THE PROCESSING OF NUMBER SCALES BEYOND WHOLE NUMBERS IN DEVELOPMENT: DISSOCIATIONS IN ARITHMETIC IN TURNER'S SYNDROME

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Abstract

The arithmetical skills in two children with Turner's syndrome [TS], each the focus of a case study, were analyzed in whole numbers and other number scales that have not been systematically explored previously, fractions, decimals, percentages and negative numbers. The intention was to identify the fractionation of arithmetical skills. The two girls with TS showed dissociations of arithmetical skill in the calculation system of whole numbers that support its modular organization. Fractionation of skills was observed in some components of the other number scales suggesting an analogous organization within these scales. The operational specificity of impairment within number scales but not others argued against a unitary arithmetical system but rather for autonomous operational scales within distinct number scales. A general model of arithmetic is proposed.

Keywords: Turner's Syndrome, models of arithmetical skills, number processing, calculation, dissociations

Introduction

There is unextensive literature on the functional architecture that characterizes normal development of whole numbers, with models outlining the step by step processing (e.g., Cipolotti, 1995; Cipolotti & Butterworth, 1995; Dehaene, 1992; McCloskey, 1992; McCloskey, Caramazza & Basili, 1985; McCloskey, Sokol & Goodman 1986). In contrast, limited systematic research is available on the processing of other numbers scales, such as fractions, decimals, percentages and negative numbers, and on their compatibility to that of whole numbers. The current study uses a case study methodology to identify the fractionation of arithmetical skills in two girls with Turner's syndrome [TS]. The intention is to identify the processes involved in using fractions, decimals, percentages and negative numbers. In addition, a direct comparison between these scales with that of whole numbers will establish whether the latter scale is a precursor of successful mastery of all or some of the other.

Models of Normal Arithmetical Skill

The development of the arithmetical system commences once children become familiar with the concept of counting. They learn and use counting principles before they acquire more complex, higher-order arithmetical skills. Arithmetical operations are then learned in terms of performing operations on sets. Addition, for instance, is perceived as joining two sets together to provide an addition sum. Arithmetical facts in the form of single-digit problems are introduced before multi-digit problems. In turn, addition and subtraction acquisition precede multiplication and division. Although addition and counting can be used to perform multiplication, multiplication in terms of one-to-many correspondence is not achieved until later on in arithmetical development. Division becomes familiar once the concept of "half" is introduced early on and plays an important role in understanding fractions and decimals. Acquisition of multiplication, division and of fractions and decimals is achieved once children understand ratios and proportions (e.g., $\frac{2}{4}$ is the same as $\frac{4}{8}$) (see Butterworth, 1999, 2005).

Available models describe the structural architecture of the developed arithmetical system of whole numbers. Following studies of acquired dyscalculia in adults, McCloskey and his colleagues (McCloskey, 1992; McCloskey et al, 1985; McCloskey et al, 1986) proposed a model that incorporated distinct subsystems each responsible for particular arithmetical abilities. Specifically in their model, they draw a distinction between number processing and calculation. In number processing, two distinct mechanisms for comprehension and production of numbers deal with verbal and Arabic numbers separately. Lexical and syntactic processing is distinct within each mechanism. The lexical system processes the individual elements in a number, and the syntactic system the order and structure of the elements. Tasks such as reading, writing, transcoding and comparison of numbers in terms of magnitude are accomplished through the number processing system. Within the calculation system, there are three mechanisms. The operation processing mechanism mediates the processing of symbols and words related to symbols (e.g., plus). The procedures mechanism applies rules for the step by step processing of operations such as addition, subtraction, multiplication and division, and the facts mechanism stores and retrieves facts (e.g., 2 x 2 = 4). A core feature of McCloskey's model is the semantic processing of numbers, with an input converted into an abstract semantic representation before being further processed in the calculation system or produced as a number. So for the number "235" the semantic form generated is {2}10EXP2, {3}10EXP1 and {5}10EXP0, where {2}, {3} and {5} are quantity representations and 10EXPn specify the power of 10 (10 in the n power). This component is common to both number processing and calculation systems.

According to McCloskey model, impairment in the semantic representations system interferes with the ability to perform all arithmetical production tasks (e.g. read numbers, verbal or written production of a calculation solution). This idea was challenged by models that have a different approach to number semantics (e.g. Cipolotti, 1995; Cipolotti & Butterworth, 1995; Dehaene, 1992). For instance, the non-semantic analogue magnitude representation system proposed in the triple-code model (Dehaene, 1992) represents a mental number line where numbers are signified as activations on different areas on the line. These are translated into quantity codes and retrieved, produced in digits (visual Arabic number form) or in words (auditory verbal word frame). According to the triple-code model, reading, writing, transcoding of numbers and simple procedures that can be solved with rote memory are not processed by the analogue magnitude representation system. In this case, impaired ability to read or write numbers should not affect the ability to produce the answer, for instance, to a complex calculation, which is processed by the latter system. There is no conclusive evidence to date with regards to whether the analogue magnitude representation system is an alternative or additional system to McCloskey's semantic representation system. Extended versions of the McCloskey model introduced asemantic routes for number processing (Cipolotti, 1995, Cipolotti & Butterworth, 1995). In these routes, a syntactic frame is formed through syntactic processing which is filled in with single-digit representations through lexical processing and gets converted directly into a spoken number word.

McCloskey's model proposes a modular architecture for the arithmetical system, with distinct subsystems, modules, responsible for different arithmetical processes (Fodor, 1983). These subsystems are semi-independent in that a defect in one subsystem, e.g. number processing, could affect a number of arithmetical skills interrelated with this system, e.g., reading and writing numbers, with other subsystems remaining intact, e.g., performing procedures (Temple, 1989; 1991; 1992).

To our knowledge, there are no models that describe the functional organization of other number scales such as, fractions, decimals, negative numbers and percentages. Studies have shown that fractions and decimals are processed componentially with the real value of these numbers not accessed automatically, and that understanding of these scales is based on understanding of integers. So, for instance, when making magnitude comparisons of fractions, the numerators are compared with each other and the denominators are compared with each other as separate integers without focusing on the numerical value of each fraction (Bonato, Fabbri, Umiltà & Zorzi, 2007; Stafylidou & Vosniadou, 2004; Vamvakoussi & Vosniadou, 2004, 2007). Similarly, negative numbers consist of components, polarity and magnitude, each represented separately. Thus, understanding of positive numbers is a prerequisite for understanding the concept of negative numbers (Fischer and Rottman, 2005; Ganor-Ster, Pinhas, Kallai & Tzelgov, 2010; Tzelgov, Ganor-Stern & Maymon-Schreiber, 2008). This would lead to an interrelationship between whole numbers and fractions, decimals and negative numbers. Although these studies support the role of whole numbers in the development of additional number scales, they nevertheless do not describe the step by step processing that is involved in mastering these number skills.

Arithmetical skills in Turner's Syndrome

Turner's syndrome [TS] is a sex chromosome disorder that affects females with an incidence of approximately 1 in 2000 live female births (Stochholm, Juul, Juel, Naeraa & Gravholt, 2006). It results from complete or partial loss of the second X chromosome. Approximately half of females with TS are diagnosed with the classic 45XO and the other half with some other karyotype, including mosaicism (45XO/46XX), isochromosome of Xq, ring X, marker chromosome X, and Y chromosome fragment (Bharath et al, 2010; Gadhia et al, 2014). Females with TS have a physical profile that includes among other characteristics short stature, lack of estrogen, absence of secondary sexual characteristics and infertility (Turner, 1938).

Girls with TS have normal intelligence with an advantage on verbal IQ [VIQ] over performance IQ [PIQ] (Hong, Kent & Kelser, 2009; Hong & Reiss, 2012; Saad et al, 2014). Language is generally spared in TS with intact reading skills, receptive and expressive language and enhanced phonological skills but impaired oral fluency and speeded naming (Temple, 2002; Temple & Shephard, 2012; see Hong et al, 2009 for a review). Nonverbal cognition, visuo-perceptual and visuo-constructional (Green et al, 2014; Hong & Reiss, 2012) and executive skills are areas of core weakness in individuals with TS. Tactile-spatial ability has been reported as intact (Temple & Carney, 1995). Within executive function, verbal inhibition has been described as impaired (Temple, Carney & Mullarkey, 1996; Temple & Shephard, 2012). Set shifting, response monitoring and planning skills have been reported as both intact (Murphy & Mazzocco, 2008; Temple et al, 1996) and impaired (Kirk, Mazzocco & Kover, 2005; Loesch et al, 2005).

Math performance difficulty is another characteristic of the TS profile (see Baker & Reiss, 2016, for a review). The literature on arithmetic in TS has concentrated mainly on whole numbers. Rovet (1993) described the psychoeducational profile of girls with TS and reported difficulty in arithmetic in these children. Their weakness does not emerge early in their arithmetical development as girls with TS show intact counting skills (Mazzocco et al, 2006; Murphy, Mazzocco, Gerner & Henry, 2006; Temple & Sherwood, 2002). Slow response times [RTs] have been reported but did not always reach statistical significance (Bruandet, Molko, Cohen & Dehaene, 2004; Simon et al, 2008; Temple & Sherwood, 2002).

More detailed studies showed that girls with TS have difficulties in some but not other areas of arithmetic. This supports the modular arithmetical system proposed by McCloskey in his model. Number processing skills have been described as intact. Girls with TS are therefore able to read and write numbers, to transcode numbers from one number system to another and they can make magnitude and parity judgements (Murphy et al, 2006; Rovet, Szekely & Hockenberry, 1994; Temple & Marriott, 1998; Temple & Sherwood, 2002).

