Can children with Gilles de la Tourette syndrome edit their intentions?\footnote{Some parts of this paper were presented in invited lectures by the first author at the University of Rochester, New York (October 1991), the Charing Cross Hospital and the Institute of Neurology (September 1992), London.}

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SYNOPSIS In this paper we describe a cognitive mechanism, the Intention Editor, which is triggered whenever there are several intentions competing in parallel with each other. This mechanism is hypothesized to be a subcomponent of a larger mechanism, the Supervisory Attentional System (SAS: Shallice, 1988) which serves inhibition in general. The Intention Editor interrupts one of several simultaneously activated intentions, preventing it from executing its action, utterance, or thought. This mechanism appears to develop during the first five to six years of life. We propose that an impairment in the development of this mechanism may account for the triad of symptoms in children with Gilles de la Tourette Syndrome (GTS): involuntary movements, involuntary utterances, and obsessive thoughts. This mechanism is tested with normal children aged 3–6 years old, and with children with GTS, in two experiments.

In Experiment 1, subjects were required to make one hand movement while inhibiting making a (different) hand movement that the other hand was simultaneously making. In Experiment 2, they were asked to say one thing while inhibiting saying something else. On both tasks, normal 6-year-olds were significantly better than normal 4-year-olds, but children with GTS performed worse than normal 6-year-olds, despite having a mean age of 12 years. These results constitute preliminary evidence for the theory that the Intention Editor is dysfunctional in GTS.

INTRODUCTION

For over 100 years, Gilles de la Tourette Syndrome (GTS) has been the subject of investigation and curiosity by neurologists and psychiatrists. Until recently, neurologists saw this disorder primarily as a disturbance of the motor system, while psychiatrists for a long time considered it to be primarily psychological and possibly even psychogenic. Recent advances have refuted both of these views (Cohen, 1991). While motor tics are one key symptom of the disorder, the other key symptom (vocal tics) and the associated feature of obsessive thinking suggest that GTS cannot be ‘simply’ a motor disorder. Similarly, the responsiveness of some of the symptoms to medication (Cohen et al., 1979; Caine, 1985; Shapiro et al. 1988) has contributed evidence for the biological basis of this disorder. Additional evidence for its biological origin has come from brain-imaging studies, which reveal abnormalities in the frontal cingulate cortex and the inferior corpus striatum (Chase et al. 1984, 1986). Leckman et al. (1991) also summarize the evidence for abnormalities in the basal ganglia in this group. Finally, a biological cause to the disorder is implicated by genetic studies, which suggest a single autosomal dominant gene with varying penetrance and a broad range of expression (Kidd, 1980; Kurlan et al. 1986; Pauls & Leckman, 1986; Robertson & Gourdie, 1990).

Surprisingly, cognitive neuropsychology has almost ignored this syndrome (Robertson & Baron-Cohen, 1994). Although psychologists have carried out standardized psychometric assessments with these patients (Shapiro et al. 1974; Incagnoli & Kane, 1981), experimental investigations into the cognitive system of patients with GTS are few and far between

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(Channon et al. 1992), and certainly there is no cognitive model of this disorder. This gap is all the more remarkable given that the syndrome is now widely regarded as a broader disorder of cognition, rather than a narrow motor disturbance. It was to fill this vacuum that the present research was begun.

**Gilles de la Tourette syndrome**

The first clear description of this syndrome was made by Itard (1825). Georges Gilles de la Tourette (1885) went on to describe eight more similar cases, emphasizing a *triad of symptoms*: multiple tics, coprolalia (unprovoked swearing) and echolalia (repetitive speech). Current diagnostic criteria for GTS require the presence of multiple motor tics, and at least one vocal tic, both of which must exceed a year’s duration and begin before the age of 21 years (DSM-III-R, AMA, 1987). The location, number, frequency, complexity and severity of the tics characteristically change over time.

Although the exact prevalence of Tourette’s syndrome is unknown, a currently accepted figure is 1 in 1000 males, and 1 in 10000 females (Leckman et al. 1991). The mean onset of the symptoms is seven years of age. It is found in all social classes and across cultures (Robertson, 1989).

