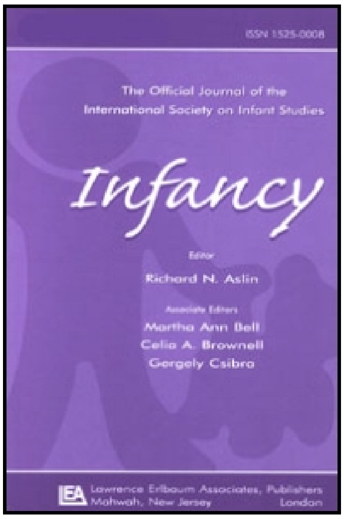

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Early Word Segmentation by Infants and Toddlers With Williams Syndrome

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Early Word Segmentation by Infants and Toddlers With Williams Syndrome

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This study tested the ability of English infants and toddlers with Williams syndrome to segment, that is, to extract from fluent speech, bisyllabic nouns that had either a strong–weak stress pattern (predominant in English), or a weak–strong stress pattern. The testing procedure was the same for both types of words: Children were familiarized with instances of isolated nouns, and then tested on their recognition of these nouns embedded in passages. In English, typically developing infants start segmenting strong–weak nouns by 7.5 months of age, and weak–strong nouns by 10.5 months. Our clinical population was able to segment strong–weak nouns, but failed, despite chronological ages above 15 months, to segment weak–strong words. These results suggest that the development of word segmentation is seriously delayed in Williams syndrome. This deficit in early phonological processing may contribute to a fuller understanding of the late lexical onset in this population, a phenomenon that had hitherto only been explained in terms of cognitive and semantic deficits.

An important aspect of mastering a language is the acquisition by infants of the lexical items that are used in their native language. This task is multifaceted. First, it

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requires that infants segment (i.e., extract) and memorize from fluent speech the sound patterns of the words used in their native language. Second, infants have to build concepts for the objects and events in the world. Finally, they need to map each sound pattern to the particular concept for which it stands. Typically developing infants have begun mastering these requirements for word acquisition by the end of the first year of life, as attested by the onset of word comprehension between 9 and 12 months (Benedict, 1979; Ingram, 1989; Oviatt, 1980), and that of production by 12 months (Barrett, 1995; Benedict, 1979; Clark, 1993; Hallé & de Boysson-Bardies, 1994; Huttenlocher, 1974; Nelson, 1973). Does such a pattern hold in infancy for a clinical population like Williams syndrome (WS) who, in adulthood, displays relatively good linguistic abilities? So far, research has shown language production and comprehension to be seriously delayed in childhood (Paterson, Brown, Gsoedl, Johnson, & Karmiloff-Smith, 1999; Singer, Bellugi, Bates, Jones, & Rossen, 1997). The cause of this delay is unknown, and no research has yet examined early speech processing in this clinical group. In this article, we focus on the hitherto unexplored ability of infants and toddlers with WS to segment words from fluent speech in order to determine whether or not problems at this level of speech processing contribute to an explanation of the initial delay in language acquisition.

WS is a rare genetic disorder occurring in 1 in 20,000 live births (Beuren, 1972; Greenberg, 1990). It is caused by a hemizygous submicroscopic deletion of some 20 contiguous genes on chromosome 7q.11.23 (Donnai & Karmiloff-Smith, 2000; Ewart et al., 1993; Tassabehji et al., 1996). Clinically, individuals with WS can be recognized by their specific facial dysmorphology (elfin-like face) and a number of physical anomalies including supravalvular aortic stenosis, hyperacusis, and dental hyperplasia (Jones & Smith, 1975; McKusick, 1988).

At the cognitive level, individuals with WS, who have a relatively low IQ, can be identified by their characteristic uneven profile. Indeed, some aspects of language performance, face processing, and social interaction seem relatively good, but many nonlinguistic functions such as spatial cognition, number, planning, and problem solving are severely impaired (Arnold, Yule, & Martin, 1985; Bellugi, Bihrle, Jernigan, Trauner, & Doherty, 1990). The unevenness of the profile is particularly striking in that although IQ in this population is generally in the 50 to 65 range, language is often surprisingly proficient in comparison.

As mentioned earlier, although language has been identified as a relative strength in the WS adult profile, its onset is severely delayed in early childhood. There is also some indication that language development in WS does not follow the typical developmental pathway, particularly with respect to the acquisition of words. First, the advantage of word comprehension over word production is smaller in WS than that found in typical development (Paterson, 2000; Singer et al., 1997). Toddlers with WS do not show evidence of better comprehension abilities than toddlers with Down syndrome who will, however, have more limited vocabularies in later life (Paterson et al., 1999). It has also been shown that some of the relationships between linguistic and cognitive milestones found in typical development and in the development of infants with Down syndrome do not hold for WS (Mervis & Bertand, 1997; Mervis, Morris, Bertrand, & Robinson, 1999). Of particular interest to this study, the data on WS showed that, in contrast to typical development, the delayed vocabulary spurt often precedes the emergence of exhaustive categorization and fast mapping by up to 12 months. Moreover, the ability to form name-based categories, which typically develops around 18 months and appears to be linked to lexical development (Nazzi & Gopnik, 2001), is still not present in toddlers with WS, in spite of their large vocabularies (Nazzi & Karmiloff-Smith, 2002; see also Nazzi & Bertoncini, 2003). Finally, it has been found that while fast mapping and mutual exclusivity constrain vocabulary acquisition in young adults with WS, the whole object and taxonomic constraints are much weaker than in controls (Stevens & Karmiloff-Smith, 1997).

