Home literacy environment and literacy outcomes in individuals with Williams syndrome and Down syndrome

J. Lettington & J. Van Herwegen

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Home literacy environment and literacy outcomes in individuals with Williams syndrome and Down syndrome

J. Lettington and J. Van Herwegen

Department of Psychology and Human Development, IOE UCL’s Faculty of Education and Society, London, UK

ABSTRACT

Background: The home literacy environment (HLE) has rarely been examined for individuals with neurodevelopmental disorders, including individuals with Williams syndrome and Down syndrome.

Method: The current study surveyed carers of individuals with Down syndrome (n = 48) and Williams syndrome (n = 18) in the United Kingdom (UK).

Results: The study reports that individuals with Down syndrome were rated higher in general reading skills and writing, while the Williams syndrome group scored higher for speaking. Yet, individuals with Down syndrome were more likely to engage in informal activities than instructional activities and the frequency of informal activities related to reading outcomes for those with Down syndrome but not Williams syndrome. Additionally, this study reports that age was not related to the HLE for both groups.

Conclusion: This is the first study to report on the HLE of individuals with Williams syndrome and supports the key role of the HLE in the development of literacy skills for individuals with Williams syndrome and Down syndrome.

Reading development has been argued to be influenced by the activities and learning that takes place in the home learning environment. The Home Literacy Environment (HLE), which includes both formal and informal literacy centred activities, are reported to be positively correlated with reading outcomes (Sénéchal & LeFevre, 2002; 2014; Sénéchal et al., 1995, 1996, 1998). Although a number of studies have examined the HLE in typically developing (TD) children, very little is known about the HLE of children with neurodevelopmental disorders, such as Down syndrome and Williams syndrome. Seeing the delay in reading abilities and reading difficulties that are often experienced by children with neurodevelopmental disorders, the focus of this study will be to gain a better understanding of the HLE for these groups to further inform interventions and educational programs.

Williams Syndrome is a rare genetic neurodevelopmental disorder caused by a microdeletion of approximately 28 genes on the long arm of chromosome 7 (Schubert, 2009). It has a prevalence rate of around 1 in 20,000 live births (Strømme et al., 2002). Individuals with Williams syndrome have a distinct behavioural phenotype that includes hypersociability, non-social anxiety, gregariousness and attention-deficit like traits (Leyfer et al., 2006).

In terms of their cognitive profile, individuals with Williams syndrome have a number of strengths and difficulties, including mild to moderate learning difficulties with average intelligence quotient (IQ) scores ranging between 42 and 68 (Martens et al., 2008). Expressive and receptive language skills are often stronger than visuospatial skills (Pezzino et al., 2018; Vicari et al., 2006). Despite low language abilities overall, expressive language skills are often better than receptive language and individuals with Williams syndrome use a wider range of figurative expressions than they can understand (Naylor & Van Herwegen, 2012; Van Herwegen et al., 2013). In addition, individuals with Williams syndrome show difficulties in a range of executive cognitive functions, such as attention, memory and problem solving (Vicari et al., 2007; Jarrold et al., 2007, Porter et al., 2007).

Down syndrome is a more common genetic neurodevelopmental disorder, which is caused predominantly by a trisomy of chromosome 21 (Silverman, 2007). Down syndrome has a prevalence rate of around 1 in 800 live births (Silverman, 2007).
Similarly to Williams syndrome, Down syndrome is characterised by a range of cognitive difficulties that include mild to moderate learning difficulties (Mervis et al., 2000) with IQ scores also ranging between 40 and 70 (Hodapp et al., 1999). The rate of cognitive development may be slow (Dunst, 1990) and development from 0 to 11 years of age is thought to be characterised by a variable cognitive profile of strengths and difficulties (Wishart, 1993). Jarrold et al. (2007) reported that individuals with Down syndrome have specific difficulties with working memory.

Receptive language and comprehension are reported to be stronger than expressive language, and expressive language is often reported to be specifically delayed in individuals with Down syndrome compared to TD children (Chapman, 1995; Chapman, 1999). Individuals with Down syndrome continue to develop their language skills in adolescence and early adulthood with increasing disparity between an individual’s ability to comprehend sentences (receptive language) and their vocabulary and sentence production (syntax; Vicari et al., 2000). Receptive vocabulary has been reported to be a strength but may be most strongly related to life experiences (Chapman, 1999; Facon et al., 1998).

The variable cognitive profile of strengths and difficulties, which is present in both disorders, has a differential effect on literacy development. The following section will review research into literacy development and literacy abilities in individuals with Williams syndrome and Down syndrome in an attempt to give an overview of the profile of reading abilities within these two disorders.

**Literacy development in individuals with Williams syndrome and Down syndrome**

Research about literacy in Williams syndrome is limited. Laing et al. (2001) examined the processes involved in learning to read. In two experiments, they looked at both “end-state of reading” and the process of learning to read in a sample of 15 children and adults with Williams syndrome (aged 9–27 years old) and reading age matched controls. Laing et al. reported the average age of the Williams syndrome group at the time of testing as 15 years 1 month while their test age on the single word reading subtest of the British Ability Scale II (Elliot et al., 1996) was recorded as 6 years 3 months. Phonological awareness (PA; which refers to the ability to recognise and manipulate spoken parts of a word including phonemes, onsets and syllables) was impaired in the Williams syndrome group and was shown to be related to reading attainment, although this effect was weaker in the Williams syndrome group compared to the control group.

