# The foundations of mathematical development in Williams syndrome and Down syndrome

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Acknowledgements

We would like to thank all of the parents and children for taking part in this project. Further thanks also go to Eleonora Gabriele for her help with data input. This work was supported by a grant from the British Academy (SG151035).

# Abstract

Background: Studies in Down syndrome (DS) and Williams syndrome (WS) have suggested that mathematical abilities are impaired. However, it is unclear which domain general or domain specific abilities impact on mathematical development in these developmental disorders. Method: The current study examined the foundations of mathematical development across participants with WS (n = 24) and DS (n = 26) compared to typically developing (TD) children (n = 26) in relation to domain general (i.e., general intelligence and visuospatial abilities) and domain specific abilities (non-symbolic and symbolic number abilities). Results: Developmental trajectories showed that mathematical abilities were delayed in line with overall mental age in DS and WS. Whilst visuospatial abilities predicted performance for DS and TD participants, this was not the case for the WS group, instead Approximate Number Sense abilities predicted mathematical development. Conclusions: These findings suggest that those with DS and WS may benefit from different mathematical intervention programmes.

*Keywords:* Mathematics; ANS; symbolic; Williams syndrome; Down syndrome

Williams syndrome (WS) is a rare congenital developmental disorder that affects approximately 1 in 20,000 life births and is caused by a micro deletion of approximately 27 genes on the long arm of chromosome 7 (Koehler, Pabst, Poher, & Kozel, 2014). It is characterized by a number of behavioural and clinical difficulties, including cardiovascular difficulties and a specific facial morphology, as well as a particular cognitive profile and average IQ scores between 42-68 (Martens, Wilson & Reutens, 2008). However, verbal abilities tend to be better compared to non-verbal abilities and this discrepancy increases with chronological age (Jarrold, Baddeley & Hewes, 1998). Research has also shown that their language development is atypical and that they use their relatively strong verbal short term memory abilities to learn verbal expressions without necessarily attaching meaning to it (Naylor & Van Herwegen, 2012). Down syndrome (DS) is a more common neurodevelopmental disorder affecting about 1 in 800 life births (Silverman, 2007). Similar to WS, individuals with DS show a general cognitive delay with average IQ scores between 50-70 (Mervis et al., 2000). However, performance on non-verbal tasks tends to be better when compared to verbal performance. Individuals with DS also show significant difficulties with short-term memory abilities and language production (Silverman, 2007). The varying overlapping strengths and difficulties in these disorders ’phenotypes allow for useful cross-syndrome comparisons and allow us to examine the impact of difficulties early on in life on phenotypical outcomes (Ball & Karmiloff-Smith, 2015; Van Herwegen & Karmiloff-Smith, 2015).

Mathematical abilities are impaired in both individuals with WS and DS (Ansari, Donlan, Thomas, Ewing & Karmiloff-Smith, 2003; Patterson, Girelli, Butterworth, & Karmiloff-Smith, 2003; O’Hearn & Landau, 2007; Brigstocke, Hulme, & Nye, 2008). In addition, older children and adults with DS perform better on a battery of mathematical ability tasks than those with WS (Paterson, Girelli, Butterworth, & Karmiloff-Smith, 2006).

Recent research in typically developing (TD) children has shown that mathematical abilities rely on a number of domain general abilities (Schneider et al., 2017), including a wide range of visuospatial skills such as mental rotation, but also spatial visualisation (the ability to mentally transform images and objects) and disembedding (the ability to extract pertinent information from a complex visual scenes), amongst others (Uttal et al., 2013). In addition to domain general skills, domain specific skills are also important for general mathematical achievement. The Approximate Number Sense (ANS), a noisy, imprecise, non-verbal system allows discrimination of large numerosities without counting or numerical symbols (Feigenson, Dehaene, & Spelke, 2004). Studies in TD children (for recent reviews and meta-analysis see Chen & Li, 2014; Fazio, Bailey, Thompson, & Siegler, 2014) have shown that ANS task performance is related to children's general mathematical skills, though conflicting results have also been found. Additional important domain specific skills include familiarity with the number system (e.g., identification on number symbols, knowledge of count word lists), flexible and adaptive strategy use when completing mathematical problems, and fast and efficient math fact retrieval (Geary et al., 2017).

Previous research has suggested that the underlying cause of mathematical difficulties in WS and DS is caused by domain specific deficits. A number of studies have suggested that ANS abilities are impaired in WS compared to TD controls, including those matched for mental age (Ansari, Donlan & Karmiloff-Smith, 2007; O'Hearn & Landau, 2007; Libertus, Feigenson, Halberda, & Landau, 2014; Paterson et al., 2006; Van Herwegen, Ansari, Xu, & Karmiloff-Smith, 2008) but not DS (Karmiloff-Smith et al., 2012; Sella, Lanfranchi & Zorzi, 2013). However, some research suggests that magnitude task performance in WS is delayed in line with mental age abilities (Abreu-Mendoza & Arias-Trejo, 2015; Rouselle, Dembour, & Noël, 2013). It has been argued that this ANS impairment explains the mathematical delays in WS (O'Hearn & Landau, 2007; Libertus et al., 2014). Others have argued that domain general abilities, such as low-level visuospatial dysfunction in WS, are the origin of their mathematical difficulties (Karmiloff-Smith et al., 2012; Rouselle et al., 2013). However, in contrast to TD children, visuospatial abilities are not a good predictor for mathematical abilities in WS (Ansari et al., 2003), perhaps due to low levels of performance (i.e. performance at floor) on visuospatial assessments and limited development of visuospatial abilities over developmental time (Van Herwegen, Rundblad, Davelaar & Annaz, 2011). In contrast, mathematical abilities in DS have been argued to be in line with language abilities (Nye, Clibbens, & Bird, 1995) and correlate with ANS abilities (Sella et al., 2013).

