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DEPARTMENT OF PSYCHOLOGY AND COGNITIVE SCIENCE

**Implementing evidence-based treatments for developmental dyslexia:  
a comparison between different approaches**

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To J.R.R. Tolkien and his “The Lord of the Ring”,  
Where Everything Began.

*“It's a dangerous business [...] going out your door.  
You step onto the road, and if you don't keep your feet,  
there's no knowing where you might be swept off to.”*

## ABSTRACT

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Developmental Dyslexia (DD) is one of the most common neurodevelopmental disorders across cultures. Children affected by DD struggle to read fluently and/or correctly, despite normal intelligence, the absence of other psychological or neurological symptoms, and standard reading education. Not only does this condition affect academic achievement, but it is also associated with a number of other negative consequences across the lifespan, such as an enhanced risk of psychological distress and mental health problems.

Despite considerable efforts to identify the underlying cause of dyslexia, agreement on a single interpretation has not yet been reached. DD is commonly described as a language-related disorder, with compromised phonological abilities being considered as the core deficit. Nonetheless, a growing body of evidence supports multifactorial models: reading is a complex cognitive process, involving not only phonological skills, but also auditory sensory processes, visual-spatial abilities, attention and memory. In this regard, various studies have documented a relationship between dyslexia and deficits in the Executive Functions (hereafter EFs), which can be defined as a cluster of general-purpose control mechanisms that modulate various cognitive sub-processes. However, the relationship between cognitive correlates and reading impairments is still a controversial issue.

The present project aims to analyse the fundamental characteristics and the efficacy of different rehabilitation methods for Developmental Dyslexia. Although the neurocognitive causes of DD are still hotly debated, researchers agree that the main challenge is the remediation, that is, how to improve children's reading fluency and accuracy.

The most common approach has been to devise sophisticated remediation programs that train sub-skills of reading, especially phonological skills and auditory perception. Despite the promising results, the improvements in these sub-skills do not automatically transfer in better reading abilities in all subjects (especially regarding reading fluency), thus giving rise to the issue related to "non-responders" or "poor responders".

Since the present data gives firm indications of the need to individualize intervention based on neuropsychological testing, the aim of this project is to investigate the efficacy of new types of treatment based on a multifactorial, probabilistic, model of the disorder.

Consequently, this project consisted of two parts: specifically, in the first study, we compared phonological-based treatment with computerized cognitive training of the executive functions (e.g., attention, working memory, planning, inhibition). The results of this study clearly pointed out an advantage both in terms of improvements in EFs and literacy skills for the group who undertook the Integrated training, i.e., the group that underwent 12 hours of Cognitive training prior to 12 hours of Phonological-based treatment.

Next, the second study aimed to explore the efficacy of a video game *Skies of Manawak* purposefully designed to train several EFs. Indeed, it has been showed that the existing treatments are not sufficiently captivating and motivating and, thus, we developed a tool *ex novo* in order to obtain overall improvement and higher chances of transfer to untrained tasks. Our goal was to investigate whether playing this video game may enhance EFs following intervention, and whether these improvements transfer to important literacy skills in typically developing (Study 2 – Part A) and dyslexic children (Study 2 – Part B). All children underwent 12 hours of training, distributed over 6 weeks, either on Skies of Manawak or on a control computerized activity (*Scratch*). Assessments upon training completion indicated greater improvements in executive functioning and reading efficiency after Skies of Manawak than after the control training in both studies. Interestingly, the advantage in reading skills was maintained in a follow-up test 6 months later and seemed to generalize to academic performance (i.e., Italian marks).

Overall findings highlighted promising effects of the training programs on children's cognition, making way for future studies investigating the underlying brain mechanisms and the factors leading to treatment success.

# CHAPTER 1

---

## DEVELOPMENTAL DYSLEXIA

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*“In the fields of observation chance favours  
only the prepared mind”  
~ L. Pasteur*

### 1. Definition and diagnostic criteria

Although Dyslexia is one of the most common neurodevelopmental disorders across cultures and it has been extensively studied over the last 50 years (Snowling, 2012), to date there is no consensus about a single comprehensible definition of this disorder (Frith, 1999). Developmental Dyslexia (DD), contrary to Acquired Dyslexia, is not a result of brain damage and it refers to a specific impairment in the acquisition and the automatization of reading skills. This term is challenging to define due to the fact that dyslexia is not a single uniform condition, in which each individual shows similar characteristics both in terms of symptoms and degrees of severity. Moreover, although it leads to a failure in achieving typical reading milestones, it has been showed to be associated with other factors (i.e. working-memory, speed processing, etc.), which makes its definition multi-layered and hard to be identified (Pennington et al., 2012; Peterson & Pennington, 2012; Peterson, Pennington, & Olson, 2013).

The following list briefly summarises the widely varying definitions of dyslexia that have emerged in the literature.

Dyslexia is described by the British Dyslexia Association (2007) as:

“A specific learning difficulty that mainly affects the development of literacy and language related skills. It is likely to be present at birth and to be life-long in its effects. It is characterised by difficulties with phonological processing, rapid naming, working memory, processing speed, and the automatic development of skills that may not match up to an individual’s other cognitive abilities. It tends to be resistant to conventional teaching methods, but its effect can be mitigated by appropriately specific intervention, including the application of information technology and

supportive counseling.” (Source: <http://www.actiondyslexia.co.uk/view-article/defining-dyslexia>) (British Dyslexia Association, The Dyslexia Handbook 2002, p.67).

In addition, the International Dyslexia Association (Lyon, Shaywitz, & Shaywitz, 2003) defines dyslexia as:

“A specific learning disability that is neurological in origin. It is characterised by difficulties with accurate and/or fluent word recognition and by poor spelling and decoding abilities. These difficulties typically result from a deficit in the phonological component of language that is often unexpected in relation to other cognitive abilities and the provision of effective classroom instruction.” (p.2) (Source: <https://dyslexiaida.org/definition-of-dyslexia/>)

Furthermore, widely used manuals for psychological disorders have defined dyslexia: the International Classification of Disorders – ICD-10 (2010) and the Diagnostic and Statistic Manual of Mental Disorders – DSM-5 (2013). The ICD-10 states the following:

“The main feature [of the specific reading disorder] is a specific and significant impairment in the development of reading skills that is not solely accounted for by mental age, visual acuity problems, or inadequate schooling. Reading comprehension skill, reading word recognition, oral reading skill, and performance of tasks requiring reading may all be affected. Spelling difficulties are frequently associated with the specific reading disorder and often remain into adolescence even after some progress in reading has been made. Specific developmental disorders of reading are commonly preceded by a history of disorders in speech or language development. Associated emotional and behavioural disturbances are common during the school-age period.”

On the other hand, the DSM-5 classifies dyslexia as one of the forms in which the Specific Learning Disorder may occur. For this reason, dyslexia is considered “an alternative term used to refer to a pattern of learning difficulties characterised by problems with accurate or fluent word recognition, poor decoding, and poor spelling abilities”. To be diagnosed with a specific learning disorder with impairment in reading, an individual must show difficulties for at least six months despite the provision of targeted interventions (*Criterion A*). A second

core feature (*Criterion B*) is that individuals with this disorder struggle to read fluently and/or correctly and their performance is well below average for age norms: this causes significant impairment in daily activities, and it may compromise academic or occupational performance. This criterion requires psychometric evidence from a series of standardised tests and battery of reading achievement (where standardised scores usually trace a perfect Gaussian curve). Reading performance can, therefore, be conceptualised as a continuum between skilled readers and poor readers (FragaGonzález, Karipidis, & Tijms, 2018). From this perspective, dyslexia can be considered as the lower part of the tail in a normally distributed model (Fawcett, 2012, Fletcher, 2009, Shaywitz et al., 1992). The cut-off point between typical and atypical reading skills is somewhat arbitrary. In Italy, the Consensus Conference on Learning Disabilities (2010) takes into consideration the international criteria and carries them further towards the development of a dimensional approach to psychological diagnosis (Vio & Lo Presti, 2014). When the distribution is normal, a performance in reading speed lower than -2 standard deviations could be considered as statistically significant.

On the other hand, there is a significant impairment with a performance in reading accuracy lower than 5° percentile (where, usually, the distribution is rectangular or reverse J-shaped, rather than Gaussian). The authors, moreover, strongly encourage clinicians to consider standard errors or confidence intervals when thinking about the score obtained from the test. Besides, we may consider the impairment significant even if the reading performance stands at around the 10° percentile or -1.8 standard deviations, but with substantial impairment in everyday life – evaluated from different sources of information – extremely impaired. This shifting paradigm, from a pure categorical to a dimensional approach in the diagnosis of dyslexia, will have beneficial effects for the special education policies. Not only the children whose reading performance satisfies all criteria will receive targeted interventions, but also children who do not entirely meet these arbitrary criteria may be eligible for special help. In addition, it is possible to categorise children with DD into three levels according to the disorder severity. *Mild*: Some difficulties showed in one or two academic domains, however mild enough that the children may be able to compensate or to operate well when appropriate accommodations or support services are provided. For example, Italian third-grade children with mild dyslexia read at a reading rate higher or equal to 1 syllable per second (syll/sec) with adequate text comprehension. *Moderate*: Marked difficulties in one or more academic domains so that the individual is unlikely to become proficient in those skills without

specialised and intensive teaching during the school years. They are in need of some accommodation at school, at home or at work, at least for part of the day, to complete activities accurately and efficiently. E.g. a reading rate higher or equal to 1 syll/sec is reached only after fourth grade. Sufficient comprehension performance is possible only for short texts. *Severe*: Severe difficulties in learning skills affecting several academic domains, so that the children are unlikely to learn those skills without individualised and specialised teaching during most of the school years. Even with appropriate accommodations and/or services, the children might still not be able to efficiently complete activities. For instance, reading performances are usually lower than 1 syll/sec in third grade, around 1 syll/sec at the end of the fifth grade or at the beginning of the sixth. Text comprehension from autonomous reading is hardly reached (Zarei, 2010; Vio & Lo Presti, 2014).

To receive a diagnosis a crucial third feature is required (*Criterion C*): even though in most of the subjects learning difficulties appear in the early school years, they may fully manifest only when the learning demands have increased in complexity and quantity, and therefore exceed the individual's capacity. For these reasons, it is useful to have at least two assessments over time: especially in the cases with minor difficulties accompanied by a good cognitive functioning, the evolutive trend is characterised by a variety of expressions of the phenotype (Raskind, Peter, Richards, Eckert, & Berninger, 2013).

Additionally, the last core diagnostic feature (*Criterion D*) is that the specific impairment in reading acquisition and automatization is not solely accounted for by low intellectual functioning (usually estimated with an IQ lower or equal to 70), visual or auditory acuity problems, other mental or neurological problems, psychosocial adversity, lack of proficiency in the language of academic instruction or inadequate schooling. This means that in order to give a diagnosis, the clinicians need firstly to exclude the presence of intellectual disabilities or a more general developmental delay. It could also happen that individuals with an intellectual functioning above the range of normative intelligence ("gifted") are able to compensate the specific difficulties in reading until the learning demands become too high (e.g. reading lengthy documents with a short deadline) or the assessment procedure too pressing (e.g. timed tests). Secondly, the subjects show neither neurological (e.g., pediatric stroke, traumatic brain injury) and psychological disorders, nor vision, hearing and motor problems (e.g. untreated myopia). Thirdly, it is essential to rule out other external factors, such as economic disadvantage or a poor educational environment. In fact, reading, as well as

the other learning domains, requires explicit instruction and is not a natural consequence of brain maturation (M. J. Snowling & Hulme, 2011). Specific brain areas (particularly the visual word form area) and their connections need to be recruited and specialized from the recognition of written patterns: reading acquisition requests a remarkable amount of brain plasticity both at a cortical and subcortical level (Dehaene-Lambertz, Monzalvo, & Dehaene, 2018; Dehaene, 2014; Dehaene et al., 2010; Fraga Gonzalez et al., 2014; Kronschnabel, Brem, Maurer, & Brandeis, 2014).

Other non-core behavioural features may be used for a diagnosis due to the fact that they are frequently found in individuals with dyslexia: internalising and externalising problems (Dahle & Knivsberg, 2014), attention deficits (Facoetti et al., 2008; Facoetti & Molteni, 2001; Lobier, Zoubrinetsky, & Valdois, 2012; Menghini et al., 2010; Vidyasagar & Pammer, 2010) and more in general impairment in the executive-function system (Menghini et al., 2010; Moura, Simões, & Pereira, 2014; Smith-Spark, Henry, Messer, Edvardsdottir, & Ziecik, 2016; Varvara, Varuzza, Sorrentino, Vicari, & Menghini, 2014). Even if it is under debate whether these features are causes, correlates or consequences of this disorder, they may provide useful hints for the diagnosis and novel paths for effective interventions.

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## **2. Associated difficulties**

As previously mentioned, the issue of adequately defining dyslexia has been highly debated for several decades. Nevertheless, only the presence of difficulties in the acquisition and the automatization of the reading process is universally accepted. Moreover, increasing evidence pointed out the neurobiological origin of this disorder: the reading impairments are likely to depend on the interaction between several genetic and environmental factors. Dyslexia seems to share several risk factors with other developmental disorders, i.e. Attention-Deficit/Hyperactivity Disorder (Moura et al., 2017) or Autism Spectrum Disorder (Frith, 2013), therefore showing substantial symptoms overlap. It is even harder to get a clear picture of this topic if we consider that dyslexia often co-occurs with other difficulties or disorders and that its symptoms expression changes over time. For now, the exact nature and the level of severity of the associated difficulties are still under debate. However, a dyslexic subject often presents some of the following symptoms: reading difficulties that could be noticeable in a highly demanding learning process, difficult identification and production of rhyme-words or problematic phonemic segmentation and blending skills (phonological

awareness); difficulties in learning and automatizing phonemes; listening and comprehension.

Consequently, children that will be diagnosed with dyslexia generally show difficulties attending to the oral language patterns within words and to accurately and efficiently identifying and manipulating them. This ability is fundamental in learning to read (and spell) fluently and accurately, even if the exact link between phonological awareness and reading acquisition may differ across orthographies (Kovelman et al., 2012; Landerl et al., 2013; Ziegler & Goswami, 2005, Goswami & Bryant, 1990). Basically, phoneme blending is at the base of reading. As time goes by, difficulties in phonological processing regard higher and more complex functions: generally students with dyslexia struggle with blending several spoken sounds in order to create a recognisable word (e.g. “glyphs”), or vice versa in remembering how to spell orthographically complex words (e.g. “embarrassment”). In addition, older students tend to skip over function words when reading aloud and to re-read sentences and passages, especially in front of lengthy texts. Reading speed and accuracy are usually impacted by frequency, length and the stress pattern of the stimuli. Lower fluency and/or lower accuracy in reading are the natural consequences of the difficulties in automatizing the processes of language decoding. Despite the fact that in regular orthographies, such as Italian, reading errors are few and often ambiguous, and the primary diagnostic feature is generally the lack of fluency, the number of errors made by dyslexic individuals is significantly higher when compared to their typically reading counterpart (Trenta, Benassi, Di Filippo, Pontillo, & Zoccolotti, 2013). Notably, the most commonly observed errors are omissions, substitutions, distortions and additions of words or parts thereof; lexicalisation, false starts, long hesitations, inversion of letters within words or words within sentences, difficulty in keeping track of the individual is reading.

In dyslexic subjects, we often find difficulties in text comprehension – the ability to grasp the meaning, recovering and processing the mental representations of what has been read. It has to be noted that problematic comprehension is not directly due to impairments in the acquisition of meaning *stricto sensu* but to the extremely difficult decoding of the written text (Oakhill, Cain, & Bryant, 2003). Children with dyslexia are therefore to be distinguished from children who show a specific impairment in comprehension (APA, 2013). These children can read within the normative limits, they have adequate phonological capacities,

but they cannot understand the meaning of written text. In this specific case, the difficulties are attributable to a general problematic in language comprehension: difficulties in inferencing and figurative language as well as in text-related processes (Spencer, Quinn, & Wagner, 2014).

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Another significant aspect of dyslexia is that it occurs in children who are usually very smart, brilliant and have a general cognitive functioning in the norm. Besides, they are often able to compensate for their specific difficulties proving high-reasoning capacities as well as vivid imagination and other talents (Shaywitz et al., 2003).

Although dyslexia is commonly considered as a very specific disorder, in the last decades, researchers have also focused on finding other distinguishing features that go beyond reading impairment. Also, this links up with several other studies that have investigated comorbid disorders (see Table 1, for an overview). The comorbidity between dyslexia and other learning disorders is well established: dyscalculia (Peters, Bulthé, Daniels, Op de Beeck, & De Smedt, 2018; Wilson et al., 2015), dysorthographia (Margari et al., 2013) and, even if to a lesser extent, dysgraphia (Döhla & Heim, 2016; Nicolson & Fawcett, 2011). Other conditions that are commonly co-morbid with dyslexia include Attention-Deficit/Hyperactivity Disorder (Moura et al., 2017; Russell & Pavelka, 2013) and Autism Spectrum Disorder (Uta Frith, 2013; Russell & Pavelka, 2013). Also, children with dyslexia may be at higher risk of receiving a diagnosis of Disruptive, Impulse-Control and Conduct Disorders (Hendren et al., 2018; Loeber, Burke, Lahey, Winters, & Zera, 2000). Furthermore, there is a growing number of studies that demonstrate the link between reading difficulties, with consequent low academic achievement, and socio-emotional aspects. Specifically, higher incidences of internalising and externalising behaviours among students with dyslexia, in comparison with their typically-reading peers, were demonstrated (Dahle & Knivsberg, 2014; Eissa, 2010; Kempe, Gustafson, & Samuelsson, 2011; Livingston, Siegel & Ribary, 2018; Mammarella et al., 2016). Whilst the former category refers to a set of problems – characterized by overcontrolled symptoms – developed and maintained within the individual such as anxiety and depression, the latter concerns a grouping of problems that result from difficulties in regulating their behaviours and emotions such as aggressive conduct problems, hyperactivity, antisocial behaviour (Merell, 2008).

Even though individuals with dyslexia have a higher risk for emotional disorders – they usually show a higher level of anxiety, lower self-esteem and they are generally less-accepted and victims of bullying – it is still unclear whether this is purely a consequence of the reading difficulties or it precedes them. On the other hand, Kempe et al. (2011) and Hendren et al. (2018) reported that conduct and behavioural disorders appear to be independent of reading failure rather than a consequence of it.

**Table 1** | An overview of comorbid conditions that commonly occur with dyslexia. Adapted from Hendren et al. (2018).

Comorbid condition	Features of comorbid group	Shared risk with RD
Attention deficit hyperactivity disorder (ADHD)	Inattention (auditory and visual); deficits in processing speed, verbal working memory, short-term phonological memory, naming speed, and central executive processes	Shared risk genes ( <i>KIAA0319</i> and <i>DCDC2</i> ); Shared structural and functional neural abnormalities; Environmental factors (smoke and miscarriage)
Autism spectrum disorder	Impaired reading comprehension	Shared risk genes ( <i>MRPL19</i> ); Comorbidity with language impairment
Disruptive, impulse-control, and conduct disorders	Externalizing behaviour	Shared cognitive risk in working memory deficit; Comorbidity with ADHD; Deficits in verbal processing/language skills
Anxiety and depressive disorders	Poor self-esteem; Internalizing psychopathology	Negative academic/social experiences; Shared familial risk factors
Other specific learning disorders	Internalizing psychopathology; Handwriting deficits	Shared cognitive risk in working memory, semantic memory, and verbal processing deficits; Deficits in rhythmic organisation

In addition, it is important to mention that several researchers (Bogaerts, Szmalec, Hachmann, Page, & Duyck, 2015; Lukasova, Silva, & Macedo, 2016; Lum, Ullman, &

Conti-Ramsden, 2013) have suggested the presence of problems related to serial-order and procedural learning (e.g., learning the alphabet, tables and months of the year), spatial and temporal relations (right/left, yesterday/tomorrow), or fine oculomotor coordination (e.g., tie their shoelaces). Besides, other cognitive functions appear impaired in dyslexia – mainly working memory (Bacon, Parmentier, & Barr, 2013; Moura et al., 2017; Smith-Spark & Fisk, 2007; Smith-Spark et al., 2016), attention (Facoetti et al., 2005; Facoetti & Molteni, 2001; Lobier et al., 2012; Menghini et al., 2010; Trichur R. Vidyasagar & Pammer, 2010) or more executive-functions (Menghini et al., 2010; Moura et al., 2014; Smith-Spark et al., 2016; Varvara et al., 2014). One possible explanation for this high comorbidity rate is that various cognitive functions are highly interrelated and linked together. For this reason, it is challenging to disentangle reading impairments from other co-morbid conditions that might be causing them; in other words, the adequate development of abilities underlying specific cognitive functions requires the previous or full concurrent functionality of other cognitive abilities. For example, phonological awareness is fundamental not only for reading but also for writing; while symbols decoding ability plays an important role also in Math proficiency. Comprehension difficulties lie in the fact that there is a mutual influence between decoding and comprehension skills: good comprehension abilities facilitate the decoding of the text and, on the other hand, if there is not correct and accurate decoding there can not be an adequate text comprehension. Moreover, an impaired executive system – the hypothesized brain process that direct, regulate and control other cognitive functions including, at a minimum, memory, attention, flexibility, inhibition, planning and problem solving – could be considered either a core deficit for dyslexia or it could be a distinct problematic that is highly co-morbid with the reading disorder.

Although it is almost impossible to find two individuals with dyslexia with the same set of symptoms or levels of severity associated to each symptom, these additional behaviours may prove to be helpful indicators on which to base a proper intervention program. At the time of the diagnosis, the prognosis is good in most of the cases if the subsequent intervention is well timed and targeted, succeeding in involving both family and teachers. This is particularly important in order to guarantee the children's well-being, preventing them from the occurrence of psychological distress. Dyslexia, in fact, co-occurs with lack of confidence and low self-esteem, demoralisation, stress and anxiety, frustration. According to statistical reports (Korhonen, Linnanmaki, & Aunio, 2014; Schulte-Körne, 2016), a substantial number of children with dyslexia show difficulties in fitting in and a higher risk of school drop-out.

Besides, an effective intervention could prevent the exacerbation of academic and relational difficulties and their progression, during adulthood, in problematic work and social integration (De Beer, Engels, Heerkens, & Van Der Klink, 2014).

### **3. Prevalence and prognosis**

The prevalence of this disorder is estimated between 3 and 17% among school-age children. Despite the high variability incidence – variability that depends on the diagnostic criteria and the characteristics of the orthographic systems (Shaywitz et al., 1998) – dyslexia is the most frequent neurodevelopmental disorder. Besides, this disorder appears to be more common in males than females (with a ratio varying between 3:1 and 2:1). However, this data could be partly due to some gender issues regarding the way of signalling subjects with learning disabilities. Since male students often show more externalising behaviours than their female classmates, teachers usually report them more frequently. When the diagnostic assessment is more cautious and is based more on rigorous diagnostic criteria rather than on the reports of the school and/or family members, the balance between males and females is higher (Arnett et al., 2017; Hawke, Olson, Willcutt, & Wadsworth, 2010; Miles, 1970).

In Italy, the diagnosis is possible only at the end of the second grade when the formal teaching of reading and writing should be concluded. However, deficits in reading prerequisites, such as language deficit, poor phonological skills and visuospatial attentional problems, may be recognised at the end of the kindergarten or during the first year of primary school. If the child's IQ is high, the disorder may not be recognised at an early age, as the child manages to compensate for its deficits. Because of this difficulty in recognising symptoms, the diagnosis is often made only when the last years of elementary schools are reached, or later. In this complex picture, it is important to remind that many students are able to develop compensatory cognitive strategies that mask the disorder and allow them to reach adequate academic performances. As a result, many people find out to be dyslexic only in adulthood.

However, to get a better prognosis, it is vital to carry out a differential diagnosis in order to disentangle between the learning disorder and normal subjective variations in academic performance that can be due to socio-economic or cultural factors. Thereafter, it is equally important to plan and implement an effective intervention program, frequently evaluating whether or not objectives have been achieved.

#### **4. Dyslexia or Dyslexias**

A key issue – not only from a theoretical point of view but also for the rationale of effective interventions – is whether there are different forms of dyslexia or not. The expressivity of this disorder is vast, involving to varying degrees different components of the reading process. For this reason, it is important to remember the parallelism between types of developmental and acquired dyslexia; as Friedmann and Coltheart (2018) stated: “[...] both developmental and acquired types of reading disorder need to be interpreted in relation to a model of intact adult skilled reading” (p.2). In other words, each component of the reading system could potentially be impaired in a child who struggles to read fluently and/or correctly as well as in an adult after brain damage.

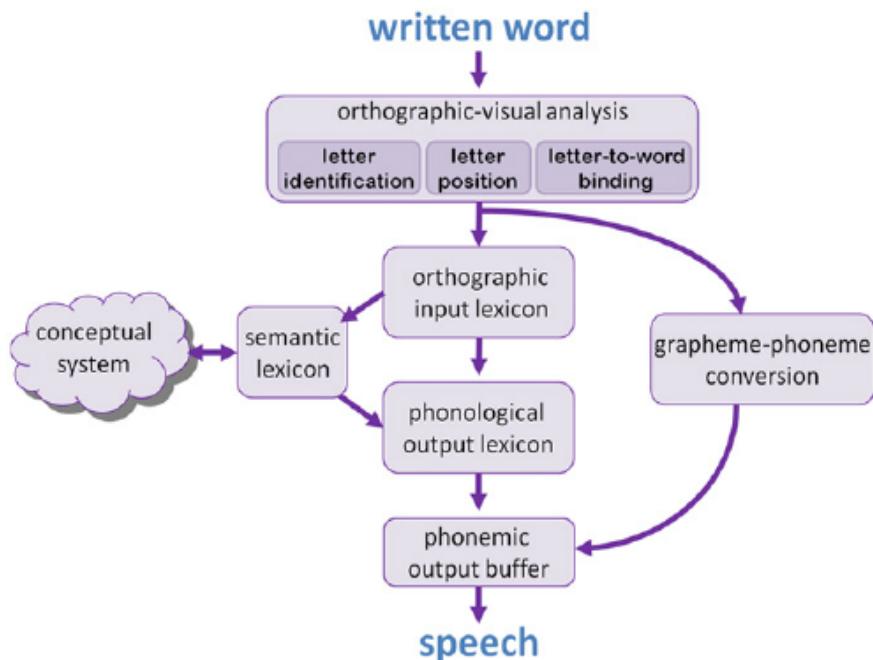
According to the dual route model for single word reading (Coltheart, 1981; Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001) outlined in Figure 1: at the beginning, the written stimuli are processed by the orthographic-visual analysis system, which is responsible for abstract letter identification, the encoding of relative letter positions within words, and binding of letters to words. After these initial processes of early analyses, the target word may be analysed via three routes:

1. the lexical route, from orthographic input lexicon to phonological output lexicon
2. the lexical-semantic route, from orthographic input lexicon to the semantic lexicon and the conceptual system
3. the non-lexical route, mechanism of conversion from graphemes to phonemes.

Considerably summarising this model, when the reader needs to read a regular, rare or novel word (e.g., a foreign word) the non-lexical or phonological route is activated, and the stimuli are read through a grapheme-to-phoneme conversion mechanism. Vice versa with words that are frequent and well-known or that have an exceptional pronunciation, the reading process is characterised by the use of the lexical or the lexical-semantic route – in the latter case recovering first the meaning of the stimuli and then retrieving its pronunciation from the lexicon.

In this regard, the types of dyslexia described in the literature are numerous, as different are the components of the reading system (Friedmann & Coltheart, 2018).

**Figure 1** | The dual route model. Source: drawn from Kohnen, Nickels, Castles, Friedmann, & McArthur, 2012.



#### 4.1. Letter position dyslexia

(Friedmann & Rahamim, 2007; Kohnen et al., 2012)

Letter position dyslexia constitutes peripheral dyslexia characterised by a selective impairment in the order of letters within words. For example, a reader with this type of dyslexia can make a migration error and confuse "form" with "from" and vice versa. Besides, the probability of making these kinds of errors is affected by the frequency of the target in comparison with the frequency of the other word: i.e. "diary" is more frequent and has, therefore, a higher probability to be read than "dairy". Other factors, such as lexicality of the error response, play a role in determining a different grade of susceptibility to within-word migration.

#### 4.2. Attentional dyslexia

(Friedmann & Haddad-Hanna, 2012) or "**Letter-to-word-binding dyslexia**" (Shallice & Warrington, 1980)

Attentional dyslexia involves a different kind of migration: letters migrate from one word to another that is closed in space, but they are adequately identified and preserve their correct

relative position within the word. This type of dyslexia seems to be due to an impaired capacity of properly focus on the target word ignoring the other irrelevant stimuli that are on the left/right or above/below the target word. As in letter position dyslexia, several factors can modulate the susceptibility to between-word migration: lexicality (these migration errors occur mainly when the migration results in an existing word), length (higher probability of migration errors with lengthy words) and similarity (words that differ in more letters are more susceptible to migration than words that differ in only one letter). Furthermore, it was demonstrated that most errors occur with final letters of the word and that it is easier to find a migration from the first to the second word.

#### ***4.3. Letter identity dyslexia or letter-identification-visual-dyslexia***

(Brunsdon, Coltheart, & Nickels, 2006)

Letter identity dyslexia is characterised by a deficit in the orthographic-visual analysis system, namely in letter identity encoding. The behavioural consequences of this type of dyslexia are generally omissions and/or substitutions of letters within words or incorrect identification of isolated letters. This clinical condition seems to differ from the associative visual letter agnosia, in which letter identification and recognition are impaired only when assessed from the visual modality and not from tactile and kinesthetic modalities.

#### ***4.4. Neglect dyslexia or Neglexia***

(Reznick & Friedmann, 2015; Friedmann & Haddad-Hanna, 2012; Nachman-Katz & Friedmann, 2010)

Neglect dyslexia consists of a selective impairment in focusing attention on a specific side of the word (typically the left). “Neglexia” has an adverse impact on word recognition (e.g. deficits in lexical decision tasks with written word) and it results in omissions and – more often – in substitutions of the neglected part that is “guessed” by the reader.

#### ***4.5. Visual dyslexia or “orthographic input buffer dyslexia”***

(Marshall & Newcombe, 1973)

This kind of dyslexia affects all the subcomponents of the orthographic-visual analysis system: letter position within the word, abstract letter identification, and binding of letters to

words. Therefore, it results in a mixture of errors characteristic of all the other peripheral dyslexia (letter position, visual, letter identity, neglect dyslexia): omissions, substitution, migration of letters within and between words, etc. To date, it is under debate whether visual dyslexia constitutes a separate disorder from the graphemic buffer dyslexia in which errors – generally not in letter identification – are more influenced by morphological features of the target.

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Whilst all the dyslexias presented earlier are peripheral, with reading difficulties that result from impairments at the level of the orthographic-visual analysis system, the following dyslexias are central, with deficits at one or more stages of the two main reading routes.

#### ***4.6. Phonological dyslexia***

(Dotan & Friedmann, 2015; Khentov-Krauss & Friedmann, 2018; Marshall & Newcombe, 1973; Valdois et al., 2003; Winskel, 2011)

A deficit in the sublexical route characterises phonological dyslexia. As a consequence, individuals with this kind of dyslexia can read all the words that are settled in the lexicon, but struggle to read non-words and new words. The sublexical route can be impaired at several levels of its stages: an impairment at the grapheme-to-phoneme conversion mechanism gives rise to the “letter-to-phoneme conversion phonological dyslexia” and it causes errors in reading isolated letters as well as non-words; difficulties in the parsing of letters typically characterise an impairment at the level of multi-letter knowledge into multi-letter graphemes (e.g. “sth”, “ch”, etc.); instead, an impairment in the phonological output buffer is at the basis of difficulties in the repetition of long non-words. Furthermore, vowel dyslexia is a selective impairment in the sublexical processing of vowels: omissions, transpositions, substitutions and additions of vowels are the typical markers of this dyslexia.

#### ***4.7. Surface dyslexia***

(Gvion & Friedmann, 2016; Job, Sartori, Masterson & Coltheart, 1984; Judica, De Luca, Spinelli, & Zoccolotti, 2002; Orsolini, Fanari, Cerracchio, & Famiglietti, 2009)

Surface dyslexia may be a consequence of several impairments in the lexical route of reading. This means that individuals who suffer from this condition are forced to read via the

grapheme-to-phoneme conversion mechanism. This is particularly evident in languages with opaque orthographies, such as English, in which reading of irregular words (e.g. “blood”) or words that contain ambi-phonics graphemes is impaired. Vice versa, in Italian it is less easy to recognise this form of dyslexia due to the high correspondence between orthography and phonology, but it is still possible, for example, in the case of words in which stress is not marked orthographically, but is lexically settled. This pattern of problematic reading may emerge as a consequence of impairments in several stages of the lexical route: the orthographic input lexicon or the access to it; the output of the orthographic input lexicon; the output to the phonological lexicon with preserved access to the semantic system. In addition, surface dyslexia can be part of a more general impairment (in most of the cases at the level of semantic) if it is caused by deficits in the semantic system, the phonological output lexicon, or its access to the phonemic output buffer.

#### ***4.8. Deep dyslexia***

(Johnston, 1983; Marelli, Aggujaro, Molteni, & Luzzatti, 2012; Temple, 2003, 2006)

Deep dyslexia occurs in the inauspicious situation of co-occurrent deficits of the two routes and this force the individual to read via meaning (especially using visual images of the words). Individuals with this kind of dyslexia produce several semantic errors (e.g. they can read “dog” instead of “cat” and vice versa) as well as morphological ones (e.g. “potato” instead of “potatoes”). Besides, non-word reading is also severely impaired.

#### ***4.9. Direct dyslexia or Access to semantics dyslexia or Hyperlexia***

(Castles, Crichton, & Prior, 2010; Zuccolotti & Friedmann, 2010)

Direct dyslexia results from disabled access to the semantic system while the lexical route between the orthographic input lexicon and the phonological output lexicon is spared. This means that individuals who suffer from this condition can read both word and non-word adequately without, however, being able to understand them.

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Despite the low – or very low – frequencies in which all these different types of dyslexia occur, the neuropsychological classification reported above has important implications. From

a clinical point of view, their relative rarities do not justify professionals using exclusively word and non-word reading tasks. In order to detect all kind of dyslexia, it is important to use adequate stimuli that are, therefore, sensible to the different form of reading impairment (including the peripheral ones). Besides, the correct identification of the type/s of dyslexia each child suffers from is fundamental to plan a tailored intervention.

The neuropsychological model of dyslexia, as well as the clinical (Boder, 1973), developmental (Frith, 1985; Seymour, 1986) and neuro-psychophysiological (Bakker, 1979, 1992) models – not described in this chapter, perfectly convey the multi-faceted nature of dyslexia. The extreme variability could be defined as the real “core deficit” of the disorder: each individual with dyslexia can be described by several symptoms; likewise, dyslexia presents a varied range of associated disorders and a multifactorial aetiology.

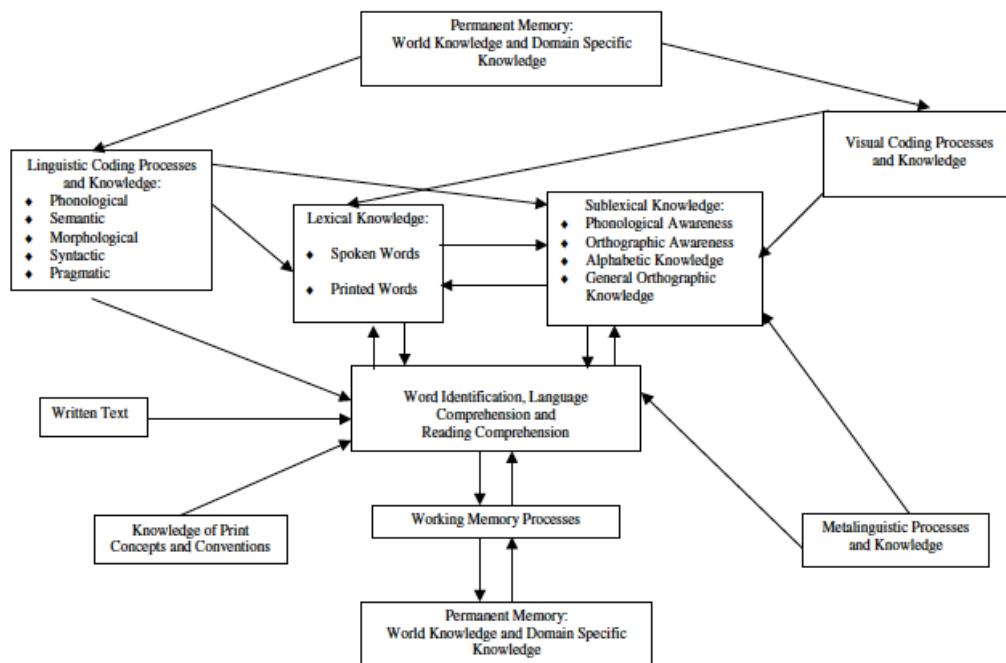
Knowing the various classifications and interpretations proposed over the years is fundamental both from a theoretical and a clinical point of view, but it is essential to approach dyslexia in a somewhat atheoretical way: individual performance must be analysed in detail, but without trying to incarcerate it in a specific clinical subtype. Several professionals have highlighted the importance of considering the individuality and uniqueness of each dyslexic. This discourse is particularly relevant if we consider all the studies conducted on the dyslexic population and aimed at demonstrating the presence of particular deficits associated with reading failure.

## **5. Aetiology: a multifactorial model**

Even though considerable effort has been expended trying to disentangle the proximal cause of this disorder, no single answer has been agreed upon. In this regard, since the end of the 1970s, the dominant approach states that dyslexia is caused by a deficit in phonological awareness (Gabrieli, 2009; Giraud & Ramus, 2013; Goswami, 2015; Peterson, Pennington, & Olson, 2013; Ramus et al., 2003; Vellutino, Fletcher, Snowling, & Scanlon, 2004; Ziegler & Goswami, 2005). According to this claim, dyslexia arises from an impaired capacity in representing, manipulating and retrieving sounds, as well as matching the letters to the word sounds (segmented in distinct phonemes). This hypothesis is supported by several neuroimaging studies that showed a link between poor input tuning into the brain regions specialized for grapheme-phoneme integration (Achal, Hoeft, & Bray, 2016; Blau, van Atteveldt, Ekkebus, Goebel, & Blomert, 2009; Clark et al., 2014; Dehaene et al., 2010; Dehaene, 2014; Vandermosten et al., 2012; J. Zhao, Thiebaut de Schotten, Altarelli, Dubois,

& Ramus, 2016). In addition to the phonological impairment, Rapid Automatized Naming (RAN) deficits are considered central in the aetiology of this disorder (Norton et al., 2014; Norton & Wolf, 2012; Wolf & Bowers, 1999). Rapid automatized naming – that reflects the efficiency of the visual-to-phonology mapping – is the speed requested to denominate out loud visually-presented stimuli whose nature can be linguistic (i.e. numbers and letters) or not (i.e. colours or familiar objects). In this view, RAN skills contribute uniquely to the aetiology of dyslexia and, therefore, it is possible to find a group of individuals with dyslexia that show an impairment in naming speed in the absence of phonological problems. Moreover, individuals that have both deficits show more severe reading problems in comparison with dyslexics with impairment only in one domain. Numerous longitudinal studies supported this theory showing that performance in this task are predictive of future reading skills (Leppanen, Aunola, Niemi & Nurmi, 2008; Lervåg, Bråten & Hulme, 2009; Lervåg & Hulme, 2009). In addition, several large-scale longitudinal studies of children at familial risk of dyslexia (Hulme, Nash, Gooch, Lervåg, & Snowling, 2015; Pennington & Lefly, 2001; Snowling et al., 2003; Snowling, Lervåg, Nash, & Hulme, 2019; Thompson et al., 2015; van Viersen et al., 2018) investigated the risk factors for later difficulties, highlighting an important predictive role of letter knowledge, phonological awareness, and RAN.

**Figure 2** | A schematic representation of the cognitive processes and the different types of knowledge required for learning to read. Source: drawn from Vellutino et al., 2004.



However, this theory was sharply criticised because the shreds of evidence in the literature do not adequately support the existence of a second independent core naming deficit (e.g. Vaessen, Gerretsen, & Blomert, 2009).

As stated in the previous paragraph, the phonological hypothesis has been the most prominent theory of dyslexia for decades. Despite this, to date there is no detailed and unique specification of the phonological deficit/s underlying this disorder. This expression, in fact, was used to pinpoint a broad range of skills related to phonological awareness (Ramus et al., 2003; Ramus & Ahissar, 2012; Ramus & Szenkovits, 2008; Ziegler & Goswami, 2005): from deficits at the lowest level (perception, manipulation and production of phonemes) through to higher ones (i.e. syllables, rhymes, spoonerism ability). Another group of researchers focused on the putative causal role of impaired access to phonological representations (Norton & Wolf, 2012). Lastly, phonological short-term memory deficits have been considered central in reading failure (Banfi et al., 2018; Majerus & Cowan, 2016). Besides, in the literature phonological awareness is not considered an efficacious risk predictor for developing dyslexia unanimously (Blomert & Willems, 2010; Helland & Morken, 2016; Peterson, Pennington, & Shriberg, 2009; Snowling, Bishop, & Stothard, 2000). In this regard, Ramus and Ahissar (2012) wrote: “Whether these components are independent or reflect a common underlying deficit remains an open question” (p.112). Additionally, contrasting evidence was collected on other tasks in which dyslexic are generally expected to perform worst by phonological hypothesis supporters: i.e. prosody perception, phonological grammar and perception/production of foreign sounds (i.e. Mundy, Carroll, Mundy, & Carroll, 2012)

Finally, as Dehaene stated in his famous book “Reading in the Brain” (Dehaene, 2009), citing the work of Ramus and colleagues (Ramus et al., 2003): “a minority of dyslexic children presents with a pronounced visual deficit and no phonological impairment” (p. 242). Namely, not all the individuals with dyslexia demonstrate a specific impairment in phonological awareness (see, for example, the case of developmental surface dyslexia; Judica et al., 2002; Shany & Share, 2011) and therefore should not be considered a necessary condition for dyslexia (Friedmann & Rahamim, 2007; Peterson & Pennington, 2012).

Furthermore, although it is clear that the majority of the dyslexics show difficulties in at least one of the tasks that assess phonological processing, it is still unclear whether phonological

deficits could be considered a consequence or a cause of dyslexia, mainly as a result of the circular relation existing between reading skills and phonological abilities (Dehaene et al., 2010). This author, in fact, highlights that:

“Phonemic awareness is an area where it is very difficult to separate causes from consequences. Do children partly become poor readers because they have trouble processing phonemes? Or is their phonemic awareness impaired because they have not yet learned to read? A vicious circle seems to close in on dyslexic children (p. 240).”

Several studies demonstrated not only the link between reading improvements and performance in phonological awareness tasks (de Gelder, Vroomen, & Bertelson, 1993; Morais, Cary, Alegria, & Bertelson, 1979), but also that the way these tasks are performed is influenced by the acquisition and automatization of the reading process (e.g., Ehri & Wilce, 1980). Finally, another problematic issue is related to phonological deficit as a single causal factor in reading failure: dyslexia co-occurs with a variety of other cognitive deficits and clinical conditions (Frith, 2013; Menghini et al., 2010; Moura et al., 2017; Moura, Simões, & Pereira, 2015; Willcutt, Doyle, Nigg, Faraone, & Pennington, 2005).

For these reasons, we are witnessing a change of perspective: several researchers stood back from a single-deficit model and started to look for other factors that can explain themselves the reading disability (Pennington et al., 2012). In general, researchers are looking for lower-level deficits that – together with impairments in phonological processing – may explain the aetiology of this complex disorder (Goswami, Power, Lallier, & Facoetti, 2015). In this regard, Ramus and Ahissar reviewed the existing literature on phonology, audition, vision and learning abilities in dyslexia (Ramus & Ahissar, 2012) finding – at least- 12 compelling theories. To our knowledge, no study was able to successfully identify and define all the different types of dyslexia and their underlying cognitive (and biological) deficits.

The aim of the following review [which, for the reasons mentioned above, is far from being exhaustive,] is to present the main hypotheses that are revitalising and, if possible complicating even further, this complex picture.

### **5.1. Learning**

Ahissar and colleagues (Ahissar, 2007; Banai & Ahissar, 2012; Oganian & Ahissar, 2012) proposed the “anchoring hypothesis”. According to this claim, individuals with dyslexia display deficit in a specific aspect of fast learning, precisely “anchoring” in auditory modality; specifically, they have difficulties in detecting sound regularities (e.g., transitional probabilities, long-distance relationships) with short windows and therefore fail to benefit from stimulus-specific repetitions. Whether such impairment leads to poor long-term representations of these regularities and to subsequent poor phonological representations is under debate, and it will require future researches. Anyway, the ability to form perceptual anchoring can be seen as a specific case of statistical learning, namely the capacity of extracting regularities that characterised a determined concept in order to predict future events.

Several studies showed a link between reading fluency and accuracy with performance levels on non-linguistic statistical learning tasks both in children (Apfelbaum, Hazeltine, & McMurray, 2013; Arciuli & Simpson, 2012; Vandermosten et al., 2012; Vandermosten, Wouters, Ghesquière, & Golestani, 2018) and in adults (Arciuli & Simpson, 2012; Frost, Siegelman, Narkiss, & Afek, 2013). Implicitly learned regularities might underlie reading and spelling acquisition by enabling detection of statistical regularities between letters (transitional probabilities between letters) and the probabilistic correspondences between phonemes and corresponding letters. Besides, some studies highlighted a link between statistical learning performances and vocabulary growth, which, in turn, may boost reading skills (Arciuli & Simpson, 2012). Moreover, Forkstam and colleagues support this claim showing that differences in brain systems associated with dyslexia partially overlap with the neural circuits involved in statistical learning (Forkstam, Hagoort, Fernandez, Ingvar, & Petersson, 2006; Petersson, Forkstam, & Ingvar, 2004). On the other hand, although numerous studies have investigated the presence of specific impairment of this domain in dyslexics (Gabay, Schiff & Vakil, 2012; Sigurdardottir et al., 2017; Vandermosten et al., 2018), the question whether statistical learning constitutes a core deficit of dyslexia remains controversial.

A third hypothesis claims that procedural learning deficits are factors causally related with dyslexia (Alvarez & Fiez, 2018; Bennett, Romano, Howard, & Howard, 2008; Folia et al., 2008; Nicolson & Fawcett, 1990; Nicolson & Fawcett, 2011; Pavlidou & Williams, 2010;

Peter, Lancaster, Vose, Middleton, & Stoel-Gammon, 2018). According to these authors, individuals with dyslexia may have difficulties in tasks whose performance is influenced by cerebellar functions and therefore are linked with the implicit learning of sequential stimuli. In fact, the cerebellum plays a fundamental role in the automation of the tasks that will be automated over time, such as cycling, driving, writing and reading. A deficit in the ability to make automatic actions would be detrimental for the acquisition of any rules, and it may cause severe difficulties in the case of reading, specifically in the grapheme-phoneme associations. Therefore, dyslexia may be due to a difficulty in acquiring and automating reading. The main supporters of this theory – Nicolson and Fawcett (2007) – proposed that at the basis of reading failure there might be an impairment in a brain system constituted by prefrontal language areas, basal ganglia and cerebellum. Few neuroimaging studies have provided evidence so far of the link between abnormalities in the cerebellum and differences in reading ability (Fernandez et al., 2015; Menghini et al., 2006; Stanberry et al., 2006). Besides, Pollack and collaborators (2015) and Stoodley & Stein (2011, 2013) failed to find any group differences in their meta-analytic studies of dyslexics versus typically reading brains.

Moreover, not all the behavioural symptoms of cerebellar impairments were found in all individuals with dyslexia (Ramus, Pidgeon, & Frith, 2003; Stoodley & Stein, 2013). Therefore, even the principal authors (Nicolson & Fawcett, 2011) consider, nowadays, the impaired cerebellar function as a secondary factor in comparison with more central deficits. Therefore it can be represented as “a more fundamental neurodevelopmental abnormality” that may lead to differences in the reading network (Stoodley & Stein, 2011).

## ***5.2. Auditory processing***

Some studies support the hypothesis of a deficit in rapid – or temporal – auditory processing (Tallal, 1980, 2004). According to this author, the phonological deficit that can be found in individuals with dyslexia is due to a difficulty in efficiently processing rapidly varying sounds. In other words, they struggle to discriminate auditory stimuli that have a short duration and are produced in rapid succession. This impairment in perceiving sounds within words would affect an accurate categorisation and recognition of the phoneme when its acoustic characteristics change (phonemic constancy). At the origin of this disturbance, there is a deficit at the basic auditory level, which, consequently, might entail an impaired temporal analysis of the phonemes, which would in turn prevent an efficient phonological decoding.

The correct mapping between letters and phonemes is, in fact, fundamental to the development of typical reading skills. Although the results of several studies supported this hypothesis (Banasich & Tallal, 2002; Fostick, Bar-El, & Ram-Tsur, 2012; Fostick & Revah, 2018; Habib, 2000; Martino, Espesser, Rey, & Habib, 2001; Rey, De Martino, Espesser, & Habib, 2002), another group of researches failed to find group differences between individuals with dyslexia and typically reading peers as well as a direct link between this deficit and phonological impairment (Ahissar, Protopapas, Reid, & Merzenich, 2000; Amitay, Ahissar, & Nelken, 2002; Breier, Fletcher, Foorman, & Gray, 2003; Bretherton & Holmes, 2003; Ramus et al., 2003). Tallal and collaborators (Tallal et al., 1996) tried to modify the wrong phonological traces in children with dyslexia, using a specific computerised training called Fast For Word (FFW): in this software it is possible to manipulate sound durations, lengthening them to make them more easily discriminable and acquirable. Other researchers used FFW with promising results (e.g. Gillam et al., 2008; Temple et al., 2003). However, these results are still unclear – especially when compared to those obtained by other research groups that perform different treatments – and the improvements in these tasks do not necessarily generalize to gains in reading performance (Cohen, Hare, Boyle, & Mccartney, 2015; Galuschka, Ise, Krick, & Schulte-Körne, 2014; Given, Wasserman, Chari, Beattie, & Eden, 2008; Marr, 2013; Strong, Torgerson, Torgerson, & Hulme, 2011).

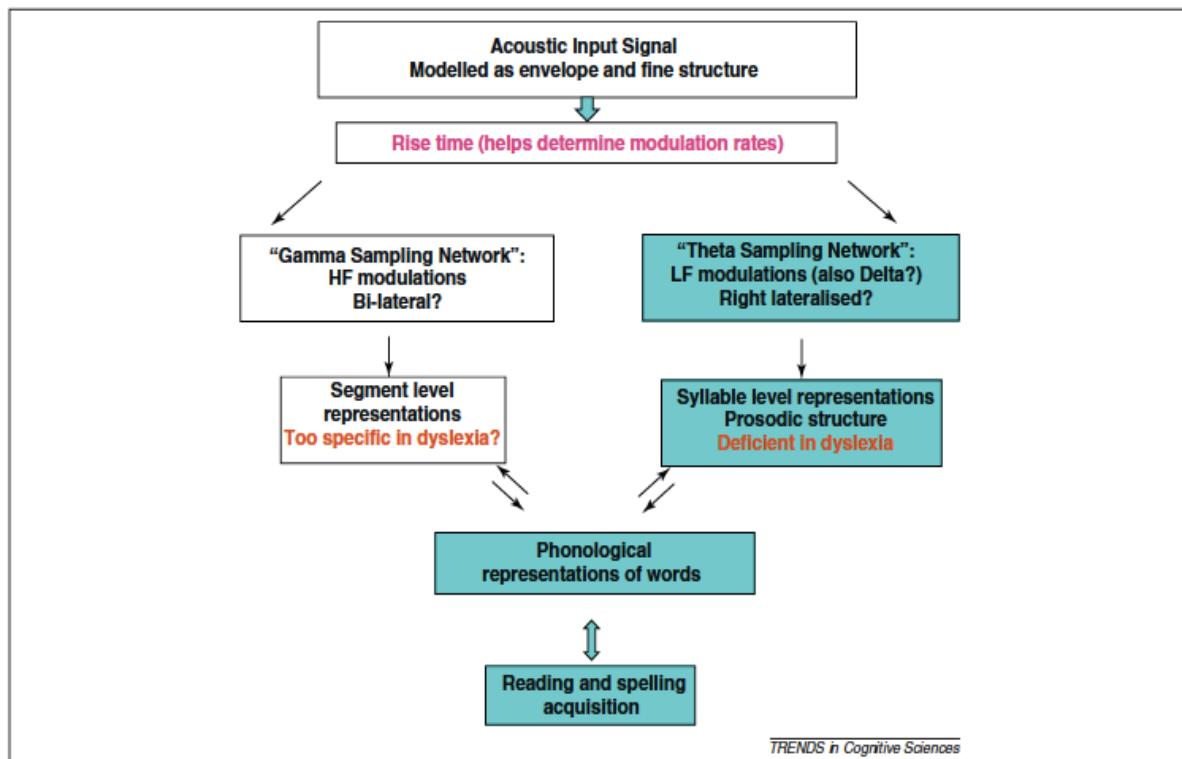
More recently, the rapid auditory processing deficit was integrated into a newer theory, the temporal sampling framework, proposed by Usha Goswami (Goswami, 2011; Goswami & Leong, 2013) and illustrated in Figure 3. Indeed, it has been suggested that the core deficit of dyslexia is an impairment in temporal sampling, namely in the “accurate perception of amplitude ‘rise time’” by the auditory cortex operating at different time scales or oscillatory frequencies (Goswami, 2011).

This theory also originates from the work of Poeppel and colleagues (Giraud & Poeppel, 2012; Poeppel, 2003, 2014; Poeppel, Idsardi, & Van Wassenhove, 2008), who suggested a role of amplitude “rise times” in enabling phase alignment. Rise times can be defined as the time interval between the first threshold crossing and the signal peak and, therefore, they denote the energy increases in the signal. Neural oscillations enable the creation of time-windows that chunk information; the phase within a cycle may serve as a reference frame for

endogenous oscillatory activity. Phase-resetting, in fact, not only allows mutually coupled oscillators to match the respective frequencies, but is also useful in aligning the phase of the oscillation to a specific reference point. According to this claim, all people with dyslexia, even if they speak different languages, display deficits in “rise time” discrimination.

**Figure 3 |** The temporal sampling framework (TSF). Deficits are highlighted in light-blue.

Source: Goswami, 2011.



The TSF assumes that individuals (both children and adults) with dyslexia display difficulties in syllabic perception with low temporal modulations (4-10 Hz range of delta-theta frequencies) which affect the detection of speech rhythm and prosody. Specifically, this theory suggests the presence of atypical delta [ $\sim 1\text{--}3$  Hz; stressed syllable rate] and theta [ $\sim 4\text{--}8$  Hz; link with syllable rate] oscillators in the right hemisphere, but also gamma [ $\sim 30\text{--}50$  Hz; correlation with attributes at the phonemic scale, i.e. formant transitions (e.g., /fa/ versus /va/)]. For example, slower rates around 2 Hz and 5 Hz are fundamental to the perception of the rhythm and its synchronisation.

In this regard, Goswami (2015) wrote: “recent theories of speech processing based on cortical oscillations identify an oscillatory hierarchy that approximately parallels the phonological hierarchy summarised here [in Figure 4]”.

Several studies supported the TSF, showing atypical functioning in the temporal integration window for syllabic parsing (Casini, Pech-Georgel, & Ziegler, 2018; Di Liberto et al., 2018; Usha Goswami, Huss, Mead, Fosker, & Verney, 2013; Power, Colling, Mead, Barnes, & Goswami, 2016; Stefanics et al., 2011). However, researchers are suggesting impairments at different auditory frequencies: e.g. Giraud & Poeppel (2012) for low frequencies (around 20 Hz) and Goswami, Gerson, et a. (2010) for higher frequencies (around 25-30 Hz). It could be possible that the nature of these impairments varies in different subgroups of people with dyslexia. Ramus and Ahissar (2012) stated: “a relative selective deficit in fast serial identification may be more characteristic of individuals who are high academic achievers and tend to be overall slow yet accurate, both in their auditory identification and in their pattern of reading difficulties”.

**Figure 4|** Phonological levels and correspondent oscillatory frequencies. Stressed syllable, syllable and phonetic rate can be considered the most relevant temporal rates from a linguistic point of view. Source: Goswami, 2015

Phonological level	Oscillatory frequency (EEG band)	Example(s)	Age at which reflective awareness develops
Intonational phrase	~1 Hz and lower	Who's a pretty boy then?	Not yet ascertained
Stressed syllable	~2 Hz (delta)	PE-ter PI-per PICKED a PECK of PICK-led PEPP-ers	Not yet ascertained
Syllable	~5 Hz ( $\theta$ )	* an-i-mal * wig-wam	2–3 years
Onset-rime	~cued by rising $\theta$ -slope	* c-at * str-eam * cl-amp	3–4 years
Phoneme	~35 Hz ( $\gamma$ )	c-l-a-m-p	With alphabetic tuition

However, a new, broader, approach has been proposed by Gori and colleagues (Gori, Cecchini, Molteni, & Facoetti, 2014; Gori & Facoetti, 2015). According to these authors, it is possible to extend these impairments in temporal processing – generally investigated only in the auditory modality – to the visual domain. In fact, efficient sequencing capacities of sound and letters within a word seem to be due to the accurate timing of auditory and visual stimuli. These capacities, in turn, rely on an efficient magnocellular or “transient” system.

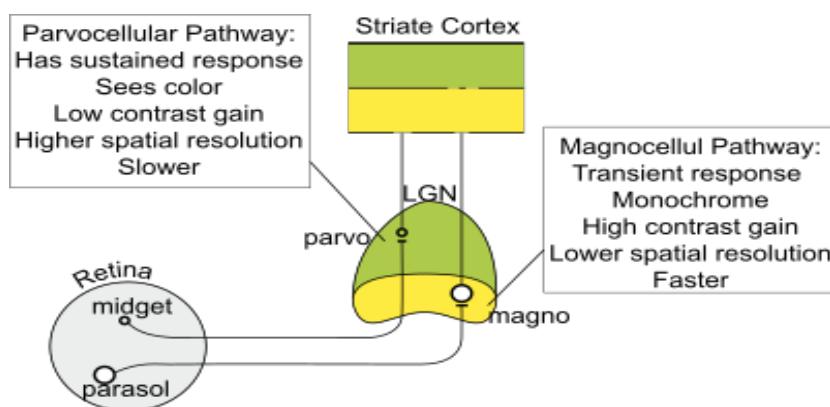
### 5.3. Visual processing

According to the first formulation of the theory (Stein & Walsh, 1997), at the basis of dyslexia there is a deficit in the magnocellular system, which is responsible for fast

processing changes in the visual modality. The properties of this system, in fact, have been extensively investigated within the visual system: it originates from the retina, specifically from the 10% per cent of the ganglions that are (up to 50 times) bigger than their counterpart, the parvo-cells (see Figure 5). Several studies pointed out that these ganglions are colour-blind and show a high response specificity to low spatial frequencies, high temporal frequencies and motion; vice versa the parvo-cells are sensitive to colour changes and respond to high spatial and low temporal frequencies (Callaway, 2005; Cheng, Eysel, & Vidyasagar, 2004; Livingstone & Hubel, 1987; Yazdanbakhsh & Gori, 2011).

