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Parallels between spatial cognition and spatial language: Evidence from Williams syndrome

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Abstract

Does the acquisition of spatial language always reflect the characteristics of non-linguistic spatial representation? We explored this question by examining spatial representation and spatial language among children and adults with Williams syndrome, a rare genetic syndrome that gives rise to a pattern of severe spatial impairment together with preserved language. The results of three experiments showed striking evidence for the preservation of axial reference systems together with fragility in representing direction within axes, in both non-linguistic judgments of object location and naming of those locations. These basic properties of the spatial representational systems were observed among WS children and adults and normally developing children, although the WS individuals exhibited noisier performance in both domains. The results indicate that non-linguistic spatial representations and spatial language share structure, even in cases of severe spatial impairment.

Keywords: Spatial representation; Language; Memory; Axes; Reference systems; Williams syndrome

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Introduction

The mental representation of spatial location is one of the most important and ubiquitous functions carried out in human cognition. It is crucial to perception, action, and language, as it provides the core of representations that support our capacity to localize and track objects over time, carry out actions on stable and moving objects, and talk about the identities and locations of objects relative to each other. Perhaps most remarkable about these functions in humans is that systems whose internal structures are radically different—such as visual perception and language—nevertheless can "talk to each other," yielding the powerful capacity to talk about what we see.

This paper explores the nature of this mapping by asking how closely tuned these two systems of spatial representation must be in order to allow conversion from what we see to what we say. We present the case of people with Williams syndrome, who have severely impaired spatial cognition together with relatively spared language, and we ask whether, despite this imbalance, there are architectural parallels between their spatial cognition and spatial language. A strong test of parallel structures should show similar architectural properties even in people who have severe spatial impairment. Moreover, if there is tight yoking between the systems, then breakdown in the non-linguistic system

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should be mirrored in spatial language. Finally, because Williams syndrome is a developmental deficit, parallels and differences in the two systems should help us to understand the extent to which the acquisition of spatial language is built upon a foundation of non-linguistic spatial cognitive structures.

To examine these issues, we focus on a core aspect of spatial representation—the reference system—which is engaged at multiple levels and by multiple spatial systems in the mind and brain (Andersen, Snyder, Bradley, & Xing, 1997; Colby & Goldberg, 1999; Landau, 2002; Logan & Sadler, 1996; McCloskey, 2001). We examine the nature of sparing and breakdown in non-linguistic representations of reference systems, and then ask what parallels and divergences, if any, are found in spatial language.

Reference systems in spatial cognition and spatial language

Humans and other mobile animals represent the locations of objects in terms of one or more reference systems which specify the location of the target object relative to some other point or points in space. Within the literature on spatial vision and spatial cognition, it is often assumed that these reference systems are represented as a set of orthogonal axes whose origin can be the retina, head, body, or other points, objects, or arrays in space (Behrmann, 2000; Colby & Goldberg, 1999; Landau, 2002; McCloskey, 2001). Because different tasks—such as pointing vs looking—can engage different reference systems, much of our everyday spatial interaction involves multiple representations of space, which must be coordinated in order for us to reach for what we see, talk about what we perceive, and so on.

Given this, it is not surprising that deficits in spatial cognition can often be traced to deficits in the ability to create and/or use reference systems. Studies of brain-damaged adults reveal that malfunctioning can occur in object-centered, ego-centered, environment-centered, and even attentional-centered reference systems (Behrmann, 2000; McCloskey & Rapp, 2000). Breakdown can also occur in the sub-components of a reference system. For example, McCloskey and Rapp (2000) described a patient whose deficit was traced to impaired direction within each axis, but not the axes themselves. Finally, reference systems can break down along lines corresponding to functional divisions within spatial cognition. For example, Milner and Goodale (1995) reported a patient who could accurately act on space, adjusting grasp for different sized objects or inserting objects in a slot, but was unable to make accurate perceptual judgments of object size or slot orientation. This separation of vision for the purposes of action vs perception has also been shown in normal adults (Bridgeman, Gemmer, Forsman, & Huemer, 2000). As a whole, the evidence shows that reference systems are highly articulated, and that they are functionally separable within the larger system of spatial cognition.

Like non-linguistic representation of location, spatial language engages multiple types of reference systems. For example, in English, the terms "top/bottom/front/back/side" engage an object-centered reference system which is stable over changes in the object's location or orientation. A different set of terms engages the geocentric reference system (e.g., "east" or "west"), and still another set of terms, including "above," "below," "left," and "right," ordinarily engages a frame of reference which is centered on another object or a part of the environment (Carlson-Radvansky & Irwin, 1993). Thus, although language is itself a highly specialized system with its own formal rules, talking about spatial location appears to be grounded by engaging frames of reference that also exist for other, non-linguistic purposes.

These parallels between non-linguistic representations of space and spatial language suggest that the two systems are homologous and at least partially yoked, and that non-linguistic representations might serve as a force shaping the nature of spatial language (Clark, 1980, 1973; Jackendoff, 1983; Landau & Jackendoff, 1993; Talmy, 1983; and see also Crawford, Regier, & Huttenlocher, 2000, although with a somewhat different interpretation). For example, Clark (1973) suggested that the importance of gravitational upright in our lives makes it plausible that linguistic terms representing the vertical axis—and particularly, vertical upright—might be privileged.

Empirical evidence for a homology between the two systems was first presented in studies by Hayward and Tarr (1995), who developed a pair of tasks designed to examine the structure of reference systems when they are engaged by perception/memory, and language. In these tasks, adult English speakers were asked to label an object's location in a small array and their responses were compared to their accuracy in remembering those same locations. Hayward and Tarr found that locations which were most consistently named by the basic spatial terms of English (e.g., above, below, right, left) were also those that were remembered best in non-linguistic memory tasks. These best-named and best-remembered locations were those that fell along the extensions of a reference object's main axes. The findings suggested to Hayward and Tarr that non-linguistic spatial representations—in particular, reference systems that are mentally imposed on a reference object—serve as an organizational basis for spatial language. The basic pattern of results has been replicated cross-linguistically (Munnich, Landau, & Dosher, 2001), showing that the use of reference systems in non-linguistic and linguistic tasks is robust over languages.

However, the yoking between the systems is not perfect. Indeed, the mature use of spatial terms actually shows some degree of separation from corresponding non-linguistic representations. For example, although terms such as ''above'' and ''below'' map to the axes of a reference object, such terms are categorical, hence they are insensitive to specific metric properties such as exact angle or distance (Landau & Jackendoff, 1993; Talmy, 1983). Consistent with this, Hayward and Tarr found that people's use of spatial terms was categorical over the distances they tested. However, they also found that people's memory for location showed *graded* effects of distance, suggesting that the parallel between language and non-linguistic systems is only partial (see also Munnich et al., 2001). Other studies suggest that prototypes in spatial language and spatial memory may at times engage quite different structures that vary depending on the cognitive requirements of a task (Crawford et al., 2000; Huttenlocher, Hedges, & Duncan, 1991; Kemmerer, 1999).

Also consistent with differences between the systems, some aspects of spatial language may be acquired without grounding in non-linguistic spatial representation. For example, the fact that ''above'' and ''below'' are antonyms is derivable from distributional properties of the two terms, and children learn that the terms are opposites before they learn the complete correct meanings of the terms (Clark, 1972). Similarly, the difference between ''between'' and ''among'' reflects the number of arguments that the spatial term takes (two vs more than two), and so could be acquired through syntactic evidence without spatial grounding. Thus, although developmental theories have typically assumed strong yoking between spatial language and non-linguistic spatial knowledge (Clark, 1973; Johnston & Slobin, 1980; Nelson, 1974), some aspects of spatial semantics might be acquired and used without reference to any specific spatial content.

In the context of these complexities, one might wonder how strong the yoking between systems must be in order to support the acquisition and use of spatial terms. One way of evaluating this issue is to examine the consequences of spatial breakdown for the acquisition of spatial language. If the two systems are strongly yoked, then we might expect breakdown in non-linguistic reference systems to entail parallel breakdown in spatial language. Alternatively, weaker or non-existent yoking would result in quite different patterns of performance between tasks tapping the two systems. We aim to address this possibility by examining spatial cognition and language in people with Williams syndrome.

The relevance of Williams syndrome

Williams syndrome is a rare (1 in 15,000) genetic defect caused by a microdeletion of material on chromosome 7. The deleted area contains the genes for elastin (ELN), Lim-Kinase (LIM-K1), and others (Frangiskakis et al., 1996; Morris et al., 1994). The syndrome has a typical phenotype, including a characteristic facial profile, various disorders of the heart and viscera, and mild to moderate retardation. Of most significance to cognitive scientists, however, is the syndrome's strikingly uneven cognitive profile: Individuals with WS exhibit profound spatial deficits together with relatively spared language.

At present, the nature of the spatial deficit is not well understood. Even within the broad domain of spatial cognition, there is unevenness, with relatively spared face recognition (Bellugi, Sabo, & Vaid, 1988; Tager-Flusberg, Plesa-Skwerer, Faja, & Joseph, 2003), object recognition (Landau, Hoffman, & Kurz, 2005; Wang, Doherty, Rourke, & Bellugi, 1995), perception of biological motion (Jordan, Reiss, Hoffman, & Landau, 2002), and some aspects of spatial language (Lakusta & Landau, 2005; Landau & Zukowski, 2003). However, there is severely impaired performance on so-called ''visuospatial-constructive'' tasks (Bellugi et al., 1988; Hoffman, Landau, & Pagani, 2003; Mervis, Morris, Bertrand, & Robinson, 1999). These tasks require a person to copy an existing design, either by drawing (Beery & Buktenica, 1967) or by assembling blocks (Elliot, 1990). Individuals with WS typically perform in the 1st percentile for their age group, with adolescents performing roughly at the level of normal 4-year-olds (Bellugi et al., 1988; Hoffman et al., 2003; Mervis et al., 1999; see Fig. 1 for some samples of copying).

The severe impairments in drawing and copying are notable because success in these tasks would seem to rest on the capacity to locate multiple objects within a common frame of reference, and to use a parallel frame of reference to assemble the parts in the copying space. The distorted copies typical of individuals with Williams syndrome could be symptomatic of their inability to mentally impose reference systems on the original array, or to construct new, analogous reference systems in the separate copying space. Severe impairment in the construction or use of such reference systems would no doubt impair many spatial capacities, including perception and action, and might also affect those aspects of spatial language which engage reference systems. Recent studies of WS toddlers suggest either delay or outright deficit in the development of reference systems used to guide visual attention to objects (Brown et al., 2003). Their later-emerging deficit in spatial construction tasks could reflect lack of basic structures, i.e., reference systems that organize and guide the ability to carry out these tasks.