The delay in the onset of lexical development in infants with WS and the subsequent atypical pathway remain to be more precisely explained. It is thus crucial to examine development at an earlier stage than language production, as already suggested by the data on early comprehension abilities (Paterson et al., 1999). To explain the atypical lexical acquisition of individuals with WS, most studies have concentrated on their deficit in cognitive and semantic abilities. By contrast, we focus here on another crucial aspect of linguistic development, that of speech perception. We selected this domain because it has been proposed that the ability of individuals with WS to learn words might rely more on good phonological and speech perception abilities than on semantics (Grant et al., 1997; Thomas et al., 2001; Vicari, Brizzolara, Carlesimo, Pezzini, & Volterra, 1996a; Vicari, Carlesimo, Brizzolara, & Pezzini, 1996). The hypothesis is that even though children with WS have problems constructing concepts, they are very good at picking up and memorizing the sound pattern of words. The problem with this proposal lies in the fact that although good phonological abilities were demonstrated for adults, this does not necessarily imply that the adult strength will also hold in infancy (Karmiloff-Smith, 1998; Paterson et al., 1999). It was thus crucial to evaluate the phonological and speech perception abilities of infants and young children with WS to determine whether or not their abilities in this domain are at least at the level of typically developing infants.

In this study, we explore the development of the ability to segment fluent speech. This ability makes possible the extraction of the individual words from the otherwise continuous speech stream. We chose this aspect of speech perception and phonology for several reasons. First, it is crucial for the acquisition of words, given that caregivers rarely present new words in isolation (Woodward & Aslin, 1990; but see also Brent & Siskind, 2001). Second, given that, for medical reasons, it is difficult to have access to very young infants with WS, we wanted to test our over 12-month-olds on abilities more "complex" than simple phonetic contrast discriminations that are present at birth in typical development. Speech segmentation was therefore a good candidate as it typically emerges around 7.5 months (Jusczyk & Aslin, 1995) and further develops up to at least 17 months (Jusczyk, Houston, & Newsome, 1999; Nazzi, Jusczyk, & Bhagirath, 1999). Third, there has recently been extensive research in this domain (limited, however, to the acquisition of English), which showed that speech segmentation is a composite ability involving the exploitation of various types of information. The typical developmental pathway of this ability has already been carefully charted against which to compare our clinical population.

Previous research had shown that infants have an early sensitivity to different types of language-specific acoustic markers of word boundaries (prosodic information: Jusczyk, Gerken, & Redanz, 1993; Turk, Jusczyk, & Gerken, 1995; phonotactic information: Friederici & Wessels, 1993; Jusczyk, Friederici, Wessels, Svenkerud, & Jusczyk, 1993; Jusczyk, Luce, & Charles-Luce, 1994; Mattys, Jusczyk, Luce, & Morgan, 1999; allophonic information: Hohne & Jusczyk, 1994). Building on these earlier results, recent studies have explored the emergence of speech segmentation abilities in typically developing "English-learning" infants, and the kinds of information that these infants use at various points in development. Importantly, this research revealed that, in English, infants start by segmenting monosyllabic words (Jusczyk & Aslin, 1995) as well as bisyllabic words with a strong–weak stress pattern (e.g., candle; Jusczyk, Houston, & Newsome, 1999), usually around 7.5 months. Given that the majority of words in English are stressed initially (Cutler & Carter, 1987), this result shows that American infants start by segmenting the (bisyllabic) words that have the predominant (strong–weak) stress pattern of their native language. The segmentation of bisyllabic words with the opposite weak–strong stress pattern (e.g., guitar) starts only around 10.5 months (Jusczyk et al., 1999b).

It has been suggested that the age difference regarding the onset of segmentation of strong–weak and weak–strong words signals developmental changes in the weight that infants give to different kinds of word-boundary information in English.

At 7 to 8 months of age, infants already seem to use both prosodic information (Echols, Crowhurst, & Childers, 1997; Jusczyk et al., 1999b; Morgan & Saffran, 1995) and statistical information (i.e., the order of occurrence of the syllables in the speech stream, see Jusczyk et al., 1999b, and Saffran, Aslin, & Newport, 1996) to retrieve words from fluent speech. More precisely, it has been suggested that they segment speech at the boundary before strong syllables (based on the use of the prosodic properties of English), and group together syllables that consistently co-occur in the speech stream (statistical/distributional information) provided they do not cross a prosodically defined boundary. Thus at 7 to 8 months, prosodic information is used first and is given more weight. This would explain why when 7.5-month-old infants hear bisyllabic words in fluent speech contexts, they extract both syllables (rather than just the strong syllable) of a strong–weak word (e.g., candle), but only the strong syllable of a weak–strong word (e.g., tar from guitar). It would also explain why, if the weak–strong word is always followed by the same weak syllable (e.g., guitar is), they missegment that sequence (as gui/taris). This is because young infants first use prosodic information to place a word boundary between the first two syllables, and then they use distributional information to group together the last two syllables (see Jusczyk et al., 1999b; and also Johnson & Jusczyk, 2001).

On the other hand, by 10.5 months of age, the weighting between prosodic and distributional information in English changes, with more emphasis placed on both distributional, as well as allophonic (Jusczyk, Hohne, & Bauman, 1999) and phonotactic cues (Mattys & Jusczyk, 2001). This shift in emphasis leads to the possibility of segmenting weak–strong English words. Hence, the evaluation and comparison of the segmentation of strong–weak and weak–strong words in English can be used as an early marker of the level of development of the segmentation ability in infants acquiring that language (similar comparisons between segmentation of predominant and infrequent word patterns might extend to other languages, although no such studies have yet been conducted).

