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Enumeration skills in Down syndrome

Francesco Sella [a], Silvia Lanfranchi [a], Marco Zorzi [b,c,*]

[a] Department of Developmental Psychology and Socialization, University of Padova, Via Venezia 8, 35131 Padova, Italy
[b] Department of General Psychology, University of Padova, Via Venezia 8, 35131 Padova, Italy
[c] IRCCS San Camillo Hospital, Venice-Lido, Italy

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ABSTRACT

Individuals with Down syndrome (DS) exhibit various math difficulties which can be ascribed both to global intelligence level and/or to their atypical cognitive profile. In this light, it is crucial to investigate whether DS display deficits in basic numerical skills. In the present study, individuals with DS and two groups of typically developing (TD) children matched for mental and chronological age completed two delayed match-to-sample tasks in order to evaluate the functioning of visual enumeration skills. Children with DS showed a specific deficit in the discrimination of small numerosities (within the subitizing range) with respect to both mental and chronological age matched TD children. In contrast, the discrimination of larger numerosities, though lower than that of chronological age matched controls, was comparable to that of mental age matched controls. Finally, counting was less fluent but the understanding of cardinality seemed to be preserved in DS. These results suggest a deficit of the object tracking system underlying the parallel individuation of small numerosities and a typical – but developmentally delayed – acuity of the approximate number system for discrimination of larger numerosities.

1. Introduction

Down syndrome (DS) is due to abnormalities on chromosome 21 and it is the most common cause of intellectual disability (Kittler, Krinsky-McHale, & Devenny, 2008). The cognitive profile of this syndrome is characterized by a relative weakness in verbal abilities, while visuo-spatial skills seem to be relatively preserved (Dykens, Hodapp, & Finucane, 2000).

It is well known that children and adults with Down syndrome (DS) exhibit several mathematical difficulties as compared to typically developing (TD) individuals (Brigstocke, Hulme, & Nye, 2008). Children with DS obtain lower scores in a wide range of tests assessing basic math knowledge, arithmetic abilities and counting skills (Buckley & Sacks, 1987; Carr, 1988; Gelman & Cohen, 1988; Porter, 1999). These mathematical deficits can be attributed to the general intelligence level or to the atypical cognitive profile of DS. In this light, it is crucial to determine whether math underachievement in DS can be related to the low level of cognitive functioning or to specific deficits in basic numerical skills. Such an investigation may provide new insights regarding the source of difference in math achievement between DS and TD individuals.

Two basic pre-verbal mechanisms have been highlighted as fundamental for numerical processing in human and non-human species: the Object Tracking System (OTS; Mandler & Shebo, 1982; Trick & Pylyshyn, 1994; Xu, Spelke, & Goddard, 2005) and the Approximate Number System (ANS; Dehaene, 1997; Feigenson, Dehaene, & Spelke, 2004; Piazza, 2010; Stoianov & Zorzi, 2012). The OTS is a domain-general system that encodes spatio-temporal characteristics of objects with a

* Corresponding author at: Department of General Psychology, University of Padova, Via Venezia 8, 35131 Padova, Italy. Tel.: +39 049 8276618.
E-mail addresses: francescosella@yahoo.it (F. Sella), silvia.lanfranchi@unipd.it (S. Lanfranchi), marco.zorzi@unipd.it (M. Zorzi).

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capacity limited to three-four items. Despite the fact that the OTS is primarily a non-numerical mechanism, it supports visual enumeration of small sets of objects. Indeed, observers can quickly, accurately and effortlessly perceive the numerosity of small sets, a phenomenon known as *subitizing* (from the Latin *subitus*, immediate; Kaufman, Lord, Reese, & Volkmann, 1949). Children with developmental dyscalculia have a reduced subitizing range and tend to adopt serial counting to determine the numerosity of small sets (Landerl, Bevan, & Butterworth, 2004; Moeller, Neuburger, Kaufmann, Landerl, & Nuerk, 2009; Schleifer & Landerl, 2011), a finding that suggests a crucial role of the OTS mechanism for numerical development (Carey, 2001; Le Corre & Carey, 2007).

When the numerosity exceeds the subitizing range (i.e., more than 4 elements) and serial counting is precluded, visual enumeration become imprecise, with a variability of response that obeys Weber's law (Dehaene, Izard, Spelke, & Pica, 2008; Stoianov & Zorzi, 2012). This pattern, which is regarded as the signature of the ANS, implies that the ability to discriminate two numerosities decreases as a function of their numerical ratio and it can be indexed by the Weber fraction, also known as *number acuity*. This ability is already active during the first year of life (e.g., six months-old infants can discriminate 8 vs 16 dots; Xu & Spelke, 2000) and it is progressively refined during childhood (Halberda & Feigenson, 2008; Halberda, Ly, Wilmer, Naiman, & Germine, 2012; Piazza et al., 2010). Crucially, number acuity has been found to correlate with mathematical achievement (Halberda, Mazzocco, & Feigenson, 2008; Libertus, Feigenson, & Halberda, 2011; Lourenco, Bonny, Fernandez, & Rao, 2012; Mazzocco, Feigenson, & Halberda, 2011a) and it is severely reduced in children with developmental dyscalculia (Mazzocco, Feigenson, & Halberda, 2011b; Piazza et al., 2010).