**Figure 5 |** Visual representation of the parvocellular and magnocellular pathways.

Source: drawn from Purves (2013)



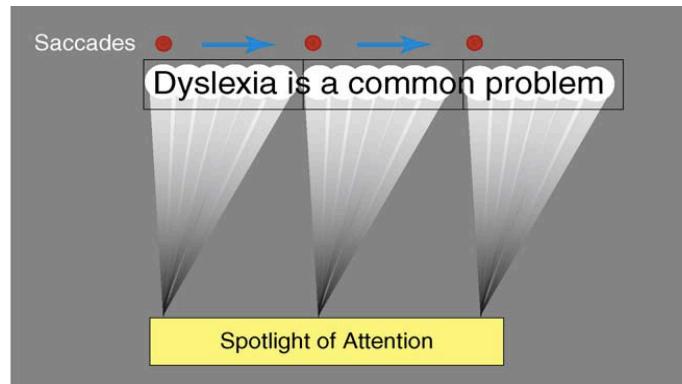
From the retina, these ganglions pass through the first and the second layers of the lateral geniculate nucleus (M-layer), project to the primary visual areas (V1-V2-V3) and also reach the parietal cortices. The intraparietal sulcus and the posterior parietal cortex, involved, among other things, in the integration of multimodal information for constructing a spatial representation of the external world, receive projections from the Magno-cells. The M pathway is also crucial for performing visual search tasks and detecting edges (Cheng et al., 2004). Since the magnocellular system ends mainly in the posterior parietal cortex, which is the brain region devoted to selective attention (Facoetti, Paganoni, & Lorusso, 2000), dominating the dorsal “where” stream, it is more often defined the magnocellular-dorsal (M-D) pathway (see below). It has been hypothesised that the ‘transient’ component of the visual system is crucial in controlling saccadic movements during reading, through the inhibition of the parvocellular system, which, in turn, is important for the correct fixation of the visual stimuli.

Indeed, an impairment in this system might provoke deficits in several functions underlying reading: ocular movements, peripheric vision, movement perception and more general attentional difficulties. Therefore, even mild impairments in this system might lead to a spatial and temporal deficit in individuals with dyslexia. In this regard, several studies support the idea of abnormalities in several loci of the M system (Stein, 2018), although their conclusion is still source of controversy (for all, Skottun, 2014; Skottun, 2005; Skottun, 2000): retina (Pammer & Wheatley, 2001), lateral geniculate nucleus (Giraldo-Chica et al., 2015; Livingstone et al., 1991), primary visual cortex (Johnston et al., 2017; Talcott et al., 1998; Talcott et al., 2002; Pina Rodrigues et al., 2017), V5/MT (Eden et al., 1996; Demb et al., 1998).

In addition, behavioural studies showed reduced performances of individuals with dyslexia in comparison with typically reading peers in a series of tasks, such as real and illusory movement, that are known to tap the M system (Gori, S., Cecchini, P., Molteni, M. & Facoetti, 2014; S. Gori & Facoetti, 2015b; Gori, Molteni, & Facoetti, 2016; Menghini et al., 2010). It is important to notice that, according to the newer interpretation of this theory, the impairments could be not only localised in the Magno-cells themselves but also anywhere along the dorsal stream. In this regard, Gori and Facoetti stated: “[...] deficits in the M-pathway could influence higher visual processing stages by the D-stream. Therefore, reading difficulties could come out due to an impaired attentional orienting system [...], which is anatomically contained in the D-stream” (Gori & Facoetti, 2015a). Therefore, dyslexia might arise from an attentional deficit, which, in turn, disrupt the visual-orthographic system (Boden & Giaschi, 2007; Bosse, Tainturier, & Valdois, 2007; Facoetti, Paganoni, & Lorusso, 2000; Facoetti, Lorusso, Cattaneo, Galli, & Molteni, 2005; Franceschini et al., 2018; Franceschini, Gori, Ruffino, Pedrolli, & Facoetti, 2012; Gori, Cecchini, Molteni, & Facoetti, 2014; Gori, Seitz, Ronconi, Franceschini, & Facoetti, 2016; Hari & Renvall, 2001; Pammer, 2014; Pammer & Vidyasagar, 2005; Ruffino et al., 2010; Valdois et al., 2003; Vidyasagar & Pammer, 2010; Vidyasagar, 1999). Using the expression coined by Hari and Renvall (2001), individuals with dyslexia seem to suffer from a “sluggish attentional shifting” mechanism. In fact, learning to read requires an accurate and rapid top-down mechanism that has been defined by Posner, Snyder, & Davidson (1980) as “a spotlight of attention” (Figure 6). This mechanism allows the selection of a specific area within the visual field in order to adequately process the stimulus, while simultaneously filtering the irrelevant and distracting

information. Indeed, the perceptual segmentation of the string into the correspondent graphemes – at the basis of the grapheme-phoneme conversion – is enabled by an efficient attentional shifting.

**Figure 6** | The “Spotlight of attention”. Source: drawn from Vidyasagar and Pammer (2010)



Attentional deficits might arise not only from impairment at the level of the magnocellular pathway, but also as a consequence of the problematic functioning of other areas involved in attentional tasks. Evidence of a sluggish attentional deployment of attention comes from the studies on temporal order judgment (Hari, Renvall, & Tanskanen, 2001), lateral masking (Martelli, Filippo, Spinelli, & Zoccolotti, 2015; Moores, Cassim, & Talcott, 2011; Spinelli, De Luca, Judica, & Zoccolotti, 2002), spatial-cueing tasks (Facoetti et al., 2006; Facoetti, et al., 2010a; Roach & Hogben, 2007), rapid multi-element presentation (Bosse et al., 2007). In support of this theory, Hari, Renvall and Tanskanen (2001) demonstrated an impaired attentional shifting mechanism entailing difficulties in processing stimuli in the left visual hemifield (left-sided mini-neglect).

Several studies are coherent with the hypothesis of an attentional weakness in the left visual field with, on the other hand, an augmented efficiency toward the right (Facoetti & Molteni; 2001; Facoetti, Turatto, Lorusso, & Mascetti, 2001; Lorusso et al. 2004).

Finally, a recent study (Franceschini et al., 2018) investigated the visuospatial attentional efficiency measuring inhibition of return (IOR) in a lexical decision task. Results showed that dyslexic adults did not display any IOR effect (highlighting a problematic attentional functioning).

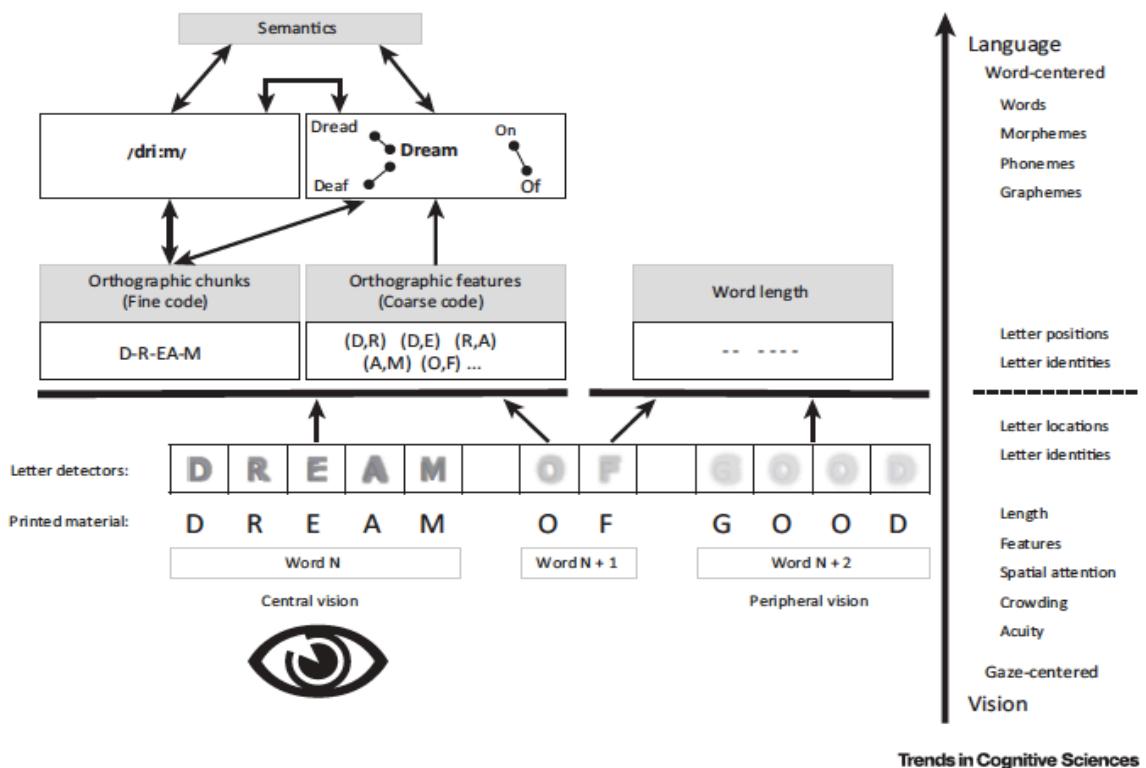
In the literature, there is accumulating evidence of a higher sensibility for dyslexics to crowding, namely the negative effect that visual distractors play towards a central target (Martelli et al., 2015; Spinelli et al., 2002; Gori and Facoetti, 2015 for a review).

The aforementioned findings support the hypothesis of a spatial and temporal attention deficit in children and adults with dyslexia, more specifically in dyslexic with poor phonological decoding skills (Gori, Cecchini, Molteni, & Facoetti, 2014; Ruffino et al., 2010). Indeed, serial attention is fundamental for graphemic segmentation and, therefore, for an efficient grapheme-to-phoneme conversion (of the sub-lexical route). However, the same authors highlighted that learning to read relies also on an efficient perception of the global scene prior to the parsing and the identification of sub-lexical orthographic units.

These results, possibly demonstrating the presence of abnormalities in the functioning of the right fronto-parietal network in individuals with dyslexia, are in line with the works of Valdois and collaborators. Specifically, they suggested that this dysfunction prevent dyslexics from having adequate simultaneous processing of multiple stimuli (visual attention span – VA span deficit). The Visual attention span was defined as “a deficit in the number of individual visual elements that can be processed simultaneously in a visual multi element array” (Lallier, Donnadieu, & Valdois, 2013; Lobier et al., 2012). The main difference with the theoretical approach described above is that the VA span deficit claims that a parallel rather than serial visual parsing and identification are impaired in dyslexia. Moreover, while the former proposes reduced attentional resources in terms of spatiality, the latter focus more on reduced quantity. However, these deficits seem to characterise the different sub-types of dyslexics and, therefore, might be no contradiction between the two accounts. Of this view are Grainger, Dufau, & Ziegler (2016) that proposed an “integrative framework” for basic process in reading alphabetic languages (Figure 7).

Within this integrative framework, an important role is played by a system specialised in parallel letter processing that assigns letter identities to different locations aligned along the horizontal meridian. Important factors are, indeed, crowding and visual acuity; they are both modulated by visuospatial attention. As Grainger and colleagues stated: “Successful development of reading skills involves the use of spatial attention to implement parallel letter processing and the efficient conversion of letters and letter combinations into their corresponding sounds. Developing mechanisms for computing letter identities, their location (relative to fixation) and their position within a word is therefore thought to be one of the keys to becoming a skilled reader”.

**Figure 7** | An architecture for orthographic processing during reading and its connection with phonology and semantics. Source: drawn from Grainger, Dufau, & Ziegler (2016)



#### 5.4. Executive system

Finally, although this hypothesis is less considered, several studies suggest impairments at the level of the Executive system in dyslexia. Regarding working memory (WM) deficits, Bacon et al. (2013) and Swanson et al. (2010, 2009) consider them one of the defining characteristics of dyslexics' reading. Numerous studies have shown disorders in both the verbal and visual-spatial components of WM in children (Menghini et al., 2011; Ghidoni and Angelini, 2008; Smith-Spark and Fisk, 2007; Brosnan et al., 2001) and in adults with dyslexia (Alloway and Alloway, 2013; Smith-Spark et al., 2003). Studies that report a deficiency in WM tests in adolescents and adults demonstrate that these problems, if left untreated, remain even in adulthood.

It has to be noted that, in dyslexics, deficits have been documented not only within the mnemonic system but also in other components of the executive domain, such as phonological and categorical fluency (Reiter, Tucha & Lange, 2005); inhibition, monitoring and “online” revision, through the administration of the Wisconsin Card Sorting Test (Helland and Asbjørgen, 2000), Stroop task (Helland and Asbjørgen, 2000; Reiter et al., 2005; Brosnan et

al., 2002; Everatt et al., 1997), Go / No-Go (Zumberge, Baker e Manis 2007) or Tower of London (Swanson et al., 2006; Reiter et al., 2005; Mati-Zissi and Zafiroglou, 2001).

Similarly, De Beni, Palladino, Pazzaglia and Cornoldi (1998) report longer response times in memory tasks associated with a higher number of intrusions (false alarms), suggesting that in dyslexic children there is a greater difficulty, compared to typical readers, to inhibit irrelevant information. In summary, different studies tend to confirm the link between executive skills and the ability to read efficiently not only in school-age children (for a recent review, see Verhoeve, Reitsma and Siegel, 2011), but also in preschool age (Davidse, de Jong, Bus, Huijbregts and Swab, 2011; Valiente, Lemery-Chalfant and Swanson, 2010; Bierman, Nix, Greenberg, Blair and Domitrovich, 2008; McClelland, Acock and Morrison, 2007, 2006).

## **6. Final remarks**

Dyslexia is a complex multi-faced disorder that is phenotypically heterogeneous and, therefore, cannot be adequately described within a single theory. According to a multifactorial model of dyslexia (Pennington, 2012), it is the whole, closely connected and communicating, of the different cognitive deficits discovered in dyslexia that causes the onset of the disorder. It is still under debate which – and especially how – these deficits are related to the aetiology of dyslexia. Thus, a substantial part of the academic word seems to agree on considering dyslexia not as a monolithic entity, but as a condition that must be distinct in two or more subtypes.

## CHAPTER 2

### COGNITIVE TRAINING OF EXECUTIVE FUNCTIONS

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*“If we knew what it was we were doing,  
it would not be called research, would it?”*

*~ Attributed to A. Einstein*

#### 1. Executive functions

Executive functions (EFs) is an umbrella term that indicates a number of interrelated cognitive processes that subserve purposeful, goal-directed behaviour (Anderson, 2001; Gioia & Isquith, 2004; Gioia, Isquith, & Guy, 2001; Huizinga, Dolan, & Molen, 2017; Miller & Cohen, 2001; Luria, 1966; Shallice, 1982). In general, as a psychological construct, EF refers to a wide range of cognitive functions that are required for the conscious, top-down control of lower level processes and that are usually associated with the activity of the prefrontal cortex and its associated circuits (Niendam, Laird, Ray, Dean, & Carter, 2013; Stuss, 2011; Zelazo & Müller, 2002). Since the executive system is highly demanding regarding the cognitive resources involved, it is activated only during the novel, non-routine and unstructured situations. In this case, no previously established routines for responding exist and, therefore, it is essential to adjust the behaviours flexibly and efficiently (Zelazo, Muller, Frye, & Marcovitch, 2003). These abilities form an essential base for dealing with novel situations, implementing new behavioural sequences and verifying that their implementation is appropriate to the context and the requests of the environment. In this regard, Diamond (2013) stated that the use of executive control requires effort and, thus, “it is easier to continue doing what you have been doing than to change, it is easier to give into temptation than to resist it, and it is easier to go on ‘automatic pilot’ than to consider what to do next” (p.1). EFs are at the base of the intelligence behaviour that distinguishes us as human beings, even though they are distinguishable from general intelligence or “g factor”. Several studies, in fact, showed that, even if there is evidence of mild/strong correlations between EFs and IQ (particularly for specific aspects of the intelligence domain such as working memory), it is

clearly possible to dissociate them (Arffa, 2007; Friedman et al., 2006; Friedman & Miyake, 2016; Kaushanskaya, Park, Gangopadhyay, Davidson, & Weismer, 2017).

This term comprises several abilities such as interference control, response inhibition, cognitive flexibility, the ability to deal with dual tasks or to switch between tasks, decision-making, temporal ordering of events, problem-solving, planning, sequencing, several aspects of attention (e.g. sustained attention) and memory (i.e. updating, maintenance and manipulation of the information), verbal and design fluency, utilization of feedback, multitasking (Burgess, Veitch, de Lacy Costello, & Shallice, 2000; Damasio, 1995; Diamond, 2013; Miyake et al., 2000; Shallice, 1988 ; Stuss & Benson, 1986 ; Stuss, Shallice, Picton, & Binns, 2005; Stuss, 2011). However, it is well known that no consensus is reached on a precise definition of EFs and clear identification and conceptualisation of their nature (e.g. unitary vs fractionated construct). Besides, Zelazo & Müller (2002) noted that this construct is at risk of theoretical incoherence due to a lack of agreement even on the terminology. Executive function/s, executive control, executive functioning, executive system and central executive are frequently used interchangeably.

For a schematic presentation of the concepts and/or components of EFs, see Table 2 drawn from Jurado & Rosselli (2007). Definitions abound for the EF concept and its composition, since researches in this field have to face several theoretical and methodological challenges. First of all, the difficulty in isolating the specific executive components evaluated by each assessment tool. Indeed, since EFs refer to higher-level processes that are essential for the control of lower-level processes, it is almost impossible to capture them directly: each ‘executive’ task requests the activation not only of higher-level abilities but also of other lower-order processes that mediate the efficiency of the EFs.

**Table 2** | Concepts and/or components of executive functions, adapted and extended from the table presented in Jurado & Rosselli (2007).

Authors	Concepts and/or components of executive functions
Lezak (1983)	Volition, planning, purposive action, effective performance
Baddeley and Hitch (1974)	Central executive, phonological loop, visuospatial sketchpad
Norman and Shallice (1986)	Supervisory attentional system
Welsh et al. (1991)	Response speed, hypothesis formulation and impulsivity control, planning

Levin et al. (1991)	Freedom from perseveration, concept formation, planning
Lafleche and Albert (1995)	Concurrent manipulation of information: cognitive flexibility, concept formation, cue-directed behaviour
Borkowsky and Burke (1996)	Task analysis, strategy control, strategy monitoring
Pennington and Ozonoff (1996)	Behavioural inhibition, planning, verbal and visuospatial WM, cognitive flexibility, phonemic and semantic fluency
Zelazo et al. (1997)	Representation, execution, planning, evaluation
Miyake et al. (2000)	Shifting, inhibition, updating
Anderson et al. (2001b, 2002)	Attentional control, cognitive flexibility, goal setting, information processing
Delis et al. (2001)	Flexibility of thinking, inhibition, problem-solving, planning, impulse control, concept formation, abstract thinking, creativity
Hobson and Leeds (2001)	Planning, initiation, preservation and alteration of goal-directed behaviour
Piguet et al. (2002)	Concept formation, reasoning, cognitive flexibility
Elliot (2003)	Problem-solving, behaviour modification in light of new information, strategies definition, sequencing complex actions
Banich (2004)	A purposeful and coordinated organisation of behaviour. Reflection and analysis of the success of the strategies employed

Besides, it is important to highlight that most of the researches focused only on the executive processes responsible, in example, for the formation of goals and strategies and the selection of relevant information, while excluding distractors that may interfere with the achievement of the target objectives. All these processes, that require conscious control of thoughts and actions without involving emotional arousal, have been termed “cold EFs” (Grafman & Litvan, 1999; Rubia, 2011; Skogli, Andersen, Hovik, & Øie, 2017; Zelazo & Carlson, 2012; Zimmerman, Ownsworth, O’Donovan, Roberts, & Gullo, 2016). On the other hand, the EFs involving more emotional component, such as the ability to delay gratification, self-regulation, experience of reward (i.e. reward sensitivity)/punishment and affective decision making, are defined as “hot EFs” (Bechara, Tranel, Damasio, & Damasio, 1996; Bechara, Tranel, & Damasio, 2000; Damasio, 1995; Grafman & Litvan, 1999; Zelazo & Carlson, 2012; Zelazo & Müller, 2002). However, it is under debate whether the distinction between hot and cold executive functions makes sense: Zelazo and Carlson (2012) and Welsh and Peterson (2014) suggested that this construct could be considered as more unidimensional in early childhood, but it diversifies over time into specific components. Nevertheless, in our

opinion, it is important to consider the tight connections between the two components: whilst the “hot EFs” regard to interpersonal and social behaviour, the “cold EFs” are more linked with information-processing mechanisms.

Finally, a critical node that remains without solution regards the theoretical model of the executive function domain. Numerous models were proposed: models that vary in the adopted theoretical perspective (some models focused on a single component of the EFs, i.e. working memory or self-regulation, whereas others considered the entire executive system), for the target population (most of the models focused on adulthood, some on childhood and adolescence, and very few provided an overall view of the entire lifespan), for the typology of data collected (some models derived primarily from clinical studies, while others resulted mainly from statistical approaches) and for the level of explanation (some models are located at the subpersonal level, whereas others at the personal level). Indeed, it is important to notice that no universal agreement has been reached on the unitary vs non-unitary nature of this construct. In the literature, unitary, fractionated, hierarchical and sequential approaches to EFs followed one another. In the next paragraphs, three models, one for each influential approach, are presented and briefly described.

### ***1.1. Unitary model of EFs – Supervisory attentional system and its revisions***

The fundamental concept at the base of the model proposed by Norman and Shallice (1986) is the Supervisory attentional system – SAS (see Figure 8); in general, the model aims at explaining how we can implement non-routine, goal-oriented behaviours by guiding the selection and maintenance of the goal-relevant task schema. According to this framework, routine or over-learned behaviours and actions are based on the activation and implementation of a schema. Each schema, which represents a learned sequence of input-output rules, is characterised by a determined level of activation from the trigger quantity (e.g. sensory input or the outcome of other schemata). The schema is selected only when the activation threshold is reached, and it is performed unless another competitive schema inhibits it. This system, named “contention scheduling”, is entirely automatic and it is used only for well-learned routine behaviours (e.g. cycling, making coffee).

On the other hand, non-routine and novel tasks require the activation of the “supervisory attentional system”, which does not exert direct attention control on the operations but modulates their activation levels that depend on external and internal stimulations. In other

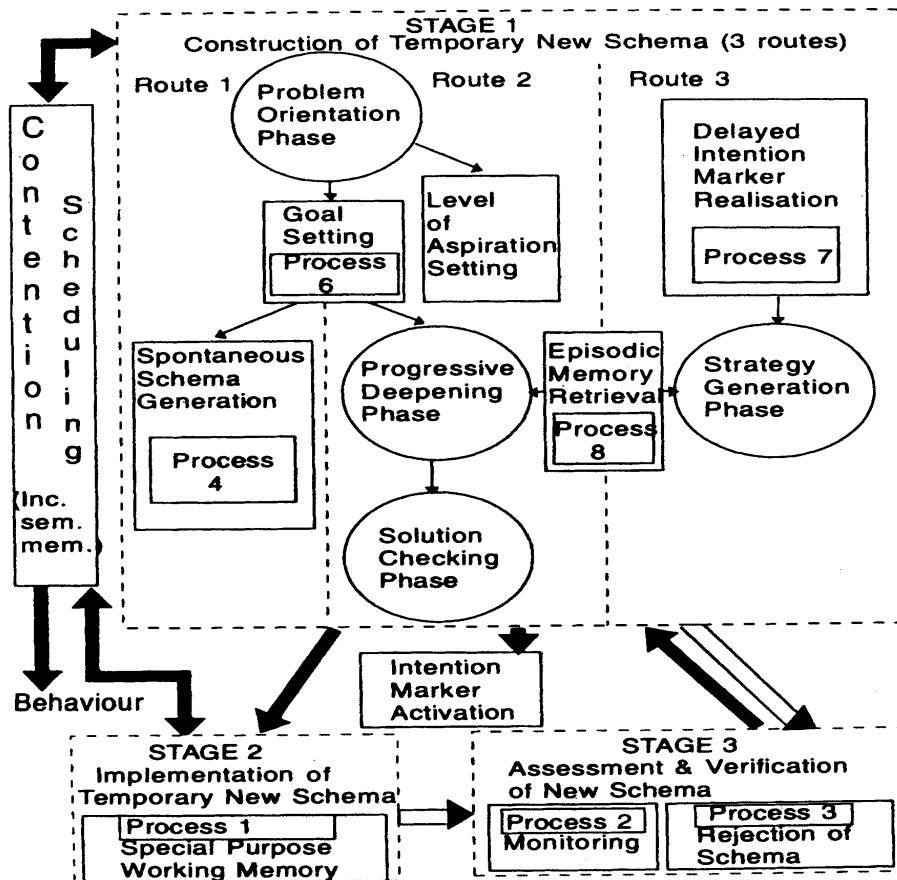
words, the SAS exerts top-down control by deactivating specific schemata and activating others in the service of higher-order objectives. These authors hypothesized that the implementation of non-routine behaviour, and therefore the activation of the supervisory attentional system, is required in five situations: 1) when planning or decision making are involved; 2) when error correction is necessary; 3) when responses are not well-learned or entirely novel; 4) when the individual is facing dangerous situations; 5) when environmental requests force the person to avoid strong habitual responses.

The initial model was extended by Shallice and Burgess (1991, 1993, 1996), mainly focusing on the fractionation of the SAS into its essential components. In this regard, Shallice and Burgess (1996, 1998) stated:

“Even if it is appropriate to view the supervisory system as a single system, it is not correct to view it as carrying out only a single type of process. Indeed, the evidence points to the existence of a variety of processes carried out by different subsystems but operating together to have a globally integrated function” (p. 1405).

Specifically, they argued that SAS behaviour could be exemplified by three main levels. Each of them brings into play determined patterns of cognitive processes and determines the activation of specific areas in the prefrontal region. The first stage is involved in strategies generation as well as schemata of actions that can be used in the short term or recycled in the future for other appropriate situations. The individual needs to adopt a problem-solving approach: he/she has to clearly define the space of the problem, identify the plausible goals, detect other different alternatives and, then, choose the most effective and efficient among them. The next step regards the implementation of the strategy and the associated schema; in this case, working memory capacity plays a fundamental role. Finally, the SAS is asked to monitor the adherence of the implemented schemata to the environmental requests: whenever the schema is not functional anymore, it could be changed or abandoned.

**Figure 8 |** An architecture of the Supervisory Attentional System (SAS) according to Shallice and Burgess (1996)



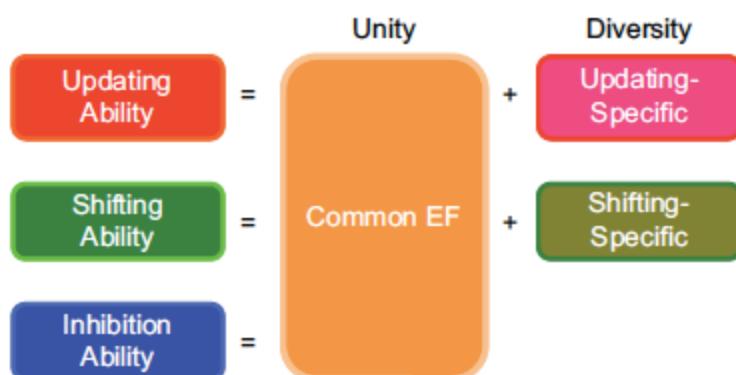
Furthermore, Stuss & Alexander (2007) entered into details in the specification of the role of the SAS. This system consists of three independent functions: 1) energization, the process of initiating and sustaining the currently relevant task schema, which is related to the SM frontal area (particularly the dorsomedial frontal cortex); 2) task setting, the process of setting a stimulus-response and contention scheduling, which involve the activation of the left dorsolateral cortex; 3) monitoring, the process of monitoring performance and modify the behaviour accordingly to the environment, which is associated with the activity of the right dorsolateral area.

### 1.2. Fractionated model of EFs – Miyake et al. (2000)

Miyake and colleagues (2000) proposed a model on the basis of an accurate and in-depth analysis of the literature. They focus on three specific aspects of executive functions: shifting, updating, and inhibition, as well as a common EF ability, which spans these components.

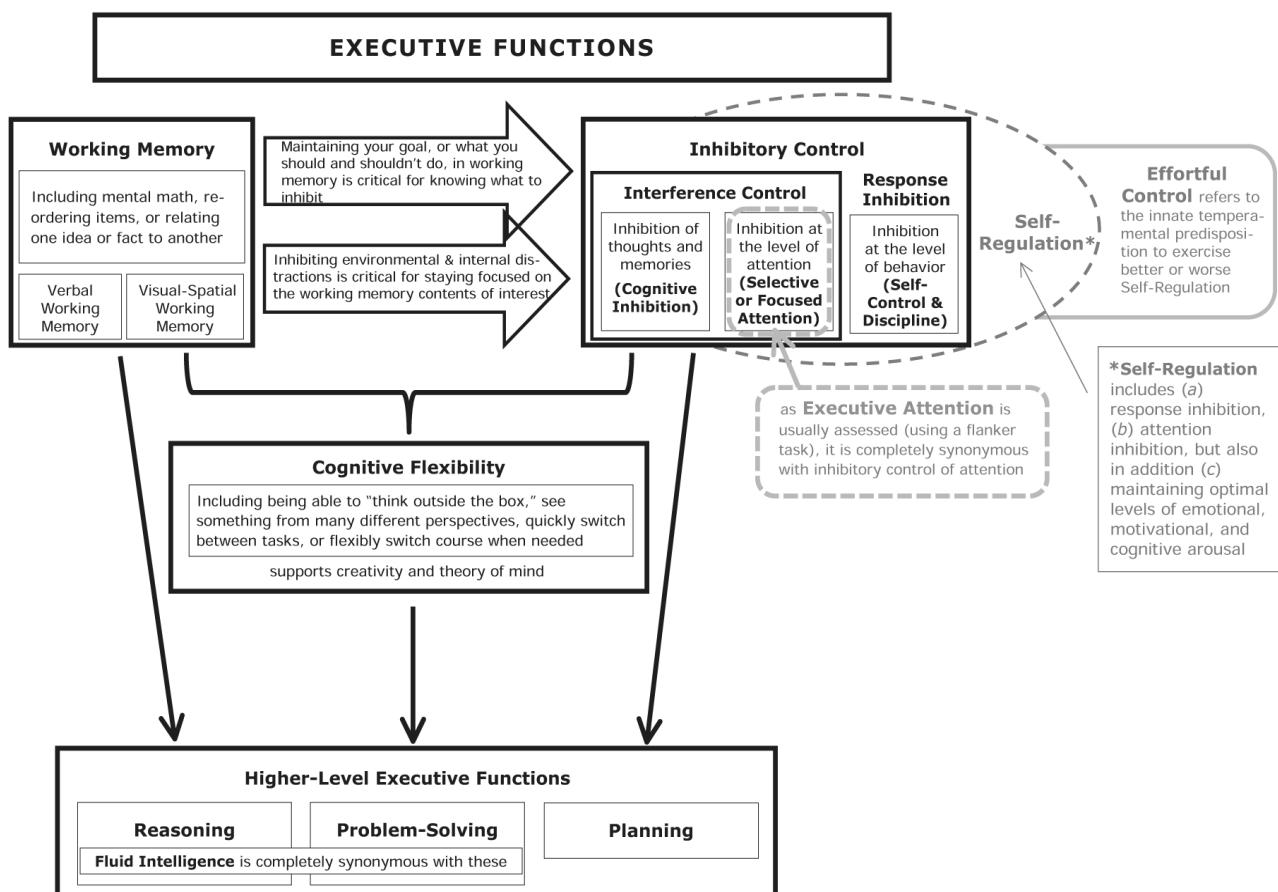
With shifting they referred to the capacity of switching flexibly between behavioural and mental tasks. Shifting or cognitive flexibility entails de-anchoring of attention from a task, operation or mental set that has become irrelevant for the attainment of the goal, subsequent active anchoring on a new mental set or operation and proactively controlling for interferences. The second executive function is the ability to update and monitor, as well as to rapidly add or delete the representations contained in working memory (updating). It is important to notice that the role of this function goes beyond the simple retention of relevant material, since this system is responsible for the active and voluntary manipulation of the information. Finally, inhibition, the last one, refers to the capacity of deliberately overriding automatic, dominant and prepotent responses. It has to be distinguished from reactive inhibition, which is associated with a negative activation and, thus, is not necessarily controlled and voluntarily decided. Based on the results of their study, Miyake and colleagues (2000) concluded that, although these components of the executive system are distinguishable, they share some underlying commonality; they can be considered “separable but moderately correlated constructs” (p.87). Since executive functions correlate with one another, it is plausible to postulate the existence of a common underlying ability. However, they also demonstrate some separability (Friedman & Miyake, 2016; Friedman, Miyake, Robinson, & Hewitt, 2011; Miyake et al., 2000; Miyake & Friedman, 2012). Also, these authors suggested two hypotheses regarding the source of commonality among these three executive functions: maintenance of goal and context information in working memory and basic inhibitory mechanism. In this view, each of the three executive components is a combination of what is common and what is specific to that function (see Figure 9 for a schematic representation).

**Figure 9** | Schematic representation of the “unity-diversity model” of EFs. Source: Miyake & Friedman, 2012.



Interestingly, Diamond (2013) agrees with Miyake and collaborators in identifying three core EFs (Figure 10): inhibition, working memory, and cognitive flexibility. Specifically, she defines inhibitory control as the capacity to “control one’s attention, behaviour, thoughts, and/or emotions to override a strong internal predisposition or external lure” (p.2). In this view, inhibition not only involves self-control or behavioural inhibition, but also interference control (selective attention - cognitive inhibition). Another core component is working memory, which “involves holding information [both verbal and visual] in mind and mentally working with it” (Diamond, 2013; p.22). On the other hand, cognitive flexibility, also called set shifting, is the capacity of “changing perspectives or approaches to a problem, flexibly adjusting to new demands, rules, or priorities (as in switching between tasks)” (p.22). Since these three EFs exist at an intermediate level of complexity, both Diamond and Miyake suggested that higher-order executive functions (i.e. reasoning, problem-solving, and planning) originate from their combination. Indeed, their models are not meant to be an exhaustive representation of the whole executive system.

**Figure 10|** A schematic representation of the executive functions and related terms. Source: Diamond (2013)



### **1.3. Sequential model of EFs – Zelazo et al. (1997, 2003)**

The work of Zelazo and colleagues (1997, 2003) is noticeable not only for the attempt of overcoming the simplistic dichotomy between cold and hot EFs, but also for the Cognitive Complexity and Control Theory (Zelazo, Carter, Reznick, & Frye, 1997; Zelazo, Müller, Frye & Marcovitch, 2003) and its revision (Cunningham & Zelazo, 2007; Zelazo & Cunningham, 2007). This theory posits that unique executive processes take place in an integrated way in order to achieve target goals and/or problem resolution. According to these authors, it is crucial to highlight the strategic and meta-cognition nature of the executive domain, contrary to the fractionated models. The model establishes four stages: representation of the problem and possible solution; planning (e.g. selection of a specific plan from among other alternatives, sequencing of actions in time); execution of the plan, that means both keeping in mind the selected plan for the duration of the action (intending) and effectively translating the plan into action (rule use); evaluation, which is activated only if the previous three end well, involves error detection and error correction processes. This model could be schematically represented in Figure 11.

**Figure 11|** A problem-solving framework for understanding executive functions as a functional construct.

Source: adapted from the original work of Zelazo, Carter, Reznick, & Frye (1997).



This model does not provide an explanation of the EFs, but it captures the dynamic interaction between more bottom-up and more top-down influences on information processing and goal-directed behaviour. Moreover, it gives important information on the development of this system, mainly focusing on the concept of “complexity”. According to this theory, the developmental change is qualitative because there is an improvement in the complexity of the rules acquired by the child from more straightforward to more complex rule systems. This theory postulates the existence of four types of rules: single stimulus-

reward associations, condition-action rules (i.e., if-then contingencies), univalent rule pairs (i.e., each stimulus has one response), bivalent rule pairs (i.e., each stimulus has two possible responses, with the correct response context-dependent). More complex rules allow the child to be more flexible in a broader range of situations. A key trigger for the shifting to newer and more complex rules is the presence of a problem or a conflict: the automatic processes are interrupted, and the child is actively reprocessing information in order to integrate seemingly conflicting rules, producing a new overarching rule system.

In conclusion, despite the large amount of alternative theoretical and empirical approaches to EFs, it is widely recognised that EFs is a construct that encompasses several sub-components. These sub-processes are distinct, but interrelated components of the executive system, and they are organised hierarchically.

## **2. Importance of EFs for (adequate) learning**

Regardless of the lack of agreement on a widely recognised model of the executive functions, growing evidence shows their importance for a successful adaptation to the human world. Indeed, EFs enable us to regulate thoughts and actions in order to achieve our objectives, by adjusting the different patterns of action to the varying environmental requests while, at the same moment, inhibiting irrelevant and inappropriate ones. Thus, an intact executive functioning is necessary for the child's cognitive, emotional and social development. Individual differences in EFs measured in childhood have been found to predict many vital aspects of human functioning such as mental health (Blakemore & Choudhury, 2006; Fraguas et al., 2014; Malloy-Diniz, Miranda, & Grassi-Oliveira, 2017; Poon & Ho, 2014; Snyder, 2014; Snyder, Miyake, & Hankin, 2015; Zimmerman et al., 2016; Zimmerman, Ownsworth, O'Donovan, Roberts, & Gullo, 2017) and physical health (Best, Nagamatsu, & Liu-Ambrose, 2014; Crova et al., 2014; Falkowski, Atchison, Debutte-Smith, Weiner, & O'Bryant, 2014; Hall, Fong, Epp, & Elias, 2008; Riggs, Chou, Spruijt-Metz, & Pentz, 2010), academic (Best, Miller, & Naglieri, 2011; Brock, Rimm-Kaufman, Nathanson, & Grimm, 2009; Gordon, Smith-spark, Newton, & Henry, 2018; Poon, 2014; Samuels, Tournaki, Blackman, & Zilinski, 2016) and occupational functioning (Barkley & Fischer, 2011; Best et al., 2009; Cramm, Krupa, Missiuna, Lysaght, & Parker, 2013; Miller et al., 2012b; Rosenberg, Jacobi, & Bart, 2017; Valiente et al., 2013), social problems (Blakemore & Choudhury, 2006; De Panfilis et al., 2013; García-Andrés, Huertas-Martínez, Ardura, & Fernández-Alcaraz,

2010; Hughes, White, Sharpen, & Dunn, 2000; Sprague et al., 2011; Tseng & Gau, 2013), substance use disorders (Ersche et al., 2012; Giancola & Tarter, 1999; Gil-Hernandez & Garcia-Moreno, 2016; Gustavson et al., 2017; Hagen et al., 2016; Nigg et al., 2006; Nixon, 2014) and eating behaviours (Groppe & Elsner, 2015; Pieper & Laugero, 2013; Tan & Lumeng, 2018). Therefore, low levels of executive functions have been hypothesised to lead to ongoing cognitive, academic and social disturbances that may interfere with the child's capacity to develop normally and interact with the surrounding environment in a socially responsible way (see, Table 3).

**Table 3** | EFs are crucial for the adequate functioning of several aspects of life. Source: Diamond et al. (2013)

Aspects of life	The ways in which EFs are relevant to that aspect of life	References
Mental health	EFs are impaired in many mental disorders, including:	
	- Addictions	Baler & Volkow 2006
	- Attention deficit hyperactivity (ADHD)	Diamond 2005, Lui & Tannock 2007
	- Conduct disorder	Fairchild et al. 2009
	- Depression	Taylor-Tavares et al. 2007
	- Obsessive compulsive disorder (OCD)	Penadés et al. 2007
	- Schizophrenia	Barch 2005
Physical health	Poorer EFs are associated with obesity, overeating, substance abuse, and poor treatment adherence	Crescioni et al. 2011, Miller et al. 2011, Riggs et al. 2010
Quality of life	People with better EFs enjoy a better quality of life	Brown & Landgraf 2010, Davis et al. 2010
School readiness	EFs are more important for school readiness than are IQ or entry-level reading or math	Blair & Razza 2007, Morrison et al. 2010
School success	EFs predict both math and reading competence throughout the school years	Borella et al. 2010, Duncan et al. 2007, Gathercole et al. 2004
Job success	Poor EFs lead to poor productivity and difficulty finding and keeping a job	Bailey 2007
Marital harmony	A partner with poor EFs can be more difficult to get along with, less dependable, and/or more likely to act on impulse	Eakin et al. 2004
Public safety	Poor EFs lead to social problems (including crime, reckless behavior, violence, and emotional outbursts)	Broidy et al. 2003, Denson et al. 2011

The aforementioned, mounting evidence demonstrates the strong link between EFs and children's achievement, classroom behaviour and psychological wellbeing.

Since kindergarten, children have already to show effortful control (i.e., attentional and inhibitory control) in activities such as call and response, listening attentively, respecting turns and complying with teacher demands. Indeed, an adequate learning process requires the

capacity to keep in mind rules and routine through an increasingly efficient working memory, the ability to follow teachers' directions without distracting and being able to delay gratifications and, lastly, the capacity to inhibit impulsive actions or irrelevant responses in favour of doing what is required by the teacher. For these reasons, for a kindergarten or a firsts-grade child, when the learning environment becomes increasingly structured, even activities such as sitting still when required, listening to the teacher while writing on the diary, get along with others could be challenging and requires much effort. As such, both hot and cold components of the executive system could be fundamental in children's capacity or demonstration of learning-related behaviours. In this regard, Jacobson and Mahone (2012) wrote: "EF skills represent an important prerequisite for learning, and provide the cognitive, affective, and behavioural foundation for a student's readiness (globally) and availability (moment-to-moment) for learning. Early school readiness includes not just knowledge of basic pre-academic concepts, but also the ability to behave as a learner."

Several studies showed that EFs are fundamental "learning to learn" skills. For example, McClelland and colleagues (McClelland, Acock, & Morrison, 2006) demonstrated a link between adaptive behaviours of kindergarten children and their academic performances at sixth grade.

Moreover, these adaptive abilities, i.e. stay on task without distracting and participate actively in learning, have been showed to be predictive of subsequent achievements even after controlling for IQ and sociodemographic factors (DiPerna, Lei, & Reid, 2007; Howse, Calkins, Anastopoulos, Keane, & Shelton, 2003; McClelland et al., 2007; McClelland, Morrison, & Holmes, 2000). Specifically, McClelland et al. (2007) found that behavioural measures of self-regulation, namely "the primarily volitional cognitive and behavioural processes through which an individual maintains levels of emotional, motivational, and cognitive arousal" (Blair & Razza, 2008, p.2), actively contribute to early mathematics and literacy abilities.

Duncan and colleagues (Duncan et al., 2007) used six large datasets from the United States, Great Britain, and Canada in order to evaluate the role of three school predictors (i.e. school-entry math and reading skills, attention and socioemotional behaviours) as indicators of later school success. Across all six studies, which took into consideration data from children between 8 and 14 years old, the most important predictors of later reading and math achievement were, in this order, early math, reading and attention performances. In 2010,

Romano and collaborators replicated these important results using two large Canadian datasets; additionally, they found (Romano, Babchishin, & Pagani, 2010) a predictive role also for socioemotional behaviours (specifically hyperactivity/impulsivity, prosocial behaviour, and anxiety/depression) for 3rd-grade math and reading marks. Indeed, a relationship characterised by reciprocity seems to link the development of emotion regulation and executive functions. In accordance with this view are the studies of several authors (García-Andrés, Huertas-Martínez, Ardura, & Fernández-Alcaraz, 2010; Jacobson, Williford, & Pianta, 2011; Liew, 2012; Liew, McTigue, Barrois, & Hughes, 2008; Rimm-Kaufman, Curby, Grimm, Nathanson, & Brock, 2009; Valiente et al., 2011, 2013; Valiente, Lemery-Chalfant, Swanson, & Reiser, 2008; Zhou, Main & Wang, 2010): they showed that early effortful control predicts adaptive and social competencies (e.g. low externalizing problems), which in turn predict their subsequent academic achievement. In this regard, Blair and Diamond (2008) state: “Emotional development and processes of emotion regulation are seen as influencing and being influenced by the development of executive cognitive functions, including working memory, inhibitory control, and mental flexibility important for the effortful regulation of attention and behaviour” (p.1).

The fundamental role played by EFs in adequate learning achievement has been confirmed by other longitudinal studies. For example, Blair and Razza (2007) suggested that the efficiency of the executive system accounted for unique variance in the academic outcomes despite IQ. Mainly, effortful and inhibitory control contribute to emergent math and literacy skills. In primary school, EFs performances are related to achievement in several subjects; specifically, working memory is related with later achievement in English and Mathematics, whereas inhibition with English, Mathematics and Science. The studies of Gropen, Clark-Chiarelli, Hoisington, and Ehrlich (2011) and Grissmer and colleagues (2010) supported the view that science learning relays on adequate executive functions, especially attentional control and inhibition. Science learning requires a constant revision of many naïve theories about causal and categorical relationships, which, in turn, involves the use of higher-order thinking and problem-solving skills. Besides, it has been suggested that the executive functions may be more related to science gains than to the other learning outcomes (Nayfeld, Fuccillo, & Greenfield, 2013).

Regarding mathematics abilities, several lines of evidence demonstrated their relationship with performances in complex working memory tasks, particularly with the ones that involve

numeric stimuli such as digit span backward (Wilson & Swanson, 2001; Wu et al., 2008; Bull, Espy & Wiebe, 2008; Bull & Sherif, 2001; Geary et al., 2000). Analysing the performances in verbal working memory of primary children, Passolunghi and collaborators (2008, 2004, 2001) highlighted their predictive role for mathematics performance. Many other studies have investigated the relationship between working memory and mathematics (see, for a review, Raghubar, Barnes, & Hecht, 2010; for two meta-analyses, Friso-Van Den Bos, Van Der Ven, Kroesbergen, & Van Luit, 2013, Peng et al., 2015). Furthermore, Bull, Jonston and Roy (1999) and Bull e Sherif (2001) found that the number of perseverative responses at the Wisconsin Card Sorting Task is negatively correlated with math scores. Children with low scores in math have low inhibition skills and substantial difficulties in changing flexibly among different strategies. These results have been reinforced by the study of Mc Lean and Hitch (1999): children with arithmetic difficulties were significantly lower in the Trail Making Task – part B, which requires cognitive flexibility. In this regard, Wei and colleagues (2018), in a 4-year longitudinal study with Chinese children, tried to disentangle the role of different components of EFs in predicting different growth parameters in mathematics (Wei, Guo, Georgiou, Tavouktsoglou, & Jordan, 2018). They showed that nonverbal IQ, speed of processing, and number sense play a role in predicting arithmetic accuracy, whereas WM was the only – among all the other EF components – significant predictor of the rate of growth in arithmetic accuracy. On the other hand, the growth in arithmetic fluency may be predicted by earlier number sense, speed of processing, inhibition, and shifting capacities. Although several studies have highlighted the relationship between cognitive flexibility and inhibition (Andersson, 2008; Blair & Razza, 2007; Clark et al., 2010; Willoughby et al., 2012; Geary et al., 2000; Gilmore et al., 2015; Cragg and Gilmore, 2014; Cragg et al., 2017; Passolunghi & Lanfranchi, 2012; Passolunghi e Siegel, 2001; Purpura et al., 2017), this latter appears to be lower in comparison with the role played by working memory.

Although most of the studies focused on the causal link between EFs and mathematics, several executive functions were found to be significantly related to gains in reading and writing (Alevriadou & Giaouri, 2015; Altemeier, Abbott, & Berninger, 2008; Brock et al., 2009; St. Clair-Thompson & Gathercole, 2007; Colè, Duncan, & Blaye, 2014; Drijbooms, Groen, & Verhoeven, 2015, 2017; Monette, Bigras, & Guay, 2011; Poon, 2018; Samuels et al., 2016; van der Sluis et al., 2007). Using the Cognitive Assessment System (CAS), Best,

Miller and Naglieri (2011) examined the relations between complex EFs and academic achievement in a large sample of students aged 5 to 17 years old. Indeed, the correlations between the scores in all the CAS scores and math-related and reading-related achievement subtests varied across ages, but the developmental pattern of the strength of these correlations was remarkably similar for overall math and reading achievement. For example, a task that requires planning abilities (i.e. “Matching Numbers”), solely with numeric stimuli, correlated as strongly with reading as with math, suggesting that these critical academic domains relay on common high-order processes such as planning, self-monitoring, inhibition and working-memory.

As already mentioned in the previous chapter, EFs seem to play a central role in the integration of visual and linguistic information. While attention is fundamental in selectively processing the target information (e.g. single graphemes and/or syllables, whole words), working memory is at the base of the automatic retrieval of linguistic information from the mnemonic system and, then, inhibition seems to influence memory both at the encoding and retrieval stages. In addition, it has been suggested that cognitive flexibility may be required in learning to read since it is helpful in building cross-modal connections between phonology and morphology and to acquire and coordinate multiple dimensions of written language such as morphology, phonology, semantics, syntax (Bierman et al., 2008; Blair & Razza, 2007; Farrar and Ashwell, 2012). In this regard, Colè and collaborators (2014) stated: “The emergence of meta-linguistic awareness, a key component of beginning reading, has been linked to concrete operational thinking, which shares features with cognitive flexibility [...]. Meta-linguistic awareness entails the switching of attention from word meaning to consider other properties of language such as phonology” (p.2).

As regards writing, several studies demonstrated a relation between executive functioning and writing, both for lower and higher levels processes (Hayes & Flower, 1980, 1996). While the first regard handwriting (i.e. automatic letter writing of the alphabet from memory) and spelling processes (Berninger & Amtman, 2003; Berninger, Richards, & Abbott, 2017), the latter are more linked with the problem-solving processes of writing, e.g. planning, translating, and reviewing (Alevriadou & Giaouri, 2015; Altemeier, Abbott & Berninger, 2008; Berninger & Amtman, 2003; Berninger, Richards, & Abbott, 2017; Drijbooms et al., 2015, 2017; Singer & Bashir, 1999). In Drijbooms’s study (2017), it was shown that the syntactic complexity of narrative writing was predicted not only by its initial level and oral

grammar skills, but also by inhibition and planning. Specifically, it seems that EFs (particularly inhibition and updating) affect the text length of the narrative directly, and indirectly, through handwriting, syntactic complexity, and story content (Drijbooms et al., 2015).

In fact, a growing body of research is adding knowledge of how executive functions also contribute to comprehension ability (Cartwright, 2002, 2007, 2012; Cartwright, Marshall, Dandy, & Isaac, 2010; Kendeou, van den Broek, Helder, and Karlsson, 2014; Kieffer, Vukovic, & Berry, 2013; Latzman et al., 2010; Tunmer and Chapman, 2013). However, it is not clear which executive component is essential for reading comprehension. Some researchers have argued that working memory (e.g., De Beni et al., 2000; Perfetti, Landi e Oakhill, 2005; Kane, Hamrick e Conway, 2005) plays a direct role, due to the requirement to keep information active while inhibiting irrelevant information and to respond to questions and draw inferences without having to revisit the text. Vice versa, other researchers consider cognitive flexibility important (e.g., Follmer & Follmer, 2017; Georgiou & Das, 2016), since reading comprehension requires the processing of phonological codes for written word recognition simultaneously with the semantic information. Finally, some researchers (e.g., Follmer & Follmer, 2017) have further argued that planning, specifically the ability to identifying, monitoring and, eventually, revising strategies (e.g. select the main ideas while skipping irrelevant information) is important for reading comprehension.

The aforementioned studies, as a whole, clearly pointed out the fundamental role played by EFs for adequate functioning, with particular relevance for the academic achievements. In addition, there is strong evidence that EF deficits – present in both the general and special education population – put children at risk of several negative life outcomes with lasting cognitive, academic, emotional and social difficulties (Best, Miller, & Naglieri, 2011; Biederman et al., 2004; Brock, Rimm-Kaufman, Nathanson, & Grimm, 2009; Clark, Prior, & Kinsella, 2002; Poon, 2014; Rosenberg, Jacobi, & Bart, 2017; Tapert, Baratta, Abrantes, & Brown, 2002; Valiente et al., 2013).

### **3. Challenges and opportunities for EFs training**

The neurobiological substrate of Executive functioning has been located in the prefrontal cortex (PFC) and a network of other cortical areas including anterior cingulate, lateral

prefrontal, medial prefrontal, and posterior parietal cortices. In addition, these complex cognitive processes are distributed in a network that involves not only cortical structures, primarily the PF, but also subcortical ones, such as the basal ganglia and thalamus. What is important to notice is that, while several studies showed the existence of executive control networks in term-equivalent and preterm infants and, thus, rudimentary EFs seem to emerge in the early years of the development, what has become more evident is that the organization of this “executive network” undergoes dramatic changes over development. Indeed, executive functions develop throughout childhood and into young adulthood, concurrent with a massive overproduction of synapsis, a prolonged period of pruning, myelination of brain regions, and recruitment and consolidation of neural networks (e.g., Stevens, Skudlarski, Pearlson, & Calhoun, 2009; Tau & Peterson, 2010). The PFC is the newest area of the brain and, probably, the most susceptible to positive or negative life experiences.

Indeed, early experiences (negative or positive) can influence PFC trajectories: this protracted developmental course appears to result in increased sensitivity to environmental influences and experience, suggesting that EF development is likely characterized by both increased vulnerability to risk and enhanced opportunities for intervention (Hackman, Gallop, Evans, & Farah, 2015; Karbach & Unger, 2014; Wass, 2015; Petanjek et al., 2011; for a thorough review see Mackey, Raizada, & Bunge, 2013).

Growing evidence, in fact, demonstrates that EFs are sensitive to different types of training. Executive functions can be improved directly, throughout training tools – usually computerised – that target one or more executive components, or indirectly, throughout other activities, such as physical exercise, martial arts, music and language training and classroom curricula.

### ***3.1. Indirect approaches***

Regarding the indirect approach, many studies have found that it is possible to promote EFs via exercise programs, even though most of them lack some criteria, such as random assignment of the groups, the inclusion of an (active) control group and the use of pre and post-training assessments. Nevertheless, some of them were able to pass peer review (Chen, Yan, Yin, Pan, & Chang, 2014; Hillman et al., 2009; Kamijo et al., 2011; Lakes et al., 2013; Lakes & Hoyt, 2004; Tomporowski, Lambourne, & Okumura, 2011; Tuckman & Hinle, 1986; Ziereis & Jansen, 2015; for reviews see Best, 2010; Diamond, 2015; de Greeff, Bosker, Oosterlaan, Visscher, & Hartman, 2018). Diamond, in her review of 2015, investigated the

evidence of the positive effects of physical activities on executive functioning. She concluded that those activities have “little or no cognitive benefit, certainly little or no improvement to the executive functions that depend on prefrontal cortex” (p.1). In addition, Diamond & Ling (2018) specified that “aerobic exercise interventions (with greater or lesser cognitive and motor skill demands), resistance training, and yoga have produced the weakest results for improving EFs of any method tried” (p.1).

Most of these ineffective trainings were realised involving older adults and consisted of resistance training or aerobic activities that require little cognitive abilities (i.e. rapid walking with or without a treadmill, riding a stationary bicycle). On the other hand, there is encouraging evidence for physical activities that involve both exercise and character development or mindfulness/meditation, or that train several motor skills, in improving executive functions (Hillman, Mcauley, Erickson, Liu-ambrose, & Kramer, 2018).

It seems very likely that the physical activities which train and challenge diverse motor and EF skills, helping the creation of a sense of social belonging and providing the child with positive emotions (i.e. joy), while increasing his/her self-esteem and self-confidence, can be the most effective in improving the EFs of young children (Diamond, 2011, 2015). For these reasons, more studies are needed for different sports (i.e. different martial arts, circus, dance). In addition, other studies should look at the role of metacognitive processes during physical activities and their regulation of children’s behaviour and executive performance (Tomporowski, McCullick, Pendleton, & Pesce, 2015). Finally, van der Niet et al. (2015) pointed out the importance of considering both the quantity and quality of physical activity in relation to cognition in children. In their study, they found a relation between the total volume of physical activity, which consisted mostly of light intensity physical activity, and children's executive functions.

As already mentioned above, an alternative approach consists in enrolling children in musical training (Moreno et al., 2009, 2011; Moreno & Bidelman, 2014) and musical training in addition to second language training (Janus, Lee, Moreno, & Bialystok, 2016; Moradzadeh, Blumenthal, & Wiseheart, 2015). Another holistic approach that seems to bring improvements in EFs regards all the activities (i.e. yoga and mindfulness) that train self-regulatory abilities (e.g., Zelazo & Lyons, 2012). For example, Purohit & Pradhan (2017) examined the effects of yoga (which involves synchronised movements through postures with breath, relaxation, breathing exercises, and sensory awareness) on orphan adolescents. After

three months of Yoga program (90 minutes per day, four days per week), the yoga group improved more in comparison with the passive control group in several executive function tasks. The first systematic review of hatha yoga efficacy (Luu & Hall, 2016) shows promising benefit for the executive functioning both of adults and children. Indeed, hatha yoga, the most widely practised form of yoga in western countries, involves bodily postures (*asanas*), breathing techniques (*pranayama*), and meditation (*dyanaa*) in order to bring not only physical health, but also mental well-being.

Analogously, mindfulness-based interventions focus on training and monitoring a moment-by-moment awareness of our thoughts, feelings and bodily sensations, without judging them. During mindfulness practice, one's attention needs to focus on the present moment in an open and non-judgemental way, rather than rehashing the past or imagining the future (Davidson et al., 2003; Kabat-Zinn, 2003).

A study comparing a mindfulness-based plus reflection program to Literacy training or Business as Usual found that preschoolers in the Mindfulness + Reflection condition showed significant improvements at the follow-up (4 weeks post-test) in comparison with the Business as Usual group (Zelazo, Forston, Masten, & Carlson, 2018). It has been suggested that mindfulness contributes directly to enhance executive processes by training sustained attention with reprocessing and conscious reflection of information and by mediating bottom-up influences such as arousal, anxiety, self-esteem and motivation (Zelazo & Lyons, 2012). Empirical evidence on (albeit mild) positive effects of mindfulness intervention on executive functions comes both from adult and children studies (Adams, 2015; Chiesa, Calati, & Serretti, 2011; Gallant, 2016; Teper & Inzlicht, 2013; Zelazo et al., 2018).

Besides, studies that have looked at the positive effects on children's EFs from school-based interventions and school curricula have failed to find consistent results. However, two curricula were showed to improve EFs in young children: Tools of the Mind (Diamond, Barnett, Thomas, & Munro, 2007; Barnett et al., 2008; but see Lillard et al., 2012 for the contrary) and Montessori (Lillard & Else-Quest, 2006; Lillard et al., 2012). The first is a curriculum inspired by the works of Vygotsky (Bodrova & Leong, 2007): the central idea is that sociodramatic play and self-regulatory speech might improve self-regulation and literacy skills. During these activities, the child has to inhibit acting out of character, remember roles (his/her own and those of the others) and, especially, adjust the behaviour flexibly and according to the improvisations of the classmates. Baron, Evangelou, & Malmberg (2016) showed that children who attend this curriculum did improve on what they practised, but

benefits did not generalise to other activities. Furthermore, they suggested that generalisation could be reached through the use of challenges of the EFs that are genuinely included in the everyday academic activities and that should have a higher ecological validity for this reason. On the other hand, evaluations of the effects of the Montessori curricula on EFs seem to paint a more positive picture, but other studies are requested in order to demonstrate which are the characteristics (i.e. an environment with reduced stress, an emphasis of character development) that truly matter in improving EFs. Both Tools of the Mind and Montessori education share some essential features with other school-based interventions complementary to the regular school curricula (Diamond, 2011): PATHS (Riggs, Greenberg, Kusche, & Pentz, 2006) and CSRP Head Start (Raven et al., 2008, 2011).

PATHS (Promoting Alternative Thinking Strategies) is a school-based intervention program that focuses on building children's competencies in self-control recognising feelings and managing interpersonal problems. This curriculum, which emphasizes the integration of affect, behaviour, and cognition via the reinforce of top-down control (i.e. verbalizing one's emotions and/or waiting before speaking or acting), has been shown to bring improvements in EFs that, in turn, modulated its effects on problem behaviours (Riggs, Greenberg, Kusche, & Pentz, 2006).

