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Citation for published version:

Stone, J, Mutch, J, Giannokous, D, Hoeritzauer, I & Carson, A 2017, 'Hurst revisited: Are symptoms and signs of functional motor and sensory disorders “dependent on idea”?', *Journal of the Neurological Sciences*. <https://doi.org/10.1016/j.jns.2017.08.3248>

Digital Object Identifier (DOI):

[10.1016/j.jns.2017.08.3248](https://doi.org/10.1016/j.jns.2017.08.3248)

Link:

[Link to publication record in Edinburgh Research Explorer](#)

Document Version:

Peer reviewed version

Published In:

Journal of the Neurological Sciences

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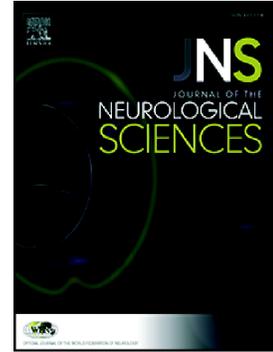
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Accepted Manuscript

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PII: S0022-510X(17)33747-4
DOI: doi: [10.1016/j.jns.2017.08.3248](https://doi.org/10.1016/j.jns.2017.08.3248)
Reference: JNS 15526

To appear in: *Journal of the Neurological Sciences*

Received date: 8 March 2017
Revised date: 22 August 2017
Accepted date: 23 August 2017

Please cite this article as: Jon Stone, Jennifer Mutch, Denis Giannokous, Ingrid Hoeritzauer, Alan Carson, Hurst revisited: Are symptoms and signs of functional motor and sensory disorders “dependent on idea”?. The address for the corresponding author was captured as affiliation for all authors. Please check if appropriate. *Jns*(2017), doi: [10.1016/j.jns.2017.08.3248](https://doi.org/10.1016/j.jns.2017.08.3248)

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Hurst revisited: are symptoms and signs of functional motor and sensory disorders "dependent on idea"?

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Keywords:

Functional neurological disorders; conversion disorder; psychogenic; Hoover's sign; Tremor entrainment; Psychogenic Movement Disorder

Abstract**INTRODUCTION:**

Symptoms and signs of functional (psychogenic) motor and sensory disorder are often said to be dependent on the patients' idea of what symptoms should be, rather than anatomy and physiology. This hypothesis has however rarely been tested.

MATERIALS AND METHODS:

Inspired by a brief experiment carried out in 1919 by neurologist Arthur Hurst we aimed to assess the views of healthy non-medical adults towards paralysis and numbness and their response to tests for functional disorders when asked to pretend to have motor and sensory symptoms.

RESULTS:

When subjects were asked to pretend they had a paralysed arm 80% thought there would be sensory loss. Of these 60% thought it would have a circumferential (functional) distribution at the wrist, elbow or shoulder. Hoover's sign of functional weakness was only positive in 75% of patients pretending to have leg paralysis with 23% maintaining weakness of hip extension in the feigned weak leg, a rare finding in neurological practice. 20% of subjects managed to continue having their feigned tremor during the entrainment test. 52% of subjects thought there was asymmetry of a tuning fork across their forehead even when no prior instruction had been given.

CONCLUSIONS:

The study confirmed Hurst's finding that non-medical people generally expect sensory loss to go along with paralysis, especially if the examiner suggests it. When present, it usually conforms to functional patterns of sensory loss. Clinical tests for functional and motor disorders appear to behave somewhat differently in patients asked to pretend to have symptoms suggesting that larger more detailed studies would be worthwhile.

Introduction

Symptoms and signs in functional neurological symptom disorders in neurology, also known as psychogenic and conversion disorders, have long been thought, in part, to be based on people's 'ideas' of how diseases might be expected to present rather than the, sometimes counterintuitive, reality of pathophysiology of the nervous system¹. This hypothesis was a common theme in late 19th writing onwards. For example Freud observed that in "hysterical paralysis" the arm is 'paralysed...according to the common idea of the arm or what we commonly designate by the word 'arm'"². This, for example, is why it is thought that patients who have functional paralysis may describe numbness in their whole leg up to the groin even though this is not a pattern seen in neurological disease (i.e. the whole leg is weak so the whole leg must be numb as well). Despite this, studies of these 'ideas' in healthy controls and the performance of diagnostic tests for functional disorders in healthy controls and patients feigning symptoms in factitious disorder and malingering are exceptionally rare.