The stark contrast between the spatial deficit of WS individuals and their relatively preserved language capacity raises an intriguing question: How might spatial language emerge, given the severe impairment in the supporting system of spatial representation? One possibility is that spatial language emerges relatively independent of corresponding spatial representations; this would suggest surprising independence of the two

Fig. 1. Sample drawings from two children with Williams syndrome, and one normally developing child matched for mental age.

knowledge systems, consistent with the idea that spatial representation and language are independent and possibly modular (Bellugi et al., 1988). However, it is also possible—even likely—that spatial language will rest on spatial representations even if they are impaired. In this case, the question is whether the two systems—linguistic and non-linguistic—will give rise to the same patterns of sparing and deficit. The answer will help us to understand how the two systems map to each other during development, and the degree to which they are yoked.

While the general profile of strength in language and deficit in spatial cognition suggests independence, it is far from conclusive. One of the principal problems is that tasks in the two domains naturally have drawn on quite different stimuli and methods, and therefore have often tapped into aspects of cognition that would be unlikely to show any relationship even if the two systems were strong coupled. For example, language tasks have tapped knowledge of syntactic form and morphology (Bellugi et al., 1988; Clahsen & Almazan, 1998; Karmiloff-Smith et al., 1997), whereas tasks showing the characteristic spatial deficit have tapped the ability to reconstruct spatial arrays (Bellugi et al., 1988; Hoffman et al., 2003; Mervis et al., 1999). While comparisons across such tasks might give insight into the developmental time courses of the systems as a whole, they cannot tell us the extent to which the two systems interact in development. The strongest evidence on mapping between the two systems would be offered by examining the same spatial organization when it is recruited over the two domains.

In the following experiments, we built on Hayward and Tarr's experiments, asking whether WS individuals show any structured representations of reference systems in non-linguistic tasks, whether these are similar to the structure emerging from spatial language tasks, and whether in either case, there is systematic breakdown that can shed light on the spatial deficit. Because Hayward and Tarr's method can reveal the mental representation of spatial structures in both memory and in language, it provides a good vehicle for examining both modes of spatial representation among WS individuals. The focus on reference systems is especially compelling in the case of WS, because their spatial impairment suggests difficulties encoding the locations of objects relative to each other. If there is severe impairment in the recruitment of reference systems, this would be an important clue to the nature of the spatial breakdown. At the same time, any breakdown observed in non-linguistic tasks can be compared to that for language tasks, providing us with evidence for sites of interaction and independence.

Experiment 1

Participants

Ten children with Williams syndrome between the ages of 8 and 14 years (M age = 10 years, 4 months, range = 8;1–14;0) participated, along with 10 normally developing children who were matched to the WS children on the basis of mental age. The normal children were between the ages of 3 and 6 (M age = 5;5 range = 3;10–6;11). The children were matched using the Kaufman Brief Intelligence Test (KBIT, Kaufman & Kaufman, 1990), which yields an overall IQ scores as well as scores for two components, Verbal and Matrices. The Verbal component requires the child to name a series of objects portrayed by black and white line drawings. The Matrices component taps conceptual abilities, requiring judgments of which pairs of objects "go together"; but there are very few items that require spatial representation. Using this measure, the children with WS are not penalized for their spatial deficit, and therefore their scores represent a fair measure of non-verbal (but non-spatial) intelligence. The score of the WS children on both components was well matched to those of the controls (*M* Verbal scores = 32, 30.6, *SE*s = 2.3, 2.5, ranges = 23–46, 20–45, respectively; *M* Matrices scores = 18.7, 18.5, *SE*s = 1.2, 1.1, ranges = 12–24, 12–22, respectively). The corresponding composite IQ scores, which reflect both raw (matched) scores and chronological age, were *M* = 74 (*SE* = 3.9, range = 55–92) and 112 (*SE* = 2.7, range = 98–123), respectively. Obviously, the IQ scores of the WS children are quite low for their chronological age, as is typical of this population (e.g., Bellugi, Bihrle, Neville, Doherty, & Jernigan, 1992; Mervis et al., 1999). This is why children were matched to normally developing controls on mental age (or raw score), not IQ.

As part of a larger research program, the children were also tested on the Pattern Construction sub-test of the Differential Abilities Scale (Elliot, 1990). This test requires the child to reconstruct a complex design composed of individual blocks and is therefore widely regarded as a hallmark task for diagnosing the spatial deficit in WS (see, e.g., Mervis et al., 1999). Of the 10 children in this study, nine scored in the 1st percentile for their age, and one performed at the 4th percentile; the mean age equivalent for the group was 4 years, 2 months. These scores are similar to those reported in other studies of individuals with WS (Bellugi et al., 1992; Mervis et al., 1999), and indicate severely impaired spatial cognition. The mean scores of the normally developing children fell at the 54th percentile, with a mean age equivalent of 5 years 4 months. Thus, on this test, which clearly marks the characteristic profile of Williams syndrome, the children with Williams syndrome performed very poorly, and even more poorly than the normally developing children who were matched for mental age on other measures.

The children with Williams syndrome were recruited with the assistance of the National Williams syndrome Association and the A.I. DuPont Hospital for Children. They were all diagnosed by a geneticist and a positive FSH test. They all lived within a $2\frac{1}{2}$ h travel range of the University of Delaware. Normally developing control children were recruited from parent groups and preschools local to the University of Delaware.

Design and methods

The design of the study was adapted from Hayward and Tarr (1995) to accommodate young children. Children viewed a sheet of paper (8.5 × 11 in.) which contained a square Reference object (1.5 in. × 1.5 in.), and a 1/2 in. dot that was located in one of 36 places on or around the square (see Fig. 2, top panel). There were no grids on the page, so location could not be determined by alignment with any explicit gridlines. The children's attention was drawn to this Standard array, and they were told "See this square? It's right here. And see this dot? It's right here" (as the experimenter pointed to each). Then they were shown a pair of Test arrays, one the Same as the Standard, and one Different from it, located side by side and below the square (Fig. 2, bottom panels). They were asked to point to the test array that had the square and the dot in "just the same place"

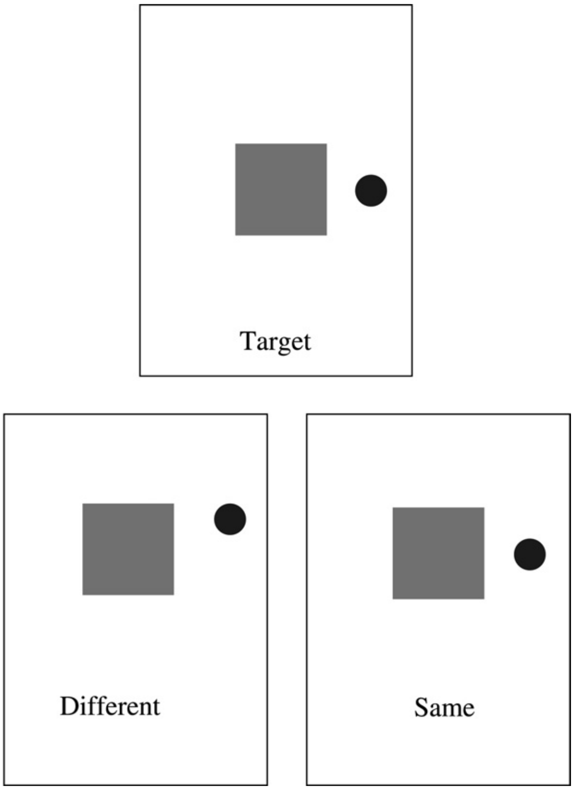

Fig. 2. Layout of test stimuli in Experiment 1. People viewed the top display and were asked to select the panel in the bottom display that showed the dot "in the same place" relative to the square reference object.

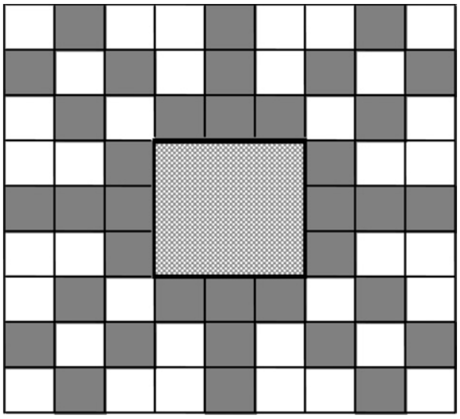

Fig. 3. Locations tested in Experiment 1. Dark boxes indicate target locations around the square. Grid lines were not present in the displays used for testing.

as in the Sample. After they pointed to one of the Test arrays, the page was turned and the next trial began.

The locations of the target dots were determined by centering a grid on the Reference object, and selecting 36 locations that were evenly distributed around it (see Fig. 3). These included (a) 12 Contact locations, adjacent to the square (three each on its top, bottom, right, and left sides), (b) eight On-Axis locations that fell on the extension of the square's principal axes at each of two distances (two each above, below, right, and left of the square, at one and two grid marks away, respectively), and (c) 16 Off-Axis locations that fell off of the square's axes, at each of two different distances (two each in the upper left, upper right, lower left, and lower right, at one and two grid marks away, respectively). Given these locations, we could examine whether the children's representation of location varied across the space. In particular, we could examine (a) any advantage for locations On-Axis compared to Off-Axis, (b) any effects of vertical vs horizontal axis or direction within axis (above or below, right or left), and (c) any effects of distance from the reference object (contact, 1, or 2 gridmarks away from it).

Each of the 36 test locations served as the Standard location on four trials, yielding a total of 144 trials. For each of the Standard locations, there were two test items, a Same array, and a Different array. The Different arrays were identical to the Standard except that the dot was displaced from its original location by 1/2 in. in each of four different directions. For target locations that were not adjacent to the square, these included two displacements diagonally upward to the left and right and two diagonally downward to the left and right. For targets that were adjacent to the square, these included the two diagonal displacements upward plus two displacements that were along the horizontal or vertical border of the square. The 144 trials were presented in random order, and left-right position of the Same and Different arrays was randomly determined and counterbalanced. The Standards and Test arrays were assembled into a loose-leaf book. After each trial, the page was turned, and the next Standard and Same/Different test pair was shown. All sessions were videotaped, and the children's responses were recorded later.

To determine whether any results were due to the absolute sizes of the arrays, a second version of the experiment was carried out in which all stimuli were reduced in size by 50%, and presented on the same size sheets and in the same loose-leaf books as the original. This version was carried out with all of the same children except one child with WS and one control, at some time after the initial study, but still during the same time frame indicated above. As will be shown, the results of the two versions were remarkably similar.

Results

Percents correct were analyzed to determine whether there were effects of (a) axial structure (on vs off axis), (b) vertical vs horizontal axis and direction within axis (V+ or "above," V− or "below," and Left/Right), and (c) distance from the reference object. The means used in these analyses are shown in Fig. 4 for each cell. As is evident from the locations of the darkest regions (those with highest proportions correct), there was a distinct advantage among both groups for locations lying on the extension of the reference object's axis. However, the children with WS showed poorer overall performance than controls and this became more apparent for locations that were farther from the reference object. It is almost as if the WS children represent "truncated" axes.

Preliminary analyses on the data for the 12 Contact locations revealed no differences over the three locations tested for each side, so these data were collapsed to yield a single score for contact with each side. Similarly, analyses comparing the different sides of the Off-Axis locations (i.e., top right/top left/bottom right/bottom left) showed no effects of side, so these scores were collapsed to yield a single score for each distance (1 vs 2 grid marks away from the reference object, as shown in Fig. 4).

