and cognitive) and is part of the “Behavioural Assessment of the Dysexecutive Syndrome for Children”²³⁵. Higher Z-scores correspond to increased cognitive difficulties relating to EF. DEX-C has less evidence regarding psychometric properties²³⁵. However the cognitive subscale of DEX-C completed by parents has a high correlation with the (Children’s Cooking Task) CCT score¹²⁰, and therefore this subscale together with the CCT were expected to detect improvement in cognitive EF impairment, which the BRIEF might not capture.

Goal Attainment Scaling (GAS) – pre-post measurement of generalization (metacognitive strategy use in untrained tasks): GAS^{376,377} was used as a generalization measure to assess if a child who has applied metacognitive strategies to meaningful activities in a rehabilitation setting is capable of applying those strategies to untrained tasks that are judged to be problematic for him/her by his/her everyday people. EF-related problems reported by the child, parents and school staff served to elaborate personalized goal attainment scales (GAS) for each child. These GAS goals were *not* trained specifically but children were repeatedly encouraged to apply metacognitive strategies to daily life. We also used “general” GAS for metacognitive strategy use and GMT application (see table 7.3 in results section). Themes of “general” goals were similar for all children, but the initial levels and expected outcome levels were specific to each child, taking into account children’s age and possibilities. The detailed procedure for goal selection, GAS elaboration and GAS levels adjustment are described in supplemental data 7.2. GAS scores were used to calculate a global T-score for each child, using Kiresuk’s formulae (see^{376,377} for details of GAS methodology). A T-score of 50 meant that the goals were overall attained as expected, and > 50 that goals were attained better than expected.

Supplemental Data 7.2: Writing procedure for GAS

Writing procedure for general GAS corresponding to metacognitive strategy use and GMT application

Eleven general GAS items for metacognitive strategy use and GMT application to daily life were created. These were sent to parents, teachers, school assistants and any other “everyday people”. For each item, everyday people had to circle the level corresponding best to the child’s actual performance relating to the goal, ranging from -2 (worse performance they could imagine) to +2 (best expected performance), 0 corresponding to the most likely performance expected after intervention.

E.g.: « XX (child name) writes on his own initiative things he/she might forget to do in a single and well-identified note-book or planner (excluding homework he/she is explicitly asked to write down by the teacher)»

-2 : never

-1 : sometimes but it is not regular or functional enough to rely on it

0 : Writes down important things to be done but you need to emphasize/repeat it is an important thing to remember to do

+1 : The important things are written without you telling the child it is important to remember

+2 : Writes all the things and without you telling the child to remember to do so

The answers to this first GAS questionnaire allowed readjustment of the scales, through the following rules: (1) Scales scoring +1 or +2 pre-intervention were removed as the performance for the item was satisfactory without intervention and the item was therefore not a goal to attain. (2) Scales that scored 0 were reformulated in order to have the pre-intervention level corresponding to -1 by fixing a more challenging 0, +1 and +2 scores (this was particularly the case for older children). (3) Scales scoring -2 or -1 were not reformulated, in order to capture a possible worsening in performance for those having -1 as the preintervention level. Therefore the initially identical scales were readjusted according to the child’s present performance relating to each metacognitive strategy use goal.

In order to fulfill the unidimensionality criteria, some goals relating to strategy use were split into two goals (e.g.: effective use of a paper note-book to compensate for PM failures was split into (1) writing down the things one might forget to do; (2) remember to look into the note book to perform the intended action). These goals were considered important goals by the researchers but *did not* take into account the personal goals of the child and his everyday people.

Writing procedure for personal GAS corresponding to EF-related problems reported by everyday people.

Two to seven additional personal GAS per child were created based on parents’, teacher’s and school assistant’s concerns. Goals were selected after analyzing the BRIEF and DEX-C questionnaires and after a one- to two-hour interview with parents and a phone interview with the teacher and/or school assistant. The first author created the GAS scales in a written form and sent them to the person the goal had been proposed by. The first answers to these personal GAS allowed, if needed, to reformulate the levels according to the same procedure as for the general GAS described above. .

All the scales, including those reformulated were sent again to the everyday people in order to check that the initial level had been worded correctly and corresponded to -2 or -1 prior to intervention. This personalized second GAS set of answers was used to calculate a pre-intervention T-score and to measure outcome at R1, R2 and R3. Because the procedure required writing, reformulating and double checking for initial level, only one baseline could be calculated for GAS.

“Christmas biscuits task” – measurement of adaptation to novelty, performed once only at the end of the intervention: EF outcome measures need to be novel to really capture EF^{165,166}. When the same task is repeated after intervention, it is more “familiar”, which can make it less demanding on EF^{367,118}. Familiarity effects increase when patients are tested on several occasions (as it is the case in our design for CCT). To get a “purer” EF measure post-intervention¹¹⁸, we developed a parallel form of the Children’s Cooking Task (CCT) for assessment at R3, involving the same number of steps and ingredients but requiring different types of ingredients and procedures. This version has no established psychometric properties. Children had to bake “Christmas biscuits”. Whilst both tasks required cooking, as children were not experienced cooks, a new recipe could not be viewed as a familiar task.

CONTROLLING FOR CONFOUNDING FACTORS.

At the beginning of the program everyday people were not informed of exactly when the intervention component would commence: from the first interview onwards all children were seen weekly, whereas the intervention started only 5-8 weeks later. We hoped to control in this way for parent’s enthusiasm for a novel rehabilitation program, which was expected to be reflected by an improvement between B1 and B2 in this study design. The intervention effect was measured comparing post intervention results (R1, R2 and R3) to the *second* baseline (B2), as this was considered a “purer” baseline eliminating the enthusiasm and novelty effect. Inconsistent answers to the BRIEF questionnaire were detected by computing the inconsistency score described in the BRIEF manual (a score >9 being a threshold to consider the questionnaire unreliable because of contradictory answers on special items serving to assess consistency of answers). Furthermore, the intervention did not significantly change the amount of time spent in rehabilitation: all children had already been attending the outpatient department for half a day a week for many years (including sports, group games and group discussion to promote socialization and language pragmatics for PB,CS,RK; analytic psychotherapy for PB, paper-and pencil neuropsychological exercises aiming at improving attention for IP) therefore the effect of the potentially confounding factor of time spent with therapist was considered likely to be negligible.

EXTERNAL INVESTIGATOR POST-INTERVENTION INTERVIEW

After the intervention, an external interviewer, who had neither been involved in the rehabilitation nor the research team called all the everyday people involved in the program. A structured interview focused on how they perceived the program, their views on applying cognitive coaching at home and at school, clarity of the cognitive coaching guide, how children reacted to the intervention and if they thought their child had improved in various domains (autonomy, school results...) even if it was a domain not included in GAS and questionnaires. The interview contained embedded questions aimed at quantifying how much the everyday people participated in the cognitive coaching and at checking if they understood the concepts that were explained to them in the cognitive coaching guide. They were asked for examples of metacognitive strategies they could recall, situations they applied them to, and were asked how often they managed to go through the child's workbook together, and if GMT posters had been hung at home. Feedback from the child was obtained informally from the first author conducting the intervention, because answering to an unknown external investigator on the phone was considered age-inappropriate.

STATISTICAL ANALYSIS AND EFFECT SIZE (ES) CALCULATION

The Saint's day task (SDT) PM scores were visually analyzed on time series graphs comparing baseline (weeks 1-4), with intervention (weeks 5-18). A two-standard deviation band (2SDB) was determined for each child based on the standard deviation of the four baseline points. Gottman and Leiblum's criterion was used: the probability that two consecutive points fall outside the 2SDB is < 0.05 . (see for details ³⁴⁴). Trends were detected by celeration lines, using the Split Middle Trend line procedure³⁴⁴. To obtain the magnitude of effect, the "nonoverlap of all pairs" (NAP) method was used ³⁷⁸, through SPSS software. For the other outcome measures, an intervention effect size (ES) was calculated for each child from B2 to R1, R2 and R3 as a standard difference between T-scores divided by 10 for the BRIEF and as a standard difference between Z-scores for the DEX-C Cognition scale. For the CCT, ES was obtained by dividing the score difference by the standard deviation of all five CCT scores of the child. Furthermore, because the CCT inevitably has a practice effect as the recipe becomes more known, which could account for improvement throughout the trials, we readjusted ES by subtracting the practice effect of each child (score change between B1 and

B2 being considered as the practice effect for that child). ES were interpreted subjectively with reference to Cohen's guidelines²⁰⁸: 0.2 = small; 0.5 = medium; 0.8 = large.

RESULTS

Five children, aged 8 to 13 years met the inclusion criteria (PB, CS, RK, IP, YR). All had sustained severe TBI at an early age, three to eleven years before the study and had a highly complex family situation. All had specialized schooling, either attending a special support class or having a school assistant. Characteristics of the participants are summarized in table 7.1. All children had a severe dysexecutive syndrome on paper-and-pencil EF tests (see table in Supplemental Data 7.3), on CCT (see B1 and B2 scores in Figure 7.2), and (apart from child IP' BRIEF score) on EF-questionnaires (see B1 and B2 in Figures 3 and 4). In contrast they were not impaired in reasoning abilities (apart from CS) or retrospective memory (see table in Supplemental Data 7.4).

One child (YR) dropped out of the study after 4 sessions. YR seemed to be unaware of his impairments, and decided that he no longer wanted to be involved in any rehabilitation. His challenging behavior (see Table 7.1) and school absconding were the main issues at the time of the study. He was however included in this pilot study initially because it was hoped that an intervention focusing on meaningful activities might be accepted by YR in contrast with all the other rehabilitation and school support he refused.

Table 7.1: Characteristics of the participants

	PB	CS	RK	IP	YR
Sex	Girl	Girl	Boy	Boy	Boy
Age at inclusion (years)	11	11	13	8	14
TBI mechanism	MVA (passenger)	Fall of metal bar on the child's head	MVA (pedestrian hit by car)	Fall of furniture on the child's head	Collision against running child
Age at injury (years)	2.5	6.5	7	5.5	2.5
Initial GCS	6	4	3	6	<7
Brain imaging	Large right hemisphere hemorrhage and edema, right parietal depression fracture	Cerebellar and right parieto-occipital lesion with depression fracture	Subdural hematoma with diffuse edema and pneumocephalus	Brain stem hemorrhage, Diffuse subarachnoid hemorrhage	Unknown
Duration of coma (days)	Unknown	1	10	6	Unknown
Time since injury (years)	9	5	6	3	11
Schooling	Ordinary school + part-time SA	Part-time special – part-time ordinary schooling	Part -time private schooling with SA, part time private lessons	Ordinary school + part-time SA	Special schooling Excluded from school half of the year for behavioral issues
Associated impairments, reported in medical records and previous neuropsychological assessments	Lack of awareness Epilepsy absences treated by carbamazepine	FSIQ 69 Impaired ToM and language pragmatics Moderately spastic equinus foot	Attention problems Left arm weakness	ADHD	Severe behavioral disorder Lack of awareness

Table continues

Table 7.1 (continued): Characteristics of the participants

Glasgow Outcome Scale	2	3	3	2	3
Family structure	Monoparental	Parents separated geographically, Sister followed-up for a transplant	Large family (10 siblings)	Monoparental family Father in prison Primary caregiver: cousin	Two parent household
Parental education (years)	father : 14 mother : 15	father : 11 mother : 11	father : 22 mother : 15	father : 7 mother : 11	father : 4 mother : 17

Note 1: MVA: Motor Vehicle Accident; GCS: Glasgow Coma Scale score; SA: School Assistant; ToM: Theory of Mind; FSIQ: Full Scale Intellectual Quotient; ADHD: Attention Deficit – Hyperactivity Disorder

Note 2: Full names of traditional paper and pencil tests: WISC: Wechsler Intelligence Scale for Children; BADS-C: Behavioural Assessment of the Dysexecutive Syndrome for Children; CMS: Children’s Memory Scale

Supplemental Data 7.3: Childhood Executive Function Battery (FEE) raw scores for participants and their age-, sex- and socio-economic status-matched controls.

	Processing speed (Stroop part 1: color naming in seconds)	Inhibitory Control (Stroop interference condition: number of uncorrected errors)	Flexibility (Barre-Joe **: time in seconds)	Planning (Labyrinths: number of errors)
PB	105*	3	405*	5*
Controls (n= 9)	77,56 ± 10,50	1,11 ± 1,17	264,11 ± 57,20	0,9 ± 1,00
CS	103*	6*	356	5*
Controls (n= 7)	70,4 ± 39,29	1,33 ± 2,07	338,29 ± 175,54	0,7 ± 0,73
RK	83	4*	380	8*
Controls (n= 14)	71,50 ± 10,14	1,00 ± 1,30	248,14 ± 51,03	0,9 ± 1,63
IP	99	4*	613*	7*
Controls (n= 13)	100,38 ± 35,88	1,46 ± 1,51	416,83 ± 95,96	1,2 ± 1,28
YR	69	15	314*	19*
Controls (n= 14)	71,50 ± 10,14	1,00 ± 1,30	248,14 ± 51,03	0,9 ± 1,63

*p<0,05
 ** Barre Joe consists of crossing 46 "Joe" characters on an A3 sheet containing 240 similar characters differing only by leg and arm position

Supplemental data 7.4: Standard scores for traditional paper and pencil tests

	WISC IV matrices	WISC IV vocabulary	6 part test BADs-C	CMS stories - immediate	CMS stories- delayed	CMS backward span	CMS words list - immediate	CMS words list - delayed
PB	9	9	10	8	5	8	9	13
CS	7	5	7	8	7	8	12	7
RK	16	7	8	4	4	10	12	16
IP	11	12	6	8	9	18	14	16
YR	11	6	7	9	8	Missing data	Missing data	Missing data

Note: WISC: Wechsler Intelligence Scale for Children; BADs-C: Behavioural Assessment of the Dysexecutive Syndrome for Children; CMS: Children's Memory Scale

Qualitative data about the program. The intervention appeared to be feasible to implement and it was reported that children enjoyed it, especially the meaningful activities and stories used in the theoretical modules. Most GMT concepts were understood by the children, although examples of personal cognitive failures were difficult to obtain. Interestingly, children seemed to consider the metacognitive strategies as exercises to practice rather than something that could be applied to other tasks. As such, they would use the strategies on theoretical modules (e.g. pausing regularly and stating the goal while sorting cards with an embedded PM task) but were reluctant to apply them to more complex and ecological activities such as cooking, judging the strategies as an additional task per-se and demonstrating no consistent application of strategies in the meaningful activities. Only the 14 year-old RK, probably the most aware and the most impaired in daily life, actually engaged with the techniques and used them whenever he noticed task similarities. CS seemed to understand only a few GMT concepts and metacognitive strategies. IP and PB seemed to lack awareness of impairments and reported not finding the intervention useful, but found the program was fun and they participated willingly.

Repeated PM measures. The Saint's Day Test was performed by 3 children. IP, aged 8, did not complete the task as he was not familiar with mobile phones, did not know how to use the internet and making a phone call to an unfamiliar person was not age-appropriate. YR dropped out of the study. Weekly PM score changes over time are shown in Figures 7.1a, 7.1b and 7.1c. During baseline, none of the children reached a score of 50%. Using a two-standard deviation band (2SDB), all children showed statistically significant progress, as all had at least two consecutive points outside their 2SDB. The best progress seemed to be made by PB. Unfortunately, when her performance was reaching 100% on week 12, she lost the charger of her mobile phone and her parents did not replace it until the end of the study giving only a medium effect (NAP = 0.47 [0.12-0.81]). CS began to make progress on the SDT only by week 8. She maintained performance until school holidays (week 17) when her performance dropped momentarily to 0 (overall medium effect; NAP = 0.74 [0.50-0.98]). RK had a very variable performance but a strong effect of intervention (NAP = 0.87 [0.713-1]). The use of the following strategies to manage the task were reported by parents and/or children: stopping all activity up to one hour before the target time and watching the clock (RK), using cues such as the view of her computer (CS).

Figure 7.1: Saint day task (SDT): prospective memory score changes over time

Note: The vertical line correspond to the beginning of the intervention. Arrows correspond to split –middle celeration lines. Dashed lines correspond to +2 and -2 standard deviation band

Figure 7.1a:

Child PB

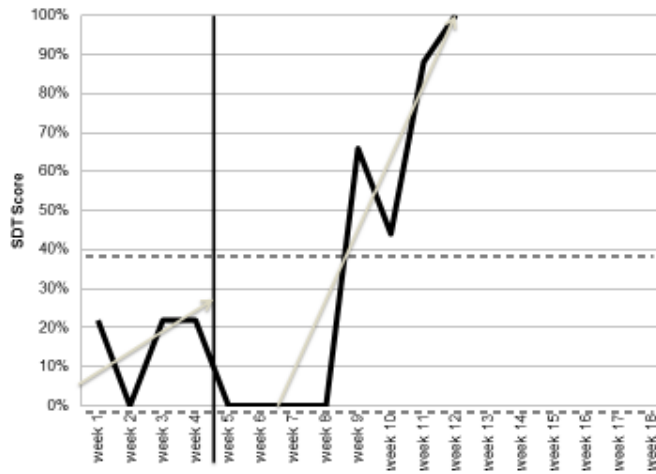


Figure 7.1b:

Child CS

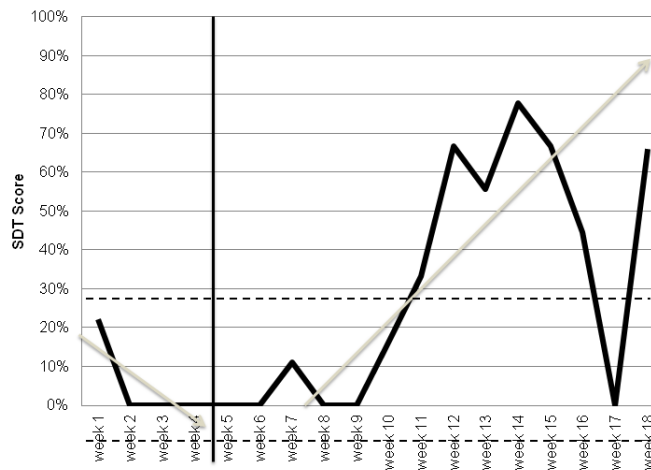
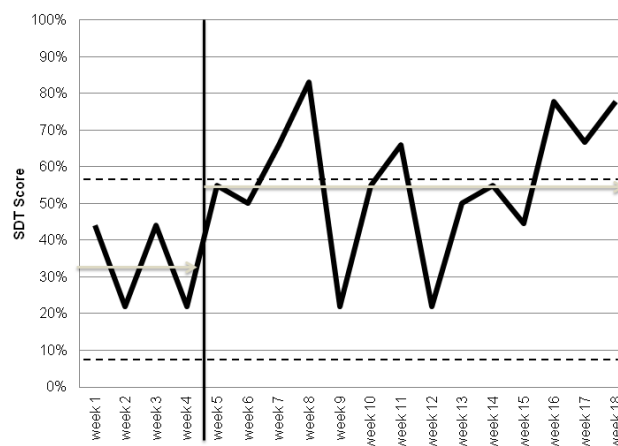


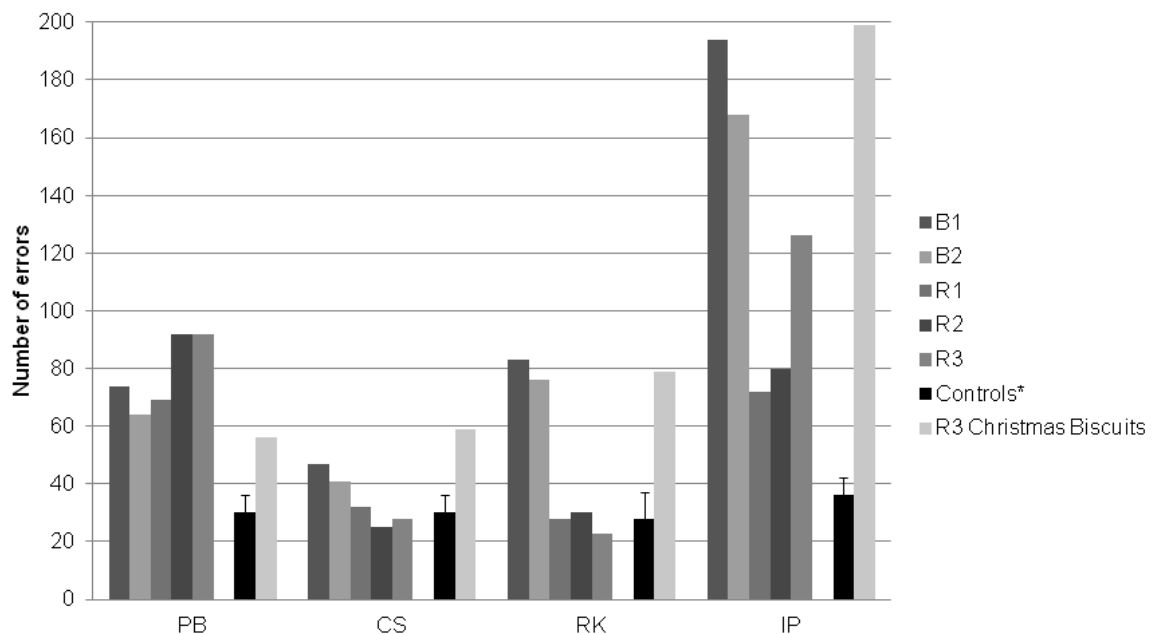
Figure 7.1c:

Child RK



Complex Cooking Task Management: Ecological EF test “Children’s cooking task” and its parallel form (“Christmas Biscuits”). All children were very impaired on the Children’s Cooking Task (CCT), scoring from 3 (CS) to 25 (IP) standard deviations below age matched controls. RK and PB required help from the examiner to finish the task, IP completed the task with nearly 200 errors, and failed the task. CS’s errors were mainly commentaries/questions on every action she undertook but she successfully finished the task.

Figure 7.2: Number of errors in the Children’s Cooking Task (CCT) changes over time



**age-matched controls, n= 8 to13 per child*

Note: B1 and B2: first and second baseline assessments; R1, R2, and R3: assessments performed at 0, 3, and 6months postintervention. In the CCT, errors include action errors as well as commentaries/questions.

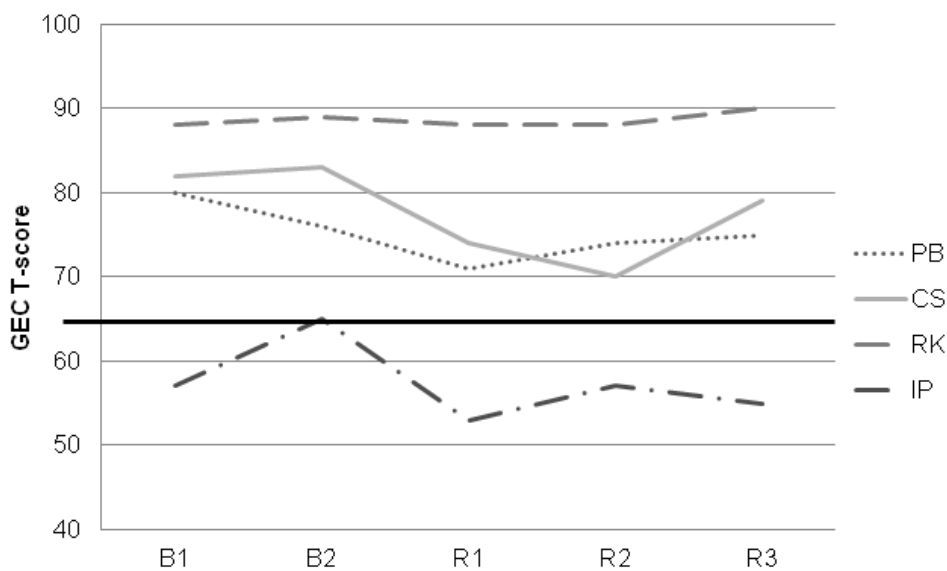
Changes in the number of errors from baseline to post intervention and follow-up for each child are presented in Figure 7.2. During baseline, all children showed some practice effect between B1 and B2. After intervention, PB *increased* number of errors. Interestingly, she was so focused on not repeating the errors from her previous trial, that she often skipped whole recipe steps, and thus forgot more ingredients/steps throughout the trials. Furthermore, from trial to trial, PB seemed more confident each time, and stated how well she knew the recipe and how easy it would be. For CS, the decrease of errors from B2 to R1 had a small effect

size (see table 7.2). Conversely, RK and IP significantly decreased the number of errors after the intervention with large effect sizes, RK showing a performance similar to controls after intervention and IP improving from 25 to 6 SD compared to controls. RK clearly used the metacognitive techniques taught in the intervention while performing the CCT (checking he finished a step before moving to the next, saying “Stop!” and thinking before adding a new ingredient...). Effects were totally maintained at 3 and 6 months for RK whereas effect progressively diminished at 3 and 6 months for IP. However, when using a completely different and unknown recipe at R3 (Christmas Biscuits) all children returned to their initial number of errors.