Bierman and colleagues (2014) randomly assigned 356 kindergarten children to an enriched intervention of the Head Start, REDI (Research-based, Developmentally-Informed) intervention or to "usual practice" Head Start classroom. The results showed that the REDI program promoted emergent literacy, learning engagement, and social-emotional competencies and reduced aggressive disruptive behaviour (especially for children who had started the school year with lower levels of EFs). Besides, children in the REDI group outperformed their peers in some EFs tasks (i.e., shifting and task orientation) and these improvements partially mediated intervention effects on phonemic decoding skills and competent social problem-solving skills. These results were replicated in further studies (Bierman et al., 2017; Nix et al., 2016), suggesting that the REDI Head Start program might improve EFs by creating a supportive interpersonal environment (e.g., establishing rules and routines in classrooms, and promoting emotion regulation).

### ***3.2. Direct approaches***

Direct training approaches typically extensively train fundamental aspects of cognition known as executive system. One approach that is very popular in these years is the use of

computerized software in order to gain improvements in one or more EFs: attention and working memory (e.g., Beck, Hanson, Puffenberger, Benninger, & Benninger, 2010; Brehmer, Westerberg, & Bäckman, 2012; Franceschini et al., 2013; Franceschini & Bertoni, 2018; Franceschini, Bertoni, GIANESINI, Gori, & Facoetti, 2017a; Galbiati et al., 2009; Holmes et al., 2010; Jaeggi, Buschkuhl, Jonides, & Perrig, 2008; Kerns, Macsween, Vander Wekken, Gruppuso, 2010; Kesler, 2008; Kesler, Lacayo, Jo, 2011; Klingberg, 2005; Klingberg, Forssberg, Westerberg, 2002; Lohaugen et al., 2011; Rabiner, Murray, Skinner & Malone, 2010; Sohlberg & Mateer, 1986, 1987; Séguin, Lahaie, Matte-Gagné, & Beauchamp, 2017; Sjo, Spellerberg, Weidner, Kihlgren, 2010; Thorell, Lindqvist, Bergman Nutley, Bohlin, & Klingberg, 2009; Van't Hooft et al., 2005), shifting (e.g., Dörrenbächer & Kray, 2018; Karbach & Kray, 2009; Kray & Ferdinand, 2013; Minear & Shah, 2008; Soveri, Waris, & Laine, 2013; Titz & Karbach, 2014), problem solving (e.g., Patel, Katz, Richardson, Rimmer, & Kilian, 2009; Suzman, Morris, Morris, & Milan, 1997; S. L. Wade et al., 2011; Shari L. Wade et al., 2017) and inhibition control (e.g., Dowsett & Livesey, 2000; Lee, Espil, Bauer, Siwiec, & Woods, 2018; Liu, Zhu, Ziegler, & Shi, 2015; Zhao, Chen, & Maes, 2018). In this regard, see Table 4 for a schematic presentation of the most used computer training tools.

**Table 4 |** Computerized training tools: description of the main features.

Training tools	Publisher	Description	Age range	Studies
Attention Process Training –3 (APT-3)	Lash & Associates Publishing/Training Inc., 2009 www.lapublishing.com	A clinician administers it along with related strategy training and generalisation activities. Targeted areas include: visual scanning/vigilance, sustained and selective attention, divided attention, inhibition).	Adolescents and adults	Sohlberg & Mateer (1987)
Pay Attention! Attention training for children	Lash & Associates Publishing/Training Inc., 2009; www.lapublishing.com	It is an adaptation of the AP and it is designed for children with attentional problems (i.e. ADD, ADHD, traumatic brain injury). It includes tasks that train sustain, selective,	4–10 years old	Sohlberg & Mateer (1987)

		divided, and alternated attention.		
COGMED	Pearson; working memory training	Computerized, massed practice trials of tasks involving attention and WM. Designed to resemble a video game, which features different components of WM.	Pre-school; middle school/ adolescent and adult	Bergman-Nutley & Klingberg, 2014; Klingberg, 2005; Klingberg, Forssberg, Westerberg, 2002; Spencer-Smith & Klingberg, 2015; Thorell, Lindqvist, Bergman Nutley, Bohlin, & Klingberg, 2009
Captain's Log	Brain Train; <a href="http://www.braintrain.com">www.braintrain.com</a>	Multi-level training activities, which aim to develop and remediate a variety of cognitive skills, including memory, attention, mental processing speed, impulse control, listening skills, problem-solving, and more.	For all ages	Boivin, Busman, Parikh et al., 2010; Rabiner, Murray, Skinner & Malone, 2010;
Brain-HQ	Posit Science; <a href="http://www.brainhq.com">www.brainhq.com</a>	An online brain-training system that has 29 online exercises that work out attention, brain speed, memory, people skills, navigation, and intelligence. It is possible to follow the training program proposed by the software or to design a personalised program.	For all ages	Mishra, Sagar, Joseph, Gazzaley, & Merzenich, 2016; Mishra, Merzenich, & Sagar, 2013; Pasqualotto et al., 2015; Pasqualotto, Venuti (in prep.)
Lumosity	Lumos Labs; <a href="http://www.lumosity.com">www.lumosity.com</a>	It is a training software that consists of mini-games intended to increase WM, attention, and a variety of other cognitive domains. Several condition-specific activities are available on an	For all ages	Hardy et al., 2015; Kesler, Lacayo, & Jo, 2011

Although there have been exciting results, few studies have successfully satisfied some high-quality standard criteria (i.e. single or double-blind, random assignment of the participants, inclusion of active/passive control group/s). Besides, very little evidence was found regarding the generalisation of training gains to other domains on which the students had not been trained. It is essential, in fact, that cognitive trainings aim not only at restoring or improving the trained executive functions, but also at generalising improvements of those training effects in other untrained cognitive functions or, broadly, other life domains (transfer effects). Barnett and Ceci (2002) proposed a taxonomy to classify transfer effects, which can be differentiated in *near* and *far*.

*Near transfer* relates to the application of learning to similar situations to the trained one, such as closely related contexts and performances that can be temporal, functional and modality near. This kind of transfer, that is easier to obtain, usually occurs when the stimulus for the original learning event is similar to the stimulus for the transfer event (Royer, 1979) and, thus, the training context and the trained behaviour are almost identical to the application context and application behaviour.

Even though near transfer is quite easy to reach through extensive practice, it is unlikely that the person would be able to flexibly adapt his/her skills to other contexts and situations. On the other hand, *far transfer* can be defined as the application of learning to situations that differ both in the context of application and in the type of required performance to those of the trained task (Laker, 1990). For these reasons, this kind of transfer requires the generalisation of the original learning event (namely the knowledge and skills acquired during the training) to different tasks or to contexts that go beyond the training setting. Even though far transfer is less likely and more difficult to reach in comparison with near transfer, it allows the child to flexibly adapt to new settings that change, as well as the ability to solve new problems, using what was learned in one context in a different one.

Although a growing number of studies showed that EFs could be improved, there is still little consensus regarding transfer effects as a result of executive training. Indeed, the multitude of researches has brought evidence for near transfer, far transfer and both near and far transfer (e.g., Bavelier, Green, Pouget, & Schrater, 2012; Diamond, 2011; Diamond & Ling, 2016;

Enriquez-Geppert, Huster, & Herrmann, 2013; Shinaver, Entwistle, & Söderqvist, 2014; Spencer-Smith & Klingberg, 2015). Also, several researchers failed to find any transfer at all, showing that training effects are limited only to the trained tasks (e.g., Melby-Lervåg & Hulme, 2013; Melby-Lervåg, Redick, & Hulme, 2016; Rapport et al., 2013; Roque & Boot, 2018; Rossignoli-Palomeque, Perez-Hernandez, & González-Marqués, 2018). In several studies, the participants did improve on the tasks in which they practised, but only on them. Also, until now little is known about the long-term effects of trainings (for how long do the improvements last) and about the conditions (i.e. refresher sessions) under which the training yields enduring benefits. Moreover, some studies (e.g., Blair & Raver, 2014; Holmes, Gathercole, & Dunning, 2009) give indications that positive effects of training might appear only later or that they can be broader/higher after a certain amount of time (i.e. six months or one year). Taken together, the current state of research seems to ask for other studies that evaluate the exact mechanisms behind training effects and their long-term and real-life transfers.

Furthermore, it is important to highlight that training studies did not vary only in the target EF and the desired learning transfer, but also in the amount of training and in the variability of training characteristics (e.g., adaptive algorithm vs non-adaptive one). It is not clear whether the positive effects of EFs training are due to the amount of practice. Most of the researches agree on the fact that a longer duration of the training produces higher results in terms of EFs gains (e.g., Diamond et al., 2007; Jaeggi et al., 2011; Jaeggi, Buschkuhl, Jonides, & Perrig, 2008; Spencer-Smith & Klingberg, 2015), but other studies found no differences between the condition with a reduced number of training sessions (with lower duration) and the condition with the double number of training sessions (that lasted more) (Stepankova et al., 2014). In this regard, Au and colleagues (2015) observed a trend for higher improvements in the shorter training sessions condition. However, adequate training, with higher chances of generalisation, seems to involve extended practice, namely a considerable repetition of the skill to be trained. For sure, future studies will have to focus more on the effects of duration, intensity and quantity of the training protocol.

Another principle regarding EFs training that seems to emerge from the past studies is that children with the poorest pre-training EFs are the ones that improve more. This is true not only for computerized training (e.g., Diamond & Lee, 2011; Karbach & Kray, 2009; Titz & Karbach, 2014), but especially for special education curricula such as Tools of the Mind (Blair & Raver, 2014) and martial arts practice (Lakes et al., 2013; Lakes & Hoyt, 2004).

Analogously, it is plausible that younger children (with higher cognitive plasticity) are more likely to have better post-test results and these gains tend to lead to transfer to other cognitive functions more extensively (Wass, Scerif, & Johnson, 2012).

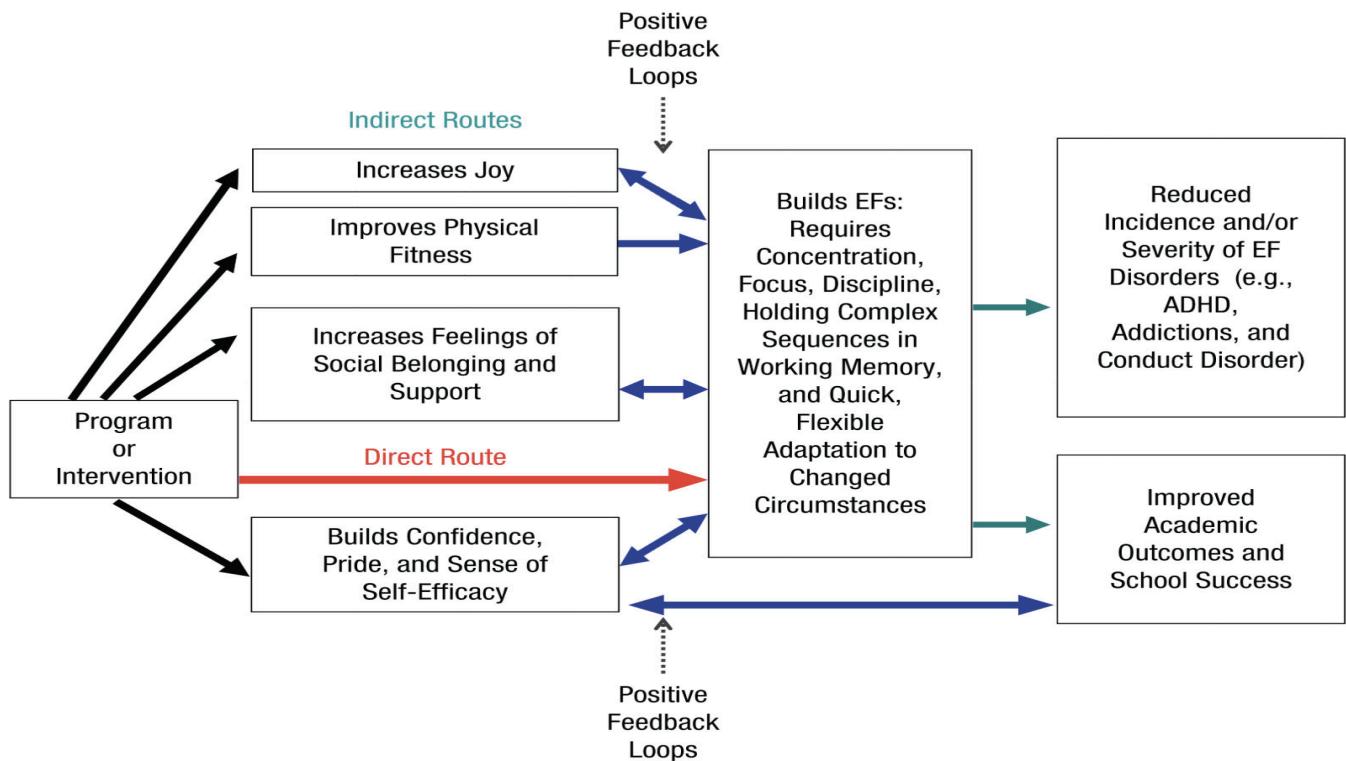
Training effects are also related to two fundamental features for thriving neuroplasticity (Anguera & Gazzaley, 2015; Mishra, Anguera, & Gazzaley, 2016; Mishra & Gazzaley, 2014): first, the presence of continuous feedbacks in order to reward the child showing whether he/she was able to go beyond his/her current level of difficulty; secondly, adaptivity of the training program in order to push the trainer to do better and to avoid boredom and loss of interest. Notably, this latter refers to a well-known concept proposed by Vigotskji (1986), the “zone of proximal development” that has been defined as “the distance between the actual developmental level as determined by independent problem solving and the level of potential development as determined through problem-solving under adult guidance, or in collaboration with more capable peers” (Vygotsky, 1978, p. 86). In this sense, an adaptive algorithm pushes the learner to go beyond what he or she can do without help trying to achieve higher performances.

Furthermore, very few studies have investigated the role of “external factors”, such as fun, motivation, self-esteem, metacognition and expectations, in determining training outcomes (Boot, Simons, Stothart, & Stutts, 2013; Burgers, Eden, Van Engelenburg, & Buningh, 2015; Foroughi, Monfort, Paczynski, McKnight, & Greenwood, 2016; Goghari & Lawlor-Savage, 2018; Katz, Jaeggi, Buschkuhl, Stegman, & Shah, 2014; Mohammed et al., 2017; Schubert, Strobach, & Karbach, 2014; Tsai et al., 2016). Diamond, for example, emphasised the importance of these components for the generalizability of trained skills to ecological settings (2012). She suggested that “the programs that will most successfully improve EFs are those that challenge EFs continually and also bring children joy and pride, give them a feeling of social inclusion and belonging, and help their bodies to be strong, fit, and healthy” (p.338). See Figure 12, for an illustration of Diamond’s model).

In conclusion, future research is still required to address more systematically the factors that influence the effectiveness of direct training and its generalisation to other learning contexts. Nevertheless, the aforementioned studies provided some intriguing promises that well-designed cognitive training is effective and that near and far transfers might be achievable under the right conditions.

**Figure 12|** Diamond's hypothesised model of effective intervention of executive functions.

Source: drawn from Diamond (2011) (p. 41).



## CHAPTER 3

### A MULTIFACTORIAL MODEL OF DYSLEXIA: EVIDENCE FROM EXECUTIVE FUNCTIONS AND PHONOLOGICAL-BASED TREATMENTS

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*“There is no greater or more important investigation of human beings than the cognition [knowledge] of the human being.”*

*~ I. Kant*

#### 1. Introduction

This research aims to analyse the fundamental characteristics and the efficacy of different rehabilitation methods for Developmental Dyslexia (DD). Even though considerable effort has been expended trying to identify the proximal cause of this disorder, no single solution has been agreed upon. On the cognitive level, the most compelling explanation is that children with DD have difficulties in processing the phonological features of words. To date, the treatment based on a systematic and exhaustive training of phoneme awareness appears to be the most efficient and the most studied (Duff et al., 2014; Melby-Lervåg, Lyster, & Hulme, 2012; Snowling, 2013; Tressoldi, Brembati, Donini, Iozzino, & Vio, 2012; Tucci, Savoia, Bertolo, Vio, & Tressoldi, 2015; Vellutino, Fletcher, Snowling, & Scanlon, 2004).

According to the theoretical model at the basis of this treatment, reading descends from the increasingly automatic recognition of groups of linguistic units (graphemes, syllables). Indeed, there is accumulating evidence of a hierarchical pattern of metalinguistic skills in the development of phonological awareness, a fundamental predictor of reading skills (for a review, Melby-Lervåg et al., 2012). In this regard, Snider (1995) wrote: “Children become aware of larger linguistic units (words and syllables) before they become aware of smaller linguistic units (phonemes)”(p.444). In this sense, phonemic awareness constitutes the last ability being developed and, in this framework, onset-rime awareness might be considered as an intermediate passage between awareness of syllables and phonemes (Konza, 2011). The phonological-based treatment aims at improving the ability to quickly recognise and automatically manipulate phonemes via several tasks: i.e. isolating, blending, segmenting, and manipulating larger (rime units-syllables) or smaller linguistic units (phonemes). The

difference between the different tasks lies on the level of complexity: “tasks assessing small phonological units (phonemes) are harder than tasks assessing larger phonological units (rime units or syllables)” (McBride-Chang, 2004 in Melby-Lervåg et al., 2012; p.323). Moreover, tasks depending upon explicit manipulation of speech sounds are harder than those requiring simply forced-choice judgments. In addition to work on phonological awareness skills and on explicit instruction in grapheme-to-phoneme correspondences, trainings that focus on rapid identification of larger linguistic units and on real words in text passages seem to be an effective rehabilitation method for poor reading fluency (Hintikka, Landerl, Aro, & Lyytinen, 2008; Huemer, Landerl, Aro, & Lyytinen, 2008; O’Brien, Wolf, Miller, Lovett, & Morris, 2011; Wolf & Katzir-Cohen, 2001). Generally, Italian children with DD struggle more with reading fluency than with accuracy and it has been proven that they are usually able to reach adequate ability to recognise or decode words, while reading fluency remains the most marked problem. Indeed, research on the development of reading speed and accuracy (Landerl, 1997; Moll et al., 2014; Tilanus, Segers, & Verhoeven, 2013; Tressoldi, Stella, & Faggella, 2001; Wimmer, 1993; Wimmer, & Goswami, 1994) clearly revealed that, in transparent orthographies, the core problem of individuals with dyslexia is the speed or automatisation of reading processes, and that reading accuracy performances tend to reach a ceiling in the last years of primary school. In this regard, O’Brien and colleagues (2011) stated: “explicit training of accuracy precedes the training of speed for each component process to achieve the ultimate goal of fluent reading” (p. 113). To date, several studies have investigated the effects of phonics training and phonological awareness training on reading fluency. Despite encouraging results, poor reading fluency seems to be resistant to intervention especially for older readers (Duff et al., 2014; Duff & Clarke, 2011; Snowling, 2013). For example, Torgesen and colleagues (2001) evaluated the effects of two one-to-one treatments that lasted 8 weeks for a total of 67 hours. These intensive training programs, which combined phonics and phonological awareness instruction, did not show significant differences in their efficacy. Both were able to improve reading accuracy (at the two-year-later follow-up, 70% of children was able to read within the average range), but these improvements did not extend to fluency. Similar results were founded in a large Cochrane review (McArthur et al., 2012) meant to analyse the effects of phonics training on English-speaking children’s reading skills. Despite the fact that phonics-based treatments seem to lead to better reading skills in comparison with other approaches, they show large effects only on non-word reading accuracy and moderate effects on word reading accuracy. The positive

effects on reading fluency were small and inconsistent (results in non-word reading fluency, measured only in one study, were in a negative direction) and did not reach a statistical significance level. In this regard, Lyon and Moats (1997) stated: "Improvements in decoding and word-reading accuracy have been far easier to obtain than improvements in reading fluency and automaticity" (p.579). Nevertheless, the National Reading Panel-NRP (2000) reported that phoneme awareness instruction was able to explain only 6.5% (10% adding letter knowledge) of the variance in reading outcomes (both accuracy and fluency), indicating that definitive and universally accepted results on phonological-based treatment have not been demonstrated yet. Besides, a large portion of students with dyslexia, even after years of treatment, not only struggles with reading fluency, but also with accuracy, showing no (or minimum) effects at the end of the intervention. The issue related to the "non-responders" or "poor responders", namely children whose reading problems are severe and persistent, is that their response, even to effective and well-implemented intervention, is poor (Snowling & Hulme, 2011). Hence, this seems to call for the dismissal of the simplistic single deficit model of this disorder. In the last few years, in fact, there has been growing evidence supporting a multifactorial model of dyslexia: the neurocognitive developmental dysfunctions in this disorder may not be limited to linguistic deficits, but also involve a more multifocal network in which the combination of different impairments in the executive system domain lead to the resulting difficulties in reading acquisition and automatisation (e.g., Menghini et al., 2010; Moll et al., 2013; Peterson & Pennington, 2012; Varvara, Varuzza, Sorrentino, Vicari, & Menghini, 2014). For these reasons, even though phonological-based interventions seem important for an effective remediation of dyslexia, they do not seem to be sufficient (Duff & Clarke, 2011).

It is, therefore, fundamental to investigate under which conditions the treatments of dyslexia are unequivocally effective. In this regard, several researchers (Denton, 2012; Griffiths & Stuart, 2013; Snowling, 2013; Snowling & Hulme, 2011; Torgesen, 2000; Torgesen, 2006) have pointed out that effective reading intervention should be explicit (i.e. direct instructions), intense (both in terms of frequency and duration) and supportive (e.g. from an academic and emotional point of view). In addition, some studies highlighted the importance of one-to-one lessons, as well as a small group of maximum 3-4 children, for effective therapy. Despite the fact that the results on recommended group size are still inconsistent, many experts agree that smaller intervention groups, providing more learning opportunities, are usually more effective in comparisons with larger groups (more than 3-4 pupils). The

choice of the group size has to be made on the basis of the single child's profile characteristics (Duff et al., 2014; Duff & Clarke, 2011; Griffiths & Stuart, 2013): e.g., for children with severe dyslexia and externalising problems, individualised treatment is preferable.

The present study aims to compare a phonological-based reading treatment with a training program of the executive functions (EFs) and to determine whether an integrated treatment (phonological-based + EFs training) has a larger remedial effect on reading skills in children with DD. The cognitive training of EFs was a computer-based training program, which focused on working memory, attention, speed of visual stimuli processing, problem solving, and inhibition. Furthermore, the aim of this study is also to explore which features (i.e. age, gender and cognitive profile of the child) relate to the intervention outcomes. Since we will compare two treatments that differ in setting (clinic-based vs home-based), we will analyse the treatments' efficacy not only in terms of statistical significance but also of clinical significance. In fact, Tressoldi and colleagues (2012) have highlighted the importance of considering efficacy under a cost-benefit view: clinic-based treatments are, usually, highly demanding concerning costs (e.g., clinic, travel expenses) and time required to reach the clinic. As Bogdanowicz & Wiejak (2016) pointed out: "a cheaper therapeutic intervention, which results in an average improvement may be preferable to effective, but expensive therapy".

## **2. Aims and Hypothesis**

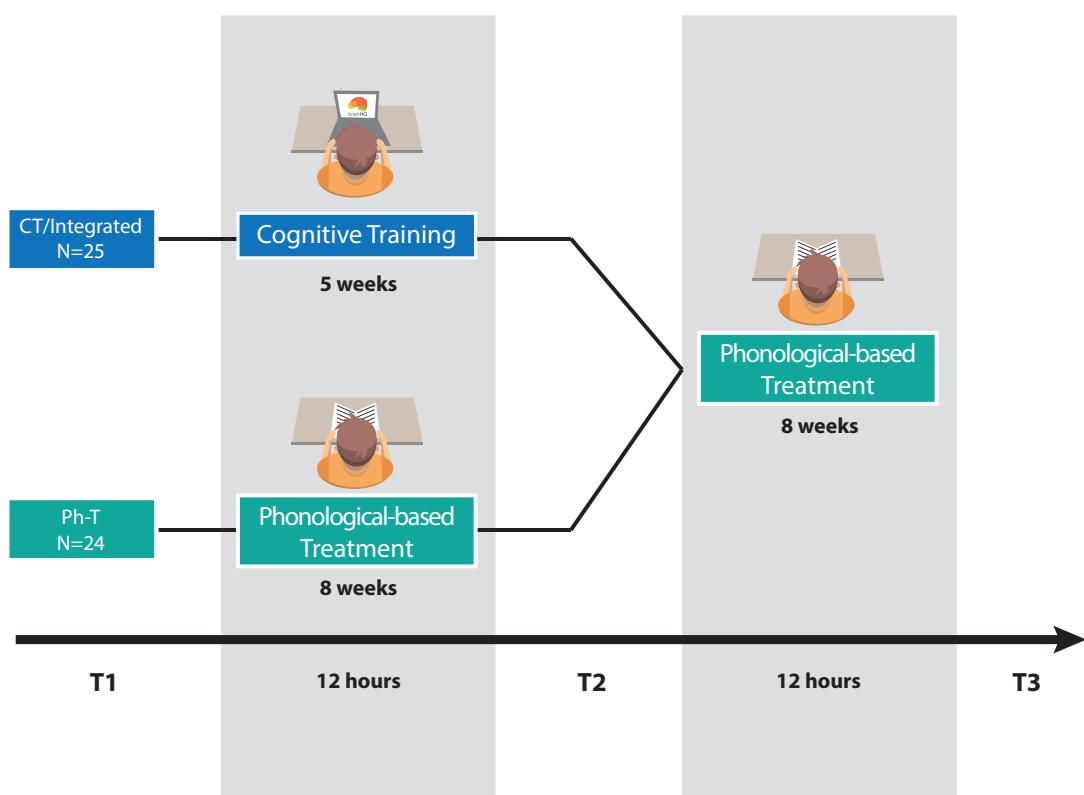
Part A. Compare different interventions for dyslexia and their effects on children with DD cognitive functioning. In particular, we will compare Phonological-based treatment (Ph-T) with Cognitive training of EFs (CT). Significant improvements in the EFs measures are expected in the Cognitive training group only.

Part B. Investigate whether an Integrated treatment (CT + Ph-T) has a more substantial remedial effect on reading skills in comparisons with the pure Phonological-based treatment (Ph-T + Ph-T). Although we expected substantial changes in reading skills for both treatments, our prediction is that only the Integrated group, i.e. the group that underwent 12 hours of Cognitive training prior to 12 hours of Phonological-based treatment, will improve on both reading accuracy and speed. Specifically we predicted that these gains will be related to improvements in the executive functions. Taking into account the clinical efficacy

criteria, we expected children with dyslexia to reach at least the improvements usually made in one year of spontaneous reading development.

In Figure 13, the study plan of the study is represented as a flowchart.

**Figure 13** | The flowchart of the present study.



### 3. Method

### *3.1 Participants*

The study design comprised two groups of children with developmental dyslexia. Participants were patients of the ODFLab (Laboratorio di Osservazione Diagnosi e Formazione - Observation Diagnosis and Training Laboratory) of the Department of Psychology and Cognitive Sciences at the University of Trento (Italy). The diagnosis of developmental dyslexia was made by a group of expert clinical psychologists according to the ICD-10 and DSM-IV inclusion and exclusion criteria. Inclusion criteria for this study were 1) a delay of at least -1.8 SD below the normative threshold in one or more of the three reading tests used (i.e., word, non-word, text), 2) the presence of reading impairment not explained by cognitive, sensorial or neurological deficits, 3) the absence of psychopathological disorders and/or other developmental disorders (e.g. ADHD). The cognitive profile of each participant was evaluated through the Wechsler Intelligence Scale for Children (WISC-IV; Wechsler,

2003; Italian version, 2012). The reading performances were assessed using the word and non-word subtests of the DDE-2 (Sartori, Job, Tressoldi, 1995) and the MT text reading test (Cornoldi et al., 1981). These two tests analyse the speed and accuracy in reading isolated (word and non-word) and non-isolated stimuli (text), respectively. In addition, each diagnosis was discussed by licenced psychologist taking into consideration the impact of this condition on children's everyday lives (i.e. school).

All parents provided informed consent to participate in this study; no differences in the expectations of the two groups of parents were created as we explained that similar interventions had been both proven effective in previous studies. After the written consent was obtained, children were randomly assigned to the two experimental groups: the cognitive training (CT) and the phonological-based treatment (Ph-T). Three children (N=2 for the CT group; N=1 for the Ph-T group) dropped out of the study because they could not keep up with the schedule of the study. Therefore, the final sample consisted of 49 subjects (N=25 for the CT group; N=24 for the Ph-T group), of which 19 Females (38.77%) and 30 males (61.22%). These percentages are consistent with a higher prevalence of this disorder observed in males compared to females (Jiménez et al., 2011; Quinn & Wagner, 2017). All children were native Italian speakers with at least two years of literacy instruction. The mean age was 8.95 (SD =1.20) years and the majority of the participants were third (n = 25; 55.50%) and second (n = 13; 26.53%) grade students.

No significant differences were found between the two groups both for demographic characteristics (i.e., socioeconomic status – SES) and literacy scores at T1. The descriptive statistics of the two groups are reported in Table 5.

**Table 5** | Demographic and cognitive characteristics (Means, SD) of CT group and Ph-T group prior to intervention. All the literacy variables are expressed in z-scores, except for phonemic awareness (percentile score).

<sup>a</sup>χ<sup>2</sup>-value; <sup>b</sup> T-value; \*sig. α= 0.05

	CT group (n=25)		Ph-T group (n=24)		Test statistic	p
	Mean	SD	Mean	SD		
Sex (F-M)	11-15		9-15		0.81 <sup>a</sup>	0.29
Age (Years)	8.91	1.19	8.98	1.21	0.02 <sup>b</sup>	0.97

SES (Family)		36.63	12.89	37.51	9.43	0.01 <sup>b</sup>	0.64
IQ (WISC-IV)		100.47	9.71	98.79	9.53	0.08 <sup>b</sup>	0.35
<b>LITERACY skills</b>							
Word	Speed	-5.30	2.62	-4.98	2.76	2.30 <sup>b</sup>	0.09
	Accuracy	-2.95	2.79	-3.02	2.81	0.28 <sup>b</sup>	0.25
Non-word	Speed	-3.39	2.27	-3.65	1.97	0.27 <sup>b</sup>	0.76
	Accuracy	-2.05	1.93	-1.97	1.74	1.94 <sup>b</sup>	0.68
Text	Speed	-2.14	0.72	-2.30	0.64	0.55 <sup>b</sup>	0.57
	Accuracy	-1.90	0.57	-1.96	0.54	0.79 <sup>b</sup>	0.88
Writing	Word	-2.79	2.56	-2.89	2.69	2.07 <sup>b</sup>	0.13
	Non-word	-1.22	1.84	-1.17	1.96	0.06 <sup>b</sup>	0.99
Comprehension		0.08	0.94	0.17	0.73	2.13 <sup>b</sup>	0.09
Phonemic Awareness		4.38	1.06	4.52	1.01	0.14 <sup>b</sup>	0.86

### **3.2 Tools**

Reading and executive functioning of all children were tested at baseline (T1) and at two other assessment points (T2-T3). All the tests were administered individually one week before treatment (T1) and within 10 days after treatment. Testing was conducted in a quiet room, and generally, each child was evaluated by the same clinician in one/two sessions lasting approximately 1.5 hours. An additional session of approximately 2 hours was performed at baseline to assess IQ. The total number of sessions, as well as the duration of each assessment session and the number of breaks, were carefully calibrated on the basis of each child's capacities and emotional wellbeing. The literacy skills assessment battery consists of reading, writing, comprehension and phonemic awareness tasks. The measures showed good test-retest reliability (Spearman's Rho higher than 0.86). In addition to reading assessment, a neuropsychological protocol assessing attention, working memory, inhibition, verbal fluency, and planning tasks was used.

The raw data were converted to z-scores according to age and/or schooling (with the exception of phonological awareness expressed in percentile).

The tests comprising the battery administered to each participant are briefly presented below.

## INTELLIGENCE

General intelligence was measured by means of the WISC-IV (2003; 2012). The WISC-IV consists of four-factor indexes in addition to the general IQ: Verbal Comprehension Index (VCI), Processing Speed Index (PSI), Working Memory Index (WMI), and Perceptual Reasoning Index (PRI).

## LITERACY SKILLS

*Reading.* Speed and accuracy of reading were assessed using two different instruments: the “Battery for the Assessment of Developmental Dyslexia and Dysorthographia” [DDE-2] (Sartori et al., 1995) and the Test of Speed and Accuracy in Reading, developed by the MT Group (Cornoldi, Colpo, & Gruppo MT, 1986).

- Word/non-word reading: Two subtests from DDE-2 were chosen in order to assess word and non-word reading. In the first subtest, children are required to read aloud 4 lists of 28 words (which are concrete or abstract), which vary in frequency (low versus high) and length (from 4 to 8 letters). In the second task, participants read 3 lists of 16 non-words, namely sequences of letters that are similar, as respecting the phonotactic restrictions of Italian language, to a real word but that is not accepted as such by native speakers. Their length could vary from 5 to 9 letters. For each task, it was possible to compute separate z-scores for speed (reading time) and accuracy (number of errors).

- Text reading: the MT text-reading task assesses reading abilities of meaningful material and it provides different scores for speed (number of syllables read per second) and accuracy (number of errors). Text length (from 250 to 500 syllables) and complexity increase with grade level with specific norms associated with each grade.

*Word/non-word Writing* was assessed through the Spelling tests of the DDE-2 (Sartori et al., 1995). These tests assess accuracy in writing words (n° 48) and non-words (n°24), expressed as the number of correct items and converted into z-scores according to age norms.

*Text comprehension* was evaluated using the comprehension MT task (Cornoldi, Colpo, & Gruppo MT, 1986). This short, multiple-choice, test provides z-scores for correctness (number of correct answers).

*Phonological awareness.* In order to determine children's levels of phonological awareness, the Spoonerism task was administered. This is a subtest of the Test CMF - “Assessment of the metalinguistic skills” (Marotta, Ronchetti, Trasciani, Vicari, 2008). The clinician pronounces two words aloud and the child is asked to swap the initial phonemes of the two

words to form two other real words. For example, the examiner presents "pane" (bread) and "collo" (neck) and the participant has to manipulate the phonemes in order to pronounce "cane" (dog) and "pollo" (chicken). The correctness (one point for each couple of new words) is presented as a percentile score.

## EXECUTIVE FUNCTIONS

*Visuospatial selective and sustained attention* was assessed by means of the "Modified Bell Test" (Biancardi, Stoppa, 1997). In this barrage test, each sheet (for a total of four sheets) contains 35 target stimuli (precisely bells) among other familiar figures (e.g. animals, houses). Participants are required to draw a line on the bells that they were able to localise. Two correctness z-scores are computed: the first one refers to the number of targets the child can find in the first 30 seconds for each sheet, the second one regards the total number of targets located in 120 seconds for each sheet.

*Auditory attention*. To assess sustained attention, the Auditory Attention Test (TAU) of the "Italian battery for ADHD" [BIA] (Marzocchi, Re & Cornoldi, 2010), was used. This task consists of counting several series of sounds at irregular intervals (50-5000 ms). In each exercise, the sequence is composed of 9 up to 15 identical sounds.

*Response Inhibition*. In order to determine motor inhibition capacities, the "Frogs" subtest of the BIA was administered (Marzocchi, Re & Cornoldi, 2010). This Go/no-Go task is a modified version of the "Walk, Don't Walk" task (TEA-Ch, Manly et al. 2001). Participants are asked to draw a sign ("the frog's jump) along a paper path each time they hear "go" sound on tape. For each sequence/path, a "no-go" sound unpredictably occurs signalling the child to stop. The two sounds are identical for the first 208 ms, and they only differ in the last part.

*Verbal memory* was estimated through the digit span subtest of the "Battery for the assessment of visuospatial memory" [BVS-Corsi] (Mammarella et al., 2008). Children are required to recall the sequence of digits pronounced by the clinician. In the first part, known as Digits Forwards, the participant has to repeat the sequence in the same order; whereas in the second part, Digits Backwards, the order has to be reversed. If the child is able to repeat correctly at least two of the three sequences, a sequence one digit longer is presented. If two trials of the same block are wrong, the clinician skips to Digits Backwards or he/she ends the task.

*Visuospatial memory*. In order to evaluate the short-term and working memory in the visual modality, the Corsi's test was used (BVS – Corsi, Mammarella et al., 2008). This test is the

visual counterpart of the Digit Span subtest: it provides both forward and backward parts, and it has identical stopping rules, number and complexity of the sequences. Children are required to tap a sequence of blocks in the same or the reverse order of the one just made by the clinician.

*Verbal fluency.* Two subtests of the “Neuropsychological Assessment Battery” [BVN] (Bisiacchi et al., 2009) were used to measure category and phonological fluency. Children are asked to generate as many words as possible (within the time limit of 60 seconds) on the basis of a specific criterion. For the verbal fluency, the initial input is a particular category (e.g. colours, animals, cities). On the other hand, in the phonological fluency task, the clinician pronounces a specific phoneme (e.g. /s/) as a starting input. Two z-scores, one for each subtest, are calculated from the number of valid words generated (excluding errors and repetitions).

*Planning.* The Tower of London test was used (Sannio, Fancello et al., 2006 Italian version; Shallice, 1982) in order to evaluate planning and problem-solving abilities. Children are presented with a model where three beads are positioned on three pegs with different heights. Participants are then asked to mentally preplan a sequence of moves of the beads in order to match the configuration of beads presented in the model. Additionally, they have to keep in mind the sequence and to execute it step by step, eventually modifying it if it is found uncorrected.

### **3.3 Procedure**

In the first part of the study (*part A*), the Cognitive Training group underwent a 5 weeks training of EFs while the Phonological-based Treatment group completed an 8 weeks phonological-based intervention. The total amount of training hours was the same (12 hours), but they differ in terms of duration and frequency of training sessions. Children of the CT group were asked to train with the cognitive training software (Brain-HQ) for approximately 30 minutes, 5 times per week. The first training session (30 minutes) was not counted as part of the training since it was aimed at the explanation of the training protocol and the functioning of the website. Except for the first training session, and an additional one in the middle of the training period that was performed at the clinic, the participants played alone in their home environment. Parents were instructed to help children at keeping the training time limit through a timer. During each training session, practice time was recorded through the software and, in average, the amount of practice time for each session corresponded to 30.67

minutes ( $SD = 2.35$ ). On the other hand, the Phonological-based Treatment was carried out in 8 individual sessions that lasted one hour and a half each. All the treatment sessions were performed at the ODFLab on a weekly basis. It is important to notice that, for each child, treatment was delivered on a one-to-one basis.

In *part B* of the study, both groups, after the second assessment point (T2), completed an additional 8 weeks Phonological-based intervention, followed by the last assessment point (T3).

The two interventions are described in the following paragraphs.

### *3.3.1. Phonological-based reading treatment*

Several types of linguistic treatment of dyslexia exist as reported by the National Reading Panel (2000) review. However, to date, the Phonological-based treatment - based on a systematic and exhaustive instruction in learning letter-sound correspondences and in manipulating sounds to form words - appears to be the most efficient (Bogdanowicz & Wiejak, 2016; Duff et al., 2014; Galuschka, Ise, Krick, & Schulte-Körne, 2014; Snowling, 2013).

Our phonological-based reading treatment relays on the guidelines provided by the Consensus Conference – Suggestions for the Clinical Practice - 2007 (CC-RPC-2007), 2011 (RC-DSA-2011) and the Consensus Conference on Learning Disorders of the National Health Institute - 2011 (CC-ISS-2011). Furthermore, it has to be highlighted that this kind of treatment has been proved effective for Italian-speaking children with DD in previous studies (Allamandri et al., 2007; Bonacina, Cancer, Lanzi, Lorusso, & Antonietti, 2015; Lorusso, Facoetti, & Bakker, 2011; Mogentale & Chiesa, 2009; Tressoldi et al., 2012; Tressoldi & Vio, 2011; Tucci et al., 2015). Most of the activities of the phonological-based reading treatment were structured using the materials included in widely used manuals (Brignola, Perrotta and Tigoli, 2012; Cazzaniga et al., 2007; Gagliardini, 2011; Perrotta and Brignola, 2000, 2014). Furthermore, we developed additional paper-and-pencils materials or other game-like activities in order to train specific aspects of interest (for some examples, see Appendix A).

In this study, all the activities – and their difficulty levels – were defined on the basis of the specific reading profile of the child. Generally, our children with dyslexia mastered basic reading precursors (i.e. letter knowledge) and seemed able to identify graphemes satisfactorily. For these reasons, the phonological-based treatment aimed at ameliorating the

reading process both in accuracy and in speed facilitating the fast recognition of syllables or other phonological units (e.g. phonemes) that constitute the key subcomponents of words (sublexical approach). In this sense, the primary objective of this treatment was to automatize the child's reading skills also through the creation and extension of the lexicon. This critical aim can be reached through a progressive reduction of the use of the grapheme-to-phoneme conversion mechanism while recognizing "on sight" groups of linguistically relevant units increasingly complex. In addition, this treatment aimed at reaching not only the best reading accuracy possible, but also to address dysfluent reading, which, for Italian children with dyslexia, is usually the main problem (e.g., Spinelli et al., 2009; Zuccolotti, De Luca, Marinelli, & Spinelli, 2014). Indeed, we decided to focus on rapid (and accurate) reading of linguistic units (usually, syllables). While at the beginning of the treatment single reading units or lists were preferred, complete texts were preferred in the last part of the intervention. Some researchers supported the idea that is better to train words in context in comparisons with training words in isolation since it leads to greater generalizability (LeVasseur, MacAruso, & Shankweiler, 2008; Martin-Chang & Levy, 2005; Tressoldi et al., 2012).

### *3.3.2. Cognitive Training*

We decided to use Brain-HQ, an online cognitive training system developed by the neuroscientist Michael Merzenich and his team, in order to develop a training protocol for executive functions. Brain-HQ consists of several short mini-games (average duration: 5 minutes). Its functionality has been proved by numerous studies, published on Posit Science (<https://www.brainhq.com/world-class-science>), which are designed to train EFs through repeated practice. According to Merzenich and collaborators (Merzenich, Van Vleet, & Nahum, 2014; Zhou, Panizzutti, de Villers-Sidani, Madeira, & Merzenich, 2011), intensive training might be fundamental in boosting new neuronal synapses, remodelling the "wiring" in brain circuits. As Merzenich stated: "That plasticity-driven growth in local "teamwork" is a critical aspect of the improvement in the processing of information supporting learning-based advances in behaviour". The constant training can bring a remapping of the cortical areas and a subsequent more automatic and straightforward application of the involved abilities. In this context, the response time should diminish, and the performance in the trained tasks should improve. This might contribute to create, in trainees, a higher sense of self-efficacy that is known to be crucial for the motivation to learn. This kind of training is inexpensive to deliver

in comparisons with the traditional clinic intervention, due to the fact that it only requires an Internet-connected computer.

This online training software has 29 online exercises, organised into six categories that are often overlapping: Attention, Brain Speed, Memory, People Skills, Intelligence, and Navigation. From this pool of exercises, we opted for a subset that best meets our research interests in order to create a 5-weeks training protocol. It has to be noted that several studies suggested that training of one or more of these cognitive functions - through traditional training methods or real video games – are effective in improving reading skills – or at least reading fluency (Benso, 2008; Dahlin, 2011; Franceschini et al., 2013; Franceschini et al., 2017b; Loosli, Buschkuhl, Perrig, & Jaeggi, 2012; Mogentale & Chiesa, 2009; Yang, Peng, Zhang, Zheng, & Mo, 2017). However, meta-analytic confirmation is still lacking, and other well-designed studies are still required (Melby-Lervåg et al., 2013, 2016).

Specifically, three exercises for each week were selected in order to train the different executive functions homogeneously. The last week of training consisted in repeating three exercises that had already been performed but were considered particularly meaningful. We tried to select engaging mini-games with the aim to make the training sessions as pleasant as possible. Moreover, we matched the exercises in order to avoid an imbalance within each session. This means that particularly demanding and tedious exercises were not performed in a row.

In conclusion, the trained cognitive functions were: (1) selective, sustained and divided attention, both in auditory and visual modality, (2) speed of visual stimuli processing, (3) fine discrimination and processing of auditory stimuli, (4) auditory and visuospatial working memory, (5) problem solving, and (6) inhibition.

In the following, a short and schematic presentation (Table 6) of the training protocol with the main characteristics of each exercise.

**Table 6**| Description of Brain-HQ exercises and their functionality.

Source: <https://www.brainhq.com/why-brainhq/about-the-brainhq-exercises>

## WEEK 1

### Divided Attention



#### TASK

Observe two figures and decide as fast and accurately as possible whether they are equal or not on the basis of a particular criterion (e.g., shape, colour). As the participant improves, the figures flash by more quickly and, in addition, the categories double up (e.g., match by colour *and* shape).

#### TRAINED FUNCTIONS

Divided attention, Cognitive flexibility, Inhibition of irrelevant information

### Hawk Eye



#### TASK

Identify the target bird (the only one that slightly differs from the others) as it appears briefly on screen (in the periphery of the visual field). As the participant improves, the stimuli become more similar, while their distance from the centre increases. Moreover, the backgrounds get more and more complex.

#### TRAINED FUNCTIONS

Visual speed and precision; UFOV (Useful field of view)

### Optic flow



#### TASK

Make rapid visual discriminations of the target shape (that is on a sign) and, at the same time, stay alert to potential hazards in the periphery of the screen. As the participant improves, the targets become more similar and the number/type of hazards increase. The navigation become more complex (night and/or bad weather), while the speed of presentation of the stimuli increases.

#### TRAINED FUNCTIONS

Visual attention; Rapid detection and identification of visual stimuli; Inhibition

## WEEK 2

### Double Decision



#### TASK

Decide which of two cars has just been flashed in the middle of the screen, while noticing where a Route 66 road sign appears in the periphery. As the participant improves, the number of distractors, their similarity and their distance from the screen increase. Furthermore, the complexity of the background grows.

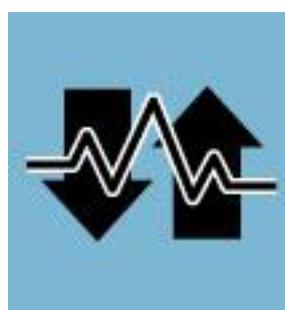
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#### TRAINED FUNCTIONS

Visual processing speed; UFOV

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### Sound Sweeps



#### TASK

Listen to two sounds that change in frequency (“sweep”) and identify their direction (up or down). As the participant improves, sound frequencies vary and the space between the two sweeps shortens.

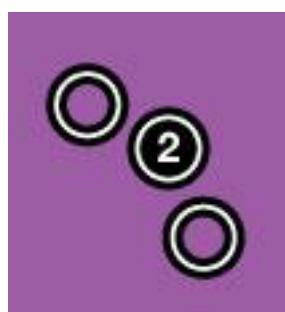
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#### TRAINED FUNCTIONS

Auditory processing speed; Auditory attention

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### Juggle Factor



#### TASK

Reconstruct a sequence of numbers (that appear briefly in the middle of circles) in the correct locations. For this reason, the participant is asked to remember not only the correct order but also the right locations. As the participant improves, the object trajectories become more complex, while the number of targets and distractors increase. In addition, the order presentation changes from forward to backward to random.

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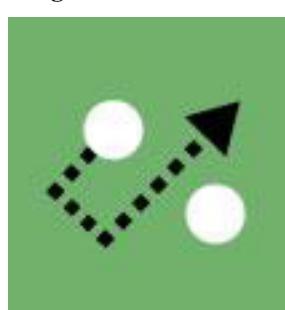
#### TRAINED FUNCTIONS

Visual attention; Rapid detection and identification of visual stimuli

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## WEEK 3

### Target Tracker



#### TASK

Track target objects as they move around the screen (Multiple object tracker – MOT task). As the participant improves, the number of objects and their visual complexity increase, while they move faster, for longer amounts of time, and over larger areas. Furthermore, the complexity of the background grows.

---

#### TRAINED FUNCTIONS

Visuo spatial attention

---

**Scene Crasher****TASK**

Pay attention and memorize the details of the scene that they will soon disappear, after they reappear, identify the added item (delayed-recognition span task). As the participant improves, the contrast between the background and the stimuli reduces, the scene presentation shortens and the display area increase.

---

**TRAINED FUNCTIONS**

Visual working memory; scanning abilities

---

**Eye for Detail****TASK**

Pay attention, remember and then identify some images that briefly flash, one at time, in different positions on screen. As the participant improves, the number of objects increases (i.e. the targets move from 3 to 5) and they become more similar. In addition, they appear over larger areas and with more complex trajectory.

---

**TRAINED FUNCTIONS**

Visual processing speed; Visual working memory

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**WEEK 4**

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**Freeze Frame****TASK**

Press the response key only when you see distractors (reinforced answer), whereas you have to inhibit the answer when you see a target (“Go/no-go task”). As the participant improves, distractors become more similar to the target, the stimulus category differs and the images appear more quickly.

---

**TRAINED FUNCTIONS**

Tonic and phasic alertness; Motor inhibition

---

**Card Shark****TASK**

Remember the sequence of visual information (the cards are added one at time and the turned over) and decide if the current card is the same presented n-times before it (“n-back” task). As the participant improves, the number of cards and their complexity (suit, number) in the sequence to remember increase, while the presentation time reduces.

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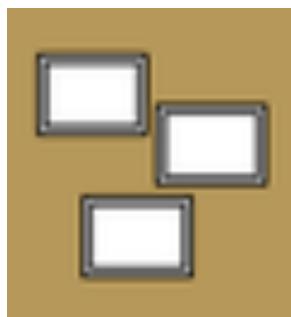
**TRAINED FUNCTIONS**

Auditory processing speed; Auditory attention

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### Syllable Stack



#### TASK

Listen to a series of syllables, and then repeat them in order (“serial memory-span” task). As the participant improves, the number of syllables increases while their frequencies decrease. Moreover, they become more similar and they are pronounced by different voices.

---

#### TRAINED FUNCTIONS

Auditory attention; Rapid detection and identification of visual stimuli

---

### WEEK 5

- Exercise 1: **Target Tracker** (Visuo-spatial attention, Attentional span)
  - Exercise 2: **Freeze Frame** (Inhibition)
  - Exercise 3: **Optic flow** (Visuo-spatial Attention)
- 

### 3.4. Statistical Analyses

#### 1) Training Effects on Literacy Skills

In order to investigate differential training effects on reading skills and on other literacy skills, a series of Analyses of Covariance (ANCOVA) were conducted. In each model, Time (T1 versus T2) was inserted as a within-subject factor and Group (CT versus Ph-T) as a between-subject factor. In order to exclude possible confounding effects, chronological age and IQ scores (Raven's CPM) at T1 and Sex were entered as covariates. Specifically, the Time X Group interaction was our primary effect of interest. We further investigate significances by means of post-hoc analyses.

Reading skills and other literacy skills were measured by means of three z-scores:

- Total Reading Speed: i.e. the average of speed scores in word, non-word and text reading;
- Total Reading Accuracy: i.e. the average of accuracy scores in word, non-word and text reading;
- Total Writing: i.e. the average of scores in word and non-word writing.

Moreover, the comprehension and phonological awareness scores were considered. In conclusion, five separate ANCOVAs were performed.

#### 2) Training Effects on Executive Functions

Vice versa, the training-related changes of the executive functions were analysed by means of a two-way mixed design (MANCOVA) with IQ and Age as covariates, Time (T1 - T2) as a

within-subject factor and Group (CT versus Ph-T) as a between-subject factor. The seven EFs tasks (Bell – TAU – Frogs – Digit span – Visuospatial span – Verbal Fluency – TOL) were inserted in the model as dependent variables. One z-score for each EFs test was taken into consideration. Therefore, for the tests that produce two scores, the average of the two was considered (e.g. digit span forward and backward).

Similar analyses, except for the time points considered (T1 and T3), were run in order to evaluate the group differences between the Integrated treatment (CT + Ph-T) and the pure Phonological based treatment (Ph-T + Ph-T).

Effect sizes are expressed as partial  $\eta^2$  coefficients (values greater than 0.14 are considered large; Cohen, 1988). Only significant results were reported.

### 3) Clinical efficacy comparison

We analysed the efficacy of the two treatments (Integrated versus pure Phonological-based) from a clinical point of view, comparing the improvements in speed and accuracy in word, non-word and text reading to clinical efficacy criteria. According to the Recommendations for the Clinical Practice - 2011 (RC-DSA-2011), a treatment should be considered effective if it modified the clinical condition more than what expected in one year of regular education and reading experience, without professional interventions. Several studies (Tressoldi, Stella, & Faggella, 2001; Tressoldi & Vio, 2008) revealed that Italian children with dyslexia, without specialist treatments, increased by around 0.3 syllables per second their single word and text reading fluency, whereas their non-word reading fluency is increased by approximately 0.15 syllables per second for each grade. Taking into account the clinical significance for accuracy in reading, an arbitrary criterion has been used according to the RC-DSA-2011: a reduction of 50% of the errors with respect to the first evaluation.

### 4) The link between improvements in Reading and Executive Functions

To explore the predictive relationships between EFs gains and reading improvements after 24 hours of treatment, two multiple regression analyses were computed for each Total Reading Scores (delta scores between T1 and T3). In the first two-step fixed-entry regression, the dependent variables were the delta scores (differences between the last assessment time point and the baseline) of the performances in Total Reading Accuracy and Total Reading Speed. Predictors were: age, IQ, sex (block 1); the reading variable at T1 (block 2). Besides, two further stepwise regressions were performed using the standardised residuals of the previous regression models as dependent variables and the improvements in the EFs test as predictors. The delta scores of the EFs tests were indexed by calculating the mean between the delta scores (T3 – T1) in each of the seven executive functions tests.

## **4. Results**

### ***4.1. Part A (T1-T2)***

#### **LITERACY SKILLS**

Reading. The ANCOVA of Total Reading Accuracy showed significant main effect of Time [ $F(1, 43) = 6.078, \eta^2_p = 0.037, p = 0.029$ ] and Time X Group interaction [ $F(1, 43) = 4.792, \eta^2_p = 0.031, p = 0.033$ ]. Post-hoc analysis showed that Ph-T group only statistically improved after intervention ( $p < 0.001$ ). In contrast, the model of Total Reading Speed showed a significant main effect of Time [ $F(1, 43) = 15.347, \eta^2_p = 0.107, p = 0.001$ ], but no significant Time X Group interaction [ $F(1, 43) = 2.941, \eta^2_p = 0.092, p = 0.078$ ].

Writing. A significant main effect of Time [ $F(1, 43) = 7.083, \eta^2_p = 0.049, p = 0.009$ ] was found. In addition, children in Ph-T group improved in writing skills as indicated by a significant Time X Group interaction [ $F(1, 43) = 5.023, \eta^2_p = 0.032, p = 0.025$ ] and by subsequent post-hoc comparisons (respectively,  $p = 0.002$  and  $p = 0.042$ ).

Comprehension. No main effects or interactions were significant, indicating the absence of an effect of any of the training regimen applied.

Phonological awareness. A significant main effect of Time [ $F(1, 43) = 5.392, \eta^2_p = 0.035, p = 0.027$ ], but no significant interactions, were found, indicating a general improvement in the presence of intervention but a lack of effect of the specific treatment.

#### **EXECUTIVE FUNCTIONS**

A Repeated measures MANCOVA was conducted to test treatment effect on EFs. The results showed there was a significant difference between CT and Ph-T groups on the executive functioning over time [Time X Group  $F(1, 43) = 8.434, \eta^2_p = 0.352, p < 0.001$ ]. Specifically, univariate tests indicated there was a significant effect of the CT on specific EFs tests: Bell [ $F(1, 43) = 7.141, \eta^2_p = 0.276, p < 0.001$ ], Digit span [ $F(1, 43) = 3.434, \eta^2_p = 0.072, p = 0.028$ ], Visuo-spatial span [ $F(1, 43) = 2.521, \eta^2_p = 0.053, p = 0.035$ ]. Conversely, no significant differences (Time X Group interaction) between the two groups were found in others EFs tests.

### ***4.2. Part B (T1-T3)***

After the last 12 hours of Phonological-based treatment, we analysed the differential effects of the two interventions taking into considerations the baseline (T1) and the last post-test assessment (T3).

## LITERACY SKILLS

Reading. After additional 12 hours of intervention, only a significant main effect of Time [ $F(1, 43) = 23.325, \eta^2_p = 0.178, p < 0.001$ ] was found for Total Reading Accuracy, highlighting gains in both groups, but no significant differences between treatments. By contrast, a significant Time effect and Time X Group interaction for the the Total Reading Speed (Time:  $F(1, 43) = 17.624, \eta^2_p = 0.152, p < 0.001$ ; Interaction:  $F(1, 43) = 13.209, \eta^2_p = 0.148, p < 0.001$ ). Post-hoc comparisons showed that that children in the Integrated group (CT + Ph-T) improved in reading fluency ( $p < 0.001$ ), while the pure Phonological-based group (Ph-T + Ph-T) did not ( $p= 0.083$ ).

Writing. Analogously, we found a significant main effect of Time [ $F(1, 43) = 9.137, \eta^2_p = 0.083, p = 0.007$ ] and a Time X Group interaction [ $F(1, 43) = 7.144, \eta^2_p = 0.078, p = 0.021$ ]. Post-hoc analyses indicated that Integrated group significantly improved in writing, while the pure Phonological-based group did not (respectively:  $p = 0.006, p=0.074$ ). Interestingly, a significant interaction Time X Age  $F(1,43) = 11.328, \eta^2_p = 0.046, p = 0.001$  was also significant, indicating that younger children showed higher gains in writing word and non-words, compared to older ones.

Comprehension. Taking into consideration the performances in the comprehension task, a trend for the Time X Group interaction was found [ $F(1, 43) = 4.026, \eta^2_p = 0.029, p = 0.061$ ] suggesting slightly higher – but not significant - improvements for the Integrated treatment compared to the Phonological-based.

Phonological awareness. Results showed a significant main effect of Time [ $F(1, 47) = 6.897, \eta^2_p = 0.046, p = 0.029$ ], but no significant interaction, indicating no significant differences between treatments.

