

Altered Attribution of Intention in Tourette's Syndrome

Clare M. Eddy, Ph.D.

Ian J. Mitchell, D.Phil.

Sarah R. Beck, Ph.D.

Andrea E. Cavanna, M.D.

Hugh E. Rickards, M.D.

Core symptoms of Tourette's syndrome are assumed to result from inhibitory dysfunction, which could also impair theory of mind. Here the authors report evidence for theory of mind difficulties: patients exhibit deficits in recognizing faux pas and understanding intentionality.

(The Journal of Neuropsychiatry and Clinical Neurosciences 2010; 22:348–351)

Tourette's syndrome is a neurodevelopmental disorder characterized by multiple motor and phonic tics. Involuntary tic-related symptoms may include the uttering of offensive language (coprolalia), and comorbid psychiatric disorders including obsessive-compulsive disorder (OCD) and attention-deficit hyperactivity disorder (ADHD) are common. One study¹ found that many patients with Tourette's syndrome experience urges to act in a socially inappropriate way (22%) or make socially inappropriate remarks including insults (30%). Failure to suppress these urges sometimes results in major social difficulties. These socially inappropriate behaviors may arise from inhibitory failure secondary to frontostriatal dysfunction. Moreover, patients with uncomplicated Tourette's syndrome (i.e., motor and phonic tics only, with no associated behavioral problems) have been found to exhibit inhibitory deficits on the Hayling Sentence Completion Task—Adapted (HSCT).^{2,3} Such inhibitory deficits could lead to impairments in theory of mind.

Theory of mind describes the ability to understand people's mental states (e.g., emotions, beliefs, intentions), which allows one to explain and predict people's actions. Inhibitory dysfunction may impair theory of

mind because appreciating another's mental state necessitates inhibition of one's own perspective. Developmental research has shown that early inhibitory control development predicts later false-belief understanding in children,⁴ while one clinical case study showed right frontotemporal damage can lead to selective impairment on theory of mind tasks requiring the suppression of one's own knowledge.⁵ Inhibitory dysfunction in Tourette's syndrome as indicated by deficits on executive tasks may contribute to theory of mind impairment, because brain regions active during these tasks are active when reasoning about others' beliefs.⁶

Developmental researchers investigating theory of mind sometimes use the "unexpected transfer" task. During this task, a target moves from one location to another, while a story character is absent. Children exhibit understanding of the absent character's false belief about the target's location from around 4 years old.⁴ Faux pas tasks are also used, which children pass between 9 and 11 years old.⁷ These tasks feature a character making a remark that they are unaware is potentially offensive. Faux pas tasks may be harder to understand because they involve both the appreciation of the perpetrator's false belief (the remark is inoffensive) and the victim's emotional response (offense). Comprehension of the perpetrator's belief about the victim's mental state involves second-order theory of mind.

One study investigated theory of mind in uncomplicated Tourette's syndrome.⁸ No deficits were evident on two tests of higher-order mentalizing skills, though this could reflect small sample size or lack of sensitivity of the theory of mind measures. The present study investigated theory of mind in Tourette's syndrome using a false-belief task⁹ and a faux pas task.⁷ Two executive tasks were used to assess inhibition (HSCT) and work-

Received October 13, 2008; revised February 9 and April 20, 2009; accepted April 24, 2009. Drs. Eddy, Mitchell, and Beck are affiliated with the Department of Psychology at the University of Birmingham in Birmingham, UK; Dr. Eddy is also affiliated with the Department of Psychiatry at Queen Elizabeth Hospital. Drs. Cavanna and Rickards are affiliated with the Department of Psychiatry at Queen Elizabeth Psychiatric Hospital in Birmingham, UK. Address correspondence to Dr. Clare Eddy, The Barberrry, 25 Vincent Drive, Edgbaston, Birmingham, B15 2FG, United Kingdom; c.eddy@bham.ac.uk (e-mail).

Copyright © 2010 American Psychiatric Publishing, Inc.

ing memory (Digit Ordering Test—Adapted¹⁰), which may affect task performance. We hypothesized that patients would exhibit deficits in theory of mind and inhibition but not in working memory.

METHODS

Participants

We recruited 16 outpatients with Tourette's syndrome (three women; mean age=32.06 years, mean education=12.94 years) from the Tourette's syndrome clinic at Queen Elizabeth Psychiatric Hospital in Birmingham. Each subject underwent a comprehensive clinical interview using the National Hospital Interview Schedule for Tourette's syndrome, a detailed semistructured interview schedule which includes personal and family histories and demographic details. For the diagnosis of various Tourette's syndrome-associated behavioral disorders, such as OCD and ADHD, the National Hospital Interview Schedule was originally developed by incorporating the relevant questions and items from the Diagnostic Interview Schedule to yield a diagnosis as per DSM-III-R and was then updated based on the DSM-IV-TR criteria.¹¹ The subjects were assessed on several occasions by a clinical neuropsychiatrist with experience in Tourette's syndrome (HR). Four exhibited comorbid OCD, two of whom had ADHD and one an anxiety disorder. A further four patients reported sub-threshold obsessive-compulsive symptoms. No patients fulfilled diagnostic criteria for oppositional defiant disorder or conduct disorder. Seven were taking medication for tics (risperidone=3, aripiprazole=2, sulpiride=1, pimozide=1). Eight healthy comparison subjects (three women; mean age=34.25 years, mean education=14.63 years) also participated.

