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PRE-PUBLICATION MANUSCRIPT

Developmental delays in phonological recoding among children and adolescents with Down syndrome and
Williams syndrome

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Running Head: Developmental delays in speech coding

Keywords: Down syndrome, Williams syndrome, phonological recoding, phonological similarity effect, word length effect, visual similarity effect, short-term memory

This study examined the development of phonological recoding in short-term memory (STM) span tasks among two clinical groups with contrasting STM and language profiles: those with Down syndrome (DS) and Williams syndrome (WS). Phonological recoding was assessed by comparing: 1) performance on phonologically similar and dissimilar items (phonological similarity effects, PSE); and 2) items with short and long names (word length effects, WLE). Participant groups included children and adolescents with DS ($n=29$), WS ($n=25$) and typical development ($n=51$), all with average mental ages around 6 years. The group with WS, contrary to predictions based on their relatively strong verbal STM and language abilities, showed no evidence for phonological recoding. Those in the group with DS, with weaker verbal STM and language abilities, showed positive evidence for phonological recoding (PSE), but to a lesser degree than the typical group (who showed PSE and WLE). These findings provide new information about the memory systems of these groups of children and adolescents, and suggest that STM processes involving phonological recoding do not fit with the usual expectations of the abilities of children and adolescents with WS and DS.

What this paper adds

Children and adolescents with Down syndrome struggle with phonological recoding

Children and adolescents with Williams syndrome struggle to a greater degree with phonological recoding

Development of phonological recoding is related to both language and cognitive abilities

Vygotsky (1987) believed that a fundamental aspect of the development of thinking involves a progression from private speech to inner speech, or in other words from non-communicative speech which often ‘helps’ problem-solving when children talk to themselves about possible actions to verbally based, internal and not spoken aloud thought processes. This progression has a crucial role in the development of cognition. More specifically, Vygotsky suggested both that inner speech provides a new code for higher level thinking and that much of adult thinking occur with inner speech. More recently, Winsler and Naglieri (2003) provided evidence for this progression by showing that children's verbal strategies progressed from overt, to partially covert, to fully covert forms with age. However, Alderson-Day and Fernyhough (2015; p 931) in their recent review have argued that the study of inner speech is ‘diffuse and largely unintegrated’ indicating a need to investigate fundamental processes, one of these is the ability to form a phonological code when processing information about visual material.

The purpose of the current study was to examine the development of an indicator of inner speech, namely phonological recoding in short-term memory (STM), among two clinical groups with contrasting STM and language profiles: Down syndrome (DS) and Williams syndrome (WS). *Phonological recoding* refer in this study to the process of recoding *non-verbal* stimuli into a phonological form, for example remembering the names of a series of pictured objects rather than remembering their visual images. This is not to be confused with the old name for decoding in reading, even if it also was called phonological recoding and involved conversion of printed visual stimuli into phonological forms.

The mechanisms underlying phonological recoding can be understood with reference to the working memory model, which was used to provide the theoretical underpinning for the current study (Baddeley, 1986; 2000; Baddeley & Hitch, 1974). One of the components of this model is the ‘phonological loop’, a temporary (1-2 seconds duration) passive storage system for speech-based information, usually regarded as the mechanism underlying verbal short-term memory. The phonological loop also contains the facility to rehearse the contents of the phonological store and keep them activated, via the ‘articulatory rehearsal mechanism’. This can be seen as a recycling mechanism that constantly enters and re-enters the phonological information into the phonological store to prevent decay. A further function of the articulatory

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rehearsal mechanism is to carry out phonological recoding, the means by which *non-verbal* information can be recoded into a verbal form and subsequently stored in the phonological loop. This form of inner speech can enhance the short-term recall of visually presented materials for which verbal labels are available. For auditorily presented speech items, there is no need for phonological recoding, because auditory items have direct access to the phonological store as phonological codes are created by the vocal input (e.g., Penney, 1989). On the other hand, visually presented items such as nameable pictures can only enter the phonological store indirectly, after a phonological code has been created.

Phonological recoding provides a strong indicator of children's potential to use inner speech, which is especially relevant to the attainment of higher level thinking in the two clinical groups considered here. Not only is phonological recoding linked to the use of inner speech, but weaknesses in phonological recoding have also been linked to delayed development of reading abilities (Palmer, 2000b), a key ability for educational progress. Furthermore, the two clinical groups' uneven cognitive profiles allow interpretation of the relative importance of cognition and language for inner speech development. We will first review the language and cognitive profiles for individuals with DS and WS, before considering how to assess phonological coding in these groups. Finally, we review previous studies on phonological recoding in individuals with DS and WS.

