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ABSTRACT

Objectives: To determine the disability, distress and employment status of new neurology outpatients with physical symptoms unexplained by organic disease and to compare them with patients with symptoms explained by organic disease.

Methods: As part of a cohort study (the Scottish Neurological Symptoms Study) neurologists rated the extent to which each new patient's symptoms were explained by organic disease. Patients whose symptoms were rated as "*not at all*", or only "*somewhat*" explained by disease were considered cases and those whose symptoms were "*largely*" or "*completely*" explained by disease controls. All patients completed self-ratings of disability, health status (SF-12) and emotional distress (HADS) and also reported their employment and state financial benefit status.

Results: 3,781 patients were recruited; 1,144 (30%) cases and 2,637 (70%) controls. Cases had worse physical health status (SF12 score 42 vs 44; difference in means 1.7 (95% CI -2.5 to -0.9)) and worse mental health status (SF12 score 43 vs 47; difference in means -3.5 (95% CI -4.3 to -2.7). Unemployment was similar in cases and controls (50% v 50%) but cases were more likely not to be working for health reasons (54% v 37% of the 50% not working) ; OR 2.0 (95% CI 1.6 to 2.4) and also more likely to be receiving disability related state financial benefits (27% v 22%; (OR 1.3, 95% CI 1.1 to 1.6)).

Conclusions: New neurology patients with symptoms unexplained by organic disease have more disability, distress and disability related state financial benefits than patients with symptoms explained by disease.

Introduction

We know that one third of neurology outpatients have symptoms, such as pain, weakness and sensory disturbance, that are not explained by recognised 'organic' disease [1,2,3,4]. But are these patients really ill? In other words, do they just have symptoms, or are such symptoms actually associated with disability and distress, and is this reflected in their employment status and receipt of disability-related state financial benefits?

A previous study we conducted of symptoms unexplained by organic disease suggested that neurology patients with this problem were at least as disabled and distressed as patients with neurological disease but it was of only a small sample (90 patients with unexplained symptoms) recruited from only one service [1]. In this study we aimed to determine the disability, distress, and employment status of more than 1,000 consecutive cases with symptoms unexplained by disease, who were prospectively identified as part of a large multicentre study of new neurology outpatients, and to compare these with those of controls from the same cohort whose symptoms were explained by neurological disease.

Methods

The Scottish Neurological Symptoms Study was a prospective, multi-centre, cohort study of NHS neurology outpatients in Scotland. Ethical approval for the study was granted by a Multi-centre Research Ethics Committee. This paper reports a case control analysis of study baseline data.

Participating clinics

Thirty six out of thirty eight consultant neurologists, working across all four Scottish NHS neurology centres, participated. Patients were recruited from their general neurology clinics (including their supervised trainee clinics) in the main Scottish neurological centres - Aberdeen, Dundee, Edinburgh, and Glasgow and associated peripheral clinics in Airdrie, East Kilbride, Falkirk, Inverness, Perth, Stirling, Vale of Leven, and Wishaw - between December 2002 and February 2004. All the clinics sampled took mainly general practice referrals with patients allocated by medical records staff according to availability of appointment. Tertiary clinics, where patients required a verified diagnosis to attend (such as multiple sclerosis clinics), and emergency clinics were excluded.

Patients

All newly referred patients at the participating clinics were potentially eligible for inclusion. The exclusion criteria were: age less than 16, cognitive or physical impairment of a degree that precluded informed consent, inability to read English, or if the neurologist identified the patient as unsuitable for the study (e.g. too distressed, terminally ill). New patients included patients with existing neurological diagnoses who had been re-referred from primary care. Patients were sent information about the study prior to their appointment with the neurologist. After the consultation they were invited by their neurologist to speak to a research assistant. Consent was obtained from patients willing to participate

Immediately following the initial consultation neurologists were asked to complete a four point Likert scale:

'To what extent do you think this patient's clinical symptoms are explained by organic disease? - "*not at all*", "*somewhat*", "*largely*" or "*completely*" [1]. Operational criteria were provided to guide ratings (web figure 1).

Measures

Following the initial neurological consultation, all participating patients completed a self-report questionnaire which included the following measures:

Demographics including age and sex.

