

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

Agata Krasny-Pacini, Jenny Limond, Jonathan Evans, Jean Hiebel, Karim Bendjelida, Mathilde Chevignard

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1 TITLE PAGE

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3 Context-sensitive Goal Management Training for everyday executive dysfunction in children after severe TBI.

4 AUTHOR NAMES AND HIGHEST ACADEMIC DEGREE

Krasny-Pacini Agata^{1,4} MD, Limond Jenny³ PhD, Evans Jonathan³, PhD, Hiebel Jean¹ MD, Bendjelida Karim⁵ MD,
 Chevignard Mathilde^{2,4}, MD, PhD

- 7 Affiliations:
- Institut Universitaire de Réadaptation Clemenceau-Strasbourg, 45 bd Clemenceau, 67082 Strasbourg,
 France Paediatric Department and Hautepierre Strasbourg University Hospital, Strasbourg. France
- Rehabilitation Department for Children with Acquired Brain Injury, Hôpitaux de Saint Maurice, 14, rue
 du Val d'Osne, 94410 Saint Maurice
- Mental Health and Wellbeing, Institute of Health & Wellbeing, University of Glasgow, Gartnavel Royal
 Hospital, 1055 Great Western Rd, Glasgow G12 0XH , Scotland, UK
- 14 4. ER 6-UPMC, University Pierre et Marie Curie Paris 6, Pitie-Salpêtrière Hospital, Paris, France
- 15 5. EMOI-TC 68, Hôpital du Hasenrain, 87 avenue d'Altkirch 68051 MULHOUSE CEDEX, France

16

- 17 Corresponding author : Agata Krasny-Pacini agatakrasny@yahoo.com; 0033671284152
- 18 Institut Universitaire de Réadaptation Clemenceau-Strasbourg, 45 bd Clemenceau, 67082 Strasbourg, France

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Context-sensitive Goal Management Training for everyday executive dysfunction in children after severe TBI.

37 **ABSTRACT**

Objectives: To assess the effectiveness of a metacognitive training intervention, based on an adapted Goal 38 39 Management Training (GMT) and on Ylvisaker's principles, on three activity domains of executive functions: (1) 40 prospective memory (PM) performance in ecological setting, (2) complex cooking task management, (3) daily 41 executive functioning (EF) at home and at school. Participants: Five children aged 8-14, who were 3-11 years 42 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 43 44 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 45 46 teacher questionnaires and Goal Attainment Scaling. Results: All children improved on the measure of PM and 47 on questionnaires of daily EF. Two children improved on the CCT but returned to their pre-intervention level in 48 a novel cooking task at follow-up. School personnel and parents' participation in the program was low. 49 Conclusions: It is feasible but challenging to use GMT in children with TBI. Further research is needed in 50 relation to how to promote generalization and how to increase the involvement of the child's "everyday 51 people" in the intervention.

- 52 *Keywords:* Executive functions, Prospective memory, Traumatic/Acquired brain injury, child, intervention,
- 53 ecological, daily life activities, Goal Management Training

54 INTRODUCTION

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Executive functions (EF) are a collection of related but distinct abilities that allow individuals to engage
efficiently in intentional, goal-directed problem-solving actions^{1,2} through conscious and effortful processing³ and
to adapt to new situations in the real world ⁴. Executive functioning (EF) deficits are a frequent consequence of
traumatic brain injury (TBI) ⁵. TBI outcome is predicted by executive functioning level⁶.

There is a lack of validated methods for EF rehabilitation in children ^{7,8,9}, although some general rehabilitation principles have proven to be useful . Ylvisaker¹⁰ 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 Braga¹¹ and Wade^{12,13} for examples of family-delivered interventions in TBI); (2) the necessity of a "context-sensitive" approach¹⁴, embedded in functional routines of everyday life and using meaningful activities rather than decontextualized exercises.

65 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 66 67 prompting devices) – though only a restricted number of situations lend themselves to such an approach; (2) 68 Training component EF skills (e.g. repeated exercises aiming at changing the brain's working memory capacity 69 for example) - though transfer to natural contexts and generalization to untrained activities effects of this 70 approach have rarely been demonstrated¹⁴; (3) Training children on specific goals (e.g. if preparing a schoolbag 71 is problematic, the child will be trained on this specific activity until the goal is achieved) – though 72 generalization to similar goals in different situations (e.g. preparing a suitcase for holidays) is often not 73 achieved; (4) Providing children with metacognitive strategies applicable to a variety of everyday situationswhilst this is effective in some adults after TBI¹⁵, there is little evidence that metacognitive training is effective 74 for children with a dysexecutive syndrome post TBI¹⁶, though research on children with other forms of brain 75 injury suggests it may be a useful approach ^{16 17}. 76

Goal Management Training (GMT)¹⁸ is one of the most studied metacognitive training programs, of which many
variants exist¹⁹. 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.²⁰ for a more detailed description). GMT was developed from Duncan's theory of "goal 81 neglect"^{21,22}, which suggests that dysexecutive patients are impaired in the construction and use of "goal lists", 82 83 necessary for goal-directed behavior. They do remember the intended goal but tend to lose sight of it as they 84 progress through a task leading to a prospective memory failure. Prospective memory (PM) (remembering to 85 carry out intended actions) tasks require retrospective memory to remember the task, but depend on EF²³ for successful goal maintenance, retrieval and implementation at the right moment. PM depends upon frontal lobe 86 integrity²⁴, with a key role for rostral prefrontal cortex (BA10)²⁵. In typically-developing children aged 6-12, 87 performance on EF tasks such as planning and switching ²⁶, working memory ²⁷, and inhibition ²⁸ is correlated 88 with PM ²⁹. PM problems are reported as a major concern for the parents of children with TBI ³⁰. PM is 89 impaired in children with TBI ^{31,32} compared to children with orthopedic injuries, even after cues are given ³³, 90 91 and even under strong incentive conditions³⁴.

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.

98 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.

- 102 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)
- 104 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.

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106 Exclusion criteria were: (1) diagnosed learning disabilities, neurologic or psychiatric condition prior to TBI; (2)

107 severe intellectual disability; (3) insufficient French language level of the child or of his/her family.

