



## Brief article

# Infants with Williams syndrome detect statistical regularities in continuous speech



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## ARTICLE INFO

## Article history:

Received 10 July 2015

Revised 9 May 2016

Accepted 16 May 2016

## Keywords:

Williams syndrome  
Statistical learning  
Language development  
Language delay  
Lexical development  
Infancy

## ABSTRACT

Williams syndrome (WS) is a rare genetic disorder associated with delays in language and cognitive development. The reasons for the language delay are unknown. Statistical learning is a domain-general mechanism recruited for early language acquisition. In the present study, we investigated whether infants with WS were able to detect the statistical structure in continuous speech. Eighteen 8- to 20-month-olds with WS were familiarized with 2 min of a continuous stream of synthesized nonsense words; the statistical structure of the speech was the only cue to word boundaries. They were tested on their ability to discriminate statistically-defined “words” and “part-words” (which crossed word boundaries) in the artificial language. Despite significant cognitive and language delays, infants with WS were able to detect the statistical regularities in the speech stream. These findings suggest that an inability to track the statistical properties of speech is unlikely to be the primary basis for the delays in the onset of language observed in infants with WS. These results provide the first evidence of statistical learning by infants with developmental delays.

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## 1. Introduction

Williams syndrome (WS) is a rare genetic disorder caused by a sporadic microdeletion of 26 genes on chromosome 7q11.23 (Hillier et al., 2003). WS has a prevalence of 1 in 7500 (Strømme, Bjørnstad, & Ramstad, 2002). It is a multisystem disorder associated with cardiovascular disease, facial dysmorphology, social disinhibition, and developmental delay (e.g., Mervis & Becerra, 2007).

WS is associated with mild to moderate intellectual disability or learning difficulty with a relative strength in language, especially concrete vocabulary (Mervis & John, 2010). However, notably, the onset of language acquisition is considerably delayed in young children with WS. For example, the median age of acquisition of 10 expressive words is 13.5 months for typically developing (TD) infants (Fenson et al., 2007). In contrast, the median age of acquisition of a 10-word expressive vocabulary is 28.2 months for children with WS (Mervis, Robinson, Rowe, Becerra, & Klein-Tasman, 2003). However, the reasons for the language delays observed in young children with WS are not known. On the one hand, it is possible that TD infants and young children with WS use the same language

learning mechanisms, but the onset of language is simply delayed for children with WS. In this case, researchers could expect to find equivalent mechanisms being used at equivalent stages of word learning, but delayed with respect to chronological age (CA). On the other hand, it is possible that language is acquired using fundamentally different mechanisms by children with WS, either throughout development or at any one point during language development. In fact, seemingly small differences in underlying cognitive mechanisms early in development could have a cascading effect on the development thereafter (Karmiloff-Smith & Farran, 2012).

Statistical learning is a domain-general mechanism that TD infants recruit for detecting words in speech, as well as other aspects of early learning (e.g., Kirkham, Slemmer, & Johnson, 2002; Krogh, Vlach, & Johnson, 2013; Romberg & Saffran, 2010; Saffran, 2003). TD 8-month-olds can exploit statistical patterns to distinguish words from sequences spanning word boundaries in fluent speech (Saffran, Aslin, & Newport, 1996). More recent evidence, based on event-related potentials, indicates that even sleeping TD newborns are sensitive to the statistical structure of linguistic input (Teinonen, Fellman, Näätänen, Alku, & Huotilainen, 2009).

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Given the intellectual disability of those with WS and the fundamental nature of statistical learning it is possible that the language delay in WS is due to a statistical learning deficit early in life. Lacking access to efficient statistical learning mechanisms early in development may put children with WS at a disadvantage in fundamental language acquisition processes such as segmenting words (Saffran et al., 1996) and associating word forms with referents (Graf Estes, Evans, Alibali, & Saffran, 2007; Lany & Saffran, 2010). To address this possibility, we used the statistical learning task previously used with 8-month-old TD infants (Saffran et al., 1996, Experiment 2) to test infants with WS who are in the early stages of language acquisition. We reasoned that failure to take advantage of the statistical information in the continuous speech stream would suggest that a statistical learning deficit may be an important contributor to the language delays observed in children with WS. Alternatively, it is also possible that infants with WS can perform statistical learning; the ability to detect distributional patterns in syllable sequences may not be impaired. If this is the case, it would suggest that the delayed onset of language associated with WS is likely caused by other factors.

