

NOTE

**Asynchrony in the cognitive and lexical development
of young children with Williams syndrome***

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ABSTRACT

The present study investigates whether five-to-six-year-old children with Williams syndrome ($N=8$) can form new object categories based on naming information alone, and compares them with five groups of typically developing children aged 2;0 to 6;0 ($N=34$ children). Children were presented with triads of dissimilar objects; all objects in a triad were labelled, two of them with the same pseudoname. Name-based categorization was evaluated through object selection. Performance was above chance level for all groups. Performance reached a ceiling at about 4;0 for the typically developing children. For the children with Williams Syndrome, performance remained below chronological age level. The present results are discussed in light of previous findings of a failure to perform name-based categorization in younger children with Williams syndrome and the persistent asynchrony between cognitive and lexical development in this disorder.

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INTRODUCTION

In typical development, lexical acquisition begins before the end of the first year of life, as attested by the onset of word comprehension around 0;10, and that of production by 1;0 (Benedict, 1979; Bates, Bretherton & Snyder, 1988; Clark, 1993; Fenson, Dale, Reznick, Thal, Bates, Hartung, Pethick & Reilly, 1994; Hallé & de Boisson-Bardies, 1994). Within the timeframe of less than a year (around 1;6–1;8), infants become expert word learners. Important progress is made in their ability to correctly remember and represent the sound patterns of words and attach them appropriately to the representation of their correct referent (Gopnik & Meltzoff, 1992; Mervis & Bertrand, 1994; Jusczyk, 1997; Hollich, Hirsh-Pasek & Golinkoff, 2000; Werker, Fennell, Corcoran & Stager, 2002; Waxman, 2003). Recent evidence (Mervis & Bertrand, 1997; Stevens & Karmiloff-Smith, 1997; Mervis, Morris & Bertrand, 1999; Paterson, Brown, Gsoedl, Johnson & Karmiloff-Smith, 1999; Laing, Butterworth, Ansari, Gsödl, Longhi, Panagiotaki, Paterson & Karmiloff-Smith, 2002; Nazzi & Karmiloff-Smith, 2002; Vicari, Caselli, Gagliardi, Tonucci & Volterra, 2002; Nazzi, Paterson & Karmiloff-Smith, 2003; Volterra, Caselli, Capirci, Tonucci & Vicari, 2003) suggests that this developmental trajectory does not hold for children with Williams syndrome, a rare genetic disorder (1 in 20,000 live births, Beuren, 1972; Greenberg, 1990) caused by a hemizygous sub-microscopic deletion of some 25 contiguous genes on chromosome 7q11,23 (Ewart, Morris, Atkinson, Weishan, Sternes, Spallone, Stock, Leppert & Keating, 1993; Tassabehji, Metcalfe, Ferguson, Carette, Dore, Donnai, Read, Pröschel, Gutowski, Mao & Sheer, 1996; see full discussion of the syndrome in Donnai & Karmiloff-Smith, 2000). In the present study, we pursue the investigation of the early development of name-based categorization in this syndrome compared to typical development.

Although language (and vocabulary in particular) is a relative strength for adults with Williams syndrome, its onset is severely delayed in early childhood (Singer, Bellugi, Bates, Jones & Rossen, 1997; Paterson *et al.*, 1999), suggesting problems in mastering the basic skills involved in lexical acquisition. Importantly, lexical development in Williams syndrome seems to be characterized by the lack of some of the relationships between linguistic and cognitive milestones found in typical development. Infants with Williams syndrome start pointing at objects after, rather than before, starting to name them (Mervis & Bertrand, 1997; Laing *et al.*, 2002). The onset and acceleration of lexical acquisition in this syndrome seems to precede by many months, rather than follow, the emergence of various cognitive abilities and biases (including categorization abilities) typically involved in lexical acquisition (Stevens & Karmiloff-Smith, 1997; Mervis *et al.*, 1999). These findings suggest that lexical development in Williams syndrome is not just delayed, but might actually follow an atypical developmental trajectory (Karmiloff-Smith, 1998).

