

Morphological patterns in Hungarian children with Williams syndrome and the rule debates

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Abstract

Williams syndrome (WMS), a rare neurogenetic disorder, has been in the forefront of research in cognitive psychology for the last 10 years. Studies of grammatical development in 14 Hungarian WMS children are presented: they were examined on tasks testing regular and irregular morphology; measures of digit span were also obtained. Results on the production of accusative and plural forms confirmed for Hungarian that regardless of the frequency of the item, inflected forms of irregulars are harder to produce, and often regularized in WMS, revealing a dissociation between the rules of grammar vs. the mental lexicon. Overall performance on the morphology task is associated with the capacity of phonological short-term memory: subjects with higher span perform better on both tasks. The specification of the surprisingly close relation of phonological short-term memory with the linguistic measures awaits further study.

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1. Introduction

Williams syndrome (WMS) is a rare (1 in 25,000) genetically based condition caused by micro-deletion of genes on the long arm of chromosome 7. Children with this syndrome are characterized by relatively preserved linguistic and social skills; they are sometimes called hypersociable because of their affective communicative style and their often indiscriminately positive approach of unfamiliar people. In contrast to their good linguistic abilities they show serious deficits in the domain of spatial cognition and motor skill learning. Despite their problems of visual–spatial organization they show surprisingly excellent performance in face recognition, which might indicate a dissociation in the involvement of the dorsal and ventral brain streams responsible for visual processing. Individuals with WMS also have specific brain morphological differences compared to

controls: decreased overall brain and cerebral volumes, disproportionate volume reduction of the brainstem with a relative preservation of cerebellar and temporo-lymbic structures (Reiss et al., 2000).

In the past decade WMS has attracted the attention of cognitive psychologists, being a population in which good linguistic skills stand in sharp contrast with serious deficits in other cognitive domains (for a survey see Bellugi, Lichtenberger, Mills, Galaburda, & Korenberg, 1999, and the special volume edited by Bellugi & St. George, 2000). Although language is a relative strength in WMS, it is not an intact faculty of the mind: both language development and linguistic performance deviate from the normal in several aspects. In our studies presented here we would like to focus on morphological development in WMS children. Our aim is to test the proposed within-language dissociation between the rule system of grammar and the associative network of the mental lexicon (e.g., Clahsen, 1999; Clahsen & Almazan, 1998), focusing on performance on regular and irregular nominal inflection in a language with a rich morphology.

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1.1. *Language in WMS*

After a late and difficult start of language development, people with WMS achieve remarkably fluent and grammatical speech by school age, with a conspicuously sophisticated and large vocabulary containing many infrequent and unusual words, and a constant urge to chat. Their linguistic skills are in sharp contrast with the general level of their cognitive abilities. The linguistic profile of WMS people is uneven, too. Besides brilliant expressive language, we often find that language comprehension is much more limited, their speech is also often irrelevant and inappropriate, and some of their words and phrases may lack semantic content. The cognitive and linguistic profiles of WMS people have received different accounts by different researchers and research groups. Ursula Bellugi and her colleagues emphasize the dissociation between language and cognition, pointing to the semantic abnormalities in WMS language (e.g., Bellugi, Lichtenberger, Jones, Lai, & St. George, 2000). Another approach draws attention to the within-language dissociation of grammatical rules and lexical processes. On this view, WMS children have a relatively intact grammar, combined with a much weaker lexical system. Therefore, regularizations are characteristic of their performance. This view is based on Pinker and Prince's (Pinker, 1991; Pinker & Prince, 1994) hybrid model of language, and is taken up by Clahsen in connection with WMS (Clahsen, 1999; Clahsen & Almazan, 1998). A third view is developed by Annette Karmiloff-Smith and her research group. Their central claim is that it is not only the representations or processes of language that are impaired in the first place, but as cognitive impairments are a result of a complex epigenetic process, language development in WMS takes a different course, so we might find deviant mechanisms even behind apparently normal performance (e.g., Karmiloff-Smith et al., 1997).

