UNIVERSIDADE FEDERAL DE MINAS GERAIS

FACULDADE DE MEDICINA

PROGRAMA DE PÓS-GRADUAÇÃO EM SAÚDE DA CRIANÇA E DO ADOLESCENTE

JÚLIA BEATRIZ LOPES SILVA

Investigation of linguistic influences on number transcoding in children

Investigação da influência de fatores linguísticos na transcodificação numérica em crianças

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Tese de doutorado apresentada ao Programa de Pós-Graduação em Saúde da Criança e do Adolescente, da Faculdade de Medicina da Universidade Federal de Minas Gerais como requisito parcial à obtenção do título de Doutora em Saúde da Criança e do Adolescente

Orientador: Prof. Dr. Vitor Geraldi Haase

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PROGRAMA DE PÓS-GRADUAÇÃO EM CIÊNCIAS DA SAÚDE SAÚDE DA CRIANÇA E DO ADOLESCENTE



FOLHA DE APROVAÇÃO

INVESTIGATION OF LINGUISTIC INFLUENCES ON NUMBER TRANSCODING IN CHILDREN.

JÚLIA BEATRIZ LOPES SILVA

Tese submetida à Banca Examinadora designada pelo Colegiado do Programa de Pós-Graduação em Ciências da Saúde, Saúde da Criança e do Adolescente, como requisito para obtenção do grau de Doutor em Ciências da Saúde, Saúde da Criança e do Adolescente, área de concentração Ciências da Saúde.

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LIST OF ABBREVIATIONS

- ADAPT A Developmental Asemantic and Procedural Transcoding Model
- AGFI Adjusted Goodness of Fit Index
- ANS Approximate Number System
- AUC Area Unider the Curve
- CFI Comparative Fit Index
- GFI Goodness of Fit Index
- IQ Inteliigence Coefficient
- KR-20 Kuder–Richardson Formula 20
- LTM Long-term memory
- **MD** Mathematics Difficulties
- MLD Mathematics Learning Disabilities
- MSLD Mathematics and Spelling Learning Disabilities
- **RAN Rapid Automatized Naming**
- Raven's CPM Colored Progressive Matrices
- **RD** Reading Difficulties
- RMR Root Mean square Residual
- RMSEA Root Mean Square Error of Approximation
- TDE Brazilian School Achievement Test
- TD Typically Developing
- w Internal Weber Fraction
- WM Working Memory

ABSTRACT

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An important skill to acquire proficiency in mathematics is number transcoding, which is the ability of converting numerical information between different systems of numerical notation. The main goal of this dissertation is to investigate the cognitive variables associated to number transcoding, focusing on the phonological processing. Changing from the Verbal to the Arabic symbolic notations involves access to phonological representations but this association has not been systematically explored yet. At first, on Chapter 1, we describe the current state of literature, regarding the most relevant cognitive models of number transcoding, its impact on mathematical learning disabilities and the association between phonological and numerical processing. We also provide a brief summary of the three studies that constitute the dissertation. On the Study 1 (Moura, Lopes-Silva, Vieira, de Almeida Prado, Wood, Haase, 2014) we report normative parameters such as mean, range values, and percentiles for first to fourth grades obtained from a large sample of school children. In addition, the diagnostic accuracy of the number-writing task in the detection of children at risk for mathematical difficulties and the influence of place-value syntax in children's achievement, were investigated. On the study 2 (Lopes-Silva, Moura, Wood, Júlio-Costa, Haase & Wood, 2014) we have found that, among the measurements of magnitude processing, working memory and phonemic awareness, only the last one was retained in regression and path models predicting transcoding ability. Phonemic awareness mediated the influence of verbal working memory on number transcoding. At last, on the study 3 (Lopes-Silva, Moura, Júlio-Costa, Wood, Salles & Haase, 2016) we have shown that all of the writing and reading tasks (single word spelling and reading as well as number reading and number writing) were significantly correlated to each other. In the regression models, phonological WM was specifically associated to word reading. Phonemic awareness was the only cognitive variable that systematically predicted all of the school skills investigated, both numerical and word tasks. The findings of this dissertation provide supporting evidence that verbal to Arabic number transcoding is a useful tool for diagnosing children with mathematical learning disabilities. Besides that, we provide an important contribution regarding the role of the phonological processing in number transcoding.

Keywords: number transcoding, dyscalculia, phonological processing, phonemic awareness

PREFACE

Research on the development of numerical abilities has gained considerable attention over the last decades. The establishment of a link between Verbal and Arabic numerical notations, namely number transcoding, is one of the most elementary skills acquired in the first years of formal schooling. Despite being considered a very basic numerical ability, number transcoding plays a prominent role in the study of numerical cognition and mathematics learning, since it is one the fundamental pillars on which more complex numerical skills, such as calculation, develop.

The present dissertation aims at investigating cognitive skills associated with number transcoding, and, more specifically, how phonological processing would be associated with it. Phonological processing is traditionally associated to reading and spelling and, more recently, some studies have demonstrated its association to mathematics, especially to multiplication (see Simmons & Singleton, 2008, 2009; DeSmedt, Taylor, Archibaldi & Ansari, 2010). The literature review described on the first chapter of this dissertation allowed us to observe that, even though number transcoding clearly has a verbal component, there are not enough studies that investigate how phonological processing could impact on transcoding. After we had identified this research question, we have performed three studies to investigate psychometric properties of a Verbal to Arabic number writing task and to disentangle the role of phonological processing in transcoding.

Structure of the dissertation:

According to the guidelines of the Post-Graduation Program in Child and Adolescent Health, from the Medicine Faculty of UFMG, this dissertation will be constituted by scientific papers. Therefore, the present dissertation has a literature review and three empirical papers. The first chapter, in addition to presenting the current state of the literature, also explains our argument and summarizes the three studies. The first one is a psychometric paper on a number writing task developed by the Developmental Neuropsychology Laboratory of UFMG, entitled "From "five" to 5 for 5 minutes: Arabic number transcoding as a short, specific, and sensitive screening tool for mathematics learning difficulties", which was published on Archives of Clinical Neuropsychology, on 2014. Afterwards, on the next chapter, we focus on the association between phonological processing. We report the results from the paper which was also published in 2014, in Frontiers in Psychology, "Phonemic awareness as a pathway to number transcoding". On this study, we discuss how phonemic awareness could mediate the influence of phonological working memory in number writing. Finally, we describe the paper "What is specific and what is shared between numbers and words", also published in Frontiers in Psychology, on 2016, which investigated the shared association of phonological processing to both numbers and words. At last, we briefly discuss how the results reported in this dissertation builds on the previous literature, as well as limitations and future directions.

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CHAPTER 1

General Introduction

The mastery of reading and writing numbers, in their verbal and Arabic form is an essential numerical skill (Lochy & Censabella, 2005). Daily activities require the communication of numerical information, such as registering a telephone number or performing calculations. Besides that, being able to manipulate numbers is one of the first steps on children's mathematical learning and it starts to be formally trained in kindergarten. The ability to establish a relationship between the verbal and Arabic representations of numbers, when a conversion of numerical symbols from one notation to the other is necessary, is called number transcoding (Deloche & Seron, 1987).

In spite of its importance, only few studies have investigated the cognitive mechanisms that underpin numerical processing in children. Children with mathematical learning disabilities reliably present difficulties with the numerical symbolic codes and transcoding is one these problems (Geary, Hamson & Hoard, 2000). Could performance in number transcoding be an indicative of arithmetic impairments? Another relevant question comes from the fact that there is a high comorbidity rate between dyslexia and dyscalculia (Landerl & Moll, 2010) and children with word reading difficulties present impairments in verbal aspects of math, such as arithmetic word problems and multiplication (Simmons & Singleton, 2008). Most studies with dyslexic children, who present phonological processing deficits, have focused on their general arithmetic performance and not in more basic number processing. Would number transcoding be one of these verbal aspects of math that could be consistently impaired by phonological processing deficits? Phonological processing may play a role in the initial encoding of the verbal input. In addition, phonological processing can interact with the capacity of verbal working memory: the higher phonological processing demands of a given numeric stimuli, fewer resources would be available to working memory (Lopes-Silva et al, 2014.). It is interesting to note that although the conversion of a verbal representation to an Arabic one is related to the quality of phonological representations, this association has not yet been systematically investigated on number transcoding. To address these questions in further detail, in this chapter, we will describe the current state of the literature regarding number transcoding and its underlying cognitive mechanisms, emphasizing open questions that needed deeper investigation. Afterwards, we present an overview of the studies that constitute this dissertation, detailing our specific goals and summarizing our main results.

The symbolic codes

The ability to use symbols to represent mental representations is one of the most remarkable landmarks in the evolution of human species. There are two main culturally-learned symbolic systems intended to represent numerical information: the verbal-numerical and the visual-Arabic systems. The Arabic code (e.g., 23) allows for exact arithmetic operations and the Verbal code (e.g., twenty-three), supported by language skills, enables the comprehension of orally presented arithmetic problems. On the Verbal code, there are interesting language-specific idiosyncrasies. In English and in Portuguese, for instance, there is a lexical order compatibility between the verbal form and its Arabic one (e.g., 48: *forty-eight, quarenta e oito*). In these cases, the order of the numerical terms corresponds to the order of the place value system. An interesting contrasting example is German, in which there is decade-unit inversion rule in number reading. Forty-eight (48), for example, is literally read as "eight forty" (*achtundvierzig*). The Arabic code is composed by a finite set of lexical units (digits from 1 to 9, and 0) that can be arranged to represent infinite numbers. This syntactic organization of digits follows the place-value principle, according to which the value that certain digit represents depends on its position on the number. The first digit

(from right to the left) is multiplied by 10° , the second to 10^{1} and so on. The number 124, for example, represents a quantity equals to $1 \times 10^{2} + 2 \times 10^{1} + 4 \times 10^{\circ}$, (or 100 + 20 + 4). The digit 0 has a special syntactic role once it denotes the absence of a given power of ten, as occurs in number with internal zeros, for example, on the number 406 ($4 \times 10^{2} + 0 + 6 \times 10^{\circ}$).

Regarding their neuroanatomic substrates, processing of Arabic numbers tend to activate the fusiform gyrus (Dehaene & Cohen, 1995), whereas the Verbal code is language-based and it is associated to left perisylvian activations, especially in the angular gyrus (Zamarian, Ischebeck & Delazer, 2009). Price & Ansari (2011) tested for the presence of a possible modality specific "Visual Number Form Area" in the fusiform gyrus (Dehaene & Cohen, 1995) investigating brain responses during the passive viewing of digits, letters and novel symbols. They found an increased activation for Arabic digits in the left central angular gyrus, but there was not any region that was specifically activated when digits were presented during short durations (50ms). Recently, Abboud, Maidenbaum, Dehaene & Amedi, (2015) investigated a sample of blind subjects using a novel visual-to-music sensory-substitution device (The EyeMusic). They have concluded that, independently of visual experience, there is greater activation in the right inferior temporal gyrus for numerical symbols compared to color and letters. The authors argue that previous studies that experienced difficulties in identifying this number-form area might have been affected by the interference of magnetic artifacts. The specificity of this area in the brain remains to be explored.

Since there is not a perfect matching between the syntax order of the Verbal and Arabic systems, converting from one notation to the other imposes certain difficulty. The next section describes relevant cognitive models that aim at describing how this process occurs.

Number transcoding models

The first systematic investigations on the convertion between these two symbolic systems were developed by the neuropsychologists Gérard Deloche and Xavier Seron (Deloche & Seron 1982a; Deloche & Seron 1982b; Seron & Deloche, 1983; Seron & Deloche, 1984). In four classical works, they have conducted a series of case studies on aphasic patients, on which they have described their performance on transcoding between the Verbal and Arabic codes. The authors found selective deficits and, therefore, argued in favor of a relative independence of the language and number domains.

Over the last years, different transcoding cognitive models were proposed, grounding their assumptions on a wide range of evidences, such as case studies in adult patients (McCloskey, Caramazza & Basili, 1985), cognitive development (Power & Dal Martello, 1990; Seron & Fayol, 1994), and computational simulations (Barrouillet, Camos, Perruchet, & Seron, 2004; Verguts & Fias, 2006). Basically, number transcoding models can be classified according to the role they attribute to semantic access while converting between different notations. "Semantic" models predict that the access to magnitude representation is a necessary step in transcoding operations while "asemantic" models predict that transcoding is independent of access to magnitude representations. It is important to note that some authors also propose simultaneously considering both semantic and asemantic transcoding routes, aiming at conciliating empirical findings (mainly from case studies) and the previous models (Cipolotti, 1995; Cipolotti &

Butterworth, 1995; Cohen, Dehaene, & Verstichel, 1994; van Loosbroek, Dirkx, Hulstijn, & Janssen, 2009). For our purposes, we will briefly describe a semantic model, derived from the broader classical model of numerical cognition proposed by McCloskey, Caramazza and Basili (1985), the Triple Code Model (Dehaene, 1992; Dehaene & Cohen, 1995), a relevant numerical cognition model, from which is also possible to presume number transcoding assumptions; and the ADAPT model (Barrouillet et al., 2004), an asemantic number transcoding model focused on the acquisition of transcoding rules.

The first important cognitive model of number transcoding was proposed by McCloskey, Caramazza & Basili (1985). According to this model, all numerical inputs are translated through the access to an amodal and abstract representation of quantity. This representation of quantity does not vary regardless of the type of numerical notation (whether the output is verbal or Arabic). Reading the number "5329", for instance, involves the translation into a thousand/hundreds/tens/units system, similar to $5 \times 10^3 + 3 \times 10^2 + 2 \times 10^1 + 9 \times 10^0$. After this process, the adequate lexical units are allocated in their respective places to formulate the adequate output. The central aspect of this model is the presence of an internal semantic representation that mediates all the possible paths between input, output and calculation mechanisms. Semantic models have been criticized because error analysis from case studies suggest that patients can present difficulties in retrieving lexical units and managing algorithmic rules (Deloche & Seron, 1982s, 1982 b; Seron & Deloche, 1983).

The Triple Code Model (Dehaene, 1992; Dehaene & Cohen, 1995) is a number processing model that postulates three numerical codes. The analogical representation would be related to the concept of "number sense": a pre-verbal system that imprecisely represents magnitudes and it is

innate. It allows for the comparison between different magnitudes and the grasping of the amount of objects in a certain set (Dehaene, 2009). Analogical number processing can be described by Weber's law (w) (Izard & Dehaene, 2008), according to which the variation that can be discriminated in the stimulus intensity is a constant fraction of the stimulus initial value. The Arabic and verbal codes, on the other hand, are culturally learned. The Arabic code (45) is the symbolic representation of magnitudes and it allows for exact arithmetic operation with multidigit numbers. At last, the verbal code (forty-five) represents the numerical information in the verbal notation. Contrary to the abstract semantic model of McCloskey, in the Triple code model there is no hierarchical organization between the different codes, they are relatively independent, which means that transcoding between verbal and Arabic codes does not imply obligatory access to numerical magnitude. This model is a representational proposal and do not describe how the transcoding between this different codes occurs.

Following a different premise, asemantic models admit that the conversion between the source code and the Arabic one is an algorithm-based procedure and do not require access to the magnitude the number represents. An important asemantic model is the ADAPT (Barrouilet et al., 2004), A Developmental, Asemantic and Procedural model for Transcoding from verbal to Arabic numerals. It accounts for the evolution of transcoding processes through practice: the experience would lead to an expansion of the numerical lexicon and improvement of conversion rules. The inputs, regardless of their numerical complexity, are codified into a phonological sequence and, then, the parsing mechanisms subdivide this sequence into smaller units to be processed by the production system (Figure 1). This production system is related to rules devoted to retrieval of information from long-term memory (LTM) (called P1 rules, responsible for retrieving "3" from its verbal form), to manage the size of digital chains (P2 and P3 rules; in

"2003", these rules create a frame of three slots) and to fill these slots (if there are any empty slots, P4 rules will fill them with 0s). Separators, such as thousands and hundreds, are used to identify the number of slots and once every segment is placed in its digital form in the chain, it is transcribed.

It is important to note that the ADAPT model can be described as a dual route model, as can be seen in Figure 1. The direct or lexical route would be associated with the writing of familiar numbers and consists of phonological encoding, working memory assisted search in long term memory, lexical retrieval, and graphic transcoding. On the other hand, the syntactic or algorithmic route would be responsible for the writing of multidigit-unfamiliar numbers by following the steps described previously.



Figure 1. Schematic representation of the ADAPT model (Barrouillet et al., 2004) adapted from Lochy & Censabella (2005)

Based on this model, one can derive two main sources of errors: lexical and syntactic. Lexical errors are due to the replacement of one lexical element by another, for instance, when "forty-five" is written as "46". Syntactic errors are related to misplacement of lexical elements in the syntactic frame; "forty-five" could be transcoded as "405". This last kind of error is frequently associated with a lack of knowledge of the place-value system and of the transcoding algorithms (Deloche & Seron, 1982a, 1982b)

The ADAPT model can be more easily operationalized in terms of cognitive-neuropsychological constructs, such as working memory, hence there is a growing body of neuropsychological literature based on this cognitive model (Camos, 2008; Zuber et al., 2009; Pixner et al., 2011; Moura et al; 2013).

Transcoding as a marker of math difficulties

Transcoding between verbal and Arabic notations imposes some difficulties in the sense that each of these numerical codes has specificities regarding their lexicon and syntax (Deloche & Seron, 1987). These difficulties are more salient in children in early schooling, who are not completely familiar with the Arabic notation yet, particularly with its place-value syntax (Geary, 2000; Camos, 2008; Moura et al., 2015). Children with math learning difficulties reliably present difficulties with the numerical symbolic codes and transcoding is one these problems (Geary, Hamson & Hoard, 2000). Previous studies by the Development Neuropsychology Laboratory of UFMG (Ferreira, Wood, Pinheiro-Chagas, Lonnemann, Krinzinger, Willmes & Haase, 2012; Moura, 2010) analyzed the performance of Brazilian typical children, with mathematics learning

disabilities (MLD), and mathematics and spelling disabilities (MSLD) in a numerical transcoding task. The task proved to be suitable for discriminating both clinical groups, as shown by the area under the ROC curve. Psychometric analysis indicated that the task was very simple, thus, new items were included that vary in length and number of rules of the ADAPT model (Moura et al, 2010). Nevertheless, we observed the lower performance of MSLD children in relation MLD children (Ferreira et al, 2012). Such difficulties exhibited by MSLD children in transcoding may be due to deficits in phonological processing, especially phonological awareness deficits, which constitute a major mechanism responsible for learning difficulties in reading and writing, which will be discussed in further detail in the next section. In these studies, it was also observed that difficulties in number writing are usually compensated by the 4th grade, which suggests that there could be a maturational lag in children with MLD. Nevertheless, it is important to note that MLD children usually continue to commit lexical errors, and this is not as frequently observed in typical children, who tend to present errors associated with the syntactic route, that also disappear by the end of primary school.

The syntactic errors presented by children can be correlated to the numerical complexity of the number, as defined by the ADAPT model. According to Camos (2008), number of 1 and 2 digits are usually lexically retrieved and do not impose a great source of difficulty to children whereas numbers that are transcoded via the syntactic route are more prone to errors. Barrouillet et al. (2004), Camos (2008), Moura et al (2013) emphasize how zeros as place-holders are especially difficult. Based on what the ADAPT model proposes, filling the blank spaces with zeros would be the last step on the Verbal to Arabic number transcoding and it is more likely to be wrongly written. A common zero-related error reported by Zuber et al (2009) and Moura et al (2013) are syntactic errors named additive and multiplicative composition. On the additive composition

errors, the child adds the zeros that correspond to the failure of following the principle of additive composition (four hundred and 7 as 4007). On the multiplicative composition errors, the child would add the one and the zero that indicate the incorrect application of the multiplicative composition principle (four hundred and seven as 41007). Children with math difficulties usually tend to present a higher frequency of these errors and continue to commit them later on development (Moura et al, 2013).

Even though transcoding difficulties are a relevant impairment presented by children with mathematical difficulties, most of the available tests in Brazil to assess math skills focus solely on arithmetic. The only standardized test we have for school achievement is the Teste de Desempenho Escolar, TDE (Stein, 1994). The TDE comprises three subtests tapping basic educational skills: single-word reading (which was not used in the present study), single-word spelling to dictation and basic arithmetic operations. The arithmetic subtest is composed of three simple oral word problems that require written responses (e.g. "John had nine stickers. He lost three. How many stickers does he have now?") and 45 basic arithmetic calculations of increasing complexity that are presented and answered in the writing format (e.g. "1+1=?" and "(-4) x (-8)=?"). The only item that investigates basic number processing skills is the first oral one, on which the child needs to decide which is larger, 42 or 28. As we have described previously, children with mathematical disabilities present basic number processing deficits that can affect formal arithmetic learning. In clinical and research contexts, it can be very informative to have a tool for evaluating number transcoding skills of children with mathematical difficulties, with reliable psychometric parameters.

Cognitive mechanisms associated with number transcoding

The nature of magnitude representations

One of the problems with the semantic model proposed by McCloskey et al. (1985) regards the nature of the abstract semantic representations of numerical magnitude. McCloskey and coworkers assume that numerical magnitude is represented by a place-value positional code. This seems intuitively implausible. An alternative is the analogic and approximate model of representing numerical magnitude on a approximate number system proposed by Dehaene (1992, 2011, Dehaene & Cohen, 1995).

The approximate number system (ANS) is a basic system that processes numerosity, responsible to directing attention towards the spontaneous extraction of an approximate number of objects in sets and it is present in several species (Piazza, 2010). Its functioning can be described according to the Weber Law, in other words, humans represent numbers in an approximate and compressed manner, in a way that two sets can only be discriminated if they differ by a certain numerical ratio. Fechner's law is another relevant psychophysical postulate to this model, according to which external numerosities are internally scaled into a logarithmic internal representation of sensation. Some authors argue that ANS development is a precondition for the acquisition of meaning comprehension from symbolic numbers because it interacts with symbolic numerical representation in a way that allows for performance of exact arithmetic (Dehaene & Changeux, 1993; Piazza, 2010). The accuracy of ANS is associated with mathematics achievement in kindergarten (Libertus, Feingenson & Halberda, 2011) as well as in elementary school (Halberda, Mazzoco & Feigenson, 2008; Inglis, Attridge, Bachelor & Gilmore, 2011; Pinheiro-Chagas, Wood, Knops, Krinzinger, Lonnemann, Starling-Alves & Haase, 2014). Impairments in the ANS are described as the core deficit in children with mathematical learning disabilities, according to the defective number module hypothesis (Butterworth, 2005; Wilson & Dehaene, 2007). It can be

thought as the domain-specific mechanism that underlies arithmetic learning. Impairments in the ANS would lead to difficulties in learning the symbolic system and, consequently, problems with arithmetics (Dehaene, 2009; Piazza, 2010; Piazza et al., 2010). We have found evidence suggesting that deficits in the ANS can be quite stable in the context of mathematical learning disabilities (Julio-Costa, Starling-Alves, Lopes-Silva, Wood & Haase, 2015; see appendix 1) Even though the ANS is important for symbolic aspects of mathematics, it does not seem to play a crucial role on number transcoding. The asemantic cognitive transcoding models (Barrouillet et al., 2004) claim that changing from one numerical notation to the other does not require access to the numerical magnitude, especially in the case of more familiar and large numbers. Furthermore, other cognitive variables have been studied as possible influences on number transcoding and will be described below.

Working memory

Working memory (WM) plays an important role in number transcoding according to the ADAPT model (Camos, 2008; Zuber et al., 2009). It is involved in the temporal storage of verbal information, lexical retrieval, and execution of the manipulations needed to generate the Arabic output. Besides that, algorithms that involve maintaining a large number of conversion rules in memory are subject to a greater number of errors (Camos, 2008). Fayol, Barrouillet & Renaud (1996) demonstrated that the number of syllables of the dictated number could account for 33% of the variance in a number writing task and they argue that this result could be associated with the limited capacity of working memory.

In number writing tasks, children with higher span in phonological working memory presented superior performance compared to those with lower span and WM correlated with the complexity

of the items to be transcoded (Camos, 2008). Zuber et al (2009) assessed 7-year-old Austrian children regarding the association of specific WM componnets and types of errors in number writing. They have concluded that inversion errors [einhundertzweiundachtzig (one hundred and eighty-two) written as 128] seemed to be associated with the central executive, whereas other syntactic errors were associated with visuospatial storage component. Pixner et al (2011) also investigated the performance of German, Italian (whose numerical system is not inverted) and Czech (which has two number systems both with and without decade-unit inversion) children. They reported that overall error rate in number transcoding was reliably predicted by their central executive index. From these studies, it could be observed that children who speak languages that have the inversion rule commit more errors, and that these errors have a negative correlation with working memory. In languages with inversion, the visuospatial working memory plays an important role on number writing, whereas the differential impact of verbal and visuospatial working memory in other numerical systems is still inconsistent (Moura et al., 2013).

The role of WM in transcoding tasks can be thus systematized in the following steps: encoding and storing the number to be transcoded; retrieving and monitoring the application of transcoding rules, and the graphic production of the numeral (Lochy & Censabella, 2005). Nevertheless, it is important to note that many of these studies described above did not control other phonological processing skills that might also demand working memory and could be confounding factors. As the ADAPT model suggests that the first step of Verbal to Arabic number transcoding is the phonological encoding of the number, more general phonological processing skills could also be possible relevant cognitive influence.

Phonological processing

Transcoding from the Verbal to the Arabic code requires the use of verbal/phonological codes, and this skill might be especially prone to dysfunction in children with developmental dyslexia or comorbid developmental dyslexia plus dyscalculia. In addition to the access to magnitude representation and working memory, phonological processing is one of the cognitive mechanisms that has been extensively studied regarding arithmetic skills. Part of this interest can be attributed to the high rate of comorbidity between dyscalculia and dyslexia (Dirks et al, 2008; Landerl & Moll, 2010).

Deficits in phonological processing are frequently found in children with pure dyslexia or in the comorbidity with dyscalculia (Vellutino et al, 2004; Simmons & Singleton, 2008). Phonological processing is traditionally associated with reading and writing acquisition. According to Wagner & Torgesen (1987), at least three phonological skills are involved in word reading: a) phonological awareness, which means the ability to perceive and manipulate phonemes that constitute words; b) phonological working memory, involved in temporary retaining of sound based representations; and c) lexical access, which is related to the retrieval of a written word from its lexical referent through the recoding into a sound-based representational system. In the last years, studies have also focused on the influence of phonological processing in mathematical achievement (Landerl, Bevan, & Butterworth, 2004; Simmons & Singleton, 2008; Landerl, Fussenegger, Moll, & Willburger, 2009) and it is hypothesized that phonological processing may be more strongly connected to the aspects of mathematics that involve verbal codes as well as Arabic number representations, such as arithmetic facts and number transcoding.

Some authors claim that the influence of phonological processing in arithmetic could be due to shared demands with working memory, since these effects can be eliminated when partialling out the influence of working memory (Swanson & Sachse-Lee, 2001; Swanson, 2004). It is important to note that the relationship between working memory and phonological processing, as well as their impact on reading and math tasks could vary according to the complexity of the instruments used (Cunningham, Witton, Talcott, Burgess & Shapiro, 2015). The impact of phonological WM, for example, might be overestimated due to influences of phonological processing that have not been controlled for in these studies. Phonological processing may play a role in the initial encoding of the verbal input.

In the Arabic number writing task, the input is verbal, hence the child must be able to differentiate between the speech sounds to correctly comprehend the verbal form that shall be transcoded into the Arabic form. Nevertheless, most studies on children with reading difficulties, which present phonological processing deficits, have focused on their general arithmetic performance, and not in more basic number processing. It is still not clear if number transcoding would be one of the verbal aspects of math that could be consistently impaired in these children.

Recently, Moll, Göbel, & Snowling (2014) investigated the neuropsychological profile of 6 to 12-year-old children with difficulties only in mathematics (MD), in both mathematics and reading (MD+RD), only in reading (RD) and typically developing (TD). The RD group did not present deficits in the nonsymbolic aspects of mathematics, yet it was particularly low on verbal tasks. Regarding number transcoding, the effects of MD and RD were comparable. This finding suggests that children with reading difficulties, who presumably present deficits in phonological processing, struggle in numerical tasks that involve the verbal code, regardless of their unimpaired approximate number system.

Rosselli, Matute, Pinto & Ardila (2006) investigated children with different subtypes of dyscalculia, including children with both dyscalculia and reading difficulties. The groups did not differ on their number writing task. One possible reason for this lack of statistical difference is that their transcoding task had only 8 items, and 2 of them could be lexically retrieved (numbers 1 and 7), so it was probably too easy for 11 year-old children. Simmons & Singleton (2009) investigated the mathematical profile of children with dyslexia and concluded that they were slower in number fact recall but did not have difficulties in place value understanding. According to the authors, place value requires children to learn rules which link the position of digits with values and it is less reliant on phonological processing. Children had to compare bags that contained a different amount of pounds, in several experimental situations, for example, on which the important distinguishing feature was the first digit (£72, £79, £81) or when the larger number islarger than a smaller number even if the first digit is smaller (e.g. £888, £999, £1002). The main problem of this result is that the place value task was mainly a symbolic magnitude comparison task and, thus, it was not a measure of number transcoding per se. More studies still need to specifically address the association between number transcoding and phonological processing.

