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Training Verbal Working Memory in Children with Mild Intellectual Disabilities: Effects on Problem-solving

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A B S T R A C T

This multiple case study explores the effects of a cognitive training program in children with mild to borderline intellectual disability. Experimental training effects were evaluated comparing pre-/post-test changes of (a) a baseline phase versus a training phase in the same participant, (b) an experimental training versus either a no intervention phase or a control training in two pairs of children matched for cognitive profile. Key elements of the training program included (1) exercises and card games targeting inhibition, switching, and verbal working memory, (2) guided practice emphasizing concrete strategies to engage in exercises, and (3) a variable amount of adult support. The results show that both verbal working memory analyzed with the listening span test and problem-solving tested with the Raven's Matrices were significantly enhanced after the experimental training.

El entrenamiento de la memoria de trabajo verbal en niños con discapacidad intelectual leve: sus efectos en la resolución de problemas

Palabras clave:
Incapacidad intelectual
Formación en memoria de trabajo verbal

R E S U M E N

Este estudio de caso múltiple explora los efectos de un programa de entrenamiento cognitivo en niños con discapacidad intelectual entre leve y límite. Se evaluaron los efectos de entrenamiento experimental comparándose los cambios pre/posprueba de (a) una fase basal frente a una fase de entrenamiento en el mismo participante y (b) un entrenamiento experimental frente a una fase sin intervención o un entrenamiento de control en dos pares de niños emparejados en el perfil cognoscitivo. Los elementos clave del programa de entrenamiento constaban de: (1) ejercicios y juegos de cartas cuyo objetivo es la atención, inhibición, conmutación y memoria de trabajo verbal, (2) práctica guiada enfatizando estrategias para realizar ejercicios y (3) un grado variable de apoyo por parte del adulto. Los resultados demuestran que tanto la memoria de trabajo verbal analizada mediante la prueba de capacidad de escucha como la resolución de problemas medida a través de las Matrices de Raven mejoraron significativamente después del entrenamiento experimental.

Working-memory has a central role in complex learning as it allows the simultaneous storage and manipulation of information. To learn a rule, for instance, individuals have to recall examples and keep them in a temporary memory store to develop an abstract schema or rule from them (Anderson, Fincham, & Douglass, 1997). Learning complex concepts also challenges working memory as one must keep active in mind the relationship between other dependent concepts. To build the concept of "son-in-law", for example, you have to keep active in mind the concepts of daughter's husband and daughter's parents and, at the same time, conceptualize the relationship between them. Working memory is involved in learning something new (Cowan, 2014), when logical or semantic connections between different elements still have to be established. Before associations are formed between the parts of a new procedure or a new concept, working memory is particularly taxed.

According to an influential multi-component model (Baddeley, 2000, 2010; Baddeley & Hitch, 1974), working memory consists of a central executive whose limited attentional control capacity is responsible for the active maintenance and processing of task-relevant information, which is temporarily held in domain-specific

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verbal and visuospatial stores or a multi-modal episodic buffer (Baddeley, 2000). The description of the central executive as a cluster of executive functions whose specific control process consists of updating the contents of working memory, switching between different tasks or procedures, inhibiting irrelevant information or actions, and coordinating multiple tasks is consistent with this model (Baddeley, 1996; Miyake & Friedman, 2012).

An increasing number of studies have examined the effects of working memory training but research exploring whether executive functions and working memory can be effectively enhanced in children with mild to borderline intellectual disability (ID) has been relatively scarce. This type of clinical population, however, would particularly benefit from evidence-based interventions targeting these cognitive functions.

In fact, it is well known that children with ID have lower performance than chronological age comparisons in most tests assessing executive functions (Alloway, 2010; Danielsson, Henry, Messer, & Rönnberg, 2012; Levén, Lixell, Andersson, Danielsson, & Rönnberg, 2008). Behavioral inhibition and interference control are particularly impaired in this population (Bexkens, Ruzzano, Collotd'Escury-Koenings, Van der Molen, & Huizinga, 2014). Studies analyzing performance in working memory tasks in children with ID show heterogeneous domain specific effects (Van der Molen, Van Luit, Jongmans, & Van der Molen, 2007) that are related to disorder-specific impairments (Jarrold, Baddeley, & Hewes, 1999; Jarrold, Purser, & Brock, 2006; Lanfranchi, Cornoldi, & Vianello, 2004). However, there is evidence that working memory (WM), particularly in the verbal domain, is weaker compared to mental age peers in most children with ID (Jarrold, Baddeley, & Hewes, 2000; Schuchardt, Gebhardt & Maehler, 2010; Schuchardt, Maehler & Hasselhorn, 2011; Van der Molen, Van Luit, Jongmans, & Van der Molen, 2009) and even in children with borderline intellectual functioning (Hasselhorn & Maehler, 2007; Henry, 2001; Henry & MacLean, 2002).

The weak development of working memory and difficulties with attentional shift (Vakil & Lifshitz-Zehavi, 2012) contribute to a range of other cognitive deficits in this developmental clinical population. First, problem-solving processes tend to be less effective if not supported by working memory. Henry and MacLean (2003), focusing on the relation between analogical reasoning and the different components of WM, found that measures tapping the central executive were the most significant predictors of arithmetic reasoning for participants with ID. Second, poor verbal working memory is likely to have detrimental effects on those processes in which language can support concepts and skills acquisition, from following instructions (Gathercole, Durling, Evans, Jeffcock, & Stone, 2008) to lexical-semantic acquisition (Baddeley, 2003). Third, there is a strong association between working memory and executive functions on one hand and academic learning on the other hand in children with intellectual disabilities (ID) and borderline intellectual functioning (Henry & Winfield, 2010; Numminen et al., 2000; Poloczek, Büttner, & Hasselhorn, 2012). Even externalizing behavior problems seem to be associated to impaired working memory in these children (Schuiringa, van Nieuwenhuijzen, Orobio de Castro, & Matthys, 2017).