As Gilles de la Tourette (1885) suggested, a triad of symptoms tends to cluster in these patients, justifying the use of the term ‘syndrome’. In the light of more recent research, we suggest that the triad be conceptualized in terms of (a) motor tics, (b) vocal tics, and (c) obsessive thinking. Different patients may manifest all or any combination of these three symptoms. We briefly summarize these three areas of abnormality as follows.

**Motor tics** The most frequent initial motor tics involve the eyes (such as eye blinking), head nodding and facial grimacing. Patients also often exhibit movements such as touching, hitting, jumping, spitting, kicking and stamping, as well as abnormalities of gait (Robertson, 1989). Finally, motor tics with specific content (such as echopraxia, or the tendency to copy seen gestures involuntarily; and copropraxia, involuntary obscene gestures) may also be present.

**Vocal tics** These are non-verbal (e.g. throat-clearing, barking, hissing and clicking) or are word related (e.g. palilalia, involuntary syllable repetition, and jargon), or are clearly verbal (e.g. explosive utterances, coprolalia, word-tics and echolalia).

**Obsessive thoughts** Many patients with GTS also show Obsessive-Compulsive Behaviour [OCB] (Yaryura-Tobias et al. 1981; Montgomery et al. 1982; Frankel et al. 1986; Green & Pitman, 1986; Robertson et al. 1988). In addition, many relatives of patients with GTS also report obsessive thoughts and compulsive actions, in the absence of tics (Kurlan et al. 1986; Pauls et al. 1986a,b; Robertson & Gourdie, 1990; Robertson & Trimble, 1991).

In this paper, we describe a cognitive model of GTS which aims to explain the above triad of symptoms in terms of an underlying cognitive deficit or deficits. Such an approach has been used in explaining other neurodevelopmental disorders, such as autism (Baron-Cohen et al. 1985; Baron-Cohen, 1990) and schizophrenia (Gray et al. 1991), and the rationale for using such an approach has been well articulated by Frith et al. (1991). Essentially, the cognitive approach tests theories of the mind’s functional mechanisms, and specifies the bridge between brain and behaviour. In the present case, we suggest that the triad of motor tics, vocal tics and obsessive thoughts may all arise as a result of an abnormality in a highly specific cognitive mechanism, the Intention Editor. The function of this mechanism is described next.

**THE INTENTION EDITOR**

Frequently, a specific mechanism may go unrecognized in the normal system until its dysfunction reveals its existence in a particular clinical population. Indeed, cognitive neuropsychology is premised on the notion that we learn about the normal through studying the abnormal (McCarthy & Warrington, 1990). Our model of GTS led us to postulate the existence of the Intention Editor as a part of the normal system (Baron-Cohen et al. 1994). Since most of our everyday psychological functioning never reveals its workings, we had taken it for granted. The internal architecture of this mechanism will not be described in detail here, but for the purposes of explicating our model, certain assumptions need to be made plain.

First, we assume that, at a certain level of
description, intentions drive much of what we do. They drive our actions, our speech, even some of our thought. Intentions are representations of future action, speech or thought. Such representations contain information specifying not only the goal-state of a future action, utterance, or thought, but also information about how to implement or reach that goal state.

Secondly, we assume that even after intentions have been accessed and activated, there is a point at which they can still be edited. As representations, they can be read, their future impact evaluated, and then either left to run or be interrupted. If the latter, they are then 'dumped' in favour of alternative intentions. Such intention editing goes on not only at the conscious level – every time we 'change our mind' – but at a preconscious level too. The assumption that intentions are separable from the actions they drive becomes clearer by a thought experiment: we can all imagine intending to do, say or think something, but then never translating that intention into action, speech or thought. In a moment of anger, you might intend to raise your fist, or intend to shout some obscenity, but instead decide to smile sweetly, say how you agree and turn on your heel. This example illustrates how actions, speech and thought are driven by intentions, but that intentions are separable from these, insofar as intentions can be terminated before they have 'run' to completion.¹

Thirdly, we assume that intentions vary in terms of their potential for triggering the Editor. That is, some intentions are dangerous (e.g. the intention to hit, etc.) and/or are socially disapproved of (e.g. the intention to swear in formal, public settings, etc.). In our model, these sort of intentions are therefore tagged with high values. Other intentions are relatively harmless (e.g. the intention to fiddle with one's hair) and/or are socially acceptable (e.g. the intention to whistle a tune while walking in public). In our model, these are therefore tagged with low values. The Intention Editor is triggered more readily by intentions with higher values.