Few studies have also investigated common brain regions for both arithmetic and phonological processing, but mostly in adults. The left temporoparietal cortex supports both types of processing (Simon, Mangin, Cohen, Le Bihan & Dehaene, 2002), but there might be a regional differentiation. Andin, Fransson, Rönnberg & Rudner (2015) report that multiplication is associated with a posterior portion of the angular gyrus wheres phonological processing recruited its anterior portion.

Phonological processing probably plays an important role in numerical processing and it is a

possible link between learning to read and write both words and numbers.

Reading and writing of words and numbers

The research regarding shared cognitive mechanisms between reading/spelling and math as well as the profile of children with pure cases of dyscalculia/dyslexia and comorbid cases has grown in order to clarify the nature of the connection between these two scholastic skills. Regarding the cognitive deficits that underlie both of these disabilities, there is consistent evidence that a deficit in phonological processing could be considered the proximal cause of reading difficulties in dyslexia (Vellutino et al 2004). When it comes to mathematical learning disabilities, the most relevant domain-specific deficit seems to be in the processing of nonsymbolic numerosities (Piazza et al., 2010). The high comorbidity rate between dyscalculia and dyslexia can be explained by the double deficit hypothesis (Landerl, Bevan & Butterworth, 2004), according to which children with both learning disabilities present simultaneous deficits in phonological processing and in the approximate number system. In contrast, Simmons & Singleton (2008) suggest the common deficit account to describe the cognitive impairments associated with both reading and math. According to these authors, math difficulties could also be caused by the phonological deficits commonly associated with dyslexia. It is assumed that the phonological representations of dyslexic children are weak, which leads to an impairment in cognitive processes that demands phonological codes. They propose the weak phonological representation hypothesis: according to it, the poorly specified nature of phonological representations could lead to poor performance on tasks that involve the retention, retrieval or manipulation of phonological codes. Because number writing requires the access to the verbal codes that represent numbers, it is logical to assume that children's phonological processing abilities will also influence their transcoding attainment. In appendix 2, we describe a case study of a child with phonological processing deficits that impact number reading and writing skills, in spite of his intact approximate number system.

Moll et al (2014) propose an additional explanation by stating that mathematics has both verbal and nonverbal components and poor performance in mathematics could be the behavioral consequence of different patterns of number processing deficits. The authors assessed a sample of 89 children ranging from 6 to 12 years old to investigate if the same behavioral outcome (such as arithmetic difficulties) could be explained by different underlying cognitive deficits. The authors concluded that children with both reading and math difficulties presented an additive profile of impairments. It is important to note that studies on RD, MD and comorbid groups frequently use different diagnostic criteria to classify children as having an impaired performance, which could lead to different impairment profiles (see Landerl et al, 2004). Jordan (2007) suggests that reading deficits aggravate, and not properly cause, math difficulties, since children with both dyslexia/dyscalculia would struggle in using language compensatory mechanisms.

Another sampling strategy to investigate the importance of reading to numerical cognition is the study of illiterate subjects. Illiteracy can be defined as the lack of word and number reading that affects the execution of simple daily living activities (Morais, Leite & Kolinsky, 2013). Research has already been carried out investigate how formal schooling and literacy influences several cognitive aspects (for review see Ardila et.al. 2010). Less attention, however, has been directed to the influence of formal education in numerical abilities. Parsons & Bynner (2005) reviewed survey data for the British National Research and Development Centre for Adult Literacy and Numeracy. They found that literacy and numeracy are relatively independent. The authors have
observed that numeracy deficits affect financial outcome and employability even when literacy is controlled for.

Nys, Ventura, Fernandes, Querido, Leybaert & Content (2013) investigated the performance of illiterates in mental calculation, transcoding and counting principles. Transcoding abilities were accessed from both Arabic to verbal and from verbal to Arabic directions. Illiterates were worse than controls in both input types. They have concluded that, in general, whenever the input was verbal, the performance from the illiterates was worse.

Martins (2016) has investigated the performance of Brazilian semi-illiterates on a number writing task, with 81 items varying in syntactic complexity and number of digits. The semi-illiterates also presented a syntactic complexity effect and the less educated were the participants, the higher the error rate in number transcoding. It is interesting to note that performance of semi-illiterate adults of normal intelligence in this task is similar to that of 2nd grade children and that the frequency of lexical errors decreases as literacy level increases. The author also assessed two other samples of ex-illiterates, who have already completed one or two years of formal schooling. These two groups did not differ between each other, suggesting that completing the first year of formal schooling already improves their transcoding skills. This finding is similar to what is reported in children, on which transcoding performance tend to improve between first and second grade, but reach a ceiling effect on the third grade (Noël & Turconi, 1999). Nevertheless, it is important to note that phonemic awareness was not accessed, because the sample could not understand the instruction of a phoneme elision task.

Evidently, illiteracy is a consequence of several factors and these results cannot be attributed

exclusively to phonological processing deficits. Nevertheless, the difficulties presented by semiilliterates in number writing is another important evidence of the association between language and transcoding skills, beyond intelligence.

From the studies described above, one can conclude that regarding number transcoding, especially Arabic number writing, the input is verbal; hence it is crucial that the child is able to differentiate and manipulate the speech sounds to correctly comprehend and transcode a number. Despite this possible impact of phonological processing on this basic number processing skill, to the best of our knowledge, there has not been studies so far that systematically investigated the association between reading and number transcoding.

The present dissertation: investigation of linguistic influences on number transcoding in children

While there is growing evidence for a relationship between phonological processing and numerical cognition, an important limitation of the current literature is that there are no studies that specifically address its impact on number transcoding. In the field of numerical cognition, phonological processing has been mostly and consistently related to arithmetical facts (Simmons, Singleton & Horne, 2008; Simmons & Singleton, 2008). Phonological processing deficits should lead to poor phonological representations and thus also impact negatively transcoding. More specifically, we set out to investigate possible associations of phonemic awareness and number transcoding. Phoneme elision requires a certain amount of working memory in the sense that the participant must hold a word in mind while determining the phonological information to be deleted (DeSmedt et al., 2010). Hence, it is important to partial out the influence of phonological working memory to examine the possibility that the association of phonemic awareness and

mathematics is due to shared demands with working memory. Additionally, there is also evidence that semantic manipulation of numerical magnitude is related to arithmetic proficiency (Halberda et al., 2008; Piazza et al., 2004, 2010; Costa et al. 2011), thus we aimed to control for its influence through a measure of the approximate number system acuity.

The three studies in this dissertation address the relevance of studies on the association of number transcoding and mathematical disabilities, as well as the underlying cognitive mechanisms of number transcoding. In the following section, we introduce our goals and the results of each study in turn.

STUDY 1: From 'Five' to 5 for 5 Minutes: Arabic Number Transcoding as a Short, Specific, and Sensitive Screening Tool for Mathematics Learning Difficulties

Specific aim: To determine reference values and psychometric properties of a Verbal to Arabic transcoding task in Brazilian school-aged children

Hypotheses: Number writing would have adequate diagnostic accuracy in the detection of children at risk for mathematical difficulties in primary school (Moeller, Pixner, Zuber, Kaufmann & Nuerk, 2011; Moura et al., 2013). Nevertheless, older children will have already probably overcome difficulties in transcoding (Ferreira et al., 2012). Besides that, creating a task with items with different level of difficulty, defined according to ADAPT model (Barrouillet et al, 2004), would lead to adequate psychometric indexes.

How we investigated it: We have conducted a psychometric study on a number dictation task, on which numbers were orally presented and the child should write the corresponding

Arabic form. This task has been previously developed in the context of a broader project on mathematical learning disabilities entitled "Endophenotyes of math learning difficulties", previously approved Ethics Research Board by the (COEP-UFMG, CAAE: 15070013.1.0000.5149). In this study, we have assessed 985 children and classified them into mathematic difficulties groups (MD) when they presented performance below the 25th percentile in the arithmetic subtest and above the 25th percentile in the spelling subtest of the TDE. Control children presented performance above the 25th percentile on both subtests. After excluding children with insufficient performance in the spelling subtest only, as well as outliers and missing cases, the final sample was composed by 683 control and 103 MD children, from 1st to 4th grades. We have assessed their number transcoding skills, using a 28-item number writing task, with numbers ranging from one- to four-digits and different level of syntactic complexity, as well as their general school achievement, with the spelling and mathematic subtest of the TDE.

Results: The number writing task presented a high internal consistency and numbers with higher syntactic complexity were more discriminative for testing purposes. The task was accurate in discriminating children with mathematics difficulties in the 1st and 2nd grades but was not as efficient in the 3rd and 4th grades. There was a significant correlation between the number of errors and the number of transcoding rules based on the ADAPT model. We also reported normative parameters such as mean, range values, and percentiles for first to fourth grades

Rationale of Study 2: Study 1 has concluded that number transcoding is a sensitive tool for discriminating children with mathematical disabilities. But what are the cognitive skills that underlie number writing performance? Study 2 pursues this question, by means of a path analysis model.

STUDY 2: Phonemic awareness as a pathway to number transcoding

Specific aim: To investigate the role of specific cognitive mechanisms underlying number transcoding, namely, general cognitive ability, verbal and non-verbal short-term and working memory, magnitude representation, and phonemic awareness. More specifically, our main goal was to investigate the relative impact of phonemic awareness on number transcoding.

Hypotheses: Working memory would contribute to number transcoding, since it is implicated in the use of transcoding algorithms (Barrouillet et al., 2004; Camos, 2008). Furthermore, verbal working memory would play a more important role, because the association of visuospatial working memory to number transcoding might be stronger in languages with decade-unit inversion (Zuber et al., 2009; Moura et al., 2013). This possible influence of verbal working memory in number writing could be mediated by phonemic awareness, since it is assumed to be an index of the quality of phonological representations. Finally, we expect that magnitude representation would not be associated with number writing if number transcoding is an asematic process.

How we investigated it: We have assessed general intelligence, verbal and visuospatial working memory, nonsymbolic magnitude comparison, phonemic awareness and Verbal to Arabic number transcoding in a sample of 172 children from 2nd to 4th grade. At first, we have looked into the general associations between these measures through Pearson's correlations. Following this, we investigated the specific impact of phonemic awareness in number writing, by means of a stepwise hierarchical regression model, with age and intelligence entered first, working memory and the Weber fraction in a second step and phoneme elision in the third.

Afterwards, we calculated path analyses including all of the previous measures to determine possible mediation effects.

Results: One third of the sample did not commit any error in the transcoding task and we arcsine transformed the error rates to approximate a normal distribution. The regression model showed that verbal working was a significant predictor of transcoding after the removal of age and intelligence effects. However, the addition of phonemic awareness in the third step lead to the exclusion of verbal working memory. Regarding the path models, when phonemic awareness was not included as a mediator between the influence of verbal working memory and number writing, the fit indexes were not acceptable. The model in which the effect of working memory was partially mediated by phonemic awareness was the most parsimonious description from the data.

Rationale of Study 3: Study 2 provides evidence that phonemic awareness is associated with number transcoding. This phonological skill has been consistently associated with reading performance (Vellutino et al., 2004). To extend this result, Study 3 investigates possible shared associations between phonemic awareness with number and word writing and reading skills.

STUDY 3: What is specific and what is shared between numbers and words?

Specific aim: To investigate shared and non-shared mechanisms involved in reading and writing of both words and Arabic numerals in school-aged children. Our main goal was to disentangle the role of phonemic awareness and its impact on lexical numerical and verbal tasks controlling for cognitive variables which may have an important impact, such as working

memory.

Hypotheses: According to the weak phonological representation hypothesis (Simmons & Singleton, 2008), phonemic awareness should impact both number writing and number reading, since these aspects of mathematical cognition involve the manipulation of verbal codes. Moreover, we hypothesize that phonemic awareness would be associated with reading and spelling skills even after controlling for the influence of intelligence and more broader reading and writing skills.

How we investigated it: Using a sample of 172 children from 2nd to 4th grade, we conducted a series of hierarchical regression models with scores of reading and writing of single words and Arabic numbers as dependent variables. As predictor variables, we investigated intelligence, phonological and visuospatial WM and phonemic awareness. In order to control for the shared variance among numerical and verbal tasks, whenever one of these tasks was set as the dependent variable, the other was inserted in the first step of the regression model, using the enter method (i.e., when number reading task was the dependent variable, word reading was included as an independent variable in the first step of the model). Intelligence and the analogous variable were included in the first step and the other cognitive variables in the second one. Raw scores were z-transformed by grade to control for any possible educational influence.

Results: Performance on each kind of number task (reading or writing) was predicted by the homologous verbal tasks (reading or spelling) and vice versa and by phonemic awareness, over and above intelligence. Phonological working memory was also significantly associated with word reading, but to a smaller extent compared to the influence of phonemic awareness.

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On the next three chapters, we will present these studies in deeper detail and, afterwards, there will be a discussion chapter, summarizing our main findings, describing our limitations and suggestions for futures studies, as well as the main clinical and research implications.

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CHAPTER 2

Study 1

STUDY 1:

From five to 5 for 5 minutes: Arabic number transcoding as a short, specific, and sensitive screening tool for mathematics learning difficulties

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ABSTRACT

Number transcoding (e.g., writing 29 when hearing "twenty-nine") is one of the most basic numerical abilities required in daily life and is paramount for mathematics achievement. The aim of this study is to investigate psychometric properties of an Arabic number-writing task and its capacity to identify children with mathematics difficulties. We assessed 786 children (55% girls) from first to fourth grades, who were classified as children with mathematics difficulties (n = 103) or controls (n = 683). Although error rates were low, the task presented adequate internal consistency (0.91). Analyses revealed effective diagnostic accuracy in first and second school grades (specificity equals to 0.67 and 0.76 respectively, and sensitivity equals to 0.70 and 0.88 respectively). Moreover, items tapping the understanding of place-value syntax were the most sensitive to mathematics achievement. Overall, we propose that number transcoding is a useful tool for the assessment of mathematics abilities in early elementary school

Keywords: Number transcoding, mathematics difficulties, screening.

From "five" to 5 for 5 minutes: Arabic number transcoding as a short, specific, and sensitive screening tool for mathematics learning difficulties

Daily activities require the communication of numerical information, such as registering a telephone number or making mental calculations. Besides that, being able to manipulate numbers is one of the first steps in mathematical learning, which begins to be formally trained in kindergarten. Learning the Arabic notation is one of the main challenges faced by young children in the first years of school, especially because of its place-value syntax (Geary, 2000). A useful tool for investigating children's knowledge of numerical syntax is the number transcoding task. This task requires the conversion of numerical symbols between verbal and Arabic numerical notations (Deloche & Seron, 1987).

The verbal number system is composed by a lexicon that designates some numbers (e.g. *five*, *eleven*), the bases by which they are multiplied (e.g., "ty" in *seventy*; *hundred*), as well as by a syntax that organizes these lexical units to represent any possible quantity. In turn, the Arabic number system possesses a lexicon of only ten elements. Its basic syntactic principle is the place-value, according to which the actual value of a digit is given by its position in the number.

The ADAPT model (A Developmental, Asemantic and Procedural Transcoding) by Barrouillet, Camos, Perruchet, and Seron (2004) accounts for this conversion from the verbal-oral to the Arabic form by means of representing information in phonological short-term memory, and by the lexical retrieval and rule application, which are driven by condition-action rules. When the lexical units in the verbal input match an Arabic form stored in long-term memory (e.g. $one \rightarrow 1$, *fifteen* \rightarrow 15), then the output is directly retrieved. Otherwise, specific rules are triggered, and operate recursively in the verbal string present in the input in order to build the correct output in the Arabic notation. The conditions that trigger a given rule can be either the class of the lexical primitives (unit, decade, hundreds, for example) or the presence of empty slots. There are eight different procedures triggered by the rules, such as "finding the positional value of the lexical primitive" (how many slots the frame must have), "filling empty slot with 0", among others. These rules are devoted to (i) the retrieval of information from long-term memory (LTM) (called P1 rules, responsible for retrieving "3" from its verbal form), (ii) to manage the size of digital chains (P2 and P3 rules; in "2003", these rules create a frame of four slots) and (iii) to fill these slots (if there are any empty slots, P4 rules will fill them with 0s).

Concerning the development of number transcoding in children, evidence suggests that the acquisition of the numerical lexicon (Wynn, 1992) and basic principles of numerical syntax (Barrouillet, Thevenot, & Fayol, 2010) are already acquired even before elementary school. During the first school years, the development of number transcoding skills is highly influenced by numerical length (quantity of digits) and syntactic complexity (quantity of transcoding rules). By the beginning of the second grade children already master the writing and reading of 2-digit numbers, showing major difficulties in the transcoding of 3- and 4-digit numbers (Camos, 2008; Moura et al., 2013; Power & Dal Martello, 1990, 1997; Seron, Deloche, & Noël, 1992). Most of these difficulties in number transcoding are scarce, and concentrated in 3- and 4-digit numbers with a more complex syntactic structure, such as the ones containing internal zeros (Moura et al., 2013; Sullivan, Macaruso, & Sokol, 1996). Therefore, numerical transcoding abilities for numbers up to four digits appear to be fully achieved in typically developing children after three years of formal education (Nöel & Turconi, 1999).

Only few studies have investigated the association between number transcoding and arithmetic achievement in school children. Examining first graders, Geary, Hoard, and Hamson (1999) and Geary, Hamson, and Hoard (2000) found a significant association between reading and writing of small numbers and formal mathematics achievement. Using a longitudinal approach, Moeller, Pixner, Zuber, Kaufmann, and Nuerk (2011) showed that, compared to working memory capacity and non-symbolic representations of numbers, the knowledge of place-value syntax in the end of first grade is the best predictor of mathematics achievement two years later. Furthermore, syntactic errors in an Arabic number writing task and the decade-unit compatibility effect in a two-digit number comparison task (Nuerk, Weger, & Willmes, 2001) have proved to be particularly important to characterize and predict mathematics achievement in children (Moeller et al., 2011).

Difficulties in number transcoding have also been observed in children with developmental dyscalculia or mathematics learning difficulties. Studies suggests that writing and reading Arabic numbers impose relevant obstacles to younger children with mathematics learning difficulties aging around 7-years-old (Geary, Hoard, & Hamson, 1999; Geary, Hamson, & Hoard, 2000). In turn, in older children (8- and 9-years-old) these difficulties in number transcoding seem to be already overcome (Landerl, Bevan, & Butterworth, 2004), or restrict to planning times (van Loosbroek, Dirkx, Hulstijn, & Janssen, 2009). This issue was investigated in deeper detail by Moura et al. (2013), using more complex transcoding tasks containing numbers with up to 4 digits, and with increasing syntactic complexity. Results revealed significant differences between children with mathematics difficulties and typical achievers, from the first to the fourth grades, in both Arabic number reading and writing, but with effect-sizes decreasing with grade. Importantly, in middle elementary grades, children with mathematics difficulties showed higher

error rates in numbers with higher syntactic complexity. Moreover, an analysis of the erroneous responses suggested that, in early elementary school, children with mathematics difficulties struggle with both place-value syntax of Arabic numbers and with the acquisition of a numerical lexicon. In middle elementary school, the difficulties observed in children with mathematics difficulties were specific to the syntactic composition of Arabic numbers. The authors thus argued that, after the first school grades, children with mathematics difficulties are able to compensate at least part of their number transcoding deficits.

In summary, the literature on number processing and mathematics difficulties indicates that transcoding tasks are sensitive to and have a good predictive validity for mathematics difficulties (Moeller et al., 2011; Moura et al., 2013). Moreover, its cognitive underpinnings have been well characterized by current information processing models (Barrouillet et al., 2004; Camos, 2008; Cipolotti & Butterworth, 1995). Nevertheless, the diagnostic properties of number transcoding remain largely unexplored. In view of the above, one may consider the usefulness of number transcoding tasks in the screening of mathematics difficulties.

To our knowledge, there is no standardized task for assessing number transcoding abilities in school children. Even though number transcoding tasks are largely used in the investigation of numerical abilities in children (Geary, Hoard, & Hamson, 1999; Geary, Hamson, & Hoard, 2000; Landerl, Bevan, & Butterworth, 2004; Moura et al., 2013) and adults suffering from neurological impairments (Deloche & Seron, 1982a, 1982b; Seron & Deloche 1983, 1984), there are no reports on reliability, validity and item properties of such tasks. In general, studies using number transcoding tasks are conducted in the context of pure experimental neuropsychology, in which psychometric properties are presumed and never explicitly investigated.

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The aim of this study is to determine reference values and psychometric properties of a verbal to Arabic transcoding task in Brazilian school-aged children. In the present study, we assessed number transcoding by means of a number dictation task, in which numbers are orally presented and the child should write them in their Arabic form. The task was previously designed in the context of a wider investigation of mathematical abilities in children (Haase, Júlio-Costa, Lopes-Silva, Starling-Alves, Antunes, Pinheiro-Chagas, & Wood, 2014; Lopes-Silva, Moura, Júlio-Costa, Haase, & Wood, 2014; Moura et al, 2013). We reported normative parameters such as mean, range values and percentiles for 1st to 4th grades obtained from a large sample of school children. In addition, the diagnostic accuracy of the number writing task in the detection of children at risk for mathematical difficulties, as well as the influence of place-value syntax in children's achievement, were investigated.

Method

Participants

The sample was constituted by children attending to first to fourth grades in both public and private schools in the Brazilian cities of Belo Horizonte and Mariana. Data collection took place in 10 schools in Belo Horizonte (7 public), and 2 schools in Mariana (1 public). In Brazil, public schools are mostly attended to by children of lower to middle socioeconomic status. All study procedures were approved by the local university ethics committee.

In total, 985 children (85% from public schools) were assessed using the following three tasks: Arithmetics and Single-word spelling subtests of the Brazilian School Achievement Test (Teste do Desempenho Escolar, TDE, Stein, 1994), and the Arabic Number-Writing Task. Testing was conducted in classrooms of 10 to 20 pupils. Children with mathematics difficulties were those with performance below the 25th percentile in the Arithmetics subtest and the performance above the 25th percentile in the spelling subtest. Children with performance above the 25th percentile in both TDE subtests were classified as controls.

Instruments

Number transcoding task

Arabic Number-Writing Task: Children were instructed to write down the Arabic numerals that corresponded to the dictated numbers (one-hundred-and-fifty \rightarrow "150"). The task was composed by 28 items with 1- to 4-digit numbers. The use of three- and four-digit numbers intended to avoid numbers with strong lexical entries. The three- and four-digit numbers were grouped into three categories according to their complexity level (low, moderate and high complexity numbers), which were defined based exclusively on the number of algorithmic transcoding rules necessary to transcode each individual item. This criterion was based upon the ADAPT model, which relates item complexity to the number of algorithmic rules necessary to transcode a number (Barrouillet et al., 2004): the more transcoding steps must be performed, the more difficult is an individual item. The administration of the Arabic Number-writing Task lasted for about five minutes in individual assessments, while in collective assessments this duration increased to about 10 to 15 minutes. One point was assigned to each correct written number. There was no interruption criteria and no time limits, and one point was attributed to each correct answer.

General school achievement

School Achievement Test: The Teste de Desempenho Escolar (TDE; Stein, 1994) is the most

widely used standardized test of school achievement in Brazil. The TDE comprises three subtests tapping basic educational skills: single-word reading (which was not used in the present study), single-word spelling to dictation and basic arithmetic operations. The word spelling subtest consists of 34 dictated words with increasing complexity. The examiner dictated a word and afterwards a sentence containing this word, and finally repeated the word once more. One point was assigned to each correctly written word. The arithmetic subtest is composed of 3 simple oral word problems that require written responses (e.g. "John had nine stickers. He lost three. How many stickers does he have now?") and 45 basic arithmetic calculations of increasing complexity that are presented and answered in writing (e.g. "1+1=?" and "(-4) x (-8)=?". One point was assigned to each correct calculation. Reliability coefficients (Cronbach's α) are around 0.8 or higher. Children are instructed to work as much as they can, without time limits.

Procedures

Testing was carried-out in group sessions in children's own schools, in a separate silent classroom. The authors administered the task along with psychology undergraduate students. Sessions took approximately one hour, starting with Arabic number writing, followed by single word spelling and arithmetics subtests of TDE. In general, children understood the tasks' instruction and could follow them adequately.

Statistical analyses

Descriptive, reliability, internal consistency and item analyses were carried out using R (R Core Team, 2013). Internal consistency was calculated using the KR-20 formula, as items were coded as dichotomous variables. ROC analyses, Mixed-ANOVA models and Pearson's correlation analyses were carried out using SPSS version 20.0.

The ANOVAs included error rates in the Arabic Number-Writing task in the three levels of syntactic complexity (low, moderate or high) as within-subjects factor, and mathematical ability (control or children with mathematics difficulties) as between-subjects factor. Whenever the assumption of sphericity was violated, the Greenhouse–Geisser correction was applied to the estimation of statistics. Finally, to approximate a normal distribution, error rates were arcsine-transformed.

To interpret the ROC analyses, we considered the criteria established by Swets (1988), according to which AUC scores above 0.7 indicate acceptable (moderate) levels of diagnostic accuracy. Moreover, interpretations were made considering the lower limits of a 95% confidence interval in order to ensure the reliability of our estimates. For the cut-off scores, we sought for the best balance between specificity and sensitivity values.

Results

Children with insufficient performance (< 25th percentile) in the spelling subtest only (n = 189) were not included in this study. Outliers (defined as 1.5 times the interquartile range below the first quartile, in each grade), and children with missing values (n = 10) in the Arabic Number-Writing Task were also excluded from the analysis.

The final sample was then composed by 786 children (55% girls), with a mean age of 9y5m (sd = 1y1m), ranging from 6 to 12 years. There were 55 children in the first grade, 249 in the second grade, 225 in the third, and 257 in the fourth grade (see Table 1 for sample sizes displayed

separately for school grade and group). The control group (n = 683) and children with mathematics difficulties (n = 103) did not differ regarding age (t[784] = -0.59, p = 0.55).

Table 1

Mean scores, internal consistency and percentile ranks in Arabic Number-writing Task according to school grade and group.

Mean Number transcoding scores												
	1st grade			2nd grade			3rd grade			4th grade		
Control children	16.80 (3.22)			24.63 (3.11)			27.60 (.67)			27.85 (.36)		
Children with MD	12.80 (3.33)			15.92 (3.12)			26.50 (1.66)			27.34 (1.15)		
Reliability	.79			.88			.46			.33		
	Overall	Control	MD	Overall	Control	MD	Overall	Control	MD	Overall	Control	MD
Sample size	55	45	10	249	224	25	225	189	36	257	225	32
Total scores	Cummulative percentiles											
8	2		10									
9	4		20									
10	5		30									
11	7		40									
12	15	9	50	1								
13	24	18	70	3		12						
14	40	33	90	4		32						
15	53	44	100	5		40						
16	55	47		6		52						
17	64	58		7		60						
18	67	62		8		72						
19	76	71		12	5	76						
20	87	84		21	14	88						
21	95	94		31	23	100						
22	100	100		37	30							
23				45	39		2		11			
24				51	46		2		14			
25				58	53		4		25	2		16
26				62	58		15	11	36	3		22
27				73	70		35	29	64	17	15	28
28				100	100		100	100	10 0	100	100	10 0

Note. numbers in brackets represents standard deviations.

In total, 55% of all children completed the transcoding task flawlessly. When analyzing groups separately, 57% of control children and 36% of children with mathematics difficulties did not commit any errors. The rate of correct items increased along with grade (Table 2, Figure 1). As percentile distributions in Table 2 suggests, the task showed a ceiling effect in the third and fourth grades.

Reliability and internal consistency of the Arabic Number-Writing Task

A high internal consistency was revealed by the KR-20 formula (r = 0.91), when examining the whole sample. When assessing school grades separately, high KR-20 indexes were observed in the first and second grades but not in higher grades (Table 2). The high internal consistency was further confirmed by a split-half analysis of the whole sample (r = 0.94).

Item analysis

Error rates per item were calculated by dividing the total amount of incorrect answers by the overall number of responses. Error rates per item category were calculated by dividing the number of incorrect responses in those items belonging to a certain category by the number of items and the total number of responses to those items. Table 2 depicts error rates for each item, and Figure 1 depicts error rates separately by grade and children's group. For individual items, error rates varied from 0 to 28%, being particularly small for one- and two-digit numbers. Among one- and two-digit numbers, the most difficult item presented an error rate of modest 3%. Two-digit numbers imposed noticeable difficulties only for first graders with mathematics difficulties. Among 3- and 4-digit numbers, higher error rates were observed in control children attending the

1st grade and children with mathematics difficulties attending both 1st and 2nd grades. Third graders with mathematics difficulties still showed some difficulties in transcoding the more syntactically complex numbers. In 4th grade, both groups showed comparable and almost flawless performance.