Although such correlational data suggest that executive functions and working memory should be a preferential target of cognitive training methods for children with mild to borderline ID, the questions of whether such functions can be effectively enhanced and whether other cognitive functions or learning processes can improve as an indirect effect of WM training are still open.

Moalli, Rota Negroni, and Vianello (2004) explored the effects of a training method focused on both teaching concepts on memory functioning and practicing specific mnemonic strategies with verbal and visuospatial tasks. Improvements from pre- to post-treatment in verbal short-term memory and visuo-spatial WM occurred in children with Down syndrome (DS) when they were compared to a control group. Training effects on verbal working memory were not explored in this study.

Van der Molen, Van Luit, Van der Molen, Klugkist, and Jongmans (2010) used a dual task in which children were asked to process the current stimuli (e.g., identifying which figure is the odd one), and remember a target item (e.g., recalling a target location across increasingly longer spans). A large group of adolescents with mild-to-borderline intellectual disabilities participated in either an adaptive or a stable training regimen with the visual dual task; a control group was trained with a single task. Results showed that children trained with dual tasks (no matter whether adaptive or stable) improved their visual WM only at follow-up testing, whereas performance with verbal WM was not affected by training in any testing phase. The authors also found transfer effects on arithmetic and story recall at follow-up, but no transfer effects on performance with the Raven's Matrices.

Söderqvist, Nutley, Ottersen, Grill, and Klingberg (2012) analyzed the effects of a training procedure combining WM and non-verbal reasoning (NVR) tasks. A sample of forty-one children with ID participated in two training groups that used the same NVR tasks but differed regarding their treatment with either adaptive or non-adaptive, computerized, visual, simple-span tasks. There was large individual variability in children's responses to intervention, and only children who made remarkable progress in the training tasks showed improved performance in verbal or visual working memory at post-testing. This study shows that progress in verbal WM after training can occur in children with ID but with highly variable individual differences.

Bennett, Holmes, and Buckley (2013) used a computerized WM training consisting of visuospatial simple and complex span tasks. Children with Down syndrome (DS) aged seven to twelve years were allocated to either the intervention program or a waiting list group. Children in the intervention group significantly improved for visuospatial WM both immediately after the training and at four-month follow-up but the training showed no effects on verbal WM. Parent ratings of behavior also showed that after training there was a highly significant reduction in difficulty with shifting behaviors for children in the intervention group.

Danielsson, Zottarel, Palmqvist, and Lanfranchi (2015) performed a training group minus control group analysis in a meta-analytic review and concluded that only mixed WM training, with both verbal and visuo-spatial components, showed significant training effects in studies involving children with ID. An analysis of the training effects distinguishing verbal and visuo-spatial short-term memory (STM) and WM showed larger effect sizes for the STM tests.

In summary, most studies involving children with ID used computerized training of visual or visuo-spatial WM (see also Pulina, Carretti, Lanfranchi, & Mammarella, 2015). Such training seems to generate remarkable enhancements especially in visual working memory though the effects tend to be medium-small in terms of effect size (Danielsson et al., 2015). Effects on verbal working memory – as assessed by dual tasks asking both processing and memorization of verbal stimuli – are rare (but see Costa, Purser, & Passolunghi, 2015; Orsolini, Melogno, Latini, Penge & Conforti, 2015; Söderqvist et al., 2012). Transfer effects of WM training to academic learning or everyday functioning are also rare (but see Bennett et al., 2013 and Van der Molen et al., 2010) whereas transfer to problem-solving and reasoning are not documented for children with ID.

Thus we are at an early stage of research on the effects of working memory training programs for children with intellectual disabilities and there is a need for group studies to identify the training conditions that are more suited to this clinical population. On the other hand, involving children with cognitive deficits in training programs that do not target academic skills cannot answer yet parents' and educators' concern that such programs may take time away from more evidence-based instructional practices. Case studies may therefore be particularly useful in such a phase to collect preliminary evidence

that the time spent in working memory training can be beneficial to other cognitive processes, such as inference and reasoning skills.

In line with this preliminary research objective, our multiple case study explores whether verbal working memory can be enhanced by training in children with a mild to borderline intellectual disability of a non-specific etiology who have a history of language delay. It also analyzes whether training verbal working memory generates a transfer effect to problem-solving and cognitive flexibility.

Method

Participants

After approval from the Ethics Committee of the Department of Developmental and Social Psychology (Sapienza University of Rome), an informed consent was asked to the parents who accepted to have their children involved in this study. Participants were children whose mild intellectual disability (IQ between 55 and 70) or borderline intellectual functioning (IQ between 71 and 85) had been diagnosed by a certified clinical psychologist within public clinics in the area of Rome who tested them with the WISC-III (Wechsler, 1991) and assessed their adaptive functioning. The children had shown wide learning disabilities since their first grade class and were assisted by a special educator who, according to the Italian law, helps the children with special needs for a varying amount of time (accordingly to the severity of their impairment) within regular classes. Children were selected by either a psychologist within the public clinic in which the diagnosis had been issued or the child's special educator. The following selection criteria were used: (a) the child had some type of language delay (see Table 1) and the psychologist or the child's special educator judged that he/she could benefit from a training targeting verbal memory, (b) the child's parents had communicated an intention to involve the child in a therapy and were deemed to be motivated to support their child's engagement in the training, and (c) the psychologist or the child's special educator judged the child to be motivated to participate at new learning experiences.

The Experimental Training

Key elements in our program included (1) specific activities stimulating attention, inhibition, switching, and verbal working memory; 2) guided practice emphasizing strategies to engage in exercises (e.g., verbalization to promote the task's goal maintenance); 3) adapting the adult's degree of support to the task difficulty and the child's level of performance; and 4) sessions starting with an initial conversation and going on with one adult's led exercise presented through PowerPoint and one card game (see examples in Table 2).

