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Psychological Therapy for Medically Unexplained Symptoms

by

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Thesis for the degree of Doctor of Clinical Psychology

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ABSTRACT

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PSYCHOLOGICAL THERAPY FOR MEDICALLY UNEXPLAINED SYMPTOMS

Catharina S Haywood, MA (Hons)

The first chapter of this thesis is a systematic review of the literature into factors that influence outcomes for people with medically unexplained symptoms (MUS), treated with cognitive behavioural therapy (CBT). A total of 14 papers were identified that met the inclusion criteria and due to the heterogeneity of results, a narrative synthesis was employed. In terms of participant characteristics, neither age, gender, comorbid depression, comorbid anxiety, or comorbid personality disorder were related to therapeutic outcomes. A greater number and intensity of symptoms were related to better outcomes. Some weak evidence was found for the impact of familial status and social difficulties, however only a limited number of articles examined those variables. In terms of therapeutic factors, individual CBT was identified as having slightly superior outcomes over group CBT and certain cognitive and behavioural factors were also identified as relevant. The findings were less clear about whether clinician profession or the number of sessions offered predict better or worse outcomes. Future research should continue to identify relevant variables involved in CBT's effectiveness in MUS and, where possible, manipulate variables experimentally.

The second chapter of this thesis is an empirical research paper examining the effectiveness of CBT and cognitive analytic therapy (CAT) at improving psychosocial, physical health, and economic outcomes for people with MUS. The paper also sought to consider whether any participant or illness characteristics predicted outcomes after treatment. CBT (N = 31) and CAT (N = 17) were no different across any of the outcomes and they were therefore grouped together as Cognitive Therapy (N = 48). A treatment as usual (TAU, N = 56) group did not receive psychological treatment and were used in analyses of economic outcomes. The Cognitive Therapy group improved significantly across all measures of psychosocial and physical health outcomes at the end of therapy compared to at the start. Neither gender, ethnicity, number of symptoms, or duration of symptoms had any impact on the improvements seen. There was no reduction in inpatient, outpatient or A&E cost at either 6, 12, or 18 months after treatment ended and between those who received cognitive therapy and those who received treatment as usual. Clinical implications and avenues for future research are discussed.

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Declaration of Authorship

I, Catharina Haywood, declare that this thesis and the work presented in it are my own and has been generated by me as the result of my own original research.

Psychological Therapy for Medically Unexplained Symptoms

I confirm that:

This work was done wholly or mainly while in candidature for a research degree at this University;

Where any part of this thesis has previously been submitted for a degree or any other qualification at this University or any other institution, this has been clearly stated;

Where I have consulted the published work of others, this is always clearly attributed;

Where I have quoted from the work of others, the source is always given. With the exception of such quotations, this thesis is entirely my own work;

I have acknowledged all main sources of help;

Where the thesis is based on work done by myself jointly with others, I have made clear exactly what was done by others and what I have contributed myself;

None of this work has been published before submission

Signed: ……………………………………………………………………

Date: ……………………………………………………………………

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Definitions and Abbreviations

MUS	Medically Unexplained Symptoms
SFD	Somatoform Disorder
PD	Personality Disorder
MDD	Major Depressive Disorder
CBT	Cognitive Behavioural Therapy
CAT	Cognitive Analytic Therapy
PMR	Progressive muscle relaxation
PCI	Psychiatric consultation
WL	Waitlist control
TAU	Treatment as usual
RCT	Randomized controlled trial
BME	Black and minority ethnic
GP	General Practitioner
DSM	Diagnostic and Statistical Manual of Mental Disorders
ICD	International Classification of Diseases
QATQS	Quality Assessment Tool for Quantitative Studies
PHQ-9	Patient Health Questionnaire – 9
GAD-7	Generalised Anxiety Disorder Questionnaire
WSAS	Work and Social Adjustment Scale
SHAI	Short Health Anxiety Inventory
CGI-SD	Clinical Global Impression Scale for Somatization Disorder
MOS SF-36	Medical Outcomes Study Short Form Health Survey
VAS	Visual analogue scale
PSC	Physical Symptoms Checklist
HADS	Hospital Anxiety and Depression Scale
IAS	Illness Attitudes Scale
SOMS-7	Screening for Somatoform Symptoms–7
EQ-5D	EuroQol Five Dimensions Questionnaire.
SCL-90-R	Symptom Checklist
FML	Psychotherapy motivation questionnaire
PHQ-15	Patient Health Questionnaire Somatic Symptom Severity Scale
WHODAS II	WHO Disability Assessment Schedule II
WI	Whiteley Index
CABAH	Cognitions about Body and Health Questionnaire
BDI	Beck Depression Inventory

DAQ	Dysfunctional Analysis Questionnaire
SPQ	Social Problem Questionnaire
IBQ	Illness Behaviour Questionnaire
SDI	Sleep Disturbance Index
BSI	Brief Symptom Inventory
KKG-I	Multidimensional Self-Report Questionnaire
SIP	Sickness Impact Profile
SF-MPQ	Short form McGill Pain Questionnaire
PCS	Pain Catastrophizing Scale
SF-36	Short Form Health Survey
STAI	State Trait Anxiety Inventory
VKP	Dutch questionnaire based on International Personality Disorder Examination

Chapter 1: Literature review into factors influencing cognitive behaviour therapy outcomes for people with medically unexplained symptoms

1.1 Introduction

1.1.1 Background

Medically Unexplained Symptoms (MUS) are physical symptoms for which no adequate medical explanation has been found, despite medical investigations having been carried out (van Dessel et al., 2014). Distinct types of unexplained symptoms can be found in many medical specialities, for instance tinnitus in ENT, non-epileptic seizures in neurology, or irritable bladder syndrome in urology, to name a few. MUS have been found to be wide spread in primary care, with between 15% and 30% of consultations being taken up by unexplained symptoms (Kirmayer, Groleau, Looper, & Dominicé Dao, 2004). While MUS is a general term, a confusing array of other labels are also used, sometimes interchangeably across the literature. For instance, conversion disorder and functional symptoms frequently appear in studies describing MUS. Moreover, MUS are often confused with the related concept of hypochondriasis (health anxiety), where an individual is preoccupied by the worry that they might have an illness, in the absence of any symptoms (Diagnostic and Statistical Manual of Mental Disorders 5th edition, DSM-5, American Psychiatric Association, APA, 2013).

In order for full somatisation disorder to be diagnosed according to both the International Classification of Diseases-10 (ICD-10, World Health Organisation, WHO, 2016) and the DSM-4 (4th edition, APA, 2000), multiple, recurrent and frequently changing unexplained physical symptoms with a history spanning several years need to be present. In contrast, the latest DSM-5 (APA, 2013) is more inclusive and requires at least one distressing somatic symptom (which no longer needs to be medically unexplained) to have been present for at least 6 months, before the new somatic symptom disorder may be diagnosed. Regardless of labelling, these symptoms are known to cause a great deal of distress and occupational impairment (Katon et al., 1991) and are associated with higher use of healthcare services (Konnopka et al., 2012). Finding acceptable and effective psychological therapies for this clinical group is therefore of great importance.

1.1.1.1 Evidence base for psychological therapies in MUS and somatoform disorders. Several reviews and meta-analyses of the existing literature have been carried out to establish the efficacy of various psychological therapies for MUS and its related conditions. The reviews highlight that CBT is studied the most and appears to be better at improving outcomes compared to waiting list controls and, to a lesser degree, compared to some other psychological therapies, however effect sizes are small and it has not always been possible to comment on the superiority of one therapy over others. For instance, a recent Cochrane review by van Dessel et al. (2014) found that CBT was better than usual care or waiting list conditions, but not enhanced care, at reducing somatic symptoms and that these improvements were maintained one year later. In a 2007 review of randomised controlled trials, Kroenke focused on somatoform disorders treated with CBT, anti-depressants, and another therapy (such as non-CBT psychotherapy or hypnosis). CBT was again studied the most, in 13 of the 34 studies, and was seen to be the most promising treatment of those investigated. Specifically, it was effective at improving at least one outcome in 11 of the 13 studies. In both cases however, there were comparatively smaller numbers of non-CBT studies with which to compare CBT. It is noteworthy also that the different reviews to date have included different diagnostic labels, with some (such as Kroenke, 2007), additionally including body dysmorphic disorder, hypochondriasis, and conversion disorder, while others (such as van Dessel et al., 2014) have excluded these. Reviews into specific syndromes, such as irritable bowel syndrome (IBS) or chronic fatigue syndrome (CFS) have demonstrated more convincing effects for CBT (see for instance Allen, Escobar, Lehrer, Gara, and Woolfolk, 2002 who studied both IBS and CFS). Indeed, while there is no guidance from the National Institute for Health and Care Excellence (NICE) on psychological interventions for MUS or somatoform disorders, it does recommend CBT or hypnotherapy for IBS (NICE, 2017) and CBT and/ or graded exercise therapy for CFS (NICE, 2007).

So far it has been shown that the most promising, if yet under-researched, psychological therapy for MUS and its related somatoform disorders appears to be CBT. The cognitive behavioural model as applied to this population (Salkovskis & Warwick, 1986) suggests that unexplained symptoms emerge when an individual pays attention to physiological sensations, such as breathlessness, and interprets their meaning in a catastrophic way. The worry about these sensations is then perpetuated as the person avoids situations which might bring about the sensations, such as climbing the stairs or doing exercise, but which in turn leads to a decrease in overall physical fitness and an increased likelihood of breathlessness after exertion. Salkovskis et al.’s (2016) adaptation of the model additionally emphasise the importance of trans-diagnostic factors including psychological inflexibility and clinical perfectionism as further maintaining factors for a person’s difficulties. The aims of CBT for this population are therefore to change a person’s cognitions and unhelpful behaviours towards their symptoms.

