Published in final edited form as:
Am J Ment Retard. 1999 November ; 104(6): 523–532.

Second-Order Belief Attribution in Williams Syndrome: Intact or Impaired?

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Abstract

Second-order mental state attribution in a group of children with Williams syndrome was investigated. The children were compared to age, IQ, and language-matched groups of children with Prader-Willi syndrome or nonspecific mental retardation. Participants were given two trials of a second-order reasoning task. No significant differences between the Williams syndrome and Prader-Willi or mentally retarded groups on any of the test questions were found. Results contrast with the view that individuals with Williams syndrome have an intact theory of mind and suggest that in their attributions of second-order mental states, children with Williams syndrome perform no better than do other groups of children with mental retardation.

One of the most striking features of children with Williams syndrome is their unusual friendliness and strong interest in other people (Beuren, Schulze, Eberle, Harmjanz, & Apitz, 1964; Dilts, Morris, & Leonard, 1990; Udwin & Yule, 1991; Udwin, Yule, & Martin, 1987). Children with Williams syndrome have been observed to be extremely sociable, charming, outgoing, and highly concerned about the well-being of others (Gosch & Pankau, 1994; Sarimski, 1997; Tomc, Williamson, & Pauli, 1990). Further, young children with Williams syndrome are particularly drawn to human faces (Mervis & Bertrand, 1997) and are highly responsive to the distress of others (Tager-Flusberg & Sullivan, 1999). This strong interest in others coupled with excellent face-processing abilities (Pezzini, Vicari, Volterra, Milani, & Ossella, 1999; Udwin & Yule, 1991) and use of language in socially relevant ways (Reilly, Klima, & Bellugi, 1991) has led some to propose that children with Williams syndrome have an intact theory of mind (Karmiloff-Smith, Klima, Bellugi, Grant, & Baron-Cohen, 1995; Tager-Flusberg, Hoshart, & Baron-Cohen, 1998). That is, they make sense of others' behavior in terms of a coherent causally related set of mental states (e.g., beliefs, desires, and intentions).

However, despite their extreme sociability and strong interest in people, children and adolescents with Williams syndrome experience great difficulty with their peers (Greer, Brown, Pal, Choudry, & Klein, 1997). By middle childhood, these children have difficulty interpreting social interactions and social cues, engage in socially inappropriate behavior, and have great difficulty making and sustaining friendships with their peers (Gosch & Pankau, 1994; Sarimski, 1997). Thus, the apparent contradiction between their extreme interest in others coupled with their social difficulty suggests that social cognition, or theory of mind, may not be an area of strength for these individuals.

To date, only a few investigators have explored theory of mind abilities in children with Williams syndrome. Although Karmiloff-Smith et al. (1995) found that the majority of children with Williams syndrome performed well on a standard first-order false belief task, methodological limitations cloud the interpretation of their results. More recently, Tager-Flusberg and Sullivan (1999; see also Tager-Flusberg, Sullivan, & Boshart, 1997) showed that fewer children with Williams syndrome pass standard false belief tasks than do matched groups of children with Prader-Willi syndrome or nonspecific mental retardation. They also performed no better on other tasks tapping theory of mind abilities at a first-order level.

Another, perhaps even more socially relevant achievement in the development of a theory of mind, is the ability to attribute second-order or embedded mental states (e.g., he thinks that she thinks). Why is this type of reasoning critical to social interactions? Being able to represent what one person thinks about what a second person thinks allows us to understand not only another's belief about the world (a first-order belief) but also to understand that person's concern about yet another person's belief about the world (a second-order belief). This sort of reasoning is necessary for any sophisticated understanding of the subtleties inherent in social interactions. Perner (1988) argued that it is at the level of second-order reasoning that social interaction can be understood as an interaction of minds where people are concerned about each other's mental states.

