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Tactile Localization on Digits and Hand: Structure and Development

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Abstract
Localization of tactile stimuli to the hand and digits is fundamental to somatosensory perception. However, little is known about the development or genetic bases of this ability in humans. We examined tactile localization in normally developing children, adolescents, and adults and in people with Williams syndrome (WS), a genetic disorder resulting in a wide range of severe visual-spatial deficits. Normally developing 4-year-olds made large stimulus-localization errors, sometimes across digits, but nevertheless their errors revealed a structured internal representation of the hand. In normally developing individuals, errors became exponentially smaller over age, reaching the adult level by adolescence. In contrast, people with WS showed large localization errors regardless of age and a significant proportion of cross-digit errors, a profile similar to that of normally developing 4-year-olds. Thus, tactile localization reflects internal organization of the hand even early in normal development, undergoes substantial development in normal children, and is susceptible to developmental, but not organizational, impairment under genetic deficit.

Keywords
developmental disorders, parietal lobe, spatial perception, visual-spatial ability, Williams syndrome

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Tactile localization on the hand and digits has provided an important model for understanding experience-dependent change in nonhuman primates (Kaas, Merzenich, & Killackey, 1983; Merzenich & Kaas, 1982; Recanzone, Allard, Jenkins, & Merzenich, 1990) and human adults (Elbert, Pantev, Wienbruch, Rockstroh, & Taub, 1995). Because digit representation can change significantly as a consequence of specific experience, tactile localization accuracy might undergo substantial change over development. However, the development of this ability—including the nature of initial cortical organization underlying tactile localization and the role of perceptual experience in its fine-tuning—remains poorly understood. Given that young children often incorrectly identify which digit on their hand has been touched (Benton, 1955), and that such finger agnosia is rare in adults, there must be developmental change in tactile localization.

Even less is understood about the genetic contributions to tactile localization on the hand. Unique insights can be gained by studying people with Williams syndrome (WS), a genetic disorder associated with a microdeletion (~25 genes) on chromosome 7q11.23. Missing genes include *LIMK1* and *CYLN2*, the former of which is thought to play an important role in spatial learning (Meng et al., 2002). Moreover, the brains of people with WS show structural abnormalities in the parietal lobe and hippocampus (Meyer-Lindenberg et al., 2004; Meyer-Lindenberg, Mervis, & Berman, 2006) and connectivity abnormalities (Marenco et al., 2007) consistent with their severe visual-spatial impairments (Mervis & Becerra, 2007). On average, affected adolescents and adults reach the level of normally developing 4- to 6-year-olds in spatial copying (Hoffman, Landau, & Pagani, 2003), attentive tracking of multiple objects (O'Hearn, Landau, & Hoffman, 2005), object recognition (Landau, Hoffman, & Kurz, 2006), and performance on visual-motor tasks (Atkinson et al., 2003; Dilks, Hoffman, & Landau, 2008).

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Such a broad spatial deficit suggests that WS is characterized by an impairment of the dorsal stream, which plays an important role in spatial representation and action (Atkinson et al., 2003; Goodale & Milner, 1992; Landau & Hoffman, 2007; Ungerleider & Mishkin, 1982). Although somatosensory representation—and tactile localization in particular—may seem functionally quite different from spatial functions such as copying, broad dorsal-stream impairment could even affect the ability to localize a stimulus on the hand. If so, people with WS could show abnormal localization or localization that mirrors the performance of much younger normally developing children (Landau, 2011; Landau & Hoffman, 2012). Such findings would suggest a genetic foundation for somatosensory development, as well as spatial development more broadly.

To examine both the normal developmental trajectory for tactile localization and its profile in a genetic disorder, we quantified stimulus-localization errors in a control group of normally developing 4- to 9-year-olds, adolescents, and adults, comparing these results with those of people with WS across a broad age range. We hypothesized that localization accuracy would undergo development in normal individuals and would be affected in the WS group as part of the overall profile of severe spatial impairment, such that people with WS would show quantitative abnormalities, qualitative abnormalities, or both relative to normally developing individuals. Both quantitative and qualitative abnormalities in people with WS have been described in a number of perceptual and cognitive domains (Ansari, Lyons, van Eimeren, & Xu, 2007; Karmiloff-Smith, 2007; Thomas et al., 2010). The developmental trajectory of normal tactile localization and its nature in people with WS may shed light both on how this important skill develops and on its genetic foundation.

Experiment 1

Method

Participants. Twenty-three participants with WS (mean age = 17 years 5 months, range: 7–32 years) and 72 control participants were tested. The control group was normally developing and included eighteen 4-year-olds (mean age = 4 years 5 months, range: 4 years 0 months to 4 years 11 months), sixteen 6-year-olds (mean age = 6 years 6 months, range: 6 years 0 months to 6 years 11 months), sixteen 9-year-olds (mean age = 9 years 6 months, range: 9 years 0 months to 9 years 11 months), and 22 adolescents and adults (mean age = 19 years 4 months, range: 15 years 11 months to 29 years 5 months). All participants with WS had the classic deletion in the Williams-Beuren syndrome region of chromosome 7q11.23, as confirmed by a fluorescent in situ hybridization test.

