Strategies and Correlates of Jigsaw Puzzle and Visuospatial Performance by Persons With Prader-Willi Syndrome

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Abstract

Some individuals with Prader-Willi syndrome exhibit strengths in solving jigsaw puzzles. We compared visuospatial ability and jigsaw puzzle performance and strategies of 26 persons with Prader-Willi syndrome and 26 MA-matched typically developing controls. Individuals with Prader-Willi syndrome relied on piece shape. Those in the control group used a different, picture-focused strategy. Individuals with Prader-Willi syndrome performed better than did the control group on an achromatic interlocking puzzle, whereas scores on puzzles with pictures (interlocking or noninterlocking) did not differ. Visuospatial scores related to performance on all puzzles in the control group and on the noninterlocking puzzle in the Prader-Willi syndrome group. The most proficient jigsaw puzzlers with Prader-Willi syndrome tended to be older and have shape-based strategies.

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Several intellectual disability syndromes are associated with unusual patterns of cognitive strengths and weaknesses. Prader-Willi syndrome, a rare genetic disorder occurring in approximately 1 in 15,000 live births, is characterized by mild to moderate intellectual disability and distinctive physical and behavioral features, including hyperphagia, increased risks of obesity, compulsivity, and other maladaptive behaviors (see Dykens, Hodapp, & Finucane, 2000). The mean IQ of people with Prader-Willi syndrome is around 70, with about 5% of scores considered "average" in typical populations (85 and above). Short-term memory may be an area of particular cognitive weakness and long-term retrieval may be relatively strong (Conners, Rosenquist, Atwell, & Klinger, 2000; Warren & Hunt, 1981). Individuals with Prader-Willi syndrome also have a significant weakness on the Sequential Processing subscale of the Kaufman Assessment Battery for Children (K-ABC) and a relative weakness on the Spatial Memory subtest of the Simultaneous Processing subscale (Dykens, Hodapp, Walsh, & Nash, 1992).

Persons with Prader-Willi syndrome have been reported to be particularly adept at assembling jigsaw puzzles, and parental reports of such skills are "supportive criteria" that lead to increased suspicion of Prader-Willi syndrome among diagnosticians (Holm et al., 1993). In the first formal study of jigsaw puzzle abilities in Prader-Willi syndrome, Dykens (2002) found that her participants who had this syndrome outperformed both typically developing, chronological age (CA) matched controls and CA- and IQ-matched controls with mental retardation. Although reasons for good puzzle assembly skills remain unclear, researchers have posited that such skills are related to visuospatial ability, a cognitive area that is considered less impaired relative to their other cognitive skills (Gabel et al., 1986). In prior research, individuals with Prader-Willi syndrome outper-

formed mixed etiology IQ-matched controls on the visuospatial subscales of a number of standardized intelligence tests and performed closer to the typical level specified by test norms than they did on other subscales (Dykens, 2002; Dykens et al., 1992). Although visuospatial ability may be considered a relative strength compared to other areas of the Prader-Willi syndrome cognitive profile, visuospatial abilities do not appear to be spared relative to those of CA-matched controls (Dykens, 2002).

Claims that visuospatial abilities are a strength in Prader-Willi syndrome are also problematic because the visuospatial parts of standardized intelligence tests used in prior studies (e.g., Object Assembly and Block Design from the Wechsler Intelligence Scale for Children-III, triangles from the K-ABC) resemble jigsaw puzzles. In these tests, participants must assemble the silhouette of an object from a set of pieces and copy a design by putting together colored shapes. The visuospatial IQ subscales, therefore, appear to test the assembly of puzzle-like stimuli (a known skill in Prader-Willi syndrome), but they do not separately assess the three main spatial abilities identified in research with typical populations: spatial perception, mental rotation, and spatial visualization (Liben et al., 2002; Linn & Petersen, 1985; Scali, Brownlow, & Hicks, 2000; Voyer, Voyer, & Bryden, 1995). *Spatial perception* involves accurately perceiving a spatial relation relative to the orientation of one's own body, and tests of this ability require that participants ignore a rotated frame of reference (e.g., the tilted bottles in the water level task described below). In *mental rotation* tasks, individuals must mentally rotate or reorient an object. *Spatial visualization* tasks (e.g., origami-like paper folding and embedded figures tests) involve multistep operations on spatial information, the use of analytic strategies, and the flexible adaptation of a set of solution procedures.

Although these three spatial domains have been examined separately in individuals with typical development, they have not been investigated in people with Prader-Willi syndrome nor have they been connected to puzzle assembly skills in persons with or without intellectual disabilities. Thus, our first aim in the present study was to identify how persons with Prader-Willi syndrome and mental age (MA) matched controls fare on spatial perception, mental rotation, and spatial visualization tasks and how these tasks relate to puzzle assembly performance in both groups.