Fourthly, we assume that the Intention Editor is only triggered when there are two or more competing intentions that have been simultaneously activated, only one of which can actually be executed. Thus, when the task is simply Do x (e.g. push the blue button), this may entail just one intention being activated. We call this a Single Intention Task. When the task requires the subject to perform a sequence of actions, one at a time, such as Do x, then do y (e.g. push the blue button, then the red button), this too may entail just one intention being activated and executed at a time. We call this a (short) Serial Intention Task. Similarly, if the task is Do x, then do y, then do z (e.g. push the red button, then the blue button, then the green button), this may still only entail one intention being activated and executed at a time, even though the sequence has lengthened. We call this a (long) Serial Intention Task. In contrast, the Intention Editor is necessarily triggered in Parallel Intention Tasks, in which the subject has to do two different things simultaneously. Examples of Parallel Intention Tasks will be given in the experiments, reported below. The different Task Types are summarized in Table 1, along with analyses of each Task Type in terms of the number of intentions that are minimally activated in each.

Our final assumption is that the Intention Editor is part of our central processes. It does not operate on any one output system (motor, speech or thought), but rather it operates on the intentions themselves.

The hypothesis we set out to test was that in children with GTS there is an abnormality in the development of this mechanism. In this paper, we report two experimental tests of this claim. This hypothesis seemed plausible in that a failure of the Intention Editor would result in actions being performed involuntarily, words and sounds being uttered involuntarily, and a difficulty in 'getting rid' of unwanted, intrusive thoughts – that is, the triad of symptoms in GTS. Moreover, we predicted this deficit would be seen only when the task involved several competing, simultaneously activated intentions (Parallel Intention Tasks), but would not be seen in control conditions (Simple or Serial Intention Tasks). In the first experiment, we tested the use of the Intention Editor in a motor task.
Table 1. Different task types

<table>
<thead>
<tr>
<th>Task description*</th>
<th>Intention analysis of each task</th>
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<tbody>
<tr>
<td>Simple intention task</td>
<td>Do x.</td>
</tr>
<tr>
<td>(Short) Serial intention task</td>
<td>Do x; then do y.</td>
</tr>
<tr>
<td>(Long) Serial intention task</td>
<td>Do x; then do y; then do z.</td>
</tr>
<tr>
<td>Parallel intention task</td>
<td>Do x and y simultaneously.</td>
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</tbody>
</table>

* The task descriptions given are for one trial only. Naturally, in a test, each of these would also include the instruction ‘then repeat, N times’.

McCarthy & Warrington (1990) discuss this task in relation to a pattern of errors Luria and other neurologists and neuropsychologists saw in patients with bilateral damage to the premotor cortex. These errors fell into 3 types: (a) non-fluent alternating; (b) non-simultaneous alternating; and (c) failure to alternate. The second and third of these error patterns are illustrated in Fig. 2.

We predicted that if children with GTS were unable to edit intentions, then they would show errors on this task. In addition, we predicted that such errors would not occur in a normal control group of children. In order to collect data with this task from normal children, we assessed a number of different age control groups. Finally, we predicted that children with GTS would not show errors on two different
control tasks. The first control task was a (short) Serial Intention Task and therefore did not necessarily involve the Intention Editor. This task was as follows: open hands, then close hands, both hands performing the same action at the same time. In formal terms, it falls under the description Do x, then do y. The second control task was a (long) Serial Intention Task. This task was one of Luria’s (1966) sequential movement tasks, the ‘fist-edge-palm’ test: using one hand, first make a fist, then hold out the hand in a rigid and vertical position, then turn the hand so that the palm faces upwards. In formal terms, it falls under the description Do x, then do y, then do z.

Subjects
We assessed five groups of children, each group containing 15 subjects. Four of these were different age groups of normal children, attending nursery and primary classes of schools in an inner city area of London. The age groups of the normal children were 3-, 4-, 5- and 6-year-olds. The fifth group comprised children with a clear DSM-III-R (APA, 1987) diagnosis of GTS who were attending the GTS clinic at the National Hospital for Neurology and Neurosurgery in London. They had been referred from all over the UK. All of these patients were diagnosed by M.R., who is well acquainted with GTS (Robertson et al. 1987, 1988, 1989, 1993; Robertson & Gourdie, 1990; Robertson & Trimble, 1991). The children with GTS ranged from 6 to 18 years of age, with a mean of 12·31 years. As will be noted, the normal control groups were younger than the clinical group. If anything, that should have given a general developmental advantage to the children with GTS. If they still performed worse on a given task than the normal controls, despite their seniority, this would constitute strong evidence in favour of a specific deficit in this group. The ages of the subjects are summarized in Table 2.