In this context, the link between vocabulary development and the emergence of name-based categorization in Williams syndrome was explored following work by Nazzi & Gopnik (2001) on typically developing children. These authors set out to determine the age at which typically developing infants, who have just been taught that two dissimilar-looking objects have the same name, will start grouping those two objects together. They found that this ability emerges between 1;4 and 1;8. Moreover, this emergence appeared to be linked to vocabulary growth, as suggested by the fact that: (a) at 1;8, name-based categorization (but not visual-based categorization) was correlated with vocabulary size, and (b) the infants aged 1;8 had significantly larger vocabularies (141 words) than those aged 1;4 (28 words). These findings thus suggest that early during lexical development, names start being attached to object categories rather than to individual objects. In other words, names start being taken as an indication of the conceptual categories to which objects belong. A recent replication of this study in French confirmed these initial results by replicating the link between the emergence of name-based categorization and vocabulary expansion over the same age period; these new results further showed that although the infants aged 1;4 failed name-based categorization, they had none the less learned the labels of the three objects (Nazzi & Pilardeau, 2003).

Nazzi & Karmiloff-Smith (2002) used the same methodology to investigate whether four-year-olds with Williams syndrome categorize objects based on visual and/or verbal cues, or whether, as found for other developmental links, vocabulary development would turn out to precede cognitive changes in this clinical population. The group of children with Williams syndrome appeared to be able to categorize objects according to visual cues, making almost no errors. However, they failed to use the verbal cues to categorize, in spite of being both chronologically much older (mean age: 4;9) as well as much more advanced in their vocabulary development (mean productive vocabulary: 504 words) than the typically developing infants aged 1;8 in the Nazzi & Gopnik (2001) study. These results suggested that, unlike for typical development, initial lexical acquisition in Williams syndrome is not linked to name-based categorization. They bring a further piece of evidence in support of the hypothesis that lexical development in this syndrome follows an atypical developmental trajectory, one that is shaped from the outset by different cognitive constraints.

The Nazzi & Karmiloff-Smith (2002) results on four-year-olds raise the question of whether individuals with Williams syndrome are ever able to use names to categorize objects, whether such development is merely delayed or fails to appear altogether. Therefore, the first goal of the present study was to test whether name-based categorization is present in somewhat older children with Williams syndrome, and if so, whether their performance reaches the level of typically-developing peers.

Our second goal was to collect cross-sectional data tracing the typical developmental trajectory of name-based categorization in toddlerhood and childhood. This was doubly motivated. First, although this ability emerges between 1;4 and 1;8, the performance of the infants aged 1;8 only reached 62.7% accuracy (Nazzi & Gopnik, 2001). Second, establishing the full typical developmental trajectory will give us more comparison points to evaluate the level of performance of the children with Williams syndrome. This trajectory approach offers an alternative to the more common practice of comparing children with Williams syndrome to typically developing controls matched on measures such as mental age (MA). It provides more insight with respect to the way in which development may have proceeded over time in a deviant fashion, even though the behavioural proficiency of the children with Williams syndrome, measured by matching controls at a specific chronological age (CA) or MA, may end up similar to that of typically developing children. These similar developmental outcomes may result from very different developmental routes, and the usual matching procedures may fail to capture this fact. Moreover, MA provides only a very general and composite measure, which conflates many different types of abilities (and often leads to discarding CA differences). Comparing developmental trajectories of specific skills thus allows us to make more fine-grained comparisons (see Thomas, Grant, Barham, Gsödl, Laing, Lakusta, Tyler, Grice, Paterson & Karmiloff-Smith, 2001, and Karmiloff-Smith, Thomas, Annaz, Humphreys, Ewing, Brace, van Duuren, Pike, Grice & Campbell, 2004, for a detailed discussion of this methodology).

METHOD

Participants

Eight children with Williams syndrome (4 males, 4 females) from monolingual English-speaking American families were recruited at the 2002 US Conference on Williams syndrome, Long Beach, CA. Their mean chronological age was 6;1 (individual ages: 4;10, 5;06, 5;10, 5;11, 6;01, 6;10, 6;11 and 6;11). One additional child refused to participate.

