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A case series of Acceptance and Commitment Therapy (ACT) for reducing symptom interference in functional neurological disorders

Christopher D. Graham*^{1,2}, Daniel J. O'Hara², Steven Kemp²

¹Leeds Institute of Health Sciences, University of Leeds, Leeds, UK, LS2 9LJ.

²Department of Clinical Neuropsychology, Leeds Teaching Hospitals NHS Trust, St. James University Hospital, Leeds, UK, LS9 7TF.

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*Requests for reprints should be addressed to Dr Christopher D. Graham, Leeds Institute of Health Sciences, University of Leeds, Leeds, UK, LS2 9LJ. E-mail: c.d.graham@leeds.ac.uk

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Abstract

There is limited high-quality evidence supporting psychological treatments for functional neurological disorders (FND), and what evidence exists suggests that the impact of such treatments could be improved. One way to increase effectiveness is to utilise approaches that can have impact across heterogeneous FND presentations. Acceptance and Commitment Therapy (ACT) targets a trans-diagnostic process called psychological flexibility, and is used effectively to integrate multidisciplinary treatments in other clinical contexts. Here, we present a consecutive case series (N = 8) of a relatively brief (6 to 10 sessions) ACT intervention, delivered face-to-face by a clinical psychologist in an outpatient neuropsychology service. Treatment aimed to reduce symptom interference and improve mood via improvements in psychological flexibility. Service users presented with a range of FND symptoms (e.g. syncope, limb paralysis, paresthesia). Following treatment, five participants showed reliable improvements in symptom interference (WSAS), two to the extent of clinical significance; four had reliable improvements in mood (CORE-10), two within the range of clinical significance. There were no reliable deteriorations in symptom interference or mood. Marked variation was apparent on the measure of psychological flexibility (AAQ-II), with four reliable improvements, three within the range of clinical significance, but also two reliable deteriorations. These promising results suggest that further investigation of an ACT approach to FND is warranted. Future studies should include measures of psychological flexibility with greater comprehensibility.

Keywords: acceptance and commitment therapy; mindfulness; functional neurological disorders; functional movement disorders; conversion disorders.

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Key Practitioner Message

- Acceptance and commitment therapy (ACT) is a trans-diagnostic treatment. This may enable impact across the psychologically heterogeneous FND population.
- This case series provides a promising evaluation of a brief ACT intervention, with the majority experiencing large improvements in symptom interference and/or mood.
- A multi-disciplinary ACT approach (i.e. also including physiotherapists, occupational therapists) may be required to affect improvements in those with the most severe symptom interference.
- This treatment approach did not have a priori assumptions regarding the cause of FND, and consequently placed emphasis on participant experiences of living with FND.

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Introduction

Functional Neurological Disorders (FND), sometimes called ‘conversion disorders’, involve neurological symptoms, such as seizures, limb weakness, and dystonia that are inconsistent with known disease pathologies but are not thought to be feigned (van der Hoeven et al., 2015). They detrimentally affect functioning, quality of life and mood (Carson et al., 2011; Vroegop, Dijkgraaf, & Vermeulen, 2013), and although some people become symptom free following diagnosis, for others symptoms persist and proliferate (Duncan et al., 2014; Gelauff, Stone, Edwards, & Carson, 2014; McKenzie, Oto, Graham, & Duncan, 2011).

It is unclear what interaction of biopsychosocial factors accounts for FND in any given case.

However, many theorise that emotional processes have primacy; suggesting that, for example, FND result from the suppression into the subconscious of traumatic experiences (Breuer, 1955), and/or emotion dysregulation (Vuilleumier, 2005). The high incidence of trauma, alexithymia (Demartini, Petrochilos, et al., 2014), psychiatric diagnoses (Bowman, 1993; Stone, Warlow, & Sharpe, 2010) and evidence of differential emotional responding in FND (Blakemore, Sinanaj, Galli, Aybek, & Vuilleumier, 2016), support such theories.

However, there is heterogeneity in presentation: some with FND do not report antecedent trauma, emotion dysregulation, or have pre-morbid psychiatric diagnosis (Stone & Edwards, 2011; van der Hoeven et al., 2015). Indeed, newer theories posit several routes to the development or maintenance of these conditions, eventually also involving attentional dysfunction, and altered sensory feedback from movement (Edwards, Adams, Brown, Pareés, & Friston, 2012).

