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Exploring the use of the Cognitive Orientation to daily Occupational Performance approach (CO-OP) with children with executive functions deficits after severe acquired brain injury: a single case experimental design study

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Abstract

Background. Children with acquired brain injury (ABI) often have cognitive and behavioral impairments that affect participation in everyday activities. Among them, executive function (EF) deficits are frequent. Cognitive Orientation to Daily Occupational Performance (CO-OP) is an individualized treatment that teaches cognitive strategies necessary to support successful performance. Few studies have examined the effectiveness of CO-OP in children with EF deficits after ABI.

Objectives. To assess whether the use of CO-OP could be of interest in children with EF deficits after ABI, to improve their occupational performance, their executive functioning in everyday life and their cognitive processes constituting EF.

Methods. This was a single case experimental study with multiple baselines across individuals and behaviors. We included 2 children at least 6 months after severe ABI. The children received 14 individual sessions of the CO-OP intervention. Each child set 3 goals by using the Canadian Occupational Performance Measure; 2 goals were trained and the third was a control goal. The achievement of the goals was measured by using repeated measures of Goal Attainment Scales (GASs). Ecological assessments of EF included the Children's Cooking Task and parent and teacher ratings of the Behavior Rating Inventory of Executive Function (BRIEF) questionnaire.

Results. Both children improved their performance on both trained goals (and, to a lesser extent, on untrained goals). We found significant improvement on tests of EF and on the BRIEF questionnaire, reflecting executive functioning in everyday life, at home and at school.

Conclusions. These results are encouraging and suggest the feasibility and effectiveness of CO-OP for children with EF deficits after ABI. They should be replicated in a larger number of cases.

Trial registration: [ClinicalTrials.gov \(NCT04718688\)](https://clinicaltrials.gov/ct2/show/study/NCT04718688)

Introduction

Childhood acquired brain injury (ABI) is the leading cause of death and a lifelong disability [1]. Cognitive and behavioural impairments are among the most disabling, and executive function (EF) deficits are frequently observed. EF deficits negatively affect participation in activities of daily living and at school [2]. Therefore, EF deficits have a direct impact on occupational performance, which refers to a person's ability to perform whatever they want to, need to or are expected to do in everyday life [3].

Many approaches have been suggested to improve EF after childhood ABI, but few have demonstrated their effectiveness [4]. However, general principles are emerging: the importance of involving parents and caregivers in cognitive coaching and the need for the most ecological and functional (context-sensitive) approaches [5–10]. In this context, Cognitive Orientation to daily Occupational Performance (CO-OP) could be an interesting approach to EF rehabilitation because it meets these principles.

CO-OP is a performance-based treatment approach for individuals who experience difficulties performing the skills they want to, need to, or are expected to perform. CO-OP is an individually tailored, active client-centered approach that engages the individual at the meta-cognitive level to solve performance problems. The effectiveness of CO-OP has been demonstrated with different populations [11–13], but little research has involved children with

cognitive deficits after ABI except Missiuna et al. [14]. Two other studies used CO-OP in samples including children/teenagers with ABI, but inclusion criteria, type of patients and objectives were very different from our current goals [15–17] (Appendix A).

Interesting results were also reported on the applicability of the CO-OP approach in 3 adults with executive dysfunction after traumatic brain injury [18]. Because EF and metacognition are immature in young children and undergo protracted maturation during childhood and adolescence, results of this study cannot be directly applied to children.

Here, we propose to complete the existing observations and specify the EF deficits. The aim of this study was to assess whether the use of CO-OP could be of interest for children with EF deficits after severe ABI. Because EF cannot be used efficiently to solve problems and metacognitive strategies can be an interesting option (if not used alone) in EF rehabilitation after childhood ABI [7,10,19], following the suggestions of the Missiuna et al. study, we hypothesized that teaching a child with ABI to explore problem solving and use the CO-OP strategies could be a useful approach to improve their occupational performance, their executive functioning in everyday life and their cognitive processes constituting EF.

Materials and methods

Participants

This study was carried out in the Rehabilitation Department for Children with Acquired Neurological Injury, Saint Maurice Hospitals, Saint-Maurice, France. It was approved by the Saint Maurice Hospitals ethics committee (no. 9, 05-18-2018). Informed parental written consent and participant assent were obtained for participants before initiating any procedure. The study was registered at ClinicalTrials.gov (NCT04718688).

Inclusion criteria were 1) age 8 to 14 years; 2) diagnosis of ABI, sustained at least 6 months previously, still attending an in- or outpatient rehabilitation program after the injury;

3) evidence of a dysexecutive syndrome on neuropsychological assessment; 4) normal vision and hearing (with appropriate correction if necessary); and 5) sufficient language skills to understand and communicate explicitly.

Exclusion criteria were 1) non-French-speaking child or parents; 2) sensory-motor or visual impairments precluding participation in the study; 3) intellectual deficit (Full-Scale Intellectual Quotient < 70) or severe impairments in comprehension, memory or attention, incompatible with understanding and choosing rehabilitation goals and participating in 45-min sessions; 4) neurological, psychiatric, genetic or learning disability diagnosed before the ABI; and 5) severe anxiety or depression symptoms.

To provide a more complete description of participants' neuropsychological profile, the following standardized tests were administered: the Wechsler Intelligence Scale for Children (WISC V) [20], subtests of the Children's Memory Scale [21], the Test of Everyday Attention for Children (TEA-CH) [22], the Tower of London test [23] and 3 subtests of the Behavioural Assessment of the Dysexecutive Syndrome for Children (BADSC) [24].

Parents and teachers were asked to answer the Behavior Rating Inventory of Executive Function (BRIEF) questionnaire and the Canadian Occupational Performance Measure (COPM) several times (if the parents were separated, we proposed parent-rated assessments to both parents individually).

Study design

A Single Case Experimental Design (SCED) with multiple baselines across individuals and behaviors was used [25–27]. SCEDs are experimental designs aimed at testing the effect of an intervention in a small number of patients (typically 1 to 3), using repeated measurements, sequential (\pm randomized) introduction of an intervention and method-specific data analysis, including visual analysis and specific statistics [25]. The repeated measures included baseline, intervention and post-intervention phases.

This design allowed for repeated measurement of goal achievement performance, to quantify the evolution in achieving chosen goals and thus the effects of CO-OP. Each patient had a specific baseline time (1.5 weeks for P1, i.e., 5 measurements; 2 weeks for P2, i.e., 6 measurements). In multiple-baselines design, the intervention is introduced sequentially to different patients. Thus, this design shows that change occurs when and only when the intervention is directed at that patient [25].

We used the Single Case Reporting Guideline In BEhavioural Interventions (SCRIBE) [28] to report the SCED (Appendix B).