1.2. *The mental lexicon of WMS people*

People with WMS generally perform at a relatively high level on standardized vocabulary tests (although their performance is still below their chronological age, see e.g., Grant et al., 1997; Jarrold, Baddeley, & Hewes, 1998; Karmiloff-Smith et al., 1998), but several tasks show that their semantic organization is different from that of normal controls. Bellugi, Wang, and Jernigan (1994) found in a semantic fluency task that people with WMS produce more infrequent words than normal controls. Another observation revealing unusual organization of the lexicon is that of Vicari, Brizzolara, Carlesimo, Pezzini, and Volterra (1996a) and Vicari, Carlesimo, Brizzolara, and Pezzini (1996b): when subjects have to reproduce words from a word list, normal controls typically reproduce more high frequency words;

in people with WMS no bias is shown towards frequent words in recall. According to the within language dissociation view (Marcus, Gary, Brinkmann, Clahsen, Wiese, & Pinker, 1995; Pinker, 1991; Pinker & Prince, 1994) the two distinct systems can be selectively impaired. On this view, Williams syndrome is an example of an intact rule system with an abnormally operating mental lexicon. In morphology this means that regularly inflected forms (e.g., talk → talked; purportedly generated by the rule system) are produced easily and correctly, but the retrieval of irregular forms (e.g., go → went; stored as a whole in the mental lexicon) is impaired, with signs of overgeneralization. In Clahsen and Almazan's study (1998) English-speaking WMS children could inflect existing regular stems virtually as well as unimpaired controls, while their performance on irregulars was poor; they often overgeneralize the regular suffix both to existing regular forms and to novel words rhyming with existing irregulars. This dissociation is also reflected in their performance on inflecting derivational forms. The results are interpreted as selective impairment of the lexical module of language, as an inability to retrieve information from subnodes of lexical entries.

1.3. *Working memory and languages organization in WMS*

Another cognitive system relevant to language in WMS is working memory. According to Baddeley and colleagues the real function of the phonological loop is not to remember familiar words but to help learn new words (Baddeley, Gathercole, & Papagno, 1998). From this point of view the rate of vocabulary development is influenced by working memory capacity. In agreement with this conception, in childhood large individual differences are found in phonological loop capacity (Gathercole & Adams, 1993). Many studies have found strong correlation between STM performance and vocabulary knowledge, and STM span was found to be a strong predictor of later vocabulary knowledge (Gathercole & Adams, 1993, 1994; Gathercole, Hitch, Service, & Martin, 1997; Gathercole, Willis, Emslie, & Baddeley, 1992). Neuropsychological evidence comes from studies of children with specific language impairment (SLI). SLI children usually lag behind their age in terms of vocabulary development (Bishop, 1992). They show poor performance on both digit span and non-word repetition tasks and recall much fewer phonologically novel names than control children (Taylor, Lean, & Schwartz, 1989). There is also an increasing amount of data concerning the association between working memory and language development in genetic syndromes associated with some mental handicap (Grant et al., 1997; Jarrold, Baddeley, & Hewes, 1999). Wang and Bellugi (1994) compared digit span in individuals with Williams and

Down syndrome, using groups matched on overall IQ. Williams syndrome children had a mean digit span of 4.6, whereas the mean span of the Down syndrome group was only 2.9.

Our Hungarian studies are relevant for several reasons. One of the central issues with regard to language is the proposed contrast between a rule-based and an item-based system, or Grammar and Lexicon within the language faculty, which, according to a strong domain specific view, are associated with different brain areas and can be selectively impaired, (Clahsen, 1999; Pinker, 1991). We show data from an agglutinative language with different stem types fit this model. Besides replicating studies adapted to a typologically different language, we are applying new methods as well in the framework of a longitudinal study, the Hungarian Williams Syndrome Research Project. We are gathering data from a single WMS subject pool on different aspects of language, spatial cognition, elementary vision, visual integration, implicit and explicit rule extraction, and memory (for some preliminary data, see Lukács, Racsomány, & Pléh, 2001).

