NOTE

A study of relative clauses in Williams syndrome*

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ABSTRACT

Despite growing empirical evidence to the contrary, claims continue to be made that the grammar of people with Williams syndrome (WS) is intact. We show that even in a simple elicited imitation task examining the syntax of relative clauses, older children and adults with WS (n = 14, mean age = 17;0 years) only reach the level of typical five-year-old controls. When tested systematically in a number of different laboratories, all aspects of WS language show delay and/or deviance throughout development. We conclude that the grammatical abilities of people with WS should be described in terms of relative rather than absolute proficiency, and that the syndrome should no longer be used to bolster claims about the existence of independently functioning, innately specified modules in the human brain.

INTRODUCTION

The myth of normal language abilities in people with Williams syndrome (WS) persists; Pinker (1999), for example, makes reference to the 'excellent language skills' of people with WS, claiming that their morpho-syntax is intact. Whilst we agree that WS is a syndrome in which language is

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surprisingly good, given the relatively poor non-verbal mental skills, it is in our view theoretically misleading and empirically inaccurate to claim that morpho-syntax, semantics, or pragmatics are intact in this clinical population (Karmiloff-Smith, 1998). What is true about the syndrome is that older children, adolescents and adults display in most cases considerably better scores on language tasks than on non-verbal tasks, and better scores on language tasks than MA-/CA-matched controls with, say, Down's syndrome. Note, however, that these are RELATIVE, not absolute advantages. This fact is overlooked when WS findings are reported in secondary sources. Indeed, there are frequent claims in the literature about a total dissociation between language and cognition in WS and a slippage from 'relative' proficiencies to absolute abilities, with the syndrome flagged as the prime example of preserved language sub-modules in the face of mental retardation (e.g. Bellugi, Wang & Jernigan, 1994; Clahsen & Almazan, 1998; Pinker, 1999).

First, it is worth noting that the non-verbal level of those individuals with WS who produce particularly good language scores is in no way so low as to suggest linguistic idiots-savants. Referring to IQ levels in a profile, rather than MA levels, can paint a very misleading picture. Take an adult with a verbal IQ of 82 and a non-verbal IQ of 53. This suggests a serious dissociation between language and cognition. However, if one calculates the same individual's MA, the result can turn out to be a verbal MA of eleven years and a non-verbal MA of seven years. It is then far less surprising that a person with a developmental disorder but an MA of seven years has relatively fluent language (Karmiloff-Smith, 1998), since normal children have fluent language as of about five years of age. A second point worth noting is the fact that each time an empirical study has actually been made with a WS population clinically and genetically diagnosed, and of sufficient number, their language turns out to be either delayed or deviant (Udwin & Yule, 1990; Mervis, Golinkoff & Bertrand, 1994; Capirci, Sabbadini & Volterra, 1996; Stevens & Karmiloff-Smith, 1996; Volterra, Capirci, Pezzini, Sabbadini & Vicari, 1996; Karmiloff-Smith, Grant, Berthoud, Davies, Howlin & Udwin, 1997; Singer Harris, Bellugi, Bates, Jones & Rossen, 1997; Jarrold, Baddeley & Hewes, 1998; Karmiloff-Smith, Tyler, Voice, Sims, Udwin, Howlin & Davies, 1998; Thal, Bates & Bellugi, 1989; Thomas, Grant, Barnham, Gsödl, Laing, Lakusta, Tyler, Grice, Paterson & Karmiloff-Smith, 2001).

The strong claims made about the intactness of morpho-syntax are sometimes supported by weak evidence. For example, the study of WS past tense on which Pinker (1999) based his conclusions was a conference poster (Bromberg, Ullman, Coppola, Marcus, Kelley & Levine, 1994) which, because never published, cannot be evaluated. Other work on WS past tense performance has argued that in WS the computational system for language is selectively spared compared to lexical look-up (Clahsen & Almazan, 1998).

But that claim was based on a very small sample (N=4), with wide individual differences. By contrast, a very different picture emerges from the results of a subsequent study in our laboratory of a much larger population of children, adolescents and adults with WS (Thomas $et\ al., 2001$). Using the same past tense task as Clahsen & Almazan, together with a second past tense elicitation task, we specifically controlled for language level in our WS population and found no unusual pattern of dissociation between regular and irregular past tense marking. Our participants looked like much younger, normal control children. Yet, clear evidence of a dissociation between computational processes and simple lexical look-up is crucial for the theoretical claims being made about Williams syndrome in the scientific and popular literature.