Five groups of typically-developing children from monolingual English-speaking American families (34 participants total) were recruited in the vicinity of the University of California at Berkeley. Their ages ranged from 2;0 (about the same age as the older infants in Nazzi & Gopnik, 2001) to 6;0 (the mean age of the children with Williams syndrome, at which age it was obvious that the task was trivial for typically developing children, see results below). Because the testing of the typically developing children was not designed to establish the existence of name-based categorization (already established in infants aged 1;8) but to trace the developmental trajectory of the evolution of performance with age, we set out to test about 6 to 8 children

per age group, which as we had estimated, turned out to be sufficient to obtain significant results for each age group. This sample of children consisted of 8 two-year-olds (4 males and 4 females), 9 three-year-olds (6 males and 3 females), 6 four-year-olds (3 males and 3 females), 6 five-year-olds (4 males and 2 females), and 5 six-year-olds (3 males and 2 females).

Stimuli

Six triads of small objects were used during the testing session (an additional triad being used during pretest). All objects were selected so that the infants would be unfamiliar with them and would not already have a name for them. All sets were made up of three very distinct objects, that all differed in shape, colour, and texture in an effort to equalize their perceptual distance.

Procedure

The procedure was identical to that used in Nazzi & Gopnik (2001) and Nazzi & Karmiloff-Smith (2002), with the exception that this time only naming trials were presented. Children were tested individually for about 10 minutes in a quiet room, in the presence of a caregiver.

After an informal warm-up period (which varied in duration and content according to the age of the participants), the child was seated on a chair across a table from the experimenter, and the testing session started. It comprised a training trial and 6 test trials. All trials were 'naming' trials that tested for categorization based on naming.

The training trial was identical to the test trials (see below) except that the presentation of the objects and the categorization question were repeated if the child's initial response was incorrect (although the child was not told the answer was incorrect). The testing phase started independently of the response provided the second time. The 2 nonwords used for the training trial were 'lep' and 'nim.'

Each test trial was composed of a presentation phase, followed by a categorization question. Each trial started with the presentation of the 3 objects, one at a time. The child was encouraged to manipulate each object for a few seconds, before placing it on the table. Within each trial, the objects were arranged on the table on a left-to-right sequence (child's perspective) in order to minimize memory load. The experimenter spoke while presenting each object, saying (for example): 'Look! A zab. This is a zab. Do you want to play with the zab? Yes, play with the zab. See this zab? All right, let's put the zab on the table. Here.' Each object was named exactly 6 times. We used three pairs of nonwords, 'douk'/'zab,' 'mora'/'pizer,' and 'nulis'/'kepod' (all three pairs were used once, in counterbalanced order, and then reused in the same order, the word of the pair being used as target being switched between the two occurrences).

After the presentation phase, the experimenter tested categorization by putting one object of the named pair in his own hand, placed at equal distance from the remaining two objects, and asking the child to give him 'the object that goes with this one.' While waiting for the response, the experimenter looked at either the child's face or the object in his hand in order to avoid influencing the child's response. After the child's response, positive feedback was provided regardless of the choice made. Successful performance corresponded to the selection of the similarly-labelled object. The order of presentation of the trials (for the first 3 trials, that order being then repeated for the last 3 trials), the position of the paired objects on the table, the side of the object picked up by the experimenter, and the pairs defined by the names were counterbalanced across participants.

Finally, note that no exact measure of productive vocabulary could be collected in the present study, because most participants were too old to be administered the MacArthur Communicative Development Inventory: Toddlers (Fenson *et al.*, 1994) as had been done in previous studies. The total number of words on that measure is 680 (plus 25 irregular word forms), reached by the time typically developing children are approximately 2;6. There are no equivalent measures of actual vocabulary size for older children, primarily because vocabularies become too large to be reliably estimated at the individual level. In our earlier study, many of the four-year-olds with Williams syndrome were at ceiling on this measure so we can presume that the present six-year-old participants' vocabulary was considerably higher. More significantly, other studies have shown that the vocabulary of older individuals with Williams Syndrome is often high, and sometimes even higher than typically developing children of the same chronological age (Grant, Karmiloff-Smith, Berthoud & Christophe, 1996; Grant, Karmiloff-Smith, Gathercole, Paterson, Howlin, Davies & Udwin, 1997; Paterson *et al.*, 1999; Grant, Valian & Karmiloff-Smith, 2002; Phillips, Jarrold, Baddeley, Grant & Karmiloff-Smith, 2004).