Note that there are crucial differences in the way prosodic and distributional information can be used for segmentation. Indeed, prosodic information is language-specific, so that infants will first need to specify the prosodic properties of their native language before being able to use such information to segment words.[1] However, once they have specified this information, they can readily use it to segment automatically and online any new incoming speech string (e.g., for English, by placing a word boundary before any strong syllable). On the other hand, the use of distributional information regarding syllable order does not require any knowledge of a specific language, and could then be present at birth, or very early in development. Hence, contrary to the use of prosody, the use of distributional information does not require any specific acquisition. Moreover, and again contrary to the case of prosodic information, the use of distributional information requires new statistical analyses for any new incoming speech string; these analyses might take much processing time and effort early in development, explaining why distributional information at 7.5 months can be used to group syllables only when these syllables do not cross a prosodically defined boundary.

This study represents a first exploration of word segmentation in a population of atypically developing infants. Using the procedure of Jusczyk and his colleagues (Jusczyk & Aslin, 1995; Jusczyk et al., 1999b), we examined whether English-speaking infants and toddlers with WS can segment strong–weak and weak–strong bisyllabic English nouns, the comparison of the segmentation of both word types in English being, as argued earlier, a marker of the level of early development of this

[1]The way infants learn the prosodic properties of their native language is still a matter of debate, in part due to the lack or rarity of data in languages other than English. Possibilities include the use of words uttered in isolation (which are not very frequent though), the use of first names and nicknames as prosodic anchors (in English, nicknames tend to have strong–weak patterns, even when the original name is not; see Jusczyk, 1997), or the use of an early sensitivity to rhythmic types (as evidenced by language discrimination; see Nazzi, Bertoncini, & Mehler, 1998; Nazzi, Jusczyk, & Johnson, 2000).

composite ability. With the strong–weak stimuli, infants and toddlers were first familiarized with two target nouns (either kingdom and hamlet, or doctor and candle), and then tested with four passages, one passage for each of the four possible targets. The same procedure was used for the weak–strong stimuli, using the nouns guitar, surprise, balloon, and device. The words used for familiarization were counterbalanced across participants in both conditions. Because WS is very rare, the participants in this study were all tested twice, once with strong–weak words, and once with weak–strong words, the order of presentation of both experiments being counterbalanced across participants. For clarity, both experiments are presented together.

Previous results with this procedure (Jusczyk & Aslin, 1995; Jusczyk et al., 1999b; Nazzi et al., 1999) established that when typically developing infants detect the occurrence of the familiarized words in the passages, they orient significantly longer to these passages than to the passages with the unfamiliarized words. As mentioned earlier, in English, typically developing infants recognize the familiarized strong–weak noun targets by 7.5 months of age and the familiarized weak–strong noun targets by 10.5 months (Jusczyk et al., 1999). Because the chronological and mental ages of our participants were above these ages (see later details), we hypothesized that if infants with WS do not have any difficulties at the speech perception/phonology level, or if such abilities are at least at the level of their mental age (as suggested by Grant et al., 1997; Thomas et al., 2001; Vicari, Brizzolara, et al., 1996; Vicari, Carlesimo et al., 1996) they should be able to segment both types of words. Alternatively, failure to segment in one or both experiments would signal problems of phonological processing.

METHOD

Participants

Nineteen infants and toddlers with WS (8 boys, 11 girls) participated in this study and were tested once with the strong–weak stimuli and once with the weak–strong stimuli (order of presentation being counterbalanced across participants). All the participants were growing up in a monolingual, British English-speaking environment. They had been clinically and genetically diagnosed with WS, using the fluorescent in situ hybridization test for the elastin gene deletion. Their chronological age ranged from 15 to 48 months, with a mean of 33 months. The children's mental age was assessed using the Bayley Scales of Infant Development II (Bayley, 1993). Their mental age ranged from 9 to 32 months, with a mean of 19 months. The parents of only 1 participant reported possible hearing problems (based, however, on poor results of a single test, which might have been the result of attention problems). One additional girl was tested but her data were not included in the analyses because she failed to display headturn responses.

Stimuli

The eight target words used in this study were four strong–weak target nouns (kingdom, hamlet, doctor, and candle), and four weak–strong target nouns (guitar, surprise, balloon, and device). All those words were those used by Jusczyk et al. (1999b), except for balloon, which replaced the original American target beret which has a strong–weak pattern in British English. The choice of using those real words (some of which could be known by our participants) rather than nonsense words was motivated, first, by the fact that we wanted to remain as close as possible to the original Jusczyk et al. (1999b) study. We also did not feel confident about introducing nonsense words because nothing is known about how they affect production in the passages (e.g., less articulation between these words and their context), nor how this could have affected segmentation. In anticipation of our analyses of the data, we asked the parents of each participant whether or not they thought their child knew the eight target words.

A female native speaker of British English recorded eight different six-sentence passages, one passage for each of the four strong–weak target nouns (kingdom, hamlet, doctor, and candle; see Table 1a), and one passage for each of the four weak–strong target nouns (guitar, surprise, balloon, and device; see Table 1b). Each noun appeared in every sentence of its appropriate passage. The passages were based on those used by Jusczyk et al. (1999b), except for the balloon passage that was constructed in a similar way. The talker was encouraged to read the passages in a lively, motherese-like voice, as if reading to a small child. The recordings were made in a sound-attenuated room. The average duration of the passages was 14.3 sec, and for each passage the first occurrence of the target noun occurred within the first second.