Arthur Hurst, a physician later at Guys Hospital, had seen large number of patients with Shell Shock at Netley and in Devon during World War 1. He was responsible for the films of "shell shock" that often appear in documentaries on the subject³. In 1920 he delivered his Croonian lectures to the Royal College of Physicians of London on functional disorders of the special senses⁴. Within it was a description of an experiment of 'hysterical anaesthesia following hetero-suggestion' in which he examined healthy individuals who had been asked to pretend that they were involved in a compensation case and had limb paralysis. When asked whether they also had altered sensation on the affected side, 22 out of 27 agreed they did, even though this did not form part of the instruction. Patterns of numbness often corresponded to those found in patients with 'hysterical' paralysis (Figure 1). Hurst concluded that many of the signs seen in 'hysteria' were a consequence of suggestion by the examiner combined with a popular idea of how paralysis and sensory loss occurs.

Inspired by this experiment, we carried out a study on healthy individuals to investigate not only their 'ideas' of limb functioning but also their response to some of the tests that are used to make diagnoses of functional disorders in neurology.

Materials and Methods

44 healthy adult volunteers (22 female, 22 male), were recruited via acquaintance of one of the authors (DG). The group was highly educated, with 36 studying for or having completed a university degree, and aged between 17 and 77 years (mean 34). None were medical or connected to medical families (e.g. spouse of health professional). None had a research interest in, or an in-depth knowledge of, functional neurological disorders. None had a history of paralysis or numbness apart from one subject who reported mild transient arm weakness following a bicycle accident that did not appear to have any impact on findings or ability to participate.

All 44 volunteers were asked a series of standardised scripted questions and examined using a standardised protocol to investigate responses both symptomatically and on examination in both a normal state and also when pretending to have paralysis, tremor or sensory loss (Table 1). Each patient was assessed individually by the same examiner (DG) who had been trained in the necessary examination techniques to the level of a "junior doctor two to three years after qualification" by a consultant neurologist (JS) based on published descriptions^{5,6}. We deliberately chose a relatively inexperienced examiner to make the study more generalisable to usual practice. NHS ethics was not required. All subjects were friends/ associates of the main investigator and consented verbally to taking part.

Results

These are shown in Table 1.

Table 1. Tests and responses of 44 healthy individuals

Test	Description of test in functional disorders	Instruction for this study	Response
Hoover's sign	Weakness of hip extension that returns to normal with contralateral hip flexion against resistance.	"Pretend to have a weak leg"	33/44 Hoovers positive 1 unable to follow examination instructions 10/44 able to maintain weakness of hip extension
Tremor Entrainment test	Tremor that either stops or entrains to an externally cued varying repetitive finger-thumb opposition rhythm. Test is also positive when subject can't copy movements.	Copy the movements with either hand	1/44 patient unable to copy movements (i.e. a positive test)
		'Pretend to have a tremor of your dominant arm. Copy the movement I am making (finger-thumb opposition) using your unaffected hand.'	16/44 unable to copy movements* 20/44 tremor entrainment* 4/44 tremor stopped* 9/44 tremor unaffected by testing*
Tuning Fork test on forehead	Vibrating tuning fork placed on each side of forehead. Asymmetry thought by some to be evidence of a functional disorder since the frontal bone is a single bone and should vibrate as one	No instruction 'any difference'?	23/44 reported asymmetry
		'...imagine that the left side of your body is numb. Would you feel any difference in vibrations on one side compared to the other?'	9/44 reported vibration more strongly on left 26/44 reported vibration more strongly on right 9/44 said symmetrical
Gait	'Imagine that your (dominant) leg is weak. Show me what it would be like to walk.'		18/44 circumducting gait, 15/44 dragging gait (with forefoot touching ground), 1/44 crouched with flexed knees, 1/44 'walking on ice' pattern, 8/44 other, 1/44 normal
Sensory symptoms in healthy people instructed to pretend to have a paralysed dominant arm	"You were in an accident and you have paralysis of your (dominant) arm". 'Would you lose feeling or sensation?' 'Please shade in the area that would be numb on this diagram.'		35/44 said the arm would also have reduced sensation 21/35 reported circumferential sleeve or glove sensory loss (17 shoulder/ 3 elbow/1 glove).

* Patients could have more than one response - e.g. unable to copy movements and the tremor stopped; The word 'right' or 'left' was substituted for dominant when appropriate

Hoovers sign was positive in 75% of subjects pretending to have paralysis with a further 23% managing to maintain hip extension weakness despite normal contralateral hip flexion. One subject failed the tremor entrainment test (unable to copy a rhythmical movement) even before being asked to pretend to have a tremor. Nine subjects managed to have a negative tremor entrainment test when pretending to have a tremor.