For the analyses of the item discriminability, item-total correlations were calculated (Table 2). One-and two-digit numbers showed low discriminability indexes (i.e. < 0.40), which are in line with the very low error rates presented by these items. In turn, three-and four-digit numbers showed higher discriminability, varying from 0.54 to 0.85, thus suggesting that numbers with higher syntactical complexity are more discriminative for testing purposes.



Figure 1 - Error rates on individual items according to children's group and school grade.

1 lie 20		ioic Nulliber-w		Error	Item-total
Item	Number	Complexity	(ADAPT)	Rate	Correlation
1	4	null	2	0.000	-
2	7	null	2	0.000	-
3	1	null	2	0.003	.076
4	11	null	2	0.003	.101
5	40	null	2	0.010	.240
6	16	null	3	0.003	.051
7	30	null	2	0.005	.253
8	73	null	3	0.034	.367
9	13	null	2	0.006	.047
10	68	null	3	0.031	.332
11	80	null	2	0.005	.266
12	25	null	3	0.000	-
13	200	low	3	0.033	.543
14	109	moderate	4	0.046	.619
15	150	low	3	0.059	.717
16	101	moderate	4	0.045	.630
17	700	low	3	0.057	.590
18	643	high	5	0.093	.755
19	8000	low	3	0.107	.632
20	190	low	3	0.080	.714
21	1002	moderate	4	0.182	.665
22	951	high	5	0.111	.747
23	1015	moderate	4	0.207	.804
24	2609	high	7	0.271	.806
25	1300	moderate	4	0.221	.851
26	3791	high	7	0.276	.788
27	1060	moderate	4	0.261	.780
28	4701	high	7	0.266	.810

Table 2The 28 items of Arabic Number-writing Task according to complexity level.

Task accuracy

Accuracy of the Arabic Number-Writing Task in discriminating children with mathematics

difficulties was estimated with ROC analysis. Accuracy of Arabic Number-Writing Task in identifying children with mathematics learning difficulties is moderate in the 1st grade and high in the 2nd grade (AUC > 0.9; Table 3). However, in the next two grades the task did not show the same efficiency, achieving only a low accuracy (AUC < 0.7) in the 4th grade.

ROC analys:	18.								
				Conf. interval (95%)					
Grade	AUC	Std. Error	p	Lower	Upper	Cutoff	Spec.*	Sens.*	
			-				-	*	
1	.791	.077	.004	.641	.941	14	.667	.700	
2	.967	.014	< .001	.940	.994	20	.762	.880	
3	.706	.053	<.001	.603	.809	27	.709	.639	
4	.582	.060	.135	.464	.699	27	.849	.281	
Global	.655	.031	< .001	.593	.716	27	.575	.650	

Table 3 ROC analysi

* Specificity, ** Sensitivity

Influence of syntactic complexity on transcoding performance

The correlation between the number of errors and the number of transcoding rules was high (r(784) = 0.83; p < 0.001). Interestingly, this correlation remains stable even after removing the effect of the quantity of digits (r(783) = 0.59; p < 0.001).

To investigate in deeper detail the influence of syntactic complexity on number transcoding, we run a series of repeated measures ANOVAs separately for each school grade, having syntactic complexity as within-subjects factor, and mathematical ability as between-subjects factor.

In the first grade, a main effect of syntactic complexity, F(2, 11) = 32.91, p < .01, MSE = 2.71, $\eta_p^2 = .38$, reflected an increase in error rates as a function of the number of syntactic rules. Contrasts showed significant differences between all three levels of syntactic complexity (low vs.

moderate: F(1, 53) = 8.37, p < .01, MSE = 1.53, $\eta_p^2 = .14$; moderate vs. high: F(1, 53) = 30.06, p < .001, MSE = 3.85, $\eta_p^2 = .36$). A main effect of group, F(1, 53) = 7.63, p < .001, MSE = .62, $\eta_p^2 = .13$, revealed higher error rates for children with mathematics difficulties. Figure 2 shows the effects of syntactic complexity for each group in the four school grades. Moreover, an interaction between syntactic complexity and mathematical ability was observed, F(1, 53) = 3.48, p = .04, MSE = .29, $\eta_p^2 = .06$. Post-hoc tests reveal differences between all levels of item complexity in control children. In turn, children with mathematics difficulties showed comparable and better performance in items with low and moderate complexity than in items with high complexity.



Figure 2 – Error rates as a function of numerical complexity and mathematical ability for each school grade.

In the second grade, main effects of mathematical ability, F(1, 25) = .153.36, p < .005, MSE = 12.85, $\eta_p^2 = .38$, and syntactic complexity, F(2, 49) = 83.44, p < .001, MSE = 6.51, $\eta_p^2 = .25$, were significant. The interaction between mathematical ability and syntactic complexity also was significant, F(2, 49) = 3.79, p < .001, MSE = .30, $\eta_p^2 = .02$. Post-hoc comparisons revealed significant differences between all levels of item complexity in both groups of children.
In the third grade, the main-effect of syntactic complexity, F(2, 446) = 17.63, p = .001, MSE = .45, $\eta_p^2 = .07$, was significant. The main effect of mathematical ability was also significant, F(1, 23) = .35.67, p < .001, MSE = .46, $\eta_p^2 = .14$, as well as the interaction between complexity and mathematical ability (F [2, 446] = .3.95, p < .001, MSE = .10, $\eta_p^2 = .02$). Post-hoc comparisons revealed in both groups of children significantly better performance in items with low complexity than in items with moderate or high complexity.

In the fourth grade, the main-effect of mathematical ability was significant, though with a smaller effect-size than in earlier grades, F(1, 225) = 15.14, p < .001, MSE = .06, $\eta_p^2 = .06$. Additionally, the effect of syntactic complexity was significant, F(2, 510) = .9.61, p < .001, MSE = .14, = .04. Numbers with moderate and high complexity did not differ in their error rates, F(1.25) = .60, p = .439, MSE = .02, $\eta_p^2 = .002$. The interaction between group and syntactic complexity was not significant, F(2, 510) = .2.30, p = .11, MSE = .03, $\eta_p^2 = .01$.

In summary, our data showed that the effects of syntactic complexity and mathematical ability on number transcoding are consistent until the 4th grade, but with decreasing effect-sizes. The magnitude of the effect of mathematical ability varied from 0.14 in 1st grade to 0.06 in 4th grade, suggesting that in later grades, children with mathematics difficulties tend to reach the performance level of control children. Likewise, effect-sizes of syntactic complexity decreased across grades, ranging from 0.38 to 0.04, so that children's difficulties in transcoding more complex numbers tend to decline with schooling. Interestingly, only in the 4th grade the interaction between mathematical ability and syntactic complexity was not significant. The

absence of this interaction can be attributed to the ceiling effect in 4th grade, so that all children exhibited low error rates irrespective of syntactic complexity.

Discussion

The purpose of this study was to examine the psychometric properties of a number transcoding task and its usefulness in screening mathematics learning difficulties in children in the early school years. The Arabic Number-Writing Task is a simple and powerful instrument for assessing children's basic number transcoding skills in early elementary school. Furthermore, the task discriminates children depending on their mathematics learning difficulties with a high degree of sensitivity and specificity. The high reliability estimates of the transcoding task are promising regarding diagnostics and evaluation of cognitive interventions in mathematics difficulties. Closer item analysis revealed a strong impact of the number of rules necessary to transcode a number correctly. Moreover, results indicate that the number of rules per item can explain most of the group differences observed between children with and without mathematics difficulties. In the following, these results will be discussed in deeper detail.

General test properties

High internal consistency coefficients were observed in the transcoding task in first and second grade children, reverting in a high precision in the characterization of individual performance (Huber, 1973). More specifically, the reliability coefficients observed in the present study in first and second grades can be considered invariant according to the criteria established by Willmes (1985) and can be used confidently to estimate confidence intervals for individual performance in the clinical context. Although one can consider the Arabic Number-Writing Task economic in its

present format, particularly because of its flexibility regarding group testing and short duration, one may desire to reduce test length, particularly because of the relatively large number of very easy one- and two-digit items (see further discussion on this merit below). Test reduction seems to us to be practically feasible since the determinants of item difficulty are well-known and the pool of suitable items in the numeric interval between three- and four-digit numbers is large enough. In this context, the number of rules necessary to transcode individual items is particularly important as a criterion for the establishment of different groups of items. Accordingly, the number of transcoding rules is a good criterion to distinguish the level of competence typical of children with and without mathematics difficulties. This can be illustrated by the interactions between item complexity vs. mathematical ability observed in first to third grades, which reflect that performance is differently affected by syntactic complexity in the two groups. An adaptive version of the task could be constructed in which the number of rules necessary to transcode an item vary in an even more fine-grained scale than that employed in the present study.

Characterization of typical and atypical development of transcoding abilities

Overall, 1- and 2-digit numbers presented very low error rates in all school grades, regardless of children's mathematics abilities. These results are in line with the literature, which indicates that even kindergartners at risk for mathematics difficulties do not have troubles in transcoding small numbers (Landerl, Bevan, & Butterworth, 2004; van Loosbroek, Dirkx, Hulstijn, & Janssen, 2009), but instead can retrieve the Arabic forms directly from their long-term memory. Moreover, two-digit numbers also showed very low error rates in all school grades but were responsible for transcoding errors in children with mathematics difficulties only in first graders, but not in higher school grades.

In contrast, 3- and 4-digit numbers accounted for a large proportion of score variability, with high error rates being observed in the 1st grade and a steady decrease in higher grades. Interestingly, 2nd grade children without mathematics difficulties showed notable difficulties in transcoding 3and 4-digit numbers. In the 2nd grade children receive the formal instruction necessary for mastering the syntax of these numbers. Moreover, children with mathematics difficulties seem to demand one year longer than control children to master the same knowledge. When analyzing the interactions between individual achievement and syntactic complexity in ANOVA models, one observes that control children attending the 3rd and 4th grades barely committed errors in the low complexity items. In turn, children with mathematical difficulties continue to present errors when they achieve the 3rd grade, although the error rates also decrease steadily over time. These findings corroborate previous evidence showing that syntactic complexity has a strong impact on error rates in transcoding tasks (Camos, 2008; Moura et al., 2013; Zuber, Pixner, Moeller, & Nuerk, 2009). A delay in the acquisition of more complex transcoding rules has already been observed in children with mathematics difficulties (Moura et al., 2013), and typically developing children with lower working memory capacity (Camos, 2008). The present results confirmed this delay in the acquisition of transcoding rules observed in children with mathematics difficulties. To which extent these errors are also attributable to reduced working memory capacity has to be investigated in future studies.

The persistence of the effect of syntactic complexity in all grades constitutes strong evidence for the prominent role of rules in elucidating transcoding abilities even in third and fourth grades. Children with mathematics difficulties struggle in learning the more complex transcoding rules, as can be inferred from wrong frame errors (Moura et al., 2013). Wrong frame errors reflect the absence of knowledge of the magnitude intrinsic to each position in the digit sequence, that is, of place-value. Several studies have related the knowledge of place-value syntax with achievement in more complex numerical abilities, such as arithmetics (Mazzocco, Murphy, Brown, Rinne, & Herold, 2013; Moeller, Pixner, Kaufmann, & Nuerk, 2009; Moeller et al., 2011). Together, these pieces of evidence indicate that more abstract levels of numerical representation such as place-value knowledge can be assessed by means of the performance in the transcoding task and reinforces its utility when trying to predict arithmetics abilities of individual children.

Together, the effect of syntactic complexity and the high correlation between error rates and the number of transcoding rules suggest that working memory is an important variable associated with number writing. Working memory capacity has been associated in the transcoding research with storing the verbal string, searching in the long-term memory for lexical entries, parsing the previously non-acquired strings and applying the procedural rules (Barrouillet et al., 2004; Lochy & Sensabela, 2005). In previous research, we have also found this association between the number of rules and working memory, which suggests an implicit link between the syntactic complexity and working memory skills (Moura et al., 2013). Camos (2008) directly addressed this issue by investigating children with different levels of verbal working memory abilities. This author found a robust association between number of transcoding rules and the number writing performance as suggested by the ADAPT model. Moreover, Lopes-Silva, Moura, Julio-Costa, Haase and Wood (2014) showed that the influence of verbal working memory on number transcoding is mediated by phonemic awareness. According to the ADAPT model, phonological encoding is the first step in the number transcoding process. Further evidence suggests that visuospatial working memory capacity may be associated to syntactic transcoding errors related to the unit-decade inversion rule present in languages such as German, Dutch and Czech (Pixner, Zuber, Hermanová, Kaufmann, & Nuerk, 2011; Zuber, Pixner, Moeller, & Nuerk, 2009). This indicates that the Arabic Number-writing task is theoretically grounded on a cognitive model with high content and construct validity.

Task discriminability

For the first time, diagnostic accuracy of Arabic Number-Writing Task was assessed by means of ROC analyses. According to established criteria (Swets, 1988), moderate and high accuracy estimates were observed in the first and second grades respectively, while in third and fourth grades the accuracy clearly insufficient. Difficulties with number transcoding might remain traceable in higher school grades, but the Arabic Number-Writing Task in its present format is too easy to discriminate mathematics difficulties. It is possible that an adaptation of the task with the inclusion of more complex 5- and 6-digit numbers would support higher group discriminability. However, it is also possible that the cognitive profile of mathematics difficulties, as measured by the Arabic Number-writing task, is not stable over time, so that difficulties experienced in early phases can be, eventually, overcome, and then new difficulties may appear (Geary, Hamson, & Hoard, 2000; Gersten, Jordan, & Flojo, 2005). If this is the case, good discriminability of number transcoding tasks regarding mathematics difficulties may be limited to the first two school grades.

Future perspectives

Further research is needed to support the use of the Arabic Number-writing task in the clinicalepidemiological and research context. Validity of the Arabic Number-writing task as a screening instrument should be established by means of longitudinal studies investigating its power in predicting mathematics learning difficulties. But the Arabic Number-writing task also can be useful in the individual clinical assessment and in theoretical research. A further development in the individual assessment context is the design of an automatic algorithm for item generation, which allows the construction of more individualized and adaptive versions of the Arabic Number-writing task (e.g. Arendasy, Sommer, Mayr, 2012). The ADAPT model provides a very valuable basis to generate items in all difficulty levels. The estimates of item difficulty obtained from large-sample studies such as the present one establish the basis for such further developments. Since transcoding tasks combine both diagnostic sensitivity and specificity regarding mathematics achievement with a solid theoretical basis of the cognitive mechanisms driving individual performance, automatic item generation may reveal to be very valuable in the construction of adaptive and flexible instruments best suitable not only to characterize individual performance but also to evaluate the impact of interventions designed to remediate the negative impact of mathematics difficulties on cognition and performance.

Practical implications

The good psychometric properties of the Arabic Number-writing Task together with its simple administration and consistent theoretical ground make of it a useful tool for assessing basic numerical skills of young children in both clinical and research contexts. The task may provide a quick and cheap way for screening first and second graders at risk of mathematical difficulties both collectively, at school, and individually, in clinical settings. The benefits of the early identification of children with possible major difficulties in mathematics are incommensurable. It enables early intervention efforts, thus minimizing future consequences of low numeracy, such as low incomes and less job opportunities (Bynner & Parsons, 1997).

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CHAPTER 3

Study 2

STUDY 2:

Phonemic awareness as a pathway to number transcoding

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ABSTRACT

Although verbal and numerical abilities have a well-established interaction, the impact of phonological processing on numeric abilities remains elusive. The aim of the study is to investigate the role of phonemic awareness in number processing and to explore its association to other functions such as working memory (WM) and magnitude processing. One hundred and seventy two children from 2nd to 4th grade were evaluated regarding their intelligence, number transcoding, phonemic awareness, verbal and visuospatial WM and number sense (nonsymbolic magnitude comparison) performance. All of the children had normal intelligence. Among these measurements of magnitude processing, WM and phonemic awareness, only the last one was retained in regression and path models predicting transcoding ability. Phonemic awareness mediated the influence of verbal WM on number transcoding. Evidence points out that phonemic awareness is responsible for a significant impact on number transcoding. Such association is robust and should be taken into account in cognitive models of both dyslexia and dyscalculia.

Keywords: phonemic awareness, verbal working memory, transcoding, ADAPT, asemantic transcoding models

Introduction

Mastering reading and writing numbers in their verbal and Arabic forms is an essential skill for daily life (Lochy and Censabella, 2005). Being able to manipulate numbers and convert them from one format into another is one of the first steps in children's mathematical learning and starts to be formally trained in kindergarten. The ability to establish a relationship between the verbal and Arabic representations of number, when a conversion of numerical symbols from one notation to the other is necessary, is called number transcoding (Deloche and Seron, 1987).

The verbal number system is linguistically structured and, although it may differ among languages, there are some common basic principles and regularities (Fayol and Seron, 2005). It is typically composed of a lexicon of single words that designate a few quantities (like five, eleven, seventy and hundred) and organized by a syntax that arranges these lexical units in order to represent any possible quantity. The two basic syntactic principles are the relations of addition and multiplication. In this sense, numbers are represented as sum relationships (e.g.: eighty-one means eighty plus one) and product relationships (e.g.: three hundred means three times hundred). The number words in Portuguese are similar to the English number words in the sense that they are also organized in lexical classes for units, decades and particulars (the -teens in English) (Wood et al., 2006).

The Arabic code is more complex and is acquired later in development (Geary, 2000). Its lexicon is composed of only a small set of different symbols (digits from 0 to 9), and the basic syntactic

principle that combines them to form all numbers is the positional value (or place-value). According to this principle, the digit's value depends on its position in the numerical string and is given by a power of base ten. Therefore, in the case of three-digit numbers, the first digit (from right to left) is multiplied by 100, the second by 101, and so on. The number 124, for example, represents a quantity equal to $1 \times 102 + 2 \times 101 + 4 \times 100$ (or 100 + 20 + 4). The digit 0 has a special syntactic role when it denotes the absence of a given power of ten, as occurs in numbers with internal zeros, for example the number 406 ($4 \times 102 + 0 \times 101 + 6 \times 100$).

One preeminent model of number transcoding is ADAPT (A Developmental, Asemantic, and Procedural model for Transcoding from verbal to Arabic numerals; Barrouillet et al., 2004). According to ADAPT, the inputs are coded into a phonological sequence and the parsing mechanisms then subdivide this sequence into smaller units to be processed by a production system. This production system is related to rules devoted to the retrieval of Arabic forms from long-term memory (LTM) (called P1 rules), to managing the size of digit chains (P2 and P3 rules, which create a frame of two or three slots) and to filling these slots (if there are any empty slots, P4 rules will fill them with 0s). Separators, such as thousands and hundreds, are used to identify the number of slots; once every segment is placed in its digit form in the chain, it is transcribed. The model accounts for the development of transcoding processes through practice: experience leads to an expansion of the numerical lexicon and improvement of conversion rules.

The ADAPT model is the only cognitive model of number transcoding which makes testable predictions regarding both working memory capacity and phonological/lexical representations

and their respective roles in the typical and atypical development of transcoding abilities. Moreover, even though it is not explicitly stated in the original publication (Barrouillet et al., 2004), ADAPT clearly emphasizes the importance of phonological encoding in the first steps of number writing production, and this has not been investigated in more detail. Because both working memory and the ability to form lexical representations of numbers and, as we assume here, phonemic awareness are related to mathematical performance, ADAPT is the only transcoding model directly examined in the present study.

Short-term memory and working memory (thereafter WM) are involved in the temporary storage of verbal information, lexical retrieval, and the execution of the manipulations to generate the Arabic output. Working memory representations are also involved in creating a sequence of digits and possibly blank spaces to be filled with subsequent procedures. It has been consistently related to number transcoding performance and error patterns (Camos, 2008; Zuber et al., 2009; Pixner et al., 2011). The role of working memory in transcoding tasks can be outlined in the following steps: encoding the number to be transcoded; monitoring the application of transcoding rules and the production of the numeral (Lochy and Censabella, 2005).

Another cognitive mechanism that may be involved in number transcoding is phonemic awareness. Phonemic awareness is the subcomponent of phonological processing which is related to the ability to perceive and manipulate the phonemes that constitute words (Wagner and Torgesen, 1987). According to the ADAPT model (Barrouillet et al., 2004), the phonological encoding of the verbal numerals is the primary step in transcoding procedures, before the use of

algorithm rules and retrieval from LTM. Therefore, limitations in phonological processing capacity may constrain the ability to transcode, particularly in the case of longer and more complex numbers. Phonological processing may also interact with the capacity of verbal working memory. The more demanding the phonological processing of numerical stimuli, the fewer resources would remain available in verbal working memory for transcoding. Although the conversion of a verbal representation to an Arabic one is related to phonological representations, this association has not yet been investigated in detail in the ADAPT model.

Krajewski and Schneider (2009) found that phonological awareness facilitates the differentiation and manipulation of single words in the number word sequence. These authors built a model of early arithmetic development that postulates three different levels: (1) basic numerical skills, in which children are already able to discriminate between quantities and to recite number words, without accessing their quantitative semantic meaning; (2) quantity-number concept, when there is a linkage between magnitudes and the number words that represent them; (3) number relationships, the point at which children understand that the difference between two numbers is another number.

According to these authors, phonological awareness (measured by phoneme synthesis and rhyming tasks) plays an important role in the first level. The authors claim that because this phonological skill is related to the ability to differentiate and manipulate meaningful segments of language, it is also important in differentiating number words ("one," "two," "three" instead of "onetwothree").

In view of the above, the aim of this study is to investigate the role of specific cognitive mechanisms underlying number transcoding such as general cognitive ability, verbal and nonverbal short-term and working memory, magnitude representation, and phonemic awareness. More specifically, our main goal was to investigate the relative impact of phonemic awareness on number transcoding. Phonemic awareness is related to reading and spelling skills (Wagner and Torgesen, 1987; Castles and Coltheart, 2004; Hulme et al., 2012; Melby-Lervå et al., 2012), and recent studies have also focused on its association with arithmetic fact retrieval and with arithmetic word problems (Hecht et al., 2001; Boets and De Smedt, 2010; De Smedt et al., 2010). Importantly, many measures of phonemic awareness, such as the phoneme elision task employed in the present investigation, require a certain availability of working memory resources. Working memory is recruited in such tasks when the participant must hold a word in mind while determining the phonological information to be deleted (De Smedt et al., 2010). Both verbal and visuospatial working memory play important roles in numerical transcoding according to the ADAPT model (Camos, 2008; Zuber et al., 2009), but no study so far has investigated the specific contribution of phonemic awareness and working memory in number transcoding tasks.

Two main hypotheses will be addressed in the present study: First, based on the central role assigned by the ADAPT model to working memory capacity (Barrouillet et al., 2004; Camos, 2008), one can argue that working memory contributes to number transcoding independently because working memory capacity is putatively implicated in the use of transformation rules and procedures employed during transcoding. Second, at least part of the influence of working memory on number transcoding should be mediated by phonemic awareness. Phonemic awareness scores are assumed to index the quality of the underlying phonological representations. These representations are related to the perception and manipulation of sound-based processes

(Simmons and Singleton, 2008); therefore, phonemic awareness performance would have an impact on verbal working memory and transcoding skills.

Materials and Methods:

The study was approved by the local research ethics committee (COEP–UFMG) and is in line with the Declaration of Helsinki. Children participated only after informed consent was obtained. Informed consent was obtained in written form from parents and orally from children.

Sample:

A total of 487 children in grades 2–4 were invited from public schools in Belo Horizonte, Brazil. Of these children, 207 (42%) children agreed to take part in this study. Testing was conducted in the children's own schools. The various tasks were presented in four different pseudo-random orders during one session that lasted approximately 1 h.

We excluded five children from the sample due to low intelligence (performance on Raven's Colored Progressive Matrices below one standard deviation). One child did not complete the entire battery and was also excluded from the analysis. Twenty-nine children were excluded from further analyses because either they had a poor R^2 on the fitting procedure to calculate their internal Weber fraction on the non-symbolic comparison task ($R^2 < 0.2$) or they showed an internal Weber fraction that exceeded the limit of discriminability of the non-symbolic magnitude comparison task (w > 0.6). The final sample comprised 172 children (55.2% girls), with a mean age of 111.84 months (SD = 10.90), ranging from 94 to 140 months.

Instruments:

The following instruments were used in the cognitive assessment: Raven's Colored Progressive

Matrices, Digit Span, Corsi Blocks, Non-symbolic magnitude comparison task, Phoneme Elision and Arabic number writing task.

(a) Raven's Colored Progressive Matrices: general intelligence was assessed with the ageappropriate Brazilian validated version of Raven's Colored Matrices (Angelini et al., 1999). The analyses were based on z-scores calculated from the manual's norms.

(b) Digit Span: Verbal short-term and working memory were assessed with the Brazilian WISC-III Digit Span subtest (Figueiredo, 2002). Performance in the forward order was considered a measure of verbal short-term memory, and the backward order was used to assess verbal working memory (Figueiredo and Nascimento, 2007). We evaluated the total score (correct trials x span) in both the forward and backward orders.

(c) Corsi Blocks: This test is a measure of the visuospatial component of short-term and working memory. It consists of a set of nine blocks, which the examiner taps in a certain sequence. The test starts with sequences of two blocks and can reach a maximum of nine blocks. We used the forward and backward orders according to Kessels et al. (2000). In the forward condition, the child is instructed to tap the blocks in the same order as the examiner, and in the backward condition, in the reverse order. We also evaluated the total scores.

(d) Non-symbolic magnitude comparison task: In this task, the participants were instructed to compare two simultaneously presented sets of dots, indicating which one contained the larger number. Black dots were presented on a white circle over a black background. In each trial, one of the two white circles contained 32 dots (reference numerosity) and the other contained 20, 23, 26, 29, 35, 38, 41, or 44 dots. Each magnitude of dot sets was presented eight times. The task comprised 8 learning trials and 64 experimental trials. Perceptual variables were varied such that in half of the trials the individual dot size was held constant, while in the other half, the size of the area occupied by the dots was held constant (see exact procedure descriptions in Dehaene et

al., 2005). Maximum stimulus presentation time was 4.000 ms, and the inter-trial interval was 700 ms. Before each trial, a fixation point appeared on the screen: a cross, printed in white, with each line 30 mm long. If the child judged that the right circle presented more dots, a predefined key localized in the right side of the keyboard should be pressed with the right hand. However, if the child judged that the left circle contained more dots, then a predefined key on the left side had to be pressed with the left hand (Costa et al., 2011). As a measure of the number sense acuity, the internal Weber fraction (w) was calculated for each child based on the Log-Gaussian model of number representation (Dehaene, 2007), with the methods described by Piazza et al. (2004).

(e) Phoneme Elision: This is a widely accepted measure of phonemic awareness (Wagner and Torgesen, 1987; Castles and Coltheart, 2004; Hulme et al., 2012; Melby-Lervå et al., 2012). The child hears a word and must say what the word would be if a specified phoneme in the word were to be deleted (e.g., "filha" without /f/ is "ilha" [in English, it would be similar to "cup" without /k/ is "up"). The test comprises 28 items: in 8 items, the child must delete a vowel, and in the other 20, a consonant. The consonants to be suppressed varied by place and manner of articulation. The phoneme to be suppressed could be in different positions within the words, which ranged from 2 to 3 syllables. The internal consistency of the task is 0.92 (KR-20 formula). (f) Arabic number writing task: To evaluate number transcoding, children were instructed to write the Arabic forms of dictated numbers. This task consists of 40 items, up to 4 dig-its (3 onedigit numbers, 9 two-digit numbers, 10 three-digit numbers and 18 four-digit numbers). The oneand two-digit numbers were classified as "lexical items" (12 items), and the other 28 items require the use of algorithm-based rules in order to be written (Barrouillet et al., 2004; Camos, 2008). This task has been used in a previous study with a comparable sample, and the consistency of this task was KR-20 = 0.96 (Moura et al., 2013).

Analysis

The differential impact of phonemic awareness and working memory on number transcoding was investigated in a hierarchical regression analysis with Arabic number writing as the dependent variable. Age and intelligence were entered first, and working memory and the Weber fraction in a second step, using the stepwise method. The phoneme elision task was entered in the model in a third step, also using the stepwise method. This allowed us to investigate the specific contribution of phonemic awareness to number transcoding performance after working memory variance was taken into account.

As a complement, path analyses, including all measures of age, intelligence, working memory and phonemic awareness were calculated, to determine the specific contribution of phonemic awareness as a mediator of the effect of working memory on number transcoding.