Reported disability (Health status): using the Medical Outcomes Study Short Form 12-item Scale (SF-12) [7]) which examines health status across eight domains and in two composite scores of physical and mental health status. We calculated the 'raw' score in each of the domains as a percentage and also the two composite scores as recommended by the authors.

Emotional distress using the Hospital Anxiety and Depression scale (HAD)[8] [9]

Employment Status and receipt of state financial benefits by patient self-report. We asked specifically whether patients were (a) in paid employment (and, if not, whether this was because of health problems); (b) in receipt of income support, unemployment benefits, disability living allowance or incapacity benefit.

Analysis

First, we calculated the number of patients in each of the four 'organicity' groups. We then amalgamated those patients whose symptoms were rated as 'not at all explained' and 'somewhat' explained by organic disease and classified them as having symptoms 'unexplained' by disease (cases) and patients who were rated as having symptoms 'largely' and 'completely' explained by disease as having

'explained' symptoms (controls) . We then compared the cases and controls using difference in means and odds ratios.

Results

Patients were recruited between 16/12/2002 and 26/02/2004. During this period a total of 4,299 new patients attended the designated clinics. Recruitment to the study is summarised in figure 1. The final sample was of 3,781 patients (88% of all attendees and 91% of all eligible outpatients) of whom 1,144 patients (30% of the total) were rated by neurologists as having symptoms 'not at all' (n=446; 12%) or only 'somewhat explained' (n=698; 18%) by disease; they were the 'unexplained' cases. The medical diagnoses given to the cases have been described in detail elsewhere [9][10]. The remaining 2,637 patients had symptoms which were 'largely' (n=940; 25%) or 'completely explained' (n=1697; 45%) by organic disease; they were the 'explained' controls. (We have also analysed the data across all four groups and include this as supplementary web table 1).

[Figure 1 about here]

Cases were, on average, four years younger and more likely to be female than controls (Table 1). They had lower scores in all SF-12 individual domains, including all the disability domains including physical and social function and physical and emotional role function. Cases also had a higher level of emotional distress on the SF-12 mental health scale and on the HADS.

[Table 1 about here]

A similarly large proportion of both cases and controls reported not being in paid employment (50% vs 50%). However of those unemployed cases were more likely to report that they were not working because of ill health (26% vs 18%). This difference was reflected in the finding that cases were also more likely to be in receipt of disability related state financial benefits (27% v 22%: OR 1.3 , 95% confidence interval 1.1 to 1.5).

Discussion

We found substantial self-reported disability, distress, and unemployment in patients with symptoms 'unexplained' by 'organic' disease. All these variables were more severe in cases than controls. Furthermore the cases were more likely to report unemployment for reasons of ill health and to be in receipt of disability related state financial benefits.

These findings clearly indicate that patients with symptoms 'unexplained' by organic disease are not merely the 'worried well'. Rather they have substantive self-reported disability, distress and associated unemployment. Whilst their degree of disability and distress was actually statistically greater than that of controls with neurological disease, this difference is around the minimum considered clinically significant [11], [12]. A more conservative interpretation of our data would therefore be that they are similar to patients with neurological disease on these characteristics.

Although cases had greater rates of health related unemployment and disability related state financial benefits than controls the majority of patients with symptoms

unexplained by organic disease were actually in work. This observation does not support anecdotal suggestions that all such patients are motivated by work avoidance and receipt of benefits.

This study has the strength of being of a large and representative sample of neurology patients. However it also has limitations: Although almost all Scottish neurologists participated in the study not all their clinics were sampled and specialised clinics such as neurovascular and memory clinics were not included; consequently patients with these disorders may be under-represented. Similarly we cannot be certain that Scottish neurological practice is similar to neurological practice round the world, although the prevalence rates of the common neurological disorders in patients attending the clinics sampled would suggest it was.

Although disability related state financial benefits are subject to independent assessment and in a small number of cases anti-fraud investigations, much of the information used to assess such claims is still based upon self-report and cannot therefore be regarded as completely objective. Furthermore, absolute rates of health related unemployment and the uptake of financial benefits may be influenced by economic conditions as well as by illness.