Our past tense study challenged the notion that aspects of morphology are differentially spared in WS. However, it remains possible that another aspect of grammar - syntax - is spared. There are several findings to doubt this, however. From previous work (Karmiloff-Smith et al., 1997), we had ascertained that people with WS scored significantly below their vocabulary age (and well below their chronological age) on a standardized test of grammar: the Test of Reception of Grammar (TROG), (Bishop, 1983). Only two subjects with WS were at ceiling (and note that ceiling is only 11 years on this test), and they turned out to be our most able subjects on non-verbal tasks also, with chronological ages of 20 and 25 years, respectively. However, one could argue that tasks such as the TROG, in which participants are asked to select a picture from an array of four that best fits a sentence, have a substantial metalinguistic component (Tyler, Karmiloff-Smith, Voice, Stevens, Grant, Udwin, Davies & Howlin, 1997; Karmiloff-Smith et al., 1998). Syntactic capacities might therefore be underestimated by such procedures, particularly if testing involves atypically developing participants for whom meta-levels of processing are difficult. Acting out paradigms also present difficulties to people with learning difficulties. Thus, despite our clear previous results, it remains possible that people with WS do have intact syntax, and that the TROG failed to reveal this competence, for reasons of task demands rather than deficient grammar.

It was to evaluate this hypothesis that the present study was designed. First, we analysed the error data on the TROG, and picked out a linguistic structure on which all our WS participants had difficulty: relative clauses. Note that a testee can be 'at ceiling' on the TROG but still have failed one syntactic block, so our two participants at ceiling also displayed difficulties with relative clauses. We then devised a simple imitation experiment whose task demands were significantly reduced compared to the picture pointing task. In this way, we hoped to assess more directly the syntactic competence of people with Williams syndrome and ascertain whether claims of intact syntax are correct.

METHOD

Participants

Fourteen children and adults (9 female, 5 male) with Williams syndrome were recruited through the Williams Syndrome Foundation (UK) as part of a larger study. Their mean chronological age was 17;11 (range 8;1–30;9). Thirty-two typically developing children were also tested. They attended a North London primary school and were drawn from three age groups, which will be referred to as the five-, six- and seven-year-old groups. These were 11 four- to five-year-olds (9 male, 2 female; mean age = 5;0, range = 4;9–5;1;11 five- to six-year-olds (5 male, 6 female; mean age = 6;0, range = 5;11–6;2);10 six- to seven-year-olds (5 male, 5 female; mean age = 7;1, range = 6;10–7;2).

All participants were monolingual English speakers and all came from a similar range of mixed socio-economic backgrounds. Basic data on each group are provided in Table 1.

Table 1. Chronological age (CA), and BPVS scores for WS and typically developing groups

Group	Mean CA (s.d.)	Mean BPVS standard score (s.D.)	Mean BPVS vocabulary mental age (s.d.)
WS (n = 14)	17;11 (7;11)	63.00 (16.55)	8;9(3;1)
5 (n = 11)	5;0(0;1)	96.82 (9.21)	4;8 (0;10)
6 (n = 11)	6;0(0;1)	100.22 (13.13)	6;0(1;2)
7 (n = 10)	7;1 (0;1)	101.20 (12.12)	7;2(1;7)

All participants were tested on the British Picture Vocabulary Scale (BPVS) (Dunn, Dunn & Whetton, 1982), which is the British equivalent of the Peabody Picture Vocabulary Test – Revised (Dunn & Dunn, 1981). The BPVS standard score is a derived measure that expresses the extent to which a participant's score is above or below the mean score for people in the same chronological age group. Like some IQ tests (e.g. Wechsler Adult Intelligence Scale, 1986), the BPVS uses a mean of 100 and a standard deviation of 15. BPVS vocabulary mental ages are also presented in the table. As can be seen, the WS participants' mean vocabulary mental age was higher (M = 8; 9) than that of the oldest control children, the seven-year-olds (M = 7; 2).

This allowed us to be confident that any sub-par performance by WS participants was not a function of a reduced vocabulary. The WS participants were also tested on the Test of Reception of Grammar (TROG) (Bishop, 1983). Their mean grammatical age was 6;8 (s.d. = 3;1).

Materials

We created two sets of test materials: Set A and Set B.

