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Parkinsonism and Related Disorders 9 (2003) 281–289

Parkinsonism &
Related Disorders

www.elsevier.com/locate/parkreldis

Tics and developmental stuttering

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Received 4 July 2002; revised 24 December 2002; accepted 26 December 2002

Abstract

Background. Developmental stuttering affects 1% of the population but its cause remains unclear. Recent PET studies of metabolism in the central nervous system suggest that it may be related to dysfunction in the basal ganglia or its connections with regions of the cortex associated with speech and motor control.

Objective. To determine the presence and characteristics of involuntary movements (IMs) in people who stutter and to investigate the hypothesis that these movements may be of a very similar nature to the IMs seen in patients with movement disorders due to basal ganglia dysfunction.

Methods. Sixteen adults with developmental stuttering and 16 controls matched for sex and age were audio–videotaped while freely speaking 300 words in conversation and reading aloud 300 words. The audio data was inspected for dysfluencies and the video data was scrutinised for the presence and characteristics of IMs.

Results. Subjects who stuttered produced more IMs than controls during free speech (354 vs 187, $p < 0.05$) and reading (297 vs 47, $p < 0.001$). Most of the IMs in both groups were tics, with a greater number of both simple and complex motor tics (CMTs) in subjects who stuttered. CMTs were more frequent than simple motor tics in those who stuttered, but not in controls. The combination of repetitive eye blink followed by prolonged eye closure was found exclusively in the stuttering group, as were simple tics consisting of eyebrow raise or jaw movement. Dystonia in the form of blepharospasm was identified in a small number of subjects who stuttered. Choreic movements were not associated with stuttering.

Conclusions. Developmental stuttering is associated with the presence of IMs that are predominantly simple and CMTs. This association suggests that tics and stuttering may share a common pathophysiology and supports the view that, in common with tics, stuttering may reflect dysfunction in the basal ganglia or its immediate connections.

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Keywords: Developmental stuttering; Basal ganglia; Involuntary movements; Tics

1. Introduction

Developmental stuttering begins in early childhood and affects about 1% of the population [1]. The cause of this form of stuttering is unknown but genetic studies suggest that it is an inherited disorder [2–4]. Stuttering may be

associated with altered neurotransmitter levels [5–7] or a defective dopaminergic pathway [8–11]. These observations, together with recent evidence using modern imaging techniques of brain function, support the proposal that the cause of stuttering may lie in dysfunction of the basal ganglia or related systems within the CNS.

1.1. Imaging studies

A study using positron emission tomography (PET) found decreased metabolism in the regions of the cerebral

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cortex associated with speech and language control (i.e. Broca's area, Wernicke's area and frontal pole) and in the left caudate nucleus during stuttering with reduced metabolism in the left caudate even during fluent speech of the subjects who stuttered [12]. Two SPECT studies showed reductions in cerebral blood flow (CBF) in regions of the cerebrum responsible for speech-motor control [13,14].

A PET study by Ingham et al. [15] found no significant difference in resting-state regional cerebral blood flow (rCBF) between adult subjects who stuttered and those who did not. Conversely, Fox et al. [16], using solo reading to induce stuttering, found widespread over-activation in both cerebrum and cerebellum, particularly on the right side. In addition, the auditory system, which is thought to support the self-monitoring of speech, lacked the predominant left-side activation found in controls. Induced fluency with chorus reading largely reversed the abnormal activation patterns. A subsequent PET study by Braun et al. [17] supported Fox et al. [16] particularly with respect to altered right-sided brain activity during stuttered speech, but found that it did not normalise during fluent speech. They also observed activation of the right basal ganglia and bilateral cerebellum during stuttering.

A recent PET study by Fox et al. [18] compared metabolism during rest, fluent and dysfluent speech and found increased rCBF in the regions of the primary motor cortex, supplementary motor cortex, Broca's area and anterior insula on the non-dominant right cortex and the non-dominant left cerebellum, with decreased activation in the primary auditory and associated areas in the right hemisphere during stuttering. They concluded that stuttering is associated with both over-activation and deactivation of these neural pathways.