Our three aims in the study presented here are all related to the debated issues of the nature of language in WMS subjects:

1. Is there a clear dissociation between regular and irregular morphology in Hungarian WMS subjects? Hungarian with its rich morphology and competing suffixation patterns provides a more suitable ground to contrast rule-based and item-based processes than languages studied previously, with more possibilities to vary and control for frequency effects.
2. We wanted to clarify possible frequency effects in morphological overgeneralizations in Hungarian WMS subjects. This has relevance to the issue whether WMS data support a dual system (Clahsen & Almazan, 1998) or basically a simple system of language representation (Thomas et al., 2001).
3. We also looked for possible relationships between phonological short-term memory and morphological performance.

2. Materials and methods

2.1. Subjects

The target group tested in this study consisted of 14 children and young adults with Williams syndrome; their mean age was 13.2 years (ranging from 5.9 to 19.6 years at the time of testing). Subjects were recruited through the Hungarian Williams Syndrome Association, and most of them were assessed in a summer holiday camp for WMS children and their families. Children were tested individually; all of them were assessed on the digit span and a morphology task. In this

paper we obtained control data on primary school 1st, 2nd and 3rd graders, altogether from 29 subjects, in the age range of 7–10. This age range broadly corresponds to the verbal mental age range of the WMS group as measured by the Hungarian version of the Peabody Picture Vocabulary Test (PPVT). The average PPVT performance of the WMS group was 95.55 (SD = 26) points, which corresponds to the average performance of normal children between the age of 84 and 120 months, according to Hungarian standard scores (Csányi, 1976).¹

2.2. Procedure

2.2.1. Verbal short-term memory

A standard measure of verbal short-term memory was taken by the digit span task. In this test, subjects hear digit sequences of increasing length and attempt to repeat them immediately. Digits were taken from those between 1 and 9, and none of them are repeated within one sequence. The score is the amount of digits in the longest sequence correctly repeated; there were two token series with each length. The subject was given the score if she/he could repeat any of them; if the subject failed both trials of one length, testing terminated.

2.2.2. Morphology task

This task contrasted regular versus irregular inflectional forms on the one hand, and frequent versus infrequent items on the other. 32 color drawn picture pairs were used in this experiment, those of Pléh, Palotás, and Lórik (2002), complemented by new picture pairs to adjust the test to our question concerning frequency effects in regular and irregular suffixation. The test had 4 items in each of the 3 regular and 4 irregular classes, 2 frequent and 2 rare. The ‘irregularity’ issue in Hungarian is related to the type of alternations a given stem undergoes. MacWhinney (1978) gives a psycholinguistic exposition of them, together with basic experimental data on their unfolding in children. Frequency estimates were based on Füredi and Kelemen (1989). Table 1 shows examples for each stem type. The first 4 lines are non-productive, ‘irregular’ items. These items are non-productive, therefore, in their case the saturation of the paradigm (how many items follow the pattern) is indicated with the numbers in the first column.

¹ Normal children are used as a reference group instead of a proper control group, as we are trying to decipher differences in the patterns of performance. The issue of proper controls is much debated in the literature: in our study choosing either age-matched or mental age-matched controls would have led to reduction in variance as most of the children in the relevant age groups perform at ceiling level on the morphology task.

Table 1
Examples of stimuli used in the morphology task

Stem class and item number	Examples	
	Frequent	Rare
1. Epenthetic <i>n</i> = 104	majom–majmok 'monkey–mon- key acc.'	bagoly–baglyot 'owl–owl acc.'
2. Lowering <i>n</i> = 71	hal–halak 'fish–fish pl.'	sÁl–sálak 'scarf–scarf acc.'
3. Shortening <i>n</i> = 222	kenyér–kenyerek 'bread–bread pl.'	bogár–bogarok 'beetle–beetle pl.'
4. v-insertion <i>n</i> = 8	kő–követ 'stone–stone acc.'	távcső–távcsövet 'telescope–tele- scope acc.'
5. 'Low V'-final	kutya–kutyát 'dog–dog acc.'	teve–tevék 'camel–camel pl.'
6. C-final	asztal–asztalok 'table–table pl.'	pingvin–ping- vinek 'penguin–pen- guin pl.'
7. 'Non-low V'-final	cipő–cipőt 'shoe–shoe acc.'	hattyú–hattyút 'swan–swan acc.'