Nineteen of the WS participants were given the Kaufman Brief Intelligence Test (Kaufman & Kaufman, 2004). Their mean composite IQ score was 82 (range: 54–94), which is representative of scores for WS participants more generally (see, e.g., Mervis, 2006).

Stimuli, design, and procedure. With their eyes closed and their palm facing up, participants were stimulated on 19 different target locations on each hand (Fig. 1a). After each stimulus, they were asked to open their eyes and to indicate the location of the stimulus by pointing to a location on their hand with a finger from their other hand. Participants' responses were recorded on previously obtained photos of their hands, displayed on a tablet PC (see Figs. 1b and 1c for samples) and visible only to the experimenter. Stimulation was delivered using a 30-g probe with a round rubber tip 7 mm in diameter. The force and duration of the stimulus application were measured post hoc by placing the stimulus probe on a computer-controlled, customized force meter; across 60 trials, the mean force was 0.28 N (*SEM* = 0.009), and the mean duration was 0.24 s (*SEM* = 0.008). The 19 stimulus locations included the distal and proximal pad of each finger (5 fingers × 2 locations), 5 palm locations just proximal to the finger pads, and 4 locations on the palm (palm center, palm base, hypothenar, and thenar eminence). Each location was tested once in each of three blocks per hand, for a total of 114 trials for each participant. Trial order was randomized within blocks.

Seventeen of the WS participants and 49 of the control participants were tested on the left hand first; the rest were tested on the right hand first.

Results

We first examined the size of stimulus-localization errors across ages, groups (WS vs. control), and hand regions. Although most localization errors on the digits occurred somewhere on the stimulated digit, we also observed a significant number of errors in which participants were stimulated on one digit and then erred by indicating a location on a different digit. Therefore, in a second analysis, we considered these *cross-digit* errors, focusing on whether there was structure in their distribution.

Stimulus-localization errors across ages, groups, and hand regions. The *x* and *y* locations of the stimulus and the response for each trial were recorded by the computer, in relation to the zero-coordinate defined by the stimulation point located at the base of the palm (i.e., B in Fig. 1a). For each participant, hand length was defined as the distance from the base of the palm to the

tip (distal pad) of the middle finger (i.e., D3d in Fig. 1a). The localization error on each trial was computed as the distance between the stimulus and response locations, normalized as a proportion of hand length. These errors were then averaged over the three trials for each stimulus location on each hand.

Figure 2 shows the average size of each participant's stimulus-localization errors across the 19 stimulus locations in each hand as a function of age for the normal control and WS groups. The average error was large for normally developing 4- to 6-year-olds (.05–.08 of the hand length) and small for normal adolescents and adults (15- to 29-year-olds; about .02 of the hand length). By contrast, the WS group showed an average error that was relatively constant across age (.06–.07 of the hand length; for left and right hands combined, $M = .066$, $SEM = .003$). The participants with WS made errors that were 3 to 4 times larger than those of the normal adults and similar in magnitude to those made by normally developing 4-year-olds. Age and error magnitude were significantly negatively correlated in the control group ($r = -.296$, $p < .001$), but not in the WS group ($r = -.021$, $p = .277$).

To fit the growth patterns over age in more detail, we used an iterative nonlinear Gaussian curve-fitting algorithm, *lsqcurvefit* (MATLAB, The MathWorks, Inc., Natick, MA):

$$\boldsymbol{y} = \alpha - \delta \times e^{-\left(\frac{(\boldsymbol{x}-\beta)^2}{\gamma^2}\right)}, \quad (1)$$

where $\boldsymbol{y}$ represents a vector of stimulus-localization errors for any given number of participants, and $\boldsymbol{x}$ represents a vector for the ages of these participants (in years); α, β, γ, and δ are free parameters (see Makous, Friedman,