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In clinical practice, people with FND are often referred for psychological therapy, medication for mood or physiotherapy. Reviews of psychological interventions have uncovered little supportive high-quality evidence (Martlew, Pulman, & Marson, 2014; Ricciardi & Edwards, 2014); however, there is an emerging evidence-base. Small-scale or uncontrolled trials of psychodynamic approaches have yielded promising (Mayor, Howlett, Grunewald, & Reuber, 2010) or mixed (Kompoliti, Wilson, Stebbins, Bernard, & Hinson, 2014) results. Trials of traditional cognitive behaviour therapy (CBT) with very small sample sizes have returned positive evaluations of (Dallocchio, Tinazzi, Bombieri, Arnó, & Erro, 2016; LaFrance et al., 2014). Regarding larger trials, a randomised control trial (RCT) of a CBT guided self-help intervention observed a short-term effect on symptom severity, with a mixed impact on mood (Sharpe et al., 2011), while a pilot RCT of higher-intensity CBT returned similar results (Goldstein et al., 2010). Thus, while promising, it seems that the impact of psychological treatments for FND could be improved.

Acceptance and Commitment Therapy (ACT) for FND

Although ACT overlaps with CBT (e.g. centrality of behaviour change) and psychodynamic treatments (e.g. relational), it emerges from differing philosophical assumptions, and consequently has some novel features (for discussion see Herbert & Forman, 2013). Most saliently, treatment is not necessarily focused on the control of unwanted emotions or on aligning beliefs with essential truths. Instead ACT aims to engender a process called psychological flexibility, defined as consisting of three components: "...the capacity to persist or to change behaviour in a way that includes conscious and open contact with thoughts and feelings (openness), 2) appreciates what the situation affords (awareness), and 3) serves one's goals and values (engagement)" (McCracken & Morley, 2014, p. 225.) ACT uses a range of methods to improve psychological flexibility: mindfulness and perspective-taking exercises are used to create the conditions for participants to choose actions that are

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consistent with their values, for example. There is strong evidence supporting ACT for improving outcomes in chronic pain (Hann & McCracken, 2014), and growing evidence in mental health conditions (A-Tjak et al., 2015) and chronic diseases (Graham, Gouick, Krahe, & Gillanders, 2016).

For a range of different reasons, several have advanced that ACT may be particularly useful in the context of FND (G. Baslet, Dworetzky, Perez, & Oser, 2015; Gaston Baslet & Hill, 2011; Cope, Poole, & Agrawal, 2017; Graham, Stuart, O'Hara, & Kemp, 2017). First, because ACT's target, psychological flexibility, is a trans-diagnostic process (Levin et al., 2014) it may have impact across the psychological heterogeneity evident in FND (Graham et al., 2017). Ergo, where FND is experienced as the result of emotion dysregulation or underlying mental health conditions, then ACT, like many other psychological therapies, is potentially applicable (Gaston Baslet & Hill, 2011; Cope et al., 2017). However, as treatment is not predicated on uncovering a psychological cause/maintaining factor for symptoms, ACT may be equally applicable to those not readily reached by existing approaches. For instance, where there is no control over symptoms and an absence of emotional antecedents/modifiers, then distress and illness beliefs may occur simply as a reflection of the context. Here ACT's focus on enhancing personally meaningful activity via openness and awareness offers scope for improvements in functioning and quality of life – and even with residual symptoms (Graham et al., 2016; Graham et al., 2017). Second, there are calls for multidisciplinary treatments for FND (Demartini, Batla, et al., 2014; Stone & Carson, 2017) and, as has been consistently demonstrated in chronic pain contexts (e.g. Vowles & McCracken, 2008), ACT offers an effective framework for integrating multidisciplinary approaches.

Thus, over a 12-month period in routine clinical practice offered ACT to a proportion of our outpatient service users with FND. Symptom interference (primary outcome), mood and

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psychological flexibility (secondary outcomes) were measured before and after treatment and analysed via calculation of reliable and clinically significant change.

Methods

Design

A case series, involving outcome data collected at pre- and post- intervention within routine clinical practice (ethics review: National Health Service [NHS] audit/service evaluation).

Participants

All included participants had a diagnosis of FND, which was first delivered by a neurologist.