Outcome measures

Main criterion (single-case experimental design)

Repeated measures of evolution in achieving chosen goals

The Goal Attainment Scale (GAS) is a criterion-referenced measure used to quantify the degree to which personal goals are achieved [19,29,30]. It was used for different purposes: 1) after identifying the goals with the COPM, the expected results for each goal were formalized using GAS; 2) the GAS assessed the stability of the control goal (i.e., untrained) in order to demonstrate the specificity of the intervention; and 3) finally, the GAS was used as the repeated measure, performed 3 times a week throughout the study (during baseline, intervention and at 1 month post-intervention).

The GASs were written by the first author (HL). All levels of the scales were to be specific, measurable, acceptable, realistic, and time-specific (S.M.A.R.T.). The occupational therapist constructed ordinal scales with incremental steps of equal intervals. Each GAS scale had to reflect a single dimension of change [31]. Several methods have been described to build GASs, with differences in the number of levels to be described [19,29,31,32]. Each

method has advantages and disadvantages [19]. Here, we chose to build 5-point GAS scales based on Steenbeek's method, using -2, no change relative to the level determined before the start of the baseline (=initial level); -1, less than expected; 0, expected goal; +1, somewhat more than expected; +2, much more than expected [31]. Indeed, no worsening of performance was expected with the CO-OP intervention.

Each expected goal was discussed and defined in collaboration with the patient [19]. The validity of each GAS was verified and validated by an independent trained expert (AK-P) as recommended for the use of GASs in clinical and research practices [33]. GAS ratings were assigned by independent therapists (having completed a 2-day theoretical and practical training course and regularly using GAS), with blinding to the patients and the phase of the study, based on a video-recording of the children performing the target goals.

Given that GAS variants do not all use the same initial level or do not describe their levels with the same degree of precision, the scales' psychometric properties cannot be compared in a valid way [19]. The Steenbeek pediatric study [31] found good to excellent interrater reliability of GAS when used by a group of trained therapists.

To control the GAS reliability, 20% of the GASs were independently rated by 2 therapists, also with blinding to the patients and the study phase.

Additional criteria

Determination of goals and measure of performance and satisfaction

We used the COPM [34]. This semi-structured interview (driven by the first author [HL] trained in use of the COPM) helps children/parents identify problematic occupations in different daily areas. The COPM was used to determine 1) (only with the child) the 3 problematic occupations identified as the most important (2 were target goals, and the third was a control goal) and 2) changes in occupational performance in self-selected goals: children and parents rated their performance and satisfaction associated with these 3

occupations (pre-, immediate- and 3-month post-intervention measures). When we use COPM in pediatrics, we can consider that the client-centered approach includes the child and the parents. It is possible to nominate all stakeholders as clients, which is why we also asked parents to rate performance and satisfaction [35].

Test–retest reliability has consistently been found well above the acceptable range in reliability studies using the COPM [36]. According to the manual, a difference of 2 points between COPM results is considered a clinically significant change [36].

To help children express and identify problematic occupations and significant goals, the OThope French pilot tool (Outil Thérapeutique pour l’Autodétermination d’Objectifs Pédiatriques en Ergothérapie [therapeutic tool for the self-determination of pediatric goals in occupational therapy] [37]) was used during the initial COPM process. This pilot tool was developed to facilitate the determination by the child of his/her problematic occupations with a visual aid (because sometimes children have difficulty stating their problematic occupations [38]). It was inspired by the Pediatric Activity Card Sort [38]. According to Perrault [37], this tool enables the child to make careful choices based on a self-evaluation of his/her capacities.

Ecological measurement of EF

The Children's Cooking Task (CCT) is an ecological standardized task designed to assess EF during actual execution of a complex task [39]. The CCT was used as a secondary outcome measure in this study to determine whether children transferred progress in problem-solving, acquired through strategies discovered with CO-OP, to the performance of a complex task, unrelated to the trained goals. This test was performed at pre-, immediate- and 3-month post-intervention by an independent therapist, trained in the task, with blinding to the child’s status and phase of the study. The CCT outcomes were the total number of errors and the task duration [40]. The CCT has been found to have good internal consistency (Cronbach

alpha=0.96) [2]; good inter-rater reliability (intraclass correlation coefficient [ICC]=0.96) [39] and test–retest reliability (ICC=0.89) [2]. The CCT also demonstrated excellent discriminant validity, because it differentiates patients from controls, even after mild injuries and EF deficits [2,41]. Finally, it demonstrated good concurrent validity (the score was moderately correlated with the perceptual reasoning index and strongly with scores of several tests assessing executive functioning) [2].

Measuring the impact of EF deficits in everyday life, in family and school contexts

The parent and teacher report forms of the BRIEF provide an ecological assessment of executive functioning by its repercussions in family and school contexts [2]. Currently, this questionnaire is the most validated and widely used for children with various congenital, developmental or acquired conditions [1] [42].

Parents answered the BRIEF at 3 times during the baseline (beginning, middle and end of baseline testing), at immediate post-intervention, and at 1 month and 3 months post-intervention. Teachers also answered the BRIEF at 3 times during baseline, and once a week during the intervention, then at 3 months post-intervention. The outcome measure was the Global Executive Composite score (GEC) T-Scores (mean [SD] 50 [10]; clinical range cutoff: T-Scores \geq 65). The questionnaires were rated by the first author (HL) following the manual instructions. The BRIEF demonstrated good test–retest stability. The stability of scores over a 2- to 3-week interval shows that the BRIEF administration can be repeated without expected variability related to the instrument itself [43].

Intervention

The intervention was included within the child’s conventional rehabilitation program. The general structure of the intervention followed the key principles described in the original CO-

OP protocol [44], with some adjustments to adapt it to the specificities of the ABI population. In the original protocol, the child sets 3 goals. Here, we concentrated on 2 rather than 3 goals because addressing 3 occupations per session seemed too ambitious given the children's cognitive profile. Among the 3 problematic occupations identified during baseline, the first 2 were addressed during sessions, and the third was a control goal.

The original CO-OP protocol consists of 10 individual 1-hr sessions. In this study, as suggested by Krasny-Pacini et al. [45], we increased the number of sessions to 14 (2 sessions per week for 7 weeks) because of the children's severe EF profile and associated deficits.

The global strategy (goal, plan, do, check [GPDC]) was taught to the child at the first session. The following sessions consisted of an iterative process of implementing the global strategy in the context of guided discovery to identify domain specific strategies (DSSs) to overcome performance "breakdowns" the children were experiencing when performing the self-selected tasks. DSSs are cognitive strategies that are specific to a particular task, part of a task or a situation, and are usually intended to be used for a short time [44]. In addition, "homework" (to be done between sessions) was assigned to encourage the application and practice of the cognitive strategies discovered during the intervention sessions, and to practice the target skills. Parents were invited to attend the sessions if it was possible for them. In any case, the global strategy, as well as the progress of the child, were explained to the parents by telephone and in writing. The principles of CO-OP were explained to teachers during a brief meeting before the start of the intervention.

The sessions were performed by the first author (HL), a trained CO-OP occupational therapist and CO-OP instructor. Given the type of intervention, no blinding was possible.