2.3. Procedure

The picture depicting an individual object shown first from each pair. After asking the child to provide the name for the object, they were shown the second picture from the pair, and were asked questions prompting either a plural ('What are these?') or an accusative ('What is the boy eating?') forms (accusative and plural questions are alternating, one is requested for each word, and the two forms are taken to be equivalent, as they result in the same stem allomorph). There was no

time limit on the response of the subject. Responses were tape-recorded. The independent variables were the stem type and the frequency of the word, the dependent variable was the correctness of the response. A response was coded as correct if it was properly inflected; it was considered incorrect if it was overregularized or unmarked.

3. Results

Fig. 1 shows errors of WMS children by regularity and frequency. In accordance with previous observations, WMS children seem to regularize exceptional items, and they err less on regulars (for previous research on the issue in Hungarian see Lukács & Pléh, 1999; Lukács, 2001).

A two-way analysis of variance on errors with the factors of REGULARITY and FREQUENCY, regularity had a significant main effect ($F_{1, 52} = 8.74, < .05$), while the frequency effect was not significant ($F_{1, 52} = 0.97, n.s.$), and the interaction of the two factors was not significant either ($F_{1, 52} = 0.46, n.s.$). Results of the analyses performed over the errors on different subtypes of regulars and irregulars only show a significant effect of frequency in the case of -v inserting stems (an irregular type).

Performance on irregulars is poorer, but interestingly enough overgeneralizations also appear in one of the regular stem classes, in consonant ending stems, specifically. In irregulars, beside the general effect of regularity there is a clear frequency effect in -v insertion stems (but none of the other irregular classes). This is also related to age: younger WMS children (under 10 years) are especially prone to overgeneralizations here, their mean errors being 0.3 and 1.6 for regulars and irregulars, re-

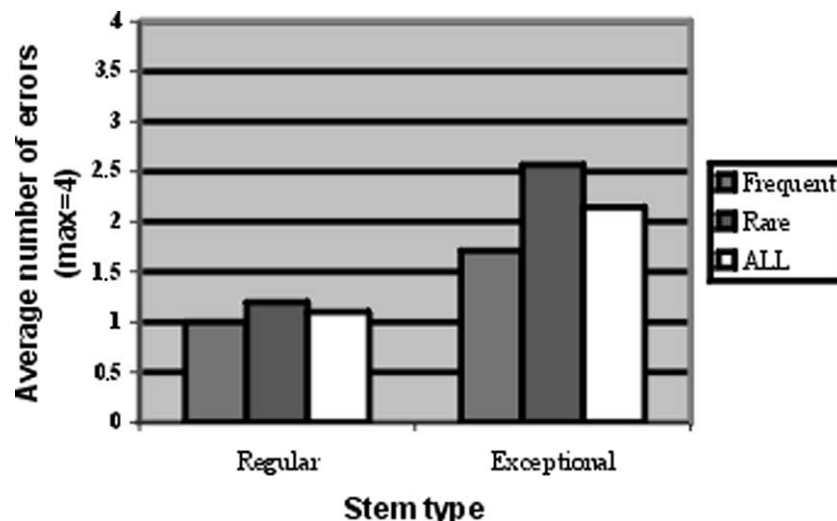


Fig. 1. Frequency and regularity effects on errors in WMS subjects.

spectively, while in the older group the values are 0.6 and 0.6 both (see Fig. 2).

A two-way analysis of variance on control data has shown a significant main effect for FREQUENCY ($F_{1,104} = 9.8$, $p < .01$), REGULARITY also had a significant main effect ($F_{1,104} = 41.7$, $p < .01$) and interaction between the two factors was also significant ($F_{1,104} = 4.1$, $p < .05$). So while WMS children did not seem to be affected by item frequency on the morphology task, frequency did prove to be a predictor of performance in control children. In irregulars, the mean error rate for frequent items was .44, and for infrequent items .96, $t = 3.85$, $p < .001$.

Some interesting relationships hold between short-term memory measures and morphological performance. Table 2 shows that low-span children made more morphological errors both on regulars and irregulars. Following our earlier division, low- and high-span subjects in the Digit span task were identified on the basis of a median division. Subjects with a span of 3 and below versus groups of 4 and more, with 8 and 6 members, respectively. This implies that working memory capacity is related to grammatical proficiency as well. Low-span WMS subjects in general had more morphological errors, but their error rate was especially high with exceptionals.