They were then routinely referred to an outpatient NHS clinical neuropsychology service,

which provides treatment that is free of charge at the point of access. An initial meeting

between a clinician and those referred to the service assessed suitability for a short-term

psychological intervention. Participants offered this ACT intervention were those who (a)

were judged as likely to benefit from a short-term outpatient psychological treatment that

went beyond simple psychoeducation, and (b) consented to participate in psychological

therapy, and (c) after explanation were interested in an ACT approach. To allow for adequate

exposure to treatment, all service users who completed at least six sessions of ACT were

included in the analysis.

Procedure

Service users were treated by a clinical psychologist with training in ACT. In line with

service parameters, they were offered up to eight weekly-to-fortnightly, hour-long face-to-

face treatment sessions. This number could be extended slightly based on need. Outcomes

were measured pre- and post- intervention using validated self-report questionnaires of

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symptom interference (primary outcome), mood, and, psychological flexibility (secondary outcomes).

Intervention

A detailed case example of **this** intervention can be found here: Graham et al., (2017). The intervention comprised assessment, formulation and treatment stages. Within the assessment, the service user's understanding of FND was elicited. The therapist discussed FND as something that is reasonably common, is produced by brain function, with some people recovering from symptoms over time. We assessed for the possibility that better psychological flexibility might reduce the symptom interference caused by FND. Assessment resulted in an individualised formulation, which was then followed by an individualised intervention. **Formulation and intervention involved functional analysis: participants were encouraged to notice clinically relevant behaviours (e.g. attempts at symptom control, rumination etc.), then to become aware of their function, consequences and effectiveness – in terms of making progress on their own values (Table 1).** The clinical conversation (relational framing, metaphors; Villatte, Hayes, & Villatte, 2015) and experiential tasks were used as vehicles for psychological flexibility (see Table 1). In line with the apparent psychological heterogeneity in FND presentations, treatment could be directed towards functioning well in the presence of FND, and/or other sources of pain (difficult social contexts, emotions, memories etc.), **given what was workable within the number of treatment sessions available.**

Measures

Work and Social Adjustment Scale (WSAS)

The WSAS (Mundt, Marks, Shear, & Greist, 2002) is a five-item measure of symptom interference ascribed to an identified condition (here FND). It has shown **acceptable psychometric properties (Mundt et al., 2002).** Each item represents an area of functioning

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(e.g. socialising, working). Responses are made on a scale from 0 (not at all impaired) to 8 (severely impaired). Scores range from 0 to 40, with higher scores indicating greater symptom interference. We made this the primary outcome because it captures improvements in functioning (which may occur even with symptoms unchanged) – as opposed to narrow measurement of symptom severity alone.

Clinical Outcomes in Routine Evaluation – 10 (CORE-10)

The CORE-10 is a ten-item measure of mood and distress (Barkham et al., 2013). Scores range from 0 to 40, with higher scores indicating greater mood disturbance. It has clinical cut-offs of >11 for general psychological distress and >13 for depression. Initial psychometric evaluations of the CORE-10 have shown strong psychometric properties, for use in psychological therapy contexts (Barkham et al., 2013). This was chosen a secondary outcome variable because mood disturbance is a frequent FND comorbidity (Stone, 2009).

Acceptance and Action Questionnaire II (AAQ-II)

The AAQ-II (Bond et al., 2011) was chosen because it is the most widely-used measure of Psychological flexibility. It has 7 items (e.g. “Emotions cause problems in my life”), and participants respond to each on a scale, ranging from 1 (never true) to 7 (always true). Total scores can range from 7 to 49, with higher scores indicating worse psychological flexibility. Quantitative studies suggest that the AAQ-II has strong psychometric properties (Bond et al., 2011). However, some have questioned the content validity and comprehensibility of the questionnaire (Francis, Dawson, & Golijani-Moghaddam, 2016; Wolgast, 2014).

Analysis

The Reliable Change Index (RCI) and Clinically Significant Change scores (CSC) were calculated from the pre- and post- intervention data gathered from each service user. These analyses use normative data (where possible) to calculate parameters indicating that, a)

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change is greater than measurement error (reliable change), and b) change is to the extent that it indicates clinical recovery (clinically significant change; (Jacobson & Truax, 1991; Morley & Dowzer, 2014). Repeated measures Cohen's d was calculated to investigate the magnitude of treatment effects. As recommended (Morris & DeShon, 2002), the calculation included pre-intervention standard deviation at both time-points. The convention for rating magnitude was: Large: $d > 0.8$; Medium: $d = 0.5 - 0.8$; Small: $d < 0.4$.