Attention. As illustrated in Table 2, our experimental training started from attention, as attention is involved in working memory (Vandierendonck, 2014), and it is known that weak attention skills are often present in children with ID, with a strong negative impact on working memory (Kirk, Gray, Riby, & Cornish, 2015). Activities in this unit asked the child to identify parts of incomplete pictures, name elements of complex scenes that were shown on the computer screen for a limited amount of time, and describe features in order to support a character's identification for the other player.

Inhibition. Activities stimulating inhibition of a dominant response asked participants to process affirmative and negative sentences to accomplish selection of target items (e.g., "The thief wears a red tie", "The thief does not have blond hair") or lexical-semantic categorization of pictures (e.g., the child is shown four pictures on the computer screen and is asked to quickly name the only picture belonging to a target category for some slides and then to name the only picture not belonging to a target category for other slides).

Switching and simple verbal working-memory tasks. Activities in this unit asked participants to practice different actions in the same exercise (e.g., looking at the picture and either saying something that was not true for that picture or saying something that was true but different from the word that was written on the top of the picture). Simple verbal working-memory tasks asked participants to recall sequences of items and accomplish, at the same time, a selection of items according to a target semantic category or other target characteristics.

Complex working memory tasks. Participants were asked either to recall information after having accomplished a different task (e.g., recalling a sentence after having judged whether that sentence was friendly or not) or to accomplish inferences (e.g., guessing the place in which a short dialogue has occurred) after the content of a short passage had been listened to and encoded in episodic memory.

Table 1. Participants' Characteristics[1] and Methods of Analyzing the Training Effects

	Participants				
	Ilaria (female)	Simone (male)	Roberta (female)	Lucrezia (female)	Dino (male)
Methods of analyzing the training effects	Baseline versus training versus follow-up assessment after interruption	Training versus waiting list		Experimental versus control training	
Age (years, months)	10, 6	10, 3	9, 6	12, 3	12, 1
IQ (WISC III)[1]	70	70	70	75	72
Language difficulties	Low lexical comprehension (IQ = 72)[1] Low verbal short-term memory (z = -1 in a word span test)[2]	Phonological delay[1] Deficit in verbal short-term memory (z = -2.3 in a word span test)[2]	Phonological delay[1] Low verbal short-term memory (z = -1 in a word span test[2])	Slow lexical access (word finding difficulties)[1] Low verbal short-term memory (z = -1.3 in a word span test)[2]	Phonological delay[1] Low verbal short-term memory (z = -1.66 in a word span test)[2]
Diagnosis	Mild Intellectual disability	Mild Intellectual disability	Mild Intellectual disability	Borderline intellectual functioning	Borderline intellectual functioning
Setting of the training	Home	School		University clinical center	

[1]Characteristics reported in this table are drawn from the reports of the participants' assessment in the clinical center that issued the original diagnosis and are based either on standardized tests (e.g. Lexical comprehension was assessed through the Italian adaptation of the *Peabody Picture Vocabulary Test*, Dunn & Dunn, 1981) or clinical observations made by the psychologists and speech therapists involved in the original diagnosis.

[2]The word span test is the initial condition of the Word Interference test (Nepsy II; Korkman et al., 2007).

Table 2. The Experimental Training Stimulating Verbal Working Memory

	Attention	Inhibition	Switching and simple verbal working memory	Complex working memory
Adult's led interaction is focused on enhancing…	Verbalization of stimuli Systematic visual exploration Sustained attention Selective attention	Maintenance of the task's goal Divided attention Selection of members of target categories.	Rehearsal strategies Task planning and sequencing Focus on relevant information Summarizing the available information Anticipation of possible sources of difficulty Generalization of approach to different tasks	
Examples of computer-presented exercises and card games	· *Animal detective:* An incomplete picture appears on the computer screen and quickly disappears. The participant is asked to recognize the animal and then identify the lacking part of the picture, selecting it from four cards. · *Monsters:* Therapist and child take turns in selecting one or more cards with monsters, describing their characteristics and communicating the precise location in which they put them. If the second player (who cannot see what the first is doing) makes the same choices as his/her companion does, the first player wins some points.	· *Characters detective:* A thief has been seen from people who describe his/her characteristics. Relying on each of such descriptions (e.g., "the thief was not a woman" or, "the thief did not wear glasses"), the participant removes images from a pool of suspects until the thief is identified. · *Category:* Each player has six cards and proceeds on a game of the goose board if he/she can play cards according to the category specified on the board box. Categories may be single or multiple (e.g., "food and furniture") and affirmative or negative (e.g., "no fruits, no clothes").	· *Guessing what:* The participant is asked to discover what the object hidden on the computer screen is by relying on the information provided by two types of characters. A wizard will say something that is opposite of the real characteristic (e.g., "if the wizard says that the thing is put on a lower part of the body, you have to think that it is put on an upper part of the body"). A pessimistic man will say something true but will add pessimistic evaluations that may distract you (e.g., "he will say that you wear this thing when it is hot, and he will add that if you do not do so, it may be very dangerous, and you can even die"). · *The dolphin game:* Players proceed with a game of the goose if they can repeat the sequence of words that has being said by the other player and add a new word according to the instruction specified on the board box. Boxes on the board ask for a fixed number of words (from 2 to 6) either starting with a given letter or belonging to a given category.	· *Stories:* Short narrative sequences are read by the adult and are also shown on the computer screen with the written text accompanied by a picture. For instance: "A hare was very proud of herself because she could run quickly. One day she said to all the other animals: - Nobody is quicker than me; nobody has the courage to race with me-". After the last sentence is read, the short passage disappears from the computer screen, and the participant is asked to produce a pragmatic judgment (e.g., "is what the hare says friendly?") and then to recall the sentence. · *Take cards and remember:* Each player has three picture cards and can take one of four picture cards on the table, following the given rules (e.g., humans can take animals, animals can take plants or fruits, plants or fruits can take objects). At the end of the round, each player attempts to recall the word that was written on each of the taken cards (e.g., the word "surprise" written under the image of a birthday cake), and if he/she manages to do so, he/she wins the cards.