The acquisition of a symbolic number system allows children to go beyond the pre-verbal number processing mechanisms. In particular, learning of the list of number words and the acquisition of a counting routine allows for accurate serial enumeration of potential infinite sets. Counting entails three basic principles (Gallistel & Gelman, 1992; Gelman & Gallistel, 1978): (i) a one-to-one correspondence between each object and the corresponding word in the counting list; (ii) a stable (and correct) order of the counting list; and (iii) identification of the last word in the counting list as the numerosity (cardinality) of the set. Proficient mastering of counting skills has an important influence on early math achievement (Jordan, Kaplan, Locuniak, & Ramineni, 2007; Passolunghi, Vercelloni, & Schadee, 2006).

The investigation of basic numerical skills in individuals with DS is relatively sparse, despite the fact that these abilities might be at the hearth of their math underachievement in comparison with typically developing children. The functioning of OTS and ANS was previously investigated by Paterson, Girelli, Butterworth, and Karmiloff-Smith (2006) in individuals with DS as compared to a group of individuals with William syndrome and to control groups matched for mental age and chronological age. Using a preferential looking paradigm, they found that children with DS (M_{age} = 30 months) did not discriminate between two and three objects, thereby suggesting a deficit in OTS. In contrast, performance of young adults with DS (M_{age} = 24 years-old) in a numerosity comparison task (using sets both within and beyond the subitizing range) was similar to that of control individuals. More recently, Camos (2009) reported that six year-old children with DS were able to discriminate between 16 and 8 dots but failed to discriminate between 12 and 8, thereby showing the classic ratio-dependent signature of ANS. Their performance was comparable to that of typically developing pupils (both MA and CA matched controls), but the limitation to two numerical ratios might have hidden potential differences in number acuity between children with DS and control groups. In summary, the ANS appears to be preserved in individuals with DS, whereas the OTS seems to be less efficient, at least in young children with DS.

A debated issue regarding counting skills in individuals with DS is whether they have a superficial or a deep understanding of counting (for a review, Abdelahmeed, 2007). On one hand, some studies suggest that individuals with DS use counting as a mere routine lacking the understanding of cardinality principle. Indeed, Gelman and Cohen (1988) maintained that children with DS learn to count by rote and often lack the knowledge of the cardinality principle. Porter (1999) also reported that children with DS can count by rote but are less efficient to detect counting errors performed by other individuals. On the other hand, different studies support the idea that individuals with DS properly understand the meaning of counting as well as the cardinality principle. For example, Caycho, Gunn, and Siegal (1991) found a similar understanding of counting principles in children with DS and control children matched for receptive vocabulary. Similarly, Bashash, Outhred, and Bochner (2003) found that children with DS were able to apply the three fundamental principles of counting in several counting contexts. Finally, Nye, Fluck, and Buckley (2001) reported a pattern of results in which children with DS demonstrated a conceptual understanding of cardinality, although they made more errors in the counting procedure. It clearly appears that the picture of the counting ability in DS is still controversial and it requires further investigation.

In the present study, we employed two numerosity match-to-sample tasks in order to evaluate the functioning of visual enumeration in children with DS in comparison to typically developing groups matched for both mental and chronological age. In both tasks, children observed a briefly presented sample numerosity and, after a delay period, a target numerosity. They had to decide whether the target numerosity was equal or different from the sample numerosity. In the dots-to-dots match-to-sample task, we assessed the ability to compare numerosities within and beyond subitizing range/OTS capacity. Our aim was to verify whether children with DS have a reduced subitizing range and to highlight possible differences in number acuity when numerosity discrimination entails larger numerosities. In the digit-to-dots match-to-sample task, the sample was an Arabic number and the target was a set of dots. Note that counting of the items in the target can yield the exact cardinality of the set to be matched with the cardinality entailed by the Arabic digit. Therefore, this task assessed the efficiency of the counting routine as well as the understanding of the cardinality principle

2. Materials and methods

2.1. Participants

Sixty-three participants from the middle socioeconomic status from northern Italy took part to the study after parents gave their informed consent. There were 21 children with DS (9 males; M_{age} = 14;2, SD = 3;4), 21 typically developing children (9 males; M_{age} = 5;4, SD = 0;6) matched for mental-age (MA), and 21 typically developing children (9 males; M_{age} = 14;2, SD = 3;6) matched for chronological age (CA). For the matching purpose, a measure of receptive vocabulary, the Peabody Picture Vocabulary Test-Revised (PPVT-R, Dunn & Dunn, 1997), was used. Moreover, in order to have also a measure of fluid intelligence Raven's colored matrices (Raven, Raven, & Court, 1998) were administered to DS and MA groups. Participants' characteristics are presented in Table 1. In order to have a fine matching between groups (Bonato, Sella, Berteletti, & Umiltà, 2012), participants with DS and MA controls also completed a standardized battery for the assessment of different aspects of basic numerical competence in young children (BIN – Batteria Intelligenza Numerica – Numerical Intelligence Scale; Molin, Poli, & Lucangeli, 2007) and one ad-hoc battery to assess their arithmetic knowledge. The BIN is composed of four subscales: the lexical subscale assesses the ability to read and write Arabic numbers as well as the ability to connect the number-word to the correct digit; the semantic subscale measures the ability to compare numerical quantities (dots and Arabic digits); the pre-syntactical scale evaluates the ability to link numbers to their quantity representation and to order multiple quantities; the counting subscale assesses the ability to recite the number–words sequence forward and backward as well as the knowledge of the order of Arabic digits from 1 to 5. In the BIN battery, the MA and DS groups had a similar performance in terms of total score as well as for three out of four subscales. The only significant difference was on the lexical subscale in which the better score for DS children is likely to reflect their longer experience with numbers symbols. In the arithmetic knowledge battery, children were requested to solve 8 non-verbal calculations (add or subtract one or more dots from a given set; 4 additions and 4 subtractions), 8 single digits arithmetic problems (4 additions and 4 subtractions), 8 arithmetic facts (4 additions and 4 subtractions). The only significant difference was on non-verbal calculation subscale in which MA children outperformed DS children.