Among other limitations was our approach of ‘lumping’ patients with symptoms unexplained by organic disease into one group. Whilst we believe that there are sound theoretical reasons for doing this [13] others may prefer to ‘split’ this group into many separate conditions. Furthermore we did not we seek to determine the factors causing the observed disability; these are likely to include a variety of factors

including but not exclusively psychiatric illness. Finally our ‘unexplained’ cases were slightly younger and more likely to be female, this may influence the reported rates of disability and distress.

Previous studies of specific diagnostic groups including psychogenic seizures [14,15, 16], psychogenic sensory loss [17] and psychogenic movement disorders [18,19] have also found that these patients have a degree of physical disability similar to that of patients with neurological disease. A retrospective Dutch study of a consultation liaison psychiatry service [20] included 544 patients who had been diagnosed with a somatoform disorder, of whom 215 had conversion disorder, and found a high rate of unemployment amongst the older patients with somatoform symptoms but not amongst younger ones. Crimlisk *et al* in a study of 64 patients with motor conversion disorder at a tertiary centre found that 77% of their cohort had held jobs prior to symptom onset but only 11% were still working at six years follow up [21]. A recent cohort study of psychogenic seizures in the West of Scotland described very high rates of unemployment (90%) and uptake of benefits (62%) [22]. We found a lower rate of unemployment in our cohort. We suspect this was due to two factors: First we distinguished between those whose unemployment was due to their health complaint and those who were unemployed for other reasons and second we studied a sample more representative of general neurology clinic attenders.

In summary, our date indicates that neurology patients with symptoms unexplained by disease are not merely the ‘worried well’ but are demonstrably ill by the usually applied criteria of disability and distress. Our data make a strong case the

development and implementation of targeted interventions for this group of patients. It is however imperative that such, potentially complex, interventions are properly tested in well designed randomised controlled trials before they are implemented. Researchers should consider including economic measures into trials as successful treatment could not only improve the quality of patients lives, but also help them to return to work.

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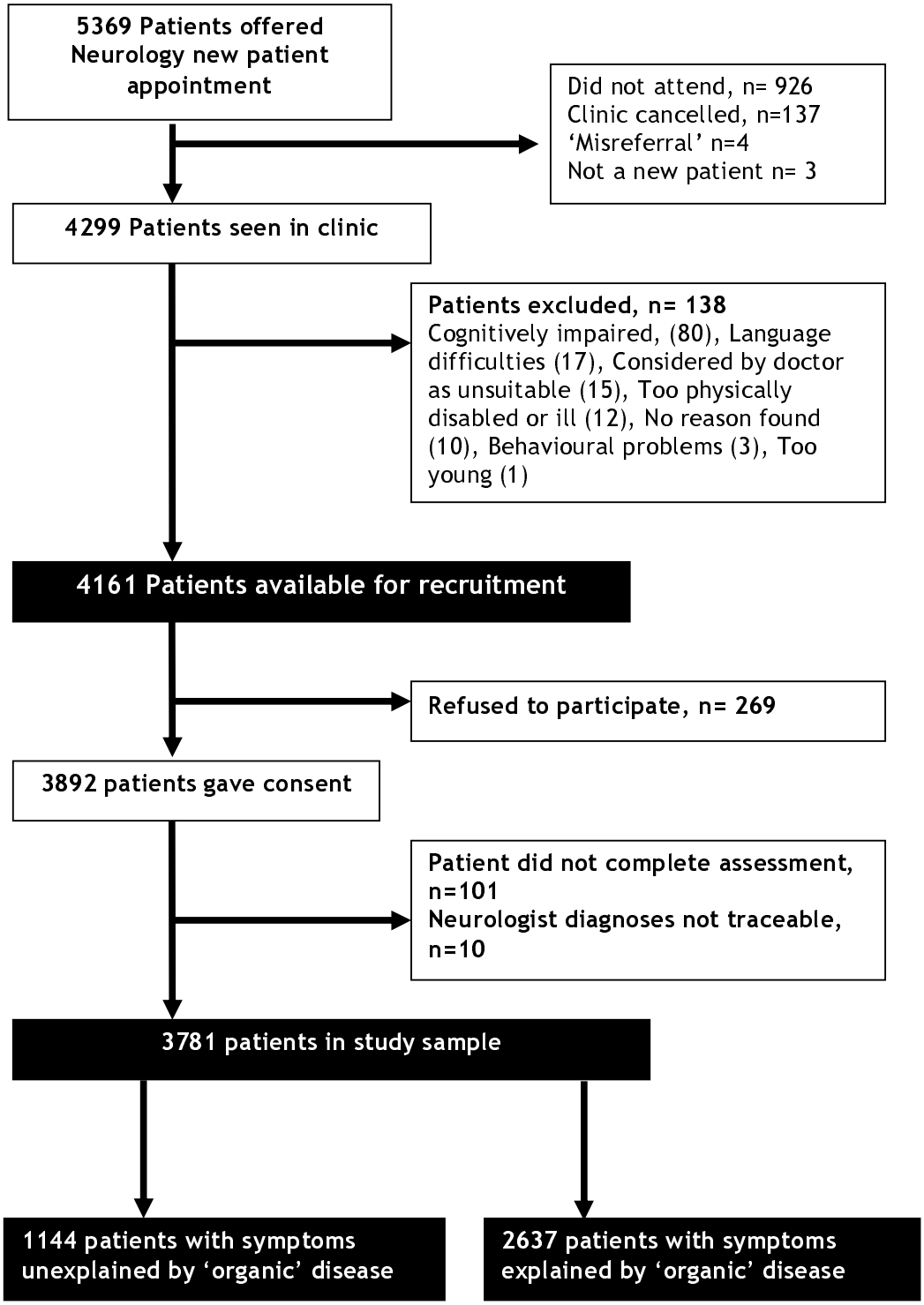
Figure 1- Flow Chart of patient recruitment into study.