TABLE 1a
Test Passages for the Strong–Weak Words

Your kingdom is in a faraway place. The prince sailed past that kingdom last summer. He saw a ghost in this old kingdom. The kingdom started to worry him. He went to another kingdom. Now the big kingdom makes him happy.
Your hamlet lies in a valley. Far away from here is an old hamlet. The kids from the hamlet often sing. Another hamlet is in the country. People from that hamlet like to farm. They live in a rather big hamlet.
The doctor saw you the other day. He’s much younger than the old doctor. I think your doctor is very nice. He showed another doctor your picture. That doctor thought you ate a lot. Maybe someday you’ll be a big doctor.
The candle that you liked has melted. She bought another candle at the shop. You put away the old candle. He gave that candle to you later. She found a place for the new big candle. Your candle is pretty and smells nice.

TABLE 1b
Test Passages for the Weak–Strong Words

Your guitar was really too fancy. There is another guitar at the back. He gave the boy a big guitar. The guitar really needs to be tuned. The man put away his old guitar. He bought that guitar from the music shop.
The surprise should be appreciated. I wonder if she got the old surprise. I think the big surprise is coming. You'll have another surprise tomorrow. Your surprise might not last too long. At least she didn't get that surprise.
Your balloon is funny and new. She gave another balloon to her son. The lady is holding an old balloon. I think that balloon belongs to him. Your dad bought himself a big balloon. The balloon on the shelf is so nice.
Your device can do more than that. I think we need another device. He likes the device that makes ice cream. That device only fixes broken things. The big device is used to sew clothes. No one will ever miss that old device.

The same talker also recorded 15 isolated occurrences in a row of each of the eight target nouns for use in the familiarization phase. Again the talker was asked to repeat the items in a lively voice and with some variation for each target noun. All these lists were 16.0 sec long.

The strong–weak noun lists and the passages containing those words were used, respectively, in the familiarization and test phase of the strong–weak experiment; the weak–strong noun lists and the passages containing those words were used, respectively, in the familiarization and test phase of the weak–strong experiment. The two experiments were run in two separate experimental sessions.

Procedure and Apparatus

The strong–weak and the weak–strong experiments were conducted following the same procedure. Each experiment was conducted in a three-sided test booth made of pegboard panels. Except for a small section of preexisting holes in the front panel used for monitoring the infant's headturns, the panels were backed with white cardboard to prevent the infant from seeing behind the panels. The test booth had a red light and a loudspeaker mounted at eye level on each of the side panels and a green light mounted on the center panel. A white curtain suspended around the top of the booth shielded the infant's view from the rest of the room. A computer terminal (Dell Optiplex GX100) and response box were located behind the center panel, out of view of the infant. The response box, which was connected to the computer, was equipped with a series of buttons that started and stopped the flashing center and side lights, recorded the direction and duration of headturns, and terminated a trial when the infant looked away for more than 2 sec. Information about the direction and duration of the headturns and the total trial duration was stored in a data file on the computer.

The headturn preference procedure was used in this study. Each child was held on a caregiver's lap. The caregiver was seated on a chair in the center of the test booth. Each trial began by blinking the green light on the center panel until the child had oriented in that direction. Then, the center light was extinguished and the red light above the loudspeaker on one of the side panels began to flash. When the child made a turn of at least 30° in the direction of the loudspeaker, the stimulus for that trial began to play. It was played until its completion, or until the child failed to maintain the 30° headturn for 2 consecutive sec (e.g., if the child turned back to the center or looked at the mother, the floor, or the ceiling).

The stimuli were stored in digitized form on the computer and were delivered by the loudspeakers (Gale 20) via an audio amplifier (Sherwood AX4050R). If the child turned away from the target by 30° in any direction for less than 2 sec, and then turned back again, the time spent looking away was not included in the orientation time. Thus, the maximum orientation time for a given trial was the duration of the entire speech sample (i.e., 16 sec). The flashing red light remained on for the entire duration of the trial.

Each experimental session began with a familiarization phase in which children heard repetitions of two of the targets on alternating trials until they accumulated 20 sec of orientation times to each (i.e., about 20 repetitions of each target word, as done by Jusczyk et al., 1999b). If the children achieved the familiarization criterion for one item, but not for the other, the trials continued to alternate until the criterion was achieved for both. Children were familiarized with either kingdom and hamlet, or doctor and candle. The side of the loudspeaker from which the stimuli were presented was randomly varied from trial to trial.

The test phase began immediately after the familiarization criterion was attained. It consisted of two presentations of each of the four six-sentence passages, leading to a total of eight test trials, each passage being presented once in each block, and the order of the different passages being randomized within each block. This number of test trials is half of what had been used by Jusczyk et al. (1999b). This change was required by the fact that our older participants would not have sat through as many trials within the same session as do younger participants. However, we had two reasons to feel comfortable that changing the number of test trials would not hinder segmentation performance. First, reduced numbers of test trials had been successfully used with older typically developing infants (e.g., eight test trials were successfully used in an experiment by Nazzi et al., 1999, with 16.5-month-olds). Second, block analyses of the results by Jusczyk et al. (1999b) had not revealed any block effects, meaning that the segmentation effect was present on all four test blocks (D. Houston, personal communication). Hence, had the typically developing infants in Jusczyk et al. (1999b) been tested on only two test blocks (like our participants), their results would have been identical.

An observer hidden behind the center panel looked through a peephole and recorded the direction and duration of the children's headturns using a response box. The observer was not informed as to the group to which the child was assigned. Moreover, both the observer and the child's caregiver wore earplugs and listened to masking music through tight-fitting closed headphones.