108 Characterization data included classical standardized tests from the Wechsler Intelligence Scale for Children

109 (WISC-IV)³⁵, from the Children's Memory Scale³⁶ and a French battery of EF for children: "Fonctions Exécutives

110 Enfant" battery (FEE)³⁷. FEE has a much larger normative data than any other EF test available in French. Each

111 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 method ³⁸ with one-tailed probability³⁹ taking
 p<0.05 criteria for significance.

114 Intervention

115 The intervention was inspired by Goal Management Training¹⁸ (GMT) but extended to follow Ylvisaker's

116 principles of involving "everyday people" in the child's social network in the cognitive coaching of the child and

117 using a context-sensitive approach prioritizing functional ecological activities.

118 Prior to the intervention, parents participated in a two to three hour informal interview during which the

119 following were discussed: 1) the child's daily life difficulties at home and at school; 2) program content and key

120 GMT concepts; 3) the need to apply the metacognitive GMT techniques at home and at school. An emphasis

121 was put on the key role parents played for the success of the intervention.

122 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

124 activities; (3) an "everyday people cognitive coaching guide". The materials included PowerPoint slides, a

125 workbook, posters for home and school use, mission sheets and the cognitive coaching guide. The 15 modules

126 (theoretical modules+ meaningful activities) of the intervention required 15-20 hours to be completed, and

127 were administered individually over a four to six-month period on a weekly basis, either in a rehabilitation

128 centre or at home.

129 (1) Theoretical modules were derived from the adult GMT PowerPoint Manual ²⁰ developed by Levine,

130 Robertson and Manly, that has already been used (with minor changes) in children⁴⁰. A new, shorter, colourfull

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131 version was created for the intervention to make materials child-friendly, age-appropriate, enjoyable and 132 simpler. For example, "slips" (referring to slips of attention) became "Oops errors". "To Do lists" and the 133 "mental blackboard" were combined in a unique "note book" concept in order to explain to the children how a 134 real paper note book can help them not to overwhelm their "mental notebook". Discussion about EF failures in 135 daily life was triggered through illustrated stories relating to school and leisure activities. In each module we 136 included a prospective memory task to be performed during the session (e.g. when you see a slide with X you 137 do Y) to encourage discussion about PM failures at the end of each session. The content-free cue "Look into 138 your mental notebook" was used as a prompt when children failed PM tasks. Throughout the training period 139 children had to complete "mission sheets", inspired from GMT between-session assignments. These were of 140 three types, introduced progressively: 1) monitoring Oops errors, their consequences and factors influencing 141 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 142 143 to manage a goal (preparing the school bag, preparing a sandwich).

144 (2) Practical modules served to practice GMT content in meaningful activities (Ylvisaker's Content-sensitive 145 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 146 147 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 activities⁴¹. The 148 149 activities involved planning, strategy generation, following steps, and monitoring of actions. Difficulty increased 150 as children progressed in the program. Similarly to the theoretical modules, the meaningful activities 151 contained naturally occurring prospective memory tasks to allow discussion about PM difficulties in real 152 activities (e.g. checking regularly if the first set of finger biscuits is cooked while preparing the next). The trainer 153 guided the children when needed, using non-specific prompts and general cues. Explicit help was only given if 154 these were not sufficient. At the end of each activity, the child was invited to review his/her performance using 155 the "mission" sheets, identify Oops errors and any effective strategy that had helped in the task, and to think 156 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.

159 (3) To encourage transfer to the child's natural contexts, we tried to involve the child's 'everyday people' as 160 cognitive coaches for their child. Everyday people were parents, teachers, school assistants and any adult the 161 parents identified as a potential cognitive coach (baby-sitter, student helping the child with homework). A 162 letter presenting the program was sent to the child's teacher and school assistant (SA), explaining briefly TBI 163 executive problems and their implications, and asking the teacher and the SA to participate by applying GMT at 164 school. The intervention content was not explained orally. We asked for a contact e-mail and a telephone 165 contact to discuss the child's difficulties and set realistic goals on goal attainment scales. The letter was sent a 166 second time after one month as the first yielded few responses, so we had responses from at least one school 167 staff member per child. Other potential 'everyday people' identified by the parents were sent a similar letter. 168 Twice a month all 'everyday people' who agreed to participate received one chapter, two-pages long, of a 169 "Cognitive Coaching Guide" that was created for the intervention. The guide was colorful, using the same 170 drawings, diagrams and analogies as the theoretical modules, explaining the rehabilitation content and 171 suggesting how to apply metacognitive strategies at school and at home. The intervention was organized in a 172 way that each metacognitive strategy was (1) first introduced during a theoretical module, (2) then practiced 173 on a meaningful activity and (3) lastly introduced to everyday people. They were sent the corresponding 174 chapter of the coaching guide that described the activity for which the child had already practiced the strategy, 175 and suggested other activities to which it could be applied (see supplemental digital data for examples). 176 Through this guide, everyday people were encouraged: (1) to use non-specific prompts for PM failures ("look 177 into your mental note book") rather than specific instructions ("you need to feed the dog") or negative 178 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"¹⁰; 179 180 (3) to prompt and help the child to fill in his/her "mission sheets" regularly; (4) to practice goal identification 181 ("state your goal") and stepwise processing in daily activities (preparing schoolbag, table setting). Parents were 182 explicitly asked to go through the metacognitive strategies of the cognitive coaching guide and to sign the

183 child's GMT workbook every week. The 'everyday people' were not requested to participate in the

184 rehabilitation session, but were advised that the therapist was available if they had any questions.

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.

189 Study design. Intervention effectiveness was assessed in two ways: (1) A Single-Case Experimental design (SCED)^{42,43}, with repeated ecological measurement of prospective memory was used to monitor progress 190 191 throughout the intervention; (2) pre-post measurement of EF with two baseline assessments 4-8 weeks apart 192 (B1 and B2), and three post-intervention assessments: immediately after the intervention (R1), and at three 193 (R2) and six months (R3) to assess maintenance of effects. Baseline was expected to be stable as children had 194 sustained their TBI at least two years earlier, minimizing chances of spontaneous recovery during the study. 195 The pre-post measurement served to capture the key issues of EF rehabilitation, namely: (1) EF performance in 196 ecological tasks; (2) transfer of training effects to natural contexts; (3) generalization to untrained tasks; (4) 197 need for novelty for a task to truly test "executive" functions.