The relationship between PA and word reading for individuals with Williams syndrome has been replicated in other studies (see Levy et al., 2003; Menghini et al., 2004). Steele et al. (2013) examined the relationship between reading ability and a range of domains such as nonverbal ability, receptive vocabulary and PA. For children with Williams syndrome, PA and letter knowledge were not powerful predictors of reading growth, possibly as it may not develop in line with their typically developing peers. Letter knowledge is taught explicitly in schools and is a focus of the teaching of early reading (Rose, 2006). However, as PA is taught more implicitly than letter knowledge, it may result in children with neurodevelopmental disorders, who may require more direct teaching of PA, being left behind.

Brawn et al. (2018) investigated the relationship between several domain-general and domain-specific skills and reading outcomes in thirty participants with Williams syndrome aged 9–39 years old. They reported the average age equivalent score for their participants as 8 years 1 month while the average chronological age was 21 years and 0 months. Reading ability was found to be associated with adaptive functioning, particularly within the communication domain. All subscales of reading were positively correlated with three cognitive domains (auditory processing, short term memory and PA) as well as community skills (measures of competency in everyday tasks involving concepts such as time, money and computer skills).

Relative to the TD population, there is limited research on the topic of reading in Down syndrome. Perhaps due to the historically low expectations of their potential reading abilities, they were often not taught to read (Laing et al., 2002). These days most children with Down syndrome in mainstream schools learn to identify simple words, yet reading comprehension abilities are often delayed (Laws et al., 2016).

In terms of reading predictors, Cossu et al. (1993) reported that PA is not always a good predictor for reading in that individuals with Down syndrome who had reading abilities in line with their TD peers performed significantly worse on a PA task. Snowling et al. (2002) reported three studies that compared the reading and PA abilities of children with Down syndrome (n = 29, aged 6–17 years) and TD children (n = 29, aged 4–6 years) of a similar reading level. It was reported that children with Down syndrome did not differ in word and non-word reading but did perform worse on syllable segmentation, rhyme and phoneme detection tasks. Phonological knowledge was found to be related to reading in both groups. For the Down syndrome group, letter-sound knowledge did not predict reading, while it did for the TD group. They suggest that individuals with
Down syndrome may not have full phonemic awareness (the ability to recognise and manipulate the smallest unit of sound, i.e., the phoneme, in spoken words) and may rely less on phonological skills for reading. This means that reading development may be atypical and develop in line with other cognitive factors compared to TD groups whose reading was found to be related to PA.

A review by Lemons and Fuchs (2010) included evidence from 20 studies and suggested that children with Down syndrome do rely on PA skills when learning to read and that some children may benefit from phonics-based reading instruction. However, a meta-analysis, including eight studies in which individuals with Down syndrome matched TD controls for word recognition level, indeed confirmed that differences in vocabulary, but not PA, were predictive of differences in nonword decoding skills (Næss et al., 2012).

In sum, although PA is not the strongest predictor for reading abilities in individuals with Down syndrome, children with Down syndrome who had better PA had a higher reading ability (Snowling et al., 2002). As PA is not the strongest predictor of reading ability in individuals with Down syndrome but language abilities seem to relate to reading outcomes, it is possible that reading development follows a qualitatively different developmental trajectory since evidence in TD groups provides indication that PA is strongly related to reading outcomes. It is important to understand these qualitative differences in order to give an insight into best practice to support reading acquisition.

Overall, studies have shown that reading abilities in Down syndrome and Williams syndrome are delayed but that there is wide variability within the reading abilities in both groups. While most research has focused on the factors associated with literacy development, little is known about good practice to support development of literacy abilities in these groups. The research would suggest that PA is related to better outcomes in reading in both groups but also that language abilities are important predictors for reading in these groups. Yet, it is unclear what strategies parents use at home to support the development of these skills and what impact this has. Additionally, little thought has been given to the effect of age on the support that parents give at home. This question is interesting, since many individuals with Williams syndrome and Down syndrome will not acquire a reading age in line with their chronological age. As such, an understanding of the home literacy environments (HLE) of individuals with Williams syndrome and Down syndrome will provide insight into what parents currently provide in terms of formal and informal activities and where future HLE programmes might be helpful.

The home literacy environment of typically developing children

The HLE includes both formal and informal literacy centred activities and resources that exist in the home, including activities related to both reading and writing. These activities include those in which children interact with adults in writing and reading situations and experiences in which children explore print on their own. The HLE has two dimensions: formal and informal literacy activities/resources. Formal literacy activities include those that are either print-centred, where the focus of the literacy activity relates explicitly to the process of reading (e.g., time spent using instructional materials, which gives a broad indication of how much time parents and carers dedicate to explicit teaching around reading processes, or guided reading with an adult), while informal literacy activities are focussed rather on implicit reading activities, where the message contained within the text (e.g., independent reading or number of books in the home) is often central (Sénéchal et al., 1998).