A study using volumetric magnetic resonance imaging [19] found anomalous anatomy in perisylvian speech and language areas of the grey matter in adults with developmental stuttering. In the first study to use diffusion tensor imaging, Sommer et al. [20] showed a significant reduction in diffusion in the left sensorimotor cortex below the laryngeal and tongue representation of the group who stuttered compared to a peer group. Packman and Onslow [21] draw attention to the possibility of there now being sufficient evidence to propose that stuttering has a definite neuroanatomical basis.

In summary, there appears to be an anatomical or functional dissociation between areas in the brain that control motor output and those that influence this output by auditory feedback and language processing. This dissociation could be due to an overactive presynaptic dopamine system with decreased metabolism in regions that normally control speech.

1.2. Neuropharmacology

Ludlow and Braun [5] proposed that stuttering may be associated with altered neurotransmitter levels. Support for

this notion comes from the finding that stuttering may be relieved by administration of the anticholinergic drug biperiden [6] and the serotonin reuptake blocker clomipramine [7]. The efficacy of the dopaminergic receptor blocking agent haloperidol in decreasing the dysfluency rate in people who stutter [8–11] implicates a defective dopaminergic pathway as a possible mechanism for stuttering. Presynaptic dopamine activity was measured with PET by Wu et al. [22] to investigate this. They found substantial increases in dopamine uptake activity in the cortex and subcortical regions associated with speech.

1.3. Acquired stuttering

Further support for the notion that stuttering is due to a deficit within the basal ganglia comes from observations of patients with acquired stuttering following penetrating missile wounds to the brain. These patients had lesions predominantly in the basal ganglia and in the connections between basal ganglia and cortex [23]. Acquired stuttering has also been noted as an occasional early symptom of Parkinson's disease (PD) [24,25] implicating possible involvement of the dopaminergic system in stuttering. The mechanism is unclear, however, as PD is associated with decreased activity of the dopamine system, while stuttering appears to be associated with an increase in activation of this system. There may be differences, however, in the type of dysfluency found in speakers with PD and those who stutter which could explain this apparent paradox [6]. Alternatively, it could be that individuals with PD who also stutter, may have transient increase in levels of dopamine from levodopa therapy leading to stuttering. This latter speculation is consistent with a study of a male with developmental stuttering and PD treated with levodopa who was found to be less fluent during 'on' periods [26]. In contrast, Shahed and Jankovic [27] found speech patterns similar to developmental stuttering in 12 patients with PD who had been childhood stutterers and whose stuttering had re-emerged after the development of the PD. In these patients, there appeared to be no consistent improvement or worsening of symptoms with levodopa therapy. Further support to the notion that stuttering is due to a deficit within or related to the basal ganglia comes from the finding that stimulation of the thalamus, for the relief of pain, has an ameliorating effect on the dysfluency in patients with acquired stuttering [28]. The thalamus is the major output relay of the basal ganglia.

1.4. Motor control

There is evidence to suggest that people with developmental stuttering have neural motor control difficulties of a more global nature than that confined to processing of speech alone. Observations include delay in time to initiate speech [29], decrease in task initiation and co-ordination of the hands [30–33], increased visual reaction times [34] and

subtle abnormality of saccadic eye movements [35,36]. Deficits have also been reported in non-motor functions including visuoperception [37] and response to cognitive stress [38].

1.5. Associated movements

Movements that do not appear to be part of facial expression or related to speech have long been described as a secondary feature of stuttering. These movements have been variously termed ancillary body movements [39], accessory features [40], non-verbal accessory behaviours [41], associated symptoms [42], secondary symptoms [43], physical concomitants [44] and non-speech behaviour [45]. They have been investigated by estimating the incidence of the individual movements [45–49] and have traditionally been viewed as an attempt to overcome the difficulty producing sounds or avoidance response to interaction with a listener [1,45,47].