Table 7.2: Outcome measures Effect Sizes (ES) comparing B2 to R1, R2 and R3			
	ES at R1	ES at R2	ES at R3
Children’s Cooking Task			
PB	-1.15	-2.90	-2.90
CS	0.33	1.09	0.76
RK	1.42	1.35	1.59
IP	1.31	1.16	0.30
Parental BRIEF			
PB	0.5	0.2	0.1
CS	0.9	1.3	0.4
RK	0.1	0.1	-0.1
IP	1.2	0.8	1
DEX-C Cognition Score			
PB	1.33	2.00	2.33
CS	0.67	1.00	1.33
RK	0.67	0.00	-0.67
IP*	2.00	1.67	1.84
IP compared to B1*	0.67	0.33	0.50
<p><i>Cohen’s rating of effect size: 0.2 = small; 0.5 = medium; 0.8 = large.</i></p> <p><i>*Because IP’s DEX-C Cognition Score at B2 was deviant (see figure 4), we also report here ES comparing post intervention outcomes to his best baseline score (B1)</i></p> <p><i>Note: R1, R2, R3: assessments performed at 0, 3 and 6 months post-intervention; BRIEF: Behavior Rating Inventory of Executive Functions; DEX-C: Dysexecutive Questionnaire for children</i></p>			

Transfer to natural contexts. At baseline, all children but one (IP) scored in the clinical range (T-scores > 65) for Global Executive Composite (GEC) scores on parental and teacher BRIEF questionnaires. *Transfer to home-context:* All parental scores were consistent (inconsistency score < 9). Immediately after the intervention 3 children (PB, CS, IP) showed a decrease on parental BRIEF scores (see figure 7.3), reflecting possibly less executive dysfunction in daily life at home. Effect sizes are reported in Table 7.2. All but one child (IP) showed a decrease on DEX-C cognition sub-scores (see figure 7.4) between the two baselines that was considered to be the enthusiasm effect we had expected due to intervention novelty. However, the decrease was accentuated much further after the intervention for all children and continued to decrease at three- and six-month follow-up for PB and for CS with large effect sizes (2.33 for PB and 1.33 for CS, see table 7.2). *Transfer to school-context:* Teacher BRIEF scores remained stable for RK and CS, were unreliable for PB and for IP (inconsistency index >9), meaning it was not appropriate to draw any reliable conclusions on EF in the school context for these children, although PB's BRIEF seemed to show significant improvement. PB was reported to have made excellent progress at school on school academic reports.

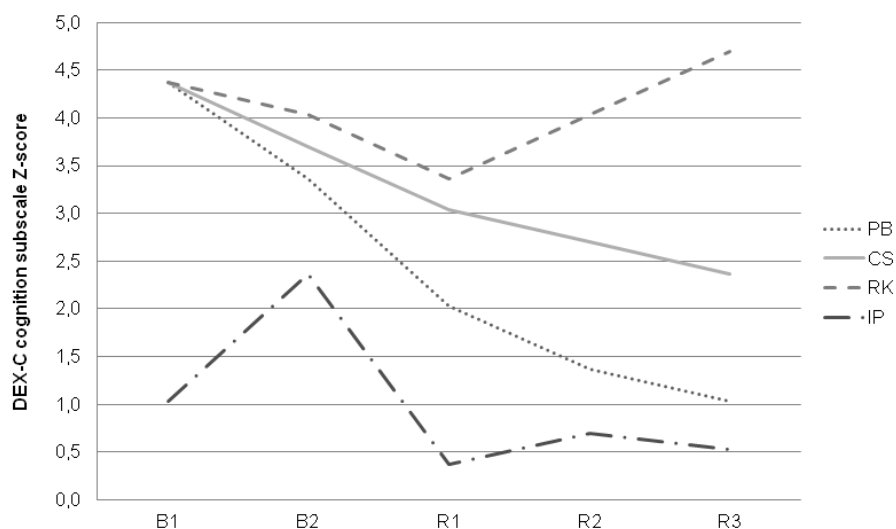
Figure 7.3: Parental BRIEF questionnaire changes of GEC Tscore over time.



Note: GEC indicates Global Executive Composite.

B1 and B2: first and second baseline assessments; R1, R2, and R3: assessments performed at 0, 3, and 6 months postintervention. The horizontal line represents the clinical cutoff score of 65.

Figure 7.4: DEX-C Cognition subscore change over time.



Note: B1 and B2: first and second baseline assessments; R1, R2, and R3: assessments performed at 0, 3, and 6 months postintervention. DEX-C indicates Dysexecutive Questionnaire for Children.

Generalization: Goal Attainment Scaling and post intervention interview.

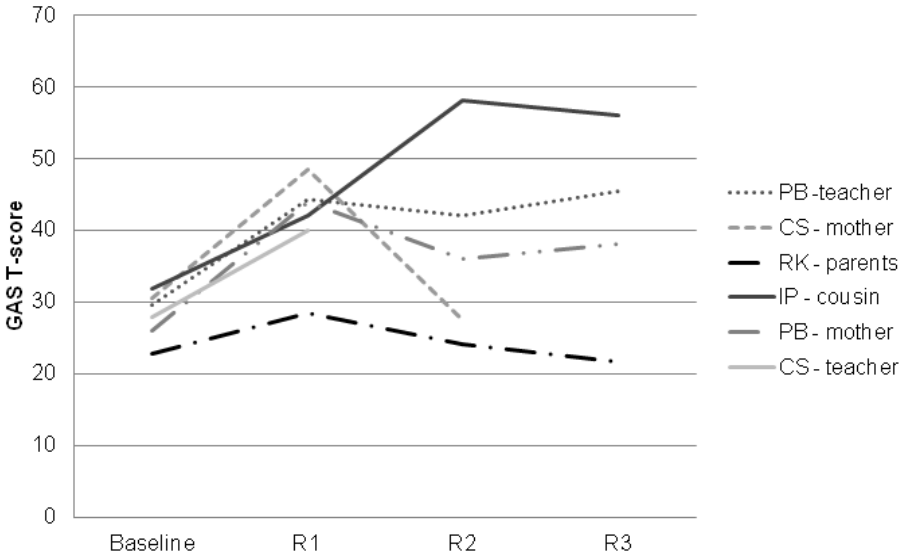
There was a high rate of missing GAS data. GAS could not be developed in collaboration with RK’s school everyday people because he attended school very rarely in that period. GAS goals were developed with IP’s teacher and school assistant but post intervention GAS forms were handed to IP who lost them (as school closed for 2 months after R1, new forms could not be obtained). CS’s teacher, with whom the goals were developed, changed after R1, explaining missing data for R1 and R2. Overall, GAS scales were obtained for at least one “everyday person” per child (see table 7.3). We were only able to agree on personal goals with one child (RK). The other three viewed the goals proposed by their everyday people as not problematic or not important.

Table 7.3: Personal and general GAS goals

<p>Examples of <u>personal GAS goals</u> corresponding to EF-related problems reported by <u>everyday people</u> (only the goals and not the full Goal Attainment Scales are reported)</p>	<p>Examples of <u>general GAS goals</u> corresponding to <u>metacognitive strategy use</u> and <u>GMT application</u> (only the goals and not the full Goal Attainment Scales are reported)</p>
<p><u>PB - mother :</u></p> <ul style="list-style-type: none"> -to forget taking antiepileptic drug less often -to lose fewer objects -to be flexible enough to change strategy if the first strategy does not work -to brainstorm for possible solutions before rushing to start a task or school exercise <p><u>PB – school assistant:</u></p> <ul style="list-style-type: none"> -to hand-in homework on time/not so late -to remember to give <i>routine</i> weekly documents to her mother -to remember <i>non-routine</i> one-off items (e.g. bring money for excursion, ask parents to sign the excursion form..) <p><u>CS – mother:</u></p> <ul style="list-style-type: none"> -to estimate if a school exercise will be hard or easy before beginning -to check school work for errors before handing it in -to check she has understood what she is supposed to do before beginning a task -to ask questions if she is not sure she understood what she is supposed to do <p><u>CS – teacher :</u></p> <ul style="list-style-type: none"> -to accept the need to check her work when she is prompted to do so -to estimate the difficulty of exercises/tasks <p><u>RK – both parents and the child:</u></p> <ul style="list-style-type: none"> -to be able to tidy up his room (without the need for someone to tell him in which order to do it) -to be less stressed about his prospective memory problems -to be able to perform an instruction made of 3 consecutive tasks (e.g.: “drink your milk, empty the dish washer and get ready to go out”). <p><u>IP – teacher and school assistant:</u></p> <ul style="list-style-type: none"> -to be able to prepare schoolbag alone -to write down information/instructions from teacher without being prompted by school assistant -to remember to check agenda to see what needs to be done 	<ul style="list-style-type: none"> -to be aware of one’s “Oops” errors (attentional slips) -to detect “Oops” errors as they occur -to stop and think before beginning a new task -to formulate a task’s main goal before beginning a task (e.g. school exercise, home activity..) -to write down the things one might forget to do -to remember to look in the note book to perform the intended action -to follow a series of steps that are given to perform a task, finishing each step before moving to another -to split complex tasks into steps and substeps -to check a task/exercise before moving on to another

All children progressed towards their goals at R1 (see figure 7.5). However, only one of the GAS scores (IP's) reached the expected goal attainment level (T-score of 50). During the external interview at the end of the study, PB's mother reported significant daily life benefits of the program: PB forgot her antiepileptic drugs much less often (by linking her breakfast orange juice with remembering to take her medications), was less often late at school and made important progress at school allowing her to continue schooling in an ordinary class with a school assistant rather than going to special education as had originally been planned. These last two improvements were not captured by the child's GAS scores because these were unanticipated positive outcomes. Some positive outcome was reported for IP by his main carer, which was consistent with a GAS score that reached 50. Parents reported some general progress in well-being at R1 for RK and CS. Most children carried on cooking after the intervention and RK was for the first time allowed to be in the kitchen alone by his parents. Parents reported that children's self-esteem increased because they could make a meal for the family.

Figure 7.5: Goal Attainment Scaling T-score evolution over time.



Note: R1, R2, and R3: assessments performed at 0, 3, and 6 months postintervention.

Participation of everyday people. Overall, the level of participation of the everyday people was very low. Mission sheets were very rarely filled in. Only RK's parents asked for feedback after the intervention. In interviews, parents, teachers and school assistants all reported that

the intervention was fun for the child and that metacognitive techniques “were useful”. However, when asked to provide examples of metacognitive strategies contained in the “Cognitive Coaching” guide they had received, seven out of ten everyday people recalled less than half of the strategies. Moreover, examples of strategy use were not always appropriate. Between-session assignments (including the simple task of helping the child to detect and write in a table when an “Oops error” had occurred) were never or rarely done. Concrete intervention content was much better followed than the abstract demand of “cognitive coaching”: one school assistant (PB’s) regularly used the paper notebook to compensate for PB’s constant PM failures relating to school goals (bring sports clothes, get a form signed), one mother (CS’s) started to cook with her daughter. Several parents reported that using the term “Oops error” helped to lower family’s tension to the child’s cognitive failures and some began using the term with their other children and themselves. All parents reported being generally too busy to apply the cognitive coaching at home. Teachers reported the children did not use the strategies at school, but had not prompted the children to do so. Both teachers and school assistants tended to emphasize the behavioral, attentional and “lack of effort” problems at school as the key problem for the child and did not consider metacognitive strategies use as a priority for the child. A lack of knowledge about TBI was identified with children’s difficulties not being seen as cognitive (“he does not try to pay attention”, “he has no friends”).

DISCUSSION

The “Context-sensitive GMT” intervention comprised of (1) an adapted Goal Management Training (GMT), (2) metacognitive strategies practice through meaningful activities, (3) a “cognitive coaching guide” for the child’s everyday people. The program was feasible to implement and apparently enjoyable for children. However, participation of everyday people was limited. Children significantly improved on the Saint Day Task (time-based prospective memory). EF performance in the ecological Children’s Cooking Task (CCT) improved in two children. Three children showed a decrease on parental BRIEF scores reflecting possibly less executive dysfunction in daily life at home. All children decreased their cognition DEX-C sub-score, suggesting that parents perceived improvement in cognitive EF impairment. There were some indications of generalization to untrained tasks in all children, but not sufficient to

achieve the expected level of achievement in EF-related GAS goals. When presented with a truly novel task (the parallel version of CCT- “Christmas biscuits”), all children dropped to their initial level of performance indicating a lack of generalization. Intervention effects persisted at three-month follow-up and were partially maintained at six-month follow-up.

One reason why participation was low for everyday people was that families had difficult situations to deal with (see Table 1), leaving little time for the cognitive coaching of their child. The chronic phase of TBI may not be the optimum time for new cognitive coaching practices to be taught to parents and others, as many habits have already settled. The cognitive coaching guide, although simply explained, was abstract and everyday people were not involved in direct training sessions, as opposed to other programs^{267,286,269}. The cognitive coaching guide was rarely used by the teachers and school assistants. It is not entirely clear why this was, but one possibility is that the relatively limited contact with the investigators (a phone call before and after the study and the rest through a written guide) was not sufficient to engage them in the intervention contrary to other school-delivered interventions²⁹⁰. This is clearly an important issue for future studies and for clinical interventions that depend heavily on a child’s everyday people for success. It is probably easier to engage school staff when the interventions are aimed at responding to their needs (especially managing behavior problems such as those reported in Feeney and Ylvisaker studies^{290,289}). In everyday clinical practice, frequent contact with the child’s everyday people (especially school staff) is often not feasible and so examining whether written information (such as our cognitive coaching guide) can facilitate intervention support from these everyday people is an important research question.

The prospective memory (PM) performance might have improved because the task became familiar and routine. However, previous studies with adults using a similar design did not show an improvement of performance with time⁹⁷. Furthermore, children performed so poorly and with so much variation from week to week (very rarely giving the Saint day within one hour of the target time) that no possible routine could have been established. We could not control for the performance of the ongoing task (activity the child was doing at the target time). It has been emphasized that PM performance needs to take into account performance in the ongoing task as well as the PM performance because PM paradigms can be considered as a dual task paradigms⁶⁵. As such it is possible that PM performance increased at the expense of ongoing activities. For one child (RK), his parents actually reported that he stopped all

activity up to one hour before the target time, watching the clock in order to perform the task (but often actually forgot the task anyway). Pausing activity to avoid missing an important phone call appointment may be considered an effective strategy in real life, albeit not for an hour beforehand.

Although ratings on the parental BRIEF questionnaire improved, better scores may not have been due to improvement in EF. Rather, it is possible there was some bias in questionnaire responses. For example, parents were involved in the training and their responses may have reflected a desire to be perceived as good cognitive coaches. Furthermore, the “home-school cognitive coaching” guide may have increased carers’ insight into the child’s difficulties, meaning that even if improvements in behavior had occurred these were balanced out with greater awareness of difficulties on the part of the carer. These issues could perhaps explain why the other outcome measures (CCT, GAS) were not consistent with BRIEF scores (e.g. the BRIEF scores of CS and PB decreased but they did not improve on the CCT and whilst RK made best progress on CCT there was no corresponding decrease in BRIEF scores). However, it is possible that metacognitive strategies are effective in a time-limited task such as the Children’s Cooking Task (CCT) but impractical in the context of daily life’s constant attentional demands, as it is an effortful, top-down process. This may explain why RK made good progress on the CCT, but did not apply the metacognitive strategies in daily life so that parents did not notice a real change in everyday life post-intervention. Alternatively it may be the case that parents and other everyday people may not have had sufficient training to enable them to support the children to implement the strategies in everyday situations consistently. Furthermore, differences between objective measures and improvements reported by parents and patients have been frequently noted^{362,65}. Correlations between parental BRIEF scores and EF classical tests^{151,153} and with the CCT¹²⁰ are typically small so the BRIEF might not have captured the children’s progress. However, all children significantly improved on the DEX-C cognition scores at R1, including IP and RK, which is consistent with earlier finding that the number of errors in CCT and DEX cognition subscales are highly correlated¹²⁰ in both adults^{357,379} and children¹²⁰ and might be a better measure of the children’s executive progress than the BRIEF.

For PB it is difficult to explain the contrast between consistency of improvement on DEX-C Cognition subscore, GAS, qualitatively reported generalization, parental BRIEF – and the

increased number of errors on CCT. This should however be interpreted with caution because all improvements were based on subjective informant reports, mostly of her mother. As PB seemed to have very poor awareness according to the therapist, the intervention might have improved her awareness rather than EF, which would explain perceived improvement in her natural contexts but not on objective measures of EF (CCT). Nevertheless, she is the child who seems to have benefited most from the intervention, with lasting effects at six months. This was unexpected as lack of insight is known to impede patients from actively engaging in rehabilitation ¹¹⁶ and is a factor of poorer outcome. Indeed, PB never found the sessions “useful”, but only “fun”. In the absence of awareness, her motivation did not seem to be to overcome her difficulties (perceived as non problematic) but rather to enjoy herself during the sessions, and through that enjoyment some implicit learning may have occurred. In children, enjoyment may be more important to an intervention’s success than awareness and our intervention seems to have fulfilled this requirement. As Bjorklund noted, “Trying something new may be a goal into itself, and the fact that it does not improve performance may be relatively unimportant to children” ²⁸⁸. This may be why children were happy to try the metacognitive strategies on paper-and-pencil tasks but showed no consistent application of strategies in the meaningful activities. The same finding has been reported in Missiuna’s study²⁸⁶ of cognitive strategy training in children with TBI: making the intervention fun was identified as being useful, whereas the “Goal-Plan-Do-Check” strategy (that is similar to GMT) and promotion of good strategy use were not. Considering together the evidence of ours and Missiuna’s studies (both on very small samples), it seems that strategy use does not come easily to children with more severe TBI and therefore may not be the best rehabilitation approach for them. In any case, strategies need to be simple, concrete and repeatedly practiced in order to benefit those children.

Usually, elaborating a goal attainment scale (GAS) serves to focus rehabilitation on that goal. Such goal-focused rehabilitation is indeed an effective approach. However this presents a methodological challenge for EF-research: when a task is trained, 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 therefore not “executive”), rather than more general problem solving and goal management processes ^{165,367}. On the contrary, daily life is full of EF-demanding tasks that require conscious, novel and effortful processing^{22,165}, without lapses into automaticity. Apparent progress after a goal-focused

training may not necessarily reflect changes in underlying executive processes needed to face daily life. Conversely, our aim was to improve children's ability to cope with new, EF-demanding situations. As an outcome measure needs to be novel (therefore untrained) to make significant demands on EF^{166,165}, personal GAS goals were *not* trained, to keep GAS as a generalization measure of EF. However, in future studies it would be more pertinent to divide child's goals into a trained set of goals (and corresponding GAS) and an untrained set of goals (and corresponding GAS) and then to focus the intervention on training the former while using the latter as an ecological generalization measure. In such an approach, main issues would be to match GAS sets for level of difficulty, child's interest and level of priority as seen by everyday people who participated in goal selection. Furthermore for GAS aiming at measuring generalization, it would be important to control how much explicit linking to these goals is done during the intervention. The intervention would probably be more effective if it combined goal-focused rehabilitation and general metacognitive training. Elaborating two sets of GAS has already been proposed by Schlosser³⁸⁰ in the concept of "control goals". This could have supported further the finding of our study that children could be trained effectively in a meaningful task such as making a chocolate cake (CCT) by combining metacognitive training and repeated cake baking but could not be trained to manage a new untrained recipe (Christmas biscuits). More broadly, in rehabilitation research, using two sets of GAS would make of GAS methodology both a powerful motor for achieving meaningful goals by focusing intervention on them and a pertinent measure of generalization. Future research should also focus on other goal-setting procedures: we did not manage to agree on EF goals with the children in our study, whereas in Missiuna's study²⁸⁶, children were able to self-identify goals using a more framed and age-appropriate goal setting system than GAS, which could be used in future studies. However, children in Missiuna's study²⁸⁶ had sustained mild to moderate TBI and were probably less impaired. Goal setting requires some basic level of awareness, which children with severe TBI often lack. Lack of awareness was identified for all children in our study except RK and seemed to be the main reason why goals could not be identified by the children. Besides, in Missiuna's study, children were allowed to choose any goal (e.g. learn a new sport) whereas we purposefully retained only EF-related goals. More in-depth assessment of awareness would bring valuable contribution for research on goal-setting procedures.

The intervention was able to improve one particular prospective memory task performed in an ecological setting (SDT), it allowed some children to perform better in a cooking task (CCT) and it resulted in some gains in daily executive functioning. However, this metacognitive training did not allow enough generalization effects to reach expected levels in EF untrained personalized goals or to manage a novel complex EF-demanding task. The aim of providing children with meta-cognitive strategies applicable to "any" situation in life is an ideal goal but is perhaps not feasible: children's ability to cope with new, EF-demanding situations of daily life may not be possible to improve with training in case of severe impairments. In those severe cases, a repeated, goal-focused rehabilitation using activities that are meaningful to the children, and *not* focusing on explicit generalization training, may be a more reasonable therapeutic option (see ²⁸⁶ for an example of effective goal-focused intervention in a small sample of children with TBI). It should also be emphasized that GMT targets more specifically the PM aspects of EF-demanding tasks and much less problem-solving abilities. This study supports a recent review²²⁹ in adults which concluded that GMT is probably more effective when combined with other interventions targeting other aspects of EF such as problem solving and initiation (see ^{119,118} for examples of such interventions in adults).

Limitations of previously published studies included: insufficient assessment of generalization³⁸¹, of specific effects on EF^{382,267}, lack of objective cognitive performance measures (using questionnaires only as the outcome measure)²⁹⁹, use of problem-solving tasks that lack ecological validity²⁹⁷, lack of demonstration of EF difficulties prior to intervention³⁷⁴ or lack of multiple baseline or follow-up in pre-post designs^{383,286}. Others focused mainly on the behavioral aspects of the dysexecutive syndrome^{290,300}. This study is to the best of our knowledge the first study that explores whether metacognitive training generalizes and helps children to adapt and manage a novel EF-demanding task and to achieve untrained goals. Of course the small sample of this study does not allow to draw general conclusions about the program efficacy. Our results must be interpreted with caution, especially because we included the most challenging population for this pilot intervention, which may have limited its effectiveness i.e. children with severe EF impairment and with three known major factors of poorer outcome^{12,14,71,77}: (1) *severe* TBI; (2) sustained at an *early age*; (3) in non-optimally functioning families (similarly to Corbett's GMT³⁷² that targeted children from low socio-economic background in Cape Town with little success). Those children are usually excluded from protocols²⁸⁶ and adult interventions using GMT usually target patients with moderate

and mild TBI^{99,119}. As this study demonstrates the feasibility of the program for a particularly complex group of children, it would be helpful to replicate this study with children who have sustained a moderate TBI and children who may have more access to everyday people for providing cognitive coaching. On the other hand, in the clinical setting, it is precisely children with severe TBI and complex family situations that are most needy of intervention and future research should focus on this group, in spite of its challenges.

For clinical use, the intervention may need further adaptation: the program may benefit from being longer to allow the children to integrate each strategy before practicing a new one, everyday people should be supported further to participate in the sessions, in a similar way to that used in Braga's study²⁶⁷. Cognitive coaching should be presented through concrete activities to be done at home and at school rather than general concepts and advice. The intervention should be more closely embedded in the family life in order to improve family participation without adding an additional family burden^{384,385}. Not all families are willing and/or capable of engaging in a family-delivered program³⁸⁵ and evaluating how to predict this prior to intervention would be of benefit to service providers. Direct contact with school staff is needed. The impact of parental metacognitive knowledge, skills and beliefs on outcomes in family-delivered interventions would also be a valuable component of future studies.

