Contrasting Profiles of Language Development in Children with Williams and Down Syndromes

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Abstract

We describe language acquisition in two distinct genetically based syndromes. Parents of children with Williams syndrome (WMS) and Down syndrome (DNS) were given the MacArthur Communicative Development Inventory (CDI), a parental report measure of child language development. Although both groups of children were found to be equally delayed according to normative standards, differential patterns of language acquisition emerged. Early in language development,, the groups were differentiated primarily by a proclivity for gesture production by the children with DNS. Later in language development,, the groups were cleaved by grammatical development, where the children with WMS displayed a significant advantage over children with DNS. These findings are striking given the marked differences observed between adolescents and adults with WMS and DNS, where participants with WMS exhibit linguistic skills superior to those of matched DNS controls despite significant but comparable cognitive deficits.

Williams syndrome (WMS), a genetically based neurodevelopmental disorder, is characterized by a unique neuropsychological profile in which language appears to "decouple" from other higher cognitive functions (Bellugi, Bihrle, Jernigan, Trauner, & Doherty, 1990; Bellugi, Bihrle, Neville, Jernigan, & Doherty, 1992; Bellugi, Wang & Jernigan, 1994; Karmiloff-Smith, Klima, Bellugi, Grant, & Baron-Cohen, 1995; Mervis & Bertrand, in press; Wang & Bellugi, 1993). Despite average IQ scores ranging from 50 to 70, adolescents and adults with WMS display surprisingly good mastery of complex linguistic structures, as compared to Down syndrome (DNS) participants matched for age and IQ. Furthermore, individuals with WMS have profound spatial cognitive deficits that exceed their levels of general cognitive impairment; a notable exception to this is their relatively unimpaired performance on tests of facial recognition (Bellugi et al., 1992, 1994; Jones, Singer, Rossen, & Bellugi, 1993). In addition, individuals with WMS tend to be quite sociable and affectively expressive (Reilly, Klima, & Bellugi, 1991; Udwin & Yule, 1990). These factors all contribute to a highly unusual neuropsychological profile exhibiting peaks and valleys of higher cognitive functioning.

The contrast between WMS and DNS goes beyond different behavioral profiles. Both syndromes have a unique genetic basis: DNS generally involves an additional chromosomeS recently has been understood as deletion of one copy of the gene for elastin on Chromosome 7, plus surrounds (Ewart et al., 1993; Morris, 1995). The incidence of WMS (1 in 25,000) is considerably rarer than that of DNS, however (1 in 600). In addition, magnetic resonance imaging studies indicate that each syndrome appears to leave its own distinct morphological "stamp" on the brain, with WMS exhibiting relatively spared frontal, limbic, and cerebellar regions, and DNS exhibiting relatively preserved basal ganglia and diencephalic structures (Jernigan, Bellugi, Sowell, Doherty, & Hesselink, 1993; Wang, Hesselink, Jernigan, Doherty, & Bellugi, 1992). Thus, multiple levels of investigation point to behavioral, neuroanatomic, and genetic distinctions between these two neurodevelopmental disorders, providing clues to the relation between genes, brain, and behavior.

A missing piece thus far has been research on the early acquisition of language and other cognitive functions, and the developmental profiles of these two syndromes. As described above, most of the research to date has examined individuals with WMS and DNS in adolescence and adulthood. Studies of younger children with WMS and DNS are of particular importance because by examining differences between the two syndromes in the early stages of cognitive and language development, insights can be obtained into the factors responsible for the very different neuropsychological profiles evidenced in the steady state. Critical questions which drive such investigations of younger children with WMS and DNS are: What happens early in development that leads these two groups to such very different end points? How do their developmental trajectories differ? By examining differential aspects of language development in WMS and DNS, the present study is one of the first and is the largest to begin to address these important questions.

A considerable amount of research has focused on language development in DNS, with investigators primarily noting "delays" rather than "deviance". Although there is general consensus that language is more impaired than other cognitive abilities in individuals with DNS, and that differences between linguistic and non-linguistic cognitive development tend to increase with chronological age, there is some controversy regarding the nature of the language deficit in DNS (cf. Chapman, 1993; Fowler, 1993). The literature indicates, however, that production deficits tend to exceed comprehension deficits, and that grammar appears to be disproportionately affected (Beeghly & Cicchetti, 1987; Beeghly, Weiss-Perry, & Cicchetti, 1990; Chapman, 1995; Fowler, 1990; Miller, 1987, 1992).

In contrast, there has been relatively little research published on early stages of language development in WMS. One study found that performance of children

with WMS on language items on the Bayley Scales of Infant Intelligence exceeded their performance on non-language items, while the reverse was true for children with DNS (Mervis and Bertrand, in press). Similarly, a study of language and symbolic gesture in two young children with WMS uncovered deviant relations between language and symbolic gesture that are consistent with the unusual relation between language and cognition in older individuals with WMS (Thal, Bates, and Bellugi, 1989). Studies also report different relations between linguistic development and purportedly linked non-linguistic cognitive development in children with WMS (e.g., lack of pointing before first referential object word; Mervis and Bertrand, in press; Goodman, 1994, 1995). These studies all involve small groups of children, yielding results that are not always consistent. One recent longitudinal study found considerable variability in language acquisition in three children with WMS (Mervis et al., 1995). While such longitudinal analyses of language acquisition are informative, they typically involve small samples. Larger samples are needed to more clearly identify global patterns of language development in WMS, and to overcome problems with variability which often plague small samples. The present study, using a cross-sectional design, is the first to involve large numbers of children with WMS and DNS in order to address these issues.

There are a number of possibilities for what the trajectories of language development could be like in WMS and DNS: a) consistent with the striking differences in the steady state, in which WMS display far more sophisticated mastery of language than their DNS counterparts, we might expect a developmental trajectory in which children with WMS are from the outset more adept at language acquisition than children with DNS; b) alternatively, based on comparable levels of general cognitive impairment, we might speculate that children with WMS and DNS are equivalently delayed in language acquisition; c) finally, due to the complex and multifaceted nature of language, differences in language development between WMS and DNS may occur across linguistic domains, within linguistic domains, or along boundaries which divide linguistic domains. This study seeks to address these possibilities, as well as the questions raised above: what happens during language development to take these two groups to such very different linguistic end points? How do their patterns of language development compare?

METHODS

Instrumentation

All data for this study were collected using the MacArthur Communicative Development Inventory (CDI), a widely used parental report measure of language development (Fenson et al., 1993, 1994).

The CDI has two scales -- a Words and Gestures scale, which assesses the onset of communication skills (for normally developing children between 8 and 16 months of age), and a Words and Sentences scale, which assess later developing communication skills, including grammatical development (for normally developing children between 16 and 30 months of age). Both scales were utilized in the present study.

Words and Gestures scale. Part 1 of the Words and Gestures scale consists of a checklist of 396 words which have been found to be the first to appear in the receptive and expressive vocabularies of normally developing English-speaking children between the ages of 8 and 16 months (Fenson et al., 1993). Next to each word item, the parent is asked to indicate if their child a) "understands" the word, or b) "understands and says" the word. The checklist is divided into 19 semantic categories: sound effects, animal names, vehicle names, toys, food items, articles of clothing, body parts, furniture, household objects, outside things and places to go, people, routines and games, verbs, words for time, adjectives, pronouns, question words, prepositions, and quantifiers. In Part 2 of the Words and Gestures scale, the child's use of intentional gestures (e.g., pointing, showing) and referential/representational gestures (e.g., putting telephone to ear) is assessed. Gestures of this type are of interest because they have been found to correlate with the onset of language comprehension and/or language production in normally developing children.

Words and Sentences scale. Part I of the Words and Sentences scale consists of a checklist of 689 words that are typically produced by normally developing English-speaking children between the ages of 16 and 30 months (this includes 396 words from the Words and Gestures scale). Next to each word item the parent is asked to indicate if their child "says" the word. The same 19 semantic categories that are found on the Words and Gestures scale are represented on the Words and Sentences scale, with two additional categories for auxiliary verbs (i.e. "helping verbs"), and conjunctions.