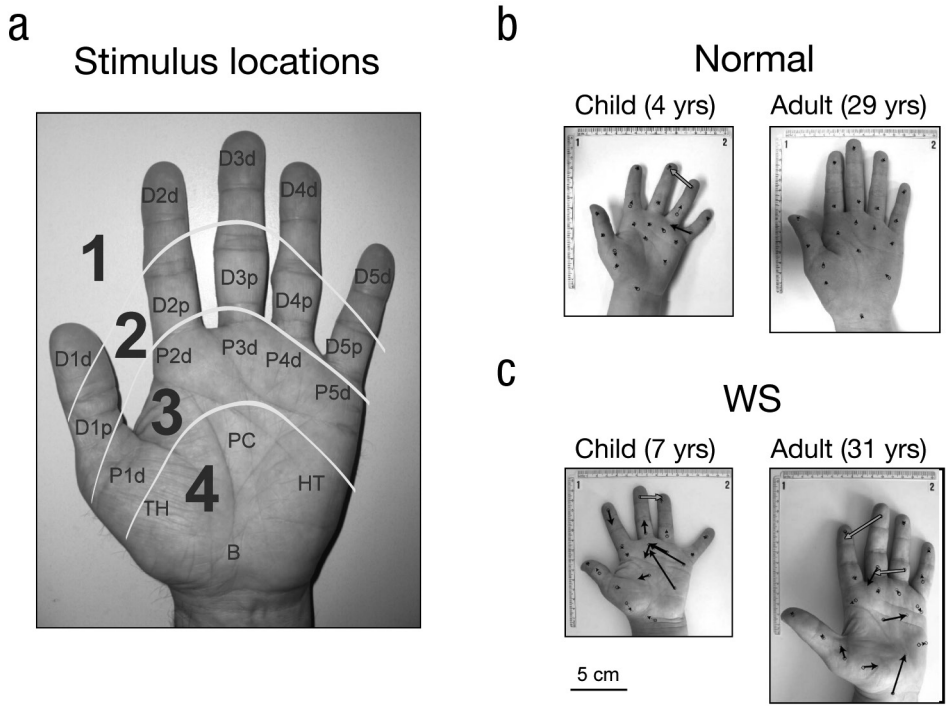

Fig. 1. Stimulus locations and examples of representative individuals' performance. The illustration in (a) shows the 19 stimulus locations in the four hand regions (1 = digit-distal; 2 = digit-proximal; 3 = palm-distal; 4 = palm-proximal). These locations are labeled by letters that denote the digits (D), the distal regions of the palm (P), the palm center (PC), the thenar eminence (TH), the hypothenar (HT), and the base of the palm (B). Further specification to the digit locations and distal locations of the palm is indicated by letters that denote distal (d) and proximal (p) locations and by numbers that denote the thumb (1), the index finger (2), the middle finger (3), the ring finger (4), and the pinky (5). For example, the stimulus location D3p represents the proximal finger pad of the middle finger. The photos on the right illustrate the performance of sample children and adults in the (b) control group and (c) Williams syndrome (WS) group. The origins of the arrows represent stimulus locations, and the endpoints, or arrowheads, represent the corresponding response locations. Thick black arrows represent localization errors with a magnitude greater than 10 mm. Gray arrows indicate cross-digit errors, in which the indicated response was on a finger other than the one that was stimulated.

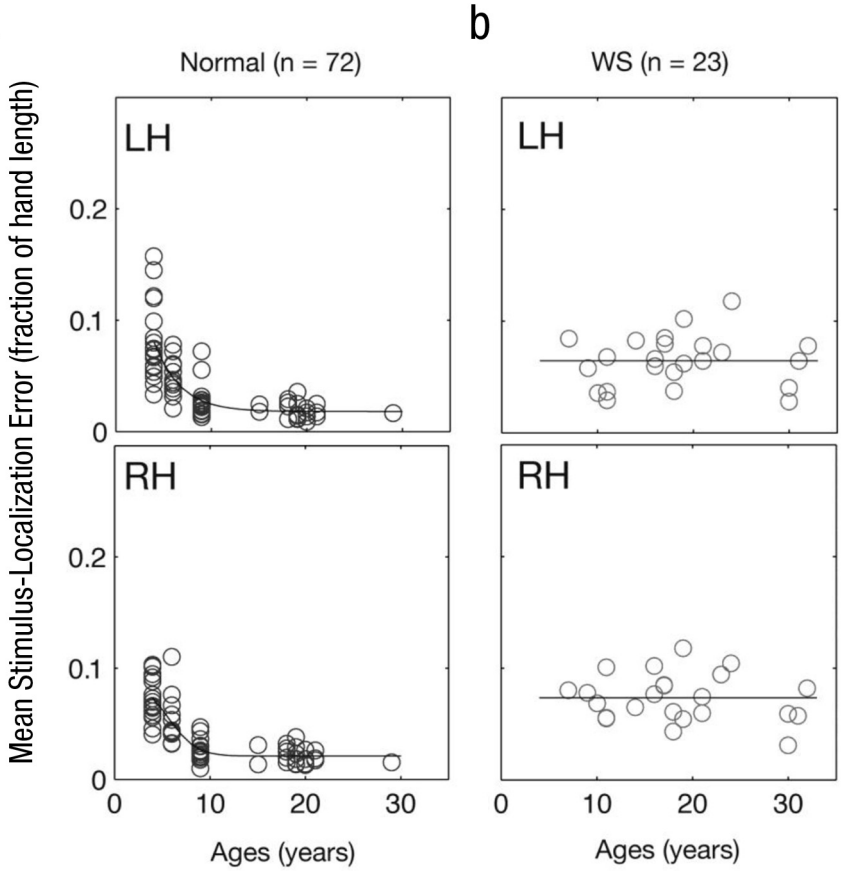

Fig. 2. Age dependence of the magnitude of the stimulus-localization errors in (a) normal participants and (b) participants with Williams syndrome (WS). Results are shown separately for the left hand (LH; top row) and right hand (RH; bottom row). Each circle represents the error of 1 participant averaged over 19 stimulus locations, which were tested three times each.