Two principal analyses were carried out. The first examined whether there was an advantage to locations On vs Off the axes. In order to examine this issue conservatively, Contact locations were omitted from analysis, leaving only locations that were at some distance from the Reference object. In addition, for this analysis, On-Axis locations were collapsed over vertical and horizontal axes. Percents correct for these locations, shown in Table 1, were entered into a 2 (Group) × 2 (On vs Off-Axis) × 2 (Distance: one vs two grid squares from the reference object) analysis of variance. The results showed main effects of all three variables. Children in

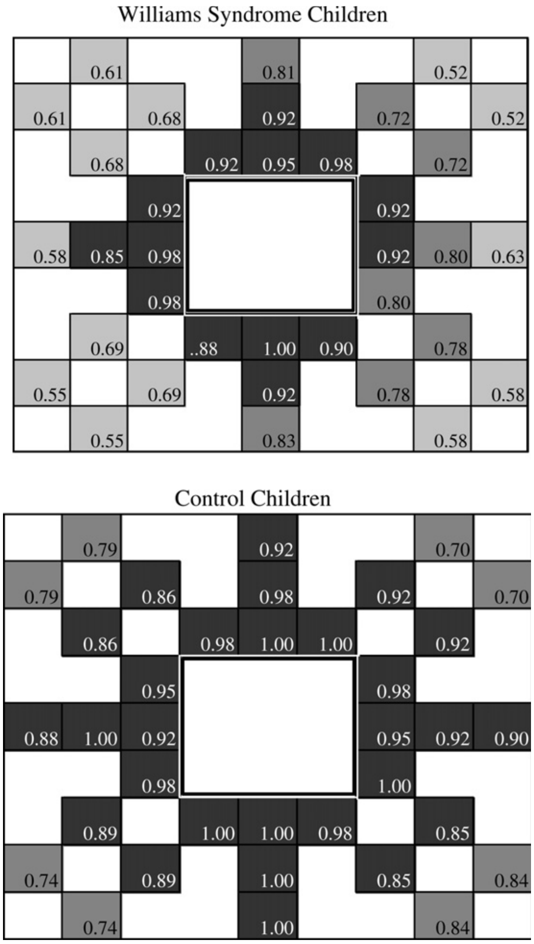

Fig. 4. Mean proportions correct for locations tested in Experiment 1.

Table 1
Proportions correct for On vs Off-Axis locations (Experiment 1)

Location:	On-Axis			Off-Axis		
Distance:	1	2	*M*	1	2	*M*
WS	.87	.71	**.79**	.72	.56	**.64**
CC	.97	.92	**.95**	.88	.77	**.83**

Table 2
Proportions correct for Contact and On-Axis locations (Experiment 1)

Location:	Contact			On-Axis					
	V	H	*M*	V			H		
Distance:				1	2	*M*	1	2	*M*
WS	.93	.92	**.93**	.92	.82	**.87**	.82	.60	**.71**
CC	.99	.96	**.98**	.99	.96	**.97**	.96	.89	**.93**

both groups performed better when the dot was located On-Axis than Off-Axis (F 1, 18 = 43.3, $p < .01$; $M = .87$, .73, $SE = .02$, .02), showing that they used the reference object's projected axes as organizing structures. They also performed better when the dot was close to the reference object (1 grid mark away) than when it was farther away (2 grid marks) (F 1, 18 = 48.4, $p < .01$; $M = .86$, .74, $SE = .02$, .03). However, the children with WS performed more poorly overall than the control children (F 1, 18 = 18.8, $p < .01$; $M = .72$, .89, respectively, $SE = .03$, .02), and their performance declined more sharply over distance, resulting in a Group by Distance interaction (F 1, 18 = 4.8, $p < .05$). Post hoc tests showed that the difference in performance for the two distances was reliably greater for the WS than the control children (Tukey's hsd = .05, $p < .05$). Part of the reason for this interaction could be the likely ceiling effect among control children.

The second analysis examined whether there were any effects *within* On-Axis locations (see Table 2). This time, Contact locations were included, in order to allow comparison across sides at three distances. We examined effects of the vertical vs horizontal axis, effects of direction within axis, and effects of distance within the axes. A 2 (Group) × 4 (Axes and Direction: above/below/left/right) × 3 (Distance: Contact, one, two grid squares away) analysis of variance resulted in main effects of all three variables. Children in both groups performed better for locations along the vertical axis (M above = .93, below = .94) than locations along the horizontal axis (M right = .85, left = .86) (F 3, 54 = 11.8, $p < .01$, Tukey's hsd = .04, $p < .05$). There were no differences due to the region's direction *within* each axis (i.e., the region "above" was no different from the region "below" the reference object, nor "left" vs "right," shown collapsed in the table). The children also performed better when the dot was in contact with the grid than when it was 1 grid mark away, and better when it was 1 grid mark away than 2 grid marks away (F 2, 36 = 19.5, $p < .01$; $M = .95$, .92, .82, respectively; Tukey's hsd = .05, $p < .05$).

Again, however, the children with WS performed more poorly overall than the control children (F 1, 18 = 10.7, $p < .01$; $M = 84$, 96, respectively; $SE = .02$, .01), and their performance declined more sharply than controls for horizontal locations and for locations at increasing distances from the reference object. The Group × Axis interaction ($F = 2.6$, $df = 3, 54$, $p = .06$) showed that the WS children's performance was not different from controls for above or below locations, but was worse than controls for both left and right (Tukey's hsd = .13, $p < .05$). The Group × Distance interaction (F 2, 36 = 7, $p < .01$) showed no difference between groups for contact, but WS children were worse than controls at both 1 and 2 grid marks away from the reference object (Tukey's hsd = .09, $p < .05$). The results confirm the impression of truncated axes, more pronounced for the horizontal axis.

As indicated in Design and methods, a second version of this task was carried out, in which all stimuli were reduced in size by 50% and presented on the same sized sheets as the originals. The same analyses as described above were carried out on these data, and the same effects emerged.[1] Thus, the findings of the original study were not due to the specific absolute sizes of the stimulus displays, and instead, reflect aspects of the children's mental representations of location.

Discussion

The results revealed several properties of the children's spatial representations. First, both children with WS and normal children showed an advantage for locations that fall along the extension of the reference object's axis, compared to locations that are off these axes. This indicates that both groups of children represent location in terms of a set of axes centered on the reference object. Locations along these axes appear to be privileged, just as they are in normal adult representations of location using similar methods (Hayward & Tarr, 1995). Both groups of children also showed an advantage for the vertical axis (compared to the horizontal), and for locations that were closer to the reference object. Neither group of children showed any advantage for regions laying at specific directions within each axis, indicating no better performance between locations "above" vs those that were "below" the reference object, nor "left" vs "right."

However, there were differences between groups, which appear to reflect breakdown on the part of children with WS for locations more distant from the reference object, and especially along the horizontal axis. Although the WS children clearly have stable reference systems for regions of space that are represented close to the reference object, the pattern of relative weakness suggests that their stably represented space may be truncated. The existence of reference frames in children with Williams syndrome is surprising, in view of the severe spatial deficits typical of this population, and the findings suggest that the spatial deficit cannot be attributed to complete absence of these mental structures.

The sparing of these reference systems—fragile as they might be—raises the question of whether they support spatial language. To the extent that they do, we would expect similar organization of axial structure to emerge in studies of spatial language—both in normal children and in children with Williams syndrome. Yoked effects would also predict fragility among WS children, specifically, overall poorer performance, disproportionate weakness along the horizontal axis, and/or fragility at locations more distant from the reference object. Non-yoked effects might emerge in a variety of ways. Spatial language might be completely spared, with no fragility analogous to that we have seen in the matching task. Or, spatial language might be much more impaired, since learning spatial terms requires the additional step of figuring out which spatial terms map onto which regions. Given these extra steps, we might expect additional disruptions in spatial language, such as lack of axial structure for axial terms, impairment in directional representation, or gross errors and confusions in the mapping.

Experiment 2 addresses these possibilities by testing children's capacity to produce and comprehend a variety of spatial terms that normally apply to the regions we have considered so far. In addition, this experiment stretched the developmental possibilities by testing adults with Williams syndrome, who are known to have quite extensive productive vocabularies—ones that largely outstrip many of their non-linguistic spatial capacities (Bellugi et al., 1988; Mervis et al., 1999). Given that WS adults' vocabularies are on average better than would be predicted by mental age, we might predict that their control of spatial terms should also outstrip the performance expected for their non-verbal mental age. If so, this might suggest that spatial term learning can proceed without parallel advances in non-linguistic spatial cognition. Alternatively, if limits in spatial vocabulary are imposed by limits in non-linguistic

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[1] The 50% reduction in the size of the stimuli for the Matching task showed the following effects, which mirrored those of the full-size test. The analyses comparing locations On-Axis to locations Off-Axis also showed the same effects as for the 100% size stimuli. There were reliable effects of Axis ($F = 61.3$, $df = 1, 16$, $p < .01$), with children performing better for locations On-Axis than Off-Axis, Distance ($F = 46.82$, $df = 1, 16$, $p < .01$), with children performing better for locations one grid mark away from the reference object than two grid marks away, and Group ($F = 12.62$, $df = 1, 16$, $p < .01$), with the WS children performing worse than controls. There was also an interaction of Distance by Group ($F = 5$, $df = 1, 16$, $p < .05$), showing that the WS children were more negatively affected than controls by locations farther away from the reference object.In the analysis of accuracy *within* the locations On-Axis, there were effects of Axis ($F = 6.27$, $df = 3, 48$, $p < .01$), with children performing better along the vertical than horizontal axis, Distance ($F = 14.55$, $df = 2, 32$, $p < .01$) with children performing better at locations closer to the reference object, and Group ($F = 4.6$, $df = 1, 16$, $p < .05$) with the WS children performing worse than controls. There was also a reliable interaction between Group and Distance ($F = 4.76$, $df = 2, 32$, $p < .05$), with WS children worse at locations farther from the reference object, and a marginally reliable interaction of Group and Axis ($F = 2.14$, $df = 3, 48$, $p = .10$), with WS children performing worse than controls for left/right, but not above/below. Overall, these results replicate the findings of the original (100% stimuli) study, and show that the deficits observed in the WS children were not due to some lower level aspect of visual processing, which might be responsive to absolute size or distance from the reference object.

representation of space, spatial language might be no better than would be expected based on non-linguistic measures of mental age.

Experiment 2

Participants

Child participants were the same as those in Experiment 1. In addition we tested 13 adolescents and adults (henceforth "adults") with Williams syndrome (M age $= 21$ years, range $= 14$–30). Their scores on the Verbal portion of the KBIT were substantially higher than the WS and control children (Adult $M\text{s} = 47.9$, range $= 21$–64, $SE = 3.1$), and their Matrices scores were also somewhat higher ($M = 21.7$, range $= 17$–29, $SE = 1.3$). The adults had reliably higher Verbal scores than each group of children ($F\ 2,30 = 13.8$, $p < .01$, Scheffe post hoc tests, $p < .01$), but did not differ from the children on the Matrices measures ($F\ 2,30 = 2.5$, $p = .09$). Hence the WS adults were comparable on non-verbal measures to both the WS and normal children, but were quite a bit stronger than these groups on the verbal measure. The composite (IQ) scores of the adults were typical of people with WS ($M = 67$, $SE = 4.3$).

