



MSc in Clinical Psychology

Functional Neurological Symptoms: Clinical Manifestations and the Effects of Interdisciplinary Rehabilitation. A Case Study

June, 2017

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Foreword and acknowledgements

This is an MSc thesis in Clinical Psychology at Reykjavík University and was written over three semesters. In the spring semester 2016, a literary review of studies in this field was turned in. The method chapter was written in the fall semester 2016 and over the spring semester 2017 data from a database was processed and this paper written.

The idea for the study was developed with Inga Hrefna Jónsdóttir, chief psychologist at Reykjalundur who is also my supervisor along with Þorlákur Karlsson. Data used in this study was extracted from a database being gathered in relation to an ongoing project at Reykjalundur about the clinical manifestation of functional neurological symptoms (FNS), effects of rehabilitation and prospects. Guðrún Karlsdóttir, head physician on the neurological interdisciplinary team at Reykjalundur, leads the project and it was approved by The National Bioethics Committee (NBC) in November 2014 with approval number 14-126 (clinical study registration number: VSNb2014090009/03.01).

The goal of the Reykjalundur project is to gather data about 30 individuals diagnosed with functional neurological symptoms disorder (FNSD) and study their outcomes after undergoing an interdisciplinary, individualized rehabilitation program. The purpose of my thesis was to perform an exploratory case study of the first four individuals that completed the program and their follow-up at six and twelve months. The emphasis was on the effects of treatment on the clinical manifestation of FNS and psychological problems. The team at Reykjalundur has gathered extensive data and the current thesis only uses a part of it related to background variables, symptoms and psychological factors. There is a great need for research into treatment options for FNSD and therefore their project is very valuable. I want to thank the members of the research team at Reykjalundur for allowing me to use their database. I especially want to thank Inga Hrefna for a rewarding collaboration and Þorlákur for his assistance.

My MSc study has been an enjoyable and long-awaited journey. On the way, I struggled with major illness. I would never have made it through without the selfless support of my husband, the patience of my three children and the encouragement of my sisters. I also want to thank the teaching staff at the MSc program and especially Jón Friðrik Sigurðsson and Áslaug Pálsdóttir for their understanding and goodwill.

Abstract

Functional neurological symptoms (FNS) are physically unexplained complaints such as hemiparesis or seizures that have similar detrimental effect on the wellbeing of patients as organic neurological disorders. Evidence based treatments for FNS are lacking although the results of using CBT and interdisciplinary treatments are promising. This exploratory multiple case study used both qualitative and quantitative measures. Qualitative measures were semi-structured interviews asking about medical conditions and stressful life events. Quantitative measures were Mini-Plus, DASS, SCQ, QOLS and IPQ-R. The study examined the manifestation of FNS and the effects of an individualized interdisciplinary treatment at Reykjalundur for four patients. The manifestation of psychological and socioeconomic factors was in concordance with previous studies. The participants had low levels of education, high incidence of stressful life events and three out of four subjects had mood or personality disorders. The rehabilitation resulted in much less FNS for all subjects and the recovery was maintained after six and twelve months. Two out of four were symptom free at twelve-month follow-up. Psychiatric diagnoses and other symptoms were fewer at twelve months than at admission and other psychological problems such as low self-esteem and poor quality of life improved. All participants had better understanding of their FNS and overall subjects had more confidence in their personal control over their illness. These results are promising, suggesting that an individualized, interdisciplinary rehabilitation program for treating FNSD is effective and improves the wellbeing of FNS patients. A larger project at Reykjalundur is currently underway.

Keywords: functional neurological symptoms disorder, conversion disorder, functional weakness, psychogenic non-epileptic seizures, interdisciplinary rehabilitation, stressful life-events, self-esteem, quality of life, illness perception

Functional Neurological Symptoms: Clinical Manifestations and the Effects of Interdisciplinary Rehabilitation

Functional neurological symptom disorder (FNSD) manifests as physical complaints concerning alterations of voluntary motor and sensory function (American Psychiatric Association, 2013). These functional neurological symptoms (FNS) are unexplained by pathophysiological or structural abnormalities or another medical or mental disorder. They must have a significant detrimental effect on the patient's social, occupational or other important functions and cause clinically significant distress.

FNSD is a wide spectrum of disorders divided into two main subtypes, psychogenic non-epileptic seizures (PNES) and functional movement disorder (FMD) (Edwards & Bhatia, 2012; Stone & Carson, 2011). FMD is categorized as functional tremors, dystonia, and myoclonus. Other subtypes of FNSD are functional weakness (FW), sensory and visual symptoms. FMD and FW can also be grouped together as functional motor symptoms (FMS) (Gelauff, Stone, Edwards, & Carson, 2013).

Hysteria, psychogenic, conversion disorder, somatization disorder, medically unexplained symptoms (MUS) and non-organic are historical terms for FNSD (Edwards & Bhatia, 2012). Many carry a stigma that is unhelpful when treating FNSD, indicating the patient's complaints are feigned. The reaction of neurological outpatients to some of these terms was explored by Stone et al. (2002) and "functional" was deemed least offensive.

The epidemiology of FNSD is problematic. FNS are often comorbid with symptoms that have a medical cause (Caplan & Nadelson, 1980; Reuber, Mitchell, Howlett, Crimlisk, & Grünewald, 2005). FNSD diagnosis requires an extensive neurological assessment making a large scale study of its prevalence difficult (Carson et al., 2012). According to Akagi and House in 2001 the incidence rate of conversion disorder is between four and twelve in 100,000 annually and the prevalence is at least 50 in 100,000 (as cited in Carson et al., 2012).

FNSD affects women two or three times more often than men (American Psychiatric Association, 2013)

The psychoanalytical ideas of conversion, disassociation and somatization of traumatic events are still the basis for modern psychiatric approaches to explaining and treating FNSD (R. J. Brown, 2004; M. Sharpe & Carson, 2001). These ideas have come under increasing scrutiny in the last decade (Edwards & Bhatia, 2012; M. Sharpe & Carson, 2001). Despite research linking FNSD to traumatic and other psychological causes, some patients have no history of traumatic events (Roelofs, Spinhoven, Sandijck, Moene, & Hoogduin, 2005; D. Sharpe & Faye, 2006).

Nevertheless, psychological and socioeconomic etiological factors cannot be overlooked. Negative life events were more common before the onset of symptoms in patients with FNS than in a control group with organically explained symptoms (Binzer, Andersen, & Kullgren, 1997). Low levels of education were significantly associated with FNS. FNSD patients were more likely to have suffered childhood trauma, sexual abuse, and reported a greater number of traumatic episodes (Bodde et al., 2009; Kranick et al., 2011; Roelofs, Keijsers, Hoogduin, Näring, & Moene, 2002; Sar, Akyüz, Kundakçi, Kiziltan, & Dogan, 2004). Studies have reported high comorbidity with psychiatric disorders such as depression, anxiety and personality disorders (Binzer et al., 1997; de Waal, Arnold, Eekhof, & Hemert, 2004; Reuber, Pukrop, Bauer, Derfuss, & Elger, 2004).