& Vierck, 1995). As a result, Equation 1 is a power function that represents the growth curve of tactile sensitivity. If growth is rapid early in development, the contribution of the exponent will be large; if there is little growth early in development, the exponent will be small: zero or close to zero.

In the control group, age had a large inverse exponential relationship to the magnitude of stimulus-localization errors: Error magnitude rapidly became smaller as age increased (Fig. 2a), which indicated significant change in the somatosensory system between childhood and adolescence. In contrast, error magnitude was fairly constant over age in the WS group (Fig. 2b). This result indicates that the development of tactile localization in people with WS is functionally arrested at an early developmental level, such that error magnitude remains similar to that of normally developing 4-year-olds. The magnitude of stimulus-localization errors was significantly correlated between the left and right hands in both the WS group ($r = .761$, $p < .001$) and the control group ($r = .825$, $p < .001$).

Because the magnitudes of the localization errors were not normally distributed in either the WS group or the control group ($ps = .001$, Lilliefors test), a Kruskal-Wallis rank-order test was used to examine differences in error magnitude between the WS group and each of the age categories in the control group. We found significant differences between the WS group and normally developing 6-year-olds and 9-year-olds, as well as normal adolescents and adults ($ps < .05$), but no difference between the WS group and the normally developing 4-year-olds ($p > .05$).

We then examined differences in error magnitudes across the four different hand regions (as defined in Fig. 1a) for each group and age category, collapsing over the two hands (Fig. 3). The distribution of errors was compared using curves fitted by a generalized-extreme-value distribution (MATLAB, The MathWorks, Inc., Natick, MA)—a family of continuous probability distribution functions that includes different asymmetric data distributions. Median values of the error-magnitude distributions were used in these comparisons when the distributions were asymmetric.