The Control Training

Control training targeted narrative memory and visuo-spatial working memory. Narrative memory was trained with both conversation and structured activities (see Table 3) asking the participant to recall personal events, verbally reconstructing the plot of a short video clip, imagining fictional events to link pictures in card games. Visuo-spatial working memory was trained with a software (Mammarella, Toso, & Caviola, 2010) targeting first immediate attention and memory of visual stimuli and then active memory involving dual tasks of maintaining and processing information. This software considers two aspects of visuo-spatial WM: the nature of the stimulus (visual, spatial-sequential, and spatial-simultaneous) and the level of attentional control, with tasks demanding a low, medium or high level of control.

Table 3. Activities Stimulating Narrative Memory in the Control Training

Conversation	The participant was asked to verbally share personal events and the therapist also produced personal narratives. The therapist used questions to support narrative expansion soliciting the child's recalling of events, acknowledged the participants evaluations, and occasionally rephrased or synthesized the child's utterances.
Recalling video clips	The therapist and the child looked at a video clip showing one episode from popular cartoons. At the end the child was asked to image that the video transformed in a picture book: "what would show the first picture?" No matter whether the answer of the child described the starting event or a subsequent one, the therapist showed a printed picture depicting the event described by the child and solicited some semantic elaboration. The therapist's subsequent questions asked the child to recall "what happened then?" and again used printed pictures to enrich the child's recalling of events.
Sentences linking different images and verb phrases	The child can use card pictures and language cards. A specific verb phrase is selected from language cards (e.g. ___ realizes that ____) and the child is asked to produce a sentence with that verb phrase including one or more card pictures. There are also special language cards with connectives such as "when", "because", "if", "and at the end" that can be used to build a longer sentence. The therapist and the child take turns in producing sentences and compete for producing the longest but "coherent" sentence.
The Goose game	The gameboard has pictures depicting fictional characters, actions, objects. Each player can proceed on the board if manages to link the events described by the previous player to the one depicted in the picture where he/she landed after throwing the dice.

Similarly to the experimental training, each session started with conversation to promote a close adult-child relationship and build a practice of sharing personal memories. After such a warming stage, there were exercises with the software stimulating visuo-spatial working memory followed by structured narrative activities.

Experimental Design

Case studies have to specify the conditions under which their results could be replicated (Wolery & Ezell, 1993). To understand whether the training could be effective regardless of the setting in which is delivered, we choose to involve the participants in a training that took place either at the child's home (Ilaria) or at school (Simone), or at a university clinical center (Lucrezia).

Studies exploring whether participants' performance can improve after cognitive training have to rule out that increasing exposure to tests, or unspecific factors, such as increased motivation, are the crucial conditions generating changes. To understand whether participants' improvements from pre- to post-training assessment could be interpreted as generated by the training itself, we choose three types of comparison, as shown in Figure 1. The first participant, Ilaria, was involved in a within-subject design by comparing the effects of a baseline condition with those of the experimental training, and then with those of a training interruption (see Figure 1). Each phase of baseline, training, and interruption consisted of eight weeks and was preceded and followed by testing.

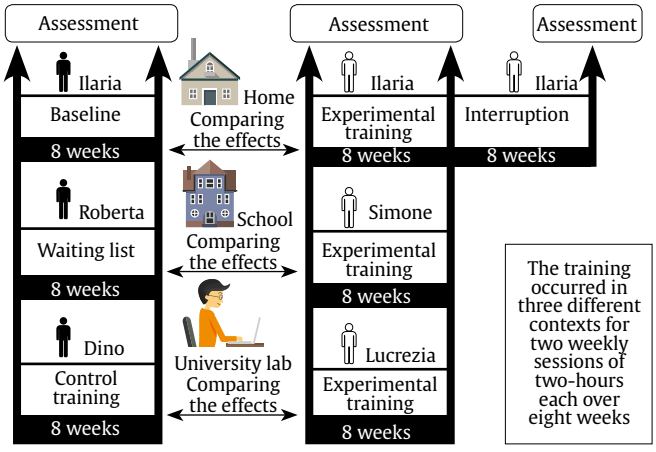

Figure 1. The Research Design.

A second participant, Simone, was involved in a between-subject design and participated at the experimental training taking place at school and lasting eight weeks. Simone's pre-/post-training improvements were compared with those of Roberta who was assessed before and after eight weeks of a "waiting list" phase. Simone and Roberta were selected in the same school, attended the same grade IV class, and showed a similar cognitive profile (see Simone versus Roberta in Figure 1).

In order to further examine training effects and distinguish them from either test familiarity effects or generic effects introduced by motivation, we involved a third participant, Lucrezia, in a between-subject design and compared her pre-/post- experimental training improvements to the pre-/post-control training changes observed in Dino. Lucrezia and Dino were selected in the same University clinical center and showed a similar cognitive profile (see Lucrezia versus Dino in Table 1 and Figure 1).

Thus our study can evaluate both test familiarity and training effects. Tests' familiarity effects were analyzed in two ways: analyzing pre-/post-test changes after either a baseline phase in Ilaria or a waiting list phase in Roberta. The experimental training effects were evaluated through three types of comparison: analyzing pre-/post-test changes after (a) a baseline versus an experimental training phase in Ilaria, (b) an experimental training versus a no intervention phase in Simone and Roberta, (and c) an experimental training versus a control training in Lucrezia and Dino. We could replicate an experimental effect three times across different participants, in line with one basic criterion recommended by Horner et al. (2005) for single-case designs.

Dependent Measures and Materials

The participants' pre-/post-test changes were analyzed assessing attention, executive functions of inhibition and switching, verbal short-term memory, verbal working memory, problem-solving, and cognitive flexibility.