Design. Ten children were tested in the strong–weak condition first, and the remaining 9 children were tested in the weak–strong condition first. For the strong–weak experiment, 10 children were familiarized with kingdom and hamlet, the other 9 with doctor and candle. During the test phase, all the children heard two blocks of the same four passages, each block containing a different random ordering of the passages corresponding to kingdom, hamlet, doctor, and candle. For the weak–strong experiment, 10 children were familiarized with guitar and surprise, the other 9 with balloon and device. During the test phase, all the children heard two blocks of the same four passages, each block containing a different random ordering of the passages corresponding to guitar, surprise, balloon, and device. Children were randomly assigned to the different conditions.

RESULTS

Familiarization Phase

It took a mean of 5.6 trials for participants to achieve the familiarization criterion in the strong–weak experiment, whereas it took them a mean of 5.4 trials to achieve the familiarization criterion in the weak–strong experiment. The difference was not significant, $t(1, 18) < 1$, two-tailed. Overall, during the whole familiarization phase, participants oriented to the lights 60.5 sec in the strong–weak experiment, and 62.7 sec in the weak–strong experiment, a difference that was not significant, $t(1, 18) < 1$, two-tailed. Hence, there was no difference between the two experiments in terms of the duration and number of trials of the familiarization phase, the only two factors that were infant-controlled.

Test Phase

For each participant, we first calculated the mean orientation times to the passages containing the familiarized and unfamiliarized nouns for both the strong–weak and the weak–strong experiments (four means per participant). All participants had mean orientation times greater than 2 sec, which ensures that (as a mean) they all heard at least one target word per test trial.

The second step of the analyses was to identify (and remove from the analyses) outliers. To do so, we calculated for each participant two recognition indexes, one for the strong–weak, and one for the weak–strong condition. Within each condition, the recognition index was obtained by subtracting the orientation times for the passages containing the unfamiliarized words from the orientation times for the passages containing the familiarized nouns. The mean and standard deviation of all recognition indexes were calculated. Outliers were defined as participants having a recognition index (on either condition) 2 *SD*s above or below the mean. Two participants met that criterion in the weak–strong condition, and their data were excluded from both conditions (CA, chronological age; MA, mental age; outlier 1: CA = 18, MA = 10; outlier 2: CA = 48, MA = 32). All analyses were then conducted on the data of the remaining 17 participants (see Table 2 for individual data).

The goal of this study was to determine whether infants and toddlers with WS could segment strong–weak words on the one hand and weak–strong words on the other hand. However, because all participants had been tested with both types of words, we first conducted a grouped analysis in the form of a 2 × 2 × 2 analysis of variance (ANOVA) on the mean orientation times with the main between-subject

TABLE 2
Individual Data for the Infants and Toddlers With Williams Syndrome Included in the Analyses, Presented in Increasing Chronological Age

			OTs (s) Strong–Weak			*OTs (s) Weak–Strong*					
Child	*CA (Months)*	*MA (Months)*	*Fam*	*Unfam*	*RI (Sw)*	*Fam*	*Unfam*	*RI (wS)*	*Order*	*CDI Comp*	*CDI Prod*
1	15	9	5,237	3,993	1,244	3,186	5,653	–2,467	12	24	9
2	22	13	7,436	3,698	3,738	7,584	9,587	–2,003	21		
3	25	14	5,833	2,325	3,508	11,183	8,297	2,887	21		
4	26	16	13,483	12,200	1,255	14,215	13,112	1,103	12	83	9
5	27	16	8,197	7,607	0,591	3,850	4,009	–0,159	12	79	29
6	28	15	5,342	3,978	1,364	5,773	6,458	–0,685	21	40	0
7	29	22	8,903	7,284	1,619	10,660	11,96	–1,299	12	294	222
8	33	19	5,185	4,420	0,765	9,673	8,253	1,420	21		
9	34	16	6,800	6,458	0,341	3,691	2,972	0,718	12		
10	34	17	5,669	4,373	1,296	7,393	6,443	0,950	21		
11	36	26	6,240	5,013	1,227	9,920	7,994	1,927	12		
12	37	20	4,518	3,430	1,088	4,109	3,260	0,849	12	116	76
13	38	19	3,840	6,981	–3,141	4,334	7,170	–2,836	21	140	48
14	42	25	10,080	6,729	3,347	4,374	5,944	–1,569	12	329	295
15	46	—	9,825	9,187	0,638	6,942	4,860	2,081	12	94	22
16	46	25	2,815	3,430	–0,62	7,154	5,628	1,526	21	181	147
17	47	23	3,688	4,557	–0,87	3,725	5,374	–1,649	21	322	58
M	33	19	6,652	5,629	1,023	6,927	6,881	0.047		155	83

Note. CA = chronological age; MA = mental age; OTs = mean orientation times; RI = recognition index; CDI = Communicative Development Inventory. Order: 12 = strong–weak condition tested first; 21 = strong–weak condition tested second.

factor of order and the main within-subjects factors of condition (strong–weak words vs. weak–strong words) and familiarity (passages with familiarized vs. unfamiliarized words). All main effects were nonsignificant: order, $F(1, 15) = 1.30$, $p = .27$; condition, $F(1, 15) = 2.08$, $p = .17$; familiarity, $F(1, 15) = 2.58$, $p = .13$, and all double and triple interactions were nonsignificant, except for the interaction between order and condition, $F(1, 15) = 7.80$, $p = .01$. To better understand this interaction, we then ran separate analyses for the strong–weak words and weak–strong words. Such analyses would also allow us to check for the possibility that participants were segmenting the words in only one of the word conditions, but with effects too small to allow for an interaction between familiarity and condition to show up in the general analysis. A further advantage of the separate analysis of the two word conditions was that it allowed us to introduce in the analyses the counterbalancing factor of group (i.e., which two familiarized words had been used during familiarization), which could not be used in the general ANOVA given that counterbalancing had been made independently in the two word conditions.