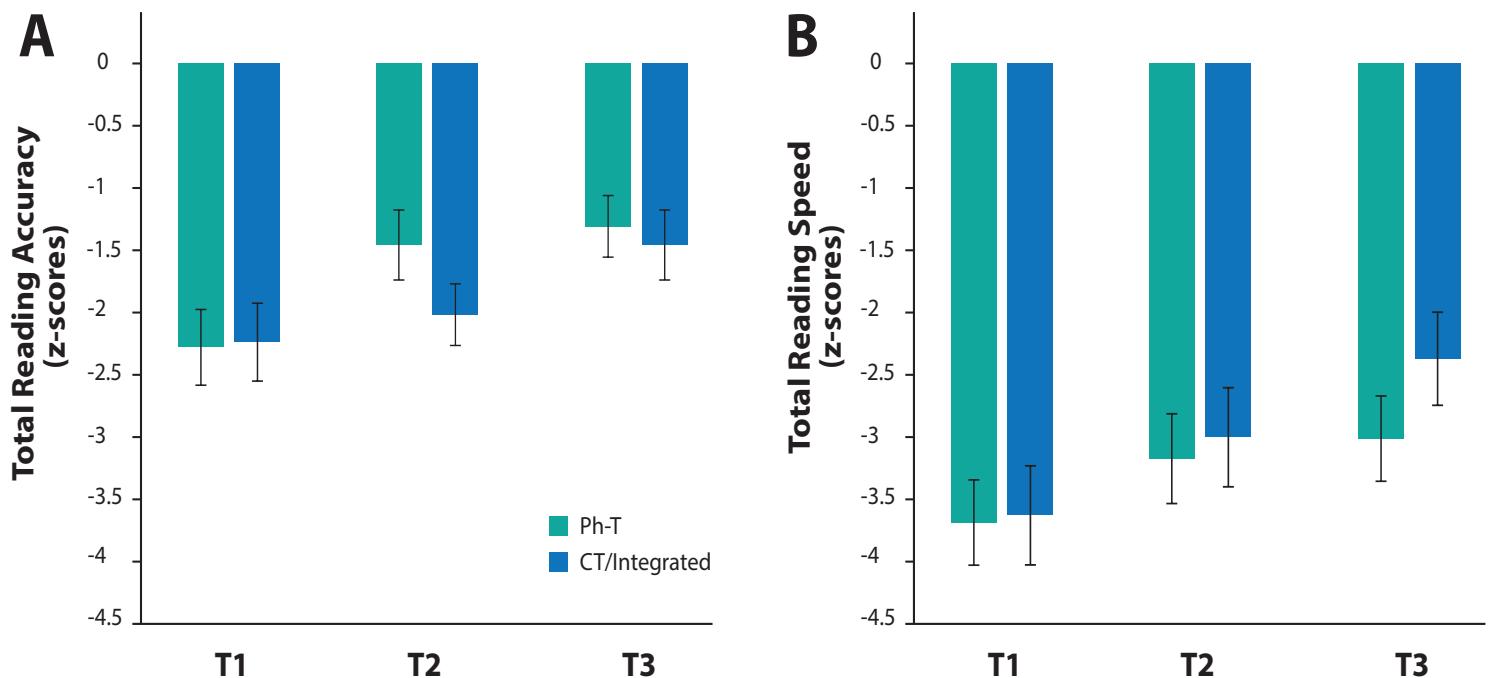
## EXECUTIVE FUNCTIONS

The results of the repeated-measure MANCOVA confirmed that children of Integrated group retained their reading advantage in the overall executive functioning as suggested by a significant Time X Group Interaction,  $F(1, 47) = 9.174, \eta^2_p = 0.371, p < 0.001$ . Interestingly, univariate tests revealed significant intervention effects not only for the Bell task [ $F(1, 43) = 15.372, \eta^2_p = 0.371, p < 0.001$ ], Digit span [ $F(1, 43) = 8.518, \eta^2_p = 0.547, p < 0.001$ ] and Visuo-spatial span [ $F(1, 43) = 7.554, \eta^2_p = 0.251, p = 0.007$ ], but also for the Verbal Fluency task [ $F(1, 43) = 4.613, \eta^2_p = 0.312, p = 0.033$ ]. On the contrary, only a main effect of Time was showed for the TAU and Frogs tasks [respectively,  $F(1, 43) = 11.941, \eta^2_p = 0.321, p <$

0.001;  $F(1, 43) = 8.903$ ,  $\eta^2_p = 0.352$ ,  $p < 0.001$ ], whereas no significant effects were found for the Tower of London (TOL) [Time:  $F (1, 43) = 0.068$ ,  $\eta^2_p = 0.001$  ;  $p = 0.421$ ].

The treatment-related changes are summarised in Table 7 and Table 8 and are represented in Figure 14.

**Figure 14** | The Total Reading Accuracy is showed before (T1), after 12 hours (T2) and at the end of the 24 hours of treatment (T3) in the Phonological-based and CT/Integrated group (**A**). Training effects (T1-T2-T3) of the Phonological-based group (Ph-T), and CT/Integrated group on Total Reading Speed (**B**). The two groups did not differ at the baseline in all the reading measurements. Error bars represent the standard errors.



**Table 7** | Summary of the mean values and standard deviations of the literacy performances at the three assessment time points for the two groups. In addition, the training effects have been reported. It has to be noted that, when all the time points are considered (T1, T2 and T3), the CT group refers to the Integrated treatment (Ct + Ph-T) while the Ph-T to the pure Phonological-based treatment (Ph-T + Ph-T).

<sup>a</sup> Time (T1–T2) X Group interaction.

<sup>b</sup> Time (T1–T3) X Group interaction.

<sup>c</sup> Test significance. \* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$ .

<sup>d</sup> Effect sizes of interaction Time X Group expressed as partial eta squared.

LITERACY SKILLS		T1	T2	F-score <sup>a,c</sup>	ES <sup>d</sup>	T3	F-score <sup>b,c</sup>	ES <sup>d</sup>
Total Reading Accuracy	CT	-2.30 (1.53)	-2.10 (1.12)			-1.47 (1.05)		
	Ph-	-2.32 (1.49)	-1.46 (1.33)	4.79*	0.31	-1.37 (1.37)	0.12	0.01
	T							
Total Reading Speed	CT	-3.61 (1.97)	-3.04 (1.96)			-2.41 (1.83)		
	Ph-	-3.64 (1.66)	-3.18 (1.73)	2.94	0.09	-3.05 (1.70)	13.21***	0.15
	T							
Total Writing	CT	-2.01 (2.2)		-1.89 (1.48)		-1.61 (1.38)		
	Ph-	-2.05 (2.32)		-1.80 (1.67)	5.02*	0.03 (1.99)	7.14*	0.08
	T							
Comprehension	CT	0.08 (0.94)	0.11 (0.94)			0.26 (0.86)		
	Ph-	0.17 (0.73)	0.16 (0.84)		0.07	0.01	4.03*	0.03
	T					0.18 (1.01)		
Phonological Awareness	CT	4.38 (1.06)	5.49 (0.98)			6.15 (1.32)		
	Ph-				0.05	0.01	0.09	0.01
	T	4.52 (1.01)	5.83 (1.36)			6.43 (1.29)		

**Table 8** | Summary of the mean values and standard deviations of the executive functions performance at the three assessment time points for the two groups. In addition, the training effects have been reported. It has to be noted that, when all the time points are considered (T1, T2 and T3), the CT group refers to the Integrated treatment (Ct + Ph-T) while the Ph-T to the pure Phonological-based treatment (Ph-T + Ph-T).

<sup>a</sup> Time (T1–T2) X Group interaction.

<sup>b</sup> Time (T1–T3) X Group interaction.

<sup>c</sup> Time X Group Interaction of the MANCOVA model.

<sup>d</sup> Test significance. \* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$ .

<sup>e</sup> Effect sizes of interaction Time X Group expressed as partial eta squared.

EXECUTIVE FUNCTIONS		T1	T2	F-score <sup>a,d</sup>		ES <sup>e</sup> <sup>c</sup>	T3	F- score <sup>b,d</sup>		ES <sup>e</sup> <sup>c</sup>
				8.434 <sup>c***</sup>	0.35 <sup>c</sup>			9.17 <sup>c ***</sup>		
Bell task (Visuo-spatial attention)	CT	-0.86 (1.09)	-0.17 (1.08)			0.32 (0.97)				
	Ph-	-0.75 (1.17)	-0.71 (1.11)	7.14***	0.28	-0.53 (1.34)		15.37***	0.37	
	T									
TAU (Auditory attention)	CT	-0.92 (1.30)	-0.41 (1.32)			-0.29 (0.69)				
	Ph-	-0.98 (1.22)	-0.57 (1.39)	1.22	0.06	-0.18 (1.54)		0.07	0.003	
	T									
Frogs (Auditory attention + inhibition)	CT	-0.28 (0.79)	-0.16 (0.77)			0.48 (0.45)				
	Ph-	-0.36 (0.84)	-0.23 (0.96)	0.94	0.05	0.23 (1.05)		0.09	0.005	
	T									
Digit span (verbal WM)	CT	-0.94 (0.86)	-0.35 (0.74)			-0.32 (0.82)				
	Ph-	-0.85 (1.03)	-0.79 (0.86)	3.43*	0.07	-0.77 (0.94)		8.52***	0.55	
	T									
Visuo-spatial span (visuo-spatial WM)	CT	-0.34 (0.78)	0.03 (0.56)			0.04 (0.71)				
	Ph-	-0.26 (0.91)	-0.09 (0.97)	2.51*	0.05	-0.06 (1.09)		7.55**	0.25	
	T									
Verbal Fluency	CT	-0.46 (1.01)	-0.26 (1.07)			0.23 (1.12)				
	Ph-	-0.39 (1.05)	-0.31 (1.14)	3.01	0.11	0.07 (1.48)		4.63*	0.31	
	T									
TOL (Planning)	CT	0.01 (1.30)	0.24 (1.08)			0.37 (0.94)				
	Ph-	0.06 (1.12)	0.21 (1.04)	0.09	0.01	0.33 (1.17)		0.07	0.001	
	T									

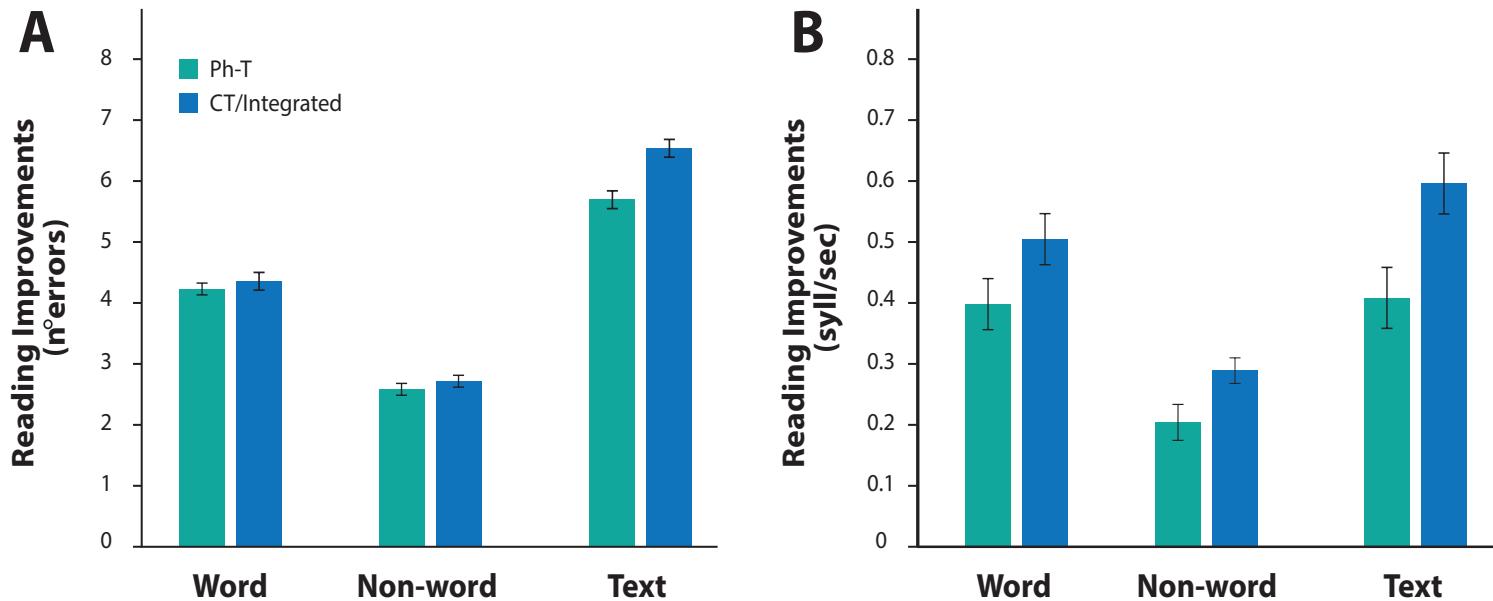
#### 4.3. Clinical efficacy comparison

In order to take into account the efficacy of the two treatments (Integrated versus pure Phonological-based) from a clinical point of view, we compared the improvements in fluency and accuracy in word, non-word and text reading for both treated groups. For Accuracy, we took into consideration the improvements between the baseline and the final evaluation (time

points T1-T3) in terms of reductions of the errors made by the children while reading. In the accuracy of text reading, the Integrated group (Accuracy Delta:  $M = 6.55$ ,  $SD = 0.64$ ) showed a larger gain compared to the pure Phonological-based group (Accuracy Delta:  $M = 5.77$ ,  $SD = 0.71$ ). Consistently, the improvements in terms of number of reading errors in word and non-word reading tasks were higher for the Integrated group (Accuracy Delta: respectively,  $M = 4.36$ ,  $SD = 0.67$ ;  $M = 2.72$ ,  $SD = 0.46$ ) than the Phonological-based group (Accuracy Delta: respectively,  $M = 4.23$ ,  $SD = 0.44$ ;  $M = 2.61$ ,  $SD = 0.37$ ). For Fluency, we compared the treatment outcomes in terms of syllables per seconds: the Integrated group showed bigger improvements than the pure Phonological-based group in the reading speed of word (Integrated:  $M = 0.51$ ,  $SD = 0.19$ ; Ph-T:  $M = 0.39$ ,  $SD = 0.22$ ), non-word (Integrated:  $M = 0.28$ ,  $SD = 0.14$ ; Ph-T:  $M = 0.21$ ,  $SD = 0.08$ ) and text (Integrated:  $M = 0.59$ ,  $SD = 0.27$ ; Ph-T:  $M = 0.41$ ,  $SD = 0.24$ ).

Furthermore, it was important to compare these gains to the clinical efficacy criteria (expected improvements without specialist treatments in each year): 0.3 syll/sec (word an text reading fluency); 0.15 syll/sec (non-word reading fluency); reduction of 50% of the errors (reading accuracy). Reading improvements (T1-T3) both for Accuracy (number of errors) and Fluency (Syllables per Second) of the two treatments are presented in Figure 15. In this study, children of both groups, after only 24 hours of treatment, managed to reach the level of clinical significance, improving more than what expected in children with dyslexia without treatment for one year. Only the Integrated group was close to reaching the clinical efficacy criterion for accuracy with a reduction of the text reading errors (in average) of 6.55 (T1: 14.93; T3: 8.38). Although it is important to specify that this clinical criterion is usually meant to apply to more extended treatments, which last around one year. Moreover, the central defining feature of the reading disorder in the Italian language, as well as other languages with shallow orthography, is the lack of reading fluency and, therefore, the more important results are those related to word, non-word and text reading speed. Specifically, 88% of children in the Integrated group improved in reading speed and, of those, 68% had an improvement higher than the clinical efficacy criterion. Furthermore, fifty-nine per cent of the Integrated participants improved in accuracy, whereas 51% improved both in speed and in accuracy. On the other hand, only 31% of the children that underwent the pure Phonological-based treatment improved in reading speed and, of those, 14% could be considered clinically improved. Approximately, 49% of these children had an improvement in accuracy, while only 29% of the Phonological-based participants demonstrated an improvement both in speed and accuracy.

**Figure 15** Improvements in Accuracy (A), expressed in the number of errors, and in Fluency (B), expressed in syll/sec, reached by the two treatments in word, non-word and text reading. Error bars represent the SE.



#### 4.4. The link between improvements in Reading and Executive Functions

Both the two-step fixed-entry regressions (Total Reading Accuracy and Total Reading Speed delta scores) were found significant. Specifically, the level of the reading variable at T1, entered last, accounted for a significant quote of variance for the amount of gain in Total Reading Accuracy ( $\beta = -.282$ ,  $F(1, 47) = 4.02$ ,  $R^2 = 0.189$ ,  $p = .031$ ) and Total Reading Fluency ( $\beta = .329$ ,  $F(1, 47) = 5.78$ ,  $R^2 = 0.283$ ,  $p = .019$ ). These results seem to suggest the children with initially poorest reading skills (both accuracy and speed) are those who gain the most at the end of the treatments. Therefore, higher reading improvement might be associated with low initial reading abilities.

Two further stepwise regressions were computed in order to explore the link between improvements (T1-T3) in Reading and Executive Functions. Only the improvements in the Bell's task and the Frogs test significantly accounted for a significant quote of the variance of Total Reading accuracy changes (13.9%,  $R^2 = 0.139$ ,  $p < 0.001$ ). In the second regression model, which took into consideration the improvements in Total Reading Fluency, only the gains in the Bell's task and the Digit span accounted for 19.6% of the variance ( $R^2 = 0.196$ ;  $p < 0.001$ ) in Total Reading Speed changes. These results support the idea of involvement of the EFs performance in the prediction of the improvements in speed and accuracy in reading.

## **5. Discussion**

Although the neurocognitive causes of Developmental Dyslexia are still debated, researchers agree that the main challenge is the treatment, that is, how to get children with DD to read more accurately words in less time. The most common approach has been to devise sophisticated remediation programs that train sub-skills of reading, especially phonemic awareness skills. Despite somewhat successful, the improvements in these sub-skills do not automatically transfer in better reading abilities. Even though extensive research has been carried out, factors leading to treatment outcomes remain speculative. Considerable effort has been spent in the last decades in order to develop an evidence-based framework for the remediation of dyslexic reading, but, still, we are in the need for “evidence to guide the more fine-grained selection of specific interventions for individual struggling readers at all ages and at all levels of reading ability” (Shaywitz, Morris, & Shaywitz, 2008, p. 466).

In this regard, the primary aim of this paper was to take a step forward in identifying effective treatments for children with DD and with different reading and neuropsychological abilities. Children with dyslexia who see their peers reading and making progress may feel that they have underachieved. As they move through the school system, problems can get worse as reading becomes more critical. As a result, their self-esteem suffers. Frequently dyslexia not only affects students' academic achievements, but it is also associated with many negative life outcomes. For these reasons, it is important to quickly start effective remediation programs, keeping in mind the complex aetiology of this disorder and taking into account the most important factors that can influence the treatments outcomes. With this aim in mind, first we compared two treatments addressing different skills: the Cognitive Training program was build to train several executive functions, mainly working memory and attention; the Phonological-based treatment consisted of a systematic and exhaustive training of phoneme awareness and other reading-related skills, as well as the rapid reading of progressively larger linguistic units. Assessment upon the first 12 hours of Cognitive Training, indicated statistically significant improvements in several EFs (visuo-spatial attention, verbal and visuo-spatial memory), but not in literacy skills. The Phonological-based Treatment had the highest observed effects in reading accuracy and writing. This result is in line with the RC-DSA-2011: the first aim of the intervention is to achieve an adequate level of reading accuracy in order to reach, then, the ultimate goal of fluent reading, which for Italian children with dyslexia often remains the principal problem.

In the second part of the study, we investigate whether an Integrated treatment (EFs training + Phonological-based) has a larger remedial effect on reading skills in children with DD

compared to the pure Phonological-based treatment. In general, the treatment effects can be regarded as satisfactory for both interventions, even though the Integrated group obtained the most positive effects. Despite the short duration of the treatment program (a total of 24 hours), the integration of the Cognitive Training and the Phonological-based treatment resulted in marked gains in visuospatial attention, working memory and verbal fluency at post-test (T3), while no significant improvements in EFs were observed in the pure Phonological-based Treatment. Importantly, these gains generalised to untrained reading abilities, such as writing and reading accuracy and speed. This is particularly important, since, in the context of transparent orthographies, the core symptom of students with dyslexia is the lack of fluent reading (Tressoldi et al., 2001; Tressoldi & Vio, 2008; Zoccolotti, De Luca, Marinelli, & Spinelli, 2014; Zoccolotti et al., 1999). Without a specific and systematic intervention, the expected increment per grade of these students is around half the increment observed in typically reading children. In most cases, children who suffer from this disorder not only struggle to identify fluently individual words, but also the speed with which they move through a connected text is extremely slow and laborious. An effortless, flowing reading is at the base of other higher-level components such as text comprehension (Fuchs, Fuchs, Hosp, & Jenkins, 2001; Jenkins, Fuchs, Van den Broek, Espin, & Deno, 2003; Klauda & Guthrie, 2008; Raghbar, Barnes, & Hecht, 2010). Indeed, a skilful reader attains proficient comprehension focusing his/her attention on the content, rather than to word decoding or to guess words from the context in which they are contained. Therefore, the automaticity with which a reader is able to decode words is almost as crucial as word reading accuracy, constituting a fundamental goal for effective remediation of dyslexia. In this study, the treatment, which combined a computerised cognitive training of EFs and a Phonological-based intervention (i.e. Integrated treatment), consistently improved several components of reading: accuracy, fluency and comprehension. These results are substantiated by the comparison between the reading gains after 24 hours of treatment and the expected gains in one year of spontaneous reading development. Only the improvements obtained after the Integrated treatment were larger than what expected for word (0.30 syll/sec), non-word (0.15 syll/sec) and text (0.30 syll/sec) reading after one year of regular education without specific and systematic intervention. The gains in reading fluency can be fully appreciated by considering that the Integrated training improved not only non-word reading, which represents the sub-lexical route functioning, but also word text reading, which also represents the ability to rapidly recognize words via the lexical route (Coltheart, 1981; Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001). Non-words have no representation in the lexicon and, thus,

need to be read using the grapheme-to-phoneme correspondence rules; instead, well-learned words, that have a visual representation in the lexicon, can be read directly. In both cases, words are not read as wholes, but are rather processed as letters or larger units in a sequence or in parallel (Dehaene et al., 2010; Pelli & Tillman, 2007). In this regards, Vidyasagar and Pammer (2010) proposed that: “attentional mechanisms controlled by the dorsal visual stream help in serial scanning of letters and any deficits in this process will cause a cascade of effects, including impairments in visual processing of graphemes, their translation into phonemes and the development of phonemic awareness” (p.57). In this study, results of the regression analyses seem to confirm a multifactorial view of reading, which is a complex process supported by a multitude of cognitive functions (Menghini et al., 2010; Moll et al., 2014; Peterson & Pennington, 2012; Varvara, Varuzza, Sorrentino, Vicari, & Menghini, 2014). Improvements in visuospatial attention and verbal memory appeared to go with improvements of reading speed, while improvements in visuospatial, auditory attention and response inhibition seem to predict improvements in reading accuracy. Hence, beyond phonological awareness, other cognitive processes, such as EFs, are needed to account for all the components of reading. Therefore, impairments in the functioning of other cognitive areas, particularly the ones related to attention, may play a causal role in reading difficulties of individuals with dyslexia (Franceschini, Gori, Ruffino, Pedrolli, & Facoetti, 2012; Gori, Seitz, Ronconi, Franceschini, & Facoetti, 2016b; Shaywitz & Shaywitz, 2008). The recognition of the role of other cognitive factors in reading does not negate the role of the phonological deficit in the aetiology of this disorder, but rather provides the opportunity to develop and validate innovative intervention programs. In this regard, the studies of Franceschini and collaborators (2013, 2017b, 2018) gave promising indications as to the efficacy of unconventional training tools such as action video games in improving reading speed of individuals with dyslexia. At this point, progresses in this field offer the hope of providing evidence-based interventions that are tailored to the specific cognitive and reading profile of each student with dyslexia. For this reasons, researchers now need to focus on disentangling how to personalize programs, also specifying the type of implementation required (i.e. clinic-based versus home-based; computerised versus non-computerized). We hypothesised that the Integrated treatment was especially suited since the traditional phonological-based intervention was preceded by a daily short-lasting intervention (30 minutes per day). In this way, we were able to work intensively and to develop a therapeutic alliance with the dyslexic children without frustrating their reading difficulties and, thus, avoiding the possible initial refuse of the clinic-based treatment.

### ***5.1. Limitations and Further Research***

Although the results of the study give clear indications on the necessity of individualising treatment programs on the basis of the specific cognitive profile of the child, several limitations of the study should be taken into consideration. Firstly, we compared two treatments that differed in settings: the Cognitive Training was a home-based activity, whereas the Phonological-based treatment was administered entirely at the clinic. Therefore, these differences should be the object of further evaluations in order to clarify the underlying processes of home-based computerised training in comparison with a clinic-based non-computerized treatment. Secondly, in the present study, we did not include a passive control group for ethical reasons. However, the use of clinical efficacy criteria based on the expected achievement of children with dyslexia per grade gave firm indications of the efficacy of both treatments. In addition, presently there is no way of clearly establishing whether the effects observed at T3 are delayed effects of the first part of the intervention versus immediate effects of the second part of the intervention. Furthermore, the results regard only efficacy in the short-term. Given the promising results of the study, especially of the Integrated treatment, follow-up testing is required in order to confirm the long-term validity and its generalisation to other important aspects of life such as the academic functioning. Thus, a follow-up will be planned in order to understand whether reading interventions result in long-term gains and individual features will be measured to investigate how they relate to intervention outcomes. The main challenge for the remediation of this disorder is not only to find the most effective remediation programs, but also to precisely select a personalized program for each child with dyslexia.

## CHAPTER 4

### SKIES OF MANAWAK: A VIDEOGAME FOR COGNITIVE TRAINING OF EXECUTIVE FUNCTIONS

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*"This is the real secret of life — to be completely engaged  
with what you are doing in the here and now.  
And instead of calling it work, realize it is play."*

*~ A. Watts*

#### 1. Introduction

In the last two decades the literature on cognitive training has grown considerably, articulated on the concept of brain plasticity (i.e., functional and structural changes). Neuroplasticity, which allows the central nervous system to reorganize itself by forming new neural connections in response to new situations or to modification in the environment, has been shown to take place in several cognitive domains and throughout the lifespan (Bavelier, Green, Pouget, & Schrater, 2012; Fu et al., 2013; Merzenich, Van Vleet, & Nahum, 2014; Gilbert et al., 2009; Merzenich et al., 2001, 2013). The basic brain mechanisms involved in this process (i.e., neurogenesis, pruning and activity-dependent synaptic plasticity) contribute crucially to the brain's ability to acquire new information, modify flexibly to changes in the environment and recover from injuries (Johnston et al., 2009). For example, studies on adults (Bottari et al., 2014) and children (Sadato et al., 2004) with early deafness showed that cortical areas deprived of afferent input, such as auditory cortex, were rewired for processing aspects of visual input.

According to Galvan (2010), cognitive training influences developmental trajectories by modulating the interaction between biological maturation - pre-specified at the genetic level - and learning. Several studies have documented that although any enriching experience consistently enhances performance, direct training is more efficient at positively affecting learning and transfer of skills. Thus, a growing number of research reports have highlighted the possibility that systematic and intensive cognitive training may improve one or more cognitive functions (e.g., Bergman-Nutley & Klingberg, 2014; Dowsett & Livesey, 2000; Holmes et al., 2010; Holmes, Gathercole, & Dunning, 2009; Klingberg et al., 2005; Spencer-

Smith & Klingberg, 2015).

Research on cognitive training has targeted not only typically developing children, but also clinical populations. Some empirical findings indeed suggested that children with learning disabilities might benefit from early intervention programs focusing on cognitive training. For instance, perceptual learning - an improvement of perceptual skills with practice - could selectively reduce the impairment of visual functions (Gori & Facoetti, 2014).

Training programs designed to strengthen executive functions may be particularly relevant for children with impaired or lagging cognitive skills, since it has been demonstrated that these abilities are crucial for a successful adaptation both in and outside school. The enhancement of these skills is fundamental for a harmonious development of the child; for example, a mature behaviour requires the ability to retain and manipulate information in the mind and to adjust the behaviour quickly and flexibly. Therefore, a functioning executive system should entail higher scholastic achievements, greater social skills and, more generally, a better quality of life.

Despite several studies showing promising results for cognitive training, the tools used for these trainings generally present some limitations (Jaeggi et al., 2008; Loosli, Buschkuehl, Perrig, & Jaeggi, 2012).

Firstly, their application is usually limited to one cognitive function, with uncertain generalizability to unrelated tasks. They are, in fact, designed to train single functions, even though the EFs work as a multi-dimensional system and it could be reasonable to include more than one EF in the intervention programs. Of primary importance is, in fact, the positive transfer of training, which is the extent to which trained skills may transfer to real life.

Furthermore, it has to be noted that not all studies included an active control group, thus resulting in potential motivational or placebo effects confounds.

Moreover, cognitive training programs tend to be less enjoyable and stimulating compared to other digital products, such as video games. Traditional training tools are designed for a generic audience and, even if they incorporate some aspect of gamification (e.g., scoring mechanisms), the training aspects have more relevance than the entertainment ones. The added value of video games is its obvious motivating characteristics: the game industry aims at products that are attractive and entertaining, aspects that usually are not central in neurocognitive trainings (Eichenbaum, Bavelier, & Green, 2014). In this regard, the structure of a rehabilitative intervention should consider emotional and motivational aspects. Given the young age of the users, these characteristics of the tool can influence compliance and motivation, increasing resistance to frustration, and therefore play a crucial role in

determining the effectiveness of a treatment, or at least affect the outcome positively (Marotta & Varvara, 2013).

In order to overcome the motivational barrier, the training must consist of challenging and variable activities that require the user to be continuously committed in adapting their own behaviour, while creating a fair sense of competition through appropriate feedbacks. A direction that seems very promising (Green, Li, & Bavelier, 2010) is to design new rehabilitation products that while being based on a deep knowledge of neuropsychological theories also provide an engaging and challenging experience. Franceschini and colleagues (Franceschini et al., 2013, 2017b) highlighted the fact that action video games could improve attention abilities and that this improvement could be generalized to better reading abilities (specifically reading speed) in children with dyslexia in shallow (i.e. Italian) and deep (i.e., English) orthographies. These studies focused exclusively on children with reading-impairment and, thus, to date little is known about attentional – or more in general - EFs training and its transfer to the general school-aged population. However, their studies provided the first evidence of the potential of video games in remediating learning disabilities.

The synergy between a playful and engaging experience and an effective cognitive training makes for a well-supported case for future research and development. Therefore, we decided to develop *Skies of Manawak* (hereafter referred to as SOM) a Sci-Fi video game for cognitive training. SOM was designed as close as possible to classic video games from the entertainment industry. The aim of the present study was to investigate whether a video game, specifically designed for training EFs, might have a positive effect not only on these functions, but also on literacy skills both in typically reading children and in children with dyslexia.

## **2. The project**

The development of the project started in January 2015 as collaboration between the Laboratory of

Observation, Diagnosis and Training (ODFLab) of the Department of Psychology and Cognitive Science and the InterAction Lab of the Department of Information Engineering and Computer Science (DISI), both of the University of Trento. This project required two years of design iterations and was divided into several stages following an approach oriented

to the participation of different stakeholders. Specifically, domain experts (clinical psychologists and cognitive scientists) and players (children from 8 to 13 years old) were involved since the beginning of the project. The former became an integral part of the development team and were involved in all design stages, from conceptualization to development. The latter participated in co-design sessions at several stages of the design process, contributing to the definition of the basic elements of the game, developing the first concepts and providing feedback during the overall development.

Starting from the experiences of other researchers - for example (Iivari & Kinnula, 2016; Khaled & Vasalou, 2014; Moser et al., 2013) - the designers had the chance to extend the participatory game design research, applying the solutions developed by others, and documenting their own experiences and reflections.

**Figure 16**| A screenshot of gameplay.



### 3. Theoretical Framework

Our aim was not only to provide children with an effective training tool, but also with an enjoyable experience of play. Several theoretical and methodological choices have been made accordingly. Video games are complex, multidimensional systems that are structured on various dimensions. Several manifold aspects should be taken into consideration in order to achieve an effective multi-dimensional design process. In this regards, Shell (2014) defined four fundamental dimensions for video game design (Elemental Tetrad). First, the *Story* is represented by the sequence of events and it provides the essential elements for understanding the objectives and keeping the player engaged. Secondly, the *Aesthetics* represent the visual and acoustic contents that the player perceives and, thus, strongly influence the first impressions of the player (Miniukovich & De Angeli, 2014) and facilitate a sustained level of immersion (Bateman, 2015; Dickey, 2015; Frome, 2007). Thirdly, the

*Mechanics* constitute the procedures and rules of the game, defining the ways in which the player is supposed to interact with the system. Finally, the *Technology* regards the whole technological apparatus (e.g., the controller) with which the player interacts.

With few exceptions (e.g., Tetris), a well-designed game can be conceptualized through the use of these four, interrelated, dimensions, which, in turn, can be better specified via one hundred design cards or lenses (Schell, 2014). It has to be noted that these elements are strongly interrelated; this means that each design choice concerning one dimension will necessarily shape constraints on the others. Since video games can be considered as the result of a multi-dimensional design process, for an effective result all these elements should co-exist symbiotically.

Another important contribution to the identification of an adequate theoretical framework for our design process is represented by the work of Hunnicke and colleagues (Hunicke, LeBlanc, & Zubek, 2004). The authors formulated the Mechanics - Dynamics - Aesthetics (MDA) framework. The *Mechanics* are described as the base components of the game, namely the basic elements of the game that, through data structures and algorithms, define the rules. The *Dynamics* embeds the emergent rules of the game. All the run-time behaviors of the mechanics acting on the base of the players' actions should be considered part of this category. The *Aesthetics* refer to the emotional responses of the player. Specifically, Hunnicke and collaborators proposed eight types of Aesthetics, which are presented in Table 9.

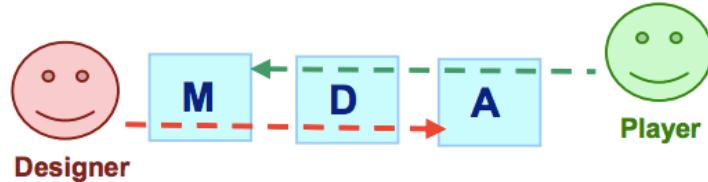
**Table 9|** The taxonomy of Aesthetics (Hunicke et al., 2004).

<b>1. Sensation</b>	<b>5. Fellowship</b>
Game as sense-pleasure	Game as social framework
<b>2. Fantasy</b>	<b>6. Discovery</b>
Game as make-believe	Game as uncharted territory
<b>3. Narrative</b>	<b>7. Expression</b>
Game as drama	Game as self-discovery
<b>4. Challenge</b>	<b>8. Submission</b>
Game as obstacle course	Game as pastime

In addition, the MDA framework focuses on the different roles of the game designers and the players in the design process. While the former specify the mechanics, which give rise to the dynamics, which in turn create the aesthetics, the latter prioritize the aesthetic of the game,

which is caused by the observable dynamics and eventually, operable mechanics. Since they are direct consumers of the product they can provide essential information about their preferences and desires. With this double perspective (Figure 17) the MDA framework facilitates a player-centered thinking in game design.

**Figure 17|** The designer and player each have a different perspective. Source: Hunicke, LeBlanc, & Zubek, 2004



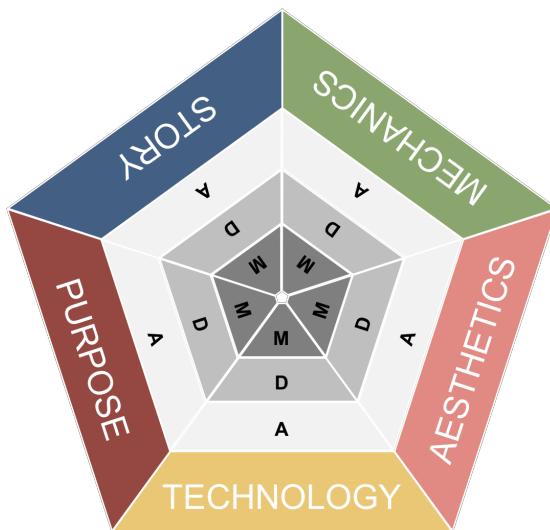
The design expert of the group selected these two frameworks due to the fact that they provide complementary perspectives on game design. The Elemental Tetrad (Shell et al., 2014) focused more on the structure of the game, providing a complete decomposition in its basic elements. Vice versa, in the MDA framework the player experience is crucial. In general, both supported the idea that the involvement of players would positively influence the overall design in terms of the purpose and the gaming characteristics.

However, despite the provision of valuable methodological contributions, the aforementioned works did not take into consideration an important feature of our video game: the *Purpose* (i.e., training of EFs). In our case, in fact, it was fundamental to reach a balance design, reconciling the gaming characteristics with the purpose. In order to fulfil this important goal, the additional involvement of the domain experts (clinical psychologists and cognitive scientists) was fundamental in identifying the cognitive requirements of the training and in organizing all the sessions in which players were involved (e.g., organization and observation of workshops).

In this regard, our work integrated the elemental tetrad (Schell, 2014) with the MDA framework (Hunicke et al., 2004), adding a new dimension related to the *purpose*. The result is the Participatory Elemental Pentad - PEP (Menestrina, 2017), a game design framework oriented to the participation of the stakeholders. The main goal of PEP (Figure 18) is to support participatory practices in game design, for the development of well-balanced video games for a purpose. This new framework aims at defining the potential contribution of the various stakeholders and their level of participation. In order to address the involvement and

contribution of stakeholders, two dimensions defined the participation space. The outer ring represents the five basic elements of games for purpose: story, aesthetics, mechanics, technology and purpose. This ring specifically clarifies the role and the contribution of various stakeholders (on) to the elements of the pentad. The internal rings define at which level the discussion takes place. This can be developed at a high level, more linked to the general gameplay experience (the A of the MDA), or deepened to the basic rules of the game (M).

**Figure 18|** The Participatory Elemental Pentad (PEP). Source: Menestrina, 2017.



#### 4. Design process

The design process consisted of 9 fundamental stages (see Figure 19). The following subsections provide a description of the main stages of the design process in relation with the theoretical framework adopted.

**Figure 19** Main stages of the design process (source: Menestrina et al., 2018). In detail, the stages involving the entire research group (designers and domain experts) are in black, whereas the stages involving designers and players are in white. Only the stages of implementation (in grey) were carried out by the game designers.



#### 4.1. Groundwork

At the beginning of the project, a series of introductory meetings spread over one month and aimed at initiating a transfer of knowledge between designers and domain experts, were carried out. The initial group was composed of eight people including several professional figures. After these initial meetings, the research group was identified: Angela Pasqualotto, clinical psychologist and cognitive scientist; Zeno Menestrina, computer scientist and game developer; Adriano Siesser, visual artist. Several meetings with the research supervisors (prof. Venuti and prof. De Angeli) were then planned.

The groundwork stage was particularly important in identifying and defining set of mini-games that train specific executive functions, whose features make them suitable for a real video game. This common dialogue was essential to define the objectives of the project, thus avoiding misalignments or misunderstandings (De Troyer, Janssens, & Vandebosch, 2013).

At the end of this stage all the parties agreed on the design goals and the activity plan.

#### **4.2. Design Space**

Following the preliminary meetings, the team envisioned a high-level game scenario integrating into the story a set of mini-games that would have addressed the cognitive training requirements. Our team, in fact, aimed at the development of a video game for a purpose where the training becomes part of the design with the same importance of elements such as aesthetics and story. Thus, a set of training tools, used in the clinic for the training of the EFs, were selected, decomposed into their structural components, translated into game mechanics and reassembled as mini-games. An example of this process is represented by the auditory working memory task (*Rekenanagi*). The original paper-and-pencil exercise was based on single letters or numbers. In the example presented in figure 20, the exercise presents a list of numbers of undefined length and the participant is required to repeat the last three elements of the series. In this regard, our team took the decision to avoid any kind of letters and numbers as material for the mini-games. The reason was two-fold: on one hand they would strongly constrain the narrative of the game (e.g., why would a player need to repeat a sequence of letters in the middle of a sci-fi game?); on the other hand, it should be the norm to avoid submitting alphanumeric materials to children with learning disorders. Therefore, numbers were easily translated into mysterious symbols in this and other mini-games.

In conclusion, the main idea at the base of this design stage was the necessity to create a structure that allowed full control over the stimulation induced by the mini-games, to ensure compliance with cognitive requirements while providing an engaging experience. The mini-games, thus, should be considered as plausible for the narrative and not be perceived as disruptive elements. In SOM, the player followed a story that at times required the engagement with mini-games - tapping different sub-domains of cognition (e.g. inhibition, planning, attention, visual and auditory working memory) – fully integrated in the story. Crucially, players travel from one flying island to another with the help of a flying creature (*Raku*). These flying sessions encompass key action video game mechanics suggested to facilitate learning and brain plasticity (Bavelier et al., 2012).

**Figure 20|** The *Rekenanagi* mini-game, an example of translation of a paper-and-pencil exercise into a mini-game.

STRUCTURAL COMPONENTS	MECHANICS	MINI-GAME
<b>ELEMENTS:</b> letters or numbers	<b>1 2 3 ...</b>	
<b>INPUT:</b> the operator construct out loud a list of elements with no pre-defined length	<b>operator:</b> “1 4 2 9 6 1”	
<b>OUTPUT:</b> the subject must keep track of the list and repeat the last three elements	<b>subject:</b> “9 6 1”	

#### 4.3. Game Ideation

In May 2015, 60 children aged between 8 and 13 years old were involved in 12 workshops. The main aim of these workshops was to provide the designers with suggestions on features desired by possible players in a video game.

Each workshop lasted approximately two hours and was hosted either at the Department of Computer Science or at the ODFLab. Typically reading and dyslexic children worked in groups composed by three to five participants and at least one researcher actively interacting with them. The choice of including both children with and without learning disorders was made because the focus of this activity was on the gaming component, not on the training purpose of the application.

A summary of the groups' characteristics alongside the location of the workshop is reported in Table 10.

**Table 10|** Summary of the participant's information of the game ideation workshops.

date	participants	group	age	location
6-May-2015	4 (4 M)	1	11	university
11-May-2015	9 (6M, 3 F)	2	8-10	clinic

12-May-2015	24 (11 M, 13 F)	6	9-10	university
13-May-2015	11 (8 M, 3 F)	3	11-13	clinic
18-May-2015	12 (8 M, 4 F)	3	11-13	clinic

The procedure was inspired by the work of Moser and colleagues (Moser, Chisik, & Tschelegi, 2014), adapted to our design space, and refined in a pilot session (N=4). The workshop was structured as following: *introductory presentation*, *design document*, *low-fidelity prototyping* and *videotaping*.

An *Introductory presentation* (10 minutes) opened the session presenting the members of the development team and the schedule of activities. It has to be noted that researchers deliberately omitted any information about cognitive training since the interest was entirely in working with the children on the gaming dimension.

The first phase consisted of the creation of a *design document* (40 minutes), sketching the conception and conceptualization of the game. The children were asked to reflect on three game elements (i.e., the character, the obstacles and the final goal) and to individually answer the following questions:

- *who?* the protagonist/s of the story;
- *where?* the spatial setting of the events;
- *when?* the temporal setting of the events;
- *what?* will the protagonist/s run into someone or something?  
(e.g., enemies and companions);
- *why?* the motive of the game events.

After the individual phase of ideas collection, a collective discussion followed. The researcher mediated the discussion and reported the main results on a blank poster where it was decomposed following the framing questions (Figure 21 reports the English translation of some ideas). The mediator then turned the blank poster and repeated the activity with the whole group, aiming at consolidating a single proposal and resolving any dysfunctional conflict about the game idea.

**Figure 21** An example of design board filled by a group of four participants. The translation is superimposed on the original.

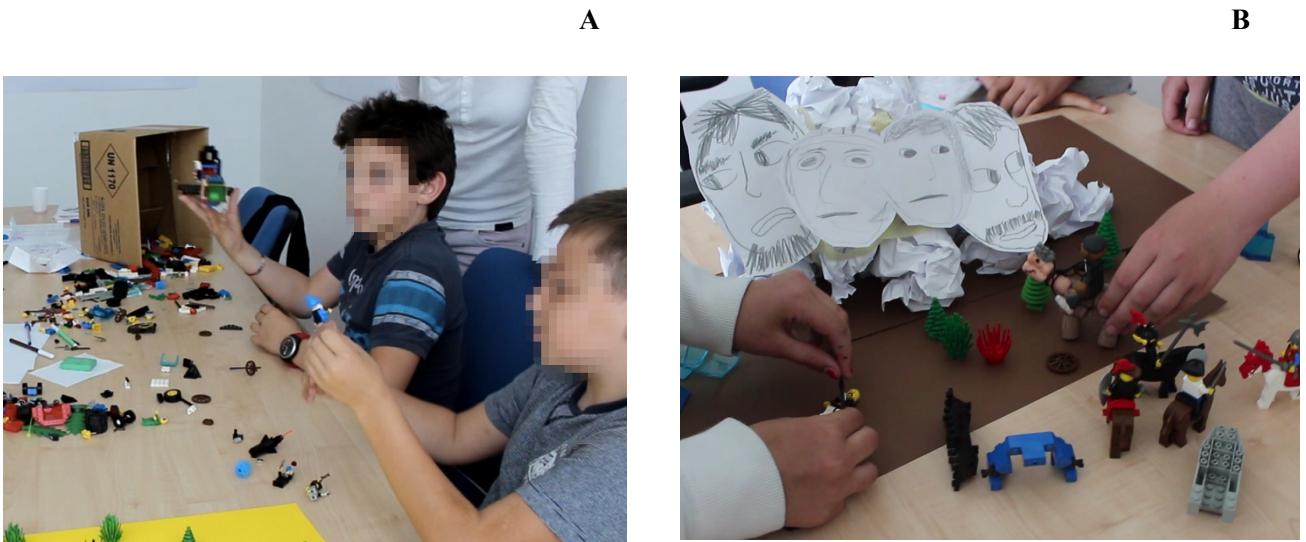
	CHARACTER	OBSTACLES	FINAL GOAL			
	WHO?	WHERE?	WHEN?	D?	WHAT?	WHY?
1	an historian, gems digger	an abandoned hospital	21h century	foes: lunatics friends: humans	find the gem and obtain the golden machine gun	
2	a merchant	the universe, everywhere	3000 A.C.	foes: commanders of the enemy planets friends: aliens, humans	find the pieces to build the time machine	
3	a scientist	a castle	in the '90s	foes: enemy's soldiers friends: no one	escape taking the potion of unlimited power	
4	a mutant human	an abandoned metropolis	the future	foes: aliens friends: no one	run away from the aliens and find the pieces to build the time machine	

A *low-fidelity prototyping* phase (30 minutes) followed: each group received a set of material (modelling clay, Lego®, cardboard and PlayMais®) and was invited to recreate a scene showing their game proposal. After a short break (10 minutes), they moved to *videotaping* (30 minutes), divided into a preparation phase, where the groups tried their scene, and into the final shoot, where each group simulated a few minutes of gameplay. See figure 22, for some examples.

These ideational workshops were useful in highlighting the patterns in the design choices of the participants. Data were classified according to a sub-set of the *lenses of game design* (Schell, 2014) and several clusters of themes were identified.

A common theme to many proposals was a story that incorporated elements of the hero's journey: a hero following a path of personal growth, which will eventually lead to a great victory. The environments had in most of the cases a fantastic component, and very often they were placed in a time frame positioned in a distant future. Another interesting element was the presence of allies or companions. Other common game components were crafting systems, upgrades, and rewards. The proposals were mainly based on the experience (the A of the MDA) of this type of mechanics, with almost no details on their actual implementation; a more in-depth discussion on the topic (in terms of M and D of the MDA) would have required an exchange of knowledge with designers.

**Figure 22|** Examples of Low-fidelity prototyping (A) and Videotaping (B)



#### 4.4. First Design

During the entire summer, the team worked on a demo of the game that was able to combine the suggestions received from the children with the results of the game concepts phase. The aim was to develop a test version of the game that showed the aesthetics, introduced the narrative and integrated some exercises. At the end of September, the team conclude a first demo of 30 minutes gameplay containing: an introduction to the mechanics of the game (e.g., commands); three scenarios, with different interactions and slight variations on the aesthetics; the integration of four cognitive exercises, three based on the translation of paper-and-pencil EF exercises and one based on theories about the cognitive benefits of action videogames (Bavelier et al. 2012; Eichenbaum, Bavelier, & Green, 2014; Franceschini et al., 2013, 2017b; Franceschini & Bertoni, 2018, Green et al., 2010).

#### 4.5. Formative Evaluation

In October 2015, in conjunction with a public event organized at regional level by our research group for the Dyslexia Awareness Week, there was a second involvement of the players in the design process. This stage had an intermediate purpose: verifying whether the current design of the game was appreciated by the players, and collecting feedbacks on possible changes and improvements. The main emphasis of the evaluation was on the ludic aspect of the game and its potential to engage the users.

The Dyslexia Awareness Week included a set of activities to be held in five schools across different cities of the region. From each school, two to four classes were invited to try SOM.

Each session lasted about an hour and was divided into two parts: a gameplay activity followed by a focus group. The two activities were held, respectively, in the computer labs of the schools and the classrooms. At least two researchers supported by at least one teacher, were present at every activity. A summary of the sessions is shown in Table 11.

**Table 11|** Participant's information of the evaluation at the primary schools.

date	participants	classes	age	location
05-Sept-2015	87 (41 M, 46 F)	4	8-11	Rovereto
06-Sept-2015	43 (19 M, 24 F)	2	8-11	Riva del Garda
07-Sept-2015	45 (22 M, 23 F)	2	8-11	Cles
08-Sept-2015	44 (26 M, 18 F)	2	8-11	Levico Terme
09-Sept-2015	39 (19 M, 20 F)	2	8-11	Trento

The evaluation of the gameplay was based on a modified version of the extended Short Feedback Questionnaire (eSFQ) created by Moser and colleagues for the rapid assessment of game experiences (Moser, Fuchsberger, & Tscheligi, 2012).

One-hour focus group in the classroom followed the administration of the questionnaire. The goal was to obtain a deeper understanding of the opinion of the players and, more importantly, to give them the opportunity to express their approval/disapproval about the game and give their suggestions about modifications and improvements on the design. The focus group started with the presentation of a poster divided according to four questions:

- *what did you dislike?*
- *what did you like?*
- *what would you change?*
- *what would you add?*

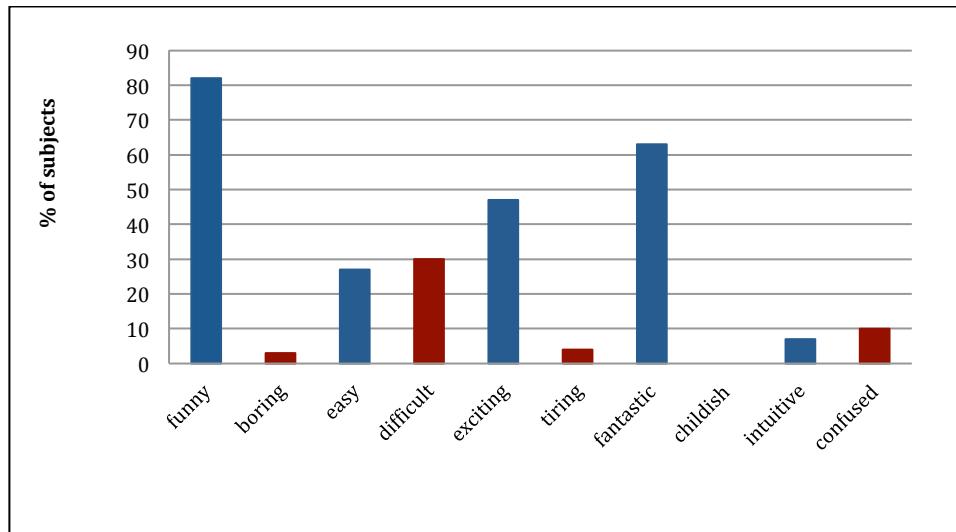
Each participant had a block of Post-it® notes: for approximately ten minutes the participants could individually note their comments and stick it to the poster and, then, in the last ten minutes the entire class was involved in the discussion about the notes.

Overall, observational data showed a strong engagement with the game. Children were often very immersed in playing. Questionnaire data collected in the school supported this general

impression. The majority of the comments confirmed themes emerged during the game ideation workshops. Many players suggested action elements, such as large quantities of weapons and enemies with diverse aesthetics and skills. They also expressed a desire for greater customization of the game, with a wider range of characters (in the demo limited to two).

Figure 23 summarizes the answers for the multiple choice question "how would you define this game?". The only dimension receiving criticism, by a relevant percentage of players (30%) was difficulty: when crossing this information with the comments from the focus group, it emerged that difficulty was indeed a critical dimension with some participants considering the game too difficult, while others considering it too simple. This dichotomy can be attributed to the lack of an automatic balance of the difficulty that could not be implemented in the demo due to the brevity of the game session. A putative explanation – derived from the observation of the playing behaviour – is that some players do not read the tutorials and, thus, had difficulties in advancing with the game (requiring the intervention of the researchers).

**Figure 23|** Mean answers (expressed in percentages) at one of the questions: "how would you define this game?"



#### 4.6 Re-design

The re-design phase aimed at simplify the tutorials (e.g., reducing the quantity of written text and adding an audible version obtained via a text-to-speech software). In addition, attention was given to increase the reliability of game balancing algorithms in order to personalize the difficulty of the mini-games to the individual and highly heterogeneous skills of participants. Moreover, the upgrade system, until then only sketched in the design document and

completely absent in the demo, was discussed within the team and included as a basic component of the game.

However, not all suggestions were directly implemented in the design. For example, many players requested more masculine and feminine characters, stereotyped according to the idea of the “muscular hero” and the “princess”. This proposal was discarded, as it clashed with our gender stereotyping concern, even though the researchers provided a larger roster of characters for both sexes. Similarly, other requests concerning violent content were strongly moderated according to the general values of the designers. In fact, in the final design of the game some aerial battles were introduced; yet all the enemies were non-organic entities, reducing the violent act of killing to the dismantlement of lifeless objects.

**Figure 24|** Three characters of the game. The one on the left is a female.



#### **4.7 Expert Evaluation**

In June 2016 the development team completed six mini-games out of the ten planned for the first release of the game. At this point, a sample of clinical therapists and cognitive scientists conducted an expert-based evaluation to verify that all mini-games fulfilled the cognitive requirements.

The experts, who had no previous knowledge of the game, played individually the six mini-games (approximately thirty minutes). At the beginning of the evaluation session, the researchers provided a questionnaire containing six identical questions, one for each exercise. The question asked to identify which executive functions were trained, presenting a multiple-selection list and the possibility to add an answer. The experts had to answer the question at the end of each mini-game, identifying the trained EFs and providing further comments. At the end of the session, the researchers conducted a short interview with the whole group to gather additional feedbacks in a collective discussion.

Results were positive confirming a match between the designers' expected EFs and the one

identified by the experts (67% of the sample in the worst case).

## **5. Final version**

The months until September 2018 were dedicated to the implementation of the last mini-games, the definition of the training algorithm and the refinement of the aesthetics.

### ***5.1 The Participatory Elemental Pentad in SOM***

At the end of the design and implementation stages, some important theoretical conclusions were made. First of all, the direct users of the game (i.e. the players) were important contributors in the design process, but, it has to be noted, their understanding of the mechanics and the dynamics (Hunicke et al. 2004) was superficial and any deeper discussion would require a transfer of knowledge between designers and players. Players, in fact, prioritized the aesthetic (how the game look and what they feel playing it), rather than focusing on how the different mechanics can contribute to the overall game experience. Most of the suggestions regarded the fundamental ideas at the base of the story (e.g., distant words in the future, fight between heroes and enemies); even if they related with some mechanics (e.g., crafting or rewarding systems), they were mostly focusing on the look and feel of these features than on the specific game rules.

Vice versa, the domain experts played a crucial part in identifying the goals for which the game is developed and in translating them into game dynamics and mechanics. Without the involvement of clinical psychologists and cognitive scientists, the designers can only rely on assumptions, which could lead to very adverse consequences on the efficacy of the game. The domain experts should keep constantly in mind the purpose of the game and their hands-on experience in typically developing and dyslexic children. However, they should be ready to compromise balancing the choices related to the purpose and the ones more link with a playful game experience.

### ***5.2. Story***

Skies of Manawak (SOM) is a Sci-Fi video game set on Manawak, a planet located in another galaxy, made habitable thanks to a terraforming process and colonized by the human race. Centuries ago, a cataclysm caused the dysfunction of gravity generators, causing diverse anomalies that disrupted the entire planet. The result was a surreal scenario where entire continents have lifted off the ground and float hundreds of meters above a land now submerged by the seas. After centuries of peaceful living, the descendants inhabit peacefully

various villages spread across the planet. One-day invaders from outer space declare war, reactivating the ancient towers containing the gravity generators, the *Kivas*, now considered sacred temples. The consequences were extreme: the floating lands begin to collapse, jeopardizing the fate of Manawak. The game begins the day of the initiation of the protagonist, when the girl/boy (the player can choose between a roster of different characters) is promoted to the role of guardian of the Rakus, creatures able to glide in the gravitational anomalies, the *Manawak*. Flying through the various nations, facing a number of challenges (i.e., cognitive exercises alternating between action and mini-games) the protagonist and her/his Raku will try to stop the invaders, deactivate the Kivas and finally put an end to the war.

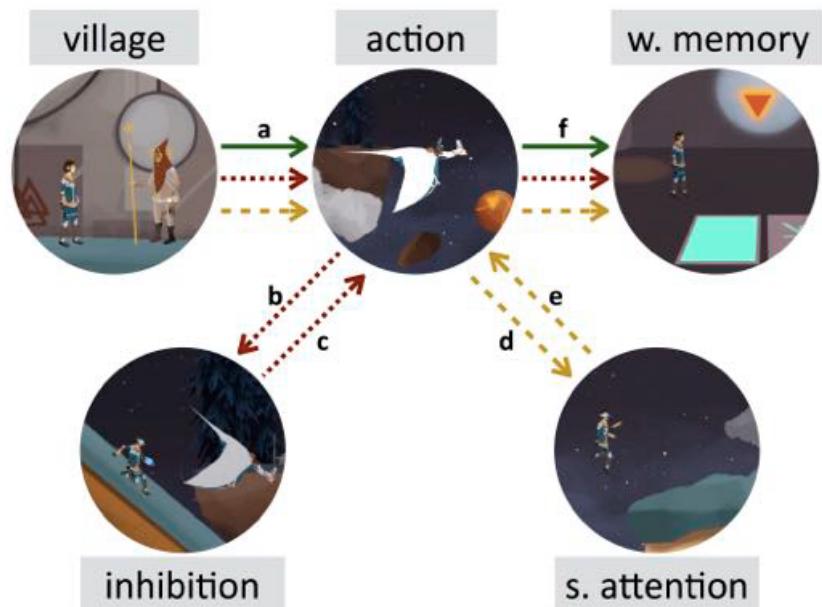
### **5.3. Mini-games**

In the final version, SOM was composed by 10 mini-games each one tapping one or more executive functions. The set of mini-games of a game module (“a quest assigned by the village chief”) is determined by the player performance and the training path: even if some events are strictly related to the main plot, everything else is generated at run-time according to the performance of the player.

Importantly, the game structure is such that players need to flight from one game module to another, training several fundamental aspects of action video games, which are known to facilitate brain plasticity (Bavelier et al., 2012). Each player pursues a different training path on the basis of the initial performance (first session): for example, the children who performed worse at the beginning of the training in a specific mini-game that trains working memory, will find more exercises that tap into that specific cognitive function.

A simplified representation of these mechanics can be found in Figure 25. In this structure the fixed points are represented by the villages and the Kivas (the player has to flight between the firsts and the latters); the sub-quests that the player must face are determined and generated by the system. If, for example, in the previous “quest” the player strongly improved in the mini-game tapping response inhibition, but had a low performance in split attention (Divided attention and Dual Task), the system generates a new quest that is structured through a path a, d, e, f (yellow path: village, first action session, split attention, second action session, working memory); on the other hand, if the player improved in this last mini-game, but performed low in response inhibition, the system generates a path a, b, c, f (red path: village, first action session, response inhibition, second action session, tower).

**Figure 25|** Example of the ramification of the quests according to the performance of the player.



In addition, the system automatically adapts the exercises difficulty level on the players performance. In other words, the training algorithm was set so that the difficulty level was increased modifying every parameter (e.g., duration and number of trials) of every mini-game. In this way the structure and overall difficulty is challenging, always maintaining a minimum level of challenge to keep the immersion (Anguera and Gazzaley, 2015). Since compliance and motivation could strongly affect the outcomes of training (Richter, Raban & Rafaeli, 2015), our goal was to provide a game in which a design carefully studied that drive game play to be both challenging and fun is combined with effective cognitive exercises.

The following subsections briefly present them, providing information in terms of the mechanics and the underlying cognitive mechanics.

### 5.3.1. *The flight – visuo-spatial selective and sustained attention*

Whenever a player needs to move from one place to another within the game, he/she has to go through the flying mini-game. This mini-game requires flying through a sky populated by different types of threats, evaluating their trajectories and dodging or shooting them. Threats can come from different directions: stones and sentinels move horizontally from the right boundary of the screen to the left one; boulders hail from the depth of the screen in the direction of the observer (three-dimensional movement). Moreover, stones and boulders change in speed and density of the clusters they form, while sentinels display different

behaviours and statistics depending on the typology they belong to. Additionally, the player is required to defeat the phoenix - the main menace of these game sessions - using a combination of tactics that requires both dodging and shooting. The system adjusts the level of difficulty of the flying session, taking previous performance into account. These sessions train different aspects of attention, particularly visual selective and sustained attention.