Consistent with the McCloskey model, several studies have reported a selective impairment in the calculation skills of TS, in line with separate operation mechanisms in arithmetical facts and procedures systems. Girls with TS are accurate at retrieving addition and subtraction facts but they take longer (Bruandet et al, 2004; Butterworth et al, 1999; Rovet et al, 1994; Temple & Marriott, 1998; Temple & Sherwood, 2002). Multiplication fact retrieval has been reported as impaired on different measures: accuracy (Rovet et al, 1994; Temple & Sherwood, 2002), speed (Butterworth et al, 1999) and pattern of errors (Temple & Marriott, 1998; see Bruandet et al, 2004, for a contradiction).

Girls with TS also make errors in performing procedures. They are significantly worse than controls at multiplication and division operations, making errors in employing the correct algorithm. Addition and subtraction performance are both intact (Temple & Marriott, 1998). Other studies have assessed procedural skills through subsets of battery tests. These studies show impairment in girls with TS when compared to age matched controls. This disappears when they are compared to IQ matched controls (Mazzocco, 1998; Mazzocco et al, 2006; Mazzocco & Hanich, 2010; Murphy & Mazzocco, 2008). There remains an issue of what constitutes an appropriate IQ match for children with uneven subtest profile and VIQ/PIQ discrepancies (Temple & Carney, 1993; Temple, 2002).

In contrast to the extensive literature on whole numbers in girls with TS, there is little systematic research on fractions and decimals. Girls with TS showed impairment in recognizing and understanding fractions and decimals (Rovet et al, 1994) although identification of decimals has also been reported as intact (Mazzocco, 2001). Within calculations, Murphy and Mazzocco (2008) showed that girls with TS were accurate in performing simple arithmetic operations with decimals and fractions. To date, the data available on these number scales is based on subtests of battery tasks. The detailed exploration that takes place in this study will generate a more comprehensive picture of their organization and modularity. In addition, using tasks that are comparable in design to those used with whole numbers will allow direct comparison between scales and will indicate if any scale is essential precursor for the mastery of others.

The current study explores the arithmetical skills of two girls with TS. The objective is to identify single and double dissociations in the two cases that illustrate the organization of the developing arithmetical system. This study utilized a case study methodology. Therefore, any dissociation in arithmetic will not necessarily generalize and may not be encountered as a common feature of TS. Specifically, the study aims to a) identify any fractionations of skills in the whole number scale to interpret in relation to McCloskey's model and or other models of normal arithmetical skill, b) investigate unexplored number scales utilizing the components included in the McCloskey model and assessing their modular nature, and c) generate models that describe the organization of these scales and the connections between them. It is expected that the two girls will show similar patterns of strengths and weaknesses in processing whole numbers as observed in past research (Bruandet et al, 2004; Butterworth et al, 1999; Mazzocco et al, 2006; Murphy et al, 2006; Rovet et al, 1994; Simon et al, 2008; Temple & Marriott, 1998; Temple & Sherwood, 2002) that can be interpreted in terms of the modular organization of the McCloskey model. Specifically, we expect to find preserved accuracy in counting and number processing skills. Ability within calculation is expected to be selective, with intact addition and subtraction fact retrieval and procedural skills, and impaired multiplication and division in both calculation systems. Investigations of competence on the other number scales are exploratory in nature and it is therefore difficult to make any predictions. However, if whole numbers skill is a prerequisite to the development of other number scales, then we expect similar areas of weakness to emerge in other number scales also. Furthermore, a connection between different number scales is anticipated. For instance, when making magnitude comparisons between positive and negative numbers, the whole numbers scale is assumed to connect to the negative numbers scale to allow comparison. The nature of the connections is part of the focus in this study.

Method

Participants

TS Individuals

Two children with TS aged 11;3 and 12;0, each was the focus of a case study. Both girls attended mainstream schools. NB has the classic 45XO karyotype. Assessment with WASI showed normal intelligence with a VIQ of 99, PIQ of 99 and FSIQ of 100. KJ has small ring X chromosome (45X/46X,r(X)) karyotype. 45XO was found in 40% of cells and the ring chromosome in 60% of cells. Assessment with WASI showed normal intelligence with a VIQ of 108, PIQ of 89 and FSIQ of 99.

Typically Developing [TD] Group

The performance of the girls with TS was compared to that of a TD group. The TD group consisted of 20 children: 9 males and 11 females. An age matched TD group was considered as more appropriate group comparison for the two TS girls in the study. They were selected based on their chronological age and school year. The cases were not explicitly matched to the TD group on the basis of IQ, given the difficulty in deriving such matches in a valid form for syndromes with uneven subtest profile and PIQ/VIQ discrepancies (Temple & Carney, 1993; see Temple, 2002 for discussion). Estimated mean FSIQ from the short WASI (Wechsler, 1999) was 106. Three exclusion criteria were applied for the TD group selection.

Children were excluded if they had a scaled score of 6 or below in one of the two subtests of the short version of the WASI, Vocabulary and Matrix Reasoning; a reading age 18 months below their CA in the Word Reading subtest of the British Ability Scale-Second Edition [BAS-II] (Elliott, Smith & McCulloch, 1996); or an equivalent age of 18 months below their CA in the Numerical Operations subtest of the Wechsler Individual Achievement Test - Second Edition [WIAT-II] (Wechsler, 2005).

Experimental Tasks

The experimental tasks administered in this study aimed to assess all components of the arithmetical system. The tasks employed addressed the core features of the McCloskey model, number processing and calculation, and their subsystems. Although the structure of the tasks followed the McCloskey model, tasks assessing these components are used to assess components of arithmetic in all other models that describe the normal development of arithmetic (e.g., Dehaene, 1992; Cipolotti, 1995; Cipolotti & Butterworth, 1995). The same categorization of tasks was followed for the other number scales. If whole numbers is a prerequisite for the development of other numbers scales the same form of tasks allows direct comparisons between these.

The tasks included paper and pencil, and computerised tasks. The computerised tasks were programmed on SuperLab 4. All stimuli appeared on the centre of the screen and responses were recorded by a build-in microphone or a button-box. Stimuli in the tasks with microphone input stayed on the screen for 4 sec regardless of the participant's response. In the button box input tasks, the stimuli disappeared from the screen immediately after a response. There was an interstimulus interval of 2 sec for both microphone and button box tasks. Computerised tasks were used to attain accuracy and RT measures. The experimenter noted the strategies participants employed on tasks of procedural calculations. In the non-computerised tasks, RTs were recorded with a stopwatch. In the majority of the tasks, the timer started immediately at the end of the instructions and stopped immediately when writing was completed on the last item of the task. Any variations in the stopwatch procedure are described within the task. The experimenter followed the exact same procedure for the two TS girls and the TD group avoiding as much as possible any group biases. There were no video recordings for any of the sessions. Given the imprecision and variations within stopwatch recordings, all RTs were rounded up to the nearest second (e.g. Temple & Marriott, 1998; Temple & Sherwood, 2002).

Whole Numbers

According to Butterworth (1999), counting is the basis for more advanced arithmetical development. Counting tasks were therefore included to assess if impairment of this skill could have influenced advanced arithmetical development in the two TS girls.

Counting

Forward and backward counting. Participants were asked to count from 1 to 20 forwards and backwards. RTs were recorded with a stopwatch. The timer started when the experimenter said "start now" and stopped when the participant said "20" for the forward counting and "1" for the backward counting.

How many? Twelve arrays of coloured stars were presented for counting aloud. There were four arrays of 1-10 stars, four of 10-20 and four of 20-30 stars. RT was not recorded in this task.

What comes next or before? (based on Paterson, Girelli, Butterworth & Karmiloff-Smith, 2006). Sixteen numbers were spoken aloud by the experimenter and participants had to say the number that followed or preceded (e.g., What comes after 79?). The list consisted of four single-digit and 12 two-digit numbers. RTs were recorded for each trial using a stopwatch, timed immediately after the question was given and stopped with the first response.

Number processing Tasks:

Production of Whole Numbers (based on Temple, 1989): Arabic and verbal numeral production systems in McCloskey's model were explored with reading, writing and transcoding Arabic numbers, and number words. Lexical and syntactic processing was assessed from responses on the numerical elements in the number (e.g., " 3" and "4" in "34") and the order and structure of the element in the number (e.g., "3" in the tens class and "4" in the ones class).

Reading Arabic numbers and number words (computerised tasks). A list of 40 numbers in Arabic form, 10 each of length 1-4 digits (e.g., 3, 78), appeared in the centre of the screen one after the other. Participants had to read aloud each number. Two practice trials preceded the test trials. The same set of numbers was presented in a different order in number word form (e.g., seventy-eight, three) for reading.

Writing Arabic numbers and number words. Participants were required to write in Arabic number form 16 of the 40 numbers presented for reading (four each of length 1-4 digits). The numbers were spoken aloud by the experimenter. RTs were not recorded in this task. In similar fashion, participants were required to write in number word form the same 16 numbers, presented in a different order.

Transcoding Arabic numbers into number words and number words into Arabic numbers. Twelve numbers from the set of 40 used for reading were presented in Arabic number form and participants were asked to write the equivalent number words (e.g., "34" to thirty-four). In similar fashion, a different set of 12 numbers was presented as number words and participants were asked to write the equivalent number in Arabic number form (e.g., "ninety-nine" to 99). An example was given. Both lists included three numbers each that were single-digit, two-digit, three-digit and four-digit. RTs were not recorded in these tasks.