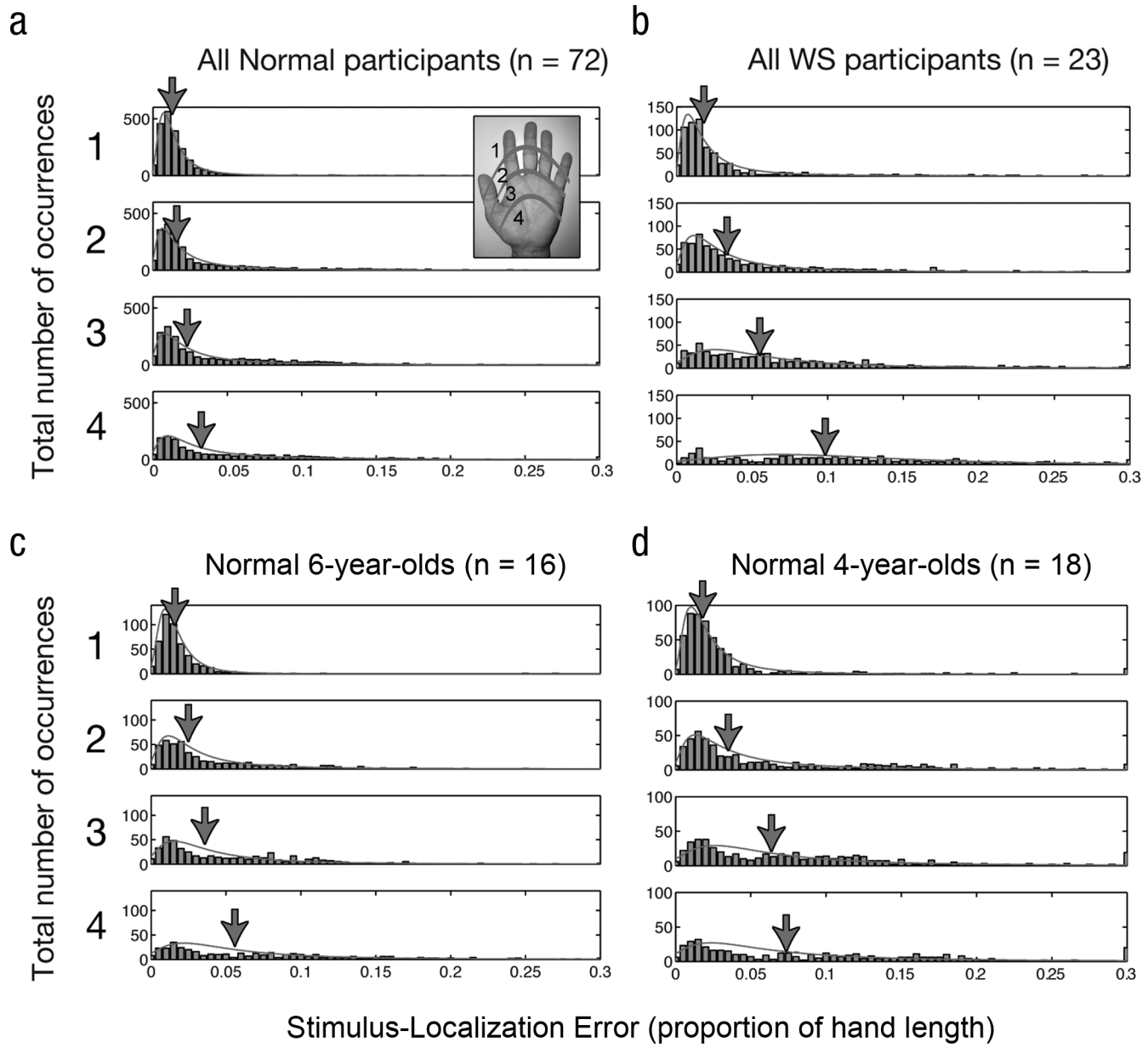

Fig. 3. Total number of stimulus-localization errors of different magnitudes in each of the four hand regions (defined in Fig. 1a). Data were combined across the left and right hands, and results are shown for (a) all participants in the control group, (b) all participants in the Williams syndrome (WS) group, (c) normally developing 6-year-olds, and (d) normally developing 4-year-olds. The gray arrows indicate the median error magnitude in each distribution. Results for normally developing 9-year-olds are not illustrated, but showed the same gradient pattern found in the control group at other ages.

We found that the error-magnitude distributions varied across ages and across hand regions in the control group (Figs. 3a, 3c, and 3d). In the WS group, the distribution also varied across hand regions (Fig. 3b). In the normal adolescents and adults, the median localization error was smallest in the fingertips (.009 of the hand length) and largest at the base of palm (.017 of the hand length). The same qualitative error gradient appeared in WS participants (.016 of the hand length at the fingertips, .099 at the base of the hand), as well as in normally developing 4-year-olds (Fig. 3d), 6-year-olds (Fig. 3c), and 9-year-olds (not illustrated). Thus, in both groups and in all age categories of the control group, the median error was smallest in the fingertips, where the mechanoreceptor

density is highest, and largest at the base of the palm, where the mechanoreceptor density is lowest (Darian-Smith & Kenins, 1980; Johansson & Vallbo, 1979).

Despite having the same qualitative error gradients, groups varied in the magnitudes of these errors (as a proportion of hand length). Pairwise comparisons showed no reliable differences between the WS participants and the normally developing 4-year-olds in any of the four hand regions (*p*s > .05, Kruskal-Wallis test). However, the WS participants differed from the normally developing 6-year-olds in error magnitude in the fingertip region, the proximal finger region, and the lower palm region (*p*s < .05). The WS participants differed from the normally developing 9-year-olds and normal adolescents and adults in each of the hand regions (*p*s < .001).

Cross-digit errors. Cross-digit errors occurred on both hands, in both the WS and control groups, and across all age categories in the control group, with the WS participants and normally developing 4-year-olds producing the highest proportions of such errors (WS group: M = .073 cross-digit errors over participants, or 101 errors total; 4-year-olds: M = .088 over participants, or 95 errors total). Normally developing 6-year-olds (M = .033 over participants, or 36 errors total) and 9-year-olds (M = .011 over participants, or 11 errors total) and normal adolescents and adults (M = .008 over participants, or 10 errors total) produced smaller proportions of cross-digit errors. The cross-digit errors were organized in a confusion matrix and normalized by the total number of errors per stimulus location to examine the distribution of error responses for each target location (see Fig. 4 for results from the WS group and normally developing 4- and 6-year-olds).

We first examined the effect of age and group on the proportions of cross-digit errors. Because the proportions were not normally distributed for either the WS (p = .005, Lilliefors test) or the control groups (p = .001, Lilliefors test), we carried out a nonparametric median rank-order test, finding a significant effect of age in the control group (p < .001), but not in the WS group (p > .05). The proportion of cross-digit errors in the WS group was significantly different from the proportion of such errors in all age categories of the control group (*p*s < .001) except for the 4-year-olds (p = .911). The proportions for normally developing 4- and 6-year-olds were significantly different from the proportions for both normally developing 9-year-olds and normal adolescents and adults (*p*s < .05 for all pairwise comparisons).

To further understand the developmental trajectory of these cross-digit errors, we fitted the proportions of errors per participant to the same iterative nonlinear Gaussian curve used in our analysis of error magnitude over age. The same pattern found in the localization errors was found in the cross-digit errors. The proportion of cross-digit errors showed a large inverse exponential relationship with age in the control group (Fig. 5a), but not in the WS group (Fig. 5b). In the WS group, the proportion of cross-digit errors remained fairly constant over age.