Results:

Thirty-three percent of the children did not commit any errors in the number transcoding task. Ninety-three percent of the children did not commit any errors on the numbers that can be lexically retrieved (items 1–12). According to what is suggested by the ADAPT model, errors rates increased with the number of rules required for number transcoding. In the numbers that required 3 transcoding rules, 50% of the children committed errors, in the 4-rules, 71.6% presented some errors, in the 5-rules, 73.3% and, finally in the more complex items (6 and 7 rules), 84.5% of the children committed, at least, one error.

Since one-third of the sample did not commit any error in the transcoding task, one may argue that they should be excluded from the sample to avoid biases in the estimation of the covariance

matrix, particularly with regard to the association between transcoding performance and other cognitive functions. To investigate the occurrence of bias, regression and path analyses were performed in the full sample and in the sample without the children with perfect score in the transcoding task. Results were numerically comparable in both regression and path analyses and their interpretation was exactly the same. For this reason, we decided to report the results obtained by analyzing the full sample.

Association between cognitive variables and transcoding ability

First, the specific impact of the different cognitive mechanisms on number transcoding was evaluated by means of hierarchical regression models. To approximate a normal distribution, error rates of the Arabic number writing task were arcsine transformed. Initially, we examined the general association between these measures through Pearson's correlations. Inspection of Table 1 reveals that the error rates observed in the number transcoding task were negatively correlated to age, intelligence, working memory, and phonemic awareness. There was also a weak positive correlation between error rates in number transcoding and the Weber fraction, which may reflect the maturation level of more general numerical skills. Moreover, phonemic awareness was significantly correlated to intelligence and working memory.

	1	2	3	4	5	6	7	8
1. Age (in months)	1							
2. Raven	23**	1						
3. Digit Span-Forward	.19*	.19*	1					
4. Digit Span-Backward	.05	.34**	.18*	1				
5. Corsi Blocks Forward	.19*	.28**	.15*	.20**	1			
6. Corsi Blocks Backward	.01	.34**	.14	.36**	.36**	1		
7. Weber fraction	19*	11	17*	19*	16*	13	1	
8. Phoneme elision	.11	.36**	.23**	.36**	.24**	.25**	13	1
9. Number Transcoding	11	17*	11	15*	10	13	.21**	36***

Table 1. Correlations between the neuropsychological measures

**. Correlation is significant at the 0.01 level (2-tailed)

*. Correlation is significant at the 0.05 level (2-tailed)

To investigate in more detail the specific impact of phonemic awareness on transcoding abilities, a hierarchical regression model was calculated (Table 2). In this model, more general determinants of cognitive development were entered first, and more specific predictors of transcoding ability were included later on, in a hierarchical fashion. In step 1, age and intelligence were included as general factors that predict school achievement, using the enter method. In step 2, the following cognitive measures were included: Weber fraction and the total scores of the forward and backward orders of Digit Span and Corsi Blocks. Last, in step 3, we included the phoneme elision score. The stepwise method was used in steps 2 and 3 to avoid redundancy and to guarantee a high degree of parsimony.

The regression model reveals that after removing the effects of age and intelligence in step 1, verbal working memory remains a significant predictor of transcoding performance in step 2. Nevertheless, the addition of phonemic awareness to the model in step 3 leads to the exclusion of verbal working memory. Phonemic awareness, along with age and intelligence, was a significant predictor of number transcoding and absorbed the impact of verbal working memory on transcoding performance. The model explains a moderate amount of variance (Table 2). Measures of the approximate number system, visuospatial short-term memory, and visuospatial working memory were not retained in the model.

Predictor	Beta	Partial t	Sig	r ² change
Intercept		10.14	<.001	
Age (months)	-0.404	-6.487	<.001	0.305
Raven	-0.225	-3.282	0.001	0.303
Digit Span-Backward	-0.089	-1.358	0.176	Excluded
Weber fraction	0.095	1.545	0.124	Excluded
Digit Span-Forward	-0.056	-0.885	0.378	Excluded
Corsi Blocks- Backward	-0.035	-0.529	0.598	Excluded
Corsi Blocks-Forward	-0.003	-0.051	0.959	Excluded
Phoneme elision	-0.337	-5.038	< 0.001	0.088

Table 2. Regression	Analysis for Number	• Transcoding	(errors arcsine.	adjusted $r^2 = 0.41$)
racie 2. regression	i indi joio i oi i taine ei	1 I millio Counting	(on one on one,		• • •

The reason to employ a hierarchical regression model in this analysis is to demonstrate the validity of the present experimental setup. By entering the measures of working memory in the regression model first we are able to replicate previous studies and thereby show that our measures of working memory were well-chosen and are associated to transcoding abilities. After completing this step of validation of well-established results, we continue the investigation showing that phonemic awareness absorbs the impact of measures of working memory on transcoding capacity. We have also calculated a regression model allowing the effect of phonemic awareness to vary simultaneously to measures of working memory, that is, with no hierarchical distinction between these variables. Results were largely comparable with those reported previously: only phonemic awareness is retained in the model along with intelligence and age (R2 = 0.64; adjusted R2 = 0.40; b = -0.02).

Describing the roles of phonemic awareness and verbal memory in Arabic number transcoding

As shown in the previous section, the influence of the verbal working memory on number transcoding is shared with phonemic awareness. Therefore, as a complement to the previous findings, path analyses including both working memory and phonemic awareness, as well as Weber fraction, were calculated in order to investigate the interplay of these variables in number transcoding.

To determine the strength of the effect of phonemic awareness on number transcoding, a sequence of models was calculated and compared. Chi-square and the approximate fit indexes root mean square residual (RMR), goodness of fit index (GFI), adjusted goodness of fit index (AGFI), comparative fit index (CFI) and root mean square error of approximation (RMSEA) were used to evaluate model quality. A non-significant chi-square indicates no significant

discrepancy between model and data. The RMR measures the ratio of residuals in comparison to the covariances expressed by the models. Values smaller than 0.10 are considered adequate. GFI, AGFI, and CFI evaluate the degree of misspecification present in the model. Usually, the best acceptable values are greater than 0.90. Finally, the Root Mean Square Error of Approximation, or RMSEA, considers the model complexity when evaluating the model fit. The RMSEA is considered acceptable when it is lower than 0.05. The Chi-square difference between models was employed to compare models with increasing numbers of free parameters. Models were calculated in the software AMOS v.19 using the maximum likelihood estimation function.

To control for the influence of developmental and intellectual levels on the path models, we calculated the unstandardized residuals of the independent variables (short-term and working memory, Weber fraction and phonemic awareness), in which the portion of variance due to age (in months) and/or intelligence was removed. These adjusted values of working memory, magnitude processing and phonemic awareness were entered as the exogenous variables in the path analyses. All the covariances between the exogenous variables were set as free (Figure 1).

Those variables with negative standardized values indicate that higher scores in these predictors lead to lower error rates in the number transcoding task. The only exception is the Weber Fraction path, in which higher values indicate poorer magnitude representation acuity and, hence, more errors in number transcoding.

Fit statistics of path models are shown in Table 3. The first and most complex model (ALL PATHS) included the two measures of short-term and working memory (forward and backward versions of Digit Span and Corsi Blocks), as well as Weber fraction and an additional Phoneme

Elision mediation path between both the forward and backward versions of the Digit Span and the number writing tasks. This model presented adequate fit indexes but is not parsimonious. Models with fewer parameters to be estimated were designed and were compared to the ALL PATHS model and to one another.

First, the NO VISUOSPATIAL model removed the paths from visuospatial memory to transcoding. Accordingly, the NO ANS model also suppressed the path from the Weber fraction to transcoding. In one further step, two models were calculated. In the first (MEDIATION PATH), the contribution of verbal working memory to transcoding is partially mediated by phonemic awareness. Finally, to determine the relevance of phonemic awareness for transcoding, in the last model, the path from Phoneme elision to Number transcoding was removed, while the direct paths from verbal working memory to transcoding were retained (NO MEDIATION). If the exclusion of any of these paths leads to a statistically significant decrease in model fit, one may conclude that the specific parameters removed from the more parsimonious version of the path model contribute substantially to model fit.

Inspection of Table 3 reveals that all models including the Phoneme Elision-mediation path reached satisfactory fit levels. Nevertheless, all models presented large residuals, as indicated by the RMR, which suggests that the variables included in the models were not sufficient to fully explain the variance in the number writing task. However, non-significant Chi-squares and the other fit measures associated with these models were largely acceptable.

Overall, the model that presented the worst fit indices was the one that excluded the Phoneme Elision-mediation path and assumed that Digit Span has a direct influence on number transcoding (NO MEDIATION). Model comparisons corroborate these results because the model NO MEDIATION presented statistically poorer fit than all other models. Its chi-square was statistically significant, and the model did not present any adequate fix indexes (Table 3). This finding suggests that phonemic awareness is a relevant predictor of transcoding performance, with substantial specific contribution. Moreover, comparisons among all other models only produced non-significant chi-square differences. Given the statistical equivalence of these models, one may select the model MEDIATION PATH, in which the effect of working memory on transcoding performance is partially mediated by phonemic awareness, as the most parsimonious description of the present data. Importantly, the association between verbal working memory and phonemic awareness. Regression values of the model MEDIATION PATH are depicted in Figure 1.



Figure 1. Path-analyses models describing the effects of WM, Weber fraction as well as

phonemic awareness in number transcoding task. Paths marked with * are significant at the level .05 and with ** are significant at the level .001.

Number transcoding model	χ^2	df	р	RMR	GFI	AGFI	CFI	RMSEA
ALL PATHS	0.793	3	0.851	1.066	0.999	0.988	1.000	0.000
NO VISUOSPATIAL	0.953	5	0.966	1.067	0.998	0.991	1.000	0.000
VERBAL PATH	2.396	6	0.880	1.067	0.996	0.981	1.000	0.000
MEDIATION PATH	4.156	8	0.843	1.071	0.993	0.976	1.000	0.000
NO MEDIATION	30.654	9	< 0.001	3.388	0.954	0.858	0.563	0.119

Table 3. Fit statistics for path models regarding Number Writing performance

Note: RMR: root mean square residual; GFI: goodness of fit index; AGFI: adjusted goodness of fit index; CFI: comparative fit index; RMSEA: root mean square error of approximation.

Discussion

The present study investigated the impact of phonological skills on a number transcoding task, and it is, to our knowledge, the first to simultaneously evaluate the relative impact of short-term and working memory, number sense and phonemic awareness on number transcoding. Our results revealed two main findings. First, we confirmed previous evidence of a verbal working memory effect on number transcoding, and, more importantly, we provided evidence of a relationship between number transcoding and phonemic awareness. Our second main finding is that the well-established relationship between verbal working memory capacity and number transcoding is mediated by phonemic awareness abilities. In the following sections, these topics will be discussed in more detail.

The impact of verbal and visuospatial working memory on Arabic number writing

The performance of children in the number writing task was far from being flawless. They present many errors on the more complex two-, three-, and four-digit items, which require more than three transcoding rules, according to ADAPT. These findings are in accordance to what has been reported in the literature regarding transcoding skills of school aged children (Moura et al., 2013) and have been interpreted as a product of working memory processes in number transcoding (Camos, 2008). However, little is effectively known about the selective impact of different components of working memory on number transcoding. To our knowledge, this was the first study to analyze this problem in greater depth. Although a specific role of the central executive function in transcoding has been suggested (Camos, 2008), the present study is the first to explore the impact of phonological and visuospatial working memory in a number writing task and distinguish them from the central executive. We provide evidence regarding the specific role of phonological working memory and, more precisely, of the quality of underlying phonological representations, by means of the phonemic awareness performance.

Working memory plays an important role in the algorithmic-based procedures of number transcoding (Camos, 2008; Pixner et al., 2011). Essentially, it is believed to be involved in the maintenance of verbal units from the verbal numbers and in managing the new digit chain. In our study, we found that better verbal working memory capacity was associated with higher number transcoding performance. Interestingly, the same does not apply to the visuospatial components of short-term and working memory, as none of them revealed an association with transcoding performance in correlation, regression or path analyses. In a previous study by Zuber et al.

(2009), the visuospatial working memory component was associated with the management of Arabic code syntax. Nevertheless, it is important to note here that the sample used in this other study was composed of German-speaking first graders, and the German number word system is different from the Portuguese system. In German, the order of the units and decades in the verbal numerals is inverted in comparison to the Arabic ones. One possibility, therefore, is that transcoding numbers in Portuguese demands less visuospatial working memory capacity than in languages with this inversion. Linguistic comparison research remains necessary to confirm this hypothesis.

Raghubar et al. (2010) reviewed evidence indicating that the influence of the subcomponents of working memory on arithmetic performance might vary according to age. The visuospatial component is recruited in earlier phases of development, while children are still learning basic mathematical concepts, whereas the phonological loop is more relevant after these skills have already been mastered. Although Raghubar et al. (2010) did not specifically discuss number transcoding, this study reviews evidence regarding the complex and dynamic nature of the relationship between working memory and math achievement. Consistent with these results, no effect of visuospatial working memory on number transcoding was observed in second- to fourth-grade children in the present study.

The relationship between verbal working memory and phonemic awareness

The first step of writing Arabic numbers from dictation proposed by the ADAPT model (Barrouillet et al., 2004) is the phonological encoding of the auditory input, which consists of verbal numerals. Nevertheless, the procedures involved in this phonological encoding are still not completely specified. Here we showed that, in addition to working memory capacity, phonemic

awareness also plays an important role in number transcoding. Our results showed that even when considering the influence of working memory and basic numerical skills on number transcoding, the predictive value of phonemic awareness abilities was substantial. This suggests that phonemic awareness is an important facilitator of the phonological encoding required in the initial steps of number transcoding.

Another aim of the present study was to clarify the influence of phonemic awareness on number transcoding. We aimed to investigate whether there is a direct influence of verbal working memory on number transcoding or if this association would be mediated by phonemic awareness. Our results presented evidence showing that phonemic awareness mediates the influence of verbal working memory in number transcoding, even after controlling for the effects of age and intelligence. In the path analyses, the removal of the Phoneme Elision-mediation path had a deleterious effect on model fit, which suggests that this parameter contributes crucially to improve the model fit.

This finding is consistent with the ADAPT model, which postulates that the first step in number transcoding would be the encoding of the verbal string into its phonological form (Barrouillet et al., 2004). This encoding phase would be followed by parsing procedures that segment these strings into smaller units. Smaller units are then sequentially processed through a production system in which verbal working memory is required for transcoding algorithms. It is possible to hypothesize that phonemic awareness would be the main cognitive precursor engaged in the phonological encoding phase that precedes further verbal working memory involvement in number transcoding.
A plausible explanation for the association between phonemic awareness and the influence of verbal working memory in number transcoding is the "weak phonological representation hypothesis" (Simmons and Singleton, 2008). According to this model, phonological processing deficits would impair the quality of phonological representations and thus affect aspects of numerical cognition that involve the manipulation of a verbal code.

The performance in verbal working memory and phonemic awareness depend on the same underlying and latent phonological representations (Hecht et al., 2001; Alloway et al., 2005; Durand et al., 2005). In our study, it was also possible to observe this association through the positive correlation between verbal working memory and phonemic awareness. Baddeley et al. (1975) had already suggested that, given that verbal short-term memory is a speech-based system, its capacity should be measured in more basic speech units, such as phonemes. Oakhill and Kyle (2000) also found that phonemic awareness (operationalized by means of phoneme elision and phoneme segmentation tasks) had a strong association with word and sentence span.

Evidence indicates that the influences of phonemic awareness and verbal working memory on literacy acquisition are both shared and unique (Mann and Liberman, 1984; Alloway et al., 2005). Factor analytical studies indicate that different types of phonological awareness tasks are loaded onto a single latent construct (Schatschneider et al., 1999). Tasks vary, however, in the additional cognitive demands they impose, regarding, for instance, working memory and other general cognitive components. According to this type of reasoning, different phonemic awareness tasks assess a common phonological processing construct plus additional varying components that change according to task demands. A task such as phoneme elision would consist then of at least two components, one tapping the phonological latent construct and the other one depending on

working memory demands. Previous studies (Oakhill and Kyle, 2000; Alloway et al., 2005) have investigated the influence of verbal working memory on phonemic awareness performance. This question, however, is rather complex and our results emphasize the importance of also investigating the other direction of this relationship. This is especially relevant regarding the interplay between verbal working memory, phonemic awareness and number transcoding skills.

Another dimension adding complexity to the relationship between phonemic awareness and verbal working memory is the child's individual level of development, which may be characterized as the degree of automatization in phonological processing. Before the child acquires expertise with phonemic awareness, a task such as phoneme elision may impose heavy demands on the central executive. As the child progressively acquires experience with phonological processing, this task can be solved in a more automatic way, freeing working memory resources for other tasks relevant for more advanced operations. If, however, the child does not acquire abilities of accurately and automatically processing the phonemic units, precious working memory resources will be less available for numerical transcoding. Accurate and automatic phonemic processing liberates sparse processing resources necessary to solve more complex tasks.

Disclosing a complex relationship among working memory, phonemic awareness and transcoding has important consequences for math achievement in general and for its disorders. School achievement in reading and/or mathematics depends on a complex interaction between general and specific cognitive factors. As the child acquires expertise in specific domains, such as phonemic and/or quantitative representations, processing resources are liberated to work in increasingly more complex activities. The accurate and automatic nature of more basic sound and

quantitative representations may thus influence the whole process of school learning, explaining variances both in achievement and in working memory. Johnson (2012) recently proposed that the occurrence of learning disabilities depends on such an interaction between specific and general cognitive factors. If a specific impairment, say in phonological or number processing, can be compensated by central executive resources, there is a smaller probability that the individual develops a learning disability. Otherwise, if executive processing resources are not sufficient to compensate or automatize basic cognitive processes, difficulties persist. This hypothesis has been explored in another report, investigating two cases of math learning difficulties (Haase et al., in press, this issue). In one case, math learning difficulties were associated with a lack of automatization and in the other case with impaired executive working memory resources.

There have been few studies that directly addressed the relationship between verbal memory and phonemic awareness during the performance of arithmetic tasks. Leather and Henry (1994) claim that both constructs share a certain amount of variance with arithmetic performance because phonemic manipulation demands arithmetical processes (for instance, phoneme elision tasks require, literally, the subtraction of a sound) and also involve working memory for the mental retention and management of verbal information. Phoneme elision tasks require both storage and processing of phoneme units because children usually hold the word in mind while deleting one sound and producing the new word with what is left (Oakhill and Kyle, 2000). Hecht et al. (2001) longitudinally investigated the role of phonological awareness in arithmetic development of children from different age ranges and found that from the 3rd to 4th grades, as well as from the 4th to 5th grades, this was the only subcomponent of phonological processing that explained the growth of performance in a standardized arithmetic task. According to the authors, the same memory resources engaged in arithmetic problem solving are also recruited in phonological

awareness tasks.

Our findings are in accordance to what was reported by Michalczyk et al. (2013). The authors also found that the simultaneous inclusion of verbal and visuospatial working memory, the central executive as well as phonological awareness in a regression model showed that only phonological awareness—none of the working memory subcapacities—had a direct impact on basic quantity-number competencies. In this study, they investigated the performance of children aged 5 and 6 in a number sequence task, in which children had to recite the number word sequence forwards up to 31 and backwards from 5. Afterwards they had to name 3 subsequent and 3 preceding number words. Even though they did not use a transcoding task, one can infer from this result that phonological awareness might mediate the relation between verbal working memory and number words knowledge. Nevertheless, as mentioned above, our study was the first one to provide evidence regarding the mediation of the effect of verbal working memory on number transcoding by phonemic awareness.

Final remarks

Mathematics encompasses a range of several different competences, such as numerical estimation, word problems, fact retrieval and number transcoding. Standardized arithmetic tasks usually assess these different abilities simultaneously and do not tap their specificities. It is important to investigate the distinct cognitive mechanisms that are associated with each of these mathematical skills. In our study, we concluded that phonemic awareness and verbal memory are directly connected to number transcoding, being important pathways between the verbal input and the transcription of the Arabic output.

The acuity of number sense, as measured by the Weber Fraction, did not influence number writing, suggesting that the assessment of numerical magnitude is not a necessary step in number transcoding. The acuity of number sense has been considered an important predictor of arithmetic performance (Halberda et al., 2008), but its relationship to number transcoding is less explored.

Although we did not explicitly assess children with learning disabilities, our results provide additional support to the hypothesis that phonemic awareness might be a cognitive mechanism that underlies both dyslexia and dyscalculia. Epidemiological studies describe high comorbidity rates between reading and mathematical difficulties: approximately 40% of dyslexics also have arithmetical difficulties (Lewis et al., 1994), and the prevalence of dyslexia and dyscalculia is similar, approximately 4–7% (Dirks et al., 2008; Landerl and Moll, 2010). The finding that phonemic awareness is related to number transcoding is useful in the comprehension of mathematical difficulties presented by dyslexic children (Haase et al., in press, this issue). We suggest that this should also be assessed in neuropsychological evaluations as well as in clinical interventions for children with learning disabilities.

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CHAPTER 4

Study 3

STUDY 3:

What is specific and what is shared between numbers and words?

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ABSTRACT

Reading and spelling performance have a significant correlation with number transcoding, which is the ability to establish a relationship between the verbal and Arabic representations of numbers, when a conversion of numerical symbols from one notation to the other is necessary. The aim of the present study is to reveal shared and specific mechanisms involved in reading and writing words and Arabic numerals in Brazilian school-aged children. One hundred and seventy two children from 2nd to 4th grades were evaluated. All of them had normal intelligence. We conducted a series of hierarchical regression models using scores on word writing and reading single words and arabic numerals, as dependent variables. As predictor variables we investigated intelligence, the phonological and visuospatial components of working memory and phonemic awareness. All of the writing and reading tasks (single word spelling and reading as well as number reading and number writing) were significantly correlated to each other. Statistics models not including general intelligence pointed to significant associations of phonemic awareness and working memory with each of the school tasks. Working memory effects guarded a certain specificity: visuospatial working memory predicts number writing and phonological working memory influences word reading. Inclusion of general intelligence in the models lead to different results, since effects of working memory disappeared or were substantially attenuated. This suggests that general intelligence and working memory share a considerable amount of variance. Associations between phonemic awareness and each of the school tasks remained. This suggests that phonemic awareness is a modular cognitive ability shared by several school tasks and might be an important factor associated to the comorbidity between dyslexia and dyscalculia.

Keywords: reading, spelling, number transcoding, phonemic awareness, intelligence

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Introduction

Reading and math performance in school-aged children are related in important ways. In this study, we set out to investigate shared and nonshared mechanisms involved in word reading/spelling and number transcoding abilities. Genetically-informed studies indicate that performance in standardized math, spelling and reading achievement tests substantially correlate at the phenotypic and genetic levels, both in typically and atypically developing children (Hart et al., 2009; Kovas et al., 2005; 2007). The discovery of genetic correlation between reading/spelling and math achievement led to the formulation of the "generalist genes" hypothesis, according to which both kinds of academic abilities share multifactorial genetic and environmental etiologies across levels of performance (Plomin and Kovas, 2005). Comorbidity rates are high between multifactorial developmental dyslexia and dyscalculia (Landerl and Moll, 2010). These data suggest that reading and mathematical abilities may depend on common cognitive mechanisms. Evidence for shared mechanisms is so overwhelming that the DSM-5 Task Force considered abolishing the distinction between specific reading and math learning disorders (Tannock, 2013).

Other evidence points to dissociations between reading and math performance. Pure cases of multifactorial dyslexia and dyscalculia hint at nonshared specific cognitive mechanisms that may characterize distinct entities or subtypes (Haase et al., 2014; Landerl et al., 2009; Rubinsten and Henik, 2006; Tressoldi et al., 2007). Analysis of family recurrence patterns in developmental dyslexia and dyscalculia indicate that besides common genetic factors, also specific ones may be involved. For example, Landerl and Moll (2010) observed cross-condition family recurrence, but recurrence rates were higher for dyslexia in families of reading disabled individuals and higher for dyscalculia in families of math disabled individuals.

Shared and nonshared mechanisms can be identified at the neural level (Ashkenazi et al., 2013). The neural networks involved in word reading and arithmetic learning are only partially overlapping. Learning of word reading has been shown to depend on a neural network consisting, among other areas, of the inferior lateral occipito-temporal cortex, posterior superior temporal cortex and adjacent inferior parietal areas, and lateral inferior prefrontal cortex (Ashkenazi et al., 2013, Dehaene, 2009). Learning of arithmetics is associated to the structural and functional integrity of a frontoparietal network converging on the intraparietal sulcus, but also including inferior lateral occipitotemporal cortex, inferior parietal cortex and hippocampus (Ashkenazi et al., 2013; Kaufmann et al., 2011; Matejko and Ansari, 2015; Moeller et al., 2011).

In a systematic review, Ashkenazi and coworkers (2013) identified both nonshared and shared neural underpinnings of reading and math learning. Word reading learning depends on the structural-functional integrity of the left hemisphere and math learning relies more heavily, but not exclusively, on right hemisphere mechanisms. Different parietal areas are also involved. Reading depends more on the inferior parietal cortex (supramarginal gyrus, angular gyrus) and math on the intraparietal sulcus. Overlapping neural components are situated at the lateral inferior occipitotemporal cortex, inferior parietal cortex (supramarginal gyrus, angular gyrus) and inferior frontal gyrus.

Which are the possible shared and nonshared mechanisms at the cognitive level of description? In order to search for an answer to this question it is important to choose outcome measures shared by reading and arithmetic. Standardized achievement tests are not good indicators because they evaluate abilities at different complexity levels, recruiting semantic and reasoning processes in

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different degrees. We focused then on some relatively low-level abilities of word reading/spelling and basic numerical processing (Arabic number reading and Arabic number writing).

These basic, decontextualized and less semantically loaded abilities have been consistently implicated with reading and math performance both in typical and atypical populations. In the case of reading, for example, it has been assumed that reading at the word level is an important precursor of more advanced mechanisms of reading comprehension (Florit and Cain, 2011; Gough, 1996). The same can be argued for arithmetics: basic knowledge and processing of verbal and Arabic numerals is an important precursor of later arithmetic learning (Moeller et al., 2011), as well as a marker of math learning difficulties (Moura et al., 2013, 2015).

Focusing on the basic abilities at the lexical and numerical processing levels, we also hope to raise the probability of uncovering meaningful patterns of association and dissociation between cognitive variables and reading/spelling- and math-related performances in early school age. According to Ashkenazi and coworkers (2013), there are three main sets of variables which are associated to reading/spelling and arithmetic performance: a) phonological processing, mainly at the phonemic level, and including rapid access to phonological representations (RAN tasks), phonological short-term memory, and phonemic awareness, is consistently associated to word reading learning (Castles and Coltheart, 2004; Melby-Lervag et al., 2012); b) numerical magnitude representations (Halberda et al., 2008, Landerl et al., 2004, Mazzocco et al., 2011; Piazza et al., 2010; Pinheiro-Chagas et al., 2014) or symbolic access to magnitudes (Rousselle and Nöel, 2007, Rubinsten and Henik, 2005, see review in DeSmedt et al., 2013) have been proposed as mechanisms underlying math learning and its difficulties; and c) working memory is relevant for both reading/spelling and math learning (Peng and Fuchs, 2014; Swanson et al.,

2006). The question is whether the verbal and nonverbal aspects of working memory differentially affect reading/spelling and math performance (Peng and Fuchs, 2014).

The picture is, however, complicated by the fact that arithmetic-related abilities are more heterogeneous than reading-related abilities. Research increasingly shows that some aspects of arithmetics and number processing may be dependent on verbal processes, sharing mechanisms with reading learning and dyslexia (DeSmedt and Boets, 2010; Simmons and Singleton, 2008) while others rely on nonsymbolic magnitude processing (Halberda et al., 2008; Landerl et al., 2004; Mazzocco et al., 2011; Piazza et al., 2010; Pinheiro-Chagas et al., 2014). For instance, phonemic awareness has been identified as a predictor of not only reading but also math abilities (Hecht et al, 2001). Foremost, among the numerical abilities dependent on verbal processes are number transcoding (Lopes-Silva et al., 2014), retrieval of arithmetic facts (DeSmedt and Boets, 2010), and word problem solving (Jordan, Hanich and Kaplan, 2003; Powell et al., 2009). Phonological representations have also been included as an important component in some models of numerical transcoding, such as ADAPT (Barrouillet et al., 2004).

Following Ashkenazi and coworkers (2013), we identify intelligence, working memory (both phonological and visuospatial) and phonemic awareness as cognitive dimensions relevant to acquiring early reading/spelling and math abilities. We will discuss the role of these general and specifics abilities in word reading/spelling and numerical transcoding.