Set A. Set A was composed of four exemplars of each of four types of complex sentence containing a relative clause. The four types – described below – were subject–subject (SS), subject–object (SO), object–subject (OS), and object–object (OO). We have used this terminology because of its long-standing use in the literature, even though most current linguistic analyses would not represent the relative clause as containing either the subject or object of the main clause. Instead, they would view the subject or object position as containing PRO linked co-referentially with the main clause subject or object.

Because we wished to make direct comparisons with our previous study of the TROG, the items for the first three types of relative clause were taken directly from that test (Bishop, 1983). We maintained the exact form and content of those items because, as mentioned above, our previous work (Karmiloff-Smith *et al.*, 1997) had indicated that these were among the most difficult for individuals with WS to comprehend. However, there are certain linguistic limitations in the design of the TROG items. For example, in the SS items, the relative clause is non-finite, whereas in each of the other types the relative clause is finite. There is also a repetition across sentences of some lexical items and of the copula 'to be'. Nonetheless, for comparative purposes it was essential to keep as close as possible to the original SS, SO, and OS items (see examples below). Since the TROG lacks OO items, we created OO exemplars similar in vocabulary and form to the other TROG items.

There were four sentence types, with four items per sentence type, examples of which are given below:

SS: The subject of the main clause is modified; the subject of the relative clause is co-referential with the subject of the main clause, e.g. *The boy chasing the horse is fat*.

SO: The subject of the main clause is modified; the direct object or prepositional object of the relative clause is co-referential with the subject of the main clause. An example of the former is *The cat the cow chases is black*; an example of the latter is *The book the pencil is on is red*.

OS: The object (direct or prepositional) of the main clause is modified; the subject of the relative clause is co-referential with the object of the main clause. An example of the former is *The dog chases the horse that is brown*; an example of the latter is *The square is in the star that is blue*.

OO: The object (direct or prepositional) of the main clause is modified; the object of the relative clause is co-referential with the object of the main clause. An example of the former is *The dog is chasing the cow the boy sees*; an example of the latter is *The pencil is on the shoe the girl has*.

Nine simple sentences, six words in length, were included as fillers in Set A (e.g. *The ring is on the finger*). A single pseudorandom order of filler (9) and experimental sentences (16) was created, with all participants tested on all 25 items.

Set B. Early results with Set A suggested that SO items were particularly difficult to imitate. We thus created a supplementary set (B) of four SO sentences with the relative pronoun that inserted, as in The cat that the cow chases is black. A single pseudorandom order of presentation of these four sentences was used. For Set B, there were no fillers. Eight of the 14 WS participants and all the controls were tested with Set B.

Procedure

For Set A, a mixed design was used, with group (four levels) as a between subjects variable, and a sentence type (four levels) as a within subjects variable. A mixed design was also used for Set B, with group (four levels) as a between subjects variable and sentence type (two levels) as a within subjects variable.

All participants were seen individually in a quiet room. The typically developing children were tested at their school. Eleven of the 14 WS participants were tested in our laboratory; of the remainder, two were tested at their school and one at home.

For the typically developing children there was a single test session. Set A (including filler sentences) was presented first, followed by the BPVS, and then by Set B. Testing of the WS participants took place over two to four sessions. The general pattern was for the BPVS and TROG to be administered during the first one or two sessions as part of a comprehensive cognitive test battery. Set A of the imitation task was presented at least a month later. Set B was presented either on a subsequent occasion, or after an interpolated task between Set A and Set B, as with the typical controls.

Administration of the elicited imitation task followed the same pattern for both groups. The experimenter said something like 'We're going to play a game where you have to say what I say. Just copy what I say. Can you say I like ice-cream?' Most participants repeated the practice sentence accurately but a few of the five-year-olds needed several practice trials to understand the task. The experimenter then presented Set A in a single pseudorandom order. Following each response, the experimenter noted whether the participant had repeated the sentence correctly and what changes, if any, had been made. All responses were audiotaped and checked later against the online scoring. Set B was administered in the same way.