Professionals no longer view FNSD as purely psychiatric disorder (de Schipper, Vermeulen, Eeckhout, & Foncke, 2014). This shift reflects the changes in DSM-5 where a preceding psychological stressor was dropped as a diagnostic criterion (American Psychiatric Association, 2013; Czarnecki & Hallett, 2012). The current understanding is that the etiological picture of FNSD is a complex interplay between biological, psychological and social factors. More complex than the 100 years old notion of conversion of “unbearable

stress” into physical symptoms. The current focus is on finding a multifactorial disease model, pointing a way to a closer collaboration between neurology, psychiatry and psychology (Edwards & Bhatia, 2012; Reuber, 2009; Reuber, Howlett, Khan, & Grünewald, 2007; Reuber et al., 2005; Stone & Carson, 2011).

Recent systematic reviews of outcome studies for FMS (Gelauff et al., 2013) and PNES (Durrant, Rickards, & Cavanna, 2011) indicate generally poor prognosis of FNSD patients. PNES patients have worse prognosis than FMS patients (Durrant et al., 2011; McKenzie, Oto, Russell, Pelosi, & Duncan, 2010; Reuber et al., 2003). A study of neurological outpatients with MUS linked poorer outcome with expectation of non-recovery, not attributing symptoms to psychological factors and receipt of social benefits (M. Sharpe et al., 2010).

The burden of FNSD is comparable and even worse than other neurological disorders (Carson et al., 2011). Compared to Parkinson’s patients, FMD patients had similar levels of disability and quality of life (QOL) and had comparable impairment in performing activities of daily life but worse mental health scores (Anderson et al., 2007). A comparison between individuals with FW and sensory disturbance and multiple sclerosis gave similar results (Stone, Sharpe, Rothwell, & Warlow, 2003). PNES patients experienced lower health related QOL and more mood difficulties than an epileptic comparison group (Szaflarski et al., 2003).

It is very important that the patients feel their problems are taken seriously by the doctor, he or she explains their symptoms to them and that they are treatable (Carson, Lehn, Ludwig, & Stone, 2016). The neurological assessment can in itself be therapeutic and some patients experience improvement of symptoms from a single consultation (Stone, 2015). Using a standardized protocol for delivering the diagnosis of PNES can reduce symptoms (Hall-Patch et al., 2010) and explaining the attacks are non-organic can reduce use of health care services (Razvi, Mulhern, & Duncan, 2012).

No current treatment options for FNSD have strong evidence based support (Martlew, Pulman, & Marson, 2014; Morgante, Edwards, & Espay, 2013). After a neurological diagnosis, FNSD patients are in most cases referred to mental health professionals for further treatment, usually a psychiatrist (Espay et al., 2009). Psychiatric treatment is commonly based on ideas of conversion and underlying trauma, being a form of psychoanalysis and antidepressant drug therapy. Uncontrolled studies have shown some beneficial effects of these types of treatment, for example psychodynamic interpersonal therapy (Hinson, Weinstein, Bernard, Leurgans, & Goetz, 2006; Reuber, Burness, Howlett, Brazier, & Grünewald, 2007). Despite its popularity empirical backing is weak (Stone & Carson, 2011).

Physiotherapy is helpful in treating FMS. Many newly diagnosed patients view physiotherapy as more relevant than psychotherapy (Edwards, Stone, & Nielsen, 2012). In a historical cohort study, patients with chronic FMD showed significantly more improvement than a control group after one week targeted motor-reprogramming protocol (Czarnecki et al., 2012).

Cognitive behavioral therapy (CBT) is an evidence-based treatment and is promising as treatment for FNSD. A review of randomized clinical trials (RCT) found CBT had the best evidence support treating somatoform disorders, including conversion disorder (Kroenke, 2007). Despite promising early results there are still few studies on the use of CBT to treat FNSD, most focusing on PNES (Hopp & LaFrance, 2012). A pilot RCT comparing CBT with standard medical care found CBT more effective in reducing seizures in PNES patients and the CBT group was more likely to be seizure free three months after treatment (Goldstein et al., 2010). A recent pilot study using CBT-based group therapy both for PNES and other FNS patients concluded it is potentially a feasible, cost effective and easily deliverable treatment option that can improve the emotional wellbeing of FNSD patients (Conwill, Oakley, Evans, & Cavanna, 2014).

In 2011, a group of healthcare professionals recommended a three-step care model. For those clients who need more extensive treatment they propose an interdisciplinary care combining more intensive psychological treatment, psychoactive medication, physiotherapy and occupational therapy (Healthcare Improvement Scotland, 2012).

Studies into the effectiveness of multidisciplinary inpatient treatments for severe FNSD have started emerging in the last decade. The treatment protocols described have in common an individualized approach containing a combination of CBT, physiotherapy and occupational therapy. Other components such as speech therapy, physiatry, neuro- and rehabilitation psychology are sometimes also employed (McCormack et al., 2014; Yam et al., 2016). A single case-study using interdisciplinary treatment for FMD, memory loss and speech impairment resulted in the patient being symptom free 13 weeks after admission and at six months follow-up (Yam et al., 2016). The outcomes of 33 FMD patients treated using a multidisciplinary approach showed significant improvement in measurements of activities of daily living and mobility (McCormack et al., 2014).

This study examines the clinical manifestation of FNS in patients undergoing an interdisciplinary treatment at Reykjalundur, especially the manifestation of psychological factors. The expectation is that the subjects have experienced negative life events, mood or personality disorders and limited education. The prediction is that FNS and psychological problems have reduced from admission to discharge and the recovery persists at six, and twelve months' follow-up.

Method

Participants

The participants were four women, admitted to rehabilitation at the neurological department of Reykjalundur in the period January-November 2015. Details of each participant are summarized in table 1. Eligible for inclusion were patients admitted for

treatment of FNSD in the age range 18 to 67. Excluded were previous participants in the rehabilitation program and those diagnosed with an organic neurological disorder.

Table 1

Summary of Socioeconomic Status, Diagnoses and Rehabilitation Duration for All Four Subjects

	Subject			
	A	B	C	D
Gender	Female	Female	Female	Female
Age range	35-39	35-39	18-22	50-54
Diagnosis ICD-10	F44.4	F44.4	F44.5	F44.4
FNS	FW*	FW	PNES**	FW
Rehabilitation (days)	45	45	50	45
Marital status	Married/domestic partnership	Divorced	Single	Divorced
Education level	Primary school	Vocational school	Primary school	Vocational and high school
Occupational status	Employed 35% 100% before illness	On sick leave, was 80% employed	Employed 10% and part time distance learning 30%	Disability
Number of children	2	1	0	2

*Functional weakness **Psychogenic non-epileptic seizures

Design

This is an exploratory multiple case study using both qualitative and quantitative measures.

Qualitative Measures

Three semi-structured medical interviews were used. The first one gathered information about socio-economic status, medical diagnosis, other suspected diagnoses, onset, duration and manifestation of FNS and other physical symptoms on admission. The second asked about the status of the FNS at discharge. The third one was used at follow-up to gather information concerning the development of the FNS and other symptoms and if the

participant had received further treatment after discharge. Additionally, a semi-structured interview was used to ask about stressful life events (SLE).