Superior jigsaw puzzle performance by persons with Prader-Willi syndrome may also be associated with specific problem-solving behaviors or strategies. Dykens (2002) reported that compared to typical controls, participants with Prader-Willi syndrome looked less at the box-top picture accompanying the puzzle, were less likely to try to force pieces together, and were more likely to start with the edge pieces. In Dykens' study, these differences, based on in-vivo observations that were not videotaped for reliability analyses, were presented as preliminary descriptions of behaviors deserving of more systematic analysis. Unusual puzzle-solving behaviors, such as failing to refer to the picture, could indicate that individuals with Prader-Willi syndrome remember the picture and do not need to look at it again or that they find it detrimental for some reason to go back and forth between the picture and puzzle pieces (for instance, if previously documented short-term and spatial memory deficits make it difficult to hold both in mind simultaneously). Alternatively, persons with Prader-Willi syndrome may simply be more attuned to the information contained in the puzzle pieces themselves. In the current study we manipulated the presence of information from the puzzle piece (e.g., shape, color) to identify properties of puzzles that are associated with enhanced performance. Despite the long-term popularity of jigsaw puzzles and their potential as a window into the development of visuospatial skills, such fine-grained analysis of puzzle features and assembly strategies has not been conducted with typically developing children or children with this developmental disability.

Because all persons with Prader-Willi syndrome are not equally proficient with jigsaw puzzles, an area of interest involves correlates of within-syndrome variability. Chronological age, for example, was modestly associated with puzzle performance in Dykens' (2002) study of people with Prader-Willi syndrome aged 5 years to adulthood (M = 14 years). Compared to others with disabilities, those with Prader-Willi syndrome are more fascinated by puzzles and have more experience with them, suggesting that increased exposure might enhance performance (Rosner, Hodapp, Fidler, Sagun, & Dykens, 2004; Sellinger, Dykens, & Hodapp, 2006). Puzzle building and strategy development could also relate to certain compulsive symptoms often noted in those with Prader-Willi syndrome (Dykens, Leckman, & Cassidy, 1996), especially the need for exactness and

getting things "just right" (in a jigsaw puzzle, it is possible to achieve an exact and perfect solution). Finally, within-syndrome variability in puzzle skills may be associated with genetic subtypes. Most cases (70%) of Prader-Willi syndrome are caused by paternal deletions of chromosome area 15q11-q13, and approximately 25% are due to maternal uniparental disomy (UPD). Both subtle and blatant phenotypic differences have been found across these subtypes, including better developed verbal skills for persons with UPD than for those with deletions, and superior visuospatial performance by persons with deletions than for those with UPD (Roof et al., 2000).

We designed the current study, therefore, (a) to identify how persons with Prader-Willi syndrome and MA-matched controls fare on tasks that tap all three major areas of visuospatial ability (spatial perception, mental rotation, spatial visualization), and how these tasks relate to jigsaw puzzle performance; (b) to identify strategies that participants in each group use to solve puzzles by varying puzzle stimuli (traditional jigsaw puzzle, achromatic (blank) puzzle, noninterlocking puzzle) and by coding videotaped puzzle-solving behaviors; and (c) to examine such participant correlates of puzzle performance as age, MA, IQ, gender, puzzle experience, compulsivity, and genetic subtype of Prader-Willi syndrome.

Method

Participants

The participants were 26 individuals with Prader-Willi syndrome (15 males, 11 females; mean CA = 20.98 years, *SD* = 12.15; mean IQ = 68.40, *SD* = 14.48) and 26 typically developing individuals (14 males, 12 females; mean CA = 6.73 years, *SD* = 1.82; mean IQ = 108.25, *SD* = 11.84). We individually matched all participants by using MA obtained from the Kaufman Brief Intelligence Test (K-BIT) (Kaufman & Kaufman, 1990). The average MA was 7.84 years (*SD* = 2.50) for the group with Prader-Willi syndrome and 7.83 years (*SD* = 2.47) for the control group. Matches were made so that each participant with Prader-Willi syndrome had a control match who was within 9 months of his or her MA (mean MA difference = 3.75 months). Typically developing control participants, recruited from a local database compiled from state birth records and by flyers distributed in the community, included 23 European Americans, 2 African Americans, and 1 Australian of European descent.

Participants with Prader-Willi syndrome included 23 European Americans, 2 Asian Americans, and 1 African American. Diagnoses were based on genetic testing, with 16 persons having paternal deletions, 5 having maternal UPD, and 5 having less common variants (2 microdeletions, 1 imprinting mutation, 1 translocation, and 1 subtype unknown, diagnosed by methylation). Participants were recruited as part of a larger, longitudinal study through local contacts and clinics as well as through the Prader-Willi Syndrome Association. The vast majority lived at home with their parents; however, some, particularly the older individuals, resided in group homes. Parents and participants were told that solving puzzles would be one of the study activities, but puzzles were not highlighted as a main focus of the research.

Procedure

Parents filled out questionnaires while their child completed the 90-minute visuospatial test battery. Parts of the sessions were videotaped and coded by a student assistant who was unaware of the study hypotheses. To optimize performance, participants were offered breaks as needed, and tasks were presented in a set order that interspersed short, hands-on (i.e., more engaging) tasks with repetitive or demanding tasks requiring verbal responses. The order was K-BIT placement tasks, water level task, jigsaw puzzles, mental rotation task, motor-free visual perception test, and Lego building. Presentation orders within task were counterbalanced and/or randomized whenever possible.

Prader-Willi Syndrome and Control Group Measures

Kaufman Brief Intelligence Test. This test allowed for MA-matching between groups. Designed for research with and screening of persons aged 4 to 90, the K-BIT consists of two subscales (Verbal and Matrices). It has been used successfully with individuals with Prader-Willi syndrome in prior studies (e.g., Ly & Hodapp, 2005).