198 Outcome Measures

199 Repeated measures of prospective memory (PM) performance - SCED design: We monitored the effectiveness 200 of the program through a weekly score on a time-based PM task. The PM task was inspired by the Fish et al. 201 phone call task ⁴⁴ that has recently been used in children ⁴⁵. 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 202 203 time, either as a text message, an e-mail or a phonecall. The child was awarded three points if correct 204 information was given within one hour of the agreed time, two points if within the day, one point if on a 205 different day and zero points if the child completely forgot about the task. The retrospective memory 206 component was controlled for by checking at each session that children remembered the task and agreed 207 times. Parents were given the timings in case the child wanted to check the target time. To encourage use of 208 mental strategies, children were asked not to use cues such as alarms, or pre-programmed text messages.

209 Parents were instructed not to give any cues or help to complete the task. The three target days and times 210 were chosen individually for each child with the parents before the first assessment to ensure: (1) that the child 211 was easily available for the task (i.e. not during school time or leisure activity); (2) that it didn't disturb family 212 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 213 214 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 al⁴⁴, days when the tasks had not been performed for another reason than PM failure, 215 216 were not used in the analysis (medical appointment at target time, no internet connection during a week-end 217 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 218 improve EF in daily life, assessment included an ecological⁴⁶ test of EF, called the Children's Cooking Task 219 (CCT)^{47,48}. In the CCT, children have to prepare a real chocolate cake and a fruit cocktail following a structured, 220 221 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 222 223 goal management and innovative higher level strategies to succeed. The Children's Cooking Task (CCT) has 224 good inter-rater and test-retest reliability, high internal consistency, as well as good discriminant and 225 concurrent validity⁴⁷. It can be performed from the age of 8. Scoring is based on the number of errors, including 226 omissions, additions, commentaries, substitution and estimation errors. Normative data is not yet available. 227 Therefore raw scores were used to track individual child changes. The number of errors in CCT made by each 228 child was compared to the number of errors made by age matched healthy controls, extracted from 229 unpublished data⁴⁹.

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 child¹⁰, 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^{50, 51}, completed by both parents

235 (transfer to home-context) and teachers (transfer to school-context); (2) a cognition subscore derived from the Dysexecutive Questionnaire for Children (DEX-C)⁵² that was completed by parents only. The BRIEF assesses 236 237 eight domains of executive functioning in the real world, which give together a Global Executive Composite 238 (GEC) score. Higher scores correspond to increased EF difficulties in daily life. A T-score superior to 65 is 239 defined as the clinical range. The BRIEF has large normative data for children aged 5 to 18, high internal consistency⁵³, good validity ⁵⁴ and good test-retest reliability⁵³ although parent-teacher agreement is only 240 moderate⁵³. It is the most commonly used questionnaire of executive functions and seems to be sensitive to 241 deficits in executive functioning in children with TBI ^{54,55,56,57}. Its relationship with common EF cognitive tests is 242 243 however inconsistent. DEX-C is a 20 item questionnaire, probing four broad areas of EF difficulties 244 (emotional/personality, motivational, behavioural and cognitive) and is part of the "Behavioural Assessment of the Dysexecutive Syndrome for Children" ⁵². Higher Z-scores correspond to increased cognitive difficulties 245 relating to EF. DEX-C has less evidence regarding psychometric properties⁵². However the cognitive subscale of 246 247 DEX-C completed by parents has a high correlation with the (Children's Cooking Task) CCT score⁴⁷, and 248 therefore this subscale together with the CCT were expected to detect improvement in cognitive EF 249 impairment, which the BRIEF might not capture.

250 <u>Goal Attainment Scaling (GAS) – pre-post measurement of generalization (metacognitive strategy use in</u>

untrained tasks): GAS^{58,59} was used as a generalization measure to assess if a child who has applied 251 252 metacognitive strategies to meaningful activities in a rehabilitation setting is capable of applying those 253 strategies to untrained tasks that are judged to be problematic for him/her by his/her everyday people. EF-254 related problems reported by the child, parents and school staff served to elaborate personalized goal 255 attainment scales (GAS) for each child. These GAS goals were not trained specifically but children were 256 repeatedly encouraged to apply metacognitive strategies to daily life. We also used "general" GAS for 257 metacognitive strategy use and GMT application (see table 3 in results section). Themes of "general" goals 258 were similar for all children, but the initial levels and expected outcome levels were specific to each child, 259 taking into account children's age and possibilities. The detailed procedure for goal selection, GAS elaboration and GAS levels adjustment are described in supplemental digital data. GAS scores were used to calculate a 260 global T-score for each child, using Kiresuk's formulae (see ^{58, 59} for details of GAS methodology). A T-score of 50 261

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meant that the goals were overall attained as expected, and > 50 that goals were attained better than
 expected.

264 "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^{60,61}. When the same task is repeated 265 after intervention, it is more "familiar", which can make it less demanding on EF^{62,63}. Familiarity effects increase 266 267 when patients are tested on several occasions (as it is the case in our design for CCT). To get a "purer" EF 268 measure post-intervention⁶³, we developed a parallel form of the Children's Cooking Task (CCT) for assessment 269 at R3, involving the same number of steps and ingredients but requiring different types of ingredients and 270 procedures. This version has no established psychometric properties. Children had to bake "Christmas 271 biscuits". Whilst both tasks required cooking, as children were not experienced cooks, a new recipe could not 272 be viewed as a familiar task. 273 Controlling for confounding factors. At the beginning of the program everyday people were not informed of 274 exactly when the intervention component would commence: from the first interview onwards all children were 275 seen weekly, whereas the intervention started only 5-8 weeks later. We hoped to control in this way for 276 parent's enthusiasm for a novel rehabilitation program, which was expected to be reflected by an 277 improvement between B1 and B2 in this study design. The intervention effect was measured comparing post 278 intervention results (R1, R2 and R3) to the second baseline (B2), as this was considered a "purer" baseline 279 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 280 281 the questionnaire unreliable because of contradictory answers on special items serving to assess consistency of 282 answers). Furthermore, the intervention did not significantly change the amount of time spent in 283 rehabilitation: all children had already been attending the outpatient department for half a day a week for 284 many years (including sports, group games and group discussion to promote socialization and language 285 pragmatics for PB,CS,RK; analytic psychotherapy for PB, paper-and pencil neuropsychological exercises aiming 286 at improving attention for IP) therefore the effect of the potentially confounding factor of time spent with 287 therapist was considered likely to be negligible.