Quantitative Measures

The Mini International Neuropsychiatric Interview-plus (Mini-Plus) is a structured diagnostic interview that includes 23 disorders. Mini-Plus has been shown to have good to excellent inter-rater reliability ($Kappa = .79 - 1.0$) and very good test/retest reliability for 14 of the 23 disorders ($k \geq .75$) (Sheehan et al., 1998). This study used an Icelandic translation of the Mini-Plus.

The Depression Anxiety and Stress Scales (DASS) is an instrument shown to have good internal consistency detecting depression ($\alpha = .91 - .96$), anxiety ($\alpha = .81 - .89$) and stress ($\alpha = .89 - .93$) (T. A. Brown, Chorpita, Korotitsch, & Barlow, 1997; Lovibond & Lovibond, 1995). Temporal stability at two weeks is also good ($r = .71 - .81$) (T. A. Brown et al., 1997). DASS has good convergent and discriminative validity (Lovibond & Lovibond, 1995). The reliability of the Icelandic translation is similar to the American version (Ingimarsson, 2010).

Robson's Self Concept Questionnaire (SCQ) is a self-report instrument designed to gauge individual's attitudes towards him or herself (Robson, 1989). SCQ has good internal consistency reliability ($\alpha = .90$) and convergent and discriminant validity (Addeo, Greene, & Geisser, 1994). SCQ was translated to Icelandic by Sóley Dröfn Davíðsdóttir but the psychometric properties of the translation are unknown (Viðar & Davíðsdóttir, 2010). British average global scores on SCQ are around 140 and *SD* is about 20. When interpreting the scores, low self-esteem is one *SD* below the average (Robson, 1989; Romans, Martin, & Mullen, 1996).

The Quality of Life Scale (QOLS) is a 16 item self-report scale that measures quality of life and has good psychometric properties. In a healthy population the total average score

is about 90 (Burckhardt & Anderson, 2003). This study uses an Icelandic translation by Pétur Tyrfingsson which has been shown to be both valid and reliable (Hrafnsson & Guðmundsson, 2007).

The Revised Illness Perception Questionnaire (IPQ-R) is a self-report questionnaire that assesses patients' representation of their illness. IPQ-R consists of seven components: identity, cause, timeline (acute/chronic and cyclical), consequences, control (personal and treatment), emotional representation and illness coherence. IPQ-R has four scales, symptoms since illness, symptoms relation to illness, views towards illness and causes of illness. Views towards illness are rated on a five point Likert scale (1 = strongly disagree, 5 = strongly agree). Internal and test-retest reliability is good and also discriminant and predictive validity (Moss-Morris et al., 2002). An Icelandic translation of IPQ-R was used (Jónsdóttir, Beck, & Hallvarðsdóttir, 2004) and reliability of the scales is good ($\alpha = .75 - .87$) (Jónsdóttir, Hreinsdóttir, Kristbergisdóttir, & Beck, in press).

Procedure

The study took place at Reykjalundur from January 2015 until December 2016. The observation points were at admission, at discharge and on follow-up after six and twelve months. On all observations, a physician interviewed the patient. A psychologist administered Mini-Plus and asked about SLEs at admission and follow-up after twelve months. A psychologist administered other scales on all observations.

All participants attended an interdisciplinary rehabilitation program at Reykjalundur. All subjects had been informed that their FNSD was non-organic and treatable before starting the program. The program was tailored for each patient based on his symptoms and impairment of abilities. It included the following components: medical and nurse supervision, education about FNS, physiotherapy, occupational therapy and cognitive behavioral therapy (CBT) both individual and group. The extent of group and individual CBT sessions is shown

in table 2. The patients came in for a day's follow-up at six and twelve months after discharge.

Table 2

Extent of the Subjects Individual and Group CBT

Subject	Group CBT	# of individual CBT sessions
A	8 mindfulness sessions	6
B	8 low self-esteem sessions	6
C	8 low self-esteem sessions	30
D	None	7

This study received approval from the NBC to use data from an ongoing study at Reykjalundur (clinical study registration number: VSNb2014090009/03.01). Approval number is 14-126 addendum 2.

Results

Subject A

Manifestations on admission. Subject A had been experiencing symptoms for seven to eleven months prior to admission. Her diagnosis was FW or hemiparesis affecting her right arm and/or leg. No specific event triggered her symptoms. She also had a diagnosis of hypothyroidism. Other symptoms experienced are listed in table 3 along with SLEs and Mini-Plus diagnoses. The participant had suffered several SLEs such as childhood sexual abuse.

Table 3

Other Symptoms, Stressful Life Events and Mini-Plus Diagnoses for Subject A

	Admission	Six months	Twelve months
Other symptoms	Speech impairment (trouble finding words)	Speech impairment (trouble finding words)	Speech impairment (trouble finding words)
	Fatigue		
	Dizziness		
	Neuralgia (right arm)		
Stressful life events	Sexual violence by a family member or someone known	N/A	Sexual violence by a family member or someone known
	Sexual violence by stranger		Sexual violence by stranger
	Sexual touching before the age of eighteen by someone at least five years older		Sexual touching before the age of eighteen by someone at least five years older
	Bullying		Bullying
	Loss of her mother as a child		Loss of her mother as a child
	Physical assault		
Mini-Plus diagnoses	Alcohol dependence and abuse (Lifetime)	N/A	Alcohol dependence and abuse (Lifetime)
	Substance dependence and abuse (Lifetime)		Substance dependence and abuse (Lifetime)
	ADHD (Attention deficit/Hyperactivity disorder) (Current)		ADHD (Attention deficit/Hyperactivity disorder) (Current)
	Antisocial personality disorder (Lifetime)		Antisocial personality disorder (Lifetime due to substance and alcohol abuse)

Depression, anxiety and stress were below clinical criteria measured on DASS. QOLS score was 100 or 10 points above the healthy average. Self-esteem was 152, about half a *SD* above a healthy average.

According to her IPQ-R identity component answers, she viewed fatigue and loss of strength as symptoms related to her FNS. Her results on the views towards illness subscale

can be seen in table 4. Subject A did not think that her illness was chronic ($M = 2.0$) or considered the consequences of her illness to be great ($M = 2.8$). She though, estimated that it had negative consequences for her closest family. The participant experienced that her symptoms were cyclical ($M = 3.5$). She believed her symptoms were controllable and the progress of her illness depended on her actions ($M = 3.8$) and was absolutely convinced that her illness was treatable ($M = 5.0$). Her comprehension of her FNS was ambiguous ($M = 3.0$) and emotional response to her illness was weak ($M = 2.5$). The main cause of her illness in her opinion was overwork but also stress.

Table 4

All Component Scores on the IPQ-R Views Towards Illness Subscale for Subject A

Component	Admission		Discharge		6 months		12 months	
	Total	<i>M</i>	Total	<i>M</i>	Total	<i>M</i>	Total	<i>M</i>
Timeline (acute/chronic)	12	2.0	11	1.8	9	1.5	12	2.0
Timeline (cyclical)	14	3.5	11	2.8	8	2.0	4	1.0
Personal control	23	3.8	26	4.3	26	4.3	26	4.3
Treatment control	25	5.0	25	5.0	25	5.0	25	5.0
Illness coherence	15	3.0	20	4.0	22	4.4	20	4.0
Consequences	17	2.8	15	2.5	16	2.7	13	2.2
Emotional representation	15	2.5	13	2.2	14	2.3	8	1.3

Treatment results. On discharge and at follow-up after six and twelve months, subject A's FNS were much less noticeable than on admission. Speech impairment was the only other symptom remaining at follow-up and she no longer mentioned assault as SLE (see table 3). Mini-Plus diagnoses did not change as seen in table 3 except the antisocial personality disorder was considered related to substance and alcohol abuse.