Spatial perception: Water level task. A multiple-choice version of Piaget and Inhelder's (1956) water level task (Figure 1) provided a measure of spatial perception (see Vasta & Liben, 1996). For each trial, participants saw drawings of five identical bottles tilted at the same angle. This angle varied

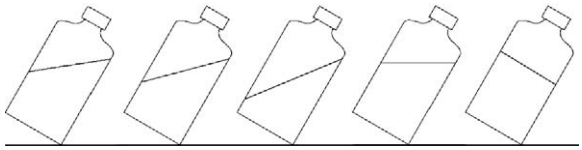

Figure 1. Water level task with bottles tilted at 45° angle. Choice 4 is correct and 5 is a "parallel bottle" choice, with the water level parallel to the bottom of the bottle instead of the tabletop.

across the four trials (15°, 30°, 45°, or 60°). A line beneath the bottles (representing the tabletop) served as a reference point against which to compare a different angled line within each bottle (representing the water level). Participants were asked to point to the bottle that "shows where the top of the water would be." Answers were scored on a scale from correct (parallel to the tabletop, 4 points) to the angle farthest from correct (0 points), for a total of 16 possible points. We also noted the number of bottles chosen in which the water level was parallel to the bottom of the bottle (a common incorrect answer—see Piaget & Inhelder, 1956).

Mental rotation task: Rotated monkeys. As a measure of mental rotation ability, we employed a version of Estes' (1998) computer-based task in which two monkeys appear on a computer screen, and participants indicate (using 2 keyboard keys) whether the monkeys are holding up the same or different arms. The monkey on the left was upright and always faced forward. The monkey on the right was rotated 0 to 180 degrees from upright in 45° increments. On some trials, this monkey was also facing the opposite direction, requiring rotation in both planes (a variation not used by Estes). After 10 practice trials, participants completed 3 blocks of 10 test trials, with backward facing monkeys appearing on 10 of the 30 total trials. Trials were presented in a pseudo-random order that prevented stimuli with the same degree of rotation from appearing on consecutive trials.

Spatial visualization: Motor-Free Visual Perception Test. During pilot testing, participants were given the entire Motor-Free Visual Perception Test (third edition, Colarusso & Hammill, 2003). At-chance scores on the latter half of the test suggested the need to shorten it. Therefore, we chose 13 items similar to standard embedded figures tasks (e.g., Witkin, 1950) as a test of spatial visualization. For instance, participants needed to locate a target shape hidden in a display of overlapping, intersecting lines and identify how many of the shapes were present. Six items from another test section required participants to mentally complete an incomplete figure and select a matching figure from an array of four choices. Another section (5 items) required participants to identify the one figure from a set of four that was different. All 25 items forming our spatial visualization test required multistep operations on spatial information and the use of analytic skills—distinguishing features of spatial visualization.

"Real-life" visuospatial task: Map reading. A modified version of Laurendeau and Pinard's (1970) map placement task was used to measure participants' ability to complete a visuospatial problem encountered in real life. The task involved a portable 3-D Styrofoam "terrain" (50 cm $\times$ 50 cm) and a matching map (27 cm $\times$ 27 cm). Four placement locations within the terrain created a continuum of difficulty due to the presence or absence of distinctive landmarks (e.g., trees, roads, or houses). To increase motivation, participants were told a story about a Lego man looking for buried treasure. They were asked to "Draw an *X* on the map where the Lego man is standing" as the man was moved to the four different terrain locations. These terrain-to-map trials required representational insight into the relation between the map and terrain as well as conversion of scale due to size differences. Next, on 4 map-to-map trials, participants saw the Lego man placed on a map and had to identify his location on a second, identical map. These trials, requiring one-to-one matching across identical representations, were expected to be easier. We scored placements using a multistep coding scheme reflecting how close the mark was to the target location (maximum per trial = 4).

"Real-life" visuospatial task: Lego building. In another measure of spatial ability on a task that occurs in real life, participants were given 5 min to assemble a duck figure from a set of 17 Lego blocks using a diagram. Each block was assigned a point for being placed in the correct row and a point for being in the correct position within the row (determined in relation to the row below) for a maximum potential score of 34 points.

Jigsaw puzzles. Participants were given 5 min to assemble 3 puzzles created for this study: a traditional 30-piece face jigsaw puzzle (Figure 2a); a blank, white, 30-piece jigsaw puzzle (Figure 2b); and a 17-piece, noninterlocking face puzzle that contained the same picture as the traditional puz-

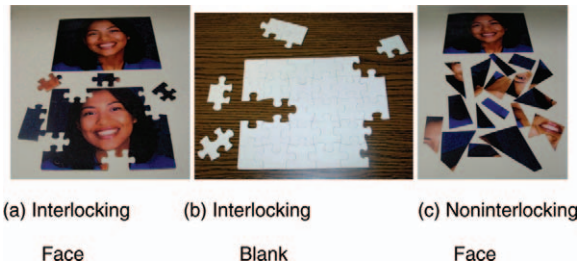

Figure 2. Puzzle pictures.

zle (Figure 2c). Individuals with Prader-Willi syndrome often have difficulty with fine- or gross-motor control; therefore, we chose not to make the noninterlocking pieces smaller and more numerous (they tended to shift when bumped). The traditional puzzle provided strong pictorial and shape cues; the blank puzzle, only shape cues; and the noninterlocking puzzle, strong pictorial cues but only weak shape cues (i.e., shape alone did not constrain the placement of most pieces). Participants could refer to a copy of the picture (the box-top picture) when completing the two face puzzles. Because a number of individuals with Prader-Willi syndrome finished the puzzles before time expired, we chose to use the number of pieces assembled in 3 minutes for comparisons of puzzle performance. Data from the full, 5-min session revealed the same pattern of performance, but effect sizes were not as strong due to the ceiling effect.