At the same time, in the control group no significant relationships were obtained comparing low- and high-span subjects. Low-span subjects (with the span of 4) had an average of 1.47 errors with irregulars, while high-span subjects (5 and over) had a mean of 1, $F < 1$. Thus, in a control group with comparable mental age morphological overgeneralization seems to be insensitive to differences in working memory. The same pattern emerges in correlations between digit span and results on the morphology task in the two groups. In WMS children, the correlation between digit span and number of errors was significant in both regulars ($r = -.52$, $p < .01$) and irregulars ($r = -.63$, $p < .02$). Control data do not show any correlation between verbal short-term memory and

Table 2
Effects of digit span differences on morphological errors in WS subjects

Stem type	Low-span	High-span	<i>F</i>	<i>p</i>
Regular	2.75	0.40	5.72	.05
Exceptional	6.25	1.60	6.61	.05
All	9.00	2.00	8.83	.01

morphological performance (regulars: $r = .05$, $p > .1$; irregulars: $r = -.04$, $p > .1$).

4. Discussion

Regarding the three issues raised in designing the experiment, our results gave the following answers. In the morphology task we obtained the usual superiority of performance on regulars over performance on irregulars, which corresponds to the proposal made by Pinker (1991) and Clahsen and Almazan (1998), that WMS people have an intact rule system and an impaired lexicon. There was no overall effect of frequency. A strong main effect of frequency would challenge this view: if performance on regulars had been affected by frequency, it would be a symptom of regulars being treated by the same memory system as irregulars. In WMS many irregulars are regularized, i.e., treated by the rule system, and show no frequency effects. Within this overall picture, a moderate item frequency sensitivity was observed in some stem types (one regular and one exceptional). Normal controls seemed to follow a similar pattern in that they also had more errors on irregulars, but in their case overregularization of irregulars decreased with age. This may be interpreted as evidence for a retarded language development in WMS as suggested by Thomas et al. (2001), at the same time maintaining a basic dual system. More detailed analysis of the data (Lukács, Pléh, & Racsmány, 2003) showed a more intricate picture overgeneralizations being not specific to the WMS group.

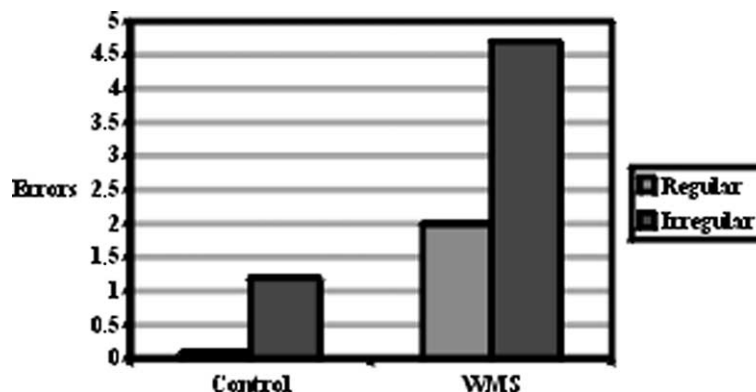


Fig. 2. Number of morphological errors in WMS and normal control subjects.

In the WMS population, working memory span of children seemed to be a more central modulator than token or type frequency of words. Working memory span which was shown to be related in WMS to the knowledge of rare words (Lukács et al., 2001), was also, and not trivially, related to performance on the morphology task. It is too early to draw conclusions, but this may suggest that grammatical proficiency has some intricate relations to working memory, too. Working memory might help to move irregular items to the item-based storage system, as a general mechanism supporting learning new words (Baddeley et al., 1998). As a more general implication of our results relevant to WMS research, we suggest that some of the non-homogeneity of WMS children on cognitive and behavioral measures (emphasized by Bellugi et al., 2000; Jarrold et al., 1998) might reduce, at least in linguistic aspects to differences in verbal working memory capacity. This is especially warranted by the fact that no similar working memory effects were observed in normal controls. Our further studies broadening the age-range within both our clinical and especially in our control samples might help to articulate this suggestion.

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