Data analysis

The results of the standardized measures were scored according to each task's manual (e.g.,

COPM, BRIEF and CCT). GAS interrater reliability was calculated for 20% of the GASs, using the Kappa coefficient (strength of agreement: <0, poor; 0.00-0.20, slight; 0.21-0.40, fair; 0.41-0.61, moderate; 0.61-0.80, substantial; 0.81-1.00, almost perfect) [46].

To analyze the results of the SCED (= GAS scores, which are the repeated judgement criterion), we performed a visual analysis of the GAS graphs, which were confirmed by statistical analysis [26]. Visual analysis was based on Fisher's conservative Dual Criteria Method [47], which is based on projecting the level line (= mean) and the baseline trend line on the intervention phase. By determining how many data points were both above the level line and above the trend line in the intervention phase, the conservative Dual Criteria Method was a visual aid illustrating the effect or non-effect of the intervention (the more points above these 2 lines, the more pronounced the effect of the intervention). The conservative dual criterion was calculated on the Manolov website: <https://manolov.shinyapps.io/Overlap/>.

Statistical analysis involved using the non-overlapping indices of the data points between the baseline and the intervention phases (the fewer overlapping points between the phases, the more likely the intervention is effective). We calculated the Nonoverlapping of All Pairs (NAP) value [48]. NAP is an estimate of the probability that a randomly selected observation from the intervention phase improves upon a randomly selected observation from the baseline phase. For an outcome where increase is desirable, the effect size parameter is $\theta = \Pr(Y_B > Y_A) + 0.5 \times \Pr(Y_B = Y_A)$. NAP was calculated as an estimate of the effect size of the intervention (whereas the Dual Criteria Method offers only a visual analysis aid). Its value ranges from 0 to 1, with a weak effect from 0 to 0.65; medium 0.66 to 0.92 and large 0.93 to 1. This was used to satisfy the SCED recommendation of using both a visual analysis and an effect-size calculation [25]. The NAP is adequate when there is no strong baseline line trend. The NAP was calculated on the Pustejovsky website: <https://jepusto.shinyapps.io/SCD-effect-sizes/>. The p-value (p value should be interpreted with caution because it is distorted by auto

correlation) was calculated on the Vannest et al. website: <http://www.singlecaseresearch.org> .

The BRIEF was not the main repeated judgement criterion, but this questionnaire was used many times throughout the study phases (see methods section above); thus, it was also possible to analyze these results statistically by using the Dual Criteria Method and NAP.

Missing data were replaced by a score corresponding to an average of the scores of the phase to which it belonged.

Result

Participants

Two patients met the inclusion and exclusion criteria at the time of the study. Their characteristics are summarized in Table 1.

Intervention process/missing data

Both patients performed all 14 sessions. No procedural changes occurred during the investigation after the start of the study. Some measurements were missing: the second baseline Parental BRIEF for P1, the ninth week Teacher BRIEF for P2, the 3-month post-intervention Mother BRIEF for P2 and Teacher BRIEF for P1 and 4 GAS measurements of the control goal for P2.

GAS inter-rater reliability

The level of reliability for GAS ratings ranged from fair to excellent/almost perfect (kappa coefficient, calculated for 20% of the ratings of each GAS): P1 goal 3: kappa=0.61; P2 goal 1: kappa=0.644; P2 goal 3: kappa=0.644; and kappa=1 for P1 goal 1 and goal 2 and P2 goal 2.

Main criterion (single-case experimental design)

Repeated measures of evolution in achieving chosen goals

An example of a GAS is in Appendix C. The GAS for both P1's intervention goals increased, up to +2 level, which was maintained over time (Fig. 1). The scores of the control goal were variable but tended to increase toward the end of the intervention, then decreased at post-intervention. Analysis of the dual criterion showed a pronounced effect of the intervention on both intervention goals (Fig. 1). The value of the NAP agreed with the dual criterion, with a medium effect of the intervention for goal 1 (NAP=0.90, $p=0.0066$, 95% confidence interval [CI] 0.62–0.98), a large effect for goal 2 (NAP=0.94, $p=0.0028$, 95% CI 0.67–0.99), and a weak effect for the control goal (NAP=0.595, $p=0.5187$, 95% CI 0.33–0.81).

For P2, all GAS levels increased, including for the control goal. Scores varied but remained at least at level 0 (target level) from the end of the intervention to the end of the 3-month post-intervention assessment. Analysis of the dual criterion showed a significant effect of the intervention, for both intervention goals and for the control goal in the last 3 weeks of the intervention. The NAP value supported these observations, with a medium effect for goal 1 (NAP=0.7619, $p=0.0543$, 95% CI 0.50–0.91), goal 2 (NAP=0.873, $p=0.0061$, 95% CI 0.61–0.96) and goal 3 (NAP=0.6842, $p=0.1815$, 95% CI 0.42–0.86).

Additional criteria

Determination of goals and measuring performance and satisfaction

Using the COPM, the children were able to identify their 3 most important problematic occupations. They were assisted in this by the use of OThope, which allowed

them to support their reflection and encouraged their thought process.

P1 and P2 showed significant clinical effects (increase of at least 2 points in performance and satisfaction scores) at both immediate and 3 months post-intervention for the goals achieved and for P2's control goal (Fig. 2). Performance and satisfaction of P1's control goal increased significantly at immediate post-intervention but was not maintained at 3 months post-intervention. For P1's parents, performance and satisfaction increased significantly for the 3 goals at immediate post-intervention and these results were maintained at 3 months post-intervention. P2's parents noted a significant increase in performance for the 3 goals (mother's missing data at 3 months post-intervention for goal 2 and control goal) and an increase in satisfaction (for the mother only).

Ecological measurement of EF

For P1, the number of errors in the CCT decreased considerably at the immediate post-intervention assessment, with a significant reduction of the deviation from the norm ($\sigma = z$ -score compared to age-matched controls = -2.91 vs -7.67 at pre-intervention, Fig. 3). At 3 months post-intervention, the number of errors increased ($\sigma = -11.58$), especially additions (useless actions) and context neglect. Task duration decreased over the last 3 testing sessions ($\sigma = -0.07$, $\sigma = 1.05$, and $\sigma = 0.8$ at pre-, immediate post-, and 3 months post-intervention). Clinical observations showed that P1 was better organized to carry out the activity during the immediate- and 3-month post-intervention sessions.

For P2, the number of errors was stable overall, throughout the testing sessions ($\sigma = -2$; $\sigma = -2.04$; $\sigma = -1.7$ at pre-, immediate post- and 3 months post-intervention, respectively). Her performance worsened over time, with many additions, environmental adherence and purposeless actions. She did not use the strategies discovered during the CO-OP sessions and reproduced the same pattern of errors. During the post-intervention test, she clearly expressed

her lack of motivation: “I don't really want to do it, because we've already done it... And I don't like this cake!”. Task duration improved over time, although she remained slower than expected for her age ($\sigma = -3.3$, $\sigma = -2.6$, and $\sigma = -1.6$ at pre-, immediate post- and 3 months post-intervention).