A study by Sénéchal et al. (1998) examined the HLE and its impact on literacy outcomes in 168 TD children (age range: 47–86 months). Parents completed a questionnaire that gathered information on their child’s literacy experiences, including storybook exposure (a measure of exposure to children’s books) and parental teaching about literacy, and a battery of assessments was administered to gather information on children’s written- and oral-language abilities. Although there was no relationship between storybook exposure and parent teaching, storybook exposure was positively associated with children’s oral-language skills (such as receptive vocabulary, listening comprehension and phonemic awareness) and direct parent teaching was positively associated with children’s written-language outcomes (concepts about print, alphabet knowledge and reading consonant–vowel-consonant words).

In a follow-up study, Sénéchal and LeFevre (2002) provided further evidence for the importance of distinct formal and informal home literacy activities. This 5-year longitudinal study, which provided an account of the HLE model, followed 110 kindergarten children until grade 3. The model postulates that storybook exposure and parent teaching are not related to each other. Storybook exposure was found to predict language (measured by vocabulary and listening comprehension) in Grade 1, which in turn predicted child book exposure and ultimately better reading at the end of Grade 3 (7–8 years). Parent teaching was found to predict emergent literacy skills in Grade 1 (i.e., awareness of others reading, interest in books, independently looking at books...
etc.), which predicted reading at the end of Grade 1 (5–6 years). In turn, this predicted reading at the end of Grade 3 as well as reading comprehension. The HLE model is displayed in Figure 1.

Authors additionally highlight that oral-language, emergent literacy skills and PA in Grade 1 are interrelated in the sense that the development of the separate factors is dependent upon the others. Sénéchal and LeFevre (2002) pointed out that storybook exposure was not a significant predictor of emergent literacy skills, known to be a strong predictor of reading outcomes (Whitehurst & Lonigan, 1998), and that these informal activities alone may not be sufficient to develop early literacy.

Hood et al. (2008), in an Australian study with 143 children (mean age = 5.36 years), showed that the distinct types of home literacy activities were differentially related to literacy and language development. Informal parent-child reading (a measure of storybook exposure) was related to vocabulary in Grade 1, while formal parent teaching activities related to letter-word identification in Preschool (a measure of reading). Hood et al. (2008) also reported that letter-word identification mediated the relationship between parent-teaching and reading measures in Preschool, Grade 1 and Grade 2.

Large scale meta-analyses have also provided support for the importance of the HLE. One such example is that of the National Early Literacy Panel (2008), which reported that informal shared reading experiences are positively associated with children’s vocabulary acquisition and knowledge about print but did not have an effect on phonemic awareness or alphabet knowledge. In a separate meta-analysis conducted by Sénéchal and Young (2008), it was reported that parent listening, and parent tutoring were related to early literacy outcomes. They concluded that informal activities are related to children’s oral language development and phonological awareness, which will eventually be important for reading comprehension (as evidenced by the aforementioned longitudinal study). Formal activities are related more directly to the acquisition of the skills necessary for understanding the mechanics of reading. However, both are important to improve phonological awareness and reading outcomes, although children are likely to need further direct teaching to develop phonemic awareness.

In sum, there is a large amount of evidence that supports the significant role the HLE plays in language and literacy development. Evidence suggests that informal experiences at home support language development, while formal experiences support early literacy and therefore later literacy outcomes. In the TD population, writing activities are additionally considered as part of the HLE and reading and writing skills are considered to impact one another’s development. While the body of HLE research provides a sound basis for drawing inferences on relationships between HLE and literacy development in typical populations, less is known about the HLE of atypically developing individuals.

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**Figure 1.** Importance of formal and informal home learning activities on reading outcomes (model based on Sénéchal & LeFevre, 2002).
The HLE of individuals with ND

Some studies have reported on the HLE of children with a neurodevelopmental disorder. For example, it has been reported that parents of children with neurodevelopmental disorders and parents of TD children do not significantly differ in the number of books in their homes, frequency of shared reading and hours viewing TV (Butz et al., 2009). The number of storybooks in the home in the Down syndrome population are also comparable to TD children (Al Otaiba et al., 2009; van Bysterveldt et al., 2013). However, some differences in the HLE of children with Down syndrome have been reported, including shorter reading times associated with difficulties with attention as well as the use of shorter texts (Schneider & Hecht, 1995), even though aspirations for literacy attainment from parents of children with Down syndrome remained high (Ricci & Osipova, 2012). In addition, parents of children with Down syndrome read to their children more often than parents of autistic children and parent teaching was positively correlated with the child’s letter name knowledge for the Down syndrome group (Westerveld & van Bysterveldt, 2017).

To date there are no studies on the HLE in individuals with Williams syndrome and thus, it is unknown how HLE differs between groups of children with neurodevelopmental disorders that have comparable learning difficulties but very different cognitive profiles. As such, it is unclear whether findings reported for individuals with Down syndrome are unique and relate to their level of neurodevelopmental impairment or whether features of the HLE are shared across different groups.

An important question that remains unanswered is the question about the impact of age on the HLE of individuals with ND. The approach to studying the HLE of typically developing individuals is arguably not directly transferable to the study of individuals with ND. One of the main measure of informal HLE in the HLE model (Sénéchal & LeFevre, 2002) is the number of children’s books in the home. This poses a problem for individuals with Williams syndrome and Down syndrome who do not typically develop a reading age in line with their chronological age and as such, their reading material may not correlate so readily with their reading age as it does in TD populations. This may be reflected in a more diverse range of reading material in line with both their chronological ages (and therefore experiences) but which also reflect their reading abilities. This study explored some of these unanswered questions and sought to provide a direction for future research.