Comprehension of Whole Numbers (Computerised Tasks): Magnitude comparison and parity judgements of Arabic numbers assessed Arabic numeral comprehension. Verbal numeral comprehension was explored with parity judgements of number words. Lexical and syntactic processing was addressed in similar fashion to the production tasks.

Magnitude comparison of Arabic numbers (derived from Temple, 1989). A set of 20 two-digit pairs of numbers was presented one at a time and participants had to indicate the larger of the two numbers in each pair (e.g., 84 42). Response was recorded by pressing on the right or left button in a button box.

Parity judgement. Ten numbers in Arabic form (e.g., 52) and 10 in word form (e.g., thirteen) (half even and half odd) were displayed randomised one at a time. Participants had to judge if each number was even or odd number by pressing an orange button named "ODD" or a white button named "EVEN" on the button box. There were two practice trials on both comprehension tasks.

Calculation Tasks: The following tasks assessed the three calculation subsystems proposed by McCloskey in his model.

Symbols of Operations

Participants were presented with a set of 20 simple arithmetical operations each with the operation symbol missing (e.g. 8 $\boxed{}$ 2=4). Participants had to write the missing symbol. RT was not recorded in this task.

Fact Retrieval for Whole Numbers (derived from Temple, 1991; Warrington, 1982)

The 45 single-digit addition operations (using all combinations of digits 1-9, paired all with smaller digits, e.g., 8+2) were displayed one at a time and participants had to produce the answer aloud as quickly as possible. In a similar way, 28 single-digit subtraction (digits 2-9, paired with all smaller digits, e.g., 7-3) and 64 single-digit multiplication operations (all combinations of 2-9 in both orders, e.g., 3x4 and 4x3) were also presented.

Procedural Knowledge of Whole Numbers

Sixteen addition operations required producing the answer in written form. The operations varied in difficulty, from single-digit (e.g., 26+3=) to multi-digit with (e.g., 46+76=) and without carrying over (e.g., 13+25=) to more complex multi-digit with (e.g., 769+22=) and without carrying over (e.g., 125+233=) numbers. The problems were presented in random order on a single sheet. RTs were recorded for the whole task with a stopwatch. In similar fashion, there were 16 operations for subtraction (e.g., 35-6=), 14 for multiplication (e.g., 324×12=) and 10 for division (e.g., 84÷21=). All operations were presented in vertical form.

Fractions, Decimals, Percentages

Number Processing Tasks:

Production of Fractions, Decimals and Percentages

Reading and writing fractions, decimals and percentages (computerised tasks). Three lists of 40 fractions (e.g., $\frac{1}{8}, \frac{7}{10}$), 40 decimals (e.g., 0.78, 0.06) and 40 percentages (e.g., 3%, 49%) were administered in Arabic form for reading. There were two practice trials. Sixteen of each of the 40 fractions, decimals and percentages were spoken aloud by the experimenter for writing in Arabic form. There were no RTs in the writing tasks.

Transcoding of fractions, decimals and percentages. Sets of 12 fractions, 12 decimals and 12 percentages were presented and participants had to write down the equivalent in one of the other number systems, specified for each task (e.g., "$\frac{3}{10}$" to 0.3, "$\frac{3}{10}$" to 30%). RTs were not recorded.

Comprehension of Fractions, Decimals and Percentages

Magnitude comparison of fractions, decimals and percentages (computerised tasks). Twenty pairs of fractions were presented one at a time and participants had to indicate with the button box which of the two fractions in each pair was the larger (e.g., $\frac{1}{16}$ $\frac{4}{5}$). In similar fashion, there were 20 pairs of decimals (e.g., 87.1 3.49) and 20 pairs of percentages (e.g., 70% 24%). Practice trials preceded the test trials.

"Odd one out" in numbers task. Twenty sets of four numbers were administered, a mixture of fractions, decimals and percentages in each set. Each set consisted of three numbers of equal magnitude and one of different magnitude. Participants had to circle the odd one out in magnitude number (e.g., 0.25 $\frac{1}{4}$ $\frac{2}{8}$ 35%). RTs were recorded for the whole task with a stopwatch.

Calculation Tasks:

Fact Retrieval for Fractions and Decimals (Computerised Tasks)

Ten additions (five fractions and five decimals) were presented. Participants had to say aloud the answer to the addition as quickly as possible. The 10 addition operations did not require procedural knowledge to produce the answer (e.g., $\frac{1}{2}+\frac{1}{2}$, 0.5+0.5). In similar fashion, there were 10 subtraction and 10 multiplication facts for fractions and decimals.

Procedural Knowledge of Fractions, Decimals and Percentages

Addition and subtraction of fractions. Twenty-four addition and 24 subtraction problems for fractions (e.g., $\frac{1}{5}+\frac{1}{5}=$, $\frac{18}{23}-\frac{15}{23}=$) were administered in a written form starting from simple single-digit and two-digit fractions with common denominators and different denominators and proceeding to mixed numbers with common denominators and different denominators. RTs were recorded for the whole task with a stopwatch.

Addition and subtraction of decimals. Twenty addition (e.g., 9.5+0.2=) and 20 subtraction problems (e.g., 48.03-31.7=) for decimals were administered in a written form starting from addition of decimals with the same number of decimal places and proceeding to those with different number of decimal places, and whole numbers. RTs were recorded for the whole task with a stopwatch. All operations were presented in a vertical form.

Multiplication of decimals. Twenty multiplication problems for decimals included ten decimals that were multiplied by 10 (e.g., 3.2×10=) and 10 by 100 (e.g., 0.65×100=). RTs were recorded for the whole task with a stopwatch. All operations were presented in a vertical form.

Calculation of percentages and of fractions. Nine single-digit percentages (e.g. 7% of 76) and 15 two-digit percentages were presented and participants had to find the percentage of eight single-digit numbers (e.g. 19% of 3), eight two-digit numbers and eight three-digit numbers. Similarly, 16 trials required calculation of fractions of numbers (e.g. $\frac{5}{8}$ of 50). These consisted of single-digit fractions, two-digit fractions and single-digit numerator/two-digit denominator fractions. RTs were recorded for the whole task with a stopwatch.

Negative Numbers

Number Processing Tasks:

Production of Negative Numbers

Reading and writing negative numbers (computerised task). A list of 40 negative numbers in Arabic number form, 1-4 digits in length, was presented for reading (e.g., -10, -492). Sixteen of the 40 negative numbers (four each of length 1-4 digits) were spoken aloud by the experimenter and participants had to write them in Arabic form. RTs were not recorded in the writing task.

Comprehension of Negative Numbers

Put numbers in order. Twenty sets of five randomly sequenced negative numbers were presented and participants were required to rewrite the numbers in order of magnitude from smallest to largest (e.g., -37, -20, -47, -82, -85). The numbers varied between -1 and -100 and each number appeared only once within the 20 sets. RTs were not recorded.

Magnitude comparison of positive and negative numbers (computerised task). Twenty two-digit pairs consisting of positive and negative numbers were presented. Participants had to judge which of the two numbers was the largest in magnitude with a button box response (e.g., 24 -32).

Calculation Tasks:

Procedural Knowledge of Negative Numbers

Addition and subtraction of positive and negative numbers. Twenty-five addition and 25 subtraction operations for positive and negative numbers, of 1-3 digits, were presented (e.g., (-37)+55= and (-36)-(-65)=). Participants were required to write the answer. All the possible combinations of positive and negative numbers were included. RTs were recorded for the whole task using a stopwatch.

Procedure

For the cases, assessment took place in four or five sessions of 2-3 hours in their house mainly at weekends. Assessment for the TD group was for 40 min once a week for approximately 10 weeks in a quiet room in their schools. The order of the tasks and procedure were kept the same for all participants. The main focus of the study was to identify dissociations in the arithmetical skills of two girls with TS to describe the organizations of the arithmetical system. There was no intention to intervene with their arithmetical development and therefore, there was no attempt in improving their arithmetical ability by training them on tasks where weakness was observed. There is no information available from teachers with regards to extra training in arithmetic the two girls with TS might have received at their mainstream school. Nevertheless, we anticipated any additional training to only have strengthen rather than weaken their ability to perform some tasks. All children received book tokens as a reward for their participation.

Analysis of Tasks

Computerised Tasks

For the microphone tasks, noise trials were eliminated and a scaled score for accuracy was calculated proportionately taking account of the number of items successfully presented. Scaled scores were calculated by dividing the number of correct responses by the total number of trials successfully presented (eliminating noise trials), and multiplying this by the overall number of trials (including noise trials).

In the computerised tasks the following RTs were dropped from analysis: incorrect responses; below 50 msec (anticipations); more than 3SD above the mean (distorting outlier); and noise trials (e.g., trials activated from background noise and responses preceded by background noise that might have activated the stimulus). When fewer than eight RTs were available from a child in a task, this child's RT data was eliminated from the analysis. In tasks with small number of trials, RTs were combined to generate sufficient number of trials. RTs were therefore combined in the fact retrieval for fractions and decimals (addition, subtraction and multiplication).

Non-Computerised Tasks

For non-computerised tasks, the main objective was to identify the speed of response on attempted trials, how long it would take a child to perform each completed trial. Omitted trials, where there was no attempt in producing an answer, were therefore excluded from the RT analysis. Scaled RTs represented an approximate RT for each trial in tasks where RT was recorded for the whole task. These were calculated by dividing the total RT for each task by the number of stimuli in the task, after taking away omissions.