Impact of EF deficits in the family and school contexts according to the BRIEF

P1's parental BRIEF scores showed a trend toward normalization of the GEC score at the immediate post-intervention assessment (baseline: GEC T-score(1) = 69, GEC T-score(2) = 72, both in the clinical range; immediate post-intervention: GEC T-score = 61, in the normal range), which was confirmed at 3 months post-intervention (GEC T-score = 55) (Fig. 4), with a large effect (NAP=1, 95% CI 1–1), which suggests decreased impact of EF deficits in the family context.

For P2's mother, the pre-intervention BRIEF score was within the low normal range (indicating slight difficulties) and remained almost stable throughout the study phases (baseline: GEC T-score(1) = 59; GEC T-score(2) = 64; GEC T-score(3) = 63; immediate post-intervention: GEC T-score = 61; 3 months post-intervention: 61.5 [treated as missing data]). This was confirmed by a medium effect (NAP=0.67, 95% CI 0.24–0.92).

For P2's father, BRIEF scores were in the low normal range at all assessments, reaching the borderline zone at post-intervention (baseline: GEC T-score(1) = 57; GEC T-score(2) = 58; GEC T-score(3) = 55; immediate post-intervention: GEC T-score = 56; 3 months post-intervention: GEC T-score = 63), with weak effect (NAP=0.28, 95% CI 0.06–0.72), which suggests a lack of effect of the intervention on executive functioning at home.

P1's teacher's BRIEF scores (Fig. 4) were in the low average at baseline and decreased (improved) throughout the study phases, reaching the normative range (except for a slight increase for the last 2 questionnaires). The visual analysis of the dual criterion did not

show any significant effect of the intervention on the BRIEF scores for P1 and this was confirmed by the NAP value showing a weak effect (NAP=0.33; $p=0.44$; 95% CI -1 to 0.375).

For P2's teacher, the BRIEF scores all remained above the clinical cut-off score (except for the first 2 measures of the intervention phase, which were slightly lower). The dual criterion showed little effect (only 3 points below the 2 lines) and the NAP value showed a weak effect (NAP=0.47; $p=0.88$; 95% CI -0.802 to 0.669).

Discussion

The aim of this study was to assess whether the use of CO-OP could be of interest for children with EF deficits after severe ABI.

Both children were receptive to how to approach problematic situations by using CO-OP. They were able to achieve the goals they had set. The effect of the intervention on their goals, measured by repeatedly administered GAS showed statistically significant results. Their occupational performance improved significantly (COPM) and the improvements in task performance were still evident at 3 months post-intervention. The performance in CCT improved for P1 immediately post-intervention (number of errors) but not for P2. Both patients performed the task faster over the study. Everyday executive functioning in the family and school context (BRIEF) suggested an improvement.

Regarding the intervention goals, 4 tasks were addressed during therapy (2 goals per child). Both children were able to achieve the goals they had set: all trained goals achieved GAS scores of 0 or higher at immediate post-intervention, with results maintained at 3 months post-intervention. A medium effect size was obtained for 3 trained goals and a large effect size for one, which indicates that changes are clinically meaningful. These results confirm those obtained by Missiuna et al. [14], in which all children improved in self-selected tasks trained during therapy sessions (although a different scale was used to measure goal

achievement). Overall, patients' attitudes were variable, generating longer confidence intervals for NAP. Further, NAP should be interpreted with caution because of the autocorrelation of SCED data (all scores relating to the same patient).

A statistically significant change in performance and satisfaction (child COPM) was achieved for 3 of 4 trained goals at immediate post-intervention and was maintained at 3 months post-intervention. These results agree with previous studies [14], although those studies included patients with different, less severe profiles, for whom EF was not investigated in detail.

The parent's performance and satisfaction scores broadly followed the same trend. Thus, teaching the child to explore problem solving and to use the CO-OP strategies may be a useful approach to improve their occupational performance.

The measurement of the control goal did not remain stable. For P1, achievement of the control goal showed variations, of lower magnitude, highly fluctuating without reaching a stable level, as compared with the 2 target goals. For P2, the improvement of the control goal was clear, even if it did not reach the level achieved with the target goals. These findings may suggest an early generalization of cognitive strategies (global and specific) discovered through CO-OP to other tasks. This hypothesis is supported by clinical observations, suggesting some degree of generalization and transfer. Being able to perform the task in another context (generalization) and to apply discovered strategies to non-target situations (transfer) are 2 of the key principles and the "ultimate" aim of CO-OP [44]. It is also one of the main difficulties encountered in experimental EF rehabilitation protocols because patients often struggle to spontaneously transfer the learned skills to untrained situations [45]. The increased scores of the control goals could be interpreted as a positive consequence in terms of generalization and transfer.

The analysis of the CCT gave contrasting results for P1: the substantial improvement noted during the immediate post-intervention test was not maintained over time. Clinical observations suggest that P1 displayed better organization skills, and he performed the task faster. He managed to maintain and control the ongoing activity, while being less impulsive. In the Krasny-Pacini et al. study [45], one patient exhibited similar results, behaving more confidently from trial to trial. This observation suggests the underlying effect of the use of the global strategy used in the intervention. However, this positive aspect was masked by the very large number of behavioral errors at the 3-month post-intervention test. For P2, the increase in number of errors from one assessment to the next was inversely related to her level of commitment to the task. Thus, we found mixed progress for the children using CO-OP, with repetition of a complex task.

Regarding EF deficits in everyday life, according to the BRIEF, for P1, difficulties appeared to be more pronounced at home than in the classroom. However, parent and teacher ratings tended to be congruent in the post-intervention phase, with scores within age-expected norms and significant progress displayed by statistical analyses in both contexts. For P2, the teacher reported more difficulties than the parents, which has been reported previously because difficulties may be more evident in a more structured and demanding environment [1]. P2's parents did not report any progress of the GEC score over time, with almost linear results, closer to the normative range than those of the teacher. The teacher, although she reported more severe deficits, qualitatively perceived significant positive changes, especially at the beginning of the intervention. Taken together, these results suggest that the intervention had a positive effect on both patients' daily-life EF deficits.

In the Krasny-Pacini et al. study [45], using a pediatric Goal Management Training metacognitive training (with some similarities to CO-OP), parent ratings of the BRIEF improved. However, the authors suggested that this finding could have been due in part to a

questionnaire response bias rather than to pure EF improvement because BRIEF ratings might have also reflected parents' desire to be perceived as good cognitive coaches.

The content of CO-OP sessions described by Polatajko et al. [44] was used easily, but in contrast with the Missiuna et al. study [14] (in which the number of 10 sessions was respected), an increase in the number of sessions (n=14) seemed necessary to achieve the goals (perhaps because the children had a more severe profile in our study). This finding is consistent with observations of Krasny-Pacini et al. [45], and similar modifications were also implemented by Dawson et al. to use CO-OP with adults with EF dysfunction after traumatic brain injury (20 sessions) [18].