There have also been inconsistent findings in research on symbolic skills in these groups. Whilst young children with DS had shorter counting sequences than TD children (Nye, Fluck, & Buckley, 2001) and were less able to detect counting errors (Porter, 1999), those with WS perform similar to controls (Ansari et al., 2003; Robinson & Temple, 2013). However, there was no difference in counting sequence knowledge between older participants with DS and WS (Paterson et al., 2006). Individuals with WS have also been shown to perform better on numerical fact retrieval as well as addition and subtraction problems compared to DS participants, and it has been argued that individuals with WS may rely heavily on rote memorization for accurate performance on these types of tasks (Krajcs, Lukács, Igács, Racsmány & Pléh, 2011). Finally, in WS, better performance was observed on a verbal magnitude comparison task when compared to a non-symbolic magnitude task (Libertus et al., 2014), suggesting that symbolic number abilities in WS are better compared to non-verbal mathematical abilities (Krajcs et al., 2011). The opposite pattern has been proposed for those with DS (Brigstoke et al., 2008).

These cross-syndrome findings would suggest that the mathematical impairments in WS and DS are caused by different domain specific factors; ANS difficulties for WS and difficulties with precise or symbolic abilities for those with DS. As this dissociation is already present from infancy (Karmiloff-Smith et al., 2012), WS and DS would provide evidence about the innate specification of dissociated modules in the brain for mathematics. However, most of these previous studies in WS and DS have taken a static approach when researching mathematical abilities, in that they have only looked at mathematical abilities in a specific age group. However, a truly developmental approach is required when studying neurodevelopmental disorders (Karmiloff-Smith 1998), in that domain general abilities, such as language and visuospatial abilities, may impact on the development of domain specific abilities and these domain general abilities may provide a better explanation for the number difficulties and abilities observed in these neurodevelopmental disorders (See Van Herwegen & Karmiloff-Smith, 2015 for a discussion). To date, there has been one longitudinal study in each disorder. These have suggested that mathematical abilities in WS plateau at the mental age of an 8-year-old (Udwin, Davies, & Howlin, 1996) and that mathematics in individuals with DS plateaus around the chronological age of 16-18 years old (Turner & Alborz, 2003).

## The current study

There is current debate in the literature on the underlying cause of mathematical difficulties in WS and DS. Previous studies examining what abilities relate to the development of mathematical abilities in WS and DS are scarce and they have often measured one potential underlying factor, rather than a range of domain general and specific abilities that have been shown to impact on mathematical development in TD populations. In addition, direct comparisons between WS and DS are rare and often use different tasks, making direct comparisons between different studies and age cohorts difficult. Knowing which factors relate to mathematical delay and how these differ between neurodevelopmental disorders will allow for the development of more effective educational programmes and interventions.

The current study is the first to focus on the development of mathematical foundation skills in individuals with DS and WS from a wide age range as well as TD children and to examine:

1) The developmental pathways of mathematical abilities in the groups: Are individuals with WS and DS delayed to a similar degree from the start of development or do the delays differ over development?
2) The interaction between mathematical development and domain general abilities (i.e., visuospatial abilities, general IQ) in the groups: Do mathematical abilities develop in line with the same or different domain general abilities in the three groups?
3) The interaction between mathematical development and domain specific abilities (i.e., symbolic and non-symbolic abilities) in the groups: Do mathematical abilities develop in line with the same or different domain specific abilities in the three groups?

Based upon the previous research, it was predicted that mathematical abilities would plateau with increasing age in DS (Turner & Alborz, 2003) and WS (Udwin et al., 1996) but that participants with DS would outperform those with WS (Paterson et al., 2006). In addition, whilst it was predicted that general mathematical abilities in WS would be in line with overall mental age and ANS abilities, mathematical abilities in DS may be related to domain general abilities and symbolic skills, but not ANS abilities.

## Method

### Participants

Twenty-four participants with WS (17 females) aged 8;00 to 51;08 years old were recruited via the Williams Syndrome Foundation, including an equal number of children under the age of 18 and adults over the age of 18. All of the participants with WS had a positive diagnosis for WS using the genetic fluorescent in situ hybridisation (FISH) test confirming the genetic deletion implicated in WS, in addition to a clinical diagnosis for WS.

Twenty-six participants with DS aged 8;08 to 49;02 (12 females) were recruited via Down syndrome support groups across the South-East of the UK. All of the DS participants had a genetic mutation on chromosome 21. Twenty-six TD children (23 females) were recruited whose chronological age fell within the range of performance scores of the two neurodevelopmental groups for the Raven's Coloured Progressive Matrices (RCPM; Raven, 2007) which measures fluid intelligence. We chose not to include TD participants of a similar chronological age as the WS and DS groups as previous studies have already shown that mathematical abilities in WS and DS are delayed and not in line with their chronological age (Ansari et al., 2003; Patterson et al., 2003; O'Hearn & Landau, 2007; Brigstocke et al., 2008). Therefore, the TD children were aged between 4;06 and 10;02 years old. All of the participants had English as a first language and none of the TD participants had a diagnosis for a learning difficulty. Further participant details can be found in Table 1.