The current study

The current study examined the HLE in two groups of individuals with ND that have similar overall learning needs with very distinct cognitive profiles: individuals with Down syndrome and Williams syndrome. Cross-syndrome comparisons are useful for tracing early domain-relevant deficits and how these may have a cascading effect on development (Karmiloff-Smith, 1998). Comparing the relationships between HLE and literacy ability within and between these groups allows for a better understanding of how cognitive profiles play a role in the HLE and further insight into the similarities and differences in the HLE between these two groups.

Consistent with the HLE model, the following hypotheses were made:

1. It was expected that respondents would highlight different strengths and weaknesses related to their child’s literacy abilities, in line with their child’s neurodevelopmental disorder with those with Williams syndrome having specific difficulties with tasks related to their poor visuospatial skills such as writing and those with Down syndrome having more problems with oral language. Similarly to previous research, it was predicted that these strength and difficulties would not relate to the child’s chronological age. Furthermore, in line with previous research (Steele et al., 2013), it was predicted that there would be no differences for parental report between individuals with Down syndrome and Williams syndrome for overall reading abilities.

2. It was anticipated that there would be no differences between the two groups of individuals with ND in types of material used and frequency of use seeing that both disorders present with literacy difficulties (Laing et al., 2001; Snowling et al., 2002).

3. It was anticipated that there would be no difference in the amount of time using formal and informal materials as both disorders present with literacy difficulties and previous studies in Down syndrome have shown high parental aspirations (Ricci & Osipova, 2012).

4. Based on results in TD children, measures of informal and formal HLE activities will be positively associated with parent reported reading abilities.

A further exploratory question driving this study was the relationship between age and the HLE. Although previous studies have shown that cognitive abilities often develop in line with mental age, little consideration has been given to the effect of age on the HLE and it is unclear whether there are any changes over developmental time. This was a key exploratory question in this study since there exists no evidence in the literature to date.
Methods

Participants

Respondents included parents and carers of individuals with Williams syndrome and Down syndrome. A total of 78 responses for individuals with Down syndrome and Williams syndrome were obtained between 2014 and 2015. Respondents who either failed to identify the age of their child or who identified the age of their child as below the age of 3 years 6 months (the average age that children in the UK start formal education; n = 12; 15%) were excluded. Of the data from 66 respondents included in the analysis, 18 (27%) responded on the behalf of an individual with Williams syndrome and 48 (73%) responded on the behalf of an individual with Down syndrome. Of the 18 individuals with Williams syndrome (age range = 3.58–36.33 years old), 11 (61%) were female. In the Down syndrome group (age range = 3.66–33.75 years old), 24 (50%) were female.

For the Williams syndrome group, 13 (72%) respondents were the mothers of the individual, and two (11%) were fathers, while three (17%) either identified as a “parent,” grandparent or failed to respond. In the Down syndrome group, 45 (94%) responders were mothers, one (2%) was a father and the remainder of the group (n = 2; 4%) did not identify their relationship to the individual with Down syndrome.

Materials and procedure

Individuals were invited to take part in an online survey that gathered data around various aspects of the HLE using Qualtrics, an online survey platform. The survey was open for just over 12 months. Participants were recruited by volunteer sampling via Twitter, through the Williams syndrome Foundation UK, Down Syndrome Association, Facebook support groups for parents and carers of individuals with Williams syndrome and Down syndrome and through existing contacts of the Child Development and Learning Difficulties Lab.

Respondents were provided with detailed information about the project and provided their written consent to take part. The survey was anonymous, and respondents were given the contact details of the researcher should they have any further questions. Respondents were able to opt out of the study at any time and to skip questions. The study had received approval by the ethics committee at University College London’s (UCL’s) Institute of Education before recruitment and data collection took place.

The questions in the survey were designed based on the HLE model (Sénéchal, 2006; Sénéchal & LeFevre, 2002). Respondents were asked to provide detailed information about the individual with neurodevelopmental disorder (ND) as well as providing contextual information regarding the child’s home life and the parental educational background. Respondents were asked to rate their child’s competency in a number of areas related to literacy and to provide details of the HLE, including number and types of material as well as frequency of use, using 4-point Likert-type scales. The survey was administered between 2014 and 2015 and was available for 12 months. The survey is available to download through the open science framework.

Data analysis

Independent samples t-tests were performed on the data to check for differences in age between groups. Given the nature of the data collected, non-parametric Mann–Whitney tests were used to examine differences between data about background characteristics of respondents and individuals with ND. Independent samples t-tests were also used to test for group differences of parent reports of how challenging individuals find various tasks (task challenge).