Part II of the Words and Sentences scale assesses the acquisition of grammar. Specifically, parents are asked if their child has begun to combine words; possible answers include "not yet", "sometimes", or "often". If the parent indicates that their child has begun to combine words they are then asked to provide examples of the three longest sentences that they have recently heard their child say. In addition, parents are provided with a checklist of nouns and verbs in both regular and irregular inflected forms in order to assess the onset of inflectional morphology. Finally, grammatical complexity is assessed by presenting 37 sentence pairs, each of which represents a minimal contrast in grammatical complexity which are typical examples of early multiword combinations. For example, some pairs index the attainment of bound morphemes (e.g., "two shoe" vs. "two shoes"; "doggie kiss me" vs. "doggie kiss*ed* me"), some index free morphemes (e.g., "baby crying" vs.

"baby *is* crying"; "cookie mommy" vs. "cookie *for* mommy"), and some index sentence embeddings and noun phrases (e.g., "don't read book" vs. "don't *want you* read that book"; "want cookies" vs. "want cookies *and milk*"). The parent simply marks "the one that sounds *most* like the way your child talks right now". The analyses presented in this study will address the production checklist, examples of the child's longest utterances and the grammatical complexity measure, as well as a parental report measure of mean length of utterance (MLU).

For both the Words and Gestures scale and the Words and Sentences scale, normative data also are provided for *associations* between the various dimensions of language development assessed by the CDI. This enables an examination of one dimension of language development in the context of another (e.g., word production in the context of gesture production; grammatical complexity in the context of word production). Naturally, there will be variability in the extent to which these dimensions are associated in different children; furthermore, the strength of associations are likely to vary at different time points in development. By providing normative information about these associations at different time points in language development, the CDI enables one to look for potential dissociations between domains of language, dissociations which not only indicate extremes of normal language development, but are particularly likely to be found in atypical populations such as WMS and DNS. The CDI provides this normative information in the form of "dissociation percentiles", which indicate where a given child "ranks" compared to the normative CDI sample. For example, for "word production relative to word comprehension," a child who scores in the 80th percentile is producing more words than 80 percent of the children in the CDI normative sample who were comprehending the same number of words as she (indicating high production relative to comprehension); accordingly, a child who scores in the 20th percentile is only producing more words than 20 percent of the children in the CDI normative sample who were comprehending the same number of words as she (indicating low production relative to comprehension). The more extreme the dissociation percentile (whether high or low), the larger the dissociation between the two domains, with 50th percentile indicating no dissociation whatsoever relative to the normative sample.

Participants

The participants, 54 children with WMS and 39 children with DNS, are part of an ongoing longitudinal investigation of language acquisition. The WMS group is comprised of 30 males and 24 females, and the DNS group is comprised of 23 males and 16 females. For the data points reported in this study, participants ranged in age from 12 months to 76 months. Participants were recruited through the Williams Syndrome Association and Down Syndrome Association (through advertisements in the national and regional newsletters), as well as through medical and other professional contacts. Because of the nature of the study, diagnostic information was acquired through parental report. Participants with DNS were included if parents indicated that diagnosis had been confirmed by chromosomal analysis. Participants with WMS were included if they had been diagnosed with Williams syndrome and did not evidence any confounding developmental abnormalities. Many of the children have been administered both the Words and Gestures scale and the Words and Sentences scale of the CDI. This paper presents cross-sectional data from the children's first data points on each scale, resulting in 74 WMS data points and 58 DNS data points.

Procedure

Through initial contact with parents, the child's approximate level of language development was ascertained in order to determine the appropriate CDI scale to administer. If parents indicated that their child was producing less than 50 words and was not yet combining words, the Words and Gestures form was sent. If parents indicated that their child was producing more than 50 words or was combining words, the Words and Sentences form was sent. If it was not clear what level of language development the child had reached, parents were asked to complete both scales. Parents were mailed the CDI along with a self-addressed, postage paid envelope for its return. Instructions for completing the CDI are stated clearly on the form itself, and a cover letter accompanied the questionnaire and provided the telephone number of a researcher who assisted parents with any questions or comments regarding the questionnaire.

RESULTS

Whole-Sample Results Across Both Scales

As a first pass through the data, pooling all data points across both forms of the CDI enabled an examination of the sample as a whole. This is important because different patterns of results on the two forms could potentially be caused by sampling effects due to having the "more advanced" children receive the Words and Sentences scale. Because of differences between the forms, the only variable on which data points can be compared across forms is language age-equivalent scores based on the normative data for the CDI. After excluding data points for which a child had both the Words and Gestures and Words and Sentences scales administered at the same age, our sample for this analysis contained 69 WMS data points and 54 DNS data points (9 data points were excluded, 5 WMS and 4 DNS, with the higher score being retained for each child). Table 1 indicates characteristics of the sample.

A one-way analysis of variance (ANOVA) yielded no differences between the two syndrome groups overall in age F (1, 122) = 0.896, *ns*. There was a trend for the children with WMS to produce larger absolute numbers of words F (1, 122) = 3.4, p = .07; however, this could be related to the fact that there were more children with WMS than DNS who completed the Words and Sentences scale, which has a higher ceiling for number of words produced (maximum is 689, versus 396 for the Words and Gestures scale). Thus, language age-equivalence, based on the CDI normative sample, is a more appropriate anchor on which to compare the two groups. Analysis by one-way ANOVA yielded no difference between the two groups in overall language age-equivalence F(1, 122) = 0.954, *ns*. The individual data points depicted in Figure 1 illustrate that overall the two syndrome groups appear to be producing similar numbers of words throughout the age range sampled here. Importantly, although there is variability, these children are on average 20 months behind their normally developing peers with regard to expressive language.

Following the initial analysis of all data points combined, the two scales were analyzed separately in order to examine the more detailed information about language development that the CDI provides. Unless otherwise indicated, analyses were conducted using one-way ANOVA, with syndrome as the independent variable.

Words and Gestures Scale: The Onset of Symbolic Communication

Overall findings. A total of 66 data points initially were obtained for the Words and Gestures Scale (34 WMS and 32 DNS). After excluding those children who were producing more than 300 words, considered to be "ceiling" for this scale, 60 data points remained (32 WMS and 28 DNS). Table 2 describes characteristics of the sample both before and after this exclusion.

The analyses discussed below included only children producing 300 words or less on the CDI Words and Gestures scale. No significant group differences were found with regard to age, number of words comprehended or number of words produced, F(1, 59) = 0.37, 1.8 and 0.07 respectively, *ns*. Group differences did emerge, however, with regard to total number of gestures, F(1, 59) = 9.9, p< .01. The children with DNS produced significantly more gestures than did the children with WMS (see Figure 2). The mean language age for the WMS group was 14.5 months, and the mean language age for the DNS group was 15.4 months F(1,59) = 0.70, *ns*. Importantly, both groups of children were delayed relative to normal children, falling well below the 10th percentile according to the CDI normative sample (Figure 3).

Relation between components of early language development. The relations among comprehension, production, and gesture in WMS and DNS also are informative, as they may yield clues to mechanisms which may differentiate the two syndromes either from one another or from normal children. As mentioned earlier, the CDI provides normative data for indices of dissociation among these three components of early language, in the form of percentile scores which indicate where a given child "ranks" relative to the CDI normative sample. In addition to enabling one to look for potential dissociations among domains of language development, these percentile scores allow for an examination of the relations among various language parameters intraindividually, rather than relying on sample means.