1.1.1.2 Variables involved in therapeutic outcomes. While CBT appears to be effective with MUS and its related conditions, it is important to consider possible mechanisms and mediators for change, so that treatment programmes may be better-targeted. There is some evidence for instance that CBT may be more effective at improving quality of life in patients with more severe and long-standing IBS symptoms (Lackner et al., 2007). Certain cognitive factors have also been seen to influence outcomes after CBT for functional somatic syndromes. For instance, Pedersen, Frostholm, Søndergaard Jensen, Ørnbøl, and Schröder (2016) demonstrated that reduced symptom catastrophizing during group CBT partially mediated the long-term treatment effects, while Christensen, Frostholm, Ørnbøl, and Schröder (2015) found improved illness perceptions during treatment to partially mediate CBT’s effect on physical health one year later. There may also be certain participant variables that could influence how well a person responds to CBT for MUS, with Cuijpers et al. (2014) finding for instance that gender did not influence whether a person responded better to CBT than to medication. It is equally important to consider whether any therapeutic factors influence CBT outcomes. For example, in depression it has been found that group and individual CBT are equally effective (Lockwood, Page, & Conroy-Hiller, 2004), while there appears to be a trend for better outcomes after individual CBT in panic and agoraphobia (Sharp, Power, & Swanson, 2004).

1.1.1.3 The gap in the literature and objectives of the current review. To the author’s knowledge no comprehensive review has yet been carried out into participant characteristics or therapeutic factors that might be involved in CBT’s success with this specific client group. Identifying mediating, moderating or predictive factors of success would enable CBT to be delivered in a way that may improve the outcomes seen so far, with clear clinical implications for clients and services. At the same time, it would enable future researchers to draw firmer conclusions about CBT’s efficacy in MUS, thus bringing the evidence base in line with that for specific syndromes such as IBS and CFS (see for instance Allen et al., 2002), as well as for other mental health conditions such as anxiety disorders (see for instance the comprehensive review by Hofmann, Asnaani, Vonk, Sawyer, and Fang, 2012). Objectives of the present review are therefore to review the evidence into participant variables found to have a mediating, moderating or predictive involvement in CBT’s success in MUS, (abridged) somatisation disorder, undifferentiated somatoform disorder, somatoform pain disorder, multisomatoform disorder, or somatic symptom disorder. The review also aims to explore the role of specific therapeutic factors on outcomes (number of sessions received, mode of delivery i.e. individual or group, profession of clinician delivering the therapy, any cognitive or behavioural factors on the part of participants).

1.2 Method

1.2.1 Inclusion and exclusion criteria

1.2.1.1 Types of studies. All outcomes studies were considered for inclusion in this review. These could be pre- post-treatment studies or longitudinal designs. Randomisation was not a requirement due to the low number of high-quality papers in this generally under-researched area. Qualitative designs or single case studies were excluded in order to be able to comment on effect sizes. Due to the author’s bilingualism, papers could be available either in English or in German and mediators, predictors, moderators, process or mechanisms needed to be included.

1.2.1.2 Participant characteristics. Minimum age was 18, while no maximum age was applied and there were no limits applied to gender or country of publication.

1.2.1.3 Diagnoses. In order to capture the current as well as the older literature, participants were included if they had a diagnosed somatisation disorder, an undifferentiated somatoform disorder, or somatoform pain disorder according to the DSM-3 (APA, 1980), DSM-4 (APA, 2000), ICD-9 (WHO, 1975), or ICD-10 (WHO, 2016). Participants were also included if they had abridged somatisation disorder, multisomatoform disorder, or chronic MUS (at least 6 months in duration). Participants were included if they had the new DSM-5 (APA, 2013) diagnosis of somatic symptom disorder, although no papers were found using this label. The related concept of hypochondriasis (health anxiety) was excluded from the present review as somatic symptoms are not usually present in this diagnosis. Similarly, specific syndromes such as fibromyalgia or chronic fatigue were excluded as a large body of literature already exists around these syndromes, including relatively recent reviews (such as Bernardy, Klose, Busch, Choy, and Häuser, 2013 or Price, Mitchell, Tidy, and Hunot, 2008). Finally, comorbidities such as personality disorders or anxiety and depression were included in the review, however the somatoform disorder had to be the primary diagnosis.

1.2.1.4 Types of interventions. Eligible studies had to investigate the effects of CBT, while variations such as cognitive therapy, relaxation therapy, biofeedback therapy, psychoeducation, and mindfulness-based cognitive therapy were excluded. Including these variations would have made it difficult to comment on CBT's impact on MUS, as the interventions would have looked different across studies. Due to the limited amount of research in this area, no restrictions were placed on the kind of control intervention, if any, that was used. Similarly, no restrictions were placed on the duration of the treatment, whom it was delivered by, and in what setting.

1.2.2 Search methods

The literature was searched in a way that allowed participant variables to emerge naturally, as opposed to exclusively searching for the most obvious variables, such as gender or the presence of comorbidity.

1.2.1.2 Electronic searches. Online databases searched were EBSCOHost (covering CINAHL, Medline, and Psych Info), ISI Web of Knowledge, and PubMed. No restrictions were placed on publication dates, however searches were restricted to academic articles and excluded conference proceedings and theses. All searches were completed on 02/12/2016.

1.2.1.3 Search terms. The following Boolean/ phrase free text terms were used: (CBT OR cognitive behav*) in all text or topic, AND (mechanism* OR mediat* OR moderat* OR process* OR predict*) in all text or topic, AND (unexplained OR MUS or MUPS OR somat*) in title.

1.2.1.4 Hand-searching. The reference lists of recent meta-analyses as well as the publication lists of relevant authors were searched for additional articles and were included in the initial number of records screened.

1.2.3 Risk of bias across studies

The grey literature was not searched as part of this literature review. Therefore, it is possible that a publication bias exists in the papers selected as any unpublished articles, which may have had non-significant results, would have been excluded.

1.2.4 Study selection

After the initial database search using the aforementioned search terms, duplicates across the different databases were removed and a final hand-search was completed. Titles and abstracts were then screened by one reviewer to exclude articles that were entirely unrelated to the topic. Those remaining were then assessed in their full-text by the same reviewer, using the criteria described earlier. Any articles which did not meet the criteria, for instance if an adolescent population had been used, were then excluded. Only those articles which fully met the criteria outlined above were included in the final synthesis.

1.2.5 Data extraction

Relevant information that was extracted about the characteristics and outcomes of each study can be found in table 1. These were the article title, author(s) and year, participants and sample size(s), design, details of the CBT intervention (i.e. individual or group, number of sessions received, profession of clinician delivering the treatment), relevant outcome measures and outcomes, effect sizes, main points of critique, and the quality rating. Quality assessments of individual papers, including risk of bias, were completed using the Quality Assessment Tool for Quantitative Studies (QATQS, Thomas, Ciliska, Dobbins, & Micucci, 2004) and was completed at the same time as the data extraction process. Additionally, effect sizes were calculated for those articles that did not report them.

1.2.6 Synthesis of results

The study findings (together with an examination of study quality and effect sizes) were arranged according to two overarching themes; participant variables involved in therapeutic outcomes and process variables involved in outcomes. As stated earlier, identifying potential factors of success would allow CBT to be delivered in a way that might improve the outcomes seen so far in the literature. A narrative synthesis was carried out due to the heterogeneity of the results. Participant variables included gender, age, familial status, education, being in receipt of disability benefits, social difficulties, number, severity, chronicity of symptoms, and psychiatric comorbidity. Process variables were the number of sessions received, whether it was individual or group therapy, clinician profession, and any additional cognitive or behavioural factors on the part of participants.

1.3 Results

As can be seen from figure 1, a total of 657 papers were obtained by searching databases using the aforementioned search terms (yielding 624 papers) and through hand searching (yielding 33 papers), reducing to 492 after database duplicates were removed. 358 titles and abstracts were excluded for being irrelevant to the topic, leaving 134 articles to be considered in full text. Of these, 120 were excluded for reasons ranging from using the wrong intervention to being unavailable in either English or German. Finally, 14 articles were selected for the present review and their summaries are presented in table 1. A narrative synthesis was carried out, due to the heterogeneity of the findings.

Figure 1.1. Flowchart of database search results for systematic literature review