Children were tested in a Production and Comprehension task after Experiment 1, and during 1–2 sessions that were held 1–3 months apart, coinciding with the children's regular visits to our lab. The adults were recruited from the Williams syndrome Association, and were tested in both tasks at the Association's biennial meeting. These participants did not carry out the axes task of Experiment 1, but they did carry out a related task which allowed us to address specific questions about their non-linguistic spatial representations (see Experiment 3). All participants carried out the Production task before the Comprehension task, to prevent them from hearing any terms that they might not use on their own.

Experiment 2A: Production task

Design and procedures

Using the same arrays that had been constructed for the Location Matching task of Experiment 1, people were shown each of the 36 Target arrays (without the Test arrays), and were given a cloze test to elicit a spatial term. Specifically, they were told, "See this dot? See this square? The dot is ______ the square?" (see Fig. 2). Prior to these instructions, the participants were told that we were going to talk about "where things are." They were shown several simple examples with objects, such as a cup on a table, and said "See this cup? This cup is where? It is … the table?" People had no trouble with the task, producing appropriate terms for each example. The 36 Target locations used in Experiment 1 were each presented for naming once, in randomized order and on separate pages of a book.

Normal adults typically use more vertical terms along the vertical axis than in Off-Axis locations, and more horizontal terms along the horizontal axis than in Off-Axis locations. They also use directionally appropriate terms within each axis (i.e., appropriate contrasts of above/below and right/left; Crawford et al., 2000; Hayward & Tarr, 1995; Munnich et al., 2001). Our question was whether children with WS would show any indication of such normal structure, and if not, how spatial breakdown would be manifested. Furthermore, we asked whether any breakdown would persist into adulthood.

Results

The productions were fully transcribed and coded, and then analyzed in much the same way as the Matching task of Experiment 1, asking whether spatial terms were applied in a way that respects axial structure, whether there were any biases across axes, and whether direction within each axis would be respected (i.e., distinguishing correctly between above/below, and left/right). We also looked for effects of distance from the reference object, anticipating that application of terms might break down for more distant locations.

Axial terms of main interest included: Vertical "positive" terms (V+, e.g., above, top, up), vertical "negative" terms (V−, e.g., below, under, down), horizontal "neutral" terms (H, e.g., next to, beside), horizontal "left" (L), and horizontal "right" (R). These categories were used for the main analyses, which examined use of axial terms. People also sometimes used contact terms (C, e.g., "touching") or proximal terms (P, e.g., "near," "far," "close to"). These do not encode axial representations, and are discussed separately below.[2] These categories captured 97% of the WS children's data, 94% of the controls' data, and 90% of the WS adults' data.[3] Coding

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[2] The analyses of these terms are discussed later in this section. One might worry that omitting these terms from the main analyses (reported below) might unfairly penalize the WS participants, who tended to use them more often than control children. However, we repeated all analyses including these terms as well as the main axial terms. The results were the same, unless noted in the text.

[3] The remaining productions for children were either missing (1%) or "other" (e.g., "right there" or uncodable comments such as "it's driving on the purple circle"). The "other" responses by adults were predominantly (71%) made to the Off-Axis areas (top left/right and bottom left/right), and used the term "diagonal" (e.g., "diagonal to the square"), altogether accounting for 42% of the adults' "other" responses.

was carried out on 20% of the productions by a second coder and the resulting reliability was 97%.

Locations On vs Off-Axis. The first analysis examined whether there was more production of target axial terms for locations that were On-Axis vs Off-Axis. For the Vertical axis analysis, we grouped together four different regions: Locations that lay (a) in the Vertical positive region and immediately surrounding the axis (On-Axis, five locations,[4] (b) in the Vertical negative region and immediately surrounding the axis (On-Axis, five locations), (c) in the Vertical positive region and Off-Axis (eight locations), and (d) in the Vertical negative region and Off-Axis (eight locations). For each of these regions, we computed the percent of times an individual produced target vertical terms (i.e., V+ terms for the V+ regions, V− terms for the V− regions). The Horizontal axis analysis used the same logic, grouping together locations that lie (a) in the Left region and On-Axis (five locations), (b) in the Right region and On-Axis (five locations), (c) in the Left region and Off-Axis (eight locations), and (d) in the Right region and Off-Axis (eight locations). This time, however, we computed the percent of times a person produced target horizontal terms (i.e., either Horizontal Neutral or Left for the left regions, and Horizontal Neutral or Right for the right regions). Note that each analysis considered the same Off-Axis regions, first computing the percent of vertical terms, then percent of horizontal terms. Both terms can be used appropriately in these Off-Axis regions, either singly (e.g., above; left) or in combination (e.g., above and to the left). Because the same regions were considered twice, we carried out two separate analyses, one for Vertical regions and one for Horizontal regions.

Table 3 shows the means collapsed over Direction for both regions. The data for each region were submitted to a 3 (Group) × 2 (On vs Off-Axis) × 2 (Direction within Axis) analysis of variance. People produced more target axial terms in the On-Axis regions than the Off-Axis regions for both Vertical and Horizontal regions (Vertical regions: $F = 22.25$, $df = 1, 30$, $p < .01$; Horizontal regions: $F = 59.15$, $df = 1, 30$, $p < .01$). This confirms the basic pattern of axial advantage that was shown in the non-linguistic task of Experiment 1. Looking within each axis, there was no interaction with Group for the Vertical Regions, but there was one for the Horizontal regions, with control children and WS adults showing a sharper advantage than WS children for On-Axis locations ($F = 3.3$, $df = 2,30$, $p < .05$, Tukey's hsd = .27, $p = .05$). This is consistent with the Experiment 1 finding of relative weakness for WS children on the Horizontal axis, and suggests that this weakness may be partially resolved among adults with WS. When this set of analyses was carried out including Proximal and Contact terms as well as axial terms, the results were the same, except that there was no interaction with Group for the Horizontal region. This shows that the WS children were capable of describing these locations, but that they often used terms such as "far" or "near," which describe global, not horizontal, proximity. We discuss this use of non-specific terms at greater length below.

Table 3
Proportions of target axial terms produced On and Off-Axis (Experiment 2A)

	Vertical regions		Horizontal regions		*M*	
	On-Axis	Off-Axis	On-Axis	Off-Axis	On	Off
WS children	.70	.49	.54	.30	**.62**	**.39**
Control children	.91	.58	.81	.25	**.86**	**.42**
WS adults	.71	.49	.75	.39	**.73**	**.44**

The Vertical region analysis also showed an interaction of Direction with On–Off Axis ($F = 13.7$, $df = 1, 30$, $p < .01$). This was due to greater accuracy for V+ than V− terms in On-Axis regions (*M*s = .88, .67, respectively), but not in Off-Axis regions (*M*s = .49, .54). The special strength of V+ terms when they were used for location On-Axis is discussed further below.

Locations within each axis. A second analysis focused only on target terms produced in the On-Axis regions, and directly compared the two axes and directions within each axis (see Table 4). There was an advantage among all groups for the Vertical positive ("above") region compared to all other regions. Thus the naming space was highly asymmetric, with the V+ region named best by all groups.

The results of the 3 (Group) × 2 (Axis: Vertical/Horizontal) × 2 (Direction) analysis of variance confirmed this impression. There was a main effect of Group (F $2,30 = 3.7$, $p < .05$), with control children producing reliably more target terms than WS children (*M*s =

Table 4
Proportions of target axial terms produced in On-Axis regions (Experiment 2A)

	Vertical regions		Horizontal regions[a]		*M*
	V+	V−	Left	Right	
WS children	.86	.54	.50	.58	**.62**
Control children	.96	.86	.80	.78	**.85**
WS adults	.82	.60	.68	.74	**.73**
M	**.88**	**.67**	**.68**	**.70**	

[a] Horizontal regions were primarily named with Horizontal Neutral terms (e.g., beside, next to) by both groups of children.

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[4] In this category were included the three contact locations for each side because adults predominantly use basic axial terms for these locations, e.g., "on top," "above").

.85, .62, respectively, Tukey's hsd = .20, $p = .05$); WS adults were in between ($M = .73$) and not reliably different from either group. When Proximal and Contact terms were included, the main effect of Group disappeared, indicating again that reasonable (if imprecise) terms such as "close" or "near" were often used by WS people to describe locations.

There was also an interaction of Axis with Direction (F 1,30 = 10.23, $p < .01$), confirming the advantage for V+ region terms relative to V− terms (*M*s = .88 vs .67), with no comparable asymmetry for the two Horizontal regions (*M*s = .68 Left, .70 Right; Tukey's hsd = .13, $p = .05$). In fact, the V+ region was named more accurately than each of the other three regions, suggesting that this region is "highlighted" in the naming space—an effect that was not seen in the non-linguistic matching task of Experiment 1, and will be discussed further below. There were no interactions with Group.

Distance along the axes. A final analysis examined whether there were any effects of distance from the reference object, as we had found in the non-linguistic task. Because there was only one response per location, we compared the sums for contact locations for each main region (V+, V−, Left, Right) to the sums for the most distant locations along each axis. People received a score of 1 for a target response at either "contact" and/or "far" locations, and 0 for a non-target response, with a maximum of 4 for "contact" and 4 for "far" locations (2 per axis). A 3 (Group) × 2 (Axis) × 2 (Distance) analysis of variance showed a main effect of Group (F 2,30 = 3.3, $p = .05$), with WS children producing fewer target responses than control children (*M*s = 1.52, 1.87, Tukey's hsd = .33, $p = 05$). There was also an effect of Distance (F 1,30 = 4.25, $p = .05$), with Contact locations eliciting more target terms than Far locations (*M*s = 1.8, 1.6, respectively) and a three-way interaction of Group, Axis, and Distance (F 2,30 = 6.16, $p < .01$). Only WS adults showed reliable differences across the locations, with Horizontal Far locations eliciting the fewest target terms, reliably worse than Vertical Far or Horizontal Near (*M*s = 1.1, 1.8, 1.9, 1.5 for H-Far, V-Far, H-Near, and V-Near, respectively, Tukey's hsd = .6, $p = .05$). The children with WS showed the same pattern (*M*s = 1.3, 1.6, 1.5, 1.7), but these differences were not reliable. The control children showed no effects of distance (*M*s = 1.8, 1.8, 1.9, 2.0, respectively).