RESULTS

For each trial, children were given a score of 1 when the chosen object was the second of the named pair, and a score of 0 otherwise. Total scores could range from 0 to 6. Results are presented in Table 1 for the Williams syndrome group and the 5 groups of typically developing children. Figure 1 presents the data of the individual participants with Williams syndrome, compared to the data of the individual typically developing children together with the typical developmental trajectory (note that two data points are almost conflated on Figure 1 as the two Williams syndrome participants aged 6;11 had the same performance of 4 correct responses).

ASYNCHRONOUS DEVELOPMENT IN WILLIAMS SYNDROME

TABLE 1. Mean number of correct naming responses (and percent correct) for the Williams syndrome and the 5 groups of typically developing children (aged 2;0 to 6;0)

	Naming responses	S.D.	p values
Williams syndrome (M=6;1)	4.63 (77.1%)	0.74	<0.001
2-year-olds	3.75 (62.5%)	1.04	0.040
3-year-olds	4.44 (74.0%)	1.01	0.001
4-year-olds	5.5 (91.7%)	0.84	<0.001
5-year-olds	5.83 (97.2%)	0.41	<0.001
6-year-olds	6 (100%)	0	<0.001

p values: 1-tailed t-test comparisons to chance value (=3).

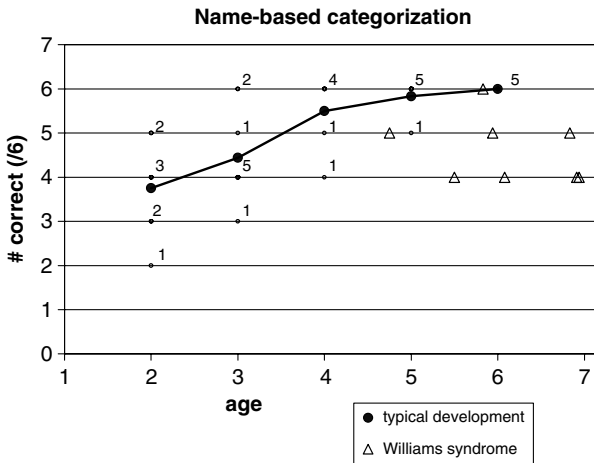


Fig. 1. Name-based categorization performance for individual participants with Williams syndrome (open triangles) and individual typically developing children (dots, the digits indicating the number of children having a given performance value at a given age). The solid line with solid circles indicates the typical developmental profile. The age of the infants is given by the horizontal axis.

An ANOVA on the categorization scores with the main effect of group (6 groups) revealed a significant effect of group, $F(5, 36) = 7.97, p < 0.001$. Comparisons of each group's performance to the chance value of 3 revealed that all groups were responding above chance level (see Table 1).

Post hoc analyses (LSD test) were conducted to specify the group effect (see Table 2). These analyses first show that performance for the typically developing infants increases significantly with age from 2;0 to 4;0, when children virtually stop making mistakes. Second, the comparisons involving

TABLE 2. Results of the post hoc analyses (LSD test) of the group effect: p-values

	WS	2 yrs	3 yrs	4 yrs	5 yrs	6 yrs
WS		0.039	0.651	0.055	0.009	0.005
2 yrs			0.088	0.000	0.000	0.000
3 yrs				0.019	0.003	0.002
4 yrs					0.483	0.318
5 yrs						0.738

the Williams syndrome group show that they perform significantly above the two-year-olds, and significantly below the four-, five-, and six-year-olds, while their performance ($M = 4.63$) cannot be distinguished from that of the three-year-olds ($M = 4.44$).

DISCUSSION

The present study first provides cross-sectional data specifying the typical developmental trajectory of name-based categorization. In previous research, this ability had been found to emerge between ages 1;4 and 1;8 (Nazzi & Gopnik, 2001). However, performance at 1;8 was only of 62.7% correct responses. It was therefore vital to chart the full developmental trajectory of this ability until ceiling was reached. The present results with two-to-six-year-olds confirm that young children use names to categorize objects: performance is above chance level for all age groups, and increases with age to achieve ceiling around 4;0.