Table 2. Numbers of OK responses to Set A sentences in each category

	Response category						
	$\overline{\mathbf{v}}$	Syn	Lex	Mor	Comb	Total OK	Mean (s.d.) OK
SS							
WS	32	6	I	I	0	40	2.86 (1.41)
5 6	30	0	2	2	I	35	3.18 (0.82)
6	39	0	2	0	0	41	3.73 (0.65)
7	34	3	I	0	0	38	3·80 (o·63)
SO							
WS	ΙI	6	I	I	0	19	1.36 (1.20)
5	17	0	2	I	0	20	1.82 (1.25)
6	27	3	0	0	0	30	2.73 (1.10)
7	22	0	I	0	0	23	2:30 (1:06)
os							
WS	18	5	4	2	2	31	2.21 (1.28)
5	6	5	0	ΙI	I	23	2.09 (1.64)
6	30	5	I	3	I	40	3.64 (0.92)
7	27	3	I	5	0	36	3.60 (0.84)
OO							
WS	8	18	2	0	8	36	2.57 (1.45)
5	14	6	2	5	4	31	2.82 (1.25)
5 6	32	6	3	0	I	42	3.82 (0.40)
7	24	6	2	I	I	34	3.40 (0.40)

Sentence types SS, SO, OS and OO are explained in the text. For participants in each group, N=14 for WS, N=11 for five-year-olds, N=11 for six-year-olds, and N=10 for seven-year-olds. Response category V= verbatim; Syn=addition of relative pronoun in SO and OO, addition of relative pronoun plus tensed auxiliary in SS, and substitution of relative pronoun in OS; Lex=change of a lexical item; Mor=change in morphology; Comb=combination of any categories except V; Total OK = total OK responses; Mean OK = mean number of OK responses out of 4 possible.

Scoring

Each response was categorized as OK or as an error. OK responses were verbatim repetitions of the target sentence, or responses that left the meaning and essential structure of the sentence unaltered and that remained grammatical. There were five subcategories of OK responses.

V. Verbatim responses were word-for-word repetitions of the target.

Syntactic change. Additions or substitutions of a relative pronoun occurred if the participants added who, which, or that or substituted (in OS sentences) who or which for that. In SS sentences, the grammatical addition of a relative pronoun also required the addition of tense, for example changing The boy chasing the horse is fat to The boy who is chasing the horse is fat. Omission of that from Set B sentences was also categorized as a syntactic change response. All of these additions, substitutions and omissions were only categorised as syntactic if the sentence remained grammatical.

Morphological change. Minor morphological changes, such as pluralising a single noun, contracting is, or changing the tense or aspect were permitted.

Lexical change. Minor lexical substitutions such as a change of determiner from the to a, or for instance a noun change from lady to woman were permitted. Permutating the terms for subject and object or for the two subjects of the main and relative clauses was not permitted.

Combination. Repetitions which included more than one of the above permissible changes were placed into this category.

RESULTS

To determine whether the four groups differed in their overall ability to imitate sentences, we compared their performance on the nine filler items. All participants found the fillers easy to imitate; most responses were verbatim or had only minor changes. There were no significant differences among the groups. Mean OK scores for the nine filler sentences ranged from 8.15 (s.d. = 2.5) for the WS participants to 9 (s.d. = 0) for the seven-year-olds.

In contrast, the groups did differ significantly in their performance on the test sentences of Set A. Table 2 shows the distribution of the different categories of OK responses for each sentence type. The mean OK responses are graphed in Fig. 1. A mixed-design MANOVA revealed significant main

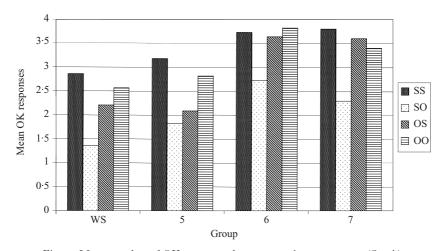


Fig. 1. Mean number of OK responses by group and sentence type (Set A).

effects of group (F(3,42) = 4.48, p < 0.01), and sentence type (F(3,126) = 26.52, p < 0.001), but no interaction (F(9,126) = 1.01, ns). Group differences were further examined using Tukey-HSD tests. Tests for group differences on individual sentence types showed that the WS group and the six-year-olds

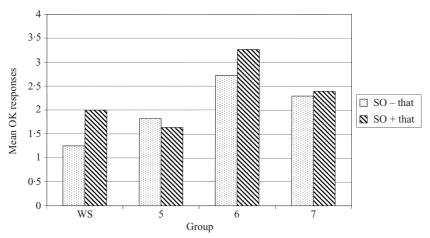


Fig. 2. Mean number of OK responses for sentence type SO, without the relative pronoun (Set A) and with the addition of the relative pronoun (Set B).

differed on SO and OO sentences. The six-year-old participants' scores for OS sentences were also significantly higher than those of the five-year-olds ($p \le 0.05$). No other group differences between scores for individual sentence types reached significance.