We next considered whether there were qualitative differences between the errors of WS and normal participants. First, we examined error *distance*, exploring

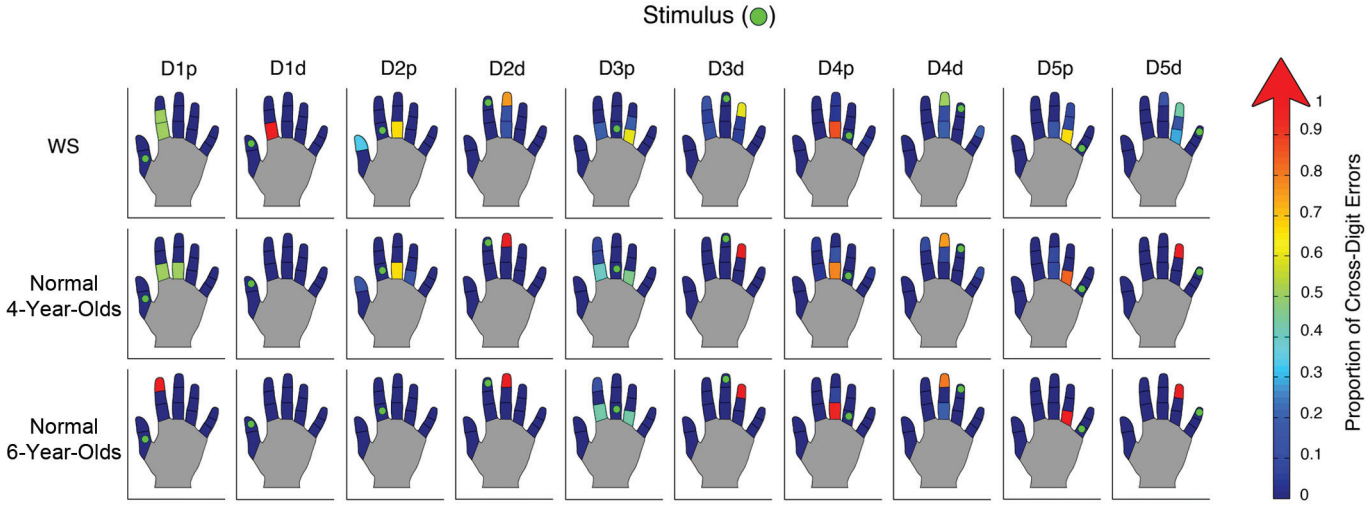

Fig. 4. Distributions of cross-digit errors across both hands in Williams syndrome (WS) participants, normally developing 4-year-olds, and normally developing 6-year-olds. Stimulus locations are indicated by green dots, and the proportion of cross-digit responses is color-coded on each response location according to the key at the right. For example, participants with WS who were stimulated at the Digit 1 proximal location (top row, left-most box) made cross-digit errors that occurred on the proximal and medial locations of the second finger, with .50 of these errors occurring at each of the two indicated locations. Note that although stimulus locations did not include the middle pads of the fingers, participants sometimes reported stimulation at these locations.

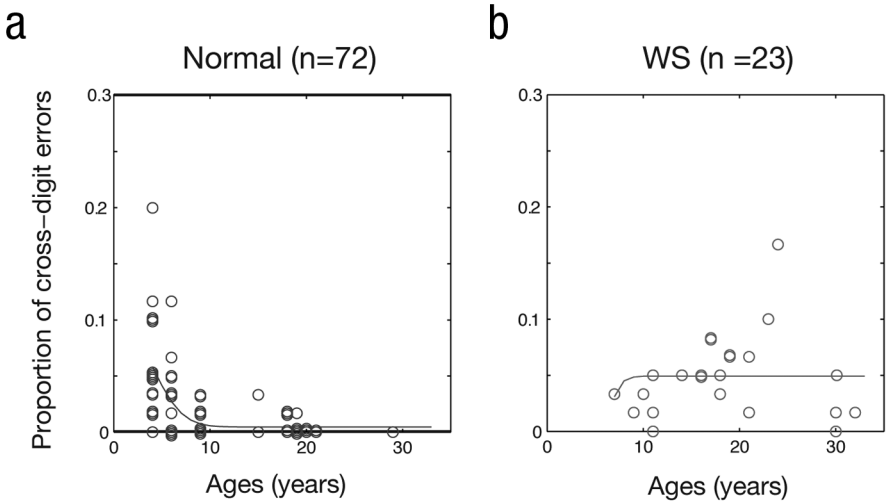

Fig. 5. Age dependence of the proportion of cross-digit errors in (a) normal participants and (b) participants with Williams syndrome (WS). Data were fitted using the same curve-fitting analysis as for stimulus-localization errors (Fig. 2a).

whether the cross-digit errors migrated to an adjacent digit or to more distant digits. Distance was determined by counting the number of horizontal or vertical units separating the target and response locations, with a unit of distance defined as one horizontal or vertical translation from the target location to another digit's proximal, medial, or distal location.[1]

In the WS group and in all age categories of the control group, cross-digit errors tended to occur on a neighboring digit, specifically, 1 unit away from the target location. One-unit errors accounted for 85% of the cross-digit errors in the WS group (86 out of 101 errors), 89% of the cross-digit errors among normally developing 4-year-olds (85 out of 95 errors), 92% of the cross-digit errors among normally developing 6-year-olds (33 out of 36 errors), and 100% of the cross-digit errors among both normally developing 9-year-olds (11 out of 11 errors) and normal adolescents and adults (10 out of 10 errors).