Strong–weak condition. Do infants and toddlers with WS segment strong–weak words? Fourteen of the 17 children had longer orientation times to the passages containing the familiarized words than to the passages containing the unfamiliarized words, a result significantly different from chance, sign test, $z(17) = 17.65$, $p = .015$. A $2 \times 2 \times 2$ ANOVA with the main factors of order, familiarity, and the counterbalancing between-subject factor of group (familiarization with kingdom/hamlet vs. doctor/candle) was conducted on the orientation times. A main effect of order, $F(1, 13) = 8.48$, $p = .012$, was found, the orientation times in the strong–weak condition being longer if the participants had been tested on that condition first ($M = 7.51$ sec, $SD = 2.75$ sec) than last ($M = 4.60$ sec, $SD = 1.41$ sec). There was also a main effect of familiarity, $F(1, 13) = 6.57$, $p = .024$, the participants having longer orientation times for the passages containing the familiarized nouns ($M = 6.65$ sec, $SD = 2.74$ sec) than for the passages with the unfamiliarized nouns ($M = 5.63$ sec, $SD = 2.51$ sec). All other effects and interactions failed to reach significance (all $F < 1$).

Do age and lexical development impact on strong-weak word segmentation? Given that most studies having explored word segmentation had been conducted on groups of prelinguistic infants taken from narrow age ranges, we decided to explore whether age and lexical knowledge influenced performance in this study. To evaluate the effect of these factors on the ability to recognize words, correlation analyses were performed between various measures of age and lexical knowledge, and the recognition indexes (i.e., for memory, mean orientation time to passages with familiarized minus unfamiliarized words).

Regarding age, correlations between recognition indexes and CA on the one hand, and MA on the other hand, were computed. Both correlations were nonsignificant, although the interaction regarding CA approached significance: for CA, $r(17) = -.45$, $p = .07$; for MAs, $r(16) = -.24$, $p = .36$. This indicates that recognition performance on the task was independent of both CA and MA.

The effect of lexical knowledge on recognition performance was evaluated in three different analyses. First, we used overall vocabulary estimates based on the MacArthur Communicative Development Inventory (CDI): Words and Gestures (Fenson et al., 1993), which were available for 11 of the 17 participants. Mean comprehension vocabulary was 155 words (range: 24–329) and mean productive vocabulary was 83 words (range: 0–295). Correlations between recognition indexes and the two vocabulary measures revealed a single significant correlation, between word comprehension and word production, $r(11) = .80$, $p = .003$. These findings show that word segmentation can be observed using this procedure with infants who have already started to learn words (as had been found by Nazzi et al., 1999, with typically developing 16.5-month-olds), but also that general level of lexical knowledge does not influence segmentation performance in this clinical group.

Second, we analyzed parental information regarding their children's knowledge of the target words. The number of parents reporting that their child knew the words doctor, candle, kingdom, and hamlet was, respectively, 4 (plus 1 being uncertain), 5 (plus another being uncertain), 0, and 0. Because of these low numbers, and because of the diversity of the individual situations (across children, some knew zero, one, or two words; for some, their known words were the familiarized words, whereas for others they were the unfamiliarized ones), it was impossible to directly determine the effect of word knowledge on recognition performance. To evaluate this effect indirectly, we assigned to each child a known word index.[2] If word knowledge plays a role in word recognition, then one would expect a correlation between the recognition index and the known word index (positive if the effect is enhancing, negative otherwise), a prediction that failed to be confirmed because of the absence of a significant correlation, $r(17) = -.002$; $p = .99$. It is then unlikely that children's knowledge of some of the words had strongly biased the recognition results of the words in the passages.[3]

Finally, the ANOVA reported earlier revealed no effect or interaction of the counterbalancing group factor (being familiarized with doctor/candle vs. kingdom/hamlet). Given that none of our participants knew kingdom or hamlet, this result

[2]Each child received 0 for each unknown word; 0.5 for each possibly known word used for familiarization, or –0.5 if it was not used in familiarization; 1 for each known word used for familiarization, or –1 if it was not used in familiarization. That index could hence vary from –2 to 2.

[3]Note also that even if word knowledge had played a role, the fact that known words were equally distributed between the two familiarity conditions (known words corresponded to familiarized words 5 times and to unfamiliarized words 4 times) would have minimized the influence of that effect on the orientation indexes at the group level.

constitutes further evidence that word knowledge is not a crucial determinant of strong–weak word recognition in this study.

Weak–strong condition. Do infants and toddlers with WS segment weak–strong words? Only 9 of the 17 children had longer orientation times to the passages containing the familiarized words than to the passages containing the unfamiliarized words, sign test, $z(17) = 0$, $p = 1$. A 2 × 2 × 2 ANOVA with the main factors of order, familiarity, and the counterbalancing between-subject factor of group (familiarization with guitar/surprise vs. balloon/device) revealed no main effects or interactions (all $F < 1$). Importantly, regarding familiarity, the participants had equivalent orientation times for the passages containing the familiarized nouns ($M = 6.93$ sec, $SD = 3.25$ sec) compared to the passages with the unfamiliarized nouns ($M = 6.88$ sec, $SD = 2.79$ sec).