Heading- Supplementary web figure 1. **Operationalised criteria for 'organicity' rating**

WHAT DO WE MEAN BY 'ORGANIC DISEASE' ?

The following table is meant as a guide for *this study* and we are aware that any divisions like this are imperfect.

Many patients have a mixture of symptoms, syndromes or disease and the final coding is your decision based on these guidelines

NOT 'Organic Disease' **for the purposes of this study**	'Organic Disease' **for the purposes of this study**
1 Tension Headache	**1** Migraine
2 Aetiologically controversial symptom 'syndromes' e.g., - Chronic fatigue syndrome - Fibromyalgia - Irritable Bowel Syndrome	**2** Any neurological disorder with a known pathological basis
3 Physiologically explained processes which are thought to be linked to emotional symptoms e.g., - Hyperventilation	**3** Neurological disorders with defined and characteristic features but without a clear pathological basis e.g., - Gilles de la Tourette syndrome - Idiopathic focal dystonia
4 Emotional disorders e.g., - Depression - Anxiety - Panic disorder	**4** Physiological explained processes NOT linked to emotional symptoms e.g., - Micturition syncope
	5 Psychotic disorder

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if there are further queries

Table 1. Disability, distress and employment status of cases with symptoms 'unexplained by organic disease' and controls with symptoms 'explained by organic disease'.

	cases	controls	Odds ratio Difference in means (95% CI)
Age, mean (SD)	43.6 (14.4)	47.5 (17.0)	-3.85 (-4.98; -2.72)
Female, % (n/N)	65.3 (747/1144)	53.6 (1414/2637)	1.63 (1.41; 1.88)
SF-12, mean (SD)[a]			
general health	42.6 (26.0)	48.7 (24.7)	-6.15 (-7.90; -4.41)
physical functioning	63.9 (38.4)	66.9 (36.8)	-2.99 (-5.58; -0.40)
role physical	50.0 (46.7)	56.1 (45.0)	-6.02 (-9.18; -2.86)
bodily pain	58.0 (35.8)	67.2 (34.5)	-9.23 (-11.66; -6.81)
social functioning	62.3 (34.7)	69.2 (33.6)	-6.91 (-9.27; -4.56)
vitality	39.5 (27.7)	44.4 (28.0)	-4.90 (-6.85; -2.96)
mental health	54.5 (25.5)	61.4 (24.0)	-6.94 (-8.63; -5.24)
role emotional	64.7 (43.1)	75.2 (38.9)	-10.48 (-13.28; -7.69)
Composite score - Physical Health Status	42.2 (12.5)	43.9 (11.7)	-1.72 (-2.55; -0.89)
Composite score - Mental Health Status	43.4 (12.1)	46.9 (11.3)	-3.53 (-4.33; -2.73)
HADS, mean (SD)[a]			
anxiety subscale	7.9 (5.0)	6.4 (4.5)	1.51 (1.19; 1.84)
depression subscale	5.9 (4.9)	4.7 (4.2)	1.21 (0.91; 1.52)
total score	13.8 (8.9)	11.1 (7.8)	2.73 (2.16; 3.30)
Not in paid employment, % (n/N)	49.5 (563/1137)	49.9 (1313/2629)	0.98 (0.86; 1.13)
…if not, was this because of health, % (n/N)[b]	54.2 (297/ 548)	37.4 (470/1258)	1.98 (1.62; 2.43)
On income support/unemployment benefit, % (n/N)	19.7 (224/1135)	16.0 (417/2612)	1.29 (1.08; 1.55)
In receipt of incapacity benefit or DLA, % (n/N)	27.0 (307/1137)	21.9 (573/2614)	1.32 (1.12; 1.55)