CHAPTER 8: METACOGNITIVE STRATEGY USE IN CHILDREN WITH SEVERE TBI

INTRODUCTION

In daily life our behaviours often implicitly reflect metacognitive knowledge. We act in ways that reflect that we are aware of how our cognitive processes work, and their limitations: double checking a bill, writing “to-do” lists when planning a week-end trip and reviewing if we have met all our goals for the day before leaving work, are examples of use of ‘metacognition’. We know that we might have made an error in the bill; we know we might forget one of the steps to make the week-end trip successful and we know we might have forgotten one of the tasks we were supposed to do at work that day. Through experience, we acquire knowledge about our cognition and implicitly learn how to overcome cognitive failures before they actually happen.

We know from developmental literature that older healthy children use strategies more often, select more effective and efficient strategies and perform better than younger children, but even young children use some strategies that reflect knowledge of their cognition²⁸⁸. Although younger healthy children don’t usually produce strategies spontaneously (termed “production deficiency”), they can be trained to use a strategy and enhance their performance as a result ²⁸⁸. They will, however, often fail to generalize a trained strategy to a new task, reverting to their nonstrategic ways ²⁶¹. Conversely, other children will use a strategy but it will not improve performance (termed utilization deficiency) because of the effortful nature of strategies: children who use a strategy (especially a new one) may not have enough “mental resources”²⁸⁸ and especially working memory (WM) space left to devote to solving the problem at hand. Furthermore children’s lack of awareness that a strategy is not helping, and generally poor metacognition, may mean they fail to recognize that the extra effort they are applying isn’t benefitting them ³⁸⁶. Children who practice strategies eventually get good at

them, and their performance improves. As utilization deficiency is short-lived, teaching strategies to children is an effective process in healthy children.

METACOGNITION AND TBI

Since Price described two patients with frontal damage acquired early in life whose lack of “insight and foresight” (i.e. metacognition) resulted in severe behavioral problems ⁴³, a number of studies have shown metacognitive impairment in children with TBI ^{81,82,83,219,220,221,84}.

Literature has shown that adults with TBI are less able to detect their errors than controls in multitasking activities ²⁴⁷ but once detected, they are able to correct them just as well as controls. However, patients with traumatic brain injury (TBI) tend to repeat errors they had detected and corrected, while controls rarely repeat errors after they have corrected them once²⁴⁷. It has been proposed that inability to self-generate feedback could be a reason why patients with TBI are described as poor at learning from their errors.

The use of metacognitive strategy training is a practice standard in adults with executive functioning deficits ¹¹⁴, however there is no evidence for effectiveness of metacognitive training for children with a dysexecutive syndrome ^{110,113,111}. Since Vygotsky’s ³⁸⁷ and Luria’s³⁸⁸ exploration of problem-solving, metacognitive strategies have been proposed to help children in problem solving: Talking aloud to solve a problem ³⁸⁷, “Goal-Plan-Do-Check”³⁸⁹ ²⁸⁶ “Stop and Think” are most well-known examples. Authors advocate metacognitive strategy use in children presenting with developmental coordination disorder^{295,389}, specific learning disabilities such as math problem-solving ³⁹⁰, and attention deficit hyperactivity disorder (ADHD), ²⁶⁹ with successful results. The few studies that have explored metacognitive strategy use in children with TBI showed more equivocal results. The “Goal-Plan-Do-Check” strategy (that is similar to Goal Management Training) and promotion of good strategy use were not identified as being useful ²⁸⁶.

Most metacognitive strategies described in the literature have not been validated. Evidence for their age-appropriateness is lacking. Many questions have not been addressed: From a developmental point of view, from what age is a child able to learn to use a particular metacognitive strategy, irrespective of brain injury status? Are brain injured children capable

of understanding, learning and using a metacognitive technique efficiently, while they are specifically impaired in EF-related functions including metacognition? Can metacognition (and especially metacognitive strategy use) be trained in children who have suffered a TBI? And from which age?

The aims of this study were (1) to investigate metacognitive strategy use after specific training (2) to identify possible candidate factors impeding successful metacognitive strategy use.

METHODS

This study was part of a larger pilot study that tested an intervention based on a context-sensitive Goal Management Training combined with ecological activity practice. Details and effectiveness of the intervention are reported elsewhere (see chapter 7)²⁰⁰. The training was administered weekly, for 15-20 hours over a 4-6 month period. As already described in chapter 7, children were taught metacognitive strategies through discussion of stories where characters presented cognitive failures (see table 8.1), introducing how those may be prevented. Then they performed exercises, first without, and then with, a metacognitive strategy as a “proof” of its usefulness, initially on paper-and-pencil exercises, then on ecological activities in the rehabilitation centre and finally on real life activities at home and school (see table 8.2 for an example).

The program used a range of functional, meaningful activities. It was consistent with Toglia’s recommendation ³⁵⁵ of using several settings to increase metacognition. Metacognitive strategies used in the training were developed from Goal Management Training and included the following metacognitive strategies to help children with planning, successful step-following, and carrying out prospective memory intentions: “Stop! What am I doing?”, “I make a plan”, “STOP! I state my goal!”, “I split the task into sub tasks”, “I check the step is finished before proceeding”, “Stop! Am I Following my plan?”, “Make a to do list”, “I write it down in my paper notebook”, “I check my paper note book”, “I look into my mental note book” (the paper notebook is taken as an analogy of the “mental” notebook bearing prospective memory intentions in one’s mind). The children also learnt to recognize and

identify factors influencing the occurrence of “Oops errors” (attentional gaps, action slips, and prospective memory failures) and to prevent its reoccurrence.

TABLE 8.1: Examples of situations and stories used as examples in the training	
<p><u>Daily life problems related to executive dysfunction:</u> e.g. putting an essay to hand in at school into a sports bag instead of a school bag.</p> <p><u>Consequences of the problems:</u> e.g. stress while looking in the school bag for the essay, feeling upset when finding the essay in the evening while getting dressed for football, getting a lower mark because the essay was handed in late...</p> <p><u>Factors that contributed to the problem:</u> e.g.: going to bed late because of writing the essay at the last minute, being in a rush when preparing the school bag and the sports bag the next day</p>	<p><u>Story illustrating “Being side-tracked from your goal”:</u> “Gavin was playing football with his friends in the field near his house, but half through the match, the ball became stuck in a very tall tree and they couldn’t get it down!!! As all his friends wanted to finish the game, Gavin suggested he go and get his own football at home that was close by. So he went home assuring his friends he’d be back in 5 minutes.</p> <p>As he got home, he began looking for the ball when the phone rang, so he answered the phone: it was his cousin John from America. He told Gavin that his parents were planning a holiday in Montgolfier in the United States and that Gavin was invited to go with them!!!</p> <p>He got so excited by the trip that he completely forgot about the match, the ball and about his friends!!! His friends had been waiting for him to arrive with the ball for an hour... and finally came to his house angry!”</p>

Table 8.2: Example of a series of exercises introducing the metacognitive strategy “STOP! State your goal”	
Exercises first without a metacognitive strategy	Do some of each task (word finding, card sorting, identifying differences between two images, number trail)
Strategy introduced	<i>“I want you to say aloud to yourself: “My main goal is to do something from each of the tasks”. I then want you to write this in your mental notebook To try and help you, I am going to say “STOP!” from time-to-time. When I say “STOP!” all I want you to do is to think about what’s on your mental notebook and whether you are on track to achieve that goal.”</i>
Exercises with a metacognitive strategy as a “proof” of its usefulness on paper-and-pencil exercises	Do some of each task (different content of previous tasks but same format and goal).
Exercises with a metacognitive strategy as a “proof” of its usefulness on ecological activities in the rehabilitation centre	Route finding and completing a series of missions (find out the length of the swimming pool, get an appointment in another department, find out the name of the doctor on duty, find a telephone number...)
Exercises with a metacognitive strategy on real life activities at home and school	Set the table for family evening meal: activity proposed to parents in a “cognitive coaching guide”, with explanation of the mental notebook and of the “STOP” cue.

The program used a range of functional, meaningful activities. It was consistent with Toggia's recommendation ³⁵⁵ of using several settings to increase metacognition. Metacognitive strategies used in the training were developed from Goal Management Training and included the following metacognitive strategies to help children with planning, successful step-following, and carrying out prospective memory intentions: "Stop! What am I doing?", "I make a plan", "STOP! I state my goal!", "I split the task into sub tasks", "I check the step is finished before proceeding", "Stop! Am I Following my plan?", "Make a to do list", "I write it down in my paper notebook", "I check my paper note book", "I look into my mental note book" (the paper notebook is taken as an analogy of the "mental" notebook bearing prospective memory intentions in one's mind). The children also learnt to recognize and identify factors influencing the occurrence of "Oops errors" (attentional gaps, action slips, and prospective memory failures) and to prevent its reoccurrence.

Children included in the study had sustained a severe TBI at least two years earlier, had a documented dysexecutive syndrome and had executive functioning difficulties in daily life as reported by parents and school staff (see chapter 7).

Three types of tasks were used to assess children's use of metacognitive strategies after the training: the Six-part test from the Behavioural Assessment of the Dysexecutive Syndrome in Children (BADS-C)²³⁵ and the Children's Cooking Task (CCT)^{120,121,201}, administered before and after the intervention, and the Organization Task, designed for this study (see below for a detailed description) administered after the intervention only. The assessment did not focus on traditional scoring of the tasks but on the strategies children used to earn a good score and the effectiveness of those strategies.

(1) In the Six-part test, children are instructed to do some of each of the six tasks in a set amount of time, respecting the rule of never doing two tasks of the same colour consecutively. As such it is very similar to some of the exercises used in the training. The Six-part test is a validated and normed test of EF, which is considered more ecological than typical EF tests, although it is still a paper and pencil test, performed in an office-based context.

(2) The CCT is a truly ecological EF test, validated for children with TBI but it does not have norms. In the CCT, children have to prepare a chocolate cake and a fruit cocktail following structured, photo-cued, child-friendly recipes contained in a cooking book along with

distracter recipes. Each recipe comprises numbered step-by-step instructions with a picture for each step, however each step can contain more than one action (e.g.: add the sugar, the vanilla sugar and mix). Not all instructions are stated, in order to assess children’s adaptability to the context (e.g.: use a straw to open and empty a carton of apple juice into a glass). The necessary ingredients and utensils are assembled on a table with the instruction sheet and the cookbook, along with distracter utensils (e.g. small bowl next to the needed mixing bowl) and distractors ingredients (e.g. salt next to the sugar). Children are instructed to perform the task with no commentary and to behave as if they were alone, not asking the examiner for help. The task has been shown to be highly sensitive to executive dysfunction as it is novel, challenging, open-ended, uses a real life context, food and utensils, requires multiple goal management and innovative and higher level strategies in order to succeed. It can be performed from the age of 8. CCT scores include the number of “errors”, and qualitative data, errors being defined as omissions, additions, substitutions – sequence errors, estimation errors and commentary-questions. Table 8.3 shows examples of behaviours counted as errors, taken from the sample of the five children in the study.

Commentaries and questions	-asks to stop the recipe because the pastry is not as it should be on the photo (PB) -tells the examiner a knife is missing (RK) -says “That’s so easy! I’m so intelligent” (IP)
Omissions	-does not add sugar (PB) -does not mix the pastry as stated at the end of step three (CS) -adds two glasses of flour instead of three (PB)
Additions	- walks around the table thinking about what to do (RK) - adds ingredients presented on the table but not required in the recipe (YR) -cleans each object every time he uses it (RK)
Substitutions – sequence errors	-starts with the wrong recipe (first recipe of workbook) (RK) -uses a small bowl instead of a salad bowl (IP) -does step three before step two (CS)
Estimation errors	-adds flour to a small bowl already full of sugar (IP) -adds all the sparkling water to the glass with water flowing out of it (RK) -adds all the baking powder without counting how much is needed (YR)

(3) The Organization Task was a multi-element task, designed for the study in which the aim is not to do something of each task as in the Six-part test but to earn as many points as possible in five minutes. Children were given a list of 12 tasks (e.g. sorting cards, learning word lists, circling verbs and colour names in a text, building blocks towers, performing simple calculations). Each task varied considerably in the time needed to complete it (5 seconds to 3 minutes per task), in difficulty and in the number of points the task gives (10 to 200 points per task). There was no correlation between the task length and number of possible points (i.e. some tasks were very short and easy and gave many points). The Organization Task was very similar to an exercise used four times in the GMT (identical in format but different in content, although both used the same materials: cards, blocks, colour pens, texts and word lists). During the training, children were taught to stop, think about the goal and make a plan before beginning an activity. They practiced how to make a plan according to the predicted difficulty of a task, judgment of the time required to complete it, effectiveness in terms of points and personal preference/strength (some children being used to sorting cards others preferring simple calculations). Tasks used were not over-trained tasks as the intervention was not a repetitive training but an exposure to metacognitive strategies. As the format of the Organization Task was identical to exercises used as proof of utility of the metacognitive strategies (see table 8.2) it allowed us to detect *spontaneous* metacognitive strategy *use* without the need to recognize task similarity because task similarity with the exercises from the training was so obvious. In case children did not use the metacognitive strategy spontaneously, however, a second version of the Organization Task (Organization Task 2) followed, in which the examiner explicitly prompted the child to use the strategy “make a plan” before beginning the task, to investigate *accuracy in the metacognitive strategies application*.

The tasks were administered in the following order after the intervention: (1) Organization Task 1; (2) Organization Task 2; (3) CCT; (4) six-part test. The following data was recorded for all tasks: spontaneous use of a metacognitive strategy, response to examiner’s prompts to use a metacognitive strategy (Organization Task 2 only), correct remembering of the metacognitive strategy technique, correctness of metacognitive strategy use, success in the task with metacognitive strategy use. The underlying question was to assess if metacognitive strategies are spontaneously used after training and if they help. Qualitative data were recorded to screen for any factor preventing effective metacognitive strategy use. After the

intervention, parents and children were interviewed as to the spontaneous or prompted use of metacognitive strategy at home to screen for reasons facilitating or preventing children from applying metacognitive strategies in their natural contexts.

RESULTS

Five children were initially included for the study. One child (YR) dropped out after four training sessions, so only the data from the four children who completed the training is presented. Pre and post intervention results of the CCT and six-part test are reported in table 8.4

Table 8.4: Scores for the Six Part Test and the Children’s Cooking Task (CCT):				
PB	CS	RK	IP	YR
Six-Part Test Standard scores before metacognitive strategy training				
10	7	8	6	7
Six-Part Test Standard scores after metacognitive strategy training				
10	7	4	9	
Number of errors on CCT before metacognitive strategy training (second baseline)				
64	41	76	168	
Number of errors on CCT after metacognitive strategy training				
69	32	28	72	
<i>Note: higher six-part test standard scores show better EF, higher number of errors in CCT shows worse EF.</i>				

As previously noted the Organisation Task did not yield a score but was only used to detect metacognitive strategy use. Detailed data of cognitive functioning of the children (and especially EF), injury and demographic characteristics can be found in chapter 7 of the thesis, table 7.1 or in the related paper²⁰⁰. Data on self-awareness (metacognitive knowledge and on-line awareness) and CCT performance are detailed in chapter 4 (and related paper²⁶⁰) and briefly repeated here in table 8.5.

CHILD PB

PB was 11. She had sustained a motor-vehicle passenger accident at the age of 2.5 years with severe brain lesions and immediate neurosurgical treatment. She attended mainstream school with a part time school assistant. She had Epilepsy absences treated by carbamazepine.

Training: PB could use metacognitive strategies correctly but she could not see metacognitive strategy usefulness and was very resentful of using them throughout the program. When strongly prompted to use strategies on simple paper-and pencil tasks (such as GMT card sorting), she interpreted the strategy per se to be the main goal and used the strategy correctly, but was so focused on the strategy rather than the task that her performance dropped compared to card sorting without strategy use. In ecological tasks, when forced to stop and check, she had difficulty noticing her errors and omissions. She viewed strategy use as an additional task and applied it as a goal of its own, to please the trainer rather than as a helpful strategy that could help her to perform an activity. Her attitude towards the training can easily be described with her usual response “I love it! I’d like to do the cooking again but please don’t make me do that stopping and checking again”.

Post-training testing: PB showed no strategy use in any of the post-training tasks. No improvement was seen in the CCT nor six-part test compared to baseline. In the CCT, she did not make the same errors (e.g. forgetting the baking powder) but increased the total number of errors, as she considered the recipe easier each time. In Organisation Task 1, after finishing two easy subtasks, she spent a considerable time reflecting on what to do next, while time was running out and finally earned very few points. Before being prompted in Organisation Task 2, she began making a plan spontaneously but only took some factors into account (mainly personal preferences) without looking at the number of points the task gave. This resulted in a slightly better, but still poor performance. When asked about the plan she made, she said she was quite satisfied with it. It was not clear if her plan was poor because it was long and fastidious to do or because she ignored the goal of earning as many points as possible in order to do enjoyable subtasks and have fun, which is consistent with her overall behavior during the months of training.

Daily life: Surprisingly, while showing no sign of strategy use in the post-training tests, family, school and PB herself reported she had made great progress after the training. She arrived at school on time much more often, without forgetting her schoolbag. Her mother found her explaining to a family friend how her brain worked, using analogies presented during the training. Moreover, PB identified effective cognitive techniques in daily life by herself, for example linking her orange juice with taking her antiepileptic drug at breakfast.

CHILD CS

CS was 11. She had sustained a severe traumatic brain injury (TBI) when a soccer post fell on her head at the age of 6.5 years, causing mainly cerebellar and right parieto-occipital lesions. She attended part-time special, part-time mainstream schooling. Her intellectual quotient was in the low average range, and she had impaired theory of mind and language pragmatics described in her previous rehabilitation reports.

Training: Discussion based on GMT stories was difficult to promote as she lacked theory of mind skills that are necessary to understand the character's thoughts. Her poor vocabulary (e.g. "consequence" of an error) limited her understanding of GMT concepts and metacognitive strategies. CS had severe reasoning difficulties. Even after 15 sessions, she could not logically order steps required to prepare a school bag or to plan a party (from easy informative drawings of party preparing steps), therefore strategic thinking and strategies like "I make a plan" were simply impossible for her. Any unexpected event requiring additional problem-solving made her fail the activity, as her flexibility was low. Conversely, once provided with an explicit strategy, she was willing to apply it, though usually without success. Her strategy use could be described as rigid, not internalized, allowing no flexibility and therefore probably not truly "meta-cognitive".

Post-training tests: CS showed no correct strategy use in any of the post-training tasks (CCT, six-part test), and performance remained stable in both the CCT and six-part test. She was used to cooking quite often with her Mum and seemed to rely heavily on her knowledge and procedural skills for completing the task, rather than using strategies or problem solving. In Organisation Task 1 she spontaneously made a plan on totally arbitrary criteria, without reading the full list of tasks and not taking into account the possible points. She chose mainly the first tasks, including card sorting, which she always performed poorly during the training. As she overestimated her capacity to sort cards, she earned no points in Organisation Task 1. In Organisation Task 2, she made a plan based on personal preferences but without any reference to the goal of earning points and obtained a poor score. CS seemed proud of having made a plan, judging it as a good performance and a goal achieved on its own, ignoring the main goal of earning as many points as possible.

Daily life metacognitive strategy use: School reported no progress and no spontaneous strategy use. Her parents reported some progress in domains that could not be linked with the metacognitive strategy training. It was difficult to obtain information from parents about strategy use in daily life.

CHILD IP

IP was 8 at the beginning of the training. A television post had fallen on his head at the age of 5.5 years, causing severe TBI. He attended a mainstream school with a part-time school assistant. He was diagnosed with ADHD, with symptoms that had probably started prior to the TBI.

Training: IP understood quickly all the metacognitive strategies, although he could not understand what they were for and was often annoyed to be asked to use them. He was protected by his carers, who confronted him as little as possible with his difficulties (e.g. managing his schoolbag preparation). He therefore did not have the opportunity to realize he was impaired and lacked knowledge about the consequences of his TBI. During training sessions, he alternated from overestimating his abilities (“I’m the most intelligent boy in my whole class, this exercise is just too easy”) to deep self-deprecation (“I can’t do it because I’m just too stupid”). He showed strong emotional reactions to his performance, including crying when he was failing a cooking recipe, inappropriate laughing, and throwing a tantrum on the floor when he could not find a solution to a problem. Associated hyperactivity symptoms, possibly present prior to TBI made it very challenging for him to concentrate on the tasks.

Post-training tests: IP showed no strategy use in the post-training tasks. When presented with Organisation Task 1, he said: “Oh, I think I need to make a plan first... No actually I don’t need a plan, it’s too easy!” and began without a plan, trying different tasks without success and earned no points. When prompted to make a plan in Organisation Task 2, he only decided which two subtasks to do first, and spent a considerable amount of time reading the tasks during the testing and obtained a poor score. Although he did not seem to use strategies, his performance on the six-part test and CCT improved.

Daily life strategy use: His teacher and school assistant reported no progress and no strategy use at school. Carers at home thought he made fewer slips and prospective memory failures,

however they could not recall a precise strategy or technique they saw him use and therefore his progress may be due to other factors than metacognitive strategy training.

CHILD RK

RK was 13. He had sustained a severe TBI following a motor-vehicle pedestrian accident at the age of 7. He attended part-time private schooling with a school assistant, part time private lessons. He suffered severe attention problems. Although he was aged 13 at the time of the study, his parents could never leave him at home alone, he was not allowed to enter the kitchen as he usually forgot to switch off the gas.

CCT: He suggested that the recipe was not feasible instead of looking for a strategy to compensate for the lack of a knife (children don't need to cut the butter but if they want to, they can use the fork's handle to cut it). His performance was very much affected by long periods of purposeless actions such as cleaning the pastry left on the mixer with a clean little spoon, cleaning the spoon with a clean wooden spoon, cleaning the wooden spoon with a fork until no clean utensils were left on the table.

Training: RK was very motivated throughout the training. His good intellectual level, rapid reasoning and motivation to be independent helped him understand efficiently the metacognitive strategies and kept his motivation up. He rapidly made the techniques his own and used them whenever he noticed task similarity with a previous module. He could apply them successfully in ecological activities. However, he finished each session exhausted and it was clear that he used great attentional resources to use the strategies and to perform the activities. After the sessions, he was too tired to participate in cognitively demanding activities in the rehabilitation centre.

Post-training tests: RK spontaneously and efficiently used the "Make a plan" technique in Organisation Tasks 1 and 2, without being prompted and took into account all variables, earning the maximum possible number of points. Moreover, strategy use was also seen on the CCT (e.g. checking he finished a step before moving to the next, saying "Stop!" and thinking before pouring a new ingredient). As a result, RK dramatically decreased his number of errors, nearly reaching age-matched controls' performance. However, after completing the CCT task, he completely failed the six-part test, decreasing his score compared to baseline.

Daily life strategy use: RK reported that, though effective, metacognitive strategies were impossible to apply in his stressful and busy lifestyle (he had ten siblings at home). RK reported hurrying to begin a task before he could make a plan or write down the necessary steps. His family reported that RK didn't apply the metacognitive strategies to home daily life situations they would have liked him to make progress in (e.g. tidying his room, crossing the road) so they did not see a real change in his behavior after the intervention. When he did use strategies at home for an activity he selected (for example to make biscuits he had practiced during the intervention for his family), he could obtain better results than before training, however his parents reported that RK was so exhausted afterwards, that his cognitive problems increased in the following activity as a result.

Data from all five children is summarized in table 8.5. For all children, the striking point was the contrast between the effectiveness of the cognitive techniques in the office based tasks (such as the GMT card sorting task) and the difficulty of using them in complex ecological activities (such as cooking): children seemed so overwhelmed with the task requirement that they forgot to use the metacognitive strategies. When children practiced the strategies, they all seemed to consider the metacognitive strategy as something additional to do rather than a way of succeeding at the task in hand. Enjoyment was the main motivating factor, irrespective of success.