DS is characterised at the genetic level by a triplicate copy of chromosome 21 (trisomy 21) as well as phenotypical characteristics such as mild to severe intellectual disabilities (Chapman & Hesketh, 2000; Pennington, Moon, Edgin, Stedron, & Nadel, 2003), precocious aging and high risk of dementia (Numminen, Service, Ahonene, & Ruoppila, 2001). It is by far the most commonly observed genetic disorder, occurring in approximately 1 in every 691 live births (Parker et al., 2010). Individuals with DS display broad weaknesses in language, including expressive vocabulary and grammar (Næss, Lyster, Hulme, & Melby-Lervåg, 2011), and phonological awareness (Næss, Melby-Lervåg, Hulme, & Lyster, 2012; Roch & Jarrold, 2008), although receptive vocabulary appears to be in line with non-verbal mental age (Laws & Bishop, 2004; Næss et al., 2011). In terms of short-term memory (STM), individuals with DS usually show relative weaknesses in verbal STM (see a range of studies including: Carney, Henry, Messer, Brown, Danielsson, & Rönnberg, 2013; Jarrold & Baddeley, 1997; Laws & Bishop, 2003; Smith & Jarrold, 2014b;

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Vicari, Marotta & Carlesimo, 2004; Wang & Bellugi, 1994), combined with relative strengths in

visuospatial STM (e.g., Carretti & Lanfranchi, 2010; Yang, Conners, & Merrill, 2014). Importantly, these relative strengths in visuospatial STM occur in comparison to verbal STM, not in comparisons to mental age, as some visuospatial abilities are mental age appropriate and others are slightly below mental age expectations (Yang et al., 2014).

WS is a rare genetic disorder which occurs in approximately 1 in every 7,500 individuals (Strømme, Bjørnstad, & Ramstad, 2002). The psychological phenotype of WS is primarily characterised by mild to moderate intellectual disabilities with a mean IQ of around 55 (e.g., Martens, Wilson, & Reutens, 2008), relatively strong verbal abilities (Howlin, Davies, & Udwin, 1998; Udwin, Yule, & Martin, 1987) and weaker non-verbal abilities such as spatial cognition and visuospatial construction (Martens et al., 2008). Often, those with WS have an outgoing, loquacious and distinctly ‘hypersociable’ personality type (Jones et al., 2000; Porter, Coltheart, & Langdon, 2007), which has been referred to as “cocktail party syndrome” (Bellugi, Birchle, Neville, Jernigan, & Doherty, 1992; Udwin & Yule, 1990). Individuals with WS are usually characterised as having relatively strong language abilities, but language is still delayed in relation to chronological age (Martens et al., 2008). There is agreement that WS involves an uneven profile of language abilities from neuroconstructivists (e.g., Karmiloff-Smith, Brown, Grice, & Paterson, 2003) and those who argue that language is a modular system (e.g., Bellugi et al., 1988, 1994), although there is disagreement about the precise nature of the profile. Several studies have shown a range of limitations in syntax, semantics and pragmatics (e.g., Grant, Valian, & Karmiloff-Smith, 2002; Laing, Hulme, Grant, & Karmiloff-Smith, 2001) but some studies have not found differences (e.g. Zukowski, 2009; Musolino, Chunyo, & Landau, 2010). Those with WS demonstrate similar levels of phonological ability compared to typically developing children matched on mental age (Vicari, Carlesimo, Brizzolara, & Pezzini, 1996) and compared to children matched on verbal and reading ability (Laing, Hulme, Grant, & Karmiloff-Smith, 2001). Individuals with WS may depend more on phonology than semantic information (Grant et al., 1997; Vicari et al., 1996), possibly due to a semantic–phonological imbalance (Thomas & Karmiloff-Smith, 2003). To establish the language profiles of the three groups (DS, WS, TD) in this investigation, information was collected about phonological abilities and receptive grammar.

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In terms of STM, individuals with WS tend to show a relative strength in verbal STM and a relative weakness in visual-spatial STM and other visuospatial skills (e.g. Carney et al., 2013; Klein & Mervis, 1999; Menghini, Addona, Costanzo, & Vicari, 2010; O'Hearn, Courtney, Street, & Landau, 2009; Vicari, Bellucci, & Carlesimo, 2003; Wang & Bellugi, 1993; but see Jarrold, Phillips, & Baddeley, 2007), a pattern that matches their uneven cognitive profile. Some researchers have argued that the impression of superior linguistic skills in those with WS is an artefact of hypersociability combined with superior verbal memory (Gosch, Stading, & Pankau, 1994; Thomas et al., 2010; Udwin & Yule, 1991).