Other responses. If the WS children and adults produced fewer axial terms than control children, what did they say? In order to examine this, all responses other than those already considered were tallied and categorized. As shown in Tables 5 and 6, these responses were quite constrained. They included terms that named (a) the wrong direction along the correct axis (e.g., "above" for a region below the square), (b) the wrong axis entirely (e.g., "beside" for a region above the

Table 5
Proportions of terms produced other than target axial terms: Vertical Axial regions (Experiment 2A)

	V+ region			V− region		
	WSC	Controls	WSA	WSC	Controls	WSA
Terms produced						
Wrong direction	.00	.00	.02	.10	.04	.03
Wrong axis	.06	.00	.04	.14	.00	.14
Contact, Proximal	.02	.00	.12	.22	.06	.17
Other	.06	.04	.00	.00	.04	0.6
Total	.14	.04	.18	.46	.14	.40

Maximum = 50 for WSC and Controls, 65 for WSA.

Table 6
Proportions of terms produced other than target axial terms: Horizontal Axial regions (Experiment 2A)

	Left region			Right region		
	WSC	Controls	WSA	WSC	Controls	WSA
Terms produced						
Wrong direction	.00	.04	.02	.00	.00	.02
Wrong axis	.06	.04	.03	.06	.02	.03
Contact, Proximal	.36	.06	.14	.34	.12	.15
Other	.08	.06	.07	.02	.08	.06
Total	.50	.20	.26	.42	.22	.26

Maximum = 50 for WSC and Controls, 65 for WSA.

square), or (c) a more general relationship of contact or proximity (e.g., "touching" the square for a contact location on its left side, or "near" the square for a location below the square).

The latter terms are less specific than the axial terms, but in general, they were used correctly. For example, in the Vertical regions, Contact terms were produced for locations that did, in fact, contact the Reference object. Thus, locations that were below the square and touching it could plausibly be described as "touching" the square. Similarly, Proximal terms such as "near to " or "far from" were appropriate (if vague) for locations that were not in contact with the square. As can be seen in Tables 5 and 6, these terms made up the bulk of the responses among WS individuals. As we already indicated, inclusion of these terms in the main analyses resulted in fewer effects or interactions with Group, suggesting that the WS participants often named locations with these terms rather than the more precise axial terms. The fact that they were largely appropriate tells us that the WS participants were competent in their description of location. But the fact that these terms were often used in lieu of more specific axial terms also hints that there may be special difficulty in coding location with axial terms (which require representing both axis and direction). We discuss this possibility more fully later.

There were only two other notable patterns. One was the production of terms with the wrong direction (V+) for the V− region, i.e., "above" or "on top of" for regions below the square. These directional errors occurred only a few times for the horizontal axis, probably because participants rarely produced directional terms (left/right) for the horizontal axis, sticking with "beside" or "next to." The other pattern was one in which terms were used for the wrong axis (e.g., "next to" for V− regions and "above" or "below" for horizontal regions). It is notable that these error types were rare in the corpus and that they occurred at some level among all groups. Moreover, others have reported that normally developing children use "next to" for vertical locations and sometimes exchange "above" and "below" (Clark, 1980; Richards, 2001; see also Coventry, Prat-Sala, & Richards, 2001). We return to this issue in the discussion.

Summary. All groups showed more target basic terms for locations that were On-Axis than Off-Axis, effects of distance from the reference object, and a special advantage for terms applied to V+ regions that were On-Axis. The first two properties echo the findings of the non-linguistic matching task of Experiment 1. The third property is new and reflects a strong asymmetry in the naming space for V+ regions On-Axis. The WS children performed more poorly overall than MA matched children and the WS adults fell somewhere in between. Both WS children and adults showed weakness relative to the control children in naming along the horizontal axis (for the WS adults, this occurred for locations most distance from the reference object). These relative weaknesses also echo the findings of Experiment 1. Finally, the WS children and adults used general terms more frequently than control children to name On-Axis locations.

Some of the differences between WS children and adults vs normal controls could be due to the pressures of a Production task. Hence we also carried out a Comprehension task which tested knowledge of the basic axial terms.

Experiment 2B: Language comprehension

Design and procedures

Subjects were shown a book of $8\frac{1}{2}$ in. × $5\frac{1}{2}$ in. white pages, each of which was blank except for a 2×2 in. solid square placed directly in the center. They were given a pen and were asked to "Put a mark ______ the square." Twenty-two terms (or compound phrases incorporating these terms) were tested, these included 14 terms that map onto locations along the vertical axis and eight that map onto locations along the horizontal axis. Within the vertical axis, seven terms encode "positive" direction and seven "negative" direction (above/below, right above/right below, way above/way below, on top of/ underneath, on the top of/on the bottom of, over/under, and higher than/lower than). The remaining eight terms map onto locations along the horizontal axis. These included four terms that are neutral with respect to direction along the axis (next to, right next to, beside, and on the side of), and four that encode direction as well (on the right of/on the left of, to the right/to the left of). We examined whether the marks for the terms were placed in regions that fell (a) along the axes of the reference object (vs Off-Axis), (b) on the correct axis (vertical vs horizontal), and (c) in the directionally appropriate location within each axial region (i.e., above/below or right/left). Terms were presented in one of two random orders, with the constraint that each pair of terms with directional mates (e.g., above/below) was queried on adjacent pages. Note that each term was tested on a new, blank page, so it was possible to place each target term's dot anywhere on the page. That is, there were no constraints introduced by marks already occupying the page.

Results

As with the Production task, the critical questions were whether people would place marks for the spatial terms in locations that were On-Axis (vs Off-Axis), whether there would be an accuracy advantage for Vertical terms compared to the Horizontal terms (reflecting the advantage of the Vertical axis), and whether, within each axis, directional distinctions would be preserved (i.e., distinguishing correctly between above/below, and left/right). Distance could not be systematically evaluated, because the task allowed marking any location on each page. Therefore, we can only informally evaluate the distribution of marks over distance.

The results were analyzed by computing, for each group of terms, the proportion of marks that were placed in the target axial regions. These target regions were designated as extending from the appropriate (top/bottom/right/left) edge of the square all the way to the edge of the page. These regions encompass the axes and allow for a small amount of room on each side. As will be seen, most of the marks did fall along the narrow region that just encompasses the axes. In addition, there were a number of marks placed inside of the square. Some of these were quite plausible for particular target terms, e.g., "on" or "over" the square (viewing it as three-dimensional), or, "on the right" of the square if placed just inside the right edge of the square. These were also counted as in the target region, but the results did not differ when these marks were not so counted.[5]

The distribution of dots for each group of terms is shown in Figs. 5 (Vertical positive/negative terms), 6 (Horizontal non-directional terms), and 7 (Horizontal left/right terms). Numbers inside each grid mark represent the numbers of marks placed in that location. A quick look tells the story: For the most part, subjects in all groups followed the region surrounding the appropriate axis for each group of terms—a strong effect of organizing axes. In addition, for the Vertical terms, people recruited the spatial region all the way up to the top (or bottom) of the page. However, there were errors on direction, especially among WS children and adults. WS children made such errors on both vertical positive/negative terms, and on right/left. WS adults and control children made the errors on right/left.

The proportions of marks placed in each term-appropriate target region are shown in Table 7. These were entered into a 3 (Group) × 4 (Axis and Direction: V+, V−, H neutral, and H marked) analysis of variance, which revealed only an effect of Axis and Direction (F $3,90 = 17.12$, $p < .01$). Post hoc comparisons showed that the Horizontal marked terms were worse than all others (Tukey's hsd = .15, $p < .05$). There was only a marginal effect of Group (F $2,30 = 2.32$, $p = .11$), but, as Table 7 shows, the WS children performed the worst and the control children performed the best, with WS adults in between. This is the same pattern that was observed in the Production task.

Other responses. We analyzed the locations of the marks placed outside of the target areas to determine whether they were placed along (a) the right axis but wrong direction or (b) the wrong axis. The overwhelming number of errors was of the first type and occurred for the Horizontal Left/Right terms among all groups (Proportions = .38, .38, and .27 of Left and Right trials for WS children, control children, and WS adults, respectively). The WS children also made several directional errors on Vertical terms, placing marks for "above" terms somewhere below the square, or vice versa (.07 of responses for V+ terms and .01 for V− terms), but the control children and WS adults made none of these error types. Errors of placement along the wrong axis were infrequent and were distributed evenly across the terms that were tested (M proportions across all term types = .06, .02, and .02 for WS children, control children, and WS adults, respectively). The only remaining pattern was a tendency for WS adults to place marks for V+ and V− terms in locations that were directionally correct (above or below the reference object), but fell outside of the target region (which was defined rather narrowly). These were responsible for .12 of the WS adults' V+ and .12 of their V− term responses.

Summary of language tasks. The results of both the Production and Comprehension tasks showed that WS children and adults were able to correctly map key spatial properties of basic spatial terms onto the spatial world. In the Production task, they produced more target axial terms for locations that were On-Axis than Off-Axis, similar to the pattern shown by control children (and normal adults, see Hayward & Tarr, 1995; Munnich et al., 2001). In the Comprehension task, they also respected the axial regions extending from the reference object, placing marks along the axes of the reference object on an entirely blank page. These findings strongly suggest that WS children and adults, like normal children, represent space in terms of reference systems that are structured by the mentally represented axes of reference objects.

This pattern also echoes the findings of the Matching task, which revealed mental representation of both vertical and horizontal axes. Additional findings of relative weakness among the WS children also mirrored findings in the Matching task. In the Production task, the WS children showed less of an advantage for On-Axis locations along the Horizontal axis than control children or WS adults; and WS adults showed weakness at a distance along the Horizontal axis. As a whole, these findings converge on the idea that there may be relative weakness in representing locations along the Horizontal axis among people with WS, although there also appears to be some development between children and adults with WS.

Despite these similarities between the Language and Matching tasks, the results were not identical. Both Lan-

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[5] Analyses were also carried out removing any credit for marks inside the square, and the results were the same. The additional credit given for "in square" marks added to the V+ scores by .12, .08, and .09 for the WS children, control children, and WS adults, respectively. They also added .02 to the Right/Left scores for the WS children and WS adults, respectively.