As Missiuna et al. noted, certain chosen goals (here: reading the time, writing legibly for example) were academic in nature, whereas children with Developmental Coordination Disorder in other studies typically chose motor-based goals [44]. This specificity requires an adaptation of the therapist and thus the intervention becomes more challenging. Working on 2 rather than 3 goals allowed to account for the severe executive profile of the children and allowed for achieving relatively complex goals (e.g., cooking tacos).

Children did not systematically seem to pick up the global strategy (goal, plan, do, check [GPDC]) as a tool for solving problems, similar to previous studies [14]; however, this was not assessed with a standardized tool. Clinically, the therapist had to frequently remind the child of the potential usefulness of GPDC during the sessions. Children may have internalized this strategy and no longer needed to say it out loud. The GPDC strategy helped establish a common structure and language throughout the sessions. More than an aid to the learning process (although P1 re-used it spontaneously in the classroom), the GPDC rather seemed to help focus, sustain attention, and control impulsivity. Similar to Missiuna et al. [14], we noted that children were able to use domain-specific strategies that could be generalized across other tasks. With the use of metacognitive strategies and guided discovery, the whole CO-OP

process seemed to improve problem-solving despite the EF difficulties. It seems an interesting option in EF rehabilitation after childhood ABI, with some adaptation because of the specificities of the EF deficits.

Study limitations

All baselines were not perfectly stabilized before starting the intervention (as should be the case in an SCED [25]) because baselines that were too long would have reduced children's motivation. Furthermore, only 2 children were included and the study should be replicated in a larger number of cases, if possible in different centres, with different therapists and with randomized baselines (according to SCED guidelines [25]). Regarding the EF assessments (CCT), they are not designed to be repeated at short intervals, and parallel versions were not available. Some of the observed progress could be due to a retest effect. Further studies should use parallel forms or use only 2 time points for those outcomes while favoring specific goal-directed outcome measures, such as GAS. For P1, the opportunity to apply CO-OP strategies at home (and the involvement of his parents) was limited because he was still an inpatient and returned home only on weekends. Given the small scale and pilot nature of this study, we did not have the means to externally control the fidelity of the intervention delivery. We trust that the therapist followed CO-OP principles.

Applicability

Rehabilitation with the CO-OP approach has been manualized and tested. Our clinical experience allows us to state that the sessions were easy to set up. The children took an active part in the sessions: despite their rather severe executive profile and associated deficits, they were enthusiastic, receptive to CO-OP concepts and motivated to achieve their goals. The fellow occupational therapists were very interested and asked to be trained in CO-OP after

this study. Educators and teachers found this approach very interesting for addressing problematic situations.

Conclusions

This pilot study, using an SCED methodology suggests that a CO-OP approach is feasible for children with disabling executive deficits after severe ABI, allowing significant improvements of occupational performance, achievement of personal goals, and increased executive functioning. Results were maintained at 3 months post-intervention, and several elements suggest some degree of generalization and transfer. The generally positive results of this study add to the small body of research in the use of problem-solving approaches and cognitive strategies to improve functional abilities in children with ABI. These results need to be supported by further studies, including more participants, to adapt the application of CO-OP to this population.

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Conflict of interest. None declared.

Legends

Figure 1. Evolution of Goal Attainment Scale (GAS) scores across the different phases.

◆ : baseline, ▲ : intervention, ● : follow-up (1-month post-intervention). Meaning of

scores: -2, initial level: -1, progress but goal not achieved; 0, goal achieved as expected; +1, goal achieved better than expected; +2, most favourable outcome. **Repeated judgment criteria: GAS scores, analysis by Dual Criteria Method.** The blue line represents the trend line of baseline, the red line represents the level line (=average) of the baseline. These 2 lines, projected in the intervention phase, allow for visualising the dual criterion: here, the more points above these 2 lines in the intervention phase, the more pronounced the effect of the intervention.

Figure 2. Evolution of child chosen goals: Canadian Occupational Performance Measure (COPM). Performance and satisfaction were rated by children and parents on a scale of 1 to 10 (1. not at all able to perform the activity (performance)/not at all satisfied with how the activity is performed (satisfaction); 10, perfectly able to perform the activity (performance)/perfectly satisfied with how the activity is performed (satisfaction). A difference of 2 points between pre- and post-intervention is clinically significant [36]. Missing data are represented by X=Not observed by the mother in the last few weeks.

Figure 3. Number of errors and task duration of the Children's Cooking Task (CCT): change over time, σ , z-score compared to age-matched controls; A. baseline, B. intervention; FU, follow-up.

Figure 4. Behavior Rating Inventory of Executive Function (BRIEF): evolution of parent and teacher ratings of the Global Executive Composite (GEC) T-score score. A, baseline, B, intervention, FU, follow-up (1 and 3 months post-intervention); the horizontal line represents the clinical cutoff score of 65.