There have, however, been alternative explanations for these movements. Their similarity to involuntary movements (IMs) seen in patients with ‘defined dystonic syndromes’ was emphasised in a study of 23 people with developmental stuttering [6]. Others have noted a relationship between stuttering and Tourette’s syndrome [50], with Joseph et al. [51] proposing that stuttering may represent a vocal tic. Abwender et al. [52] also suggest a pathogenetic relationship between developmental stuttering and Tourette’s syndrome after identifying tics in 11 of 22 adults with developmental stuttering. It is pertinent to note other possible relationships between stuttering and Tourette’s syndrome; a male to female ratio of 3:1 in the incidence of both developmental stuttering [1] and tics [53] and familial inheritance patterns found in both disorders [1,53,54].

If stuttering is caused by some defect in the basal ganglia it would not be unreasonable to conclude that the associated IMs are also a consequence of the same defect and hence a primary feature of the stuttering phenomenon. It is of interest then, that stuttering has increased prevalence in the families of patients with idiopathic torsion dystonia, a genetically determined movement disorder considered due to a basal ganglia abnormality [55].

1.6. Purpose of study

There is evidence as discussed above from a variety of sources that stuttering is a result of disordered motor control. If the movements accompanying stuttering are similar to the IMs seen in patients with basal ganglia disorders, it would be an attractive proposition that dysfunction of the basal ganglia might be linked to or be the cause of developmental stuttering. This study sought to identify and characterise the IMs in stuttering and consider their potential relationship with basal ganglia dysfunction.

2. Methods

2.1. Subjects

Ethical approval for this study was obtained from the Canterbury Ethics Committee, with informed consent obtained from all subjects. The experimental group comprised 16 subjects with developmental stuttering and without diagnosed neurological or psychiatric condition. English was their preferred language. Subjects were included in the study if they produced three or more stuttering dysfluencies per 100 words of conversational speech or reading. Stuttering dysfluencies were defined as prolongation and repetition of sound or syllable as defined in Riley’s Stuttering Severity Instrument [44].

The study subjects (mean age = 41.7, SD = 14.6, range = 15–67 years) were matched by age (± 5 years if adult and ± 1 year if under 20 years) and gender with a fluent healthy peer to form the control group (mean age = 41.3, SD = 15.8, range = 16–70 years). There were 11 males and five females in each group. Eight of the subjects who stuttered and two of the control subjects had immediate or extended family members who also stuttered. Of the subjects who stuttered, three were classified as very mild, six as mild, six as moderate, none as severe and one as very severe on the Riley’s Stuttering Severity Instrument [44]. None were on medication for stuttering.

2.2. Procedure

Subjects were seated comfortably in a quiet room in front of a plain background. A videocamera (Sony Hi8 CCD-TR910E) was placed on a tripod 3 m away from the subject. A lapel microphone (Sony Electret ECM-T110) was attached to the subject’s clothing 15–20 cm away from the mouth. A reading passage of 350 words was taken from a popular magazine, enlarged and mounted on a stand and placed at a distance of 1.0–1.5 m in front of the subject depending on his/her preferred reading distance.

Subjects were asked to read the passage aloud in their normal voice and speed whilst being audio–videotaped. This was followed by 5 min of conversational speech with the subject asked to speak about their job, family or interests.

The reading task consisted of the middle 300 words of the reading passage. The speech task consisted of 300 words of free speech, taken by transcription from the middle of the 5 min of conversational speech.

Each individual movement of the face, head and upper body considered involuntary and abnormal was identified and described according to location and duration by a neurologist (TJA) with a specialist interest in movement disorders. Full subject data from both free speech and reading tasks were presented randomly for analysis. The audio was turned off and the neurologist was not made explicitly aware of which data were from which group.

Movements were identified only if they were not considered to be part of normal facial expression (e.g. smiling) or movement (e.g. eye blinks) or gesture (e.g. head movements in agreement or disagreement) and not part of the mechanics of speech. Individual movements were grouped into those of the upper face (prolonged eye closure, repetitive eye blinking, brief eyebrow arching, and movements of the eyes to each side or upwards), lower face (facial grimace, mouth grimace, repetitive lip movement, lip smacking, lip pursing, tongue protrusion, forceful swallowing and jaw shuddering), movements of the head and neck (head jerking and sustained movements of the neck to either side), or movements of the upper limbs (arm jerking or hand scratching).