Early studies of typically developing children showed that an understanding of second-order mental states emerges between the ages of 5 to 7 (Hogrefe, Wimmer, & Perner, 1986; Leekam, 1990; Perner & Howes, 1992;Perner & Wimmer, 1985). In a later studySullivan, Zaitchik, and Tager-Flusberg (1994) showed that younger children can also pass second-order tasks. These investigators introduced a simpler second-order task that substantially reduced the information-processing demands of the task. Their task reduced the number of characters, episodes, and scenes and included a deception context in which one actor deceives another to conceal a gift. Sullivan et al. found that nearly half of their sample of preschool children (ages 4.08 to 5.25 years) could attribute and justify second-order beliefs, suggesting that children are capable of second-order reasoning well before the age of 7. This task has also been used successfully with atypical children. Using the same procedure, Tager-Flusberg and Sullivan (1994) found that 58% of participants with autism and 67% of participants with mental retardation between 8 and 22 years of age were able to attribute second-order beliefs.

Second-order reasoning by individuals with Williams syndrome has been directly investigated in only one study. Using the Sullivan et al. (1994) procedure, Karmiloff-Smith et al. (1995) found that 7 out of 8 individuals with Williams syndrome (88%), ranging in age from 9 to 23 years, were able to attribute second-order mental states. However, because of the small sample size, and the absence of a comparison group of individuals with mental retardation due to other causes, it would seem important to reopen the question of whether younger children and adolescents with Williams syndrome are genuinely spared in their ability to understand second-order belief.

Thus, our goal in the present study was to explore second-order reasoning in a group of children with Williams syndrome. We compared the performance of children with Williams syndrome to two other groups: children with mental retardation without specific known etiology and children with PraderWilli syndrome, another well-defined genetic disorder. The three groups were matched on age, IQ, and language ability as measured by a receptive vocabulary test (Peabody Picture Vocabulary Test-Revised—PPVT-R (Dunn & Dunn, 1981). The inclusion of two well-matched comparison groups allowed us to assess whether children with Williams syndrome are indeed spared in their ability to attribute second-order mental states relative to other children with mental retardation who are of similar developmental level. If children with Williams syndrome are indeed spared in their ability to attribute second-order mental states, we would predict that they would perform better that IQ- and language-matched groups of children with Prader Willi syndrome and children with mental retardation due to other causes (nonspecific mental retardation).

Method

Participants

Three populations participated in this experiment: children with Williams syndrome, Prader Willi syndrome, and nonspecific mental retardation. Table 1 presents the characteristics of

these groups. The first group included 22 children (13 females, 9 males) with Williams syndrome. Williams syndrome is a contiguous gene disorder caused by a hemizygous submicroscopic deletion of chromosome 7 at band q11.23 that contains the *elastin* gene (Mervis, Morris, Bertrand, & Robinson, 1999). All of the children with Williams syndrome had previously been clinically diagnosed as having this disorder. Referrals from the New England Williams Syndrome Association and the National Williams Syndrome Association were used to identify and contact local families. Eleven children with Williams syndrome who attended the 1996 National Williams Syndrome Association Family Conference also participated in this study.

The second group included 14 children with Prader Willi syndrome (4 females, 10 males). This disorder is caused by the loss of paternally expressed genes in the q11-q13 region of chromosome 15 (Butler, 1990, 1994). All of these children had previously been clinically diagnosed as having Prader Willi syndrome. The IQ distribution among individuals with Prader Willi syndrome is similar to individuals with Williams syndrome, but they do not have a particularly uneven cognitive profile (Dykens, 1999; Dykens, Hodapp, Walsh, & Nash, 1992; Gabel, Tarter, Gavaler, Colder, Hegedus, & Maier, 1986). Referrals from the Prader-Willi Association of New England were used to recruit and contact local families. Eight children with Prader Willi syndrome who attended the 19th annual Prader-Willi Syndrome National Conference in 1997 also participated in this study.

The third group included 13 children with nonspecific retardation (7 females, 6 males). They were drawn from special education classes in local public schools. Their mental retardation was of unknown etiology.