288 External investigator post-intervention interview. After the intervention, an external interviewer, who had 289 neither been involved in the rehabilitation nor the research team called all the everyday people involved in the 290 program. A structured interview focused on how they perceived the program, their views on applying cognitive 291 coaching at home and at school, clarity of the cognitive coaching guide, how children reacted to the 292 intervention and if they thought their child had improved in various domains (autonomy, school results...) even 293 if it was a domain not included in GAS and questionnaires. The interview contained embedded questions aimed 294 at quantifying how much the everyday people participated in the cognitive coaching and at checking if they 295 understood the concepts that were explained to them in the cognitive coaching guide. They were asked for 296 examples of metacognitive strategies they could recall, situations they applied them to, and were asked how 297 often they managed to go through the child's workbook together, and if GMT posters had been hung at home. 298 Feedback from the child was obtained informally from the first author conducting the intervention, because 299 answering to an unknown external investigator on the phone was considered age-inappropriate.

300 Statistical analysis and effect size (ES) calculation. The Saint's day task (SDT) PM scores were visually analyzed 301 on time series graphs comparing baseline (weeks 1-4), with intervention (weeks 5-18). A two-standard 302 deviation band (2SDB) was determined for each child based on the standard deviation of the four baseline 303 points. Gottman and Leiblum's criterion was used: the probability that two consecutive points fall outside the 304 2SDB is < 0.05. (see for details ⁴²). Trends were detected by celeration lines, using the Split Middle Trend line 305 procedure⁴². To obtain the magnitude of effect, the "nonoverlap of all pairs" (NAP) method was used ⁶⁴, 306 through SPSS software. For the other outcome measures, an intervention effect size (ES) was calculated for 307 each child from B2 to R1, R2 and R3 as a standard difference between T-scores divided by 10 for the BRIEF and 308 as a standard difference between Z-scores for the DEX-C Cognition scale. For the CCT, ES was obtained by 309 dividing the score difference by the standard deviation of all five CCT scores of the child. Furthermore, because 310 the CCT inevitably has a practice effect as the recipe becomes more known, which could account for 311 improvement throughout the trials, we readjusted ES by subtracting the practice effect of each child (score 312 change between B1 and B2 being considered as the practice effect for that child). ES were interpreted subjectively with reference to Cohen's guidelines⁶⁵: 0.2 = small; 0.5 = medium; 0.8 = large. 313

314 **RESULTS**

315 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 316 317 schooling, either attending a special support class or having a school assistant. Characteristics of the 318 participants are summarized in table 1. All children had a severe dysexecutive syndrome on paper-and-pencil 319 EF tests (see table 1bis in Supplemental Digital Content), on CCT (see B1 and B2 scores in Figure 2), and (apart 320 from child IP' BRIEF score) on EF-questionnaires (see B1 and B2 in Figures 3 and 4). In contrast they were not 321 impaired in reasoning abilities (apart from CS) or retrospective memory (see table 1ter in Supplemental Digital 322 Content).

323

table 1 : attached at the end of the paper

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 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.

329 Qualitative data about the program. The intervention appeared to be feasible to implement and it was 330 reported that children enjoyed it, especially the meaningful activities and stories used in the theoretical 331 modules. Most GMT concepts were understood by the children, although examples of personal cognitive 332 failures were difficult to obtain. Interestingly, children seemed to consider the metacognitive strategies as 333 exercises to practice rather than something that could be applied to other tasks. As such, they would use the 334 strategies on theoretical modules (e.g. pausing regularly and stating the goal while sorting cards with an 335 embedded PM task) but were reluctant to apply them to more complex and ecological activities such as 336 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 337 338 impaired in daily life, actually engaged with the techniques and used them whenever he noticed task

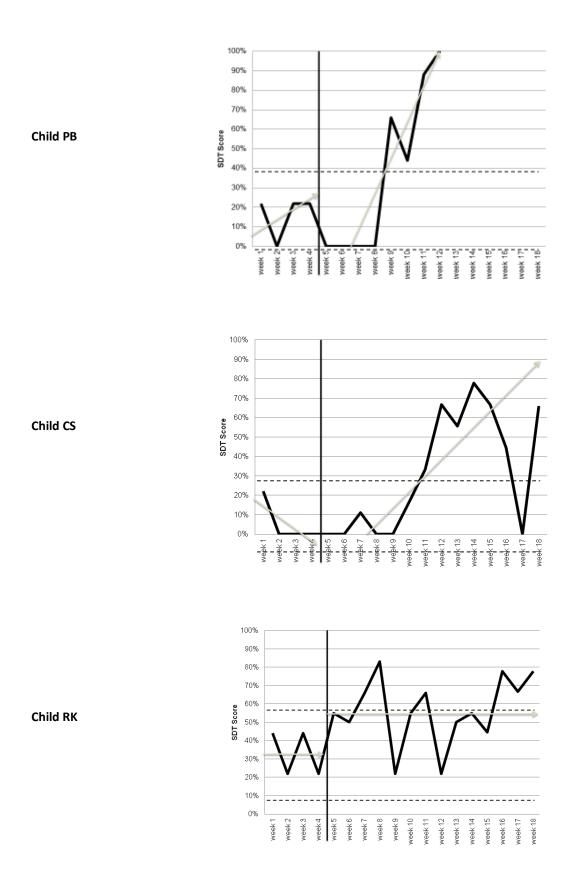
similarities. CS seemed to understand only a few GMT concepts and metacognitive strategies. IP and PB
 seemed to lack awareness of impairments and reported not finding the intervention useful, but found the
 program was fun and they participated willingly.

342 Repeated PM measures. The Saint's Day Test was performed by 3 children. IP, aged 8, did not complete the 343 task as he was not familiar with mobile phones, did not know how to use the internet and making a phone call 344 to an unfamiliar person was not age-appropriate. YR dropped out of the study. Weekly PM score changes over 345 time are shown in Figure 1. During baseline, none of the children reached a score of 50%. Using a two-standard 346 deviation band (2SDB), all children showed statistically significant progress, as all had at least two consecutive 347 points outside their 2SDB. The best progress seemed to be made by PB. Unfortunately, when her performance 348 was reaching 100% on week 12, she lost the charger of her mobile phone and her parents did not replace it 349 until the end of the study giving only a medium effect (NAP = 0.47 [0.12-0.81]). CS began to make progress on 350 the SDT only by week 8. She maintained performance until school holidays (week 17) when her performance 351 dropped momentarily to 0 (overall medium effect; NAP = 0.74 [0.50-0.98]). RK had a very variable performance 352 but a strong effect of intervention (NAP = 0.87 [0.713-1]). The use of the following strategies to manage the 353 task were reported by parents and/or children: stopping all activity up to one hour before the target time and 354 watching the clock (RK), using cues such as the view of her computer (CS).