Intelligence is an important long-term predictor of school achievement assessed with standardized omnibus tests (Deary et al., 2007). One could ask if intelligence would not only be associated with achievement at the higher and more complex levels of performance. However,

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correlations are higher with intelligence for reading comprehension than for word decoding (Nobre and Salles, 2014) and a large body of literature also implicates intelligence in the acquisition of early visual word decoding skills (Juel et al., 1986; Stanovich et al., 1984, Tunmer and Nesdale, 1985). The association of intelligence and word reading achievement is observed inclusively in the intellectually disabled population (Levy et al., 2002).

The role of intelligence in number reading and writing has been the focus of less research. However, extant data indicate that general cognitive ability is significantly associated with Arabic number reading and dictation in the school-age population (Lopes-Silva et al., 2014, Moura et al., 2013). Probably, intelligence is important for learning words and number reading and writing skills because at the age children are involved with these tasks, this represent quite an accomplishment, a novel and challenging task that mobilizes their best available cognitive resources.

Regarding visuospatial working memory, Zuber and coworkers (2009) verified a specific association between Corsi blocks and a number writing task in 7-year-old German speaking children. As it is known, the verbal numeric notation in the German language is characterized by the inversion between units and decades in two-digit numbers. The authors observed that over 50% of the errors made by the children had a syntactic nature, involving the inversion between units and decades. These results were confirmed by Pixner et al. (2011) in Czech-speaking children, a language that uses both direct and inverted systems for naming two-digit numbers. They showed that Czech children committed more syntactic errors when they wrote numbers in the inverted form and these errors were associated to visuospatial working memory. Furthermore, Moura et al. (2013) observed that the Corsi Blocks task had a moderate correlation with a number

writing task and a weak, but significant, correlation with a number reading task.

The association between visuospatial working memory and reading/spelling is unclear and more studies are required. A recent investigation explored the complete working memory profile of children with poor reading ability (Dawes et al., 2015). Results showed that these children had low performance in the phonological loop and central executive components, but typical abilities in the visuospatial sketchpad. Additionally, a study with school age children with and without reading difficulties revealed no influence of visuospatial working memory on reading development (Cormier and Dea, 1997).

Phonological working memory is the mechanism involved in the temporary storage and manipulation of verbal items (Baddeley, 2007). Concerning the development of mathematical abilities, it has been argued that phonological working memory is recruited in basic skills such as counting (Nöel, 2009) and in calculations based on procedural strategies (Hecht, 2002; Imbo and Vandierendonck, 2007; McKenzie et al., 2003), but not on fact retrieval (Seyler et al., 2003). Furthermore, phonological working memory is also predictive of later mathematics achievement, as discussed above. Phonological working memory skills have also been consistently related to single word reading performance (Alloway et al., 2005; Leather and Henry, 1994; Oakhill and Kyle, 2000). A recent meta-analysis showed persistent deficits in phonological working memory in children with reading disability, independently of chronological age and intelligence (Kudo et al., 2015).

Phonological awareness can be investigated by means of tasks that demand the distinction between the sounds that constitute words, such as rhyme detection and blending isolated sounds to create words (Lewkowicz, 1980). Recently, Cunningham et al. (2015) investigated how the nature of the phonological task and (a) the linguistic nature of the stimuli, (b) the phonological complexity of the stimuli, and (c) the production of a verbal response, can influence the relationship between the task and reading. They have argued that the production of a verbal response is important for the task to be a good predictor of decoding. In this study, we will use a phoneme elision task, in which the child is required to say what a word would be after deleting a certain phoneme and, furthermore, the child should verbally emit the response.

A question that demands further discussion lies in the fact that the phonological complexity of the stimulus might be a confounding factor to mask the association to reading (Cunningham et al., 2015). One important hypothesis is that phonological working memory may act as a mediator between phonological awareness and reading performance, as measures of phonological awareness generally involve phonological working memory resources (Dufva et al., 2001). In the specific case of phoneme elision, there is the need of a conscious access to phonological representations and this may lead to special demands on access mechanisms (Ramus and Szenkovits, 2008). Furthermore, the importance of phonemic awareness in reading/spelling and math skills might be overrated due to phonological working memory influences which would then support the hypothesis of an access deficit in dyslexia (Boets et al., 2013). It is important then to simultaneously investigate the influence of both variables to have more information on the specific correlates of each of them.

Children with developmental dyslexia, who perform poorly on phonological processing tasks, such as phonemic awareness, frequently exhibit deficits in mathematics. According to the weak phonological representation hypothesis (Simmons and Singleton, 2008), phonological processing

deficits would impair aspects of mathematical cognition that involve the manipulation of verbal codes, but do not impair those that are not verbally coded, such as approximate addition and nonsymbolic magnitude comparison. Nevertheless, only few studies (see: Lopes-Silva et al, 2014; Michalczyk et al, 2013) have investigated the association between phonological processing and number transcoding.

The aim of the present study is to investigate shared and specific mechanisms involved in reading and writing words and Arabic numerals in school-aged children. Our main goal was to disentangle the role of phonemic awareness and its impact on lexical and numerical tasks controlling other cognitive variables which may have an important impact. We hypothesize that even after controlling for the influence of intelligence and broader reading and writing skills, phonemic awareness would be an important predictor since all of these tasks involve some sort of verbal processing. In order to investigate that, we conducted a series of hierarchical regression models using scores of reading and writing of single words and Arabic numbers tasks, as dependent variables. As predictor variables we investigated intelligence, the phonological and visuospatial components of working memory and phonemic awareness skills.

Material and Methods

The study was approved by the local research ethics committee (COEP–UFMG). Children participated only after informed consent was obtained. It was obtained in written form from parents and orally from children.

Participants

We have assessed 207 children from the 2nd to 4th grades of public schools from Belo

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Horizonte, Brazil. Data collection took place in the participants' schools. We excluded 1 child who did not complete the entire battery, 4 children due to low intelligence (performance on Raven's Colored Progressive Matrices below one standard deviation from the mean), and 30 children were excluded from further analyses because either they had a poor R^2 on the fitting procedure to calculate their internal Weber fraction on the non-symbolic comparison task or their Weber Fraction exceeded the limit of discriminability of our task (w > 0.6). The final sample was constituted by 172 children with ages ranging from 7 to 11 years (mean = 8.86[0.96] years), 55.2% girls.

Instruments

At first, the intelligence (Raven's CPM), word spelling (Brazilian School Achievement Test -TDE) and number transcoding (Number writing task) were evaluated in small groups of approximately 9 children. Subsequently, we tested number transcoding (Number reading task), word reading (reading subtest of Brazilian School Achievement Test - TDE), phonemic awareness (Phoneme Elision) and working memory (Corsi Blocks and Digit Span).

1. Raven's Colored Progressive Matrices: General intelligence was assessed with the ageappropriate Brazilian validated version of Raven's Colored Matrices (Angelini et al., 1999).

2. Brazilian School Achievement Test: The Teste de Desempenho Escolar (TDE, Stein, 1994) is the most widely used standardized test of school achievement in Brazil. The TDE comprises three subtests: arithmetics, single-word spelling and single-word reading. Norms are provided for school-aged children between the first and sixth grades. The arithmetic subtest is composed of three simple orally presented word problems and 35 written arithmetic calculations of increasing complexity. The spelling subtest consists of dictation of 34 words of increasing

syllabic complexity. The single-word reading subtest of the TDE consists of 70 stimuli, which must be read aloud by the individual participant. Reliability coefficients (Cronbach α) are 0.87 or higher. Children are instructed to work as hard as they can, without time limits. In this study we used the data only of reading and spelling subtests.

3. Arabic number writing: To evaluate number transcoding, children were instructed to write the Arabic forms of dictated numbers. This task consists of 40 items, up to 4 digits (3 one-digit numbers, 9 two-digit numbers, 10 three-digit numbers and 18 four-digit numbers). The one- and two- digit numbers were classified as "lexical items" (12 items), and the other 28 items require the use of algorithm-based rules in order to be written (Barrouillet et al., 2004; Camos, 2008). This task has been used in previous studies with a comparable sample, and the consistency of this task was KR-20 = 0.96 (Moura et al., 2013; Lopes-Silva et al., 2014; Moura et al., 2015).

4. Arabic number reading - A total of 28 Arabic numbers with one to four digits were printed in a booklet and presented to the children one at a time. The children were instructed to read them aloud. The three- and four-digit numbers were grouped into three categories according to their complexity, indexed by the number of transcoding rules established by the ADAPT model. The three and four-digit numbers were chosen to avoid presenting numbers with very strong lexical entries and to maintain the focus on syntactic complexity. This task has been used in previous studies, and the consistency of this task was KR-20 = 0.92 (Moura et al., 2013).

5. Phoneme Elision - This is a widely accepted measure of phonemic awareness (Wagner and Torgesen, 1987; Castles and Coltheart, 2004; Melby-Lervå et al., 2012). The child hears a word and must say what the word would be if a specified phoneme in the word were to be deleted (e.g., "filha" without /f/ is "ilha" [in English, it would be similar to "cup" without /k/ is "up"). The test comprises 28 items: in 8 items, the child must delete a vowel, and in the other 20, a consonant. The consonants to be suppressed varied by place and manner of articulation. The

phoneme to be suppressed could be in different positions within the words, which ranged from 2 to 3 syllables. This task has been used in previous studies with a comparable sample (Lopes-Silva et al., 2014), and the internal consistency of the task is 0.92 (KR-20 formula).

6. Corsi Blocks - This test is a measure of the visuospatial component of working memory. It is constituted by a set of nine blocks which are tapped, in a certain sequence, by the examiner. The test starts with sequences of two blocks and can reach a maximum of nine blocks. We used the forward and backward orders according to Kessels et al. (2000). In the forward condition, the child is instructed to tap the blocks on the same order as the examiner, in the backward condition, in the inverse order. We evaluated the total score (correct trials x span) in both the backward order.

7. Digit Span - Verbal short-term memory was assessed with the Brazilian WISC-III Digits subtest (Figueiredo, 2002). Performance in the forward order was considered a measure of phonological short-term memory, and the backward order was used to assess verbal working memory. We also evaluated the total scores of the backward order.

Results

Raw scores were standardized according to school grades using z scores. By doing so, we aimed at controlling for any possible educational influence on children's performance. At first, we investigated the association between the tasks using Pearson's correlations. Afterwards, we performed four hierarchical regression models with each of these reading and writing skills as dependent variables to investigate which cognitive abilities would predict their performance. In order to control for the shared variance among numerical and verbal tasks, whenever one of these tasks was set as the dependent variable, the other was inserted in the first step of the regression model, using the enter method (i.e.: when number reading task was the dependent variable, word reading was included as an independent variable in the first step of the model). In the second step, phonological and visuospatial working memory and phonemic awareness were included as predictors. The stepwise method was used in this second step to avoid redundancy and to guarantee a high degree of parsimony. We then included intelligence in the first step to investigate whether the influence of the other cognitive variables would continue to be significant.

Associations between cognitive variables and reading and writing processes

To explore the general pattern of association between the cognitive variables and reading and writing of numbers and words, we investigated the correlations between them (Table 1).

As can be seen in the table below, all of the writing and reading tasks (TDE word reading and spelling, as well as number reading and number writing) were significantly correlated (all r's > 0.55, p < 0.001). Word reading and word spelling were highly correlated to each other, as well as number reading and number writing. Intelligence also presented correlations between 0.30 and 0.40 with all of the variables. Both phonological processing tasks correlated with the verbal and numerical ones. Correlations for phoneme elision were in the 0.37 to 0.69 range, and correlations for digit span were in the 0.27 to 0.39 range. Corsi Blocks did not present significant correlations to the word reading task.

	Word Writing	Word Reading	Number Writing	Number Reading	Phoneme Elision	Digit Span	Corsi Blocks
Raven	0.449**	0.313**	0.401**	0.411**	0.371**	0.359**	0.329**

Table 1: Correlations between the neuropsychological measures

Word Writing	1	0.719^{**}	0.593**	0.562**	0.557**	0.317**	0.155*
Word Reading		1	0.503^{**}	0.598**	0.693**	0.395**	0.149
Number Writing			1	0.762**	0.483**	0.271**	0.244**
Number Reading				1	0.602**	0.287**	0.238**
Phoneme Elision					1	0.366**	0.253**
Digit Span						1	0.354**
Corsi Blocks							1

**. Correlation is significant at the 0.01 level (2-tailed).

*. Correlation is significant at the 0.05 level (2-tailed).

Specific predictors of reading and writing skills

To further explore the association between these variables and to have a more fined-grained perspective regarding the predictive power of them on each of the verbal and numerical skills, we conducted separate hierarchical regression models.

We calculated regression models including intelligence and the analogous variables in the first step and the other cognitive variables in the second one. By doing so, we aimed at investigating the pattern of association between these variables, once we had controlled for more general cognitive skills. Variance for each kind of number task (e.g.,reading or writing) was predicted by the homologous tasks with words (e.g.,reading or writing) and vice-versa(r^2 ranging from 0.36 to 0.41)and by phoneme elision (r^2 ranging from 0.03 to 0.17). As can be seen in Tables 2–5, phoneme elision was the only task that was a significant predictor in the four hierarchical models. It is important to note that phonological WM was associated to word reading, but its influence was much smaller compared to phonemic awareness.

Table 2: Regression analysis for Number reading (adjusted $r^2 = 0.449$).

Predictor	Beta	Partial t	Sig	r ² change
Intercept		-2.387	0.018	
Raven	0.196	3.194	0.002	

Word Reading	0.327	4.142	< 0.001	0.413
Digit Span	-0.03	-0.469	0.64	Excluded
Corsi Blocks	0.055	0.904	0.367	Excluded
Phoneme elision	0.303	3.747	< 0.001	0.045

Table 3: Regression analysis for Word reading (adjusted $r^2 = 0.539$).

Predictor	Beta	Partial t	Sig	r ² change
Intercept		0.441	0.660	
Raven	-0.034	-0.575	0.566	
Number reading	0.279	4.143	< 0.001	0.363
Digit Span	0.151	2.614	0.010	0.018
Corsi Blocks	-0.099	-1.746	0.083	Excluded
Phoneme elision	0.482	7.136	< 0.001	0.168

Table 4: Regression analysis for Number writing (adjusted $r^2 = 0.390$).

Predictor	Beta	Partial t	Sig	r ² change
Intercept		-5.357	< 0.001	
Raven	0.138	2.042	0.043	
Word Spelling	0.421	5.557	< 0.001	0.374
Digit Span	0.019	0.291	0.804	Excluded
Corsi Blocks	0.096	1.509	0.867	Excluded
Phoneme elision	0.197	2.71	0.007	0.026

Table 5: Regression analysis for Word Spelling (adjusted $r^2 = 0.465$).

Predictor	Beta	Partial t	Sig	r ² change
Intercept		4.421	< 0.001	
Raven	0.187	2.984	0.003	
Number writing	0.369	5.557	< 0.001	0.405
Digit Span	0.046	0.737	0.462	Excluded
Corsi Blocks	-0.087	-1.454	0.148	Excluded
Phoneme elision	0.309	4.721	< 0.001	0.070

Discussion

In the present study, we investigated the cognitive variables that underlie the performance of reading and writing skills for both numbers and words in a sample of Brazilian school-aged children. Our results can be summarized into two mains topics: the specific influence of phonemic awareness on reading and writing words and numbers and the impact of non-verbal intelligence on them. We have found a prominent role of phonemic awareness which was consistently associated to all the reading and writing modalities we have assessed. As far as we know, this is the first study to simultaneously investigate these four skills and the cognitive variables related to them.

Phonemic awareness as a common mechanism shared between reading and writing of both numbers and words

Phonemic awareness is an important underlying factor of reading acquisition (Ehri et al., 2001) and deficits in it are associated to reading disabilities and dyslexia (Lyon et al., 2003). A puzzling aspect of this association lies in the fact that there is a reciprocal relationship between

phonological processing and reading skills: when children begin to read, their reading skills become the best predictor of their own reading development (Bell et al., 2003). In this study, we aimed at controlling this confounding variable to be able to analyze the interplay between numbers and words. To do so we investigated the influence of cognitive variables in word reading, for example, by controlling for the impact of number reading.We have found a consistent role of phonemic awareness in both number and word reading and writing, after controlling for other cognitive variables. Even though the relation of phonemic awareness and word reading skills is well documented in the literature (Vellutino et al., 2004), the association between phonemic awareness and word spelling is less robust. Nevertheless, the interaction between phonological processing and other cognitive skills, such as syntactic awareness and naming-speed, are taken as evidence in favor of an integrative hypothesis (Plaza and Cohen, 2003). In our study, we have found that phonological WM was an important predictor of single word reading but not spelling, A possible reason for that lies in the fact that in the spelling subtest of Brazilian School Achievement Test, children hear the word three times: first the examiner dictates only the word itself, afterward inside a sentence, and then the isolated word again. Since the child hears the word so many times, other variables that are associated to spelling skills, such as orthographic rules, may play a more important role and this should be investigated in further studies. Despite this, reading and spelling are highly correlated [0.71 in our study and from 0.77 to 0.86 according to Ehri (1997)] and both have phonemic awareness as a common underlying mechanism. Regarding number transcoding and phonological processing skills, there is even less investigation. Even though mathematical and reading/spelling disabilities have a high comorbidity rate (Landerl and Moll, 2010) there are not so many studies that deeply investigate what is shared in this most basic level: single word and number writing and reading. One plausible argument to explain the comorbidity is the weak phonological representation hypothesis

which explicitly states that any aspect of numerical cognition that is associated to a verbal code would be impaired in dyslexics, since they would have fuzzier phonological representations. According to the ADAPT model of number writing, one of the first steps in the verbal to Arabic number transcoding is phonological encoding (Barrouillet et al., 2004). Phonemic awareness might be a distal source of influence on this phonological step in the model. Regarding number reading, the picture is less clear: the procedural steps between Arabic to verbal number transcoding have been described in terms of intermediary semantic representations which can have a verbal-linguistic component (Power and Dal Martello, 1990). Nevertheless, cognitive models of number reading do not usually take linguistic processes into consideration. One can assume, however, that phonemic awareness might also be an important linguistic mechanism associated to this modality of transcoding. The association between spelling and word reading to mathematics disabilities depends on the cutoff criteria used to define them. Most studies have investigate associations between word reading and arithmetic, especially in dyslexic samples (Boets and De Smedt, 2010; Göbel and Snowling, 2010). Landerl and Moll (2010) investigated the comorbidity rates between Spelling Disabilities (SD); Reading Disabilities (RD), and Arithmetic Disabilities (AD) in a large population- based sample of elementary school children. They reported an interesting finding: the rate of comorbidity between AD and RD decreases when children are defined based on a more stringent criteria, whereas the rate between arithmetics and spelling remains constant. The interplay between reading and writing of numbers and words is rather complex and one should always have in mind that it depends on the measures and criteria used. It is also important to emphasize that the power of phonemic awareness as an indicative of literacy skills changes according to grade (Moll and Landerl, 2009): our results should be cautiously interpreted and circumscribed to Portuguese-speakers who are on the second to fourth grades.

The contribution of Intelligence to Reading and Writing

The use of intelligence as a covariate in studies of children with learning disabilities has been criticized (Dennis et al., 2009). According to these authors, the high correlation of IQ and school performance is underspecified, as intelligence is both a predictor and an outcome variable. The dual role of IQ and its correlation with school achievement increases the risk of statistical distortions caused by regression to the mean. According to this line of reasoning, including IQ-related measures as covariates is not only irrelevant but also an improper conduct.

Another line of argumentation is also defensible. Estimates of general cognitive ability have been repeatedly found to be among the best predictors of school achievement and other psychosocial outcomes (Strenze, 2007; Deary and Johnson, 2010). According to this perspective, not all IQ-related measures are equally influenced by educational experience and socioeconomic status. Some measures, such as vocabulary, are heavily dependent on educational and reading experience, while others such as the Raven's CPM are more related to fluid general intelligence (Carpenter et al., 1990) and are less dependent on educational experience. The fluid Intelligence (Raven's CPM) is closely related with WM in childhood and this relation is primarily explained by the executive component of WM (Sbicigo et al., 2014).

Mastery of word and number processing at the lexical level is an enormous task for children at early school age. There is evidence, for example, that mastery over Arabic number dictation is reached only after 3–4 years of schooling in typically developing children (Moura et al.,2013, 2015). Associations between general cognitive ability and reading are higher for reading comprehension ($r^2 = 0.44$) than for word decoding ($r^2 = 0.05$, Shatil and Share,2003). But general cognitive ability is not irrelevant for reading at the word level. This is corroborated by higher prefrontal activation levels in beginning readers than in older, more proficient, children and adults (Schlaggaretal.,2002; Turkeltaubetal., 2003). According to Mayes et al.(2008), IQ tests can predict academic achievement, but its predictive power is increased when phonological WM and visuo-motor integration are also included.

Early school abilities related to word and number reading and writing may depend on both domain-general and domain-specific cognitive abilities. Sources of influence are both shared and non-shared across codes and tasks. Phonological and visuospatial WM tasks could be uniquely associated, respectively, to verbal lexical and numerical tasks. But these effects disappear or are attenuated when general non-verbal intelligence is covaried (the zero-order correlation between number writing and digit span is r =0.271; p<0.001 and the partial correlation controlling for intelligence decreases to r = 0.149; p = 0.052). Phonemic awareness seems to represent a shared source of variance, common to both kinds of codes and tasks, exerting effects over and above general intelligence.

Conclusion and implications

These findings suggest that phonemic awareness may be considered as a domain-specific cognitive mechanism which is strongly associated to reading and writing of both numbers and words. From these results, one can infer that phonemic awareness is a mechanism shared by numerical and verbal domains, which might also be a candidate associated to the high comorbidity rate between dyslexia and dyscalculia. It is important to note that this specific mechanism is related to number and word reading and writing and should, therefore, be taken

into account in intervention models.

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CHAPTER 5

General Discussion

Discussion

The present dissertation aimed at investigating the cognitive underpinnings of number transcoding in Brazilian school-aged children and how phonological processing could be associated with it. This chapter will summarize our main findings and discuss limitations and next steps.

In study 1, we report the psychometric properties of a Verbal to Arabic number writing task. We showed that this is a rather simple and fast task to administer, yet it is a powerful tool to screen for mathematics difficulties in early elementary school. In our opinion, this is an important contribution to the literature on numerical cognition, since we lack instruments to assess mathematical skills, both in research and clinical contexts. This study also provides the set of items with their adequate psychometric properties, as well as normatives parameters, that can be easily administered to groups of pupils by their teachers.

Afterwards, in the next two chapters, we have described two studies that investigated the specific association of phonemic awareness and number transcoding. We concluded that this phonological skill might mediate the influence of working memory in number writing and could be a shared mechanism among measures of math and reading/spelling.

Our findings complement previous research on number transcoding in children, which is relatively scarce. At first glance, one could claim that number transcoding is an easy and intuitive skill that do not impose a great source of difficulty to children; and that this could be the reason why it has not been widely investigated. Nevertheless, as mentioned above, this can be one of the first signs presented by children with mathematical learning disabilities. An important point that deserves deeper investigation is the fact that linguistic features also impact on number transcoding, since there are different numerical systems, and findings from different languages may not be generalized. For example, two digit-numbers transcoding may be especially hard for French-speaking children ("quatre-vingtet un"/for-twenty on = "81") or to other languages like German ("Einundachtzig" one and eighty = "81") characterized by decade-unit inversions. Our findings show that, even though the Portuguese numerical system is quite regular, oral verbal to Arabic transcoding may be difficult for children, especially for the ones with mathematical learning difficulties and should be taken into account in neuropsychological assessments.

More especially, we provide additional evidence regarding the association between phonological processing and number transcoding, since most of previous studies have focused on the impact of phonological skills in other verbal aspects of mathematics, such as arithmetical facts or oral problems (see Simmons & Singleton, 2008, 2009; Boets & DeSmedt, 2010). In studies 2 and 3, we conclude that Verbal to Arabic number transcoding can also be influenced by children's phonological skills, especially by phonemic awareness. Our findings are in line with the weak phonological representation hypothesis (Simmons & Singleton, 2008), according to which phonological processing deficits impair aspects of mathematics that rely on the manipulation of verbal codes (e.g. counting speed, number fact recall), whilst other aspects of mathematics that are less reliant on verbal codes (e.g. estimation, subtizing) are unimpaired. We expand this impact of poor phonological representations to number transcoding. Future studies should explore this impact in further detail: in the context of neuropsychology, it would be quite informative to explore this at the neural level by means of the investigation of possible shared activity in the left temporo-parietal areas and angular gyrus on both phonological and numerical tasks.

The specific role of phonemic awareness in number writing is still controversial. It is possible to raise the question that the ability to perceive and manipulate phonemes should not be relevant to number transcoding. A hypothesis that support this association is that phonemic awareness could be a distal index of the quality of children's phonological representations, a skill that affects all sorts of verbal tasks, including the ones associated with numerical processing (Simmons & Singleton, 2008). Nevertheless, future studies should also include the other subcomponents of phonological processing to investigate possible specific influences. Lexical access, for instance, is quite relevant for both arithmetic facts and reading fluency. According to Geary (1993), deficits in representing and retrieving semantic information from long term memory could be related to the comorbidity between dyslexia and dyscalculia. Regarding number transcoding, deficits in lexical access could, for example, lead children to commit lexical errors, due to the wrong retrieval of the corresponding Arabic form from a certain spoken number. In every language, there are some numbers which are phonologically similar and strategies are created to orally differentiate between them. An interesting example in German is "zwei" (two) and "drei" (three). When dictating a phone number, for instance, people could say "zwo" for "zwei", to avoid mistakes. The same is observed in Portuguese, for "três" (three) and "seis" (six). Frequently, people would say "meia" instead of "seis" to prevent misunderstandings. (In appendix 2, we report a case of a child with phonological processing deficits associated with this type of error). Moreover, it is clear that the precise comprehension of the phonological structure of the verbal number is crucial for retrieving its correct corresponding lexical Arabic form. Phonological awareness could, therefore, be strongly associated with lexical retrieval.

Regarding phonological short-term and working memory, its role on numerical cognition is

widely investigated. According to Ramussen & Bisanz (2005), the phonological buffer correlates to many sorts of arithmetic problems (both verbal and nonverbal, with or without contextirrelevant information). On the field of numerical transcoding, many studies (Barrouillet et al., 2004; Camos, 2005; Zuber et al., 2009; Moura et al., 2013), emphasize the relevance of phonological short term and working memory for Verbal to Arabic number transcoding.

According to Swanson (2004), although developmental increases in WM can co-occur in phonological processing (e.g. phoneme elision), these two processes operate independently. Swanson (2004) argues that WM contributes to arithmetic problem solving beyond phonological processing in children between 8 and 11 years old. Our results show a slightly different pattern. Even after partialling out the influence of phonological working memory and approximate number system, the influence of phonemic awareness in number transcoding remained significant. One possible explanation for these deviant results is that the phoneme elision task of Swanson (2004) might have been too easy for the children because they had to delete the beginning and final phoneme, whereas, in our task, children also had to delete phonemes in the middle of the words. Hence, the task used by this other study could be insufficiently sensitive and specific to identify phonological deficits. It is important to note that these authors were not specifically investigating number transcoding. Children needed to solve arithmetic problems, which were quite complex, in the sense that they had extraneous information [an example extracted from her paper: "Darren found 15 pinecones (assignment). He threw 5 pines cones back (assignment). Darren uses pine cones to make ornaments (extraneous). How many pine cones did Darren keep (question)?"]. These problems may involve other executive functions, such as inhibition of irrelevant information or monitoring components of WM (manipulation and recombination of material).

In order to disentangle the specific role of phonological working memory and phonemic awareness in number writing, it would be interesting to have a phonemic awareness task that does not require so much the retention and manipulation of phonological information. This is quite a challenge, since elementary school children who are learning to write multi-digit numbers tend to have ceiling performance on more simple phonological awareness tasks.

Another point that deserves further discussion is the quarrel between semantic and asemantic models of number transcoding. Damian (2004) establishes a parallel regarding the semantic access proposed by reading cognitive models and number transcoding. Regarding reading models, it is accepted that the conversion between the written word and its phonological form can happen without access to its meaning (Seindenberg & McClelland, 1989). Analogically, it is possible to infer that the conversion between the verbal-oral format of a number does not require access to the magnitude represented by it. The ADAPT model (Barrouillet et al 2004) supports the hypothesis that the conversion between these two codes is not mediated by the numerical semantics In general, the model postulated the existence of an encoding process of the verbal-oral input into a phonological form and the subsequent analysis of its segments by means of a production system. This production system is based on condition-action procedures. Nevertheless, the question if there is semantic access in Verbal to Arabic number transcoding remains unanswered, since the investigation of this issue can be methodologically challenging. There has not been a consensus so far regarding the nature of this possible semantic activation and how to operationalize this measure. In Study 2, we have taken the Weber Fraction as an index of the acuity of the approximate number system, and it was not associated with number transcoding. However, it is possible to argue that this is not a proper way to measure the semantic activation in number writing.