To summarise, individuals with DS show relative weaknesses in language and verbal STM abilities and relative strengths in visuospatial STM, whereas individuals with WS show relative strengths in language and verbal STM abilities and relative weaknesses in visuospatial STM. Even though not all studies are in complete agreement, there is a consensus that the language and cognitive profiles in these groups are uneven. After this review of the language and cognitive profiles for individuals with DS and WS, a description of the methodology of assessing phonological recoding will be follow before a review of previous studies on phonological recoding

In order to assess phonological recoding, a picture memory span task was employed in the current study (e.g. Henry, Messer, Luger-Klein, & Crane, 2012). This involved children remembering lists of nameable pictures. Four different types of pictures were used: those with one-syllable phonologically similar names, those with longer names (three to four syllables), those with visually similar images (e.g., all long and thin shapes, presented at 45 degrees), and a control group of one-syllable words that did not share common characteristics with one another. The measures of phonological recoding involved the phonological similarity effect (PSE; calculated as the difference between performance for control pictures minus performance for phonologically similar pictures) and the word length effect (WLE; calculated as the difference between performance for control pictures minus performance for longer named pictures). These assessments were based on the assumption that when phonological recoding occurs, this produces two types of effects within the phonological store: (1) errors increase for phonologically similar items because of sound confusion within the phonological store; and (2) recall is reduced for items with longer names because long words occupy more space in the time-limited phonological store. There is also the possibility of visual

Developmental delays in phonological recoding coding strategies being used (visual similarity effect, VSE; this was calculated as the difference between performance for control pictures minus performance for visually similar pictures). However, the findings about visual coding have been inconsistent across studies (e.g., Henry et al., 2012), so the VSE was included in our assessments, but was not the focus of the research.

The PSE has been attributed both to confusion between similar sounding items in the phonological store (Baddeley, 1986; 2007) and confusion at the recall or reconstruction/redintegration stage (Cowan, Sauls, Winterowd, & Sherk, 1991; Hasselhorn & Grube, 2003). Both of these confusion effects, however, would imply phonological recoding. The word length effect (WLE) has been attributed to the fact that longer words might impair verbal rehearsal and speech output processes (e.g., Baddeley, Thomson, & Buchanan, 1975; Cowan et al., 1991; Henry, 1991) due to longer articulatory durations and the time-limited nature of the phonological store. These articulatory rehearsal and speech output processes can only be used if the participant first employs phonological recoding. Related to this Jarrold and Hall (2012) have challenged the assumption that verbal STM involves rehearsal. Consequently, it is important to note that the current study is predicated on the assumption that the presence of both PSEs and WLEs with visually presented/recalled materials implies that phonological recoding has taken place because these effects are not possible for visual representations and can only arise if phonological recoding has been utilized (Baddeley, Lewis, & Vallar, 1984). After this description of the methodological aspects, a review of the previous studies on phonological recoding is provided.

In typically developing children, phonological recoding in visual short-term memory (STM) tasks develops gradually with age as a voluntary strategy employed to improve recall, with the PSE first emerging at approximately 6 years (e.g. Conrad, 1971; Henry et al., 2012; Palmer, 2000a). Palmer (2000a) suggested a four-stage developmental progression in strategic approach to picture span tasks among typically developing children, starting from ‘no strategies used’ at around 3-4 years, progressing to the use of visual methods of remembering at 5 years, then to dual visual and phonological recoding after 5-6 years, and finally to predominantly phonological recoding between 6-8 years. The precise transition point to phonological recoding remains uncertain and probably emerge gradually over a reasonably lengthy time period (Palmer, 2000a). Somewhat later developmental trajectories have been put forward for the WLE with visually

Developmental delays in phonological recoding presented materials (e.g., Hitch, Halliday, Dodd, & Littler, 1989). Research on phonological recoding in individuals with neurodevelopmental disorders has found similar transition ages: 7 years for those with autism spectrum disorders (Williams & Jarrold, 2010) and mental ages of 6 years for those with intellectual disabilities (Henry, 2008).

Most previous studies examining PSE and WLE in individuals with DS and WS have used a verbal STM task procedure with verbal presentation of items and verbal serial recall. Since the items are already presented in a phonological form, conclusions about the voluntary use of phonological recoding is not possible because such coding is not necessary for ‘automatic’ entry into the phonological store (Baddeley, 1986). The demand for speech output prior to serial verbal recall (e.g. Cowan et al., 1991; Henry, 1991) also requires the participant to produce verbal codes to recall the items. Therefore, such codes should not be regarded as ‘spontaneously’ produced as they are demanded by the task procedure. Hence, studies with verbal presentation and verbal recall methods are not relevant for phonological recoding. This is the case for studies that have found effects for individuals with DS (PSE: e.g., Hulme & Mackenzie, 1992; Kittler, Krinsky-McHale, & Devenny, 2004; Smith & Jarrold, 2014a, and WLE Jarrold, Cowan, Hewes, & Riby, 2004) and where no effect has been found (PSE: Varnhagen, Das, & Varnhagen, 1987), and studies that have found effects for individuals with WS (PSE: Vicari et al., 1996, and WLE: Jarrold, Cowan, Hewes, Riby, 2004). The only study, to our knowledge, with nonverbal presentation and response methods for individuals with DS found no PSE (Lanfranchi, Toffanin, Zilli, Panzeri, & Vianello, 2014).