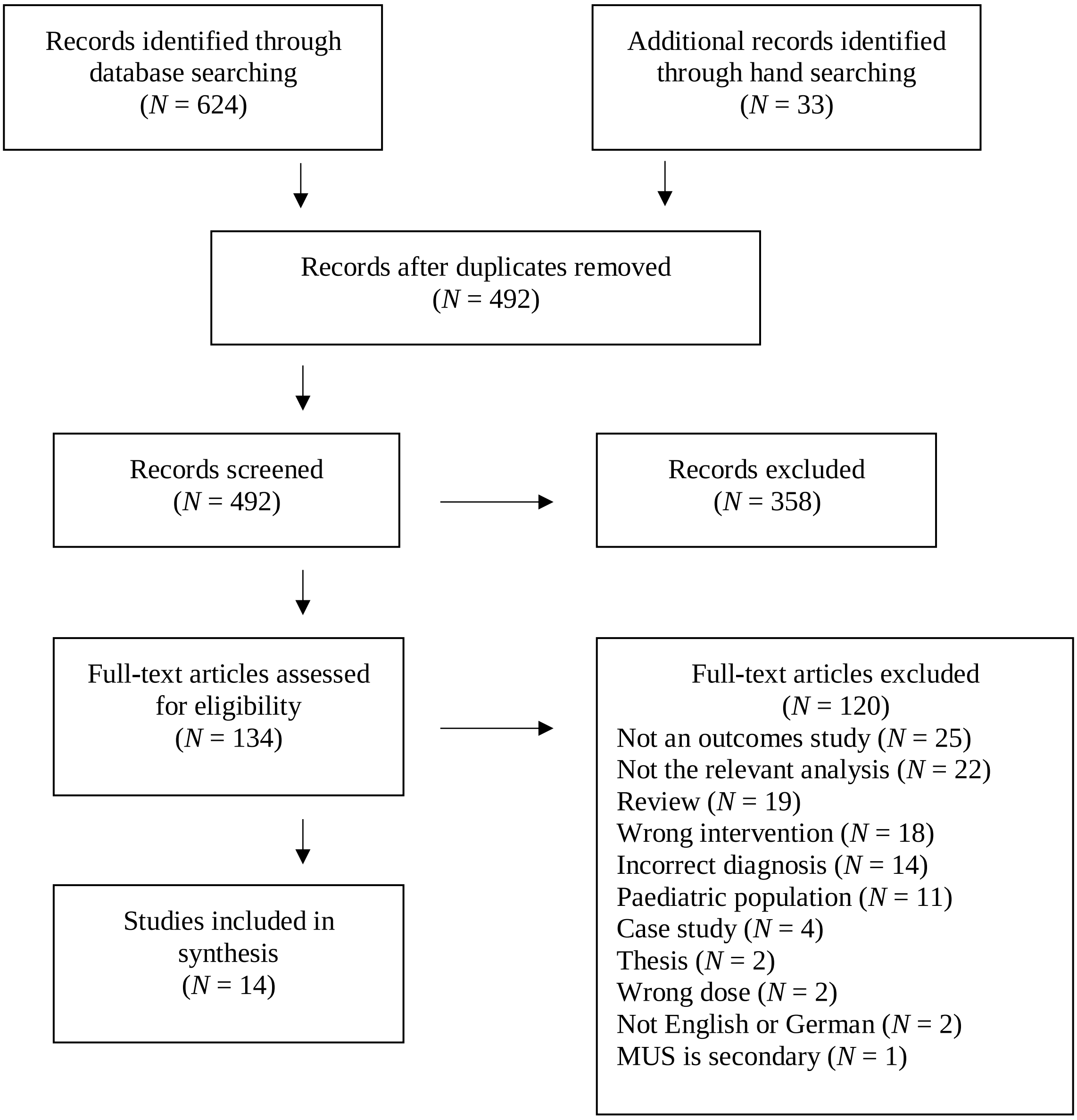


Table 1.1.

Main characteristics and findings from the fourteen articles included in the literature review

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Allen, Woolfolk, Escobar, Gara, & Hamer (2006)	Diagnosis of somatisation disorder CBT plus PCI $N = 43$ PCI $N = 41$	Longitudinal, randomized controlled trial	Average 10 individual CBT sessions delivered by psychological therapists	CGI-SD Physical functioning subscale of MOS SF-36 Somatic Symptom Diary Severity of somatic symptoms on VAS	Somatic symptom severity significantly reduced 15 months after baseline in CBT+PCI group vs psychiatric consultation alone ($p < 0.001$) On CGI-SD, CBT more likely to be rated by clinician as ‘much/ very much improved’ than only PCI at all time points ($p = 0.002$ at all times) At 15 months continued to be associated with greater improvement in self-reported physical functioning ($p = 0.02$) and greater decrease in health costs ($p = 0.01$)	$d = 0.661$ At 3months $d = 1.167$, 9 months $d = 1.35$, 15 months $d = 1.4$ Physical functioning $d = 0.336$, self-report severity $d = 0.412$	+ Good design + Low attrition rates - Self-referral to study	Moderate A Weak B Strong C Mod D Mod E Strong F Strong

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Arnold, De Waal, Eekhof, Assendelft, Spinhoven, & Van Hemert (2009)	Diagnosis of undifferentiated somatoform disorder TAU plus CBT *N* = 31 TAU *N* = 34	Longitudinal, clinical controlled study	Average 5 individual CBT sessions delivered by GPs	Symptom severity and self-rated improvement on VAS PSC HADS MOS SF-36 IAS Healthcare utilization from GP records and self-report	CBT plus care as usual no significantly better outcomes on any measure. In both groups number of symptoms improved significantly ($p <$ 0.05) Number of sessions not associated with improvements in symptoms 'Treatment by a mental health professional' was not associated with the improved outcomes	Data not available	+ Control of possible confounding variables and bias - High drop-out rates	Strong A Mod B Mod C Strong D Mod E Strong F Mod

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Bleichhardt, Timmer, & Rief (2005)	Diagnosis of somatoform disorder Responders (previously improved by at least 1/3 of a standard deviation) $N = 79$ Non-Responders (who previously did not improve) $N = 83$	Cohort analytic study	Individual and group CBT delivered by mental health staff	SOMS-7 EQ-5D SCL-90R HADS German version FMP	Responders significantly fewer symptoms at end ($p = 0.002$) and one year later ($p = 0.004$) compared to non-responders. One year later intensity also significantly reduced for responders ($p = 0.017$) Positive expectations of improvement predicted short- and long-term reduction of no. of symptoms ($p = 0.004$ and $p = 0.025$). Claiming for disability benefits predicted poorer outcomes in short-term Less subjective suffering at end predicted long-term improvement to health-related quality of life ($p < 0.001$) More motivation correlated with reduction in no. of symptoms in short- ($p = 0.01$) and long-term ($p = 0.005$)	Short-term symptom number $d = -0.477$, long-term symptom number $d = -0.452$, long-term symptom intensity $d = -0.369$ Short-term expectations $d = -0.453$, long-term $d = 0.367$ Disability benefits short-term somewhere between 0.34 $r = -0.43$ $r = 0.20$ short-term, $r = 0.23$ long-term	+ Regression analyses indicate impact of therapy - Additional therapies given e.g. PMR - Poor generalisability to outpatient settings	Strong A Strong B Mod C Strong D Mod E Strong F Strong

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Haggarty, O'Connor, Mozzon, & Bailey (2016)	Diagnosis of somatisation High somatising *N* = 138 Low somatising *N* = 404	Cohort analytic study	Average 7.3 individual CBT sessions delivered by mental health staff	PHQ-15 WHODAS II	PHQ15: Significant improvement in symptom severity by 41.3% for high somatising ($p < 0.0001$) and of 39.6% for low somatising ($p < 0.0001$) (both compared to general population) WHODASii: Significant decrease by 44.1% in disability for mental illness ($p < 0.0001$) among high somatising group and a 51.2% decrease ($p < 0.0001$) among low somatising group	$d = 1.8$ and $d = 0.98$ $d = 0.92$ and $d = 0.77$	+ Controlled for selection bias +Design allows for greater confidence in cause and effect of therapeutic impact + Good generalizability - Fidelity to treatment may have been varied - No follow-up - No control group although compared to community norms	Weak A Strong B Mod C Weak D Mod E Strong F Weak

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Heider & Zaby (2011)	Diagnosis of undifferentiated somatoform disorder, somatisation disorder, pain disorder, and conversion disorder CBT *N*=106	Prospective, longitudinal cohort study	8 sessions of group CBT delivered by mental health staff	HADS-D SOMS-7	Higher initial number and intensity of somatoform disorder predicted better therapeutic outcome (reduction of symptoms), in short- (number and intensity $p < 0 .05$) and long-term (number and intensity $p < 0.0001$) Neither comorbid anxiety or depression predicted the reduction in symptoms	Short-term reduction in symptom number $r = 0.24$, long-term $r = 0.39$ Short-term reduction in intensity $r = 0.39$, long-term $r = 0.45$	+ Multiple regressions add strength to correlations - No control/ WL group - Lacks generalisability to other settings - Variability in treatment programme - Short follow-up	Strong A Mod B Mod C Strong D Mod E Strong F Strong

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Hiller, Fichter, & Rief (2003)	Diagnosis of somatoform disorder CBT $N = 172$ Clinical comparison (other DSM-IV diagnoses) $N = 123$ Waitlist control $N = 295$	Longitudinal controlled treatment study	15-20 hours of weekly individual and group CBT (mean inpatient period 58.6 days) delivered by mental health staff	SOMS WI CABAH BDI DAQ Medical and billing records for healthcare use	Somatoform and clinical comparison group improved significantly in physical symptom distress, health anxiety, dysfunctional beliefs about body and health, depression, and psychosocial impairment (all $p < 0.01$) in short-term but not at follow-up	Depressive symptoms in somatoform group: $d = 0.86$, psychosocial impairment: $d = 0.68$ somatic symptoms $d = 0.49$, hypochondriasis $d = 0.58$, dysfunctional attitudes $d = 0.30$	+ Good use of waitlist control to reduce individual differences + Objective measure of healthcare use - CBT not adapted to MUS - Additional interventions given e.g. PMR	Moderate A Strong B Mod C Weak D Mod E Strong F Strong
					Waitlist period showed no change apart from on BDI ($p < 0.01$)	$d = 0.10$		
					Somatoform and other mental disorders had significant reductions in outpatient and inpatient cost (analysis not given) and reduction in sick leave cases among the somatoform group ($p < 0.05$)	Data not available		

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Leibbrand, Hiller, & Fichter (1999a)	Diagnosis of somatoform disorder with anxiety and MDD *N* = 18 With PD *N* = 17 With MDD, anxiety and any PD *N* = 39 Non-somatoform control *N* = 10	Longitudinal case control	Duration of inpatient treatment from 60.1 days to 66.6. Individual and group CBT delivered by mental health staff	WI SCL-90R DAQ CABAH	Significant improvement for all 5 groups across all measures ($p < 0.01$), no significant differences in outcomes between somatoform patients with comorbid Axis 1 and those with PD Combining comorbidity of MDD and ANX with comorbidity of axis II, no significantly worse outcomes than for somatoform patients with axis I comorbidity or non-somatoform controls	Effect sizes were largest on 4 of 6 measures for those with most severe level of comorbidity (WI $d = 0.7$, BDI $d = 1.2$, SCL $d = 0.7$ and DAQ $d = 1.1$)	+ Good generalizability to other settings - Large variation in sample sizes across groups and unclear whether controlled for statistically - Additional interventions were offered e.g. PMR	Strong A Mod B Mod C Strong D Mod E Strong F Strong