**Figure 26** Two flight sessions showing a cluster of meteors (upper part) moving toward the Raku, and boulders (lower part) moving toward the camera (left). Some of the sentinels populating the skies of Manawak and a phoenix (right)

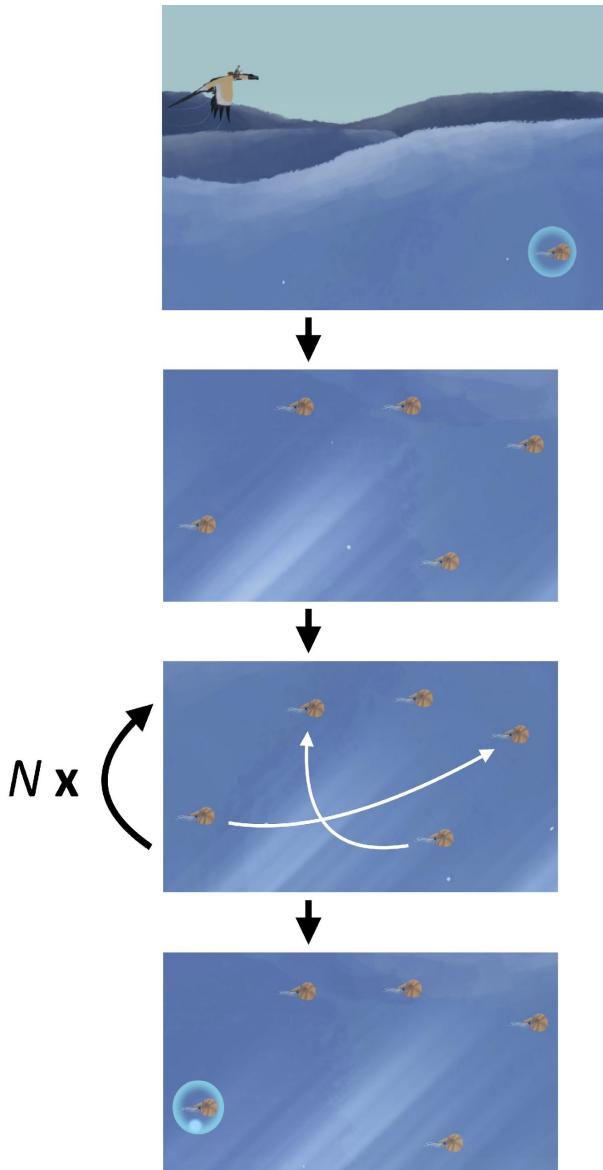


### 5.3.2. Fishing - visual attentional span and working memory

Whenever the player's health points drop to zero during the flight, he/she needs to reach the surface of the sea to gather food and recharge the health points (Figure 26). The fishing session is a gamified version of the Multiple Object Tracking task (MOT), where the participant is asked to keep trace and identify of a subset of targets (fishes) that move randomly among a group of identical non-targets. The target appears on the screen at the beginning of the session and a light-blue circle surrounds it. After few seconds, other identical fish appear and the group starts to shuffle. At the end of the movements, the player is required to identify the target/s. The persistence of the target, the total number of stimuli and the number and speed of the random movements of the stimuli is based on the level difficulty.

The fishing session was designed to train divided visual attention, namely the prolonged ability to distinguish targets from identical-looking distractors, while successfully tracking them over time. Similarly to the MOT task, this mini-game is also likely to load on a working memory component (Lavie et al., 2018).

**Figure 27|** A fishing session: the first nautilus appears; it is shuffled with the others; finally, the player has to identify the correct one.

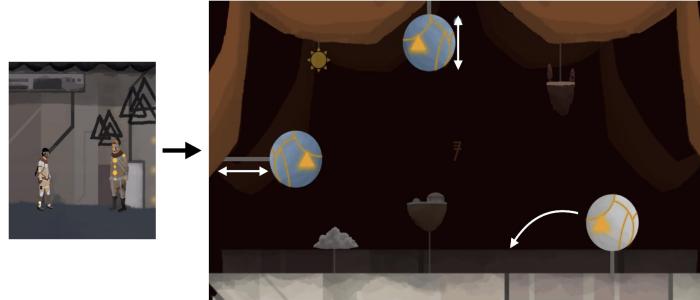


### 5.3.3. Secret island, the shooting gallery - Visual attention divided and selective

This mini-game (Figure 28) displays similar mechanics than the shooting galleries of amusement parks: the player has to anticipate the trajectories of the sentinels and, using the mouse, has to shoot them. The total number of sentinels, their speed and the duration of the session are proportional to the level of difficulty, which is adjusted according to the user performance during the previous session. The shooting gallery was designed to train several aspects of attention. In fact, the player must pay attention to the entire screen and, meanwhile,

discriminate targets coming from various unpredictable areas of the screen among distractors.

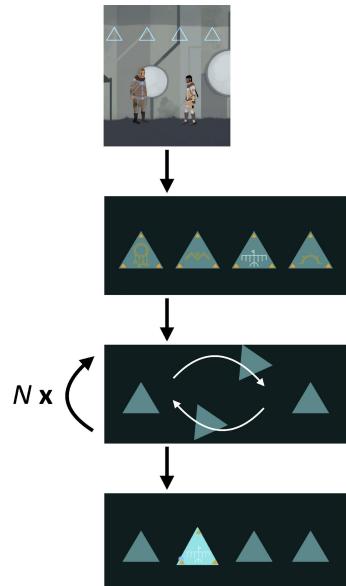
**Figure 28|** In the shooting gallery the fake sentinels move around the screen. The player has to click (shoot) on them in order to gain points.



#### 5.3.4. Secret island, the shell game - Visual selective attention and spatial tracking

This mini-game, together with the “shooting gallery”, is located in a secret island that can be reached by the players if they want to gain additional points can be spent for upgrades (Figure 29). It was inspired by the *shell game*, or similar gambling games where three or more identical objects are placed on a surface. Firstly, a card with the target symbol on it is shown to the player, secondly up to six cards are added to the screen and thirdly the cards are covered and they start to shuffle. The aim of the player is to follow and ultimately identify the target card among other, identical cards. This mini-games trains attention and memory capacities. The number of cards and speed of shuffle varies according to the level of difficulty, based on performance in previous sessions.

**Figure 29|** A shell game session: the cards are revealed and shuffled; the player has to identify the correct one.

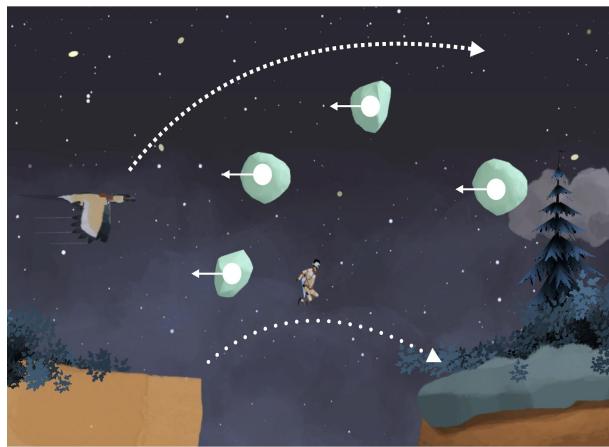


### *5.3.5. Meteors and islands - Divided attention and Dual Task*

In this mini-game the player needs to find one of the keys required to open the Kivas. In order to do this, the player has to control both the protagonist and the Raku. In fact, while the former has to jump (using the jump button) from one island to another, the Raku has to dodge the meteors (using the keyboard arrow keys). The difficulty is updated based on the total loss of health points, adjusting the speed of the characters, the density of meteors, the distance between the islands and the total duration of the session.

Since the player needs to rapidly switch focus between the two characters, this session mainly trains divided attention.

**Figure 30|** The player has to control the jump of Hoa'manu, while moving the Raku to dodge the meteors.



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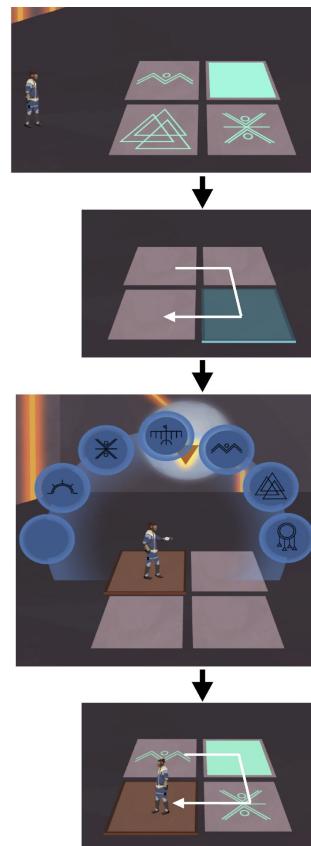
### *5.3.6. Kiva, the security system - Visuo-spatial working memory*

During the game (Figure 31), the player is asked to deactivate several Kivas (gravitation-generating towers that are now threatening the planet). The player needs to find the right combination of symbols for every room to access the elevator, reach the last floor, and deactivate the tower by destroying its energy panel. Each combination corresponds to a matrix of tiles placed at the centre of the room; based on the difficulty, the tiles can form a 2X2 or a 3X3 matrix. The combination is shown in two steps. The persistence of the symbols and the path lighting speed are determined by the difficulty of the level.

The security system was designed to train visual working memory. The player has to keep track of both the displayed symbols and the path: at first, every tile shows a symbol and then, the symbols disappear and the tiles are illuminated, one by one, forming a path. To deactivate the security system, the participant has to fill an empty table with the symbols shown before, following the correct order of answer.

This mini-game trains visual short-term memory while the player has to remember the symbols and the path; additionally she/he has to combine these two kinds of information to output the correct answer.

**Figure 31|** An example of security lock. Firstly, Kiva shows the symbols and the path of answer; then, Hoa'manu must replicate it.



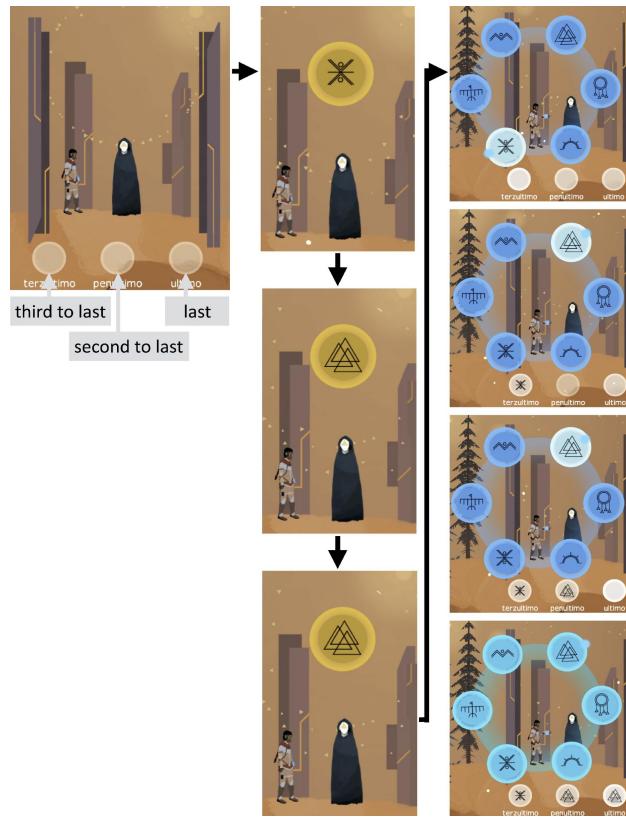
### 5.3.7. Rekenanagi - Visual working memory and Updating

This is a ritual that the protagonist must perform to summon his flying creature (“Raku”). The summoning is structured into several steps (Figure 32): the shaman presents a series of symbols, one after the other. An algorithm calculates the length of the string and the persistence of each symbol according to the level of the game (symbols are presented randomly). Subsequently, the player has to answer, selecting symbols so as to reproduce the sequence of the last  $n$  symbols presented. The  $n$  is determined according to the difficulty (maximum three) and notified to the user using a visual clue (bottom grey circles, in Figure 32) at the beginning of the ritual.

This mini-game was designed to train visual working memory capacities: the player has to keep track of the symbols summoned by the shaman, working on short-term memory. Due to the fact that the length of the string is undefined, she/he has to continuously update

information in order to then report the last  $n$  symbols of the list.

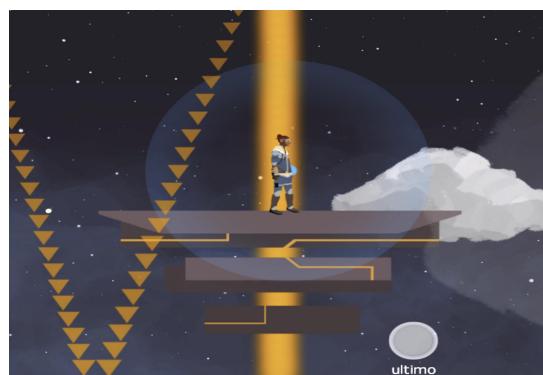
**Figure 32** A session of the Rekenanangi: the Uka starts the ritual and shows the combination (left and central columns); the player (Hoa'manu) repeats the combination (right column).



#### 5.3.8. The call – Auditory working memory

This mini-game is a simplified version of the working memory Rekenanangi mini-game, yet based on auditory stimuli. Only three sounds are used: a low, a medium and a high pitch sound, each one being associated with a symbol. The player is required to trace the emitted sounds, manipulating this information in his/her memory system and answer by selecting the last  $n$  symbols of the series.

**Figure 33** The process is similar to the Rekenanangi, but based on auditory stimuli.

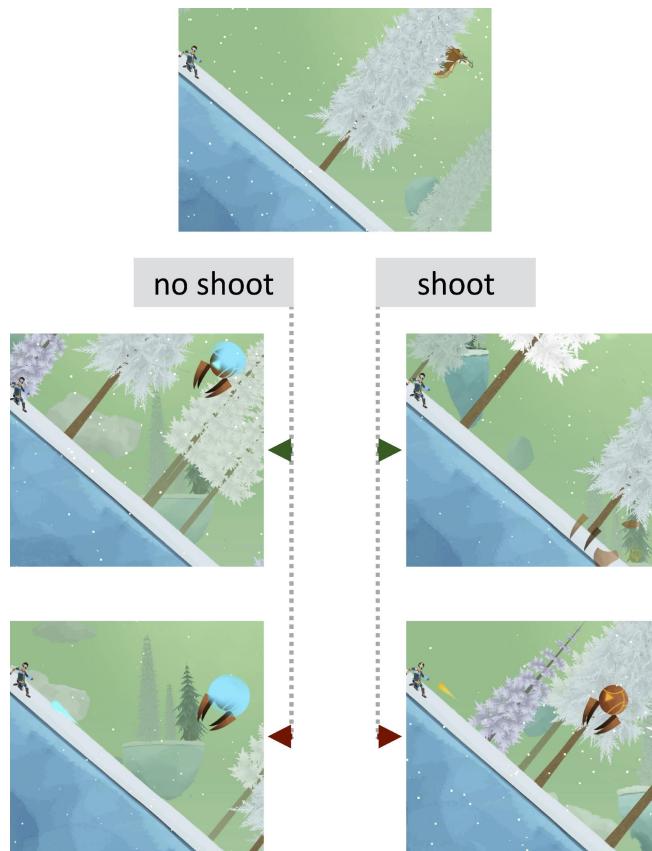


### *5.3.9. The falling island - Inhibition and Sustained attention*

This mini-game is a transposition of the classic Go/no-Go task, which tests the ability to inhibit a response (Figure 34). Players are instructed to make a button response anytime they hear a “go sound” (a roar emitted by the Raku) and to not respond to any “no-go sounds” (a roar that is identical to the go sound in the first part, but which ends with a low pitch). The low pitch roars (“no-go sounds”), which are less frequent than the “go sounds”, indicate the presence of shielded sentinels: the player has to inhibit the shooting response in order to avoid the rebound of their bullet.

The system determines the number of sentinels, the maximum time available to press the button and the total duration of a session on the basis of the difficulty level.

**Figure 34|** Examples of possible reactions by the player: the upper part shows the right answer for the shielded (left) and shooting (right) sentinels; the lower part shows the wrong ones.



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### *5.3.10. Kiva - the energy system*

The objective is to solve a series of puzzles (four per mini-game session) based on gravitational mechanics (Figure 35). The player has to use the different objects and modify the canvas in such a way that the glowing ball, starting from the entry point, will end up

reaching the exit point.

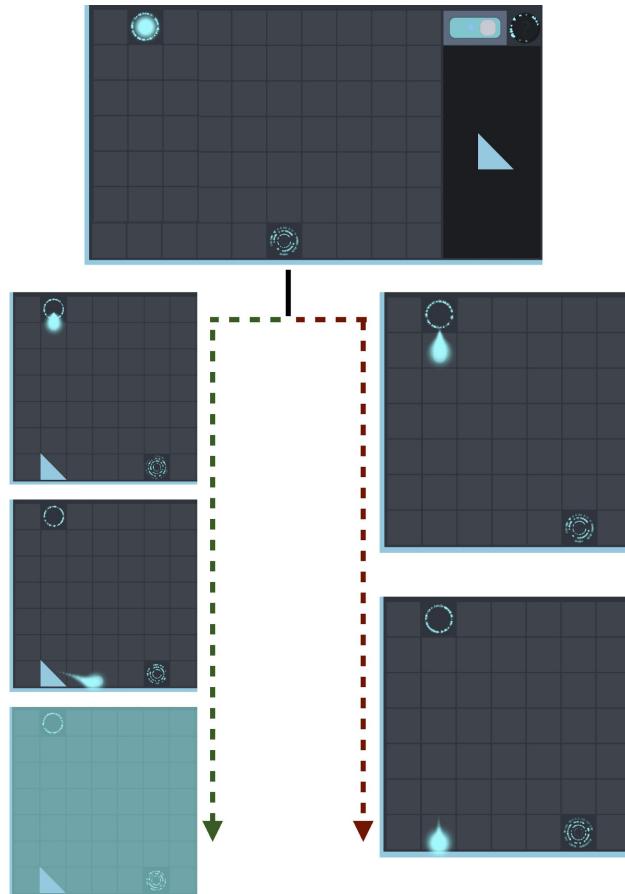
The mini-game comprises four different type of objects:

- physical objects: they modify paths and trajectories of the ball;
- gravitational field: it attracts or repulses any ball;
- laser: it destroys any ball on contact;
- antimatter ball: it has similar shape and behaviour to the energy ball. If it reaches the exit point, the player fails the puzzle.

The difficulty increases with each new level; if the player cannot solve a puzzle she/he can use, after a fixed waiting time, a maximum of three hints, which will provide a visual clue of the solution.

The mini-game was designed to train planning skills: the player has to carefully understand the cause-effect relationships of the various objects in relation to the ball, and plan their usage and location accordingly.

**Figure 35**| The second puzzle of the energy panel. With no intervention by the player, the energy ball cannot reach the exit point (right part). However, adding a physical object to change the trajectory of the ball, the puzzle is solved (left part).



## CHAPTER 5

# ENHANCING READING SKILLS IN TYPICALLY READING AND DYSLEXIC CHILDREN WITH “SKIES OF MANAWAK”, A VIDEO GAME FOR COGNITIVE TRAINING

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*“The essence of knowledge is, having it, to apply it;  
not having it, to confess your ignorance”*  
~ Confucio

### 1. Introduction

In recent decades, growing evidence has accumulated on the possibility of altering the cognition for the better through cognitive training. Despite promising results, these past studies showed some limitations: several studies in the field of EFs training have only focused on the training of one specific executive component and, in addition, most of them investigated only the near transfer effects without taking into consideration the far transfer and the long-term effects of the training. Moreover, it should be mentioned that not all studies included an active control group, thus resulting in potential confounds related to motivation or expectations (e.g. placebo effects). Furthermore, no research has been found that surveyed attentional training (e.g., action video games) in the general school-aged population since, up to now, the action video game studies all focused on reading-impaired children.

Therefore, the aim of the present study was to investigate whether Skies of Manawak (SOM), a video game that combine the training of several executive skills within one same gamified intervention, might have a positive effect not only on these functions, but also on literacy skills. On account of this fact, we first decided to test training effects through classic, standardized paper and pencil tasks in a school-based study with typically developed children (Study 2 – part A). Secondly, our goal was investigating whether SOM may enhance EFs following intervention, and whether these improvements transfer to reading skills in children with dyslexia (Study 2 – part B). All children underwent 12 hours of training, distributed over 6 weeks, either on Skies of Manawak or on a control game (Scratch), as fun and as engaging as the former.

Specifically, we expect to find higher improvements in EFs, which are specifically addressed

by the video game and are extended to literacy skills, for the training group compared to the control group both for the typically reading and dyslexic children.

In the next part of the chapter, the two studies are presented.

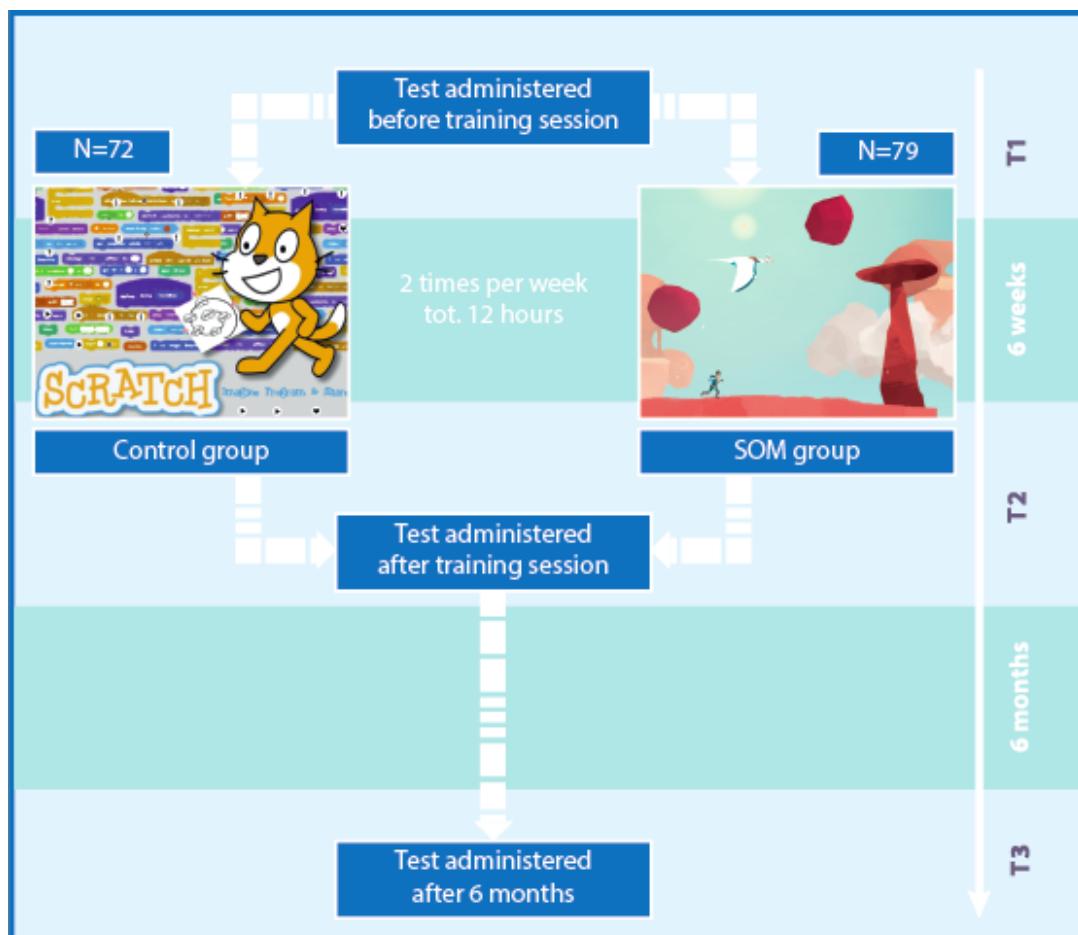
## 2. STUDY 2 – part A

### Method

#### 2.1. Study design and participants

The effects of the training program were assessed in a study involving 151 typically developing Italian children, conducted in a school in the north of Italy (Veneto). Participants, who attended the 3<sup>rd</sup>, 4<sup>th</sup>, 6<sup>th</sup> or 7<sup>th</sup> grade, were divided into a training group ( $n = 79$ , 39 females) and a control group ( $n = 72$ , 41 females), which completed two different 6-weeks training programs. In Figure 36, the study plan is represented as a flowchart.

**Figure 36** The flowchart of Study 2 - part A.



The experimenter ensured that participants were focused and that they were motivated, by

providing encouragement according to their individual performance. Children in the first group were trained on our videogame, Skies of Manawak (SOM), 2 times per week, with training sessions of one hour each (see Figure 37, for a picture of a game session). Participants in the active control group received a computer-based training on coding using *Scratch*, a programming language developed by the Lifelong Kindergarten (MIT) Group, which allows children to create interactive stories, games, animations, and simulations (Resnick et al., 2009). All the information regarding SOM can be found in chapter 4, while a description of the training protocol with Scratch will be presented in the next paragraph (5.2.2.). Training duration and frequency (1 hour per day, twice a week for 6 weeks) were matched across the two groups. Comparing the training effects in the experimental group to those observed in the active control group allows controlling for other variables that can affect results such as test-retest effects, familiarity with the assessment environment, developmental and schooling effects.

At the end of the school year (6 months after the end of training), we carried out a follow-up evaluation, the objectives of which were threefold: 1) implement a complete double-blind evaluation; 2) control for test-retest effects; 3) assess the persistence of the aforementioned improvements in literacy skills and EFs performance.

**Figure 37|** Children playing Skies of Manawak.



The study was quasi-experimental, due to the fact that it was not possible to randomly assign participants in the two groups. However, any statistical differences were found between the demographic and neuropsychological variables at baseline of the two groups, with the exception of comprehension (see Table 2). The two groups were matched in age (SOM group:  $M = 10.39$  years ( $SD=1.48$ ); control group:  $M=10.30$  ( $SD=1.45$ );  $t_{(1\ 149)}=0.422$ ,  $p=0.674$ ), sex (SOM group: 39 Female; control group: 41 Female;  $t_{(1\ 149)}= 0.868$ ,  $\chi^2 =0.351$ ).

Criteria for inclusion in the analysis were: (i) normal or corrected to normal visual acuity; (ii) no Attention Deficit Hyperactivity Disorder diagnosis; (iii) no diagnosis of Learning Disorder or no reading delay in word and non-word reading tests; (iv) no diagnosis of Intellectual Disability. In fact, in the general intelligence test (Raven, 1994), children of both groups had a performance within the normal range (above 10th percentile (SOM group:  $M=107.32$  ( $SD = 9.73$ ), control group:  $M = 106.85$  ( $SD = 9.69$ );  $t_{(1\ 149)} = 0.422$ ,  $p=0.767$ ). Players with intellectually different profiles –e.g. children with learning disorders ( $n=21$ ) or with borderline intellectual functioning ( $n=10$ ) - played SOM like any other classmate, however their data were not included in the sample.

All the children's parents gave written informed consent after an extensive description of the research study. At the end of the study, participants were thanked for their time and effort with a small gift.

The descriptive statistics of the main variables that were identified at the first assessment time point (T1) are presented in Table 12 (Demographic and cognitive characteristics), Table 13 (Literacy skills performance), Table 14 (Executive functions performance).

**Table 12|** Demographic and cognitive characteristics (Means, SD) of SOM group and Control group prior to intervention.

<sup>a</sup> $\chi^2$ -score, <sup>b</sup> F-score; \*sig.  $\alpha= 0.05$

	SOM		CONTROL		Test	p
	Mean	SD	Mean	SD		
Sex (f-m)	39-40		41-31		0.87 <sup>a</sup>	0.35
Age (years)	10.39	1.48	10.30	1.45	0.004 <sup>b</sup>	0.67
IQ (Raven's matrices)	107.32	9.73	106.85	9.69	0.06 <sup>b</sup>	0.77

**Table 13|** Means and standard deviations at baseline (T1) of literacy measures of participants of the two groups (SOM and Control). All the literacy variables are expressed in z-scores. <sup>a</sup> $\chi^2$ -score, <sup>b</sup> F-score; \*sig.  $\alpha= 0.05$

	SOM		CONTROL		Test	p
	Mean	SD	Mean	SD		
<b>LITERACY skills</b>						
Word Reading	Speed	-0.30	0.81	-0.31	0.86	0.30 <sup>b</sup>
	Accuracy	-0.41	0.84	-0.36	0.89	0.29 <sup>b</sup>
Non-word Reading	Speed	-0.24	0.71	-0.28	0.76	0.28 <sup>b</sup>

	Accuracy	-0.12	0.85	-0.06	0.74	2.05 <sup>b</sup>	0.67
Text Reading	Speed	-0.02	0.72	-0.10	0.70	0.43 <sup>b</sup>	0.53
	Accuracy	0.12	0.57	0.13	0.64	0.81 <sup>b</sup>	0.92
Writing	Word	-0.70	1.98	-0.65	1.69	2.16 <sup>b</sup>	0.86
	Non-word	-0.11	1.25	-0.11	1.17	0.06 <sup>b</sup>	0.98
Comprehension		0.23	0.73	-0.05	0.95	4.53 <sup>b</sup>	0.04*

**Table 14**| Means and standard deviations at baseline (T1) of executive functions performance of participants of the two groups (SOM and Control). All the literacy variables are expressed in z-scores. <sup>a</sup> $\chi^2$ -score; <sup>b</sup> F-score; \*sig.  $\alpha=0.05$

		SOM		CONTROL		Test	p		
		(n=79)		(n=72)					
		Mean	SD	Mean	SD				
<b>EXECUTIVE FUNCTIONS</b>									
Digit span	Forward	-0.63	0.89	-0.71	0.73	1.13 <sup>b</sup>	0.57		
	Backward	-0.24	0.88	-0.22	0.87	0.06 <sup>b</sup>	0.83		
Visuo-spatial span	Forward	-0.36	1.39	-0.44	0.91	15.07 <sup>b</sup>	0.67		
	Backward	-0.26	1.15	-0.25	1.06	1.03 <sup>b</sup>	0.95		
Bells (barrage) task	Speed	-0.15	1.03	-0.18	0.91	0.29 <sup>b</sup>	0.84		
	Accuracy	-0.14	0.86	-0.22	0.95	0.59 <sup>b</sup>	0.59		
TOL		0.10	0.94	-0.04	0.96	0.24 <sup>b</sup>	0.35		

## 2.2. Scratch – The Control Activity

Scratch is a free visual programming language developed by the Lifelong Kindergarten Group at the MIT Media Lab, that was launched in May 2007 and, nowadays, it is available and used in more than 50 different countries. This online program aims at teaching coding, computer science, and computational thinking particularly to elementary and middle school children. Users of different ages can enjoy in creating projects (i.e. make animations and games) that differ in complexity using a simple block-like interface (see Figure 38 for an idea) and, furthermore, can share these projects with other young designers from all round the world in the online community. Scratch is a very popular software due to the fact that it is free, with an appealing aesthetic and it easy to use (it is possible to download it in ordered to use in contexts with no internet access): until now it has been used by hundreds of thousands of users that have created and shared more than 6 million projects.

**Figure 38|** Scratch's interface



A well-written manual (<http://scratched.gse.harvard.edu/guide/download.html>) helps parents, teachers and other education specialists in building training curricula that support computational creativity and computational thinking (Brennan, Balch, & Chung, 2011). Computational thinking could be defined as the ability “to think like a computer scientist and being able to apply this competence to every field of human endeavor” (Corradini, Lodi, & Nardelli, 2017). In this regard, Brennan & Resnick (2012) developed a computational thinking framework in order to describe the learning processes that take place when children design and interact with Scratch. Specifically, this software allows activities that “encourage exploration of key *computational thinking concepts*” designers employ as they program such as sequence, loops, parallelism, events, conditionals, operators, data. Moreover, Scratch is supposed to boost several important *computational thinking practices*, particularly abstracting, iterating, modularizing, testing and debugging. Finally, the last important dimension of this framework is represented by *computational perspectives*, namely the perspectives that designers form about the world around them and about themselves such as expressing, connecting, and questioning. In general, it is possible to agree that computational thinking could be considered as a particular type of thinking for problem-solving. Children who are programming with Scratch need to learn how to formulate the problem they want to solve: a processing agent will then execute them. In example, one of the first steps is understanding that a particular activity or task could be conceptualized as a sequence of single steps that can be carried out by the program (“sequences”), that some activities can be the result of other - previous – steps (“events”) or can happen at the same time of others (“parallelism”) or that it is possible to make decisions based on certain conditions, i.e. if-else

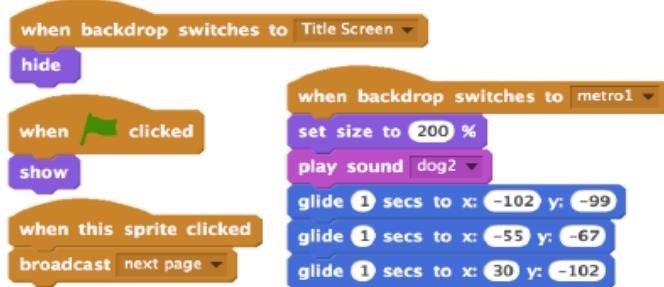
(“conditions”). Moreover, in Scratch it is possible to implement “loops”, aka the mechanisms for running a sequence multiple times. In addition, teachers use it as a tool to teach important concepts such as mathematical, logical, and string expressions: Scratch, in fact, supports a range of mathematical operations and string operations.

Figure 39| An example of a Scratch’s code.



An appreciable element of novelty is represented by the possibility of learning in high motivational context that allows the direct experimentation of what has been learned. Children can learn and store in their memory important concepts of different subjects (i.e. science, history, geography) through a game or a story they created utilizing their imaginations. Usually, the creation process starts from an idea, and then a prototype is created, tested, and corrected in case of errors or imperfections and, finally, submitted to the judgment of other peers/teachers in order to be able to further improve it. It is a continuous – positive -spiral: have an idea, create the project, think to new ideas from which further projects will be implemented and so on. In our studies, we developed in synergy with the teachers a curriculum based on the activities proposed by the Creative Computing Curriculum Guide (Brennan, Balch, & Chung, 2011) and the Beri & Boscaini’s Book (2016). The main aim of the curriculum, after an initial fase of practice (Unit 0) was to introduce children to the key computational concepts (described above) through step-by-step tutorials (Unit 1). In addition, children learned how to create stories through digital storytelling (Unit 3): designing characters, developing scenes, creating conversations and other interactions between characters. Sixth and seventh-grades students were able to extend the practice with the creation of an animated Christmas card. For a further description of the activities and some examples, see Appendix B.

**Figure 40|** An example of a story scene with the related code.



### 2.3. Procedures

All children were tested before and after the video game training in a quiet room in their school. Baseline (pre-training) assessments were completed one week prior to the start of treatment, and outcome (post-training) assessments one week after the end of the treatment. Testing was conducted by three different clinicians. Tasks were administered in a pseudorandom manner; IQ and comprehension abilities were always assessed in a collective session, whereas the other tests were administered individually.

Both the experimental group and the active control group were pre-tested for visuo-spatial attention, auditory and visual working memory, planning and literacy skills (T1).

All measures, except for IQ, were re-assessed at the second measurement point, within a week from the end of training (T2). Furthermore, at the end of the training all the children completed a questionnaire aimed at investigating their motivation, the perceived difficulty and appreciation of the video game, as well as their personal evaluation of the training. After 6 months – approximately at the end of the school period – a follow-up (T3) evaluation was done.

While at T2 testing was conducted by three different clinicians, with only two of them fully unaware of each child's treatment allocation (SOM versus Control), at T3 all experimenters were entirely blind to the group each participant belonged to, as well as to the research question of the study. Our aim here was to prevent biases due to observer-expectancy effects. In order to check for test-retest effects that can be caused by the use of the same paper-and-pencil tests through all the assessment sessions, at T3 we chose to administer two additional tests: a text-reading task that measures reading abilities of meaningful material and a barrage task that evaluates visuo-spatial attention. Both are part of the Neuropsychological Assessment Battery for childhood and adolescence (Bisiacchi et al., 2005).

Therefore, the assessment battery at the follow-up consisted of a selection of the tests previously used (reading tasks, bells task and TOL) and two additional tests (text-reading and barrage task).

#### **2.4. Tools**

The instruments comprised in the initial diagnostic battery (T1) are detailed below. Furthermore, the feedback questionnaire, specifically designed for assessing the gaming experience (T2), is presented. In conclusion, the two additional tests administered at the follow-up (T3) are described.

#### *INTELLIGENCE*

Fluid intelligence (Gf) was measured using a non-verbal multiple choice IQ test: the Coloured Progressive Matrices - CPM (Raven, 1994) for children until 11 years-of-age and the Standard Progressive Matrices – SPM for the older children. Raven's matrices assess the ability to reason and solve problems by analogy and forming perceptual relations, irrespective of culture, language and formal schooling.

#### *EXECUTIVE FUNCTIONS*

*Attentional control (visuo-spatial selective and sustained attention)* was estimated through the “Test delle Campane Modificato” (Modified Bells Test, Biancardi, Stoppa, 1997). In this barrage test, children are asked to localize drawings of bells among distractors (familiar figures such as houses, horses). The stimuli are pseudo-randomly organized and each sheet contained 35 target stimuli. Two types of z-scores are obtained: the score that is given by the sum of the targets (bells) found in the first 30 seconds for each of the four sheets that composed the test (fast score), and the score obtained counting the total number of targets

found in 120 seconds (slow score).

*Verbal memory* To assess short-term memory and WM a digit span subtest from the “BVS - Corsi - Batteria per la valutazione della memoria visiva e spaziale” (Battery for the assessment of visual and spatial memory”, Mammarella et al., 2008)), was used. Participants hear a sequence of digits and are required to recall the sequence first in the same order (Digits Forwards) and later in the reverse order (Digits Backwards), with increasingly longer sequences being tested in each trial. Each correct sequence is worth one point (with a maximum of 28: 14 points for each subscore series). If both trials of a pair of sequence are coded wrong, the examiner skip to Digits Backwards.

*Visuo-spatial memory (Short-term and Working)* was evaluated by Corsi’s test in both forward and backward versions. This test is part of the “BVS - Corsi - Batteria per la valutazione della memoria visiva e spaziale” (Battery for the assessment of visual and spatial memory” (Mammarella et al., 2008)). It has the same structure (e.g. stopping rule, number of sequences, etc.) of the Digit span subtest. Participants were asked to mimic the psychologist while he/she is tapping a sequence of up to nine identical blocks. The backward version requires the subject to repeat the sequence back in order.

*Planning* To determine planning and problem solving abilities the Tower of London test was used (Sannio, Fancello et al., 2006 Italian version; Shallice, 1982). Participants were presented with a model where three beads are positioned on three pegs with different heights. Children were then asked to manipulate beads from a predetermined starting position to match the configuration of beads in the model.

## LITERACY SKILLS

*Reading skills* were assessed through two different tasks:

- “Batteria per la valutazione della dislessia e disortografia evolutiva” (Battery for the assessment of Developmental reading and spelling disorders, Sartori et al., 1995). Children were asked to read aloud 4 lists of 28 words and 3 lists of 16 non-words. Speed (in seconds) and errors (each incorrect word or non-word was counted as one error) z-scores were computed for single word and non-word reading.
- “Prove di lettura MT per la scuola elementare-2” (Reading tests for primary school, Cornoldi et al., 1998) and “Nuove prove di lettura MT per la scuola media inferiore” (New reading tests for secondary school, Cornoldi and Colpo, 1995). These age-standardized text-reading tasks were used to measure ecological-context reading. It provided separate score for reading fluency and errors.

*Writing* was evaluated through the Spelling tests of the “Batteria per la valutazione della dislessia e disortografia evolutiva” (Battery for the assessment of Developmental reading and spelling disorders, Sartori et al., 1995). These tests give correctness scores (expressed as number of correct responses) in writing words and non-words.

*Text comprehension* was assessed using “Prove di lettura MT per la scuola elementare-2” (Reading tests for primary school, Cornoldi et al., 1998) and “Nuove prove di lettura MT per la scuola media inferiore” (New reading tests for secondary school, Cornoldi and Colpo, 1995). This is a multiple choice test that can be administered in a collective form. The results of the tests were expressed as z scores according to age norms.

### STUDENTS' WELL-BEING

Social emotional aspects were took into consideration administering the “Questionnaires for evaluating school well-being and identifying risk factors”- QBS 8-13 (Tobia and Marzocchi, 2015). The main aim of *QBS 8-13* is to analyse the well-being of students aged between 8 and 13.

The child/pre-adolescent questionnaire, which comprises 27 items, examines several aspects of school-well-being: satisfaction and recognition, relationships with teachers, relationships with schoolmates, emotional attitude at school, sense of self-efficacy and causal attribution processes. The results of the five indexes are expressed in standardized scores (T-scores).

### FEEDBACK QUESTIONNAIRE

Children completed a feedback questionnaire (see APPENDIX x) at the end of the training. The structure was based on the Short Feedback Questionnaire (eSFQ) created by Moser and colleagues for the rapid assessment of game experiences (Moser et al., 2012).

Our questionnaire includes items concerning the self-evaluation of the enjoyment of the training, the perceived difficulty and how much they liked/disliked playing. In addition, SOM questionnaire has a set of closed questions aimed at identifying the most liked, disliked and difficult mini-games, with the possibility to specify the reasons. The final part consists in an open question design to collect general feedbacks and suggestions for improvements.

### FOLLOW-UP ADDITIONAL TESTS

*BVN - text reading-task.* This task is part of the “Batteria per la Valutazione Neuropsicologica per l’età evolutiva – BVN” (Battery for the Neuropsychological Assessment) proposed by Bisiacchi and colleagues (2005). The child is asked to read aloud a

short text passage as fast and as correctly he/she can. The final score (the transformed in z-score) represents the number of correct syllables that the participant was able to read in one minute of time.

*BVN - barrage task.* Similarly, this barrage task, which assess selective visuo-spatial attention, is part of the BVN (Bisiacchi et al., 2005). In this task, children are required to draw a line on the target stimuli (a square with two small lines in the middle) that they were able to localize in one minute. The distractors are represented by other squares that differ for the orientation and the position of the two lines. The task is composed by only one sheet, in which the stimuli are organized. One z-score is computed (total number of targets found in 60 seconds).

## **2.5. Statistical Analyses**

Following 12h of training in their assigned game (our own video game, Skies of Manawak - SOM versus the control commercial game Scratch), administered over a 6-weeks period, performance in the same cognitive domains was re-assessed, in order to estimate the effects of the administered training regimens on literacy skills and executive functions performances, giving rise to a first set of analyses focusing on comparing T1 and T2. Then we asked whether the intervention effect may be long lasting by comparing T1 and T3 on the restricted set of measures collected at T3.

For all analyses, group differences were examined by means of Analyses of Covariance (ANCOVAs/MANCOVAs, on the basis of the number of dependent variable considered). More specifically, a series of repeated measures general linear model (GLM) analyses were conducted to evaluate training effects with *Time* as a within-subject factor and *Group* (SOM versus Control) as a between-subject factor. The Time X Group interaction was our primary effect of interest. In case of a significant Time X Group interaction, further post-hoc analyses were run.

In order to exclude possible confounding effects, *chronological age* and *IQ scores* (Raven's CPM) at T1 were entered as covariates and *Sex* as a between-subject factor for all MANCOVA/ANCOVA analyses. With the exception of IQ scores, all participants' performance measures were transformed into z-scores on the basis of age norms.

In addition, similar analyses, but on time points T1 and T3, were run in order to evaluate the group differences between the two group at the follow-up six months later.

Because of slightly different tasks administered at T1 and at T3 to assess reading and attention, a Pearson correlation was run to determine the relationship between the BVN-text

reading score and performance in the text-reading task at T3. Analogously, a similar correlation was run between the two barrage tasks.

Furthermore, in order to evaluate the generalization of potential improvements to the academic domain, we computed two separate ANCOVA for marks obtained in Italian and Mathematics at five time points (pre-training, post-training, follow-up 6, 12 and 18 months later).

Moreover, three steps, fixed entry, multiple regression analyses were computed to explore the linear relationship between literacy improvements ( $\Delta$  T1-T2 reading speed, reading accuracy, writing: dependent variables) and EF improvements (block 3:  $\Delta$  T1-T2 EFs) taking into considerations covariates effects (block 1: age, IQ, sex) and the level of the literacy variables at pre-test (block 2).

In order to investigate the link between the improvements in literacy skills and students' wellbeing at pre-test, we performed three, one for each literacy variable ( $\Delta$  T1-T2: reading speed, reading accuracy, writing) three-step, fixed entry, multiple regression analysis only on SOM group.

Finally, descriptive statistics on the feedback questionnaire were conducted in order to assess children's appreciation of the training, potentials elements of criticalities and suggestions for improvements.

## **2.6. Results**

First, we report below the analyses corresponding to our main hypothesis, which concerns the impact of training from pre-test at T1 to post-test at T2. We then consider whether any effects of training may be long lasting by comparing the pre-test at T1 to our follow-up session 6 months later at T3.

We consider the impact of training separately on literacy skills and executive functions. Within literacy, reading speed and accuracy plus writing and comprehension measures were considered; within executive functions, attention, short-term memory and planning were considered separately.

### **T1-T2**

#### **LITERACY SKILLS**

*A) Speed of Reading.* A 2x2x3 MANCOVA with Time (T1, T2) and Measure (Word, Non-word, Text) as within-subject factors, and Group as between-subject factor was carried out. A significant Time effect was observed ( $F(1,145)= 6.485$ ,  $p = 0.012$ ,  $\eta^2_p = 0.043$ ), but more

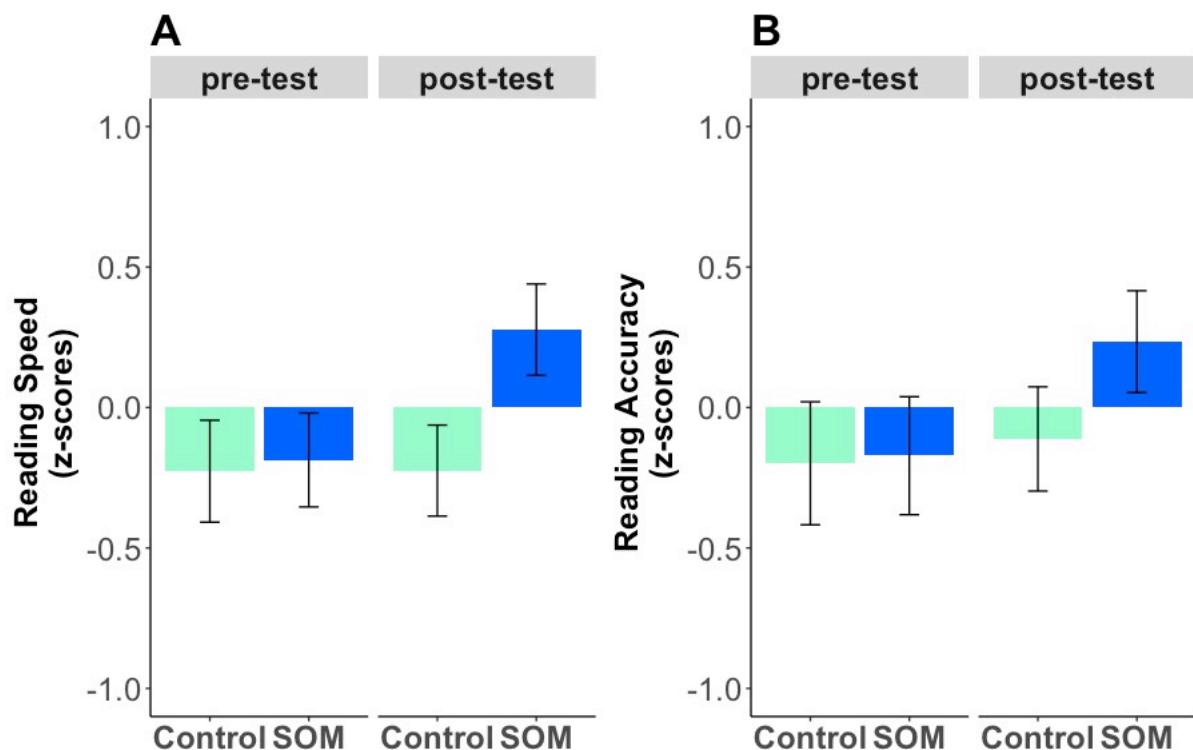
importantly a Time X Group interaction [ $F(1, 145) = 53.869, \eta^2_p = 0.271, p < 0.001$ ] was found indicating greater improvement after training in reading speed in the SOM group than in the control group. Post-hoc analyses confirmed that children in the SOM group significantly improved their reading time ( $p < 0.0001$ ), whereas the Control group did not ( $p=0.967$ ). A significant triple interaction Time X Group X Measure [ $F(2, 144) = 5.352, \eta^2_p = 0.069, p = 0.006$ ] reflected that children in the control group displayed worse text reading speed at T2 ( $\Delta T1-T2: -0.23, SD= 0.75; p = 0.013$ ), while they significantly improved their word reading speed ( $\Delta T1-T2 : 0.23, SD= 0.59; p = 0.044$ ).

Taking into consideration the effects of the covariates, a significant interaction between Time and Age [ $F(1, 145) = 9.744, \eta^2_p = 0.063, p = 0.002$ ] revealed that, regardless of the group, younger children showed higher results at the post-test evaluation.

*B) Accuracy of Reading.* Analogously, when considering reading accuracy, the Time X Group interaction was significant [ $F(1, 145) = 17.645, \eta^2_p = 0.108, p < 0.001$ ], with improvements observed in SOM group only (post-hoc analyses revealing a significant difference between T1 and T2 measures in SOM  $p < 0.001$ , but not in the control group  $p=0.934$ ). No group differences between the three reading measures at T2 were observed [ $F(2, 144) = 0.091, \eta^2_p = 0.001, p = 0.913$ ].

Results of reading speed and accuracy are presented in Figure 41.

**Figure 41|** Performance in reading speed (A) and accuracy (B), expressed in z-scores, were measured before (T1) and after (T2) SOM and Control trainings. Only SOM group improved significantly after training. Error bars represent confidence intervals (CI).



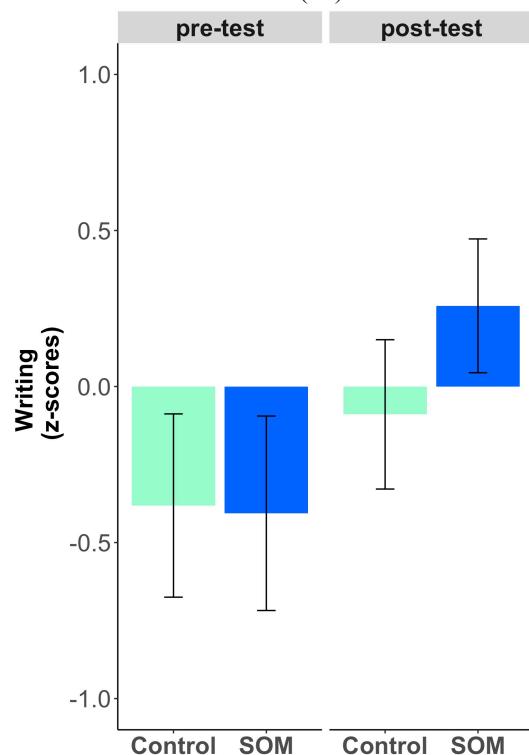
*Writing.*

A MANCOVA with Time as within-subject factor and Group as between-subject factor was carried out on the two measures of writing inserted as a within-subject factor (Measure: Word Writing, Non-word Writing). A significant Time effect was observed ( $F(1,145)= 32.627$ ,  $p < 0.012$ ,  $\eta^2_p = .184$ ). More importantly, a significant difference between SOM and Control group on writing capacities over time [Time X Group  $F(1, 145) = 9.691$ ,  $\eta^2_p = 0.063$ ,  $p < 0.002$ ] was also found. Post-hoc comparisons revealed differences between T1 and T2 for both groups (SOM group  $p < 0.0001$ , control group  $p = 0.001$ ).

Taking into consideration the effects of the covariates, significant interactions between Time and IQ [ $F(1, 145) = 8.807$ ,  $\eta^2_p = 0.057$ ,  $p = 0.004$ ] and Time and Age [ $F(1, 145) = 15.062$ ,  $\eta^2_p = 0.094$ ,  $p < 0.001$ ] revealed that, regardless of the group, children of third grade and with lower IQ at pre-test showed higher results at the post-test evaluation.

Results of writing are presented in Figure 42.

**Figure 42|** Writing performance, expressed in z-scores, was measured before (T1) and after (T2) SOM and Control trainings. Only SOM group statistically improved after training. Error bars represent confidence intervals (CI).

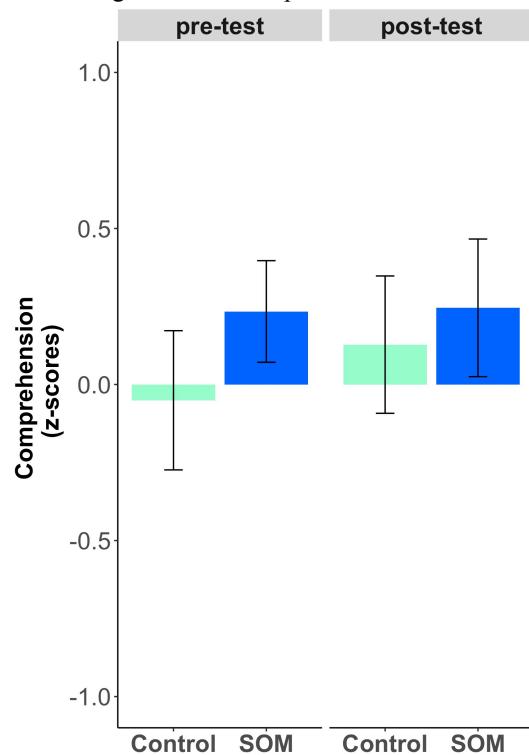


*Comprehension.* Text comprehension improvements were analysed using a 2 (Time: T1, T2) X 2 (Group: SOM, Control) analysis of Covariance (ANCOVA). The results demonstrated no differences in the effects of the training with regard to Text comprehension [Time X Group

$$F(1, 145) = 1.127, \eta^2_p = 0.008, p = 0.290].$$

Results of comprehension are presented in Figure 3.

**Figure 43** Comprehension performance, expressed in z-scores, was measured before (T1) and after (T2) SOM and Control trainings. Error bars represent confidence intervals (CI).



**Table 15** Training effects of SOM group ( $n = 79$ ) and control group ( $n = 71$ ) on literacy skills performance [Means ( $SD$ )]. In addition, the delta scores (T2-T1) are reported for both groups.

<sup>a</sup> Time (T1–T2) X Group interaction.

<sup>c</sup> Test significance. \* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$ .

<sup>d</sup>Effect sizes of interaction Time : Group expressed as partial eta squared.

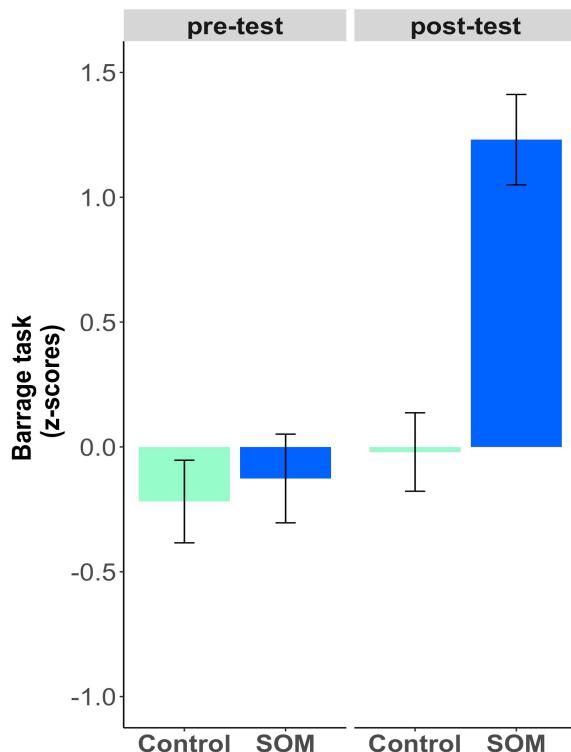
	SOM			CONTROL			<i>F</i> -score <sup>a</sup>	<i>p</i> <sup>c</sup>	ES <sup>d</sup>
	T1	T2	$\Delta$	T1	T2	$\Delta$			
Reading Speed	-0.19 (0.65)	0.28 (0.63)	0.46 (0.37)	-0.23 (0.57)	-0.22 (0.50)	0.00 (0.43)	53.86	<0.001	0.27
Reading Accuracy	-0.14 (0.56)	0.17 (0.47)	0.31 (0.44)	-0.10 (0.54)	-0.09 (0.41)	0.00 (0.46)	17.64	<0.001	0.11
Writing	-0.41 (1.39)	0.26 (0.96)	0.66 (0.89)	-0.38 (1.25)	-0.09 (1.01)	0.29 (0.72)	9.69	0.002	0.06
Comprehension	0.23 (0.73)	0.25 (0.98)	0.01 (0.93)	-0.05 (0.95)	0.13 (0.94)	0.18 (1.13)	1.13	0.290	0.01

## EXECUTIVE FUNCTIONS

*Visuo-spatial Attention.* A 2x2x2 MANCOVA with Time (T1, T2) and Outcomes (fast and slow score) as within-subject factor, and Group as between-subject factor was carried out. A significant Time effect was observed ( $F(1,145)= 4.713, p = 0.032, \eta^2_p = .031$ ), but more importantly a Time X Group interaction [ $F(1, 145) = 53.869, \eta^2_p = 0.271, p <0.001$ ] was found highlighting a larger gain in SOM group compared to Control group (post-hoc analyses revealing a significant difference between T1 and T2 measures in SOM  $p < 0.0001$ , but not in the control group  $p=0.073$ ). No Time X Group X Outcomes interaction was found significant [ $F(1, 145) = 0.137, \eta^2_p = 0.001, p = 0.711$ ] showing, thus, no differences between the two scores (fast and slow score). Results of visuo-spatial attention are presented in Figure 44.

Taking into consideration the effects of the covariates, a significant interaction Time X Outcome X IQ [ $F(1, 144) = 4.294, \eta^2_p = 0.04, p = 0.029$ ] was also found revealing that children with higher IQ (above 100) improved more in the fast score of the barrage task ( $\Delta$  T1-T2: 1.03, SD= 1.06;  $p < 0.001$ ).

**Figure 44|** Visuo-spatial performance, expressed in z-scores, was measured before (T1) and after (T2) SOM and Control training. Only SOM group statistically improved after training. Error bars represent CI.