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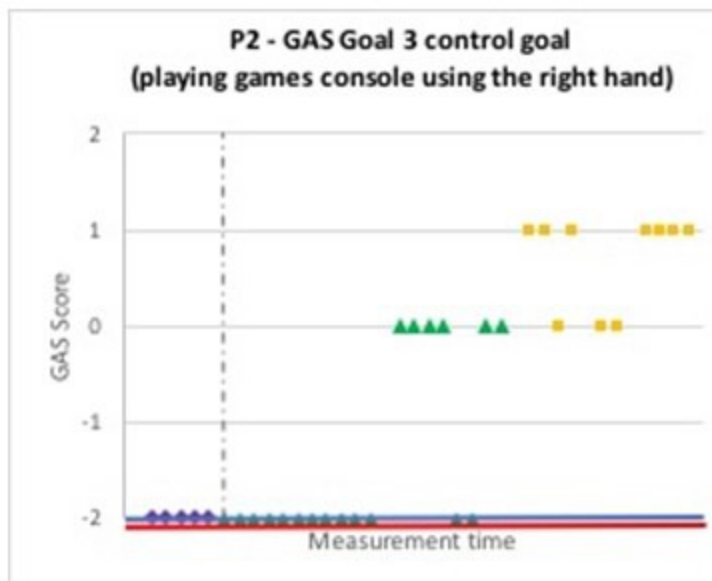
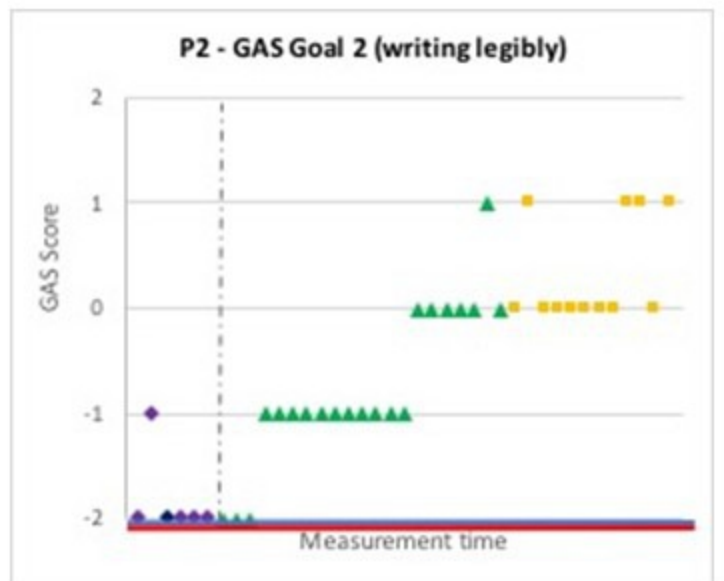
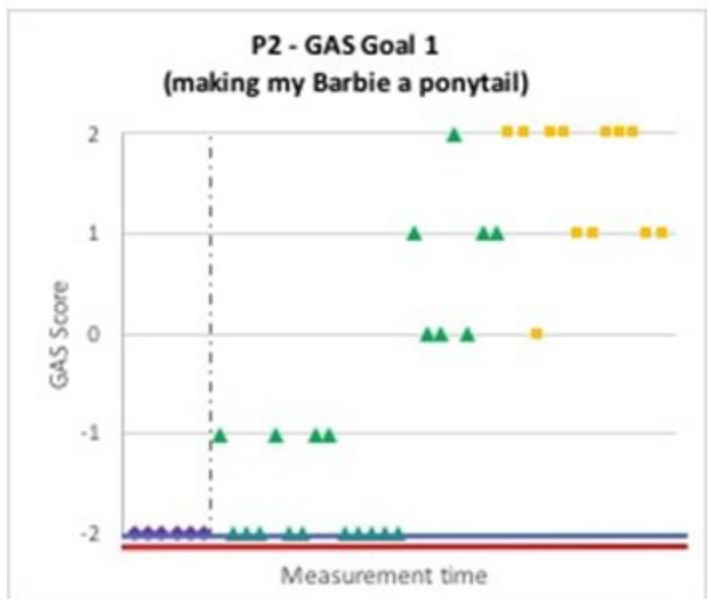
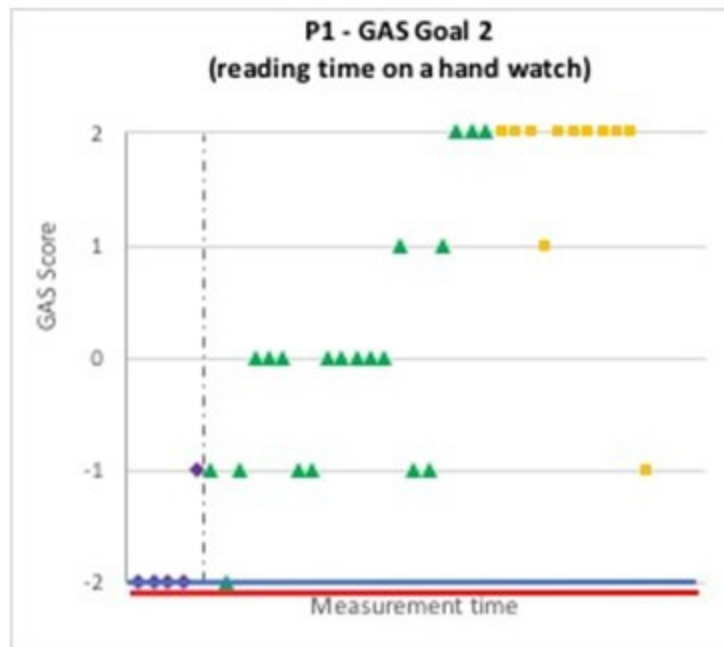
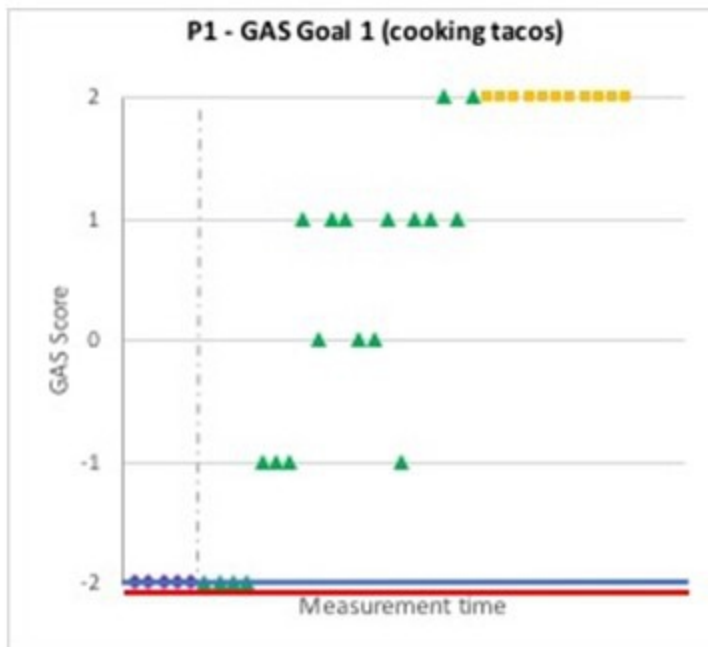
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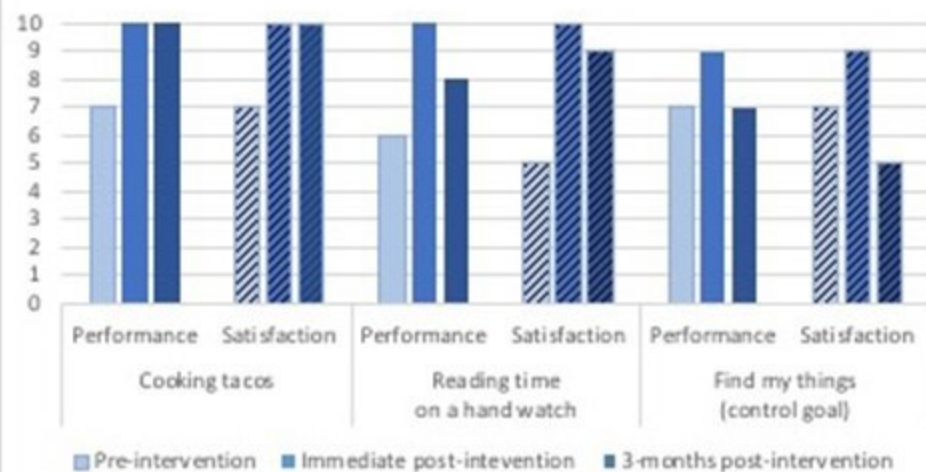
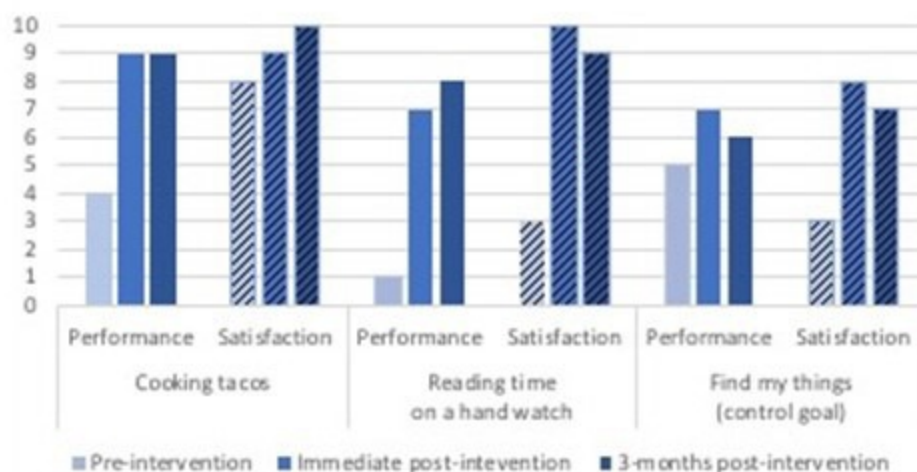
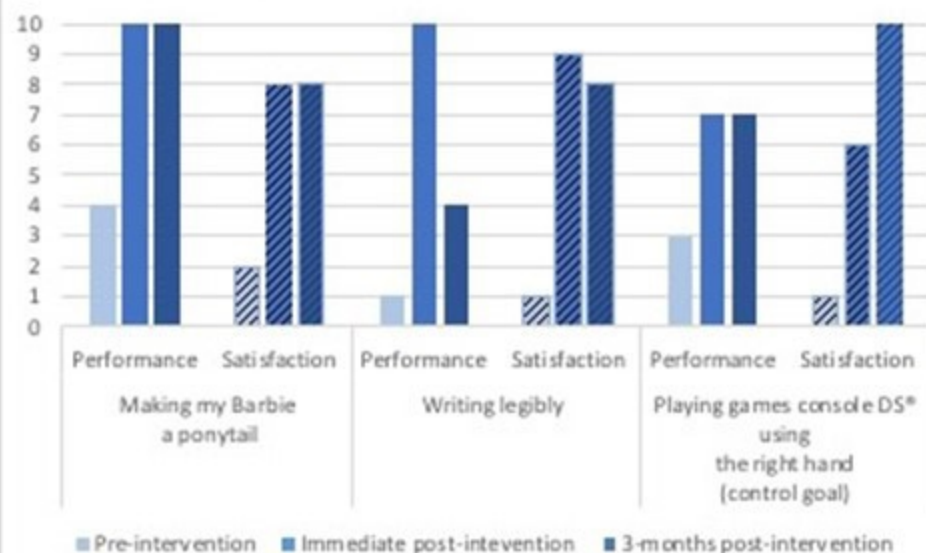
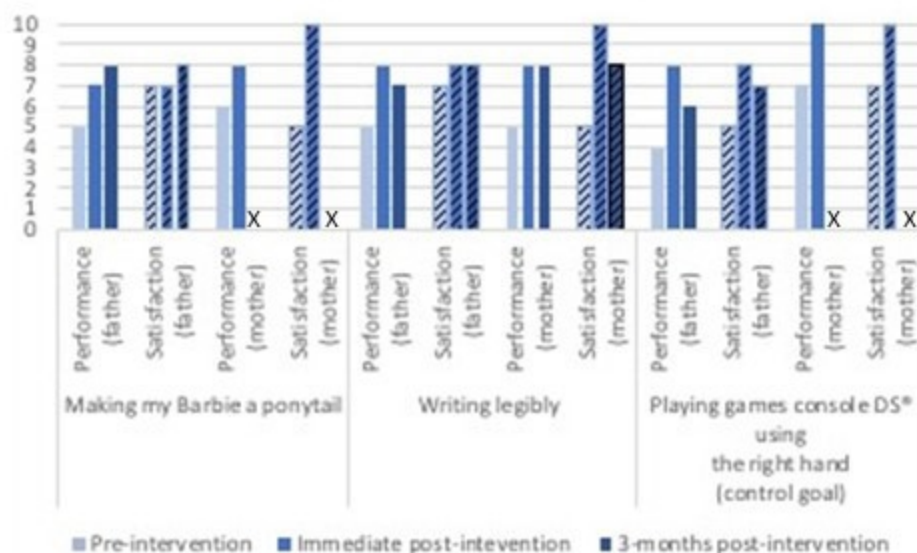
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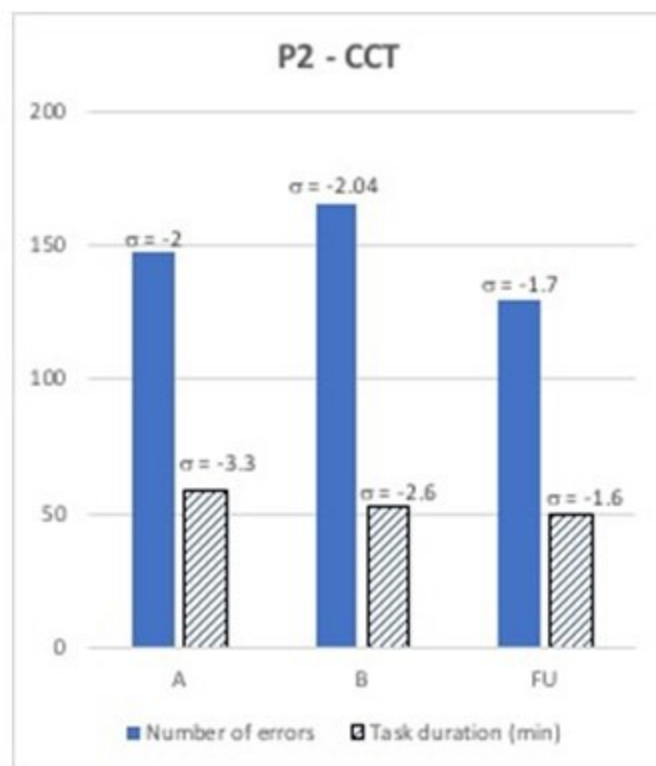
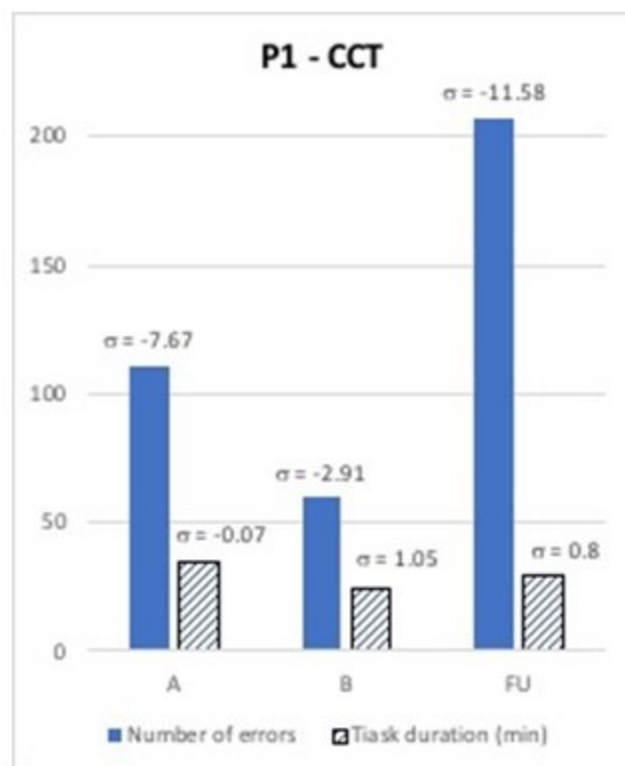
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P1 - Self assessment COPM**P1's Parents - COPM****P2 - Self assessment COPM****P2's Parents - COPM**