355

Figure 1-3: 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



358 Complex Cooking Task Management: Ecological EF test "Children's cooking task" and its parallel form

359 ("Christmas Biscuits"). All children were very impaired on the Children's Cooking Task (CCT), scoring from 3

- 360 (CS) to 25 (IP) standard deviations below age matched controls. RK and PB required help from the examiner to
- 361 finish the task, IP completed the task with nearly 200 errors, and failed the task. CS's errors were mainly
- 362 commentaries/questions on every action she undertook but she successfully finished the task.

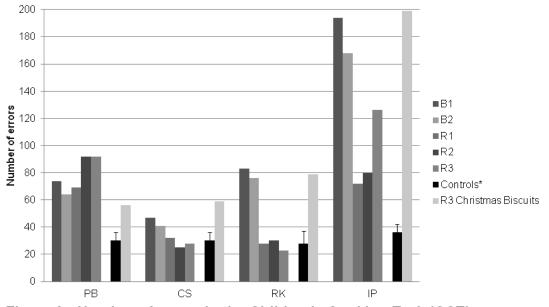


Figure 2 : Number of errors in the Children's Cooking Task (CCT): changes over time (* age-matched controls, n= 8 to 13 per child). *Note 1: B1 and B2: first and second baseline assessments; R1, R2, R3: assessments performed at 0, 3 and 6 months post-intervention. Note 2: : In CCT, errors include action errors, as well as commentaries/questions.*

- 363
- 364 Changes in the number of errors from baseline to post intervention and follow-up for each child are presented
- in Figure 2. During baseline, all children showed some practice effect between B1 and B2. After intervention,
- 366 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.
- 368 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 2).
- 370 Conversely, RK and IP significantly decreased the number of errors after the intervention with large effect sizes,
- 371 RK showing a performance similar to controls after intervention and IP improving from 25 to 6 SD compared to

372 controls. RK clearly used the metacognitive techniques taught in the intervention while performing the CCT

373 (checking he finished a step before moving to the next, saying "Stop!" and thinking before adding a new

- 374 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
- 376 Biscuits) all children returned to their initial number of errors.

ES at R1	ES at R2	ES at R3					
Children's C	ooking Task						
PB -1.15 -2.90 -2.90							
0.33	1.09	0.76 1.59 0.30					
1.42	1.35						
1.31	1.16						
Parenta	al BRIEF						
0.5	0.2	0.1					
0.9	1.3	0.4 -0.1 1					
0.1	0.1						
1.2	0.8						
DEX-C Cogr	nition Score						
1.33	2.00	2.33					
0.67	1.00	1.33					
0.67	0.00	-0.67					
2.00	1.67	1.84					
0.67	0.33	0.50					
	Children's C -1.15 0.33 1.42 1.31 Parenta 0.5 0.9 0.1 1.2 DEX-C Cogr 1.33 0.67 0.67 2.00	Children's Cooking Task -1.15 -2.90 0.33 1.09 1.42 1.35 1.31 1.16 Parental BRIEF 0.5 0.2 0.9 1.3 0.1 0.1 1.2 0.8 DEX-C Cognition Score 1.33 2.00 0.67 1.00 0.67 0.00 2.00 1.67					

*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)

377

379

Table 2: Outcome measures Effect Sizes (ES) comparing B2 to R1, R2 and R3.

Inventory of Executive Functions; DEX-C: Dysexecutive Questionnaire for children

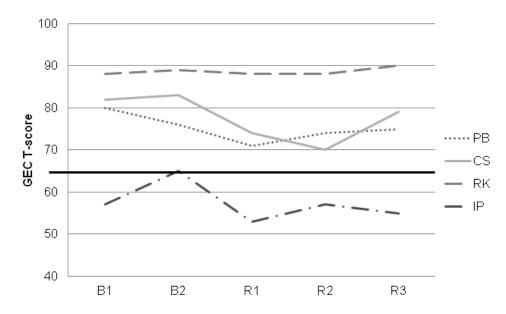
- 380 Transfer to natural contexts. At baseline, all children but one (IP) scored in the clinical range (T-scores > 65) for
- 381 Global Executive Composite (GEC) scores on parental and teacher BRIEF questionnaires. Transfer to home-
- 382 *context:* All parental scores were consistent (inconsistency score < 9). Immediately after the intervention 3
- 383 children (PB, CS, IP) showed a decrease on parental BRIEF scores (see Figure 3), reflecting possibly less
- 384 executive dysfunction in daily life at home. Effect sizes are reported in Table 2. All but one child (IP) showed a

³⁷⁸ Note: R1, R2, R3: assessments performed at 0, 3 and 6 months post-intervention; BRIEF: Behavior Rating

385 decrease on DEX-C cognition sub-scores between the two baselines that was considered to be the enthusiasm 386 effect we had expected due to intervention novelty. However the decrease was accentuated much further 387 after the intervention for all children and continued to decrease at three- and six-month follow-up for PB and 388 for CS with large effect sizes (2.33 for PB and 1.33 for CS, see table 3). Transfer to school-context: Teacher BRIEF 389 scores remained stable for RK and CS, were unreliable for PB and for IP (inconsistency index >9), meaning it was 390 not appropriate to draw any reliable conclusions on EF in the school context for these children, although PB's 391 BRIEF seemed to show significant improvement. PB was reported to have made excellent progress at school on 392 school academic reports.



Figure 3: Parental BRIEF questionnaire changes of GEC Tscore over time.



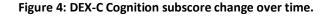
394

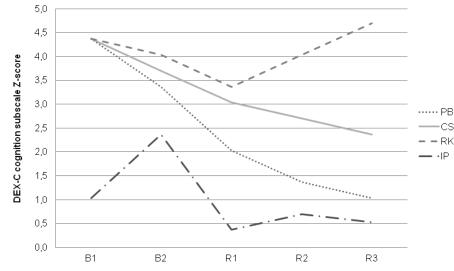
395 Note: GEC indicates Global Executive Composite.