On all observation points, she did not show any signs of depression, anxiety or stress on DASS. Her self-esteem remained good and even showed signs of improvement and she continued to estimate her QOL as above average (see figure 1).

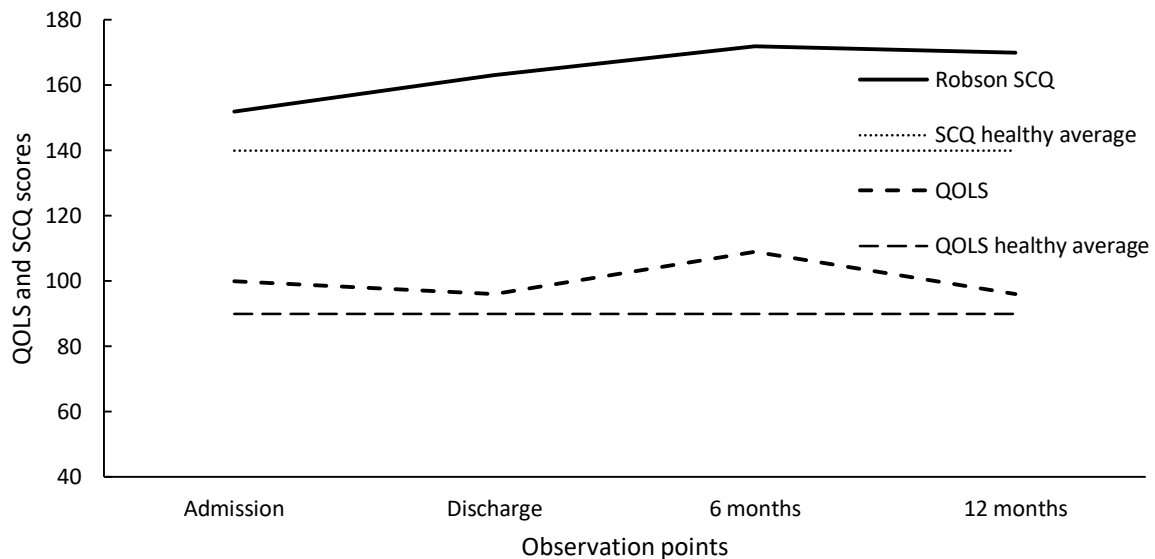


Figure 1. SCQ and QOLS for subject A across all observation points.

Her IPQ-R identity component results at discharge showed that in addition to earlier symptoms she considered weight loss to be related to her illness. She did not mention that symptom at follow-up. At twelve months, she named dizziness along with the symptoms she listed on admission.

Three components on the views towards illness scale changed markedly at discharge and follow-up (see figure 2). Her illness coherence improved ($M = 3.0$ to 4.0). As time passed, subject A became convinced that her symptoms were not cyclical ($M = 3.5$ to 1.0) and she had even stronger belief in her personal control of her FNS ($M = 3.8$ to 4.3).

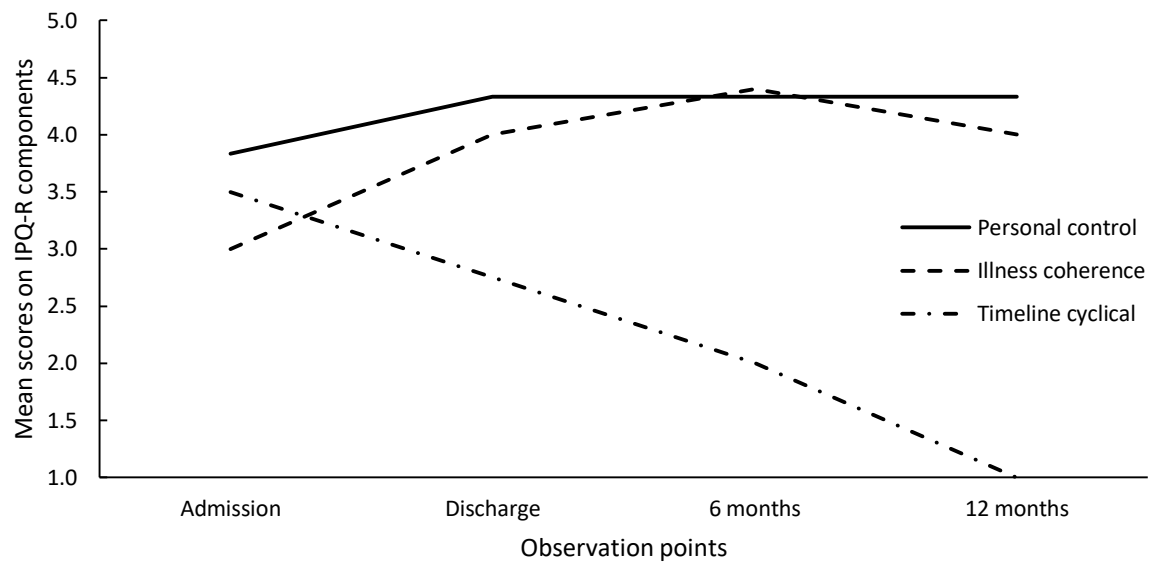


Figure 2. Mean scores on three components from the views towards illness subscale of IPQ-R for subject A.

At discharge and follow-up, she continued to attribute here FW to stress but traumatic events appeared as a new main cause for her illness.

Subject B

Manifestations on admission. Subject B's symptoms had been present three to six months prior to admission. Their onset was without any obvious cause. Her symptoms were FW in both legs (paraparesis). She had several other symptoms, SLEs and Mini-Plus diagnoses that are described in table 5. Furthermore, she was diagnosed with hypothyroidism.

Table 5

Other Symptoms, Stressful Life Events and Mini-Plus Diagnoses for Subject B

	Admission	Six months	Twelve months
Other symptoms	Fatigue	Fatigue	Fatigue
	Pain (legs)	Pain (legs, seldom)	
	Dizziness	Dizziness	
	Sleep disturbance		
	Sensory disturbance		
Stressful life events	Bullying	N/A	Bullying
	Loss of a close relative		
	Sexual touching before the age of eighteen by someone at least five years older		
Mini-Plus diagnoses	Major depressive episode (Recurrent/not current)	N/A	Major depressive episode (Recurrent/not current)
	Hypochondriasis (Current)		
	Attention deficit disorder/ADD (Current)		

Subject B's scores on DASS did not show any signs of depression, anxiety or stress. Her QOL was 74 or 16 points below the healthy average. On SCQ, here self-esteem was 87 or 2.65 *SD* below the average. This indicated a very low self-esteem and a diminished QOL.

On the IPQ-R identity component, she believed that pain, fatigue, stiff joints, sleeping difficulties, dizziness and loss of strength were symptoms related to her FW. All scores for subject B on the views towards illness are listed in table 6.