TABLE 8.5: Summary of participants' use of Metacognitive Strategies (MSs)

	A ge	Awareness (see chapter XX for details)	Behavior during MS training program	CCT (see chapter 4 and 7 for details)	Six-part test and Organisation Tasks	Daily life
PB	11	Good basic level of intellectual awareness Poor higher level of intellectual awareness and on-line awareness	“It’s not useful but it’s fun” Could use MSs correctly during training but resentful to use them throughout the program	Focusing on not repeating previous errors but making more errors each time as she considered the recipe easier each time. Never repeated same errors but forgot another step or ingredient each time.	Did not apply MSs even when prompted Did not see MSs utility	Paradoxically, MSs application at home and at school; new MSs created by herself and applied with success.
CS	11	Good intellectual (both basic level and higher level) Poor on-line awareness	Poor comprehension and abstract reasoning preventing good understanding and use of the MSs. MS use was rigid, non-truly internalized, allowing no flexibility and therefore probably not truly “meta-cognitive”.	Good performance. Errors were mainly commentaries and questions throughout the recipe while the instructions state to perform the task without asking for help.	Tried to apply MSs but without success. Viewed MSs application as a goal on its own, irrespective of outcome (utilization deficiency)	No effect
IP	8	Moderate awareness, intellectual awareness slightly better than on-line awareness	Quick understanding of MSs but thought he did not need them. “I know a strategy for this but this task is too easy to apply it” Alternated from overestimations of his abilities to deep depreciation	Estimation errors Hyperactivity Behavioral and emotional manifestations Failure to make the cake	Did not want to apply MSs even when prompted.	No effect
RK	13	Good awareness	Motivated but overwhelmed by attentional demands of the task	Endless Perseveration Fluctuation in attention level Difficulty in strategy search and poor flexibility	Applied MSs spontaneously with success to Organisation 1 and 2 and CCT. Failed to apply MSs on the last task (six-part test) and fails.	Effective in short time activities. “Too tiring and time consuming to apply in daily life”

DISCUSSION

After 15-20 hours of metacognitive training, CS used metacognitive strategies inappropriately, without success and without understanding them; IP had learned how to use strategies but did not understand the need to use them; RK learned strategies well and they improved his performance but he could apply them only to time-limited activities and not throughout the day, making them impractical and therefore ecologically of little use. PB showed a paradoxical response with no strategy use in the rehabilitation setting while using them in her natural contexts.

Our results from children PB and CS are consistent with Bjorklund's "strategy utilization deficiency" ^{288,261} described in young children: because of the effortful nature of strategies, children who use a strategy (especially a new one) may not have enough "mental resources"²⁸⁸ and especially WM space left to devote to solving the problem at hand (their focus of attention is on the strategy rather than on the task). Developmental research found a relation between strategy use and WM ^{391,392}. As such CS concentrated on the strategy rather than the task and all her WM was used by the strategy rather than the task, and she did not improve performance with the strategy. Also, during the training, most children resorted to usual nonstrategic ways when they had to move from a paper and pencil task to a more ecological task. Furthermore CS's lack of awareness that the strategy was not helping did not allow her to recognize that the extra effort she was putting in didn't result in any benefit ³⁸⁶. If she had practiced the strategies for long enough, her performance might perhaps improve as utilization deficiencies are short-lived in typically-developing children. There remains the question of whether this is also the case for children whose EF and metacognitive abilities have been compromised by a TBI. Bjorklund also described a "strategy mediation deficiency" (children who can't manage to apply a strategy) and CS's rigid and inappropriate application of strategies might have been due also to a mediation deficiency. Our results are also in line with Bjorklund's finding that the main requirement for children to benefit from strategy training is to have fun while learning to use the strategies, irrespective of their success.

Metacognitive strategy use is a top-down process and thus costly in attentional resources. Young children process information less efficiently than older children do, making them less

likely to use a strategy spontaneously and less likely to benefit from the imposition of a strategy ²⁸⁸. It has been hypothesized that having a detailed, elaborated knowledge base results in faster processing for domain-specific information, which in turn results in more efficient processing and greater availability of mental resources. The relationship between knowledge and strategy use has been demonstrated in many domains^{256,261,288}. Knowledge may influence performance in three ways: (1) by increasing the accessibility of specific items; (2) by the relatively effortless activation of relations among sets of items (without the need to implement an effortful strategy); (3) by facilitating the use of deliberate strategies. The multiple cognitive impairments of included children may have increased WM load and contributed to obtaining little benefit from strategy use. Furthermore, the effortful nature of metacognitive strategy application may make it impractical for use in daily life and may limit its effectiveness given the low attentional resources TBI children experience (RK being an example of this limitation). Therefore, before providing the child and the family with metacognitive strategy training, its ecological validity should be assessed on real life activities. A metacognitive technique effective in an office may not be efficient in daily life and in the natural context of the child.

Preserved metacognition/self-awareness has been linked to the ability to learn the use of compensatory strategies ^{210,393,394,395,396,397}. However it has also been reported that patients can use compensatory strategies without a prerequisite level of awareness ³⁹⁸: Sohlberg's study indicated a dissociation between behavioral and perceptual indicators of awareness - patient K increased his independence in grocery shopping but without any indicators of increased insight (measured by caregiver ratings and judgments of photographs depicting cognitive failures where patient K had to judge if they would likely occur in his life). Sohlberg had proposed that for more severely impaired subjects, it may be more productive to tap into implicit learning and procedural knowledge without addressing declarative knowledge for why the compensatory strategy is important. PB seems to confirm the dissociation between lack of awareness and progress in the problematic behavior in Sohlberg's patient K. PB is particularly interesting because she lacked on-line and anticipatory awareness and did not find the strategies useful, nor did she apply them effectively during rehabilitation sessions but she applied them at home. Some authors suggested that implicit learning plays an important role in on-line awareness. An implicit mechanism is thought to guide behavioral responses in the

absence of conscious awareness of current state³⁹⁹ and is an important component of awareness models accounting for tacit awareness (evidenced by task avoidance and behavioral adaptability in the absence of conscious awareness of deficits). Patients with impaired implicit sequence learning have been shown to have more impaired awareness than those with intact implicit learning⁴⁰⁰. One possible explanation is that learning during a task depends upon the integrity of an implicit anticipatory/ predictive mechanism, that may contribute to on-line awareness, which is mediated by frontal circuitry and diminished or abolished after neuropathology⁴⁰¹. PB might have progressed through this implicit learning. “Just because something is not registered in consciousness does not mean that it is not registered at all or that it cannot influence ongoing experience”⁴⁰² (p. 217). An individual, who demonstrates poor intellectual awareness at a verbal level, may have some awareness of a deficit that can be observed during task performance. Anosognosic patients indicate that they appreciate their deficits at some level of awareness even though they do not explicitly acknowledge them⁴⁰³. Clinicians need to be alert to observations that suggest implicit awareness. Areas of implicit knowledge can be capitalized on in treatment by either training tasks at a nonverbal or procedural level or by trying to make implicit knowledge accessible to consciousness²¹¹.

Finally, IP’s emotional instability may also have affected how he responded to metacognitive strategy training. Knowledge and beliefs about one’s capabilities are based on past experiences in similar situations²¹¹. Beliefs regarding one’s self are the products of experience, but they are also involved in the construction of experiences⁴⁰⁴. Beliefs and perceptions of one’s capabilities can create biases in the processing and interpretation of information that can enhance or impede cognitive functioning. They can influence activities one chooses to engage in, selection of strategies, what is monitored, the degree of effort and persistence, and commitment to goals. Thus, inaccurate beliefs regarding one’s capabilities can limit cognitive performance by their disrupting effects on self-monitoring and self-regulatory processes. Larson²⁴⁸ examined the relative contributions of negative affect and cognitive sequelae to performance monitoring dysfunction following severe TBI using event-related potentials. Negative affect was associated with decreased amplitude of error-related negativity that is thought to correspond to implicit error detection. Cognitive sequelae predicted positive deflection amplitude that is thought to reflect conscious error detection. In

another study Larson⁴⁰⁵ found that impairment in performance monitoring had a disproportionate effect when negative affect is overlaid on severe TBI (event-related potential results revealed disproportionately smaller error-related negativity amplitudes in participants with TBI relative to controls as a function of negative affect). In future, negative affect and more generally self-perception and task orientation^{406,407} should be addressed during training in order to maximize intervention trainings.

FACTORS IMPEDING METACOGNITIVE STRATEGY USE

Based on data from these case-studies and from literature on awareness, metacognitive strategy use, and metacognitive and EF development, we identified potential causes impeding children to use effectively metacognitive strategies in daily life. These are presented in table 8.6 together with possible factors contributing to failure at each step and possible ways to remediate them that would need to be explored in future research.

TABLE 8.6: Hypotheses for potential causes impeding children’s use of metacognitive strategies (MS) in daily life and possible ways to remediate them

Necessary requirements for effective MS use	Child	Factors contributing to failure of the requirement	Potential useful principles to remediate to the failure of the requirement
“I’ve been confronted with my problem” ⁶	IP	Lack of knowledge Young age	-Parents information: parents may not realise their child who sustained an ABI is more impaired than peers and therefore not realize that they give more support to their child than to healthy children -Child confronted to situations parents usually manage for him: high levels of support usually provided by family and professionals to ensure that the child experiences success at all times might in turn prevent opportunities for the child to develop awareness ⁸³ .
“I’m aware I have a problem” (Crosson’s lower level of intellectual awareness)	YR	“Organic” Awareness, neurologically-based awareness	Low to high confrontational methods (see Ylvisaker for examples ⁸³): Ylvisaker et al presented awareness intervention methods ranging from low confrontation (e.g. self-evaluation, daily conversational interactions, peer teaching...) to high-confrontation (e.g. presentation of low test results, self-observation of video-tapes performance on a difficult task). They emphasized that interventions should be chosen according to child’s age, emotional fragility, consequences of unawareness and the resources available in the family.
“...And I realize its consequences” (Crosson’s higher level of intellectual awareness)	PB	Systematic corrections from environment, minimising of the consequences	-Discussion about <i>the consequences</i> of cognitive failures -Confrontation to consequences instead of systematic correction of failures before consequences appear.
“I accept that I have a problem “	YR	Denial	Psychological support
“I understand a metacognitive strategy technique”	CS	Weakness in vocabulary, reasoning, global intellectual weakness	Training basic cognitive skills: Knowledge base, Vocabulary before trying higher-order interventions such as MS trainings (see the “Pediatric Neurocognitive Intervention model” ⁴⁰⁸ for a practical hierarchy of interventions to use)
“I recognize the metacognitive strategy as useful”	IP, PB	Examples and practice on decontextualized paper-and-pencil exercises, without sufficient repetition for utilisation deficiency to reduce.	Practice on meaningful, child-chosen tasks ²⁸⁶ Repetition for Implicit/Procedural learning ³⁹⁸ Enough practice to gain some automaticity in the strategy and to gain effectiveness
“I recognize task similarity”		Little experience (young children)	Train for Transfer on many different tasks, in different contexts (school, home) with explicit generalization training ³⁵⁵ .

TABLE 8.6: Hypotheses for potential causes impeding children’s use of metacognitive strategies (MS) in daily life and possible ways to remediate them (continued)

<p>“I remember to use the technique”/ “I initiate the MS”</p> <p>Automatic/procedural prompted versus Recognizing task similarity</p>	RK	<p>Memory impairment</p> <p>Initiation impairment</p>	<p>Content-free cueing ⁹⁷</p> <p>Prompts</p> <p>Linking an object with a plan ¹¹⁸</p>
<p>“I detect my errors”/ (On-line Awareness)⁴</p>	YR	<p>Error monitoring deficit</p> <p>Overwhelmed Attentional and EF resources</p> <p>Negative affects</p>	<p>Task Orientation training ⁴⁰⁷</p>
<p>“I can predict the task will be difficult for me” (Crosson’s anticipatory awareness)</p>	PB IP	<p>No habit of detecting task difficulty, thus no motivation for using MS compensatory strategies</p>	<p>Include difficulty prediction and review in all tasks ²⁶⁶</p>
<p>“I manage to focus both on the task and the strategy”¹</p>	PB CS	<p>Working memory limited capacity</p> <p>Totally unknown task increasing WM load</p>	<p>Increasing task familiarity</p> <p>Allow to practice MS without increase performance in task (i.e. allow time for utilization deficiency as it is short-lived) ²⁸⁸</p>
<p>“I like the task”/ “I’m motivated to do it”/ “I believe I can do it”³</p>		<p>Negative affect</p>	<p>Training task orientation (see Rath for an example in adults⁴⁰⁷)</p>
<p>I apply it correctly</p>	CS	<p>Executive functions impairments, mediation deficiency</p>	<p>Use the MS on simple tasks before proceeding to attention-demanding and novel tasks with unfamiliar materials</p>
<p>“I can adjust MS to the situation”</p>	CS	<p>Impairment in cognitive flexibility</p>	<p>Practice on various tasks, in various situations and contexts ²⁷⁷</p> <p>Include problem-solving and encourage strategic thinking</p> <p>Allow unpredicted situations during practice, occurring naturally in natural contexts instead of office-based protected artificial context</p>
<p>“I have time to apply it”</p>	RK	<p>Real life time constraints</p>	<p>Allow more time on task, self-paced study</p>
<p>“I have enough attentional resources to apply it”</p>	RK	<p>Effortful character of MS</p>	<p>Choose time-limited, important tasks rather than expecting metacognitive strategy use throughout the whole day</p> <p>Choose specific time for soliciting attention for MS use (e.g.: when child is left alone)</p>
<p>“I find the MS is fun”⁵ (Bjorklund’s criterion of the necessity of a strategy to be fun rather than useful in children)</p>	PB	<p>“Selection by pleasure”: only MS that children enjoy or that are not too demanding will be used</p>	<p>Allow the children to practice MS even if MS use is not followed by an immediate positive outcome</p>

TABLE 8.6: Hypotheses for potential causes impeding children’s use of metacognitive strategies (MS) in daily life and possible ways to remediate them (continued)

<p>“I can integrate my present performance with my past experiences”²</p>	<p>RK</p>	<p>Impairment in episodic and/or autobiographical memory</p>	<p>-Training episodic memory utilization to improve planning and EF⁴⁰⁹⁴¹⁰ (see the model of self-awareness in children²⁶⁰ in chapter 4) -Reinforce memory of successful strategy use and of failures without effective strategies by reviewing task performance and (for patients using routinely memory note books) making notes of successful task management.</p>
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The main limitation of this study is the lack of data on control children i.e. how would typically developing children have reacted to the Organisation task, and in particular to the prompt of making a plan before starting? It is not possible to conclude if the children in our study did not use the strategies because of their TBI or because of their age (i.e. it may not be age-appropriate to teach children metacognitive strategies).

CONCLUSIONS

Children in our pilot study seemed to have benefited very little from metacognitive strategy training. If the pGMT program has any effect, metacognitive strategy teaching is probably not an important active ingredient. This should be explored further by comparing typically developing children's response to a metacognitive training with our data - which could not be done here. Patterns of failure of strategy use, however, were similar to developmental data on younger children who show mediation and utilization deficiencies in strategy use, mainly because of limited WM space. This likely to be similar to the deficits in the children in our study who had impaired EF and WM, which makes them closer in cognitive abilities to younger children. Further research is needed to confirm our results and those of Missiuna²⁸⁶ that children with TBI do not benefit easily from metacognitive strategies and to identify why. Possible explanations presented in this chapter are only tentative and further research is needed to test those hypotheses, especially because some are based on the adult literature and may not apply to younger children. It is important to remember that children are not small adults and that adult interventions may not be developmentally coherent. "Although from an adult perspective the number-one reason to use a strategy is to improve performance, this may not reflect the child's point of view. Trying something new may be a goal unto itself, and the fact that it does not improve performance may be relatively unimportant to children"²⁸⁸.

CHAPTER 9: GENERAL DISCUSSION

SUMMARY OF FINDINGS

Part one of this PhD explored EF and related impairments after childhood severe TBI. Chapter 1 confirmed that EF dysfunction is worse when the injury occurred during period of rapid development of EF and showed that children injured 10-12, at a period of slower EF development, may be less vulnerable to the effects of the TBI. Chapter 1 also raised the issue of the ecological validity of EF paper and pencil tasks, which are known to underestimate EF impairment in daily life.

Goal Neglect: Chapters 2 and 3 confirmed impairment in prospective memory that persist at seven years post injury, and into adulthood (chapter 3) following childhood severe TBI. Similarly, to Duncan's model of goal neglect (GN), primacy of instructions impacted PM as instructions given second (make a fruit cocktail, tidy up the kitchen...) were more likely to be forgotten than the main instruction of making a chocolate cake (see chapter 2). Motivation seemed to impact PM as younger children were as good as adolescents and young adults in remembering a motivating PM task (entering a draw for winning an entry in an attraction park, chapter 3), although we did not compare it to a control condition (similar task but with low motivation). There seemed to be an age effect in typically developing controls (i.e. the older the children, the better they remembered to make the fruit cocktail and to tidy up after finishing the cake, see chapter 2), but this effect was not present in children with TBI, as those aged 14-20 performed close to typically developing controls aged 8-10. It may be possible, however, that goal neglect was influenced by poorer working memory and attentional resources that made the ongoing task more difficult for children with TBI, rather than a true PM effect (see discussion chapter 3).

Chapter 4 presented a model of anosognosia post-TBI in children, based on dissociated scores in three ways of assessing self-awareness. It proposed directions in cognitive rehabilitation

based on which component of self-awareness is impaired and alerted to the inappropriateness of metacognitive approaches in children who are too young or too anosognosic.

Part two of this PhD investigated ways to remediate EF dysfunction. First two literature reviews identified the best approaches for children (chapter 5) and reviewed evidence of the effectiveness of interventions based on Duncan's goal neglect model (GMT – chapter 6) in adults. GMT alone had no proof of effectiveness but it was effective when coupled with personal goal setting, external cueing or prompting to apply GMT to the current task, personal homework to increase patients' commitment and training intensity and ecological and daily life training activities rather than paper-and-pencil, office-type tasks. Based on this literature review, a paediatric GMT intervention (pGMT), was developed from the adult GMT, and drawing on Ylvisaker's "context-sensitive" approaches identified as an important factor for EF interventions in chapter 5.

Chapters 7 and 8 - the intervention component of this PhD- tested the efficacy of pGMT on EF, PM and on the adaptability to new EF-demanding situations. They showed a limited effectiveness of the intervention, with gains on some outcome measures but inconsistency across outcome measures (children improving either on the ecological CCT test or on EF questionnaires). Most importantly, it showed that progress after an intervention may be incorrectly attributed to a reduction of executive deficits, when in fact it reflects rather the learning of tasks that make them less demanding in terms of EF: children who made progress on the CCT, came back to their pre intervention number of errors and presented a similar dysexecutive behaviour when a parallel version of CCT, using a totally unknown recipe, was used. Chapter 8 explored further if metacognitive strategies (such as those in the pGMT) could be taught to children and proposed that it is necessary to take into consideration the stage of EF development, which may limit the ability of younger children to benefit from these types of interventions.

STRENGTH AND CHALLENGES OF THE PHD

This PhD used the largest (to our knowledge) cohort of children with severe TBI (n=65) to explore EF and PM. Larger cohorts of childhood TBI published so far usually include heterogeneous samples of children with a range of TBI severity, where children with severe TBI are usually relatively few compared to the rest of the group^{161,42,71,162,123,79,19,14,72}. Further, most samples focus on a restricted age range. Children in this sample were aged 0-15 years at injury, and were included prospectively immediately after uniformly severe accidental TBI. They were followed up seven years after TBI (for the PM assessment). PM assessment included ecological contexts and meaningful tasks. Using both performance-based tests of EF and ecologically-valid questionnaires to assess EF, EF scores were likely to be representative of true EF impairment. Age at injury effects were explored using age at injury periods rather than simplified linear relationships. The sample was representative of the TBI population in the Paris region, as children whose parents did not speak French were not excluded, contrary to other studies¹⁶¹. As part of non-French speaking and low education, parents are likely to have more difficulty to apply for help and understand counselling regarding their child because of language restrictions, it is important that those children are not excluded from trials.

Apart from PM explored in chapter 3, most results of the TG2 cohort still need to be exploited, which will give in the future valuable and unique information about long-term outcome specifically after severe TBI, which so far has been explored mostly in small samples. Further this study will provide unique and reliable information about transition into adulthood of children who sustained a severe TBI, as one third of the sample was aged eighteen or more at the seven year follow-up.

The intervention part of the PhD used a series of relevant outcome measures, that allowed a comprehensive evaluation of EF impairment before and after intervention. It used the N of 1 trial methodology that was adapted for a pilot study in a research domain where very little has been rigorously evaluated and proven to be effective. Outcome measures included creative ways to better reflect the complexity of EF, and especially the use of Goal Attainment Scales as a generalisation measure. The use of a parallel form of the cooking task, allowed capturing adaptation to novelty after intervention, which is rarely explored in EF rehabilitation studies,

while it constitutes the heart of EF. The use of two baselines and a blinded beginning of intervention allowed to better control for enthusiasm effects unrelated to the content of the intervention. The intervention included a range of activities chosen to promote generalisation, both paper and pencil exercises and more ecological and meaningful activities. A big effort was made to involve school staff and families in the training (although it proved difficult). It was relatively long (15-20 hours). It probably included the most challenging population for rehabilitation, i.e. children with severe TBI, sustained at a young age (and at a rapid developmental stage of EF), presenting with a severe dysexecutive syndrome (as reflected by the pre intervention BRIEF scores), experiencing extremely complex family situations. The comprehensive assessment of children's self-awareness and use of an external (unbiased) investigator to assess family and school staff perception of the intervention and compliance allowed to better explore factors limiting intervention success.

Finally, this PhD opened a series of future possible research, especially on themes that remain under-explored in childhood TBI, such as self-awareness, prospective memory and adaptation to novelty. This work also highlighted the major importance of everyday implication of families and school staff in children's rehabilitation.

DEFINING WHAT IS AN EFFECTIVE INTERVENTION

Part 2 of PhD examined the general question in rehabilitation: what is an effective intervention? We identified five criteria for intervention effectiveness, of which most were addressed in chapter 7. An effective intervention is one that shows:

1. Effects in daily life, in meaningful, ecological activities (i.e. not only on paper and pencil tests disconnected from the patient's reality). This implies the need to use ecologically valid tests, collaborative goal setting, and requires the family to judge the success of an intervention. Of particular usefulness in this PhD were tools such as Goal Attainment Scaling^{411,412,413}, and ecological tests¹⁸⁰ such as the Children's Cooking Task^{121,120}.
2. Maintenance of effects at long-term follow-up (with or without on-going support, booster sessions). Long-term follow-up is essential in children, as a function that was not mature at the time of intervention may mature differently because of the

intervention. The TGE 1 (chapter 1) and TGE 2 (chapter 3) studies showed how ongoing cognitive development is affected in the long term after childhood TBI and how consequences are not predictable. Therefore, in children particularly, the effectiveness of an intervention should be assessed in the long term (i.e. more than the six months post-intervention used here). Furthermore, an intervention may seem effective immediately after the training, while in fact it accelerated the development of a cognitive skill that would have developed anyway. For this reason, effectiveness should be compared in relation to usual care and education in the long term. Finally, there is the possibility that the intervention displaces another more beneficial activity, that would have helped cognitive development (e.g.: sports, physical activity^{319,322,323,318-321}, traditional martial arts³²⁵, special school curricula^{310,311,312,313,314}).

3. Generalisation to untrained tasks requiring the same cognitive skill (e.g. does practicing stepwise processing and use of a checklist to prepare school bag generalise to preparing suitcase for a holiday OR does learning to make a chocolate cake generalise to another recipe²⁰⁰, as in the GMT intervention with a parallel form of CCT).
4. Transfer to the natural contexts of the child. Progress seen in the rehab centre should be seen at school and at home and not just in the office-based rehabilitation session. Chapter 8 compared the use of metacognitive strategies during ecological activities and during office-based paper-and pencil tasks and found important discrepancies and difficulties applying the strategies in real word situations, which was confirmed for most children by teachers and parents, using context-specific questionnaires (BRIEF teacher and parental forms).
5. Specificity of treatment effects. Although generalisation is an aim, interventions also need to show some specificity, in order to prove that gains are not due to general cognitive stimulation, but to the key ingredients of the intervention. Any stimulation of a child may produce some gains in EF compared to no intervention. In order to explore what is effective (and what should be given priority by government funding and priority in the children's busy timetables), future research needs to demonstrate

specific effects that are superior to general stimulation. Specificity of the intervention was not assessed, although it would have been possible to include, for example, a retrospective memory task that was not expected to show any progress.

CHALLENGES IN COGNITIVE INTERVENTION RESEARCH

Research on EF interventions for children faces a number of challenges that have only partially been addressed in this PhD.