2.2. Tasks

In the dots-to-dots match-to-sample task (Fig. 1, *panel a*), each trial began with a fixation cross in the middle of the screen for 500 ms immediately followed by a blank screen for 150 ms. Thereafter, a sample set of dots was shown in the middle of the screen for 200 ms and immediately replaced by a mask for 100 ms. Then, after 1000 ms of black screen, a target set appeared and participants reported whether the target set had the same or a different numerosity with respect to the sample set by pressing the left or the right button of the keypad, respectively. The time allowed to provide a response was 8000 ms, otherwise the program skipped to the next trial and the response was categorized as missing. The target set had the same numerosity of the sample set (match condition) in half of the trials whereas in the other half the target numerosity was minus one or plus one dot with respect to the sample set (non-match condition). When the sample set numerosity was one dot or nine dots, the target in the non-match condition was two dots or eight dots, respectively. The size of the dots and their spatial arrangement was randomly selected at each trial (for both sample and target); moreover, the sample and the target sets had opposite contrast polarity (white dots and black dots, respectively). This ensured that participants could not base their judgments on visual cues but had to extract numerosity information from the display. There were 12 trials for each numerosity from 1 to 9 in the sample set, yielding a total of 108 trials.

The digit-to-dots match-to-sample task (Fig. 1, *panel b*) had the same structure of the dots-to-dots match-to-sample task except for one feature: instead of a sample set, an Arabic digit ranging from 1 to 9 was shown in the middle of the screen for

Table 1
Mean scores (standard deviations in parentheses) of intelligence tests, numerical battery (BIN) subtests and arithmetic tests for MA and DS children. The result of the statistical comparison between the two groups (t-values) are reported in the last column.

Measures	MA M(SD)	DS M(SD)	$t(40)$
PPVT-R	59(11)	56(13)	0.62
Raven	17(3)	14(4)	2.59*
BIN semantic	18(2)	17(3)	1.61
BIN lexical	16(6)	21(3)	3.44**
BIN counting	30(7)	32(10)	0.41
BIN pre-syntactical	15(5)	14(4)	0.51
BIN total score	79(18)	85(18)	1.09
Non-verbal calculation	4(1)	3(2)	2.22*
Arithmetic problems	2(1)	2(1)	0.45
Arithmetic facts	2(1)	3(2)	1.69
Arithmetic knowledge total score	8(2)	7(5)	0.17
Years of school	1(1)	12(3)	13.72**

* $p < 0.05$.
** $p < 0.01$.

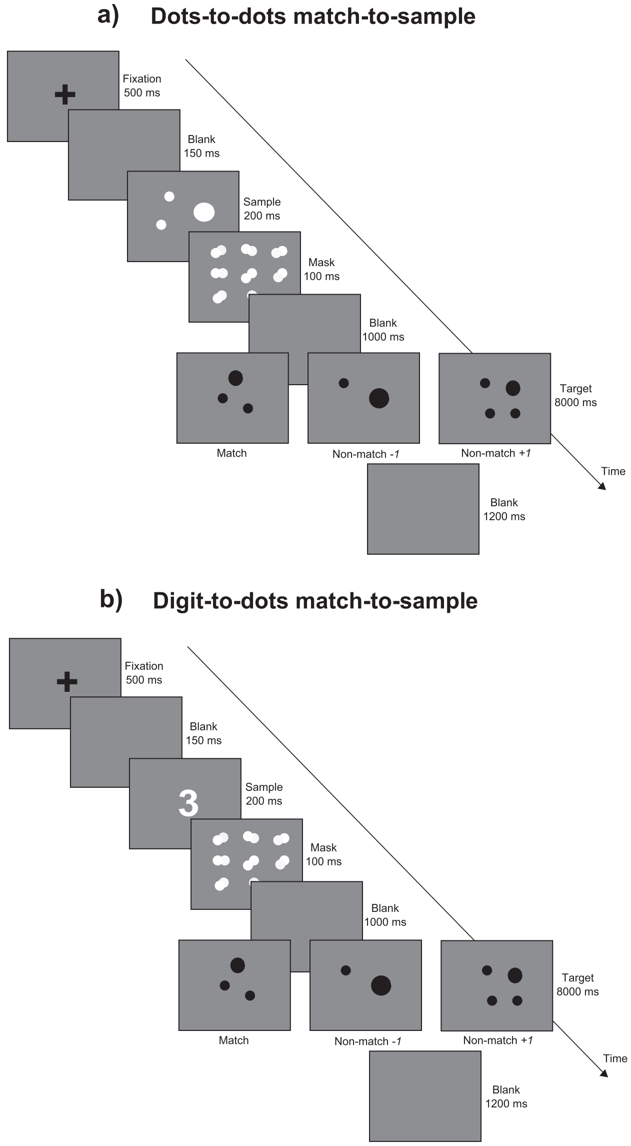

Fig. 1. In both tasks, participants decided whether the numerosity in the sample set was the same (match condition) or different (non-match condition) as compared to the target set. In the dots-to-dots match-to-sample task (*panel a*), the sample was composed of dots which whereas, in the digit-to-dots match-to-sample task (*panel b*), the numerosity of the sample was represented by an Arabic digit.