Classification of IMs was performed by two neurologists (TJA and IMD), both specialists in movement disorders. Full data of the two tasks were again presented randomly and with audio off. If necessary, the videotape was replayed until a consensus decision was made. The neurologists were asked to review each individual movement previously identified, decide if the movements were isolated or part of a more complex series of movements, then classify them as definitely normal, probably abnormal, or abnormal. The probably abnormal and abnormal movements were then further classified by body part according to the following:

- (i) myoclonus; movements faster and briefer than normal and appearing as sudden shock-like movements
- (ii) simple tic; abrupt, sudden, brief, isolated movements [56]
- (iii) complex motor tic; more co-ordinated and complicated movements than simple motor tics [56] and occurring simultaneously or in sequence
- (iv) stereotypy; co-ordinated, patterned, rhythmic, purposeless but seemingly purposeful movements or postures that occur repetitively [57]
- (v) mannerism; a gesture peculiar or unique to the individual that may at times seem stereotypical [57]
- (vi) chorea; irregular, purposeless, non-rhythmic and non-patterned movements that flowed randomly from one part of the body to another, and were unpredictable in time and distribution
- (vii) dystonia; slower, sustained movements [56].

Many movements originally identified as involuntary movements (IMs) in the first section of this study by the single neurologist were deemed to be part of a movement complex in the subsequent classification exercise by the two neurologists. These were called complex motor tics (CMTs) and included stereotypies and mannerisms deemed as abnormal.

2.3. Statistical analysis

The data was considered to be non-parametric, despite being interval, because it was highly skewed (particularly in

the stuttering group) and there were substantial differences in variances between the two groups. The Wilcoxon matched-pairs test (one-tailed for comparison between experimental and control subjects and two-tailed for within-group comparisons) was used for inferential statistics.

3. Results

3.1. Involuntary movements during free speech

All sixteen experimental subjects and 15 control subjects exhibited IMs during free speech. Subjects who stuttered had more IMs than controls (overall total of 354 vs 187 with a mean number per subject of 22.1 vs 11.7, $p = 0.013$) during free speech. Differences were found in number of prolonged eye closures (41 vs 1, $p = 0.009$) and head jerks (46 vs 8, $p = 0.026$). Marginal differences were found in repetitive eye blinking (46 vs 19, $p = 0.071$) and mouth grimacing (13 vs 5, $p = 0.078$) (Table 1).

Table 1
IMs during free speech task

	Stuttering group			Control group			<i>p</i>
	<i>S</i>	Total	Range	<i>S</i>	Total	Range	
<i>Movements in upper face</i>							
Prolonged eye closure	7	41	0–18	1	1	0–1	**
Semi-prolonged eye closure ^a	7	21	0–10	8	32	0–11	
Repetitive eye blink	10	46	0–10	4	19	0–10	~
Brief eyebrow arch	12	32	0–6	13	32	0–7	
Prolonged eyebrow arch	3	11	0–6	6	9	0–3	
Eyes right	6	3	0–6	1	2	0–2	
Eyes left	6	21	0–6	4	15	0–12	
Eyes up	3	10	0–6	0	0		
<i>Movements in lower face</i>							
Facial grimace	4	17	0–8	5	8	0–3	
Mouth grimace	7	13	0–3	3	5	0–3	~
Repetitive lip movement	2	3	0–2	0	0		
Lip smack	1	1	0–1	0	0		
Lip purse	4	21	0–8	1	1	0–1	
Tongue protrusion	4	13	0–8	4	8	0–4	
Forceful swallow	1	1	0–1	0	0		
Jaw shudder	1	14	0–14	0	0		
<i>Movements of head and neck</i>							
Head jerk	7	46	0–12	8	8	0–1	*
Sustained head right	3	3	0–1	7	8	0–2	
Sustained head left	3	6	0–3	4	6	0–3	
<i>Movements of upper limbs</i>							
Right arm jerk	4	6	0–2	3	4	0–2	
Left arm jerk	6	15	0–5	6	17	10–6	
Right hand scratch	2	5	0–4	4	7	0–3	
Left hand scratch	4	5	0–2	3	5	0–2	
Total movements	16	354	0–55	15	187	0–21	*

~ $p < 0.1$, * $p < 0.05$, ** $p < 0.01$; *S*, number of subjects exhibiting the IM; total, total number of IMs in the group; range, the range in number of IMs exhibited by each subject in the group.