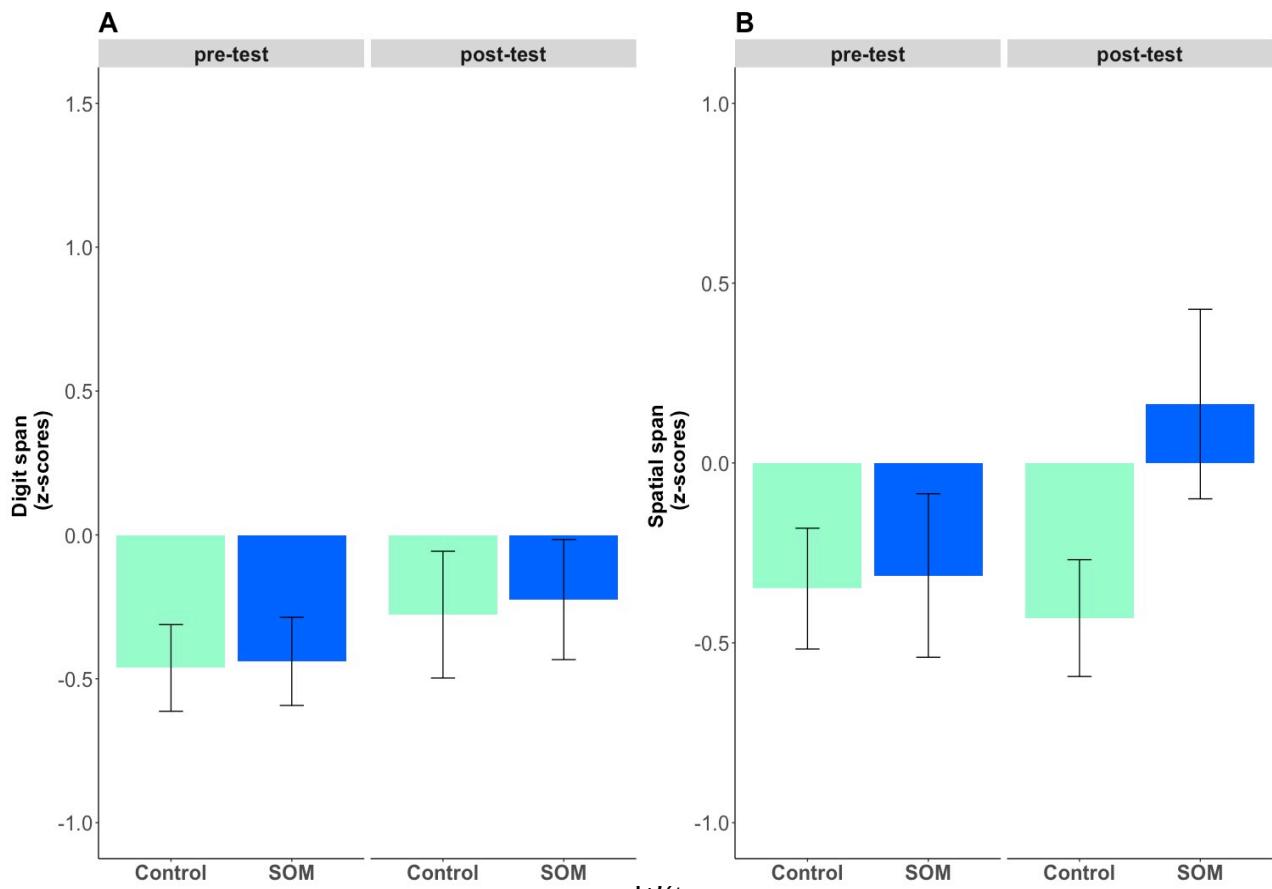
*Verbal and visuo-spatial memory.* Improvements in short-term memory were analysed by means of a 2 (Time: T1, T2) X 2 (Group: SOM, Control) X 4 (Task: Digit span Forward,

Digit span Backward, Visuo-spatial span Forward, Visuo-spatial span Backward) analysis of Covariance (MANCOVA). The results showed that, after training, there were greater improvements in memory performance in the SOM group than in the control group [Time X Group  $F(1, 145) = 5.832$ ,  $\eta^2_p = 0.039$ ,  $p = 0.017$ ]. Post-hoc comparisons revealed improvements in memory for children trained with SOM only ( $p = 0.009$ , control group  $p = 0.173$ ).

However, it has to be noted a marginally significant triple interaction Time X Group X Task [ $F(3, 143) = 2.528$ ,  $\eta^2_p = 0.050$ ,  $p = 0.06$ ]. Specifically, univariate tests indicated there was a significant training effect on the visuo-spatial span, as indicated by a significant Time X Group interaction [ $F(1, 145) = 6.110$ ,  $\eta^2_p = 0.039$ ,  $p = 0.015$ ] and by subsequent post-hoc comparisons (significant improvements for children in SOM group  $p = 0.001$ , but not for children in the control group  $p=0.344$ ). In contrast, assessments of verbal memory (Digit span forward and backward) before and after training revealed no significant differences between the two groups (post-hoc comparisons: SOM  $p = 0.029$ ; control  $p = 0.068$ ).

Results of verbal and visuo-spatial short term and working memory are presented in Figure 45.

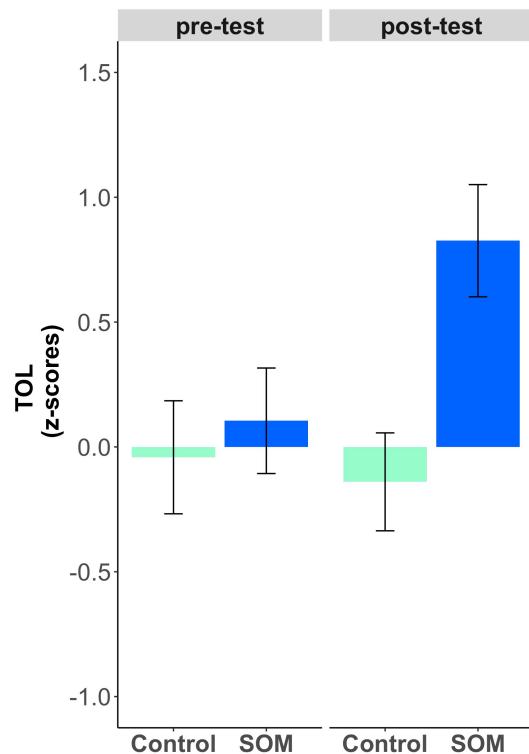
**Figure 45|** Performances in verbal and visuo-spatial working memory, expressed in z-scores, were measured before (T1) and after (T2) SOM and Control trainings. Error bars represent confidence intervals (CI).



*Planning.* Finally, an ANCOVA with Time as within-subject factor and Group as between-subject factor was carried out on the planning measure (Tower of London task). Significant differences between training conditions were again observed [Time X Group interaction,  $F(1, 145) = 26.982$ ,  $\eta^2_p = 0.157$ ,  $p < 0.0001$ ]. Post-hoc comparisons highlighted an enhancement in planning for children trained with SOM only ( $p < 0.0001$ , control group  $p = 0.311$ ). These improvements, however, were driven by improvements in females. Indeed, a significant triple interaction Time X Group X Sex was found [ $F(1, 145) = 7.852$ ,  $\eta^2_p = .051$ ,  $p = 0.006$ ], with females in SOM group presenting large gains in planning ( $p < 0.0001$ ), whereas females in the control group did not ( $p = 0.123$ ). Performance in TOL improved for males of the SOM group ( $p = 0.027$ ), but not for the one in the control ( $p = 0.688$ ).

Results of planning are presented in Figure 46.

**Figure 46|** Planning performance, expressed in z-scores, was measured before (T1) and after (T2) SOM and Control trainings. Only SOM group statistically improved after training. Error bars represent confidence intervals (CI).



**Table 16|** Training effects of SOM group ( $n = 79$ ) and control group ( $n = 71$ ) on executive functions performance [Means ( $SD$ )]. In addition, the delta scores (T2-T1) are reported for both groups.

<sup>a</sup>Time (T1-T2) X Group interaction.

<sup>c</sup>Test significance. \* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$ .

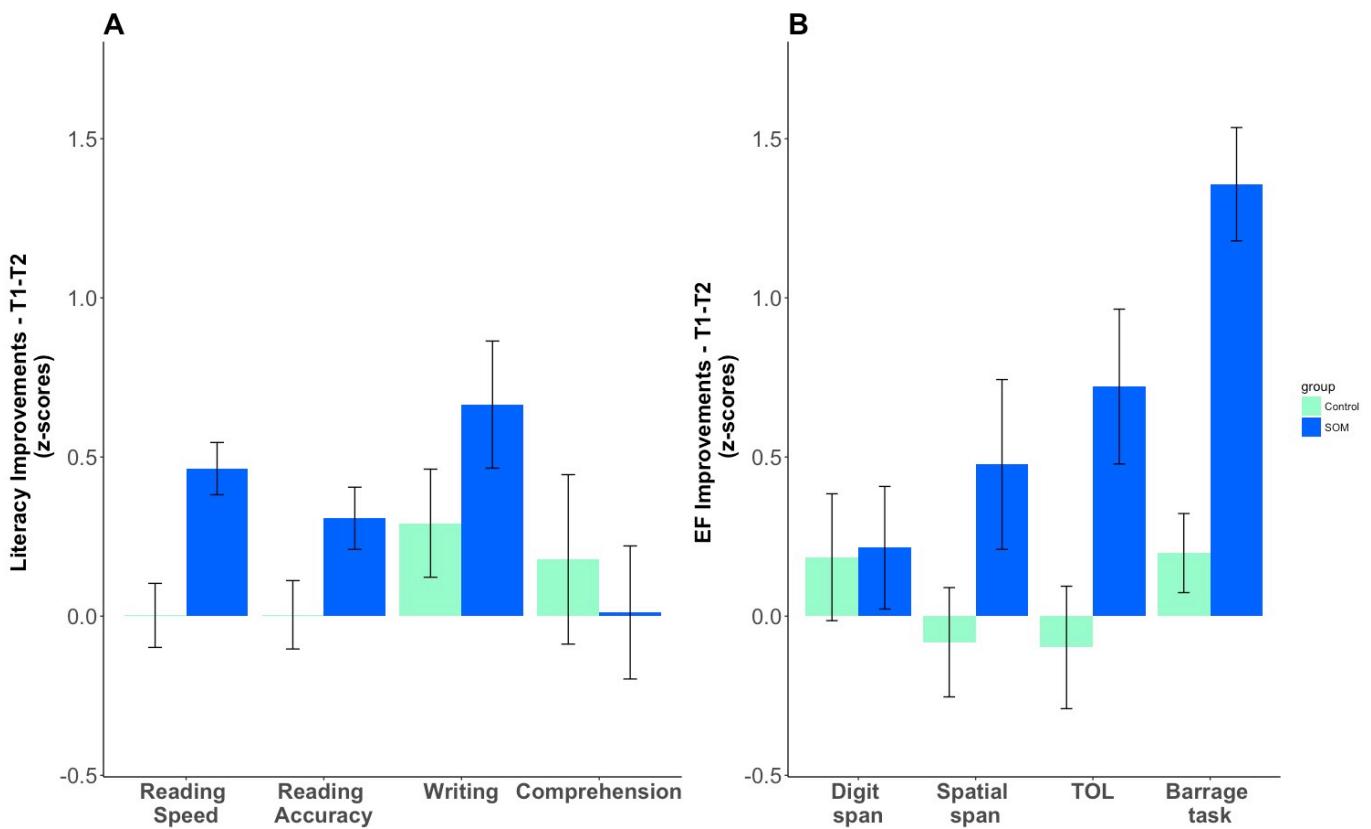
<sup>d</sup>Effect sizes of interaction Time : Group expressed as partial eta squared.

	SOM			CONTROL			<i>F</i> -score <sup>a</sup>	<i>p</i> <sup>c</sup>	ES <sup>d</sup>
	T1	T2	Δ	T1	T2	Δ			
Bells (barrage) task	-0.13 (0.79)	1.23 (0.81)	1.36 (0.80)	-0.22 (0.70)	-0.02 (0.67)	0.20 (0.53)	109.23	<0.001	0.43
Digit+spatial span	-0.38 (0.69)	-0.03 (0.87)	0.35 (0.80)	-0.41 (0.51)	-0.35 (0.61)	0.05 (0.59)	5.83	0.017	0.04
TOL	0.10 (0.94)	0.83 (1)	0.72 (1.09)	-0.04 (0.96)	-0.14 (0.83)	-0.10 (0.82)	26.98	<0.001	0.16

In addition, the means of the delta score (difference in the performances between post-training and pre-training) in literacy skills and executive functions, obtained by the two groups, can be found in Figure 47.

**Figure 47|** Means of Δ scores (T1-T2) in literacy skills and executive functions, obtained by the two groups.

Error bars represent CI.



### T1-T3

In order to check for test-retest effects that can be caused by the use of the same paper-and-pencil tests through all the assessment sessions, at T3 we administered two additional tests: a

text-reading task and a barrage task. A Pearson correlation was run to determine the relationship between the BVN-text reading score and performance in the text-reading task at T3. There was a strong, positive correlation between the two text-reading performances, which was statistically significant ( $r = 0.653, n = 151, p < 0.001$ ). Furthermore, a strong correlation between the two barrage tasks was also found ( $r = 0.970, n = 151, p < 0.0001$ ). Finally, we examined whether the aforementioned improvements were maintained after 6 months, by means of Analyses of Covariance (ANCOVAs) or MANCOVAs in the case of multiple dependent variables.

## LITERACY SKILLS

### *Reading.*

*A) Speed of Reading.* Maintenance of the improvements in reading speed were analysed by means of a 2 (Time: T1, T2) X 2 (Group: SOM, Control) X 3 (Measure: Word, Non-word and Text reading) MANCOVA. A significant Time effect was observed ( $F(1, 145) = 7.024, p = 0.009, \eta^2_p = 0.046$ ), but more importantly a Time X Group interaction was significant [ $F(1, 145) = 53.360, \eta^2_p = 0.269, p < 0.001$ ]. Post-hoc comparisons showed that children in the SOM group maintained their advantage in reading fluency ( $p < 0.0001$ ), while children in the control group did not demonstrate a significant difference between the two assessment points ( $p = 0.743$ ). A significant triple interaction (Time X Group X Measure) was again observed [ $F(1, 144) = 6.995, \eta^2_p = 0.089, p = 0.001$ ] due to children in the control group displaying worse text reading speed at T3.

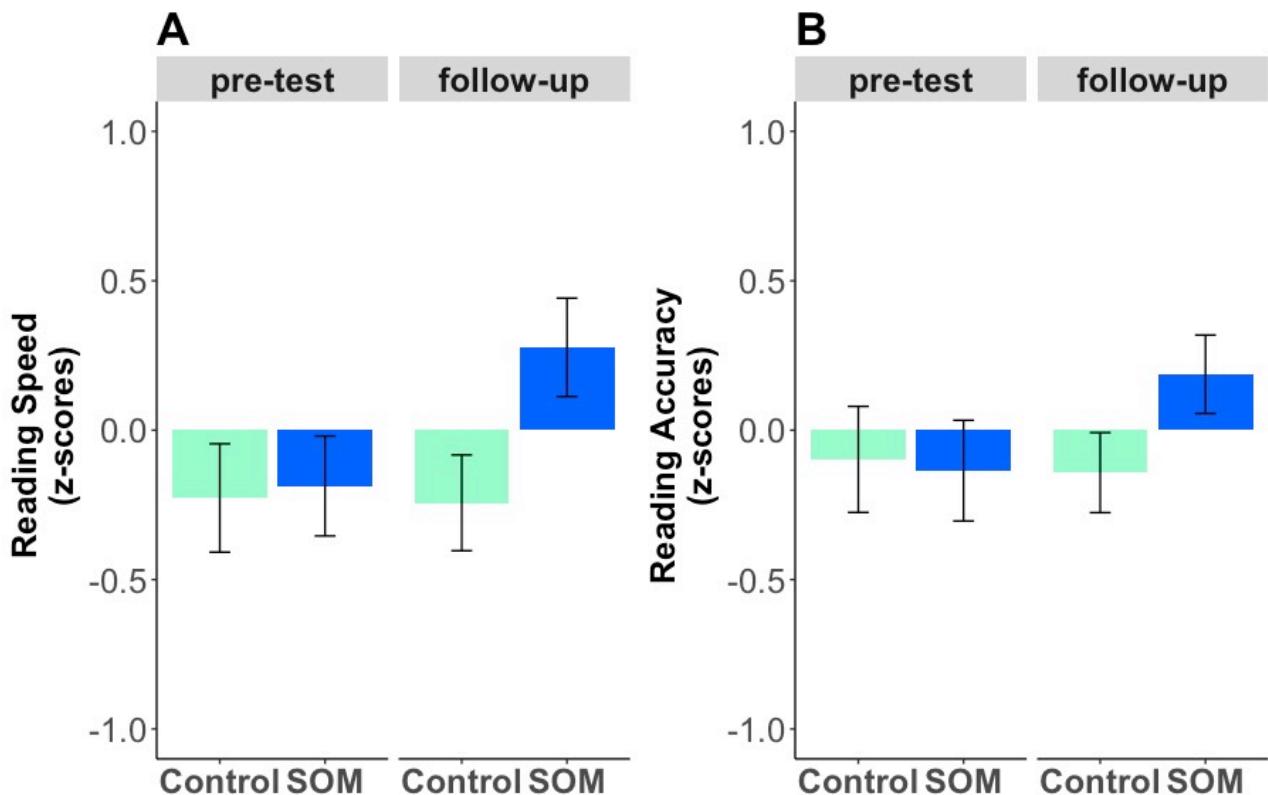
Taking into consideration the effects of the covariates, a significant interaction between Time and Age [ $F(1, 145) = 10.270, \eta^2_p = 0.066, p = 0.002$ ] revealed that, regardless of the group, younger children showed higher results at follow-up. In addition, results showed a significant Time X Measure X Age interaction [ $F(1, 144) = 4.549, \eta^2_p = 0.059, p = 0.012$ ], reflecting that only grade 3 children, displayed better reading speed at all the reading measures at T3 (word  $\Delta$  T1-T3: 0.38, SD= 0.66;  $p < 0.001$ ; non-word  $\Delta$  T1-T3: 0.30, SD= 0.53;  $p = 0.010$ ; text  $\Delta$  T1-T3: 0.44, SD= 0.64;  $p < 0.001$ ). Vice versa, older children (grade 4-6-7), regardless of the group, did not show statistically significant improvements in text reading ( $\Delta$  T1-T3: -0.03, SD= 0.82;  $p = 0.653$ ).

*B) Accuracy of Reading.* The same MANCOVA model was performed using the three measures of reading accuracy (Word, Non-word and Text reading) as dependent variables. Children in SOM group retained their gains in reading accuracy at follow-up, as pointed out by a significant Time X Group interaction [ $F(1, 145) = 28.084, \eta^2_p = 0.162, p < 0.001$ ] and

by subsequent post-hoc comparisons (significant differences for children in SOM group  $p < 0.0001$ , but not for children in the control group  $p=0.413$ ).

Results of reading speed and accuracy are presented in Figure 48.

**Figure 48** Performances in reading speed (A) and accuracy (B), expressed in z-scores, were measured before (T1) and 6 months after the end of the SOM and Control trainings (T3). Error bars represent confidence intervals (CI).



**Table 17** Training effects of SOM group ( $n = 79$ ) and control group ( $n = 71$ ) on literacy skills performance [Means ( $SD$ )]. In addition, the delta scores (T3-T1) are reported for both groups.

<sup>a</sup>Time (T1–T3) : Group interaction.

<sup>c</sup>Test significance. \* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$ .

<sup>d</sup>Effect sizes of interaction Time : Group expressed as partial eta squared.

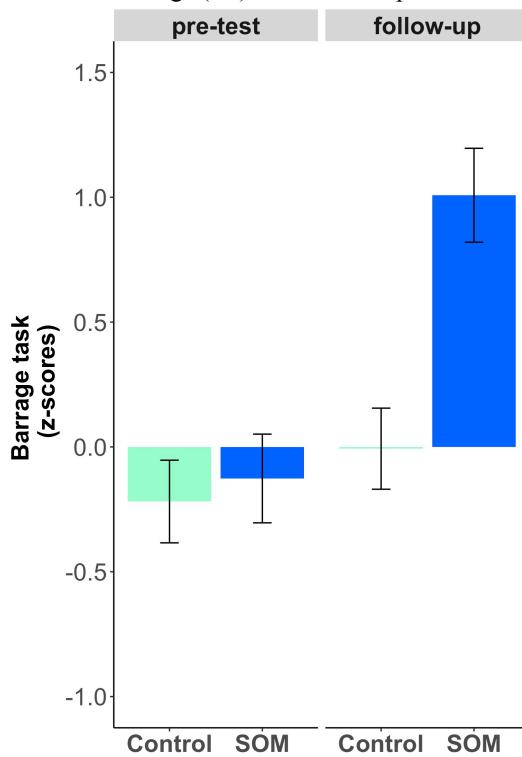
	SOM			CONTROL			$F$ -score <sup>a</sup>	$p$ <sup>c</sup>	ES <sup>d</sup>
	T1	T3	$\Delta$	T1	T3	$\Delta$			
Reading Speed	-0.19 (0.65)	0.28 (0.74)	0.46 (0.37)	-0.23 (0.57)	-0.24 (0.50)	-0.02 (0.44)	53.86	<0.001	0.27
Reading Accuracy	-0.14 (0.56)	0.19 (0.44)	0.32 (0.40)	-0.10 (0.54)	-0.14 (0.40)	-0.04 (0.45)	17.64	<0.001	0.11

## EXECUTIVE FUNCTIONS

*Visuo-spatial attention.* A 2x2x2 MANCOVA with Time (T1, T3) and Outcome (fast and slow score) as within-subject factors, and Group as between-subject factor was carried out. At follow-up, children in the SOM group confirmed higher performance in visuo-spatial attention, as indicated by significant Time X Group interactions [ $F(1, 145) = 58.758, \eta^2_p = 0.288, p < 0.0001$ ] and by subsequent post-hoc comparisons. (significant improvements for children in SOM group,  $p < 0.0001$ , but not for children in the control group,  $p = 0.095$ ). Taking into consideration the effects of the covariates, a significant interaction Time X Outcome X IQ [ $F(1, 144) = 4.311, \eta^2_p = 0.04, p = 0.029$ ] was also found, again showing that children with higher IQ (above 100) improved more in the fast score of the barrage task ( $\Delta T1-T3: 0.91, SD= 1.02; p < 0.001$ ).

Results of visuo-spatial attention are presented in Figure 49.

**Figure 49|** Visuo-spatial performance, expressed in z-scores, was measured before (T1) and 6 months after the end of the SOM and Control trainings (T3). Error bars represent confidence intervals (CI).

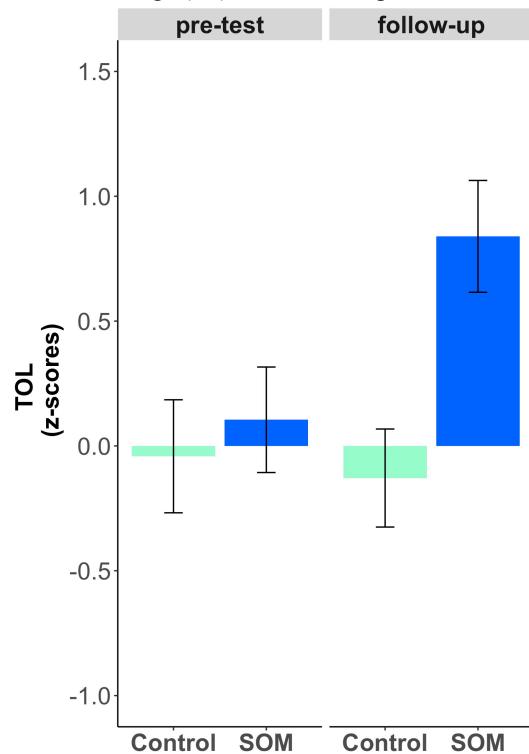


*Planning.* Maintenance of the training effects in planning skills were analysed using an ANCOVA with Time as within-subject factor and Group as between-subject factor. Comparing T1-T3, significant differences between the two groups were again observed [Time X Group interaction,  $F(1, 145) = 27.312, \eta^2_p = 0.159, p < 0.0001$ ]. Post-hoc comparisons confirmed higher gains for SOM group after 6 months ( $p < 0.0001$ , control

group  $p = 0.369$ ) compared to the control group.

Specifically only Female in the SOM group obtained and maintained better results in comparison with Control group [triple interaction Time X Group X Sex was found  $F(1, 145) = 7.905, \eta^2_p = 0.052, p = 0.006$ ]. Indeed, females in SOM group showed higher performances at follow-up ( $p < 0.0001$ ), whereas females in the control group did not ( $p = 0.149$ ). Performance in TOL improved for males of the SOM group ( $p = 0.023$ ), but not for the one in the control ( $p = 0.647$ ). Results of planning are presented in Figure 10.

**Figure 50|** Planning performance, expressed in z-scores, was measured before (T1) and 6 months after the end of the SOM and Control trainings (T3). Error bars represent confidence intervals (CI).



**Table 18|** Training effects of SOM group ( $n = 79$ ) and control group ( $n = 71$ ) on executive functions performance [Means ( $SD$ )]. In addition, the delta scores (T2-T1) are reported for both groups.

<sup>a</sup> Time (T1–T3) : Group interaction.

<sup>c</sup> Test significance. \* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$ .

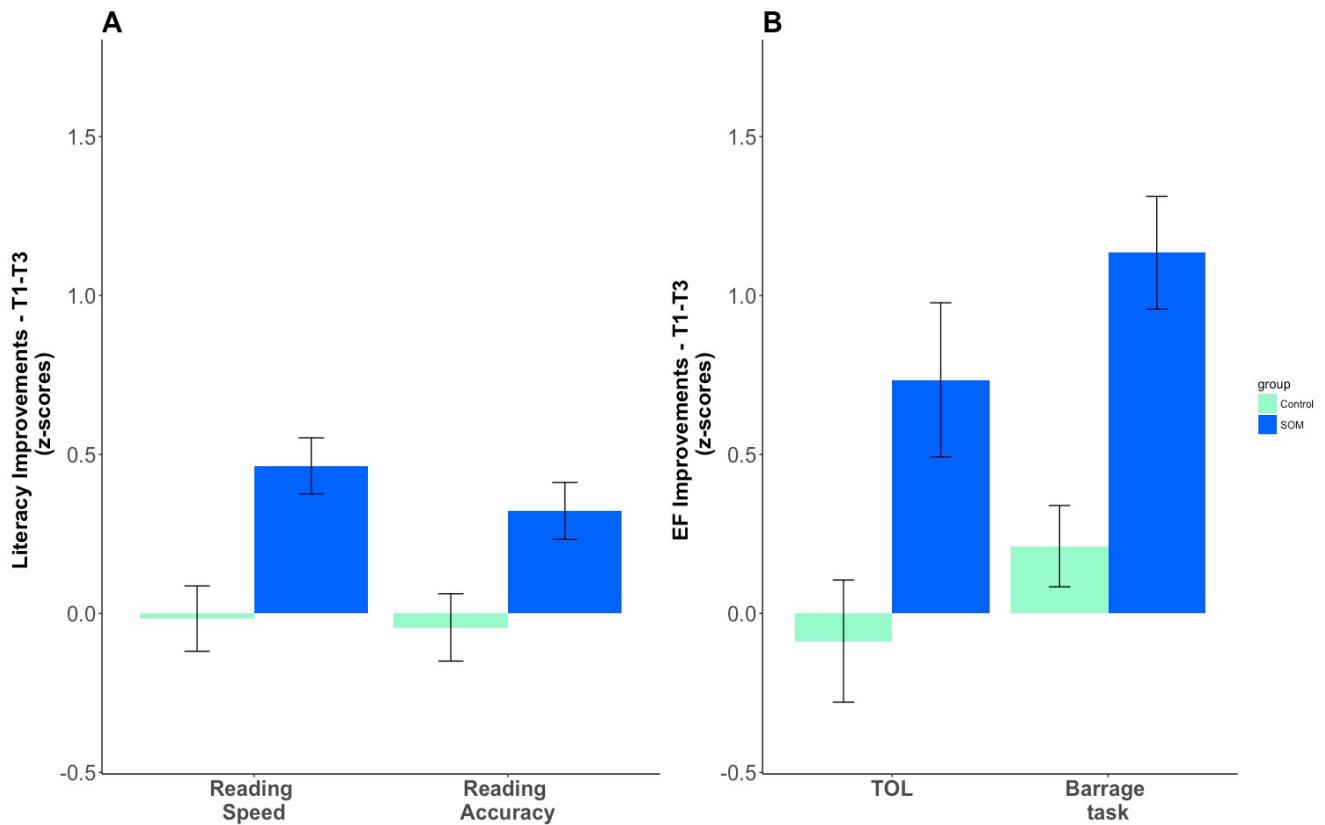
<sup>d</sup>Effect sizes of interaction Time : Group expressed as partial eta squared.

	SOM			CONTROL			<i>F</i> -score <sup>a</sup>	<i>p</i> <sup>c</sup>	ES <sup>d</sup>
	T1	T3	Δ	T1	T3	Δ			
Bells (barrage) task	-0.13 (0.79)	1.01 (0.84)	1.13 (0.79)	-0.22 (0.70)	-0.01 (0.69)	0.21 (0.54)	109.23	<0.001	0.43
TOL	0.10 (0.94)	0.84 (0.99)	0.73 (1.08)	-0.04 (0.96)	-0.13 (0.84)	-0.09 (0.82)	26.98	<0.001	0.16

Finally, the means of the delta score (difference in the performances between the follow-up and the baseline) in literacy skills and executive functions, obtained by the two groups, are presented in Figure 51.

**Figure 51|** Means of  $\Delta$  scores (T1-T3) in literacy skills and executive functions, obtained by the two groups.

Error bars represent CI.



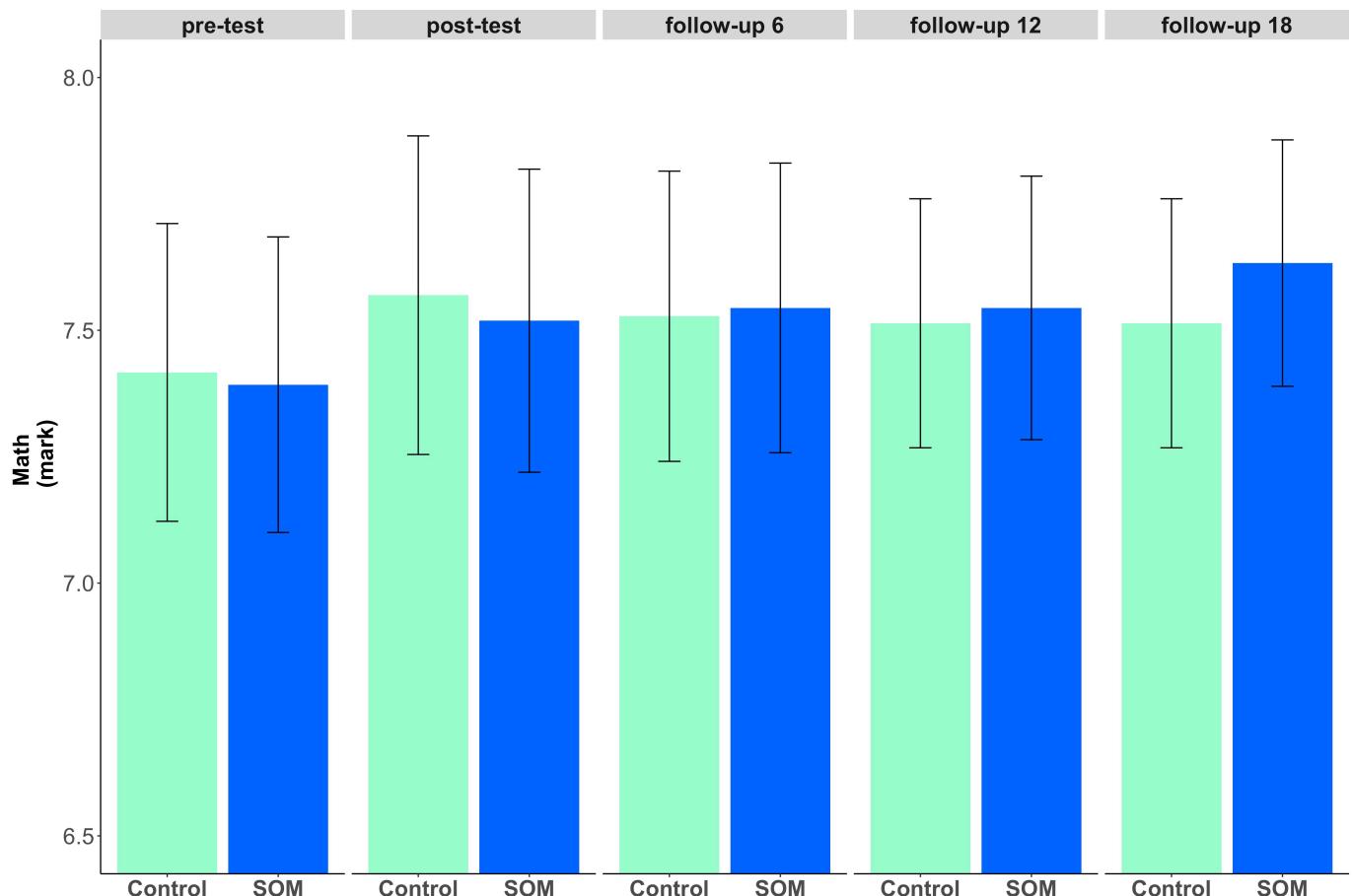
### *Academic performance*

The generalization of training effects to two important academic domains, such as Mathematics and Italian performance, were analysed by means of two ANCOVAs.

*Mathematics.* A 5x2 ANCOVA with Time (pre-training, post-training, follow-up 6, 12 and 18 months later) as within-subject factor, and Group as between-subject factor was carried out. No significant main effect of Time [ $F(4, 142) = 1.649, \eta^2_p = 0.011, p = 0.160$ ] and Time X Group interaction [ $F(4, 142) = 0.778, \eta^2_p = 0.005, p = 0.54$ ] were found, indicating no significant differences between the two groups at the different time points. As regards the covariates effect, a significant Time X Age interaction was observed [ $F(4, 142) = 5.683, \eta^2_p = 0.038, p < 0.001$ ].

Results of performance in Mathematics in the five time points are presented in Figure 52.

**Figure 52** Performance in Mathematics, measured as the marks obtained by children of the two groups (SOM-Control) at the five time points (pre-training, post-training, follow-up 6, 12 and 18 months after the end of the training). Error bars represent confidence intervals (CI).



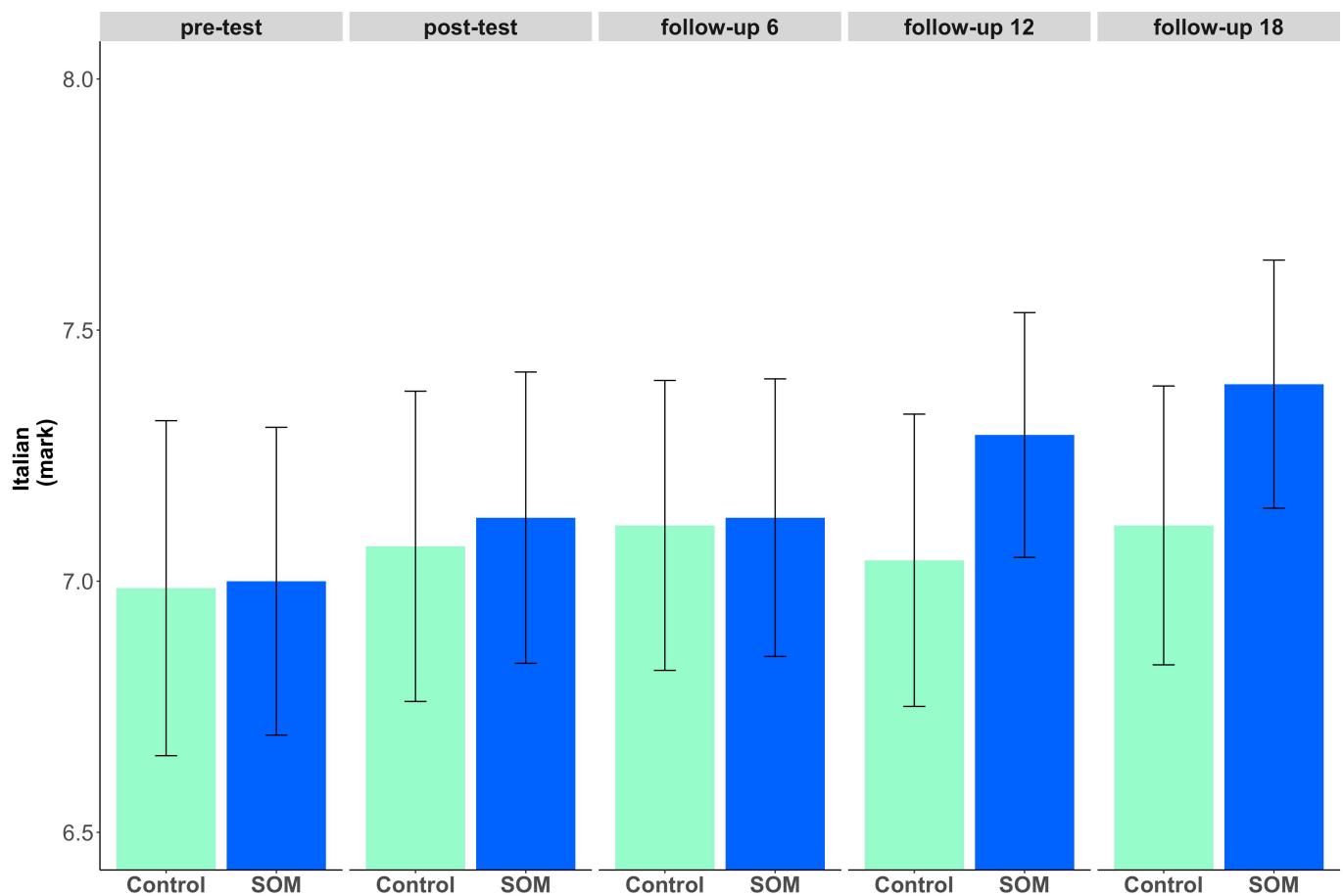
*Italian*. The ANCOVA model presented before was used also with the mark in Italian as dependent variable. Interestingly, a significant Time X Group interaction [ $F(4, 142) = 3.448$ ,  $\eta^2_p = 0.23$ ,  $p = 0.008$ ] was found indicating greater improvement in children of the SOM group in the performance in Italian compared to the children in the control group. Post-hoc analyses confirmed that, 18 months after the end of the training, children in the SOM group significantly improved their marks in Italian ( $p = 0.027$ ), whereas the Control group did not ( $p=0.967$ ). Taking into consideration the effects of the covariates, a significant interaction between Time and Age [ $F(1, 145) = 5.024$ ,  $\eta^2_p = 0.033$ ,  $p = 0.556$ ] revealed that, regardless of the group, younger children showed higher results at the post-test evaluation.

Three polyserial correlations were computed in order to assess the relationship between the improvements in literacy skills (speed of reading, accuracy of reading and writing) after training and the marks obtained in Italian by children in SOM group at the last time point (T5). Overall, there was a mild positive correlation between gains in reading speed (delta score T1-T2) and performance in Italian at the follow-up conducted 18 months after the end

of the training. This positive correlation ( $r = 0.215$ ,  $p = 0.047$ ) indicates that those children in SOM group who improved the most in reading fluency were the ones that showed the highest gains in Italian 18 months later.

Results of performance in Italian in the five time points are presented in Figure 53.

**Figure 53|** Performance in Italian, measured as the marks obtained by children of the two groups (SOM-Control) at the five time points (pre-training, post-training, follow-up 6, 12 and 18 months after the end of the training). Error bars represent confidence intervals (CI).



#### ***Link between Literacy and Executive functions improvements***

In order to investigate the link between the improvements between literacy skills and EFs we performed three, one for each literacy variable ( $\Delta T1-T2$ : reading speed, reading accuracy, writing) three-step, fixed entry, multiple regression analysis on the entire sample of participants. We took into consideration only the EFs variables for which a significant Time X Group interaction was found: planning, visuo-spatial working memory and attention. The delta score of planning was calculated using the difference between T1 and T2 performance in the “Tower of London” task, while the visuo-spatial working memory delta using the improvements (T2-T1) in the digit span-backward task. In addition, visuo-spatial attention

changes were indexed by calculating the mean between the delta scores ( $T_2 - T_1$ ) in fast and slow score of the barrage task.

*Reading speed.* The dependent variable was the reading speed changes between  $T_1$  and  $T_2$  ( $\Delta T_1 - T_2$  reading speed), and the predictors were: age, IQ, sex (block1); reading speed at  $T_1$  (block 2), EFs changes between  $T_1$  and  $T_2$  (block 3). These variables significantly predicted reading speed changes,  $F(7, 143) = 9.018, p < .0001, r^2 = 0.306$ , and only visuo-spatial attention changes added statistically significantly to the prediction,  $p < 0.001$ . However, we replicated this analysis on the two groups separately and results evidenced that link between reading speed improvements and visuo-spatial attentional improvements are entirely due to the fact that the groups have different means at  $T_2$ . Considering the SOM group only, in fact, only reading speed at  $T_1$  significantly predict reading speed changes [ $F(4, 74) = 3.268, p = 0.016, r^2 = 0.150$ ], while the EFs variable did not [ $F(7, 71) = 1.875, p = 0.086, r^2 = 0.156$ ] as showed by a non significant R-squared change ( $F(3, 71) = 0.165, p = 0.920$ ).

*Reading accuracy.* Consistently, results of the regression were significant when we considered the entire sample [ $F(7, 143) = 15.849, p < .0001, r^2 = 0.437$ ], but not when the analysis was performed on the two groups separately. Results for SOM group highlighted a predictive role of reading speed at pre-test [ $F(4, 74) = 13.430, p < .0001, r^2 = 0.421$ ], but not of the EFs variables [r-squared change: 0.002; ( $F(3, 71) = 0.064, p = 0.979$ )].

*Writing.* Analogously, these variables significantly predicted writing changes,  $F(7, 143) = 18.282, p < .001, r^2 = 0.472$ , only when the entire sample was took into consideration. Considering the SOM group only, only reading speed at  $T_1$  significantly predict reading speed changes [ $F(4, 74) = 21.751, p < 0.001, r^2 = 0.540$ ], while the EFs variable did not [r-squared change: 0.025; ( $F(3, 71) = 1.347, p = 0.266$ )].

Therefore, when considering SOM group only, none of The EFs changes significantly accounted for a significant quote of variance of literacy variables changes.

### ***Link between literacy improvements and students' well-being at pre-test***

We analysed the predictive role of wellbeing levels of SOM children at  $T_1$  (satisfaction in the school environment, emotional attitude towards school, sense of self-efficacy in literacy skills changes ( $\Delta T_1 - T_2$  reading speed and accuracy, writing)). After controlling for age, IQ and sex (block 1) and the literacy variable at  $T_1$  (block 2) by using a series of three-step fixed-entry multiple regression analysis, students' well-being at pre-test ( $T_1$ ) entered last significantly predicted improvements only in reading accuracy: [ $F(7, 69) = 8.682, p < 0.001, r^2 = 0.468$ ] Specifically, only initial levels of children' sense of self-efficacy added statistically

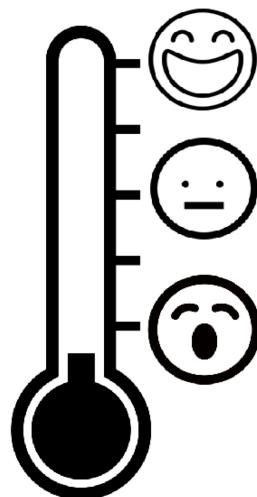
significantly to the prediction ( $p = 0.038$ ), indicating that children who started with lower levels of self-efficacy improved more in reading accuracy after training.

### ***Feedback questionnaire***

Descriptive analyses of the feedback questionnaire demonstrated that the training was well received: 82% of the children ( $n=79$ ) indicated that they liked to play with SOM, while 75.5% ( $n=72$ ) enjoyed the activities with Scratch. The “Fun-o-meter” (see Figure 54), a captivating Likert scale from 1 to 5 for the evaluation of fun - recorded a score of 4.57 out of 5 for SOM and a 3.99 for Scratch. An unpaired t-test revealed no statistically significant difference between the two questionnaires ( $p$ -value = 0.243). Similarly, there were no statistically significant differences in the level of appreciation between males and females in both groups ( $p$ -value = 0.451).

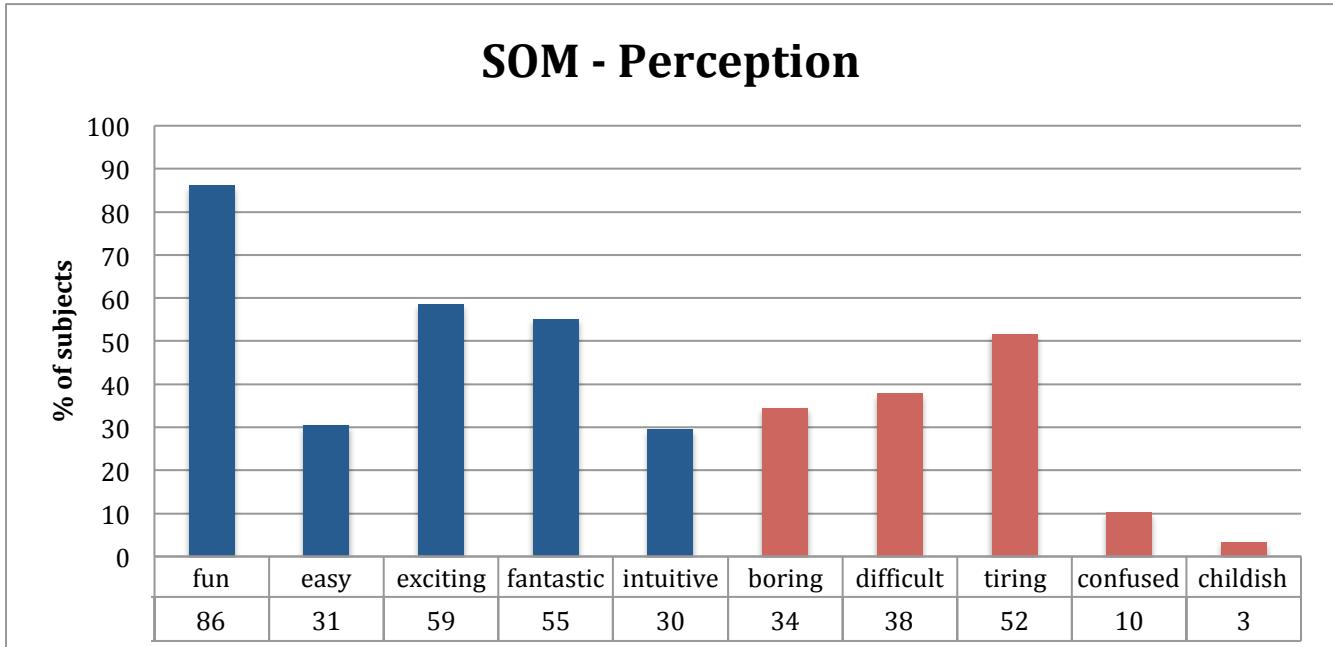
**Figure 54|** The “funometer”.

The statement “It was super-fun” corresponds to the fifth point of the Likert scale, while “It was boring” to zero.



The general perception (*How would you define this game?*) was positive in both groups. The appreciation of the game decreased with age, with the older children choosing an almost equal amount of positive and negative items. Additionally, the majority of the children judged the difficulty level of the training as appropriate (79.4% SOM; 81.6% Scratch). Both Skies of Manawak and Scratch were considered relatively difficult, but nonetheless challenging and not too boring.

**Figure 55** Overall positive and negative items of the perception question on Skies of Manawak.



Regarding SOM, many children highly appreciated the aesthetics (91.4%) and would have suggested the game to a friend (87.9%). Furthermore, a high percentage (about 95%) expressed a strong desire to play a second episode of the adventure. Table 19 shows the questions and the average response.

**Table 19** Questions and related means of answers on the appreciation of Skies of Manawak.

Question	Mean and SD of participants answers (0 = no/never, 2 = yes/often)
Would you suggest it to a friend?	1.67 (0.45)
If you could take it home, would you like to play it?	1.53 (0.29)
Would you like to play to a second episode?	1.94 (0.83)
Was it difficult?	0.78 (0.89)
Did you like the graphics?	1.74 (0.21)
While you were playing, were you curious about what could happen next?	1.79 (0.33)
Was it challenging?	1.41 (0.58)
Did you feel bored?	0.64 (0.75)
Compared to the beginning, do you feel to be more	1.89 (0.24)

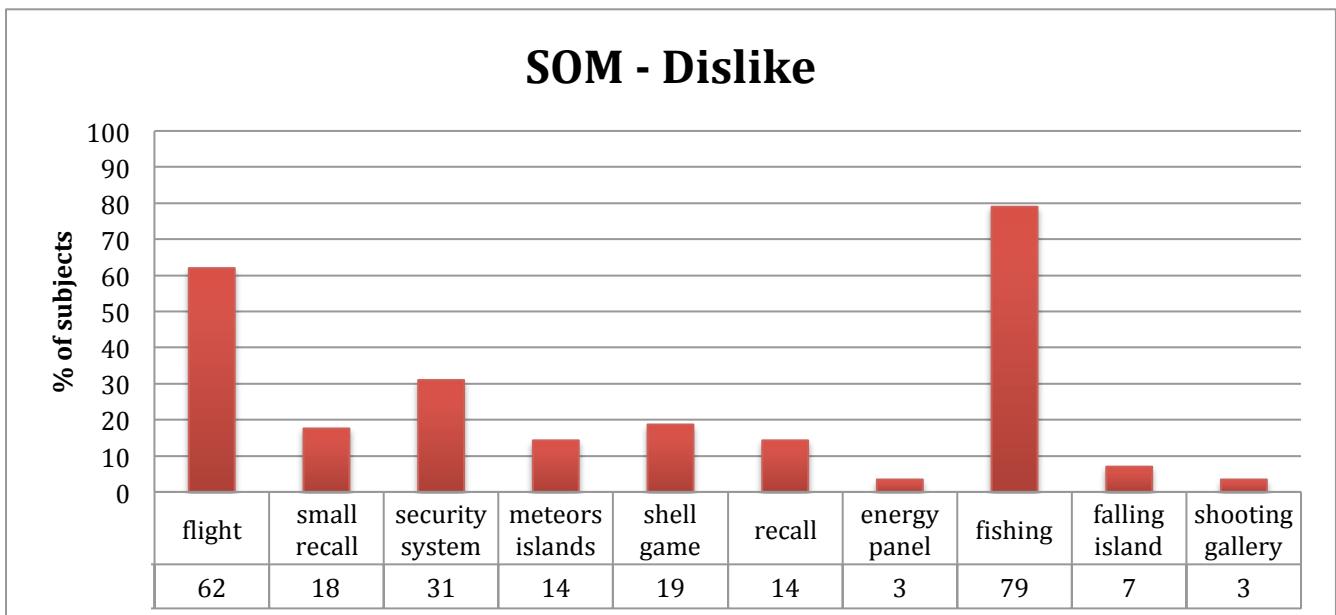
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competent in playing this game?

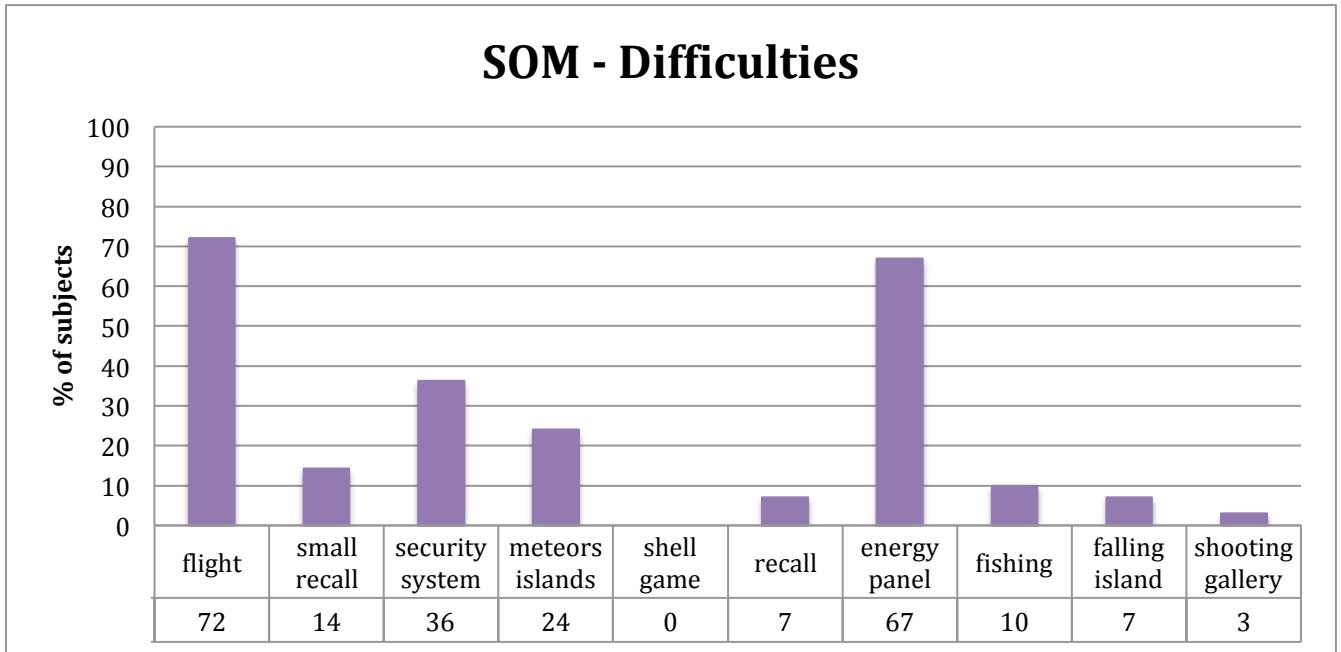
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With respect to the mini-games, only the Fishing session was particularly disliked (41%) (for details, see Figure 56). Despite it shares similar mechanics with the Shell game, the latter was not perceived negatively by most of the students and this could be due to differences in the settings. In fact, while the Fishing mini-game is activated every time the player loses all the health points during the flight sessions, the Shell game was set on a secret island and was particularly appreciated due the mystery and secrecy that enriched the overall experience. In addition, the less skilled players – with lower level of attention skills - ended up playing the Fishing more frequently, probably disrupting the flow of the game session. A significant correlation between the players disliking the Flight mini-game and identifying it as one of the most difficult was found ( $t = \text{inf}$ ,  $df = 88$ ,  $p\text{-value} < 1.2\text{e-}16$ ,  $\text{cor} = 0.96$ ). Figure 57 shows to the most difficult mini-games.

**Figure 56|** Overall answers to the most disliked mini-games.



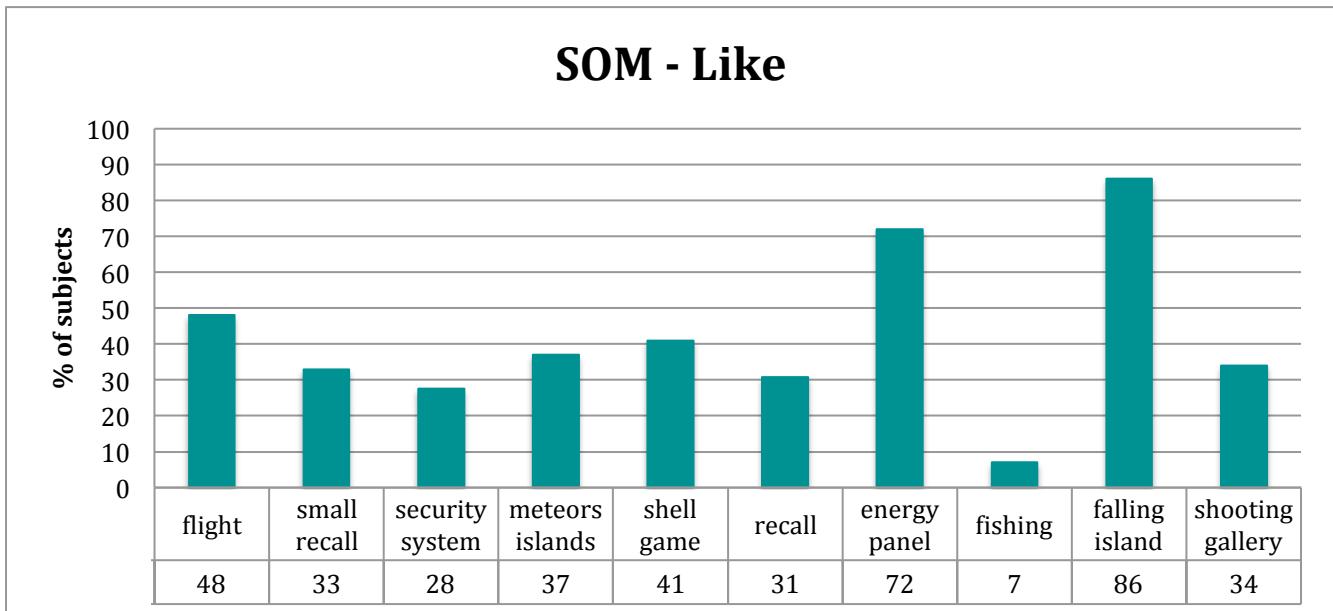
**Figure 57** Overall answers to the most difficult mini-games.



The mini-games that the players liked the most were the flight (50%) and the energy panel (70%), as shown in Figure 58. Particularly, males (88%) appreciated the flight mostly due to its variety (e.g., several types of enemies) and the type of challenge, less tied to cognitive exercises and more similar to action video games. Vice versa, females (91%) where the ones that liked most the energy panel, allegedly as a result of the possibility to forecast while thinking of different scenarios (and how to react appropriately to them). In general, both males and females appreciated the mini-game for the freedom to experiment their own solutions, but males tended to apply more of a trial-and-error approach.

To further explore the issue of a higher females' appreciation of the mini-game, a point-biserial correlation was run to determine the relationship between the appreciation of the planning mini-game in females (1 means that the mini-game was considered the favourite one, 0 means that the mini-game was not selected as the favourite) and improvements in the planning performance at T2 as measured with the Tower of London task. Results showed a mild, yet significant, positive correlation between the two measures ( $n = 39$ ,  $r_{pb} = .21$ ,  $p = 0.024$ ).

**Figure 58** Overall answers to the most liked mini-games.



## 2.7. Final remarks

As expected given the specific focus of Skies of Manawak in training attention, short-term memory and planning, major improvements in these areas were observed following 12h of game play, whereas no such changes were reported in the control, Scratch playing group.

Moreover, assessment upon training completion indicated improvements not only in attention and EFs, but also in reading and writing skills, after SOM yet not after the control training Scratch. Crucially, the advantages in EFs and literacy performance were maintained at a followup test 6-months later.

### **3. STUDY 2 – part B**

#### **Method**

Given the promising results of the previous study of SOM training, in this second study we assessed the effects of this training regimen in children with dyslexia. Specifically, we expected that only the children in the SOM group would improve in executive functioning, and that these improvements would generalize in better reading skills.

##### ***3.1. Study design and participants***

30 Italian-speaking children (mean age 10.10; SD 1.47, range 7.5 –13 years; 10 Females) with dyslexia who had been referred by the ODFLab (Laboratorio di Osservazione Diagnosi e Formazione - Observation Diagnosis and Training Laboratory) of the Department of Psychology and Cognitive Sciences at the University of Trento (Italy) participated our study. A control group of 25 Italian-speaking children (mean age 10.24; SD 1.39, range 8–13 years; 8 Females) with dyslexia was recruited in the same school of Study 2 - part A (Veneto, North of Italy).

Children in the first group trained with Skies of Manawak (SOM) for a total of 6 weeks: 4 times per week, with training sessions of 30 minutes each. Participants attended two training sessions (at the beginning and in the middle) at the ODFLab, while, the rest of the time, they played alone in their home environment. Vice versa, children of the control group received the training on coding using *Scratch* (Resnick et al., 2009). The training sessions were held during school time for two hours a week, for 6 weeks. Participants in the active control group received a computer-based training on coding using. Training duration (6 weeks) and the total amount of training (12 hour) were matched across the two groups. In Figure 59, the study plan is represented as a flowchart.

In both cases, the technological requirements were twofold: Internet access to download the file containing the video game, the possibility of using a computer with adequate characteristics (i.e. version of Windows system equal or higher to Windows 7), a mouse and (suggested) headphones.

