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Attention and sensation in functional motor disorder

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ABSTRACT

Functional motor disorder (FMD), also called psychogenic motor disorder or conversion disorder, describes impairments of motor function where there is no evidence of organic disease. The diagnosis is usually confirmed by positive clinical signs, such as Hoover's sign, in which normal power returns when attention is diverted away from the affected limb. This suggests that selective attention is an important determinant of these functional symptoms. The present study is the first specifically to explore the shifting of spatial attention in relation to the side of FMD. We tested 14 patients with unilateral functional upper limb weakness on three tasks requiring detection of visual targets close to the affected or unaffected hand, or touches to the hand itself. Targets were preceded by central cues promoting voluntary shifts of attention, or peripheral cues promoting automatic shifts. We observed a reduced response to visual and/or tactile targets on the affected side in around half of the patients, by comparison with age-matched controls, indicating that some degree of detection cost often accompanies FMD. Additionally, although the patient group showed normal cueing effects on the visual tasks, they had a unilateral absence of cueing effect on the affected side in the tactile task. Consideration of the data in the context of recent theory suggests that the abnormality may be not in the shifting of attention itself, but rather in the *consequences* of attending to the affected side. Specifically, the expected cueing effects may be absent on the affected side, because attention to a functionally weak limb increases the perception of the symptom, including any reduced sensory response. This preliminary research suggests promising new lines of investigation into the role of attention, and particularly somatic attention, in FMD.

1. Introduction

Functional neurological symptom disorder, also known as conversion disorder, is a condition in which there are one or more symptoms of altered voluntary motor or sensory function, causing distress or impairment, and with clinical findings incompatible with organic disease (American Psychiatric Association, 2013). Functional motor disorder (FMD) is a common reason for referral to neurology clinics, and a common cause of disability in working age adults (Carson et al., 2000, 2011; Stone et al., 2010b). Studies in Scotland suggest that FMD is more common than multiple sclerosis, and that it causes similar levels of disability but with greater psychological comorbidity (Carson et al., 2011; Stone et al., 2010a).

Clinical tests for FMD often use distraction to reveal normal muscle power, or a change in the frequency or character of a tremor, when the person's attention is drawn elsewhere (Daum et al., 2013; Stone and Carson, 2015). For example, one of the most reliable positive features of functional leg weakness is Hoover's sign, in which a weakness of hip extension returns to normal power when attention is drawn away from the affected side, by asking the patient to focus on flexing their other hip against resistance (Ziv et al., 1998). A cohort study of patients with suspected stroke estimated that Hoover's sign is moderately sensitive (63%) and highly specific (100%) to FMD (McWhirter et al., 2011). These clinical findings are mirrored by experimental work suggesting that automatic aspects of motor control may be preserved in FMD, with impairment evident in more explicit movement tasks in which attention is focused on the action (Pareés et al., 2013).

Abnormal attentional focus has long been thought core to functional neurological disorders, though different ideas about the nature of the abnormality have emerged over time. Janet (1907; as reviewed by Edwards, 2016) believed that such symptoms arose from a 'retraction of the field of consciousness', implying a withdrawal of attention from the

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affected body part. This basic idea, that an insufficiency of attention is responsible for a loss of function, was echoed by later theorists (Ludwig, 1972; Whitlock, 1967), and drew some support from findings of reduced somatosensory evoked potentials in hemianaesthesia (Halliday, 1968; Hernandez-Peon et al., 1963). However, the idea is hard to reconcile with the clinical observation that functional motor symptoms are improved by the diversion of attention elsewhere.

A more recent, and somewhat opposite, proposal is that attention to the affected site or symptom may be a necessary, though not sufficient, condition for FMD and other functional neurological disorders (Edwards, 2016; Edwards et al., 2012). Specifically, focused attention in combination with a strong prior expectation of a symptom, whether positive (e.g. tremor) or negative (e.g. sensory or motor loss), might over-ride normal bottom up signals, causing the expected symptom to become a reality. If the symptom thereby manifests whenever attention is turned to it, the patient might have the subjective impression that it is continuous. Consistent with this, Pareés et al. (2012) found that patients with functional tremor, but not those with organic tremor, grossly over-estimate the proportion of time for which their tremor is present. Coding of gaze position in observational video-recordings suggests that patients with functional tremor spend far more time looking at their tremor than do those with organic tremor (~ 80% vs ~ 25%) (van Poppelen et al., 2011); and functional imaging suggests that FMD is associated with increased activation of brain areas implicated in self-monitoring (Bell et al., 2011; de Lange et al., 2007). It is therefore possible that FMD may be associated with *too much*, rather than too little attention to the affected site.