^a Brief eye closure (i.e. blinks) were not included as these were deemed part of normal facial expression/movement.

3.2. Involuntary movements during reading

Subjects who stuttered had a greater number of IMs (overall total of 297 vs 47, mean number per subject of 18.6 vs 2.9, $p = 0.001$) during reading. Two of the subjects who stuttered and four of the controls did not have IMs when reading. Twelve types of IMs were found solely in the stuttering group although some were quite infrequent. Four IMs were found to occur with a significantly greater prevalence in the stuttering than in the control group: repetitive eye blinking (10 vs 0, $p = 0.034$), mouth grimacing (12 vs 0, $p = 0.034$), head jerks (55 vs 7, $p = 0.043$) and right arm jerks (17 vs 1, $p = 0.014$). Brief eyebrow arching was marginally different (60 vs 18, $p = 0.063$) (Table 2).

Table 2
IMs during reading task

	Stuttering group			Control group			<i>p</i>
	S	Total	Range	S	Total	Range	
<i>Movements in upper face</i>							
Prolonged eye closure	0	0		0	0		
Semi-prolonged eye closure ^a	1	7	0–7	0	0		
Repetitive eye blink	4	10	0–6	0	0		*
Brief eyebrow arch	8	60	0–23	8	18	0–7	
Prolonged eyebrow arch	3	8	0–5	2	7	0–5	
Eyes right	0	0		0	0		
Eyes left	0	0		0	0		
Eyes up	2	5	0–4	0	0		
<i>Movements in lower face</i>							
Facial grimace	2	5	0–4	0	0		
Mouth grimace	4	12	0–5	0	0		*
Repetitive lip movement	2	25	0–23	0	0		
Lip smack	0	0		0	0		
Lip purse	4	10	0–5	0	0		
Tongue protrusion	4	9	0–3	3	9	0–5	
Forceful swallow	1	6	0–6	0	0		
Jaw shudder	2	52	0–44	0	0		
<i>Movements of head and neck</i>							
Head jerk	7	55	0–21	4	7	0–4	*
Sustained head right	1	1	0–1	0	0		
Sustained head left	2	2	0–1	0	0		
<i>Movements of upper limbs</i>							
Right arm jerk	6	17	0–9	1	1	0–1	*
Left arm jerk	6	9	0–4	4	5	0–2	
Right hand scratch	1	1	0–1	0	0		
Left hand scratch	0	0		0	0		
Total movements	14	297	0–52	12	47	0–14	**

* $p < 0.05$, ** $p < 0.01$; S, number of subjects exhibiting the IM; total, total number of IMs in the group; range, the range in number of IMs exhibited by each subject in the group.

^a Brief eye closure (i.e. blinks) were not included as these were deemed part of normal facial expression/movement.

Table 3
Overall classification and frequency of IMs

	Stuttering group		Control group	
	S	Total	S	Total
<i>Free speech</i>				
Simple motor tics	13	42	10	29
CMTs	15	149	12	42
Dystonia	3	13		
Chorea	1	1	1	1
<i>Reading</i>				
Simple motor tics	11	31	2	2
CMTs	14	121	4	8
Dystonia				
Chorea			1	1

S, number of subjects who exhibited one or more of the IM; total, total number of IMs in the group.

3.3. Overall classification of involuntary movements

By far the greater majority of IMs in both subject groups were classified as motor tics, both simple and complex. There were no instances of myoclonus, and infrequent dystonic and choreic movements (Table 3). Of the 96% of IMs classified as tics in the group that stuttered, 21% were classified as simple tics, with 79% being CMTs.