Three dissociation percentile scores were examined for the Words and Gestures scale: 1) word production level given the child's word comprehension, 2) gesture level given the child's word comprehension, and 3) gesture level given the child's word production. Table 2 provides the means and standard deviations for these variables (some children (2 DNS, 5 WMS) had missing values for the production/comprehension variable, because the dissociation percentiles can not be derived reliably for those children near the floor or ceiling of the normative tables). Results indicated that relative to normally developing children, on average children with WMS and DNS had similar relations between word comprehension and word production, F(1, 52) = 0.151, *ns*. That is, relative to normal children at the same comprehension levels, the WMS group was on average at the 63rd percentile for word production, while the DNS group was at the 60th percentile. Two-tailed binomial tests indicated that many more children in the WMS group were above the 50th percentile than expected by chance (21/27 were above 50th percentile, p = .004) and somewhat more children in the DNS group were above the 50th percentile than expected by chance (18/26 were above 50th percentile, p = .05). A chi square analysis revealed no significant difference between groups on this parameter X^2 (1, N = 53) = 0.5, *ns*.

The relation between word comprehension and gestures and between word production and gestures was different for the two syndrome groups, however. Results indicated that relative to normally developing children, children with DNS on average had significantly more gestures given their word comprehension and word production levels than did children with WMS. F(1, 59) = 10.2 and 11.3 respectively, p < .01 for both analyses. Furthermore, the children with DNS in this sample gestured more than most normal children do at similar comprehension and word production levels. The DNS group was on average at the 77th percentile for gestures relative to normal children at the same comprehension levels, and at the 80th percentile relative to children at the same production levels, while the WMS group was at the 55th percentile for gestures relative to normal children at the same comprehension and production levels. Two-tailed binomial tests indi-

cated that many more children in the DNS group were above the 50th percentile on these variables than expected by chance (25/28 were above 50th percentile, $p<.0001$, while no more children in the WMS group were above or below the 50th percentile than expected by chance (14 of 32 were below 50th percentile, $p = ns$; a chi square analysis revealed significant differences between groups on these parameters $X^2(1, N = 60) = 8.03$ for both analyses, $p< .01$. Figure 4 depicts the relationships among word production, word comprehension, and gesture for the two syndrome groups.

Differences between children producing less than 50 words and children producing more than 50 words. Because of the relatively large word production range in the samples, it is instructive to look at children in the earliest stages of word production (<50 words) separately from those who have larger productive vocabularies (>50 words). Productive vocabulary of 50 was used as a cutoff because prior to this lexical level most children are at the one-word stage, which is considered to be a fairly homogeneous stage of language development (e.g., Nelson, 1973). As it turns out, results were different in the two subsamples, and different from the sample as a whole. Table 3 lists characteristics of the two subsamples.

In the subsample of children with productive vocabularies less than 50 words, those with DNS tended to comprehend more, $F(1, 36) = 3.8$, $p = .06$; produced significantly more words, $F(1, 36) = 14.4$, $p < .001$, and gestured more, $F(1, 36) = 8.6$, $p < .01$ than did the children with WMS. In contrast, in the subsample of children with productive vocabularies greater than 50 words, word comprehension and word production were not different in the two syndrome groups, $F(1, 22) = 0.846$ and 0.577 respectively, *ns*, while the DNS participants still gestured significantly more than those with WMS, $F(1, 22) = 13.1$, $p < .01$ (see Figure 5). These differential findings in the children producing less than 50 words versus those producing more than 50 words may have been influenced in part by the presence of a number of WMS data points in our sample from older children who were still in the very first stages of language development. It is possible that these older children may in fact not have "classic" WMS (i.e., deletion of one copy of the elastin gene), or may be more affected (i.e., may have a larger deletion). Studies are currently under way to tease apart such variability in the phenotypic presentation of WMS. Nevertheless, these findings suggest the possibility that in the earliest stages of language acquisition children with DNS may have an advantage over children with WMS, an advantage which attenuates as the children acquire larger vocabularies (and subsequently begin to develop grammar). This finding, if replicated, would be extraordinary in that it would represent a complete reversal of the later linguistic profile of individuals with WMS and DNS.

Summary of indings from Words and Gestures scale of the CDI.

Taken together, the current findings indicate that there are minimal differences between children with WMS and children with DNS early in language development, with the notable exception of gestures, in which the communicative abilities of children with DNS outstrip those of children with WMS. At this earlier stage of communicative development, if anything, children with DNS may have an overall advantage over children with WMS, an advantage which fades as productive vocabulary increases. Although there is considerable variability, both groups of children are significantly delayed in their language development. We now turn to the next stage in language acquisition, the development of grammar. As we will see, it is here where the relative advantage shifts.

Words and Sentences Scale: The Emergence of Grammar

Overall findings, word production. A total of 58 data points initially were obtained for the Words and Sentences Scale (35 WMS and 23 DNS). After excluding those children who were producing more than 600 words, considered to be "ceiling" for this scale, 48 data points remained (27 WMS, 21 DNS). Table 4 describes characteristics of the sample both before and after this exclusion.

Before excluding data points from those children at ceiling on the scale, results indicated that the children with WMS produced significantly more words than those with DNS, $F(1, 57) = 4.41$, $p < .05$. Without these data points, this difference diminished to a trend, $F(1, 47) = 2.12$, $p = .15$. The children who completed this form of the CDI were on average 15 months older than those who completed the Words and Gestures form, and they were producing on average 200 more words. The mean language age for the WMS group was 22.7 months, and the mean language age for the DNS group was 21 months, $F(1, 47) = 2.5$, *ns*. Thus, this more linguistically advanced group was still quite delayed in language development, with both syndrome groups falling well outside of typical developmental limits. The analyses discussed below utilized those children producing 600 words or less on the CDI Words and Sentences scale. When the children at ceiling were included, the differences that emerged in grammatical development were magnified.

Grammatical development. The CDI provides several measures of grammatical development. As described earlier, grammatical complexity is indexed by a checklist of 37 pairs of contrasting phrases, for which the parent is asked to indicate "which sounds the most like what the child is producing now". The score on

this measure of complexity is the total number of word pairs for which it is indicated that the child is currently saying the more complex phrase of the pair. This measure of grammatical complexity has been shown to correlate strongly with laboratory measures of mean length of utterance (MLU) in normally developing children (r= .88 at 20 months, r= .76 at 24 months; Fenson et al., 1994). As depicted in Figure 6, the children with WMS achieved significantly higher scores on this grammatical complexity measure of the CDI than those with DNS, $F(1, 47) = 7.9$, $p < .01$.

Examining a child's mean length of utterance (MLU) has a long history in the child language literature and has been widely used as an indicator of a child's level of grammar (Brown, 1973; Miller, 1981). Accordingly, another index of grammatical development on the CDI is the mean length in morphemes of the three longest utterances (M3L) that the child has produced recently according to the parent (see Fenson et al., 1994, for a discussion of this measure as an index of MLU). In our sample of children with WMS and DNS, strong differences in the M3L emerged (see Figure 6). Children with WMS produced significantly longer utterances than their DNS counterparts, F (1, 43) = 12.9, $p < .001$, complementing the finding of more complexity in the speech of children with WMS as compared to children with DNS. Sample sentences from matched WMS and DNS participants shown in Figure 7 highlight these differences.

Relation between word production and grammar. The differences in grammar between children with WMS and children with DNS are striking. Nevertheless, because of the trend for children with WMS to say more words than those with DNS, it could be argued that this difference is what accounts for the difference in grammatical complexity and phrase length. Fortunately, the CDI provides normative data for indices of "dissociation" between word production and the complexity measure, and between word production and M3L. As with the dissociation measures for the Words and Gestures Scale, these variables allow for an intraindividual comparison of word production and grammar, which is more appropriate than using group means. Two such variables were used for examining grammatical development on the Words and Sentences Scale, each expressed in the form of a percentile score: 1) sentence complexity given the child's word production, and 2) M3L given the child's word production. As described earlier, the dissociation percentiles indicate where a given child "ranks" compared to the normative CDI sample.