Results

The eight service users presented with at least one of the following FND symptoms: arm tremor, leg weakness/paralysis, propriospinal myoclonus, paresthesia, visual disturbance, syncope. Most were female, and ages ranged from 18 to 65 years. Many reported co-morbid health or mental health conditions. As evident in Figure 1, pre-intervention functional impairment was generally quite high, the average being slightly higher than a functional movement disorder sample included in a previous trial of physiotherapy ($M = 24.7$, $SD = 7.9$; Nielsen et al., 2016). All service users scored above both cut-offs (psychological distress, probable depression) on the CORE-10 (Figure 2). As shown in Figure 3, the initial psychological flexibility of the group was similar a sample collected from an outpatient counselling service ($M = 28.34$, $SD = 9.92$; Bond et al., 2011). Service users attended between six and 10 treatment sessions (inclusive).

On average, improvement in FND symptom interference was of a large magnitude ($d = 1.02$), with five (of eight) experiencing reliable improvements - two to the extent that it indicated clinical recovery (Figure 1). There were no reliable deteriorations in symptom interference. Similarly, a large improvement in mood was noted ($d = 1.70$); four service users showed reliable improvements, two to the extent of clinical recovery, with no reliable deteriorations (Figure 2). Improvement in psychological flexibility was of a medium magnitude ($d = 0.77$),

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with reliable improvement evident in four cases, two to the extent that it indicated clinical recovery. However, two reliable deteriorations - one to the extent of clinical deterioration - were evident (Figure 3). Those four who experienced an improvement in psychological flexibility also experienced improvements in symptom interference and mood; while the two with a reliable decline in psychological flexibility reported no change in symptom interference or mood. There was no clear pattern of difference in the number of treatment sessions attended between those who did (6-10 sessions) and did not (8 -10 sessions) experience reliable improvements in the primary outcome.

Discussion

We offered ACT for FND due to its theoretical applicability across the psychological heterogeneity apparent within this group, and demonstrated effectiveness in similar contexts (A-Tjak et al., 2015; Hann & McCracken, 2014). This pragmatic case series provided a promising initial evaluation of this brief intervention. It was notable that most service users had reliable improvements in symptom interference and/or mood, with many to the extent of clinical significance. Indeed, these within group improvements were of a large magnitude, and there were no reliable deteriorations. Nonetheless, a key characteristic of the smaller group of non-responders on the primary outcome ($n = 3$), was that they reported extremely severe functional impairment at presentation ($\geq 35/40$ on the WSAS). This is consistent with other clinical reports showing that those with extremely severe symptoms are less likely to respond to a brief psychological intervention alone (Davison, Sharpe, Wade, & Bass, 1999). This group may have required a longer duration of treatment or an integrated multidisciplinary approach (i.e. involving physiotherapy, and other treatments.) We advance that, as is done effectively in chronic pain contexts (Vowles & McCracken, 2008), ACT might be used to integrate such multidisciplinary treatments for FND.

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Marked variability of response was evident on our measure of psychological flexibility (AAQ-II), with large improvements and also two reliable deteriorations. Although deteriorations in psychological flexibility may have occurred, we suspect that the comprehensibility of the AAQ-II questionnaire best explains this phenomenon. The AAQ-II involves esoteric concepts - for example, “My painful memories prevent me from having a fulfilling life”. Such concepts may be difficult to understand until one is socialised to the treatment model, potentially leading to a worsening of score as understanding improves, as well as vice versa. Future researchers might consider using psychological flexibility measures that appear to have greater comprehensibility - for example the CompACT questionnaire (Francis et al., 2016).

Many psychological approaches to FND make assumptions that specific psychological factors cause or maintain symptoms (e.g. Russell et al., 2016; Dallochio et al., 2016).

Therapy then targets these psychological factors to directly ameliorate symptoms.

Alternatively, ACT does not require a priori assumptions regarding FND aetiology, and focuses first on living well in a difficult context. This allows consideration to be given to a broad range of psychological factors and responses to living with FND. Consequently, closely guided by each participant’s experiences of living with FND we aimed to help them enact behaviours that were in line with their own personal values. For instance, where the function of participant behaviour appeared to include uncovering an alternative cause for their symptoms, their attention was directed to the consequences of such behaviour. Cause-finding behaviours could be helpful: noticing the possible influence of emotions on symptom experiences, or seeking further applicable treatments. These behaviours could also be unhelpful – mirroring the stagnation of worry and rumination, increasing symptom-focused attention or leading to ineffective use of healthcare services.