Given that the large majority of cross-digit errors were at 1-unit distance, we further classified 1-unit errors according to *direction*, to determine the frame of reference that best explained them. Rapp, Hendel, and Medina (2002) showed that tactile localization errors in 2 patients with lesions involving somatosensory cortex were characterized by a shift toward the midline of the hand, a pattern suggesting a *hand-based* reference system. We therefore examined whether errors migrated toward a hand midline or toward a body midline, the latter of which would instead indicate a body-based reference frame. For the hand-based reference system, we assumed a midline bisecting the upturned palm at the middle finger and categorized errors as migrating toward or away from this midline. In the case of stimulation to the thumb or pinky, cross-digit errors necessarily were in a direction toward the midline of the hand; for completeness, these errors were also included.

If the reference frame was hand based, stimulation to the proximal pad of the ring finger would be expected to yield reports of stimulation to the proximal pad of the middle finger, rather than to the pinky. In contrast, and given that testing was carried out with hands in a palms-up position, a body-based reference system would be expected to yield shifts toward the body midline (e.g., stimulation to the proximal pad of the ring finger would yield reports of stimulation to the proximal pad of the pinky, rather than to the middle finger). Alternatively, if a body-based reference system with a canonical palms-down representation of the hand was adopted, stimulation to the proximal pad of the index finger would be expected to yield reports of stimulation to the proximal pad of the thumb, rather than to the middle finger.

A hand-based frame of reference accounted for most 1-unit cross-digit errors (as depicted in Fig. 4): 97% of errors (83 out of 86) in the WS group, 99% (84 out of 85) among normally developing 4-year-olds, 100% (33 out of 33) among normally developing 6-year-olds, 100% (11 out of 11) among normally developing 9-year-olds, and 100% (10 out of 10) among normal adolescents and adults. By comparison, the body-centered, palm-up pattern accounted for 45% of 1-unit cross-digit errors (39 out of 86) in the WS group, 22% (19 out of 85) among normally developing 4-year-olds, 21% (7 out of 33) among normally developing 6-year-olds, 36% (4 out of 11)

among normally developing 9-year-olds, and 30% (3 out of 10) among normal adolescents and adults. Finally, the body-centered, palm-down pattern accounted for 55% of 1-unit cross-digit errors (47 out of 86) in the WS group, 78% (66 out of 85) among normally developing 4-year-olds, 79% (26 out of 33) among normally developing 6-year-olds, 64% (7 out of 11) among normally developing 9-year-olds, and 70% (7 out of 10) among control adolescents and adults.[2] In sum, a hand-based reference system accounted for almost all of the 1-unit cross-digit errors and best explains those data for the WS group and all age categories of the control group.

Experiment 2

The improvement in tactile localization with age in the control group and the lack of improvement with age in the WS group raises the question of whether the relatively poor performance of younger normally developing children and people with WS could have been due to general task demands, including failure to understand the task, memory limitations, or difficulties in pointing to the hand locations. To examine this possibility, we tested the ability of normally developing 4-year-olds and people with WS to point to stimulus locations on photos of their hands.

Method

Participants. Ten people with WS (mean age = 18 years, range: 9–33 years) and 11 normally developing 4-year-olds (mean age = 4 years 5 months, range: 4 years 3 months to 4 years 11 months) completed Experiment 2. Eight of the participants with WS and none of the normally developing 4-year-olds had also completed the task in Experiment 1.

Stimuli, design, and procedure. The task in Experiment 2 was identical to the task in Experiment 1, except that, using the probe, the experimenter touched a photo of each participant's hand while that participant watched. Participants were instructed to point to the location on the photo that the probe had touched.

Results

Localization errors were analyzed as in Experiment 1. Both the WS participants and the normally developing 4-year-olds showed significantly larger mean error magnitudes in Experiment 1 (.066 and .071 of the hand length, respectively) than in Experiment 2 (.023 and .028 of the hand length, respectively; Kruskal-Wallis independent-samples test, ps < .001). There was also no significant effect of age on the magnitude of localization errors in the WS group in Experiment 2 (left hand: $r = -.55$, $p = .083$; right hand: $r = -.41$, $p = .205$).

The WS group made no cross-digit errors, and the 4-year-olds made only a small number (12 out of 660 responses). The different patterns of performance between the tactile and control tasks indicate that the relatively poor performance of the WS participants and the normally developing 4-year-olds in Experiment 1 was not likely due to misunderstanding the task or to limitations in motor or memory skills.

General Discussion and Conclusions

We observed three striking patterns for tactile localization through development and under genetic deficit. First, we found that tactile localization accuracy develops gradually in normally developing children, reaching adult levels by age 10 to 12. This maturational endpoint is similar to that of tactile spatial acuity (Bleyenheuft, Cols, Arnould, & Thonnard, 2006). Second, we found that, by contrast, at all ages people with WS performed at the level of normally developing 4-year-olds; their ability to localize tactile stimuli did not improve with age. We ruled out the possibility that their tactile localization errors were due solely to task misunderstanding or limitations in motor or memory skills: Both participants with WS and normally developing 4-year-olds showed significantly smaller errors in a nontactile, comparable visual control task. The large localization errors in people with WS, from childhood through adulthood, are consistent with the findings that they show developmental arrest in a variety of dorsal-stream functions, including visual-spatial construction, attentive tracking, object recognition, and visual-manual action (Landau & Hoffman, 2012). Our finding that the spatial deficit extends to tactile localization is striking, suggesting that the missing genes in WS play a broad role in the growth of capacities involving spatial representation.