## 2. Method

### 2.1. Participants

Eighteen 8- to 20-month-olds (12 females, 6 males) with genetically-confirmed WS and normal or corrected-to-normal vision and hearing participated in this study. All participants had classic-length deletions as determined by FISH or qPCR. The racial/ethnic background of the participants was: 13 White/Non-Hispanic, 3 White/Hispanic, and 2 biracial Non-Hispanic (1 African American/White, 1 American Indian/White). Mean chronological age (CA) was 15.5 months ( $SD = 4.1$ , range: 8.1–20.6 months). Mean Mullen Scales of Early Learning (MSEL; Mullen, 1995) Early Learning Composite (ELC; similar to developmental quotient, DQ) was 64.1 ( $SD = 9.7$ ), indicating mild developmental delay. ELC standard scores ranged from 51 (moderate developmental delay) to 85 (low average for the general population). The mean of 64.1 was almost identical to that for a sample of 144 infants and toddlers with WS reported by Mervis and John (2010). Thus, the performance of the participants on the MSEL in the present study is consistent with that expected for young children with WS. Seven additional infants were excluded for the following reasons: fussiness ( $n = 1$ ), lost interest ( $n = 2$ ), distracted during test phase ( $n = 2$ ), outlier [attending the maximum trial length on 6 of 8 test trials ( $n = 1$ )], and computer error ( $n = 1$ ).

### 2.2. Materials

#### 2.2.1. Vocabulary assessment

The 396-word vocabulary checklist from the MacArthur-Bates Communicative Development Inventory: Word and Gestures (CDI-WG; Fenson et al., 2007), a parent report measure, was used to assess receptive and expressive vocabulary. The CDI-WG was normed for infants aged 8–18 months in 1-month intervals. Fenson et al. provide percentiles separately for receptive and expressive vocabulary sizes. Age equivalents were determined based on the vocabulary size at the 50th percentile for a given chronological age.

#### 2.2.2. Statistical learning stimuli

We used familiarization and test stimuli modeled on those used by Saffran et al. (1996, Experiment 2). The familiarization stimulus consisted of a 2-min stream of speech in an artificial language composed of four trisyllabic nonsense words (*pabiku/tibudo/golatu/tibudo/daropi/pabiku...*). The speech stream was spoken by a synthesized female voice and offered no acoustical cues to word

boundaries, such as pauses or stress. Each nonsense word was repeated 44–48 times in a pseudo-random order with the constraint that no item was repeated twice in succession. There were four test trial stimuli: 2 “words” (*pabiku, tibudo*) and 2 “part-words” (*pigola*, formed from the across-word sequence *daropi/golatu*, and *tudaro*, formed from the across-word sequence *golatu/daropi*). Within the words, the transitional probability from one syllable to the next was 1.0 at each syllable transition; the order was fully consistent. In contrast, within the part-words, the transitional probability from syllable 1 to syllable 2 was 0.33 and from syllable 2–3 was 1.0. Thus, the word and part-word test items differed in their statistical structure, but both occurred in the speech stream. In each of the test items, the word (or part-word) was spoken by the same synthesized voice repeatedly with a brief pause between each repetition of the word or part-word.

We made two slight modifications to the visual stimuli used by Saffran et al. (1996): (1) Instead of using a blinking light to get infants’ attention prior to familiarization and each test trial, a movie clip of a laughing baby was presented on the monitor in front of them. (2) A video of a rotating orange teardrop on a white background was presented throughout the entire familiarization phase and during each test trial.

### 2.3. Procedure

Each mother filled out the CDI-WG vocabulary checklist within one day of when her child completed the statistical learning procedure.

The procedure for the statistical learning task was similar to that used in Saffran et al. (1996, Experiment 2). In this task, participants were familiarized with 2 min of a continuous stream of speech made up of four three-syllable nonsense words presented repeatedly. The only cues to word boundaries were the probabilities with which syllables co-occurred, which were high within words and low between words. In the test phase, participants were presented with two kinds of three-syllable stimuli: “words” and “part-words.” The word stimuli were composed of syllables that consistently occurred together in sequence during familiarization. The part-word stimuli were composed of the final syllable of one word paired with the first two syllables of another word. Longer listening times to part-word test trials compared to word test trials would be taken as evidence that participants were sensitive to the statistical information in the speech stream.

For the present study, two modifications were made: (1) Both the familiarization and test auditory stimuli were presented from speakers in front of the infant. Thus, looking times were measured based on infants’ visual fixations toward the screen in front of them. (2) To give participants enough time to process the test stimuli, the maximum duration of each test trial was increased by 5–20 s.

At the time of testing, each participant was seated on an adult’s lap approximately 120 cm from a Panasonic 50” color plasma screen (1024 × 576 pixels) in a dimly lit room. The adult was instructed not to talk, point, or otherwise influence the infant. The adult, who was either an undergraduate student from another laboratory or a parent, was not aware of the research hypothesis. No data had to be excluded because an adult interfered with testing.

A closed circuit Canon VC-C50i camera hidden just below the center of the plasma screen was connected to a 15” JVC monitor in an adjacent control room, allowing the experimenter to view the infant and the direction of the infant’s eye gaze. Habit X software (Cohen, Atkinson, & Chaput, 2004) running on a Macintosh Power Mac G5 was used by the experimenter to control stimulus presentation and calculate listening times based on the experimenter’s key presses. The experimenter was unable to hear

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