An argument used as an evidence for the asemantic models is that the errors patterns found for 2and 3-digit numbers change whether they are dictated in isolation or they are part of a larger number (ex: 400 and 3400). The Semantic models predict that the type of errors in transcoding numbers should remain unchanged when these numbers are either dictated in isolation or part of a larger number whereas according to the ADAPT model, there should be different errors when numbers are presented in isolation or in larger numbers (Barrouillet et al., 2004). Future studies should analyze if there will be differences in latency and accuracy between lexically retrieved units when they are separately written or inside larger numbers, as a measure of semantic influence on number writing.

Finally, future studies should also focus on more fine-grained measures of number transcoding. Just like in reading research, on which the importance of assessing fluency is extensively debated (see Wimmer, 2006), studies on number writing should also consider the transcoding speed. Children could differ regarding the lexical access or present a speed/accuracy trade-off. Most studies on number transcoding report exclusively accuracy and this outcome measure may mask difficulties children could have regarding the automatization and fluency in number writing. An interesting strategy would be to investigate temporal patterns of transcoding for Arabic numerals, which could provide information regarding retrieval and procedural strategies in number writing. Van Loosbroek, Dirkx, Hulstijn & Janssen (2009) were pioneers in investigating a sample of children with arithmetical disabilities and controls in a computerized number writing task, from which they derived a measure of planning time. The authors concluded that children with arithmetical disabilities were slower in planning the writing of Arabic digits. Interestingly, they

also assessed reading and vocabulary measures and children with arithmetical disabilities were also worse on these tests. It would be interesting to investigate if these differences in phonological skills could be associated with number writing fluency.

At last, our studies focused on typically achieving children. Investigating a sample of dyslexic children, who have deficits in phonological processing could be quite informative regarding the role of phonemic awareness in number transcoding. Most studies of numerical cognition in dyslexia have focused on arithmetic fact fluency (Geary, Hamson, & Hoard, 2000; Simmons & Singleton, 2008; Simmons & Singleton, 2009; Boets & De Smedt, 2010). The verbal aspects of mathematics, such as counting and exact arithmetic are specifically prone to being impacted by the poor quality of children's phonological representations (Simmons & Singleton, 2008). Number writing clearly depends on the phonological encoding of the numbers and studies with dyslexic samples should also deepen the research regarding their transcoding skills.

Regarding clinical purposes, this dissertation demonstrates the importance of assessing number transcoding in the screening of children with mathematics learning disabilities. Our results provides evidence to build theories related to education practices of mathematical development. It is clear that teaching the basic of symbolic representations in mathematics is extremely relevant for learning more complex tasks, such as calculation. Taken together, the studies in this dissertation provide further evidence regarding the importance of research on basic number processing and educational neuroscience related to mathematics learning. Besides that, we also provide evidence regarding the relevance of investigating phonological skills while assessing arithmetical achievement. More specifically, this dissertation contributes to future models of the development of arithmetic processing and its relationship to phonological processing. Practitioners should also train phonological skills in dyscalculia neuropsychological rehabilitation as well as in school activities, since improving this could result in better reading and math performance.

Next steps

The research agenda on linguistic influences on number transcoding offers a wide range of possible approaches. I personally aim at continuing to explore how language can affect number transcoding but, this time, exploring linguistic differences.

Bahnmueller, Huber, Nuerk, Göbel & Moeller (2015) suggested a possible association between number processing and orthographic processing strategies in different languages. Eye-tracking analyses indicated that German-speaking adults had a higher probability of using a sequential processing strategy and English-speaking participants a parallel processing strategy. These differences may be associated with the distinct patterns of phonemic-graphemic correlations in the orthography of the two languages (Landerl, Wimmer, & Frith, 1997), as well as with the units-decades inversion characteristic of the German language. The German orthography is considerably more regular than English is. German-speakers may thus reliably resort to a sequential phonological-orthographic processing of stimuli, which is required by the unitsdecades inversion. English-speakers more frequently use a parallel processing strategy. Given the characteristics of English orthography, sequential processing is both less efficient and less required, as there is no units-decades inversion in this language. However, Bahnmueller et al. (2015) did not actually measured reading behavior of their participants. Therefore, I aim at picking up on this research and evaluating the proposed influences of differences in reading behavior on multi-digit number processing by assessing Brazilian and German participants on both number processing and reading behavior for the first time.

Concluding remarks

In this chapter, we have outlined how our results aid to provide a more complete picture of the number transcoding importance to mathematics performance. This dissertation provides evidence that support the relevance of investigating transcoding in the context of mathematics learning disabilities. We also demonstrate how number transcoding can be investigated from many different angles and approaches and emphasize the need of more research on this extremely relevant topic of numerical cognition.

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APPENDIX 1

Stable measures of number sense accuracy in math learning disability: Is it time to proceed from basic science to clinical application?

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ABSTRACT

Math learning disability (MLD) or developmental dyscalculia is a highly prevalent and persistent difficulty in learning arithmetic that may be explained by different cognitive mechanisms. The accuracy of the number sense has been implicated by some evidence as a core deficit in MLD. However, research on this topic has been mainly conducted in demographically selected samples, using arbitrary cut-off scores to characterize MLD. The clinical relevance of the association between number sense and MLD remains to be investigated. In this study, we aimed at assessing the stability of a number sense accuracy measure (w) across five experimental sessions, in two clinically defined cases of MLD. Stable measures of number sense accuracy estimate are required to clinically characterize subtypes of MLD and to make theoretical inferences regarding the underlying cognitive mechanisms. G. A. was a 10-year-old boy with MLD in the context of dyslexia and phonological processing impairment and his performance remained steadily in the typical scores range. The performance of H. V., a 9-year-old girl with MLD associated with number sense inaccuracy, remained consistently impaired across measurements, with a nonsignificant tendency to worsen. Qualitatively, H. V.'s performance was also characterized by greater variability across sessions. Concomitant clinical observations suggested that H. V.'s difficulties could be aggravated by developing symptoms of mathematics anxiety. Results in these two cases are in line with the hypotheses that at least two reliable patterns of cognitive impairment may underlie math learning difficulties in MLD, one related to number sense inaccuracy and the other to phonological processing impairment. Additionally, it indicates the need for more translational research in order to examine the usefulness and validity of theoretical advances in numerical cognition to the clinical neuropsychological practice with MLD

Keywords: developmental dyscalculia, mathematical learning disabilities, number sense,

single-case study, Weber fraction

In this article, we approach the question of the cognitive underpinnings of math learning disabilities (MLD) from a clinical, idiographic perspective. Basic research has proposed that number sense, or the accuracy in approximately and nonverbally discriminating the numerical magnitudes of sets of objects, may be causally linked to math performance, both in typical and atypical populations (Halberda, Mazzocco, & Feigenson, 2008; Mazzocco, Feigenson, & Halberda, 2011; Piazza et al., 2010; Pinheiro-Chagas et al., 2014). If the hypothesis of a number sense representational deficit in developmental dyscalculia is valid, then at least some children with math learning difficulties should consistently exhibit impairments in this ability, and number sense accuracy should be reliably measurable and predictive of their patterns of numerical processing and arithmetic difficulties.

Mathematical learning disability or developmental dyscalculia is a specific, severe, persistent disability in learning arithmetic, not primarily attributable to factors such as low intelligence, gross neurosensory impairment, emotional disorders or lack of proper education and opportunity (American Psychiatric Association, 2000, 2013). MLD constitutes a heterogeneous group from the neurocognitive point of view (Wilson & Dehaene, 2007).

Subtyping of MLD or dyscalculia dates back to Kosc (1974) and Geary (1993), and it is based on the underlying cognitive dysfunctions. We refer to a proposal made by Wilson and Dehaene (2007). These authors distinguish four subtypes of MLD. The first subtype is caused by a deficit in number sense, either degraded representations of approximate numerosities (Landerl, Bevan, & Butterworth, 2004) or a deficit in accessing approximate quantities from symbolic representations (Rousselle & Noël, 2007). The second subtype depends on dysfunctional verbal mechanisms, mainly phonological processing (phonological lexical access, phonological shortterm memory and phonemic awareness; De Smedt, Taylor, Archibald, & Ansari, 2010; Lopes-Silva, Moura, Júlio-Costa, Haase, & Wood, 2014). A deficit in executive functioning and/or working memory underlies the third subtype (Raghubar, Barnes, & Hecht, 2010; Van de Weijer-Bergsma, Kroesbergen, & Van Luit, 2015). A fourth, more tentative subtype, is also proposed with underlying spatial-attentional deficits (Venneri, Cornoldi, & Garuti, 2003; Verdine et al., 2014). Besides these, motivational and emotional factors are associated with math performance (Haase et al., 2012).

Subtype identification is based on the presence of associated deficits (Wilson & Dehaene, 2007). Cases of impairments in number sense processing without other accompanying impairments are classified as pure MLD or developmental dyscalculia. Cases without impairments in number sense but with deficits in phonological processing are classified under the verbal subtype. This could correspond to the math impairment observed in developmental dyslexia. There may be comorbidities when one individual exhibits impairments in more than one mechanism (Landerl & Moll, 2010).

Number sense is the ability to approximately represent and manipulate magnitudes (Dehaene, 2007). One way to operationalize number sense is to assess the ability to rapidly and approximately discriminate differences in the numerical magnitude between two sets of objects (Piazza et al., 2010; Pica, Lemer, Izard, & Dehaene, 2004). The accuracy in discriminating magnitudes can be measured by the internal Weber fraction, from now on referred to as "w" (Dehaene, 2007).

A lower w-value means that the individual needs smaller differences between dot sets to discriminate their magnitude without counting, so the higher the w-value, the lower the precision (see Halberda, 2006). The scaled inverse relationship between discriminability and magnitude difference is known as the distance effect (Dehaene, 2007). In addition, this same difference between the magnitudes is inversely proportional to the individual reaction time to choose which is the largest set of dots (Izard & Dehaene, 2008). This psychophysical effect is known as Weber's Law (Halberda, 2006). The smaller the numerical distance between two sets, the greater the reaction time and the higher the error rate. This relationship is characterized by a constant, w, which provides an estimate of the minimally discriminable numerical difference and the resolving power of the underlying system or accuracy of the number sense (Dehaene, 2007). Some evidence indicates that the number sense improves throughout life (Piazza & Izard, 2009) and is related to typical and atypical math achievement (Costa et al., 2011; Halberda et al., 2008; Mazzocco et al., 2011; Pinheiro- Chagas et al., 2014). However, not all authors have been able to replicate findings of an association between approximate number system (ANS) accuracy and math performance (Chen & Li, 2014; De Smedt, Noël, Gilmore, & Ansari, 2013; Fazio, Bailey, Thompson, & Siegler, 2014).

Knowledge on the connection between number sense and arithmetic learning has mainly been gained in the context of basic research, especially through cross-sectional demographic screening of participants with arbitrary cut-off scores (e.g., Mazzocco et al., 2011, Piazza et al., 2010, Pinheiro-Chagas et al., 2014). Although theoretically relevant, the clinical significance of the association between number sense accuracy and math performance is complex and subject to considerable theoretical and empirical dispute (see meta-analyses in Chen & Li, 2014; Fazio et

al., 2014). More detailed, clinical analyses concerning the stability of number sense accuracy measures in single cases and its relevance to the cognitive deficits underlying MLD are missing.

To elucidate a possible causal role of number sense accuracy in MLD, its estimates should be reliable across measurements. Toward this end, we conducted repeated measures of number sense accuracy in previously reported cases of two children with different MLD profiles, one associated with phonological impairment and dyslexia and the other with number sense deficit (Haase et al., 2014). Our predictions are that, if number sense accuracy is not involved with math difficulties in dyslexia, its estimates should reliably remain in the typical range across measurements. Otherwise, if number sense accuracy is to play a role in some cases of dyscalculia, its estimates should remain reliably impaired across measurements.

Methods

Participants

The two cases investigated here have been reported in a previous study, which more deeply discusses their cognitive profiles (Haase et al., 2014). At the time of assessment, H. V. was a 9-year-old girl and G. A. was a 10-year-old boy. They both came to our clinical attention because of persistent math learning difficulties.

H. V. was diagnosed with developmental dyscalculia. Her difficulties in math were restricted to deficits in number sense and arithmetic facts retrieval. H. V.'s cognitive profile was high intelligence (percentile in Raven's Colored Progressive Matrices = 99), excellent performance in phonological processing, visuospatial, and executive functions tasks. Moreover, H. V. scored below the mean in the nonsymbolic magnitude comparison task and in arithmetic problems.

Clinically, H.V. had persistent difficulties in learning math, in telling time on analog and digital display watches and in estimating/comparing object sets (e.g., telling if a bookshelf had more or fewer books than another). At school, she used her fingers as a support to solve even simple arithmetic operations. She was attending the third grade at a private school and had a supportive family. No behavioral problems were reported.

G. A. was diagnosed with developmental dyslexia and MLD, especially regarding verbal math aspects, such as transcoding and word arithmetic problems. His intelligence was in the mean range (percentile in Raven's Coloured Progressive Matrices = 75). G. A.'s deficits were more prominent in phonological processing tasks (digit span, pseudoword repetition, and reading). Furthermore, G. A. had other general difficulties in motor dexterity and executive functions. His performance in the nonsymbolic magnitude comparison task and in arithmetic facts was average. At school, G. A. had problems in learning math, reading/spelling, and overall in verbal aspects of school performance. He was attending the third grade at a public school. Clinically, G. A. presented difficulties with attention (for more case details, see Haase et al., 2014).

Procedures

Informed consent was obtained in written format from the children's parents and orally from the children. Research instruments and procedures complied with the Helsinki declaration and had previously been approved by the local research ethics review board. H. V. and G. A. were evaluated in a university outpatient clinic for math learning difficulties. Both children completed repeated measures of a nonsymbolic number comparison task, for which the internal Weber fraction (w) was estimated. They were also assessed regarding their addition and subtraction

calculation skills at the beginning and at the end of the experiment in the school year's second semester.

Repeated assessments of w and addition/subtraction were performed concomitantly to intervention programs. Intervention programs were customized and adapted to the performance level and main symptoms of each child. H. V.'s program focused on single digit multiplication abilities and G. A. worked on numerical transcoding abilities. Each intervention was structured in two pre- and post-test sessions, with 18 training sessions in between. The concepts and the procedures of single digit multiplication (H. V.) and numerical transcoding (G. A.) were discussed with the participants and taught using concrete materials and exercise sheets of increasing complexity. The intervention was based on errorless learning principles and a token economy system was implemented. A detailed analysis of these interventions is beyond the scope of the present article, and will be the subject of a future report.

Assessment of intelligence

General intelligence was assessed with the age-appropriate Brazilian validated version of Raven's Colored Matrices (Angelini, Alves, Custódio, Duarte, & Duarte, 1999). The analyses were based on z-scores calculated from the manual's norms.

Assessment of number sense accuracy

H. V. and G. A. were presented with the nonsymbolic comparisontask (Costa et al., 2011; Pinheiro-Chagas et al.,2014) in five weekly sessions. They completed the task twice in each session to the best of their capacities, without any feedback, with a total of 10 measures of their number sense abilities (w). In the nonsymbolic magnitude comparison task, the participants were instructed to compare two simultaneously presented sets of dots and to indicate which set contained the larger number of elements (Costa et al., 2011; Pinheiro-Chagas et al., 2014). Black dots were presented on a white circle over a black background. On each trial, one of the two white circles contained 32 dots (reference numerosity), and the other circle contained 20, 23, 26, 29, 35, 38, 41, or 44 dots. In order to assess discrete nonsymbolic numerical magnitude in dot comparison tasks, it is important to control for the possibility of using continuous dimensions as a hint to set numerosity. To control for this possibility, a trade-off between dot size and total area occupied by the dots was adopted in the confection of stimuli (Dehaene, Izard, & Piazza, 2005). The task comprised eight learning trials and 64 experimental trials. Stimulus remained on the screen until the child pressed a button or for a maximum duration of 4000 ms, and the intertrial interval was 700 ms. Between trials, a fixation point appeared on the screen-a cross, printed in white, with 30-mm-long arms. Estimations of the internal Weber fraction (w) or the standard deviation of the Gaussian distribution of the underlying numerosities were used as an index of number sense accuracy. The calculation of the Weber fraction was conducted in R software, using the following equation, according to methods proposed by Piazza, Izard, Pinel, Le Bihan, and Dehaene (2004):

$$p[R \in (r, r+dr)] = \frac{e - \frac{(r - Log[n])^2}{2w^2}}{\sqrt{2\pi}w} dr.$$

Assessment of basic calculation skills

In order to control for the stability of their arithmetic profile, we also report the results of a basic arithmetic operations task at two different time points. The task consisted of addition (27 items) and subtraction (27 items) operations for individual application, which were printed on separated sheets of paper. The children were instructed to answer as quickly and as accurately as they could, with the time limit per block being 1 min. These tasks have been used in previous studies (Costa et al., 2011; Haase et al., 2014), and their reliability is high (Cronbach's alphas > .90). Performance on multiplication operations is not reported as a control measure because it was the focus of the intervention program for one of the children (H. V.).

Results

At the time that they were originally assessed, w-values of H.V. and G. A. were, respectively, 0.42 and 0.21. (Haase et al.,2014). Repeated estimates of w were more stable for G. A.than for H. V., being respectively equal to 0.25, 0.20, 0.34,0.25, 0.23, 0.25, 0.21, 0.32, 0.47 and 0.38 for G. A. and 0.39,0.47, 0.41, 0.53, 0.74, 0.70, 0.75, 0.48 and 0.59 for H. V.

In the nonsymbolic comparison task, we calculated the R^2 of the fitting procedure to estimate w for each child. Low R^2 values (< 0.2) indicated that the children's performances were not well described by the Log-Gaussian model of number representation (Piazza et al., 2004). The first measure of H.V.'s third session was excluded, since it did not fit the model used to calculate the Weber fraction.

To investigate the stability of number sense measures, we performed a linear regression analysis, using session as independent variable and the Weber fraction as dependent variable for each participant (Figure 1). Results indicated that practice did not reduce the w-values, as tested by one-sided regression models, F(7) = 3.83, p = .95 for H. V. and F(8) = 4.72, p = .97 for G. A.

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Figure 1. H. V. and G. A. repeated measures of Weber fraction.

Additionally, H. V., who clinically presented more difficulties in processing numerosities (e.g. difficulties in telling time on analog and digital displays and estimating/ comparing object sets), qualitatively demonstrated a higher dispersion in the Weber fraction over the sessions than G. A., as indicated by the regression analysis error measures (residual standard error of H. V. = 0.12; residual standard error of G. A. = 0.07). When we compared the mean of all H. V. and G. A. measures, H. V. presented a significantly worse Weber fraction, t(12) = -5.20, p < .001.

Non-significant two-sample tests for equality of proportions with continuity correction indicated that the performance on two untrained kinds of calculation (addition and subtraction) did not change from pre-test to post-test (Table 1).

GA's assessment		\mathbf{V}^{2} (re value)	HV' assessment		\mathbf{V}^2 (re volue)
1^{st}	2^{nd}	$X^{-}(p-value)$	1^{st}	2^{nd}	$X^{-}(p-value)$
12 12	12/12	0.0 (p = ns)	11/12	8/12	1.0 (p = ns)
6/15	7/15	0.0 (p= ns)	11/15	9/15	0.2 (p= ns)
10/12	10/12	0.0 (p= ns)	7/12	4/12	0.7 (p= ns)
	GA's ass 1 st 12 12 6/15 10/12	GA's assessment 1 st 2 nd 12 12 12/12 6/15 7/15 10/12 10/12	GA's assessment X^2 (p-value) 1^{st} 2^{nd} X^2 (p-value)12 1212/120.0 (p = ns)6/157/150.0 (p = ns)10/1210/120.0 (p = ns)	GA's assessment X^2 (p-value) HV' assessment 1^{st} 2^{nd} X^2 (p-value) 1^{st} 12 12 12/12 0.0 (p = ns) 11/12 6/15 7/15 0.0 (p = ns) 11/15 10/12 10/12 0.0 (p = ns) 7/12	GA's assessment 1^{st} X^2 (p-value)HV' assessment 1^{st} 2^{nd} X^2 (p-value) 1^{st} 2^{nd} 12 1212/120.0 (p = ns)11/12 $8/12$ 6/157/150.0 (p = ns)11/15 $9/15$ 10/1210/120.0 (p = ns) $7/12$ $4/12$

Table 1 Longitudinal Comparison of Calculation Skills of H. V. and G.A.
Discussion

In this study, we investigated the stability over time of w, an estimate of number sense accuracy, in two children with MLD. In one case, H. V., pure developmental dyscalculia occurred in the context of a number sense impairment. In the other case, G. A., developmental dyscalculia occurred in association with developmental dyslexia with phonological processing impairments and normal w. Repeated assessments indicated that estimated w remained stable in both H.V. and G. A. and far from the range of normality in H. V. Qualitative observation indicated that for H. V., there was a nonsignificant tendency to worsen performance across measurements. Performance on simple addition and subtraction operations remained stable on two assessments over a period of 6 months.

Results in these two cases indicate that the internal Weber fraction could be a stable measure apparently insensitive to practice, especially considering that no feedback was presented. Qualitatively, the performance of H. V. presented greater variability from one session to another with a statistically nonsignificant tendency to become less accurate over sessions. As H. V.'s tendency to lower accuracy was not statistically significant, one cannot be sure of its clinical and theoretical significance. This result could be ascribed to motivational factors, since the girl already presented a difficulty in magnitude discrimination. Concomitant clinical observation indicated that H.V. was developing mathematics anxiety symptoms, and this could have influenced her performance.

Across the experimental sessions, it was observed that H. V. became less engaged in the study and reported doubts on her ability to solve the task. Her hands became wet and she also reported being bullied by colleagues and teachers at school because of her math difficulties. These results suggest that, although stable, internal Weber fraction (w) assessment may be influenced by motivational factors, such as math anxiety. Youngsters with MLD frequently display symptoms of internalizing and externalizing psychopathology (Auerbach, Gross-Tsur, Manor, & Shalev, 2008). It would be important, then, to further elucidate a possible role of motivational factors on their performance.

Mathematics anxiety is a feeling of tension, apprehension, or fear specifically directed toward math activities or math cognitions, that is known to impair mathematics performance (Chinn, 2007). Ma and Xu (2004) showed that low math performance is predictive of higher levels of math anxiety. Additionally, psychological treatment of math anxiety has been reported to improve math performance (Hembree, 1990). The clinically observed development of math anxiety symptoms in H. V. underscores the complexity of MLD. MLD may result from distinct cognitive mechanisms, and deficits may interact with the individual's perceptions and emotional reactions.

In spite of his mathematical difficulties, G. A. did not present an impairment in number sense as his performance remained stable across measurements and comparable to that of matched controls (see Haase et al., 2014). He remained engaged while performing the task and no evidence of mathematics anxiety was clinically observed. His pattern of math difficulties was related to poor performance in arithmetic tasks that required verbal engagement, such as number transcoding and word problems (Haase et al., 2014). Phonological processing impairments may thus be a causal factor, besides number sense, contributing to math learning disabilities. Children with dyslexia who do not present number sense deficits are frequently impaired in verbal aspects of mathematics (Simmons & Singleton, 2008, 2009).

In this study we observed that the patterns of number sense performance in two children with distinct forms of MLD remained stable. The number sense performance of G.A. remained consistently in the normal range, in line with his phonological processing impairments. The number sense accuracy of H. V. remained consistently very low and variable with a nonsignificant tendency to worsen. These results are compatible with the existence of different subtypes of MLD, and also with their relative stability over time.

The observation of temporal stability of number sense measures is at odds with other research. All studies showing improvement of w after training employed some kind of feedback procedure (DeWind & Brannon, 2012; Odic, Hock, & Halberda, 2014), which was not employed in our study. In the DeWind & Brannon (2012) study, significant improvement occurred only from the first to the second sessions. Feedback was removed after the sixth session, and performance remained stable thereafter. Together with our results, this suggests that feedback may be important to improving number-sense-related performance.

To our knowledge, no previous research has addressed the temporal stability of number sense accuracy estimates in clinically defined MLD. Temporal stability of number sense impairment and preservation is of considerable theoretical importance. The role of number sense impairments in MLD has been the focus of considerable dispute. Evidence of degraded approximate numerical representations in dyscalculia comes from studies showing difficulties in nonsymbolic number comparison tasks (Landerl et al., 2004; Mazzocco et al., 2011; Piazza et al., 2010). Other researchers observed impaired symbolic and spared nonsymbolic number processing in dyscalculia, indicating difficulties in assessing intact nonsymbolic magnitude representations from symbolic numbers (Noël & Rousselle, 2011; Rousselle & Noël, 2007). There is no conclusive position regarding this topic so far (Chen & Li, 2014; Fazio et al., 2014). Our results suggest that number sense impairments may play a role in some cases, such as H.V.'s, but not in others, such as G. A.'s.

Research on the association between number sense accuracy and arithmetic learning has been mainly conducted with demographically selected samples. Clinical research is still incipient. Our work underlines the need for more translational research, investigating the possibilities of clinical application of important theoretical advances in numerical cognition research. Single case studies can play this bridging role. Single case studies allow observations across time of the dynamic evolution of symptoms and the complex interactions between causal factors and clinical expression.

In conclusion, it can be said that the study contributed the following pieces of information: (1) the internal Weber fraction (w) can provide a stable measure of number sense accuracy in MLD children; (2) impairments in number sense accuracy may be used to characterize different subtypes of MLD (one associated with impairments in number sense accuracy, which may occur in an isolated or non-isolated form, and another associated with phonological impairments usually observed in developmental dyslexia); and (3) theoretically informed case studies may be useful in uncovering the cognitive deficits underlying developmental disorders as well as

validating current clinical diagnosis criteria, and even providing insights into treatment perspectives.

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APPENDIX 2

Contributions from specific and general factors to unique deficits: two cases of mathematics learning difficulties

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ABSTRACT

Mathematics learning difficulties are a highly comorbid and heterogeneous set of disorders linked to several dissociable mechanisms and endophenotypes. Two of these endophenotypes consist of primary deficits in number sense and verbal numerical representations. However, currently acknowledged endophenotypes are underspecified regarding the role of automatic vs. controlled information processing, and their description should be complemented. Two children with specific deficits in number sense and verbal numerical representations and normal or above-normal intelligence and preserved visuospatial cognition illustrate this point. Child H.V. exhibited deficits in number sense and fact retrieval. Child G.A. presented severe deficits in orally presented problems and transcoding tasks. A partial confirmation of the two endophenotypes that relate to the number sense and verbal processing was obtained, but a much more clear differentiation between the deficits presented by H.V. and G.A. can be reached by looking at differential impairments in modes of processing. H.V. is notably competent in the use of controlled processing but has problems with more automatic processes, such as nonsymbolic magnitude processing, speeded counting and fact retrieval. In contrast, G.A. can retrieve facts and process nonsymbolic magnitudes but exhibits severe impairment in recruiting executive functions and the concentration that is necessary to accomplish transcoding tasks and word problem solving. These results indicate that typical endophenotypes might be insufficient to describe accurately the deficits that are observed in children with mathematics learning abilities. However, by incorporating domain-specificity and modes of processing into the assessment of the endophenotypes, individual deficit profiles can be much more accurately described. This process calls for further specification of the endophenotypes in mathematics learning difficulties.

Keywords: endophenotype, mathematics learning difficulties, number sense, verbal numerical representations, phonological processing, dyslexia

Introduction

The cognitive underpinnings of arithmetic are highly complex (Rubinsten and Henik, 2009). One proposal is that arithmetic requires three types of symbolic and nonsymbolic number representations (Dehaene, 1992). The most basic form of numerical representation is nonsymbolic, analogic and approximate and corresponds to the number sense or the ability to discriminate numerosities. This ability can be described by Weber–Fechner's law, which measures the precision of the internal representation of numbers (Moyer and Landauer, 1967; Izard and Dehaene, 2008; Piazza, 2010). Precise numerical magnitude representations are related to phonologically and orthographically coded verbal numerals and visually based Arabic numerals (Dehaene and Cohen, 1995).

The number sense acuity is predictive of math achievement in both typical (Halberda et al., 2008; Mazzocco et al., 2011a) and disabled individuals (Piazza et al., 2010; Mazzocco et al., 2011b). Moreover, general cognitive resources are also involved in number processing, and calculations involve visuospatial abilities (Venneri et al., 2003), finger gnosias (Costa et al., 2011), phonological processing (De Smedt and Boets, 2010; De Smedt et al., 2010), working memory and executive functions (Camos, 2008; Pixner et al., 2011; Zheng et al., 2011).