200 ms and immediately replaced by a mask for 100 ms. Participants reported whether the numerosity of the target matched or mismatched the number indicated by the sample (Arabic digit) by pressing the left or the right button of the keypad, respectively. There were 25 trials of training for each task in order to get the participants more familiar with the task demands.

2.3. Procedure

Participants sat in a quiet room approximately 60 cm from a 16-in. monitor. MA and DS met one to one with the experimenter for three sessions of approximately 30 min each. The assessment of mental age, using Raven's colored matrices and PPVT-R, was performed during the first session. The two computerized tasks were administered during the second and third session, respectively, with order of the tasks counterbalanced across participants. A typically developing child was included in the mental age control group when his/her raw scores on the PPVT-R was within 4 points (in either direction) of the score of the corresponding kid with DS. Similarly, a typically developing kid was included in the chronological age group when his/her chronological age was within 4 months (in either direction) of the corresponding kid with DS. CA children completed the two computerized tasks in counterbalanced order in one experimental session.

3. Results

We analyzed the data in a series of mixed and one-way ANOVAs. The Greenhouse–Geisser correction was applied in case of missing sphericity in the data. Follow-up statistical comparisons, given the reduced number of participants, were based on non-parametric analyses. The planned contrasts were two-tailed and the *p*-values were corrected for multiple comparisons analysis using the Bonferroni formula.

3.1. Dots-to-dots match-to-sample task

In order to perform a fine-grained analysis of the results, we categorized the trials into nine different conditions based on the number of dots in the sample set and in the target set, that is 1 vs 2, 2 vs 3, 3 vs 4, 4 vs 5, 5 vs 6, 6 vs 7, 7 vs 8, 8 vs 9 and 9 vs 9. For instance, the condition 1 vs 2 included the trials with one dot in the sample set and one or two dots in the target set and those trials with two dots in the sample set and one dot in the target set. Similarly, the 2 vs 3 condition included the trials with two dots in the sample set and two or three dots in the target set and those trials with three dots in the sample set and two dots in the target set. We applied the same logic to all other combinations. We note that the conditions 1 vs 2 and 8 vs 9 had 15 trials for each subject, the condition 9 vs 9 had 6 trials, whereas all other conditions had 12 trials.

In the examination of data, we discarded from the analysis participants who produced more than 15% of missing responses. An excessive number of missing responses may denote poor attention which could undermine the validity and reliability of the administered task. Moreover, given the dichotomous modality of response, we expected that participants exceeded the chance level at least in the easiest condition, namely 1 vs 2 dots. Accordingly, we removed participants who had a mean percentage of correct responses in this condition that did not significantly exceed the chance level according to a binomial test (i.e., 11 out of 15 correct responses for $p \leq 0.05$). This procedure reduced our DS group to 14 participants (8 males; M_{age} = 14;8 years, SD = 3;0 years; $M_{verbalMA}$ = 5;2, SD = 0;11 year; $M_{visuospatialMA}$ = 5;4 years, SD = 1;5 months) whereas MA children remained the same. One CA kid did not complete the task thus the resulting sample was composed by 20 individuals (8 males; M_{age} = 5;4 years, SD = 7 months). Nevertheless, DS and MA group were still matched for mental age, while DS and CA groups remained matched for chronological age ($p_s > 0.05$). We then calculated the mean percentage of correct responses for each condition removing from the computation the missing responses and responses faster than 200 ms (i.e., anticipations). We analyzed percentage of correct responses in a 8[1] [Condition: 1 vs 2, 2 vs 3, 3 vs 4, 4 vs 5, 5 vs 6, 6 vs 7, 7 vs 8, 8 vs 9] × 3 [Group: DS, MA, CA] mixed ANOVA with Condition as within-subjects factor and Group as between-subjects factor (Fig. 2). The main effect of Condition, $F(7, 364) = 43.36$, $p < 0.001$, and the main effect of Group, $F(2, 52) = 25.39$, $p < 0.001$, were both significant. The Condition by Group interaction was also significant, $F(14, 364) = 4.47$, $p < 0.001$. Importantly, the interaction remained significant, $F(7, 231) = 3.15$, $p = 0.004$, also when we included only DS and MA in the mixed ANOVA. Planned Mann–Whitney comparisons (two-tailed, Bonferroni adjusted to alpha level of 0.05/8 = 0.006) revealed that participants with DS showed a worse performance for conditions 2 vs 3 and 3 vs 4 in comparison to MA children (respectively, $Z = 2.77$, $U = 67$, $p = 0.006$, $r = 0.47$; $Z = 2.73$, $U = 67$, $p = 0.007$, $r = 0.46$).