The data for the study were analysed using the modified t-test developed by Crawford and Howell (1998). This test allows comparison of an individual's performance against that of a small normative group and estimation of the rarity or abnormality of the individual's performance (Crawford & Howell, 1998; Crawford & Garthwaite, 2002). Dissociations between performances were analyzed following Crawford, Garthwaite and Porter's (2010) method. This method assesses if a case's performance meets criteria for dissociation taking into consideration the correlation between tasks in the normative group. Although multiple comparisons were performed, relative to a small sample size, Bonferroni corrections were not applied to decrease Type I error because this increases Type II errors (Perneger, 1998). TS is a genetic disorder and it is predicted on the basis of the literature that where there is a difference the girls with TS will perform worse than the TD group. Nevertheless, a more conservative two-tailed analysis was performed.

Results

Whole Numbers

Counting

Table 1 and 2 show statistical analysis and results for accuracy and RTs in counting. Both NB and KJ showed intact performance, being able to count forwards and backwards and had good knowledge of the sequence of numbers. NB was significantly slower than the TD group in counting and performed more slowly than any of the controls. KJ was as fast as the TD group.

[Tables 1 about here]

Number Processing of Whole Numbers:

Table 2 and 3 show results for accuracy and RTs in production and comprehension of whole numbers. Both NB and KJ were able to produce numbers in both Arabic and number word form as accurate and as fast as that of the TD group. Transcoding and number comprehension performance was also intact for accuracy. Both girls needed significantly more time than any of the TD children to make magnitude comparisons but not for parity judgement.

[Tables 2 and 3 about here]

Calculation of Whole Numbers:

Symbols of Operations

NB identified 18 (90%) and KJ all (100%) missing symbols of the 20 operations correctly. The TD group identified 19.85 (99.3%) (SD=0.37) symbols. Although NB identified significantly fewer symbols than the TD children ($t=4.88$, $p<0.001$), her performance was nevertheless of high accuracy. KJ was as accurate as the TD group.

Fact Retrieval for Whole Numbers

NB and KJ used finger and verbal counting strategies to produce some answers, evidence that fact retrieval was not occurring in some cases but fact deduction. Table 4 shows the results for correct responses on fact retrieval tasks for whole numbers including trials where finger counting occurred and then excluding those trials.

[Table 4 about here]

Both NB and KJ were able to generate correct responses to all problems for addition and subtraction facts without errors and most problems for multiplication facts, performance as accurate as the TD group. In a further analysis of accuracy eliminating trials where fact deduction had occurred, NB retrieved significantly fewer addition, subtraction and multiplication facts than any of the TD children. Although KJ retrieved significantly fewer addition and subtraction facts than the TD group mean, she nevertheless performed at the bottom end of the TD range. Her performance in retrieving multiplication facts was as accurate as the TD group.

NB was significantly slower than any of the TD children in multiplication but not in addition and subtraction when generating answers using finger counting and retrieval. A less conservative one-tailed analysis, taking into consideration the slow RTs in both counting and fact retrieval reported previously in studies with TS girls (e.g., Bruandet et al, 2004; Temple & Sherwood, 2002), indicated speed impairment also in generating addition answers (p=0.05). In contrast, KJ was as fast as the TD group. Neither NB nor KJ was significantly different from the TD group regarding RTs on any of the fact tasks when analysis was based on retrieved only trials (see Table 5).

[Table 5 about here]

Procedural Knowledge of Whole Numbers

Tables 4 and 5 also show results for accuracy and RTs in solving addition, subtraction, multiplication and division operations. Figure 1 shows individual scores on the TD group's performance on all operations. NB's performance in addition operations was significantly worse and outside the TD group range but as fast as the TD group. KJ was as accurate and as fast as the TD group. There was no irregularity observed in the procedure KJ followed to add numbers. NB started her addition from right to left, carrying over numbers where appropriate. However, when the two addends had uneven number of digits she added the units with each other, the unit with the tens and the unit with the hundreds.

Both NB and KJ were significantly worse than any of the TD children in subtracting numbers. Both NB and KJ started their procedures from right to left but they both had difficulty in borrowing numbers. Like additions, when the number of digits was uneven, NB subtracted the bottom unit from the top unit, the bottom unit from the top tens, and the bottom unit from the top hundreds. When borrowing was required NB reversed the order of the numbers to make the subtraction possible (e.g. *517-8*="371", where she subtracted 8-7=1 instead of following the borrowing procedure). KJ also had difficulty with the borrowing procedure. When she had to borrow numbers she put a 0 in the answer claiming that the subtraction was impossible. Both NB and KJ were as fast as the TD group.

Neither NB nor KJ differed from the TD group in multiplication operations in both terms of accuracy and RTs. NB was as accurate as the TD group also in performing divisions. Although KJ solved significantly fewer division operations, she nevertheless performed at the bottom end of the TD range. They were both as fast as the TD group. NB counted up the divisor using her fingers to see how many times it went in the dividend. Some of the TD children also used her strategy. KJ said that she was judging how many times the divisor went into each number of the dividend.

The differences observed in NB's performance between operations fitted the criteria for classical dissociations for addition and division ($t(19)=3.68$, $p=0.002$), subtraction and multiplication ($t(19)=2.82$, $p=0.01$) and subtraction and division ($t(19)=4.67$, $p=0.001$). The difference in KJ's performance between addition and subtraction operations also fitted the criteria for classical dissociation, $t(19)=2.10$, $p=0.05$. All other dissociations did not reach significance ($p>0.05$).

NB showed a single dissociation between retrieving arithmetical facts and carrying out procedures of operations. NB had difficulty in retrieving multiplication facts but was able to carry out multiplications. This fulfilled the criteria for classical dissociation ($t(19)=6.94$, $p=0.001$).

[Figure 1 about here]

Fractions, Decimals, Percentages

Number Processing of Fractions, Decimals and Percentages

Table 6 shows accuracy in tasks of number processing for fractions, decimals and percentages. Table 7 shows RTs where available in these tasks.

[Tables 6 and 7 about here]

Both NB and KJ were as accurate as the TD group in reading and writing fractions and percentages. NB read significantly fewer decimals than any of the TD children, making syntactic errors (e.g., 0.78 read as "zero point seventy eight"). Her performance was intact in writing decimals. The difference in NB's scores between reading and writing decimals did not fulfil the criteria for a classical dissociation ($t(19)=0.07$, $p=0.94$). The dissociations however between reading fractions and decimals ($t(19)=2.19$, $p=0.04$) and whole numbers and decimals (Arabic numbers: $t(19)=2.11$, $p=0.05$ and number words: $t(19)=2.15$, $p=0.04$) were significant. The difference in scores between reading decimals and percentages was not possible to analyse as the TD group showed ceiling performance in reading percentages (constant). Although KJ read fewer decimals than the TD group this was not significant because of the variability in the TD group performance. She was accurate at writing decimals. Similarly, both NB and KJ transcoded less fractions into decimals and percentages correctly than the TD group performance not significantly different due to variability in the TD group. Speed of response was as fast as the TD group in producing fractions, decimals and percentages for both girls.

In the magnitude comparison tasks, both NB and KJ were as accurate and as fast as the TD group. Both girls with TS and the TD group had difficulty in performing the "odd one out" task. NB identified the numbers of different magnitude in 5 out of 20 sets (25%) and KJ in 7 out of 20 sets (35%). The TD group had a mean of 11.55 (57.8%) (SD=4.76 NB's and KJ's performance was not significantly different from the TD group because of the variability in the TD group performance (NB: t=1.34, p=0.20; KJ: t=0.93, p=0.36).

NB had a scaled RT of 8 secs and KJ of 4 secs. The TD group had a mean scaled RT of 8.40 secs (SD=2.26). Both NB and KJ were as fast as the TD group in identifying the odd one out number in each successful set (KJ: t=1.90, p=0.07).

Fact Retrieval for Fractions and Decimals

Table 8 shows scores for accuracy in fact retrieval for fractions and decimals. Both NB showed difficulty in retrieving fraction facts only producing 1 or 2 facts correctly. Nevertheless, only her performance on subtraction facts for fractions reached significance and it was also outside the TD group range. KJ's performance was similar to that of the TD group.

Similarly, in retrieving facts for decimals, NB failed to retrieve any subtraction and multiplication facts correctly and her performance was outside the TD group range. KJ performed within the TD group range on all tasks of decimals. Only the dissociation observed in NB's performance between addition and multiplication facts for decimals fitted the criteria for a classical dissociation, t(19)=2.17, p=0.04.

[Table 8 about here]

There were insufficient correct trials to merit a RT analysis on fraction and decimal facts.

Procedural Knowledge of Fractions, Decimals and Percentages

Fractions. Table 9 shows scores for accuracy of procedures with fractions and decimals. NB and KJ failed to add and subtract any fractions. However, some TD children also shared the same difficulty (t=0.87, p=0.40 and t=1.26, p=0.22, respectively). All the errors NB and KJ made were procedural errors, they added and subtracted numerators with each other and denominators with each other (e.g. $\frac{1}{5}+\frac{1}{5}=$ "$\frac{2}{10}$").

[Table 9 about here]

In adding fractions, NB had a scaled RT of 16 secs and KJ of 12 secs. The TD group had a mean scaled RT of 17.25 secs (SD=12.61). In subtracting fractions, NB had a scaled RT of 14 secs and KJ of 12 secs. The TD group had a mean scaled RT of 17.40 secs (SD=12.74). Neither NB nor KJ was significantly different from the TD group in adding (NB: t=0.10, p=0.92; KJ: t=0.41, p=0.69) or subtracting fractions (NB: t=0.26, p=0.80, KJ: t=0.41, p=0.68).