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Leibbrand, Hiller, & Fichter (1999b)	Diagnosis of somatoform disorder without comorbid PD $N = 35$ With cluster C PD $N = 16$ With multiple cluster C PDs $N = 12$ Non-somatoform controls, no PD $N = 11$ With any comorbid PD $N = 8$ With at least one cluster A or PD $N = 13$	Longitudinal case control	Number of sessions not stated. Individual and group CBT delivered by mental health staff	SOMS-7 WI BDI CABAH	Outcomes for all groups (SFDs and controls) improved significantly for somatic symptoms ($p < 0.001$). No significant difference in improvement between SFD with/ without PDs and controls with/ without PDs Significant improvements in depression, hypochondriasis, catastrophizing, beliefs about bodily weakness, beliefs about autonomic sensations (all $p < 0.001$), and intolerance of bodily complaints ($p < 0.01$). No significant difference in these outcomes between different groups.	Overall sample $d = 0.2$ Overall sample, depression $d = 0.7$, hypochondriasis $d = 0.4$, catastrophizing, $d = 0.3$, beliefs about bodily weakness $d = 0.3$, beliefs about autonomic sensations $d = 0.4$, intolerance of bodily complaints $d = 0.2$	- Sub-classifications among two groups not comparable - Large variation in sample sizes across groups and unclear whether controlled for statistically	Moderate A Mod B Mod C Weak D Mod E Strong F Strong

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Lidbeck (1997)	Diagnosis of somatisation disorder CBT $N = 32$ Waiting list control $N = 17$	Longitudinal controlled prospective study	8 group CBT sessions delivered by GPs	SPQ IBQ HADS SDI Medication usage self-report`	Significant improvement after CBT compared to control for physical illness perception and somatic preoccupation ($p < 0.05$ post treatment and $p < 0.01$ at 6 months) Significant reduction in hypochondriasis ($p < 0.05$ at 6 months although not immediately post treatment), and medication usage $p < 0.05$ post treatment and $p < 0.05$ at 6 months	Post-treatment $r = 0.53$ and at 6 months $r = 0.42$ Hypochondriasis at 6 months $r = 0.52$, medication use post treatment $r = 0.39$, and at 6 months $r = 0.41$	+Good attendance once treatment started - Potential selection bias with large drop-outs of 25% before start - Did not assess concurrent psychiatric disorder rigorously	Strong A Mod B Strong C Strong D Mod E Strong F Strong

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Martin, Rauh, Fichter, & Rief (2007)	Diagnosis of at least two MUS in last six months CBT *N* = 70 Standard medical care control *N* =70	Longitudinal randomised clinical trial	1 group CBT session delivered by mental health staff	BSI SOMS7 WI BDI Internal control subscale of KKG-I Medication use Healthcare use	Number of healthcare visits reduced more in CBT group than control ($p < 0.05$) Medication use decreased more in CBT group ($p < 0.05$) Greater reduction in somatisation severity in CBT group ($p < 0.05$). Number of symptoms improved in both groups, but not significantly more for CBT	$\eta^2 = 0.031$ $d = 0.42$ $\eta^2 = 0.048$	+ Ethical considerations where control group was offered CBT group 6 months later - Measure of healthcare use subjective and relied on participant memory	Moderate A Mod B Strong C Strong D Mod E Weak F Strong

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Moreno, Gill, Magallón, Bauzá, Roca, del Hoyo, & Garcia-Campayo (2013)	Diagnosis of abridged somatisation disorder Individual CBT $N = 56$ Group CBT $N = 64$ TAU $N = 48$	Longitudinal randomised controlled trial	10 individual sessions and 10 group sessions delivered by psychologists	SOMS Severity of somatic symptoms on VAS Hamilton Anxiety Rating Scale Hamilton Depression Rating Scale	Individual and group CBT better than TAU at all time points ($p < 0.001$) for both number of symptoms and severity	No. of symptoms TAU vs group CBT post treatment $d = 2.29$, 6 months $d=0.46$, 12 months $d=1.99$. TAU vs individual CBT post treatment $d = 3.44$, 6 months $d = 2.47$, 12 months $d = 2.74$ Symptom severity TAU vs group post treatment $d = 2.00$, 6 months $d = 1.29$, 12 months $d = -0.02$. TAU vs individual post treatment $d = 2.77$, 6 months $d = 2.54$, 12 months $d = 1.75$	+Direct comparison of group vs. individual +Large sample sizes and good effects - Unvalidated and unreliable depression scale - Unclear whether CBT applied in same way to group as individual	Moderate A Mod B Strong C Strong D Mod E Weak F Mod
					Individual CBT greater improvement in no. of symptoms compared to group CBT and TAU at 6 months ($p < 0.001$)	Individual CBT at 6 months $d = 2.10$		

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Speckens, Spinhoven, van Hemert, Bolk, & Hawton (1997)	Diagnosis of MUS CBT *N* = 39 Optimised care control *N* = 40	Longitudinal Randomised controlled trial	Median 11 sessions individual CBT sessions delivered by GP trained in CBT and a behavioural therapist	Self-report Likert scale for frequency and intensity of symptoms HADS SIP IAS WI	Change in hypochondriacal cognitions related to change in psychological distress (2.15[0.53-3.77]) Hypochondriacal cognitions at six months predicted level of psychological distress one year later (2.62[0.50-4.74]) Higher levels of illness behaviour at baseline predicted poorer outcomes one year later (2.15[0.22-4.09])	*d* = 1.47 *d* = 1.61 *d* = 1.47	- Questionable reliability and validity of self-report scale for symptoms - Apparent lack of comparison with the TAU group	Moderate A Strong B Strong C Strong D Mod E Weak F Strong

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Yoshino, Okamoto, Doi, Horikoshi, Oshita, Nakamura, … Yamawaki (2015)	Diagnosis of somatoform pain disorder $N = 34$	Longitudinal cohort study	12 session of individual CBT delivered by psychiatrists with CBT experience	SF-MPQ Pain intensity on self-report VAS PCS BDI-II SF-36 STAI	Pain intensity VAS ($p < 0.001$), pain catastrophizing ($p < 0.001$), trait anxiety ($p < 0.01$), depression ($p < 0.01$), bodily pain ($p < 0.001$) improved significantly after treatment compared to their own WL control period Pain intensity ($p < 0.01$), pain catastrophizing ($p < 0.01$) and depression ($p < 0.01$) remained improved one year later Greater changes in pre-post pain catastrophizing scores were correlated with greater improvements in affective pain, depression, anxiety, all $p < 0.01$	Pain intensity VAS $d = 0.94$, pain catastrophizing $d = 0.79$, trait anxiety $d = 0.36$, depression $d = 0.36$, bodily pain $d = 0.70$ Pain intensity $d = 1.07$, pain catastrophizing $d = 0.58$, depression $d = 0.36$ Affective pain $r = 0.61$, depression $r = 0.51$, anxiety $r = 0.59$	+ Use of waiting list control group, although relatively short of 30 days - Potential selection bias and questionable generalisability to rest of clinical population	Moderate A Weak B Mod C Strong D Mod E Strong F Strong

Author(s)	Sample	Design	Details of CBT intervention	Relevant measures	Main findings	Effect sizes	Main critiques	Quality rating
Zonneveld, van Rood, Kooliman, Timman, Spijker, & Busschbach (2012)	Diagnosis of undifferentiated somatoform disorder CBT *N* = 162	Longitudinal randomised controlled trial	13 sessions of group CBT, not stated who delivered it	SF-36 SCL-90R VKP	More psychological symptoms, fewer personality disorder characteristics, presence of psychiatric history, better mental health related QoL at baseline all predicted short-term CBT outcomes $p = 0.003$ and explained 15% of the variance and psychological symptoms had the strongest association	$d = 0.39$	+ Prevented phishing by focusing on predictors based on existing literature - Personality measure may not have been sensitive enough	Moderate A Mod B Strong C Weak D Mod E Strong F Mod

Note. CGI-SD = Clinical Global Impression Scale for Somatization Disorder. MOS SF-36 = Medical Outcomes Study Short Form Health Survey. VAS = visual analogue scale. PSC = Physical Symptoms Checklist. HADS = Hospital Anxiety and Depression Scale. IAS = Illness Attitudes Scale. SOMS-7 = Screening for Somatoform Symptoms–7. EQ-5D = EuroQol Five Dimensions Questionnaire.SCL-90-R = Symptom Checklist. FML = Psychotherapy motivation questionnaire. PHQ-15 = Patient Health Questionnaire Somatic Symptom Severity Scale. WHODAS II = WHO Disability Assessment Schedule II. WI = Whiteley Index. CABAH = Cognitions about Body and Health Questionnaire. BDI = Beck Depression Inventory. DAQ = Dysfunctional Analysis Questionnaire. SPQ = Social Problem Questionnaire. IBQ = Illness Behaviour Questionnaire. SDI = Sleep Disturbance Index. BSI = Brief Symptom Inventory. KKG-I = Multidimensional Self-Report Questionnaire. SIP = Sickness Impact Profile. SF-MPQ = Short form McGill Pain Questionnaire. PCS = Pain Catastrophizing Scale.SF-36 = Short Form Health Survey. STAI = State Trait Anxiety Inventory. VKP = Dutch questionnaire based on International Personality Disorder Examination.CBT = Cognitive behavioural therapy. TAU = Treatment as usual. WL = Waitlist control. PCI = Psychiatric consultation. PMR = Progressive muscle relaxation. PD = Personality disorder. MDD = Major depressive disorder. SFD = Somatoform Disorder.
+ = strengths. - = limitations
Quality rating categories A = Selection bias. B = Design. C = Confounders. D = Blinding. E = Data collection methods. F = Withdrawals and drop-outs

As table 1.1 shows, most papers were rated as of moderate quality (eight papers), while five were rated as strong and only one was rated as being weak on the QATQS scale (Thomas, Ciliska, Dobbins, & Micucci, 2004). Common strengths across papers included particularly robust data collection methods with reliable and valid measures being used, as well as high follow-up rates of 80% or greater. In contrast, common reasons for a drop in quality ratings were due to a lack of control of confounding variables, such as additional concurrent therapies, as well as a lack of blinding of participants due to the nature of the intervention. Moreover, some results will be interpreted with particular caution as although predictor variables were referred to, the analysis in some articles was only correlational, calling into question the degree of impact those variables have on outcomes.