None of the normal subjects were receiving any special education, and they were thus assumed to be of normal intelligence. The children with GTS were assessed using three subtests of the WISC-R or WAIS-R (block design, vocabulary and picture completion), as well as a test of language development (the BPVS, or British Picture Vocabulary Scale [Dunn et al. 1982]). The three IQ subtests were selected on the basis that one was verbal, one non-verbal and a third (block design) a task in which children with GTS had been reported to have problems (Incagnoli & Kane, 1981). Full IQ assessment was not given, in order to keep testing to a minimum, as these young subjects were also being used in several other research and clinical trials. All of these subjects were within the average to superior range on these three subtests (scaled scores between 10 and 18).

Method
Each child was tested individually in a quiet room, either in their school (in the case of the normal children) or in a laboratory at the Institute of Neurology (in the case of the children with GTS). The experimenter sat opposite the child, and in the Experimental Task, said ‘I would like to see if you can do this with your hands. Watch me first. Keep one hand open, and
Table 2. Chronological ages (in years) of subjects

<table>
<thead>
<tr>
<th>Group (N = 15 in each)</th>
<th>Mean CA</th>
<th>s.d.</th>
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<tbody>
<tr>
<td>3-year-olds</td>
<td>3.57</td>
<td>0.28</td>
</tr>
<tr>
<td>4-year-olds</td>
<td>4.43</td>
<td>0.27</td>
</tr>
<tr>
<td>5-year-olds</td>
<td>5.49</td>
<td>0.27</td>
</tr>
<tr>
<td>6-year-olds</td>
<td>6.49</td>
<td>0.31</td>
</tr>
<tr>
<td>GTS</td>
<td>12.31</td>
<td>3.52</td>
</tr>
</tbody>
</table>

one hand closed, then change over, so that now this one is open, and this one is closed. Then change back again. Try to do it 10 times. Remember, as you open one hand, close the other one at the same time. OK? Off you go.' The experimenter then counted how many continuous, fluent, simultaneous alternations the subject could produce.

If the subject succeeded in producing 10 fluent, simultaneous alternations, that was the end of the task. If the subject stopped after fewer than 10 alternations, or made errors within the 10 alternations, he or she was asked to have another go, again being shown the correct method. Subjects were given a maximum of 5 trials. On each trial, the experimenter recorded how many fluent, simultaneous alternations the subject had produced. For the later analysis, each subject's best score was used.

Each child with GTS was also given the two control tasks, described earlier. The tasks were given in random order. In fact, all subjects with GTS achieved perfect performance (10 repetitions) on these control tasks.

Results

Table 3 shows the mean best score for each group, on the experimental task. As can be seen, there is a clear progression among the normal groups, from 3 through to 6 years of age. Three-year-olds were significantly worse than 4-year-olds ($t = 2.4, 28$ df, $P < 0.01$) and 4-year-olds significantly worse than 5-year-olds ($t = 3.9, 28$ df, $P < 0.01$), though the 5- and 6-year-olds did not differ ($t = 1.2, 28$ df, $P > 0.05$). The five-year-olds were the youngest normal group to show ceiling performance (10 fluent, simultaneous alternations in a row), although this was not attained on the first trial by the majority of 5-year-olds. In contrast, all but 3 of the 6-year-olds were able to produce 10 correct alternations on the first trial, the other 3 doing so on the second trial. As predicted, the children with GTS made more errors than either the 5- or the 6-year-olds (GTS × 5-year-olds, $t = 4.5, 28$ df, $P < 0.001$). Finally, all three error types occurred, both among the normal children and the children with GTS. The results are also shown graphically in Fig. 3.