3.2. Digit-to-dots match-to-sample task

In this task, visual enumeration was restricted to the target set because the sample was a symbolic number. Accordingly, we analyzed the percentage of correct responses and the reaction times as a function of the numerosity of the target set (see Fig. 3). The responses of children with DS were particularly slow and the time at their disposal (deadline after 8 seconds from target onset) was in some cases insufficient, thereby resulting in a large number of missing responses for trials with the largest numerosities. To ensure that each condition had a sufficient number of valid trials, we excluded from the analyses the trials with 7, 8, and 9 a target numerosities. Thereafter, we followed the same data cleaning procedure used for the dots-to-dots match-to-sample task. This procedure reduced the DS group to 12 participants (7 males; M_{age} = 13;8 years, SD = 3;5 years; $M_{verbalMA}$ = 5;0, SD = 6 months; $M_{visuospatialMA}$ = 5;1 years, SD = 1;3 year) and the MA children group to 19 participants (7 males; M_{age} = 5;3 years, SD = 8 months), whereas the CA children remained the same. Nevertheless, DS and MA group were still matched for mental age, while DS and CA group remained matched for chronological age. We calculated the mean percentage of correct responses for each target numerosity removing from the computation the missing responses and responses faster than 200 ms (i.e., anticipations). These were submitted to a 6 [Target Numerosity: 1, 2, 3, 4, 5, 6] × 3 [Group: DS, MA, CA] mixed ANOVA with Target Numerosity as within-subjects factor and Group as between-subjects factor (Fig. 3, *upper panel*). The main effect of Target Numerosity was not significant, $F < 1$, whereas the main effect of Group was significant, $F(2, 49) = 16.25$, $p < 0.001$. The Target Numerosity by Group interaction was significant, $F(10, 245) = 3.51$, $p = 0.001$. Nevertheless, when we ran the same analysis only with the DS and MA groups, the main effect of Group, $F(1, 29) = 3.91$, $p = 0.057$, and the interaction Target Numerosity x Group were no longer significant, $F(5, 145) = 1.77$, $p = 0.15$.

In order to investigate the fluency of counting, we calculated the individual slope of the linear regression with mean reaction times as dependent variable and the target numerosity as predictor (Fig. 3, *lower panel*), separately for subitizing range (i.e., 1, 2, 3) and counting range (i.e., 4, 5, 6). We analyzed the individual RT slopes in a 2 [Range: subitizing,

[1] Condition 9 vs 9 was not included in the analysis.

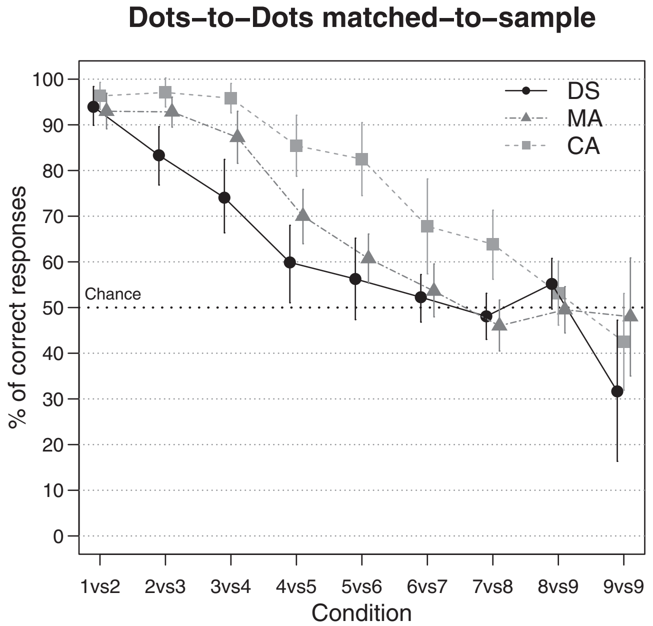

Fig. 2. Mean percentage of correct responses as a function of the conditions (error bars indicate 95% CI; dotted black line indicates the chance level).

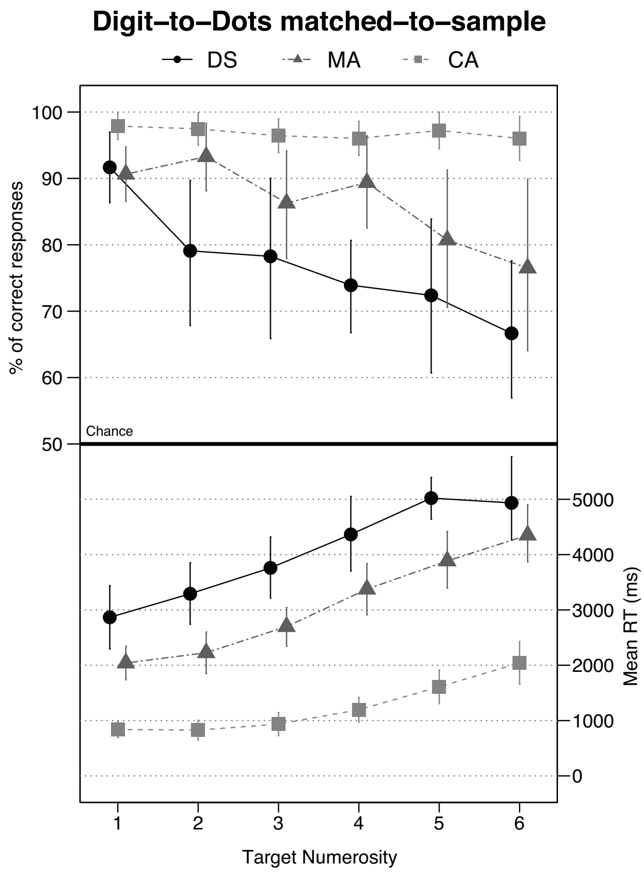

Fig. 3. *Upper panel*: Mean percentage of correct responses as a function of the numerosity of the target set (error bars indicate 95% CI; solid black line indicates the chance level). *Lower panel*: Mean reaction times as a function of the numerosity of the target set (error bars indicate 95% CI).

Table 2
Correlations between accuracy in the experimental tasks and tests scores (general intelligence, numerical and arithmetic knowledge).