Attention. Selective and sustained visual attention was evaluated with the Bells Test (Biancardi & Stoppa, 1997), in which the participant must cancel 35 pseudo randomized bells found on a horizontal sheet of paper mixed with another 315 figures. The bells are located in seven columns, three in the right visual field, three on the left, and one in the centers. As the participant's task is to locate the bells and cross them out in the shortest possible time, and to repeat the search across four sheets, we used two main scores: the number of bells crossed in the first 30 seconds of each of the four sheets and the total number of bells crossed in the 120 seconds allowed for each of the four sheets. The first score is likely to involve selective attention; the second score taps the participant's capability of sustaining attention effectively to the same visual search target.

Inhibition and switching. In this timed test of the Nepsy II battery (Korkman, Kirk, & Kemp, 2007), the ability to inhibit automatic responses in favor of novel responses and the ability to switch between response types is assessed. In the Naming phase of the task, the participant looks at a series of black and white shapes (circle and square) or arrows (pointing up and down) and names either the shape or the direction. In the Inhibition phase, the child names the same symbols but is asked to apply the non-target label (e.g., saying "square" for a circle or "up" for an arrow pointing down). In the Switching phase, the child is asked to say the correct name for black symbols but to apply the non-target label if the symbol is white (e.g., "down" for a white arrow pointing up or "circle" for a white square). The completion time and the total number of mistakes (including self-corrections) are evaluated for naming, inhibition and switching.

Verbal short-term memory. A Forward digit span (Gugliotta, Bisiacchi, Cendron, Tressoldi, & Vio, 2009), in which the examiner reads a list of numbers – a digit per second – and the participant must immediately repeat them back, was used to evaluate verbal short-term memory. The starting point in the task is a three-digit list, and the span is increased until the participant fails in all three lists of the same span. The score is the highest span in which the child manages to correctly repeat two out of three lists of that span. Verbal short-term memory was also tested with a word span using the first part of the Word Interference test from the Nepsy II. The child is presented in an auditory manner with blocks of words increasing in span (from two to five) and is asked to repeat them in the same order. The number of blocks correctly repeated is the task score.

Although the experimental training did not target verbal-short term memory with specific activities, exploring whether some changes are induced in such function will allow us to better understand the underlying nature of verbal working memory improvements, clarifying whether they are related to a more effective primary memory for verbal information or a stronger central executive enabling children to cope with dual tasks in the verbal

domain.

Verbal working memory. Verbal working memory was assessed with both a simple and a complex dual task. The simple task, Backward digit span (Gugliotta et al., 2009), is similar to a Forward digit span in the presentation of the items and score assignment, but at the end of each sequence, the participant is asked to recall the presented digits in the reverse order. The complex dual task is the Listening span test, an Italian adaptation (Pazzaglia, Palladino, & De Beni, 2000) of the Daneman and Carpenter's (1980) task consisting of sentences that are auditorily presented in blocks of increasing span (from two to six). The participant is asked (i) to judge the plausibility of each sentence (state whether it is true or false) and (ii) to recall the last word of each sentence, in the correct order, at the end of each block. The total number of words correctly recalled *in order* provides one type of score. For instance, if a subject is presented with a six-span block and recalls the last word of the third and fourth sentences in the right order, the score in this block would be 2. Further types of score are the number of errors with sentence judgements and the number of intrusion errors. Intrusion errors consist of recalling words that do not occupy the sentence ending position (e.g., recalling "football" instead of "mountain" for the sentence *Football is a sport that you can only practice in a high mountain*), and a high number of intrusion errors is an indicator of difficulties in inhibiting irrelevant information.

Problem-solving. The Raven's Colored Matrices (Raven, Court, & Raven, 1992) were used with the primary school participants and the Standard Progressive Matrices (Raven, 1989) consisting of 60 items were administered to the two 12-year-old children.

Cognitive flexibility. The Animal Sorting test from the Nepsy II (Korkman et al., 2007) was used to assess concept formation and the ability to flexibly shift from one concept to another. The child sorts pictures cards as quickly as possible into two groups of four cards each, using self-initiated criteria. The score is the number of correct different categories in which the participant sorts the pictures cards.

Procedure

The children's parents were interviewed by a therapist who illustrated the treatment and made explicit that it would target "basic abilities" – such as verbal working memory or narrative memory – rather than academic skills. At the end of the interview parents signed an informed consent.

Therapists were 5th year developmental psychology students undertaking an intensive training course in which they could follow the guiding principles of each type of experimental and control training activity and practice simulations of adult-child verbal interaction. Therapists were supervised by the first author on a regular basis and there was a checklist asking them to report observations on the child´s performance in each training session. Each training activity had written instructions which therapists were instructed to follow.

In the baseline phase of Ilaria (see Figure 1) the therapist visited the participant at home and engaged for eight weeks in two weekly sessions of one hour play and conversation with the objective of building an adult-child warm relationship. This was followed by an

Table 4. Difference in Standard Scores Between Post- and Pre- assessments

		Visual attention		Executive attention		Verbal short-term memory		Verbal working memory		Problem-solving	Cognitive flexibility
		Selective attention	Sustained attention	Inhibition	Switching						
				Combined score	Combined score	Forward Digit Span	Forward Word Span	Backward Digit Span	Listening Span Test	Raven's Matrices	Animal Sorting
Ilaria	Difference post/ pre-test after the baseline condition	1.55[&]	0.46[&]	0	-0.33	0[&]	0.66	0.90[&]	0	0.49	0.33[&]
	Difference post/ pre-test after the experimental training	0.41	0.11	0.67	1.00*^	0	0.66	0	2.17**^	1.50*^	0.66
	Difference post/ pre-test in the follow-up	0.40	0.21	0	0.66	0	0	0	0	0.39	0.33
Simone	Difference post/ pre-test after the experimental training	0	0.11	1.00*	-0.66	0.48	1.66*	0.50[&]	1.88*^	1.43*	1.66*
Roberta	Difference post/ pre-test after the waiting list period	-0.03	0.47	2.00**^	1.66**	0	0	0.05[&]	-0.62	1.48*	0
Lucrezia	Difference post/ pre-test after the experimental training	2.09^	3.12^	1.33*^	1.00*	-0.85[&]	0	0[&]	1.18**^	1.53*[&]	0.66[&]
Dino	Difference post/ pre-test after the control training	1.52	2.00	-2.00 [&]	0.33	0	0	0.90[&]	0.51	0.40	2.33**^