Table 6

All Component Scores on the IPQ-R Views Towards Illness Subscale for Subject B

Component	Admission		Discharge		6 months		12 months	
	Total	<i>M</i>	Total	<i>M</i>	Total	<i>M</i>	Total	<i>M</i>
Timeline (acute/chronic)	19	3.2	16	2.7	20	3.3	19	3.2
Timeline (cyclical)	10	2.5	12	3.0	11	2.8	16	4.0
Personal control	20	3.3	20	3.3	18	3.0	15	2.5
Treatment control	18	3.6	19	3.8	18	3.6	20	4.0
Illness coherence	13	2.6	19	3.8	9	1.8	17	3.4
Consequences	25	4.2	23	3.8	24	4.0	22	3.7
Emotional representation	21	3.5	22	3.7	21	3.5	17	2.8

Subject B tended to think her illness would last a long time even though she was uncertain if it was chronic ($M = 3.2$). Generally, she did not think her FNS were cyclical ($M = 2.5$). She agreed that her illness had negative consequences for her life and those around her ($M = 4.2$). She was uncertain if she had personal control over her FW ($M = 3.3$) but her believe in the treatability of her illness was stronger ($M = 3.6$). Her illness coherence was weak ($M = 2.6$). Her FW appeared to cause her emotional distress ($M = 3.5$) and she experienced depression, anger, anxiety and fear when thinking about it. She believed the main causes of her FNS were faulty nerve impulses along with overwork and incorrect physical exertion. She also mentioned chance or bad luck.

Treatment results. At discharge and six months, subject B's FNS were still present although much less noticeable than on admission but were no longer present at twelve months' follow-up. After discharge, she continued in physiotherapy. Other symptoms and SLEs were fewer on follow-up and recurring major depressive episodes (not current) was the only remaining psychiatric diagnosis (see table 5).

Her DASS scores showed no signs of depression, anxiety or stress. The subject assessed her QOL at discharge and follow-up, more than 80 or somewhat better than on

admission (74) but still below the healthy average (90). This is shown in figure 3 and how her self-esteem grew steadily from low (87) at the beginning to normal (136) at the end of observation.

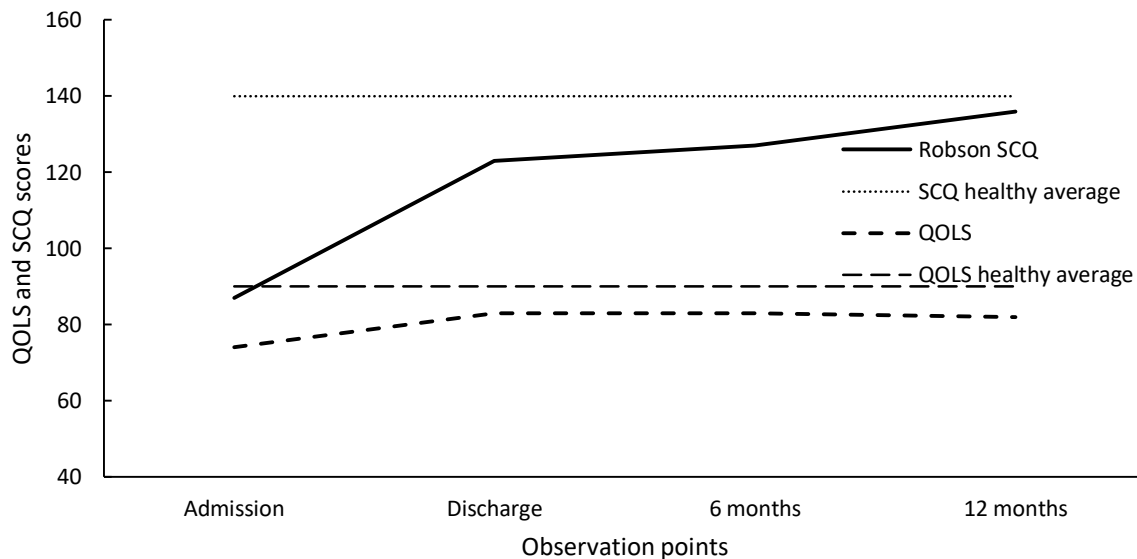


Figure 3. SCQ and QOLS for subject B across all observation points.

The symptoms related to subject B's FNS on IPQ-R fluctuated a lot on the identity component from one point to the next. Constant were pain, fatigue and loss of strength. At discharge, she only mentioned these three. At six months, she named five symptoms, stiff joints and dizziness in addition to the other three. At twelve months, she no longer complained about dizziness. This corresponded fairly well with the symptoms she described in her medical interview except that at twelve months' follow-up she only mentioned fatigue to the doctor.

Subject's B IPQ-R results on the views towards illness subscale indicated changes in four of the components (see figure 4). She had less belief in her personal control over her FNS ($M = 3.3$ to 2.5). Her illness coherence improved and was much better at discharge ($M = 3.8$) than at admission ($M = 2.6$) but was unstable. At six months, it fell to a mean of 1.8

but went up again to a mean of 3.4 at twelve. From admission ($M = 2.5$) to twelve months' follow-up ($M = 4.0$) she appeared to become more convinced that her illness was cyclical. The emotional impact of the illness appeared to lessen as emotional representation went from a mean of 3.5 to a mean of 2.8 after twelve months.

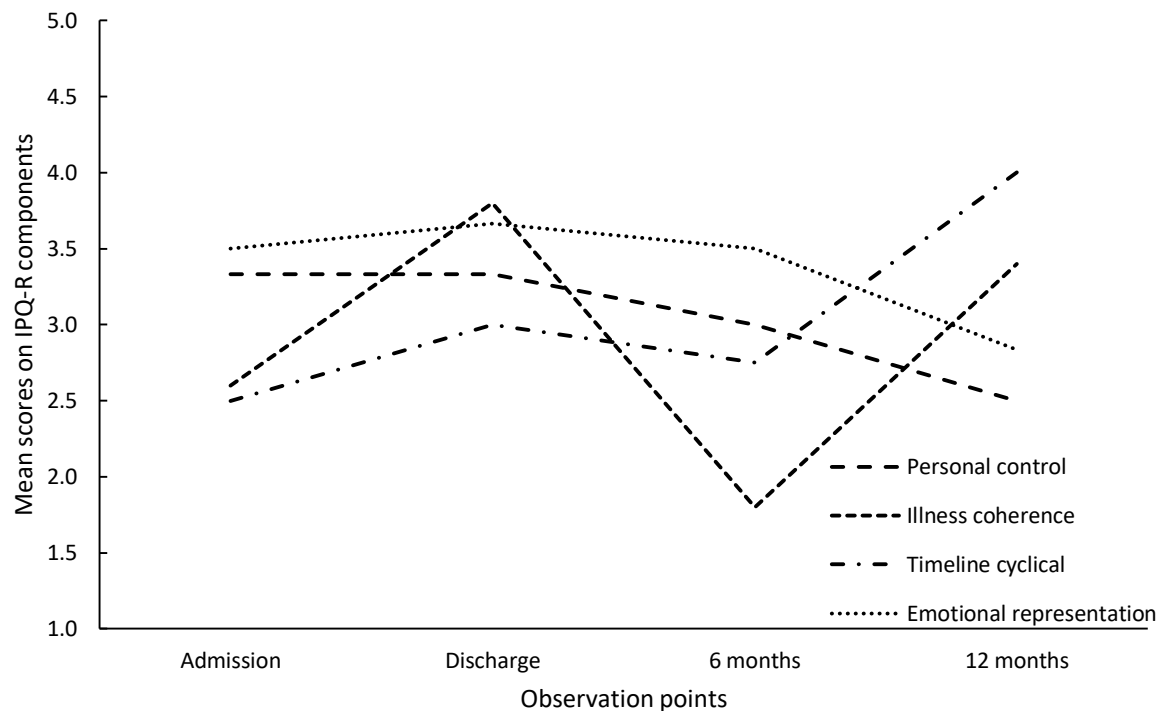


Figure 4. Mean scores on four components from the views towards illness subscale of IPQ-R for subject B.