Measures	Dots-to-dots match-to-sample				Digit-dots match-to-sample	
	Accuracy within subitizing (1vs2–3vs4)		Accuracy beyond subitizing (4vs5–8vs9)		Accuracy (1–6 target numerosity)	
	MA ($n = 21$)	DS ($n = 14$)	MA ($n = 21$)	DS ($n = 14$)	MA ($n = 19$)	DS ($n = 12$)
PPVT-R	0.17	0.51	−0.18	0.12	0.13	0.68*
Raven	−0.10	0.77*	0.10	0.63*	0.36	0.30
BIN total score	0.57**	0.54*	−0.20	0.08	0.01	0.10
Arithmetic knowledge total score	0.33	0.58*	−0.23	0.18	0.03	0.44

* $p < 0.05$.
** $p < 0.01$.

counting] × 3 [Group: DS, MA, CA] mixed ANOVA with Range as within-subjects factor and Group as between-subjects factor. The main effect of Range, $F(1, 49) = 18.82$, $p < 0.001$, and the main effect of Group, $F(2, 49) = 3.24$, $p = 0.047$, were both significant. The interaction between Range and Group was also significant, $F(2, 49) = 6.22$, $p < 0.01$. Therefore, we run two one-way Kruskal–Wallis (non-parametric) ANOVAs on the individual RT slopes for subitizing and counting ranges, using Group as between subjects factor. The effect of the Group was significant only for the slope of the subitizing range, $\chi^2(2, N = 52) = 18.57$, $p < 0.001$. Planned comparisons (Bonferroni adjusted to alpha level of 0.05/3 = 0.016) revealed that the CA group had a smaller RT slope in the subitizing range ($M = 48$ ms, $SD = 153$) as compared to MA group ($M = 327$ ms, $SD = 328$), $Z = 3.39$, $U = 74$, $p < 0.001$, $r = 0.54$, and DS group ($M = 450$ ms, $SD = 434$), $Z = 3.63$, $U = 29$, $p < 0.001$, $r = 0.63$. Conversely, the DS group had a RT slope that did not differ from that of the MA group, $Z = 1.5$, $U = 151$, $p = 0.141$, $r = 0.27$.

3.3. Correlation analysis

We run a correlational analysis in order to highlight the relation between the performance in the computerized tasks, general intelligence, numerical competence and arithmetic score (Table 2). We found a significant correlation between accuracy in the subitizing range and BIN battery total score in both MA and DS children. Accuracy in DS children was also correlated to Arithmetic knowledge (total score) and to general IQ (Raven score). Estimation accuracy only correlated with Raven score for the DS group, whereas the ability to correctly count in the digit-to-dots match to sample task only correlated with Peabody score for DS children.

4. Discussion

In the present study, we employed two delayed match-to-sample tasks in order to evaluate visual enumeration skills in children with DS as compared to typically developing individuals matched either for mental or chronological age. In the dots-to-dots match-to-sample task, participants had to enumerate visual sets with numerosities both within and beyond the subitizing range. This task allowed us to systematically evaluate the functioning of both the OTS and the ANS. The most striking result is that performance of children with DS in the subitizing range was impaired even in comparison to the MA group. The accuracy level decreased with increasing numerosity within the subitizing range, whereas the accuracy of both MA and CA group was at ceiling. Thus, participants with DS displayed a ratio dependent effect within the subitizing range, as if they relied on the ANS for enumerating small numerosities, whereas MA and CA deployed the OTS to accurately individuate the number of objects in the sets. This finding supports the hypothesis of an impaired OTS in DS and it is consistent with the results of Paterson et al. (2006) who tested young children with DS using a habituation paradigm and found a specific deficit in the discrimination of small quantities (2 vs 3 elements).

It is worth noting that recent studies have highlighted that subitizing and visual short term memory share the same cognitive resources, thereby suggesting a common reliance on the OTS (Cutini, Sella, & Zorzi, submitted for publication; Piazza, Fumarola, Chinello, & Melcher, 2011). In this light, further support to the hypothesis of OTS impairment in individuals with DS comes from studies on memory, and specifically from the finding of a specific deficit in visual short term memory in DS (Carretti & Lanfranchi, 2010; Carretti, Lanfranchi, & Mammarella, 2013; Lanfranchi, Carretti, Spanò, & Cornoldi, 2009). Therefore, an impaired OTS might be responsible for atypical performance in both visual short term memory and enumeration tasks.

Discrimination of larger numerosities, beyond the subitizing range, showed the typical ratio dependent effect – the key signature of ANS functioning – in both DS and typically developing groups. Importantly, numerosity discrimination in DS individuals was less accurate in comparison to CA children but similar to MA controls. This finding suggests that the development of ANS acuity in DS follows a trajectory that is aligned with mental rather than chronological age. In other words, children with DS show a severe delay in ANS acuity with respect to their chronological age, in contrast to the conclusions of the study by Camos (2009). Given that number acuity has been recently found to correlate with mathematics achievement (Halberda et al., 2008; Mazzocco et al., 2011a) and to be impaired in children with developmental dyscalculia (Mazzocco et al., 2011b; Piazza et al., 2010), it is conceivable that its delayed development contributes to math underachievement in children with DS. Moreover, our finding fits well with the observation that the numerical abilities of children with DS (including symbolic number processing, as measured by the standardized battery) were aligned with their mental rather than chronological age.

In the digit-to-dots match-to-sample task, the numerosity of the sample set was represented by an Arabic digit and participants compared its numerical value with the cardinality of the target set. The performance of children with DS showed a pattern that is consistent with the use of a serial counting routine. Notably, RTs increased systematically as a function of target numerosity, with a slope that did not differ from that of MA controls.