Table 1 about here

## Materials

*Mathematical Achievement* was measured using the Numerical Operations sub-test from Wechsler Individual Achievement Test (WIAT; Wechsler, 2005). This task assessed written math calculation skills including basic skills, basic operations with integers, geometry, algebra, and calculus. Participants were presented with a worksheet and asked to complete as many operations as they could, without any time limitations. The task was terminated once the participant produced six consecutive incorrect answers. The total number of correctly completed items was used as a raw score.

*General Intelligence* was assessed using the RCPM. This task includes 36 trials in which the participant had to identify which picture out of 6 options completed a pattern.

Participants completed all 36 trials and the total number of correct answers was used to calculate a raw score.

*Visuospatial abilities* were measured using the Pattern Construction Task (PC task) from the British Ability Scales-II (Elliot, Smith, & McCullock, 1997). Participants were presented for the first 6 trials with two-dimensional yellow and black foam squares and then with three-dimensional plastic yellow and black sided cubes and asked to replicate patterns from a book. Ability scores were used to assess performance on this task (see Van Herwegen et al., 2011 for a similar approach).

*Non-symbolic number abilities* were measured using a classic non-symbolic ANS Task (See Gebuis and Reynvoet, 2011 for a similar task). In this computerised task participants were presented with a set of blue and red dot presentations on the left and right of the screen. The dot presentations included between 5 and 28 dots and the dot presentations in each trial included either ratios 0.5, 0.6, 0.7 or 0.8 distributed across 48 trials. In half of the trials dot size correlated with the number of dots (i.e., congruent trials) and in the other half of the trials there was no systematic relationship between dot size and the number of dots (i.e., incongruent trials). The presentation with ‘more ’dots was counterbalanced, appearing on either the left or right side of the screen. Stimuli were only presented for 1500 msec in order to prevent participants counting the dots. Participants were asked to select the dot presentation that had ‘more ’by pointing or saying the colour of the dot set.

Prior to the actual ANS task, participants were administered a practice task which showed two dot presentations that had a ratio difference of 1/3 between them. In this training task participants received up to 24 training trials (or until they have 8 consecutive trials correct). Participants received feedback when they picked the incorrect answer (see Negen & Sarnecka, 2015 for a similar approach).

*Symbolic number abilities* were assessed using a range of counting and recognition tasks that measure number familiarity. Participants were asked to count out loud to 20 starting from 1. The highest number correctly counted was recorded (max. = 20). Next, participants were asked to count backwards from 20. Again, the number of correctly backward counted numbers was recorded (max. = 20). Finally, participants were asked to count onwards from 25 and given 1 point if they could do so (see Patterson et al., 2006 for a similar approach). Recognition of digits was assessed by showing participants flashcards with numbers 1 to 20 in random order. Participants were asked to name the digit. The total number of digits correctly named was recorded (max. = 20). For all tasks, participants' final answer was counted when they corrected themselves. An average percent number familiarity score was calculated by averaging the percent correct across the four tasks.

## Procedure

Parents were provided with detailed information about the project and gave written consent, whilst verbal assent was obtained from all children. This project was approved by the xxxx Ethics Committee.

The tasks were presented in random order to participants and participants were assessed one-on-one with a researcher in a quiet room at the university. The total testing session took about 1 hour, breaks were taken as often and long as required.

## Data analysis plan

Cross-sectional developmental trajectories were used to examine the development of mathematical abilities in each group (see Thomas et al., 2009 for full details of this approach). First, the relationship between mathematical ability and chronological age (CA) were assessed by modelling developmental trajectories for each group separately using ANCOVA with age as covariate and mathematical abilities as dependent outcome. Before running the ANCOVA, we took the youngest age in the TD group and subtracted this age from the CA of all other data-points in all three groups to make sure meaningful comparisons could be made. Trajectories can only be compared if the independent trajectories are significant, due to the fact that unreliable trajectories can either be caused by the fact that there is a true zero trajectory or because there is a non-systematic trajectory (see Thomas et al., 2009 for a discussion). Therefore, only when CA explained a significant amount of variance in mathematical performance in each group, the trajectories from the three groups were compared using cross-sectional ANCOVA tests. This allowed investigation of any developmental differences between the groups for the *onset of development*, i.e., differences at the youngest age or lowest performance measured, and *rate of development*, i.e., the slope of the trajectory. In the second phase, these analyses were repeated but mathematical performance was plotted against performance on  domain general tasks (i.e., general intelligence and visuospatial abilities) and domain specific tasks (non-symbolic and symbolic number abilities), in order to investigate whether domain general and/or domain specific abilities were a better predictor for general mathematical abilities. Again before running the ANCOVA with performance scores, we took the lowest performance score and subtracted this score from the data-points in all three groups to ensure that meaningful comparisons could be made. Outliers were identified using Cook's distance (i.e., above or below three times the mean). Finally, in order to ensure that none of the results were caused by performance at floor (i.e., the lowest score available) and ceiling level (i.e., highest score available), each of the analyses was repeated excluding performance at floor- and ceiling level. Only when excluding these scores, or excluding the outliers, changed the results were these analyses reported.