When a tuning fork was placed on either side of the forehead with the question 'any difference?' 52% of subjects described asymmetry. When asked to pretend that their left side was numb this rose to 80%.

When subjects were instructed to pretend that they had a paralysed arm, 80% thought there would be sensory loss as well when asked a question about that. Of these 60% thought it would have a circumferential distribution either at the wrist, elbow or shoulder.

When subjects were asked to have a paralysed leg and asked to walk 34% had a 'dragging gait' typical of functional leg weakness although 41% did have a circumducting gait as seen in 'organic' hemiparesis.

Discussion

Clinical tests for functional disorders are based on demonstrating internal inconsistency and incongruity with patterns of organic disease. In practice, this is generally believed to depend, in part, on an expectation that they have more in common with what people think might happen if you have a symptom, than what actually does when you have a disease. Those tracing the history of this often arrive upon Ross Reynolds' 1869 paper, '*Paralysis and other disorders of motion and sensation dependent on idea*'¹. Confirmation of illness spreading purely by idea can be seen perhaps most overtly in mass outbreaks of hysteria and culture bound disorders such as Latah⁷.

Recent work provides a neurobiological and computational framework for understanding this "inferential leap" between ideas held, implicit predictions and the symptoms experienced^{10,11} and recent imaging studies show neural system connections between imaging and executing an action or emotion^{8,9}.

We assume that patients undergoing testing of vibration sense across the forehead should feel the stimulus symmetrically because it is a single bone yet a remarkable 52% of subjects in this study thought it was asymmetric even without any instruction. In fact this accords with some other studies which have shown that this particular sign is not that reliable^{12,13}. In recent years an increasing number of studies have looked at the sensitivity, specificity, validity and interobserver agreement of these tests and found that some do appear to be reasonable^{13,14}. None, of course, distinguish functional disorders from feigning.

Most subjects in our study thought that their paralysed arm would also be numb when asked if was the case (80%). This is very close to the 81% of subjects that Hurst found in his original experiment. Hurst believed that at least some patients with "hysterical" anaesthesia developed it after being asked a leading question on the topic although he believed that others did complain of it symptomatically prior to any medical contact. His view however was that all such patients had a prior organic condition that had led to a phenomenon of "autosuggestion". Suggestion is hard to avoid when carrying out sensory testing. Our question 'any difference?' was enough to induce it, and some measure of implicit and explicit sensory deficit in volunteers pretending to be paralysed would be instructive. In over half (60%) of cases in this small sample, people's ideas of numbness associated with weakness were compatible with the most well-known description of functional anaesthesia (sleeve/glove). This is in keeping with long held notions that there is a 'popular idea' about what numbness is. However, this argument is tempered by a recognition that the remaining 40% of participants described heterogenous sensory change associated with paralysis. Sensory testing in functional disorders is especially prone to error and the positive signs that have been described are sensitive but lack specificity. Hurst found that all seven subjects in whom he suggested sensory loss associated with distal paralysis reported glove and stocking loss. In Hurst's study though subjects were asked to act out where the sensory loss would be rather than imagine and draw (as in our study).

We have extended Hurst's experiments by testing positive signs of functional motor disorder in subjects pretending to be paralysed or have a tremor. Our clinical experience of Hoover's sign is that patients with functional leg weakness only very rarely maintain hip extension weakness when contralateral hip flexion induces normal hip extension automatically. This only happened once in 60 patients with a Hoover's sign in a case control study carried out by one of the authors¹⁵. The fact that 23% of

patients pretending to be paralysed appeared to be able to do this suggests this could be investigated further as a sign of factitious disorder or malingering. Our own experiments show that it is possible to maintain hip extension weakness with only a little practice. In the tremor entrainment test, we assume that everyone can copy a simple movement made by the examiner, so their failure to do so should indicate the presence of a functional tremor. Yet one subject failed to do this, emphasising the need for further studies of the sensitivity and specificity of clinical signs used in making a positive diagnosis of functional neurological symptom disorders.

The data from this small pilot study are limited to the single observer who was deliberately junior in training. Someone more experienced may obtain different results. There was also a relatively high educational level among the subjects who were also known to the examiner which could have influenced the results, although none had a health professional background or specific knowledge of functional neurological symptom disorders.