Videotapes of participants assembling the puzzles were coded for two key behaviors: (a) whether participants started with edge pieces or inside pieces (scored as the first 5 pieces of each of the 2 interlocking puzzles that participants tried to assemble) and (b) the number of looks that the participants directed toward the box-top picture (for the 2 puzzles with pictures). Edge-piece coding was not done for the noninterlocking puzzle; it did not contain obvious edge pieces because all edges were straight. Coding of the number of looks to the picture was done for the full 5 min because this provided more instances of a relatively low incidence behavior. This choice could have underestimated the number of looks that would have been made by individuals who finished the puzzles early, had they more puzzles to do; however, most were individuals with Prader-Willi syndrome who performed best on the traditional jigsaw puzzle while making almost no looks at the picture and whose looking scores, therefore, would be unlikely to change regardless of the timeframe coded. One coder scored all of the videos, and a second coder scored 50% of them. There was a high degree of interrater reliability on the edge pieces coding for the jigsaw/face and jigsaw/blank puzzles, Spearman rs = .94 and .92, respectively, as well as for the number of looks to the jigsaw/face puzzle and noninterlocking/face puzzle, rs = .81 and .83, respectively.

Parental measures. Parents completed a Leisure Activities Questionnaire and the Yale-Brown Obsessive-Compulsive Scale (Goodman et al., 1989). The Leisure Activities Questionnaire was used to ascertain whether puzzle assembly skill and visuospatial scores were related to experiences with a wide array of activities (e.g., playing computer games; doing hidden figures puzzles such as *Where's Waldo* and *Highlights*). Parents rated their child on 50 common activities using a 5-point Likert scale, ranging from 1 (*never does activity*) to 5 (*does activity daily*). As a way to look at past experience, parents also answered yes or no to the question, "Has your child ever been very interested in this activity?" To determine whether either of our groups pursued or avoided the spatial activities from the list, 5 typically developing adults with psychology backgrounds, blind to the purpose of the study, selected the 10 questionnaire items that they considered the most taxing and another 10 they considered to be the least taxing in terms of spatial ability. On the 10 items receiving the most selections in each category, we summed scores for each of our participants to create indexes of their interest in spatial and nonspatial activities.

The Yale-Brown Obsessive-Compulsive Scale was used to determine whether compulsive symptoms, known to be part of the Prader-Willi syndrome phenotype, related to participants' skill at assembling jigsaw puzzles. This scale is used to assess the severity of obsessions and compulsions, independent of the number and type manifested. It is composed of Likert scale items from Obsessions and Compulsions subscales and has been widely used in previous research on Prader-Willi syndrome (e.g., Dykens et al., 1996; Holsen & Thompson, 2004). The number and severity of compulsive symptoms were used in data analyses.

Results

Visuospatial Abilities and Relations to Jigsaw Puzzles

We used t tests, which revealed that the MA-matched typically developing group scored signif-

icantly higher than those with Prader-Willi syndrome on every measure of spatial ability (see Table 1 for means and *SD*s) except the water level task, on which neither group's scores differed from chance. On the latter task, children in the control group systematically chose bottles with the water level parallel to the bottom (resulting in the lowest score) or completely avoided them and performed well on the task; 10 of 26 control participants, but only 2 of 26 in the group with Prader-Willi syndrome, chose these parallel bottles on the majority of trials, χ^2 (1, N = 52) = 6.93, p = .008. The pattern of choices in the group with Prader-Willi syndrome was more consistent with random selection (11 of 26 chose one parallel bottle, the number expected by chance). Both groups scored significantly above chance on the other multiple-choice measures of visuospatial ability (the Motor-Free Visual Perception Test and the Mental Rotation Task).

More consistent and stronger correlations emerged between the visuospatial tasks and jigsaw puzzle scores in the typically developing group than in the Prader-Willi syndrome group. As shown in Table 2, correlations in the typically developing group were found between all tasks and scores on the traditional and noninterlocking puzzles with pictures, whereas correlations in the Prader-Willi syndrome group were smaller and mostly involved the noninterlocking puzzle.

Strategies: Jigsaw Puzzles

A 2 (group) × 3 (puzzle type) repeated measures ANOVA, with puzzle type as a within-subjects variable, revealed a significant main effect of puzzle type, F(2, 96) = 40.94, $p < .001$, and a significant interaction, F(2, 96) = 6.04, p = .003, but no main effect of group. Both groups performed best on the traditional, 30-piece jigsaw/face puzzle, with the Prader-Willi syndrome group somewhat, but not significantly, ahead of the typically developing group, 11.72 pieces (*SD* = 7.32) to 8.84 pieces (*SD* = 5.22). However, on the jigsaw/blank puzzle, individuals with Prader-Willi syndrome assembled almost twice as many pieces, 5.73 (*SD* = 5.71) versus 3.00 (*SD* = 3.23), t(49) = 2.09, $p < .05$, effect size (ES; Cohen's d with Hedges correction) = .58. In contrast, scores on the noninterlocking/face puzzle favored the typically developing group, although the difference did not reach significance (6.20 pieces, *SD* = 4.00, and 4.77 pieces, *SD* = 3.15, respectively). These distinctive patterns were also revealed by nonparametric tests. The group with Prader-Willi syndrome scored highest on the jigsaw/face puzzle, followed by the jigsaw/blank puzzle, and lowest on the noninterlocking/face puzzle, Friedman test of rank scores, χ^2 (2, N = 25) = 25.55, $p < .001$. Typically developing children, however, scored highest on the jigsaw/face puzzle, then the noninterlocking/face puzzle, and lowest on the jigsaw/blank puzzle, Friedman χ^2 (2, N = 25)= 22.52, $p < .001$.