Respondents were able to report on measures of the characteristics of the HLE on Likert-type scales. These were then analysed using independent samples Mann–Whitney tests. Since respondents were able to indicate the number of materials in the home in ratio data (i.e., the amount of various types of materials), these data were analysed using a two-way repeated measures analysis of variance (ANOVA), which tested for differences in amounts of materials at home. Reading ability was rated by parents on a 4-point Likert-type scale (with respondents indicating degree of difficulty on a scale) and analysed using independent samples Mann–Whitney tests. Correlations between scales were measured using Spearman’s Rho correlational analysis. Age was defined by school phase, yielding four groups. Data on the characteristics of the HLE (as indicated by a 4-point Likert-type scale) were therefore compared across age groups using non-parametric Kruskal–Wallis tests. As a secondary analysis of the relationship between age and the HLE and to test whether there was a ceiling effect of age, individual data were grouped by ability (defined by parent report) and mean age was compared across groups using a one-way ANOVA.

Results

Background of the parents and individuals with ND

A description of the characteristics of the carers and individuals can be found in Tables 1 and 2. The number of respondents per question differed throughout as response to each question was not mandatory.
Table 1. Socio-economic status (highest level of formal education) of the parent respondents \((n = 66)\).

<table>
<thead>
<tr>
<th>Socio-economic status</th>
<th>Williams syndrome n (%)</th>
<th>Down syndrome n (%)</th>
<th>Total actual responses</th>
</tr>
</thead>
<tbody>
<tr>
<td>Secondary school / GCSEs</td>
<td>0 (0)</td>
<td>4 (8)</td>
<td>4 6</td>
</tr>
<tr>
<td>A-Level</td>
<td>1 (6)</td>
<td>3 (6)</td>
<td>4 6</td>
</tr>
<tr>
<td>College / vocational training</td>
<td>4 (22)</td>
<td>13 (27)</td>
<td>17 26</td>
</tr>
<tr>
<td>Bachelor’s degree</td>
<td>9 (50)</td>
<td>19 (40)</td>
<td>28 43</td>
</tr>
<tr>
<td>Master’s degree</td>
<td>3 (16)</td>
<td>6 (13)</td>
<td>9 14</td>
</tr>
<tr>
<td>Doctoral degree</td>
<td>0 (0)</td>
<td>3 (6)</td>
<td>3 4</td>
</tr>
<tr>
<td>Missing data</td>
<td>1 (6)</td>
<td>0 (0)</td>
<td>1 1</td>
</tr>
</tbody>
</table>

Note: missing data refers to "no response."

Independent samples Mann–Whitney tests revealed there was no significant difference in SES between groups as measured by highest level of formal education of the respondent, \(U = 365.0, p = .498\).

There was no significant difference between the age of the individuals in the Williams syndrome group \((\text{Myears} = 12.4, \text{SD} = 9.6)\) and the Down syndrome group \((\text{Myears} = 10.1, \text{SD} = 6.1)\); \(t(64) = 1.17, p = .248\). There was additionally no significant difference in current phase of education between groups, \(U = 277.0, p = .955\). About half of the data from both the Williams syndrome group (61%) and the Down syndrome group (50%) came from primary school aged individuals. There was no significant between group difference in the type of schooling that individuals received, \(U = 335.0, p = .211\). There was no significant difference between groups about whether the individual with Williams syndrome or Down syndrome had speech and language therapy (SLT), \(U = 384.0, p = .093\).

**Hypothesis 1: reports of ability and degree of challenge**

Respondents to the survey were asked to judge how challenging individuals with Williams syndrome and Down syndrome tend to find the following areas of learning on a Likert scale from 1 to 7, with one representing not challenging and seven representing very challenging. Areas of learning included sight word reading, phonics, reading comprehension, writing, speaking and listening. Independent samples \(t\)-tests were performed to ascertain if there were any group differences in the aforementioned ratings. The results of these tests can be found in Table 3.

As can be seen in the Table 3, significant differences were found in the writing domain with carers of individuals with Williams syndrome judging writing as more challenging on average than the carers of individuals with Down syndrome. Additionally, carers of individuals with Down syndrome rated speaking more challenging on average than carers of individuals with Williams syndrome. There were no differences for sight word reading or phonics between the two groups. In addition, as can be seen in Table 4, these reported difficulties did not relate to the participants’ ages, with the exception of age correlating positively with listening in the Down syndrome group.

**Measures of Reading ability**

Respondents judged the current level of reading ability for the individuals with Williams syndrome and Down syndrome by selecting from a descriptive 4-point Likert-type scale of increasing complexity (see Figure 2).

Independent samples Mann–Whitney tests revealed a significant difference in respondents’ ranking of individual’s reading level, \(U = 161.5, p = .015\), with the Down syndrome group \((M = 2.79, SD = .86)\) receiving a higher reading scores than the Williams syndrome group \((M = 2.08, SD = .954)\). As shown in Figure 2, most participants with Williams syndrome did not have reading levels beyond a few words.

As can be seen in Table 4, there was no relationship between areas of difficulty and the reported reading difficulties.
ability level for individuals with Williams syndrome. For those with Down syndrome, there was a positive correlation between reading ability and difficulties with listening. However, seeing the positive relation between listening and age, this might suggest that as individuals with Down syndrome get older, they have better reading abilities but also more listening difficulties (Figure 3).