The pattern of responses across the different sentence types was similar in the WS and the typically developing groups of participants. As Fig. 1 shows, the WS group most resembled the five-year-old typical controls. These two groups were similar in their overall level of performance as well as in their pattern of responding. This was despite the fact that the WS group had a BPVS test age (8;9) and a TROG test age (6;6) greater than the chronological age of the five-year-olds.

The SS sentences were easiest for the WS and five-year-old participants, followed by OO sentences. Both OS sentences and SO sentences were difficult for both groups, with SO sentences slightly harder. Length did not account for difficulty. The difficulty order (SO > OS > OO > SS from hardest to easiest) did not match the length order (OO > OS > SO > SS from longest to shortest). With the exception of SS sentences, which were both the easiest and shortest, the orders were reversed. The order of difficulty of SO, OS and SS sentences for WS participants mirrored that found in their TROG comprehension data. The percentages of incorrect comprehension responses were 59 % for SO sentences, 32 % for OS sentences and 27 % for SS sentences.

A second analysis compared imitations on Set B - SO sentences with the relative pronoun *that* – to imitations of SO sentences in Set A that did not contain a relative pronoun. A mixed design MANOVA had group as a between-subjects variable and presence or absence of *that* as a within-

subjects variable. Both main effects and the interaction showed clear trends although they did not reach conventional levels of significance. The presence of *that* tended to help imitation (F(1,36) = 3.98, p = 0.054). The groups performed differently overall (F(3,36) = 2.55, p = 0.071). There was a slight tendency for an interaction between group and presence of *that* (F(3,36) = 1.91, p = 0.146). As Fig. 2 shows, the presence of *that* improved performance, particularly for the WS group. Tables 2 and 3 illustrate that the WS group

Table 3. Numbers of OK responses to SO sentences — that (from Set A) and to SO sentences + that (Set B) in each category

SO -that	V	+RP		Lex	Mor	Total OK	Mean (s.d.) OK
WS	8	2		0	I	10	1.52 (1.30)
5	17	0		2	I	20	1.82 (1.25)
6	27	3		0	0	30	2.73 (1.10)
7	22	0		I	0	23	2.30 (1.06)
SO +that	\mathbf{V}	-RP	Subst.RP	Lex	Mor	Total OK	Mean (s.d.) OK
WS	15	I	0	0	0	16	2.00 (1.60)
5	10	8	0	0	0	18	1.64 (1.69)
6	27	7	2	0	0	36	3.52 (1.13)
7	19	3	0	2	0	24	2.40 (1.21)

The two SO sentence types are explained in the text. The numbers of participants in each group are as in Table 1 except that in the WS group n=8. Response category V= verbatim; +RP= addition of relative pronoun; -RP= omission of relative pronoun; Subst.RP= substitution of relative pronoun (*what* for *that*); Lex= change of a lexical item; Mor= change in morphology; Total OK = total OK responses; Mean OK = mean number of OK responses out of 4 possible.

both added pronouns more frequently for Set A and deleted them less frequently for Set B than any of the typically developing groups tested. Table 3 shows that only one OK response to the Set B sentences in the WS group involved omission of *that* (6 % of their OK responses), compared with 33 % of the responses of the five-year-olds, 19 % of the six-year-olds and 13 % of the seven-year-olds.

Error analysis

An analysis of errors across all the sentence types was then carried out. This revealed that the WS participants rarely added a relative clause inappropriately, although this error was quite common among the six- and seven-year-olds (0.9% of the total number of errors made by WS participants, 1% of those made by five-year-olds, 27% for six-year-olds and 31% for seven-year-olds). The WS participants also rarely omitted a relative clause inappropriately (only 4/25 of their errors to OS sentences compared to 16/21

in the case of five-year-olds; the six- and seven-year-olds did not make this error).

All groups' errors included changes that resulted in another type of relative clause structure (21% of WS errors, 10% for five-year-olds, 16% for sixyear-olds and 27% for seven-year-olds). Another large category of error was simplification of the sentence structure by omission of a verb or verb phrase, or an entire clause. This strategy was more common among the WS participants than in the other groups, accounting for 37% of their errors compared with 14%, 3% and 6% of those of five-year-olds, six-year-olds and seven-year-olds respectively.