Surprisingly, given the central role of attention in theories of FMD, there has been very little direct study of attention in such patients. Roelofs et al. (2003) adapted the well-established cueing techniques of Posner (1980) to investigate selective spatial attention in eight patients with FMD of the upper or lower limbs. Visual targets were preceded, at a variable delay, by a predictive central arrow (endogenous cue) or the non-predictive appearance of a box at a potential target position (exogenous cue). FMD patients were overall slower to respond than matched controls, and showed muted effects of endogenous cues at the shortest cue-target delay (150 ms). Patients also failed to show clear *inhibition of return* following exogenous cues at longer cue-target delays (550 ms). Inhibition of return is the transition of cue validity effects, from facilitation at short delays (< ~ 350 ms) to inhibition at longer delays (> ~ 350 ms). That is, a non-informative exogenous cue initially enhances detection at the cued location, but as the person then re-orients away from the cue, detection becomes relatively faster at non-cued locations. This secondary reorienting after an exogenous cue is arguably a *voluntary* process, so an absence of inhibition of return in FMD could be taken as further evidence for impaired voluntary control of attention, with automatic attention relatively preserved (Roelofs et al., 2003).

The focus of the above study was on the distinction between voluntary and automatic shifts of attention. Even so, it is surprising that no analyses were made of the effect of target side in relation to the side of symptoms. These well-established cueing tasks potentially provide a rich framework for an assessment of asymmetries of attention, toward or away from the affected side, as proposed by several theories of FMD. There are many possible permutations of these tasks, combining different cueing and response methods, and sensory modalities, and we could sample only a subset in the present study. We decided to focus on voluntary and automatic attention to the visual space around an affected or unaffected hand, and on voluntary attention to *tactile* stimulation of the hand. Given that different theories have suggested either under- or over-attention to the affected site, we did not have any strong *a priori* commitment to the direction of any asymmetry. Rather, we proposed an initial, exploratory assessment of lateralized visual and tactile detection and attention in patients with unilateral upper limb FMD.

2. Method

2.1. Participants

Patients were recruited from neurology or neuropsychiatry outpatient clinics, who were between the ages of 18 and 75, and had a diagnosis of functional motor disorder with unilateral upper limb weakness, made by a Neurologist (JS) or Neuropsychiatrist (AC) following DSM-5 criteria (American Psychiatric Association, 2013). Patients were excluded who had a diagnosis of dementia, learning disability or a comorbid psychiatric disorder, for whom there was any clinical suspicion of factitious disorder, or who did not speak English.

Fourteen FMD patients (8 female, 6 male) took part. Twelve were right-handed by self-report, six of whom had left-sided weakness and six of whom had right-sided weakness. Two were left-handed by self-report, one of whom had left-sided and one of whom had right-sided weakness. The mean age of the FMD group was 39 years (SD 9.9, range 23–53), and the median symptom duration was 3 years and 3 months (range 5 months – 21 years). All patients had positive evidence of functional limb weakness (Daum et al., 2013), and clinical investigations that had shown no signs of organic pathology. Upper limb weakness was self-reported as present continuously. Grip strength and tapping frequency were reduced in the affected compared with the unaffected side in all patients tested. On sensory examination (see Procedure for details), two patients additionally reported reduced sensation in the weak limb (patients FMD01 and FMD02). Patient characteristics, including prescribed medications, are reported in Table 1.

Fourteen healthy control (HC) participants (8 female, 6 male) were recruited from patients' families and from the University of Edinburgh Psychology volunteer panel. Eight were right-handed, five were left-handed and one was 'ambidextrous' by self-report (this participant was considered as left-handed in controlling for hand-dominance at ANOVA). The mean age of the HC group was 40 years (SD 14.0, range: 18–64).

2.2. Procedure

This study was conducted with approval from the relevant NHS Research Ethics Committee and the University of Edinburgh Psychology Research Ethics Committee, and written informed consent was obtained for each participant.

Each participant attended the University of Edinburgh Visuomotor Laboratory once. For patients, not controls, the attention tasks were preceded by assessment of maximum tapping rate (best of three ten second trials), hand dynamometer grip strength (best of three attempts), and tactile sensation. Sensation was tested using a standard neurological examination, with light touch and pinprick (neurotic) stimulation of upper limbs, including hands (stimulation of each dermatome). All participants then performed three cued target detection tasks to assess: visual exogenous attention (vX task); visual endogenous attention (vN task); and tactile endogenous attention (tN task). The vX and vN tasks were performed first, with the order alternated between participants within each group; the tN task was performed last.

Each task was performed on a widescreen monitor (active display 530 × 298 mm; resolution 1920 × 1080; refresh rate 60 Hz), tilted backwards by ~ 70° so that it faced up from the table toward the participant, at a viewing distance of ~ 60 cm. A schematic diagram of example stimulus displays is shown in Fig. 1. The participant sat with their hands resting palm-down on the screen surface, with each index fingertip abutting a white outline box (35 mm square), which acted as a placeholder for visual targets. These boxes were centred 134 mm to left and right of a white outline diamond (17 mm side length), which had a 1 mm white fixation point at its centre, coinciding with the horizontal centre of the screen. In the tactile task, the participant had a mechanical 'tapper' attached by medical tape to the medial side of the distal