# Results

**A o**ne-way ANOVA indicated that the groups differed significantly in CA; *F*(2,73)= 20.369, $p < .001$, $\eta^2_p$= .96 with post-hoc pairwise comparisons showing that the TD group was significantly younger than the WS ($p = .001$) and the DS group ($p < .001$), but that there was no difference between the WS and the DS group ($p = .635$). However, a one-way ANOVA did not show any difference between the three groups for RCPM raw scores; *F*( 2,70)= 1.047, $p = .357$, which suggests that the groups have similar fluid intelligence abilities and that the groups can be compared when collapsed over age.

## Mathematical performance in relation to chronological age

As shown in Figure 1, while WIAT scores increased significantly with increasing CA in the TD group; *F*(1, 25) = 59.098, $p < .001$, $\eta^2_p$= .711, there was no systematic relationship between the scores and increasing CA in either the WS group; *F*(1, 23) = 1.184, $p = .288$, $\eta^2_p$= .051 or the DS group; *F*(1, 23) = .028, $p = .869$, $\eta^2_p$= .001.

As CA was not a reliable predictor for performance in the neurodevelopmental groups, WIAT scores were evaluated using an ANOVA collapsed over age. This comparison showed that, although WIAT scores in the WS group (mean: 8.63, SD = 3.91) and the DS group (mean: 8.29, SD = 3.43) were lower than in the TD group (mean: 11.27, SD = 6.32), these group differences were not significant; *F*(2, 73) = 2.950, $p = .059$, $\eta^2_p$= .077.

## Mathematical performance in relation to domain general and domain specific abilities

For each group, the RCPM could not be completed by one participant due to participant refusal or lack of concentration. Plotting WIAT scores against RCPM scores showed that there was a reliable trajectory for the TD group; *F*(1, 24) = 14.604, $p < .001$, $\eta^2_p$= .388, the WS group; *F*(1, 22) = 21.346, $p < .001$, $\eta^2_p$= .504, and the DS group; *F*(1, 22) = 12.417, $p = .002$, $\eta^2_p$= .374. As there was a reliable trajectory within each group, the developmental trajectories of each group were compared. There was an overall effect for RCPM scores, in that, overall the model was significant; *F*(1, 70) = 40.368, $p < .001$, $\eta^2_p$= .383. However, there was no effect of group; *F*(2, 70) = 1.196, $p < .309$, $\eta^2_p$= .035 or a significant interaction between group and RCPM; *F*(2, 70) = 2.419, $p = .097$, $\eta^2_p$= .069. This demonstrated that there were no differences in the onset or the developmental rate of mathematical abilities between the three groups when performance was plotted against RCPM scores (see Figure 2).

Figure 2 about here

For the PC task, data from 5 participants was missing across the three groups. This was due to lack of concentration which meant that the testing session was finished early or participant refusal to complete the task. In addition, two participants with WS performed at floor level. As excluding these participants who performed at floor from the analyses changed the results, the final reported analyses excludes these two participants. There was a reliable trajectory when WIAT scores were plotted against PC-test scores in the TD group; *F*(1, 21) = 14.067, $p = .001$, $\eta^2_p$= .413, and in the DS group; *F*(1, 21) = 12.321, $p = .002$, $\eta^2_p$= .381 but not the WS group; *F*(1, 20) = 2.814, $p = .110$, $\eta^2_p$= .129. As there was no reliable trajectory for the WS group (see Figure 3), only performance in the DS group could be directly compared to the TD group. This comparison showed, although visuospatial abilities were a reliable predictor for mathematical abilities; *F*(1, 43) = 24.507, $p < .001$, $\eta^2_p$= .380, there was no significant effect of group; *F*(1, 43) = 1.753, $p = .193$, $\eta^2_p$= .042 or significant interaction of group by PC-task scores; *F*(1, 43) = 2.735, $p = .106$, $\eta^2_p$= .064. This shows that the rate and the onset of development of mathematical abilities in the DS and the TD group was the same when performance was plotted against visuospatial abilities. Comparison of the visuospatial abilities collapsed over age between the three groups, using an ANOVA, showed that there was a significant effect of Group; *F*(2, 64) = 11.160, $p < .001$, $\eta^2_p$= .265. As can be seen in Table 2, the WS group displayed substantially poorer performance when compared to the TD ($p < .001$) and the DS group ($p = .050$) and the DS performed similar to the TD group ($p = .076$).

Figure 3 about here

Table 2 about here

For non-symbolic abilities, all participants managed to achieve 8 trials correct on the training task and all participants scored 5 out of 6 trials correct for the easiest 0.5 ratio on the actual ANS task. In contrast to the TD group; $F(1, 25) = .051$, $p = .824$, $\eta^2_p = .002$ and the DS group: $F(1, 23) = 1.863$, $p = .186$, $\eta^2_p = .078$, there was a reliable trajectory in the WS group; $F(1, 21) = 9.783$, $p = .005$, $\eta^2_p = .328$ when WIAT scores were plotted against non-symbolic number abilities as measured by the ANS test (see Figure 4). A one-way ANOVA collapsing the groups over age showed that there was no difference between the three groups; $F(2, 71) = .877$, $p = .420$, $\eta^2_p = .025$ for non-symbolic number abilities (see Table 2 for descriptive data).