Discussion

On Luria's Hand Alternation Task, normal 5- and 6-year-olds showed ceiling performance, after a relatively protracted development of this skill in the preceding 2 years. In contrast,

Table 3. Mean best score of each group (maximum = 10) on the Luria Hand-Alternation Task, Experiment 1

<table>
<thead>
<tr>
<th>Group (N = 15 in each)</th>
<th>Mean best score</th>
<th>s.d.</th>
</tr>
</thead>
<tbody>
<tr>
<td>3-year-olds</td>
<td>2.6</td>
<td>2.6</td>
</tr>
<tr>
<td>4-year-olds</td>
<td>5.5</td>
<td>3.7</td>
</tr>
<tr>
<td>5-year-olds</td>
<td>10</td>
<td>0</td>
</tr>
<tr>
<td>6-year-olds</td>
<td>10</td>
<td>0</td>
</tr>
<tr>
<td>GTS</td>
<td>6.7*</td>
<td>2.9</td>
</tr>
</tbody>
</table>

* GTS × 5-year-olds, $P < 0.001$.

Fig. 3. The Luria Hand Alternation Test (mean best score, maximum = 10).
children with GTS, despite being considerably older, made significantly more errors than the 5- and 6-year-olds. Given the similar deficits on this task by Luria’s patients with damage to the frontal cortex, this would implicate a role for frontal pathology in children with GTS. There is some evidence for frontal abnormalities in GTS, from neuroimaging studies (Chase et al. 1984, 1986). The results from Experiment 1 also constitute preliminary evidence in support of the hypothesis that these children have an abnormality in the development of the Intention Editor. However, by itself this result might simply indicate a motor deficit, not a central impairment. This is particularly important because the task is quite complex in motor terms. In Experiment 2, we therefore employed an equivalent non-motor task, described next.

EXPERIMENT 2: THE YES AND NO GAME

Whereas in the experimental task in Experiment 1 the subject had to execute one intention while inhibiting another in the motor domain, in Experiment 2 we tested the subject’s ability to do the same in the domain of speech. We used a well-known game that is seen in parent–child play as well as child–child interaction, but which had not been used as a formal experimental procedure before, to the best of our knowledge. In this game, the subject is told that to win the game he or she must not say ‘Yes’ or ‘No’ in response to anything the experimenter might ask. Since these words are the usual ways of answering closed questions, the subject has to inhibit the strongly activated intention to say these words, and instead think of alternative ways to word their answer. For example, the experimenter might ask a subject called Jeffrey ‘Is your name Jeffrey?’, to which the subject could reply ‘It is’, or ‘That’s right’, etc. While this task does not parallel the motor task in all of its overt respects (e.g. in speech it is impossible to produce two observable responses simultaneously, while in movement this is possible), they are nevertheless similar in one key respect: they are both Parallel Intention Tasks, in that they both involve two intentions being activated simultaneously, one of which is edited and the other of which is translated into action.

Subjects
The 15 subjects from the GTS group in Experiment 1 took part in this, as did the 15 subjects from each of the 4- and 6-year-old normal groups. The 3- and 5-year-old normal groups were not included, as Experiment 1 had suggested that if the Yes and No Game was tapping the same central mechanism, the developmental shift of interest in the Yes and No Game was likely to be seen between age 4- and 6-years-old. One normal six-year-old from Experiment 1 was not available for testing in Experiment 2, so he was replaced with a new subject of the same chronological age (CA). This did not, therefore, affect the mean CA of the 6-year-olds.

Method
Subjects were tested individually, either on the same occasion as Experiment 1, or on a separate day. The experimenter said ‘We are going to play a game called the Yes and No Game. Would you like to play? Good. Now, the rules are that if you say Yes or No, then you lose. But if you don’t say Yes or No to any of my questions, then you win. Let’s see if you can do this, shall we? OK. First, let’s have a practice’. At this point, five training questions were asked, in order to check that the subject understood the rules. After feedback and correction, the subject was then told the game was starting for real now. The experimenter then asked questions (with flexible content, adapted to the circumstances of the individual subject), and continued asking questions until the subject made an error, or until the subject had answered 10 questions without saying Yes or No. This scoring system paralleled that used in Experimental 1. Thus, if the child said Yes or No before 10 correct responses had been given, this was deemed to end one trial, and a new trial was started. The maximum number of trials given was 5. Once 10 correct responses were given, the trial and the Experiment ended. The experimenter recorded the number of questions answered correctly in each trial, the subject’s best score, as well as the trial number in which the best score was achieved.

