Loss of sensory attenuation in patients with functional (psychogenic) movement disorders

Isabel Pareé, Harriet Brown, Atsuo Nuruki, Rick A. Adams, Marco Davare, Kailash P. Bhatia, Karl Friston and Mark J. Edwards

1 Sobell Department of Motor Neuroscience and Movement Disorders, UCL Institute of Neurology, Queen Square, London, UK
2 The Wellcome Trust Centre for Neuroimaging, University College London, London WC1N 3BG, UK
3 Department of Information Science and Biomedical Engineering, Kagoshima University, Kagoshima, Japan

Correspondence to: Dr Mark J. Edwards
Sobell Department,
UCL Institute of Neurology
Queen Square,
London WC1N 3BG, UK
E-mail: m.j.edwards@ucl.ac.uk

Functional movement disorders require attention to manifest yet patients report the abnormal movement to be out of their control. In this study we explore the phenomenon of sensory attenuation, a measure of the sense of agency for movement, in this group of patients by using a force matching task. Fourteen patients and 14 healthy control subjects were presented with forces varying from 1 to 3 N on the index finger of their left hand. Participants were required to match these forces; either by pressing directly on their own finger or by operating a robot that pressed on their finger. As expected, we found that healthy control subjects consistently overestimated the force required when pressing directly on their own finger than when operating a robot. However, patients did not, indicating a significant loss of sensory attenuation in this group of patients. These data are important because they demonstrate that a fundamental component of normal voluntary movement is impaired in patients with functional movement disorders. The loss of sensory attenuation has been correlated with the loss of sense of agency, and may help to explain why patients report that they do not experience the abnormal movement as voluntary.

Keywords: sensory attenuation; agency; functional movement disorders; attention; force-matching
Abbreviations: FMD = functional movement disorder; HADS = Hospital Anxiety and Depression Scale; PDI = Peters Delusions Inventory

Introduction

Patients with functional motor symptoms account for about half of patients with functional neurological symptoms (Stone et al., 2010). Diagnosis is made on the basis of positive clinical characteristics: the dysfunctional movement requires attention to manifest, and when the patient is distracted, the dysfunction improves or disappears. The necessity of attending the movement for the dysfunction to manifest might be expected to be associated with a strong sense of ‘voluntariness’ or agency for the movement. However, patients report that the abnormal movement is involuntary. If these patients are not feigning, why do they experience a movement that appears objectively to be voluntary as involuntary?

Sensory attenuation is a phenomenon whereby the intensity of sensation caused by self-generated movement is reduced (Blakemore et al., 1998, 2000; Shergill et al., 2003). A common example of this is the observation that one cannot tickle oneself. Sensory attenuation can be understood as a decrease in the (attentional) gain of the sensory consequences of one’s own actions. Under an active inference model of human movement generation (Brown et al., 2013), this reduction in gain is necessary to allow movement to occur, as it allows predictions of movement to be...
resolved by generating actual movements via spinal reflex arcs rather than being explained away by sensory evidence that the movement has not yet occurred. The experience of sensory attenuation is reported to be important in labelling movements as self-generated and a loss of sensory attenuation has been associated with a loss of agency for movement (Blakemore et al., 2002).

In the experimental setting, sensory attenuation has been most commonly assessed with the force matching paradigm (Shergill et al., 2003, 2005). Here, subjects are asked to match a force delivered to their finger; either by pressing directly on their own finger with the other hand, or by operating a joystick that, via a non-linear transform, causes a robot arm to press down on their finger. Healthy subjects consistently generate more force than required when directly pressing on their finger compared with using the joystick. It has been proposed that the excess force exerted in the first condition reflects sensory attenuation of the sensory consequences of self-generated movements, something not present in the second condition, where the non-linear transform between movement and sensation disrupts the sense of agency. As a loss of sensory attenuation improves force-matching, poor performance is difficult to feign in this paradigm, and also provides an opportunity to assess a key psychophysical property of movement that is experienced as voluntary. Here, we hypothesized that patients with functional movement disorders (FMDs) would have abnormal sensory attenuation for movement.

Material and methods

Participants

Fourteen patients with a diagnosis of clinically established or documented FMD (Fahn and Williams, 1988) and 14 healthy participants matched with respect to gender, age, and handedness (Oldfield, 1971) were recruited. Patients with FMD involving upper limbs or with sensory abnormalities were excluded. The study was approved by the local ethics committee. Participants gave their written consent to take part in the studies, which were carried out in accordance with the Declaration of Helsinki.