Subject's B ideas about the causes of her illness fluctuated. Constant were stress and bad luck but she also seemed to be looking for environmental causes for her illness such as bacteria or pollution. At twelve months, she listed bad luck as the main cause, followed by bacteria and stress.

Subject C

Manifestations on admission. Subject C had been suffering from PNES (fainting) three to four years prior to admission. She believed her illness was onset by a head injury

caused by an accident. She did not have a diagnosis of other diseases. She complained of several other symptoms, had experienced numerous SLEs and had five psychiatric diagnoses on Mini-Plus as listed in table 7.

Table 7

Other Symptoms, Stressful Life Events and Mini-Plus Diagnoses for Subject C

	Admission	Six months	Twelve months
Other symptoms	Fatigue	Fatigue	Fatigue (controllable)
	Sleep disturbance	Sleep disturbance	
	Dizziness		Dizziness
	Colon spasms		Colon spasms
	Pain (head and back)		
	Sensory disturbance		
	Memory impairment		
Stressful life events	Sexual violence by a family member or someone known	N/A	Sexual violence by a family member or someone known
	Sexual violence by stranger		Sexual violence by stranger
	Bullying		
Mini-Plus diagnoses	Major depressive episode (Recurrent/not current)	N/A	Major depressive episode (Recurrent/not current)
	Panic disorder (Lifetime)		Panic disorder (Lifetime)
	Agoraphobia (Current)		Agoraphobia (Current)
	Social phobia (Current/past month)		
	ADD (Current)		

Subject's C DASS scores at admission indicated mild depression (12), moderate anxiety (14) and no stress (14). At the beginning of treatment, her QOLS score was 57 showing that she estimated her QOL as poor and self-esteem score on SCQ was low at 76 or 3.2 SD below the healthy average.

Subject C thought seven of the items on the IPQ-R identity component related to her PNES; breathlessness, fatigue, headaches, upset stomach, sleep difficulties, dizziness and loss of strength. Table 8 show subject C's scores on the views towards illness subscale. She experienced her illness as acute ($M = 2.2$) and cyclical ($M = 4$). She rated the consequences ($M = 3.5$) of her PNES as negative for her life. She had some belief in her personal control ($M = 3.5$) and agreed that her symptoms were treatable ($M = 3.8$). Her illness coherence was low ($M = 1.4$), reflecting a little understanding of her PNES. She had a negative emotional representation of her illness ($M = 3.7$). It caused her depression, worry and anger. The most important causes of her illness in her opinion were head injury caused by an accident, rape and stress.

Table 8

All Component Scores on the IPQ-R Views Towards Illness Subscale for Subject C

Component	Admission		Discharge		6 months		12 months	
	Total	<i>M</i>	Total	<i>M</i>	Total	<i>M</i>	Total	<i>M</i>
Timeline (acute/chronic)	13	2.2	9	1.5	13	2.2	13	2.2
Timeline (cyclical)	16	4.0	15	3.8	18	4.5	20	5.0
Personal control	21	3.5	23	3.8	20	3.3	28	4.7
Treatment control	19	3.8	24	4.8	20	4.0	24	4.8
Illness coherence	7	1.4	14	2.8	16	3.2	20	4.0
Consequences	21	3.5	16	2.7	16	2.7	20	3.3
Emotional representation	22	3.7	13	2.2	20	3.3	15	2.5

Treatment results. Subject C's PNES was still present but much less noticeable at all observations after admission. She continued to see a psychologist outside Reykjalundur and a personal physical trainer. At follow-up, other symptoms, SLEs and psychiatric diagnoses were considerably fewer than on admission (see table 7).

Subject C's emotional state through the observation period was unstable (see figure 5). When discharged her DASS scores showed no signs of depression, anxiety or stress. She was going through emotional difficulties at six months since her DASS was 34 on depression, 36 on anxiety and 38 on stress. Her emotional condition had improved markedly at follow-up at twelve months but she did show signs of moderate anxiety (10).

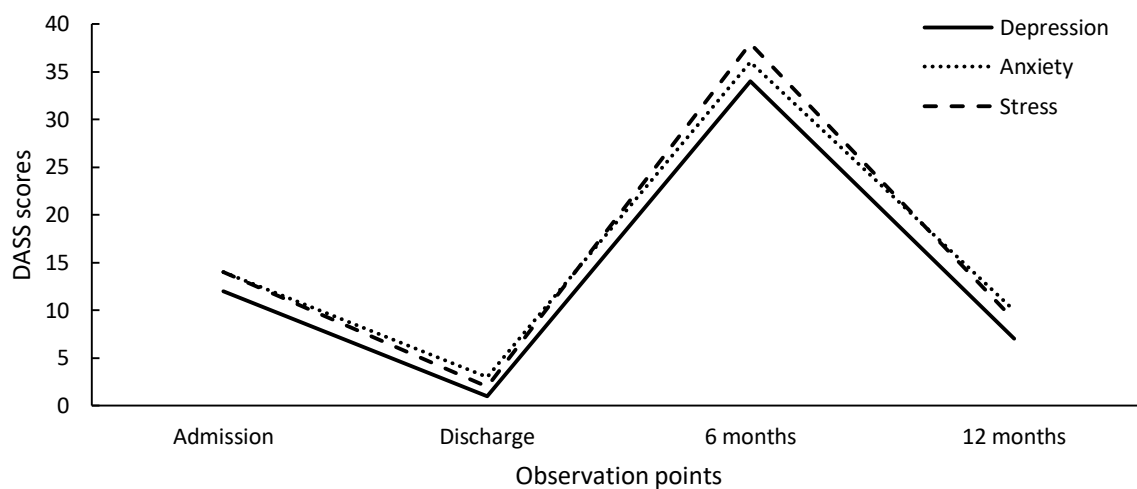


Figure 5. DASS scores for subject C across all observation points.

Instability was also reflected in Subject C's QOLS and SCQ scores (see figure 6). When discharged her QOLS score had risen to a healthy average (91). During her episode at six months, her score dropped to 55. At that time, she estimated her QOL as poor but that estimate changed to 80 at twelve months. Her SCQ showed a similar pattern, rising to a normal level (132) at discharge, falling drastically (75) during her emotionally troubled period and then recovering at twelve months (126).