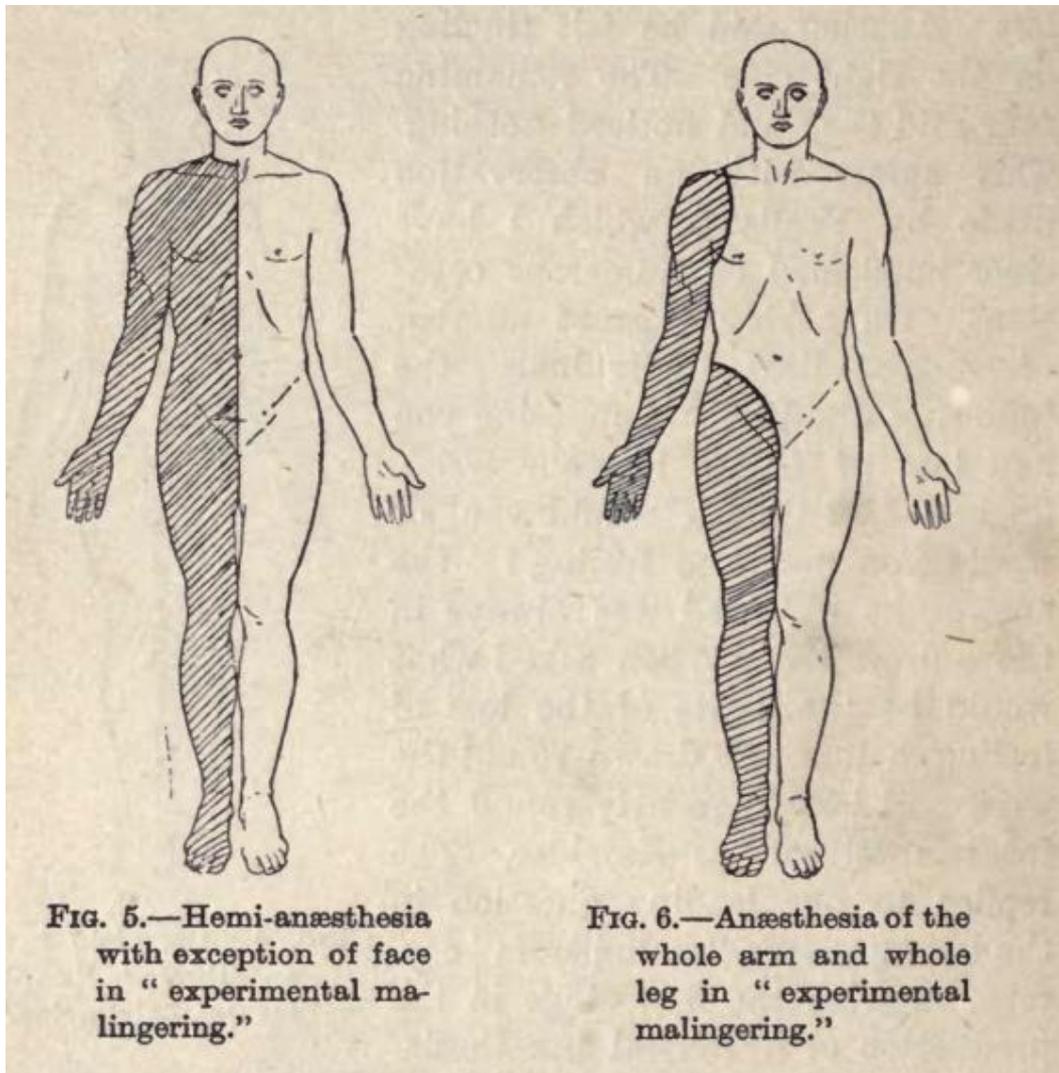
We think this kind of study does have value, however, in attempting to calibrate these tests in patients with functional disorders, disease and perhaps in patients who have factitious or malingered symptoms. Larger studies with videoed interviews, multiple raters of different seniority and blinding of the examiner to the patient would be instructive about how reliable our tests for functional disorders are. Finding out where people get their ideas about neurological disease and to what extent the symptoms can be experienced by the participant as voluntary or involuntary may help explore further whether there are common 'ideas' of illness which patients with genuine functional disorders may be susceptible to.

Acknowledgements: JS is supported by an NHS Scotland NRS Career Fellowship. IH is supported by an ABN/Patrick Berthoud Clinical Research Fellowship.

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Figure 1. Depiction of areas of sensory loss in medical student asked to pretend to be paralysed. from Arthur Hurst's 1920 book - '*The psychology of the special senses and their functional disorders*'⁴



Highlights

- Ideas about neurological symptoms often don't correlate with what would be expected anatomically.
- Non-medical people generally expect sensory loss to go along with paralysis.
- Such sensory loss is similar to that seen in functional sensory disturbance.
- Tests for functional disorders may behave differently in patients pretending to have symptoms.

ACCEPTED MANUSCRIPT