Third, we found two remarkably systematic patterns of localization and error that were common to both young normally developing children and people with WS. One of these patterns was an accuracy gradient with error magnitude increasing from fingertips to palm. This gradient was more pronounced in the WS group and normally developing 4-year-olds than in older normally developing children or normal adolescents and adults. The gradient is similar to other known tactile gradients; for example, higher spatial acuity in the fingertips, compared with the palm, is due to higher mechanoreceptor density in the fingertips (Darian-Smith & Kenins, 1980; Johansson & Vallbo, 1979). The second systematic pattern was found for cross-digit errors. Such errors most frequently involved reporting that a digit adjacent to the stimulated digit had been touched. Moreover, these errors tended to migrate toward the midline of the hand (rather than toward the

body), which suggests that the digits are represented in a hand-centered reference frame.

These findings reveal that the representation of digits in somatosensory cortex not only is organizationally constrained by age 4, but also undergoes refinement over development, which results in increased precision in distinguishing individual digit locations. These findings also reveal that, unlike normally developing individuals, individuals with WS do not undergo full developmental refinement of the hand representation, exhibiting quantitative and qualitative error patterns similar to those of normally developing 4-year-olds. In combination with existing evidence of experience-dependent plasticity in somatosensory cortex (Schwenkreis et al., 2007; Wang, Merzenich, Sameshima, & Jenkins, 1995), these findings indicate that developmental changes in the representation of the human hand are linked to both genes and experience.

The mechanisms by which the missing genes in WS cause the broad spatial disorder are unknown. However, the missing genes *LIMK1* and *CYLN2*, which regulate cytoskeletal assembly in neurons, are ubiquitous and therefore likely to affect both peripheral and central nervous systems. The tactile localization deficit in WS may be based on abnormalities of the peripheral afferent neurons responsible for mechanoreception, especially Slowly Adapting type 1 (SA1) afferent neurons, which likely contribute to precise localization of tactile stimuli (Hsiao, Johnson, & Yoshioka, 2003; Johnson & Yoshioka, 2002). Alternatively, if cortical somatotopic organization is distorted in WS, this might result in a lower discriminability of the spatial locations of tactile stimuli.

Our findings indicate that tactile localization undergoes a lengthy developmental trajectory in humans, reaching adult levels only well after age 9. This may seem surprising; however, prolonged developmental trajectories have also been documented for other aspects of spatial representation, for example, flexible use of reference systems (Nardini, Atkinson, Braddick, & Burgess, 2008) and global visual shape processing (Scherf, Behrmann, Kimchi, & Luna, 2009). Moreover, the substantial developmental course we have suggested for the somatosensory system is consistent with the few available studies on somatosensory development in nonhuman primates (Krubitzer & Kaas, 1988; Pons, Wall, Garraghty, Cusick, & Kaas, 1987).

To our knowledge, this is the first detailed report on the somatosensory representation of hand and digits in normally developing children, its change over age, and the impact of missing genes on this representation. These findings can form the foundation for further study regarding the precise roles of genes and experience in the development of one of human beings' most important assets: the brain's representation of the hand.

Author Contributions

Design of the experiments was carried out by T. Yoshioka, B. Rapp, and B. Landau; testing was carried out by M. R. Dillon and B. Landau; analyses were carried out by T. Yoshioka, M. R. Dillon, G. C. Beck, and B. Landau; writing and final editing was done by T. Yoshioka, M. R. Dillon, B. Rapp, and B. Landau.

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Declaration of Conflicting Interests

The authors declared that they had no conflicts of interest with respect to their authorship or the publication of this article.

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Notes

1. Although target locations were restricted to the proximal and distal pads of each finger, occasionally participants reported stimulation to the middle segment of another finger. These errors were rare (WS participants: 12 out of 101; normally developing 4-year-olds: 7 out of 95; normally developing 6-year-olds: 1 out of 36; normally developing 9-year-olds: 1 out of 11; normal adolescents and adults: 0 out of 10). For our analysis of the distance and direction of cross-digit responses, we grouped these middle-pad responses with either distal or proximal responses depending on the target location: For distal target locations, a cross-digit middle-pad response was grouped with cross-digit distal responses; for proximal target locations, a cross-digit middle-pad response was grouped with cross-digit proximal responses.

2. For these three frames of reference, the percentages of errors sum to more than 100% because some errors are consistent with more than one frame of reference (e.g., a shift from the proximal pad of the index finger to the proximal pad of the middle finger follows both hand-centered and body-centered, palm-up reference frames).

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