1.3.1 Participant variables and therapeutic outcomes

1.3.1.1 Gender. Three of the five articles which explicitly assessed for an impact of gender on outcomes after CBT found that gender was not correlated with improved symptoms or psychosocial improvements either in the short- or the long-term (Heider & Zaby, 2011; Speckens, Spinhoven, van Hemert, Bolk, & Hawton, 1997; Hiller, Fichter, & Rief, 2003). The two papers that did find women to benefit more than men both yielded only small effects for the relationship (Bleichhardt, Timmer, & Rief, 2005; Haggarty, O'Connor, Mozzon, & Bailey, 2016) and both papers acknowledged a significant gender imbalance of more females at baseline. It is unclear whether this imbalance was controlled for statistically, which could account for the finding that women fared better after therapy. However, women are over-represented in the literature, as MUS and somatoform disorders are more common in females (van Dessel et al., 2014). Overall, the quality of the five papers included here was either strong or moderate, while the aforementioned paper by Haggarty et al. (2016) was rated as weak due to particularly high drop-out rates, although it is unclear whether there was a gender difference in drop-outs. While acknowledging the limited number of articles in this review which cover gender as a variable of interest, it appears that the strongest evidence points towards gender not being correlated with the positive impact that CBT has for individuals with MUS.

1.3.1.2 Age. The four papers included in this review which considered age as a variable of interest were all in agreement that age was not correlated with positive outcomes (Heider et al., 2011; Hiller et al., 2003; Martin, Rauh, Fichter, & Rief, 2007; Speckens et al., 1997), although Speckens et al. (1997) did not report any statistical analyses to support their assertion that ages were similar across both groups at the start. Martin et al. (2007) improved on the correlations above by statistically controlling for age and demonstrating that it did not change the improvements seen in somatisation severity, outpatient visits and medication use after CBT. Three of the articles were rated as being of moderate quality (Martin et al., 2007; Hiller et al., 2003; Speckens et al., 1997), while one was rated as particularly strong (Heider et al., 2011). Reductions in quality were due to poor validity and reliability of outcome measures and a lack of control of concurrent treatments being offered. As already highlighted, the present review limited its search to papers which involved participants aged 18 or over, while no upper limit was set. While some of the included articles did not state whether an upper age limit was set, there were no participants over the age of 70, while others limited their age criteria to between 64 and 70. Therefore, conclusions from the present review cannot be extended to individuals younger than 18 or over 70.

1.3.1.3 Familial status. The findings in this area are extremely limited and make it difficult to draw conclusions about whether familial status (having children or being a guardian) has an impact on CBT's efficacy for people with MUS. The only study which made mention of this found that it was not correlated with later outcomes, at least not psychosocial outcomes, at the end of treatment (Hiller et al., 2003). However, the article made no mention of whether it was correlated with an improvement in physical symptoms after treatment. Nonetheless, the article was rated as being of moderate quality, with a reduction in scores due to the possibility of a confounding influence of concurrent medical treatment.

1.3.1.4 Education. Two papers included education as a variable of interest, however the findings were conflicting. One (Heider et al., 2011) did not find education to be a significant predictor for any outcomes while Hiller et al. (2003) found that those with a lower level of education at baseline improved significantly more in psychosocial functioning than those with a higher level of education. The two papers were rated differently in terms of quality; the latter was of moderate quality due to possible confounding influences of concurrent interventions, while that by Heider et al. (2011) was particularly strong. Moreover, while Hiller et al. (2003) had a sample of 295 (more than double that of the former study) data were not available with which to calculate effect sizes, making it all the more difficult to say with confidence which findings are relevant.

1.3.1.5 Being in receipt of disability benefits. This was also poorly covered by the literature and was only explored by one of the studies in this review, although it was of strong quality (Bleichhardt et al., 2005). This study suggested that being in receipt of disability benefits predicted poorer outcomes in the number of symptoms after CBT, at least in the short-term. However, upon closer inspection of the results the relationship was in fact not significant in an earlier correlational analysis carried out by the authors. This would call into question the findings of a mediating relationship between claiming disability benefits and having worse outcomes after CBT.

1.3.1.6 Social problems. It is difficult to draw any firm conclusions about whether more social difficulties (such as housing, financial, employment, relationships) predict poorer outcomes in CBT for people with MUS, as only one study in this review explored this concept (Lidbeck, 1997). The authors found that among those who received treatment, those with greater initial severity of social problems also had significantly higher somatisation, anxiety, and depression at the end of treatment and at 6 months, compared to those with lower initial social problems. These correlations represent large effects, although it is unclear whether the authors controlled for baseline somatisation, anxiety, and depression. Moreover, while the paper was rated as strong in quality and used a validated and reasonably reliable measure (Corney & Clare, 1985; Corney, 1988) to assess the concept, it is arguably highly subjective and may not accurately reflect a person’s degree of social difficulties. Significantly, the study also suffered large early withdrawal rates before treatment, although specific reasons for these were not stated.

1.3.1.7 Number/ severity/ chronicity of symptoms. Two of the three papers which examined these variables strongly suggest that the greater the number or intensity of a person’s unexplained symptoms, the better the outcomes to symptoms after CBT (Bleichhardt et al., 2005; Heider et al., 2011). This is both in the short- and the long-term and carries intermediate to large effects. Similarly, Haggarty et al. (2016) found that high and low somatizers improved equally well after CBT. The only paper which did not find such a relationship (Speckens et al., 1997) used a less robust and unvalidated measure of symptom frequency, while the others used the well-validated and reliable SOMS-7 (Rief & Hiller, 2003). It is possible that the less robust measure was not sensitive enough to accurately capture the extent of the symptoms, making their conclusions more questionable. However, it is less clear whether one of these variables is more important than the other, or what the impact might be if a person has both numerous and intense symptoms. Speckens et al. (1997) further found that the duration of symptomatology was not associated with outcomes one year later, although it did not make it clear how chronicity was assessed (for instance through medical notes or as self-report).

1.3.1.8 Psychiatric comorbidity. Six papers examined the effect that psychiatric comorbidities might have on CBT outcomes. These broadly fell into two categories; mood disorders (specifically anxiety and depression) and personality disorders. Most of the articles indicated that neither comorbid anxiety nor depression had an impact. Bleichhardt et al. (2005) found that comorbid anxiety and depression were no different across those whose symptoms improved after CBT versus those who did not improve, although the effect size for therapy success in the first instance was small. Likewise, Leibbrand, Hiller, and Fichter (1999a) found no significant overall differences in any treatment outcomes (including physical symptoms, hypochondriasis, and thoughts about body and health) across the different comorbidities. Moreover, effect sizes indicated that none of the groups with comorbid depression and anxiety, personality disorder, or a combination of both had any worse outcomes compared to somatoform participants with other minor axis I comorbidities or even non-somatoform controls. Furthermore, Heider et al. (2011) found neither comorbid depression nor anxiety at the start predicted the reduction in somatic symptoms. In contrast, Hiller et al. (2003) found comorbid depression to predict better symptom outcomes and comorbid anxiety to predict greater improvements to dysfunctional attitudes after CBT, compared to those without the diagnosis. One possible explanation for this discrepancy is that in the aforementioned study the clinical comparison group had significantly higher rates of depression at the start of therapy than the somatoform group, which would question the validity of that particular result. The study was also of moderate quality, with a reduction due to the confounding influence of concurrent therapies being given. The former three articles were rated as particularly strong in quality, notably for their control of confounding variables, their use of validated and reliable measures, and their low rates of withdrawal and drop outs.

The impact of comorbid personality disorders was explored in three studies and on the whole, it appears that they do not adversely affect outcomes. The studies by Leibbrand et al. (1999a; 1999b) found that those with a comorbid personality disorder did not have significantly worse outcomes for somatic symptoms, hypochondriasis, and catastrophizing beliefs than those with axis I disorders (Leibbrand et al., 1999a) or those without personality disorders (Leibbrand et al., 1999b). In Leibbrand et al. (1999b), effect sizes were moderate for somatic symptom improvement and somewhat better for depression and hypochondriasis outcomes. A trend was noted for slightly poorer outcomes in somatic symptoms and hypochondriasis where there were multiple personality disorders. There were large differences across sample sizes in the aforementioned studies however, particularly across the different personality disorder subcategories and it is unclear whether these differences were controlled for. In contrast, Zonneveld et al. (2012) found that having fewer personality disorder characteristics at baseline predicted better short-term CBT outcomes for physical quality of life. However, this finding was only true 6 months after therapy ended and not immediately after or in the long-term, suggesting the finding was not stable over time and questioning its reliability. It appears then that comorbid mood and personality disorders may not negatively impact on outcomes of CBT for MUS.