396 B1 and B2: first and second baseline assessments; R1, R2, and R3: assessments performed at 0, 3, and 6 months

397 postintervention. The horizontal line represents the clinical cutoff score of 65.

398





400

401 Note: B1and B2: first and second baseline assessments; R1, R2, and R3: assessments performed at 0, 3, and 6
 402 months postintervention. DEX-C indicates Dysexecutive Questionnaire for Children.

403 Generalization: Goal Attainment Scaling and post intervention interview.

404 There was a high rate of missing GAS data. GAS could not be developed in collaboration with RK's school

405 everyday people because he attended school very rarely in that period. GAS goals were developed with IP's

406 teacher and school assistant but post intervention GAS forms were handed to IP who lost them (as school

407 closed for 2 months after R1, new forms could not be obtained). CS's teacher, with whom the goals were

408 developed, changed after R1, explaining missing data for R1 and R2. Overall, GAS scales were obtained for at

409 least one "everyday person" per child (see table 3). We were only able to agree on personal goals with one

- 410 child (RK). The other three viewed the goals proposed by their everyday people as not problematic or not
- 411 important.

Examples of personal GAS goals corresponding to EF-	Examples of general GAS goals			
related problems reported by everyday people (only the goals	corresponding to metacognitive strategy use			
and not the full Goal Attainment Scales are reported)	and GMT application (only the goals and not			
	the full Goal Attainment Scales are reported)			
<u>PB - mother :</u>	-to be aware of one's "Oops" errors (attentional			
-to forget taking antiepileptic drug less often	slips)			
-to lose fewer objects	-to detect "Oops" errors as they occur			
-to be flexible enough to change strategy if the first strategy does	-to stop and think before beginning a new task			
not work	-to formulate a task's main goal before			
-to brainstorm for possible solutions before rushing to start a	beginning a task (e.g. school exercise, home			
task or school exercise	activity)			

<u>PB – school assistant:</u> -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)	 -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
<u>CS – mother:</u> -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	-to check a task/exercise before moving on to another
<u>CS – teacher :</u> -to accept the need to check her work when she is prompted to do so -to estimate the difficulty of exercises/tasks	
 <u>RK – both parents and the child:</u> -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"). 	
 <u>IP – teacher and school assistant:</u> -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 	

412 Table 3: Personal and general GAS goals

413

414 All children progressed towards their goals at R1. However, only one of the GAS scores (IP's) reached the

415 expected goal attainment level (T-score of 50). During the external interview at the end of the study, PB's

416 mother reported significant daily life benefits of the program: PB forgot her antiepileptic drugs much less often

417 (by linking her breakfast orange juice with remembering to take her medications), was less often late at school

418 and made important progress at school allowing her to continue schooling in an ordinary class with a school

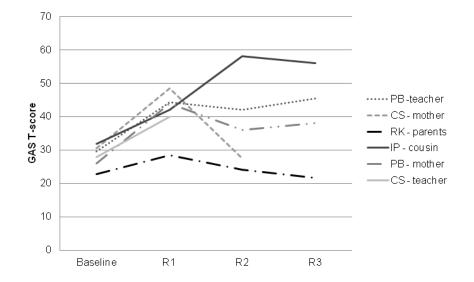
419 assistant rather than going to special education as had originally been planned. These last two improvements

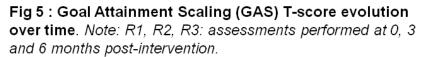
420 were not captured by the child's GAS scores because these were unanticipated positive outcomes. Some

421 positive outcome were reported for IP by his main carer, which was consistent with a GAS score that reached

422 50. Parents reported some general progress in well-being at R1 for RK and CS. Most children carried on

- 423 cooking after the intervention and RK was for the first time allowed to be in the kitchen alone by his parents.
- 424 Parents reported that children's self-esteem increased because they could make a meal for the family.





425 426 Participation of everyday people. Overall, the level of participation of the everyday people was very low. 427 Mission sheets were very rarely filled in. Only RK's parents asked for feedback after the intervention. In 428 interviews, parents, teachers and school assistants all reported that the intervention was fun for the child and 429 that metacognitive techniques "were useful". However when asked to provide examples of metacognitive 430 strategies contained in the "Cognitive Coaching" guide they had received, seven out of ten everyday people 431 recalled less than half of the strategies. Moreover examples of strategy use were not always appropriate. 432 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 433 followed than the abstract demand of "cognitive coaching": one school assistant (PB's) regularly used the paper 434

435 notebook to compensate for PB's constant PM failures relating to school goals (bring sports clothes, get a form 436 signed), one mother (CS's) started to cook with her daughter. Several parents reported that using the term 437 "Oops error" helped to lower family's tension to the child's cognitive failures and some began using the term 438 with their other children and themselves. All parents reported being generally too busy to apply the cognitive 439 coaching at home. Teachers reported the children did not use the strategies at school, but had not prompted 440 the children to do so. Both teachers and school assistants tended to emphasize the behavioral, attentional and 441 "lack of effort" problems at school as the key problem for the child and did not consider metacognitive 442 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"). 443

444 **DISCUSSION**

The "Context-sensitive GMT" intervention comprised of (1) an adapted Goal Management Training (GMT), (2) 445 446 metacognitive strategies practice through meaningful activities, (3) a "cognitive coaching guide" for the child's 447 everyday people. The program was feasible to implement and apparently enjoyable for children. However, 448 participation of everyday people was limited. Children significantly improved on the Saint Day Task (time-based 449 prospective memory). EF performance in the ecological Children's Cooking Task (CCT) improved in two children. 450 Three children showed a decrease on parental BRIEF scores reflecting possibly less executive dysfunction in 451 daily life at home. All children decreased their cognition DEX-C sub-score, suggesting that parents perceived 452 improvement in cognitive EF impairment. There were some indications of generalization to untrained tasks in 453 all children, but not sufficient to achieve the expected level of achievement in EF-related GAS goals. When 454 presented with a truly novel task (the parallel version of CCT- "Christmas biscuits"), all children dropped to 455 their initial level of performance indicating a lack of generalization. Intervention effects persisted at three-456 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 ^{11,66,67}. The cognitive 461 462 coaching guide was rarely used by the teachers and school assistants. It is not entirely clear why this was, but 463 one possibility is that the relatively limited contact with the investigators (a phone call before and after the 464 study and the rest through a written guide) was not sufficient to engage them in the intervention contrary to other school-delivered interventions 68. This is clearly an important issue for future studies and for clinical 465 466 interventions that depend heavily on a child's everyday people for success. It is probably easier to engage 467 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^{68 69}). In everyday clinical practice, frequent 468 469 contact with the child's everyday people (especially school staff) is often not feasible and so examining whether 470 written information (such as our cognitive coaching guide) can facilitate intervention support from these everyday people is an important research question. 471