First, some challenges are relevant to cognitive rehabilitation in general: the lack of validated and reliable outcome measures, or insufficient responsiveness of measures, lack of availability of recommended tests¹⁵² in French. These problems were partially overcome by the use of composite EF scores in chapter 1, by transforming all scores into Z-scores but some of the tests used did not have evidence of validity/reliability and the conclusions of the TGE1 study should therefore be interpreted with caution.

Second some additional challenges are specific to a paediatric population:

- insufficient age range of most tests, requiring use of different tests that makes overall comparisons with studies in the adult literature difficult.

- heterogeneity of the paediatric population making it very difficult to match children with controls in a RCT (i.e. EF evolution dependent on age, age at injury, family functioning, associated deficits, severity of injury.).

- children's busy timetable that requires caution in scheduling time-consuming interventions that may displace other valuable activities, socialization opportunities, or rest for children experiencing high fatigability post TBI.

- difficulty in obtaining permission to recruit healthy controls for comparison. This was a particular issue for the awareness study in chapter 4 and in the pGMT intervention in chapters 7 and 8, as we did not manage to compare children with TBI to typically-developing controls. In the TGE 2 study (chapter 3), controls could be recruited, which was very helpful in interpreting the results.

Finally, some challenges relate to EF. Most EF tasks involve complex, overlapping and multi-faceted functions that tap both executive and non-executive processes¹⁶². Isolating the influence of these processes is difficult^{20,122} and “purely executive” tasks do not exist¹⁶². Therefore, it is always difficult to decide if the progress observed post intervention is truly

“executive”. Conversely a deficit or lack of progress may be impacted by the patients’ impaired general cognition (general intellectual skills, visual-spatial processing, processing speed, memory). In children especially (and in adults too ^{39,40}) intelligence is inextricably linked to EF³⁶⁻³⁸ and it is therefore difficult to appreciate how much other functions are impaired because of EF deficits and how much EF are impaired because of impaired general cognition. In this PhD it was decided not to control for IQ in the EF tests, contrary to other studies¹²⁵ (given the overlap in constructs and hence shared variance between tests of IQ and tests of EF, controlling for IQ would potentially remove a lot of variance that one might also associate with executive functions).

SCED METHODOLOGY TO EXPLORE INTERVENTION EFFECTIVENESS

Single-Case Experimental designs (SCEDs), used in this PhD in part 2, use repeated, systematic measurement of an outcome variable within and across different conditions and sequential (\pm randomized) introduction of an intervention. Each single case provides its own control for the purposes of comparison thus avoiding the issue of unmatched control patients. Sequential multiple baselines allow documentation of the intervention effect (consistent change in level after intervention is introduced), specificity (lack of change on non-target measures, excluding therefore a general recovery hypothesis) as well as potential generalization effects (in related although not directly trained behaviours). Further they allow us to analyse differential responses to treatment, and factors influencing outcome (changes in level not directly related to intervention). SCEDs are now considered as level 1 evidence by the Oxford Centre for Evidence-based medicine, and hopefully the future will see many well designed SCEDs in paediatric cognitive interventions instead of costly, controlled trials using heterogeneous groups, unevenly matched groups and of insufficient power.

The Single-Case Experimental design in this PhD allowed an in-depth study of intervention effect and of factors mediating effectiveness: comprehensive assessment of self-awareness during the intervention, observation of metacognitive strategy use in different types of exercises, comparisons of questionnaire-reported executive gains versus performance-based EF in ecological (CCT) and paper and pencil (FEE) objective tasks. Such comprehensive assessment is not possible using large RCTs, where the outcome measures are usually few

and often consist of questionnaire that is quicker and less costly to administer (for examples see Wade et al.^{299,414}, Kurowski et al.²⁹⁸). When conducting a pilot intervention SCEDs are particularly useful, in order to screen the intervention for effectiveness, for factors influencing this effectiveness and to better adapt the intervention before eventually running a larger costly trial¹.

LIMITATIONS OF THE PHD

Limitations of each study are discussed in the corresponding chapters. More generally, the main limitation of this PhD is the preliminary nature of the findings (pilot data only) for the intervention (chapter 7 and 8) and the self-awareness component (chapter 4) that could not be explored further in a larger group or replication study, within the timescale for completion of this PhD.

The Anosognosia model especially, proposed in chapter 4, was based on a very small number of cases (N=5) combined with an extensive review of the literature. Therefore, firm conclusions as to the degree of anosognosia in children due to the TBI cannot be drawn at this stage because of unavailable data on developmental immaturity of typically developing controls matched in age. Therefore, the model is tentative but nevertheless allows to clinicians to have a broader view of self-awareness component processes and assessment tools other than just questionnaire discrepancy scores. This important piece of work provides a strong rationale for further research, which could investigate self-awareness in a large study using the Children's Cooking Task. Especially the steps needed to gain awareness (see figure 4.3) could be compared between children with TBI and typically developing age-matched controls, to investigate in which steps children with TBI experience difficulties, and therefore which steps should be trained.

In the light of the results of part 2 of the PhD and of the literature review of EF interventions, adapting an adult intervention (GMT) to children was not a developmentally optimal approach. It allowed, however, to precisely point the importance of providing interventions

¹ We are grateful to Pr. Alain Yelnik for suggesting to run this pGMT pilot study before a controlled trial, when he reviewed the pGMT intervention ISPEN-SOFMER grant.

that are coherent with the child's developmental stage and with family needs. Although adapting an adult intervention to children is often used in neuropsychology, a better approach could be to adapt and use interventions developed in other fields (psychiatry and learning disabilities especially).

More collaborations with specialists of developmental disabilities (e.g. ADHD, autism) is needed with the aim of adapting existing effective EF interventions from these conditions to children with TBI rather than adapting adult TBI EF interventions to children. The latter carries a high risk of being developmentally inappropriate, irrelevant or ineffective. There are several promising methods in pedo-psychiatric literature with children with developmental disabilities: Cogmed ©, "Unstuck and on Target" are examples of intervention that could be adapted in the future to children with TBI.

METHODOLOGICAL LIMITATIONS

LIMITATIONS IN THE WAY THE SCED METHODOLOGY WAS USED

In this PhD, SCED methodology lacked some of the quality criteria that have been published since the study was performed (see "RoBiNT" scale⁴¹⁵ or CONSORT extension of N of 1 trials, "CENT"^{342,416,417}):

The design would have been methodologically stronger if the onset of the intervention had been staggered (i.e. producing different baseline lengths for each child) and the starting date randomized across subjects. In the pGMT study, all children began the intervention at the same moment i.e. after 4 weeks of baseline, meaning that PM performance may have improved because of task learning, or due to an external factor that occurred after the 4 weeks of baseline, rather than specifically as a result of the intervention. Sequential randomized introduction of the intervention would have controlled for these confounding effects.

Intervention fidelity was not monitored. Although the use of PowerPoint slides ensured the program was delivered in the same way to all children, the amount and type of support during exercises was not precisely defined prior to the intervention, and we did not precisely specify use of learning methods such as errorless or errorful learning (that have been shown since our study was published to influence GMT effectiveness in adults¹⁰¹).

The study used simple quasi-statistical methods to interpret effects (split middle trend lines, two-standard deviation bands, non-overlap of all pairs). In recent years a number of alternative methods of statistically analysing SCED data have been developed and could have been used, though the results would be unlikely to be different given the pattern of data obtained.

OUTCOME MEASURES

Choosing reliable and valid outcome measures is extremely important but challenging, and most chapters of this PhD reflect this issue. The poor psychometric properties of performance-based EF test used in chapter 1 did not allow demonstrating an impairment in EF while all scores and sub-scores of questionnaires showed important impairment and lack of recovery at 2 years post TBI. Similarly, in chapter 2, the Rivermead Behavioural Memory Test, did not differentiate children with ABI from typically-developing controls (based on norms), while their impairment on the ecological test of EF (the CCT) showed an important impairment, with older children and adolescents with TBI performing like young healthy children. The self-awareness study that used three different ways for assessing self-awareness found dissociated scores and different awareness profiles that would have been missed if the classically used approach had been used (i.e. discrepancy score on a questionnaire rated by the patient and a proxy).

All these are examples of the difficulty of measuring EF-related impairment, and highlight the possibility of erroneous conclusions if outcome measures are not chosen carefully. Particularly striking was the lack of sensitivity of NEPSY-1 tests of EF (used in chapter 1) that were (until 2 years ago) the main EF tests used for assessing EF in France, while the psychometric properties (and especially discriminant validity) are poor or unreliably explored in the NEPSY-1 manual. This may be why, fortunately, a second edition of NEPSY has been published recently.

This PhD tried to overcome those challenges and to develop and use novel or original ways of assessing EF:

- (1) Use of paper and pencil tests of EF based on precise neuropsychological models that take into account confounding factors: the new battery of EF in French developed by

Arnaud Roy and colleagues (“Fonctions Executives Enfant” –FEE⁴¹⁸) was used in the GMT intervention to better characterize children’s EF impairments, although an improvement on paper and pencil task was not expected and although the paper-and-pencil tasks of FEE alone lack evidence of ecological validity (so the FEE battery includes the BRIEF). The FEE battery has been carefully designed and tested by the FEE group and has a much stronger scientific base than the classical EF tests that were used in the TGE1 study (chapter 1). For example, FEE’s Stroop tests takes into account children’s speed of processing (i.e. speed of colour naming) and reading ability (i.e. speed for colour names reading) to adequately infer the interference (Stroop effect) produced by asking the child to state the ink colour of colour words (e.g. The word BLUE written in red colour should be stated as RED). The classical Stroop tests, used in TGE1, based on the adult Stroop tests on the other hand, did not take into account reading difficulties or slowness in colour naming: children with poor reading abilities had therefore less difficulty to state the ink colour, as they had less difficulty inhibiting the automatic reading of colours (impairment underestimated) or on the contrary, were slow both for reading and naming and their Stroop effect was confounded by impaired speed of processing (overestimating impairment).

- (2) Use of ecologically valid tests, both performance-based (such as the CCT) and questionnaire-based (such as the BRIEF or DEX-C questionnaires): the advantage of ecological performance based tests is to measure directly and objectively EF performance on tasks, and obtain valuable qualitative data (see chapters 4 and 8). But the drawback is the time needed for administration (45-90 minute for the CCT). Questionnaires on the other hand do not require therapist time for administration, are quick to obtain and refer to a range of activities (at school, at home, during leisure time) not assessed during a performance based test, but they require parents to be fluent in French (which was the main difficulty in the TGE1 study, see chapter 1). They may lack objectivity and be highly dependent on enthusiasm effects in novel interventions (as seen by some children improving between the two baselines in the GMT intervention, because parents where not informed that the intervention had not started yet). They may also be influenced by personal factors of the person answering the questionnaire, such as anxiety and depression. Most of all, they are highly

dependent on proxy's willingness to answer questionnaires, and ability to answer accurately (in the pGMT study some teachers did not return the questionnaires, some questionnaires could not be used because the inconsistency of scores, reflecting contradictory answers, was too high). The strength of the pGMT intervention was to have used both approaches. The contradictory results (most children showing progress either on questionnaires or on the CCT) means that it is difficult to determine the true effectiveness of the intervention.

- (3) Taking into account the novelty aspect of EF: EF outcome measures need to be novel to really capture EF^{165, 166}. When the same task is repeated after intervention, it is more “familiar”, which can make it less demanding on EF³⁶⁷ 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. Familiarity effects increase when patients are tested on several occasions (e.g.: pre, post, follow-up...). As the whole purpose of EF tests is to prevent lapses into automaticity and promote conscious, novel and effortful processing^{165, 22}, apparent progress after EF interventions may not necessarily reflect changes in underlying executive processes. In the pGMT intervention, children may have improved on CCT because cooking a chocolate cake became less novel and therefore less demanding on their EF, as they repeated the test five times (two baselines, immediately after intervention, at three and six months). For this reason, we used another recipe, “Christmas biscuits”, designed to be a parallel form of the Children’s Cooking Task (CCT) for the at 6 month assessment, involving the same number of steps and ingredients but requiring different types of ingredients and procedures. Although we did not explore the equivalence of difficulty of both recipes and although this parallel form has no established psychometric properties, it allowed us to observe children’s reactions and use of metacognitive strategies, when faced with a new situation. In the pGMT study, it showed that faced with a new situation, most children did not use metacognitive strategies taught during the intervention, and presented with a severe dysexecutive syndrome (with purposeless actions, perplexity, difficulty in problem solving and lack of flexibility) similar to their pre intervention CCT performance. Even parallel forms of the same test may not overcome the novelty problem as the

content may be new but the format is not³⁶⁷. For this reason, we then worked on another parallel form of CCT that was explored in Christine Alzieu's Masters thesis⁴¹⁹, directed by the PhD candidate, that used the same presentation, number of steps and number of different objects, but not food. In this parallel version children had to make a rain stick and a maraca (two percussion instruments). Errors were counted in the same way as CCT and pilot preliminary data for typically developing children and children with high functioning autism suggested it could be a potential parallel for the CCT.

- (4) Use of Goal Attainment Scaling to measure generalization: Most often, GAS scales are set for goal-focused rehabilitation where the goal is directly trained and GAS represents the degree of progress towards a goal. In this PhD, GAS was used as a measure of generalization²⁰⁰: i.e. after a training relating to a cognitive EF, GAS scales assessed if training lead to gains in daily life (e.g. GAS relating to activities relying on executive functions such as preparing a schoolbag, putting things in order in one's room), without specifically addressing these goals. A valuable approach in the future, could be to use both a set of trained goals (and corresponding GAS) and a set of untrained goals (and corresponding GAS) and then to focus the intervention on training the former while using the latter as an untrained ecological generalization measure. Further it has been proposed that "control" goals³⁸⁰ (and related GAS scales) that are *not* expected to show progression are used, in order to demonstrate the specificity of an intervention (i.e. the client does not just progress on all goals because of general cognitive stimulation or goal-driven motivation, but progresses on the specific goal that is trained or that relates to the trained function needed to achieve an untrained generalization goal).

GAS is a controversial outcome measure in clinical trials. Some authors argue that GAS cannot be used as an outcome measure in studies testing the effectiveness of an intervention because of a series of methodological problems: (1) unknown clinimetric qualities of GAS scales used in a given study due to their idiosyncratic nature⁴²⁰; (2) subjective scoring, especially if not all levels of the scale are formulated or if descriptions are not precise enough; (3) risk of choosing goals that are not clinically relevant or too easy/too difficult to attain⁴¹³

and therefore do not represent a meaningful or realistic change in function; (4) ordinal (rather than interval) nature of GAS scales⁴²¹ and the lack of equidistance between GAS levels which cannot be controlled for⁴²²; (5) the use of a T-score that uses subjective values, especially a subjective weighting of GAS scores and a ρ coefficient assumed to be 0.3 which has not been confirmed in the literature^{423,421,380}. A major drawback of GAS methodology is that it is highly dependent on the ability of the GAS setting team/person to generate valid, reliable and meaningful scales. It has even been proposed that GAS is more a measure of how adequately a therapist can foresee outcome than an outcome measure itself^{380,424,425,426,427}. A trial result may depend more on how GAS were written than on the effectiveness of the intervention itself. As suggested by Ruble et al.: “If GAS scores are higher in the experimental conditions [...]one could argue that the targeted outcomes as scaled using GAS were less difficult and easier for [clients] in the experimental group to achieve compared to the control group; that skills were written in more measureable terms and thus easier to be observed and coded in the experimental groups; or that the intervals between each scaled description were unequal and favoured the experimental group.”⁴¹³ (p3).

Some authors, on the other hand, proposed recommendations for GAS when it is used in research: (1) including a GAS training program^{428,420,429,430}; (2) establishing all goals prior to randomization^{431,432} or blinding the goal-setter to the patient’s treatment/control status⁴³³; (3) testing of interrater reliability for initial and post intervention GAS rating^{428,432}; (4) GAS scoring by a blind examiner⁴³² who is independent from the team that set the goals^{434,435,432,436} and independent from the therapist providing intervention^{428,430}; (5) the use of “control goals” that are not targeted by the intervention³⁸⁰; (6) evaluation of the patients on two different GAS scales developed by independent therapists (i.e. treatment success must be independent of how the goals were formulated)^{376,437,438}; (7) goal-setting by a group (rather than a single therapist or the patient alone), in order to avoid overly simple or unrealistic goals⁴³⁹.

Based on these criticisms of GAS, and on the reviewer’s suggestion during the reviewing process of chapter 7 (the pGMT intervention), we proposed recommendations for GAS use in research⁴⁴⁰(see table 9.1), that were published in Archives of Physical Medicine and Rehabilitation in January 2016. In relation to those recommendations, there were some limitations in the way that GAS was used in the pGMT intervention. There was potential for

examiner bias with the persons implicated in the intervention completing the ratings (as families and schools were expected to act as cognitive coaches). Importance/relevance, difficulty, unidimensionality of goals, their specificity to the intervention content, the equidistance of GAS levels was not assessed by an external judge. Measurability was not formally assessed but was not strong given the questionnaire-based rating (rather than performance-based measurement of goal attainment). Neither inter-rater nor intra-rater reliability were evaluated. T-scores were used while the GAS scales cannot be considered as interval data.

However, some recommendations were met: Goals were set collaboratively with the child's everyday people (family, school assistants, and teachers). All five levels were precisely described for the rating. Most goals corresponded to activity and participation domains and goals were included in the published paper (although no full GAS with the five levels was included). Definitions of levels were verified with parents and schools and readjusted after the first baseline (although the readjustment was not based on an objective, performance-based, verification of initial level regarding the goal but based on family and school appreciation of current level on the goal). A time-frame for attaining goals was specified as immediately post intervention (but aiming at maintaining effects at 6 months). Researchers were aware of potential biases in GAS scales and therefore issues of unidimensionality, specificity and relevance of goals were taken into consideration (although not checked externally).

TABLE 9.1: Goal Attainment Scaling (GAS) methodology quality appraisal for rehabilitation efficacy studies⁴⁴⁰	
Quality appraisal items	Item description
Content Validity	
Collaborative goal setting	The client/family is included in goal selection, when possible and appropriate. If goals are therapist-chosen, they rely on a comprehensive assessment (and when possible, a client/family interview), identifying key target domains for intervention.
Relevance/importance	GAS scales have been verified by an external judge to check for the relevance of chosen goals and to check if GAS levels represent clinically meaningful change.
ICF classification of goal types	GAS themes correspond to functional domains. Authors report the ICF domain the GAS relate to.
Specificity	GAS scales have been verified by an external judge to check for specificity to the aim of the intervention. If GAS is used as a generalization measure to untrained goals, GAS should be specific to the function the intervention is aiming to improve.
Reliability	
Reliability of scale construction	
Equidistance of levels	GAS scales have been verified by an external judge to check if difficulty from one level to the next is roughly equal.
Pre-intervention performance	Pre-intervention performance has been verified and corresponds to initial level described in the scale. Pre-intervention score is comparable across groups (same number of clients starting from -2 and -1).
Attainability/ difficulty	GAS scales have been verified by an external judge to check for their difficulty/attainability.
Time-specificity	Authors specify if/how longer-term goals were adapted to the specific time frame of the research study. In the case of multiple assessment, authors specify which assessment was taken as the target moment for goal achievement.
Reliability of scale rating	
Inter-rater reliability (IRR)	Inter-rater reliability of GAS scales is reported.
Criteria affecting IRR	
Precise description of all levels	Five GAS levels have been precisely described pre-intervention for each scale.
Measurability	GAS scales have been verified by an external judge to check for measurability. Subjective and general goals are transformed into more objective and measurable goal attainment indicators.
Unidimensionality	GAS scales have been verified by an external judge to check for unidimensionality.
Context of measurement	Context of performance measurement is clearly defined (prompts, cueing, support, amount of help/guidance, location...) and is controlled for during GAS rating. OR Changes in context are carefully manipulated across the GAS levels, with one change per level at a time ⁴¹¹ .
Other criteria	
Training	Researchers setting the GAS with the client and verifying GAS have received training in writing GAS, have practiced GAS writing, are aware of potential sources of bias in GAS, and are experienced in the goal domain/population.
Examiner bias	The person scoring the GAS at the end of the intervention is independent from the team who set the GAS (and independent from the team that provided the intervention although the latter is not a GAS specific criterion).
Statistical analysis	Ordinal nature of GAS scales is preserved using non parametric statistics (rank tests, medians, boxplots).
Example of GAS	One (or more) example of a typical GAS full scale, extracted from the trial, is provided. A list of chosen goals is reported.
Criteria are grouped in accordance to the clinimetrics they mostly impact, although some items may impact both reliability and validity. If GAS is used in a controlled trial, GAS scales should not only be checked but also compared between groups, similarly to the methodology proposed by Ruble et al. ⁴¹³ For large trials, we propose (as for IRR) that at least 20% of GAS scales be checked. <i>ICF: International Classification of Functioning; IRR: inter-rater reliability</i>	

CHALLENGES IN USING GAS IN RESEARCH

Appreciation of goal attainment by clients may be subjective and is affected by a series of factors (self-awareness, denial, memory, high involvement in goal pursuit) that can bias the perceived attainment of a goal. On the other hand, GAS was always intended to be a person-centred measure and the risk of insisting upon an observable and measurable goal (see Discussion Table1) is to exclude the family's appreciation of goal attainment. The challenge for clinicians and researchers is therefore to understand and analyse the child's (and/or his/her families) goal and transform it into an observable, objective, performance-based measure that can then be used to discuss goal attainment with the client. For example, a general goal of "improving my memory" expressed by a client can be transformed into a GAS scale that measures memory functioning in real life situations agreed with the family (e.g. number of medicines taken on time without prompting; number of failed-to-deliver messages from school in one week). Scoring GAS based on simple interview or patient-filled forms should be avoided when possible. Creativity should be used to link subjective goals in difficult domains to more objective goal attainment indicators. For example for a GAS scale on anger management or use of social skills, rather than asking the client to recall how often she/he felt he could cope with his anger or effectively use social/language skills^{423,441}, an effort should be made to choose indirect indicators of goal attainment such as a review of controlled versus overt anger at the end of a day by the family rating of social/language skill (how many times he/she initiated/contributed to conversation or was understood) after regular naturally occurring events (outing, family dinner). It is probably the most challenging part of GAS methodology but it has been demonstrated that it is feasible to have GAS that are both person-centred in the choice of goal and objective in the formulation of GAS scales (see Steenbeek et al. for an example in motor domains⁴³⁷). Literature (and especially SCED-literature) offers growing ingenious methods for assessment of domains not directly accessible to classical performance-based objective measures. These methods include use of smartphone reminders to self-assess goal attainment at regular periods to decrease memory bias (see⁴⁴² for an example), use of naturally occurring situations monitored in real life by family/proxy to decrease self-awareness bias (see⁴⁴³ for an example), use of objective behavioral measures that are thought to reflect the underlying psychological (e.g.: happiness⁴⁴⁴) or cognitive (e.g.: functioning at school^{290,291}) target goals. A reasonable compromise between scientific rigor

and person-centred approach could be to have for each child at least one ecological patient-centred GAS that could be interview-/questionnaire-based (with the risk of being less reliable) and at least one performance-based GAS (with the risk of being less ecological). In all cases, Kiresuk, who first proposed the GAS method, recommended to “anchor scale points with behavioral or other evidence that will be meaningful to the client and readily scored by the rater” (p 31)⁴⁴⁵.

FACTORS MEDIATING EFFECTIVENESS THAT COULD APPLY TO NEW INTERVENTIONS

Specific factors limiting the pGMT intervention effectiveness are reported in the relevant chapters (7 and 8). More generally, the following factors mediating effectiveness were identified that could apply to any new intervention in the future:

- (1) Younger children (before 13 of age at least) may not have developed sufficient metacognitive skills to benefit from the truly “metacognitive” part of the training (i.e. they could be taught to use systematically or when prompted some strategies such as that the “STOP-STATE your goal-SPLIT-CHECK”, but they were unable to truly analyse their cognition and anticipate the need for compensation for their executive dysfunction spontaneously). The question remains to determine if they were unable to do so because of the cognitive consequences of their TBI, because of their age or both. Only future studies including typically developing controls of various age could clarify this important point.
- (2) Contrary to adults needing good awareness to benefit from metacognitive strategy training, the main requirement for children was to have fun while learning to use the strategies, irrespective of their success. Making interventions fun is key for effective EF rehabilitation.