Table 4 provides the means and standard deviations for the dissociation variables, which are depicted graphically in Figure 6. As with the Words and Gestures Scale, some children had missing values for these variables (7 DNS, 4 WMS for production/complexity, 1 DNS, 3 WMS for production/M3L) because the percentiles cannot be derived reliably for those children near the floor or ceiling of the normative tables. Results indicated that relative to normally developing children, children with WMS displayed more grammatical complexity $F(1, 36) = 6.9$, $p < .05$, and had a longer M3L, $F(1, 42) = 4.1$, $p < .05$, for their level of word production than did children with DNS (Figure 6). In fact, the children with WMS were on average no different than normally developing children with regards to the relation between the numbers of words they produced and their grammatical development; if anything, they tended to produce longer utterances than do most normally developing children at their lexical levels (16 of 23 children with WMS were above 50th percentile for M3L relative to word production, two-tailed binomial probability = .06; 9 of 20 children with DNS were above 50th percentile, $p = ns$; $X^2(1, N = 43) = 2.65$, $p = .10$. In contrast, the children with DNS were on average quite different from normally developing children in this regard; if anything, their speech was marked by far less grammatical complexity than that of most normally developing children at their lexical levels (11/14 children with DNS were below 50th percentile for grammatical complexity relative to word production, two-tailed binomial probability = .04; 13 of 23 children with WMS were above the 50th percentile, $p = ns$; $X^2 (1, N = 37) = 4.37$, $p < .05$).

Combining Words: An additional index of language development.

Combining words is a critical stage of language development in normal children, although little is known about this important linguistic milestone in atypically developing populations. The CDI assesses word combinations by having parents indicate if their child is combining words "often", "sometimes", or "not yet". To create "dissociation percentiles" from the CDI normative database, percentile ranks for word production were generated for each "level" of word combinations (E. Bates, personal communication 1/94). In other words, a child who was combining words "sometimes" would receive a percentile rank based on how his word production level compared to the normative children who also were combining words "sometimes". A high percentile rank would indicate that he was producing more words than most normal children are when they are at the point where they are only combining words "sometimes" (i.e., he may be considered to be a linguistically "late" combiner); conversely, a low percentile rank would indicate that he was producing fewer words than most normal children are when they are at the point where they are combining words "sometimes" (i.e., he may be considered to be a linguistically "precocious" combiner). While these measures are admittedly "rough", they do enable an examination of this important index of language development.

As it turned out, however, no differences were revealed in the relation between word combinations and

word production levels in WMS and DNS, $F(1, 47) = 1.5$, *ns*. On average, children with WMS were at the 52nd percentile relative to normally developing children, while children with DNS were at the 42nd percentile. Two-tailed binomial tests indicated that for both groups, no more children were above or below the 50th percentile than expected by chance (16 of 27 children with WMS were above the 50th percentile, $p = ns$, 9 of 21 children with DNS were above the 50th percentile, $p = ns$); a chi-square analysis also revealed no significant difference between groups on this parameter, $X^2 (1, N = 48) = 1.27$, *ns*.

Summary of findings with Words and Sentences scale of the CDI. In the group of older and more linguistically advanced children, those with WMS were producing more words than those with DNS. This finding could be an extension of results from the Words and Gestures scale, which indicated progressive improvement once language gets under way in WMS. Importantly, however, when higher production levels were controlled for individually using normatively based dissociation percentile scores, significant differences in grammatical development persisted. Regardless of number of words produced, children with WMS displayed grammatical skills far superior to their DNS counterparts. Moreover, once they reached this level of linguistic development, the children with WMS appeared to display a normal grammatical developmental trajectory relative to word production, whereas the children with DNS continued to evidence delayed grammatical development (see Figure 8). It is quite intriguing that despite their relative language delay, children with WMS not only surpass children with DNS in grammatical development, but they may actually begin achieving grammatical milestones at a normal rate. Equally intriguing is the strong dissociation between lexical and grammatical development in DNS, suggesting a deviant pattern of language development that has not been reported for other groups. These differential patterns will be explored in more detail.

DISCUSSION

In this study, we sought to gain information regarding the emerging linguistic abilities of children with WMS as compared to DNS against a large sample of normative data acquired through the MacArthur Communicative Development Inventory. The data reveals that, despite variability, initially both groups of children are equally delayed in acquisition of words (an average of two years' delay for both groups). First words appear in these children at about the same time that non-retarded children begin to combine words that they already have in their lexicon. This equivalently delayed language acquisition in WMS and DNS is surprising, because it is not at all predictive of the later differences these two syndrome groups evidence in linguistic abilities.

Despite equivalent language delay, two intriguing differences did emerge between groups, namely, an early gesture advantage for the DNS group, and a later grammatical advantage for the WMS group. The proclivity of the children with DNS for gesturing was a very robust finding. A preference for gestural expression over verbal expression in children with DNS has been noted previously in the literature (see Miller, 1987 for a brief discussion). However, as sign language is a communicative modality that is widely taught to young children with DNS (Miller, 1987, 1992), "gestures" on the CDI perhaps could have been confounded with use of signs in this study. Nevertheless, it is possible that their apparent "overgesturing" may be a compensatory strategy used by the children with DNS for their delayed word production. Furthermore, that children with DNS may be relatively good at extracting sensory detail from the visual communicative context may relate to findings of significantly better visual-spatial short-term memory in adolescents with DNS as compared to age- and IQ-matched adolescents with WMS (Wang & Bellugi, 1994).

In contrast, several investigators have noted that children with WMS appear to be selectively "agestural"; they do not evidence the communicative gestures that normally developing children do prior to the onset of first words (Mervis & Bertrand, in press), and they display significantly fewer gestures during free play than age-matched children with DNS and language-matched normal controls (Goodman, 1994, 1995). Furthermore, Thal et al. (1989) reported that 2 young children with WMS displayed dissociations in symbolic gesture that were unlike anything observed in normal children or children with specific language delay. That the current study failed to find evidence of impoverished gesturing by children with WMS could be due to the way in which the CDI assesses gesturing, which may not be as sensitive as observational or laboratory measures. For example, the CDI assesses the number of gestures the children have in their "gestural lexicon", rather than frequency of use of gestures.

Whereas children with DNS may compensate for their poverty of spoken language by the use of gestures, children with WMS may compensate by their "affective style", tending to be overly engaging with social partners. For example, Bertrand, Mervis, and colleagues (1993) have noted that children with WMS spend an inordinate amount of time focused on an adult partner's face, relative to normally developing children. Older children and adolescents with WMS are similarly captivated by social partners; their narratives are rich and complex, containing a variety of devices for engaging the listener (Reilly et al., 1991; Bellugi, Jones, Harrison, Rossen & Klima, 1995). Individuals with WMS are not merely adept at "reading" affect from a social partner, however; recent studies demonstrate that

both children and adults with WMS are able to infer another's emotions or mental state without the aid of affective prosodic or facial cues (Singer, Delehanty, Reilly, & Bellugi, 1993; Karmiloff-Smith et al., 1995). Whatever reasons may underlie the differences in gesture between WMS and DNS, a better understanding of the relation between language development and gesture (an area which is currently under investigation by many researchers) will help to elucidate the significance of these differences.

Regarding the early grammatical abilities of children with WMS, because our research has found remarkably preserved grammatical skills in adolescents with WMS, we speculated how early in language development this advantage/preservation would evidence itself. The present study provides evidence to support preservation of early grammatical skills for children with WMS who have advanced beyond the earliest stages of language development, in marked contrast with their DNS counterparts, who are much slower to develop grammar. Thus, even when no advantage was found in the total number of words produced by children with WMS, they were clearly superior to the children with DNS in their grammatical achievements. It is worth noting that the differences in language acquisition between WMS and DNS could have emerged at any point, including first words. The fact that grammatical development is what differentiates the two groups is an extremely provocative finding, given their differing linguistic profiles evidenced later in life. It is also quite surprising that in the domain of language we observe an early profile of marked delay in WMS which is not predictive of the rich and complex linguistic abilities seen later in development. Further studies of language development throughout childhood are under way to link this early profile with the adolescent and adult profile (Jones, Rossen & Bellugi, 1995; Jones, et al., 1993; Singer, Jones & Bellugi, 1995).