**Hypothesis 2: characteristics of the HLE**

**Materials in the HLE**
Carers were asked to indicate the amount and type of materials that they used to aid the development of their child’s literacy. Respondents could select from a list of pre-defined materials and had the option of adding more types (see Figure 3). The number reported below represents the actual amount of each type of material in the home of individuals. The types of materials were divided into formal and informal materials. A two-way repeated measures ANOVA was performed in order to examine any between (disorder) and within (formal / informal materials) group differences in amounts of materials. There was a significant within-subject difference in the amount of materials, $F(1, 60) = 92.13, p < .001$, where both groups were reported as having more formal materials (Williams syndrome group: $M = 4.8$, $SD = 1.48$; Down syndrome group: $M = 4.81$, $SD = 1.2$) than informal materials (Williams syndrome group: $M = 3.13$, $SD = 74$; Down syndrome group: $M = 2.9$, $SD = 1.2$). There was no significant main effect of Group; $F(1, 60) = .149, p = .701$, and no interaction of Group and Type of materials, $F(1,60) = .443, p = .508$.

**Hypothesis 3: frequency of formal and informal activities**

**Frequency of formal activities**
Respondents were asked to select the amount of time their child spends reading with another person and daily time spent using formal materials, both are measures of formal home literacy activity, according to HLE. Details of responses can be found in Figure 4.

Mann–Whitney tests revealed that there was a significant difference between groups in terms of how ability level for individuals with Williams syndrome. For those with Down syndrome, there was a positive correlation between reading ability and difficulties with listening. However, seeing the positive relation between listening and age, this might suggest that as individuals with Down syndrome get older, they have better reading abilities but also more listening difficulties (Figure 3).

**Table 4.** Spearman’s correlation of age and reading ability with reported degree of task challenge.

<table>
<thead>
<tr>
<th></th>
<th>Challenge sight word reading</th>
<th>Challenge phonics</th>
<th>Challenge comprehension</th>
<th>Challenge writing</th>
<th>Challenge speaking</th>
<th>Challenge listening</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Age (Williams syndrome)</td>
<td>-.445</td>
<td>-.063</td>
<td>.187</td>
<td>.458</td>
<td>.166</td>
<td>.150</td>
</tr>
<tr>
<td>2. Reading ability (Williams syndrome)</td>
<td>-.211</td>
<td>-.364</td>
<td>-.229</td>
<td>.475</td>
<td>.166</td>
<td>.434</td>
</tr>
<tr>
<td>3. Age (Down syndrome)</td>
<td>-.187</td>
<td>.098</td>
<td>-.053</td>
<td>-.290</td>
<td>.047</td>
<td>.356*</td>
</tr>
<tr>
<td>4. Reading ability (Down syndrome)</td>
<td>-.300</td>
<td>.122</td>
<td>-.262</td>
<td>-.203</td>
<td>.086</td>
<td>.444**</td>
</tr>
</tbody>
</table>

Note – * $p < 0.05$, ** $p < 0.01$.

![Figure 2](image.png)

*Figure 2. The percentage of responses from parents describing the reading ability of their child.*
much time individuals with Williams syndrome and Down syndrome spent reading with another person, $U = 218.0$, $p = .007$, with the Down syndrome group ($M = 2.81$, $SD = .80$) receiving a higher score than the Williams syndrome group ($M = 2.19$, $SD = .66$). There was also a significant difference in how much time was spent using formal materials daily, $U = 183.5$, $p = .024$, with the Down syndrome group ($M = 2.6$, $SD = 1.09$) receiving a lower score than the Williams syndrome group ($M = 3.36$, $SD = .84$).

**Frequency of informal activities**

Respondents were asked to select from a Likert-type scale of increasing complexity, the amount of time the individual with Williams syndrome and Down syndrome spent...
reading on their own daily and the number of adult and children’s books at home. Both measures are consistent with the HLE model of informal activities. Details of the responses can be found in Figure 5.

Mann–Whitney tests revealed that there were significant groups differences across both informal home learning measures. Individuals with Down syndrome were reported to read significantly more on their own \( (M = 2.47, SD = .98) \) than those with Williams syndrome \( (M = 1.71, SD = .85) \), \( U = 222.5, p = .005 \). In addition, carers reported more books in the home for the individuals with Down syndrome \( (M = 4.0, SD = .956) \) than the Williams syndrome group \( (M = 3.31, SD = .95) \), \( U = 234.0, p = .019 \).
Hypothesis 4: associations between HLE and Reading outcomes

Categorical data from the survey was analysed using non-parametric correlational analysis to seek out relationships between factors of the HLE and literacy outcomes in both groups. Results of this analysis can be found in Table 5. Whilst for the Williams syndrome group, there were no significant associations between HLE and reading abilities, time spent reading alone did relate to reading abilities for those with Down syndrome. In both groups, time spent reading alone related to reading with others as well.
Table 5. Spearman’s correlation of factors measuring reading ability, formal activities (FA) and informal activities (IA) for Williams syndrome and for Down syndrome

<table>
<thead>
<tr>
<th>For Williams syndrome</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Reading ability</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2. FA – Time spent reading with another person</td>
<td>.261</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3. FA – Time spent using instructional activities</td>
<td>.494</td>
<td>.093</td>
<td></td>
<td></td>
</tr>
<tr>
<td>4. IA – Time spent reading alone</td>
<td>.327</td>
<td>.526*</td>
<td>.016</td>
<td></td>
</tr>
<tr>
<td>5. IA – Number of books at home</td>
<td>.490</td>
<td>.051</td>
<td>-.017</td>
<td>-.377</td>
</tr>
</tbody>
</table>

For Down syndrome

| 1. Reading ability    |   |   |   |   |
| 2. FA – Time spent reading with another person | -.128 |   |   |   |
| 3. FA – Time spent using instructional activities | -.165 | .164 |   |   |
| 4. IA – Time spent reading alone | .437** | .296* | .124 |   |
| 5. IA – Number of books at home | .288 | -.146 | .110 | .252 |

Note: *p < 0.05, **p < 0.01.