Form M of the PPVT-R, a receptive vocabulary test, was administered to all participants. We also administered either the Differential Abilities Scales (Elliot, 1990) or the Kaufman Brief Intelligence Test (Kaufman & Kaufman, 1990) as a general measure of cognitive level. The Differential Abilities Test yields verbal and nonverbal domain scores as well as an overall composite score, or General Conceptual Ability, which is equivalent to a Full-Scale IQ score. The Kaufman Brief Intelligence Test yields verbal and nonverbal domain scores as well as an overall IQ score. For 6 of the children with nonspecific mental retardation we were unable to administer either test; therefore, recent IQ information was taken from school records. For all of these children, the school records included scores from a recently administered Wechsler Intelligence Scale for Children-Revised, which includes verbal and performance domain scores as well a Full-Scale IQ. The Full-Scale equivalents yielded by these three IQ measures have been shown to be highly correlated (Elliot, 1990; Kaufman & Kaufman, 1990). Analyses of variance confirmed that at a group level there were no significant differences among the three groups on chronological age (CA), PPVT-R mental age (MA) and standard score, and Full-Scale IQ, suggesting that the participants were well-matched. Although Williams syndrome is characterized by a particular cognitive profile in which vocabulary is generally higher than other cognitive skills, especially visual—spatial skills, whereas neither Prader Willi syndrome nor mental retardation due to other causes are associated with a distinctive profile, in each population there is considerable variability. For example, Pezzini et al, (1999) did not find that all of their participants with Williams syndrome showed the typical profile described in the present study. Investigators of Prader Willi syndrome have reported higher nonverbal IQ (Curfs, Wiegers, Sommers, Borghgraef, & Fryns, 1991) and higher verbal IQ in this population (Borghgraef, Fryns, & Van den Berghe, 1990). We took advantage of this intrapopulation variability to select participants who would, at least at a group level, provide a good match.

Procedure

Two second-order belief stories based on a task developed by Sullivan et al. (1994) were presented to the children. For example, in one story, a father deliberately misinforms his

daughter Molly about her birthday gift (a bike) because he wants to surprise her. However, unbeknownst to her father, Molly discovers her present. Thus, the father *does not know* that Molly *knows* about her present. Later in the story, when speaking to the girl's grandmother, the father is asked whether the girl *knows* what she is getting (second-order knowledge) as well as what she *thinks* she is getting for her birthday (second-order belief). For each story, children were presented with two control questions (reality and linguistic control) and a first-order false belief question. After each of these control questions, if the child gave the wrong response, corrective feedback was provided. At the end of the story two test questions (second-order ignorance and second-order belief) were presented, and participants were asked to justify their responses to the second-order belief question. The two stories were presented in counterbalanced order and were acted out with three-dimensional props and dolls (for children younger than 10 years) or were accompanied by four colored drawings depicting the characters and main story events (for children older than 10 years). The appendix provides a complete example of one of the stories used in this study. Children were tested individually by two experimenters. Responses were audiotaped and later transcribed and checked by a second coder.

Response Coding

Responses to the control and test questions were scored as correct or incorrect. Percentage agreement between coders calculated on a subset of children's responses (50%) was 100%. Justifications were coded into the following categories based on those used by Sullivan et al. (1994). Two raters independently coded a subset (30%) of justification responses. Agreement was calculated at 97%.

A. *Appropriate.* responses that demonstrated an appreciation of the deception involved in the story or of the character's knowledge states or exposure to relevant information. (a) Explicit Second-order reasoning: The child explicitly mentions the mental states of both characters; for example, "Dad doesn't know that Molly knows about the bike." (b) Implicit second-order reasoning: The child refers to one character's mental states as well as the second characters exposure to relevant information; for example, "Dad doesn't know that Molly saw the bike." (c) Deception: The child makes explicit reference to the surprise or deception; for example, "Dad wants the bike to be a surprise" or "She doesn't know that he's tricking her about the bike." (d) Communicated information: The child mentions relevant information communicated between characters; for example, "Because Dad told Molly she was getting a video game."

B. *Inappropriate:* responses that indicated that the child did not understand the relevant story information. (a) First-order reasoning: The child mentions the first order belief of the main character; for example, "Because Molly knows she's getting a bike." (b) Story facts: The child mentions story facts; for example, "Because Molly wants a bike." (c) Nonsense/No response.