Standard scores preceded by a minus sign mean that the post-test standard score was lower than the pre-test one.
*Standard scores with a reliable change index of 1.96 or greater and that equate to 95% confidence interval.
**Standard scores with a reliable change index of 2.58 or greater and that equate to 99% confidence interval.
^Standard scores changed from being below the mean (i.e., -1 or lower) in the pre-test to be within the normal limits in the post-test (i.e., -0.7 or higher)
[&]Standard scores were within the normal limits in the pre-test

experimental training phase of eight weeks consisting of two weekly sessions of two-hours each, in which the child was engaged with the experimental training activities. Then there was an interruption phase of eight weeks that coincided with the summer vacations and was followed by a follow-up assessment. The person who tested Ilaria was the same person who met her once a week in the baseline condition and was then involved as a therapist in the experimental training (N.L., the fourth author).

Simone was involved in a school-based experimental training consisting of two weekly sessions of two-hours each over eight weeks that took place in a primary school laboratory. In the first weekly session the child was involved with computer-presented training exercises (see Table 2) and interacted individually with a therapist. In the second weekly session Simone was involved with the card games of the training and interacted with another child with learning disabilities along with the therapist. Simone's pre-/post-test changes were compared to those of Roberta, who participated at a "waiting list". The two children were assessed before and after either the training or the "waiting list" phase by the same person, who was not involved in Simone's training.

Lucrezia and Dino were involved in the experimental and control training respectively that took place in a University clinical center and were delivered by the same therapist. Both experimental and control training consisted of two individual two-hour weekly sessions over eight weeks, in which the child interacted with the therapist in a room at the center. The two participants were assessed before and after training by the same person (N.L.) who had not been involved in their treatment.

"Near" and "far" transfer training effects. The first issue explored by our study is whether there are "near transfer effects" of the experimental training, asking whether specific cognitive functions (i.e., executive functions of inhibition and switching, verbal WM) that have been directly stimulated by the training improve in the post-treatment assessment. The tests assessing such cognitive functions consist of tasks quite different from the training activities. For instance, verbal working memory was assessed through two tests: backward digits recall and the listening span test. There was no training activity asking participants to implement the manipulations required by these two tests (e.g., recalling items in a reverse order or recalling in the correct order the last word of sentences that have been first judged true or false). As tests and training activities consist of different tasks, this allowed us to assess a true *transfer* effect. As the tests' tasks involved the cognitive functions that have been directly stimulated by the training, *near* transfer effects can be detected.

Our second issue was whether the experimental training had transfer effects for other than trained cognitive functions. Whereas the experimental training stimulated central executive attention, working memory, and inferential processes in the verbal domain, we assessed problem-solving and concept formation in the visual domain, through Raven's Matrices and a cognitive flexibility test (Animal Sorting from the Nepsy II; Korkman et al., 2007), respectively. Selecting tests in a domain that was not specifically stimulated by the training allowed us to evaluate the transfer potentiality of the experimental training.

Analyzing reliable pre-/post-test changes. To analyze a training effect we have to ask if a participant's improvement from pre- to post-test is significant or is just a variation stemming from the imperfect reliability of the chosen test. The reliable change index method (RCI; see Bauer, Lambert, & Nielsen, 2004; Jacobson & Truax, 1991; Norup, Spangsberg Kristensen, Poulsen, & Mortensen, 2017) was used to determine the statistical significance of change with eight-week test-retest interval that occurred either for a baseline, a waiting list, or a training phase. RCI allows you to determine who has changed reliably and is calculated as $(X_2-X_1)/S_{diff}$, where X_1 and X_2 are the individual's observed scores in the pre- and post-test, and S_{diff} is the standard error of the difference between the two test scores. The standard error of the difference takes into account the test standard deviation (*SD*) and the test reliability (rel), as follows: *SD**sqrt(2)*sqrt(1-rel). Only if the RCI is 1.96 or greater the difference is statistically significant (1.96 or 2.58 equates to the 95% or 99% confidence interval respectively). Following Norup et al. (2017), we also distinguished between post-test scores showing only a reliable change and those scores that changed from being outside the normal limits in the pre-test to be within normal limits in the post-test. We will refer to such type of scores as "clinically significant change". Such change would suggest that a training is effective in supporting an internal process of learning and development bringing specific behavioral parameters within the normal range.

Results and Discussion

We converted each raw score into a standard score, considering the chronological age norms of each test. The difference in standard score between the post- and the pre-training assessment of each test was then computed, as shown in Table 4. Marked with one or two asterisks are the scores showing statistically significant reliable change index. We will refer to such scores as "reliable" changes, meaning that the difference post-/pre-test is not "just statistical noise resulting from the lack of the perfect reliability of the chosen assessment instruments" (Norup et al., 2017).

To compute the standard error of the difference between the post- and the pre-test scores the tests' reliability was considered by selecting it from the English manual of the Nepsy II (Korkman et al., 2007). For the Raven's Matrices we considered the minimum reliability of .80 emerging from Carlson and Jensen (1981). For the listening span test we considered the reliability reported in Pazzaglia et al. (2000). Reliability of the attention test was not available and the significance of the pre-/post-test difference could not be computed for this test.

Marked with "^" in Table 4 are the pre-/post-test scores that not only show a reliable change but also improved from being outside the normal limits in the pre-test to be within normal limits in the post-test. We will refer to such type of scores as "clinically significant change".