472 The prospective memory (PM) performance might have improved because the task became familiar and 473 routine. However, previous studies with adults using a similar design did not show an improvement of performance with time⁴⁴. Furthermore, children performed so poorly and with so much variation from week to 474 week (very rarely giving the Saint day within one hour of the target time) that no possible routine could have 475 476 been established. We could not control for the performance of the ongoing task (activity the child was doing at 477 the target time). It has been emphasized that PM performance needs to take into account performance in the 478 ongoing task as well as the PM performance because PM paradigms can be considered as a dual task paradigms²³. As such it is possible that PM performance increased at the expense of ongoing activities. For one 479 child (RK), his parents actually reported that he stopped all activity up to one hour before the target time, 480 481 watching the clock in order to perform the task (but often actually forgot the task anyway). Pausing activity to 482 avoid missing an important phone call appointment may be considered an effective strategy in real life, albeit 483 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

487 coaches. Furthermore, the "home-school cognitive coaching" guide may have increased carers' insight into the 488 child's difficulties, meaning that even if improvements in behavior had occurred these were balanced out with 489 greater awareness of difficulties on the part of the carer. These issues could perhaps explain why the other 490 outcome measures (CCT, GAS) were not consistent with BRIEF scores (e.g. the BRIEF scores of CS and PB 491 decreased but they did not improve on the CCT and whilst RK made best progress on CCT there was no 492 corresponding decrease in BRIEF scores). However, it is possible that metacognitive strategies are effective in a 493 time-limited task such as the Children's Cooking Task (CCT) but impractical in the context of daily life's constant 494 attentional demands, as it is an effortful, top-down process. This may explain why RK made good progress on 495 the CCT, but did not apply the metacognitive strategies in daily life so that parents did not notice a real change 496 in everyday life post-intervention. Alternatively it may be the case that parents and other everyday people may 497 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 498 499 parents and patients have been frequently noted^{70,23}. Correlations between parental BRIEF scores and EF classical tests^{54,55} and with the CCT ⁴⁷ are typically small so the BRIEF might not have captured the children's 500 501 progress. However, all children significantly improved on the DEX-C cognition scores at R1, including IP and RK, 502 which is consistent with earlier finding that the number of errors in CCT and DEX cognition subscales are highly correlated ⁴⁷ in both adults^{71,72} and children⁴⁷ and might be a better measure of the children's executive 503 504 progress than the BRIEF.

505 For PB it is difficult to explain the contrast between consistency of improvement on DEX-C Cognition subscore, 506 GAS, qualitatively reported generalization, parental BRIEF – and the increased number of errors on CCT. This 507 should however be interpreted with caution because all improvements were based on subjective informant 508 reports, mostly of her mother. As PB seemed to have very poor awareness according to the therapist, the 509 intervention might have improved her awareness rather than EF, which would explain perceived improvement 510 in her natural contexts but not on objective measures of EF (CCT). Nevertheless she is the child who seems to 511 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 ⁷³ and is a factor of poorer 512 outcome. Indeed PB never found the sessions "useful", but only "fun". In the absence of awareness, her 513

514 motivation did not seem to be to overcome her difficulties (perceived as non problematic) but rather to enjoy 515 herself during the sessions, and through that enjoyment some implicit learning may have occurred. In children, 516 enjoyment may be more important to an intervention's success than awareness and our intervention seems to 517 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" ⁷⁴. This may be why 518 519 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 study⁶⁶ 520 521 of cognitive strategy training in children with TBI: making the intervention fun was identified as being useful, 522 whereas the "Goal-Plan-Do-Check" strategy (that is similar to GMT) and promotion of good strategy use were 523 not. Considering together the evidence of ours and Missiuna's studies (both on very small samples), it seems 524 that strategy use does not come easily to children with more severe TBI and therefore may not be the best 525 rehabilitation approach for them. In any case, strategies need to be simple, concrete and repeatedly practiced 526 in order to benefit those children.

527 Usually, elaborating a goal attainment scale (GAS) serves to focus rehabilitation on that goal. Such goal-focused 528 rehabilitation is indeed an effective approach. However this presents a methodological challenge for EFresearch: when a task is trained, its familiarity may make it less demanding on EF as it is likely to require the 529 530 application of learned knowledge and task-specific procedures (which may have become automatic therefore 531 not "executive"), rather than more general problem solving and goal management processes ^{60,62}. On the contrary, daily life is full of EF-demanding tasks that require conscious, novel and effortful processing ^{3,60}, 532 without lapses into automaticity. Apparent progress after a goal-focused training may not necessarily reflect 533 534 changes in underlying executive processes needed to face daily life. Conversely, our aim was to improve 535 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^{61,60,} personal GAS goals were not trained, to keep GAS 536 537 as a generalization measure of EF. However, in future studies it would be more pertinent to divide child's goals 538 into a trained set of goals (and corresponding GAS) and an untrained set of goals (and corresponding GAS) and 539 then to focus the intervention on training the former while using the latter as an ecological generalization 540 measure. In such an approach, main issues would be to match GAS sets for level of difficulty, child's interest

541 and level of priority as seen by everyday people who participated in goal selection. Furthermore for GAS 542 aiming at measuring generalization, it would be important to control how much explicit linking to these goals is 543 done during the intervention. The intervention would probably be more effective if it combined goal-focused 544 rehabilitation and general metacognitive training. Elaborating two sets of GAS has already been proposed by 545 Schlosser⁷⁵ in the concept of "control goals". This could have supported further the finding of our study that 546 children could be trained effectively in a meaningful task such as making a chocolate cake (CCT) by combining 547 metacognitive training and repeated cake baking but could not be trained to manage a new untrained recipe 548 (Christmas biscuits). More broadly, in rehabilitation research, using two sets of GAS would make of GAS 549 methodology both a powerful motor for achieving meaningful goals by focusing intervention on them and a 550 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 study⁶⁶, children 551 were able to self-identify goals using a more framed and age-appropriate goal setting system than GAS, which 552 could be used in future studies. However, children in Missiuna's study⁶⁶ had sustained mild to moderate TBI 553 554 and were probably less impaired. Goal setting requires some basic level of awareness, which children with 555 severe TBI often lack. Lack of awareness was identified for all children in our study except RK and seemed to be 556 the main reason why goals could not be identified by the children. Besides, in Missiuna's study, children were 557 allowed to choose any goal (e.g. learn a new sport) whereas we purposefully retained only EF-related goals. 558 More in-depth assessment of awareness would bring valuable contribution for research on goal-setting 559 procedures.