The present study also brings new information regarding the ability of children with Williams syndrome to use linguistic names to categorize objects together. Our results show that as a group, the children perform significantly better than chance. Moreover, inter-individual variation was low, and for all children, the number of correct responses was superior to the number of incorrect responses. It thus appears that, unlike four-year-olds with Williams syndrome, five-to-six-year-olds with that disorder do finally display some ability to use names to group objects into categories. Taken together with Nazzi & Karmiloff-Smith (2002), the present study highlights developmental changes in the categorization abilities of children with Williams syndrome, in the form of the gradual emergence of name-based categorization.

The emergence of name-based categorization in Williams syndrome, however, remains very delayed compared to typical development, as it only seems to emerge between 3;0 to 4;0 and 5;0 to 6;0, as opposed to between 1;4 and 1;8 in typical development. This developmental delay is further attested by the fact that even though our participants with Williams

syndrome performed above chance level, their performance remained significantly below that of their chronological age controls. *Post hoc* analyses suggested that their performance at 6;0 is only at about the level of typically developing three-year-olds.

Finally, the emergence of name-based categorization is also delayed compared to word learning in Williams syndrome. In typically developing children, name-based categorization first emerged between 1;4 and 1;8, in concert with a marked increase in vocabulary, and at a time when overall vocabularies were typically less than 150 words. In contrast, combining the current study with our earlier results for children with Williams syndrome (Nazzi & Karmiloff-Smith, 2002), name-based categorization only emerged between 3;0 to 4;0 and 5;0 to 6;0, when they already have large vocabularies. Indeed, the three-to-four-year-olds with Williams syndrome tested by Nazzi & Karmiloff-Smith (2002) had a mean of over 500 words, so that the five-to-six-year-olds in the present study could be expected to have even higher vocabularies. Rather than emerging when children have small vocabularies (<150 words), this ability emerged in the children with Williams syndrome at a time when they were likely to have more than 500 words. The present study thus adds a new cognitive ability (name-based categorization) to the list of cognitive abilities (exhaustive categorization, ability to attach new names to unnamed objects, lexical constraints such as the whole object or the taxonomic constraints) whose emergence is thought to be linked to the acceleration of lexical acquisition around age 1;6 in typical development, but takes place later than the vocabulary spurt in Williams syndrome (Mervis & Bertrand, 1997; Stevens & Karmiloff-Smith, 1997; Mervis *et al.*, 1999; Laing *et al.*, 2002). The delay in the emergence of these cognitive abilities creates an asynchrony between cognitive and lexical developments in this syndrome, which makes it impossible for these cognitive abilities to be used for early lexical acquisition (before, e.g., 5;0 to 6;0 for name-based categorization).

This lack of synchrony in the relative timing of the emergence of these various cognitive and lexical abilities, which is consistent with the proposal that lexical acquisition in Williams syndrome follows an atypical trajectory, raises the question of the nature of the early lexical acquisition mechanisms used by these infants. A proposal found in the literature, based on adult data (Vicari, Brizzolara, Carlesimo, Pezzini & Volterra, 1996; Vicari, Carlesimo, Brizzolara & Pezzini, 1996; Grant *et al.*, 1997; Thomas *et al.*, 2001), is that individuals with Williams syndrome acquire words by relying on proficient phonological/speech perception abilities. In a first attempt to test this proposal in infants, Nazzi *et al.* (2003) found that one-to-four-year-olds with Williams syndrome were unable to segment from fluent speech some word patterns (iambic bisyllabic words) that start being segmented as early as 0;10 in typical development (Jusczyk, Houston & Newsome, 1999). The

identification of these early speech perception difficulties contradicts the hypothesis that lexical development in Williams syndrome relies on proficient phonological/speech perception. At present, the early mechanisms that are actually used to learn words in Williams syndrome remain to be identified and we believe that a focus on infants and toddlers is the best way to approach this question.

In summary, we now have evidence suggesting that name-based categorization emerges in Williams syndrome between 3;0 to 4;0 and 5;0 to 6;0, i.e. much later than in typical development in which we found almost no errors beyond the age of 4;0. This confirms that name-based categorization is yet another example of a linguistic task on which infants and children with Williams syndrome are not, contrary to earlier claims, spared. This delayed emergence and particularly its lack of synchronization with lexical acquisition further suggests that early lexical acquisition in Williams syndrome follows an atypical developmental trajectory in which asynchrony may play a vital causal role.

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