Thus, despite their slower and slightly less accurate performance, children with DS showed a level of counting proficiency that is adequate for their mental age. Therefore, our results are relevant for the debate on whether children with DS master the cardinality principle or they learn to count by rote without a deep understanding of counting (Bashash et al., 2003; Caycho et al., 1991; Gelman & Cohen, 1988; Porter, 1999). In this regard, it is important to highlight that the understanding of cardinality was a necessary condition for properly accomplish the task, because the cardinality of the target set had to be compared to the numerical magnitude conveyed by the Arabic digit in the sample.

Finally, the correlation analysis (although limited by the small sample size) suggested that the OTS capacity, as measured in the dots-to-dots match to sample task, was related to basic numerical competence in both DS and MA children. Interestingly, the DS and MA group did not differ in their performance in the BIN battery, thereby suggesting a greater sensibility of the computerized task with respect to the paper and pencil battery in highlighting significant differences between the two groups (Bonato & Deouell, 2013). Moreover, general IQ (Raven score) of DS children correlated with their accuracy both within and beyond the subitizing range, as well as with their arithmetic performance.

5. Conclusions

In summary, our results suggest that children with DS have a specific deficit in OTS capacity. In contrast, number acuity (supported by the ANS) and the understanding of cardinality, though severely delayed in children with DS with respect to their chronological age, were adequate for their mental age. Thus, mathematics underachievement in children with DS is likely to stem from weakness in these basic numerical skills that are thought to be foundational to mathematical learning.

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References

Abdelahmeed, H. (2007). Do children with Down syndrome have difficulty in counting and why. *International Journal of Special Education, 22*, 1–11.

Bashash, L., Outhred, L., & Bochner, S. (2003). Counting skills and number concepts of students with moderate intellectual disabilities. *International Journal of Disability, Development and Education, 50*(3), 325–345.

Bonato, M., & Deouell, L. Y. (2013). Hemispatial neglect: computer-based testing allows more sensitive quantification of attentional disorders and recovery and might lead to better evaluation of rehabilitation. *Frontiers in Human Neuroscience, 7*, 162.

Bonato, M., Sella, F., Berteletti, I., & Umiltà, C. (2012). Neuropsychology is nothing without control: A potential fallacy hidden in clinical studies. *Cortex, 48*, 353–355.

Brigstocke, S., Hulme, C., & Nye, J. (2008). Number and arithmetic skills in children with Down syndrome. *Down Syndrome: Research and Practice,* 74–78. http://dx.doi.org/10.3104/reviews.2070

Buckley, S., & Sacks, B. (1987). *The adolescent with Down syndrome: Life for the teenager and for the family*. Portsmouth, UK: Portsmouth Polytechnic.

Camos, V. (2009). Numerosity discrimination in children with Down syndrome. *Developmental Neuropsychology, 34*, 435–447.

Carey, S. (2001). Cognitive foundations of arithmetic: Evolution and ontogenesis. *Mind & Language, 16*, 37–55.

Carr, J. (1988). Six weeks to twenty-one years old: a longitudinal study of children with Down's syndrome and their families. *Journal of Child Psychology and Psychiatry, 29*, 407–431.

Carretti, B., & Lanfranchi, S. (2010). The effect of configuration on VSWM performance of Down syndrome individuals. *Journal of Intellectual Disability Research, 54*, 1056–1058.

Carretti, B., Lanfranchi, S., & Mammarella, I. (2013). Spatial-simultaneous and spatial-sequential working memory in individuals with Down syndrome: the effect of configuration. *Research in Developmental Disability, 34*, 669–675.

Caycho, L., Gunn, P., & Siegal, M. (1991). Counting by children with Down syndrome. *American Journal on Mental Retardation, 95*(5), 575–583.

Cutini, S., Sella, F., & Zorzi, M. (2013). Visual short-term memory load disrupts subitizing limit, submitted for publication.

Dehaene, S. (1997). *The number sense: How the mind creates mathematics*. New York, NY: Oxford University Press.

Dehaene, S., Izard, V., Spelke, E., & Pica, P. (2008). Log or linear? Distinct intuitions of the number scale in Western and Amazonian indigene cultures. *Science (New York, N.Y.), 320*(5880), 1217–1220.

Dunn, L. M., & Dunn, L. M. (1997). *Peabody picture vocabulary test* (3rd ed.). Circle Pines, MN: American Guidance Service.

Dykens, E. M., Hodapp, R. M., & Finucane, B. M. (2000). *Genetics and mental retardation syndromes*. New York: Paul H Brookes.

Feigenson, L., Dehaene, S., & Spelke, E. (2004). Core systems of number. *Trends in Cognitive Sciences, 8*(7), 307–314.

Gallistel, C. R., & Gelman, R. (1992). Preverbal and verbal counting and computation. *Cognition, 44*, 43–74.

Gelman, R., & Cohen, M. (1988). Qualitative differences in the way Down syndrome and normal children solve a novel counting problem. In L. Nadel (Ed.), *The Psychobiology of Down's Syndrome*. Cambridge, MA: MIT Press.

Gelman, R., & Gallistel, C. R. (1978). *The child's understanding of number*. Cambridge, MA: Harvard University Press.

Halberda, J., & Feigenson, L. (2008). Developmental change in the acuity of the "Number Sense": The approximate number system in 3-, 4-, 5-, and 6-year-olds and adults. *Developmental Psychology, 44*(5), 1457–1465.