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This experiential and cause-agnostic stance may mean that services users who do not accept a psychological explanation for FND could not fully partake in, and benefit from, ACT.

Nonetheless, although we posit that ACT may have application across FND sub-groups, given the breadth of heterogeneity in FND other psychological treatments might be most suitable for certain presentations (Agrawal, Gaynor, Lomax, & Mula, 2014). Also, some with FND may not require or want psychological therapy.

Methodological limitations

Using pre- post- intervention data collected from clinic we calculated RCI and CSC scores.

This analysis method allowed us to control for measurement error as an explanation for treatment effects. However, as we did not use an experimental design we could not exclude further threats to the validity of our findings (e.g. regression to mean, placebo etc.; Smith, 2012). This, alongside several other factors (e.g. therapist-condition confound, lack of ACT fidelity measurement) means that further testing is required before ACT might be considered an unequivocally effective psychological approach to FND. Due to the pragmatic nature of this study we did not collect information to further characterise our cohort (e.g. demographics, proportion offered ACT, participant beliefs about the cause of FND etc.) Also, our sample may have been selective because participants had to progress through several assessment stages before they could partake in treatment. Finally, the exclusive reliance on standardised self-report questionnaires is a limitation. Although some might argue that clinician ratings of change in outcomes would have been a useful addition to the assessment battery, we suggest that the addition of behavioural data (e.g. idiographic measurement of the frequency of enactment of behaviours consistent with values) may be more informative, and this would also be consistent with the treatment model.

Conclusion

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This case series advances ACT as a promising psychological intervention for reducing symptom interference and improving mood in FND. Larger controlled studies are now required to further test the hypothesis that ACT is an efficacious treatment for this group, while more comprehensive measurement is required to investigate whether improvement occurs via psychological flexibility, as implied by the treatment model.

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Table 1

Some examples of ACT methods that were used, arranged by the targeted aspect of psychological flexibility (informed by a range of sources, mostly Harris, 2009; Hayes, Strosahl, & Wilson, 1999; Villatte et al., 2015; Wilson & DuFrene, 2009).