To summarise, phonological recoding begins to develop at about 6 years in typically developing children, but there is mixed evidence for phonological recoding (PSE and WLE) in individuals with both DS and WS. There are also methodological problems with interpreting evidence for phonological recoding where verbal presentation and recall methods have been used. Therefore, to track the development of spontaneous phonological recoding, the current study used non-verbal presentation and response methods, ensuring that phonological recoding could only be adopted as a voluntary strategy (Henry et al., 2012). Picture memory span was assessed using nameable pictures, and with picture pointing as recall method to remove all verbal demands from the experimental procedure. Groups of participants with DS and WS were compared to children with typical development (TD), and all groups had comparable mental ages of about 6

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Our hypotheses were as follows. Evidence for phonological recoding (PSE and WLE) should be found for typically developing children, based on the previous research (Henry et al., 2012). For children with DS no PSEs are predicted, which is in line with the only study we could find that used a suitable non-verbal procedure to examine PSEs (Lanfranchi, Toffanin, Zilli, Panzeri, & Vianello, 2014). Because of the lack of relevant WLE studies for this group we assumed similar patterns for WLE and PSE, i.e., no PSE or WLE for individuals with DS. In the absence of relevant studies for children with WS, we predicted both PSEs and WLEs based on this group's relative language and verbal STM strengths.

To confirm the expected pattern of STM and language performance (i.e. relatively stronger verbal scores for those with WS and relatively stronger visuospatial scores for those with DS) in the participant groups, a selection of STM, phonological awareness and language tasks were included.

2. Material and methods

2.1. Participants

The sample comprised 105 participants, across three groups, 25 children and adolescents with WS (15 girls), 29 children and adolescents with DS (16 girls), and 51 typically developing (TD) children (24 girls). Chronological and mental ages for the groups can be found in Table 1. Bonferroni corrected *t*-tests showed no significant differences between the groups with DS and WS on chronological age and no significant differences between any of the three groups on mental age (all *ps* > .5). Given the groups' different cognitive profiles, it was not possible to match all groups on both verbal and non-verbal abilities separately. However, the matching was relatively close since there were no group differences between any of the groups on nonverbal abilities (all *ps* > .2), and for verbal abilities there was only a significant difference between the group with DS and the group with WS. To ensure this did not affect the results, additional analyses with verbal and nonverbal abilities as covariates were used.

Participants with WS were recruited through the UK branch of the Williams Syndrome Foundation; participants with DS were recruited through the Down Syndrome Association. TD children were recruited

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Ethical approval was given by the Research Ethics Committee at the appropriate University. Written consent was obtained from organisations involved in recruitment and parents, and also from participants, directly prior to testing. All were told, before giving written consent, that they were taking part in a project about how different children think, that their answers would be stored anonymously, that the tasks had nothing to do with anything they did at school, and that they could opt out at any time. Table 1 gives further characteristics of the sample. A liberal definition of adolescence is used in this article, following Arone (2014) who argues that adolescence continues longer than 18 years of age. In this study, 3 individuals with DS and 5 individuals with WS were older than 18 years. For readability reasons we call the participants ‘children and adolescents’. Twenty-one participants were excluded due to the following criteria: mental age higher than 100 months which means that they were older than the expected range for phonological recoding development (one with WS and eight TD), IQ score outside one SD range (85-115) for the TD group (10 individuals), and problems in understanding the instructions (1 with DS and 1 with WS).

Table 1 about here

2.2. Procedure and Tasks

Testing of the individuals with WS and DS took place at their homes. TD participants were either tested at home or at school. Home-tested participants undertook one or two sessions, on the same day. TD participants tested at school undertook three or four sessions to fit into school timetables. All participants

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were tested in a quiet area, in order to maximise concentration levels. Effective testing time was about 60-75 minutes for most participants, and varied depending on performance on the span tests. Breaks were taken when needed and both the number of them and the length of them varied depending on the individual's needs.

All participants started with the Stanford-Binet Abbreviated Battery (ABIQ) test, a short version of the Stanford-Binet IQ test battery (Fifth Edition; Roid, 2003) which contains separate non-verbal and verbal IQ sub-tests. The Stanford-Binet Technical Manual (Roid, 2003) reports strong mean reliability coefficients for the ABIQ (internal, TD 5-8 year-olds: $r = .91$; test-retest, TD 2-20 year-olds: $r = .85$). Next, participants completed the phonological recoding-related memory test (further details below).