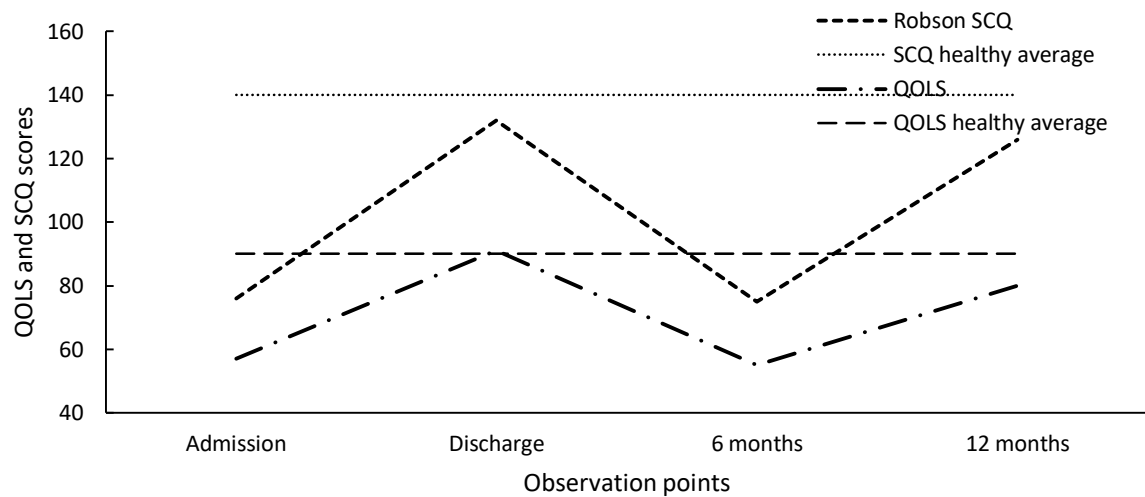


Figure 6. SCQ and QOLS for subject C across all observation points.

Subject C identified a number of symptoms as related to her illness on IPQ-R but constantly named, breathlessness, fatigue, headaches, sleep difficulties, dizziness and loss of strength. At all observation points after admission, she also named pain.

The five components on the views towards illness subscale that showed significant changes for subject C are shown in figure 7. The components that follow the subject's emotional swings are emotional representation, treatment and personal control. Overall, emotional representation decreases ($M = 3.7$ to 2.5) and belief in treatment ($M = 3.8$ to 4.8) and personal control ($M = 3.5$ to 4.7) increases. Her illness coherences increased steadily from a mean of 1.4 to 4.0 . She went from agreeing ($M = 4.0$) to strongly agreeing ($M = 5.0$) in the cyclical nature of her PNES. Stress and accident were constant causes for her illness but during and after her episode, she also named emotional state as a cause. At twelve months, she named chance or bad luck as well.

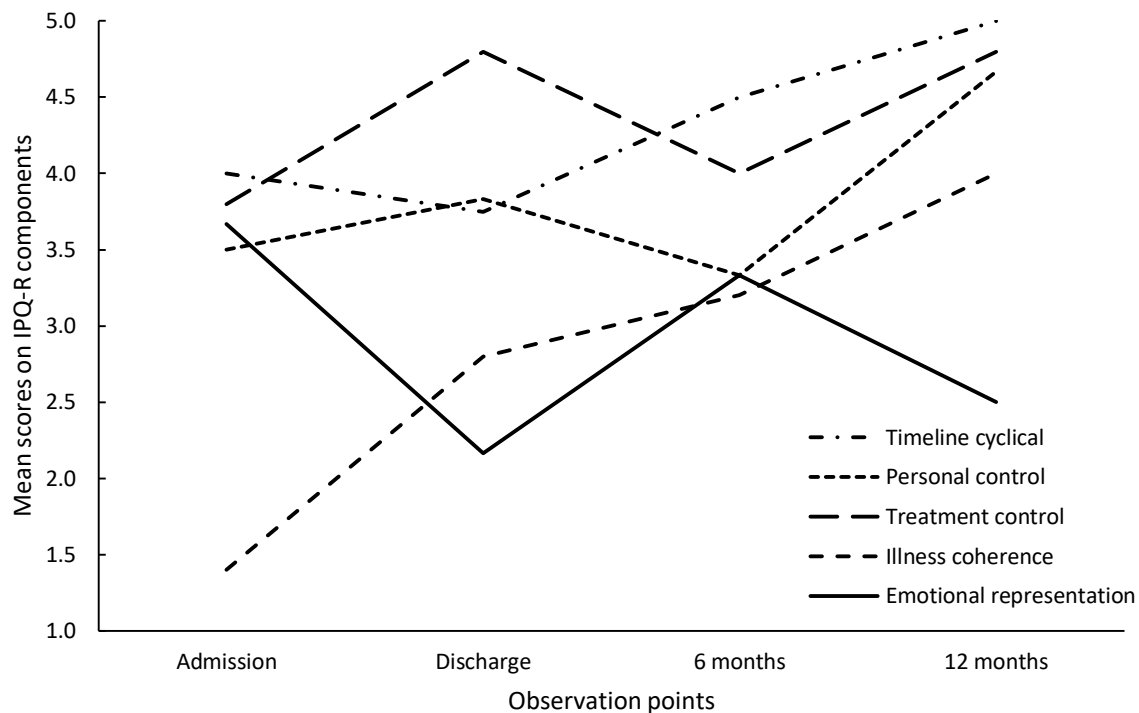


Figure 7. Mean scores on five components from the views towards illness subscale of IPQ-R for subject C.

Subject D

Manifestations on admission. Subject D had been experiencing FNS seven to eleven months prior to rehabilitation. Her symptoms were FW or hemiparesis affecting her left arm and/or leg. Her symptoms first emerged following a prolonged period of stress and after witnessing a fatal automobile accident where she knew the deceased. She was diagnosed with type 2 diabetes and Hashimoto's thyroiditis. She experienced other symptoms and SLEs but met no diagnostic criteria on Mini-Plus (see table 9). Correspondingly, her DASS scores showed no signs of emotional difficulties. She estimated her QOL (96) and self-esteem (138) as good.

Table 9

Other Symptoms, Stressful Life Events and Mini-Plus Diagnoses for Subject D

	Admission	Six months	Twelve months
Other symptoms	Fatigue	Fatigue	Loss of bladder control
	Pain (headaches, shoulder)	Loss of bladder control	Memory impairment
	Sleep disturbance	Sensory disturbance (numb fingers)	
	Memory impairment		
Stressful life events	Loss of her mother and two miscarriages	N/A	Loss of her parents
	Witnessing a serious accident		
	Assault by a family member or someone known		
Mini-Plus diagnoses	None	N/A	None

Subject D related all nine symptoms she experienced from onset to her FNS on the IPQ-R identity component. These were pain, weight loss, fatigue, stiff joints, sore eyes, headaches, sleep difficulties, dizziness and loss of strength. Her scores on the views towards illness subscale are listed in table 10. She did not experience her FW as chronic ($M = 1.8$) or cyclical ($M = 2.0$). She was overall ambiguous about consequences of her illness ($M = 3.3$) but believed strongly in personal and treatment control ($M = 5.0$). She did not have good understanding of her FNS ($M = 3.0$) or show a strong emotional representation ($M = 2.5$). She named stress, overwork and her own behavior as the main causes of her FW.