Do age and lexical development impact on weak–strong word segmentation? As with the strong–weak words, we evaluated whether age and level of vocabulary development influenced weak–strong word recognition. Regarding age, correlations between recognition indexes and CA on the one hand, and MA on the other hand, were both nonsignificant: for CA, $r(17) = .20$, $p = .43$; for MA, $r(16) = .17$, $p = .54$. This indicates that weak–strong word recognition performance was independent of both CA and MA.

Regarding lexical knowledge, we conducted the same analyses as those conducted for the strong–weak words. First, correlations between recognition indexes, number of comprehended and produced words, conducted for the 11 children for whom we had CDI data, revealed a single significant correlation, between word comprehension and word production, $r(11) = .80$, $p = .003$.

Second, parental information regarding their children's knowledge of the target words was used to calculate a (weak–strong) known word index for each participant. The number of parents reporting that their child knew the words guitar, surprise, balloon, and device was, respectively, 4 (plus 1 being uncertain), 0 (but with 3 uncertain), 13 (plus another being uncertain), and 0. These known word indexes were correlated to the recognition indexes. The correlation was not significant, $r(17) = -.043$, $p = .87$, suggesting that recognition was not strongly influenced by children's knowledge of some of the words.[4]

Third, the fact that the ANOVA reported earlier revealed no effect or interaction of the counterbalancing group factor (being familiarized with balloon/device vs. guitar/surprise), and more participants knew the words balloon and device (13, plus 1 uncertain) than the words guitar and surprise (4, plus 4 uncertain)

[4]Again, even if word knowledge had played a role, the fact that known words were equally distributed between the two familiarity conditions (eight were presented in the familiarized condition, and nine in the unfamiliarized one) would have minimized the influence of that effect on the orientation indexes at the group level.

constitutes further evidence that word knowledge is not a crucial determinant of weak–strong word recognition.

GENERAL DISCUSSION

The main goal of this study was to contribute to a fuller understanding of the serious delay in lexical onset in infants and toddlers with WS, despite their relative linguistic proficiency in later childhood and adulthood. Given that it has been suggested that lexical acquisition in this clinical population might rely on good phonological and speech perception abilities, we investigated the ability of these infants and toddlers to segment words from fluent speech. Using the procedure previously used to study typically developing infants, we succeeded in obtaining data from a relatively large clinical population of infants and toddlers with a very rare genetic disorder. Much of the published work on WS covers groups of only a few participants, whereas we tested 17 infants and toddlers whose chronological ages ranged from 15 to 46 months, and mental ages ranged from 9 to 26 months. We examined the segmentation of two types of bisyllabic words: nouns with the predominant (strong–weak) stress pattern of English, and nouns with the opposite (weak–strong) pattern. The former words start being segmented at 7.5 months, the latter at 10.5 months by infants developing typically in an English-speaking environment (Jusczyk et al., 1999b).

This study first provides evidence for the segmentation of strong–weak nouns by infants and toddlers with WS. Indeed, similar to what had been found for typically developing infants (Jusczyk & Aslin, 1995; Jusczyk et al., 1999b), our participants demonstrated longer orientation times to passages containing words they had just been familiarized to over passages containing nonfamiliarized words. This performance seemed stable over the age range (in terms of both CA and MA), and did not appear to be influenced by lexical development in general (number of comprehended or produced words), or knowledge of the specific target words used in our study. This stability suggests that even our youngest participants, who still had a limited number of words, could do the task and already displayed some ability to retrieve the sound patterns of the words of their native language. Hence, even though our experiment cannot determine whether or not the time course of the emergence of strong–weak word segmentation is identical to or delayed (both in terms of CA and MA) compared with typical development (due to the lack of availability of infants with WS younger than 7.5 months), it does show that segmentation abilities have already started to develop at the onset of lexical acquisition in this clinical population.

In contrast, our results failed to provide evidence for the segmentation of weak–strong words in these infants and toddlers, as they did not show longer orientation times to the passages containing the familiarized weak–strong words over the passages with the nonfamiliarized weak–strong words. Moreover, performance with the weak–strong words did not change with (chronological and mental) age, general lexical development, and especially knowledge of the specific weak–strong targets used here. This absence of performance improvement over the developmental period tested suggests that even the older participants were failing to retrieve the weak–strong words in the passages. Given the ages of our participants, this signals an important delay in the onset of weak–strong segmentation in this clinical population, compared to its emergence at 10.5 months in typical development.

The parallel between the strong–weak and the weak–strong conditions, and the fact that the same infants could segment the strong–weak words, suggests that the failure to segment the weak–strong words is due to the stress pattern difference. However, could other reasons account for that failure? Although the same procedure was used, two methodological elements might have differed between the conditions: prior knowledge of the target words by the participants and characteristics of the familiarization phase. Both of these explanations can be discarded. First, the fact that general lexical knowledge, and knowledge of the actual words used as targets, did not appear to have influenced segmentation in either experiment suggests that this factor cannot account for the difference in the outcomes of the two experiments. Second, even though that might not have been the case, given that the achievement of the familiarization criterion was infant-controlled, the mean duration and number of trials of the familiarization phases were identical in both conditions. This leaves the difference in terms of stress pattern (strong–weak vs. weak–strong) as the most likely interpretation of the present results, and suggests that our participants genuinely had difficulties segmenting the weak–strong words in our study.