An overview of all concepts, which tests that were used to assess them, which variables and which measures for all tests analysed in the results section can be found in Table 2. Phonological and visuospatial short-term memory (STM) and phonological awareness tests were then administered. These were counterbalanced within groups: half the participants in each group undertook the phonological STM test first (Word List Recall, WLR, from the Working Memory Test Battery for Children, WMTB-C, Pickering & Gathercole, 2001) then the visuospatial STM test (Block Recall, BR, also from the WMTB-C); the other half the opposite order. The WMTB-C manual reports strong mean internal ($r = .60$) and test-retest ($r = .80$) reliability coefficients among typical children for WLR; similarly for BR (mean internal, $r = .55$, and test-retest, $r = .63$ reliability coefficients). Counterbalancing also applied to two phonological awareness tasks (from the Phonological Abilities Test, PAT, Muter, Hulme, & Snowling, 1997), these were Rhyme Production (RP) and Rhyme Detection (RD). The PAT manual reports that scores taken from TD 4-7 year-olds give strong internal (RD: $r = .87$; RP: $r = 0.83$) and test-retest (RD: $r = .80$; RP: $r = .65$) reliability coefficients for both subtests. All participants ended the test battery with the Test for Reception of Grammar 2 (TROG-2, Bishop, 2003). The TROG-2 manual reports norms, derived from TD individuals aged 4-16, which show strong internal ($r = .88$) and parallel-form ($r = .67$) reliability.

Participants' use of phonological recoding was assessed using a specially-devised touch screen computer memory measure, programmed and developed using the E-Prime software package. This involved four conditions, each consisting of the presentation and recall of a different set of black and white line

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drawings. Nine pictures, sized at 200*150 pixels, were used in each condition. All of the pictures are listed here for each condition: 1) control – single-syllable words which did not rhyme (cake, chair, shoe, bus, leaf, frog, ring, clown, drum), 2) visually similar – similar-looking (long and narrow) single-syllable items, depicted at the same orientation (bed, fish, nail, key, sock, tie, pen, knife, brush), 3) long names – pictures with three- and four-syllable names (bicycle, teddy-bear, umbrella, television, elephant, butterfly, ladybird, tele-phone, banana), and 4) phonologically similar – single-syllable words which shared the same vowel sound (the most important feature for phonological similarity, Nimmo & Roodenrys, 2004) and were near or exact rhymes (can, lamp, hat, van, pan, ant, cat, bat, fan). For examples of pictures, see Appendix 1. Item sets were matched as closely as possible in terms of mean age of acquisition of object names, imageability, frequency, familiarity, and name agreement (Morrison, Chappell, & Ellis, 1997). Please see Henry et al. (2012) for full details. Note that ratings were not available for two items in the phonologically similar set (fan, can), and one item in the long-name list (teddy-bear), but were nevertheless included due to constraints in selecting appropriate materials. The order of the four conditions was randomised across participants.

Prior to each condition, the experimenter explained that the participant was going to be shown a new set of pictures. The Experimenter displayed each item on the screen and named it – repeating the process for each of the nine pictures in the relevant set. This was to encourage children to use the expected names for each picture, although note that participants were not asked to name pictures themselves to avoid priming phonological recoding. Each memory trial commenced with the target picture/s being presented for a duration of 1500ms. Pauses between picture presentations, during which time the screen was blank, were 500ms. Participants were then simultaneously shown all nine pictures from that set, arranged in a 3*3 array, and required to touch the pictures they had seen, in the order in which they had been presented. This was a span test, with span level (i.e. the number of pictures presented prior to recall) ranging from 1 to 9, with progress dependent on achieving four out of six trials correct at any given level. Since there were a limited number of pictures, participants saw the same picture many times throughout the test period. How many times depended on achieved span level. To ensure that the instructions were understood by the participants, practice trials at span lengths 1 and 2 were conducted. Participants were required to correctly answer two trials at each of these levels before undertaking the test proper. If wrong answer was given, the instructions

Developmental delays in phonological recoding were repeated again. After the practice trials, no feedback was given. The span level was scored as the highest level that was passed plus .25 for each correctly recalled list on the ‘failed’ span level. For example, passing span level 2 and getting 3 trials on span level 3 correct would give a score of 2.75. This scoring ensured that all participants were allocated a sensitive score that represented their maximum performance on each item set.

2.3. Statistical Procedures and Design

Given the lack of significant differences in mental age, we first examined differences between the groups (TD, DS, WS) on the STM, phonological awareness and language measures to ensure that the groups varied on these dimensions as would be expected based on the literature. One-way analyses of variance (between groups factor ‘Group’: TD, DS, WS) were followed up with Bonferroni corrected *t*-tests to assess the inter-group comparisons.

In order to investigate phonological recoding, we assessed the memory span data. A 4x3 analysis of variance was conducted with performance on the span tasks as a dependent measure. The within group variable ‘Picture Type’ had four conditions; Control, Long, Phonologically similar, and Visually similar. The between group variable ‘Group’ had three conditions; TD, DS and WS. One-way Anovas for each group were used to investigate planned comparisons between the Control picture type condition and the other Picture Type conditions. The alpha level was set at .05 for all statistical tests used throughout the study, which mean that the familywise Bonferroni corrected alpha level was .017 when three comparisons were made. The statistical assumptions for Anova were met and no corrections were required.