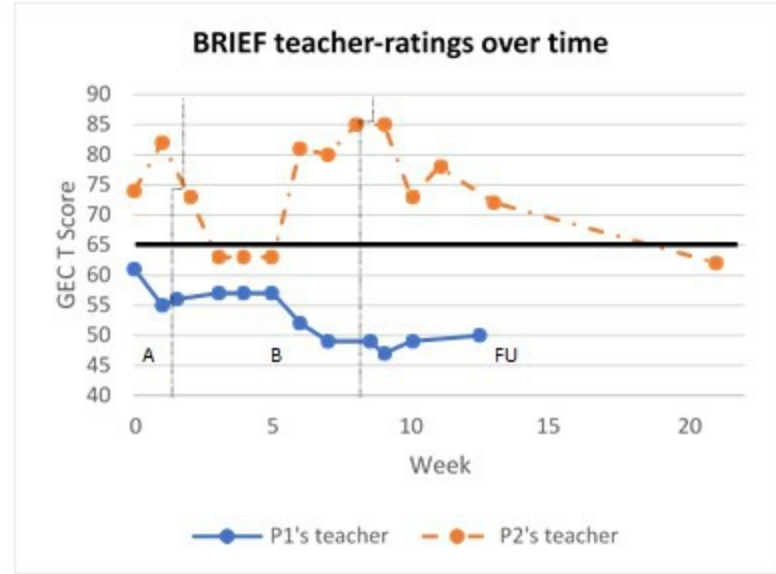
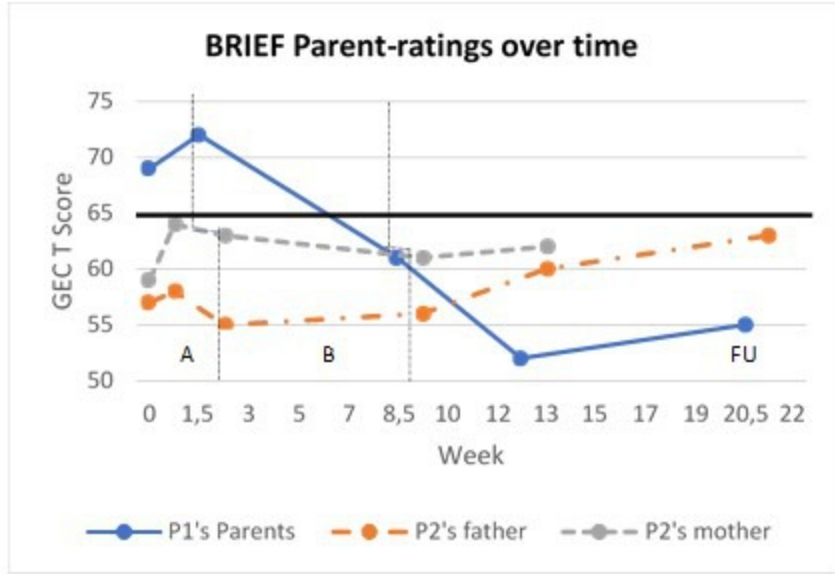