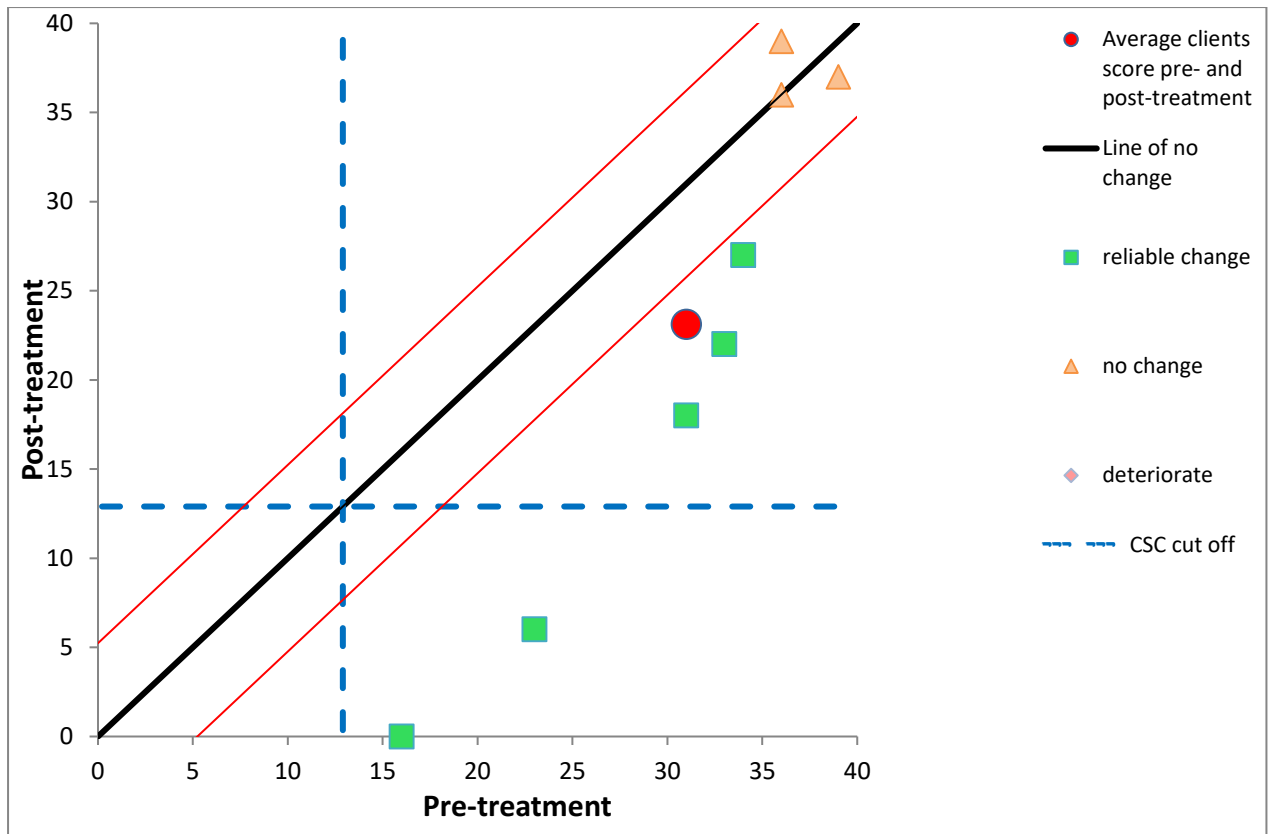
| Aspect of psychological flexibility | Therapy Methods |
|--|---|
| <p>Engagement</p> <p>Consciously aligning <i>one's</i> behaviour with <i>one's</i> values</p> | <p><u>Example functional analysis*</u> Behaviour: Avoidance of an important social event. Antecedent: Invitation to a social situation, accompanied by fear and thoughts regarding others seeing self as ill or infirmed; Consequent: less anxiety, fewer thoughts of self as ill (also boredom and lack of progress on one's values); Function: experiential avoidance of anxiety and thoughts regarding changed identity; Contingency: negatively reinforced via reduction in anxiety.</p> <p><u>Clinical conversation</u> "When you feel most alive, what are you doing, or not doing?" "Did you notice whether that was a step towards, or away from, what's important to you?" "What would you want your life to look like five years from now...with FND?"</p> <p><u>Experiential Tasks</u></p> <ul style="list-style-type: none"> - 90th birthday party: Asking participants to bring to mind their 90th birthday and to consider what they would want to be said about how they had lived their life. - Values and action discrepancy: writing down/exploring the importance of different values and how often participants are choosing behaviours that are consistent with these. - Smallest possible step: Identifying and consistently enacting the smallest possible steps (activities) that can be done now to get back on track with one's values. |
| <p>Openness</p> <p>Conscious and open contact with thoughts and feelings (emotions and bodily sensations)</p> | <p><u>Example functional analysis*</u> Behaviour: Worry/rumination on cause of FND. Antecedent: Noticing a worsening of a symptom; Consequent: excessive attention towards symptoms (to the exclusion of other meaningful aspects of life); Function: Gain certainty/truth regarding cause/prognosis of symptoms; Contingency: Historically, problem-solving negatively reinforced in reducing discomfort (e.g. relationships, work etc) and positively reinforced via achievement (e.g. work, school, from parents.)</p> <p><u>Clinical conversation</u> "...it sounds like those painful thoughts and feelings are around a lot of the time - almost like living with an annoying flatmate?" "...and when those painful memories come to mind, what happens next? What do you do when they are present?"</p> <p><u>Experiential Tasks</u></p> <ul style="list-style-type: none"> - Workability exercise: noticing where attempts to control, or ameliorate painful thoughts, feelings and sensations, are themselves interfering with values-consistent behaviour. - Practicing slowing down and attending to painful thoughts and feelings as they arise within the session, within the relationship with the therapist. |