3.4. Movements classified as simple tics

IMs classified as simple tics were recorded according to body region during free speech and reading (Table 4). Simple tics of the eyebrows were found exclusively in the stuttering group during free speech (total of 5, $p = 0.033$). Simple tics of the jaw were also found exclusively but to a smaller extent in this group. No significant differences in the number of simple tics involving other body regions were found between the two groups. In all, there were 13 subjects in the stuttering group and 10 subjects in the control group who exhibited simple tics during free speech, with a marginal difference found in the overall number between groups (42 vs 29, $p = 0.056$) (Table 4).

In contrast, the stuttering group had considerably more simple tics than controls during reading (31 vs 2, $p = 0.003$), with 11 subjects in this group but only two in the control group exhibiting simple tics. The two controls each had one simple tic either of the arm or eyebrows, while subjects in the stuttering group had simple tics in various regions of the upper body, particularly of the eyebrows, face and mouth (Table 4).

3.5. Movements classified as complex tics

There were more CMTs in the stuttering group than in controls during both free speech (149 vs 42, $p = 0.005$) and reading (121 vs 8, $p = 0.001$). Since there were a wide

Table 4
IMs classified as simple tics

Body region	Stuttering group		Control group		<i>p</i>
	<i>S</i>	Total	<i>S</i>	Total	
<i>Free speech</i>					
Repetitive eye blink	5	11	2	6	
Eyes	1	1	1	3	
Eyebrows	4	5	0	0	*
Face or mouth	2	2	4	6	
Jaw	2	3	0	0	
Head	6	15	4	4	
Upper limbs	4	5	6	10	
Total	13	42	10	29	~
<i>Reading</i>					
Repetitive eye blink	1	5	0	0	
Eyes	0	0	0	0	
Eyebrows	3	9	1	1	~
Face or mouth	3	8	0	0	~
Jaw	1	1	0	0	
Head	2	3	0	0	
Upper limbs	3	5	1	1	
Total	11	31	2	2	**

S, number of subjects who exhibited one or more tics; total, total number of IMs in the group; ~ $p < 0.10$, * $p < 0.05$, ** $p < 0.01$.

variety of CMTs, the data were collapsed into five categories, defined largely by body region: upper face, lower face, head tilt, head jerk and upper limb jerk for simplification (Table 5). The stuttering group exhibited

more CMTs in the upper face (mean 9.8 vs 2.4, $p = 0.005$), head jerks (2.8 vs 0.6, $p = 0.023$) and upper limb jerks (1.3 vs 0.1, $p = 0.029$) during free speech (Table 5). The combination of repetitive eye blinking followed by prolonged eye closure was seen only in the subjects who stuttered, with five of these subjects showing this pattern. Only one subject in this group exhibited no CMTs during the speech task, although this subject had a single simple tic. In contrast, four of the control group had no CMTs and one of these, the youngest, had no other IMs.

The differences between the two groups in CMT prevalence were even more apparent during reading. The stuttering group exhibited more CMTs than controls in the upper face (mean 6.2 vs 0.4, $p = 0.001$), lower face (2.2 vs 0, $p = 0.002$), head tilt (1.9 vs 0, $p = 0.004$), head jerk (2.9 vs 0.3, $p = 0.02$) and upper limb jerk (1.8 vs 0.2, $p = 0.013$) (Table 5). Unlike free speech, however, there was no combination of repetitive eye blinking with prolonged eye closure in either group of subjects during reading.

3.6. Movements classified as dystonia

A number of dystonic movements, characterised as blepharospasm, combined with other IMs in a CMT were seen in three subjects who stuttered, with the CMT persisting for up to 15 s (for example, one of these consisted of a sequence of eyes elevation, eyebrows elevation, prolonged and squeezing eye closure i.e. blepharospasm, accompanied by a series of head jerks). Other types of dystonia were not seen.