In addition, in order to be selected, children had to be diagnosed as dyslexic by a licenced psychologist or an infant neuropsychiatric, according to the ICD-10 and DSM-IV inclusion and exclusion criteria. Indeed, they had to show a marked reading delay – i.e., at least 1.8 SD below the normative data – in reading speed and/or accuracy in at least one of the three clinical measures - i.e. word, non-word and text reading - (Cornoldi et al., 1981; Sartori, Job,

Tressoldi, 1995). Additional inclusion criteria were: (1) absence of sensorial, neurological and emotional deficits; (2) absence of comorbidity with other developmental disorders (e.g. ADHD); (3) non-verbal IQ within or above the normal limits, measured by the WISC-IV scale (WISC-IV; Wechsler, 2003; Italian version, 2012).

Parents were informed of the research procedure and authorized the participation. Both parents and children were unaware of the aim of the training to avoid differences in the expectations. Participants appeared to be highly motivated to play either with SOM or Scratch. Furthermore, participants were requested not to play other video-games until the end of the study. To check training fidelity, parents of children that trained with SOM at home were asked to fill a form with the amount of daily exercise. In addition, the researcher monitored the training through phone/e-mail interview at the end of each week. After the training, each child received a small gift.

**Figure 59|** The flowchart of Study 2 - part B.



Despite the fact that participants were recruited in two different settings, statistical analyses did not show significant differences at baseline between the two groups in the demographic and neuropsychological variables (for details, see Table 21, 22, 23). All participants performed within normal limits (i.e., IQ above 85) in the WISC-IV scale (SOM group:  $M=99.23$  ( $SD = 9.24$ ), control group:  $M = 100.12$  ( $SD = 7.65$ );  $p=0.703$ ). Furthermore, the SOM and Control groups were matched in chronological age (SOM group:  $M = 10.10$  years, ( $SD=1.47$ ); control group:  $M=10.24$  ( $SD=1.39$ );  $p=0.721$ ), Socioeconomic status – SES

(SOM group: M =38.03 (SD=12.04); control group: M=37.40 (SD=12.67); p=0.851), and sex (SOM group: 10 Female; control group: 8 Female;  $\chi^2 = 0.921$ ).

Three children of the SOM group and two of the Control group dropped out of the study because they could not start (N=2) or they could not keep up with the schedule of the training (N=2). In conclusion, the final sample consisted of 55 subjects (N=30 for the SOM group; N=25 for the Control).

The descriptive statistics of the main variables assessed at the baseline (T1) are presented in Table 20 (Demographic and cognitive characteristics), Table 21 (Literacy skills performance), Table 22 (Executive functions performance).

**Table 20**| Demographic and cognitive characteristics (Means, SD) of SOM group and Control group prior to intervention. <sup>a</sup> $\chi^2$ -score, <sup>b</sup> F-score; \*sig.  $\alpha= 0.05$

	SOM		CONTROL		Test statistic	p		
	(n=30)		(n=25)					
	Mean	SD	Mean	SD				
Sex (f-m)	10-20		8-17		0.01 <sup>a</sup>	0.92		
Age (years)	10.10	1.47	10.24	1.39	0.13 <sup>b</sup>	0.72		
SES	38.03	12.04	37.40	12.67	0.04 <sup>b</sup>	0.85		
IQ (WISC-IV)	99.23	9.24	100.12	7.65	0.15 <sup>b</sup>	0.70		

**Table 21**| Means and standard deviations at baseline (T1) of literacy measures of participants of the two groups (SOM and Control). All the literacy variables are expressed in z-scores. <sup>a</sup> $\chi^2$ -score; <sup>b</sup> F-score; \*sig.  $\alpha= 0.05$

LITERACY skills	SOM		CONTROL		Test statistic	p		
	(n=79)		(n=72)					
	Mean	SD	Mean	SD				
Word Reading	Speed	-4.18	3.07	-4.24	2.88	0.01 <sup>b</sup>		
	Accuracy	-2.12	2.48	-2.20	2.15	0.14 <sup>b</sup>		
Non-word Reading	Speed	-2.36	2.41	-2.30	1.85	0.01 <sup>b</sup>		
	Accuracy	-1.14	1.74	-1.17	1.46	0.00 <sup>b</sup>		
Text Reading	Speed	-1.89	0.89	-1.92	1.09	0.02 <sup>b</sup>		
	Accuracy	-0.72	1.90	-0.94	1.85	0.18 <sup>b</sup>		
Writing	Word	-4.29	4.58	-4.20	4.41	0.01 <sup>b</sup>		
	Non-word	-0.50	0.90	-0.45	0.86	0.04 <sup>b</sup>		

Comprehension	-0.39	0.93	-0.31	0.85	0.10 <sup>b</sup>	0.76
Phonological Awareness	3.10	2.01	3.16	1.52	0.02 <sup>b</sup>	0.90

**Table 22**| Means and standard deviations at baseline (T1) of executive functions performance of participants of the two groups (SOM and Control). All the literacy variables are expressed in z-scores, <sup>a</sup> $\chi^2$ -score, <sup>b</sup> F-score; \*sig.  $\alpha=0.05$

		SOM (n=79)		CONTROL (n=72)		Test statistic	p
		Mean	SD	Mean	SD		
<b>EXECUTIVE FUNCTIONS</b>							
Digit span	Forward	-0.93	1.05	-0.98	1	0.03 <sup>b</sup>	0.86
	Backward	-0.33	1.16	-0.47	0.92	0.22 <sup>b</sup>	0.64
Visuo-spatial span	Forward	-0.51	1.22	-0.64	1.30	0.14 <sup>b</sup>	0.71
	Backward	-0.19	1.01	-0.16	0.90	0.01 <sup>b</sup>	0.93
Bells (barrage) task	Slow score	-0.52	1.14	-0.50	1.10	0.01 <sup>b</sup>	0.93
	Fast score	-0.36	1.29	-0.51	1.41	0.16 <sup>b</sup>	0.69
Phonological Fluency		-0.90	0.98	-0.76	1.13	0.23 <sup>b</sup>	0.64
Inhibition		-1.94	2.56	-1.95	1.82	0.00 <sup>b</sup>	0.99
TOL		0.21	0.98	0.16	0.87	0.04 <sup>b</sup>	0.84

### 3.2. Procedures

Literacy and executive functioning of all participants were tested at 2 to 7 days before the start of training and re-tested between 2 and 5 days after the end of training. Children who trained with SOM were evaluated in two testing sessions carried out on two different days at the ODFLab (Department of Psychology and Cognitive Science, University of Trento). Children of Control group were assessed in two testing sessions in the local school. If the general intelligence of a child was assessed more than one year before the start of training, an additional testing session was carried out, before the others, to have an update cognitive profile for each child. Testing was conducted by two different clinicians in a quiet room. It has to be noted that tasks were administered in a pseudorandom manner, however clinicians tried to alternate tests assessing literacy skills with EFs tasks. In the school, comprehension abilities were assessed in a collective session both at pre-test and post-test, whereas the other tests were administered individually. All measures, with the exception of general intelligence, were re-assessed at the second measurement point, within a week from the end of training.

### 3.3. Tools

The neuropsychological battery comprises the same tests used in Study 2 - part A, with the exception of the IQ that was measured with the WISC-IV scale (Wechsler, 2003; Italian version, 2012). Therefore, it involved word, non-word and text reading tasks (Sartori, Job & tressoldi, 2007), word and non-word writing (Sartori, Job & tressoldi, 2007), and text comprehension (Cornoldi, Colpo & Gruppo, 1998). As regards to EFs tasks, the neuropsychological battery comprised tests targeting visuo-spatial attention (Biancardi, Stoppa, 1997), auditory and visual working memory (Mammarella et al., 2008), and planning skills (Shallice, 1982; Italian version: Sannio, Fancello et al., 2006). At the end of training, both children and parents completed the feedback questionnaire aimed at investigate their appreciation of the video game and their self-assessment of training effects.

In addition, we evaluated phonemic awareness, phonological fluency and response inhibition with the following tasks:

*Phonemic Awareness.* The phonological processing subtest of the NEPSY-II, which stand for “Developmental Neuropsychological Assessment – second version” (Korkman, Kirk, & Kemp, 2007a, 2007b; Italian version: Urgesi, Campanella & Fabbro, 2011) was used to assess phonemic awareness. This subtest is composed of two phonological processing: Word Segment Recognition and Phonological Segmentation. In the first task (i.e. Word Segment Recognition), the child is required to repeat a word and then to create a new word by omitting a syllable or a phoneme. For example, the instructions for children were: “Your task is to repeat the word I have just said (“carte” /'karte/ [cards]) without the sound /k/”. The answer of the child should be “arte” (art). Therefore, this task requires the ability of correctly identify words from word segments (test of elision). In the second task, the examiner pronounced one word and the child is asked first to repeat the word and then to create a new word by substituting one phoneme (e.g., “Change /t/ in /s/. In order to change “morts” [dead] with “morso” [bite]). One point was assigned for each item correctly modified and the total raw score was converted into standard score on the basis of age norms.

*Phonological fluency* was estimated through a subtest of the “Neuropsychological Assessment Battery” -BVN (Bisiacchi et al., 2009). Children are asked to generate as many words as possible (within the time limit of 60 seconds) on the basis of a given phoneme (/c/, /s/, /p/) for each of the three trials. A z-score was calculated from the number of valid words generated (excluding errors and repetitions).

*Inhibition.* Response inhibition capacities were assessed via a subtest of the NEPSY-II

battery (Korkman, Kirk, & Kemp, 2007a, 2007b; Italian version: Urgesi, Campanella & Fabbro, 2011). The task consists of three subtasks: Naming, Inhibition and Switching. The child is asked to pay attention to a series of black and white shapes or arrows and to name them (Naming) or to name either the shape or direction (Inhibition) or an alternate response (Switching), depending on the colour of the shape or arrow. This timed subtest, thus, assesses not only the ability to inhibit automatic responses in favour of novel responses, but also the capacity of switching flexibly between response types.

### **3.4. Statistical analyses**

We first run the analyses corresponding to our main hypothesis, which regards the effects of the administered trainings on literacy skills and executive functions performance. Group differences were analysed by means of Analyses of Covariance (MANCOVA/ANCOVA).

To exclude the possible confounding effects of variables such as *chronological age*, *Intelligence (IQ)* measured through the WISC-IV scale), and *Socioeconomic status* of the family (SES) at the baseline, they were entered as covariates for all the MANCOVA/ANCOVA analyses conducted. Moreover, possible gender effects were taken into consideration inserting *Sex* as a between-subject factor. With the exception of the IQ scores and the Phonological Awareness scores, all participants' performance measures were transformed into z-scores (mean = 0; standard deviation = ± 1) on the basis of age norms. Both the IQ and the Phonological Awareness scores were standardized, but they differed in the scale used: the former has a mean of 100 and a standard deviation of ± 15, the latter has a mean of 10 and a standard deviation of ± 3.

Specifically, the series of repeated measured MANCOVA/ANCOVA, which aimed at evaluating the impact of training regimens from the baseline evaluation to the post-test, were run with Time (T1-T2) as a within-subject factor and Group (SOM - Control) as a between-subject factor. The main effects of Group or Time were reported only if significant. The Time X Group interaction was the primary effect of interest and in case of a significant result; further post-hoc analyses (paired-sample *t*-tests) were used to test for differences in performance of the two groups between the time points. No corrections for multiple testing were applied.

The effects of training in the two groups were analysed separately on literacy skills and executive functions. Within literacy, speed and accuracy of reading, writing, comprehension and phonological awareness measures were taken into consideration; whereas within EFs,

attention, short-term memory, verbal fluency (phonological fluency), inhibition and planning were considered separately.

Furthermore, we compared the improvements in fluency and accuracy in word, non-word and text reading for both treated groups with the developmental trend for reading. This with the aim of taking into accounts the efficacy of the two treatments (SOM versus Control) from a clinical point of view.

Moreover, three steps, fixed entry, multiple regression analyses were computed to explore the linear relationship between literacy improvements ( $\Delta$  T1-T2 reading speed, reading accuracy, phonological awareness: dependent variables) and EF improvements (block 3:  $\Delta$  T1-T2 EFs) taking into considerations covariates effects (block 1: age, IQ, sex) and the level of the literacy variables at pre-test (block 2).

Finally, descriptive statistics on the feedback questionnaire were conducted in order to assess children's appreciation of the training, potentials elements of criticalities and suggestions for improvements.

### **3.5. Results**

#### LITERACY SKILLS

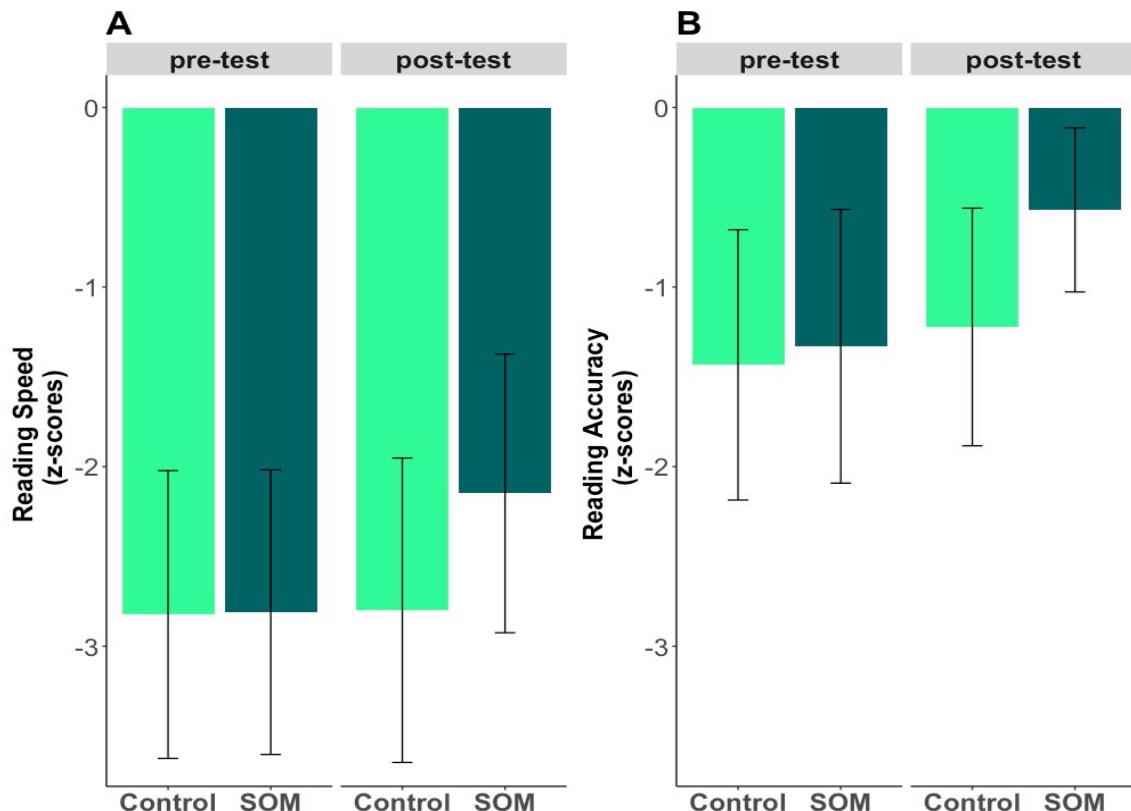
*A) Speed of Reading.* Improvements in reading speed after trainings were analysed by means of a 2 (Time: T1, T2) X 2 (Group: SOM, Control) X 3 (Measures: Word, Non-word and Text reading speed) MANCOVA. A significant Time effect was observed ( $F(1,48)= 4.770$ ,  $\eta^2_p = 0.090$ ,  $p = 0.034$ ), revealing improvements in the speed of reading across the two groups. More importantly a Time X Group interaction was significant [ $F(1, 48) = 17.906$ ,  $\eta^2_p = 0.272$ ,  $p < 0.001$ ]. Post-hoc analyses confirmed that children in the SOM group significantly improved their reading time ( $p < 0.001$ ), whereas the Control group did not ( $p=0.759$ ). No significant triple interaction [ $F(2, 47) = 5.352$ ,  $\eta^2_p = 0.069$ ,  $p =0.006$ ] were found between the improvements in the two groups in the different reading measures.

Taking into consideration the effects of the covariates, although exploratory, significant interactions between Time and IQ [ $F(1, 48) = 11.134$ ,  $\eta^2_p = 0.188$ ,  $p = 0.002$ ] and Time and Sex [ $F(1, 48) = 4.377$ ,  $\eta^2_p = 0.084$ ,  $p = 0.042$ ] reflected that, regardless the group, male children with lower IQ (measured at T1) showed higher results at the post-test evaluation.

*B) Accuracy of Reading.* The same MANCOVA model was run considering reading accuracy as dependent variable. Despite the fact that results failed to show any significant main effects or interaction, it has to be noted the Time X Group interaction [ $F(1, 48) = 3.928$ ,  $\eta^2_p = 0.076$ ,  $p = 0.053$ ], revealing higher improvements – yet not statistically significant - in SOM group.

Post-hoc analyses highlighted a significant difference between T1 and T2 measures in SOM  $p = 0.003$ , but not in the control group  $p=0.098$ . No group differences between the three reading tasks at T2 were observed [Time X Group X Measures:  $F(2, 47) = 0.701$ ,  $\eta^2_p = 0.501$ ,  $p =0.029$ ]. Results of reading speed and accuracy are presented in Figure 60.

**Figure 60|** Performance in reading speed (A) and accuracy (B), expressed in z-scores, were measured before (T1) and after (T2) SOM and Control trainings. Error bars represent confidence intervals (CI).

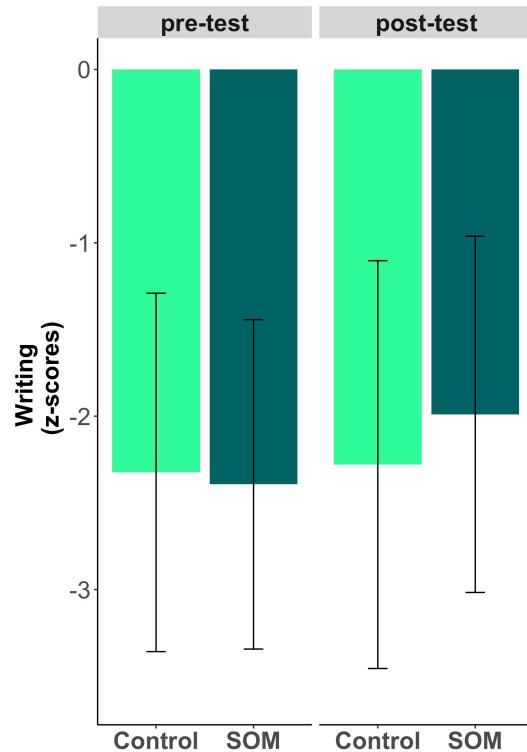


### Writing.

A 2x2x2 MANCOVA with Time (T1, T2) and Measures (Word Writing, Non-word Writing) as within-subject factor, and Group as between-subject factor was carried out. No main effect of Time and Time x Group interaction [ $F(1, 48) = 0.583$ ,  $\eta^2_p = 0.012$ ,  $p = 0.449$ ] were found significant. Results of writing are presented in Figure 61.

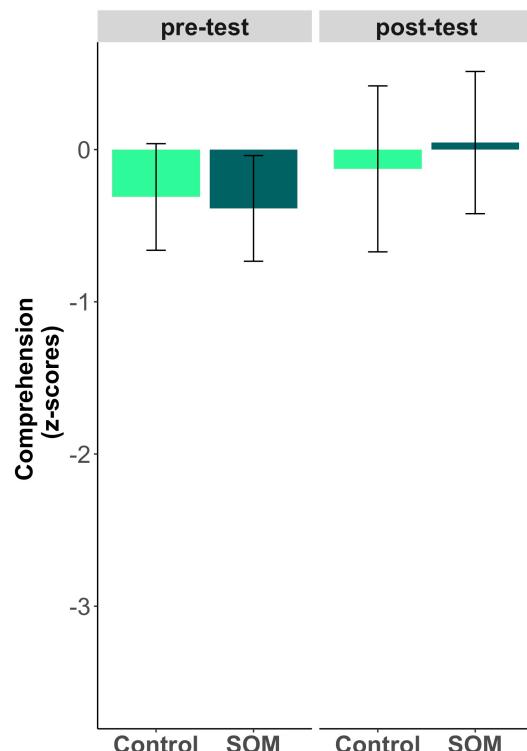
Moreover, as regards to covariates effect, a significant interaction between Measure and Sex was observed [ $F(1,48)= 14.535$ ,  $p < 0.001$ ,  $\eta^2_p = 0.232$ ] revealing that males, regardless the time point considered, obtained worst performance in word writing.

**Figure 61|** Writing performance, expressed in z-scores, was measured before (T1) and after (T2) SOM and Control trainings. Only SOM group statistically improved after training. Error bars represent confidence intervals (CI).



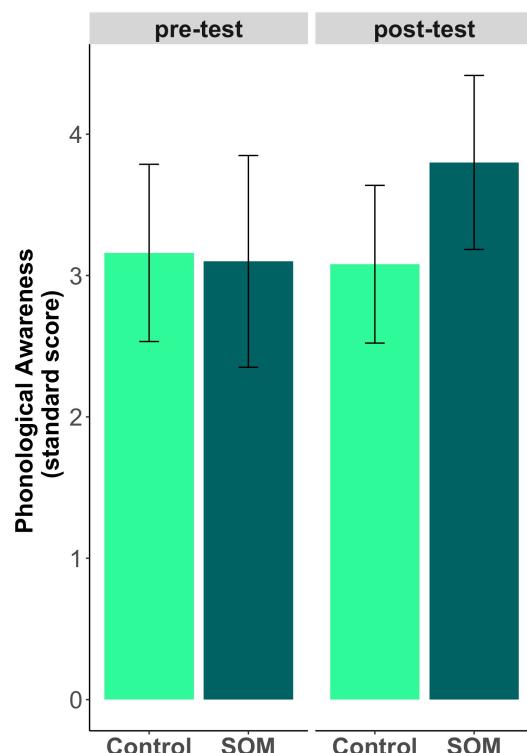
*Comprehension.* Text comprehension improvements were analysed using a 2 (Time: T1, T2) X 2 (Group: SOM, Control) analysis of Covariance (ANCOVA). The results revealed no differences in the effects of the training with regard to Text comprehension [Time X Group  $F(1, 48) = 0.733, \eta^2_p = 0.015, p = 0.396$ ]. Results are presented in Figure 62.

**Figure 62|** Comprehension performance, expressed in z-scores, was measured before (T1) and after (T2) SOM and Control trainings. Error bars represent confidence intervals (CI).



*Phonological Awareness.* Finally, an ANCOVA with Time as within-subject factor and Group as between-subject factor was carried out on the performance in the phonological awareness task. Significant differences between training conditions were observed [Time X Group interaction,  $F(1, 48) = 7.017$ ,  $\eta^2_p = 0.128$ ,  $p = 0.011$ ]. Post-hoc comparisons highlighted an enhancement in phonological awareness skills of children trained with SOM ( $p = 0.003$ ), whereas the control group did not show any significant improvement ( $p = 0.538$ ). Results in the phonological awareness task are presented in Figure 63.

**Figure 63|** Phonological awareness skills, expressed standardized score (mean= 10; SD= 3), were measured before (T1) and after (T2) SOM and Control trainings. Error bars represent confidence intervals (CI).



**Table 23|** Training effects of SOM group ( $n = 30$ ) and control group ( $n = 25$ ) on literacy skills performance [Means ( $SD$ )]. In addition, the delta scores (T2-T1) are reported for both groups.

<sup>a</sup> Time (T1-T2) X Group interaction.

<sup>c</sup> Test significance. \* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$ .

<sup>d</sup>Effect sizes of interaction Time : Group expressed as partial eta squared.

	SOM			CONTROL			$F$ -score <sup>a</sup>	$p$ <sup>c</sup>	ES <sup>d</sup>
	T1	T2	$\Delta$	T1	T2	$\Delta$			
Reading Speed	-2.81 (1.79)	-2.15 (1.77)	0.66 (0.62)	-2.82 (1.66)	-2.80 (1.75)	0.02 (0.38)	17.91	<0.001	0.27
Reading	-1.33	-0.57	0.76	-1.44	-1.22	0.21	3.93	0.053	0.08

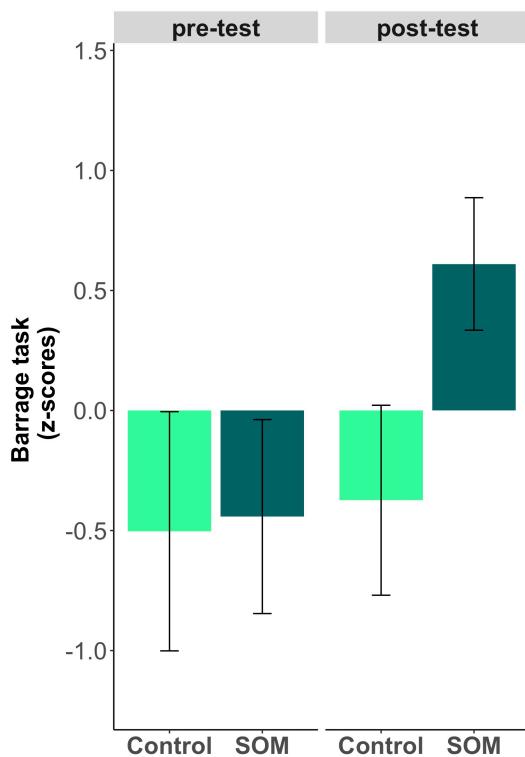
	(1.70)	(1.01)	(1.26)	(1.35)	(1.05)	(0.61)			
Writing	-2.39 (2.54)	-1.99 (2.75)	0.40 (1.30)	-2.32 (2.50)	-2.28 (2.85)	0.05 (1.20)			
							0.58	0.449	0.01
Comprehension	-0.39 (0.93)	0.05 (1.25)	0.43 (1.26)	-0.31 (0.85)	-0.13 (1.32)	0.18 (1.24)	0.73	0.396	0.02
Ph. Awareness	3.10 (2.01)	3.80 (1.65)	0.70 (1.18)	3.16 (1.52)	3.08 (1.35)	-0.08 (0.64)	7.02	0.011	0.13

## EXECUTIVE FUNCTIONS

*Visuo-spatial Attention.* A 2x2x2 MANCOVA with Time (T1, T2) and Outcomes (fast and slow score) as within-subject factors, and Group as between-subject factor was carried out. A significant Time X Group interaction [ $F(1, 48) = 25.893$ ,  $\eta^2_p = 0.350$ ,  $p < 0.001$ ] was observed revealing larger improvements in SOM group. Post-hoc analyses confirmed a significant difference between T1 and T2 measures in SOM ( $p < 0.001$ ), but not in the control group ( $p=0.172$ ). No Time X Group X Outcomes interaction was found significant [ $F(1, 48) = 0.137$ ,  $\eta^2_p = 0.000$ ,  $p = 0.966$ ] showing, thus, no differences between the two scores (fast and slow score).

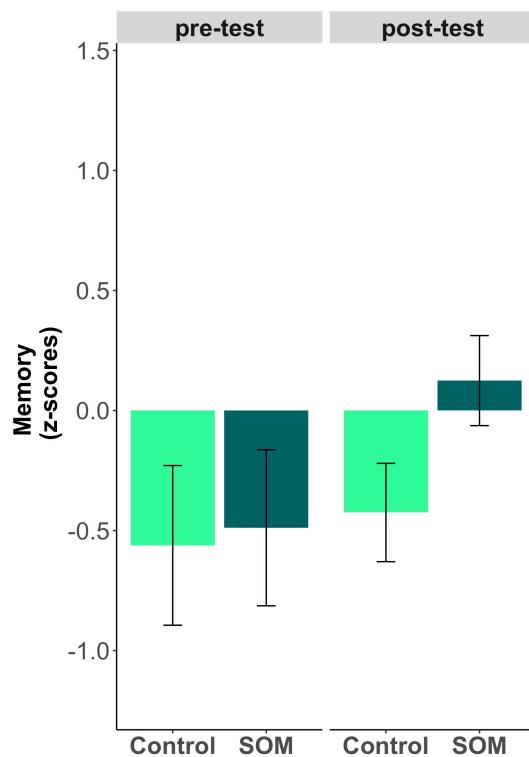
Results of visuo-spatial attention are presented in Figure 64.

**Figure 64|** Visuo-spatial performance, expressed in z-scores, was measured before (T1) and after (T2) SOM and Control trainings. Only SOM group statistically improved after training. Error bars represent confidence intervals (CI).



*Verbal and visuo-spatial memory.* Improvements in short-term memory were analyzed by means of a 2 (Time: T1, T2) X 2 (Group: SOM, Control) X 4 (Measure: Digit span Forward, Digit span Backward, Visuo-spatial span Forward, Visuo-spatial span Backward) analysis of Covariance (MANCOVA). The results revealed larger gains in memory performance in the SOM group compared to the control group [Time X Group  $F(1, 48) = 9.745$ ,  $\eta^2_p = 0.169$ ,  $p = 0.108$ ]. Post-hoc comparisons confirmed improvements children trained with SOM only ( $p < 0.001$ , control group  $p = 0.173$ ). No significant interaction between Time, Group and Memory Measure was found significant [ $F(3, 46) = 0.789$ ,  $\eta^2_p = 0.046$ ,  $p = 0.506$ ]. Results of verbal and visuo-spatial short-term and working memory are presented in Figure 65.

**Figure 65|** Performance in verbal and visuo-spatial working memory, expressed in z-scores, were measured before (T1) and after (T2) SOM and Control trainings. Error bars represent confidence intervals (CI).

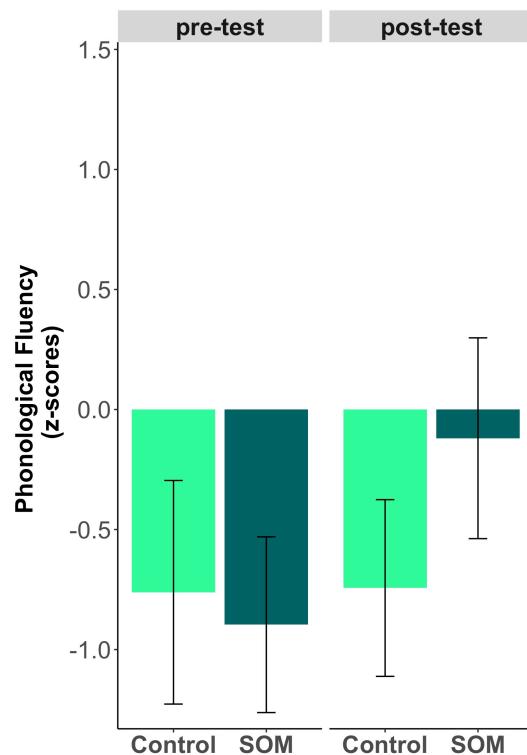


Taking into consideration the effects of the covariates, although exploratory, significant interactions between Measure and Age [ $F(3, 46) = 3.128$ ,  $\eta^2_p = 0.169$ ,  $p = 0.035$ ], Measure and IQ [ $F(3, 46) = 4.264$ ,  $\eta^2_p = 0.218$ ,  $p = 0.010$ ], and Measure and SES [ $F(3, 46) = 8.619$ ,  $\eta^2_p = 0.360$ ,  $p < 0.001$ ] were found reflected that, regardless time there is no differences between the memory measures for children of grade 3, while for older children digit span forward is significantly lower than the other measures (mean difference between digit span

backward: -0.51,  $p = 0.018$ ; spatial span forward:-0.55,  $p = 0.007$ ; spatial span backward: -0.93,  $p < 0.001$ ). In addition, in children with lower IQ (below or equal to 100) and in children with lower socioeconomic status (below or equal to 37.5), digit span forward was significantly below digit span backward (IQ: -0.59,  $p = 0.046$ ; SES: -0.85,  $p < 0.001$ ) and spatial span backward (IQ: -0.78,  $p = 0.004$ ; SES: -1.13,  $p < 0.001$ ). Moreover, a significant Time X Age [ $F(3, 46) = 7.583$ ,  $\eta^2_p = 0.136$ ,  $p = 0.008$ ] indicating that younger children (grade 3 and 4) improved more.

*Phonological Fluency.* An ANCOVA with Time as within-subject factor and Group as between-subject factor was carried out on the phonological fluency task. Significant differences between training conditions were again observed [Time X Group interaction,  $F(1, 48) = 8.940$ ,  $\eta^2_p = 0.157$ ,  $p = 0.004$ ]. Post-hoc comparisons highlighted an enhancement in the phonological fluency task for children trained with SOM only ( $p < 0.001$ , control group  $p = 0.896$ ). Results in phonological awareness are presented in Figure 66.

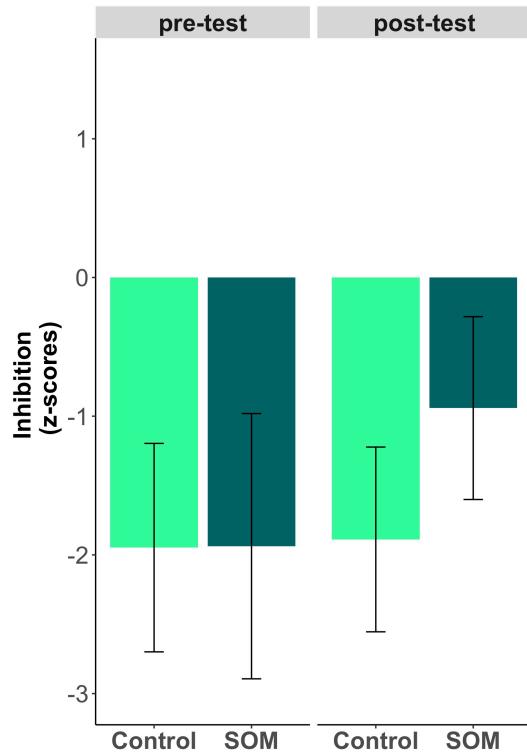
**Figure 66|** Performance in phonological awareness, expressed in z-scores, were measured before (T1) and after (T2) SOM and Control trainings. Error bars represent confidence intervals (CI).



*Inhibition.* The same ANCOVA model was run considering inhibition skills as dependent variable. Time X Group interaction was significant [ $F(1, 48) = 8.413$ ,  $\eta^2_p = 0.149$ ,  $p = 0.006$ ],

with improvements observed in SOM group only. In fact, post-hoc analyses revealed that only dyslexic children trained with SOM ( $p = 0.002$ ) significantly improved their response inhibition capacities (control group,  $p = 0.721$ ) Results are presented in Figure 67.

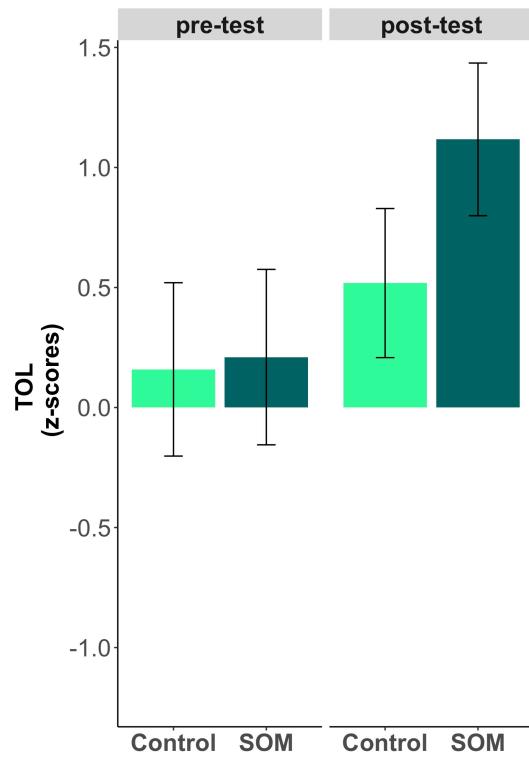
**Figure 67|** Performance in inhibition, expressed in z-scores, were measured before (T1) and after (T2) SOM and Control trainings. Error bars represent confidence intervals (CI).



*Planning.* Improvements in planning abilities (measured via the Tower of London task) were analysed using a 2 (Time: T1, T2) X 2 (Group: SOM, Control) analysis of Covariance (ANCOVA). A significant Time X Group interaction [ $F(1, 48) = 4.750$ ,  $\eta^2_p = 0.034$ ,  $p = 0.090$ ] was found revealing higher improvements in SOM group. Post-hoc analyses found significant difference between T1 and T2 planning measure in both SOM ( $p < 0.001$ ) and control group ( $p= 0.025$ ).

Results of planning are presented in Figure 68.

**Figure 68** Results in planning, expressed in z-scores, was measured before (T1) and after (T2) SOM and Control trainings. Only SOM group statistically improved after training. Error bars represent confidence intervals (CI).



**Table 24** Training effects of SOM group ( $n = 30$ ) and control group ( $n = 25$ ) on executive functions performance [Means ( $SD$ )]. In addition, the delta scores (T2-T1) are reported for both groups.

<sup>a</sup> Time (T1–T2) X Group interaction.

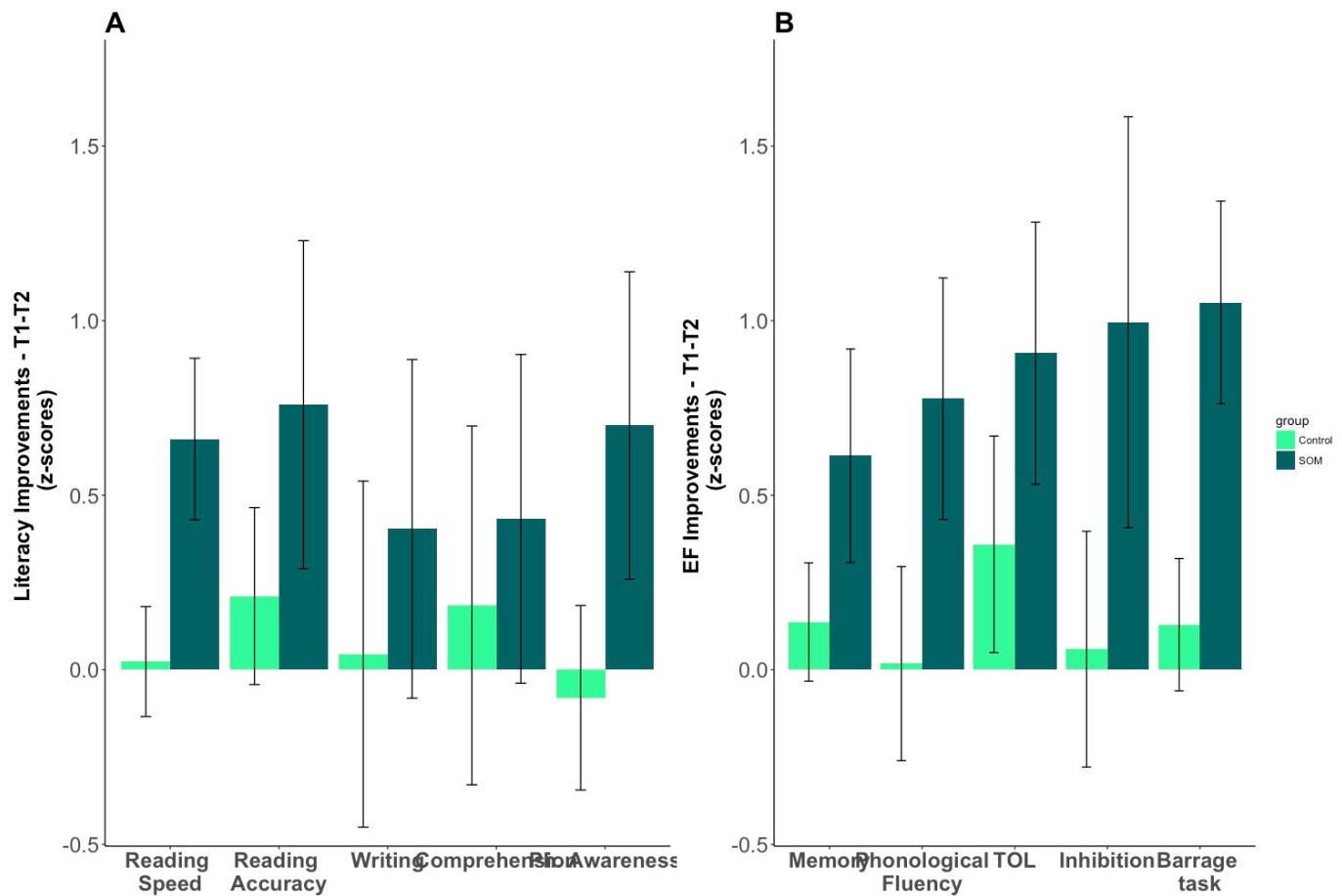
<sup>c</sup> Test significance. \* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$ .

<sup>d</sup> Effect sizes of interaction Time : Group expressed as partial eta squared.

		SOM			CONTROL			<i>F</i> -score <sup>a</sup>	<i>p</i> <sup>c</sup>	ES <sup>d</sup>
		T1	T2	$\Delta$	T1	T2	$\Delta$			
Bells task	(barrage)	-0.44 (1.08)	0.61 (0.74)	1.05 (0.78)	-0.50 (1.21)	-0.37 (0.96)	0.13 (0.46)	25.89	<0.001	0.35
Digit+spatial span		-0.49 (0.87)	0.12 (0.50)	0.61 (0.82)	-0.56 (0.81)	-0.42 (0.50)	0.14 (0.41)	9.75	0.003	0.17
Ph. Fluency		-0.90 (0.98)	-0.12 (1.12)	0.78 (0.93)	-0.76 (1.13)	-0.74 (0.89)	0.02 (0.67)	8.94	0.004	0.16
Inhibition		-1.94 (2.56)	-0.94 (1.77)	0.96 (1.58)	-1.95 (1.82)	-1.89 (1.61)	0.06 (0.82)	8.41	0.006	0.15
TOL		0.21 (0.98)	1.12 (0.85)	0.91 (1.01)	0.16 (0.87)	0.52 (0.75)	0.36 (0.75)	4.75	0.034	0.09

In addition, the means of the delta score (difference in the performance between the post-training and the baseline) in literacy skills and executive functions, obtained by the two groups, can be found in Figure 69.

**Figure 69|** Means of  $\Delta$  scores (T1-T3) in literacy skills and executive functions, obtained by the two groups. Error bars represent CI. It has to be noted that all the scores are expressed in z-scores with the exception of the Phonological Awareness task (standard score with mean=10;  $ds = \pm 3$ ).



### Clinical efficacy comparison

This study aimed at estimating the enhancements in the reading abilities from a clinical point of view. In this regard, it is possible to conclude that an intervention is clinically effective only if it was able to reduce the specific deficits (e.g., the lack of reading fluency) more than what is expected in one year of regular education and reading experience, without specialist treatments (RC-DSA-2011). For Accuracy, we took into consideration the improvements between the baseline and the final evaluation (time points T1-T2) in terms of reductions of the errors made by the children while reading. For Fluency, we compared the treatment

outcomes in terms of syllables per seconds.

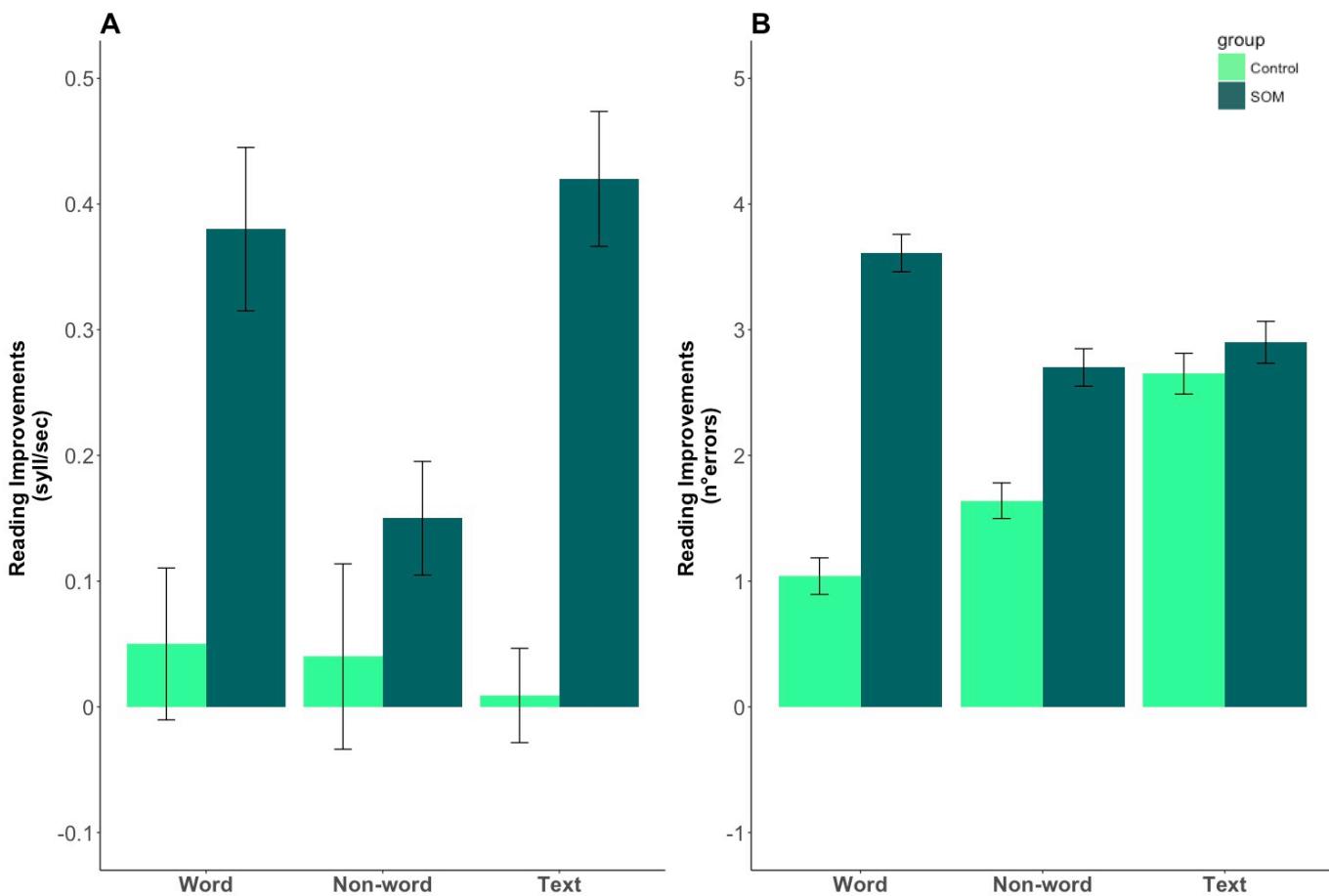
We, therefore, compared the reading improvement of the two training programs with the developmental trend in reading skills for children with dyslexia. According to the studies of Tressoldi, Stella, & Faggella (2001) and Tressoldi & Vio (2008), Italian children with dyslexia, without specialist treatments, increased by around 0.3 syllables per second their single word and text reading fluency, whereas their non-word reading fluency is increased by approximately 0.15 syllables per second for each grade. In addition, taking into account the clinical significance for accuracy in reading, an arbitrary criterion has been used according to the RC-DSA-2011: a reduction of 50% of the errors with respect to the first evaluation.

Reading improvements (T1-T2) both for Fluency (Syllables per Second) and Accuracy (number of errors) of the two trainings are presented in Figure 70.

Neither groups had a reduction of the number of errors higher than 50%. However, this clinical criterion is usually meant to apply to treatments that lasted more (approximately, from 6 months to one year). Nevertheless, 18 children of SOM group improved in reading accuracy and, of those 50% (9 children) manage to decrease their number of errors the point of reaching this clinical criterion, whereas only one child of the Control group (4%) reached it.

On the other hands, only children of SOM group (mean 0.38 syll/sec) did improve their word reading speed more than the mean improvements expected in a dyslexic child (0.30 syll/sec) after 1 year of spontaneous reading development (Control group: 0.05 syll/sec). Analogously, the SOM group showed larger improvements than the control group in the reading speed of non-word (SOM: 0.15 syll/sec; Control: 0.04 syll/sec). The improvement in non-word decoding speed of SOM group was the same of what expected in one year without specialist treatment. Finally, improvements in the speed of text reading for SOM training were 0.42 syllables per second, resulting bigger than the developmental trend (0.30 syll/sec). Specifically, 80% of children in the SOM group improved in reading speed and, of those, 16 children had an improvement higher than the clinical efficacy criterion. Vice versa, only 32% of the children that trained with Scratch (Control activity) improved in reading speed and none of them could be considered clinically improved.

**Figure 70|** Improvements in Fluency (A), expressed in syll/sec, and in Accuracy (B), expressed in the number of errors, reached by the two treatments in word, non-word and text reading. Error bars represent the SE.



### ***Link between Literacy and Executive functions improvements***

Three-step fixed entry, multiple regressions were performed on the whole sample ( $n = 55$ ) to predict the change in reading skills between T1 and T2. Specifically, in the first model, the dependent variable was the change in reading speed (i.e., the delta of the mean between the speed score in word, non-word and text reading task). Similarly, the dependent variables of the second and the third model were the change in reading accuracy and phonological awareness respectively. The predictors were: age, IQ, sex (block 1); the reading variable at T1 (block 2); attention and EFs changes (block 3). We took into consideration the following EFs variables: visuo-spatial attention, short-term memory, inhibition, phonological fluency and planning. The visuo-spatial attention changes were indexed by calculating the mean between the delta scores ( $T2 - T1$ ) in the fast and slow score of the barrage task, while the short term memory changes using the mean of the delta scores in the four measures (digit span forward and backward, visuo-spatial span forward and backward).

**Reading speed.** The dependent variable was the reading speed changes between T1 and T2 ( $\Delta T1-T2$  reading speed), and the predictors were: age, IQ, sex (block1); reading speed at T1

(block 2), EFs changes between T1 and T2 (block 3). These variables significantly predicted reading speed changes,  $F(9, 45) = 2.865, p = 0.009, r^2 = 0.363$ , and only phonological fluency changes added statistically significance to the prediction,  $p = 0.048$ . However, we replicated this analysis on the two groups separately and results evidenced that link between reading speed improvements and EFs improvements were due to the fact that the groups have different means at T2. Considering the SOM group only, in fact, results were not significant [ $F(9, 20) = 1.478, p = 0.223, r^2 = 0.40$ ].

*Reading accuracy.* Vice versa, results of the regression were significant when we considered the entire sample ( $n=55$ ) [ $F(9, 45) = 11.418, p < .0001, r^2 = 0.695$ ] and when we specifically considered the SOM group ( $n=30$ ) [ $F(9, 20) = 6.050, p < .0001, r^2 = 0.731$ ] highlighting a predictive role of the EFs changes. Specifically, only the coefficient related with inhibition changes was close to reach significance ( $p = 0.06$ ).

*Phonological Awareness.* Consistently, statistically significantly predicted reading speed changes,  $F(7, 143) = 18.282, p < .001, r^2 = 0.472$ , only when the entire sample was took into consideration. Considering the SOM group only, only reading speed at T1 significantly predict reading speed changes [ $F(4, 74) = 21.751, p < 0.001, r^2 = 0.540$ ], while the EFs variable did not [r-squared change: 0.025; ( $F(3, 71) = 1.347, p = 0.266$ )].

Consistently, these variables significantly predicted phonological awareness changes,  $F(9, 45) = 4.825, p < 0.001, r^2 = 0.491$ , and only visuo-spatial attention changes added statistically significance to the prediction,  $p = 0.002$ . These results were confirmed by the analysis on the groups separated [SOM:  $F(9, 20) = 4.428, p = 0.001, r^2 = 0.666$ ; barrage task:  $p = 0.001$ ].

Therefore, regression analyses highlighted the role played by specific EFs (i.e. visuo-spatial attention and inhibition) on the improvements in performance in reading accuracy and phonological awareness task after training.

### ***Feedback questionnaire***

A feedback questionnaire was administered following the end of training in order to investigate the general appreciation of the game of players and their parents. Therefore, two different versions - one for children and one for parents - of the questionnaire used in Study 2 - part A was created via Google Forms.

### ***Players***

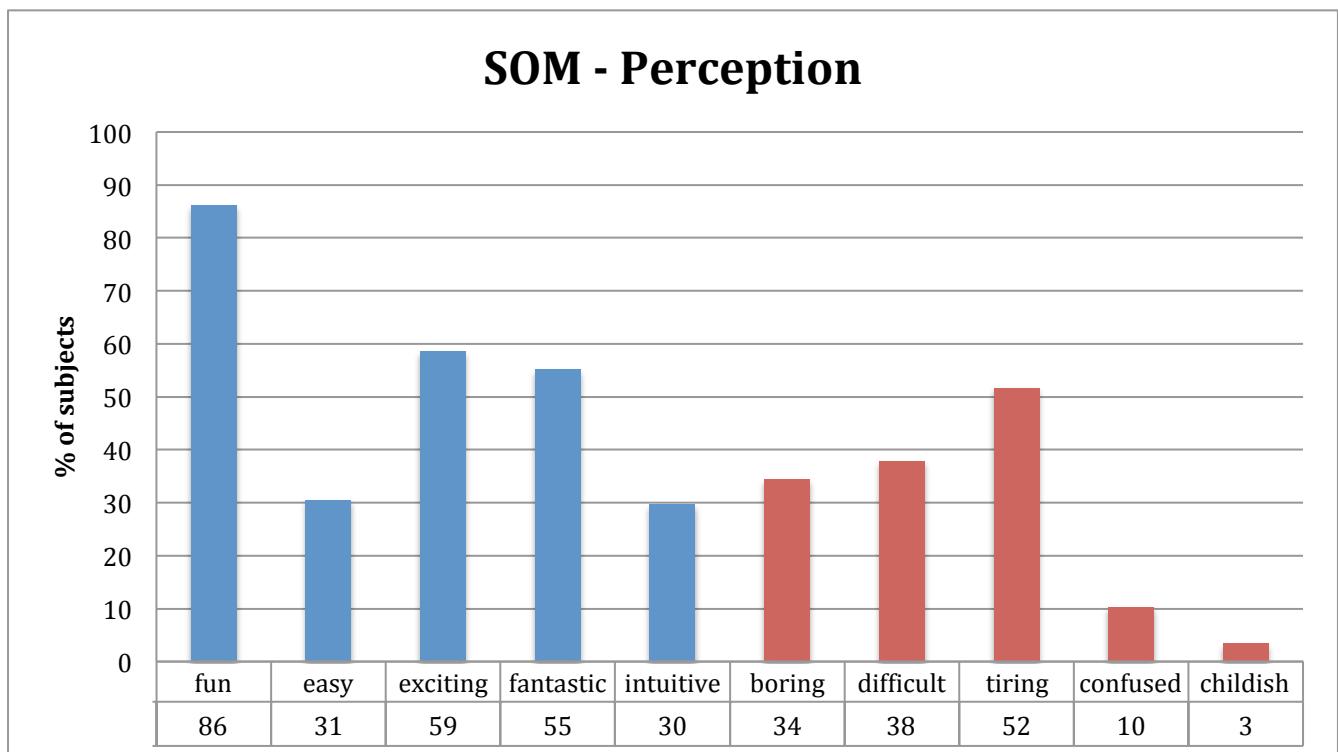
The questionnaire for players could be divided into two parts: the first aimed at evaluate the level of engagement during the training, while the second focused more on the self-

assessment of children's capacities after the training. In the last part, we created an open question for general comments or suggestions. Children's comments were transcribed, grouped by common themes (e.g., frustration for the frequent repetition of some mini-games, particularly the fishing mini-game) and discussed among the members of the design group. Questionnaire replies were received from 29 participants of SOM group and 23 of the Control group.

Crucially, no statistically significant differences were found in the level of appreciation of the game, as measured with the "funometer", between children of the two groups ( $p$ -value = 0.137). The "Funometer" recorded a score of 4.36 out of 5 for SOM and a 4 for Scratch. Analogously, an unpaired t-test revealed no statistically significant differences between the questionnaires of males and females in both groups ( $p$ -value = 0.342).

Children of both groups appreciated the training activities; in Figure 71 are specifically presented the answers of SOM players to the question "How would you define this game?". The most chosen items among the positive ones were "Fun"(86% of the players) and "Exciting" (59%) were the most chosen positive items; on the other hand tiring (52%) and difficult (38%)were the most chosen negative ones.

**Figure 71|** Overall positive and negative items of the perception question on Skies of Manawak.



Furthermore, the results from the questions on engagement confirmed an overall appreciation

of the game. The majority of the players liked the graphics (82.7%) and they would have suggested the game to a friend (65.5%). Many players expressed a general interest with respect to the advancement of the adventure (75.9%) and to a second episode (86.6%). As regards the self-assessment of the gaming skills, most of the children declared that, despite some difficulties at the beginning, they were able to improved their performance in the after 12 hours of training (89.6 %). Table 25 shows the questions and the average response of SOM group.

**Table 25|** Questions and related means of answers on the appreciation of Skies of Manawak (N=29).

Question	Mean of participants answers (0 = no/never, 2 = yes/often)
Would you suggest it to a friend?	1.71 (0.56)
Would you like to play to a second episode?	1.93 (0.39)
Was it difficult?	1.78 (0.98)
Did you like the graphics?	1.85 (0.27)
While you were playing, were you curious about what could happen next?	1.59 (0.45)
Was it challenging?	1.70 (0.28)
Did you feel bored?	0.75 (0.77)
Compared to the beginning, do you feel to be more competent in playing this game?	1.94 (0.64)

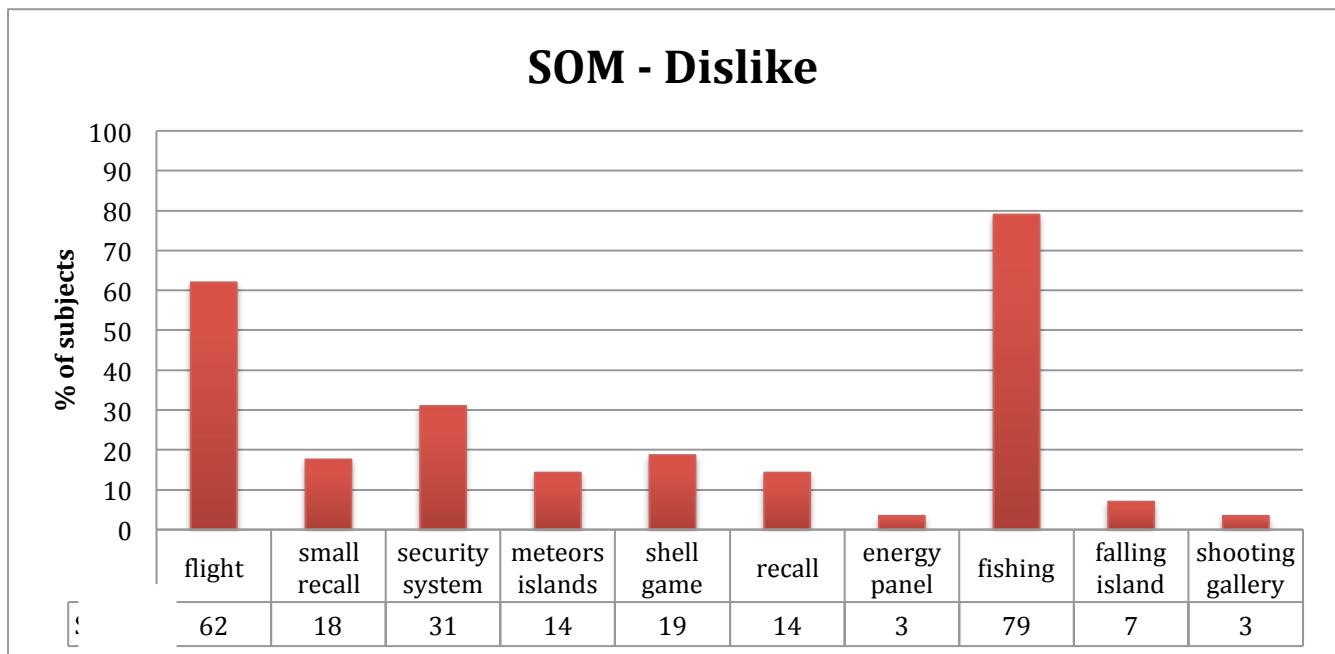
In regard to the self-assessment of their cognitive abilities:

- 72.4% of children reported an improvement in attentional skills.
- 51.7% thought to be able to keep in mind instructions more efficiently.
- 68.9% believed to be less susceptible to distractions and to be able to understand task instructions better.