Decimals. NB added and subtracted correctly significantly fewer decimals than the TD group (t=3.23, p=0.004 and t=2.59, p=0.02, respectively). Her performance was outside the TD group range on additions and on performing subtractions of decimals with different number of decimal places. In contrast, KJ was as accurate as the TD group in both addition (t=0.41, p=0.68) and subtraction (t=0.36, p=0.72). Figure 2 shows individual scores on the TD group's performance on all decimal operations. In adding decimals, both girls started the procedure from right to left carrying over numbers where required. NB failed to add 0 to make up decimal places following an incorrect procedure. In subtracting decimals, she showed some basic knowledge of borrowing procedures but in the majority of the subtractions, she reversed the order of the numbers to make the subtraction possible. KJ borrowed numbers appropriately but failed to add 0 on the decimals with fewer decimal places and as a consequence she did not follow the borrowing procedure on these occasions.

NB failed to multiply any decimals ($t=1.88$, $p=0.07$, marginal). KJ's performance level was also low but not statistically significant to that of the TD group ($t=1.36$, $p=0.20$). Both girls performed at the bottom end of the TD range in this task (see Figure 2). NB followed a similar procedure used in addition and subtraction to multiply decimals. She multiplied the units with each other, the digits of the tens column with each other and so on. If a decimal had more digits than the multiplicand 10, she multiplied 1 with the tens and hundreds column. KJ also followed the same procedure as NB.

NB's single dissociations in procedures of decimals were not significant (addition-multiplication: $t(19)=0.90$, $p=0.38$; subtraction-multiplication: $t(19)=0.55$, $p=0.59$).

[Figure 2 about here]

NB and KJ were as fast as the TD group on all operations with decimals. In addition, NB had a scaled RT of 21 secs and KJ of 14 secs. The TD group had a mean of 14.30 secs ($SD=4.10$; NB: $t=1.60$, $p=0.13$). In subtraction, NB had a scaled RT of 19 secs, KJ of 15 secs and the TD group had a mean scaled RT of 16.60 secs ($SD=5.49$; NB: $t=0.43$, $p=0.67$; KJ: $t=0.28$, $p=0.78$). Finally, in multiplication, NB had a scaled RT of 8 secs and KJ of 7 secs. The TD group had a mean scaled RT of 8.50 secs ($SD=5.93$; KJ: $t=0.25$, $p=0.81$).

Percentages. Neither NB nor KJ was able to calculate percentages. The TD group scored a mean of 6.20 ($SD=5.52$) in this task. Some children in the TD group also shared the same difficulty with the two TS girls. To explore whether the TS girls had any knowledge of percentages they were asked some basic questions on percentages (e.g. "How much would you get if you could eat 100% of the cake?", "How much would you get if you could eat 50% of the cake?"). NB and KJ showed basic understanding of percentages.

Calculation of fractions. NB refused to do the task. KJ did not perform any calculations correctly. The TD group calculated a mean of 3.25 (SD=4.41) fractions. Both KJ and the TD group performed poorly in this task (t=0.72, p=0.48).

Negative Numbers

Number Processing of Negative Numbers

Table 10 shows accuracy and RTs on tasks of production and comprehension of negative numbers. NB, KJ and the TD group all performed close to ceiling in reading and writing negative numbers. Both girls were as fast as the TD group in reading.

In the "put numbers in order" task although NB's and KJ's performance was not significantly different from the TD group. They all shared the same difficulty in reordering sets of negative numbers correctly. All the incorrect responses were an incorrect reordering of the negative numbers as positive numbers.

Both NB and KJ performed significantly worse than the TD group in magnitude comparison of positive and negative numbers. All errors for NB resulted from incorrect magnitude decision for a negative number when its absolute value was bigger than that of the positive number. NB responded significantly slower and her RT was outside the TD group range whereas KJ was as fast as the TD group in this task. The dissociation between magnitude comparison and reading (writing was constant with SD=0) of negative numbers was significant for NB (t(19)=4.43, p=0.001) but not for KJ (t(19)=0.06, p=0.95).

Impaired comprehension of negative numbers in the two girls with TS was not comparable to their intact ability to understand whole numbers, fractions, decimals and percentages. The difference in performance between the magnitude comparison tasks for whole numbers and that for negative numbers fitted the criteria for classical dissociation for both girls (NB: t(19)=2.41, p=0.03 and KJ: t(19),=2.16, p=0.04). This was also significant between parity judgements and magnitude comparison of negative numbers for NB (t(19)=3.09, p=0.01 but KJ: t(19)=1.74, p=0.10; an one-tailed analysis indicated classical dissociation p=0.05). Performance comparison on magnitude judgement tasks between the other number scales showed a significant or marginally significant dissociation between negative numbers and percentages (NB: t(19)=13.15, p=0.001 and KJ: t(19)=6.10, p=0.001), and between negative numbers and decimals for both girls (t(19)=3.53, p=0.002, KJ: t(19)=1.95, p=0.07). It only reached significance for NB between negative numbers and fractions (t(19)=4.38, p=0.001, KJ: t(19)=1.06, p=0.30).

[Table 10 about here]

Procedural Knowledge of Negative Numbers

Addition and subtraction of positive and negative numbers. Table 11 shows scores on addition and subtraction of positive and negative numbers. NB refused to do these tasks. KJ attempted to perform all additions but failed to be accurate in any item (t=1.67, p=0.11 and t=0.72, p=0.48, respectively). Some children in the TD group shared the same difficulty as NB and KJ in performing calculations of negative numbers. KJ followed the rules of addition and subtraction of whole numbers and calculated the absolute value of the negative and positive numbers ignoring the negative signs.

[Table 11 about here]

KJ had a mean scaled RT of 14 secs in additions and a mean scaled RT of 15 secs in subtractions. The TD group had a mean of 18.35 (SD=8.31) and of 15.65 secs (SD=7.59) respectively. There was no significant difference for either additions (t=0.51, p=0.62) or subtractions.

Discussion

This study reports results on the arithmetical skills with whole numbers in two girls with TS. The objective was to discuss the extent to which performance is compatible with the McCloskey model of arithmetical skill. This is the first study that systematically explored dissociation of skills on previously unexplored number scales (fractions, decimals, percentages and negative numbers). The dissociations that were observed provided evidence for the structural organization of the processing of these number scales. The results further support the modular organisation of the arithmetical system for whole numbers and give preliminary evidence of a comparable organization for the other number scales.

Implications for whole number scale

Both girls with TS were accurate on all counting tasks. Only NB showed a speed of accessing impairment on forward and backward counting similar to the one reported in previous group studies in TS (Bruandet et al, 2004; Simon et al, 2008; Temple & Sherwood, 2002). Therefore, any difficulties in their more advanced arithmetical development, at least with regards to accuracy, cannot be attributed to counting impairment (Butterworth, 1999; Butterworth, 2005).

Both girls with TS showed intact number processing accuracy for whole numbers, findings that are consistent with the literature (Murphy et al, 2006; Rovet et al, 1994; Temple & Marriott, 1998; Temple & Sherwood, 2002). No dissociations emerged in these tasks and so the data from the present study does not support modularity within the number processing system.

With regards to the calculation system, results generally supported the core features of McCloskey's model. Performance by NB confirms the distinction between procedural and arithmetical fact retrieval skills. Comparable to previous TS (Temple & Marriott, 1998) and patient case reports (Hittmair-Delazer, Semenza & Denes, 1994; Hittmair-Delazer, Sailer & Benke, 1995; Kaufmann, 2002; Kaufmann, Lochy, Drexler & Semenza, 2004; Temple, 1991), NB was impaired in retrieving multiplication facts but she was intact in performing multiplication procedures. These results suggest that acquisition of single-digit operations during arithmetical development precedes that of higher order multi-digit operations (see Butterworth, 2005). NB's performance indicates that her difficulties do not arise from a failure to acquire full development of calculation skills for whole numbers. Rather, in line with McCloskey's model, there is a specific disruption in the fact retrieval mechanisms for multiplications.

McCloskey's model also incorporates distinct mechanisms for each operation in both the facts and the procedural operations systems. The data from the two girls do not support this distinction for the fact retrieval system. It is important, though, to note that when finger-counting strategies were used, which involve reconstruction rather than retrieval, both NB and KJ were able to perform at TD level on these tasks. Therefore, reconstruction could have underlain the slow RTs but intact accuracy observed in previous studies in TS girls (Bruandet et al, 2004; Butterworth et al, 1999; Rovet et al, 1994; Temple & Marriott, 1998; Temple & Sherwood, 2002) masking relevant dissociations of performance that are compatible with models of normal arithmetical skill. Decomposition and strategy reliance in arithmetic has been previously reported in non-TS individuals (e.g., Hittmair-Delazer, Semenza and Denes, 1994; Kesler, Menon & Reiss, 2005; Kesler, Sheau, Koovakkattu & Reiss, 2011; Mazzocco &

Hanich, 2010). Future studies should take the possibility of reconstruction into account on arithmetical tasks.

The existence of separate mechanisms for each operation in the procedures operation system is supported by single dissociations. NB was able to perform multiplication and division but not addition and subtraction procedures, and KJ performed addition but not subtractions operations. Addition and subtraction skills are acquired earlier than the more complex and higher-order multiplication and division skills in arithmetical development. NB's impaired ability to perform additions and subtractions was not due to unfamiliarity or difficulty of the tasks as she was able to perform the more complex multiplication and division procedures. Rather, it provides evidence for a disruption in the distinct procedures mechanisms that are involved in addition and subtraction in McCloskey's model.