Table 1. Participant characteristics.

	Participant 1 (P1)	Participant 2 (P2)
Sex and age	9-year-old boy	11-year-old girl
Type of injury /diagnosis	Severe traumatic brain injury in a motor vehicle accident (pedestrian hit by a car)	Arterial ischemic stroke (large superficial and deep left middle cerebral artery territory)
Associated impairments	Hearing aid corrected hearing loss after the accident	Right hemiparesis with poor right-hand function in a right-handed girl (re-lateralized on the left for most activities of daily living and writing)
Schooling	Specialized classroom with a small number of students	Specialized classroom with a small number of students
Time since injury (months)	7	11
Sibling	3-year-old sister	14-month-old paternal half-brother; 1-month old maternal half-brother
Family structure	Two-parent household Father: temporary worker in building business Mother: housewife	Parents divorced. P2 lives alternatively with each parent Father: site manager Mother: nursing aid
Intellectual ability (WISC V) (composite indices; mean [SD] 100 [15])		
Verbal comprehension index	95	84
Perceptual reasoning index	94	94
Working memory index	88	82
Processing speed index	80	80
CMS (indices; mean 100 [15])		
Visual immediate memory	/	103
Visual delayed memory	/	113
Verbal immediate memory	/	59
Verbal delayed memory	/	51
Delayed recognition learning	/	84
Attention/concentration	93	84

TEA-Ch (standard scores; mean 10 [3])		
Sky search (selective visual attention):		
Targets found	14	5
Time per target	9	6
Attention score	7	6
Score (sustained attention)	10	3
Creature counting (flexibility):		
Accuracy	/	14
Timing score	/	2
Sky search (divided attention)	1	1
Score DT (auditory attention)	10	1
Same world (inhibition)	/	4
Opposite world (inhibition)	/	2
Tower of London test (SD)		
Number of achieved attempts	-0.7	-1.2
Total execution time	-0.6	-3.2
BADS-C (standard scores; mean 10 [3])		
- Six parts test	8	6
- Zoo Part 1	10	7
- Zoo Part 2	1	12
- Water test	6	1
Child chosen goals	Goal 1: outline the ingredients and steps needed to make tacos (and cook). Goal 2: read the time on a hand-watch. Goal 3 (control goal): find my belongings in a general way (loses/doesn't find his belongings in his schoolbag, his suitcase, his bedside table etc.).	Goal 1: to put my Barbie's hair in a ponytail . Goal 2: write legibly (according to BHK* criteria) so that others understand me. *BHK: Échelle d'évaluation rapide de la dysgraphie chez l'enfant (children's rapid writing assessment scale; Charles, Soppelsa et Albaret, French adaptation, 2004) Goal 3 (control goal): play game console using my right hand to press the Y, A, B buttons.

CMS, Children's Memory Scale; BADS-C, Behavioural Assessment of the Dysexecutive Syndrome for Children; TEA-Ch, Test of Everyday Attention for Children; WISC, Wechsler Intelligence Scale for Children