- (3) The cognitive coaching guide for parents and school was inappropriate. It was not read, some parents lacked the capacity to understand the strategies and foresee their possible benefit in everyday life. Most families had challenging problems to deal with, on top of their child's cognitive and behavioural problems. All families reported the lack of time or energy to read the guide. Making regular phone calls to parents, teachers and school assistants may have been more beneficial for the intervention effectiveness, and may also have reduced parental distress and provided support to school staff, which are often overwhelmed and misinformed as to the reasons of the child's difficulties (especially challenging behaviours). Face-to face interviews may have been also useful, but given the families busy time table, home visits may have been the best option. More modern format interventions such as web-based programs^{298,300}, on-line coaching with hands-free kit²⁷⁰, or smartphone apps may be more enjoyable and more motivating for families. Written supports are perhaps outdated. The format of interventions may be as important as the content itself.
- (4) Intervention duration, although quite long, was probably insufficient. To allow children to automatize the metacognitive strategies, a lot of repeated practice is needed which is feasible probably only if undertaken by schools and families. This emphasizes the need to better implicate families in the children's rehabilitation care, in a format that would be perceived as useful and motivating by families, by addressing their needs before proposing any kind of specific cognitive coaching.
- (5) Ylvisaker proposed that "targeting EF goals throughout the day in the context of everyday activities and routines is better than targeting these goals in highly specialized therapy or instructional periods."²⁶⁶ (p 243). Therefore, the usual format of individual sessions is probably inappropriate and research should rather move towards sessions at school, in the classroom, using usual exercise and teaching school staff to teach and use the strategies in everyday activities. It is interesting to note that EF support that has been proven to be effective in children with TBI^{289-291,291,446} at school has found enthusiasm among teachers who began to use them with the rest of the classroom as well.

(6) Determining rehabilitation priorities based on children’s associated deficits (e.g. CS poor vocabulary, IP’s end YR’s behavioural problems) and family priorities (more security for RK) is inescapable. The Pediatric Neurocognitive Intervention (PNI) model offers a useful framework for planning interventions (see figure 9.1). The pGMT intervention is, according to the PNI model, a level C/D intervention, while most children perhaps needed more basic interventions first to address the psychosocial and systemic foundations (see table 7.1 showing the difficult family situations and challenging behaviour that needs support first). In the PNI model, core executive skills such as inhibition, sequencing and working memory should be addressed first, which is consistent with developmental successive maturation of EF (see Diamond’s model in introduction). CS for example had important sequencing impairment and therefore planning exercises such as putting in order steps to organize a birthday party was impossible. More generally her poor working memory, vocabulary, semantic knowledge (and poor overall intellectual capacity) did not allow her to understand the strategies, the exercises and to practice both the task and the strategies (she did not have enough mental resources to concentrate on both).

Figure 9.1: Pediatric Neurocognitive Intervention (PNI) Model⁴⁰⁸

	Level	Cognitive Impairments	Intervention Aim	Examples of Interventions
<p>Level D Skills & Interventions</p>	D	Specific cognitive skills e.g. episodic memory, visual processing, language skills	Compensatory strategies to be used independently	Training in the use of e.g.; elaborative encoding, retrieval strategies, visual compensations
<p>Level C Skills & Interventions</p>	C	Evaluative skills e.g. metacognition, supervisory processes, and reasoning	Training to support general cognitive functioning	Training of e.g. goal management skills, prospective reminding, "stop and think"
<p>Level B Skills & Interventions</p>	B	Core skills e.g. working memory, inhibitory control, processing speed, and sequencing	Remediation of skills	Intensive practice e.g. working memory, attention process, and speed training
<p>Level A Skills & Interventions</p>	A	Semantic knowledge, adaptive functioning and specific cognitive skills (e.g. episodic memory)	Compensatory strategies, cued and supported by others	Providing techniques such as precision teaching, errorless learning, elaborative encoding and rehearsal
<p>Psychosocial and Systemic Foundations</p>	<p>Psychosocial and Systemic Foundations - Supporting health needs, sensory impairments, pragmatic and social care issues (e.g. visual processing, diet, exercise, financial, and practical resources). Addressing systemic factors (e.g. family chaos). Ensuring positive behavioral support for challenging behavior. Accessing parenting skills training to ensure development of emotional competence. Providing psychotherapy for mood disorders.</p>			

PERSPECTIVES AND FUTURE DIRECTIONS FOR PAEDIATRIC EF REHABILITATION

Although this PhD allows only tentative recommendations due to the pilot nature of the intervention data (chapters 4, 7, 8), the following recommendations are proposed based on literature reviews (chapters 3 and 4) and the pilot data:

1. There is evidence to encourage collaboration of neuropsychologists with schools, training of school team and of parents in understanding and intervening with EF, while it is less clear that engaging children in EF interventions decontextualized from the school and home environment is effective ^{277,326}.
2. There is growing research on family involvement in cognitive rehabilitation ^{267,269,447, 299,384}. Targeting the family at some level of the intervention seems necessary, ranging from giving family support ⁴⁰⁸ to actively engaging the family in cognitive rehabilitation²⁶⁷. However this may be challenging and not all families are able to engage²⁰⁰. In all cases, it is important that family priorities and goals are met, that intervention-related burden is evaluated and that it is not greater than perceived gains.
3. Promising programs that offer support to families in order to improve daily EF have been published and translating these into French and adapting them to the French culture may be a reasonable option (e.g.: TOPS, CAPS and I-Interact Programs in chapter 5)
4. The PNI model (se figure 9.1) should be used more systematically, not only in clinics but also in research (e.g. a patient's basic unmet needs should be an exclusion criterion for higher-order EF intervention).
5. Adapting interventions from developmental disabilities to children with TBI is more likely to be of benefit than adapting adult interventions to children. Developmental coherence of interventions is key for success (if not, there is a risk of training more elaborate skills, while the basic ones, needed to benefit from the training, have not developed/recovered yet).

6. A strong emphasis should be put in training school assistants and teachers in simple support (such as in papers by Feeney et al.^{289–291,446}) rather than investing money to increase specialized rehabilitation outside school.
7. Rather than testing intervention success on isolated components of EF, more open-ended and ecological tasks should be used¹⁸⁰. Questionnaires should be used in association with performance-based testing to assess intervention impact at school and at home but also obtain objective measures of EF.
8. Finally, it may be inappropriate to reduce children's after-school leisure (e.g. sports, music) in order to provide cognitive rehabilitation sessions as there is growing research on the benefits of sports on EF development (in typically developing children however). "Training may even lead to negative effects if the activities it displaces are more beneficial than the training itself"²⁶⁴ (p9). As concluded by Diamond (referring to typically-developing children), "Stress, loneliness and not being physically fit impair prefrontal cortex function and EFs. The best approaches to improving EFs [...] will probably be those that (a) engage students' passionate interests, bringing them joy and pride, (b) address stresses in students' lives, attempting to resolve external causes and strengthen calmer, healthier responses, (c) have students vigorously exercise, and (d) give students a sense of belonging and social acceptance, in addition to giving students opportunities to repeatedly practice EFs at progressively more-advanced levels. The most effective way to improve EFs [...] is probably not to focus narrowly on these alone, but also address children's emotional and social development and children's physical development"³¹⁵ (p7).

FINAL CONCLUSIONS

Executive functions are impaired after childhood severe TBI, but impairment is often underestimated due to inadequate outcome measures and failure to assess EF related impairments such as prospective memory and self-awareness. Prospective Memory, in particular is rarely assessed, while it is commonly impaired after childhood TBI, with a deficit that persists into adulthood and that shows important implications for daily life.

Self-awareness is also rarely assessed in children and is a multi-component construct that can mediate the engagement in rehabilitation and effectiveness of interventions. Cooking tasks provide a feasible, enjoyable and ecologically valid way for studying on-line awareness. Prediction and evaluation of performance are important to assess but also to practice, and should be included during activities at school and at home. For intellectual awareness (also called metacognitive knowledge), stories seem a particularly well-suited assessment format. Brain-storming about personal examples of cognitive failures elicited by stories (or by another support including questionnaires) allows one to determine whether children rely on semantic awareness, autobiographical awareness or both for their responses. The remember/know paradigm and explicit requirement to provide phenomenological details can be used to differentiate between autobiographical and semantic awareness. Assessing anticipatory awareness remains a challenge but is most likely to be valid during ecological activities at home, school or during rehabilitation activities using real life settings and activities such as cooking, without prompting and without explicit knowledge of the child being assessed.

Assessments using (1) ecological performance-based tests; (2) daily life questionnaires of EF; (3) novel tasks assessing children's capacity to adapt to new situations; (4) tests built on solid theory and referring to precise models; (5) personalized measures of generalisation such as Goal Attainment Scaling are needed to adequately understand a child's impairment and to assess change after an intervention. The choice of adequate outcome measures and experimental design will make it possible to attribute change over time to an intervention over and above any general improvement arising from enthusiastic participation in a research study. Engaging the child's "everyday people" in EF coaching proved to be challenging and

should be systematically considered and achieved as best as possible, in order to improve application of adequate strategies in the home environment.

Interventions based on metacognition (also called metacognitive training) do not seem from the pilot data of this PhD to be the best option to train young children with severe EF impairments, and especially those most anosognosic or the youngest (before mid-adolescence or even adulthood when metacognition matures). Adapting interventions from developmental disabilities to children with TBI may be more beneficial than adapting adult interventions to children. Developmental coherence of interventions is key for success (if not, there is a risk of trying to train more elaborate skills, while the basic ones, that are needed in the training, have not developed/recovered yet). A strong emphasis should be put on training school assistants and teachers in simple support for developing EF. Targeting the family at some level of the intervention (personalized for each family) seems necessary, ranging from giving family support, to actively engaging the family in cognitive rehabilitation. However, this may be challenging and not all families are able to engage. In all cases, it is important that family priorities and goals are addressed, that intervention-related burden is evaluated and that it is not greater than perceived gains.

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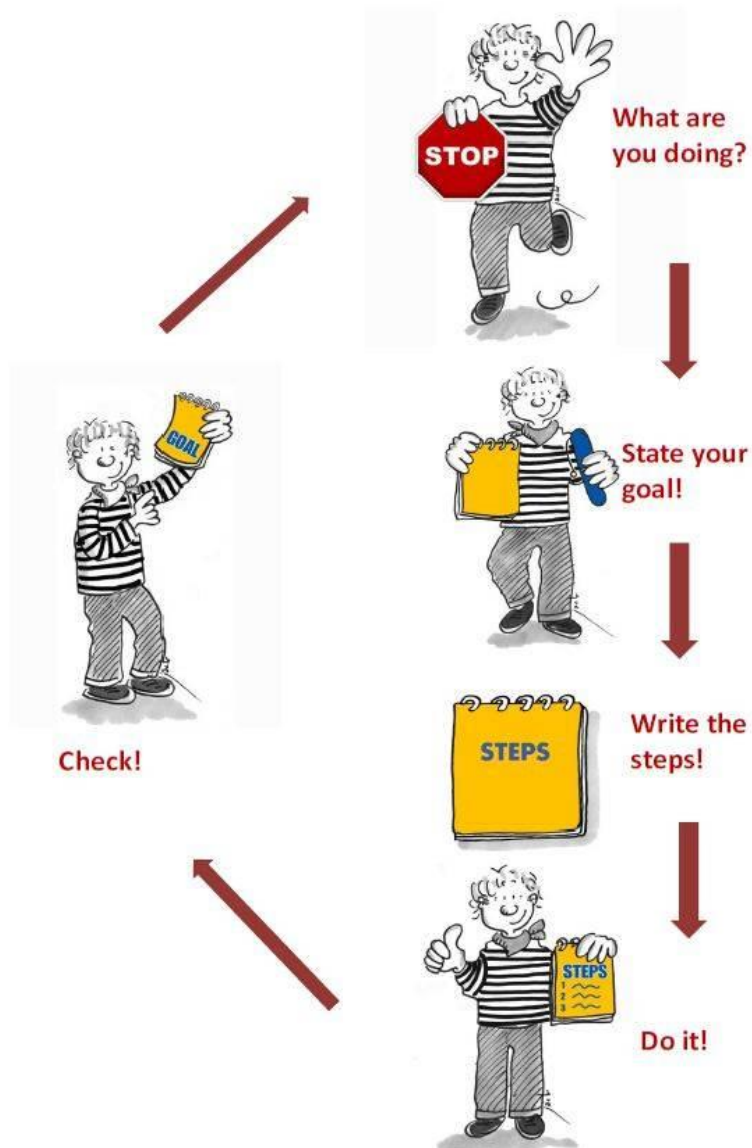
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APPENDIX

EXAMPLE OF PGMT MATERIALS



FRENCH ADAPTED VERSION OF CHAPTER 5: " REEDUCATION DES FONCTIONS EXECUTIVES CHEZ L'ENFANT"

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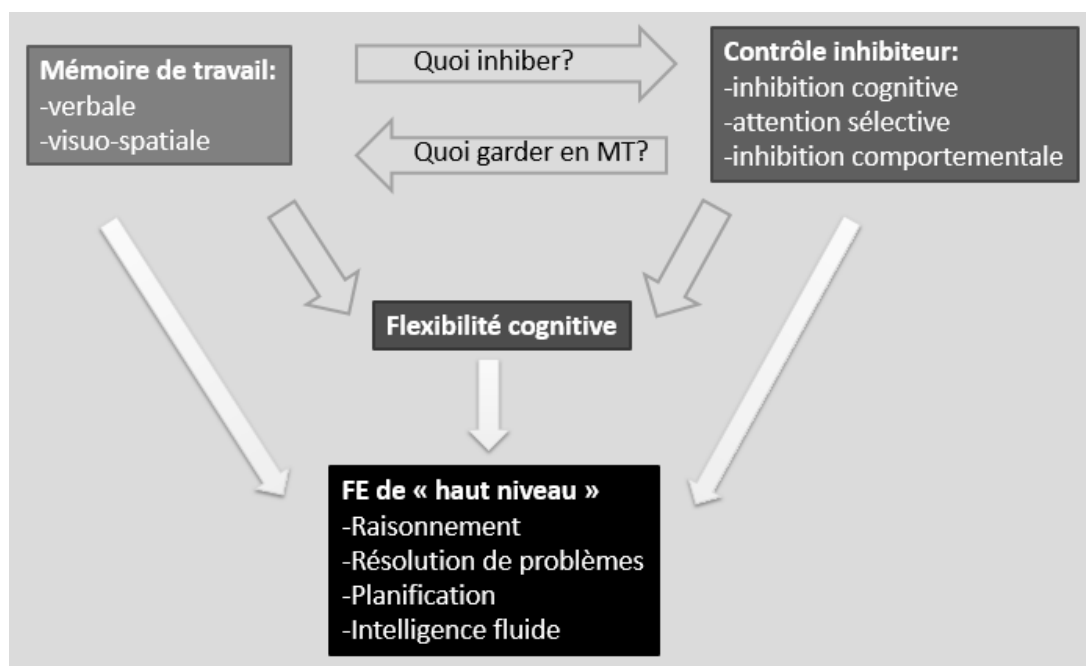
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Note préliminaire : cet article porte sur la rééducation des fonctions exécutives (FE) chez l'enfant dysexécutif en général, bien que les exemples soient majoritairement tirés de la littérature sur les enfants cérébro-lésés et très peu de la littérature de troubles développementaux tels que le TDAH et l'autisme. Cet article ne présente pas une revue exhaustive de la littérature sur ce thème large, au sein duquel pourtant peu d'approches sont clairement validées.

DEFINITIONS DES FONCTIONS EXECUTIVES

La définition des FE varie selon les modèles, selon les auteurs, mais il est généralement admis que qu'il existe trois FE centrales^{25,26} : *l'inhibition*, aussi appelée contrôle inhibiteur, *la mémoire de travail* (MT) et *la flexibilité cognitive*. Le modèle développemental des FE de Diamond²⁴ est particulièrement utile pour comprendre le lien entre ces FE et la manière dont elles s'influencent mutuellement au cours du développement progressif des FE chez l'enfant (voir figures introduction.1 et annexe.2) : la MT soutient le contrôle inhibiteur (il faut garder en mémoire ce qui est important et approprié pour la tâche en cours et ce qui est à inhiber), mais le contrôle inhibiteur soutient à son tour la MT en évitant que l'espace limité de la MT soit encombré d'information inutiles pour la tâche en cours. La flexibilité cognitive requiert des compétences de MT et de contrôle inhibiteur et permet de changer de perspective, en chargeant en mémoire de travail un autre point de vue, tout en inhibant le point de vue précédent. A partir de ces 3 FE *centrales*, les autres FE dites *de haut-niveau* se construisent, telles que la planification, la résolution de problèmes et le raisonnement³⁵. Contrairement à l'adulte où l'intelligence est supposée être indépendante des FE, l'intelligence fluide est assimilée chez l'enfant aux FE de haut niveau²⁴ car de nombreuses études ont montré leur lien inextricable³⁶⁻³⁸.

Figure annexe.1 : Modèle des Fonctions Exécutives de Diamond, traduit et simplifié.



PRATIQUES ACTUELLES DANS LA PRISE EN CHARGE DES FE

Beaucoup de stratégies ont été proposées pour aider les enfants dysexécutifs et de nombreux manuels et articles ont été publiés pour guider les professionnels ²⁷¹⁻²⁷⁵. Beaucoup de ces approches sont prometteuses, mais la plupart n'ont jamais été validées. La plupart des techniques et stratégies utilisées reposent sur des données issues de travaux réalisés chez l'adulte ⁴⁴⁸ ou chez l'enfant sain. De plus l'âge développemental de l'enfant et la particularité de la pathologie rééduquée (déficits associés ne permettant pas forcément de compenser les FE comme chez l'enfant au développement typique) sont rarement pris en compte dans le choix des techniques proposées.

En pratique courante, quatre grands types d'approches sont utilisés pour aider les enfants cérébro-lésés dans leur fonctionnement exécutif, chacune ayant ses limites: (1) Utilisation d'adaptations de l'environnement et d'aides techniques (ex : utilisation de systèmes de rappel

électronique tels le « Neuropage »^{278,279}, qui a montré des effets bénéfiques sur les difficultés de planification et de mémoire prospective, et qui peut actuellement être facilement remplacé par les smartphones, omniprésents donc accessible à tous). L'inconvénient de ce type d'approche est qu'il suppose d'avoir préprogrammé l'aide technique et préparé l'environnement, et l'utilisation de ce moyen utile et simple se limite principalement à certaines situations prévisibles; (2) Entraînement spécifique de fonctions exécutives élémentaires isolées (MT, inhibition...), mais l'effet de généralisation de ces entraînements au fonctionnement exécutif dans la vie quotidienne a rarement pu être démontré; (3) Entraînement des enfants sur des tâches précises dans lesquelles ils sont en difficulté à cause de leur syndrome dysexécutif (ex : préparer son cartable) en rendant ces tâches routinières, voire automatiques et donc nécessitant moins d'implication des FE ; (4) Fournir aux patients des stratégies métacognitives applicables à une variété de situations, y compris des situations nouvelles et imprévues. Ce dernier type d'approche est efficace chez les adultes¹¹⁴, mais il y a peu d'arguments actuellement pour l'appliquer chez les enfants (surtout en cas de dysfonctionnement sévère)^{200,260,286}. En effet chez l'enfant, la métacognition [terme se référant à une analyse de sa propre cognition pendant l'exécution d'une tâche, mais aussi à la juste connaissance des tâches qui risquent d'être difficiles et qui nécessiteront une stratégie de compensation²¹¹] est pauvre et ne se développe suffisamment qu'à l'âge adulte. Cependant, l'utilisation de stratégies simples enseignées aux enfants par des exercices répétés, visant à leur apprendre quand utiliser cette technique (donc pas une technique "métacognitive" à proprement parler, malgré l'utilisation du terme par certains auteurs d'études portant sur de jeunes enfants), peut être utilisée et semble prometteuse, même avant que les enfants aient un niveau métacognitif développé^{108,269,287}.

CE QUI EST VALIDE

Il y a globalement très peu (voire pas) de méthodes de rééducation scientifiquement validées des FE chez l'enfant. En dehors de la mémoire de travail (voir paragraphe sur le Cogmed® plus loin) et de l'attention (qui ne sera pas traitée dans cet article), les revues internationales de la littérature ne rapportent pas de méthode validée de rééducation des FE chez les enfants cérébro-lésés¹¹⁰⁻¹¹³. Le problème majeur de la plupart des rééducations est l'absence de généralisation à des tâches non travaillées et l'absence de transfert des acquis à des contextes différents de celui du cadre de rééducation (Ylvisaker, 2005). La rééducation cognitive des

enfants en général serait plus efficace sur la MT, la mémoire et l'attention que sur les FE, mais les biais méthodologiques rendent difficile l'appréciation de la taille des effets ¹⁰⁷.

Cependant des principes généraux émergent de la recherche actuelle:

1. Le niveau « challenge » (difficulté d'exercices rapidement croissante dès qu'un niveau est atteint) semble indispensable pour obtenir des progrès ²⁶⁴. Les exercices doivent donc s'adapter au niveau de l'enfant ^{24,265} à tout moment (ce qui est le cas de la méthode Cogmed® par exemple).
2. La rééducation doit prioriser le Contexte et le Contenu (« Context and Content-sensitive approach ») ^{277,290,294,356}: choix des exercices les plus écologiques et les plus fonctionnels possibles (plutôt que des exercices décontextualisés de la « vraie » vie familiale et scolaire de l'enfant) et administration de la rééducation sur les lieux de vie de l'enfant autant que possible (pour faciliter la généralisation et le transfert).
3. Implication de la famille et de l'école dans la rééducation (voir paragraphe spécifique plus loin)
4. La durée de l'intervention doit être longue si l'on veut avoir une généralisation des effets et un maintien de l'efficacité sur le long terme ¹¹².

UNE APPROCHE INTERDISCIPLINAIRE ET DEVELOPPEMENTALE EST NECESSAIRE DANS LA PRISE EN CHARGE DES FE DE L'ENFANT

Les interventions visant à améliorer les FE devraient être envisagées lorsque les fonctions cognitives plus « basiques », pré-requises aux FE ont suffisamment mûri (ou récupéré), ou sont compensées. Le modèle « Paediatric Neurocognitive Interventions » (modèle PNI) est un cadre utile pour choisir les priorités des prises en charge ²⁶². Une approche interdisciplinaire ²⁶³ est essentielle pour s'assurer que l'intervention proposée soit appropriée dans le contexte des difficultés physiques, cognitives, comportementales, sociales et psychologiques de l'enfant.

Il est indispensable que les rééducations proposées soient cohérentes du point de vue développemental. Ceci est particulièrement important pour les FE. Les lobes frontaux arrivent à maturité tard dans la vie, au début de la 3^{ème} décennie ²⁵⁸ et les FE sont toujours en cours de

maturation pendant l'adolescence et au début de l'âge adulte ⁴⁴⁹. Pour cette raison les rééducations construites spécifiquement pour des adultes (et adaptées à l'enfant) ont peu de chance d'être bénéfiques pour les enfants, en particulier si elles requièrent des fonctions cognitives qui ne sont pas attendues pour un enfant, telles que la métacognition (Krasny-Pacini et al., 2015). D'autre part, l'utilisation de stratégies se développe progressivement pendant l'enfance et par conséquent des stratégies efficaces chez les adultes peuvent être inefficaces chez l'enfant ^{261,288}.

STRATEGIES DITES « METACOGNITIVES »

DEVELOPPEMENT DE STRATEGIES CHEZ L'ENFANT

Les stratégies sont définies comme des opérations mentales volontaires, orientés vers un but, ayant pour objet de résoudre un problème ²⁸⁸. D'un point de vue développemental, les jeunes enfants peuvent utiliser des stratégies, mais les enfants plus âgés et les adolescents les utilisent plus souvent, sélectionnent des stratégies plus efficaces, et arrivent, mieux que les plus jeunes, à améliorer leur performance. Même si les plus jeunes ont des difficultés à mettre au point une stratégie (ce que les développementalistes appellent «déficience de production de stratégie»), ils peuvent être entraînés à utiliser une stratégie, et améliorer leur performance grâce à elle ²⁸⁸. Cependant, ils ne seront en général pas capables de généraliser cette stratégie à une tâche nouvelle, qu'ils auront tendance à résoudre de façon peu stratégique ²⁶¹. D'autres jeunes enfants n'améliorent pas leur performance malgré une stratégie adéquate (ce qu'on appelle « déficience d'utilisation de stratégie »), en général du fait de limitations de leurs capacités cognitives, en particulier de la MT et de la métacognition.