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| | |
|---|---|
| <p>Awareness</p> <p>Appreciation of what the present situation affords</p> | <p><u>Example functional analysis*</u></p> <p>Behaviour: Dissociation.</p> <p>Antecedent: encountering stimuli associated with stressful early experience; Consequent: less present in/less attentive to the present; Function: Learned protective behaviours, experiential avoidance of present unwanted memories and feelings; Contingency: negatively reinforced as method for avoiding re-experiencing difficult memories and feelings.</p> <p><u>Clinical conversation</u></p> <p>“It sounds like you are saying that your thoughts about yourself change quite often, almost like the weather...what’s the weather like now?”</p> <p>“...do you notice what feelings/thoughts/possibilities are here right now...?”</p> <p>“If an older you – say you in your eighties – walked into this room, what would he see?”</p> <p><u>Experiential Tasks</u></p> <ul style="list-style-type: none"> - Notice who is noticing: a mindfulness task that involves noticing that one can observe thoughts and feelings. - Mindfulness of the present moment: practicing attending to a range of experiences in the present moment. - Perspective taking: using deictic framing to view a clinically significant event from differing perspectives (person, place, and time). |
|---|---|

*Note. Behaviours such as those described in these examples can have a number of different functions, dependent on person and context. Thus, these behaviours do not always have the functions described in these examples, and none of the described methods are implicitly or exclusively ‘maladaptive’.

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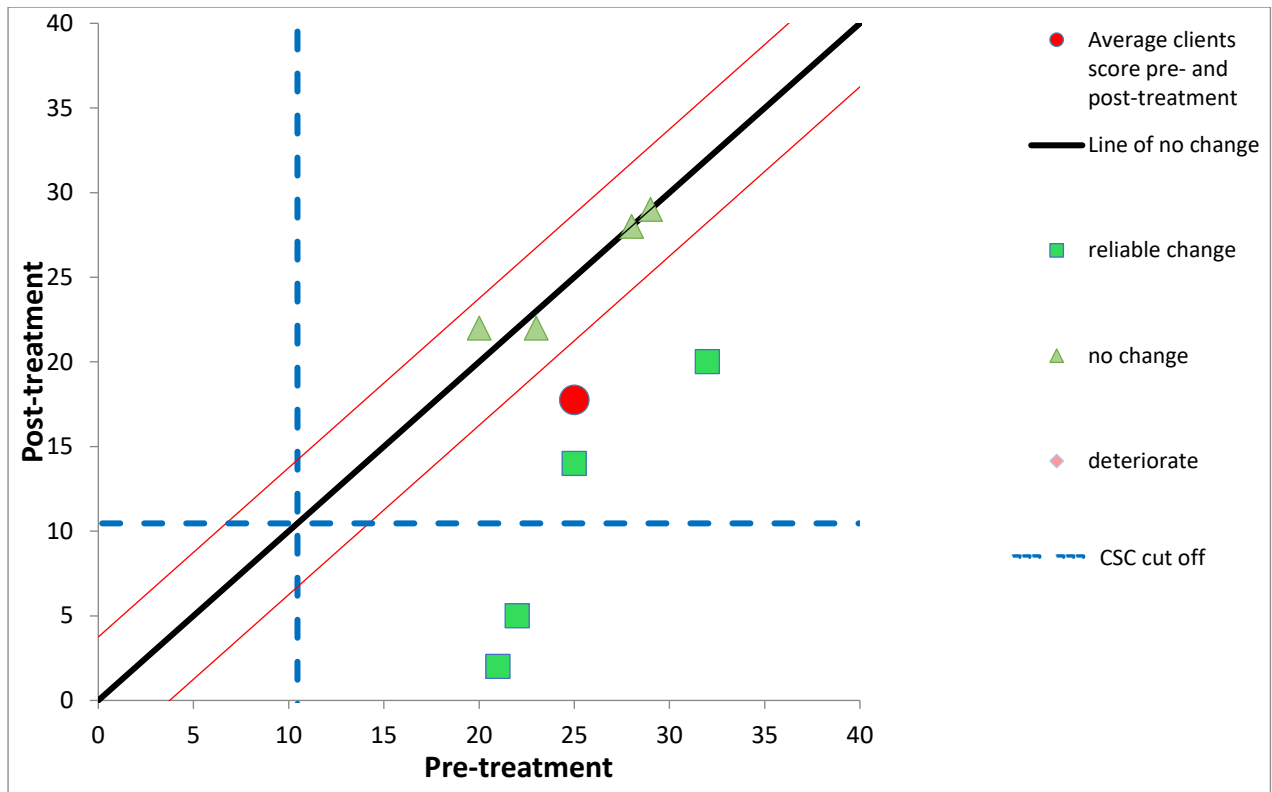


^a WSAS requires the presence of a condition, thus normative data not applicable; consequently, based on the guidance of Morley & Dowzer (2014), Criterion A was used to indicate clinically significant change.