Strategies: Puzzle Assembly Behavior

A 2 (group) × 2 (puzzle: jigsaw/face vs. jigsaw/blank) repeated measures ANOVA, with the dependent variable the number of edge pieces out of the first 5 pieces attached, revealed a main effect of group, F(1, 48) = 34.81, p = .001, a main effect of puzzle type, F(1, 48) = 55.82, $p < .001$, and no interaction. Individuals with Prader-Willi syndrome were more likely than the typically developing controls to start with the edge pieces for both kinds of interlocking puzzles. The use of edge pieces increased for both groups on the blank jigsaw puzzle, where shape was the only information that could aid assembly (jigsaw/face:

Table 1. Visuospatial Score Comparisons by Group

	Prader-Willi syndrome		Typically Developing		Comparison
Task	Mean	*SD*	Mean	*SD*	Effect size[a]
Mental rotation	18.0	4.0	21.4	5.7	.68*
Motor-Free Visual Perception Test	12.8	3.7	15.8	3.7	.80**
Water level	7.3	3.0	7.6	4.7	.08
Placement task	19.4	6.8	23.4	5.8	.63*
Lego task	11.2	9.8	22.4	13.3	.95*

[a]Effect sizes are Cohen's d with Hedges correction.
*$p < .05$ (2-tailed). **$p < .01$ (2-tailed).

Table 2. Correlations Between Components of Visuospatial Ability/Puzzles

Group/Puzzles	MRT[a]	WLT[b]	MVPT[c]	Jigsaw/blank	Nonlock/Face
Prader-Willi					
Jigsaw/face	.44*	.18	.24	.73**	.55**
Jigsaw/blank	.34	.20	.23		.53**
Nonlock/face	.32	.48*	.61**		
Typically developing					
Jigsaw/face	.45*	.52**	.58**	.30	.82**
Jigsaw/blank	−.00	.16	.27		.38
Nonlock/face	.67**	.71**	.76**		

Note. All correlations are nonparametric Spearman correlations.
[a]Motor-Free Visual Perception Test (spatial visualization). [b]Water level task (spatial perception). [c]Mental rotation task (mental rotation).
*$p < .05$ (2-tailed). **$p < .01$ (2-tailed).

Prader-Willi syndrome = 2.72 edge pieces, SD = 2.26 and typically developing = 1.20, SD = 1.94; jigsaw/blank: Prader-Willi syndrome = 4.73 edge pieces, SD = 0.72 and typically developing = 3.88, SD = 0.88).

When examining the number of looks participants directed toward the box-top picture (for the two puzzles with pictures), we found that a 2 (group) × 2 (puzzle type) repeated measures ANOVA revealed a main effect of puzzle type on the number of looks toward the picture, $F(1, 48) = 20.74$, $p < .001$, a marginal effect of group, $F(1, 48) = 3.29$, $p = .076$, and no interaction. Because SDs were larger than means, we conducted further group analyses using the nonparametric Mann-Whitney U test, which indicated that the individuals with Prader-Willi syndrome looked less at the box-top picture than did the typically developing group for both the interlocking/face puzzle, 0.72 looks (SD = 1.70) versus 2.32 looks (SD = 2.59), $U = 151.0$, $p < .001$, and the noninterlocking/face puzzle, 3.12 looks (SD = 4.82) versus 4.58 looks (SD = 3.24), $U = 200.0$, $p < .05$.

Group strategy differences. A discriminant function analysis was conducted to determine whether four puzzle-solving behaviors would predict group membership. The four predictors were (a) number of edge pieces used for the jigsaw/face puzzle, (b) number of edge pieces used for the jigsaw/blank puzzle, (c) number of looks to the picture for the jigsaw/face puzzle, and (d) number of looks to the picture for the noninterlocking/face puzzle. The overall Wilks' lambda was significant, $\Lambda = .590$, $\chi^2(4, N = 50) = 24.29$, $p < .001$, and in each group, 20 out of the 25 individuals were correctly classified. Thus, these four variables, reflecting only two puzzle-solving behaviors, correctly classified 80% of the sample.

"Expert" Effects

In an effort to account for the rather large variability in puzzle scores, we examined a subset of individuals with Prader-Willi syndrome who stood out as having higher scores than did their counterparts whose scores were mediocre. The top third of individuals with Prader-Willi syndrome all completed or nearly completed the jigsaw/face puzzle during the 5-min session. Because the typically developing group had only 2 individuals who were equally successful, we considered those who scored in the top third of each group experts. We first assessed participant characteristics that might be associated with Prader-Willi syndrome expert status and then compared puzzle performance strategies across the 9 Prader-Willi syndrome and 10 typically developing experts (the difference in number was due to a tied score in the typically developing group).