Il est intéressant de noter que, contrairement aux adultes qui ont besoin d'une bonne connaissance de leurs difficultés/troubles (et de la difficulté de la tâche) pour bénéficier de l'apprentissage de l'utilisation d'une stratégie, il semblerait que les enfants sains, eux, ont avant tout besoin que l'apprentissage de la stratégie soit ludique, motivant et intéressant, indépendamment de son succès/bénéfice. Surtout ils ont besoin de la pratiquer de façon suffisamment répétée pour que la déficience d'utilisation se résorbe.

Reste la question de savoir s'il en est de même chez les enfants cérébro-lésés, dont les FE et la métacognition sont déficientes, et si l'apprentissage de stratégies dans cette population

particulière est une bonne technique de remédiation. Dans l'étude de Krasny-Pacini et al. ²⁰⁰, les enfants appliquaient volontiers les stratégies du Goal Management Training sur les tâches papier crayon, dans un contexte de bureau de rééducation et les trouvaient effectivement amusantes (par exemple s'arrêter régulièrement pour redire l'objectif de la tâche lorsqu'ils voyaient le « bonhomme STOP »), mais cette stratégie n'améliorait pas forcément leur performance. De plus, lorsque la même stratégie était ensuite appliquée à des activités plus complexes et écologiques (cuisine, jeu de piste, jeu de déduction...), ils n'appliquaient plus les stratégies apprises. L'étude de Missiuna a mis en évidence le même phénomène ²⁸⁶: rendre les séances ludiques et plaisantes semblait utile, tandis qu'enseigner la stratégie "Goal-Plan-Do-Check" et essayer de promouvoir son application l'était peu (alors même que cette approche est efficace dans d'autres pathologies développementales comme la dyspraxie). Peut-être que justement cette méthode de rééducation est efficace dans la dyspraxie développementale car il s'agit d'une stratégie mettant en jeu l'utilisation des FE (au moins partiellement efficaces chez ces enfants) pour compenser les difficultés de réalisation de gestes spécifiques, contrairement aux enfants cérébro-lésés, chez qui des troubles spécifiques et probablement plus sévères des fonctions exécutives ne permettent pas l'utilisation efficace de cette stratégie pour mener à bien diverses tâches de la vie quotidienne.

Dans tous les cas, chez les enfants, les stratégies doivent être simples et concrètes, et leur apprentissage doit être accompagné, supervisé et répété, contrairement aux adultes cérébro-lésés pour qui on peut se contenter d'expliquer une stratégie pour la voir appliquer spontanément et efficacement (du moins chez des patients ne présentant pas des troubles trop sévères des FE) ^{98,119,285}.

STRATEGIES DITES METACOGNITIVES UTILISEES CHEZ L'ENFANT DYSEXECUTIF CEREBRO-LESE

Différentes stratégies à apprendre et automatiser, utilisées chez des enfants cérébro-lésés ont été publiées : (1) "Goal-Plan-Do-Review" ²⁸⁹⁻²⁹¹, contextualisé dans la vie quotidienne de l'enfant (Ylvisaker & DeBonis, 2000; Ylvisaker & Feeney, 1998) ; (2) "Goal Plan Do Check" ²⁸⁶ utilisé dans le programme pour enfants dyspraxiques CO-OP "Cognitive Orientation to Daily Occupational Therapy" ²⁹⁵; (3) "Stop, State your Goal, Split Task into subtasks, Do it, Check" du Goal Management Training ^{98,200}; (4) "Nature Planning

Representing Monitoring”²⁸⁷. Même si la phrase-type varie, l’idée est toujours la même: devant une tâche nouvelle, éviter de commencer de suite impulsivement, mais s’arrêter, analyser la nature de la tâche et déterminer quel est le but à atteindre, élaborer un plan d’action composé d’étapes, l’appliquer étape par étape, tout en vérifiant régulièrement si ce le plan fonctionne et si l’on ne s’éloigne pas du but à atteindre et, enfin, vérifier à la fin de tâche si aucune étape n’a été oubliée et si le but est atteint. L’approche de Feeney et Ylvisaker est celle qui a montré dans plusieurs études de bonne qualité, menées en méthodologie expérimentale de cas uniques multiples, des effets bénéfiques, et qui a pu être répliquée. A noter que dans ces études, la stratégie n’était pas simplement enseignée au cours de séances de rééducation déconnectées du milieu scolaire, mais (et c’est le point fondamental de ces études), elle était relayée aux enfants au quotidien par tous les enseignants à l’école, qui étaient formés et supervisés par les investigateurs de l’étude. De plus, avant de débiter une tâche ou un exercice, les enfants étaient invités à estimer la difficulté de la tâche et le temps qu’elle prendrait. Après l’avoir terminée, ils étaient invités à analyser ce qui avait fonctionné et ce qui n’avait pas fonctionné dans le déroulement de la tâche, et à en ré estimer la difficulté a posteriori.

IMPLICATIONS DES PARENTS ET DE L’ECOLE

Les interventions doivent être longues et suffisamment intensives pour permettre d’entraîner un progrès. Ce caractère intensif a peu de chances d’être atteint si la rééducation est administrée de façon classique en séances hebdomadaires de neuropsychologie, ergothérapie ou orthophonie. C’est pourquoi Ylvisaker a proposé d’adopter plutôt une approche visant à former les « *everyday people* » (personnes au contact quotidien avec l’enfant : parents, auxiliaires de vie scolaire, aide maternelle, maitres d’école...) à devenir des « coach cognitifs » pour l’enfant. Pour Ylvisaker, il est préférable de travailler les objectifs de rééducation pendant la journée, dans le contexte des activités habituelles et des routines avec les « *everyday people* », plutôt que de travailler sur ces objectifs dans un environnement spécialisé et structuré, déconnecté de la vie quotidienne de l’enfant ²⁹⁴.

Idéalement il faudrait donc inclure les parents dans la prise en charge, comme « coach cognitifs », en les formant au fonctionnement cérébral, au concept de FE et à leur utilité / leur rôle dans les activités de la vie quotidienne, aux difficultés spécifiques de leur enfant, et à la

façon dont ils peuvent l'aider concrètement au quotidien à développer ses FE ^{268,269,298,303}. L'engagement approprié des familles est parfois difficile à obtenir ²⁰⁰, surtout si les parents ont eux-mêmes des difficultés exécutives à minima ou de faibles compétences de métacognition. Les études montrant le plus d'efficacité sur les FE sont actuellement celles qui ciblent les interactions parents-enfants, et il est intéressant de noter que, contrairement à ce qu'on pourrait prévoir, ce sont les familles les défavorisées qui bénéficient le plus de ce type d'intervention (ex : programme "I-Interact" ²⁷⁰).

L'implication des parents et des intervenants à l'école semble, à ce jour, le meilleur format pour obtenir une intensité suffisante, par la répétition qui peut ainsi être obtenue dans les activités concrètes et routinières du quotidien. Là encore la culture française est bien éloignée de ce type d'approche. L'Education Nationale n'est pas toujours ouverte à des propositions innovantes personnalisées par élève et les professionnels ne sont pas toujours prêts à transmettre leur savoir à des personnes tout-venant. Nous sommes frileux lorsqu'il s'agit de donner plus de « pouvoir » aux parents et de leur permettre de choisir leurs priorités pour leur enfant. Pourtant, de plus en plus dans les autres pays, les approches « family-centered » ^{267,450,451} ont montré une efficacité supérieure aux approches classiques où la famille n'est que le récipiendaire passif du savoir des thérapeutes. L'étude de Braga et al. est une très belle démonstration de ce type d'approche dans le domaine cognitif: dans cette étude, des enfants traumatisés crâniens étaient randomisés dans 2 formats de rééducation différents : (1) une rééducation délivrée classiquement en centre de rééducation par une équipe multidisciplinaire de thérapeutes, 2 heures par jour, 5 jours par semaine ; ou (2) une rééducation réalisée au domicile, poursuivant les mêmes objectifs de rééducation, via des exercices intégrés dans la vie quotidienne de l'enfant au domicile, avec /sous la supervision des parents, en vue d'atteindre chacun des objectifs (moteur, fonctionnel et/ou cognitif). Il est fondamental de préciser que dans ce deuxième cas, les parents étaient entraînés, soutenus, accompagnés et corrigés très régulièrement par les thérapeutes, d'abord au cours d'un séjour avec leur enfant en centre de rééducation et par des visites des thérapeutes au domicile. Les enfants et leurs parents revenaient tous les 15 jours au centre de rééducation pour des séjours de réévaluation de la situation, afin de déterminer si les objectifs avaient été atteints et d'en fixer de nouveaux le cas échéant. Les parents disposaient systématiquement d'un classeur de fiches spécifiques et personnalisées avec des supports visuels d'exercices (ne requérant pas de capacité de

lecture, ce qui était une donnée importante de cette étude incluant des enfants dans des parties reculées du Brésil où beaucoup de parents pouvaient être analphabètes). La difficulté et le type d'exercices étaient personnalisés et régulièrement (tous les 15 jours) réadaptés par les thérapeutes selon les progrès des enfants, et les fiches d'exercices remises à jour. Les fonctions exécutives n'ont pas été évaluées spécifiquement dans cette étude mais le deuxième format de rééducation s'est révélé significativement plus efficace sur les progrès et moteurs et cognitifs (évalués par un test de Wechsler). Il était également moins cher et plus apprécié des familles et des thérapeutes.

QUELQUES EXEMPLES DE METHODES PROMETTEUSES DE REEDUCATION DES FONCTIONS EXECUTIVES

Le « **Cogmed Working Memory Training®** » est l'entraînement de la mémoire de travail le plus utilisé et ayant fait l'objet de très nombreuses études, principalement chez l'enfant avec Trouble Déficitaire de l'Attention/Hyperactivité (TDA/H). Cogmed ® peut être recommandé, bien que certaines données soient contradictoires en raison de biais méthodologiques de certaines études [voir pour une synthèse les revues de la littérature de ¹⁰²⁻¹⁰⁴]. Les données de la littérature montrent que Cogmed ® peut améliorer la MT verbale et visuo-spatiale et la mémoire à court terme visuo-spatiale chez les enfants avec TDA/H (voir étude de Holmes et al. 2010, qui a utilisé des tâches non travaillées comme critère de jugement) et les enfants à faible niveau de MT [chez qui on est parvenu en plus à améliorer la mémoire à court terme verbale dans une étude ¹⁰⁵]. Certaines études mettent en évidence des gains persistants à un an ¹⁰⁶. Les données sur le transfert aux capacités de mathématiques et de lecture sont par contre contradictoires ^{105,106}.

Chez les enfants cérébro-lésés, Cogmed ® améliore la mémoire à court terme visuo-spatiale (et cet effet est maintenu 3 mois après l'intervention). La lecture s'améliore à 3 mois uniquement. A ce jour il n'y a pas à notre connaissance d'études montrant l'efficacité de Cogmed ® sur les FE proprement dites chez les enfants cérébro-lésés. L'avantage de cette méthode est qu'elle peut être effectuée par l'enfant seul, devant un ordinateur, à l'heure et dans le lieu qui arrange la famille, sous la supervision régulière (éventuellement à distance par téléphone) d'un thérapeute. Le thérapeute reçoit un feedback de la compliance au programme

et des progrès de l'enfant, même s'il n'assiste pas aux séances. Une version française existe, commercialisée par Pearson. Le programme s'auto-adapte aux performances de l'enfant, créant toujours le niveau « challenge » recommandé dans la littérature. C'est l'une des méthodes ayant le plus grand nombre de publications et sa validité est robuste, sous réserve que les objectifs fixés soient bien adaptés à ce qu'elle peut apporter. A notre connaissance, aucune étude n'a démontré son efficacité sur l'amélioration de l'administrateur central et elle ne peut être recommandée actuellement que pour l'objectif d'améliorer la mémoire à court terme, ou la MT visuo-spatiale et verbale. Le niveau de preuve de la méthode Cogmed ® est supérieur à celui de tous les autres entraînements informatisés, que l'on classe hélas souvent ensemble.

L'équipe de Wade et Kurowski a développé plusieurs méthodes de rééducation administrées à distance par internet pour les enfants cérébro-lésés et leur familles : le **TOPS** [(« **Teens On-Line Probel Solving**, ²⁹⁹⁻³⁰¹] qui a ensuite donné naissance à deux autres méthodes: le **CAPS** (**Counsellor-Assisted Problem Solving**, ^{298,303} et le **I-INTERACT** ²⁷⁰. Ces méthodes s'attachent plus à donner à la famille les outils nécessaires pour favoriser la récupération après lésion cérébrale, qu'à entraîner les fonctions défaillantes chez l'enfant/adolescent. Elles offrent toutes les trois une base familiale solide sur laquelle d'autres rééducations peuvent éventuellement s'ajouter et elles ont, à ce jour, un niveau de preuve supérieur aux réentraînements cognitifs administrés dans un bureau par un professionnel seul sans participation de la famille. Elles ont été testées dans de grandes études de bonne qualité méthodologique (dont certaines randomisées contrôlées) et ont montré un effet bénéfique en situation écologique (notamment sur le questionnaire *Behavior Rating Inventory of Executive Function* - BRIEF¹⁴⁵, évaluant le fonctionnement exécutif au quotidien).

- La particularité du CAPS et TOPS est de s'adapter aux rythmes et besoins des familles, qui peuvent choisir quand débiter la session. Les thèmes des modules incluent la résolution de problèmes, l'auto-régulation, l'implantation et le monitoring d'objectifs spécifiques, ainsi que des modules visant davantage le comportement et les aspects psychologiques (rester positif, utiliser de manière optimale la communication verbale et non verbale, gérer les crises, contrôler le comportement). Les familles peuvent également choisir parmi une série de modules optionnels (douleur, sommeil, interactions avec la fratrie, habiletés sociales...). L'ensemble de l'intervention dure

environ 6 mois, selon le rythme des familles. L'ensemble de la famille participe aux sessions. Le CAPS a montré son efficacité chez les adolescents de plus de 14 ans avec un maintien des effets, et même une augmentation des effets par rapport au groupe contrôle à long terme.

- Le I-Interact s'adresse aux parents et enfants âgés de 3 à 9 ans, présentant au premier plan des difficultés exécutives comportementales (difficultés de contrôle inhibiteur, d'autorégulation) et constitue un « télé-coaching » des parents, administré en situation de jeu, à la maison (que l'on pourrait comparer au programme télévisé « super nounou »).

Le programme adulte **CRT** (“**Cognitive Remediation Therapy**”) semble permettre d'obtenir des gains sur la flexibilité chez l'adolescent présentant une anorexie accompagnée de troubles neuropsychologiques ²⁸⁴, mais les méthodologies des études utilisant la CRT ont souvent des biais (utilisation d'un critère de jugement très proche des exercices ayant été utilisés pour la rééducation, et manque de mesures de généralisation). A notre connaissance, il n'y a actuellement pas d'étude convaincante chez les enfants plus jeunes ou dans d'autres pathologies.

“**Unstuck and On Target**”³⁰⁴ (tr: « débloqué et focalisé sur la cible ») est une intervention utilisée auprès d'enfant présentant des troubles du spectre autistique sans retard mental global associé. Administrée à l'école en 28 sessions de 30-40 minutes, cette méthode vise la flexibilité, le « *big picture thinking* » (voir l'ensemble plutôt que les détails) et la planification. Elle intègre les parents et les professeurs non spécialisés pour relayer l'intervention au cours de la journée et elle a montré sa supériorité par rapport à un entraînement des compétences sociales seules. De plus, de façon surprenante, une généralisation aux compétences sociales, pourtant non entraînées spécifiquement par la méthode, a été obtenue dans cette étude ²⁶⁸.

INTERVENTIONS EFFICACES CHEZ L'ENFANT AU
DEVELOPPEMENT TYPIQUE QUI MERITERAIENT D'ETRE
INTEGREES DANS DES PROGRAMMES DE REEDUCATION

Contrairement au manque d'études sur la rééducation des FE chez les enfants présentant une pathologie ou un développement atypique, chez l'enfant sain au développement typique, plusieurs interventions ont démontré une capacité à améliorer les FE ^{265,315,452}.

Certains programmes implantés dans les écoles [ex.: « Tools of the Mind » ³¹⁰, « Montessori » ³¹¹ « PATHS: Promoting Alternative Thinking Strategies »³¹², « Chicago School Readiness Project » ^{314 313}] et délivrés par les enseignants habituels des enfants, préalablement éduqués à la méthode, ont montré qu'il est possible de promouvoir le développement des FE dans les écoles non spécialisées [voir les revues de Diamond ³¹⁵ et ³¹⁶]. Cependant comme les compétences dans les FE et la réussite scolaire sont corrélées, il est difficile d'établir le lien direct de causalité entre ces programmes et le développement propre des FE ³¹⁷. Ce type de programmes à l'école semble actuellement plus recommandable pour les enfants dysexécutifs que les approches délivrées dans un environnement clinique (centre de rééducation, cabinet d'orthophonie...).

Les professionnels sont souvent amenés à donner des conseils éducatifs aux parents d'enfants présentant des troubles dysexécutifs. Ces conseils sont trop souvent basés sur leur expérience personnelle, et pas suffisamment sur les techniques ayant prouvé une efficacité. A défaut de guidelines validées chez l'enfant dysexécutif, il pourrait être utile d'utiliser ce qui est validé chez l'enfant sain. Le site internet australien « Elever les enfants » (www.Raisingchildren.net.au) liste les pratiques éducatives testées et efficaces et peut être utile pour conseiller les familles.

Une approche très prometteuse mais non (ou très peu) exploitée en France est la pratique d'activité physique pour améliorer les FE. Une série d'études bien menées a montré les effets bénéfiques de l'exercice physique sur la cognition en général ³¹⁸⁻³²¹, et les FE en particulier. L'activité physique soutenue ^{319,322,323}, les sports à « challenge cognitif », qui stimulent le développement de stratégies, comme le tennis ³²⁴, ou les arts martiaux traditionnels ³²⁵ sont des exemples de sports ayant montré une amélioration des FE (indépendamment des effets métaboliques), et qui seraient à recommander aux enfants dysexécutifs. Hélas, en France de manière générale, lors de la prise en charge d'enfants en situation de handicap, qui sont souvent lents et fatigables, qui ont des séances de rééducation organisées sur le temps scolaire, lors de l'établissement des Projets Personnalisés de Scolarisation, la première

activité à être supprimée des emplois du temps est l'activité physique et sportive, afin de « caser » une séance de rééducation ou d'alléger le nombre d'heures de cours de l'enfant. Ceci est particulièrement dommage, si l'on considère que l'activité physique est l'activité qui a actuellement le niveau de preuve le plus élevé en termes de développement des FE. La fatigabilité des enfants ayant un handicap moteur associé entraîne souvent l'utilisation d'un fauteuil roulant (parfois électrique) à l'école, ce qui peut contribuer à créer un cercle vicieux de déconditionnement à l'effort, baisse des fonctions exécutives et attentionnelles et fatigabilité cognitive accrue. Il serait urgent de prendre ces éléments en compte et d'inclure un réentraînement à l'effort avec des exercices aérobies dans les programmes multidisciplinaires dédiés aux enfants dysexécutifs, et d'évaluer leur bénéfice.

Comme souligné par Diamond, «le stress, la solitude et la mauvaise condition physique handicapent la fonction du cortex préfrontal et les FE. Les meilleures approches pour améliorer les FE [...] seront (a) celles qui aident les enfants à s'engager dans ce qui les passionne et leur apporte joie et fierté, (b) celles qui prennent en compte les éléments stressants de la vie des enfants, en essayant d'en résoudre les causes externes et en renforçant les réactions plus saines et calmes, (c) celles qui font pratiquer vigoureusement des exercices et sport, (d) celles qui donnent aux enfants un sentiment d'appartenance et d'acceptation sociale, tout en leur donnant des opportunités de pratiquer de manière répétée les FE à des niveaux progressivement plus difficiles. La meilleure manière d'améliorer les FE n'est pas de se focaliser étroitement sur celles-ci, mais de s'occuper du développement physique, émotionnel et social de l'enfant »³¹⁵.

DES METHODES NON REEDUCATIVES A L'ETAT DE RECHERCHE, QUI POURRAIENT DANS LE FUTUR S'ASSOCIER A LA REEDUCATION

Le Neurofeedback⁴⁵³ a montré des effets intéressants dans certaines études sur des populations avec autisme et TDAH⁴⁵⁴⁻⁴⁵⁶, mais nécessite davantage de recherche. Son application aux enfants cérébro-lésés n'a pratiquement pas été évaluée.

La recherche sur la stimulation trans-crânienne magnétique répétitive (rTMS) ou par techniques similaires (type tDCS) en est actuellement à une étape de recherche uniquement^{457,458}. Les effets à long terme doivent être explorés pour s'assurer de l'innocuité de la

méthode, mais il pourrait s'agir de méthodes d'avenir, particulièrement en phase aigüe après lésion cérébrale de l'enfant.

LIMITES METHODOLOGIQUES DES ETUDES EVALUANT LES PROGRAMMES DE REMEDIATION DES FE CHEZ L'ENFANT

Les limites de la plupart des méthodes de rééducation publiées sont nombreuses.

1. Beaucoup d'études utilisent le même type d'exercices pour l'entraînement et l'évaluation post rééducation. Elles permettent de prouver que les enfants s'améliorent sur ces tâches, mais ne permettent aucune conclusion sur l'efficacité de la rééducation sur les FE (et leur utilisation au quotidien). Une tâche évaluant les FE doit être nouvelle, non apprise, non automatisée, sans quoi elle n'évalue plus les FE en tant que telles ^{22,229,367}. De plus, à force d'utiliser en rééducation des exercices inspirés du « Stroop » ou de tests de classement de formes (Wisconsin Card Sorting Test, exercice du Cognitive Remediation Therapy), nos tests d'évaluation risquent de perdre en validité et cela peut fausser les résultats des bilans, car les tâches ne seront plus nouvelles pour l'enfant.
2. Pour les FE on ne devrait utiliser un test qu'une fois si on veut évaluer l'adaptation à une situation non routinière. Dans l'étude du Goal Management Training par exemple ²⁰⁰, les enfants semblaient s'être améliorés sur la tâche écologique du gâteau au chocolat ^{120,121} mais lorsqu'ils ont été confrontés à une nouvelle recette ayant le même nombre d'étapes, le même nombre d'ingrédients et présentée sous la même forme dans le même lieu, ils revenaient au nombre d'erreurs qu'ils avaient faites avant la rééducation. La généralisation à des tâches non travaillées, et surtout à des tâches écologiques et au contexte de l'enfant est insuffisamment évaluée, or elle constitue l'élément clé permettant de dire qu'une rééducation est réellement efficace.
3. Le transfert au contexte de l'enfant et la généralisation à des tâches écologiques sont insuffisamment évalués. Les critères de jugement ne peuvent être constitués uniquement d'exercices papier-crayon.
4. Les études portant sur des approches globales qui pourraient avoir un effet sur les FE, n'évaluent pas souvent le progrès sur le FE proprement dites ^{267,382}.

5. Souvent dans les études de grande puissance (incluant de nombreux enfants dans le cadre d'essais randomisés contrôlés) le critère de jugement principal est un questionnaire rempli par les parents ²⁹⁹, avec toute la subjectivité inhérente à ce type d'évaluation. L'aveugle des parents par rapport à la rééducation testée pourrait compenser en partie ce biais. Dans les études de plus petit effectif, un critère objectif serait nécessaire.
6. Les exercices de résolution de problèmes utilisés sont souvent éloignés de la vie de tous les jours de l'enfant et manquent de validité écologique ²⁹⁷.
7. Le design des études est souvent pauvre, avec uniquement des mesures avant et après rééducation, ne permettant pas d'exclure que l'effet observé soit lié à un effet retest ou à un effet développemental indépendant de la rééducation, surtout si l'étude ne comporte pas de groupe contrôle ^{286,383}.