1.3.2 Process variables and therapeutic outcomes

1.3.2.1 Number of sessions. Twelve papers stated the average number of CBT sessions that participants received, from as few as a single session up to 16 sessions, with a median of 8. In two studies (Hiller et al., 2003; Leibbrand et al., 1999b), the number of sessions received was not explicitly stated, but rather the duration of the inpatient stay was presented, with the former offering around 17 hours of therapy weekly over the course of a mean inpatient stay of 58.6 days, while the latter offered an unknown quantity of therapy during an average inpatient stay of 63.35 days. In those cases, the number of sessions received was most likely higher than in the other papers presented. Only one paper explicitly considered whether the number of sessions predicted outcomes in some way. While it was not possible to calculate effect sizes, Arnold et al. (2009) found the number of sessions did not predict the improvements to symptoms, health anxiety, physical functioning, or healthcare use. However, it is noteworthy that they did not find CBT to be superior to the TAU group but that both groups improved. The four articles with large effects for somatic symptom improvement ranged in number of sessions from 7.3 (Haggarty et al., 2016) to 12 (Yoshino et al., 2015). Three articles demonstrating intermediate effects offered 8 and 10 sessions (Heider et al., 2011 and Allen, Woolfolk, Escobar, Gara, & Hamer, 2006 respectively) as well as the far more intensive 15-20 hours offered weekly to inpatients by Hiller et al. (2003). The only article to have found small effects for improvements to somatic symptoms was that by Martin et al. (2007), who offered only a single session. It is clear that there is no discernible trend in terms of how many sessions exhibited better effects. This would suggest that the number of sessions received do not make a clear difference to outcomes, while it can be concluded that a single session is unlikely to bring about meaningful change to a person's symptoms. Alternatively, it is possible that the different ways in which CBT was delivered across studies, with slightly different procedures and protocols, may have in part explained the large range of effect sizes seen. The different effect sizes may have also been influenced by potentially confounding variables. For instance, several studies (Bleichhardt et al., 2005; Hiller et al., 2003; Leibbrand et al., 1999a; 1999b), while primarily offering CBT, acknowledged that some participants received additional therapeutic interventions, such as progressive muscle relaxation or medication, without stating whom this applied to and without controlling for it. Finally, no studies offered between two and five sessions, making it impossible to say whether these session lengths would have yielded similar results to those seen above or not.

1.3.2.2 Mode of delivery. Of the fourteen papers included, five reported on individual CBT (Allen et al., 2006; Arnold et al., 2009; Haggarty et al., 2016; Speckens et al., 1997; Yoshino et al., 2015), four described group CBT (Heider et al., 2011; Lidbeck 1997; Martin et al., 2007; Zonneveld et al., 2012), another four described mixed group and individual CBT for inpatients (Bleichhardt et al., 2005; Hiller et al., 2003; Leibbrand et al., 1999a; 1999b), while one article directly compared group with individual CBT (Moreno et al., 2013), although not all articles made it possible to calculate effect sizes. For those that did, there was a visible spread of small, intermediate, and large effect sizes across the different modes of delivery for the primary outcome of somatic symptoms. Despite this spread of effect sizes however, there was one apparent trend, namely that there were fewer instances of small effect sizes when CBT was delivered individually as opposed to in a group. Moreover, the only study in the present review which directly compared the two (Moreno et al., 2013) found that individual CBT enjoyed visibly larger effect sizes than group CBT for reductions in symptom number and severity, as well as depression and anxiety. Individual CBT also showed a significantly greater improvement in symptom number reduction at 6 months compared to group CBT, which carried a large effect. There are some caveats to this however; it is entirely possible that the differences in effect sizes are an artefact of the way in which CBT was delivered across studies, where protocols may have varied greatly. Sample sizes across studies had also been identified as varying wildly, ranging from 34 to 542 total participants, making it difficult to draw firm conclusions about whether individual CBT was indeed superior to group CBT. It would have been of interest to compare the drop-out rates across both individual and group CBT designs, in order to explore whether one mode of delivery was more acceptable to participants that the other. Moreover, certain inpatient studies offered a mixture of individual and group CBT in unknown quantities, as well as a smaller but nonetheless undefined number of additional therapies, including progressive muscle relaxation and occupational therapy (Bleichhardt et al., 2005; Hiller et al., 2003; Leibbrand et al., 1999a; 1999b). Nonetheless, their spread of effect sizes was just as heterogeneous as the spread seen across studies investigating either individual or group CBT, suggesting the impact of mixing group with individual CBT, or even of offering a small amount of additional input to certain participants may not have disadvantaged these studies too much.

1.3.2.3 Profession of clinician. Thirteen of the fourteen included studies made reference to the profession of the person delivering CBT. Seven papers described CBT being delivered by mental health staff (Bleichhardt et al., 2005; Haggarty et al., 2016; Heider et al., 2011; Hiller et al., 2003; Leibbrand et al., 1999a; 1999b; Martin et al., 2007), three by General Practitioners (GPs, Arnold et al., 2009; Lidbeck, 1997; Speckens et al., 1997), two by psychological therapists (Allen et al., 2006; Moreno et al., 2013), and one by a psychiatrist (Yoshino et al., 2015) although not all of these papers made it possible to calculate effect sizes. There was a large range of effect sizes seen across the different clinician professions, with small, intermediate, and large effects found in most clinician groups. What did emerge however was that psychological therapists did appear to have particularly strong results at least for short-term symptom improvement, although their effects reduced in one study (Allen et al., 2006) and eventually disappeared in another (Moreno et al., 2013), which was not a trend seen among other clinicians. Interestingly, Arnold found CBT to be no better than TAU at improving symptoms when delivered by GPs, and that both groups improved significantly after the intervention. There may be several explanations for the degree of spread seen in effect sizes. One of the studies involving psychological therapists (Allen et al., 2006) used a more basic assessment tool for symptom severity, which only requires a 7-point scale response, while most other studies (including the second with psychological therapists) opted for the valid and reliable SOMS-7 measure (Rief & Hiller, 2003). Studies also varied in their diagnostic inclusion criteria, particularly among mental health staff studies, with one only requiring a minimum of two different MUS in the last six months (Martin et al., 2007), while another required a full DSM-IV diagnosis (Heider et al., 2011). It is possible that the former set of participants may not have had as severe or enduring difficulties as the latter group to begin with, making the potential for change quite different among the two. Among the three papers that considered CBT delivered by GPs only one of these looked at symptom outcomes and another (Arnold et al., 2009) did not allow for effect sizes to be calculated.

1.3.2.4 Additional cognitive or behavioural factors. Three of the fourteen included articles considered the potential impact that cognitive or behavioural factors might have had on the positive outcomes achieved by CBT. Expectations of improvement were covered by two papers, although the results were conflicting. Allen et al. (2006) found that such expectations were not correlated with outcomes, as the CBT group and the control group did not differ significantly in terms of their treatment expectations, while Bleichhardt et al. (2005) found that positive expectations of improvement actively predicted significant short- and long-term reductions of physical symptoms. While both studies used reliable and valid measures with very similar Cronbach α‘s (Devilly & Borkovec, 2000 and Schneider, Basler, & Beidenherz, 1989 respectively), the ability of the CEQ as used by Allen et al. (2006) to predict therapeutic outcomes is less certain (Devilly & Borkovec, 2000). However, as Bleichhardt et al. (2005) did not make it possible to calculate effect sizes, it is difficult to conclude either way whether expectations of improvement predict better outcomes in MUS. Motivation was also found to be involved in the improvements seen from CBT with this client group, at least by the one study which investigated the concept. Bleichhardt et al. (2005) identified that a greater motivation to engage in therapy correlated with a significantly greater reduction in symptoms both short- and long-term, although the effect sizes were small. The same study also found that a reduced level of subjective suffering at the end of treatment predicted long-term improvements to health-related quality of life, which carried a large effect. However, it is worth bearing in mind the study’s earlier finding that claiming disability benefits also predicted poorer outcomes. It is possible that the perceived risk of participants losing their disability benefits due to improvements in their functioning may have influenced therapy motivation, although this was not explored further in the study.

Finally, particular health-related cognitions and behaviours were explicitly examined as potential predictors of treatment outcome by just one paper. Speckens et al. (1997) found that not only was a change in health anxious cognitions related to a significant change in psychosocial distress, but that a greater degree of hypochondriacal cognitions at six months predicted more psychological distress one year later. Both of these carried large effects. Moreover, the same study discovered that higher levels of illness behaviours at baseline predicted poorer outcomes one year later, with a large effect. While the findings are persuasive in terms of their effect sizes, it is important to note the questionable choice of a simple symptom measure, when the well-validated and reliable SOMS-7 (Rief & Hiller, 2003) could have been used and which is likely to have been more sensitive to clinical levels of MUS. Drawing firm conclusions is made more difficult considering these results come from only a single article.

1.4 Discussion

1.4.1 Summary of evidence for participant variables and therapeutic outcomes

Gender did not appear to influence the improved short- or long-term outcomes to physical and psychosocial difficulties after CBT. While the finding is at odds with a review by Kleinstäuber, Witthöft, and Hiller (2011), who found that gender moderated the impact that psychotherapy had on outcomes, it is important to note the small number of effect sizes they found per outcome. Their review was also not as stringent in its inclusion criteria around diagnosis, additionally including specific syndromes such as IBS and CFS. The present findings are however in line with the research into CBT's use in depression for instance, where a large scale meta-analysis by Cuijpers et al. (2014) found no evidence that men or women respond better or worse to CBT than to medication. These are highly relevant findings for healthcare services supporting people with MUS, as it suggests that men are just as likely to benefit from a psychological intervention for their symptoms as women are, despite women being more likely to seek support for mental health difficulties in the first instance (Department of Health, 2008). This in turn ought to motivate policy makers to encourage more men to access psychological therapy for their physical symptoms. Likewise, age was not found to mediate the improved outcomes that were seen after CBT, although these findings cannot be extended to children and adolescents or older adults over 70. Indeed, a completely separate literature exists into the efficacy of CBT for specific syndromes such as chronic fatigue in adolescents (see for example Stulemeijer et al, 2005). Nonetheless, the present finding is significant for services where individuals of a range of ages may be referred for psychological support and where both they and their clinicians may be reassured that outcomes are promising, regardless of age. Adaptations are likely to be required, such as when explaining to children how emotions, thoughts, bodily sensations and behaviours are linked, or when involving an older adult's carer as co-therapist. Familial status also did not appear to influence improved psychosocial outcomes after CBT, however this was based on only a single study and it is difficult to draw firm conclusions from this. In some ways, the lack of a correlation is surprising, as more social support has been linked to better CBT outcomes for instance in post-traumatic stress disorder (Fredette, El-Baalbaki, Palardy, Rizkallah, & Guay, 2016), while moderating the effect of CBT on quality of life improvements in older adults (LaRocca & Scogin, 2016). However, having children, especially when they are young, arguably also has the potential to be a significant life stressor at times and may therefore still have a mediating effect on outcomes.