4. Results

Means and standard deviations for all study variables can be found in Table 3; Table 4 summarises key statistical analyses and group comparisons.

Insert Tables 3 and 4 about here

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As can be seen in Table 4, the group with DS showed lower performance compared to the TD group on the phonological awareness and language measures (expressive and receptive measures of rhyme production and detection respectively, and receptive grammar) and on the verbal STM measure, but did not differ on visuospatial STM. The group with WS had the opposite pattern, with lower performance compared to the TD group only on the visuospatial STM measure. These profiles were as expected, and are consistent with the established literature demonstrating weak verbal STM, phonological awareness and language skills in those with DS, together with weak visuospatial STM in those with WS.

A Picture Type by group analysis of variance showed a main effect of Picture Type, $F(3, 306) = 7.17, p < .001, r = .27$, and a Picture Type by Group interaction, $F(6, 306) = 2.26, p = .04, r = .21$, but no significant main effect of Group ($p = .78, r = .07$). The interaction was analysed further according to our predictions with planned contrast comparisons. These analyses, for the TD group, showed higher performance in the control condition compared to all the other conditions (long, $F(1, 50) = 14.11, p < .001, r = .47$, phonologically similar, $F(1, 50) = 21.65, p < .001, r = .55$, visually similar, $F(1, 50) = 5.67, p < .01, r = .38$. For the group with DS, performance in the control condition was significantly higher than in the phonologically similar condition, $F(1, 50) = 10.49, p < .01, r = .52$ (for the nonsignificant comparisons long and visually similar the p -values and effect sizes were $p = .18, r = .26$ and $p = .64, r = .05$ respectively). For the group with WS, no significant differences were found between picture types (long: $p = .93, r = .02$; phonologically similar: $p = .55, r = .12$; and visually similar: $p = .12, r = .31$). This means that the TD group showed a PSE, a WLE and a VSE; the group with DS showed a PSE; and the group with WS showed none of these effects. A planned comparison on group differences for the control condition confirmed that there was no significant group effect ($p = .21$). This rules out the possibility that the pattern of results was due to performance differences for control pictures.

The analyses above were repeated with the Stanford-Binet verbal score and Stanford-Binet non-verbal score as covariates. The interaction and all planned comparisons followed the same pattern of significance as above. The only deviation from the pattern was the main effect of Picture Type, which became non-significant ($p = .10$).

4. Discussion

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This study provided evidence for phonological recoding in children and adolescents with DS, but not in children and adolescents with WS. These findings were contrary to the expected pattern based on the profiles of strengths and weaknesses in terms of verbal STM and language in our groups: those with DS showed significantly weaker verbal STM, phonological processing and language skills (rhyme production, rhyme detection and receptive grammatical ability) compared to mental age level (i.e. the TD comparison group); whereas those with WS showed verbal STM, phonological and language skills in line with their mental age level. As expected, based on previous literature, children in the TD comparison group showed evidence for phonological recoding (PSE and WLE). Consequently, the findings suggest that for the children and adolescents with DS, their lower STM and language abilities were not a barrier to phonological recoding; whereas the language-related and STM abilities of the WS group were not sufficient to enable phonological recoding to occur in most of the children and adolescents.

The evidence for phonological recoding in the group with DS, but not in the group with WS was contrary to our hypotheses. This raises interesting questions about why this should occur, which have implications for our understanding of the relationship between phonological abilities and phonological recoding. Given that the children and adolescents in the group with DS, as in other research, had significantly lower language and phonological abilities than the mental age matched TD group, it appears that phonological recoding can occur in children and adolescents with a range of language and phonological abilities. This suggests that a high level of phonological abilities like rhyme detection and production, are not necessarily prerequisites for development of phonological recoding. Further research involving children with lower levels of phonological abilities is needed to investigate whether there is minimum level of phonological ability to support phonological recoding.

The findings from the group with WS, who had language and phonological abilities which did not differ significantly from the TD group, also support the argument that the development of phonological recoding may be independent of other phonological abilities. In the group with WS there were no significant differences in performance across the four picture span tasks to indicate any form of strategic memory coding, yet the TD group, who had similar language and phonological abilities, demonstrated

Developmental delays in phonological recoding evidence of PSE and WLE. This all suggests that phonological recoding may *not* occur in some individuals with developmental disorders, despite the presence of relatively advanced language/phonological abilities.

In children with typical development, the precise transition point to phonological recoding remains uncertain and may emerge gradually over a reasonably lengthy time period (Palmer, 2000a). Somewhat later developmental trajectories have been put forward for the WLE with visually presented materials (e.g., Hitch, Halliday, Dodd, & Littler, 1989). Our findings suggest that the development of phonological recoding in children and adolescents with DS, and particularly WS, starts at a higher mental age, takes longer time to develop or is atypical in nature (Karmiloff-Smith et al., 1997). Consequently, the PSE and WLE may be present in individuals with learning impairments who have higher mental ages than 6 years.