Prader-Willi syndrome puzzle experts did not differ from their lower-scoring counterparts in gender, degree of obesity (body mass index), leisure activity, or Yale-Brown Obsessive-Compulsive Scale scores. Experts were, however, older (Ms = 27.09 vs. 16.24 years), $t(23) = 2.46$, $p < .05$. Relative to others with Prader-Willi syndrome, experts had similar Matrices scores on the K-BIT (Ms = 66.00 vs. 61.22), $t(23) = 0.86$, $p = .40$, but significantly lower K-BIT Vocabulary domain scores (Ms = 62.11 vs. 81.31), $t(23) = 2.82$, $p <$

.01, even after controlling for age. Experts did not differ from nonexperts in genetic subtype of Prader-Willi syndrome; the expert group included 6 persons with deletions, 2 with UPD, and 1 with an imprinting mutation.

We used a 2 (Prader-Willi syndrome experts vs. typically developing experts) × 3 (puzzle type) repeated measures ANOVA to examine the number of puzzle pieces placed and found significant main effects of group, $F(1, 17) = 8.030$, $p = .01$, and puzzle type, $F(1, 17) = 87.29$, $p < .001$, and a significant interaction, $F(1, 17) = 22.08$, $p < .001$. As shown in Figure 3, experts with Prader-Willi syndrome excelled at both the face and blank puzzles with interlocking pieces (i.e., puzzles in which shape information constrained the placement of pieces). Their advantage disappeared on the noninterlocking puzzle, however, when shape information was limited. The connection between adherence to a particular strategy and jigsaw puzzle performance is also seen at the individual level. Using the difference in scores between the jigsaw/blank puzzle (highlighting shape as a strategy) and the noninterlocking/face puzzle (highlighting the picture), we categorized each expert as scoring better on one puzzle or the other (none scored the same). In the Prader-Willi syndrome group, 7 of the 9 jigsaw-puzzle experts showed the shape pattern, whereas only 2 of the 10 typically developing experts did, a difference that was significant by Fisher's Exact test, $p = .023$. The other 8 typically developing experts showed the "picture" pattern. Therefore, most experts adhered to their group's typical strategy. Among the nonexperts, however, the pattern was mixed: in each group, approximately half ($n = 8$ per group) used a picture strategy, with the rest employing either no clear strategy (same score on both puzzles, $n = 4$ per group) or a shape strategy (ns = 4 Prader-Willi syndrome, 3 typically developing). (One participant from each group did not complete one puzzle and could not be classified.)

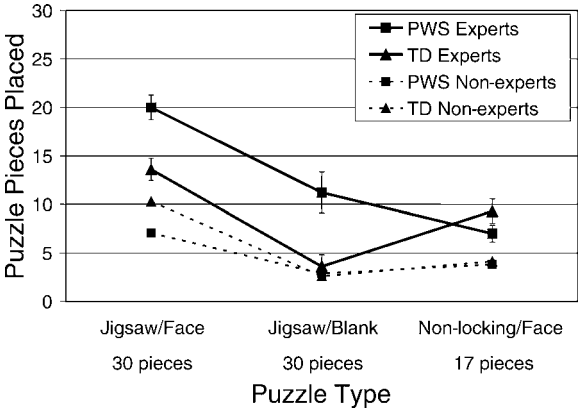

Figure 3. Number of pieces placed by experts and nonexperts from the typically developing and Prader-Willi syndrome groups in 3 min.

Correlates of Puzzle Assembly

We examined several possible correlates of puzzle performance: CA, MA, IQ, gender, everyday experience with visuospatial and puzzle-like activities, and in the group with Prader-Willi syndrome, genetic subtype and compulsivity. As shown in Table 3, CA and MA correlated with puzzle building for the typically developing group, but not for those with Prader-Willi syndrome. There were no effects of gender or IQ on puzzle scores or visuospatial measures.

Genetic subtype analyses were exploratory due to small numbers, with t tests showing no significant differences in puzzle scores between persons with paternal deletions versus maternal UPD. Those with deletions had, on average, a 4-piece advantage over those with UPD on the jigsaw/face puzzle (Ms = 13.20 pieces vs. 9.20); differences were modest on the other puzzles (jigsaw/blank: UPD = 7.00, deletion = 6.50; nonlocking/face: UPD = 3.60, deletion = 4.63).

No significant relations were found between puzzle scores and parental reports of everyday visuospatial leisure activities, including puzzle building. There also were no significant correlations between puzzle ability and Yale-Brown Obsessive-Compulsive Scale total scores in the group

Table 3. Correlations of CA, MA and Puzzle Scores

Group/Puzzle	MA	CA
Prader-Willi syndrome		
Jigsaw/face	.21	.22
Jigsaw/blank	.28	.16
Nonlocking/face	.40*	.00
Typically developing		
Jigsaw/face	.57**	.78**
Jigsaw/blank	.12	.24
Nonlocking/face	.76**	.90**

Note. All correlations are nonparametric Spearman correlations.
*$p<.05$. **$p<.01$ (two-tailed).

with Prader-Willi syndrome, although a moderate correlation was noted between the jigsaw/face puzzle and the supplementary item "needs to perform acts until 'just right'," $r(23) = .45$, $p = .03$.