Figure 4 about here

When plotting WIAT performance against symbolic number test scores, there was a reliable trajectory for the TD group; $F(1, 25) = 23.280$, $p < .001$, $\eta^2_p = .492$, the WS group; $F(1, 23) = 5.137$, $p = .034$, $\eta^2_p = .189$, and the DS group; $F(1, 23) = 12.467$, $p = .002$, $\eta^2_p = .362$ (see Figure 5). A number of participants in the TD group (n= 8), the WS group (n= 10) and the DS group (n= 3) performed at ceiling, exclusion of these participants only changed the findings for the WS group. Thus, the developmental trajectories for the three groups could not be compared. A one-way ANOVA, collapsed over age, showed a significant difference for number familiarity scores between the three groups; $F(1, 73) = 3.523$, $p = .035$, $\eta^2_p = .090$ but the effect size was very small and pairwise comparisons showed no significant differences between the groups (all *p*s > .05) (see Table 2).

## Discussion

The current study examined the development of mathematical abilities and what factors contribute to this development in individuals with WS and DS across a wide age range by building developmental trajectories. In line with previous studies, mathematical performance in both WS and DS group was not in line with their chronological age (Brigstocke et al., 2008; Paterson et al., 2006; Udwin et al., 1996). This shows that individuals with WS and DS, regardless of their age, struggle with their basic number skills (i.e., ANS abilities and symbolic abilities which form the foundation of mathematical abilities). Importantly, previous research with typically developing children has indicated that issues with basic number skills may impact on their level of independence and dealing with money (Duncan et al., 2007; Watts, Duncan, Siegler, & Davis-Kean, 2014).

Still, mental age, as measured by performance on the RCPM task, was a predictor of mathematical abilities for all three groups. There were no differences in the development of mathematical abilities between the three groups when performance was plotted against RCPM scores. In addition, our results showed that mathematical abilities related to RCPM scores, even in adulthood. Previous studies have shown that RCPM scores continue to improve with increasing CA (Van Herwegen, Farran & Annaz, 2011). Therefore, the current finding suggests that mathematical abilities may continue to increase over development, although not in line with CA. This contrasts with previous findings from longitudinal studies that have suggested that mathematical abilities plateau in DS (Turner & Alborz, 2003) and WS (Udwin et al., 1996) or studies in adults that have shown that those with DS achieve higher mathematical abilities than those with WS (Paterson et al., 2006). One of the reasons why the results of the current study differ from previous research is the inclusion of a very large age range comprising cross-sectional data from both children and adults with DS and WS. This is in contrast to the narrow age ranges used in previous studies, thus the current study was able to capture more of the spectrum of development in the groups. The suggestion that mathematical abilities continue to improve across the life span in line with MA but not CA in both WS and DS suggest that intervention and training programmes could potentially continue to make a positive impact on independence and life skills of people with DS and WS, even in older individuals. In addition, knowing that mathematical abilities in DS and WS develop in line with general cognitive abilities can be informative for educational practitioners and parents, in terms of what can be expected from children with WS and DS in the classroom. However, the results from the current study need to be followed up with results from longitudinal studies (e.g., WISDOM, see Van Herwegen, Purser & Thomas, 2019), in order to confirm that mathematical abilities do indeed continue to improve in line with MA.

The current study not only examined whether, and at what rate, mathematical abilities in WS and DS develop but also whether this development was related to domain general and domain specific abilities that have previously been shown to be important for mathematical abilities in typically developing populations. A better understanding of what domain general and domain specific abilities relate to mathematical abilities in these groups can provide further insight into what intervention strategies might be beneficial and whether individuals with WS and DS may benefit from the same training programmes.

In the DS and TD, but not the WS group, visuospatial abilities reliably predicted mathematical abilities and there were no differences for the rate or onset for mathematical development between the TD and DS group when performance was plotted against visuospatial abilities. Previous research in TD children has shown that visuospatial abilities are important for the development of mathematical abilities (Uttal et al., 2013), especially for the development of a number line or the understanding of how numbers relate to each other. The absence of a reliable trajectory in the WS group was not caused by participants with WS being at floor on this task as some participants with WS achieved rather high ability scores on the PC task. This finding supports previous studies that have suggested that visuospatial abilities do not predict mathematical abilities in WS (Ansari et al., 2003) and that development of mathematical abilities in WS is atypical and follows a different developmental trajectory.

Domain specific abilities were measured by a non-symbolic ANS task and a number familiarity score for symbolic abilities. Performance on the symbolic number familiarity predicted mathematical abilities in the DS group and the TD group but not the WS group. Previous studies have often argued that symbolic abilities are a particular weakness in individuals with DS (Nye, Clibbens & Bird, 1995). However, the current study shows that counting and digit recognition abilities in DS are not necessarily lower compared to what is expected for their overall mental age or compared to other populations with similar mathematical difficulties, such as WS. The finding that mathematical abilities relate to symbolic knowledge in the DS but not the WS group suggest that improving counting and number recognition abilities would allow individuals with DS to obtain better mathematical skills.

In contrast to the TD and DS groups, mathematical abilities related to ANS abilities in the WS group, even though ANS abilities did not differ between the three groups. These results support previous studies that have found that ANS abilities are delayed but in line with mental age ability and those that have suggested that weaker ANS abilities may cause mathematical delays in WS (Libertus et al., 2014; O’Hearn & Landau, 2007; Van Herwegen & Karmiloff-Smith, 2015).