Table 5
CMTs classified according to body region during free speech and reading

Body region	Number								
	Stutterers				Controls				<i>p</i>
	<i>S</i>	Mean	Median	Range	<i>S</i>	Mean	Median	Range	
<i>Free speech</i>									
Upper face ^a	15	9.8	6.5	0–35	11	2.4	2.0	0–11	**
Lower face ^b	3	0.3	0	0–2	0	0	0	0	~
Head tilt ^c	10	2.0	1.0	0–8	7	1.0	0	0–5	~
Head jerk ^d	10	2.8	1.0	0–17	5	0.6	0	0–5	*
Upper limb jerk ^e	6	1.3	0	0–8	2	0.1	0	0–1	*
<i>Reading</i>									
Upper face	13	6.2	3.0	0–21	4	0.4	0	0–3	**
Lower face	11	2.2	1.0	0–18	0	0	0	0	**
Head tilt	11	1.9	1.0	0–10	0	0	0	0	**
Head jerk	7	2.9	0	0–14	3	0.3	0	0–3	*
Upper limb jerk	7	1.8	0	0–13	2	0.2	0	0–2	*

~ $p < 0.10$, * $p < 0.05$, ** $p < 0.01$; *S*, number of subjects with CMTs in the particular body region; mean, mean number of CMTs exhibited in the group; median, median number of CMTs exhibited in the group; range, the range in number of CMTs exhibited by each subject in the group.

^a Repetitive eye blinking, prolonged eye closure, sustained or repetitive eyebrow raising or face grimace.

^b Mouth grimace, jaw shudder, contraction of platysma muscle or unusual tongue protrusion.

^c Retrocollis, torticollis or antecollis.

^d A single sharp jerk of the head on the neck, or a series of rapid nodding movements of the head.

^e Elevation or backward thrust of a shoulder, rapid abduction of an arm from the shoulder, rapid flexion of an elbow.

3.7. Movements classified as chorea

Choreic movement was identified once in one subject in each group during free speech and in one control subject during reading.

4. Discussion

This study demonstrates that adults with developmental stuttering have a higher number of involuntary or associated movements than their fluent peers and confirms similar findings in children [45]. We have further observed that the vast majority of IMs are simple and CMTs. These observations lead us to suggest that stuttering may be a tic disorder.

Only a few dystonic movements around the eyes (blepharospasm) were evident in subjects who stuttered. In contrast, Kiziltan and Akalin [6] considered the IMs seen in their male subjects with developmental stuttering to be similar to those seen in dystonic syndromes. They suggested that stuttering might be a focal or segmental action dystonia. Clearly there has been a difference in interpretation of the phenomenology of the movements observed between the present study and that of Kiziltan and Akalin. There is a need for further studies to be done in order to resolve this disparity.

The movements classified as tics in this study were very similar to those described as body tics by Lees [58], being abrupt, jerky and repetitive movements involving discrete muscle groups including eye winking, eye rolling, eyebrow raising, nasal flaring and mouth grimacing. In the present study, tics of the eyes, eyebrows, grimacing of the face or mouth and jerking of the head or arms were found to occur more in the stuttering group than controls, with movements of the mouth and jaw peculiar to this group during free speech. During reading, tics around the eyes, grimacing of the mouth and face and some sustained postures of the neck were also peculiar to the group who stuttered. Movements classified as CMTs comprised IMs occurring simultaneously or in sequence. Stereotypies, in contrast are, 'patterned, repetitive, purposeless movements that are performed the same way each time' [59]. They have usually been associated with conditions such as obsessive-compulsive disorders, Tourette's syndrome, schizophrenia, autism and mental retardation, although they can be seen in normal individuals particularly during periods of stress or anxiety [59]. Distinguishing between stereotypies and some other movement disorders can be difficult. Tan et al. [59] drew attention to the similarity between complex tics and stereotypies. In the present study we included stereotypies deemed as abnormal in the category of movements classified as CMTs because the movement patterns were so similar and difficult to distinguish apart.

It is notable that simple and complex tics were also present in the control group, albeit at a considerably lower

frequency. Conture and Kelly [45] also found considerable overlap in the type of movements observed in their group of children who stuttered and the matched group of fluent controls, although the authors did not attempt to classify the movements in any way. It might be questioned whether movements classified in our study were true IM's or whether some could have been normal emotional facial expression. The difficulty in classifying tics [53] must be acknowledged as a potential criticism of this study. However, we believe the classifications are valid since this was done by two experienced neurologists with a special interest in movement disorders, as well as the data being presented randomly and with the audio turned off. Thus the classification was done in a blinded fashion so that the neurologists did not know whether a subject or control was stuttering or not [46]. This methodological approach ensured that any misclassification of normal expressive movements as associated (abnormal) movements was equal across the groups, thus not influencing the results and subsequent conclusions.