By contrast, the WS participants were no more likely to fail to respond, or to respond with unintelligible utterances than other groups (7.8% of their errors could be categorised in this way, and 3%, 13% and 2% of five-, six-and seven-year-olds errors respectively).

DISCUSSION

Four aspects of our findings merit particular focus. First, older children and adults with WS were significantly impaired in their ability to correctly repeat relative clause sentences. Their performance was below that of typically developing six- and seven-year-olds. They were most similar to five-yearolds in both their overall number of errors and in the pattern of errors across the different types of relative clauses. This was the case despite their chronological age range of 8;0 to 30;0 years, their vocabulary mental age of almost 9;0, and their comparable ability to repeat the unembedded filler sentences. Thus, the difficulty experienced by WS participants cannot be attributed to vocabulary or simple memory limitations for repeating a string of words. It is syntactic structure that poses the difficulty for them. Our findings suggest that individuals with WS are seriously delayed in syntactic development, even into adulthood. This is compatible with a now growing number of studies that have revealed impaired grammatical development in people with Williams syndrome (Thal et al., 1989; Udwin & Yule, 1990; Mervis et al., 1994; Capirci, Sabbadini, & Volterra, 1996; Volterra et al., 1996; Karmiloff-Smith et al., 1998; Thomas et al., 2001).

Second, despite their poorer performance, the participants with WS showed the same profile of relative difficulty across the various relative clause types as the typically developing children. All four groups were affected by the syntactic structure of the sentences that they were trying to imitate, finding SO sentences hardest and SS sentences easiest. Notably, none of the groups treated the sentences as unstructured lists. Although the psycholinguistic factors that determine relative difficulty of different types of relative clauses are still unclear, our results show that those factors operate in a similar way for WS as for typically-developing individuals. Whatever the interaction of syntactic and cognitive processes in the comprehension and

production of relative clause sentences, it is an interaction that seems to operate similarly for WS and normal individuals, even though it is seriously delayed in WS.

Third, the WS participants were especially aided in their imitations of SO sentences by the provision of an overt relative pronoun. They imitated 'the book that the pencil is on is red' more easily than 'the book the pencil is on is red'. This too indicates a receptivity to features of the syntactic structure of the sentences. An overt marker makes the relation between the matrix and embedded clause syntactically clearer, which may in turn help clarify the conceptual connections among the sentence constituents. Overt markers are likely to be important under conditions of processing or conceptual strain. Normal adults who listen to sentences through white noise, for example, are aided in retrieval when complementizers like *that* are present rather than absent (Valian, 1980). In contrast to typically developing young children, older children and adults with WS may be unable to extract meaning from syntactically complex sentences without a great deal of syntactic support.

Fourth, and relatedly, the WS group differed in an intriguing way from the typically developing young children. They were more likely to insert relative pronouns in their repetitions of Set A. In SS, SO, and OO – the three sentence types where pronouns could be added – WS participants added a pronoun more often than any other group (see Table 2), without, however, making other additions or substitutions at a higher rate than any of the control groups. Although the insertion of pronouns demonstrates syntactic sensitivity, it also indicates a deficit in processing, since the task was to repeat the sentence as given. It seems that listeners with WS have enough syntax to know where to insert a helpful pronoun but cannot then distinguish their recoded sentence from the target.

These two features of the data – that WS participants were aided by relative pronouns and that they inserted pronouns – suggest that WS people are especially dependent on overt markers in order to process multi-clause sentences. However, even with such markers, their performance remains seriously impaired through into adulthood. Future experiments could test the interaction of conceptual and syntactic factors by examining other types of complex sentences and varying both the presence or absence of syntactic support and the presence or absence of pragmatic/contextual support.

How, then, should we characterize the syntactic abilities of people with WS? Our data show that, compared to typically developing children, older children and adults with WS are impaired in their level of functioning but similar in the kinds of relative clauses that they find hard and easy to imitate. However, it is clear that even in adulthood, individuals with WS do not have intact sentence comprehension and repetition abilities; their performance looks like that of the youngest group of controls, the five-year-olds, but they do show sensitivity to syntactic structure.

Our experiment demonstrates the need to investigate at a fine-grained level how syntactic and cognitive factors are interwoven in the language processing of people with WS. If claims about syntactic modularity require intact syntactic performance or a complete dissociation between syntax and mental age, then individuals with WS clearly do not buttress that case. But our results suggest that the behaviour of people with Williams syndrome is far too complex simply to be summarized as either intact or impaired.

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