The relationship between symbolic and non-symbolic number abilities and further mathematical abilities is still hotly debated within the TD literature (see Schneider et al., 2017). However, it has been argued that whilst non-symbolic number abilities might be predictive early on in mathematical development, symbolic number abilities might become a better predictor once children start formal education (Xenidou-Dervou, Molenaar, Ansari, van der Schoot, & van Lieshout, 2017). The fact that most children in the TD group would have started formal education already, could thus explain why performance on the ANS task was not predictive for mathematical development, in contrast to symbolic abilities. The same explanation might explain why there was no significant trajectory for ANS task and mathematical abilities in the DS group. However, the current study, by using a developmental approach for the first time, shows that mathematical development in WS is atypical, in that there was still a significant trajectory for when mathematical abilities were related to ANS abilities. Children with dyscalculia or mathematical learning difficulties have been shown to have similar ANS deficits. In addition, individuals with dyscalculia also show a significant relationship between ANS and mathematical abilities later on in life (Piazza, 2010; Mazzocco et al., 2011b). It has been argued that their mathematical learning difficulties originate from a core deficit in the ANS, in that the meaning of symbolic numerals is acquired by mapping number words and Arabic digits onto the pre-existing approximate number representations (Dehaene 2001). Therefore, the findings that symbolic number abilities in WS were lower compared to TD participants of a similar mental age, but related to mathematical abilities, in addition to the delayed and significant relationship between ANS and mathematical abilities, support the suggestion that verbal abilities and strong rote memory abilities might compensate for mathematical difficulties in the WS group (Ansari et al., 2003; Krajcs et al., 2011). Therefore, it is possible that individuals with WS are learning symbolic information, without necessarily associating these symbols with meaning.

In sum, the current results show that, although mathematical abilities are delayed, they keep developing in both individuals with DS as well as WS in line with their overall mental age. However, development of mathematical abilities in WS also are atypical, while for individuals with DS they are merely delayed. The atypical development in WS is evidenced in that there was no meaningful relationship between mathematical abilities and visuospatial abilities, in contrast to a meaningful relationship between mathematical and ANS abilities. Both of these findings suggest an atypical development of a mental number line or an understanding of how numbers relate to each other.

These findings have important implications for interventions and educational training programmes in that those with WS might benefit from ANS and symbolic number knowledge training programmes, whilst for those with DS improving their visuospatial abilities, in addition to symbolic abilities, might impact on their mathematical abilities. However, the current study did not examine how the different domain general and domain specific abilities *interact* over time and the current cross-sectional results should be replicated by longitudinal studies. It is still possible that basic-level abilities, such as eye movement planning in WS or the sustained attention difficulties in DS can explain both the domain general and domain specific difficulties observed in WS and DS (Karmiloff et al., 2012; Van Herwegen, 2015). Similar to all research, we did not assess all domain specific and domain general abilities relevant to mathematical development, due to the limited amount of time available. Future studies might also want to include more complex symbolic number abilities, such as ordinality and cardinality knowledge, as a large number of participants in the TD and WS group performed at ceiling levels. Also, the ANS task has been shown to rely on executive function (EF) and inhibition abilities (Clayton & Gilmore, 2015). As the current study did not assess EF or inhibition abilities, future studies may want to examine how these functions impact on ANS performance, especially in the WS group. Finally, previous studies in those with mathematical learning difficulties have shown that different factors may predict mathematical difficulties for different individuals (Costa, Nicholson, Donlan & Van Herwegen, 2018) and thus future studies, including larger and longitudinal samples may allow for further examination of these individual differences for mathematical difficulties in WS and DS.

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Table 1.

*Participant characteristics by group, including mean chronological age (CA) in months and mean raw score from Raven's Coloured Progressive Matrices (RCPM) as well as minimum and maximum score from RCPM.*

| | Age | | RCPM | | | |
|---|---|---|---|---|---|---|
| Group | Mean | *SD* | Mean | *SD* | *Min* | *Max* |
| Typically developing | 76.46 | 19.58 | 17.40 | 6.01 | 5 | 26 |
| Williams syndrome | 232.96 | 144.73 | 15.61 | 4.57 | 7 | 25 |
| Down syndrome | 262.21 | 129.75 | 15.17 | 6.27 | 4 | 25 |

Table 2.

*Participant performance (mean raw score and SD) per group for visuospatial abilities (PC task), non-symbolic number abilities (ANS task) and symbolic number abilities (number familiarity score).*

| | PC task | | | ANS task | | | Number Familiarity | | |
|---|---|---|---|---|---|---|---|---|---|
| Group | N | Mean | *SD* | N | Mean | *SD* | N | Mean | *SD* |
| Typically developing | 22 | 102.68 | 24.28 | 26 | 37.15 | 6.48 | 26 | .77 | .23 |
| Williams syndrome | 21 | 67.09 | 25.74 | 22 | 36.32 | 5.65 | 24 | .86 | .18 |
| Down syndrome | 22 | 85.63 | 24.07 | 24 | 34.75 | 7.15 | 24 | .70 | .21 |

Figure 1.