Results

Table 2 shows the means (and standard deviations) for each group for the control and test questions. There were no significant group differences on the control questions. A one-way ANOVA with group as the between-subjects variable was used to examine performance on the first-order belief question. There were no significant group differences.

To explore performance on the second-order test questions, we conducted a 3 X 2 mixed design ANOVA with group as the between-subjects factor and question type (second-order ignorance, second-order belief) as the within-subjects factor. This analysis revealed a significant main

effect of question type, $F(2, 46) = 15.72$, $p < .001$ - A post-hoc paired samples *t* test revealed that for all children, the second-order ignorance question proved easier than the second-order belief question, $t(48) = 3.90$, $p < .001$, which is consistent with previous findings using a similar procedure with both typical (Sullivan et al., 1994) and atypical populations (Tager-Flusberg & Sullivan, 1994). The main effect of group and the interaction of group and question type were not significant.

We then looked at the number of children passing the second-order ignorance and belief questions, where *passing* was defined as answering correctly on both trials of the test questions. Table 3 shows the number and percentage of children in each group passing each test question. Chi-square analyses revealed that the groups performed equivalently on the second-order ignorance question. On the false belief question, significantly more of the children with Williams or Prader Willi syndrome passed than did the children with nonspecific mental retardation, χ^2 (1, $N = 35$)=328, $p < 07$, and χ^2 (1, $N = 27$) = 504, $p < .03$, respectively.

Justifications

Children's responses to correct second-order belief responses are presented in Table 4. There were no differences among the groups in the use of the various justification responses. As can be seen, the majority of correct second-order belief responses across all three groups were appropriately justified. The highest proportion of appropriate justifications involved explicit mention of the deception in the story (e.g., "Because he's tricking her with the bike") or reference to the main character's knowledge state relative to the other characters exposure to relevant information (e.g., "Because he doesn't know that she saw the bike"). This suggests that the majority of children's correct belief responses were not due to guessing, but were reasoned responses.

However, a substantial proportion of the children in all three groups gave inappropriate justifications to correct belief responses (ranging from 21% to 36%). This raises questions about how to interpret the responses of those children who were unable to appropriately justify their answers. One possibility is that these children were simply guessing, and their inappropriate justifications reflect a genuine lack of understanding. On the other hand, it is possible that they were capable of second-order reasoning but were unable to make their understanding explicit. To clarify this issue, we examined the pattern of performance across both trials of the belief question, looking at whether more of the inappropriate justifications were given by children who passed only one trial. We found that children passing only one trial of the belief question provided the vast majority (75%) of the inappropriately justified responses, suggesting that these children may have been guessing on the second-order false belief question.

Finally, we looked at whether there were significant differences in CA, Full-Scale IQ, or PPVT MA between passers and failers within each group. For the Williams syndrome and nonspecific mental retardation groups, none of these variables revealed differences between those children who passed and those who failed the second-order belief question. Among the children with Prader Willi syndrome, passers had significantly higher PPVT MAs than did the failers, $r(14) = .67$, $p < .008$.

Discussion

The main finding in this study was that the children with Williams syndrome performed at a comparable level to two comparison groups matched on language and IQ levels in attributing second-order mental states. For participants in all three groups, the second-order ignorance question was significantly easier than the second-order belief question. Over two thirds of the participants with Williams syndrome and Prader Willi syndrome and 62% of those with