[a] Sample size varies between 1134 and 1143 in the not at all/somewhat explained group and between 2612 and 2633 in the largely/completely explained group.
[b] Note- denominator is those who reported not being in paid employment n= 1806

Supplementary Data

Heading for Supplementary Web Table 1. **Health and employment status showing comparison between patients across all four 'organicity' groups.**

	Total cohort	Not at all explained	Somewhat explained	Largely explained	Completely explained	χ2 (3df)	p-value
N	3781	446	698	940	1697		
Age	3781	41 (14)	45 (15)	47 (16)	48 (18)	62.9	<.0001
Male, N (%)	3781	141 (32)	256 (37)	395 (42)	828 (49)	58.6	<.0001
SF12 general health	3775	43.3 (26.5)	42.1 (25.6)	46.7 (25.2)	49.9 (24.4)	53.8	<.0001
SF12 physical functioning	3776	65.4 (38.7)	62.9 (38.1)	66.2 (37.0)	67.3 (36.6)	6.4	0.09
SF12 role physical	3773	49.9 (46.9)	50.1 (46.6)	54.0 (45.1)	57.2 (44.9)	16.1	0.001
SF12 bodily pain	3773	58.4 (35.3)	57.7 (36.2)	61.7 (36.3)	70.3 (33.1)	90.4	<.0001
SF12 social functioning	3775	62.7 (35.0)	62.1 (34.7)	66.1 (34.7)	70.9 (32.9)	45.9	<.0001
SF12 vitality	3775	40.9 (28.2)	38.6 (27.3)	42.3 (27.6)	45.6 (28.2)	35.1	<.0001
SF12 mental health	3773	54.9 (24.9)	54.3 (26.0)	57.9 (24.9)	63.4 (23.2)	84.8	<.0001
SF12 role emotional	3770	66.6 (42.5)	63.4 (43.5)	70.6 (41.4)	77.7 (37.2)	72.1	<.0001
SF12 composite score Physical Health Status	3762	42.3 (12.6)	42.2 (12.5)	43.3 (12.0)	44.3 (11.5)	17.6	0.0005
SF12 composite score Mental Health Status	3762	43.8 (11.5)	43.2 (12.5)	45.3 (11.8)	47.9 (10.8)	98.6	<.0001
HAD anxiety	3765	7.9 (5.0)	7.9 (5.0)	7.2 (4.7)	6.0 (4.3)	119.5	<.0001
HAD depression	3763	5.8 (4.7)	6.0 (4.9)	5.2 (4.4)	4.4 (4.0)	69.2	<.0001
HAD total	3755	13.7 (8.7)	13.9 (9.0)	12.5 (8.4)	10.3 (7.4)	115.7	<.0001
Not in paid employment, N (%)	3766	237 (53)	337 (49)	479 (51)	837 (50)	3.4	0.3
…if not was this because of health, N (%)	1806	117 (58)	180 (52)	181 (41)	289 (35)	49.9	<.0001
In receipt of income support unemployment benefit, N (%)	3747	92 (21)	132 (19)	173 (19)	244 (15)	15.3	0.002
In receipt of incapacity benefit or DLA, N (%)	3751	119 (27)	188 (27)	193 (21)	380 (23)	12.7	0.005