Research question 5: age and the HLE

To ascertain whether there was an effect of age on the above factors, a series of non-parametric Kruskal-Wallis tests were performed on the data with educational cohort (early years/primary/secondary/post-16) as the grouping variable. Results revealed that the distribution of time spent reading with another person was the same across groups, $H(3) = 4.74, p = .192$, the distribution of time spent using formal materials was the same across groups, $H(3) = 2.35, p = .502$, the distribution of time spent reading on their own was the same across the different age groups, $H(3) = 2.88, p = .41$ and the distribution of number of books at home was the same across the different age groups, $H(3) = 4.13, p = .248$.

In order to ascertain whether there was an effect of age on reading ability, participants were grouped by reading ability, yielding four groups ("not reading yet," “reading less than 20 words,” “reading simple books and passages,” and “reading difficult words and longer texts”) and mean age was compared across groups. In the Down syndrome group, there was no significant difference in age between groups, $F(3,42) = .681, p = .569$. Similarly, in the Williams syndrome group, there was no significant difference in age between groups, $F(3,12) = 3.248, p = .074$. However, one-tailed non-parametric Spearman’s rho correlations do show a relationship between reading ability and age for those with Williams syndrome, $r(13) = .612, p = .013$ and for those with Down syndrome: $r(47) = .457, p = .001$.

Discussion

This study focused on the HLE of 66 individuals with Williams syndrome and Down syndrome to gain an understanding of the frequency of activities at home as well as materials in the HLE and how these relate to literacy outcomes. While this sample was relatively small in comparison to other literature on the topic of the HLE, this paper reported on individuals with neuro-developmental disorders, including one very rare one, and reflect typical group sizes in this field (Kozel et al., 2021). This was the first study to investigate the HLE of individuals with Williams syndrome and the first study to directly compare the HLE of individuals with Williams syndrome and Down syndrome to see how the HLE relates to the uneven cognitive profiles of these groups. As such, this study was exploratory and largely descriptive in nature.

Background characteristics of participants and respondents

There were no significant differences between the groups for SES and the type of schooling or current phase of education. A large proportion of the individuals in both groups received SLT (96%), which is consistent with the language difficulties that both groups experience (Pezzino et al., 2018; Martens et al., 2008) as well as previous studies that have examined the support that individuals with Williams syndrome and Down syndrome receive within the school (Van Herwegen et al., 2019).

Strengths and difficulties

Respondents rated how challenging individuals with ND found a series of proficiencies that broadly reflected written- or oral-language skills. There were no group differences in parent reports of sight word reading, phonics, comprehension or listening. Consistent with previous research, respondents rated writing to be a significant challenge for individuals with Williams syndrome. It is well documented that individuals with Williams syndrome have difficulties with visuospatial skills, fine motor skills and co-ordination abilities, which all relate to writing (Mayall et al., 2021), and may account for the difficulties in writing skills reported for those with Williams syndrome. For individuals with Down syndrome, oral language abilities were reported as being a significant weakness. This is an unsurprising result given the vast amount of evidence supporting the delays in oral language development that reported in Down syndrome (Pezzino et al., 2018). Parental reports confirmed that the two groups showed the expected uneven cognitive profiles, sometimes referred to as spikey profiles reflecting diverse areas of strength and difficulty, that are likely to impact on early literacy abilities. Research in TD children and those with literacy difficulties has highlighted the critical importance of
oral language for early literacy development (Lervåg et al., 2018) and it has been shown that oral language difficulties are often a predictor of later reading difficulties (Burgoyne et al., 2019). In addition, whilst reading instruction has a positive effect on writing abilities, writing instruction and abilities also improve reading outcomes (Graham & Hebert, 2011), as both require linguistic knowledge (Schoonen, 2019). Yet, the strengths and difficulties reported in Williams syndrome and Down syndrome did not relate to the reading profile in those with Williams syndrome and there was only a significant relationship between reading abilities and listening difficulties for those with Down syndrome. However, this discrepancy can probably be explained by the increasing listening difficulties with age and increasing reading abilities with age.

In contrast to previous studies that assessed reading abilities directly (Steele et al., 2013), parents of individuals with Down syndrome rated their child’s reading abilities higher compared to parents of individuals with Williams syndrome. As the current study did not directly measure reading or wider cognitive abilities in the two groups, it is unclear whether participants differed in terms of overall cognitive abilities from previous studies or whether carers overestimated their child with Down syndrome’s reading abilities. The latter seems less likely seeing that the overall cognitive profile of the two groups reported by the carers matched the strengths and difficulties in studies that directly assessed these profiles. One possibility is that there has been a shift towards inclusive education with currently more individuals with Down syndrome being schooled in mainstream school compared those with Williams syndrome (Van Herwegen et al., 2018). Although there were no significant differences between the two groups for type of schooling and the majority of individuals in this study were school aged, it is still possible that different school experiences and expectations may have affected the different academic outcomes between the two groups (Buckley et al., 2002). An alternative explanation could be that the HLE of these two groups differ, which was further explored in this study.