Additional evidence in support of the suggestion that stuttering may be a tic disorder comes from the similarities in the pharmacological effects on tics and stuttering [8]. Antidopaminergic agents, such as haloperidol, have been used to alleviate both tics [58–60] and stuttering [8,10]. Serotonin reuptake blocking drugs such as reserpine and clomipramine have also been used successfully for tics [61] and stuttering [7,62]. Dopamine plays a critical role in the circuits of the basal ganglia [63], whilst serotonin influences mood through the limbic system, also now thought to be influenced by the basal ganglia [63,64].

In addition to the suppression of tics by dopamine receptor antagonists, there is other evidence for basal ganglia dysfunction in Tourette's syndrome. This includes neuro-imaging studies using MRI [65], SPECT [66] and PET [67] showing an abnormality in structural and functional relationships between the left and right sides of the brain, particularly in the basal ganglia region.

Tics and developmental stuttering share a number of other characteristics. There is a similar typical age of onset of between 3 and 8 years in otherwise normal children who present with tics [58] and developmental stuttering [1]. Further, there is often spontaneous remission of tics [58] and stuttering [1] during early adolescence. Additionally, there is a high incidence (16% compared with 4% in age-matched controls) of stuttering in ticquers [58]. Both ticquers and people who stutter have more tics and stutters, respectively, during times of anxiety, anger and self-consciousness [58]. Alarcón and Lees [50] reported a young man who had stuttered since the age of 5 years, had concomitant tic-like facial movements, and subsequently developed behavioural disorders. They suggested that the movements bore a close resemblance to those seen in ticquers and concluded that there may be a link between Tourette's syndrome and stuttering. Josephs et al. [51] supported this notion by suggesting that stuttering seen in a 35 year old man with Tourette's syndrome may represent a vocal tic. In summary,

the similarities in age of onset, behaviour of symptoms, and effect of certain neurotransmitter drugs on symptoms, support a link between tics and stuttering.

Caruso [31] has reviewed a growing body of data which suggests that stuttering is a neuromotor deficit involving temporal control of both speech and non-speech movements. He suggests that, because of the nature of these motor difficulties (timing and co-ordination of speech, eye and finger movements), there may be deficient functioning in areas such as the supplementary motor areas (SMA) and/or the basal ganglia. There are direct connections between the basal ganglia and the SMA [63] and both of these areas are important to speech and motor control [31]. Caruso's suggestion is supported by findings in a study by Mulligan et al. [46]. This study identified abnormal movements (i.e. not normal facial expression) accompanying speech in adults who stuttered and compared them to a normally fluent control group. The findings that some of the abnormal movements accompanied both stuttered and fluent speech in people who stutter supports the notion that stuttering and the accompanying movements are due to altered function in a motor control system wider than that of speech motor control alone. Recent studies using PET implicate the SMA and basal ganglia in motor control including speech production and stuttering [16,17,22].

The findings from imaging studies, the effect of antidopaminergic drugs on both movement disorders and stuttering, and the observation in this study that the movements accompanying stuttering resemble tics collectively suggest that stuttering and its associated movements is a tic disorder due to basal ganglia dysfunction or possibly a more widespread dysfunction of the dopaminergic system. Future research is required to learn whether the occurrence of IMs would decrease with whispering, choral reading, delayed auditory feedback or other frequency enhancing techniques, or indeed, with treatments that address stuttering. In addition, it will be important to explore further similarities between tics and stuttering by investigating whether IMs can be suppressed in a similar manner to tics.

Acknowledgements

We wish to thank Dr Chris Frampton for assistance with statistical analysis of the data. This study was supported by a grant from the Canterbury Medical Research Foundation.

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