560 The intervention was able to improve one particular prospective memory task performed in an ecological 561 setting (SDT), it allowed some children to perform better in a cooking task (CCT) and it resulted in some gains in 562 daily executive functioning. However, this metacognitive training did not allow enough generalization effects to 563 reach expected levels in EF untrained personalized goals or to manage a novel complex EF-demanding task. The 564 aim of providing children with meta-cognitive strategies applicable to "any" situation in life is an ideal goal but 565 is perhaps not feasible: children's ability to cope with new, EF-demanding situations of daily life may not be 566 possible to improve with training in case of severe impairments. In those severe cases, a repeated, goal-567 focused rehabilitation using activities that are meaningful to the children, and not focusing on explicit

568 generalization training, may be a more reasonable therapeutic option (see ⁶⁶ for an example of effective goal-569 focused intervention in a small sample of children with TBI). It should also be emphasized that GMT targets 570 more specifically the PM aspects of EF-demanding tasks and much less problem-solving abilities. This study 571 supports a recent review¹⁹ in adults which concluded that GMT is probably more effective when combined with 572 other interventions targeting other aspects of EF such as problem solving and initiation (see ^{76,63} for examples 573 of such interventions in adults).

574 Limitations of previously published studies included: insufficient assessment of generalization⁷⁷, of specific 575 effects on EF^{78,11}, lack of objective cognitive performance measures (using questionnaires only as the outcome measure)¹², use of problem-solving tasks that lack ecological validity⁷⁹, lack of demonstration of EF difficulties 576 577 prior to intervention⁴⁵ or lack of multiple baseline or follow-up in pre-post designs^{80,66}. Others focused mainly on the behavioral aspects of the dysexecutive syndrome^{68,13}. This study is to the best of our knowledge the first 578 579 study that explores whether metacognitive training generalizes and helps children to adapt and manage a 580 novel EF-demanding task and to achieve untrained goals. Of course the small sample of this study does not 581 allow to draw general conclusions about the program efficacy. Our results must be interpreted with caution, 582 especially because we included the most challenging population for this pilot intervention, which may have 583 limited its effectiveness i.e. children with severe EF impairment and with three known major factors of poorer outcome^{81,57,56,82}: (1) severe TBI; (2) sustained at an early age; (3) in non-optimally functioning families (similarly 584 585 to Corbett's GMT⁴⁰ that targeted children from low socio-economic background in Cape Town with little success). Those children are usually excluded from protocols⁶⁶ and adult interventions using GMT usually target 586 587 patients with moderate and mild TBI^{20,76}. As this study demonstrates the feasibility of the program for a 588 particularly complex group of children, it would be helpful to replicate this study with children who have 589 sustained a moderate TBI and children who may have more access to everyday people for providing cognitive 590 coaching. On the other hand, in the clinical setting, it is precisely children with severe TBI and complex family 591 situations that are most needy of intervention and future research should focus on this group, in spite of its 592 challenges.

593 For clinical use, the intervention may need further adaptation: the program may benefit from being longer to 594 allow the children to integrate each strategy before practicing a new one, everyday people should be 595 supported further to participate in the sessions, in a similar way to that used in Braga's study¹¹. Cognitive 596 coaching should be presented through concrete activities to be done at home and at school rather than general 597 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^{83,84}. Not all families are willing and/or capable 598 of engaging in a family-delivered program⁸⁴ and evaluating how to predict this prior to intervention would be of 599 600 benefit to service providers. Direct contact with school staff is needed. The impact of parental metacognitive 601 knowledge, skills and beliefs on outcomes in family-delivered interventions would also be a valuable

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component of future studies.

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800 SUPPLEMENTAL DIGITAL DATA

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Examples of he	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					

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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
- 806 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.
- 811 *E.g.: « XX (child name) writes on his own initiative things he/she might forget to do in a* 812 single and well-identified note-book or planner (excluding homework he/she is explicitly
- 813 asked to write down by the teacher)»
- 814 *-2 : never*
- 815 *-1 : sometimes but it is not regular or functional enough to rely on it*
- 816 0: Writes down important things to be done but you need to emphasize/repeat it is an 817 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
- 820 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.
- 829 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
- 834 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.

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	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
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	father : 14	father : 11	father : 22	father : 7	father : 4
(years)	mother : 15	mother : 11	mother : 15	mother : 11	mother : 17

Table 1: Characteristics of the participants

Note: 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: 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

	Processing speed (Stroop part 1: color naming in seconds)	Inhibitiory 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)	$77,56 \pm 10,50$	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	$338,29 \pm 175,54$	0,7 ± 0,73
RK	83	4*	380	8*
Controls (n= 14)	$\textbf{71,50} \pm \textbf{10,14}$	$\textbf{1,00} \pm \textbf{1,30}$	$\textbf{248,}\textbf{14} \pm \textbf{51,}\textbf{03}$	$\textbf{0,9} \pm \textbf{1,63}$
IP	99	4*	613*	7*
Controls (n= 13)	$\textbf{100,38} \pm \textbf{35,88}$	$\textbf{1,}\textbf{46} \pm \textbf{1,}\textbf{51}$	$\textbf{416,83} \pm \textbf{95,96}$	1,2 ±1,28
YR	69	15	314*	19*
Controls (n= 14)	$\textbf{71,50} \pm \textbf{10,14}$	$\textbf{1,00} \pm \textbf{1,30}$	$\textbf{248,}\textbf{14} \pm \textbf{51,}\textbf{03}$	$\textbf{0,9} \pm \textbf{1,63}$
*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

Table 1 bis: Childhood Executive Function Battery (FEE) raw scores for participants and their age-, sex- and socio-economic status-matched controls.

	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

Table 1 ter: Standard scores for traditional paper and pencil tests

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