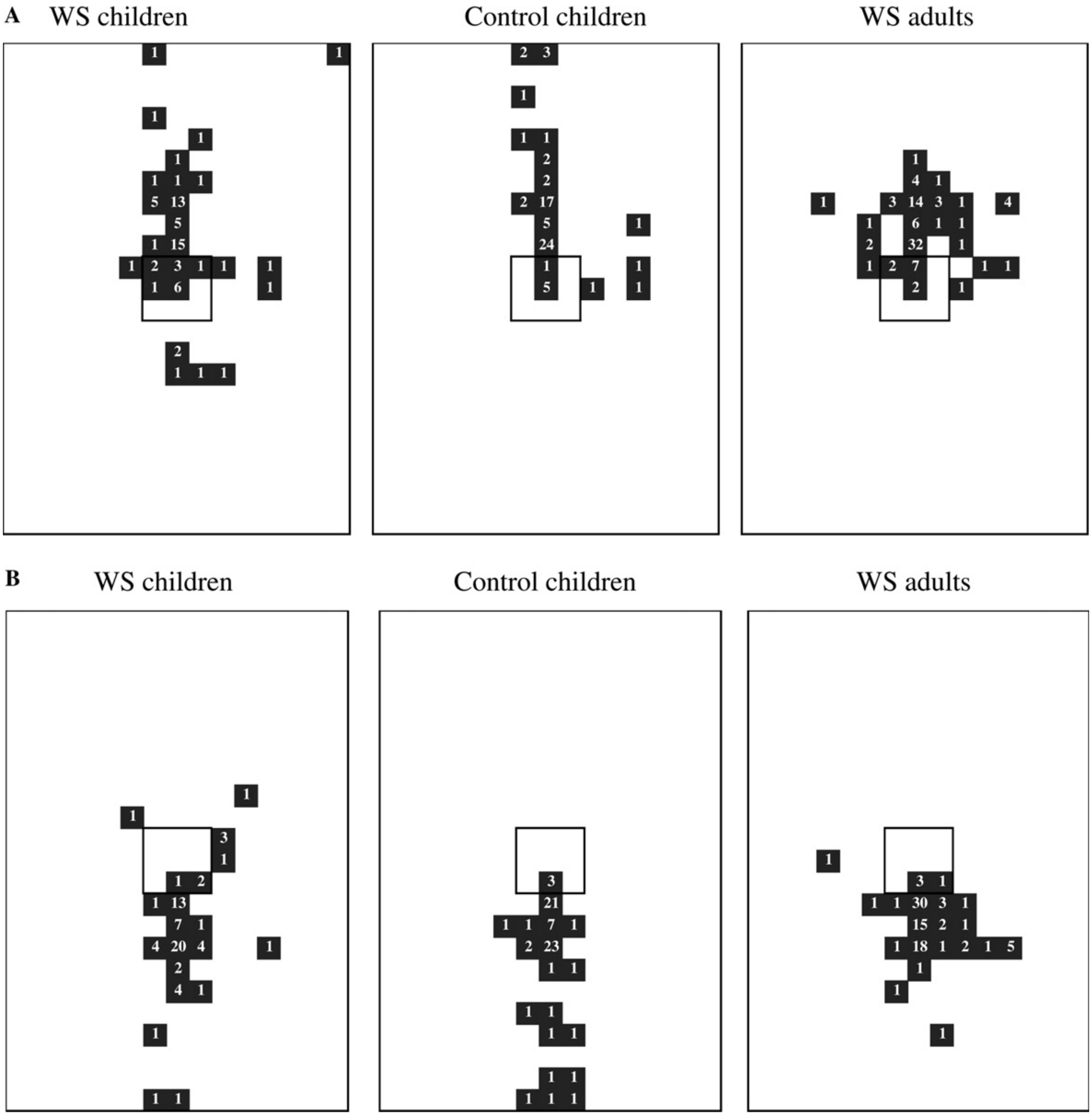

Fig. 5. Distribution of marks placed by participants in the Comprehension task. (A) Vertical Positive terms. (B) Vertical Negative terms.

guage tasks—but not the Matching task—revealed difficulty in encoding direction among all groups. The Production task showed a strong bias in favor of the Vertical positive region, and WS children (but not controls or WS adults) sometimes erred by extending the V+ terms to the V− region. The Comprehension task did not show this V+ bias, but did reveal difficulties among all groups in getting direction correct for terms Left and Right.

What is the source of the directional coding problem? One possibility is that directional coding within an axis is difficult in both linguistic and non-linguistic tasks. Although we did not observe any difficulty in directional coding in the Matching task of Experiment 1, this may not be telling. Importantly, the "Different" stimuli in the Matching task of Experiment 1 were never displaced from the target location by a major directional change along an axis. Therefore, people would not need to encode a contrast in direction between the target and distractors in order to accurately match the dot's location, making the Matching task a poor diagnostic for any directional problems. In order to determine whether directional encoding is difficult in a non-linguistic matching task, we would need a non-linguistic task that specifically evaluates people's ability to represent

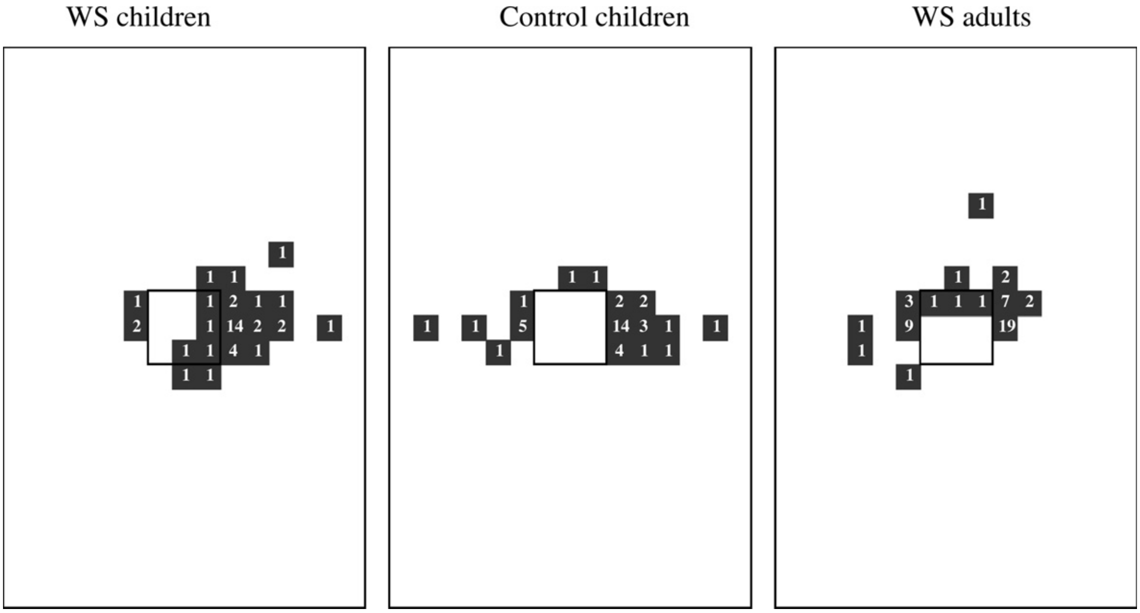

Fig. 6. Distribution of marks placed by participants in Comprehension task. Horizontal non-directional terms.

and remember both axial and directional coding. This was the purpose of Experiment 3.

Experiment 3

Participants

The same individuals participated as in Experiment 2, with the exception that one of the WS children was replaced with a new subject, as were two of the controls, all due to attrition. Of those who participated in both experiments, three of the WS children and five of the controls completed this experiment within a month of Experiment 2, and the remaining children completed the experiment within a year. In addition, 11 of the 13 adolescents and adults from Experiment 2 participated, all at the Biennial WS Association meeting. At the time of test, the WS children had a Mean age of 11;6 (range = 8;9–14;3) and the controls had a Mean age of 5;10 (range = 3;11–7;2). The scores of the WS children were still well matched to those of the controls using the KBIT (*M* Verbal scores = 33.5, 32.7 *SE*s = 2.3, 2.4; Matrices scores 18.3, 18.8, *SE*s = 1.4, 1.6). As reported in Experiment 2, the WS adolescents and adults had verbal scores that were reliably higher than the controls. The new children were drawn from the same sources as described in Experiment 1.

Design and methods

The principal goal of this experiment was to determine whether people would have difficulty encoding direction within an axis when tested in a non-linguistic task. Following the general design of Experiment 1, people were asked to match the location of a target Standard array to one of two Test arrays, one the Same and one Different from the Standard. Different arrays showed the dot at the same distance from the Reference object, located either (a) along the same axis, at the opposite end, or (b) along a different axis (see Fig. 8 for an example of a "Same Axis" trial). For example, if the Standard showed a dot located 2 grid marks *above* the square, then the Different array would display the dot *either* (a) 2 grid marks *below* the square (Same Axis, Different direction, as in Fig. 8), or (b) 2 grid marks to the *right* or *left* of the square (Different Axis). If the Standard was to the left of the square, then Different arrays would show the dot either (a) to the right of the square (Same Axis, Different direction) or (b) above or below the square (Different Axis). If people have difficulty representing direction within an axis, then they should make more errors confusing locations that lie on the same axis (but a different direction) than on a different axis.

Subjects viewed eight target locations, all aligned with the vertical or horizontal axis of the square reference object. The eight locations included four in contact with the reference object (aligned with the center of the top and bottom, right and left sides) and four that fell along the same axes, but two grid-marks away from the square. Each of these Standards was shown eight times with two Test arrays, one Same and one Different. Half of the Different arrays showed the dot along the Same Axis (but in the opposite direction), and half showed it along the Different Axis, with choice of direc-

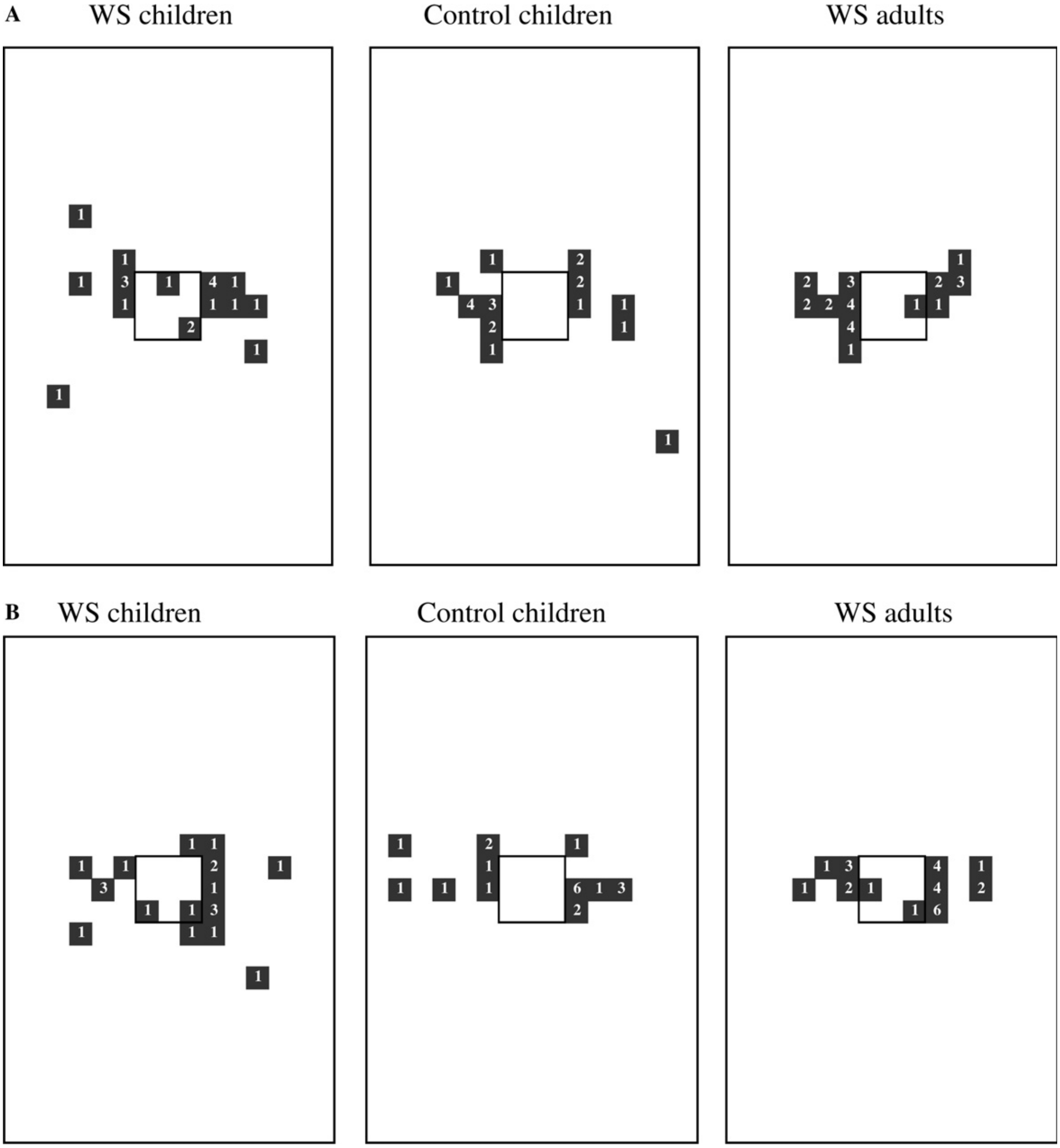

Fig. 7. Distribution of marks placed by participants in Comprehension task. (A) Horizontal "left" terms. (B) Horizontal "right" terms.