Discussion

The results presented here suggest that persons with Prader-Willi syndrome do not have a general strength in visuospatial ability that helps explain their fascination or ability with jigsaw puzzles. The findings highlight key differences in puzzle-solving strategies between typically developing participants and those with Prader-Willi syndrome, reflected both in their performance on various puzzles and in their puzzle-solving behaviors. Visuospatial and puzzle assembly scores, as well as strategy differences, shed new light on this aspect of the Prader-Willi syndrome behavioral phenotype and suggest new research directions.

In examining a proposed link between visuospatial ability and skill in jigsaw puzzle assembly (Dykens, 2002), we used a test battery that tapped the three major components of visuospatial abilities: spatial perception, mental rotation, and spatial visualization. Participants with Prader-Willi syndrome scored significantly lower than did the MA-matched typically developing group on almost every measure of spatial ability. Their spatial scores were not related to assembling jigsaw puzzles. These were unexpected findings, considering previous indications of a relative strength in visuospatial ability. Unlike earlier investigators, however, we used tasks that did not resemble jigsaw puzzles and that tapped the full range of visuospatial processes as they are now being studied in the typically developing population (e.g., Liben et al., 2002). The use of such a wide range of visuospatial tasks makes it unlikely that the relatively poor showing of our participants with Prader-Willi syndrome is a spurious result. This conclusion is supported by results for the typically developing group, whose scores on the traditional jigsaw puzzle correlated with all spatial measures. Thus, the data from typically developing participants mirrors the expected outcome, indicating that the spatial tasks were appropriate for the MA of participants and that these spatial domains reflect the skills used by typically developing individuals to assemble jigsaw puzzles. The results, therefore, suggest the need to rethink the assumption that visuospatial ability is a true strength in the Prader-Willi syndrome cognitive phenotype and that it is responsible for the unusual skills in puzzle assembly shown by some individuals with this syndrome. The processes or skills that underlie jigsaw puzzle abilities in Prader-Willi syndrome seem atypical, a conclusion supported by evidence of their discrepant puzzle-solving strategies.

We examined strategies by manipulating puzzle stimuli and by observing puzzle-solving behaviors. On the traditional jigsaw puzzle, group differences in behaviors suggested by Dykens' (2002) preliminary observations were supported. Compared to the typically developing group, participants with Prader-Willi syndrome looked at the box-top picture significantly less often and were much more apt to start with the edge pieces—the plain, blue background of the puzzle. They fared much better at building puzzles with traditional jigsaw shapes, including the blank puzzle. They seemed to need shapes and interlocking components to perform exceptionally well. However, participants with Prader-Willi syndrome were able to perform at a level that was not significantly different from the MA-matched typically developing control group on the noninterlocking puzzle, which shares features with items on standardized IQ measures (e.g., the need to assemble nonlocking pieces to match a sample shape). This result is, therefore, consistent with earlier reports that individuals with Prader-Willi syndrome score closer to test norms on visuospatial subscales than on other standardized subscales (Dykens, 2002; Dykens et al., 1992). In addition, scores on two visuospatial measures (water level and Motor-Free Visual Perception Test) were related to scores on the noninterlocking puzzle for the group with Prader-Willi syndrome as well as the typically developing group. When participants with Prader-Willi syndrome could not use the shape strategy and needed to rely on other information, those with better visuospatial skills (as assessed by these two measures) were more successful.

The typically developing group had a markedly different pattern of behaviors and results. They tended to look at the picture more often and to start with inside pieces (those that included parts of the face). Reliance on the use of pictorial information as a strategy is consistent with the typically developing group's scores across the three puzzles (i.e., they performed best on the puzzles that had pictures, with or without interlocking pieces).

Strikingly, the two puzzle-solving behaviors—looking at the box-top picture and starting with

edge or inside pieces—correctly classified 80% of participants in each group. These behavioral differences are, thus, strongly associated with each group and hold promise for future research on puzzle-solving strategies in persons with and without disabilities.

The group differences were most obvious in a subset of participants whom we deemed "expert" in their puzzle-solving abilities. Experts adhered to a clear, well-defined strategy (shape-based *or* picture-based), despite the fact that neither strategy was uniformly successful across puzzle type. The success of either strategy depends on characteristics of a given puzzle.

A closer look at puzzle characteristics may suggest why individuals with Prader-Willi syndrome choose the strategy they do. Traditional jigsaw puzzles contain both surface features (e.g., shape and color of the pieces themselves) and content information (the subject of the box-top picture). It is quite possible, however, to rely successfully on only one kind of information. For example, a puzzle with repeating content (such as identical windows on a building) would be more difficult to complete relying exclusively on a picture-based strategy; in this case, a surface feature from individual pieces (e.g., piece shape) may be the most informative cue. For the blank puzzle, shape was the only source of information available to specify the correct location of a piece, and participants with Prader-Willi syndrome (especially the experts) did quite well. The typically developing group's content-based, picture-focused strategy might be considered more complex or advanced because it requires interpretation of the box-top picture and the integration of surface information, such as color. Yet the Prader-Willi syndrome strategy of relying primarily on shape may often be more effective because shape constrains the possible placement locations for each piece. Individuals relying on a shape strategy also benefit from the appearance of similar shapes (e.g., edges; pieces with particular configurations of protrusions and holes) across different jigsaw puzzles. In contrast, the pictures on puzzles vary widely. Therefore, a shape-based strategy may be easier for individuals with Prader-Willi syndrome to apply when encountering new puzzles. Sensitivity to piece shape—perhaps even implicit or explicit awareness of categories of shapes—may help them to efficiently assemble puzzles.