Questionnaires

We administered the Hospital Anxiety and Depression Scale (HADS) (Zigmond and Snaith, 1983) to assess mood. To estimate the non-verbal intelligence level of participants, the 12-item short form of Raven’s Progressive Matrices Test was used (Raven, 1977).

Delusional ideation was assessed using the Peters Delusions Inventory (PDI-21) (Peters et al., 2004). Total scores range from 0 to 336, with higher scores reflecting higher delusional ideation.

Procedure

Detailed description of the materials and the experimental design is provided in the online Supplementary material. Each participant was tested in a single experimental session consisting of two main conditions. They were asked to match a target force delivered by the arm of a small robot, either by pressing on themselves with their right index finger on the left index finger (self-condition) (Fig. 1A) or by manipulating horizontally a second robot which controlled the output of the other robot that applied a force vertically to the left index finger (Fig. 1B). Five different target forces (16 trials of each), increasing in increments of 0.50 Newton (N) from 1 N to 3 N, were randomly presented in both conditions. All subjects completed a total of four blocks of 20 trials each (80 trials in total) for each condition. The order of conditions was counterbalanced across participants. None of the applied forces was experienced as painful either in patients or healthy participants.

Our measure of sensory attenuation was the ratio between the matched force and the target force (ratio > 1 indicating generation of excessive force). This measure was averaged across trials to give the mean attenuation for each force level and condition.

Statistical analysis

SPSS Statistics software (version 21.0.0) was used for the analysis. Normality of errors was assessed by using the Kolmogorov-Smirnoff test. When not normally distributed, the data were subjected to a log10 transformation. P-values for categorical variables were calculated with the use of Fisher’s exact test. Mann-Whitney U test was used to compare differences for numerical data in baseline characteristics.

A three-way mixed design ANOVA was used to compare the results of the main experiment with Condition (self versus external) and Force (1 N, 1.5 N, 2 N, 2.5 N, 3 N) as within subject variables and Group (patients versus healthy participants) as a between-subject factor. For post hoc explorations, we conducted a two-way mixed design ANOVA for self and external conditions, respectively, with Force (1 N, 1.5 N, 2 N, 2.5 N, 3 N) as within subjects variables and Group (patients versus healthy participants) as a between-subjects factor. Finally, we analysed patients and healthy participants separately with a two-way repeated measures ANOVA with Condition (self versus external) and Force (1 N, 1.5 N, 2 N, 2.5 N, 3 N) as within subject variables. We calculated potential associations between IQ, PDI-21 and HADS and the amount of sensory attenuation (calculated as the mean of the ratios for each force level in the self-condition) by using Pearson’s correlation. Statistical significance of P < 0.05 was assumed.

Results

Baseline characteristics of the participants are shown in Table 1. Patients and healthy participants were matched for Raven’s and PDI scores. Most patients were females and had functional fixed dystonia of the lower limbs as FMD. Clinical features included an acute onset and rapid escalation of the symptoms after a minor injury. Most presented a placebo response after receiving botulinum toxin injections (n = 7) or following examination under anaesthesia (n = 2) and dystonic symptoms disappeared for a period
of time to recur later on in five patients. Two patients were taking gabapentin, two were taking tramadol and one patient was taking a combination of amitriptyline, carbamazepine and clonazepam. None of the patients had been previously treated with antipsychotic medication.

A three-way mixed design ANOVA showed a significant Condition × Group interaction ($F = 6.54, \text{df} = 1, P = 0.017$), indicating that the effect of the self and external condition was different in patients compared to healthy participants. Post hoc exploration of this interaction revealed that this was due to patients having significantly less sensory attenuation than healthy controls in the self-condition ($F = 8.47, \text{df} = 1, P = 0.007$) but no significant difference from healthy controls in the external condition ($F = 0.145, \text{df} = 1, P = 0.706$). Raw data are represented in Fig. 2 and individual data for each participant and condition are shown in Fig. 3.

When we analysed patients alone, we found no significant differences in their performance when self and external conditions were compared ($F = 2.62, \text{df} = 1, P = 0.129$). In contrast, healthy controls significantly overestimated the force required in the self-condition compared to the external condition ($F = 26.64, \text{df} = 1, P < 0.001$).

We found no significant correlation between the amount of SA and duration of symptoms ($r = 0.007, P = 0.98$), HADS ($r = 0.29, P = 0.91$), Raven’s score ($r = 0.147, P = 0.62$), or PDI-21 total score ($r = -0.67, P = 0.82$).