*Mathematical abilities as measured by WIAT (raw scores) plotted against chronological age (CA: reported in months) for each of the groups (TD: typically developing; WS: Williams syndrome; DS: Down syndrome).*

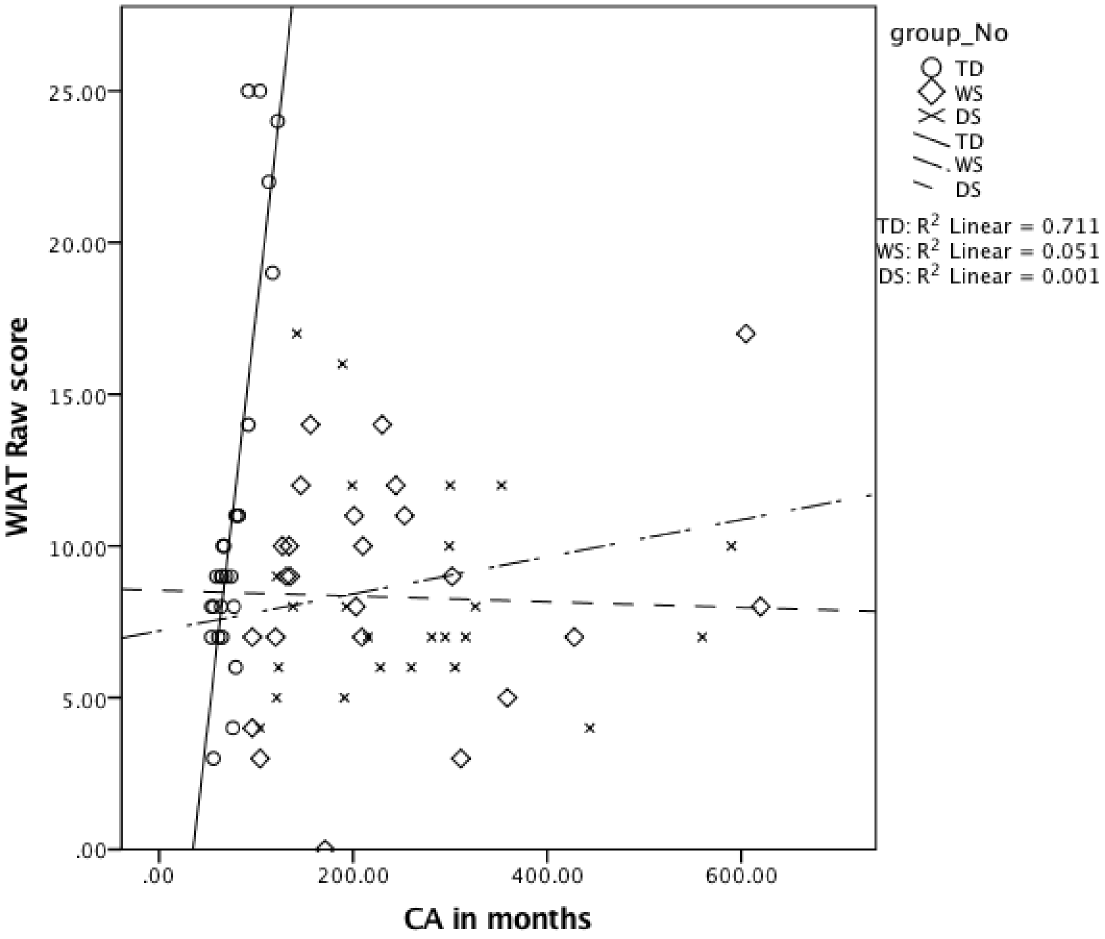

Figure 2.

*Mathematical abilities as measured by WIAT (raw scores) plotted against RCPM raw scores for each of the groups (TD: typically developing; WS: Williams syndrome; DS: Down syndrome).*