Procedure

Informed consent was obtained after the procedure was explained to participants, who were tested in the consulting room or at home. Executive tasks were presented first. Tasks were alternated within executive and theory of mind blocks, resulting in four procedure orders. The tasks were administered and scored by an experienced psychological researcher (CME) who was unblinded to the study hypotheses and group assignment.

Hayling Sentence Completion Task—Adapted² Participants completed sentences with a single word (e.g., "the dog

chased the cat up the . . ."). For part A, initiation, completion words had to fit the sentence (e.g., "tree"). In part B, inhibition, completion words should make sense but not be obvious (e.g., "mountain"). Two practice sentences were followed by 10 sentences in each part.

Digit Ordering Test—Adapted¹⁰ Scores reflected the number of digits presented in random order that could be correctly recalled in ascending order.

Theory of Mind Vignettes⁹ Participants were read four unexpected transfer style vignettes (e.g., "Andrew is in bed. Susie goes to the shops and while she's away, Andrew goes to the school."). Each vignette was read twice and followed by four questions: counterfactual ("Where would Andrew be if he hadn't gone to the school?"); false-belief ("Where does Susie think Andrew is?"); memory ("Where was Andrew at the start?"); and reality ("Where is Andrew now?"). There were two forced choice responses, the target's original and current locations. Vignettes were presented in a set order, but questions were counterbalanced in pairs (first and second false belief or counterfactual; third and fourth memory or reality).

Faux Pas Task⁷ Participants were read eight vignettes. Four test vignettes described a potentially offensive faux pas (e.g., "Jill has moved to a new house and bought new curtains. Lisa says the curtains are horrible."). Four control stories involved no faux pas. The first two questions assessed recognition of faux pas ("Did someone say something they should not have?" If yes, "Who was it and what did they say?"). If the faux pas was identified, two further probes asked, "Why shouldn't they have said that?" and "Why do you think they did say it?" Another question checked recall ("What had Jill just bought?"). Finally, a belief question was asked about all test vignettes ("Did Lisa know that Jill had chosen the curtains?").

RESULTS

Data analysis employed two-tailed Mann-Whitney U tests and Spearman's rho correlation coefficients.

The results are shown in Table 1. Comparison subjects performed at ceiling on the Hayling Sentence Completion Task while six patients made errors, but this difference did not quite reach statistical signifi-

TABLE 1. Patient and Comparison Performance on the Executive Tasks, Theory of Mind Vignettes, and Faux Pas Task

Measure	Patients (n=16)		Comparison (n=8)		Analyses	
	Mean	SD	Mean	SD	Mann-Whitney U	p
Hayling Sentence Completion Task (inhibition)						
Part B times	4.402	2.599	2.143	0.568	13	0.005**
Part B errors	0.562	0.892	0	0	40	0.052
Digit Ordering Test—Adapted (working memory)						
Max digit span	6.219	1.016	6.313	0.258	54.5	0.548
	%	Fraction	%	Fraction	Mann-Whitney U	p
Theory of mind vignettes						
All errors	7.03	9/256	0	0/128	44	0.083
Faux pas task						
Recognition errors	14.84	19/128	3.13	2/64	58	0.017*
Belief errors	26.56	17/64	3.13	1/32	30	0.021*
Fact recall errors	10.94	14/128	9.34	6/64	61	0.843

*significant at 0.05
**significant at 0.01

cance. However, patients took significantly longer than comparison subjects to respond to inhibitory items, indicating possible inhibitory dysfunction. No significant difference was found between patients and comparison subjects on the Digit Ordering Test—Adapted.

The patient group made nine errors on the theory of mind vignettes, and though comparison subjects performed at ceiling, this difference was not statistically significant. Patients made errors on counterfactual, memory, and reality questions, providing no evidence for a specific deficit in false belief.

Patients were significantly poorer than comparison subjects at recognizing faux pas, but their recall of factual information contained in the vignettes was not significantly different.

More errors were made by patients than comparison subjects on faux pas belief questions, even on occasions when they identified faux pas. When failing to attribute a false belief to the perpetrator, patients often inferred the offensive remark was intentional. In such cases, explanations for the faux pas remark included anger, jealousy, or negative personality traits such as “nasty,” “mean,” “a bitch,” or “sarcastic.”

No significant correlations were evident for patients’ performance on executive and theory of mind measures.

Patients were grouped according to whether they reported obsessive-compulsive symptoms. These groups did not differ for performance on the Hayling Sentence Completion Task (times: Mann-Whitney U=8, p=0.079; errors: Mann-Whitney U=24.5, p=0.361), Digit Ordering Test—Adapted (Mann-Whitney U=31.5, p=0.0957),

theory of mind vignettes (Mann-Whitney U=20.5, p=0.138), or faux pas task (recognition: Mann-Whitney U=31.5, p=0.955; belief errors: Mann-Whitney U=19, p=0.148; fact recall: Mann-Whitney U=21, p=0.219). However, in relation to the nonsignificant findings it may be noted that sample size meant power value may be estimated at below 0.20.