After the main analyses had been completed, while exploring potential connections between strategies and visuospatial skills, we found an interesting set of correlations that may provide a clue regarding the use of the shape strategy. For the group with Prader-Willi syndrome, scores on the Motor-Free Visual Perception Test (our measure of spatial visualization) were negatively correlated with the number of edge pieces (of the first 5) placed on the blank jigsaw puzzle, $r = -.46$, $p < .05$, but positively correlated with looks to the box-top picture on the noninterlocking puzzle, $r = .56$, $p < .01$. Thus, the individuals who had poorer scores on spatial visualization were the ones who were (a) more likely to use the shape strategy on the challenging blank jigsaw puzzle and (b) less likely to look at the box-top picture on the noninterlocking puzzle (when pictorial information was much more helpful than shape). Use of information from the box-top picture would appear to involve spatial visualization—multistep mental operations on spatial information and the use of analytic strategies—because it requires holding the picture in mind while identifying matching pictorial information on individual pieces and analyzing the relative position of pictorial elements. It is possible that individuals who have difficulty with spatial visualization do not find looking at the picture helpful. They may benefit more by concentrating on a simple strategy of matching piece shapes, rather than trying to attend to and integrate pictorial information at two levels.

We also examined correlates of puzzle performance, with an eye toward possible explanations for within-syndrome variability. Indeed, the analyses of the expert groups confirm that a number of persons with Prader-Willi syndrome excel at jigsaw puzzles, but that this special skill is not present in all individuals. Contrary to expectations, few significant correlates emerged between puzzle performance and participant characteristics. As predicted, in the group with Prader-Willi syndrome, we found a modest correlation between jigsaw puzzle score and needing to perform acts until "just right." Counter to our expectations, however, performance was not related to parental reports of previous experiences with puzzles or to visuospatial or puzzle-like daily activities. Experts in both groups were older than nonexperts (Yet, paradoxically, experts in the group with Prader-Willi syndrome scored lower on the Vocabulary subtest of the K-BIT than did the Prader-Willi syndrome nonexperts). Assuming that age is a proxy for opportunities to construct

puzzles, practice may indeed contribute to more proficient performance. Even so, experience by itself is unlikely to explain the strategy differences between experts from the two groups. If experience promotes the development of a shape-based strategy, then the most experienced (and therefore skilled) participants should share similar strategies regardless of group. In fact, our study showed the opposite: The most skilled participants in each group had strategies that were the most different.

This study had several limitations that relate both to Prader-Willi syndrome and to challenges inherent in jigsaw puzzles. First, the number of participants with each of the genetic subtypes was small, limiting our ability to use subtype as a predictor of puzzle performance. However, with the previous research indicating that those with UPD tend to perform less well on puzzles and on intelligence tests tapping visual or spatial functioning (Dykens 2002; Roof et al., 2000; Whittington et al., 2004), it is noteworthy that 2 of our Prader-Willi syndrome experts had UPD. Further subtype comparisons are needed with larger numbers of persons with UPD and Type I and Type II deletions.

Second, our age range in the Prader-Willi syndrome group was quite wide due to the difficulty of recruiting members of such a rare population; therefore, our typically developing age range was also somewhat wide. Chronological age (i.e., experience), logically important for the development of puzzle skills, was an important factor for puzzle performance in the typically developing group (but not for the Prader-Willi group). It is conceivable that lack of experience in the younger typically developing children may have resulted in reduced puzzle scores and, perhaps, affected the strategies of this group, which had younger individuals overall. Of interest, both groups, including the youngest children, knew and could use the other group's preferred strategy (at least to some extent), when other information was lacking. For instance, the typically developing children did use edge pieces more on the blank puzzle than on the jigsaw/face puzzle. We are planning a follow-up study in which a group of CA-matched adult participants will be included to control for possible contributions of age and experience.

In the current study we were able to manipulate only some of the properties of puzzles. Besides the presence or absence of pictures and piece shapes, puzzles also differ in their use of repeating patterns, colors, contours, textures, appeal of visual stimuli, and even dimensionality, as 3-D puzzles are now readily available. Indeed, variability in puzzles themselves likely underlies our smaller effect sizes compared to those found by Dykens (2002). In future research, we will examine whether individuals with Prader-Willi syndrome use other piece-based information (e.g., color), and how the use of "local" cues, such as piece shape or color (versus information from the picture) might relate to reported short-term memory deficits (Conners et al., 2000; Warren & Hunt, 1981).

The current results reveal that persons with Prader-Willi syndrome do not appear to have exceptional general visuospatial abilities that are responsible for their jigsaw puzzle skills. They do, however, have a pronounced, shape-based puzzle-solving style that differs from typically developing controls and serves them well on interlocking (even achromatic) puzzles. If we assume that different strategies emerge based on underlying skills, then understanding these strategies can serve as indicators of consistent between- and within-group differences in the processing of visual information. Although further work is needed on genetic, developmental, and other reasons for variability in puzzle abilities, this study begins to disentangle an unusual aptitude shown by some persons with Prader-Willi syndrome.

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