Enfin, à partir de quel niveau de preuve peut-on considérer qu'une rééducation est efficace ? L'amélioration d'une tâche travaillée est rarement le reflet d'un vrai progrès des FE mais plus souvent d'un apprentissage de la tâche, qui la rend plus routinière et donc moins dépendante des FE.

Même lorsque les enfants sont évalués par une tâche différente de celle utilisée en rééducation, bien souvent ces tâches sont des tâches papier crayon très éloignées de la vie des enfants, qui manquent de validité écologique. Idéalement un bon critère de jugement de progrès exécutif serait une tâche ayant une bonne validité écologique (voir pour une revue¹⁸⁰, dont l'effet retest est connu et le plus faible possible. Les questionnaires tels le BRIEF, sont un complément utile, mais ne devraient pas constituer le seul critère de jugement. Les tests utilisant la réalité virtuelle ⁴⁵⁹, le test du gâteau au chocolat ^{120,121} sont des exemples de tests écologiques valides et sensibles, mais qui ont pour l'instant des normes insuffisantes. Dans le futur, le développement de versions parallèles de ce type de tests permettrait de garder leur caractère « exécutif » ^{22,229,367}, c'est-à-dire mesurant l'adaptation à la nouveauté, la recherche de stratégie, au cours d'un traitement non automatique. Une autre approche consiste à quantifier la réussite d'objectifs personnalisés de rééducation par des méthodes de type « Goal Attainment Scaling » - GAS ^{376,377,380,460} qui peuvent être un critère de jugement intéressant, surtout dans des populations hétérogènes. Une rigueur dans le choix du critère d'atteinte de l'objectif est nécessaire, surtout en situation de recherche ^{413,440}.

Dans tous les cas, pour être efficace, tout protocole de rééducation doit promouvoir et évaluer la généralisation à des activités non travaillées, le transfert à d'autres contextes et situations que le lieu et cadre de la rééducation (et en particulier l'école et la maison) et le maintien à long terme des effets ^{111-113,329}.

RECOMMANDATIONS ET CONCLUSION

Beaucoup de manuels/méthodes/exercices visant l'amélioration des FE (et le travail cognitif en général) sont commercialisés, desquels la plupart n'ont pas montré d'efficacité supérieure à des activités non spécifiques (voir des amis, faire du sport...). Les professionnels doivent être prudents avant d'acheter et d'utiliser ce type de matériel, et vérifier s'il a fait l'objet de publications utilisant une méthodologie sérieuse. Les professionnels doivent rester critiques devant les interventions largement diffusées alors que certaines sont chères, inefficaces (ou d'efficacité non prouvée) et chronophages. Il peut y avoir une perte de chance à participer à des séances de rééducation cognitive au détriment d'activités physiques, de loisirs ou d'activités qui permettraient un développement de la socialisation, d'un sentiment de satisfaction issu de la réalisation d'une activité investie voire d'une passion, ou plus simplement de phases de repos permettant une diminution des facteurs de stress qui influencent négativement le développement des FE. Au vu de la littérature publiée, il n'est pas éthique de diminuer les loisirs de l'enfant (sport, musique, etc.) au profit de séances de rééducation dérivées de méthodes adultes (telles le Goal Management Training, le Problem Solving Therapy) qui, pour le moment n'ont pas montré d'efficacité franche chez l'enfant. « La rééducation peut avoir des effets délétères si elle remplace des activités plus bénéfiques à l'enfant que la rééducation elle-même » ²⁶⁴.

Les rééducations doivent être répétées, ludiques, représenter toujours un challenge et ne pas être totalement déconnectées de l'environnement écologique de l'enfant (maison, école) et pour ce faire, l'implication des parents et des intervenants des écoles (enseignants, auxiliaires de vie scolaire, etc.) semble à ce jour le meilleur format pour obtenir une intensité suffisante, par la répétition qui peut ainsi être obtenue. Il y a actuellement un meilleur niveau de preuve pour les méthodes visant à encourager la coopération des neuropsychologues avec les écoles, l'entraînement des équipes à l'école, l'éducation des parents et la formation des auxiliaires de vie scolaire, que pour les méthodes utilisant des interventions décontextualisés de l'environnement naturel de l'enfant, administrées sur des séances hebdomadaires dans un

bureau ^{277,326}. Par contre, l'implication de certaines familles et l'interaction avec les enseignants dans certaines écoles peuvent se révéler problématiques et dans ce cas, il faut faire preuve d'inventivité et une intervention « décontextualisée » dans un bureau de rééducateur pourra se révéler nécessaire pour donner à l'enfant des stratégies et des techniques dont on espère qu'il pourra se les approprier pour les automatiser ultérieurement. Dans tous les cas le contenu de ce type d'approches devrait être davantage évalué, et l'utilisation de méthodes validées privilégiées (ex : utilisation de Cogmed® pour la mémoire de travail, utilisation d'exercices à difficulté croissante et adaptative). Une combinaison des deux approches semble raisonnable à envisager.

Les méthodes de rééducation devraient davantage s'appuyer sur ce qui est utilisé dans d'autres pathologies développementales (ex : adapter une méthode utilisée dans l'autisme ou le TDA/H aux enfants cérébro-lésés) plutôt que d'essayer d'adapter des rééducations d'adultes cérébro-lésés aux enfants, qui risquent d'être inappropriées du point de vue développemental, inefficaces ou non pertinentes pour les difficultés rencontrées par ces enfants.

D'une manière générale, tout protocole de rééducation doit promouvoir et évaluer la généralisation à des activités non travaillées, le transfert à d'autres contextes et situations et le maintien de l'effet à long terme des effets obtenus ^{111-113,329}.

L'idée reçue que seules les études de grande ampleur (peu réalisables en rééducation cognitive de l'enfant) peuvent être publiées est fautive: devant le manque de méthodes validées, de bons journaux internationaux (*Journal of Head Trauma Rehabilitation*, *Developmental Neurorehabilitation*, *Brain Injury* par exemple) publient régulièrement des études même de petit effectif, des études pilotes, ou des cas uniques expérimentaux (méthode N-of-1 trial), du moment que la méthodologie utilisée est appropriée et rigoureuse. La publication de résultats de ce type contribuerait à avancer plus vite dans ce domaine difficile où nous manquons d'outils validés.

ENGLISH ABSTRACTS OF PUBLISHED OR SUBMITTED CHAPTERS

CHAPTER 1: EXECUTIVE FUNCTIONS FOLLOWING SEVERE CHILDHOOD TRAUMATIC BRAIN INJURY – THE TGE PROSPECTIVE LONGITUDINAL STUDY: AGE AT INJURY VULNERABILITY PERIODS

Submitted to the Journal of Head Trauma Rehabilitation, in march 2016

Introduction: Executive functioning (EF) impairment is a major predictor of overall outcome following traumatic brain injury (TBI). While TBI severity is known to be a factor of poorer outcome, most studies include a majority of children with mild and moderate TBI. The aims of this study were to estimate EF impairment after childhood severe TBI and to explore factors predicting EF outcome. Secondary aim was to compare recovery trajectories across age at injury groups.

Methods: Prospective longitudinal study of children (N=65) who sustained severe TBI, tested on EF at 3, 12 and 24 months, by performance-based tests and questionnaires.

Results: Children had significant impairment on working memory, inhibition, attention and global EF, with little or no recovery at 24 months. For flexibility and performance-based EF score, children were impaired at 3 months only and improved to reach normal scores by 12 months. No impairment was found on planning. Global EF was predicted at 3 and 24 months by Glasgow Coma Score and parental education. Children aged 10-12 at injury showed better EF outcome and better EF progress than the other age groups. Coma duration predicted EF at 3 months but was no longer a significant predictor at 24 months.

CHAPTER 2: ECOLOGICAL PROSPECTIVE MEMORY ASSESSMENT IN CHILDREN WITH ACQUIRED BRAIN INJURY USING THE CHILDREN'S COOKING TASK

Published in Developmental Neurorehabilitation Journal

Ecological prospective memory assessment in children with acquired brain injury using the Children's Cooking Task. Krasny-Pacini A, Servant V, Alzieu C, Chevignard M. **Dev Neurorehabil.** 2015 Jul 29:1-6.

Prospective memory (PM) has been shown to be impaired in children with acquired brain injuries (ABI) and is a major concern for parents. Few studies have addressed this issue and most used tasks that are not ecologically valid. The aims of this study were (1) to explore if children who have sustained an ABI suffer PM impairment, measured both by the Children's Cooking task (CCT) PM score and using the two PM subtests of the Rivermead Behavioural Memory Test (RBMT), and (2) to explore if the CCT PM score is sensitive to developmental changes in PM in typically developing children and in children with ABI. Fifty-four children with ABI and 33 typically developing controls participated in the study. Children with ABI had significantly lower PM scores and poorer performance in the CCT than their typically developing peers. PM scores increased significantly with age, indicating developmental progress of PM performance.

CHAPTER 3: PROSPECTIVE MEMORY SEVEN YEARS AFTER CHILDHOOD SEVERE TRAUMATIC BRAIN INJURY - THE TGE 2 COHORT

To be submitted

Objective: to investigate long term outcome in prospective memory (PM), seven years after childhood severe traumatic brain injury (TBI), in a prospective longitudinal cohort. Design: inception cohort. Setting: individuals from the general community, with severe TBI identified seven years earlier from a single, regional, University pediatric neurosurgical intensive care unit. Participants: 76 young individuals (aged 7-22): 39 patients with a severe accidental TBI (defined as Glasgow Coma Score GCS of 8 or lower), aged 0-15 years at injury, and 37 population-based controls individually matched on age, gender and parental education. Main

outcome measures: three novel short PM tasks varying in the delay (long versus short), motivation (high versus low motivation) and context (ecological versus office-based). Interventions: Not applicable. Results: individuals with sTBI showed significantly poorer PM than matched controls in the two low-motivation PM tasks: (1) The ecological long-delay task consisting of sending a letter on a rainy day, showed significant differences both in its PM component ($p=0.047$, odds ratio = 2.6), and the retrospective memory component ($p=0.01$, odds ratio= 3.7). (2) The non-ecological short-delay task consisting of taking off post-its while identifying facial emotions showed a significant medium effect ($p=0.004$, $r=0.34$). PM performance on the high motivation task did not differ from controls. Conclusions: PM is impaired in individuals long-term post childhood sTBI. High motivation conditions may enhance PM in individuals with sTBI. Key words (MeSH): brain injuries, child, cohort studies, memory

CHAPTER 4: SELF-AWARENESS ASSESSMENT DURING COGNITIVE REHABILITATION IN CHILDREN WITH ACQUIRED BRAIN INJURY: A FEASIBILITY STUDY AND PROPOSED MODEL OF CHILD ANOSOGNOSIA

Published in Disability and Rehabilitation Journal

Self-awareness assessment during cognitive rehabilitation in children with acquired brain injury : a feasibility study and proposed model of child anosognosia. Krasny-Pacini A, Limond J, Evans J, Hiebel J, Bendjelida K, Chevignard M. **Disabil Rehabil.** 2015;37(22):2092-106.

Children with brain injuries and development disabilities can have impaired awareness due to both developmental immaturity and organically based unawareness/anosognosia. Although awareness of deficits is thought to be essential for effective cognitive rehabilitation, there is a lack of clinical models of awareness in children. This paper presents a model of cognitive awareness in children, based on dissociations found in a pilot study of five single cases of children with traumatic brain injury, aged 8 to 14, undergoing metacognitive training. Comprehensive assessment of children's awareness of cognitive impairments was performed using three types of awareness measures showing large awareness variability. We propose a framework for awareness assessment in cognitive rehabilitation, drawing on developmental,

neuropsychological and learning disability literature. The model emphasizes the role of on-line error detection in the construction of autobiographical memories that allow a child to build a self-knowledge of his/her strengths and difficulties. Implications for rehabilitation are discussed.

CHAPTER 6: GOAL MANAGEMENT TRAINING FOR REHABILITATION OF EXECUTIVE FUNCTIONS: A SYSTEMATIC REVIEW OF EFFECTIVENESS IN PATIENTS WITH ACQUIRED BRAIN INJURY.

Published in Disability and Rehabilitation Journal

Goal Management Training for rehabilitation of executive functions: a systematic review of effectiveness in patients with acquired brain injury. Krasny-Pacini A, Chevignard M, Evans J. **Disabil Rehabil.** 2014;36(2):105-16. Epub 2013 Apr 18. Review.

PURPOSE: To determine if Goal Management Training (GMT) is effective for the rehabilitation of executive functions following brain injury when administered alone or in combination with other interventions.

METHOD: Systematic review, with quality appraisal specific to executive functions research and calculation of effect sizes.

RESULTS: Twelve studies were included. Four studies were "Proof-of-principle" studies, testing the potential effectiveness of GMT and eight were rehabilitation studies. Effectiveness was greater when GMT was combined with other interventions. The most effective interventions appeared to be those combining GMT with: Problem Solving Therapy; personal goal setting; external cueing or prompting apply GMT to the current task; personal homework to increase patients' commitment and training intensity; ecological and daily life training activities rather than paper-and-pencil, office-type tasks. Level of support for GMT was higher for studies measuring outcome in terms of increases in participation in everyday activities rather than on measures of executive impairment.

CONCLUSION: Comprehensive rehabilitation programs incorporating GMT, but integrating other approaches, are effective in executive function rehabilitation following brain injury in adults. There is insufficient evidence to support use of GMT as a stand-alone intervention.

CHAPTER 7: CONTEXT-SENSITIVE GOAL MANAGEMENT TRAINING FOR EVERYDAY EXECUTIVE DYSFUNCTION IN CHILDREN AFTER SEVERE TBI.

Published in the Journal of Head Trauma Rehabilitation

Context-sensitive goal management training for everyday executive dysfunction in children after severe traumatic brain injury. Krasny-Pacini A, Limond J, Evans J, Hiebel J, Bendjelida K, Chevignard M. **J Head Trauma Rehabil.** 2014 Sep-Oct;29(5):E49-64.

Objectives: To assess the effectiveness of a metacognitive training intervention, based on an adapted Goal Management Training (GMT) and on Ylvisaker's principles, on three activity domains of executive functions: (1) prospective memory (PM) performance in ecological setting, (2) complex cooking task management, (3) daily executive functioning (EF) at home and at school. **Participants:** Five children aged 8-14, who were 3-11 years post severe traumatic brain injury (TBI), experiencing severe EF difficulties in daily life. **Design:** Single-case experimental design, and assessment of EF twice prior to intervention, post-intervention, 3- and 6-months post-intervention. Progress was monitored by a weekly ecological PM score. The effect on EF was assessed using the Children's Cooking Task (CCT). Transfer to the child's natural context was assessed by parental and teacher questionnaires and Goal Attainment Scaling. **Results:** All children improved on the measure of PM and on questionnaires of daily EF. Two children improved on the CCT but returned to their pre-intervention level in a novel cooking task at follow-up. School personnel and parents' participation in the program was low. **Conclusions:** It is feasible but challenging to use GMT in children with TBI. Further research is needed in relation to how to promote generalization and how to increase the involvement of the child's "everyday people" in the intervention.

LONG SUMMARY OF THE PHD IN FRENCH

Fonctions Exécutives, Memoire Prospective et Métacognition chez l'enfant traumatisé crânien: évaluation, caractérisation et rééducation.

Le traumatisme crânien (TC) sévère de l'enfant est un enjeu de santé publique de par sa fréquence (incidence annuelle du TC, toutes gravités confondues, estimée à 864/100 000, dont environ 10% de TC sévères) et les séquelles générées. Même si on assiste souvent à une relativement bonne récupération sur le plan moteur, des déficits cognitifs sévères et invalidants sont fréquents, concernant de nombreux domaines, dont les fonctions exécutives (FE). Les FE se définissent comme un ensemble de fonctions cognitives élaborées intervenant dans le comportement intentionnel, organisé, volontaire, dirigé vers un but et la gestion des activités non routinières. A l'heure actuelle, aucune rééducation spécifique des FE n'a scientifiquement prouvé son efficacité chez l'enfant cérébro-lésé.

Cette thèse explore deux aspects particuliers des FE : la métacognition (capacité à réfléchir à sa propre cognition, monitorer sa pensée, la contrôler et être conscient de ses limites lors de l'exécution d'une tâche) et la mémoire prospective, c'est à dire la capacité à se rappeler ce que l'on doit faire à un moment donné. Ces deux aspects sont particulièrement importants pour les techniques de rééducation fondées sur le modèle de la négligence du but (« goal neglect »), utilisé dans cette thèse. Ce modèle considère que syndrome dysexécutif et difficultés de mémoire prospective sont des présentations similaires du phénomène du « goal neglect ». Le « Goal Management Training » (GMT) se fonde sur ce modèle et tente de réduire le syndrome dysexécutif en améliorant le maintien en mémoire prospective de l'objectif principal d'une tâche donnée. Le GMT utilise la métacognition pour compenser les difficultés exécutives et lutter contre la négligence du but, et semble efficace chez les adultes dysexécutifs cérébro-lésés. Chez l'enfant, dont la métacognition ne mature que très tard (à l'adolescence voire au début de l'âge adulte), ce type d'approche pourrait donc ne pas être

approprié. D'autre part il y a actuellement très peu de données sur les troubles de la mémoire prospective chez les enfants cérébro-lésés.

La première section de cette thèse explore les aspects controversés ou incomplètement connus concernant l'atteinte des FE après TC de l'enfant : les périodes de vulnérabilité et les altérations des FE en fonction de l'âge de survenue du TC (chapitre 1), les conséquences d'un TC sévère sur la mémoire prospective, notamment en contexte écologique (chapitres 2 et 3), les conséquences du TC sur les capacités métacognitives (chapitre 4). Cette première partie permet premièrement de constater que les FE sont plus sévèrement atteints lorsqu'elles sont dans une période de développement rapide et que l'utilisation de tests papier-crayon (pourtant largement utilisés pour évaluer, dépister et suivre le syndrome dysexécutif de l'enfant) sous-estiment largement les conséquences du TC sur la vie quotidienne (chapitre 1). Deuxièmement, elle confirme les conséquences du TC et du syndrome dysexécutif sur la mémoire prospective, en montrant que les difficultés de mémoire prospective sont de sévérité telle que les adolescents cérébro-lésés ont des difficultés similaires à celles rencontrées par les jeunes enfants sains, âgés de 8-10 ans (chapitre 2) et que ces difficultés persistent à 7 ans post TC (chapitre 3). Le chapitre 4 présente les résultats pilotes d'une évaluation multimodale de la métacognition et montre des dissociations de l'atteinte de ses différents composants chez des enfants cérébro-lésés. Il soulève les difficultés potentielles liées à l'utilisation des rééducations métacognitives chez de jeunes enfants, particulièrement s'ils sont anosognosiques du fait de leur TC. Ce chapitre propose également un modèle d'anosognosie de l'enfant, permettant de mieux adapter la rééducation cognitive aux capacités métacognitives de l'enfant.

Suite à la mise en évidence des effets d'un TC survenu dans l'enfance sur les FE et la mémoire prospective, la section 2 de cette thèse tente de proposer des moyens d'améliorer ces fonctions. Le chapitre 5 présente un état des lieux des interventions visant à améliorer les fonctions exécutives chez l'enfant. Le chapitre 6 (revue systématique de la littérature) examine l'efficacité du *Goal Management Training* chez l'adulte cérébro-lésé, dans l'objectif de pouvoir ensuite l'adapter à l'enfant. Il propose une revue critique des études publiées en proposant des critères d'évaluation spécifiques à prendre en compte dans les études testant une intervention de rééducation des FE. Les chapitres 7 et 8 décrivent l'adaptation du GMT à l'enfant et en explorant son efficacité sur la mémoire prospective et les FE, et en particulier

les capacités d'adaptation à une tâche nouvelle. Cette étude confirme la difficulté de rééduquer les FE, et alerte sur le risque de juger efficaces des interventions au cours desquelles les sujets s'améliorent par effet d'apprentissage des tests et effet d'enthousiasme, sans amélioration de leurs réelles capacités à utiliser efficacement leurs FE en situation de tâche nouvelle. Elle alerte également sur la nécessité de mieux prendre en compte les aspects développementaux chez l'enfant cérébro-lésé, concernant l'anosognosie (basé sur le chapitre 4) et l'utilisation de stratégies métacognitives (chapitre 8).

En conclusion, les difficultés exécutives et de mémoire prospective après TC de l'enfant sont souvent sous-estimées du fait de l'utilisation d'évaluations inadéquates ou trop peu « écologiques ». Les rééducations basées sur la métacognition ne semblent pas être le meilleur moyen d'améliorer les FE, en particulier chez les enfants jeunes, présentant une anosognosie importante et des troubles sévères des FE.

Résumé :

Fonctions Exécutives, Mémoire Prospective et Métacognition chez l'enfant traumatisé crânien (TC): évaluation, caractérisation et rééducation.

La première section permet premièrement de constater que les fonctions exécutives (FE) sont plus sévèrement atteintes lorsqu'elles sont dans une période de développement rapide et que l'utilisation de tests papier-crayon sous-estime largement les conséquences du TC sur la vie quotidienne. Deuxièmement, elle confirme les conséquences du TC et du syndrome dysexécutif sur la mémoire prospective. Enfin, elle présente les résultats pilotes d'une évaluation multimodale de la métacognition et montre des dissociations de l'atteinte de ses différents composants chez des enfants cérébro-lésés. Elle propose également un modèle d'anosognosie de l'enfant, permettant de mieux adapter la rééducation cognitive aux capacités métacognitives de l'enfant. La section 2 de cette thèse tente de proposer des moyens d'améliorer les FE. Une revue systématique de la littérature examine d'abord l'efficacité du Goal Management Training (GMT) chez l'adulte cérébro-lésé. Le travail expérimental décrit ensuite l'adaptation du GMT à l'enfant. Cette étude confirme la difficulté de rééduquer les FE, avec des résultats contradictoires entre les critères de jugement (certains enfants s'améliorant sur la tâche écologique de « gâteau au chocolat » et d'autres sur les questionnaires de FE). De plus, quand ils étaient confrontés à une nouvelle recette de cuisine, les enfants revenaient à leur comportement dysexécutif, suggérant que la fonction d'adaptation à la nouveauté n'avait pas progressé. Enfin les enfants utilisaient peu les stratégies métacognitives du GMT. La discussion alerte sur la nécessité de mieux prendre en compte les aspects développementaux chez l'enfant cérébro-lésé, concernant l'anosognosie et l'utilisation de stratégies métacognitives. Elle propose également des critères de jugement et de méthodologies innovants pour la recherche sur la rééducation des FE de l'enfant. En conclusion, le TC de l'enfance provoque des difficultés de FE qui persistent à l'âge adulte et pour lesquelles, à l'heure actuelle, il y a peu de traitements dont l'efficacité soit clairement démontrée.

Mots clefs: Fonctions Exécutives, Mémoire Prospective, Traumatisme Crânien, enfant, écologique, anosognosie, métacognition, Goal Management Training

Executive Functions, Prospective Memory and Metacognition in children with traumatic brain injury (TBI): assessment, characterization and rehabilitation

Part one of this PhD explored executive functions (EF) and related impairments after childhood severe TBI. Executive dysfunction was worse when the injury occurred during the period of rapid development of EF. Paper and pencil tasks underestimated EF impairment in daily life. Impairment in prospective memory persisted at seven years post injury, and into adulthood. A model of child anosognosia, and directions for cognitive rehabilitation interventions, are proposed based on a study of self-awareness after childhood TBI. Part two of this PhD investigated methods of remediating EF dysfunction. A systematic review found that Goal Management Training (GMT) is effective in adults when coupled with personal goal setting, external cueing, personalised homework to increase patients' commitment and training intensity and daily life training activities. Based on this literature review, a paediatric GMT intervention was adapted and tested. The intervention showed limited effectiveness, with inconsistency across outcome measures. Children did not apply the metacognitive strategies taught during the training. Furthermore, there was no evidence of generalisation of task-specific improvements to a novel, parallel version of the secondary outcome measure (the Children's Cooking Task). The adapted GMT did not seem developmentally coherent for younger children. It is clear that deficits in EF arising from TBI continue to be an important target for rehabilitation interventions in children. Proposals on how the use of novel innovative outcome measures and methodologies may improve the field of EF rehabilitation in children with TBI are presented.

Keywords: Executive functions, Prospective memory, Traumatic/Acquired brain injury, child, intervention, ecological, self-awareness, anosognosia, metacognition, goal neglect, Goal Management Training