DISCUSSION

Patients with Tourette’s syndrome made errors on theory of mind tasks despite unimpaired working memory and accurate recall of factual information contained in the vignettes. Theory of mind deficits were most apparent on faux pas tasks, where patients were specifically impaired on belief questions and inappropriately assumed the faux pas was intentional. This pattern of deficits was not associated with the presence of obsessive-compulsive symptoms and did not correlate with inhibitory problems as shown by the Hayling Sentence Completion Task. The pattern, however, is similar to that seen in patients with frontal-variant frontotemporal dementia.¹²

Belief deficits on the faux pas task may have occurred because the task involves attributing intentions. Thus, patients’ difficulties may reflect not a deficit in theory of mind competence but rather a difference in application. Patients may be capable of understanding other beliefs but apply theory of mind reasoning differently in certain social situations. Developmental research shows that when belief and outcome information conflict, adults’ judgments are determined primarily by the be-

lief while young children may fail to integrate beliefs and intentions and make judgments based on outcome alone.¹³ Negative consequences therefore lead to negative attributions about an actor, regardless of whether the outcome was intended.

Frontostriatal dysfunction may reduce patients' cognitive resources, leading to difficulties with the cognitively demanding task of reasoning about beliefs relative to reasoning based on consequences. Orbitofrontal and medial prefrontal activity has been linked to reasoning about others' intentions.¹⁴ These regions are also active when processing first- or third-person perspective and transgressions of social norms, along with the

anterior cingulate gyrus, the temporal poles, and precuneus.¹⁵ Activity in these regions may vary depending on whether transgressions are considered intentional or unintentional.¹⁶ Changes in these brain regions could be associated with alterations in the attribution of intentions in Tourette's syndrome.

Overall, these findings suggest that social difficulties may arise in Tourette's syndrome from a lack of understanding of the intentionality of social actions. Although the small sample size may lead to caution about the generalizability of these findings as core characteristic features of Tourette's syndrome, further investigation is clearly merited.

References

1. Kurlan R, Daragfari C, Como PG, et al: Non-obscene socially inappropriate behavior in Tourette's syndrome. *J Neuropsychiatry Clin Neurosci* 1996; 8:311-317
2. Burgess PW, Shallice T: Response suppression, initiation, and strategy use following frontal lobe lesions. *Neuropsychologia* 1996; 34:263-273
3. Channon S, Pratt P, Robertson MM: Executive function, memory, and learning in Tourette's syndrome. *Neuropsychology* 2003; 17:247-254
4. Flynn E: The role of inhibitory control in false belief understanding. *Infant Child Dev* 2007; 16:53-69
5. Samson D, Apperly IA, Kathirgamanathan U, et al: Seeing it my way: a case of a selective deficit in inhibiting self perspective. *Brain* 2005; 128:1102-1111
6. Saxe R, Schulz LE, Jiang YV: Reading minds versus following rules: dissociating theory of mind and executive control in the brain. *Soc Neuroscience* 2006; 1:284-298
7. Baron-Cohen S, O'Riordan M, Stone V, et al: Recognition of faux pas by normally developing children and children with Aspergers syndrome or high-functioning autism. *J Autism Dev Disord* 1999; 29:407-418
8. Channon S, Sinclair E, Waller D, et al: Social cognition in Tourette's syndrome: intact theory of mind and impaired inhibitory functioning. *J Autism Dev Disord* 2004; 34:669-677
9. Apperly IA, Samson D, Chiavarino C, et al: Frontal and temporoparietal lobe contributions to theory of mind: neuropsychological evidence from a false-belief task with reduced language and executive demands. *J Cogn Neurosci* 2004; 16:1773-1784
10. Werheid K, Hoppe C, Thone A, et al: The adaptive digit ordering test: clinical application, reliability, and validity of a verbal working memory test. *Arch Clin Neuropsychol* 2002; 17:547-565
11. Robertson MM, Eapen V: The national hospital interview schedule for the assessment of Gilles de la Tourette syndrome and related behaviors. *Int J Methods Psychiatr Res* 1996; 6:203-226
12. Gregory C, Lough S, Stone S, et al: Theory of mind in patients with frontotemporal dementia and Alzheimer's disease: theoretical and practical implications. *Brain* 2002; 125:752-764
13. Young L, Saxe R: The neural basis of belief encoding and integration in moral judgment. *Neuroimage* 2008; 40:1912-1920
14. Blakemore SJ, den Ouden H, Choudhury S, et al: Adolescent development of the neural circuitry for thinking about intentions. *Soc Cogn Affect Neurosci* 2007; 2:130-139
15. Cavanna AE, Trimble MR: The precuneus: a review of its functional anatomy and behavioral correlates. *Brain* 2006; 129:564-583
16. Berthoz S, Armony JL, Blair RJR, et al: An fMRI study of intentional and unintentional (embarrassing) violations of social norms. *Brain* 2002; 125:1696-1708