Taking into consideration the single mini-games, the fishing was again particularly disliked (79%), followed by the flight (62%, see Figure 72 for the percentages of the other mini-games). Many of our participants reported difficulties in paying attention to stimuli presented in the periphery of the visual field and in processing stimuli that were difficult to anticipate both from a temporal and a spatial point of view (72% of players perceive the fight as difficult, see Figure 74). A correlation between the players disliking the Flight mini-game and

identifying it as one of the most difficult was again found ( $p$ -value < 0.014, cor = 0.43). For these reasons, at least at the beginning of the training, the less skilled players were not able to obtain an adequate performance in the flight (“action part”) and, thus, they ended up playing the fishing mini-game (multiple object tracking task) more often. This mini-game was not perceived as difficult in itself (only 10% of the players), but since it was activated every time the player loses all the health points during the flight, it may have caused a disruption to the flow of the game session. This hypothesis is supported by the fact that the shell game, which shared the same mechanisms of the fishing, was perceived negatively only by 6 players. Most of the children appreciated this last mini-game due to the fact that it was set on the secret island, surrounded by mystique.

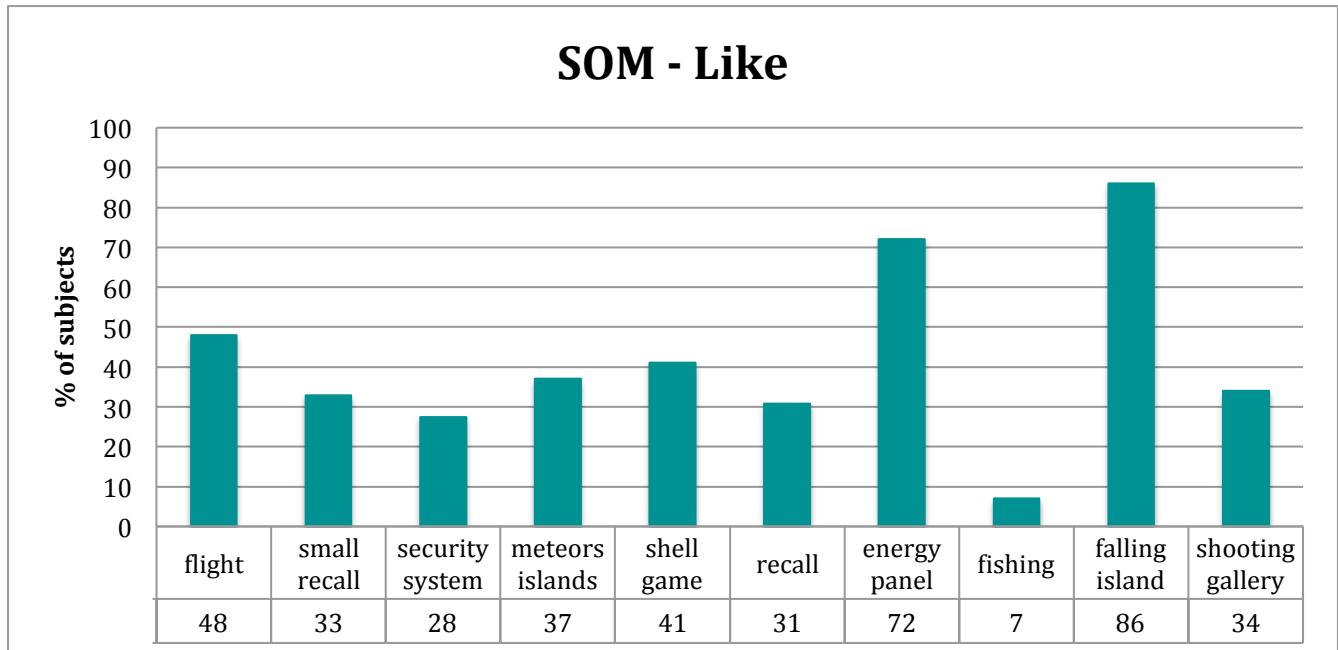
**Figure 72** Overall answers to the most disliked mini-games.



The mini-games that the players liked the most, as shown in Figure 73, were the falling island (86%) and the energy panel (72%). While children liked the falling island due to its simple mechanic (i.e. inhibit the predominant response of shooting at the sentinels), the energy panel was appreciated mainly because of the ability to freely experiment with various solutions and observe the chain of reaction.

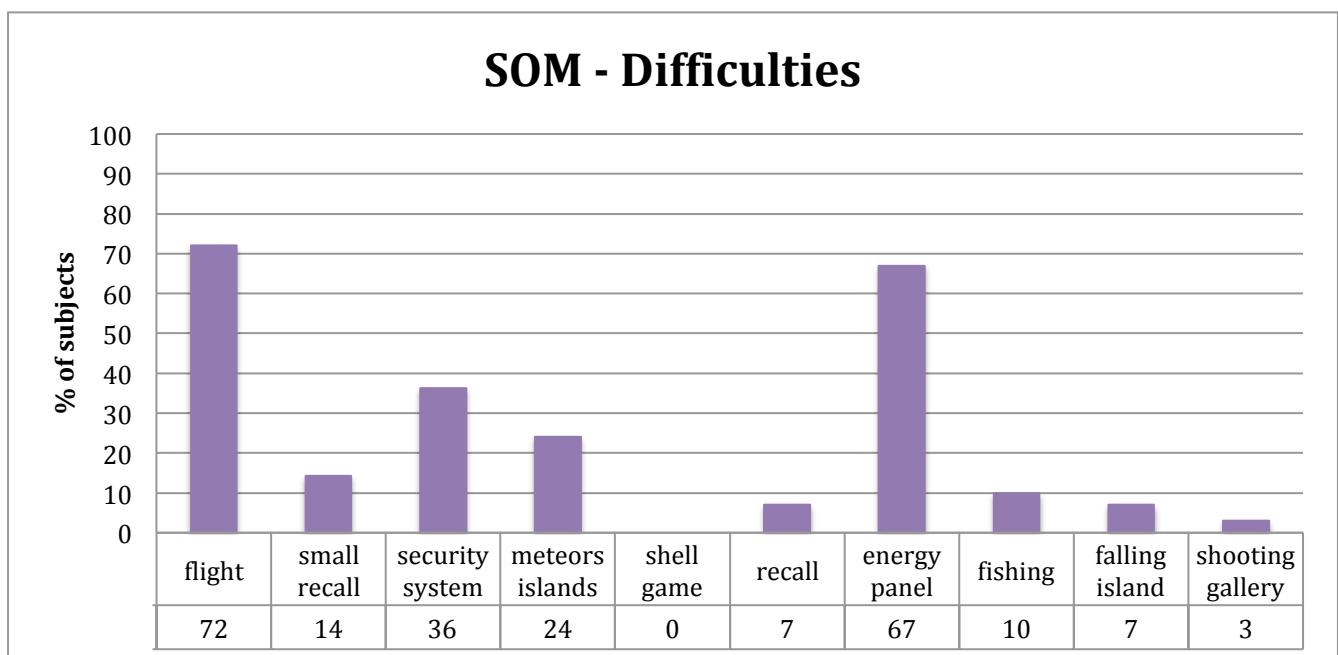
The flight was appreciated by 48 % of the players because it was perceived as challenging (due to its variety and the type of challenge), less tied to cognitive exercises and more similar to commercial action video games.

**Figure 73**| Overall answers to the most disliked mini-games.



Again a, mild, yet significant correlation between the players liking the energy panel mini-game and identifying it as one of the most difficult was found ( $p\text{-value} < 0.021$ ,  $\text{cor} = 0.27$ ). Any other correlation on liked, disliked and difficult mini-games had no significant result.

**Figure 74**| Overall answers to the most disliked mini-games.



The last question of the questionnaire (“Would you have any suggestion for the second episode of Skies of Manawak?”) stimulated the players to propose various ideas. Without

going into details, the players' proposals focused on the reward and the customization features. The children expressed a strong desire to receive a greater number of rewards; these rewards should not only be used to boost the characters, but also their appearance. Beyond that, they suggested additional content such as new characters, vehicles, enemies and environments. Particularly, some of them complained that the characters were not enough sexually characterized. As highlighted in the previous chapter, this last objection is against our design principles and, thus, will not be taken into account in a new re-design of the video game.

Overall, Skies of Manawak was appreciated by the players, but many of them did not fully understand the story. Therefore, for some of them the structure of the game was perceived as a mere continuous repetition of the mini-games.

### *Parents*

A questionnaire was administered at the end of the study in order to have feedback on their perception of the training experience. As for the players' version, we received replies from 29 participants of SOM group (24 mothers, 5 fathers) and 23 of the Control group (all mothers).

In regard to their evaluation of the gaming experience:

- 72.4% of parents claimed that their children were motivated to trained with SOM and that they enjoyed the activity.
- 58.6% considered the training easy.
- 65.5 % considered the training not excessively burdensome or boring.
- 86.2% thought that the child was able to train alone without supervision, while only 44.3% of the parents let their children trained autonomously.

Finally, the training was seen as a useful rehabilitation activity by 93,1% of the parents.

In addition, positive was the feedback regarding the question "Would combine this type of training with traditional rehabilitation activities?" with 26 affirmative answers.

Lastly, it emerged that:

- 82.8% of the parents saw an improvement in their children's reading speed, while 86.2% in reading accuracy.
- Additionally, for 79,3% of the parents their son/daughter seemed to have a more positive attitude towards the reading task.
- 82,7% considered their children improved in attention, whereas 72,4% noticed improvements in memory capacities as well, keeping in mind instructions more efficiently (51,7%).

- Finally, 79,3% believed their children to be less susceptible to distractions and be able to understand task instructions better.

In conclusion, the overall appreciation for the training activity was high: they complimented on this initiative both for the innovative idea of involving a real video game that could be played at home, and for its utility since it was able to improve usually underdeveloped skills in their children. Specifically, they reported improvements in children's focus capacities and in resistance to frustration. Some parents reported initial difficulties of their children in understanding how to overcome to obstacles of some levels but, after they made it through, many of the children were happy and satisfied. Overall, parents rated the program as having a pretty high acceptability and satisfaction.

Furthermore, the majority of parents (75.8%) asked for a second chapter of the adventure.

### **3. Final Remarks**

The results (before vs. after EFs training) showed that after intervention, the dyslexic children who trained with SOM presented better performances compared to the control group both in EFs measures (i.e., visuo-spatial attention, short-term memory, phonological fluency, inhibition, planning) and in reading efficiency (specifically, the speed of reading).

### **4. Discussion**

#### ***4.1. Efficacy***

The main aim of these studies was to investigate the efficacy of a video game specifically designed to train several executive functions, starting from growing evidence of a strong association between impairments in the executive domain and educational and social difficulties (Diamond et al., 2013).

On account of this fact, we evaluated the effects of the training regimens (SOM versus the control activity Scratch) through standardized paper and pencil tasks in a school-based study with typically developed children (Study 2 - part A). In a second study we involved children with dyslexia to investigate whether SOM may enhance EFs following intervention, and whether these improvements transfer to reading skills in children with dyslexia (Study 2 - part B).

Overall, results of Study 2 - part A demonstrated the benefits of a specific video game intervention on executive functioning in typically developing children. Skies of Manawak was indeed able to boost visuo-spatial attention and visuo-spatial working memory. The

reported benefits, which followed 12 hours of training distributed over several weeks, were maintained 6 months after the end of training. Gains in planning abilities were observed in girls playing Skies of Manawak, though not in boys. Importantly, improvements in cognitive areas not directly trained by SOM game were found as well: reading skills were enhanced, both in terms of speed and accuracy. In addition, significant gains were found in writing capacities. Such untrained skills were found to be improved immediately after the end of training, and the gains were maintained at follow-up. Crucially, no comparable improvements were observed as an outcome of the active control intervention, i.e. children playing the off-the-shelf video game Scratch, which teaches computer-programming bases.

Recent evidence indicates that there might be a higher chance of far transfer if the training program targets higher-level EF processes instead of focusing on lower level cognitive processing (Dahlin, 2013; Lustig, Shah, Seidler, & ReuterLorenz, 2009; Noack, Lövdén, Schmiedek, & Lindenberger, 2009; for a review on WM training: Titz & Karbach, 2014). In this regard, Dahlin and colleagues (2008) claimed that both near and far transfer would occur only when the task object of training and the new task involve overlapping cognitive functions and brain areas (Dahlin et al., 2008).

Furthermore, a generalization of training effects to important academic domains, such as Italian performance, was indicated by a mild correlation between the improvements in reading speed after training and the marks in Italian 18 months after the end of the intervention. Importantly, transfers to Italian performance – which had not been part of the training regime – were not evident at the conclusion of the training, but only after one year and a half. Thus, this study has not only indicated that EFs and literacy skills might be improved by training, but it also highlighted that these training-related benefits transferred to new, complex task, that composed the Italian evaluation.

The hypothesis that higher executive functions may be related to better academic performance, such as in the case of reading, is supported by numerous studies taking an inter-individual differences approach to evaluate the relation between various competences in childhood (DiPerna, Lei, & Reid, 2007; Howse, Calkins, Anastopoulos, Keane, & Shelton, 2003; McClelland et al., 2007; McClelland, Acock, & Morrison, 2006; Romano, Babchishin, & Pagani, 2010).

As concerns the second study, children with dyslexia who received the training with SOM (12 hours) improved significantly on all of the executive functions – as evaluated through classic paper and pencil tests. The children demonstrated improvement not only in visuo-

spatial attention, short-term/working memory and inhibition, but also on higher-level EFs. In fact, the response inhibition task involves both the capacity of inhibit predominant responses and cognitive flexibility, phonological fluency task assesses lexical access ability and updating ability, and the Tower of London task mainly evaluate working memory, interference control, and planning.

Moreover, assessment upon training completion indicated greater improvements in reading speed after Skies of Manawak than after the control training Scratch. In other words, dyslexic children in SOM group read significantly faster after a relatively short training. Regarding the accuracy of reading, a marginally significant result was found providing indications of a positive effect also on the correctness of reading.

These results are substantiated by the comparison between the reading gains after training regimens and the expected gains in one year of spontaneous reading development. Only children who trained with SOM posted larger improvements than what expected for word (0.30 syll/sec) and text (0.30 syll/sec) reading speed after one year of regular education without specific and systematic intervention (gains of SOM group in word reading: 0.38 syll/sec; text reading: 0.42 syll/sec). The improvement in non-word reading speed (decoding speed) of SOM group was the same as expected in one year without specialist treatment (0.15 syll/sec).

Finally, multiple regression analyses uncovered a link between improvements in literacy and improvements in EFs following SOM, specifically suggesting a potential transfer path from training of visuo-spatial attention to phonemic awareness and from response inhibition to reading accuracy.

Taken together, the results of the two studies pointed out that SOM training improved fundamental components of the executive system as demonstrated by the significant improvements on performance during standardized paper and pencil tests. Former studies indicating that EF skills and the levels of activation of their correlated brain areas can be changed for the better by task-repetition or training (Bavelier, Green, Pouget, & Schrater, 2012; Fu et al., 2013 Merzenich, Van Vleet, & Nahum, 2014; Gilbert et al., 2009; Merzenich et al., 2001, 2013). In fact, cognitive remediation therapy influences developmental trajectories by modulating the interaction between biological maturation - pre-specified at the genetic level - and learning (Galvan, 2010). Therefore, intensive and systematic training of EFs might positively affecting learning and the transfer of these skills by increasing the activation of the frontal lobes and other brain areas fundamental for the executive

functioning.

However, many past studies (e.g., Bergman-Nutley & Klingberg, 2014; Dowsett & Livesey, 2000; Holmes, Gathercole, & Dunning, 2009; Klingberg et al., 2005; Spencer-Smith & Klingberg, 2015) focused only on one specific executive function, most commonly working memory and inhibition, despite the fact that the EFs work as a multi-dimensional system and it would be reasonable to include more than one EF in the intervention programs. Indeed, the transfer effects of the aforementioned studies were narrow, often limited to trained EFs.

Skies of Manawak, on the other hand, was purposefully designed to train multiple EF components in order to obtain overall improvement and higher chances of generalization to untrained tasks.

Moreover, cognitive training programs are usually less enjoyable in comparison with other digital products, such as video games (Eichenbaum, Bavelier, & Green, 2014). In order to overcome the motivational barrier, Skies of Manawak consists of challenging and variable activities that require the children to be continuously committed in adapting their own behaviour, while creating a fair sense of competition. Due to the high adaptability of the video game, each player pursues a different training path: for example, the children who performed worse at the beginning of the training in a specific mini-game that trains working memory, will find more exercises that tap into that specific cognitive function. Vice versa, children that already possessed adequate levels of a specific skill will be trained less on that specific skill in favour of others. In this regard, the overall appreciation for the training activity was high in terms of entertainment, engagement and motivation for the children who played with SOM. Interestingly, in typically developing children (Study 2 - part A), a predictive role of the initial levels of childrens' sense of self-efficacy in the improvements of reading accuracy was found. Moreover, planning skills were enhanced in girls playing SOM but not in boys. These data can be linked with the results of the debriefing questionnaires where girls (and not boys) showed greater appreciation of the planning mini-game compared to other mini-games.

In conclusion, the results of the studies provided promising evidence on the application of gamified tools to enhance executive functions and reading abilities in children with and without dyslexia.

#### ***4.2 Limitations and Further Research***

Some limitations regarding the participants and the design of the two studies should be taken into consideration. Firstly, it was not possible to randomly assigned participants to the two

experimental groups. Secondly, due to technical difficulties it was not possible to obtain data on video-game improvements. Thirdly, the effects of the training regimen were only compared to those of a control activity (Scratch) and, thus, no comparison with a passive group was made. As a consequence, it was impossible to investigate maturation effects and, therefore, we used data from another study (Tressoldi, Stella, & Faggella, 2001). Lastly, for the last study, we did not conduct the kind of follow-up assessment that is important to investigate the long-term validity of the training and its generalisation to other important aspects of life such as the academic functioning.

For these reasons, a randomized controlled trial with a larger sample of children with dyslexia and a waiting list group that does not receive any kind of specialized treatment, and with the possibility of keeping trace of the participants' improvement in the targeted abilities, would be a useful next step in the exploration of the efficacy of this video game in positively affecting children's cognition.

## CHAPTER 6

### GENERAL DISCUSSION AND FUTURE DIRECTIONS

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*“Home is behind, the world ahead,  
and there are many paths to tread  
through shadows to the edge of night,  
until the stars are all alight.”*

*~ J. R. R. Tolkien*

Bridging the gap between etiological theories and effective interventions remains an important challenge for research on dyslexia, a deficit affecting 3-7% of the school-age population, and being associated with several negative consequences throughout life (Landerl et al., 2013). In fact, the impairments associated with this disorder might negatively influence the possibility for dyslexic children to reach their potential in life, as it undermines the base of adequate functioning in our society dominated by the written word (Pape, Bjørnsgaard, Westin, Holmen, & Krokstad, 2011). Reading, and more in general literacy skills, were indeed declared a fundamental human right by the United Nations Convention on the Rights of the Child (2005; 2013) and, thus, should be object of specific and timely intervention programs. These important recommendations should apply not only to the specific reading disorder, or more broadly to the poor readers (i.e. children whose reading performance lie at the low-end of the distribution), but also to other neurodevelopmental disorders (e.g. Attention-deficit hyperactivity disorder or Autism Spectrum Disorder) that share the symptom of lack of automatization of reading.

As already mentioned at the beginning of this dissertation, the diagnosis of dyslexia is still based on behavioural symptoms (i.e. reading fluency and accuracy below the norms), but it remains unclear which are the cognitive underpinnings of the reading impairments. Dyslexia is commonly described as a language-related disorder, with compromised phonological abilities being considered as the core deficit. Nonetheless, a growing body of evidence indicates that areas of deficit may be multiple, interacting and best understood in a probabilistic framework (Pennington et al., 2012; Ziegler et al., 2008; Ziegler, Perry, &

Zorzi, 2014). It has, in fact, been demonstrated that deficits in several determinants (e.g. phonological, visual, and executive domains) could underlie developmental dyslexia as highlighted by longitudinal cohort studies spanning a large array of cognitive abilities (Schatschneider et al., 2004; Franceschini, Gori, Ruffino, Pedrolli, & Facoetti, 2012; Gori, Seitz, Ronconi, Franceschini, & Facoetti, 2016; Carroll et al., 2016). Particularly, accumulating evidence indicates that dyslexia could be associated with impairments in EF skills. The “umbrella term” of EFs includes selective attention, self-control, working memory (WM), cognitive flexibility, reasoning and planning (Diamond & Ling, 2018a). This concept generally refers to a group of interrelated, top-down processes that are fundamental to accomplish a particular goal in a flexible manner (Diamond, 2013; Miyake, Friedman et al., 2000). EFs are, therefore, crucial for adequate development and seem to be more predictive of academic success than other variables, such as IQ or socioeconomic status (Blair & Razza, 2008; DiPerna, Lei, & Reid, 2007; Howse, Calkins, Anastopoulos, Keane, & Shelton, 2003; McClelland et al., 2007; McClelland, Moffitt et al., 2011; Moffitt, 2012; Morrison, & Holmes, 2000). An enhancement of Executive Functions is, thus, to be considered fundamental for harmonious development, satisfactory scholastic achievement, better-functioning social skills and better quality of life.

In this regard, there has been a mounting interest in the last decade – sometimes for commercial reasons - in trying to power EFs to “remediate deficits, improve academic performance, improve productivity, increase the likelihood of healthy choices and quality of life, and head off, slow, or reverse cognitive decline during aging” (Diamond & Ling, 2018b; pag. 1). Several different methodologies were designed in order to obtain significant improvements in one or more executive functions. These fundamental functions can be trained using a direct approach, throughout training tools – usually computerised – that target one or more executive components, or indirectly, throughout other activities, such as physical exercise, martial arts, music and language training and classroom curricula. Surprisingly, mindfulness programs that involve movement (i.e. Taekwondo, Tai chi, and Quadrato motor training) has shown by far the highest improvements in the executive domain (Diamond & Ling, 2018a). However, it has to be noted that a relatively small number of studies has been carried out on this specific topic compared to the more classical computerized training. In this regard, several research projects have shown that systematic and intensive computerized training (CogMed) could improve a number of EFs such as working memory, attention, mental flexibility and problem solving abilities (Holmes et al. 2009, Klingberg et al. 2005, Thorell et al. 2009). In addition, some evidence has been found for task-switching training

(Karbach & Kray, 2009), an integration of interactive games and computerized training (Mackey, Hill, Stone, & Bunge, 2011) and two school-based interventions, the Chicago School Readiness Project (CSRP; Raver et al. 2008, 2011) and Promoting Alternative Thinking Strategies (PATHS; Riggs et al. 2006). Promising, yet not consistent among the different studies, promising results have also been seen with two other curricula based on Tools of the Mind (Diamond, Barnett, Thomas, & Munro, 2007; Barnett et al., 2008) and Montessori (Lillard & Else-Quest, 2006; Lillard et al., 2012).

Overall, the aforementioned studies have demonstrated the possibility to enhance one or more EFs and that these improvements generalize to one measure that was not the direct object of the training.

Consequently, a debate the domain has been facing in recent years is whether training EFs may result in greater reading abilities in school-aged children with or without dyslexia. Empirical findings suggest that children with learning disabilities might benefit from early intervention programs focusing on training EFs. For example, Gori and Facoetti (Gori & Facoetti, 2014) found that perceptual learning - an improvement of perceptual skills through exercise - could specifically improve visual abilities, whose impairment characterizes some neurodevelopmental disorders such as dyslexia. In addition, Franceschini and colleagues have highlighted how Italian (2013) and English-speaking children (2017b) suffering from developmental dyslexia, a specific reading disability, may benefit from action video game training (but see Łuniewska et al., 2018). Playing action video games is known to foster several attentional components (Bejjani et al., 2014; Cardoso-Leite & Bavelier, 2014; Green & Bavelier, 2012; Green, Li, & Bavelier, 2010; see Bediou et al., 2018 for an extensive meta-analysis of action video game impact on perceptual, attentional, and cognitive skills). Another example comes from working memory training (Blakey & Carroll, 2015; Karbach et al., 2015), although meta-analytic confirmation is lacking (Melby-Lervåg et al., 2013). Melby-Lervåg and colleagues did not find convincing evidence of generalization of WM training to other cognitive skills, but only near transfer to other working memory and attentional tasks.

However, despite the fact that evidence for a positive answer is starting to emerge, as a few types of training schemes have indeed been suggested to increase reading speed and/or reading accuracy, several of the previous studies present some limitations.

Not all the studies used random assignment, and included pre- and post-intervention measures and active control group, resulting in potential confounds related to motivation or expectations (e.g. placebo or Hawthorne effects). In addition, most of the existing

computerized training tend to focus on the training of single functions, thus ignoring the heterogeneity of many neurodevelopmental disorders, dyslexia *in primis*. Moreover, EFs should be considered as a multi-dimensional system and it could be reasonable to include more than one EF in the intervention programs. Finally, the action video game studies all focused on reading-impaired children, and therefore little is known about attentional training and transfer in the general school-aged population.

Therefore, despite the fact that many cognitive training companies advertise their products as extremely effective for a wide set of cognitive and emotional functions, there is a glaring lack of evidence that computerized training are able to consistently improve EFs and that these improvements generalize to other – untrained – domains.

In an effort to overcome scientific inconsistencies and disagreements, this research aims to analyse new evidence-based treatment methodologies based on a multifactorial, probabilistic, model of dyslexia.

## **1. Efficacy**

In the first study, we developed a training program that combined phonological-based intervention and training of the EFs. The two types of interventions should not be seen as conflicting, but rather as complementary. After 24 hours of Integrated training, dyslexic children improved in visuo-spatial attention, working memory and verbal fluency at post-test (T3), while no significant improvements in EFs were observed in the pure Phonological-based Treatment. Importantly, these gains transferred to untrained reading abilities, such as writing and reading efficiency. In fact, despite the short duration of the treatment program, the integration of the Cognitive Training and the Phonological-based treatment resulted in significant gains in both reading accuracy and speed. Specifically, children of the Integrated group posted higher improvements in reading fluency than expected in one year of development without specialized interventions. In conclusion, the treatment effects can be regarded as satisfactory for both interventions, even though the Integrated group obtained the most positive effects due to the fact that combining the two methods helped children with dyslexia to maximize the chance of enhancing their reading skills. Therefore, it is possible to train EFs, and the improvements in these fundamental skills seem to positively affect reading and writing.

These results are supported by the findings of the second experiment with typically developing (Study 2 - Part A) and dyslexic children (Study 2 - Part B), in which the efficacy of a unique video game (*Skies of Manawak*) purposefully designed to train several EFs was evaluated. Our goal was to develop a new training tool in which effective training and a playful and engaging experience can coexist in order to maximize its effectiveness and to have higher chances of transfer to untrained tasks. Assessment upon training completion (12 hours) revealed that only children that trained with SOM significantly improved their EFs, while children of the control group (*Scratch*) did not. Interestingly, the EF tasks shared few surface features with - but similar underlying cognitive demands as - the mini-games of the video game. This clearly highlighted that the improvements in performance was due to a real transfer and not to test-retest effects (e.g., increased familiarity with the specific task or with the examiner).

Specifically, in Study 2 - Part A the improvements were observed not only in visuo-spatial attention and working memory, but also on a higher-level executive function (planning), as evaluated through the Tower of London task. In addition, results of the second study, in which a broader assessment battery was used, demonstrated the positive effects of training on attention, working memory, inhibition, phonological fluency and planning. In both studies, the greater improvements (as demonstrated by the effect sizes of the Time X Group interactions) were found in the tasks that involve complex, multi-component measures. As Diamond and Ling pointed out in their recent review (2018b): “Often the benefits of an intervention are only seen, or are seen most clearly, on outcome measures that push the limits of participants’ EFs” (pag. 28). In fact, the response inhibition task involves both the capacity of inhibit predominant responses and cognitive flexibility, phonological fluency task assesses lexical access ability and updating ability, and the Tower of London task mainly evaluate working memory, interference control, and planning. Therefore, despite the obvious limitation that it is impossible to isolate which particular EF skill improved, these tasks are often useful in detecting outcome differences between the training groups (e.g., Albinet, Boucard, Bouquet, & Audiffren 2010; Alesi, Bianco et al., 2016; Diamond et al., 2007; Hillman, Pontifex et al., 2014; Manjunath & Telles, 2001; Schmidt, Jäger, Egger, Roebers, & Conzelmann, 2015; Westendorp, Houwen et al., 2014).

Another important result clearly emerged from all the studies of this research: children who are more behind on EF performance benefited the most from the training (both the Brain-HQ and the SOM training). This is in line with the literature in the field (e.g., attention: Flook,

Smalley et al., 2010; working memory capacity: Holmes et al., 2009; inhibitory control: Drollette, Scudder et al., 2014; EFs overall: Holmes, Gathercole et al., 2010; Klingberg et al., 2005). For example, Fedewa and Ahn's (2011) demonstrated in their meta-analysis that the mean effect size in the different studies on physical activity was twice as large for children with cognitive impairments as for typically-developing ones. In addition, for many variables a link with chronological age was found: younger children displayed the largest gains at the end of the training. Reasons can be found in higher levels of plasticity, but also in a higher level of motivation towards the training activity. An example can be found looking at the improvements in the planning performance at T2 and the level of appreciation of the mini-game, as evaluated in the feedback questionnaire: only typically-reading females (not males) significantly improved in planning capacities and females were the ones that most liked the planning mini-game. However, the link between motivation and training improvements mainly comes from the observations of the clinicians and, thus, should be object of further research.

No effects were found regarding IQ and socioeconomic status. Several studies have demonstrated that general intelligence do not predict the responsiveness of children to intervention (Frijters et al., 2011; Fuchs & Young, 2006; Stuebing et al., 2015; Stuebing, Barth, Molfese, Weiss, & Fletcher, 2009). Vice versa, it has been shown that individuals who are socio-economically disadvantaged, particularly in childhood, tend to benefit more from EF interventions than children with more socio-economic advantages (Blair & Raver, 2014; Lillard & Else-Quest, 2006; Mackey, Hill, Stone, & Bunge, 2011). A possible explanation of the lack of significant effects of socioeconomic levels in these studies can be found in the low variability of our participants: they all came from two regions in the north of Italy, presenting (with few exceptions) a SES above the Italian average.

In addition to clear improvements in the executive functioning, post-training assessments indicated greater improvements in reading efficiency after Skies of Manawak than after the control training Scratch. In other words, both typically developing (Study 2 - Part A) and dyslexic children (Study 2 - Part B) that underwent SOM training read significantly faster. Analogously, a significant result was found in reading accuracy for children with typical development and a marginally significant result for dyslexic children, thus providing firm indications of a positive effect also on the correctness of reading.

The results of SOM training, as well as the Integrated treatment, are substantiated by the comparison between the reading gains after training regimens and the expected gains in one

year of spontaneous reading development. Only children of these intervention groups made equal or larger improvements than expected for reading fluency after one year of regular education without specific and systematic intervention.

The aforementioned findings are notable, considering existing work focusing on the educational outcomes of video games training in childhood. Franceschini and colleagues (2013, 2017b) found enhancements only in reading speed in Italian and English-speaking dyslexic children, following brief training in an action video game (child-friendly mini-games from Rayman's Raving Rabbids game, bearing action video games characteristics). However, in these two studies reading accuracy remained unchanged.

Interestingly, the advantage in reading skills obtained after SOM training by typically reading children was maintained in a follow-up test 6 months later and appear to generalize to academic performance (i.e., Italian marks). Numerous studies (DiPerna, Lei, & Reid, 2007; Howse, Calkins, Anastopoulos, Keane, & Shelton, 2003; McClelland et al., 2007; McClelland, Acock, & Morrison, 2006; Romano, Babchishin, & Pagani, 2010) supported the hypothesis that higher EFs may be related to better academic performance (i.e., reading).

Finally, multiple regression analyses uncovered a link between improvements in literacy and improvements in EFs following intervention in the studies with dyslexic children, but not with typically developing ones. These exploratory results suggested a potential transfer path from training of EFs (particularly visuo-spatial attention and inhibition) to literacy proficiency.

## 2. Feasibility

The results from this research suggest that the EF training programs were feasible to administer both in a school and home-based setting. Nonetheless, some further considerations should be made. Firstly, training with Brain-HQ was appreciated by most of the children; however, this positive framework was counterbalanced by some complaints from children and parents. If on one hand the computer-based intervention immediately appealed to most of them, Brain-HQ was in the end considered boring and difficult by some children. Thus the training had to be enforced by the parents and, in some cases, was considered more as additional homework than a playful diversion. This attitude is well exemplified in the words of a child participant (age 10) that dropped the training after one week and a half: “[...] *I don't know if I would rather prefer to do this again or my math homework*”. Therefore, for some children (especially the ones with low resistance to frustration) the appeal of the

gamification elements quickly vanished, whereas the performance anxiety induced in some of them tended to last. Overall, the interactive system of rewards did not appear to sufficiently motivate all of them.

On the other hand, feedback on Skies of Manawak was extremely positive indicating the importance of a stronger game design in order to provide a captivating and motivating experience. The majority of parents were satisfied with the video game and would like to suggest this intervention to other parents who had children with dyslexia. Furthermore, both parents and children asked for a second chapter of the adventure (respectively, 75.8% and 86.6%).

However, it has to be mentioned that even with SOM, some players did not fully understand the story, perceiving the training as a mere continuous repetition of the mini-games. Therefore, a possible re-design phase should create more immediate rewards (e.g., more prizes and more varied ones) and a greater customization of the content. Another possibility could be to add other mini-games on the secret islands and to expand the variety of the environments; these two elements, in fact, were particularly appreciated and softened the sense of repetitiveness of the structure of the training.

In addition, some further reflections should be made: many children that trained with Brain-HQ and SOM at home were completely left without supervision by the parents. Specifically, many of these parents reported differences between the attitude that their children had towards the computerized training tool: when they were alone they easily lost the motivation, while they really enjoyed the sessions with the clinician at the ODFLab. These observations strongly support the role of a supportive mentor, who believes in the program and the ability of participants to succeed. It could be the case that technology itself should not be considered sufficient to positively affect cognition and emotion. For example, when Cogmed training (WM) was compared with other training curricula characterized by a significant trainer-participant interaction, not many differences between the groups were observed (Gray et al., 2012; van der Donk et al., 2015). In this regard, de Jong (2014) hypothesized that the motivational component could have played a fundamental role in determining the benefits of the Cogmed training.

In addition, in the study with a school-based setting, children were able to work together with other classmates toward a common goal (e.g., to beat the villain at the end of SOM adventure). As Diamond and Ling observed (2018b): “Some of the best results for improving

EFs have come from programs that build feelings of community and connections with others” (p.125).

Many of the players particularly appreciated the fact that what had looked nearly impossible at the beginning of the training, became easier and more satisfying at the end (after hours of practice). Indeed, Skies of Manawak consisted of challenging and variable activities that require the children to be continuously committed to adapting their own behaviour, constantly keeping the child in his/her “zone of proximal learning” (Vygotskij, 1978). Due to the high adaptability of the video game, each player pursues a different training path. Therefore, children realized that they were able to accomplish results they had never thought possible, enhancing their self-esteem and sense of self-efficacy.

However, the character and quality of interpersonal aspects were not directly taken into account in this research. Further studies need to be planned in order to systematically evaluate the role of these important variables in the benefit of a program. In this regard, Diamond (2016; 2018) predicted that an EF program that “brings joy, builds self-confidence, and enhances social well-being” should be able to produce more significant improvements than a program focusing only on challenging EFs.

### **3. Conclusion**

The current project addressed the important issue of finding efficacious evidence-based treatments for developmental dyslexia on one hand, and of proposing a new training tool for typically developing and dyslexic children on the other. The design of each intervention program was carefully based on current neuropsychological findings that indicate the need to consider multifactorial models at the base of an individual’s adequate functioning. Evaluation of their efficacy clearly showed advantages in using EF training to improve not only EFs, but also literacy skills for the better. In addition, phonological-based treatment might not be considered enough to achieve the best results, at least in some children.

Future work will include a longitudinal evaluation of the training effects also in dyslexic children. It will be interesting to assess Skies of Manawak efficacy in children speaking other languages, such as English or French, to understand whether benefits are seen in an opaque orthography, in which phonological deficits are expected to play a larger role.

In addition, it might be useful to expand the number of mini-games adding some exercises that train phonological awareness. The phonological training should focus on enhancing children's capacity of synchronizing to auditory rhythmic stimuli.

Finally, the results obtained from these first evaluations form a promising basis for more individually based training. Therefore future research should more carefully address the cognitive profile of each child beyond language and into the executive domain – in order to better capture the many possible underlying deficits of reading or, more broadly, academic difficulties. Furthermore, a new re-design of Skies of Manawak should focus on developing a user interface accessible not only to researchers, but also to clinicians in order to reach the best training algorithm on the basis of the pattern of strengths and weaknesses of each child. The main challenge for the remediation of this disorder is not only to find the most effective remediation programs, but also to precisely select a personalized program for each child with dyslexia.

The hope is that a shared theoretical framework can in turn illuminate evidence-based practical guidelines for treatment.

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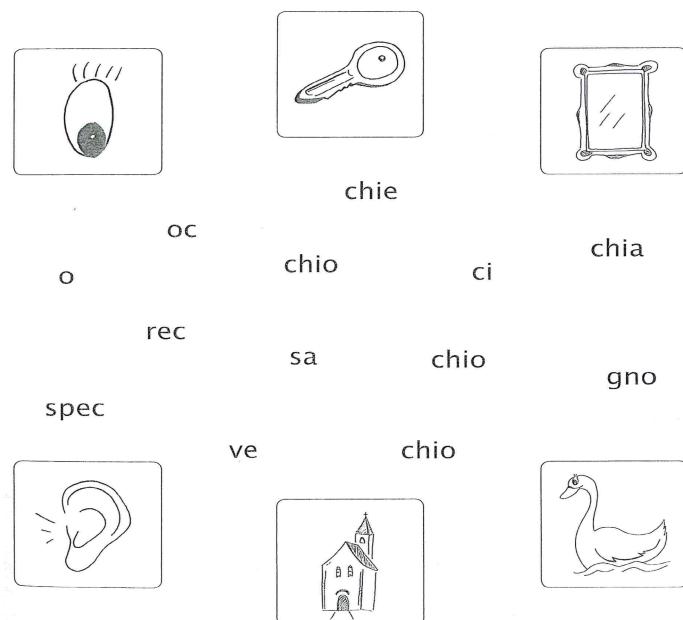
## APPENDIX A

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**PHONOLOGICAL-BASED TREATMENT:** examples of the activities proposed.

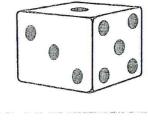
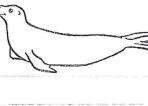
Form words (some of them are represented in pictures) with the syllables presented below.

Source: "Dislessia e Trattamento sublessicale" [Dyslexia and sublexical treatment] of Cazzaniga and colleagues (2007).



Read as fast and correctly as possible the following syllables proceeding from left to right.

Source: "Occhio alle parole" [Watch words] of Brignola, Perrotta and Tigoli (2012).

BA	BE	BI	BO	BU	
BI	BU	BA	BE	BO	
BE	BO	BU	BA	BI	
CA	CHE	CHI	CO	CU	
CO	CHI	CA	CU	CHE	
CHI	CO	CU	CHE	CA	
CIA	CE	CI	CIO	CIU	
CIO	CI	CIU	CIA	CE	
CIU	CIA	CE	CI	CIO	
DA	DE	DI	DO	DU	
DO	DU	DI	DA	DE	
DI	DA	DO	DE	DU	
FA	FE	FI	FO	FU	
FO	FI	FU	FA	FE	
FA	FE	FO	FU	FI	

Read as fast and correctly as possible the following words proceeding from left to right.  
Source: “Lettoscrittura vol.6” [Literacy skills vol.6] of Gagliardini (2011).

angolo	ordina	arne
uncino	invade	dine
ondata	alzare	urbano
indice	invito	ultimo
urlare	ungere	altare
angelo	albero	antico
anfora	orfano	intero

Read as fast and correctly as possible the following non-words proceeding from left to right.  
Source: “Lettoscrittura vol.6” [Literacy skills vol.6] of Gagliardini (2011).

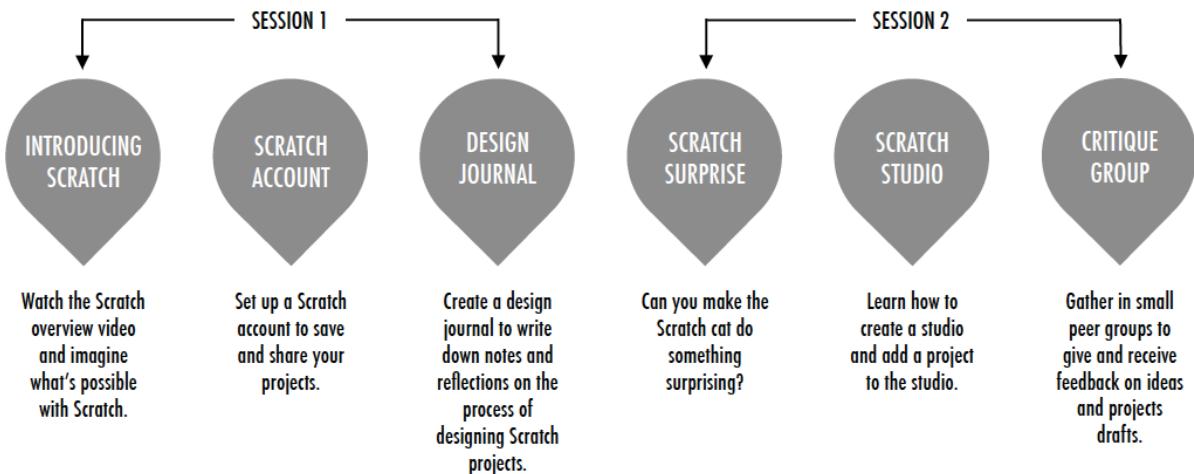
verba	sando	zalbo
rinzo	dulda	tolsa
pilco	mompe	nerzi
tilte	rurda	filce
darbo	tenuv	sampi
zilte	cunba	domci
rirtu	vesmo	borde
nenza	tirne	mantu

## APPENDIX B

### SCRATCH PROTOCOL

(source:<http://scratched.gse.harvard.edu/guide/download.html>)

#### UNIT 0 – GETTING STARTED



N.b. For time reasons, we skipped the “design journal part” and, it has to be noticed that some classes were not able to do the “critique group”.

Example of Unit 0 activity:

**UNIT 0 ACTIVITY**

## SCRATCH SURPRISE

SUGGESTED TIME  
15-30 MINUTES

**ACTIVITY DESCRIPTION**

- Help students open the Scratch project editor by navigating to the Scratch website at <http://scratch.mit.edu>, signing in to their Scratch accounts, and then clicking on “Create” at the top of the page. Optionally, have the Scratch Surprise handout and Scratch Cards available to guide students during their explorations.
- Give students 10 minutes to explore the Scratch interface in an open-ended way. Prompt students with, “You have 10 minutes to make something surprising happen to the Scratch cat.” Or, “Take 10 minutes to explore the interface fearlessly. What do you notice?” Encourage students to work together, ask each other for help, and share what they are figuring out.
- Ask for 3 or 4 volunteers to share with the entire group one thing that they discovered. Optionally, after the volunteers have shared, offer several challenges to the students:
  - Did anyone figure out how to add sound?
  - Did anyone figure out how to change the background?
  - Did anyone figure out how to get help with blocks?

**RESOURCES**

- Scratch Surprise Handout
- Scratch Cards  
<http://scratch.mit.edu/help/cards>

**REFLECTION PROMPTS**

- + What did you figure out?
- + What do you want to know more about?

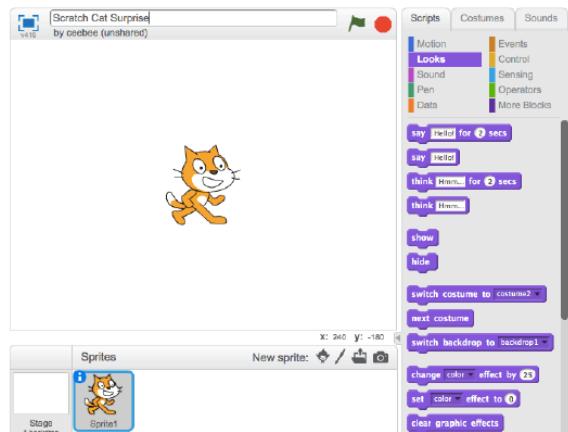
**REVIEWING STUDENT WORK**

- + Do students know how to initiate a new project?
- + Do students understand the basic mechanism of snapping Scratch blocks together?

# SCRATCH SURPRISE

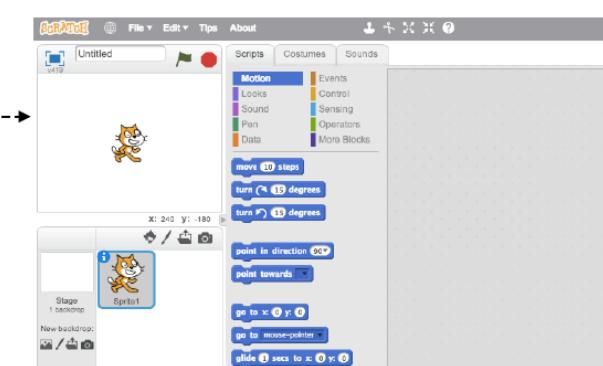
CAN YOU MAKE THE SCRATCH CAT DO SOMETHING SURPRISING?

In this activity, you will create a new project with Scratch and explore different Scratch blocks to make the cat do something surprising! What will you create? ----->

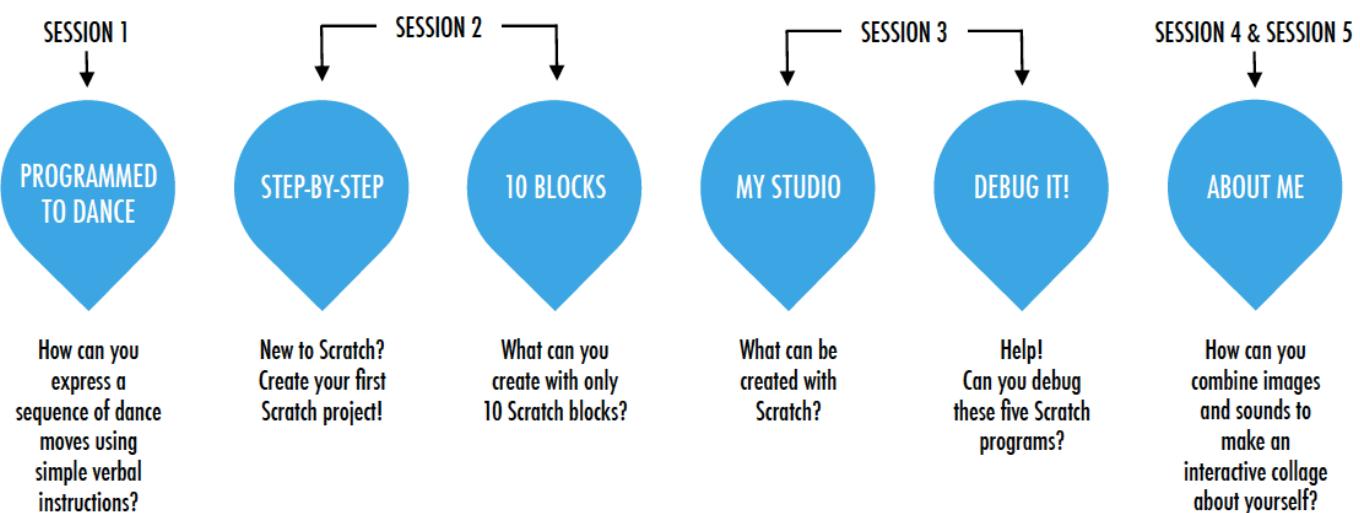


## START HERE

- Go to the Scratch website: <http://scratch.mit.edu>
- Sign into your account.
- Click on the "Create" tab located at the top left of the browser to start a new project.
- Time to explore! Try clicking on different parts of the Scratch interface to see what happens.
- Play with different Scratch blocks! Drag and drop Scratch blocks into the scripting area. Experiment by clicking on each block to see what they do or try snapping blocks together.



## UNIT 1 – EXPLORING



Example of Unit 1 activity:

**UNIT 1 ACTIVITY**

# ABOUT ME

SUGGESTED TIME  
45–60 MINUTES

**OBJECTIVES**  
By completing this activity, students will:  
+ become familiar with a wider range of Scratch blocks  
+ be able to create an open-ended Scratch project that is an interactive digital representation of their personal interests

### ACTIVITY DESCRIPTION

- Introduce students to the concept of the interactive collage, a Scratch project that represents aspects of themselves through clickable sprites. Optionally, show interactive project examples from the About Me studio.
- Have students sign in to their Scratch accounts and open a new project. Optionally, have the About Me handout and Scratch Cards available to provide guidance. Give students time to create an About Me interactive collage Scratch project, encouraging them to build up their programs by experimenting and iterating.
- Allow students to share their works-in-progress with others. We suggest pair-share: have students share and discuss their projects in pairs. Optionally, invite students to add their projects to the About Me studio or a class studio.
- Ask students to think back on the design process by responding to the reflection prompts in their design journals or in a group discussion.

### RESOURCES

- About Me handout
- About Me studio  
<http://scratch.mit.edu/studios/475470>
- Scratch Cards  
<http://scratch.mit.edu/help/cards>

### REFLECTION PROMPTS

- + What are you most proud of? Why?
- + What did you get stuck on? How did you get unstuck?
- + What might you want to do next?
- + What did you discover from looking at others' About Me projects?

### REVIEWING STUDENT WORK

- + Do projects make creative use of sprites, costumes, looks, backdrops, or sound?
- + Are projects interactive? Can users interact with various elements within the project?

# ABOUT ME

HOW CAN YOU COMBINE INTERESTING IMAGES AND SOUNDS TO MAKE AN INTERACTIVE COLLAGE ABOUT YOURSELF?

Experiment with sprites, costumes, backdrops, looks, and sounds to create an interactive Scratch project – a project that helps other people learn more about YOU and the ideas, activities, and people that you care about.

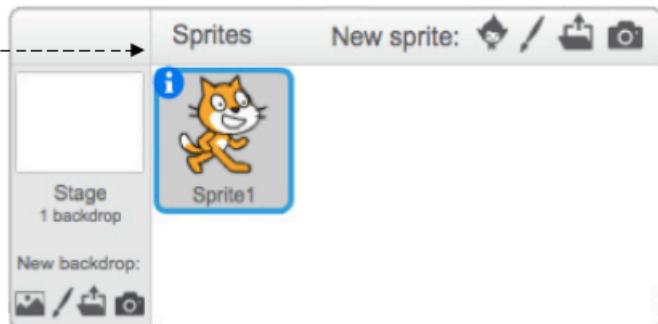


## START HERE

- Create a sprite.
- Make it interactive.
- Repeat!



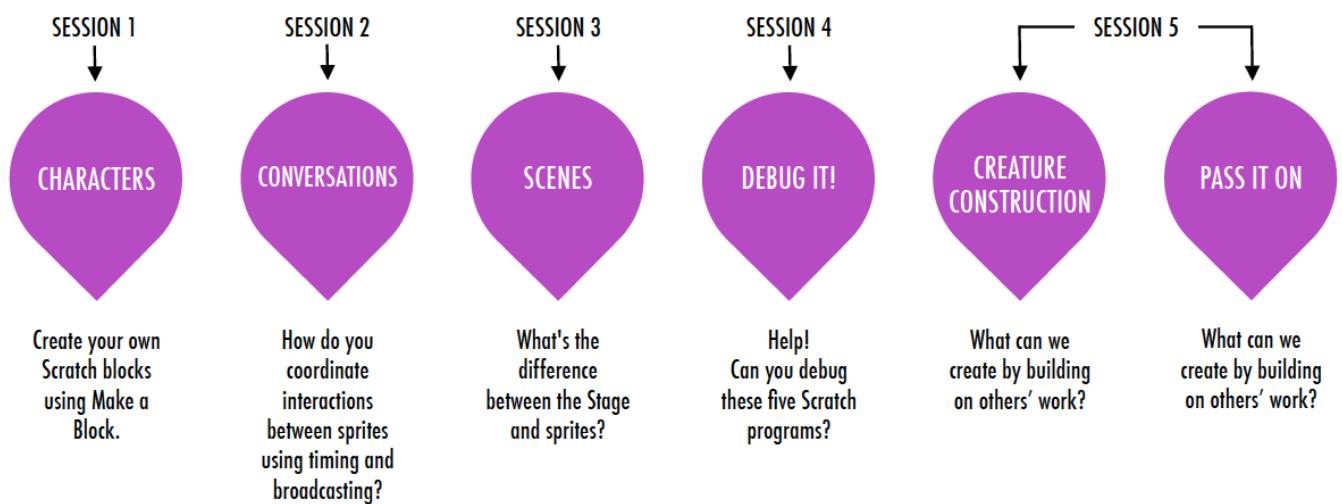
Make your sprite interactive by adding scripts that have the sprite respond to clicks, key presses, and more!



## THINGS TO TRY

- Use costumes to change how your sprite looks.
- Create different backdrops.
- Try adding sound to your project.
- Try adding movement into your collage.

## UNIT 3 – STORIES



Example of Unit 3 activity:

### UNIT 3 ACTIVITY

# CHARACTERS

SUGGESTED TIME  
30–45 MINUTES

**OBJECTIVES**

By completing this activity, students will:

- + experiment with defining behaviors for characters using Scratch's Make a Block feature
- + gain more familiarity with the computational concepts of events and parallelism and the practice of experimenting and iterating

#### ACTIVITY DESCRIPTION

- Optionally, show example projects from the Characters studio and have the Characters handout available to guide students.
- Give students time to create their own Scratch blocks using the Make a Block feature found in the More Blocks category. Help them design two sprites or "characters" that each have two behaviors. Optionally, conduct a walkthrough of the Make a Block feature together as a class.
- Allow students to share their characters and behaviors with one another. We suggest the design demo activity: invite a few students to present their work to the class and demonstrate how they implemented the Make a Block feature. Optionally, have students add their projects to the Characters studio or a separate class studio.
- Ask students to think back on the design process by responding to the reflection prompts in their design journals or in a group discussion.

#### RESOURCES

- Characters handout
- Characters studio  
<http://scratch.mit.edu/studios/475545>

#### REFLECTION PROMPTS

- + How would you explain Make a Block to someone else?
- + When might you use Make a Block?

#### REVIEWING STUDENT WORK

- + Do projects include two sprites that each have two behaviors using the Make a Block feature?
- + Can students explain how to use the Make a Block feature to each other and to you?

# CHARACTERS

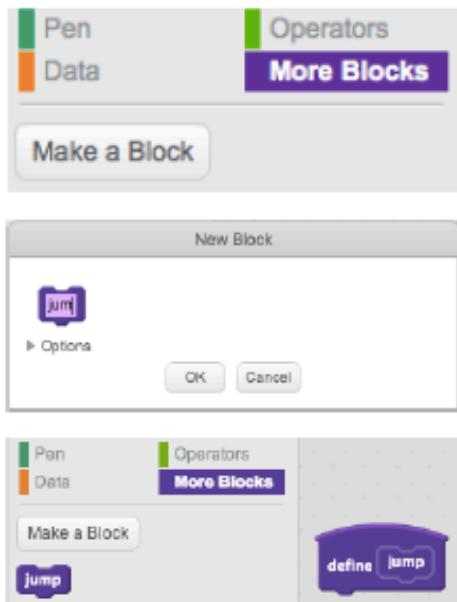
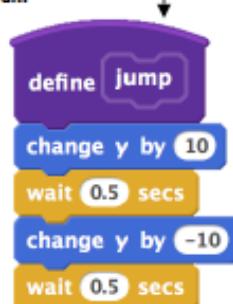
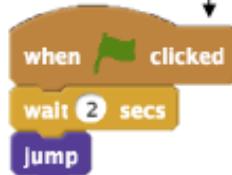
DO YOU WANT TO CREATE YOUR OWN SCRATCH BLOCKS?

Experiment with the Make a Block feature in Scratch! In this project, you will create your own blocks that define two behaviors for two different characters.



## START HERE

- Choose from the library, paint, or upload two sprite characters.
- Click on the Make a Block button in the More Blocks category to create and name your block.
- Add blocks under the Define block to control what your custom block will do.
- Experiment with using your block to program your characters' behaviors.
- Repeat!



## APPENDIX C

## **FEEDBACK QUESTIONNAIRE**

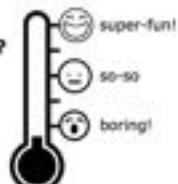
WHAT DO YOU THINK OF SKIES OF MANAWAK?

I am   and I am  years old.

number \_\_\_\_\_

- ① How much fun did you have playing the game?

Fill the thermometer until your fun level



- ③ How would you describe this game?

Tick nine or more words

110

exciting

第二部分

### confusing

easy

difficult

ANSWER

### **Intuitive**

### **childish**

- ③ For each question tick an answer

- Would you suggest it to a friend?       yes     maybe     no
  - If you could take it home, would you play it?       often     sometimes     never
  - Would you like to play the second episode?       yes     maybe     no
  - Did you find it complex to play?       often     sometimes     never
  - Did you like the aesthetics (drawings, colours, ...)?       yes     no opinion     no
  - While you were playing, were you curious to find what was next?       yes     no opinion     no
  - Would you consider this game (positively) challenging?       often     sometimes     never
  - Did you feel bored?       often     sometimes     never
  - Compared to the beginning, do you feel to be more skilled in playing the game?       yes     no opinion     no

- Now that you have completed Skies of Manawak, you feel to be improved ...

For each question tick an answer

- Do you feel more focused? yes    maybe    no
  - Do you think you remember more easily the instructions that you receive? often    sometime    never
  - Do you feel less distracted? yes    maybe    no
  - Do you think you understand better what you have to do in the various tasks? often    sometime    never

① What did you NOT like playing Skies of ManaWak?

Tick only one box and write your answer below



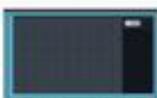
Flying with the Raku and fighting sentinels



Perform the ritual with the Uka



Calling the Raku on the Tower (sounds)



Solve the puzzle of the tower's panel



Memorize symbols and order of the tiles



Feed the Raku



Jump with the character and avoid the meteors with the Raku



Shooting while sliding only when you hear the right sound



The game of the triangles on the mysterious island



The shooting gallery on the mysterious island

Why? Tell us what you think by writing your answer below

② What did you like playing Skies of ManaWak?

Tick only one box and write your answer below



Flying with the Raku and fighting sentinels



Perform the ritual with the Uka



Calling the Raku on the Tower (sounds)



Solve the puzzle of the tower's panel



Memorize symbols and order of the tiles



Feed the Raku



Jump with the character and avoid the meteors with the Raku



Shooting while sliding only when you hear the right sound



The game of the triangles on the mysterious island



The shooting gallery on the mysterious island

Why? Tell us what you think by writing your answer below

① What did you find difficult playing Skies of Manawak?

Tick only one box and write your answer below



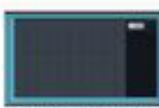
Flying with the Raku and fighting sentinels



Perform the ritual with the Uka



Calling the Raku on the Tower (sounds)



Solve the puzzle of the tower's panel



Memorize symbols and order of the tiles



Feed the Raku



Jump with the character and avoid the meteors with the Raku



Shooting while sliding only when you hear the right sound



The game of the triangles on the mysterious island



The shooting gallery on the mysterious island

Why? Tell us what you think by writing your answer below

② Would you have any suggestion for the second episode of Skies of Manawak?