It was not possible to conclude, based on the articles included here, whether education has an impact on later CBT outcomes. One might have expected that a lower education at baseline would predict poorer outcomes from CBT for MUS. For instance, Hyphantis, Goulia, and Carvalho (2013) compared healthy controls with participants who had explained chronic illnesses and found that less education in both groups was positively correlated with greater somatic symptom severity. It would have been useful to know whether the studies which were based in specific MUS services (as was the case with Hiller et al., 2003) had comparable education rates to the rest of the population, not just between treatment and control groups. More conclusive findings would be relevant for services as it would indicate whether adaptations are needed to the way in which CBT is delivered, depending on a person's level of education.

Being in receipt of disability benefits was found by one (Bleichhardt et al., 2005) to predict poorer outcomes, although this article may not have conducted appropriate statistical analysis on the correlation. This calls into question any conclusion that can be drawn, although it would have been useful for services to have an answer to this question. For instance, if claiming disability benefits were indeed to have a negative impact on how well CBT works for individuals with MUS, it would be important to find out why this might be; is for instance diminished motivation involved in explaining the relationship. The question is highly relevant when one considers the high degree of occupational impairment that is associated with MUS (Katon et al., 1991) and supporting individuals to get back into paid employment is a priority for governments. Greater social difficulties more broadly were also found to correlate with significantly poorer outcomes at the end of treatment and at 6 months, compared to those with lower initial social difficulties (Lidbeck, 1997). However, there were large early drop-out rates and it is plausible that those with greater social difficulties (such as housing or financial) may have been more likely to drop out for practical reasons, which might have biased the samples to begin with. There is an emerging evidence into the relationship between, for instance, poverty and poorer mental health (Elliott, 2016), however there does not appear to be any investigation of whether such social problems mediate CBT outcomes in mental health conditions. One might expect lower socioeconomic status to impact on how easily a person accesses CBT, particularly in countries where the therapy is provided by a national health service. If it were the case that having greater social difficulties predicts a poorer response to CBT for one’s medically unexplained symptoms, further research into how this might be happening would be crucial. This in turn would enable clinicians to tailor their intervention if need be, for instance by taking into account the person’s more limited resources. On a broader scale, governments might be encouraged to invest further in social support, as successful treatment of MUS with psychological therapies would be hugely cost effective.

The current literature also suggests that a greater number and intensity of symptoms at the start predicts greater improvements after CBT. The length of time that symptoms have been present for does not appear to impact on outcomes, although these findings are more tentative. It remains unclear whether any one of these variables are more important than the others, or what the outcome might be if a person has numerous, intense and long-standing symptoms compared to someone with a different presentation. It would be motivating to know that CBT may be highly effective even if a person exhibits numerous, intense, or long-standing symptoms, particularly considering that for many, these symptoms will have been present in their lives for some time (Rief & Hiller, 1992). In contrast, having a comorbid anxiety, depressive, or personality disorder does not significantly impact on how well a person responds to CBT for their unexplained symptoms. Comorbidities have also been investigated in other domains, such as in panic disorder. For instance, the results of a review by Mennin and Heimberg (2000) were less certain regarding the impact of comorbid depression, but found a clearly detrimental effect of comorbid personality disorders on CBT outcomes for panic. As the findings from the present review are based on a limited number of studies, the concepts are worth exploring further. For instance, in the included articles there was no additional analysis of whether the chronicity or severity of these comorbidities had any effect. This would be relevant for clinical settings, where individuals with MUS have high rates of mood and personality comorbidity (Henningsen, Zimmermann, & Sattel, 2003; Nater et al., 2010). The present findings overall are relevant to services in the UK especially, which usually divide themselves according to whether it is a mental health or a physical health problem. However, this might result in a person with both mental and physical health comorbidities to fall between services where these may be unsure about who should treat the person. The present findings would indicate that outcomes are promising when CBT is offered for unexplained symptoms, regardless of whether the person has an additional mood or personality disorder.

1.4.2 Summary of evidence for process variables and therapeutic outcomes

There was a large range in terms of the number of sessions provided, with no clear pattern in terms of effect sizes for the different studies, such that it is difficult to say whether there is an optimum number of sessions for greatest impact. However, it is unlikely that a single session is adequate for this client group. This might indicate that the number of sessions a person receives does not necessarily impact how well they will respond to treatment, suggesting the presence of other factors. For instance, research has shown the importance of elements such as the therapeutic relationship (described in Leahy, 2008), collaborative working (described in Dattilio & Hanna, 2012), and use of homework tasks (see for instance Kazantzis, 2000 and Kazantzis, Deane, & Ronan, 2000) in improving outcomes of CBT. It stands to reason that it may not be entirely down to how many sessions are offered, but instead how these are used in collaboration with the client. Indeed, there does not appear to be any NICE guidance on how to offer psychological interventions for people with somatoform disorders or unexplained symptoms. For CFS and IBS for example, CBT is recommended although no advice is given about how many sessions are required, just that one should follow “the usual principles” of CBT (NICE, 2007; NICE, 2017). Being able to deliver effective CBT with fewer sessions would be of interest to services, as it would cut waiting times and reduce cost. However, the findings above would argue that the number of sessions are not in fact predictive of better outcomes, other than that a single session is unlikely to bring about meaningful change.

In terms of the mode of delivery, overall the findings suggest a trend for individual CBT as having slightly better outcomes than group CBT for MUS. Considering the wider literature, it has been found in depression that individual and group CBT are in fact comparable in their efficacy and that both are superior to no treatment at all (Lockwood, Page, & Conroy-Hiller, 2004), while in panic and agoraphobia the two forms of delivery also achieved statistically comparable improvements compared to a waiting list control (Sharp, Power, & Swanson, 2004). However, the latter study found that *clinical* significance of outcome was significantly greater after individual therapy than both group therapy and waiting list control, which is somewhat more in line with the present review’s finding. It is possible that certain conditions lend themselves more or less to individual or group therapy and confirming whether individual or group CBT is more effective for people with MUS or somatoform conditions is highly relevant for services and policy makers, so that the intervention can be delivered in the most cost-effective way.

There did not appear to be any apparent trend in terms of whether particular clinician professions brought about greater improvements over others. This might suggest that it does not matter so much who delivers the treatment. However, upon closer inspection of effect sizes, when CBT was delivered by psychological therapists, short-term outcomes appeared particularly strong, while symptom severity improvements then reduced in the long-term. Moreover, the only study in which CBT was no better than TAU was a study in which the treatment was delivered by GPs. A recent meta-analysis of 3225 participants across 20 papers by Gerger, Hlavica, Gaab, Munder, and Barth (2015) supports the above findings to some extent. They found that psychological interventions were more effective at reducing somatic symptoms when they were delivered by psychological therapists than by GPs, although there was no difference in terms of physical functioning and psychological symptoms between the two clinicians. Due to the meta-analytic nature of that study, it is likely that its findings are a more accurate reflection of clinician impact than the present review. However, before one can conclude from the present findings about whether the profession of the clinician impacts on the efficacy of the therapy, it is important to note that many more papers included here examined CBT’s effectiveness as delivered by mental health staff, while very few studies included psychological therapists and even fewer GPs or psychiatrists. If indeed it is the case that psychological therapists bring about more effective change (even if just short-term) than GPs for instance, it would certainly merit further funding of secondary care psychological services, as opposed reducing these in favour of GPs offering the treatment in primary care.

In terms of cognitive or behavioural factors on the part of participants, it was not possible to conclude whether expectations of improvement influenced CBT outcomes, however it would be intuitive to suspect that higher expectations would predict better outcomes. This was demonstrated in a meta-analysis by Constantino, Arnkoff, Glass, Ametrano, and Smith (2011) of 8016 participants receiving a range of psychotherapies for different conditions. They found a small but positive effect for the association between greater pre-therapy expectations with improved therapy outcomes, although their risk of publication bias advises caution in drawing firm conclusions. Nonetheless, if expectations of improvement also predict better outcomes for MUS then clinicians and services should set up appropriate expectations from the very beginning. This is all the more significant in MUS, where clients may enter therapy with the belief that their symptoms would disappear completely, when in fact the aim of CBT would be to reduce the distress associated with these symptoms. The current literature review also appears to suggest that greater motivation is related to a greater reduction in physical symptoms in the short- and long-term after CBT for MUS. This is in line with the wider literature suggesting that motivation may be associated with treatment outcome after CBT. Specifically, the stage that a person is at within the stages of change model (Prochaska, & DiClemente, 1984) predicts their likelihood to drop out of treatment (Dozois, Westra, Collins, Fung, & Garry, 2004). If motivation were a significant predictor of outcomes in MUS, then assessing for this would be hugely important for services. By establishing early on where a person is within the stages of change model, whether it be at pre-contemplation where they may not be aware of their problematic behaviours, or at the preparation stage where they may already be taking small steps, the intervention can be offered in the most timely manner. It may mean for some that the intervention is postponed until the person is in the preparation phase, thus avoiding unnecessary input that is not made use of. Finally, in the present review both health anxious thinking styles and illness behaviours strongly predicted poorer outcomes one year later. This would emphasise the need for clinicians to target these thoughts and behaviours early on in CBT. Indeed, Christensen et al. (2015) compared completers of CBT group therapy with a treatment as usual group and found that an improvement in illness perceptions after therapy partially mediated the effects of CBT on physical health outcomes.