In addition, the subjects with GTS were given a control task in which they simply had to say Yes, No, 10 times in a row. This is a (short)
Serial Intention Task, since in formal terms it falls under the description Do x, then do y. There were 10 trials, in random order. Any errors made were recorded. It was chosen in order to test the prediction that this would not present any difficulty for subjects with GTS, given that it could be carried out without having to activate two different intentions simultaneously.

Results

All of the subjects with GTS were at ceiling on the control task. Table 4 shows the mean best score on the Experimental Task for each group. As can be seen, there is again a clear progression among the normal groups, from 4 to 6 years of age. Four-year-olds were significantly worse than 6-year-olds ($t = 3.4, 28 \text{ df}, P < 0.001$). All but 4 of the 6-year-olds were able to produce 10 correct alternations on the first trial, the others doing so on the second or third trial. We assume that 6-year-olds reflect the adult level of competence on this task. Again, as predicted, the children with GTS made more errors than the 6-year-olds (GTS x 6-year-olds, $t = 3.9, 28 \text{ df}, P < 0.0005$). The 4-year-olds did not differ from the children with GTS ($t = 0.1, 28 \text{ df}, P > 0.9$). The results are also shown graphically in Fig. 4.

Table 4. Mean best score of each group (maximum = 10) on the Yes and No Game, Experiment 2

<table>
<thead>
<tr>
<th>Group (N = 15 in each)</th>
<th>Mean best score</th>
<th>S.D.</th>
</tr>
</thead>
<tbody>
<tr>
<td>4-year-olds</td>
<td>6.5</td>
<td>3.9</td>
</tr>
<tr>
<td>6-year-olds</td>
<td>10</td>
<td>0</td>
</tr>
<tr>
<td>GTS</td>
<td>6.7*</td>
<td>3.3</td>
</tr>
</tbody>
</table>

* GTS x 6-year-olds, $P < 0.0005$.

Discussion

Once again, both developmental changes and pathology were identified on this task. There was an improvement between ages 4 and 6 in the normal groups, paralleling changes observed on the Luria Hand Alternation Task. Children with GTS, despite their chronological age advantage, were significantly worse than normal 6-year-olds at inhibiting an intention to say one thing (e.g. ‘Yes’) while executing an intention to say something else (e.g. ‘That’s right’). This was not due to failure to understand the rules of the game, as subjects often showed their awareness that they had made an error, by laughing and expressing ‘annoyance’ at their mistakes. Neither was it due to either perseveration, or an inability to follow simple instructions, or an inability to participate in a game, as their almost perfect performance on the control task demonstrated. It was only the Parallel Intention Task, which activates two different intentions simultaneously, and which is, therefore, held to require the Intention Editor, that produced the deficit in subjects with GTS. These results confirm the hypothesis that the deficit detected in Experiment 1 is not specifically motor in nature, but reflects a central system which operates on a range of output systems. We discuss this possibility in more detail in the final part of the paper.

GENERAL DISCUSSION

In this paper we have presented evidence suggesting that children with GTS suffer from a specific cognitive deficit in the Intention Editor, as reflected in two Parallel Intention Tasks, in the motor and language domains. While these
Can GTS patients edit their intentions?

tasks differ in several aspects, they are formally comparable in that they both require the child to inhibit one intention while executing another simultaneously activated intention. In both experiments, children with GTS performed at a level significantly lower than normal 6-year-olds (though not significantly different from normal 4-year-olds).

In entertaining the assumption that these tasks tap this proposed mechanism, it is also necessary to consider alternative interpretations. Perhaps the most obvious alternative is the notion that these errors simply reflect a general loss of inhibition. We agree that a loss of inhibition must be part of the explanation, but loss of inhibition of what? It cannot simply be a loss of the ability to inhibit actions, pure and simple. If this was all the deficit amounted to, then children with GTS should also have had difficulty with the (long) Serial Intention (control) Task in Experiment 1, which they did not. Similarly, one might have expected them to show an inability to stop performing a repetitive task when given an unexpected ‘Stop!’ command. In fact, all of our subjects were able to stop a finger tapping task immediately when so instructed. They were also able to pass the control task in Experiment 2. Rather, this deficit appeared to be a loss of the ability to inhibit one intention while simultaneously executing another. It is in this sense that there is a failure in editing. In addition, the general ‘loss of inhibition’ hypothesis does not capture the important distinction between actions and intentions.