Halberda, J., Ly, R., Wilmer, J. B., Naiman, D. Q., & Germine, L. (2012). Number sense across the lifespan as revealed by a massive Internet-based sample. *Proceedings of the National Academy of Sciences of the United States of America, 109*(28), 11116–11120.

Halberda, J., Mazzocco, M. M. M., & Feigenson, L. (2008). Individual differences in non-verbal number acuity correlate with maths achievement. *Nature, 455*(7213), 665–668.

Jordan, N. C., Kaplan, D., Locuniak, M. N., & Ramineni, C. (2007). Predicting first-grade math achievement from developmental number sense trajectories. *Learning Disabilities Research & Practice, 22*(1), 36–46.

Kaufman, E. L., Lord, M. W., Reese, T., & Volkmann, J. (1949). The discrimination of visual number. *American Journal of Psychology, 62*, 496–525.

Kittler, P. M., Krinsky-McHale, S. J., & Devenny, D. A. (2008). Dual-task processing as a measure of executive function: A comparison between adults with Williams and Down syndromes. *American Journal of Mental Retardation, 113*(2), 117–132.

Landerl, K., Bevan, A., & Butterworth, B. (2004). Developmental dyscalculia and basic numerical capacities: A study of 8- and 9-year-old students. *Cognition, 93*, 99–125.

Lanfranchi, S., Carretti, B., Spanò, G., & Cornoldi, C. (2009). A specific deficit in visuospatial simultaneous working memory in Down syndrome. *Journal of Intellectual Disability Research, 53*, 474–483.

Le Corre, M., & Carey, S. (2007). One, two, three, four, nothing more: an investigation of the conceptual sources of the verbal counting principles. *Cognition, 105*(2), 395–438.

Libertus, M. E., Feigenson, L., & Halberda, J. (2011). Preschool acuity of the approximate number system correlates with school math ability. *Developmental Science, 14*(6), 1292–1300.

Lourenco, S. F., Bonny, J. W., Fernandez, E. P., & Rao, S. (2012). Nonsymbolic number and cumulative area representations contribute shared and unique variance to symbolic math competence. *Proceedings of the National Academy of Sciences of the United States of America, 109*(46), 18737–18742.

Mandler, G., & Shebo, B. J. (1982). Subitizing: An analysis of its component processes. *Journal of Experimental Psychology: General, 111*, 1–22.

Mazzocco, M. M. M., Feigenson, L., & Halberda, J. (2011a). Preschoolers' precision of the approximate number system predicts later school mathematics performance. *PLoS ONE, 6*(9), e23749.

Mazzocco, M. M. M., Feigenson, L., & Halberda, J. (2011b). Impaired acuity of the approximate number system underlies mathematical learning disability (dyscalculia). *Child Development, 82*(4), 1224–1237.

Moeller, K., Neuburger, S., Kaufmann, L., Landerl, K., & Nuerk, H. C. (2009). Basic number processing deficits in developmental dyscalculia. Evidence from eye-tracking. *Cognitive Development, 24*, 371–386.

Molin, A., Poli, S., & Lucangeli, D. (2007). *Batteria Intelligenza Numerica [Battery for Numerical Intelligence]*. Trento: Ed Erickson.

Nye, J., Fluck, M., & Buckley, S. (2001). Counting and cardinal understanding in children with Down syndrome and typically developing children. *Down Syndrome: Research and Practice, 7*(2), 68–78.

Passolunghi, M. C., Vercelloni, B., & Schadee, H. (2006). The precursors of mathematics learning: Working memory, phonological ability and numerical competence. *Cognitive Development, 22*, 165–184.

Paterson, S. J., Girelli, L., Butterworth, B., & Karmiloff-Smith, A. (2006). Are numerical impairments syndrome specific? Evidence from Williams syndrome and Down's syndrome. *Journal of Child Psychology and Psychiatry, 47*, 190–204.

Piazza, M. (2010). Neurocognitive start-up tools for symbolic number representations. *Trends in Cognitive Sciences, 14*(12), 542–551.

Piazza, M., Facoetti, A., Trussardi, A. N., Berteletti, I., Conte, S., Lucangeli, D., et al. (2010). Developmental trajectory of number acuity reveals a severe impairment in developmental dyscalculia. *Cognition, 116*(1), 33–41.

Piazza, M., Fumarola, A., Chinello, A., & Melcher, D. (2011). Subitizing reflects visuo-spatial object individuation capacity. *Cognition, 121*(1), 147–153.

Porter, J. (1999). Learning to count: A difficult task? *Down Syndrome: Research and Practice, 6*(2), 85–94.

Raven, J., Raven, J. C., & Court, J. H. (1998). *Coloured progressive matrices*. Oxford: Oxford Psychologists Press.

Schleifer, P., & Landerl, K. (2011). Subitizing and counting in typical and atypical development. *Developmental Science, 14*, 280–291.

Stoianov, I., & Zorzi, M. (2012). Emergence of a "visual number sense" in hierarchical generative models. *Nature Neuroscience, 15*, 194–196.

Trick, L. M., & Pylyshyn, Z. W. (1994). Why are small and large numbers enumerated differently: A limited-capacity preattentive stage in vision. *Psychology Review, 101*, 80–102.

Xu, F., & Spelke, E. S. (2000). Large number discrimination in 6-month-old infants. *Cognition, 74*(1), B1–B11.

Xu, F., Spelke, E. S., & Goddard, S. (2005). Number sense in human infants. *Developmental Science, 8*(1), 88–101.