"Near" Transfer Effects

Table 4 shows the results on selective and sustained attention: out of the four children who could receive the effects of either experimental (Ilaria after the training, Simone, Lucrezia) or control training (Dino), only two (Lucrezia and Dino) showed a marked improvement. It is clear that the experimental training did not induce specific effects on attention, at least when such function is assessed through a visual search task.

Focusing on executive attention, we considered for Inhibition and Switching the standard scores that combine errors and completion time (combined scaled scores). Test familiarity effects occurred for Roberta who remarkably improved for Inhibition; for Switching she changed from being unable to pass the familiarization phase in the pre-test to get a score corresponding to 1 standard deviation below the mean in the post-test. We attributed to Roberta's initial assessment of Switching the same pre-test score she showed for Inhibition and thus her pre-/post-test difference turned out to be a reliable change. Test familiarity effects, that could have been observed in two children (Ilaria after the baseline, Roberta after the waiting list period) were only observed in Roberta, whereas experimental training effects occurred for three out of three children (Ilaria after the training, Simone and Lucrezia). Dino, who was involved in the control training, did not show any improvement with executive functions of Inhibition and/or Switching.

Table 5. Participants' Performance in the Listening Span Test Analyzed with Standard and Raw Scores

		Listening span test		
		Number of words correctly recalled in sequence	Number of errors in judging sentences plausibility	Number of intrusion errors
Ilaria	Initial assessment	-1.09 (raw score:11)	-0.7 (raw score: 2)	-0.12 (raw score: 1)
	After the baseline condition	-1.09 (raw score: 11)	-0.7 (raw score: 2)	-0.6 (raw score: 0)
	After the experimental training	1.08 (raw score: 20)	-0.01 (raw score: 1)	-0.12 (raw score: 1)
	Follow-up assessment	1.08 (raw score: 20)	-0.01 (raw score: 1)	-0.12 (raw score: 1)
Simone	Initial assessment	-2.22 (raw score: 5)	1.7 (raw score: 3)	4.62 (raw score: 10)
	After the experimental training	-0.34 (raw score: 14)	1.7 (raw score: 3)	1.03 (raw score: 3)
Roberta	Initial assessment	-1.39 (raw score: 9)	-0.05 (raw score: 1)	1.03 (raw score: 3)
	After the waiting list period	-2.01 (raw score: 6)	1.7 (raw score: 3)	3.6 (raw score: 8)
Lucrezia	Initial assessment	-1.77 (raw score: 15)	0.89 (raw score: 4)	3.14 (raw score: 5)
	After the experimental training	-0.59 (raw score: 22)	0.10 (raw score: 2)	1.63 (raw score: 3)
Dino	Initial assessment	-2.78 (raw score: 9)	0.10 (raw score: 2)	3.89 (raw score: 6)
	After the control training	-2.27 (raw score: 12)	-0.29 (raw score: 1)	7.65 (raw score: 11)

Interpreting the Findings concerning Verbal Working Memory

Assessing verbal working memory with backward digits span failed to show reliable training or test familiarity changes in the participants. Conversely, when verbal working memory was assessed with the listening span test robust training effects were observed. None of the three participants who could show either test familiarity (Ilaria after the baseline, Roberta after the waiting list period) or control training effects (Dino) showed a reliable or clinically significant improvement in the listening span test. On the contrary, each of the three children involved with our experimental training (Ilaria, Simone, and Lucrezia) showed a reliable and clinically significant change. In other words, each child started with a dysfunctional performance in the listening span test's initial assessment and then reached a score within the normal range in the post-training assessment (see Table 5). In the follow up assessment Ilaria maintained the improved performance with the listening span test.

There were two types of evidence suggesting that the participants' improvements involved working memory. First, the ability to recall more words in sequence in the listening span test did not occur at the cost of a less effective sentence processing. As shown in Table 5, in which raw and standard scores for performance in the listening span test are reported for each participant, there was no increase of errors in sentence judgements in the children who improved after the experimental training. An enhanced ability to control interference when recalling words was also observed (see a decrease of intrusion errors in Ilaria after the training, Simone, and Lucrezia). These findings suggest that after the experimental training children were more skilled in implementing the dual task of judging the semantic plausibility of the current sentence and keeping in memory the last word of the previous sentences.

A second type of evidence clarifying the underlying nature of children's improvement with the listening span test was the participants' stable ability to store a sequence of words or digits in short-term memory. As verbal short-term memory did not increase after the experimental training, as shown in Table 4 (only Simone showed a reliable change in the words span test), this suggests that the improved performance in the listening span test was not generated by an enhanced capacity of the short-term verbal store. In other words, children were not more skilled in memorizing words but more able to direct attentional resources towards the two parallel goals of judging the plausibility of each sentence and memorizing the sentence's last word.

Focusing on Dino, who was involved in a control training targeting both visuospatial working memory and verbal recalling with narrative memory tasks, it was observed that his performance with the listening span test did not improve. This suggests that simply practicing the recall of verbal information from long-term memory or being trained with dual tasks in a non-verbal domain may not be effective to enhance verbal working memory.

If participants' improved performance with the listening span test involves a strengthened working memory, how can we explain the lack of improvements in performance with the backward digits span? To answer such a question we should emphasize that words memorization may depend on *cumulative rehearsal* in the listening span test. The participant encodes in memory the last word in a sentence after having judged the semantic plausibility of such sentence; this process is repeated for the subsequent sentences and word traces in memory can be strengthen by a cumulative rehearsal (e.g., silently repeating the first and second memorized word before attending to the third sentence). In the backward digits span the participant has to first keep in memory a sequence of digits and then recalling them in a reverse order. The effectiveness of processing the digits' order reversal is a function of the sequence of items that is kept in the short-term memory store. As the short-term store has not been strengthened in our participants, manipulating digits in a reverse order did not undergo any improvement. Another factor explaining the different results of backward digits span and the listening span test may be that the participants' pre-test scores were much lower for the latter and that our training program could more easily improve lower scores.