The phenotypic presentation of mathematics learning disability and developmental dyscalculia (DD) is heterogeneous and includes a combination of the cognitive mechanisms that underlie arithmetic (Geary, 1993; Wilson and Dehaene, 2007). Because there are no consensual cognitive or biological markers, DD is operationally defined as persistent and severe difficulties in learning math in children of normal intelligence, that cannot be attributed to neurosensory impairment, sociodemographic, and emotional factors, or lack of adequate educational

experiences (American Psychiatric Association, 2000; World Health Organization, 2011). The nosological complexity of DD is compounded by its frequent comorbidity with other disorders, such as dyslexia (Landerl and Moll, 2010) and attention- deficit-hyperactivity disorder (ADHD, Gross-Tsur et al., 1996). Comorbidity can be explained by chance co-occurrences or by shared underlying mechanisms. The present evidence is still insufficient to decide about the role of comorbidity in characterizing DD (Rubinsten and Henik, 2009).

One possible way to solve the conundrum of DD's nosological validity is to consistently characterize implicated cognitive mechanisms as endophenotypes, in other words, as intermediate constructs between the interacting environmental and genetic etiologies and the phenotypic expression (Bishop and Rutter, 2009). A reliable endophenotype of number sense impairment has been gradually emerging (Piazza et al., 2010; Mazzocco et al., 2011b). However, restricting the definition of DD to individuals with more basic number processing impairments related to a number sense or number module (Reigosa-Crespo et al., 2012) would exclude from the domain of coverture of DD children and adolescents whose math learning difficulties could be persistent and of varying degrees of severity but associated with other cognitive mechanisms, such as phonological processing disorders (De Smedt and Boets, 2010).

Moreover, cognitive mechanisms that underlie math achievement and are potentially implicated in math learning difficulties could be classified as domain-specific or domain-general (Butterworth and Reigosa, 2007). Math-specific cognitive mechanisms include number sense (e.g., symbolic and nonsymbolic number comparison and estimation, number line estimation) and knowledge of the number system (Cowan and Powell, 2013). Domain-general mechanisms associated with math achievement and underachievement include phonological processing (Hecht et al., 2001), intelligence, processing speed, working memory, and executive functions (Cowan and Powell, 2013). It is increasingly recognized that DD can thus be characterized as primary, associated with number sense deficits, or secondary, associated with domain-general factors (Price and Ansari, 2013, for similar conceptions, see also Rubinsten and Henik, 2009; Reigosa-Crespo et al., 2012).

We argue that, in addition to being influenced by primary and secondary cognitive factors, the achievement profile of kids who struggle to learn math could also be affected by the nature of the information processing strategy that is deployed. An important research tradition in cognitive psychology, which dates back at least to Shiffrin and Schneider (1977), distinguishes between automatic (data-driven, bottom–up, effortless) and controlled (concept-driven, top–down, effortful) processing (Hasher and Zacks, 1979; Logan, 1988; Birnboim, 2003).

Evidence is still accumulating and is often inconsistent, but there are data that support impairments of both automatic and controlled processing in math learning difficulties. Impairments in the rapid automatized naming (RAN) of numbers (Bull and Johnston, 1997), a lack of the congruency effect in the number-size interference task (Rubinsten and Henik, 2005), and impairment in symbolic (with sparing of nonsymbolic) number comparisons (Rousselle and Noël, 2007) have been interpreted as evidence for an automatization deficit in DD. Impairments of several subcomponents of the central executive in DD have often been described (Bull and Scerif, 2001; van der Sluis et al., 2004; Geary et al., 2007; Raghubar et al., 2010, see also Kaufmann et al., 2004; de Visscher and Noël, 2013). This literature indicates that math achievement could be associated with both domain- specific and domain-general cognitive factors. Moreover, these two dimensions could interact with different modes or strategies of information processing according to the nature of the task.

In general, it is possible to say that researchers agree as to the cognitive factors that are implicated in math learning difficulties. Disagreement arises when the relative importance of each factor or their possible interactions or lack of interaction are considered. One possibility is a multiple-deficit model, according to which math learning difficulties are the epigenetic outcome of multiple interacting mechanisms (Cowan and Powell, 2013). Another possibility is that different types of DD are explained by impairments in different non-interacting endophenotypes. One of the most important endophenotypes that is implicated in dyscalculia is a number sense or a number module deficit (Reigosa-Crespo et al., 2012). Single-case studies of individuals with math learning difficulties could constitute an opportunity to test these concurrent models of cognitive impairments in dyscalculia.

Although not without its critics (Thomas and Karmiloff- Smith, 2002), the logic of doubledissociation in cognitive neuropsychology has also been applied in the context of developmental disorders, to more specifically characterize the endophenotypes that are implicated (Temple, 1997; Temple and Clahsen, 2002; White et al., 2006a,b; de Jong et al., 2006, 2009). In cognitive neuropsychology, it is generally assumed that if two cognitive processes double-dissociate or present complementary patterns of spared and impaired functions in two different patients, then this pattern is an indication of different underlying neural substrates (Temple, 1997).

A possible double-dissociation in the field of learning disabilities is the case of the underlying cognitive mechanisms of DD and dyslexia. Evidence indicates that children with DD could be selectively impaired in number sense tasks, while dyslexia impairs phonological processing

(Rubinsten and Henik, 2006; Landerl et al., 2009). Analysis has been performed on a series of single-case-generated evidence that is compatible with this interpretation (Tressoldi et al., 2007). The sole occurrence of DD and the sole occurrence of dyslexia, when associated with different cognitive profiles, suggest that these two disorders constitute distinct entities. At least in certain cases, the co-occurrence of DD and dyslexia could represent a true comorbidity, without a shared etiopathogenic variance (Landerl and Moll, 2010).

Double-dissociation logic has also been used to refine the phenotype of DD, characterizing subtypes that are related to impairments in specific cognitive components. A double dissociation has been observed in Arabic number processing. A case described by Temple (1989) presented a specific difficulty in reading Arabic numbers. The opposite difficulty of writing Arabic numbers was found by Sullivan et al. (1996). Similar to what is observed in adults with acquired acalculia, Temple (1991) demonstrated the existence of a double dissociation between procedural calculation impairment and a fact retrieval deficit. Specific fact retrieval deficits were later corroborated by Temple and Sherwood (2007) in a group study. Two additional single-case studies described specific impairments in math facts retrieval, uncovering a role for executive function and automatization in the deficits (Kaufmann, 2002; Kaufmann et al., 2004; de Visscher and Noël, 2013). Moreover, more complex interactions between magnitude processing and procedural knowledge also can be observed in the carry over operation when solving addition problems (Klein et al., 2010). A number sense deficit impairing cardinality and sparing ordinality was observed in an earlier case described by Ta'ir et al. (1997).

This line of reasoning suggests, then, that single-case studies that use double-dissociation logic could play an important role in clearing the complexity that underlies phenotypic manifestations

of DD and in establishing the relevant endophenotypes. Investigations on the number sense endophenotype using contemporary experimental measures are missing in the single-case literature. In this study, the aim is to contrast the patterns of cognitive deficits in two children at approximately 10 years of age with persistent math learning difficulties that are associated with distinct cognitive profiles. H.V., a 9-year-old girl, has math learning difficulties in the context of number sense inaccuracy, while G.A, a 10-year-old boy, presents math difficulties that are associated with developmental dyslexia and a phonological processing disorder. Neither of the children fulfilled the criteria for a more severe math learning disorder or disability. Instead, they were classified as having math learning difficulties, in other words, a performance below the 25th percentile on a standardized achievement test (Mazzocco, 2007). Performance on the Arithmetic subtest of the WISC-III was also not impaired in either of the children. Notwithstanding spared psychometric performance on achievement and intelligence tests, these two children presented persistent difficulties in specific domains of arithmetic, which were severe enough to cause low grades and to justify clinical referral.

The two cases were considered for analysis because of the comparable ages, similar sociodemographic backgrounds, normal or above average intelligence and impairment patterns that were suggestive of specific deficits in math learning difficulties. Standard neuropsychological assessment revealed specific impairments in the number sense in H.V. and in phonological processing in G.A. A more detailed assessment followed these observations.

Both domain-general and domain-specific cognitive mechanisms were included in the assessment (Butterworth and Reigosa, 2007; Cowan and Powell, 2013). Specific math assessment was based on two widely used cognitive models (McCloskey et al., 1985; Dehaene

and Cohen, 1995). In the numerical domain, the following assessments were performed: numerical transcoding, calculation, simple word problems and the approximate number system (ANS).

Selection of domain-general assessments included the following functions: general intelligence (Deary et al., 2007), working memory (Geary et al., 2007; Raghubar et al., 2010), and executive functions (van der Sluis et al., 2004). Moreover, we used both non-numerical (Victoria Stroop, Strauss et al., 2006) and numerical stimuli (Five-digits Test, Sedó, 2007) when testing executive functions and interference (see the rationale in Raghubar et al., 2010). Some aspects of our assessment protocol deserve further discussion. Phonological processing has been implicated in math learning (Hecht et al., 2001), mostly in the context of developmental dyslexia. A specific subtype of verbal dyscalculia has even been proposed (Wilson and Dehaene, 2007). Notwithstanding its theoretical plausibility, there is scarce evidence for a visuospatial subtype of dyscalculia (Geary, 1993; Wilson and Dehaene, 2007). Impairment of more executive aspects of visuospatial processing in math achievement has been reported, mostly in the context of the socalled nonverbal learning disability (Venneri et al., 2003). Wilson and Dehaene (2007) consider the possibility that impairments in the ANS and deficits in visuospatial attention could constitute two different subtypes of dyscalculia. It is important then to assess visuospatial and visuoconstructional abilities to check for the possibility of a nonverbal learning disability (Venneri et al., 2003; Fine et al., 2013). Finally, assessment of finger gnosias and motor dexterity were obtained because of their association with math learning difficulties (Costa et al., 2011; Lonnemann et al., 2011). Finger gnosias can underlie finger counting, which is an important offloading mechanism that liberates working memory resources at the beginning of formal math learning (Costa et al., 2011). Motor impairment could provide clues regarding the presence of minor brain insult (Denckla, 1997, 2003; Batstra et al., 2003).

Methods

Considering the hypothesis that modes of information processing interact with the domainspecificity of stimuli in the genesis of learning difficulties, we employed tasks assessing automatic and controlled processing in both general and math-specific domains. General automatic processing was assessed using RAN of colors in the Victoria Stroop test. Numerical automatic processing was assessed by means of RAN of digits and speeded counting in the Fivedigits Tests, nonsymbolic and symbolic number comparison tasks and by retrieval of arithmetic facts. Domain-general controlled processing was tapped by backward Corsi blocks span and the color-word interference phase of the Victoria Stroop test. Controlled processing in the numerical domain was evaluated with the backward Digit span and Inhibtion and Switching tasks of the Five-digits Test, as well as by word problems and working memory-dependent items in the numerical transcoding tasks. A simple reaction time task and the Nine-hole Peg Test were used to control, respectively for more basic aspects of alertness and motoric function.

CASE REPORTS

H.V. and G.A. were selected from cases at an outpatient facility for mathematical learning disabilities in Belo Horizonte, Brazil. Parents gave their written informed consent. In addition, informed consent was orally obtained from the children. Anamnestic information was obtained from the mothers of the two children.

H.V.

H.V. is a well-adjusted girl from a middle-class and supportive family, attending the third grade

at a private school. She had just completed 9 years of age by the time of evaluation. H.V. had difficulties in telling time on analogic and digital displays and estimating/comparing object sets (e.g., telling if a bookshelf had more or fewer books than another). She struggled to learn the math facts, to understand the place-value system and to solve word math problems. She uses fingers as a support to perform even the most simple additions and subtractions. Her learning difficulties are highly specific to math because her intelligence and achievement in other domains are above the average expected for her age. No major developmental problems were reported.

G.A.

G.A. is a well-adjusted boy from a middle-class and supportive family, who was 10 years and 2 months at the time of the neuropsychological assessment. He was attending the third grade at a public school. During his infancy, G.A. was submitted to several ear canal draining procedures that were related to recurrent otitis media. After the last surgery, his hearing and speech improved. His hearing is now normal and he was re-evaluated by a speech therapist who confirmed he has already improved from his previous difficulties. However, occasionally, he still mispronounces some of the more complex words, those that are less frequent and multi-syllable words that have consonantal clusters.

G.A. was referred due to early and persistent difficulties with reading/spelling and math. His reading/spelling difficulties are severe. His math difficulties are milder but are also persistent and are mostly related to word problem solving. Clinically, G.A. presents difficulties with attention. A tentative diagnosis of ADHD was made by another clinician.

Procedures

First, a general neuropsychological assessment was conducted, and the performances of both H.V. and G.A. were compared to available published norms. Table 1 lists the neuropsychological tests and their sources. Afterward, an experimental study was conducted to specifically investigate math cognition in both cases. In the experimental investigation, the performances of H.V and G.A. were compared to two control groups that were individually matched by gender, educational level, age, and socioeconomic status. In Brazil, the type of school is an important indicator of socioeconomic status because private schools generally offer better instruction than public schools (Oliveira-Ferreira et al., 2012). For this reason and because of the age differences between the two patients, separate control groups were used for the comparisons. The controls were selected among the participants of a population-based research project on math learning difficulties that was approved by the local ethics review board. Parents gave written informed consent, and the children gave their oral consent.

Domain	Test	References
Psychosocial functioning	CBCL—Child Behavior Checklist responded to by parents	Achenbach et al., 2008; Rocha et al., 2012
School achievement	TDE—Teste de Desempenho Escolar (School Achievement Test)	Stein, 1994; Oliveira-Ferreira et al., 2012
Intelligence	Raven's colored progressive matrices	Angelini et al., 1999
	We chsler intelligence scale for children 3° ed.	Figueiredo, 2002
Motor dexterity	9-HPT: Nine-hole peg test	Poole et al., 2005
Visuospatial abilities	Copy of the Rey–Osterrieth complex figure	Oliveira, 1999
Short-term and working memory	Corsi blocks	Santos et al., 2005
	WISC-III digits	Figueiredo and do Nascimento, 2007
	Auditory consonantal trigrams	Vaz et al., 2010
Phonological processing	Phoneme elision task	Lopes-Silva et al., 2014

 Table 1 | Neuropsychological instruments

	Pseudoword repetition	Santos and Bueno, 2003
	Pseudoword reading	Same stimuli as in Santos and Bueno (2003)
Executive functions	Color-word interference in the Victoria stroop	Charchat-Fichman and Oliveira, 2009
	Five-digits test	Sedó, 2004

The test performance of both cases was compared either to normed values, in the general neuropsychological assessment, or to the reference given by their individually selected control groups, in the math-cognitive assessment. Different statistical procedures that were based on psychometric single-case analysis (Huber, 1973; Willmes, 1985), one person vs. small sample comparisons (Crawford et al., 2010) and criterion-oriented methods (Willmes, 2003), were employed in these comparisons.

H.V.'s performance was compared to that of a group of 8 girls [mean age = 113 (SD = 3) months] from 3rd grade of a private school in Belo Horizonte, Brazil. All of them had intelligence performance that was well above the mean (percentile ranks in the Raven's Colored Progressive Matrices ranged from 70 to 95) and no learning difficulties. G.A.'s performance, in turn, was compared to that of 17 boys [mean age = 117 (SD = 4) months] from the 3rd grade of two public schools in Belo Horizonte, Brazil. The percentile ranks in the Raven' Colored Progressive Matrices of this control group ranged from 50 to 99, which was comparable to that of G.A.'s.

Instruments

In the following section, the more specific cognitive tests and tasks will be described in greater detail.

Brazilian school achievement test (TDE; Stein, 1994)

The TDE is a standardized test of school achievement (Oliveira- Ferreira et al., 2012) and comprises arithmetic, single-word spelling, and single-word reading. Specific norms are provided for school-age children between the second and seventh grade. Reliability coefficients (Cronbach α) of TDE subtests are 0.87 or higher. Children are instructed to work on the problems to the best of their capacity but without time limits.

Nine-hole peg test (9-HPT, Poole et al., 2005)

The 9-HPT is a timed test in which nine pegs should be inserted and removed from nine holes in the pegboard with the dominant and non-dominant hand. The pegboard is placed horizontally in front of the child, in such a way that the compartment that contains the pegs is on the side of the hand to be tested, while the compartment with the holes is on the contralateral side. Children must pick up one peg at a time. The test is performed two times with each hand, with two consecutive attempts with the dominant hand followed immediately by two consecutive attempts with the non-dominant hand. The scores were calculated based on the mean time for each hand.

Handedness ascertainment

Lateral preference was investigated by means of tasks that examine the ocular, hand, and foot preference based on Lefèvre and Diament (1982). The child was instructed to look through a hole, to kick and to throw a ball, three times each. The result was given by the side the child had chosen most of the time.

Right-left orientation test

This test is based on Dellatolas et al. (1998). It has 12 items of right and left body part

recognition that involves simple commands regarding the child's own body, double commands (direct and crossed) toward the child's body, and pointing commands to single lateral body parts of an opposite-facing person. The score system is based on the number of correctly pointed parts of the body. Internal consistency was assessed with the Kuder–Richardson reliability coefficient, which was high (KR-20 = 0.80) (Costa et al., 2011).

Finger localization task

This 24-item task was also based on Dellatolas et al. (1998), and it was used to assess finger gnosia. It consists of three parts: (1) localization of single fingers touched by the examiner with the hand visible (two trials on each hand); (2) localization of single fingers touched by the examiner with the hand hidden from view (four trials on each hand); and (3) localization of pairs of fingers simultaneously touched by the examiner with the hand hidden from view (six trials on each hand). A total score (that ranged from 0 to 12) was calculated for each child as well as the total score, which was the sum of the total from both hands. The internal consistency of this task is high (KR-20 = 0.79) (Costa et al., 2011).

Phoneme elision task

This test is a widely accepted measure of phonemic awareness (Wagner and Torgesen, 1987; Castles and Coltheart, 2004; Hulme et al., 2012; Melby-Lervåg et al., 2012). The child listens toa word and is expected to say how it would be if a specified phoneme were deleted. (e.g., "filha" without /f/ is "ilha" in English it would be "cup" without /k/ is "up"). The test comprises 28 items: in 8 of them, the child must delete a vowel, and in the other 20, a consonant. The consonants to be suppressed varied according to the place and manner of articulation. The phoneme to be suppressed could be in different positions of the words, which ranged from 2 to 3 syllables. The internal consistency of the task is 0.92 (KR-20 formula) (Lopes-Silva et al., 2014).

Victoria stroop task (Charchat-Fichman and Oliveira, 2009)

The Victoria Stroop task is a measure of executive function (Strauss et al., 2006). The subject is presented with three cards, each containing six rows of four items. In the first card (color), the task is to name quickly the color of 24 rectangles, which can be green, yellow, blue, or red. In the second card (word), the task is to name the colors of common words printed in green, yellow, blue, or red, ignoring their verbal content. On the third card, the stimuli are color names that are printed in an incongruent color that is never the same color as the word that is printed. The task is to name the color in which the word is printed (e.g., when the word "blue" is printed in red, the subject must say "red"). For each of the three conditions, the time to complete the naming of all of the stimuli was recorded. Additionally, the interference score (Stroop-Effect) was calculated as the quotient between the time score for the incongruent (third card) and the color (first card) conditions.

Five digits test

The Five Digits Test was validated and standardized in Spanish and English by Sedó (2004, 2007) as a measure of speeded counting, Arabic number reading, and inhibition and set shifting. Similar sets of stimuli are used across tasks. Automatic processing is assessed through speeded tasks of counting randomly presented star sets (up to five) and reading Arabic digits (up to five). Controlled processing is assessed through inhibition and set-shifting tasks. In the inhibition task (choosing), the child must count the number of Arabic digits instead of reading them. In the set-shifting condition (switching), the child switches from counting the number of Arabic digits in

most trials to reading them when a frame surrounds the stimulus set.

The numeric and arithmetic tasks for the experimental study have been employed in previous investigations (Costa et al., 2011; Ferreira et al., 2012; Júlio-Costa et al., 2013) and are described below.

Simple reaction time

The computerized simple RT task is a visual detection task that is used to control possible differences in the basic processing speed that is not related to numerical tasks. In this task, a picture of a wolf (height 9.31 cm; length = 11.59 cm) is displayed in the center of a black screen for a maximum time of 3000 ms. The participant is instructed to press the spacebar on the keyboard as fast as possible when the wolf appears. Each trial was terminated with the first key press. The task has 30 experimental trials, with an intertrial interval that varies between 2000, 3500, 5000, 6500, and 8000 ms.

Nonsymbolic magnitude comparison task

In the nonsymbolic magnitude comparison task, the participant was instructed to compare two simultaneously presented sets of dots and to indicate which set contained the larger number (see Figure 1). Black dots were presented on a white circle over a black background. On each trial, one of the two white circles contained 32 dots (reference numerosity), and the other circle contained 20, 23, 26, 29, 35, 38, 41, or 44 dots. Each magnitude of dot sets was presented 8 times. The task comprised 8 learning trials and 64 experimental trials. Perceptual variables were randomly varied such that in half of the trials, the individual dot size was held constant, while in the other half, the size of the area occupied by the dots was held constant (see exact procedure

descriptions in Dehaene et al., 2005). The maximum stimulus presentation time was 4000 ms, and the intertrial interval was 700 ms. Between each trial, a fixation point appeared on the screen—a cross, printed in white, with 30 mm in each line. If the child judged that the right circle presented more dots, then a predefined key localized on the right side of the keyboard should be pressed with the right hand. In contrast, if the child judged that the left circle contained more dots, than a predefined key on the left side had to be pressed with the left hand.





Symbolic magnitude comparison task

In the symbolic magnitude comparison task, Arabic digits from 1 to 9 were presented on the computer screen (height =2.12 cm; length = 2.12 cm). The visual angle of the stimuli was 2.43° in both the vertical and horizontal dimensions. Children were instructed to compare the stimuli with the reference number 5. Digits were presented in white on a black background. If the presented number was smaller than 5, the child had to press a predefined key on the left side of the keyboard, with the left hand. If the stimulus was higher than 5, then the key to be pressed

was located at the right side and should be pressed with the right hand. The number 5 was never presented on the computer screen. Numerical distances between stimuli and the reference number (5) varied from 1 to 4, each numerical distance being presented the same number of times. Between trials, a fixation point of the same size and color of the stimuli was presented on the screen. The task comprised 80 experimental trials. The maximum stimulus presentation time was 4000 ms, and the intertrial interval was 700 ms.

Simple calculation

This task consisted of addition (27 items), subtraction (27 items), and multiplication (28 items) operations for individual applications, which were printed on separate sheets of paper. Children were instructed to answer as fast and as accurately as they could, with the time limit per block being 1 min. Arithmetic operations were organized at two levels of complexity and were presented to children in separated blocks: one consisted of simple arithmetic table facts and the other consisted of more complex facts. Simple additions were defined as those operations that had results of below 10 (i.e., 3 + 5), while complex additions had results between 11 and 17 (i.e., 9 + 5). Tie problems (i.e., 4 + 4) were not used for addition. Simple subtraction comprised problems in which the operands were below 10 (i.e., 9 - 6), while for complex subtractions, the first operand ranged from 11 to 17 (i.e., 16 - 9). No negative results were included in the subtraction problems. Simple multiplication consisted of operands (i.e., 2×7 , 5×6), while for the complex multiplication, the result of the operands ranged from 24 to 72 (6×8). Tie problems were not used for multiplication. Reliability coefficients were high (Cronbach's $\alpha > 0.90$).

Simple word problems

Twelve arithmetical word problems were presented to the child on a sheet of paper while the examiner read them aloud simultaneously to avoid a reading proficiency bias. There were six addition and six subtraction items, all of them with single-digit operands and results that ranged from 2 to 9 (i.e., "Annelise has 9 cents. She gives 3 to Pedro. How many cents does Annelise have now?"). The child had to solve the problems mentally and write the answer down in Arabic format as quickly as possible, and the examiner registered the time that was taken for each item. Cronbach's α of this task was 0.83.

Arabic number reading task

Twenty-eight Arabic numbers printed in a booklet were presented one at a time, to the children, who were instructed to read them aloud. The item set consists of numbers up to 4 digits (3 onedigit numbers, 9 two-digit numbers, 8 three-digit numbers, and 8 four-digit numbers). There were 12 numbers that could be lexically retrieved, 5 numbers that required three transcoding rules according to the ADAPT model (Barrouillet et al., 2004) to be correctly read, 6 numbers with four rules and 5 numbers with more than five rules. The internal consistency of the task is 0.90 (KR-20 formula) (Moura et al., 2013).

Arabic number writing task

Children were instructed to write the Arabic form of dictated numbers. This task is composed of 40 items, with up to 4 digits (3 one-digit numbers, 9 two-digit numbers, 10 three-digit numbers, and 18 four-digit numbers). The one- and two-digit numbers were classified as "lexical items" (12 items), and the other 28 items were subdivided according to the number of transcoding rules based on the ADAPT model (Barrouillet et al., 2004; Camos, 2008). There were six numbers that require 3 rules, nine numbers that require 4 rules, six numbers with 5 rules, five

numbers with 6 rules, and two numbers with 7 rules. The internal consistency of this task is 0.96 (KR-20 formula) (Moura et al., 2013).

Results

General cognitive assessment

Results of the CBCL reported by their respective mothers were in the normal range in all of the subscales (T-scores in the single subscales ranged from 37 to 45 in H.V. and from 36 to 54 in G.A. Scores above 70 are considered to be clinical). This finding indicates that both children have adequate levels of psychosocial functioning, according to their mothers. The results of the intelligence test are exhibited in Figure 2, while Figure 3 depicts comparative results in the two cases for the general neuropsychological assessment compared to norms from the original publications. H.V. shows a performance in the upper bound of normal intelligence (Raven's PR = 99, FSIQ = 120, VIQ = 116, and PIQ = 121), and G.A. shows average intelligence (Raven's PR = 75, FSIQ = 87, VIQ = 89, and PIQ = 89).



FIGURE 2. H.V.and G.A.performances in WISC-III. *Marked statistical significance at the level p < 0.001. Note: as H.V.'s standardized Block Design score was below the mean in the first assessment, this subtest was repeated two years later (gray dot). The new standardized score in Block Design was equal to 10.



FIGURE 3. H.V.and G.A. performances in intelligence, motor dexterity, visuospatial, short-term and working memory, executive functions, and phonological processing tasks. 9-HPT,Nine Hole Peg Test; ROCF, Rey-Osterrieth Complex Figure; ACT,Auditory consonantal trigrams; PRp,Pseudoword Repetition; PRd,Pseudoword Reading; PE,Phoneme Elision; VS,Victoria

Stroop.Clinical score < -1.5 SD.

Statistical comparisons between both children in the subtests that measure the verbal and performance IQs (Huber, 1973; Willmes, 1985) reveal significantly higher scores for H.V. in the subtest Information (Z = 2.95; p = 0.016), Similarities (Z = 3.33; p = 0.004), Arithmetic (Z = 2.58; p = 0.05), Vocabulary (Z = 4.87; p = 0.00001), Figure Assembly (Z = 2.36; p = 0.01) and Coding (Z = 5.59, p = 0.00001). These results disclose a general pattern of higher scores in H.V. than in G.A. regarding tasks that demand more from verbal IQ but not as much regarding performance IQ.

Performance on the TDE (Brazilian School Achievement Test) was below the 25th percentile in both cases for Arithmetic. H.V.'s accuracy percentage was 29% (raw score = 11, grade mean = 16 grade, SD = 3.39) and G.A.'s was 36% (raw score = 14, grade mean = 16, grade SD = 3.39). The 25th percentile criterion is used as a lenient cut-off and is sensitive to math learning difficulties (Mazzocco, 2007; Landerl and Kölle, 2009; Landerl et al., 2009). Performance on the single word Reading and Spelling subtests of the TDE were normal for H.V. and below the 25th percentile for G.A. G.A. solved 14 out of the 35 items of the Spelling subtest correctly. In some items, he excluded phonemes (especially /r/, regardless of its mode or place of articulation), and in others, he confused phonemes that have similar sounds (such as v/f; m/n; b/d; and s/c). He clearly presented a phonological writing pattern, but he still lacks the mastery of the alphabetical principle. In the Reading subtest, G.A. could read 55% of the single words (raw score = 39, grade mean = 64.75, SD = 4.67), and his reading was extremely slow. He struggled at reading consonant clusters.

Regarding motor dexterity in the 9-HPT, H.V. did not present any major difficulties, whereas

G.A.'s score was on the adopted clinical range, which means that he was significantly slower than would be expected for his age range according to Poole et al. (2005) (Figure 2). Both children presented right hand dominance (Lefèvre and Diament, 1982) as well as normal right-left orientation (Dellatolas et al., 1998) and finger gnosias (Dellatolas et al., 1998) (Figure 2). Neither of the children presented visuospatial constructional deficits.

On the phonological processing tasks, G.A. was significantly worse on all of the tests that were used, while H.V. presented typical scores. G.A. presented difficulties in storing and reproducing pseudowords as well as in reading them. In addition, he was not able to grasp the grapheme-phoneme correspondence principle that is needed to perform the phoneme elision task.