nonspecific mental retardation passed two trials of the ignorance question. This relative ease of the second-order ignorance question is consistent with previous findings using this same method with both typically developing 4 to 6 year olds (Sullivan et al., 1994) and atypical populations (Tager-Flusberg & Sullivan, 1994). On the other hand, only 45% of the children with Williams syndrome, 57% of the children with Prader Willi syndrome, and 13% of those with nonspecific mental retardation passed both second-order belief question trials. These findings show that on the simplest measure of second-order belief attribution available, children with Williams syndrome are not spared relative to other groups of children with borderline or mild mental retardation. Furthermore, the relatively poor performance of the children with Williams syndrome on the second-order belief question does not reflect "success" in the domain of theory of mind. Sullivan et al. (1994) found that the majority of their preschool-age sample (ages 4 to 6 years) passed second-order belief. Yet, despite the significantly higher mental and language age levels of our participants with Williams syndrome, only about half of them demonstrated the capacity for second-order reasoning. This finding suggests that these children are not spared in this domain relative to their overall cognitive or linguistic profile. All three groups of children with developmental disorders who participated in the present study performed worse than did much younger typically developing children, despite their higher verbal MAs. This poorer performance needs to be explored further because it suggests that other factors may influence higher order social reasoning, as measured in this theory of mind task. Performance on this second-order false belief task requires children to integrate information across the entire narrative and draw inferences from this information. Thus, the task entails a significant working memory load and attention to the sequence of events in the story. These kinds of executive function task components may adversely influence the performance of children with developmental disorders.

The findings reported here on children with Williams syndrome contrast with those reported by Karmiloff-Smith et al. (1995), who found that most of their participants with Williams syndrome passed the second-order task. One possible explanation for this discrepancy is the criterion for success on this task. Although in the present study children had to respond correctly on two trials of the second-order belief question to receive a passing score, it is unclear what Karmiloff-Smith et al. considered to be passing performance. It is possible that subjects received only one trial of the test question, which would have made passing much easier than in the present study. In fact, if we look at the performance of the children with Williams syndrome on only the first trial of the second-order belief question, 68% scored correctly (compared to 45% on two trials), which is closer to the success rate reported by Karmiloff-Smith and her colleagues. Another possibility is that our use of props and pictures to support the narrative may have diverted the attention of the children with Williams syndrome, who perhaps find it easier to listen to stories without such distractions (Karmiloff-Smith, personal communication).

Furthermore, because only a subset of 8 participants with Williams syndrome received the second-order task in Karmiloff-Smith et al's (1995) study, it may be that their participants with Williams syndrome were, on average, older than ours and may have included some adults. It may be that eventually most people with Williams syndrome are able to pass second-order theory of mind tasks, indicating that they are not, in an absolute sense, impaired in this domain. Although we did not find that our older participants with Williams syndrome were more likely to pass than were our younger participants, it is still likely that development in this domain continues through adulthood. Studies of typically developing children suggest that social experience with both siblings and peers promotes the development of false belief understanding (Lewis, Freeman, Kyriakidou, Maridaki-Kassotaki, & Berridge, 1996; Perner, Ruffman, & Leekam, 1994). An understanding of mental states, which are especially important in social interactions, is acquired in part through experiences with more knowledgeable members of the culture (Bruner, 1990; Dunn, 1994). Children and adolescents with developmental disabilities

are likely to have more limited social experiences outside the family. As they get older, they often have more social opportunities, especially when they leave school and have more contacts within the community. Through such experiences, they are likely to develop a more advanced understanding of mind, including the ability to attribute second-order beliefs.

The findings from our study indicate that many children and adolescents with mental retardation, including Williams syndrome, Prader Willi syndrome, and nonspecific.mental retardation, have difficulty with second-order reasoning, particularly understanding that one person may hold a false belief about what a second person knows about a situation. These kinds of deficits in social understanding have important ramifications for their ability to interpret the kinds of complex social situations that mark everyday peer interactions. It is, thus, not surprising that children with mental retardation, including highly sociable children with Williams syndrome, have such difficulty with social relationships in middle childhood and adolescence. Future research should be focused on developing interventions that will help to remediate the kinds of social cognitive deficits that we have identified in this study.

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APPENDIX Script for Second-Order Belief Task

This is a story about Molly and her dad. Today is Molly's birthday and she's having a big party tonight. For Molly's big present, Dad is surprising her with a new bike that he has hidden out in the garage. See? *(Experimenter points to bike.)* Here is the surprise bike.

Molly and Dad are in the kitchen talking about her birthday. Molly says, "Dad, I really want a new bike for my birthday." *Now remember,* Dad wants the bike to be a big surprise. So, he says, "Sorry, Molly, I didn't get you that. I got you a great video game instead.'