Table 10

All Component Scores on the IPQ-R Views Towards Illness Subscale for Subject D

Component	Admission		Discharge		6 months		12 months	
	Total	<i>M</i>	Total	<i>M</i>	Total	<i>M</i>	Total	<i>M</i>
Timeline (acute/chronic)	11	1.8	12	2.0	14	2.3	10	1.7
Timeline (cyclical)	8	2.0	4	1.0	8	2.0	6	1.5
Personal control	30	5.0	30	5.0	23	3.8	30	5.0
Treatment control	25	5.0	25	5.0	23	4.6	25	5.0
Illness coherence	15	3.0	25	5.0	20	4.0	25	5.0
Consequences	20	3.3	18	3.0	20	3.3	18	3.0
Emotional representation	15	2.5	12	2.0	15	2.5	12	2.0

Treatment results. When discharged and at six months, subject D's FNS were still present but much less noticeable but were no longer present at twelve. She continued in physiotherapy but had stopped at twelve months. Other symptoms and SLEs decreased at follow-up and she still did not meet any Mini-Plus criteria (see table 9). DASS showed no emotional troubles at discharge and follow-up. Her self-esteem and QOL remained good.

The subject D's symptoms on the IPQ-R identity component decreased from nine at admission to five at discharge and four at six months' follow-up. At twelve months, she related seven symptoms to her illness, which did not correspond to the medical interview. She consistently considered pain, fatigue, stiff joints and loss of strength as related to her FW.

There were little changes in her illness representation on IPQ-R during the observation period (see table 10), except those shown in figure 8. Her belief in personal control ($M = 3.8$) waned somewhat at six months but was again very strong at twelve ($M = 5.0$). Her illness coherence improved from being a mean of 3.0 on admission to 5.0 at twelve months. From the beginning of treatment, subject D consistently believed stress, overwork and her own behavior caused her illness.

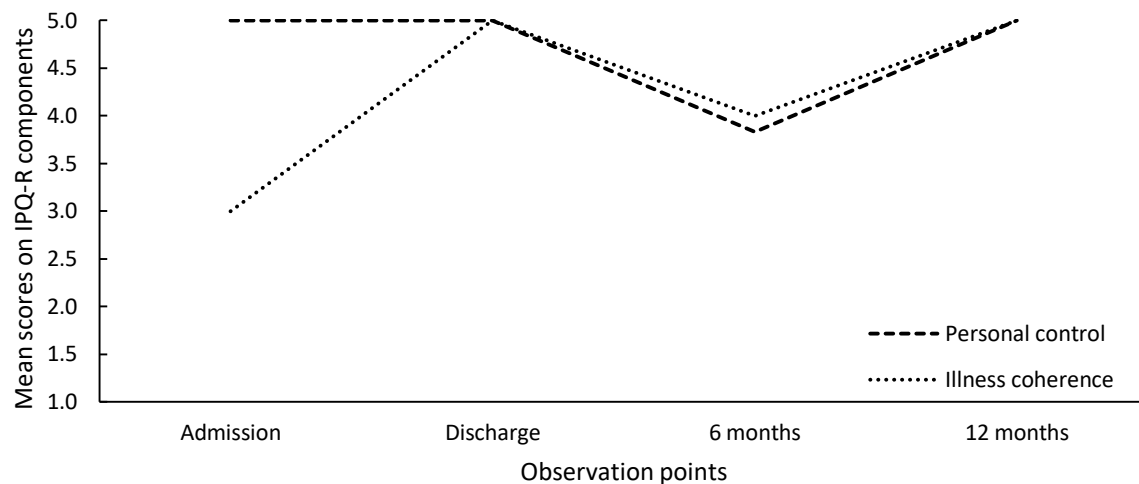


Figure 8. Mean scores on two components from the views towards illness subscale of IPQ-R for subject D.

Discussion

The main outcome of this study is that the interdisciplinary rehabilitation resulted in an improvement for all participants. All had much less noticeable symptoms at discharge and two were free of FNS at twelve months. No relapses were observed during the follow-up and, therefore, the recovery was maintained. Other symptoms and psychiatric diagnoses decreased across the observation points. The self-esteem on SCQ of all participants improved noticeably. The self-esteem of the participants with low self-esteem at admission rose to near healthy average. The QOL of the two participants who estimated it as poor at admission experienced it as considerably better at twelve months. These results suggest that the subjects' dealing with psychological problems at admission showed recovery that was persistent at twelve months.

All participants gained illness coherence, indicating that they understood their FNS better. Three out of four participants gained confidence in their personal control and all believed their FNS were treatable. No participant perceived the FNS as chronic and that perception did not change over the observation period. It was varied how cyclical the subjects

experienced their FNS, two gained and two had less belief. All evaluated the consequences and the emotional representation of their illness as less at twelve months than at admission. The IPQ-R views towards illness components give valuable insights into how a patient perceives his or her illness. It is recommended to use it to guide treatment to strengthen factors such as personal control and illness coherence that can be helpful for recovery. It was not possible to detect any distinct patterns in the subjects' answers on the identity and causes IPQ-R components related to their recovery.

All subjects had difficult life experiences. Two had a history of childhood sexual abuse. Three had suffered sexual violence and bullying. One had a history of physical violence. Interestingly, even with their history of SLEs, none of them met the diagnostic criteria for posttraumatic stress disorder on Mini-Plus. Nevertheless, their history corresponded with earlier studies of the relation between FNSD and traumatic life events. The same goes for psychiatric diagnoses since three of them had mood or personality disorders. Despite this and the fact that they were dealing with FNSD only one subject showed signs of depression, anxiety and stress on DASS during the observation period. All four had low levels of education, likewise linked to FNSD in earlier studies. It is noteworthy that all three subjects with FW were also diagnosed with thyroid dysfunction. This variable should be controlled for in future studies to estimate its relationship to FW.

In this study, there is no comparison group of FNSD patients that did not undergo the interdisciplinary rehabilitation. Such a comparison group could have been attained by using the waitlist of FNSD patients at Reykjalundur. It was not possible to ascertain if the difference was significant on the quantitative measures between observation points because the sample size was too small. It is also not clear which treatment components were effective. It would have been interesting to ask the subjects which components they felt were most helpful. Furthermore, it is unclear which factors brought about the full recovery of two

subjects after twelve months. It is possible that something other than the treatment effects contributed to this result. It is impossible to generalize from these results for four female cases to a larger population of FNSD patients.

The positive side of this study was that it used a number of different measures for psychological factors and followed the subjects for a whole year. It is probable that the good recovery of the subjects is related to their belief in the treatability of their FNS and increased understanding of their illness. The facts that the subjects' problems were acknowledged, they were informed that their FNS was treatable and the subjects were treated the same as other patients at Reykjalundur have probably increased the believability of the rehabilitation program. Also, the program was individualized making it possible to address each subject's specific problems which should have given the subjects more faith in the treatment. The subject with PNES showed more instability in the follow up than the FW subjects indicating that her prognosis was worse than the FW subjects as studies have shown.

This study suggests that an individualized, interdisciplinary rehabilitation program for treating FNSD is effective and improves the wellbeing of FNS patients. A larger study is currently underway at Reykjalundur and it will be interesting to see if the results from a larger group of subjects will echo the results of this exploratory multiple case study. FNSD is a seriously disabling condition for its sufferers. These results are promising and give FNS patients hope they can recover from their burden.

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