Table 7
Proportions of terms placed in correct regions in the Comprehension task (Experiment 2B)

Terms	Vertical regions		Horizontal regions		*M*
	V+	V−	Neutral	Right/Left	
WS children	.80	.86	.78	.32	**.68**
Control children	.94	.94	.95	.55	**.85**
WS adults	.82	.82	.84	.67	**.79**
M	**.84**	**.87**	**.86**	**.53**	

tion counterbalanced. The 64 trials were presented in one of two random orders on a 19-in. Sony Trinitron monitor with a 1024/768 pixel display. The size of the displays was similar to those used in Experiment 1, with a $1\frac{3}{8}$ in. square reference object, and 3/8 in. dots as the target figures.

Because it was unclear how difficult it would be for the children to match locations that differed as coarsely as the ones used here, a pilot experiment was carried out, in which the children were tested with both Standard

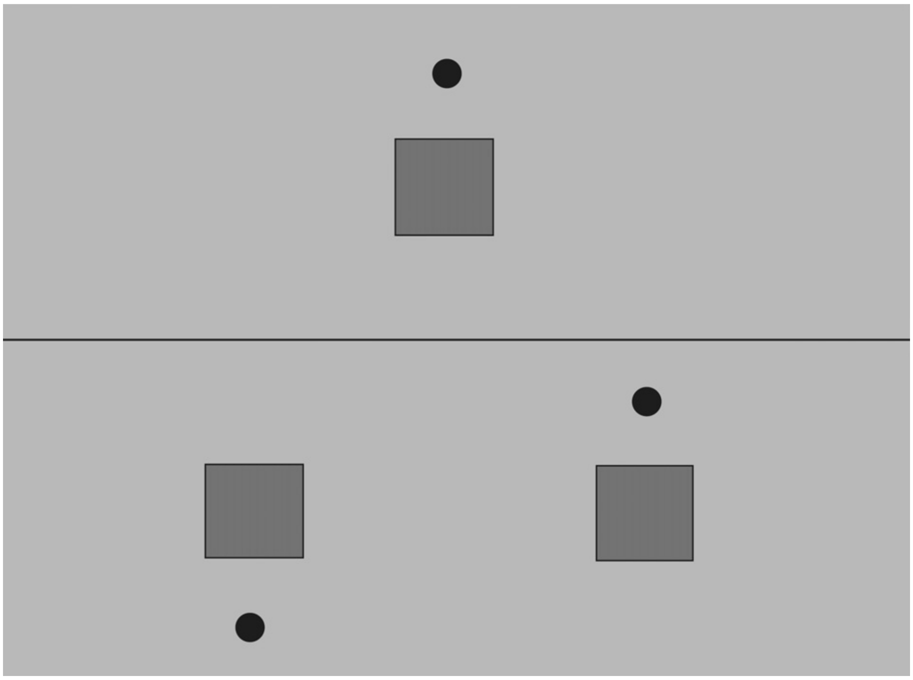

Fig. 8. Layout of panels for a "Same Axis" trial in Experiment 3. Participants viewed the top panel, which either stayed on the screen (No-Delay) or was removed for 1 s (Delay), followed by presentation of the lower panel with the Same (right panel) and Different (left panel) choices. In the "Different Axis" trials, the Different choice changed the axis, in this case resulting in location to the left or right of the square. See text for further detail.

and Test arrays simultaneously present on the computer monitor. Children viewed the Standard on the screen alone, and when they were ready, the experimenter clicked the mouse to bring up the two Test items for the trial. Results showed ceiling performance (reported below), and hence, the main experiment was carried out with a delay. In this Delay condition, participants viewed the Standard, and when they agreed that they were ready, the experimenter clicked the mouse, making the Standard disappear, followed by a 1-s delay before the Test items appeared. In order to prevent or disrupt verbal encoding during the delay, the experimenter began to read numbers as soon as the participants said they were ready. Participants were instructed to shadow these numbers aloud from the "ready" signal until the test items appeared (about 3 s). All people complied with these instructions.

Results

Proportions correct in the No Delay condition (children only) were entered into a 2 (Group) × 2 (Target's Axis: Vertical or Horizontal) × 2 (Target's Direction: Positive or Negative, with Left designated as "Positive") × 2 (Trial type, Same or Different Axis) analysis of variance with the last three factors within-subjects. Performance was close to ceiling, but the controls still performed better than WS children (Ms = 98, 92, F 1,16 = 5.9, $p < .05$).[6] There was also an effect of Axis (F 1,16 = 9.2, $p < .05$), modulated by a Group × Axis interaction (F 1,16 = 9.2, $p < .05$). The latter reflected poor performance by the WS children along the Horizontal Axis, that is, when targets were to the left or right of the reference object ($M = 87.8$, compared to 96.2 along the Vertical, and $M = 98$ for control children along each axis). This relative weakness along the Horizontal axis echoes the results of Experiments 1 and 2. However, it is also possible that the lack of similar effect among controls in the present experiment could be due to a ceiling effect, especially given the Vertical axis advantage already shown in Experiment 1. There was no effect of Trial type or Direction in this condition.

Performance in the Delay condition was analyzed as above, this time using the three groups as the between-subjects factor (see Table 8 for means). There was a main effect of Group (F 2,28 = 3.5, $p < .05$), with the WS adults and the control children both performing alike, and better than the WS children (Ms = 81.1, 80.6, 67.9, respectively). Group did not interact with any factor. There were main effects of Target Axis (F 1,28 = 7, $p < .05$), Target Direction (F 1,28 = 5.3,

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[6] Data from one control child in the No Delay condition were lost by computer failure, so there were nine controls in this analysis.

Table 8
Proportions correct on axis matching task with delay (Experiment 3)

Trial type	Axis and direction (target location)			
	V+	V−	Left	Right
Same axis, new direction				
WS children	.79	.64	.58	.65
Control children	.91	.64	.66	.79
WS adults	.92	.56	.80	.78
M	**.87**	**.61**	**.68**	**.74**
Different axis				
WS children	.75	.71	.65	.68
Control children	.91	.85	.81	.88
WS adults	.94	.84	.77	.88
M	**.87**	**.80**	**.74**	**.81**

$p < .05$), and Trial type (F 1,28 = 17.1, $p < .01$), subsumed by a three-way interaction (F 1,28 = 7.3, $p = .01$). As can be seen in the table, all regions except the V+ region showed better performance on trials where the distractor lay on a Different Axis from the target. For example, targets that were "below" the reference object were more likely to cause errors when the distractor was "above" the square than when it was "left" or "right," and targets that were "left" (or "right") of the square were associated with more errors when the distractor array was "right" (or "left," respectively) than when it was "above" or "below." There was no reliable interaction of these factors with group (F 2,28 = .31, $p > .73$). In general, then, all groups of subjects made more errors when they had to choose between two locations that were on the same axis, but lay in different directions from the target. The fact that these errors did not appear in the No Delay condition suggests that the problem does not lie in perceptual representations of the locations, but in their storage in memory, albeit over a very short period of time.

The exception was the Vertical positive region, where there seemed to be no difficulty storing both axis and direction. This pattern is reminiscent of the special strength of this region in the Production task, and we consider below how the patterns in the two tasks might be related. For now, we note that any difficulties in representing direction seem to persist into adulthood for individuals with Williams syndrome, who performed at the same level as 5-year-old normally developing children.

General discussion

The results of three experiments show preservation and breakdown in the use of spatial reference systems among children and adults with Williams syndrome, as well as normally developing children. The findings point to three different themes. One is the importance of a fundamental axial structure across the three groups and across the linguistic and non-linguistic tasks. The findings demonstrate the strong organizing effects of reference systems even among children and adults who have severe spatial cognitive deficits. The second is the difficulty of directional coding, also seen across both linguistic and non-linguistic tasks and among all groups, and the special status of the vertical positive region, which was seen in both the Production task and in the non-linguistic matching task of Experiment 3. The third concerns the consequences of spatial breakdown in non-linguistic representation for the acquisition and use of spatial language. The results shed light on how even a fragile non-linguistic spatial system might support spatial language; and some ways that spatial language might be partially independent of this anchoring.

Perhaps the most striking finding of the experiments was the preservation of the reference system as an organizing structure across both non-linguistic and language tasks and among all groups. In the first experiment, children matched locations along the extension of the reference object's axes more accurately than locations off these axes. In the second experiment, people produced and comprehended basic spatial terms more accurately and more often along the reference object's axes. The pattern in the matching task was shown by WS children and normally developing children between the ages of 3 and 6; the pattern in the language tasks was shown among these groups and also by WS adults. These findings are, to our knowledge, the first demonstration of such detailed parallel effects in linguistic and non-linguistic tasks among children, normal or impaired. They are remarkably similar to the patterns shown by normal adults and therefore suggest that the reference system is a rock-bottom representational structure that serves to organize children's perception and memory for location, as well as their acquisition of basic spatial terms. The reference system also appears to be part of the spatial representational system of children and adults with WS. Surprisingly, despite the classic profile of severe deficit in some aspects of spatial cognition, people with WS apparently construct and use reference systems to represent an object's location, as well as to talk about those locations. These findings strongly support the idea that the two systems are yoked.

The second finding that emerged was the relative difficulty of encoding direction. The first evidence of this emerged in the language tasks, which naturally require that people encode direction in order to make appropriate distinctions between above/below and right/left. Children with WS made production errors in which they sometimes produced vertical positive terms for regions that were below (V−) the reference object. These substitutions occurred only in the youngest control child (who was 3;10 years), and never occurred in the WS adults, suggesting that these groups had sorted out the terms that apply to the vertical axis as well as the directional distinctions within the axis. However, directional errors occurred frequently along the horizontal axis for all groups in the Comprehension task, indicating that, although all groups had sorted out the set of terms that neutrally apply to the horizontal axis, they had not yet sorted out the direction of the non-neutral (right/left) terms.