Another trend in early language development which has been noted by researchers and parents of children with WMS is that some of the children seem to say more than they actually comprehend. Although the current study did not completely replicate these findings (the WMS children in our sample were on average in the 63rd percentile for word production relative to word comprehension), we have acquired numerous anecdotes from parents that do attest to this phenomenon. In fact, a number of parents have indicated that on the CDI Words and Gestures form, in addition to the word checklist columns *understands* and *understands and says*, they need a separate column for *says, but does not understand*! Further investigation of this possibility clearly is needed.

Although this study contributes to existing knowledge about language acquisition in atypical populations by providing extensive information about language development in large samples of children with contrasting genetic disorders, there are several limitations that must be kept in mind. First, the cross-sectional nature of the study is able to uncover patterns of language development in the two populations, but not developmental trajectories. Longitudinal studies, currently under way in our laboratory, will complement the findings presented herein and enable us to better address developmental trajectories in the two syndrome groups. Second, the fact that the current data are based on parental report rather than experimental observations could potentially introduce some bias, particularly if parents have a tendency to overestimate their child's linguistic capabilities. Numerous studies, however, have documented the validity of the CDI for assessing language development through parent report (see Fenson et al., 1994), and any parental bias that did exist would not be expected to differ between syndrome groups. Third, although the mailing procedure by which the data were collected enabled us to amass the largest sample of children with WMS ever studied, this technique has inherent limitations such as lack of control over accuracy of the data and homogeneity of the sample. We believe, however, that this limitation is offset by the large sample sizes, which are quite unusual for studies of such rare genetic syndromes. In fact, this study provides the largest sample of children with WMS in this developmental range that has ever been studied, providing crucial information that complements ongoing observational studies in our own lab and others. Furthermore, the basic developmental trends identified in this study are compatible with other observational studies (e.g., Chapman, 1995; Goodman, 1994, 1995; Mervis and Bertrand, in press; Mervis et al., 1995).

A final note pertains to unavoidable sampling issues facing a cross-sectional study such as the present one. In determining where to "dive in" to our assay of language development in a syndrome as rare as WMS, we distributed the CDI to as many parents as we could contact. When we examined our returns, there appeared overall to be more variability in the WMS group than in the DNS group. This could be due to differences in base rates of the syndromes, such that a fuller range of the WMS population was sampled than of the DNS population. The WMS sample was drawn from a national sample while the DNS sample was drawn from a local sample, which could also confer more variability on the WMS sample. Finally, although both syndromes have clear clinical manifestations, it is only in the past year that a genetic probe for WMS has become clinically available, which enables children to be identified as having WMS at an earlier age than when we completed the collection of data for this study. Because of this, it is possible that some children in our sample may not have classic WMS; studies of variability in the phenotypic expression of WMS are currently under way, and should help address sampling issues in the future. Longitudinal studies with matched samples of WMS and DNS are also under way to confirm and expand the current findings.

CONCLUSIONS

This study represents one of the largest investigations to date of emerging language in two genetically based neurodevelopmental disorders, and provides the largest group of young children with WMS ever studied. Despite striking differences in the linguistic abilities of adolescents and adults with WMS and DNS, the results presented herein indicate that both syndrome groups are equally delayed in the onset of language. Early in language development, the groups are differentiated primarily by a proclivity for gesture production by the children with DNS. Later in language development, the groups are cleaved by grammatical development, where the children with WMS display a significant advantage over children with DNS. That individuals with WMS may display normally developing language with the advent of grammar, while those with DNS display what could almost be termed "agrammaticism", highlights the importance of grammar for human language, and raises intriguing questions about genetic influences on brain and language development. This study is one of the first to examine these broader issues of language development in WMS as compared with DNS. Longitudinal studies are under way to assist in identifying developmental trajectories for patterns which may differentiate language acquisition processes in the two syndromes. The relation between gestures and words in WMS and DNS is also being examined in more detail, as well as characteristics of the early lexicon in WMS and DNS as compared to normally developing children. Expanding these investigations to include other atypical populations, such as autistic children and children with focal lesions (e.g., this volume), will provide further opportunities to view variation within and across components of early language, thereby enhancing our understanding of language development and its neural underpinnings.

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Table 1.

Sample Characteristics: Both CDI Scales Combined

	WMS (n=69) M (SD)	DNS (n=54) M (SD)
Age (months)	41 (14)	39 (11)
Word Production	217 (222)	150 (172)
Language Age (in months)	19.5 (6.8)	18.4 (5.2)
Language Delay (in months)	21.5 (12.4)	20.3 (8.9)

Note: M = mean, SD = standard deviation; All comparisons n.s. (at p = .05)

Table 2.

Sample Characteristics: Words and Gestures Scale

	All Subjects			Production <300 words		
	WMS (n=34) M (SD)		DNS (n=32) M (SD)	WMS (n=32) M (SD)		DNS (n=28) M (SD)
Age (months)	34 (11.2)		34 (9.7)	34 (11.5)		32 (9.1)
Word Compreh.	173 (115)	**	224 (118)	163 (111)		201 (107)
Word Production	77 (101)		93 (119)	61 (82)		56 (69)
Gesture Production	35 (15)	*	47 (14)	34 (14)	*	45 (14)
Comp/Prod %	63 (29.3)		60 (26)	63 (29.3)		60 (26)
Comp/Gest %	57 (30.5)	*	79 (20.2)	55 (30.5)	*	77 (20.9)
Prod/Gest %	56 (32.7)	*	81 (21.1)	55 (33)	*	80 (22.1)

Note: M = mean, SD = standard deviation

* $p<.05$

** $p<.10$

Table 3.
Words and Gestures Scale: Subjects Producing Fewer vs. Greater than 50 Words

	Production <50 words			Production >50 words		
	WMS (n=19) M (SD)		DNS (n=18) M (SD)	WMS (n=13) M (SD)		DNS (n=10) M (SD)
Age (months)	34.6 (14.4)		28.8 (8.14)	33 (5.7)	**	38.5 (7.5)
Word Compreh.	96 (82.2)	*	154 (96.7)	260 (65.9)		286 (67.2)
Word Production	5.3 (6.1)	*	21.2 (17.2)	143 (70.8)		118.6 (83.3)
Gesture Production	27 (13)*	40 (15)	44 (8.4)	*	55 (4.6)	
Comp/Prod %	51.5 (30.7)		51.8 (27.8)	83 (11.6)		76 (12.4)
Comp/Gest %	55 (30.8)	**	72 (23.3)	57 (31.3)	*	87 (11.6)
Prod/Gest %	55 (33.2)	**	75 (25.8)	55 (34)*	90 (6.9)	

Note: M = mean, SD = standard deviation
* $p<.05$
** $p<.10$

Table 4.
Sample Characteristics: Words and Sentences Scale

	All Subjects		Production <600 words	
	WMS (n=35) M (SD)	DNS (n=23) M (SD)	WMS (n=27) M (SD)	DNS (n=21) M (SD)
Age (months)	47 (13.5)	47 (8.7)	45 (13)	46 (7.7)
Word Production	366 (208) *	252 (195)	280 (152)	215 (159)
Gram. Complexity	13 (14) *	3 (7.8)	7 (9.1) *	1.2 (2.2)
Prod/Complex %	52 (23.7) *	32 (22.2)	52 (23.7) *	32 (22.2)
MLU	6.2 (4) *	3.4 (2.3)	4.9 (2.4) *	2.9 (.84)
Prod/MLU %	64 (26.3) *	47 (30.8)	64 (26.3) *	47 (30.8)