NB's single dissociation between subtraction and multiplication is difficult to explain in terms of models that contained single-digit multiplication system that is distinct from that of subtraction (Dehaene, 1992; Dehaene & Cohen, 1995; 1997; Dehaene, Piazza, Pinel & Cohen, 2003). These models argued that in multiplication, rote verbal memory is also used to retrieve facts that are required to perform multi-digit operations, whereas subtraction is always performed by counting or semantic elaboration and does not involve rote fact retrieval. Thus, it is claimed that the procedures that are used for multiplication and subtraction differ on the basis of their dependency upon rote fact retrieval. The results for NB contradict this theory as she was accurate at multiplication operations, despite impairment in multiplication fact retrieval. In line with McCloskey's model, the data for NB argue for a distinction between factual and procedural mechanisms.

The two girls with TS showed intact number processing for whole numbers but their selective impairment within calculations was compatible with the modular structure of the arithmetical system. The data within calculations strengthens McCloskey's functional architecture of the procedures mechanisms. In addition, it highlights the importance of taking reconstruction into consideration when assessing the arithmetical skills. Fractionations in the arithmetical system can be masked by strategy reliance.

Implications for number scales beyond whole numbers

There is no previous literature available to describe the processing that underlies number scales beyond whole numbers. The observed dissociations of skills in tasks assessing unexplored number scales, fractions, decimals, percentages and negative numbers provided evidence for analogous functional architecture in these scales to that of whole numbers.

Fractions and Percentages. KJ had intact number processing for fractions and percentages, reading, writing and making magnitude judgments correctly, but she was unable to produce a single solution in calculations involving fractions and percentages. These data confirm the distinction between number processing and calculation skills in these scales, in line with McCloskey's model. Some children in the TD group, however, shared these difficulties when performing these tasks. It is therefore possible that inability to perform these tasks could be due to the unfamiliarity or difficulty of the tasks rather than to impaired skill. The type of errors that both NB and KJ made in performing addition and subtraction of fractions are in line with the componential processing of fractions (Bonato et al, 2007). Both NB and KJ added and subtracted numerators with each other, and denominators with each other, without focusing on the numerical value of the fraction.

Decimals. NB had difficulty in reading decimals. The type of errors she made in reading decimals indicates distinct mechanisms within the number production system for decimals similar to the ones described by McCloskey in his model. NB made syntactic errors in reading decimals but showed preserved lexical processing. If the decimal organisation system is analogous in architecture to that of whole numbers, then this type of errors suggests impairment in the syntactic processing mechanism of the spoken verbal decimal production system with the lexical processing mechanism intact.

Within calculations, a single dissociation supports the existence of autonomous operations in the fact retrieval mechanism for decimals compatible with the one outlined by McCloskey in his model for whole numbers. NB was able to retrieve addition facts but not multiplication facts for decimals. The tasks employed to assess fact retrieval of fractions and decimals included an insufficient number of trials to provide a clear pattern of performance in these tasks. Future studies should employ a more adequate design to assess fractionations of fact retrieval skills in these scales.

KJ was able to perform additions and subtractions of decimals but showed difficulty in performing multiplications. Arguably, this finding suggests distinct operations mechanisms in the procedures system of the decimals number scale also. Some children from the TD group showed a similar difficulty on this task. KJ’s poor performance could therefore reflect a difficulty to perform multiplications of decimals because they are more complex skill to acquire than additions and subtractions.

Negative Numbers. NB and KJ had impaired number comprehension for negative numbers. The errors they made in the magnitude comparison tasks as well as the intact development of the comprehension system for whole numbers suggest that the impairment was located in the comprehension system for negative numbers. Their ability to read and write negative numbers was intact. NB’s dissociation between reading and making magnitude comparisons of negative numbers implies distinct systems for comprehension and production of negative numbers. It also indicates that reading and writing of negative numbers could be achieved through a direct asemantic route in line with other arithmetical models (e.g., Dehaene, 1992; Cipolotti, 1995; Cipolotti & Butterworth, 1995). Similar to fractions, number processing system for negative numbers could also include both a semantic and an asemantic route for processing skills.

Although the data in the current study does not demonstrate all possible fractionations of skills in fractions, decimals, percentages and negative numbers in line with the modular scales for whole numbers, it nevertheless, provides preliminary evidence for some distinct components in these scales that support their modular structure. There should be further investigations in the future of whether dissociations in these scales, in the calculation system in particular, reflect a fractionation of skills rather than a lack of familiarity with the task employed. This study is the first step towards the development of models that describe the structure of scales beyond whole numbers.

General Model of arithmetic: dissociations between number scales

The findings of the current study provided evidence against a unitary arithmetical scale. Instead it was consistent with the autonomous representation of arithmetical scales with interconnections between them. Evidence in favour of a distinction between number scales is provided by the ability of the two girls with TS to perform tasks on some number scales but not on others. This data is innovative as there has not been an attempt in the past literature to make direct comparison between number scales addressing their independent or interrelated nature and describe these differences in a general model of arithmetic.

Whole numbers and other scales. Comprehension accuracy for whole numbers was intact for both NB and KJ. However, NB and KJ were both significantly impaired on tasks of comprehension of negative numbers. They were impaired both in accuracy of magnitude comparisons of positive and negative numbers and in accuracy of putting negative numbers in order of magnitude. Given the intact comprehension accuracy of whole numbers, this argues for a distinction between the number comprehension systems that underlie negative numbers and those that underlie whole numbers. In this study, understanding of whole numbers did not benefit understanding of negative numbers, as a precursor of successful mastery of the latter system (Fischer and Rottman, 2005; Ganor-Ster et al, 2010; Tzelgov et al, 2008). It does not, however, mean that comprehension of whole numbers is not a prerequisite for the development of comprehension of negative numbers. Both girls showed a speed of processing difficulty in making magnitude comparisons for whole numbers. NB had a comparable difficulty in speed of processing for negative number comprehension. Slow RTs in understanding whole numbers could have therefore affected performance in negative numbers in these girls (accuracy and RTs for NB, and accuracy for KJ). Negative numbers are represented to the left of a mental number line when interrelated with positive numbers (Fischer, 2003; Fischer & Rotmann, 2005; Prather & Boroditsky, 2003; Shaki, & Petrusic, 2005). All errors made by the two girls with TS related to the directional component of negative numbers rather than the absolute value of the lexical representations. The two girls with TS in this study showed difficulty in representing negative numbers as an extension of the mental number line of positive numbers. The data supports the autonomous nature of these number scales with possible interconnectivity between their subsystems to facilitate accomplishment of certain arithmetical skills.

Data from both KJ and NB also support the distinction of whole numbers from decimals. NB was able to read whole numbers but showed impairment in reading decimals. This single dissociation argues for a separate syntactic processing in the production system for whole numbers and decimals.

Although there is no evidence in this study of a separate fractions scale to that of whole numbers, the data from KJ suggest interconnections between the systems that are involved. The strategies that KJ used in calculating fractions, for instance, adding numerators with each other and denominators with each other, are in line with the componential processing view (Bonato et al, 2007). It appears that knowledge from whole numbers was applied to the processing of fractions.

Fractions, decimals and percentages. Although fractions and decimals are related mathematical concepts, they are nevertheless distinct number scales. Data from NB supports a distinction between number processing for fractions, percentages and decimals. She was able to read fractions and percentages but she had difficulty in reading decimals. This is compatible with distinct production scales for reading decimals from those utilised to read fractions and percentages, particularly in terms of syntactic processing as highlighted by the errors made.

Negative numbers and other number scales. Performance by the two girls with TS also supports the independence of the negative numbers scale. NB and KJ had an impaired comprehension system for negative numbers, yet NB had intact comprehension system for fractions, decimals and both girls for percentages. These findings support a distinction between number processing systems that underlie negative numbers and those that underlie the other number scales.

There is therefore evidence that supports the autonomous representation of number scales. The two girls with TS performed well in some tasks but showed difficulty in the same tasks on other number scales. In addition, the type of errors the two girls made in performing some of the tasks confirms interdependence between number scales. For instance, both girls with TS made incorrect magnitude decisions for negative numbers when their absolute value was bigger than that of positive numbers. They applied knowledge from whole numbers in making decision for negative numbers. Furthermore, in calculating fractions, both girls added or subtracted numerators with each other and denominators with each other applying procedure rules from whole numbers. This is the first study that systematically explored the relation between different number scales. Based on the performance of the two girls with TS, a general model of arithmetic with semi-independent autonomous number scales is proposed (see Figure 1). The McCloskey model of whole numbers is assumed to interact with all numbers scales. The current study does not illustrate the role of whole numbers as a prerequisite for the successful processing on decimals and percentages but considering that these are related mathematical concepts to fractions (e.g., $\frac{1}{4}$, 0.25, 25%), a similar architecture is proposed. In the general model of arithmetic, it is suggested that fractions, decimals and percentages are connected with each other to perform arithmetical tasks such as transcoding from one number scale to another. Transcoding between these number scales is a more complex procedure than in whole numbers (Arabic number to number word and vs) as it requires connection between mechanisms for different number scales. Although transcoding between these scales was included in the current study, the TS girls' performance does not specify the processing that takes place when translating the semantically equivalent values between scales. We only nevertheless assume there is a connection between different number scales to attain translation of a number from one scale to another. Further studies need to identify more fractionations in the arithmetical system to describe the mechanisms included in each number scale and which of these are involved in the connections between scales.