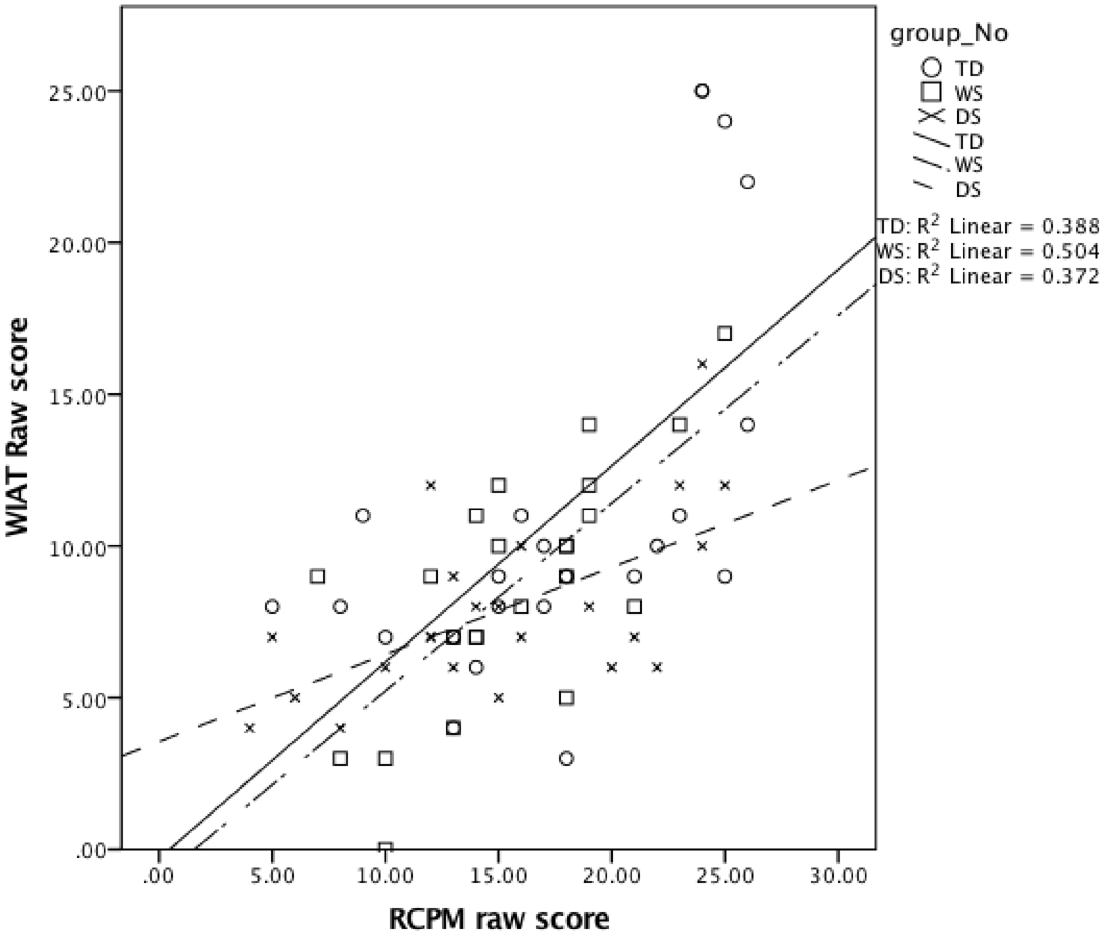

Figure 3.

*Mathematical abilities as measured by WIAT (raw scores) plotted against visuospatial ability scores as measured by the PC task (PC Ability) for each of the groups (TD: typically developing; WS: Williams syndrome; DS: Down syndrome).*

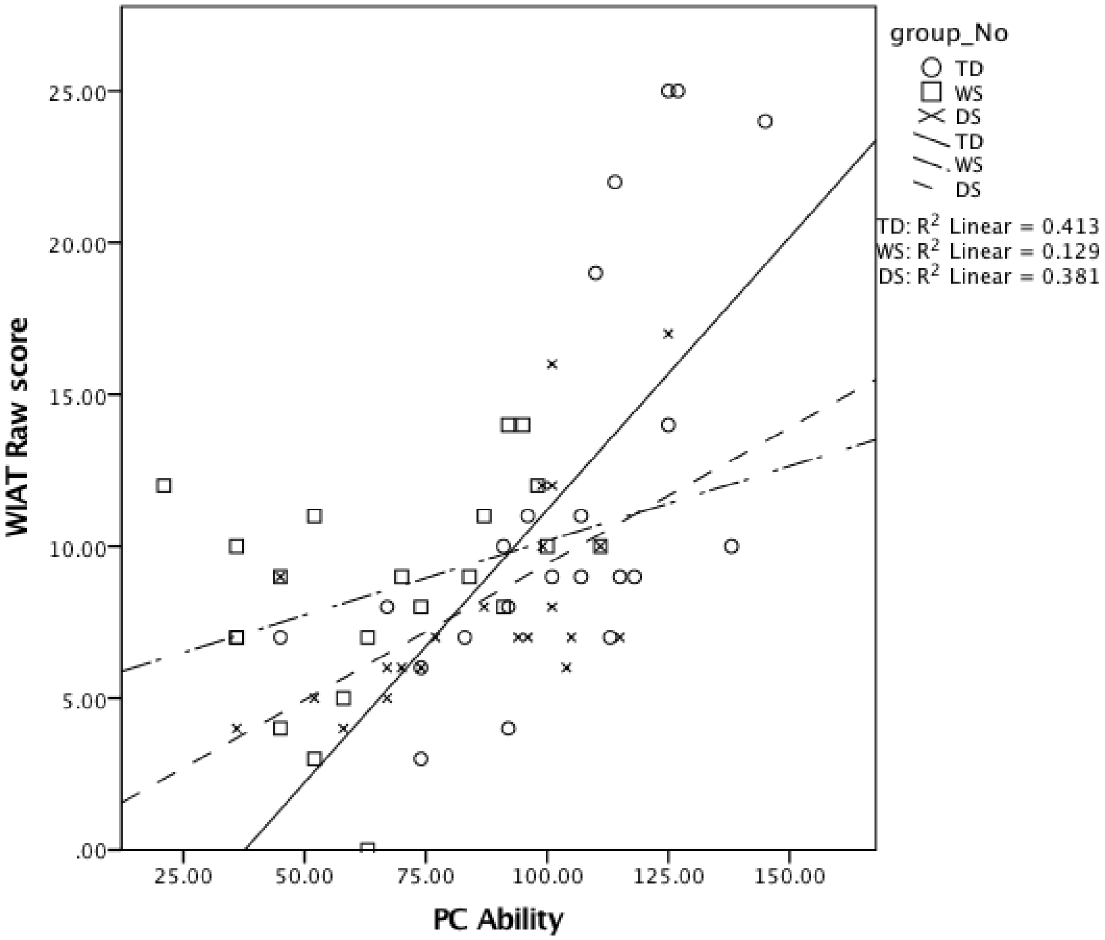

Figure 4.

*Mathematical abilities as measured by WIAT (raw scores) plotted against non-symbolic number abilities as measured by the ANS task performance (ANS Total) for each of the groups (TD: typically developing; WS: Williams syndrome; DS: Down syndrome).*