Note: M = mean, SD = standard deviation

* $p<.05$

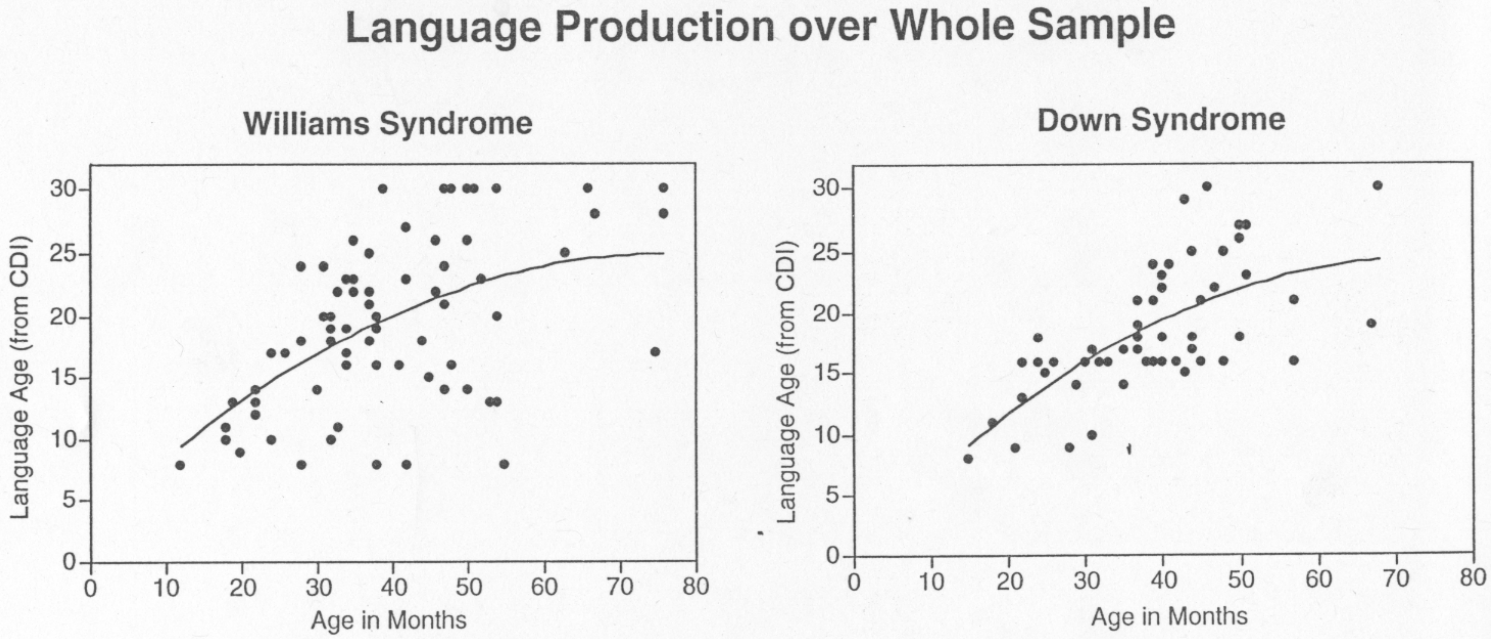


FIGURE 1 Word production across whole sample (both Communicative Development Inventory [CDI] forms combined). Language age is based on CDI normative data for word production.

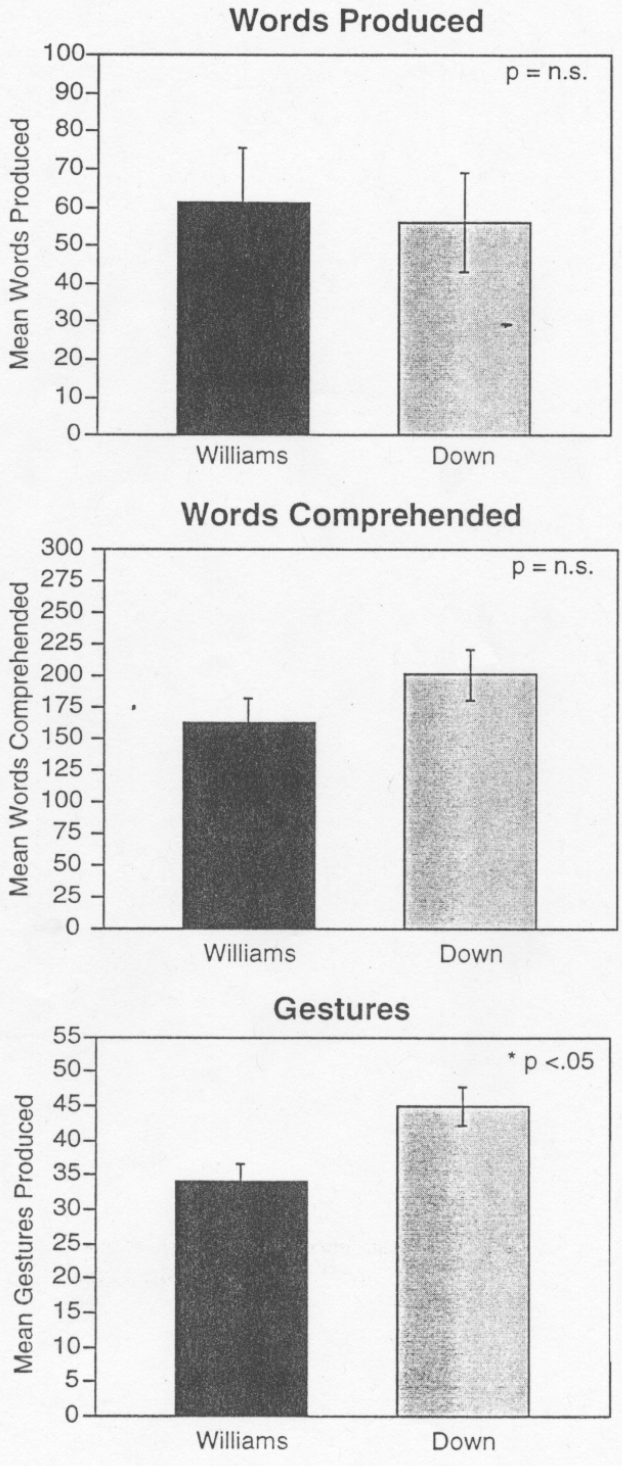


FIGURE 2
Mean words produced (top), words comprehended (middle), and gestures produced (bottom) for children who completed the CDI Words and Gestures form.