### Discussion

Here, we demonstrate that patients with FMD have a loss of sensory attenuation in a force-matching task compared with healthy controls. Healthy control subjects consistently overestimated the force required in the self-condition, whereas patients did not, and were thus more accurate in their force estimation performance.

Despite the high prevalence of patients with functional symptoms in clinical practice and the associated levels of disability and healthcare costs, research and clinical development has been limited in comparison to disorders with a similar health impact. One probable cause of this lack of activity is the pervasive view amongst neurologists that such symptoms are commonly feigned (Kanaan et al., 2009). The other dominant explanation—that symptoms represent repressed psychological trauma—is not well implemented in the brain and a considerable explanatory gap exists regarding how psychological disturbance might be translated into the clinical symptoms experienced by patients with FMD.

One way in which to close this ‘explanatory gap’ is to study the mechanism of symptom production, rather than risk factors. Experimental evidence has demonstrated some key abnormalities in patients with functional symptoms. These include abnormalities that can be interpreted as abnormal self-directed attention (Roelofs et al., 2003; Parees et al., 2012a), abnormalities of...
probabilistic decision-making (Parees et al., 2012b), and abnormal activation of motor areas related to emotional stimuli (Voon et al., 2011). However, evidence that would confirm or refute the hypothesis that the sense of agency for movement is abnormal in these patients is more indirect.

To date, four studies have aimed to explore agency for movement in these patients. In a study using functional MRI, a relative reduction in activation of the right inferior parietal lobule was found in patients with functional tremor comparing activation patterns while they were tremoring and when they were voluntarily producing tremor (Voon et al., 2010). Although the right inferior parietal lobule is considered to be important in generating a sense of agency, these data only indirectly address the question of the reduced sense of agency in patients with FMD. Two studies have shown that patients with FMD judged the feeling of intention to move significantly closer to the action of moving compared to control participants and had a decreased action-effect binding when making voluntary movements compared with healthy volunteers (Edwards et al., 2011; Kranick et al., 2013). However, both studies rely on subjective self-report and are clearly susceptible to important biases. Finally, a recent study has shown that patients with functional paresis display distinct EEG markers compared to feigners (Blakemore et al., 2013).

Unlike previous studies, the paradigm used here provides a more direct demonstration that a key component of self-generated movement related to the sense of agency differs from healthy controls. Temporal and spatial offsetting between the movement and its sensory consequences causes a gradual decline in sensory attenuation (Blakemore et al., 1999). Thus the degree of sensory attenuation is proposed to index in some manner the feeling of ‘voluntariness’ of movement. Conversely, lack of sensory attenuation has been proposed to reflect a lack of sense of agency for self-generated movement (Shergill et al., 2005).

As highlighted in experiments with force-matching in patients with schizophrenia (Shergill et al., 2005), the behaviour of patients with FMD in this study is in fact more accurate than controls, making it hard to explain the results as a general consequence of having a chronic illness or as being feigned. It seems highly unlikely that patients would be aware of which condition to manipulate. Also, it would seem very difficult to deliberately override a physiological bias towards less accurate force-matching in the self-condition and to generate the observed pattern of force generation at each target force as presented in the figures with such small variability. We cannot, of course, completely dismiss this as a possibility. However, we believe these data reflect, instead, a neurobiological process that can lead to a loss of agency for normal movement and could therefore explain the paradox of movement that appears to be voluntarily generated, but is not experienced as such.

What is the likely mechanism of sensory attenuation, and why is it disrupted in FMD? We have argued elsewhere (Brown et al., 2013) that sensory attenuation is fundamentally an attentional phenomenon, as attending to a sensory channel is essentially the process of turning up the volume or gain of that channel (Feldman and Friston, 2010; Moran et al., 2013). Note that attention in this sense does not equate to the voluntary allocation of attention. It is

| Table 1 Demographic and clinical characteristics of the participants |
|------------------|------------------|------------------|
| Age (years)      | 38.1 (30–67)     | 34.5 (29–58)     | 0.12 |
|                  | Male 1           | 4                | 0.32 |
|                  | Female 13        | 10               |      |
| Handedness n     | Right 12         | 14               | 0.48 |
|                  | Left 2           | 0                |      |
| Type of FMD, n   | Fixed dystonia 10| NA               |      |
|                  | Paroxysmal       | 1                | NA   |
|                  | movement disorders | 1 | NA |
|                  | Functional tics 1| NA               |      |
|                  | Functional       | 1                | NA   |
|                  | palatal tremor   | 1                | NA   |
|                  | Functional       | 1                | NA   |
|                  | hemifacial spasm | 1                | NA   |
| HADS total score | Median (range) 13 | 4 (0–28)        | 0.002|
|                  | Raven’s score 10 | 11 (9–12)       | 0.94 |
|                  | PDI-21 score 12.5| 12.5 (0–30)     | 0.98 |