"Far" Transfer Effects

Focusing on problem-solving, out of two children who could show test familiarity effects (Ilaria after the baseline, Roberta after the waiting list period), only Roberta made a reliable change in performance with the Raven's Matrices. Conversely, each of the three children involved with the experimental training made a reliable change in performance with the Raven's Matrices (but only two of them had a post-test score within the normal limits). Concept formation, on the other hand, showed a reliable change in Simone and a clinically significant improvement in Dino, who was involved with the control training.

Thus despite the fact that the experimental training stimulated working memory and inferential processes through activities in the verbal domain, each of the three children involved with the experimental training showed reliable changes in performance with

the Raven's Matrices. This finding suggests that our experimental training allowed participants to address a problem-solving task in the visual domain with higher attention control and better monitoring of ongoing procedures.

Test Familiarity versus Training Effects

We represented in Figure 2 the number of scores whose change in the post-test was "reliable". We focused on those 6 tests that allowed us to compute a *reliable change index* and identify near and far transfer training effects. If test familiarity was to explain a substantial part of participants' changes in the post-test we should expect such influence to underlie in a rather similar way each participant. However, the trend suggested by the findings illustrated by Figure 2 is different: out of the 15 reliably significant increments in the post-test, 11 are shown by the three children assessed after the experimental training and 4 by the three children assessed after either baseline, or a waiting list phase, or the control training. We ran the Fischer's Exact test on the data reported in Table 6 and found that differences were statistically significant (two-tailed p-value = .0234).

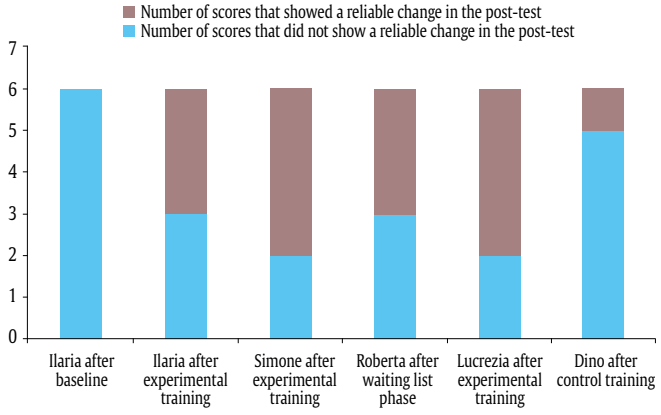

Figure 2. Number of Scores that Showed or did not Show a Reliable Change in the Post-test.

Table 6. Data for the Fisher's Exact Test

	Number of scores observed after the baseline, waiting phase and the control training	Number of scores observed after the experimental training
Reliable change in the post-test	4	11
Non-reliable change in the post-test	14	7

Conclusions

In this exploratory multiple case study, working memory was trained along with executive functions of inhibition and switching. The training method consisted of several different tasks which stimulated participants' control of attention, capacity of alternating different procedures, and ability of implementing complex processing (e.g., semantic categorization, pragmatic judgments, inferences) on memorized verbal information. The use of language as a "tool" to orient attention to task materials, focus and recall instructions, and anticipate sources of difficulty was systematically promoted in training. Children made practice of specific exercises and games and, at the same time, were guided to use strategies to implement tasks. Such type of complex training – in which variation largely prevailed on repetition and strategies co-occurred with practicing exercises – succeeded in significantly improving verbal working memory. Three children with mild to borderline intellectual disabilities who initially had an impaired performance with the listening span test, after eight weeks of training showed a performance within the normal limits in the same test.

We were interested in exploring whether training working memory could allow children to implement problem-solving in the visual domain in a more effective way even if this had not been directly stimulated by training. We did find that an enhancement of problem-solving with the Raven's Matrices was more likely after the experimental training than our control conditions. We were not interested in distinguishing whether children's improvement in problem-solving was related to a more strategic approach to the task (e.g., better visual scanning of all the items before selecting a solution), or a strenghtened capacity to construct visuo-spatial relations. For this reason we avoided the phrase "fluid intelligence" in reference to performance with the Raven's Matrices and preferred to interpret children's higher scores in this task as indicators of an increased capacity to coordinate cognitive processes to address a problem-solving task.

Thus, finding that the children observed in this study could substantially enhance their performance with the Raven's Matrices suggests that training working memory might be a preliminary step to enhance complex cognitive processes. A tentative conclusion of this study is that skills-based interventions involving coordination between task-relevant cognitive processes (Kearns & Fuchs, 2013) might be more effective if prepared by preliminary working memory training. Instruction focusing strategies for understanding or writing texts, solving mathematical problems, and building novel concepts is likely to be facilitated in children with mild intellectual disabilities if their verbal working memory has been enhanced.

Limitations

The conclusions of this study are based on a very small number of children and should obviously be contextualized taking into account the specific characteristics of participants. First, children involved in this study had a history of language difficulties that was not associated to a severe short-term memory deficit. Thus the conditions that limit verbal working memory malleability remain to be identified in future case studies. Second, participants were selected in this study because they were deemed to be motivated to participate at training and their engagement could also be supported by their parents' motivation.

Motivation, along with beliefs about the nature of intelligence, seems to affect the degree of transfer to reasoning skills in individuals involved in working memory training (Jaeggi, Buschkuehl, Shah, & Jonides, 2014). Thus an overt assessment of motivation and beliefs about learning and intelligence should be included in future research. A further limitation is that we assessed retention of training effects after a phase of training interruption only in one case (Ilaria). It remains to be explored whether enhanced verbal working memory and problem-solving are long-term training effects. Eventually, the training transfer effects could have been even larger than those found in our study if training had stimulated both verbal and visual working memory, as suggested by some meta-analysis studies (Danielsson et al., 2015; Schwaighofer, Fischer, & Bühner, 2015).

Conflict of Interest

The authors of this article declare no conflict of interest.

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