The HLE of individuals with Down syndrome and Williams syndrome

Consistent with the HLE model (Sénéchal & LeFevre, 2002), this study was concerned with the type of materials and type of activities used by carers at home to support their child’s literacy development. Research has linked formal activities with written-language outcomes, while informal activities have been linked to oral-language outcomes and in some instances with comprehension at a later stage of literacy development (Sénéchal, 2006). Seeing the oral language difficulties in Down syndrome, it could be argued that informal activities are very important and that for Williams syndrome formal activities are key to support their writing difficulties. However, it is important to note that individuals with Williams syndrome, especially young ones, have oral language difficulties as well. Indeed, the current study did find that those with Down syndrome engaged more in informal HLE activities and those with Williams syndrome engaged more in formal activities, despite there being no differences between groups for the total overall number of materials at home. Indeed, 81% of the Down syndrome group reported spending some time reading on their own, whilst 47% of the Williams syndrome group reported as spending “no time reading on their own” each day.

However, both groups reported significantly more formal instructional materials at home compared to informal materials. This perhaps reflects carers’ eagerness to support early literacy development and alludes to underestimating the long-term effects of informal aspects of the HLE. Despite this, there was an association between informal HLE activities and reading outcomes in the Down syndrome group but not the Williams syndrome group. The HLE model provides strong evidence of the important role that informal aspects of the HLE have on oral language and reading comprehension and there are important implications for the promotion of the aspects of the informal HLE.

Together these findings suggest that the Williams syndrome group has a high degree of parental involvement in both formal and informal reading activities, which was less the case for the Down syndrome group. This reflects the learning needs of the individuals with Williams syndrome who were reported to have lower reading abilities and needed more formal support.

Age and the HLE

One of the driving questions of this study was the effect of age on the HLE. It is well documented in research that individuals with Williams syndrome and Down syndrome follow a different developmental trajectory in comparison to their TD peers (e.g., Laing et al., 2001; Steele et al., 2013). Indeed, the current study also found that, despite reading abilities improving with chronological age, the support that individuals receive at home, as the number of formal activities and informal activities, remained stable across the different age groups. Similarly to Ricci (2011) who only evaluated the HLE for those with
Down syndrome, this suggests that carers recognise the support individuals with Williams syndrome and Down syndrome require at home, regardless their chronological age. As the HLE of typically developing children has rarely focused on children older than 8 years of age, it is not clear, how the findings in Down syndrome and Williams syndrome would relate to TD populations.

Limitations and future research

This data draws on carers’ reports and thus caution should be taken when interpreting the results. However, the strengths and difficulties of the individuals with Williams syndrome and Down syndrome as reported by the carers matched those from previous studies and there were no differences in how difficult those with Williams syndrome and Down syndrome find phonics or sight word reading. The current study does not provide any insight into the quality of the activities. As such, future studies are required that use standardised literacy and cognitive measures as well as include observations of the activities in the HLE in order to understand how both cognitive abilities and the HLE relate to individual differences in HLE activities and reading outcomes. Additionally, this study represented a smaller number of individuals with Williams syndrome compared to Down syndrome. This does reflect the prevalence of these disorders with Williams syndrome only occurring 1 in 18,000 live births compared to 1 in 800 for those with Down syndrome. The current study did not include a group of TD children and does not provide insight into whether carers of individuals with ND spend more time on formal activities than carers of TD individuals. Seeing these limitations, findings from this study are tentative.

Still, this study provides important information about age and the HLE, which has not been reported before, and it was found that age does not appear to affect the degree of support that is in place for individuals with Williams syndrome and Down syndrome. This is not surprising seeing that development is rarely in line with age in these populations. Indeed, the strengths and difficulties reported by parents did not relate to their child’s age. However, the current study did not directly measure the cognitive abilities of participants with Williams syndrome and Down syndrome and thus, future studies should include cognitive abilities of individuals with Williams syndrome and Down syndrome to further explore how cognitive functioning impacts on the HLE and seek to employ more sensitive measures to tease out the minutiae of qualitative differences that exist for these distinct populations.

Conclusion

This study was the first to explore the HLE of individuals with Williams syndrome and the first to compare the HLE of individuals with Williams syndrome and Down syndrome. This study shows that individuals with Down syndrome have higher reported reading abilities and that their reading abilities relate to the amount of reading they do on their own. Indeed in terms of HLE environment, despite both groups having similar resources, individuals with Down syndrome engaged less in formal activities compared to those with Williams syndrome. This may suggest that differences in HLE might impact on reading outcomes. However, further research in this area is required as it unclear if these differences are caused by better reading abilities for those with Down syndrome or whether differences in HLE lead to different reading abilities. However, age did not appear to affect the HLE. Seeing that both groups have reported difficulties with reading and writing, these findings illustrate that educational practitioners should aim to highlight the importance of HLE to parents, especially the informal aspects of the HLE, particularly for those with Williams syndrome. Further research in the HLE of those with neurodevelopmental conditions is required.

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