[Figure 3 about here]

Overall Conclusions

The current study explored the arithmetical competence of two cases with TS and identified fractionations within some components of the arithmetical system. Dissociations of performance in the calculation tasks of whole numbers supported the modular organization of the arithmetical system as described by McCloskey in his model. Performance on other unexplored number scales, fractions, decimals, percentages and negative numbers showed similarities to the McCloskey model in the way these scales are organized. There are no models that describe the functional architecture of these unexplored number scales, and data from the present study is the first systematic evidence of how numbers in these scales are processed. This study is an important first step in describing number scales beyond whole numbers. Future studies could delineate these number scales in more detail, including more cases or groups of girls with TS, and adapt McCloskey's model to describe their processing step by step. The data in the present study clearly indicates the autonomous representation of number scales with interconnections between them.

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Table 1 Counting: Accuracy and RTs

Accuracy	NB	KJ	TD	NB t (p)	KJ t (p)
How many: counting	12/12	12/12	11.75 (0.55)	-	-
How many: cardinality	12/12	12/12	11.80 (0.52)	-	-
Number next/before	15/16	16/16	15.80 (0.41)	-	-
RTs (secs)					
Forward	17**‡	6	7.45 (3.47)	2.69 (0.01)	0.41 (0.69)
Backward	18**‡	8	8.45 (3.25)	2.87 (0.01)	-
Number next/before	1.13 (0.52)	1.2 (0.56)	1.32 (0.21)	0.88 (0.39)	0.56 (0.58)

**p≤0.01; ‡Outside TD group range

Table 2 Number processing of whole numbers: Accuracy

Accuracy	NB	KJ	TD Mean (SD)
Reading Arabic numbers	40/40	40/40	39.90 (0.31)
Reading number words	40/40	40/40	39.75 (0.44)
Writing Arabic numbers	16/16	16/16	16.00 (0.0)
Writing number words	16/16	15/16	15.30 (1.26)
Transcoding Arabic numbers into number words	12/12	11/12	11.80 (0.41)
Transcoding number words into Arabic numbers	12/12	12/12	12.00 (0.0)
Magnitude comparison of Arabic numbers	19/20	20/20	19.55 (0.61)
Parity judgement	20/20	20/20	19.50 (1.57)

Table 3 Counting and number processing of whole numbers: RTs

Reading	NB secs	KJ secs	TD secs	NB t (p)	KJ t (p)
Arabic numbers	959 (270)	996 (166)	923 (102)	0.34 (0.73)	0.70 (0.49)
Number words	929 (138)	1079 (339)	955 (114)	0.22 (0.83)	1.06 (0.30)
Comprehension					
Magnitude comparison	1985 (347)** ‡	1690 (256)* ‡	1153 (228)	3.56 (0.002)	2.30 (0.03)
Parity judgement	2420 (1078)	2162 (618)	1690 (1206)	0.59 (0.56)	0.38 (0.71)

*p≤ 0.05; **p≤ 0.01; ‡Outside TD group range

Table 4 Calculation of whole numbers: Accuracy

Facts (finger counting)	NB	KJ	TD Mean (SD)	NB t (p)	KJ t (p)
Addition	45/45	45/45	44.7 (0.57)	-	-
Subtraction	28/28	28/28	27.65 (0.59)	-	-
Multiplication	62/64	61/64	60.05 (4.40)	0.43 (0.67)	0.21 (0.84)
Facts (retrieval only)					
Addition	31/45**‡	39/45**	44.30 (1.75)	7.42 (0.001)	2.96 (0.008)
Subtraction	13/28**‡	24/28*	27.25 (1.41)	9.86 (0.001)	2.25 (0.04)
Multiplication	23/64**‡	61/64	59.05 (4.80)	7.33 (0.001)	0.40 (0.70)
Procedures					
Addition	6/16**‡	14/16	14.90 (1.33)	6.53 (0.001)	-
Subtraction	7/16**‡	9/16**‡	13.75 (1.48)	4.45 (0.001)	3.13 (0.005)
Multiplication	5/14	5/14	7.85 (2.74)	1.02 (0.32)	1.02 (0.32)
Division	4/10	2/10*	6.90 (2.12)	1.34 (0.30)	2.26 (0.04)

*p≤0.05; **p≤0.01; ‡Outside TD group range

Table 5 Calculation of whole numbers: RTs

Facts (finger counting)	NB msecs	KJ msecs	TD msecs	NB t (p)	KJ t (p)
Addition	2702 (1435)	2620 (1588)	1872 (472)	1.72 (0.10)	1.55 (0.14)
Subtraction	3130 (864)	3006 (1422)	2176 (760)	1.23 (0.24)	1.07 (0.30)
Multiplication	5688 (4717)* ‡	2610 (1201)	3210 (1163)	2.08 (0.05)	0.50 (0.62)
Facts (retrieval only)					
Addition	2031 (872)	2399 (1536)	1852 (456)	0.38 (0.71)	1.17 (0.26)
Subtraction	2813 (823)	2664 (1174)	2143 (693)	0.94 (0.36)	0.73 (0.47)
Multiplication	2580 (917)	2610 (1201)	3134 (1083)	0.50 (0.62)	0.47 (0.64)
Procedures					
Addition	12	10	9.10 (3.26)	0.87 (0.40)	0.27 (0.79)
Subtraction	18	11	13.50 (4.19)	1.05 (0.31)	0.58 (0.57)
Multiplication	24	24	37.20 (18.71)	0.69 (0.50)	0.69 (0.50)
Division	36	23	27.60 (18.45)	0.44 (0.66)	0.24 (0.81)

*p≤0.05; ‡Outside TD group range

Table 6 Number processing of fractions, decimals and percentages: Accuracy

Reading	NB	KJ	TD Mean (SD)	NB t (p)	KJ t (p)
Fractions	37/40	32/40	35.30 (4.65)	0.36 (0.73)	0.69 (0.50)
Decimals	20/40**‡	30/40	37.20 (5.70)	2.95 (0.008)	1.23 (0.23)
Percentages	40/40	40/40	40.00 (0.0)	-	-
Writing					
Fractions	16/16	15/16	15.45 (0.69)	-	-
Decimals	15/16	16/16	15.90 (0.31)	-	-
Percentages	16/16	16/16	15.90 (0.31)	-	-
Transcoding					
Fractions-Decimals	2/12	3/12	6.95 (3.72)	1.30 (0.21)	1.04 (0.31)
Decimals-Fractions	7/12	8/12	7.45 (3.63)	-	-
Fractions-Percentages	3/12	2/12	6.70 (3.57)	1.01 (0.32)	1.29 (0.21)
Percentages-Fractions	9/12	10/12	9.10 (3.02)	-	-
Decimals-Percentages	12/12	12/12	10.15 (2.11)	0.86 (0.40)	0.86 (0.40)
Percentages-Decimals	12/12	12/12	9.40 (2.70)	0.94 (0.36)	0.94 (0.36)
Magnitude comparison					
Fractions	17/20	11/20	15.95 (4.21)	0.24 (0.81)	1.15 (0.27)
Decimals	19/20	19/20	18.15 (4.52)	-	-
Percentages	20/20	19/20	19.50 (1.36)	-	-

**$p \leq 0.01$; ‡Outside TD group range

Table 7 Number processing of fractions, decimals and percentages: RTs

Reading	NB msecs	KJ msecs	TD msecs	NB t (p)	KJ t (p)
Fractions	1399 (254)	1279 (260)	1451 (541)	0.09 (0.93)	0.31 (0.76)
Decimals	1268 (299)	1270 (247)	1228 (219)	0.18 (0.86)	0.19 (0.85)
Percentages	1047 (250)	1043 (187)	1067 (173)	0.11 (0.91)	0.14 (0.89)
Magnitude comparison					
Fractions	3635 (1422)	3060 (993)	3382 (1281)	0.19 (0.85)	0.25 (0.81)
Decimals	1907 (489)	1531 (313)	1661 (433)	0.55 (0.59)	0.29 (0.77)
Percentages	1424 (314)	1008 (191)	1160 (248)	1.04 (0.31)	0.59 (0.56)

Table 8 Fact retrieval of fractions and decimals: Accuracy

Addition facts	NB	KJ	TD Mean (SD)	NB t (p)	KJ t (p)
Fractions	1/5	0/5	3.05 (1.85)	1.08 (0.30)	1.61 (0.12)
Decimals	1/5	3/5	3.20 (1.36)	1.58 (0.13)	-
Subtraction facts					
Fractions	0/5*‡	1/5	4.00 (1.59)	2.46 (0.02)	1.84 (0.08)
Decimals	0/5**‡	2/5	3.50 (1.15)	2.97 (0.008)	1.27 (0.22)
Multiplication facts					
Fractions	2/5	1/5	3.80 (1.64)	1.07 (0.30)	1.67 (0.11)
Decimals	0/5**‡	2/5*	4.20 (1.01)	4.06 (0.001)	2.13 (0.05)

*$p \leq 0.05$; **$p \leq 0.01$; ‡Outside TD group range

Table 9 Procedures for fractions and decimals: Accuracy

Addition of fractions	NB	KJ	TD Mean (SD)
Common denominator	0/12	0/12	4.50 (5.04)
Easy to find LCD	0/6	0/6	0.55 (0.89)
Difficult to find LCD	0/6	0/6	0.05 (0.22)
Total score	0/24	0/24	5.10 (5.71)
Subtraction of fractions			
Common denominators	0/12	0/12	7.10 (5.41)
Easy to find LCD	0/6	0/6	0.25 (0.79)
Difficult to find LCD	0/6	0/6	0
Total score	0/24	0/24	7.35 (5.70)
Addition of decimals			
Same number of decimal places	8/10	10/10	9.35 (1.04)
Different number of decimal places	0/10**‡	9/10	8.40 (2.14)
Total score	8/20**‡	19/20	17.75 (2.95)
Subtraction of decimals			
Same number of decimal places	4/9	7/9	6.95 (1.99)
Different number of decimal places	0/11**‡	5/11	6.35 (2.01)
Total score	4/20*	12/20	13.30 (3.51)
Multiplication of decimals			
Decimals × 10	0/10	4/10	7.70 (3.91)
Decimals × 100	0/10	0/10	6.70 (3.74)
Total score	0/20	4/20	14.40 (7.49)