Figure 1

Graph illustrating changes in symptom interference (WSAS) across the period of the intervention^a

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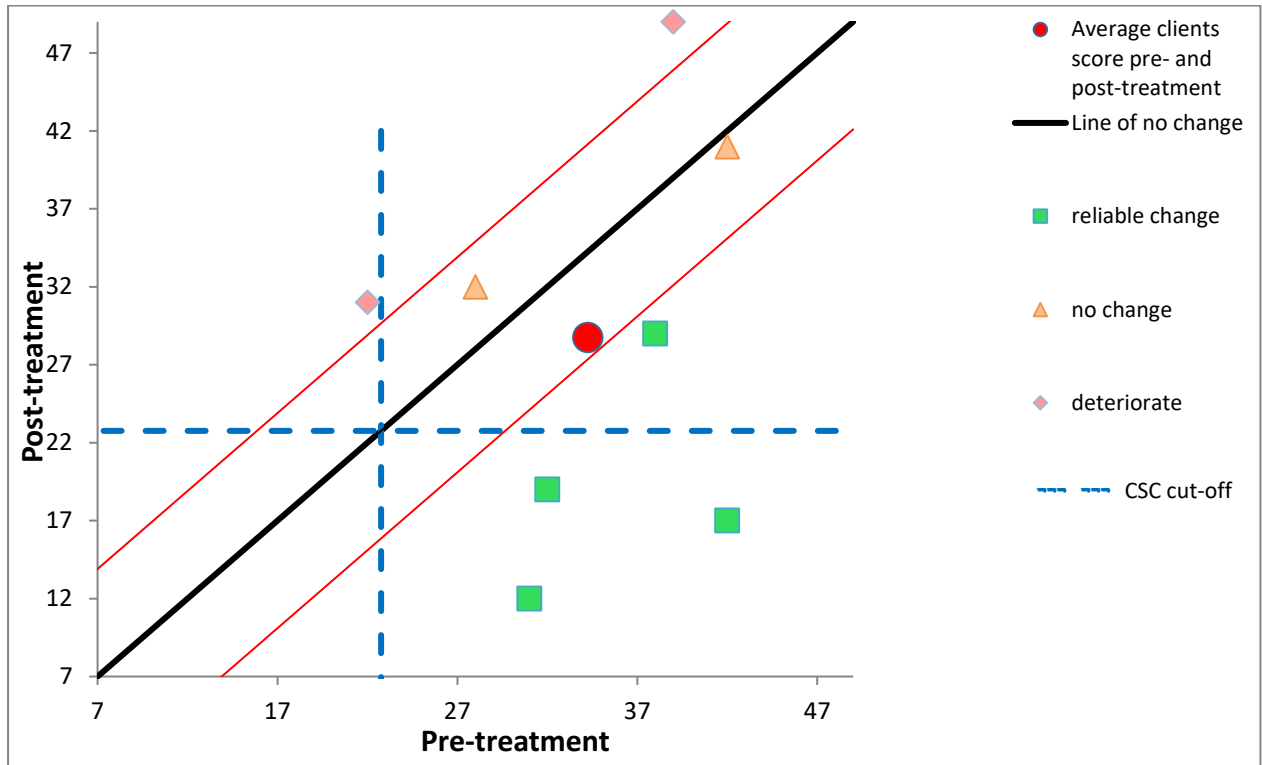
^b Cronbach's alpha, clinical and non-clinical norms from Connell & Barkham (2007; $\alpha = 0.90$; clinical norms: $M = 19.70$, $SD = 7.70$; non-clinical norms: $M = 4.70$, $SD = 4.80$)

^cAlthough Criteria B suggested by Morley & Dowzer (2014), Criterion C used as it is a more conservative value for clinically significant change, and commensurate with cut-offs suggested by Connell & Barkham (2007)

Figure 2

Graph illustrating changes in mood (CORE-10) across the period of the intervention^{b,c}

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Criterion C

^dCronbach's alpha, clinical and non-clinical norms from Bond et al., (2011; $\alpha = 0.88$; clinical norms: $M = 28.34$, $SD = 9.92$; non-clinical norms: $M = 18.53$, $SD = 7.52$).

^eCommensurate with the advice of Morley & Dowzer (2014), Criterion C used to identify clinically significant change.

Figure 3

Graph illustrating changes in psychological flexibility (AAQ-II) across the period of the intervention^{d,e}