Figure 2 Results of the force-matching paradigm. Healthy controls (dashed blue line) significantly overestimated target forces in the self-condition compared with patients with functional motor symptoms (dashed red line). There were no differences in the external condition between healthy controls (solid blue line) and patients (solid red line). Because representing the standard error of the mean for each condition by using error bars was difficult to fit in the graph and acknowledging that our data are discrete, we use colour shadows for an easier demonstration.
well established that FMD patients’ motor symptoms require (specific) attention for their maintenance, but perhaps as has been reported in patients with somatization disorder (Robbins and Kirmayer, 1991), this group also have a generalized increase in attention to their bodies. We therefore propose that a generalized increase in body-focused attention in patients with FMD means that they do not attenuate the sensory consequences of their actions, and furthermore, that this makes them more likely to lose a sense of agency for their actions. This conclusion raises two crucial questions, which we seek to answer in future work. First, is the loss of sensory attenuation in FMD patients a ‘trait’ present before and after their motor symptoms? Second, what further factors are required to transform this trait into a state loss of agency for a particular action? It is of note that a proportion of patients develop spread of functional symptoms over time, and while the initial onset of symptoms is commonly associated with a trigger, spread of symptoms is often spontaneous. It is also of interest that one can generate new functional symptoms by actively directing attention towards the body during directed manoeuvres that are performed when examining a patient. Underlying this process may be at least three types of factors: a ‘predisposing’ loss of sensory attenuation, a ‘precipitating’ incident generating illness expectations, and a ‘perpetuating’ further increase in attention to bodily symptoms (Edwards et al., 2012).

Loss of sensory attenuation in patients with schizophrenia has been correlated with the presence of delusions of control (Shergill et al., 2005). Despite this, we do not suggest that the mechanism of FMD and schizophrenia are similar. Psychotic symptoms are not a feature of patients with FMD and questionnaires probing delusional beliefs in FMD do not reveal differences from healthy controls. It is likely that reductions in sensory attenuation in schizophrenia and in FMD have different primary causes, though the final common pathway is the same.

We acknowledge limitations to our study. First, the sample size is small and we cannot exclude that in a larger cohort data may have greater statistical efficiency. Secondly, most patients in our sample suffered from functional fixed dystonia, which is the second most common FMD (Edwards and Bhatia, 2012). Our failure to include patients with functional tremor, the most common FMD, is because tremor commonly involves upper limbs and this was an exclusion criterion for the study. However, we do not believe there to be systematic differences in pathophysiology between different functional motor symptoms, as different functional symptoms commonly co-occur in the same patient. Thirdly, we did not find the same amount of sensory attenuation in healthy controls compared with previous studies. One possible explanation is that the experimental set-up differed from that used in previous literature. Finally, although there was no correlation between HADS score and sensory attenuation in our study, we cannot rule out that this could emerge as a confounding factor in a larger study sample. Also, we cannot rule out a potential systematic bias in some unmeasured cognitive

Figure 3 Individual data for each of the applied forces (N). Each participant is represented on the horizontal axis and the ratio between the matched force and the target force for each condition is represented on the vertical axis.
or psychological factor that was not detected by the Raven’s score or HADS.

**Acknowledgements**

We would like to thank all participants who generously participated in this study and Paul Hammond for technical assistance. We also thank Lourdes Pareés, who also gave her kind help with some figures in this paper.

**Funding**

This work was performed at University College London Hospitals/University College London who received a proportion of funding from the Department of Health’s National Institute for Health Research Biomedical Research Centres funding scheme; Dr Isabel Parees was funded by an EFNS scientific scholarship 2012; Dr Atsuo Nuruki was funded by JSPS KAKENHI Grant Number, 23700608 and 25560298 Dr Marco Davare is funded by the BBSRC (UK); Prof Kailash P. Bhatia is funded by grants by the Dystonia Society UK and the Halley Stewart Trust. Dr Mark J. Edwards is funded by National Institute for Health Research (6187).

**Supplementary material**

Supplementary material is available at *Brain* online.

**References**