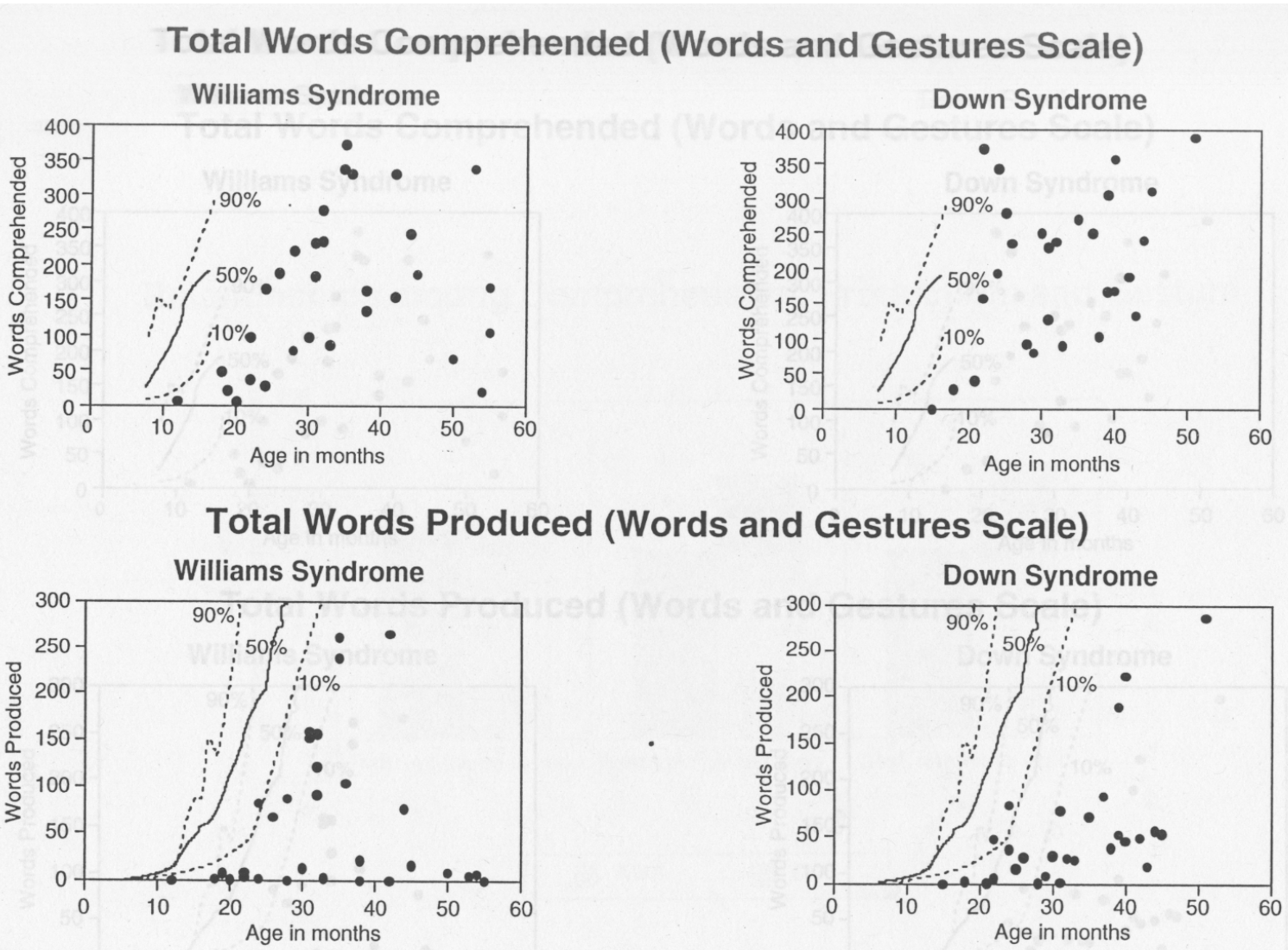


FIGURE 3

Individual data points from children who completed the CDI Words and Gestures form. Words comprehended are depicted in the top two figures, words produced in the bottom two figures. Solid and dashed lines indicate percentiles based on CDI normative data.

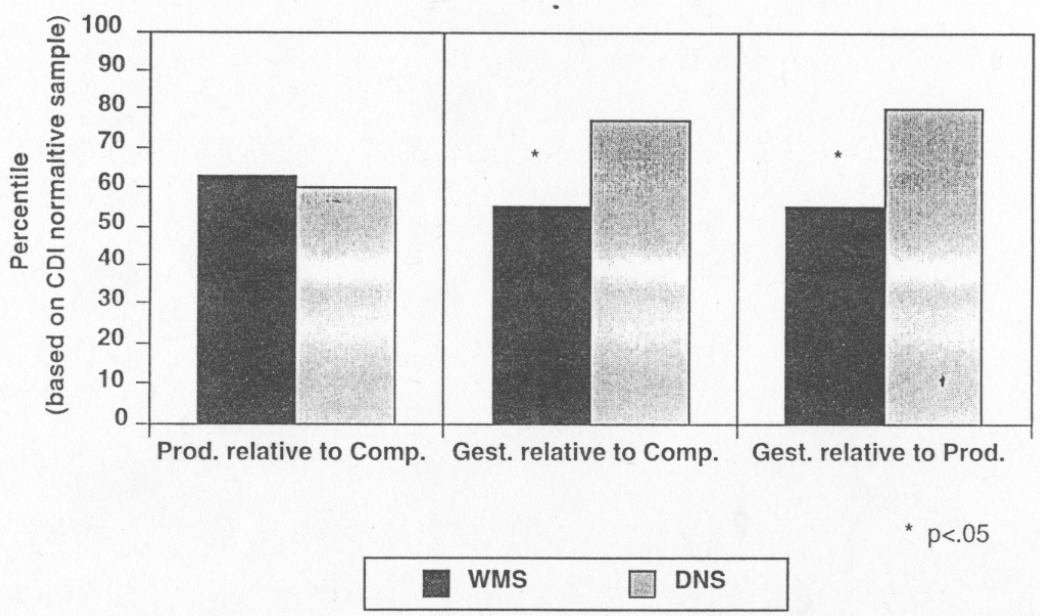


FIGURE 4

Relations among word comprehension, word production, and gesture production for Words and Gestures form. Percentile indicates mean dissociation percentile score based on CDI normative sample. Higher percentile scores indicate children were producing more words (or gestures) than children in the CDI normative sample at the same levels of word comprehension (or word production). 50th percentile is the mean for CDI normative sample.

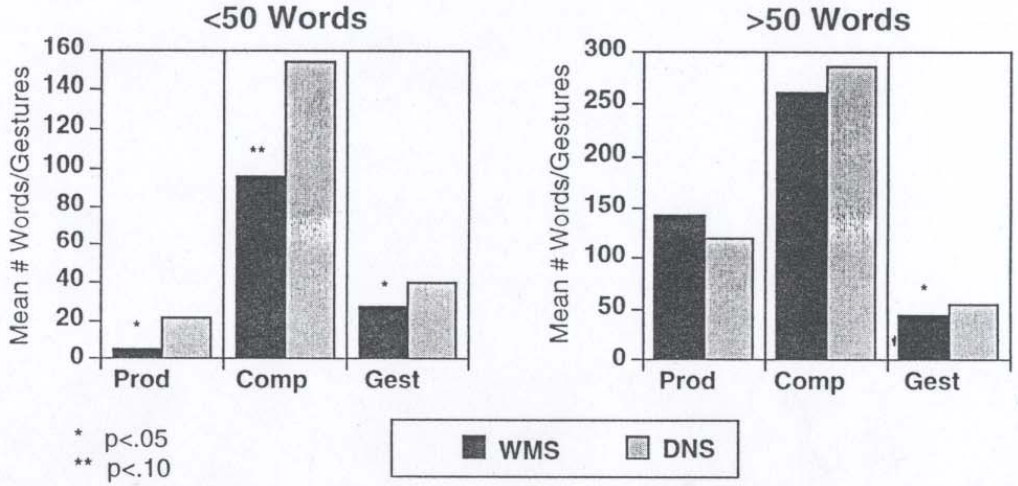


FIGURE 5
Mean levels of word production, word comprehension, and gesture production for children producing less than 50 words (left panel) and more than 50 words (right panel) on the CDI Words and Gestures form.