A second possibility is that errors on these tasks reflect poor speed-accuracy budgeting. Kelso (1984) has used tasks similar to the Luria Hand Alternation Task with normal subjects, and shown that as soon as hand alternation has to be performed above a critical speed, subjects start moving them in synchrony instead. He refers to this as a ‘phase shift’, a widespread phenomenon in motor coordination (e.g. the shift from trotting to galloping in horses). Might it be that with development normal children simply learn how fast they can go without provoking a phase shift, while both younger normal children and children with GTS are poor at making this assessment (Nigel Harvey, personal communication)? Although we did not ask our subjects to go as quickly as they could, this hypothesis prompted us to try inserting an instruction in the Hand Alternation Task to go slowly, but we still found deficits in the group with GTS. Independently, Adele Diamond (personal communication) found that 3- and 4-year-olds showed similar problems on the Hand Alternation Task even when asked to go very slowly. We therefore suspect that speed-accuracy judgements were not the cause of errors on this task in the group with GTS, though this remains to be systematically tested. Similarly, in the Yes and No Game, speed was not a consideration at all.

A third possibility is that the deficits shown by the children with GTS simply reflect attentional problems during the experimental task, and that the control tasks were somehow simpler in terms of their attentional demands. However, given that our subjects with GTS were able to pass the (long) Serial Intention Task in Experiment 1, which if anything contained a longer sequence of information to be held in memory than the Parallel Intention Task, we do not consider attentional demands can account for their poor performance on Parallel Intention Tasks. It would be worthwhile for future studies in this area to investigate if such deficits persist when task complexity is reduced even further.

Apart from alternative interpretations of the data, it is also important to explain how the Intention Editor relates to other cognitive mechanisms that have been described. Perhaps the most closely related mechanism is Frith & Done’s (1989) Intention Monitor. Frith & Done suggest that the Intention Monitor checks if an action driven by a ‘willed intention’ (a goal or plan) achieved what it set out to achieve, or whether error-correction (Rabbitt, 1966) is required. Similarly, it monitors if actions that are externally driven (by a ‘stimulus intention’) are successful. Finally, the Intention Monitor keeps track of which of these two routes generated any given action. Frith (1987) argues that in schizophrenia there are impairments in the Intention Monitor, leading Type I schizophrenic patients for example to misclassify an action generated by a willed intention as being generated externally by a stimulus intention, or Type II schizophrenic patients for example to fail to link willed intentions with actions.

We can be sure that children with GTS do not have a deficit in the Intention Monitor, in that
they are fully able to report when they have made a mistake, they can correctly identify the locus of responsibility for their mistakes, and they do not show any of the ‘negative signs’ of Type II schizophrenia. While it is likely that the Intention Editor is intimately associated with the Intention Monitor, it has its own distinct function: editing competing instructions. Thus, while the Intention Monitor’s key function is to track the source of an intention (internal v. external, or willed v. stimulus) and its effects, the Intention Editor’s key role is to select among competing willed intentions.

Another related system that is important to discuss is Shallice’s (1988) Supervisory Attentional System (SAS). To understand the SAS, we must first mention what Shallice calls the Contention Scheduling System (CSS) which responds to external stimuli and repeats relevant, routine actions. Reaching out (without looking) to pull the light cord on entering the bathroom would be a good example (that Shallice cites) of such a routine action programme, triggered simply by walking into your bathroom. The SAS oversees the CSS, by activating or inhibiting alternative schemata. The primary function of the SAS is to respond to novelty. To continue the previous example, if you know that the light bulb in the bathroom blew yesterday, then on entering the bathroom the SAS should inhibit the CSS from executing the routine action of reaching out for the light cord. Shallice (1988) has proposed that the SAS is impaired in patients with frontal lobe syndrome, resulting in disinhibition of actions.