Not only do our findings have implications for understanding the relationship between phonological abilities and phonological recoding, they also have relevance for debates about developmental delay and deviance in the context of individuals with intellectual disabilities (Dodd, 2011; Karmiloff-Smith et al., 1997; Zigler & Balla, 1982). The development of language in children with DS often appears to involve morphosyntax developing more slowly than other abilities, particularly the acquisition of receptive vocabulary (Buckley 1993; Fowler, 1990; Chapman & Hesketh, 2000; Rutter & Buckley, 1993) and it also has been argued that their language development is deviant (Perovic, 2006; Ring & Clahsen, 2005; Sanoudaki & Varlokostam 2014). Our findings about phonological recoding in children with DS provide support for the idea of a delay in relation to typical vocabulary development: the group with DS had a delay in phonological recoding, and what appeared to be the presence of limited recoding strategies (i.e. the absence of WLE). Although this profile is consistent with previous research about slower development of some language abilities such as receptive abilities that are associated with vocabulary, it also is the case that that an uneven profile where phonological recoding is found to be in advance of expected language abilities suggests deviance from typical development

The issue of delay and deviance has a long history of debate in relation to individuals with WS, particularly for morphosyntactic abilities (e.g. Karmiloff-Smith et al., 1997), and our findings from the

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group with WS are consistent with the idea of a deviant pattern of development. The group with WS had similar STM and phonological/language abilities to the TD group, yet there was no evidence of phonological recoding; nor was there evidence for the use of any other memory coding strategies. Therefore, in line with Karmiloff-Smith et al., individuals with WS showed a different developmental pathway, and weak connections between language development and related cognitive processes. Thus, the development of phonological recoding appears to be deviant in both clinical groups, but the nature of the deviance is different in each group.

It is important to consider several methodological and theoretical issues in relation to the findings. Jarrold and his colleagues have suggested that low memory span performance can make it difficult to detect performance decrements for phonologically similar pictures in children and adolescents with WS (Jarrold & Citroën, 2013; Jarrold, Danielsson, & Wang, 2015). The key argument against this, as an explanation of the current findings, is the absence of a group effect on the control items for memory span performance. This suggested that all three groups had similar levels of memory span, yet the groups with DS and TD nevertheless showed the PSE and WLE performance decrements. Inspection of the mean memory span scores across groups for each Picture Type reveals that scores ranged from 1.68 to 2.28 pictures. Previous research has found significant performance decrement effects at this level of span performance (e.g. Henry et al., 2012), suggesting that our findings are robust. This may be because we were careful to include six memory span trials at each list length and to adopt a sensitive scoring method that gave credit for partial successes on higher list lengths.

A further consideration raised by Jarrold and Citroën (2013) concerns whether scaling effects mean that individuals with lower span scores show relatively smaller performance decrements on phonologically similar or other items. This remains a possibility, and needs to be taken into account before any claims are put forward that *increases* in PSE size might be related to increasing levels of phonological recoding and/or verbal rehearsal. Thus, the current data only warrant the conclusion that in samples of children and adolescents with average mental ages of 5:11 (DS), 6:2 (WS) and 6:2 (TD), the PSE was found in groups with DS and TD only.

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Another potential explanation of the finding of a PSE in the group with DS was that this group had a slightly lower average mental age (five years 11 months) and a smaller range in mental age (37 months vs 41 months for typical and 49 months for WS). In fact, the somewhat lower mental age (and range) would act against finding the PSE in this sample. It is also important to consider whether matching the groups on an overall measure of mental age was the best approach. Comparing individuals with DS and WS is always difficult because differing verbal and non-verbal cognitive profiles mean that matching is problematic. In order to take this into account, analyses of covariance which included raw scores for verbal and non-verbal subtests in our IQ measure were reported. The findings were the same, giving reassurance that these results were not an artefact of the matching criteria.

For children with WS, the absence of PSE and WLE is relevant to the debate about the nature of their uneven language profile (e.g., Bellugi et al., 1988, 1994; Karmiloff-Smith, Brown, Grice, & Paterson, 2003). Phonological recoding could be an addition to abilities that are weak compared to other abilities, like grammar and phonological skills. Future studies should investigate if the weaker phonological recoding abilities could be related to, or are an effect of, other weaker language abilities not measured in this study, using a wider language test battery. Furthermore, investigations are needed to determine whether the absence of phonological recoding is associated with limitations in other forms of inner speech (e.g. Winsler & Naglieri, 2003).

Finally, the findings about a lack of PSEs and WLEs (and, hence, absence of strategic phonological recoding) suggest that interventions designed to improve the memory strategies of individuals with WS could benefit their recall of information. For children and adolescents with DS, who showed only PSE but not WLE, it is still an indication that memory coding strategies could be improved by a similar intervention. The use of spontaneous strategies and change of strategies are areas where individuals with intellectual disabilities in general have problems (Belmont & Butterfield, 1971; Bray, Fletcher, & Turner, 1997), and this strengthens the case for appropriate interventions.