Words and Sentences Scale Overall Results

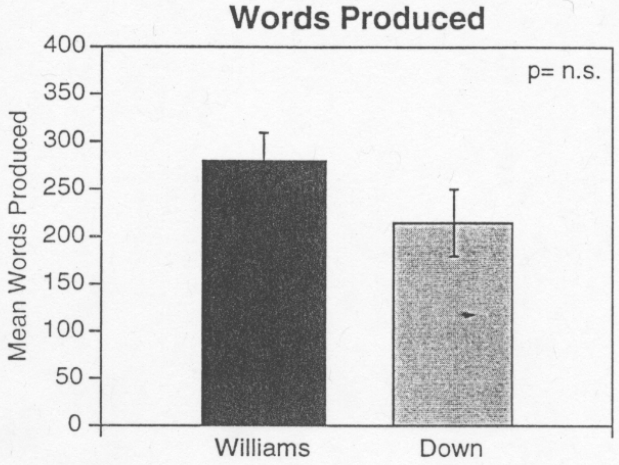
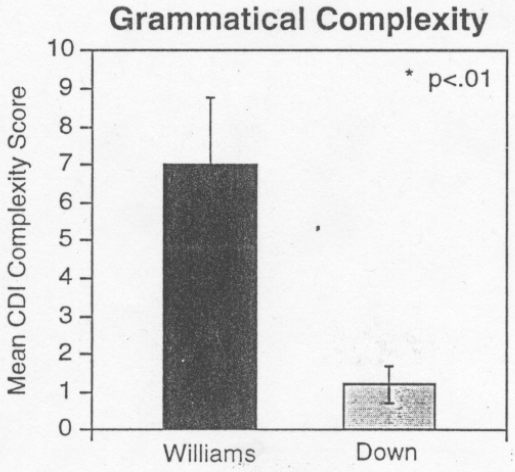
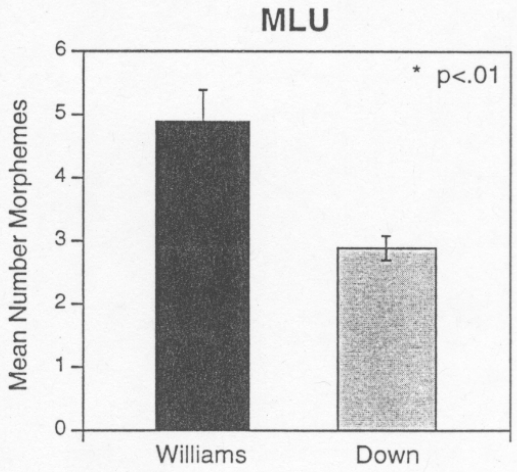
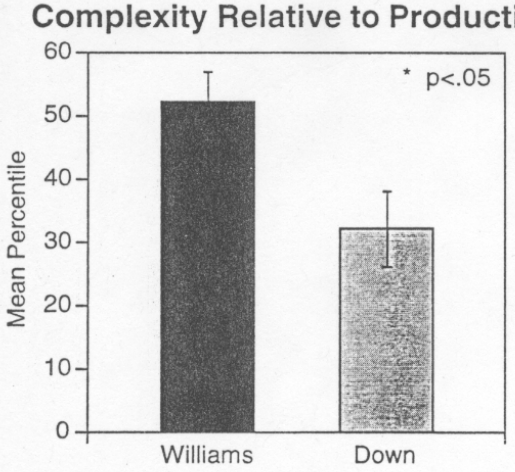
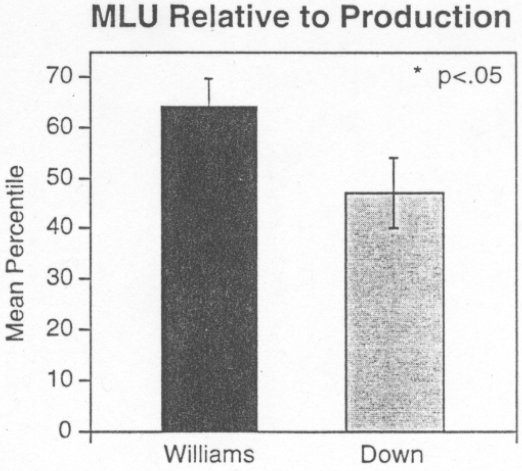


FIGURE 6
Mean levels of word production (top panel), grammatical complexity (middle left panel), and MLU (middle right panel) for children who completed the CDI Words and Sentences form. Bottom two panels depict mean percentile dissociation scores for grammatical complexity relative to word production (left) and MLU relative to word production (right). Higher percentile scores indicate children had more grammatical complexity in their language (or longer MLUs) than children in the CDI normativesample at the same levels of word production. The 50th percentile is the mean for CDI normative sample.









Examples of 3 Longest Sentences

Williams Syndrome Age: 3 years; 3 months	Down Syndrome Age: 3 years; 10 months
Production 610 Words	601 Words
Mamma, need *to* pick *up* toy*s*, vacuum floor.	Gonna go car.
I go *my* room get *one* book bring *out here*.	Matt want bottle.
Please *have some* grape*s* *in my* cup *right now*.	Here-ya-go / Hold me.

Williams Syndrome Age: 3 years; 5 months	Down Syndrome Age: 4 years; 2 months
Production 434 Words	426 Words
Karen read *a* book *with* Daddy.	Baby go bye-bye.
Karen go *on* turtle *in the* pool.	Stop baby don't.
I like *a* bottle please.	Three, four, five.

FIGURE 7
Sample sentences from children matched for word production on CDI Words and Sentences form highlight differences between children with WMS and children with DNS in grammatical complexity and MLU.

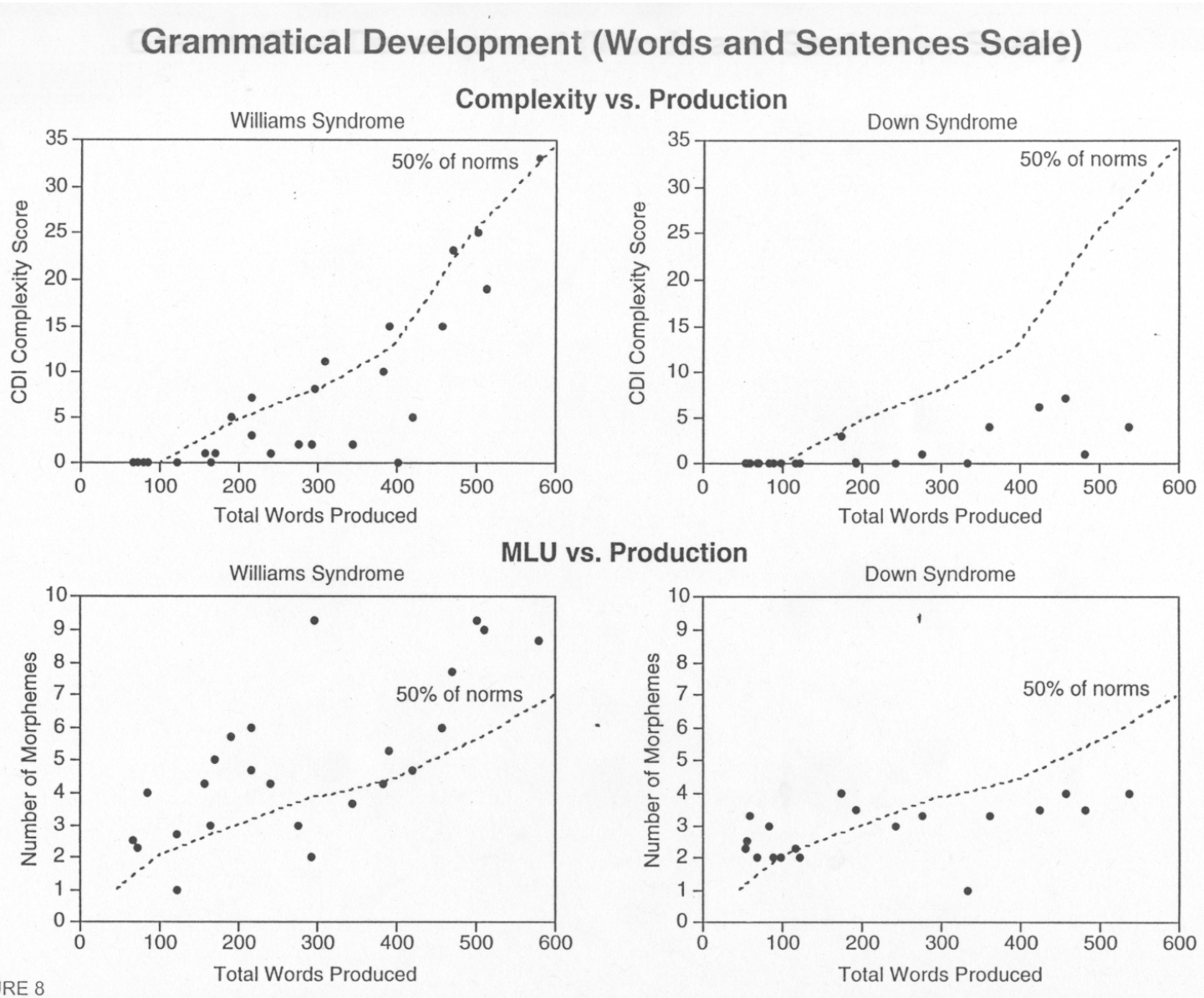


FIGURE 8

Individual data points from children who completed the CDI Words and Sentences form. Grammatical complexity plotted against word production is depicted in top two figures, MLU is plotted against word production in the bottom two figures. Dashed lines indicate the 50th percentile based on CDI normative data.