Again, we can be sure that children with GTS are not generally impaired in the SAS in that they switch to new actions in an action sequence, and are not simply stimulus-driven in their actions. However, it seems likely that the Intention Editor is a special component within the SAS. It is not used for inhibition in general (unlike the SAS), but is a highly specific mechanism, dedicated to editing out one of several intentions that are competing in parallel. We suggest that: a next important step will be to test children with GTS using traditional ‘frontal lobe’ neuropsychological tasks which are held to tap the SAS more systematically in the future, in order to establish if the deficit in children with GTS is specific to the Intention Editor, or is general to all tasks with SAS involvement. It will also be essential to offer a task analysis of such tests in terms of the levels of Task Type suggested in Table 1, in order to specify which of the traditional ‘frontal lobe’ neuropsychological tasks might also require the Intention Editor.

Before closing, it is worth thinking about how a deficit in the Intention Editor might underlie the key symptoms in the behaviour of children with GTS. We have seen how it comes into operation in laboratory tasks involving simultaneously competing intentions. In the real world, such tasks might include talking politely to someone who you think little of (editing the intention to use impolite language), or sitting quietly while working (editing the intention to jump up or move around). Motor and vocal tics, as well as coprolalia and copropraxia, can be seen as exactly the kinds of behaviours one would expect to see if this mechanism failed.

What about the relationship between the Intention Editor and thought? By thought, we mean the process of thinking about particular topics, either intentionally or unintentionally. Normal subjects experience unwanted, intrusive thoughts (Rachman et al. 1976), but report being able to dismiss such thoughts fairly efficiently. Once again, we assume that such an ability in the normal person reflects the functioning of the same mechanism, editing out one intention and selecting an incompatible alternative. Patients with Obsessive Compulsive Disorder (OCD) frequently complain that an unpleasant thought (e.g. ‘I’ve left the gas taps on, and the family is going to die’) cannot be ‘got rid of’ (Rachman & Hodgson, 1980). That patients with GTS frequently also have OCB may be further evidence that the abnormality in the Intention Editor can affect not just motor and verbal behaviour, but thought itself. This specific prediction that the same mechanism might underlie this triad of symptoms requires further testing, but one possibility is that in subjects where the Intention Editor deficit only affects thought, the only clinical sign will be OCB/OCD. In contrast, in subjects where the Intention Editor deficit affects several output systems, the clinical signs may include motor and vocal tics, and OCB/OCD.

It is clear that, to the extent that the data reported here implicate an abnormality in the Intention Editor, this was not found in all of the children with GTS. There are several possible
explanations for this. First, since the disorder is a developmental one, any degree of developmental delay or deviance in the functioning of this mechanism may change with age. Indeed, to test if there is some development in the Intention Editor in patients with GTS, we are currently testing an adult sample of patients. This will also help answer if the impairments in the Intention Editor are a necessary feature of GTS. Secondly, the mechanism may be open to damage to varying degrees of severity, or in different ways in different patients. It may even suggest the needs for subgroups within the overall diagnostic category of GTS. A third possibility is that the Intention Editor interacts with other psychological factors (such as stress) in complex ways. Such a possibility might account for widely reported within-subject variations in the severity of the symptoms (Cohen et al. 1988). Finally, given that the disorder can take an episodic form, there is a possibility that the cognitive dysfunction may also be episodic, rather than the all-or-none variety. Future work needs to test these different explanations.

Finally, as regards the normal development of the Intention Editor mechanism, we have demonstrated that perfect performance on either the Luria Hand Alternation Task or the Yes and No Game is not seen until 5 or 6 years of age. The question arises as to why 2- and 3-year-old normal children, whose Intention Editor is not yet fully mature, nevertheless do not show GTS symptoms. It may be that, in certain respects, young normal children are actually quite "disinhibited". In other respects, young normal children may be quite different in their development to children with GTS. Such possibilities remain to be examined systematically. A full understanding of the normal development of the ability to inhibit intentions is still a long way off, but children with GTS may allow us to glimpse the workings of the brain-based cognitive mechanisms that subserve development in this domain, starkly displayed in their dysfunctional state.

We are grateful to Donald Cohen for inspiring this research, and for constructive discussions of Tourette syndrome over many years. We would like to thank the staff at the following schools for their cooperation with this research: Camberwell Day Nursery, Helena Day Nursery and Comber Grove School. We are also indebted to Michael Trimble for his support and guidance in this work. We have also benefited from discussions about this work with Jeffrey Gray, Nigel Harvey, Chris Frith, Dick Passingham, John Harrison, Adele Diamond, Oliver Chadwick and Derek Bolton.

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