This finding raises two major developmental issues. First, we need to specify the reasons for this performance failure with the weak–strong words. Possible causes can be found by comparing our participants' global performance to typical development. Our present pattern of results (segmentation of strong–weak words, but failure with the weak–strong words) is comparable to what has been found for typically developing English-speaking 7.5-month-olds (Jusczyk et al., 1999b). As discussed earlier, it is considered that this pattern signals (a) the predominant use of prosodic information to segment fluent speech, resulting in placing word boundaries before any strong syllables; and (b) the weaker use of distributional information, that can be used to attach to the strong syllable of a word the weak syllable that follows it (allowing for the segmentation of strong–weak words), but not the one that precedes it (which would be necessary to retrieve weak–strong words). Hence, one possible interpretation of these results is that although infants and toddlers with WS can use prosodic markers of word boundaries, they have difficulties exploiting distributional information (i.e., grouping together consecutive syllables). Future research will test this interpretation, adapting some of the experiments conducted on this issue by Jusczyk et al. (1999b) and Saffran et al. (1996).

Second, we need to understand how, given the absence of evidence for weak–strong word segmentation in our study, children with WS ultimately manage to acquire weak–strong words, especially given that some of our participants actually knew a few weak–strong words (13 participants knew the target word balloon).[5] A possibility is that the observed segmentation failure is a performance issue. Children with WS might have some ability to use distributional information, and their difficulties might result from the fact that they need more time and evidence than they were given in this study. It is then possible that in everyday situations, with more time, more occurrences of the new words, and more motivation and visual evidence, children with WS can use distributional information to segment and learn some weak–strong words. Alternatively, children with WS might really be incapable of using distributional information to segment weak–strong words. In this case, they would need to learn these words through processes other than segmentation, for example, by memorizing these words when they are uttered in isolation. At the present time, it is not possible to choose between these two alternatives, but our future planned studies will directly target the use of distributional cues by these children, which should provide new data related to this issue.

At this point, we would like to underline a few methodological considerations. First of all, this is the first time that the headturn preference procedure (classically used in typical developmental speech perception research) has been used to study atypical development. Our successful use of the procedure with infants and children with WS provides encouragement for further employment of this procedure with this and other atypical populations to characterize early speech and language development. It would, for instance, be interesting to test other atypical populations to determine whether the effect observed in this study is a syndrome-specific deficit of WS, or a syndrome-general problem (e.g., by testing infants and toddlers with Down syndrome, who have often been used as a comparison group with WS).

Second, uses of the headturn preference procedure with typically developing infants have normally proved impossible beyond the second birthday, due to older infants becoming quickly bored and literally walking away from the experimental booth. It was a gamble to test toddlers with WS up to 4 years of CA, and yet their testing proved to be unproblematic. This might have been due to the fact that their MAs were lower, or they may be more willing to do the task than typically

[5]The CDI did not prove very useful to quantify our participants' knowledge of weak–strong words, and compare it to their knowledge of strong–weak words, as it includes only very few weak–strong words (8 weak–strong words only, as opposed to 388 strong–weak words). Balloon and hello were the two more frequent weak–strong words among our participants.

developing infants (individuals with WS are very sociable). Whether it will be possible to use this procedure with toddlers and young children from other atypical populations remains an empirical issue.

Third, although some studies of speech segmentation have established word segmentation in infants who had already started to speak (segmentation of weak–strong verbs by 16.5-month-olds; Nazzi et al., 1999), the studies that we have used for comparison with our participants were conducted with prelexical infants. Could the fact that (some of) our participants had rather large vocabularies have impacted the outcome of our experiments? To evaluate this, we correlated performance in both experiments with size of comprehension and productive vocabularies; both correlations were nonsignificant. Hence, knowing words in general did not influence performance in either experiment; that is, it neither influenced the successful segmentation of strong–weak words nor the absence of segmentation of weak–strong words. For these reasons, it is unlikely to have determined why strong–weak words were segmented and weak–strong words were not. Rather, we argue that the explanation is more likely to reside in the fact that segmentation is a phonological process involved in lexical acquisition, but it operates prior to and independently of the lexical level.

To conclude, using for the first time in atypical development the headturn preference procedure, we have explored the ability of infants and toddlers with a genetic disorder to segment words from fluent speech. The aim was to determine whether the serious initial delay of infants and toddlers with WS to establish a lexicon might in part be due to speech perception and phonological problems. Our study demonstrates that young children with WS display difficulties in the domain of speech perception corresponding to the extraction from fluent speech of some of the sound patterns of words. They could segment words with the predominant strong–weak stress pattern of English, but failed to show evidence of the segmentation of the opposite weak–strong stress pattern, suggesting a marked delay in the onset of the ability to segment weak–strong words. Whether or not segmentation of these words emerges at a later age in WS is a question to be explored by future research with a different procedure adapted to the testing of children older than 4 years. These results then converge with those of many recent studies demonstrating that, contrary to strong claims in the literature (e.g., Pinker, 1994), the language of individuals with WS is far from intact. More specifically, our study points to the fact that delays in early vocabulary acquisition are not solely due to the conceptual or semantic problems usually invoked (Mervis & Bertrand, 1997; Mervis et al., 1999; Nazzi & Karmiloff-Smith, 2002; Stevens & Karmiloff-Smith, 1997). It also challenges claims that early word acquisition in this clinical group is grounded on good speech perception and phonological abilities, claims based on results showing that adults and older children with WS rely more heavily on phonology than semantics (Grant et al., 1997; Vicari, Brizzolara, et al., 1996; Vicari, Carlesimo, et al., 1996). As already stressed in our earlier work on other aspects of the WS phenotype (Karmiloff-Smith, 1998; Paterson et al., 1999), our newly identified deficit in this clinical group suggests that it cannot be taken for granted that patterns of processing strengths in adulthood are already in place in early development.

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