The finding that vertical and horizontal terms were acquired without the correct directional distinctions is consistent with literature showing that young children often set up semantic fields before fully comprehending individual terms within the field. For example, children know that "red" and "blue" are color terms before they have sorted out which hues they encode (Bartlett, 1978; Carey, 1982). In addition, terms "come" and "go" are often used interchangeably before sorting out the specific deixis (Clark & Garnica, 1974), and "before" and "after" are used interchangeably by 3-year-olds before specific temporal direction is learned (Clark, 1971). Of course, terms "right" and "left" are classic cases of directional confusion in children (Piaget & Inhelder, 1967). Thus the WS developmental profile shows errors that are quite similar to those found among normally developing children. However, it is also clear that our WS children were delayed in their representation of axial terms relative to normal MA matches, having yet to sort out the vertical axis. Second, directional errors were prolonged into adulthood in WS, despite overall high vocabulary scores as measured by the standardized (KBIT) test. It is important to note that this test taps into object naming only, and thus may not shed much light on spatial language of the sort we have examined. However, the presence of directional errors among WS adults suggests developmental arrest in this aspect of spatial language, since directional errors for right/left are usually resolved in normal children by around age 9.

The other manifestation of directional difficulty was seen in the delayed non-linguistic matching task of Experiment 3. There, all groups showed poorer performance when they had to encode and make choices based on direction within an axis, rather than just the axis of the location. Importantly, this difficulty was observed only in the delay condition of the task, but not in the no-delay perceptual matching task. This suggests that the locus of directional difficulty is not in the perceptual system, but rather, in visual-spatial memory. There is good independent evidence that visual-spatial memory is impaired in WS (Jarrold, Baddeley, & Hewes, 1999). Elsewhere we have found that WS children have special difficulty encoding and remembering directional relationships *within* objects, again consistent with the findings of the present paper (Hoffman et al., 2003).

The finding of directional fragility is also consistent with several studies on visual attention that have shown the independence of axes and direction within axes. For example, Carlson-Radvansky and Jiang (1998) asked people to verify a target's location, and found negative priming from axes that were available on a previous trial, but had not been selected (i.e., used). This priming was observed to operate along the entire axis of the reference frame, including both endpoints, even though only one end had been primed (see also Logan & Sadler, 1996). Moreover, McCloskey and Rapp (2000) showed that axes and direction can be dissociated by neurological impairment. They studied a young woman who had a developmental spatial impairment which led her to systematically err in ballistic reaching for targets. Her reaches preserved the axis along which the object was located, but not the direction. These findings are consistent with the present ones, which further suggest that difficulties in representing direction (but not axis) may be reflected in the development of linguistic and non-linguistic spatial representations.

In addition to these asymmetries in representation, we found evidence of special status for the vertical positive region. The first piece of evidence emerged in the Production task, where we found that this region received the sharpest and most accurate use of target spatial terms. Specifically, the region above the reference object was named with appropriate basic terms more frequently than any other region by all groups, suggesting it is the region that is most firmly anchored by linguistic terms. This special status of the V+ region also emerged in the non-linguistic task of Experiment 3: It was the only region where we did not see the relative difficulty of encoding direction within the axis.[7]

What is the root of this phenomenon? One possibility is that the non-linguistic task was actually carried out using linguistic encoding. Given that the V+ region also was named most accurately, this would seem to be the natural explanation. However, several facts argue against this. First, the presence of a verbal distractor during the task should have disrupted any linguistic encoding that people might have tried. Second, there is no reason to suppose that the same explanation would not hold for V− locations, that is, that people could have linguistically encoded these locations (even as "not above") and thereby remembered them better. But this region did not show a special status, and was like both horizontal regions in showing relative difficulty in remembering direction. Third, the lack of group effects or interactions in Experiment 3 suggests that the asymmetry in the representational space exists both for

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[7] The privileged nature of the V+ region in the Production task also argues against the idea that the errors in this task were due to some general difficulty assigning roles to the figure and ground object. If this were true, one would expect errors be randomly distributed across both directions of an axis.

people who make linguistic errors for locations on the vertical dimension (as did the WS children) and those who do not (control children and WS adults).

One possible explanation, which is admittedly speculative, is that the V+ region is indeed special in spatial representation, whether linguistic or non-linguistic. Evidence for this possibility comes from several sources. First, Klatzky, Clark, and Macken (1973) tested 3- and 4-year-olds' ability to learn nonsense terms that were presented as names for the positive and negative ends of dimensions for size, height, length, and thickness. For example, "ruk" might refer to long objects and "dax" to short ones. They found that children learned the terms for the positive ends faster and with fewer errors, suggesting that across domains, there is some special status to regions named by positive terms in a spatial dimension. These authors proposed that the ease of learning positive terms is rooted in conceptual asymmetries, rather than sheer frequency of use of the terms. Second, there is evidence from the neuropsychological literature that the vertical positive region is less prone to neglect than the vertical negative region, just as there is a tendency for horizontal neglect to be less often right-sided than left-sided (Pitzalis, Spinelli, & Zoccolotti, 1997). The possibility that such an asymmetry in non-linguistic spatial representations supports the early and more robust acquisition of vertical positive terms echoes the speculation made by Clark (1973). That gravity and our upright posture inevitably leads to functional asymmetries in our representation of space (see also Shepard & Hurwitz, 1984).

The final issue concerns the consequences of spatial breakdown among people with WS for the acquisition and use of spatial language. Much of our discussion has focused on the remarkable similarity between the spatial systems of WS children and adults, to those of normal children and adults. These similarities tell us that even a fragile non-linguistic spatial system can support the acquisition of spatial language, and speaks strongly in favor of the idea that the two systems are yoked as children come to learn the meanings of certain spatial terms. Yet there were also differences in performance. What do these differences tell us?

One set of differences reflected overall performance decrements, especially between WS children and the normally developing controls. Some of these, such as overall poorer performance on the matching and language tasks, could be the result of noisier representations, increased uncertainty and/or differences in the response strategies used in situations of high uncertainty. Other differences suggest a real developmental difference in the robustness of the axial representations used for language. For example, the WS children—but not the normal children nor the WS adults—had difficulties with spatial terms encoding the vertical axis as well as the horizontal axis. The normal children and the WS adults showed similar problems only for terms along the horizontal axis, suggesting they had already straightened out the directions along the vertical axis. The fact that WS adults tended to perform better than the WS children, and were closely comparable to the normally developing controls, suggests that some of these difficulties do get resolved over development in WS. But it is also sobering to recognize that the WS adults did not perform better than normal 5-year-olds, often confusing left and right, for example. This suggests developmental arrest in WS adults for certain aspects of spatial language.

Another set of differences was more specific and spoke to the nature of the breakdown in Williams syndrome, as well as the consequences of spatial breakdown for learning and using spatial terms. One difference argues for the direct effects of spatial breakdown on spatial language. In the non-linguistic matching task of Experiment 1, WS children showed weakness along the horizontal axis and at a distance from the reference object. The WS adults were not tested in Experiment 1, but they also showed relative weakness along the horizontal at a distance in the Production task of Experiment 2. The parallel here should be treated with caution, as the results came from different tasks. However, the parallel is notable because accurate responses at a distance from a reference object require spatial integration over a relatively long distance—something that may be problematic for WS individuals, accounting for their pronounced difficulty in spatial construction tasks.

Current research on visual-spatial integration in Williams syndrome is mixed, with some studies reporting impairment at even relatively low levels of the visual system (Kovacs, Kozma, Feher, & Benedek, 1999) but others finding sparing in integration coupled with difficulties in segmentation (Jordan et al., 2002; Ogbonna, Palomares, Landau, Egeth, & Hoffman, 2004; Palomares, Landau, Hoffman, & Egeth, 2003; Reiss, Hoffman, & Landau, 2003). These results are based on basic visual tasks such as perception of illusions, so it is not clear whether they are directly applicable to the tasks used here. However, the findings of our present study suggest there may be particular difficulties in integrating over long distance locations along the horizontal axis. This could reflect a basic asymmetry characteristic of the human visual-spatial cognitive system, or it could reflect a weakness particular to WS. In either case, failure to integrate over more distant locations might lead to difficulty in naming locations that depend on such integration, such as those at a distance from a reference object. Simply put, if there is greater uncertainty in representing an object's location, then it will be more difficult to accurately name it. Such difficulties could persist into adulthood if reference systems are noisy among WS individuals.

A final difference, however, points out that spatial naming is not a direct and simple mapping from non-linguistic spatial representations. Recall that in the Production task, WS children and adults produced fewer target axial terms than normal children. Instead, they tended to produce more general spatial terms for those locations where normal children were overwhelmingly using target axial terms such as above, below, beside, and next to. As we noted, the more general terms are perfectly good descriptors for these locations, so we are not arguing for a specific deficit in the linguistic use of spatial terms. However, there may be theoretically important reasons for the development of distinctly different styles of spatial term use. We can think of several. One is that, because WS people have more fragile non-linguistic spatial representations, they have more uncertainty about location and hence more uncertainty about whether a given axial term is appropriate. Producing a more general (but appropriate) term would be an understandable solution to this problem. Another possibility is that the axial terms are more difficult to retrieve than general terms, and that errors in retrieval are therefore more likely with these terms. Although we have no particular reason to believe that axial terms are specifically prone to retrieval problems, there is much evidence that WS children and adults have lexical retrieval problems (Vicari, Carlesimo, Brizzolarra, & Pezzini, 1996; Thomas & Karmiloff-Smith, 2003). If so, this could also account for the fact that there were no group differences in performance on the Comprehension task. A third possibility is that WS people develop a style of spatial term use that is consistent with the style they are exposed to: Caregivers who know about the spatial impairment in WS people might be prone to describing spatial layouts using more global spatial terms, anticipating that these will be relatively easy to understand.

In some sense, the patterns of errors shown in the non-linguistic and language tasks were quite similar: In both, the WS children showed decreased accuracy (or target term use), but the same architectural patterns, including evidence for axial structure and directional difficulties. However, the error patterns for the language task were also clearly a product of what the system provides: Language is forgiving, in the sense that it provides for alternative plausible responses when and if a specific locational term is not available. Terms such as "near" and "far," while not as informative as "above" or "below" for certain locations, can still afford speakers and comprehenders a reasonable opportunity to communicate effectively with each other. The availability of such alternatives may, in the case of spatial language among people who are spatially impaired, provide the "look" of perfect normalcy, even if it does not exactly match the output of a person who is not so impaired.

These differences highlight the fact that, in the end, spatial language operates under different constraints from the variety of non-linguistic systems, including visual attention, action, perception, and construction. These constraints naturally stem from both the architecture of the intact system and its available solutions when something goes awry. The inherent differences in spatial architectures will naturally lead to differences in potential for breakdown and patterns of error. This suggests that we should consider constraints on spatial systems not only in terms of their normal operation, but also in terms of the ways in which they might deviate when put under pressure.

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