5. Conclusions

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To conclude, it was confirmed that the group with WS showed relative strengths in language and verbal STM, whereas the group with DS showed relative impairments. Contrary to predictions, the presence and absence of phonological recoding did not follow the language profiles in the three groups. Children and adolescents with DS showed a PSE effect, suggestive of phonological recoding, whereas those with WS showed no evidence for phonological recoding or any other strategy use. By contrast, children with typical development of a similar mental age, showed the expected PSE, WLE and VSE, indicating the use of phonological recoding and visual memory strategies. The findings suggest that the development of phonological recoding is not always directly related to language and cognitive abilities, but further research is needed to better understand the nature of these relationships. The need for interventions targeting strategy use in memory coding is discussed.

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Developmental delays in phonological recoding

Table 1. Chronological and mental age (in months) together with verbal and nonverbal ability raw scores as measured by Stanford-Binet for the three groups; Children with typical development, children and adolescents with Down syndrome and children and adolescents with Williams syndrome.

Test	Typical			Down			Williams		
	<i>M</i>	<i>SD</i>	Range	<i>M</i>	<i>SD</i>	Range	<i>M</i>	<i>SD</i>	Range
Chronological age	71.1	17.1	48-115	174.8	35.7	129-262	178.5	39.2	98-245
Mental age	74.3	12.7	55-96	70.5	9.1	55-92	73.5	13.9	49-98
Verbal ability	23.3	2.97	18-30	22.1	2.86	16-29	24.8	3.84	17-34
Nonverbal ability	15.5	4.43	8-25	14.9	2.80	10-22	13.7	4.23	8-23

Developmental delays in phonological recoding

Table 2. Overview of all the concepts, tests, variables, and measures analysed in the results section.

Concept	Test	Variable	Measure
Short-term memory	Working Memory Test Battery for Children	Block recall	Total trials correct
		Word list recall	Total trials correct
Phonological awareness	Phonological Abilities Test	Rhyme Production	Raw score
		Rhyme Detection	Raw score
Grammar	Test for Reception of Grammar 2	Grammar	Raw score
Phonological recoding	Picture span test	Control span	Span score
		Phonological span	Span score
		Visual span	Span score
		Long span	Span score

Developmental delays in phonological recoding

Table 3. Means and standard deviations on all tests for the typical, DS and WS groups.

Test	Typical		Down		Williams	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Short-term memory						
Block recall	20.0	5.1	20.8	4.7	15.5	4.2
Word list recall	15.4	3.3	13.1	2.9	15.2	3.5
Phonological awareness/grammar						
Rhyme detection	7.65	2.51	5.69	3.19	6.88	3.43
Rhyme production	6.47	3.92	3.31	4.27	4.84	2.97
TROG	8.61	4.25	3.97	2.06	7.20	4.91
Picture span						
Control span	2.28	1.07	1.99	.63	1.97	.63
Phonological span	1.75	.75	1.68	.50	1.91	.60
Visual span	1.95	1.06	1.95	.51	1.82	.68
Long span	1.90	.82	1.83	.50	1.96	.61

Developmental delays in phonological recoding

Table 4. Anovas and testwise Bonferroni corrected *t*-tests for comparison between children with typical development (TD), children and adolescents with Down syndrome (DS) and William's syndrome (WS) on different measures of short term memory and language. A positive value on the *t*-test means that the first group in the heading had higher score than the second group.

Test	<i>ANOVA</i>	<i>TD vs DS</i>	<i>TD vs WS</i>	<i>DS vs WS</i>
Short-term memory				
Block recall	$F(2, 104) = 5.00, p < .001, r = .09$	$t(102) = -0.72, ns$	$t(102) = 3.89, p < .001$	$t(102) = 4.09, p < .001$
Word list recall	$F(2, 104) = 9.96, p < .01, r = .16$	$t(102) = 3.06, p < .01$	$t(102) = 0.29, ns$	$t(102) = -2.34, ns$
Language				
Rhyme detection	$F(2, 104) = 4.10, p < .05, r = .07$	$t(102) = 2.86, p < .05$	$t(102) = 1.07, ns$	$t(102) = -1.48, ns$
Rhyme production	$F(2, 104) = 6.48, p < .01, r = .11$	$t(102) = 3.56, p < .01$	$t(102) = 1.75, ns$	$t(102) = -1.47, ns$
TROG	$F(2, 104) = 12.75, p < .001, r = .20$	$t(102) = 5.04, p < .001$	$t(102) = 1.46, ns$	$t(102) = -2.99, p < .01$

Developmental delays in phonological recoding

Appendix 1

Examples of visually similar pictures: Fish and Nail.

Examples of phonologically similar pictures: Can and Fan.

Developmental delays in phonological recoding

Examples of pictures with longer names: Elephant and Butterfly.