First-order belief question: What does Molly think Dad got her for her birthday?

(If correct: That's right, Molly thinks she's getting a video game; If incorrect: But remember, Molly thinks she's getting a video game)

Reality control question: What did Dad *really* get her for her birthday?

(If correct: That's right, Dad wants to surprise Molly with a bike; If incorrect: But remember, Dad wants to surprise Molly with a bike.)

Then Molly says to Dad, "Okay. Well, I'm going over to my friend's house. I'll be home later." On her way out, Molly goes to the garage to get her umbrella because it's raining. In the garage, Molly finds her new bike! She says to herself, *"Yes!* Dad didn't get me a video game. He really got me a bike." Dad *does not* see Molly go out to the garage and find the new bike.

Linguistic control question: Does Dad know that Molly saw her bike in the garage?

(If yes: Hut remember, Dad did not see Molly go out to the garage and find the bike.)

Later, Molly's grandmother comes over for the party. She goes into the kitchen. Grandma asks Dad, "Does Molly know what you got her for her birthday?"

Second-order ignorance question: What does Dad say?

Now remember, Dad *does not know* that Molly saw what he got her for her birthday. Then Grandma says to Dad, "What does Molly think you got her for her birthday?"

Second-order belief question: What does Dad say?

Justification: Why does Dad say that?

Table 1

Participant Characteristics by Group

				PPVT-R								
	CA			MA			Standard score			Full-Scale IQ		
Group[a]	Mean	*SD*	Range	Mean	*SD*	Range	Mean	*SD*	Range	Mean	*SD*	Range
WMS (*n* = 22)	11.58	2.67	8.17–17.25	8.56	2.33	4.67–15.50	78	13	46–107	65	11	45–96
PWS (*n* = 14)	12.08	2.75	5.67–17.08	8.79	3.00	3.25–13.08	77	14	55–101	70	10	55–91
NMR (*n* = 13)	11.42	3.25	6.42–15.25	7.00	1.50	4.75–9.58	67	17	43–96	72	11	50–86

[a]WMS = Williams syndrome. PWS = Prader Willi syndrome, NMR = nonspscified mental retardation. PPVT-R = Peabody Picture Vocabulary-Revised.

Table 2

Mean Scores and *SDs* on Control and Test Questions by Group

	WMS		PWS		NMR	
Questions	**Mean**	***SD***	**Mean**	***SD***	**Mean**	***SD***
Control						
Reality	1.59	0.67	1.57	0.85	1.77	0.60
Linguistic control	1.64	0.73	1.86	0.53	1.77	0.44
Test						
First-order belief	1.09	0.92	1.07	0.92	0.62	0.77
Second-order ignorance	1.59	0.73	1.64	0.63	1.46	0.78
Second-order false belief	1.14	0.89	1.36	0.84	0.62	0.77

Note. Maximum score = 2. WMS = Williams syndrome, PWS = Prader-Willi syndrome, NMR = nonspecified mental retardation.

Table 3

Number (and Percentage) of Participants Passing Second-Order Ignorance and False Belief

	Second-order			
	Ignorance		False belief	
Group	Freq.	%	Freq.	%
Williams syndrome	16	73	10	45
Prader-Willi syndrome	10	71	8	57
Nonspecific mental retardation	8	62	2	13

Table 4

Frequency of Appropriate and Inappropriate Justifications to Correct Second-Order False Belief Responses by Group

Response	WMS		PWS		NMR	
	Freq.	%	Freq.	%	Freq.	%
Appropriate						
Explicit second-order	1	4	0	0	0	0
Implicit second-order	5	20	4	21	0	0
Communicated information	4	16	0	0	1	13
Deception	6	24	11	58	5	63
Inappropriate						
First-order reasoning	1	4	1	5	0	0
Story facts	2	8	1	5	0	0
Nonsense	2	8	0	0	1	13
No response	4	16	2	11	1	13

Note. WMS = Williams syndrome, PWS = Prader-Willi syndrome, NMR = nonspecific mental retardation.