Both children presented difficulties in the phonological short-term memory task (forward digit span), but in both cases, scores in the phonological working memory tests (backward order of the Digit Span as well as the Auditory Consonantal Trigrams) fell into the expected ± 1.5 SD range (Figure 2). This specific difficulty on the forward order of the Digit Span was mild, and it can be attributed to attentional lapses (Strauss et al., 2006). G.A. presented a better performance on the forward order of the Corsi Blocks compared to the backward, and H.V. showed the opposite pattern. However, both of their spans were in accordance to what would be expected for their age range. The performance of both children was in the typical range for the Victoria Stroop task. G.A.'s performance was in the clinical range for all of the subtests of the Five-digits test, those that involve more automatic processing (speeded digit reading and counting) as well as those that require executive functioning (inhibition and shifting). H.V. presented only a specific impairment that involved counting skills on the Five-digits Test, which will be discussed in more
detail below ¹.

Math cognitive assessment and computer tasks

Results of the computerized and math-cognitive tasks are shown in Tables 2, 3 for H.V. and in Tables 4, 5 for G.A. and their respective control groups.

In the simple reaction time task, H.V. did not show any impairment. In contrast, she responded faster than the average of her group. In the symbolic number task, the picture is different. Although H.V. was significantly slower than her control group, her response accuracy was slightly higher than that of controls in a type of speed-accuracy trade-off. Moreover, the performance of H.V. in the nonsymbolic task was markedly impaired in comparison with her control group. While the reaction times were comparable to the group average, the accuracy was very poor, especially for the more difficult numerical ratios. These deficits added to the picture that was formed by a speeded counting impairment in the Five-digits Test. The results of the number processing tasks suggest that there was a specific impairment in the number sense acuity in the presence of relatively spared numerical symbolic abilities.

H.V.'s performance was substantially impaired in complex addition and multiplication operations. Her performance was comparable to the control group in simple word problems (Table 3). H.V. can solve simple addition and subtraction operations as accurately as expected

¹ 1After the neuropsychological assessment, both children initiated interventions based on cognitive-behavioral techniques to reduce math-anxiety symptoms and also to improve self-efficacy. Strategies such as problem-solving, self-monitoring, and self-reinforcement are coupled with errorless learning, allowing the kids to have experiences of academic success. Simultaneously,we also use instructional and training interventions that focused on number processing and arithmetic components that were considered to be impaired in each child. H.V. and G.A. have been participating in individual intervention programs for 4 semesters, 2 hs week. Their families also received counseling by means of a behavioral training program for one semester, once a week. During this time, H.V. has not obtained improvements in her number sense acuity,but she considerably improved in solving addition and multiplication problems. She has not automatized fact retrieval yet. H.V. does her homework with a pocket calculator. Initially, G.A. received training in text processing abilities and improved in arithmetical word problem solving.He also obtained substantial improvement in his transcoding abilities

according to her age. In complex addition operations, H.V. presents more difficulties when compared to her control group. Interestingly, these difficulties could not be observed in complex subtraction tasks. Moreover, in comparison to controls, H.V. shows systematic difficulties when solving simple and complex multiplication operations, which can be interpreted as a more general deficit in fact retrieval. No deficits were observed in simple word problems with onedigit operands, the solution of which depends on text comprehension; these problems can be solved by counting procedures. She solved all of the problems correctly but took considerably more time to reach the correct results. Performance on number transcoding of three- and fourdigit numerals was comparable to the control group (Table 3). These results are summarized in Table 3.

Table2.Descriptive data and comparison between the control groups and H.V., in the alertness and number sense tasks.

Domain	Task	Controls (n=08)		II V	Modifie		7.00	Estimated %	
assessed		Mean	SD	п.v.	d t test	р	Z-CC	pop. below H.V.	
Alertness	Simple manual reaction	/13 3	32.0	381.3	_0.92	0.30	-0.97	10%	
	time	415.5	52.7	561.5	-0.72	0.37	-0.77	1770	
Number sense	Symbolic Magnitude								
	Comparison								
	Response time*	771.6	154.8	1153.3	2.32	0.05	2.47	97%	
	Weber Fraction	0.27	0.17	0.12	-0.83	0.21	-0.88	22%	
	Nonsymbolic Magnitude								
	Comparison								
	Response time*	1035.5	199.5	1003.2	-0.15	0.88	-0.16	44%	
	Weber Fraction	0.29	0.06	0.42	2.24	0.04	2.17	4%	

ZCC: magnitude effect index calculated by the difference between the scores of the control group and the single case with a 95% CI (Crawfordetal., 2010); *time in milliseconds.

Domain assessed	Task	Controls (n=08)		H.V.	Chi-	n
	1 6 651X	Mean	SD		square	P
	Basic arithmetic operations					
	Simple addition(12)	11.88	0.35	9	1.30	0.25
<u>C</u> !1.	Complex Addition (15)	13.88	2.23	7	5.45	0.02
Simple	Simple Subtraction (12)	10.50	1.69	11	< 0.01	1.00
calculation	Complex Subtraction (15)	8.50	3.25	4	1.68	0.19
	Simple Multiplication (15)	13.13	2.64	4	11.18	0.00
	Complex Multiplication (13)	6.25	3.45	0	5.81	0.02
Simple word problems	Math Word Problems (12)	10.50	2.35	12	0.18	0.67
Verbal-Arabic	Arabic number writing task(40)	38.00	3.22	40	0.51	0.47
transcoding	Arabic number reading task (28)	27.75	0.71	28	< 0.01	1.00

Table 3 | Descriptive data and comparisons between control groups and H.V. in the Simple calculation, Simple word problems, and Verbal-Arabic transcoding tasks (df = 1).

In the simple reaction time task, G.A. did not show any impairment but instead showed average performance (Table 4). In the symbolic number task, G.A. responded tendentially slower and much less accurately than his control group. In contrast, G.A. presented both average response latency and average accuracy in the nonsymbolic number comparison task. In the number processing tasks, G.A. experienced considerable difficulties in tasks that use the symbolic notation and verbal procedures, such as speeded counting, speeded digit reading, transcoding and symbolic magnitude comparison (up to nine). G.A.'s pattern of impairment in the math tasks contrasts with that of H.V. Difficulties in the symbolic number processing tasks in G.A. are at odds with a normal Weber fraction.

G.A.'s difficulties with the symbolic processing were also corroborated by his lower

performance in the transcoding tasks.

In the number writing task, G.A. committed 14/40 errors. G.A. presented three lexical errors (all of them were related to phonological resemblance between the trial and the number written by him) and eleven syntactic ones (seven being related to adding internal zeros and four to deleting a digit). Fifty-two percent of his control group did not commit any error. From the eight children who did, one committed eleven errors, one presented five errors, one committed two errors and the other five children made only one single mistake.

The lexical mistakes by G.A. clearly have a phonological bias. In Portuguese, the numbers "three" and "six" sound very similar ("três" and "seis," respectively), as well as "seven hundred" and "six hundred" ("setecentos" and "seiscentos"). Moreover, the syntactic errors of G.A. always involved the addition principle (overwriting rule, Power and Dal Martello, 1990; Moura et al., 2013). G.A. wrote the number 643 as 646 and 4701 as 400601. His performance on the number reading test also corroborates his difficulties with place value understanding. He read the number "2000" as "two hundred" and "1013" as "one hundred thirteen." On two items, he decomposed the numbers: 567 was read as "five and sixty seven" and 5962 as "fifty nine and sixty two." Nevertheless, the mistakes made by G.A. cannot be easily attributed to a lack of knowledge of the rules of additivity in number transcoding.

G.A. was able to transcode correctly five out of eleven complex numbers with syntactical zeros (e.g., "109," "902," "1060," "1002," and "7013") but failed to transcode numbers of comparable complexity ("101" \ge 11, "1015" \ge 10015, "2609" \ge 20069, "4701" \ge 40601, "1107" \ge 2067, and "7105" \ge 715). Therefore, the poor transcoding performance of G.A. is compatible with deficits

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in phonological representations combined with problems with concentration and monitoring capacity. Evidence for a deficit in knowledge about the structure of the Portuguese verbal number system was not obtained.

Difficulties with simple word problems were more severe. G.A. did not show any impairment in solving addition, subtraction and multiplication problems when compared to controls, except for a single result that indicated lower performance while solving simple addition tasks (Table 5). This pattern is consistent with the mother's report that G.A. acquired the arithmetic facts after struggling with them for a while. However, the verbal nature of G.A.'s difficulties becomes explicit again, when considering his attainment of simple word problems. From 12 problems, G.A. solved only 4 correctly, responding sometimes with absurd values, which suggested that he was guessing. His performance on word problems was almost six standard scores below that of the controls. In summary, the results of the math cognitive investigation suggest that G.A.'s difficulties in learning math can be attributable to his comorbid reading learning disability.

Domain assessed	Task	Controls (n=17)			Modified			Estimated
		Mean	SD	G.A.	t test	р	Z-CC	% pop. below G.A.
Alertness	Simple manual reaction time	423.8	82.3	447.9	0.29	0.39	0.29	39%
Number sense	Symbolic Magnitude Comparison							
	Response time	983.1	249.4	1344.9	1.41	0.09	1.45	9%
	Weber Fraction	0.21	0.14	0.78	3.96	< 0.001	4.07	99%
	Nonsymbolic Magnitude							
	Comparison							
	Response time	1276.3	294.9	1038.2	-0.79	0.22	-0.81	39%
	Weber Fraction	0.28	0.10	0.21	-0.68	0.25	-0.70	25%

Table 4 | Descriptive data and comparison between control groups and G.A. in the alertness and number sense tasks.

ZCC: magnitude effect index calculated by the difference between the scores of control group and single-case with a 95%CI (Crawford et al., 2010); *time in milliseconds.

		Controls (n=17)			Chi-	
		Mean	SD	G.A.	square	p
	Basic arithmetic operations					
	Simple addition(12)	11.71	0.69	6	4.78	0.03
C' 1	Complex Addition(15)	10.12	3.06	5	2.26	0.13
Simple	Simple Subtraction(12)	10.41	2.15	8	0.46	0.50
calculation	Complex Subtraction(15)	5.18	3.21	2	0.87	0.35
	Simple Multiplication(15)	9.71	4.58	5	1.84	0.18
	Complex Multiplication(13)	2.35	2.12	0	0.85	0.36
Simple word problems(12)	Math Word Problems	10.82	1.19	4	5.98	0.01
Verbal-Arabic	Arabic number writing task(40)	38.65	2.78	26	10.94	<.001
transcoding	Arabic number reading task(28)	27.59	0.87	16	11.61	<.001

Table 5. Descriptive data and comparison between control groups and G.A. in the Simple calculation, Simple word problems and Verbal-Arabic transcoding tasks (df=01)

Discussion

In the present study, we selected two cases that had relatively specific impairment patterns from an outpatient clinic for mathematical learning disorders and conducted a detailed neuropsychological and cognitive assessment with the aim of characterizing possible endophenotypes. Specificity of the impairments is corroborated by the fact that both children were of average or above average intelligence and did not present impairments in visuospatial and visuoconstructional processing, as assessed by the Rey figure copy and Block Design subtest of the WISC. In the following, we will discuss the extent to which the neuropsychological profile of H.V. and G.A. fitted specific endophenotypes, as predicted in the literature. Difficulties in H.V. are specific, severe and persistent and were restricted to an inaccurate number sense and to the acquisition of arithmetic facts, which reflected mostly on multiplication operations. H.V. is curious and motivated to learn, except for mathematics. H.V. has difficulties in memorizing even the simplest arithmetic facts, but she is highly skilled in finger counting. The single abnormally lower score observed in the general neuropsychological assessment was in the forward version of the digit span. An excellent performance was observed in reading-related phonological processing tasks, such as pseudoword repetition, pseudoword reading and phonemic ellison. No abnormalities were observed in executive function tasks.

One might wonder why the performance of H.V. in the subtest "counting" of the Five-digits Test of executive functions was so low and discrepant from her general level of performance on this test. The subtest counting is a speeded task in which one has to count how many stars are printed on a series of cards that display sets of one up to five stimuli. The difficulties with the speeded counting of stars presented by H.V. reflect much more a deficit in the apprehension of nonsymbolic magnitude information under time constraints. This pattern contrasts with her resourceful use of strategies to compensate for her difficulties in other tasks that do not require nonsymbolic number processing. One of her favorite compensatory strategies for solving even the simplest arithmetic problems is finger counting. Once sufficient time is allowed, H.V. can find the correct response by finger counting. Her difficulties are accentuated in speeded tasks that require automatic retrieval.

The deficits in fact retrieval that are presented by H.V. cannot be attributed to a reduced capacity of verbal working memory or phonological awareness because H.V. shows high levels of competence in these two cognitive functions. However, the deficits in the numerical and arithmetic abilities of H.V. are compatible with generally imprecise or poor numerical representations: on the one hand, the deficits of H.V. in multiplication tasks suggest impairment in the retrieval of appropriate information from memory. On the other hand, the high value of the Weber fraction observed in nonsymbolic magnitude comparison suggests a very inaccurate ANS.

In contrast to our expectations, the profile of H.V. does not fit a typical endophenotype that is characterized by a number sense deficit (Wilson and Dehaene, 2007; Noël and Rousselle, 2011). Although H.V. presents low acuity in nonsymbolic magnitude comparison, this deficit is not present in the symbolic version of the task. More importantly, a substantial deficit in arithmetic operations—particularly in subtraction—was not observed. In contrast, H.V. presented some deficit in complex addition operations, but no sign of a deficit was observed in simple or complex subtraction operations. Moreover, a substantial deficit in multiplication operations (simple as well as complex) cannot be accounted for by a deficit in the number sense alone, but suggests the presence of difficulties for automatizing the retrieval of multiplication facts.

G.A.

G.A. presented persistent but milder difficulties in learning math in the context of developmental dyslexia with severe associated phonemic processing deficits. In the case of G.A., math learn ing impairments were observed in transcoding operations as well as in very simple one-digit word problems. G.A. presented deficits in all phonological processing tasks: digit span, pseudoword repetition and reading as well as in phoneme elision. Although his intelligence is normal, difficulties were also observed in motor dexterity and in all subtests of the five-digits procedure, both those tapping automatic (speeded counting, speeded digit reading) and those assessing controlled processing (inhibition, set shifting). Moreover, a borderline performance was also

observed in the forward and backward Digit and backward Corsi spans.

G.A. showed a less pronounced deficit in numerical and arithmetical abilities than H.V. The acuity of his representation of magnitude was comparable to controls, as measured by the nonsymbolic magnitude comparison task. In contrast, in the symbolic magnitude comparison task, G.A. committed many more errors and was marginally slower than his control group. Although G.A. presented lower levels of performance than controls in the simple addition operations, no other difference was observed in simple or complex addition, subtraction or multiplication operations. This pattern indicates that G.A. can retrieve from memory the correct responses to simple operations and employ the correct procedures to execute more complex addition and subtraction operations. However, in comparison to the controls, G.A. was much less successful when solving word problems. G.A. also presented substantially more difficulties in transcoding tasks in comparison to his peers, especially regarding phonological representations, concentration and monitoring capacity.

The profile of G.A. fits only partially a typical endophenotype that is characterized by a verbal and symbolic deficit. Although G.A. presents low acuity in symbolic magnitude comparison, simple word problems and impaired performance in transcoding tasks, this deficit does not extend to the retrieval of multiplication facts. It is still a matter of debate to what extent multiplication facts are stored in a typical verbal format (Varley et al., 2005; Benn et al., 2012). However, deficits in verbal numerical information processing have, very often, been associated with deficits in fact retrieval (De Smedt and Boets, 2010; De Smedt et al., 2010).

G.A. also presents severe problems with motor dexterity, which are assessed with the 9-HPT,

which deserve consideration. Sensorimotor impairments are a frequent concomitant of specific learning disorders observed both in dyslexia (White et al., 2006a,b) and in dyscalculia (Costa et al., 2011; Lonnemann et al., 2011). Minor sensorimotor dysfunction was observed in 87% of dyslexic children with an IQ higher than 85 (Punt et al., 2010). In this context, they are not interpreted as a causal mechanism that is implicated in learning difficulties, but as markers or co-localizers of brain insult (Denckla, 1997, 2003; Batstra et al., 2003). Whatever the cause of G.A.'s present learning difficulties, it also impaired his neurological functions in a more widespread manner, as shown by the relatively severe reduction in motor dexterity. Because the motor difficulties were comparable in both hands, no inferences can be made regarding lateralization of the underlying pathological process, other than the left-hemisphere dysfunction that is connected to developmental dyslexia.

In our view, the sensorimotor deficits could be responsible for his deficits in other tasks as well. G.A.s performance in both the Block Design subtest and Rey's Figure copy were situated from 0.7 to 1 standard deviations below the mean, which suits his WISC-FSIQ of 90. Moreover, a qualitative assessment of G.A.'s performance in the Block Design subtest and Rey's Figure copy indicate that his relative difficulties originate from the motor dexterity and executive components that are mobilized to solve these tasks and do not reflect impairments in apprehension or reproduction of visuospatial configurations. Further corroboration of these findings comes from the Raven. There, G.A. reached a score that was higher than average. In our view, such a level of performance on the Raven cannot be reached when simple visuospatial processing is impaired.

The difference between G.A.'s scores on the WISC and Raven can be attributed to an interaction between test and individual characteristics. Compared to the Raven, the WISC-III imposes

greater demands on verbal and scholastic abilities. Performance on several WISC tasks is also time constrained. We believe that G.A.'s relatively lower performance on the WISC can be explained by his reading and academic difficulties as well as by impairment in motor dexterity and processing speed. This pattern is especially salient on the Coding subtest, which is the test that presents the worst performance. Difficulties with the Coding subtest can also be related to G.A.'s impairment with respect to the symbolic transcoding tasks (Strauss et al., 2006).

Specific deficits in automatic vs. controlled numerical processing?

Comparisons of the endophenotypes as predicted by the current literature (Wilson and Dehaene, 2007; Noël and Rousselle, 2011) and the individual cases of H.V. and G.A. yield apparently frustrating results because the performance of H.V. and G.A. on the arithmetic tests partly contradicts the general expectation of more or fewer specific deficits in the number sense and verbal numerical representations, respectively. One possible interpretation of these results is that paradigmatic cases that regard specific endophenotypes can be very difficult to find. Although the initial assessment of H.V. and G.A. suggested number sense and verbal deficits, a more detailed examination revealed, in both cases, a less precise picture. Similar difficulties encountered by other authors (e. g., Tressoldi et al., 2007), suggest that only a small proportion of all of the cases of mathematics difficulties can reveal more pure forms of endophenotypes. This finding raises the question about the proportion of cases of mathematics difficulties that can actually be assigned with confidence to one or another subtype of this disorder. If it is low, then the general approach of endophenotypes might prove to be ineffective. Although our case design does not allow a direct investigation of this question, in this section, we will discuss one possible reason why endophenotypes can be indeed valuable in the investigation of mathematics difficulties.

One could propose that the severe deficits of H.V. solving multiplication problems while simultaneously being capable of solving complex subtraction problems are a result of compensatory strategies, such as finger counting. Finger counting could be more effective for subtraction than for multiplication operations because the multiplication operations usually have much higher numbers as the answers, which are much more difficult to reach by counting. Assuming that this reason explains H.V.'s performance, the discrepancy between her performance and the typical results that are expected according to the number sense endophenotype should be due to relatively trivial differences between prototypical profiles and individual cases, without more profound consequences for the refinement of the theoretical framework of mathematical learning disorders.

The same conclusion can be reached when analysing the discrepancy between G.A.'s performance and a verbal numerical endophenotype. Deficits in calculations should be expected, especially when the problems are more complex, rely more strongly on a verbal code, and the ability to use verbal number representations is as limited as in the case of G.A. However, this expectation was not confirmed by the results. Once more, one can attribute the discrepancy between the observed performance and typical endophenotypes to some individual compensatory resource, which is always plausible in individual cases and is frequently reported in clinical observations (Temple and Clahsen, 2002; Thomas and Karmiloff-Smith, 2002).

Moreover, the cognitive-neuropsychological approach to developmental disorders has been criticized on the grounds of the dynamics of the developing brain (Thomas and Karmiloff-Smith, 2002). Early acquired lesions or genetic dysfunctions can induce varying degrees of

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reorganization in the cognitive relevant brain processes. In exceptional cases, clear-cut structural-functional correlations, which are similar to the ones encountered in adults, are observed in cases of dysfunction in the developing brain (e.g., Temple, 1989, 1991; Sullivan et al., 1996; Ta'ir et al., 1997). In most cases of early acquired or genetic disorders, clinical-anatomical correlations are attenuated by several neuroplastic and compensatory processes.

Interestingly, there is an aspect of the performance of both H.V. and G.A. that could account for the patterns of the results observed in the respective cases without resorting to weak accounts that are based on typicality. The pattern of performance presented by H.V. reveals deficits in different numerical representations, which usually can be operated in an automatic or effortless fashion. The definition of the ANS, for example, involves an intuition for magnitudes and the capacity to activate it in a very automatic way (Dehaene, 1992; Verguts and Fias, 2008; Hyde, 2011). Moreover, the capacity to retrieve arithmetic facts appears to be a very automatic process as well (Domahs and Delazer, 2005; Zamarian et al., 2009). Such a specific deficit in the automatic access to information regarding, on the one hand, the ANS, and on the other hand, multiplication facts can account for the apparently discrepant deficits that are presented by H.V. A core deficit in subtraction operations of comparable difficulty.

On the other hand, the patterns of deficits presented by G.A. are suggestive of difficulties with a more executive and effortful processing of numerical representations as well as with some aspects of effortless processing. The spared performance of G.A. in all arithmetic operations is compatible with this view because the problems employed in the present study never had operands that were larger than two-digits, with which G.A. has had sufficient experience in the

past. In contrast, the transcoding task employed much larger numbers. This more complex part of the verbal numerical system is learned for the first time exactly in the grade that G.A. was attending during his assessment. This finding is suggestive that G.A. still needs substantial executive resources to employ correctly the transformation rules that are necessary to transcode those numbers (Barrouillet et al., 2004; Camos, 2008; Moura et al., 2013). More detailed analysis of G.A.'s poor transcoding performance reveals no evidence for a deficit in knowledge about the structure of the Portuguese verbal number system. In contrast, G.A.'s error pattern is indicative of severe problems with phonological representations, concentration and monitoring capacity. Accordingly, orally presented word problems can also be more challenging for G.A. because a good capacity in verbal working memory is necessary to select relevant information from these problems and then operate with them until the correct result is obtained.

Support for this interpretation of H.V. and G.A. endophenotypes comes also from the analysis of the Five-digits Test results (see Figure 2). The Five-digits Test is well-suited to perform this comparison because the stimuli and task context are preserved, while the cognitive demands in terms of automatic and controlled processing vary (see also van der Sluis et al., 2004). On the one hand, H.V. presents difficulties with speeded counting but does not present difficulties with the inhibition- and shifting- demanding tasks. G.A., on the other hand, encounters difficulties in all aspects of the task, which requires both automatic and controlled processing. G.A.'s pattern of performance in the Five-digits Test is similar to the pattern observed by van der Sluis et al. (2004) on an equivalent numerical task in children with math learning difficulties and both math and reading learning difficulties. Interactions between processing speed and working memory impairments have been observed in several studies of both typically developing children (Berg, 2008) and children with math learning disability (Bull and Johnston, 1997). Moreover, disorders

of automatization and procedural learning have also been implicated in learning disabilities of both reading (Menghini et al., 2006) and arithmetic (Lonnemann et al., 2011). Our results suggest that, in some cases, difficulties can be more related to the automatic or effortless processing, with possible compensation through more controlled strategies (H.V.), while in other cases, difficulties could be mixed or impairing more heavily controlled forms of processing (G.A.).

Overall, these results suggest that the search for endophenotypes could be more complex than originally expected, but not useless. In contrast, endophenotypes could be the only way to disclose more precise details on the nature and extension of mathematics difficulties. The current models of mathematics difficulties (e.g., Rubinsten and Henik, 2009) treat the different subtypes of math difficulties as members of a class of disorders that have different natures, which are nevertheless at more or less the same hierarchical level of organization of the cognitive system. This model has been proven to be useful but requires better specification.

One might consider the role of good executive functioning resources as a compensatory mechanism in developmental disorders. Johnson (2012) has proposed a role for executive functions in compensating for developmental neurogenetic impairments. According to this view, impairments in more basic and modularly organized aspects of information processing, such as phonological processing and number sense, can be compensated for if they are not sufficiently severe or if the individual has good executive functioning resources. The expression of symptoms that lead to diagnosis would occur in cases in which specific processing deficits are severe or when executive functioning resources are not sufficient to meet the environmental demands. The pattern of deficits presented by H.V. and G.A. are in line with these arguments.

While H.V. was able to mobilize resources from executive functions and compensate for many of her deficits in number processing, the same could not be observed in the case of G.A.

Moreover, H.V.'s case also suggests that, in addition to executive functions, a more basic level of task automatization should be considered to be a bridge between domain-specific and domain-general cognitive impairments that contribute to math learning difficulties. This topic has received less consideration in the literature (however, see van der Sluis et al., 2004; Chan and Ho, 2010).

Automatic and controlled processing are two dimensions of cognitive abilities that interact with domain-general and -specific factors, and the neurobiological basis of these processes should also be examined in more detail. Contemporary models of skill learning and automatization assume that, in the initial steps of learning, higher demands on processing are imposed over the fronto-parietal circuits that underlie cognitive control (Schneider and Chein, 2003). With practice, the typical focus of activity is shifted from anterior cortical regions to posterior ones and to the striatum. Another assumption is that this anterior-to-posterior shift in activity is domain-general because this circumstance has been observed with several motor and cognitive tasks. The extant literature largely supports these assertions (Patel et al., 2012). Similar observations have been made in the domain of numerical cognition. Interference effects in a number-size interference task are related to activation in frontal areas, while the distance effect is associated with activation in parietal areas, including the intraparietal sulcus (Kaufmann et al., 2005). Learning arithmetic facts is followed by a shift of the activation focus from frontal and intraparietal areas to the left angular gyrus (Zamarian et al., 2009). Developmentally, children usually activate more widespread areas during mental calculations, including frontal regions (Kawashima et al., 2004; Rivera et al., 2005). In adults, the focus of activity is more concentrated on posterior areas (Kaufmann et al., 2008, 2011; Klein et al., 2009).

Available evidence on the neurocognitive underpinnings of skill learning and automatization allow us to tentatively predict structural-anatomical correlations of automatic and controlled processing impairments in math learning difficulties. Numerical-specific automatic processing deficits, such as the deficits presented by H.V., should be related to impairments in parietal areas, including connections to the intraparietal sulcus. A broader pattern of dysfunction, encompassing the frontal areas, should be observed in cases such as G.A., in whom controlled processing is also impaired. Obviously, math learning difficulties that are associated with dyslexia also imply malfunctioning of perisylvian areas.

Results of the present paper have important implications for future research. The first implication is the need to include both domain-specific and domain-general measures to fully describe the range of manifestations and impairments in math learning difficulties (Cowan and Powell, 2013). Moreover, the neuropsychological test batteries that are used to assess math learning should fairly measure both automatic or effortless processing and effortful or controlled processing. Because comorbidity with ADHD can explain impairments in working memory and executive functions, ADHD symptoms should also necessarily be controlled. Otherwise, it is not possible to draw straightforward conclusions on how working memory/executive functions determine more general performance difficulties compared to numerical-specific deficits (Willburger et al., 2008). Tasks that assess working memory and executive functions should also be presented in two formats, using non-numerical and numerical stimuli (Raghubar et al., 2010). Another important implication is the need to assess more automatized number processing, such as RAN.

Finally, we believe that the present study contributes to underline the importance of single-case research in clarifying the role of distinct endophenotypes in dyscalculia research.

In the present study, we demonstrated that automatic and controlled information processing is one valid and necessary axis of investigation when characterizing the multitude of cognitive deficits that are associated with math difficulties, which can conciliate apparent discrepancies between individual and typical endophenotypes with respect to math difficulties. This approach constitutes a more general level of description of cognitive deficits as that originally adopted by other authors in previous studies. In summary, phenotypic manifestations of learning disabilities are compounded by impairments in both specific and general information processing mechanisms. Math-specific factors, such as number sense, and math-nonspecific cognitive factors, such as phonological processing, interact with general aspects of information processing, such as controlled processing and automatization. Math-specific and more general information processing deficits and automatic and controlled information processing deficits therefore represent orthogonal but interacting dimensions of the same disorder. In this sense, symptoms would be apparent when general or specific compensatory mechanisms are overloaded or not sufficient to meet the environmental demands in cases of more specific impairments. Impairments in restricted, specific domains could explain the unique difficulties, while impairment in more general mechanisms could be related to the degree and form of phenotypic expression via compensatory mechanisms.

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