**$p \leq 0.01$; ‡Outside TD group range; least common denominator (LCD)

Table 10 Number processing of negative numbers: Accuracy and RTs

Accuracy	NB	KJ	TD Mean (SD)	NB t (p)	KJ t (p)
Reading -ve numbers	40/40	39/40	39.85 (0.37)	-	-
Writing -ve numbers	16/16	16/16	16.00 (0.0)	-	-
Put numbers in order	0/20	0/20	8.90 (10.10)	0.86 (0.40)	0.86 (0.40)
Magnitude comparison of +ve and -ve numbers	11/20**	15/20*	19.50 (2.01)	4.13 (0.001)	2.19 (0.04)
RTs (msecs)					
Reading -ve numbers	990(217)	880(230)	959 (165)	0.18 (0.86)	0.47 (0.65)
Magnitude comparison of +ve and -ve numbers	2268(590)** ‡	988(342)	899 (174)	7.68 (0.001)	0.50 (0.62)

*p≤0.05; **p≤0.01; ‡Outside TD group range; -ve: negative; +ve: positive

Table 11 Calculation of negative numbers: Accuracy

Accuracy	KJ	TD Mean (SD)
Addition of +ve and -ve numbers	0/25	13.10 (7.64)
Subtraction of +ve and -ve numbers	3/25	7.15 (5.64)

-ve: negative; +ve: positive

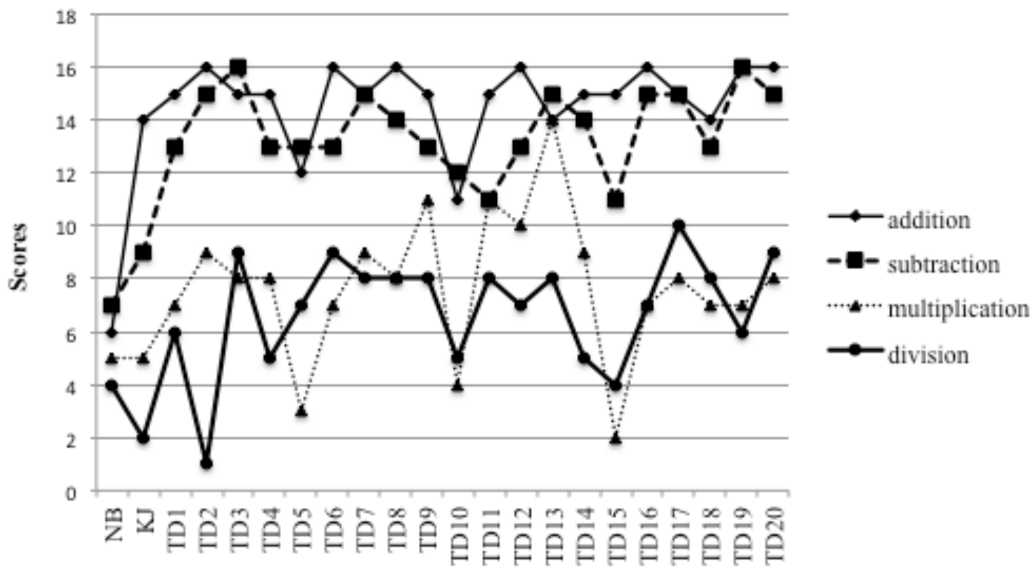

Figure 1. TS and TD group individual scores on procedures of whole numbers.

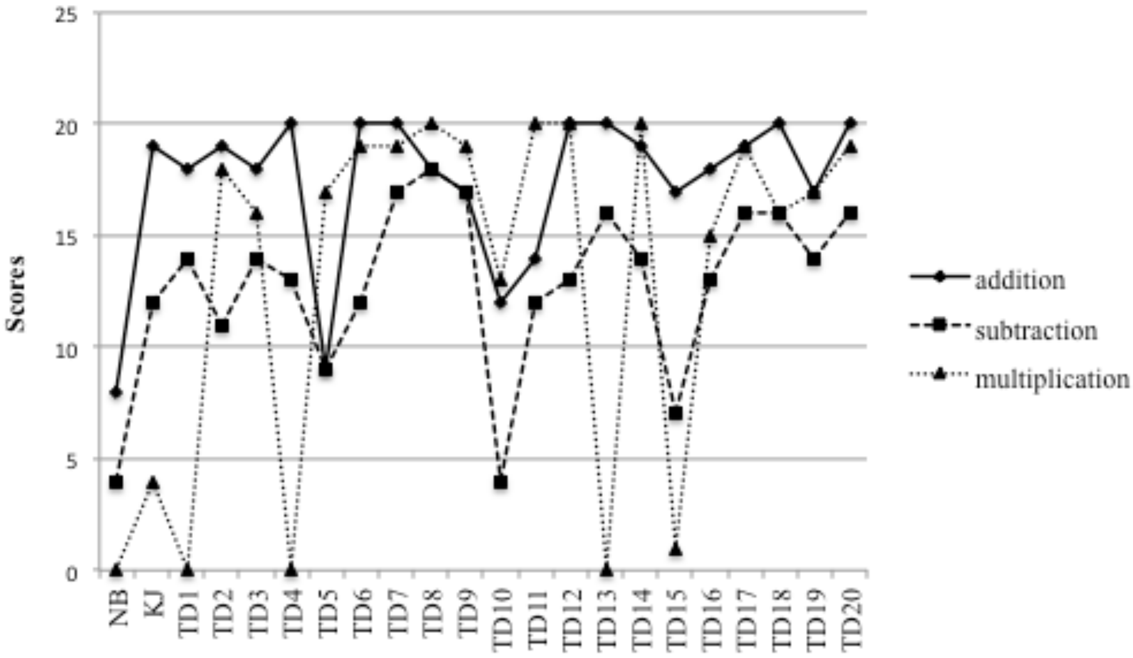

Figure 2. TS and TD group individual scores on procedures of decimals.

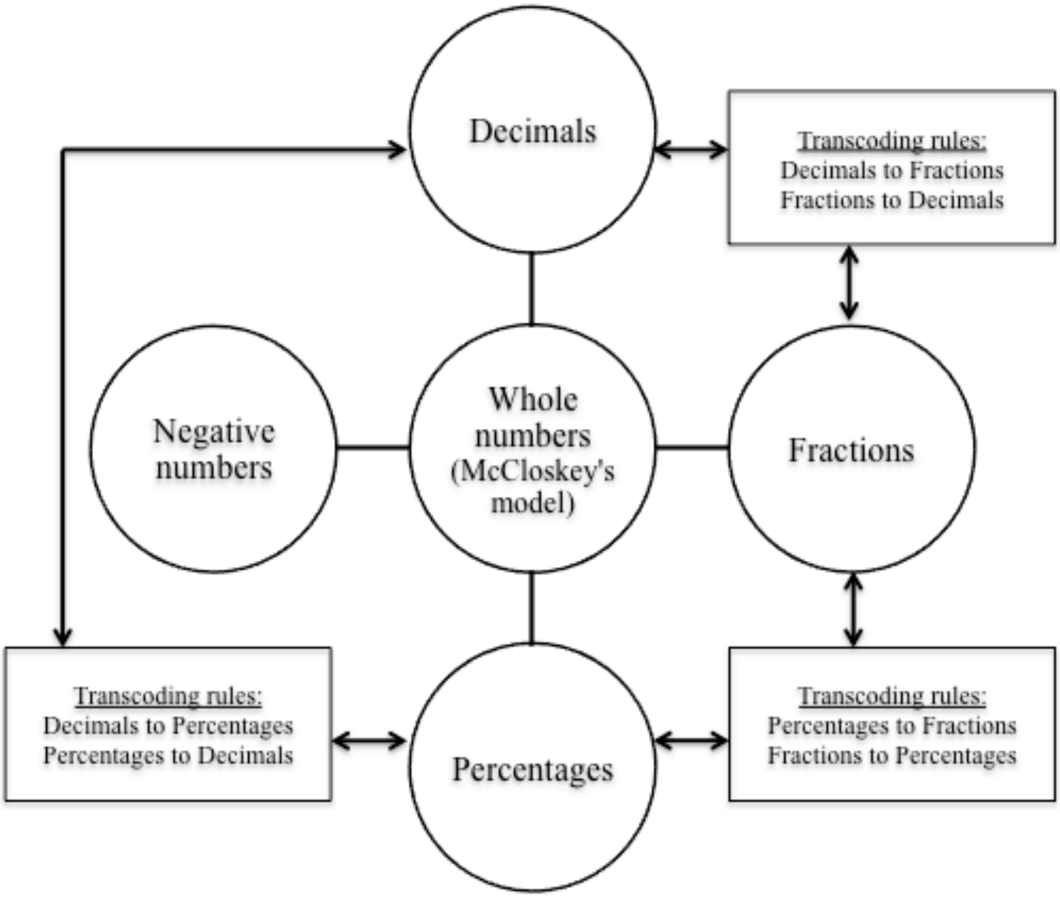

Figure 3. A general model of arithmetic.