1.4.3 Limitations of the present review

Firstly, only a single reviewer was used to evaluate papers against the inclusion and exclusion criteria, affecting the reliability of the review. Similarly, while every effort was made to be objective when rating papers for quality, it is noteworthy that the present review overall appeared to rate quality favourably. This is in comparison to the review by van Dessel et al. (2014) who examined non-pharmacological interventions for MUS and were considerably less impressed by the quality of their included papers. Regarding the search terms that were applied in the present review, it is possible that they were overly restrictive when finding moderators, predictors or mediators of CBT outcomes. For instance, including ‘regression’ as an additional search term might have included papers that carried out regression analyses and found relevant variables that way. Indeed, while hand-searching papers from reference lists, some potentially relevant articles emerged which did not explicitly use the terms ‘mediator’, ‘moderator’, or ‘predictor’. However, the hand-search itself was extensive, so the final papers included here are representative of the literature.

Moreover, the present article is not a meta-analysis and effect sizes were instead commented on in a more qualitative manner. Finally, a shortcoming across several studies, notably those in inpatient settings, was the potentially confounding impact of other therapies (such as medication or progressive muscle relaxation) being offered alongside CBT. Indeed, Kleinstäuber et al. (2011) in their meta-analysis identified through moderator analysis that a lack of control of concomitant treatments does influence effect sizes.

1.5 Conclusions

The present review aimed to highlight convincing predictors, mediators or moderators of CBT's apparent success with MUS and Somatoform Disorders. Age and gender did not appear to predict better or worse outcomes once a person had entered therapy. Having a comorbid mood anxiety, depression, or personality disorder also did not seem to impact on how well a person would respond to CBT. Moreover, individual CBT may be superior to group CBT, at least in terms of effect sizes, however it is important to note that this review could not use meta-analysis due to the heterogeneity of the data. Perhaps counter-intuitively, the number and intensity of physical symptoms appeared to predict better outcomes and future research would benefit from exploring whether additional variables are at play to explain this relationship, such as motivation. Despite the findings above, there appears to be a distinct lack of conclusive research into predictors of outcomes for CBT when offered to patients with unexplained physical symptoms and much of what was described was correlational in nature. There was a particular dearth of articles examining familial status, education, and social difficulties, which resulted in those conclusions being much more tentative. Notably, no paper examined whether ethnicity mediates therapeutic outcomes, which is surprising considering how under-represented ethnic minorities are in services such as IAPT (Clark, 2011). Likewise, there did not appear to be any convincing trend in terms of whether clinician profession or number of sessions were linked to larger effects, although it appeared that a single session of CBT was unlikely to be sufficient. Kleinstäuber et al. (2011) supplements these uncertain findings by indicating that the number of sessions and profession of the therapist do in fact impact on outcomes after psychotherapy. Future studies should use the latest DSM-5 criteria of Somatic Symptom Disorder (SSD, APA, 2013), which no longer places a requirement on the symptoms being unexplained. It is likely that this lack has been due to an absence of a suitable diagnostic tool to assess for SSD and such a tool has only become available recently (the Somatic Symptom Disorder-B Criteria Scale, Toussaint et al., 2016). Future studies should continue to identify relevant variables using validated and reliable measures, state their intervention clearly, carry out longitudinal and randomised controlled trials or similar and, where possible, manipulate variables experimentally. For instance, a study might directly compare whether offering one session, five sessions or sixteen sessions is more effective for MUS, or whether a person's ethnicity will predict how much they will benefit from CBT.

Chapter 2: Empirical paper

The effectiveness of cognitive therapy for medically unexplained symptoms

2.1 Introduction

Medically Unexplained Symptoms (MUS) are physical symptoms for which no medical explanation has been found, despite the often-extensive investigations and tests that are carried out (van Dessel et al., 2014). Many medical specialities appear to have their own distinct unexplained symptoms; irritable bowel syndrome in gastroenterology; fibromyalgia in rheumatology; tinnitus in ENT; atypical facial pain in dentistry; chronic pelvic pain in gynaecology, to name just a few. Moreover, it is telling that even the name for medically unexplained symptoms varies across the professions and throughout the literature and includes somatisation, functional disorder, conversion disorder, hypochondriasis, among many others, illustrating the widespread but uncertain nature of medically unexplained symptoms.

Multiple unexplained symptoms lasting several years are needed in order for full somatisation disorder to be diagnosed according to the International Classification of Diseases-10 (ICD-10, World Health Organisation, 2016) and the Diagnostic and Statistical Manual of Mental Disorders (4th edition, DSM-4, American Psychiatric Association, APA, 2000). The latest DSM-5 (5th edition, APA, 2013) only requires a single somatic symptom to be present for at least 6 months and it no longer needs to be unexplained in order for the new somatic symptom disorder to be diagnosed. For the purpose of the present study, the earlier label of MUS will be used throughout, as it is in line with much of the literature to date and corresponds more closely to the time at which the present data were collected. Kirmayer, Groleau, Looper, and Dominicé Dao (2004) argue that the term MUS does not in fact refer to a specific disorder, but instead describes a social and clinical predicament which the person finds themselves in. Despite remaining unexplained, these symptoms can be highly disabling and are capable of causing a great deal of functional impairment (van Dessel, Leone, van der Wouden, Dekker & van der Horst, 2014). Indeed, it has been shown that the more unexplained symptoms that a patient reports, the greater the level of emotional distress and functional impairment that is experienced (Katon et al., 1991). MUS are particularly common in primary care, where it is estimated that between 15% and 30% of all consultations are for medically unexplained symptoms (Kirmayer et al., 2004). It is perhaps not surprising that they are also linked to high healthcare use and subsequent cost (Konnopka et al., 2012).

2.1.1. Psychological theories of MUS

A number of theories have been put forward to explain these symptoms, including social constructionist (Kleinman & Becker, 1998) and early psychodynamic perspectives (Breuer & Freud, 1955). Two theories are particularly relevant to the present evidence base however, namely attachment theory and the cognitive behavioural model. Despite their limitations and far from being mutually exclusive, these two theories may combine well to explain some of the developmental and perpetuating issues of MUS and its related conditions. Adshed and Guthrie (2015) describe how the attachment system is activated at times of distress, resulting in reassurance-seeking behaviours (such as crying out). In infancy, the care-giver would respond to these behaviours and gradually the infant would develop expectations about whether they would consistently receive care when distressed. As the infant matures, these care-giver experiences form the basis of beliefs about the self as being worthy of care and whether others can be relied upon to provide care. These beliefs are highly relevant for physical health conditions, where feeling worthy of care and believing that others would respond to our ‘cries’ for help would impact on how we experience these symptoms and relate to others. There is some compelling evidence for the role of attachment styles in MUS, as demonstrated for instance in a study by Taylor, Mann, White, and Goldberg (2000) of 2042 consecutive attenders in primary care. Those with MUS were 2.47 times more likely than those with organic physical illness to have an insecure attachment and a later study (Taylor, Marshall, Mann, & Goldberg, 2012) additionally found that this attachment style was related to frequent attending in primary care generally. One critique of the earlier Taylor et al study is that a self-report measure of attachment was used, which they acknowledged yielded quite different results to interview based assessments. However, a study by Waller, Scheidt, & Hartman, 2004, which applied a widely-used interview-based measure, similarly found secure attachment styles in as few as 26% of a MUS sample, compared to 60% of controls. Importantly however, the model does not explain how the symptoms themselves may have developed.

The most prominent theory that attempts to explain in greater detail how a person might develop MUS is the Cognitive Behavioural model (Salkovskis & Warwick, 1986). The theory posits that unexplained symptoms occur when a person brings their attention to and catastrophically misinterprets physical sensations. Such sensations might be the result of normal bodily responses, such as a racing heart after exercise, or due to a minor illness such as a cold (Deary, Chalder & Sharpe, 2007). A vicious cycle may then take hold as the person seeks reassurance from others and from medical tests and alters their normal behaviours when they experience physical symptoms of any kind, such as by avoiding exercise. A recent adaptation of this model (Salkovskis et al., 2016) emphasises the additional importance of certain trans-diagnostic factors such as psychological inflexibility and clinical perfectionism as further maintaining factors. However, as Brown (2013) points out, the cognitive-behavioural model also has its limitations, most notably as it is less able to explain some of the more pronounced neurological experiences, such as unexplained blindness. Nonetheless, both attachment theory and the cognitive behavioural model appear to combine well in explaining MUS, with the former providing an insight into their maintenance within relationships, while the latter suggests possibilities for their origins, while also demonstrating maintenance cycles.

2.1.2 Cognitive behavioural therapy for MUS

As will be demonstrated below, the most widely researched psychological therapy for MUS and its related disorders appears to be Cognitive Behavioural Therapy (CBT). CBT enables individuals to manage their difficulties better by changing the way they think and behave. However, despite CBT's firm evidence base for use in various physical and mental health conditions (see for instance a large-scale review of meta-analyses by Hofmann, Asnaani, Vonk, Sawyer, and Fang, 2012), its efficacy in MUS is less well established and the evidence base surrounding it is still emerging, with mixed quality research and small effect sizes. A recent Cochrane review (van Dessel et al., 2014) considered 21 randomised- or cluster-randomised controlled trials since 2005. The papers included a total of 2658 participants diagnosed with MUS generally or with a somatoform disorder more specifically and who were also receiving psychological therapy of some sort. Inclusion and exclusion criteria for the review were stringent and so resulted in the selection of more studies evaluating the effectiveness of some form of CBT (fourteen studies), than other therapies. Two studies evaluated behaviour therapies, two more evaluated mindfulness, while two studies examined psychodynamic therapies. One more study included participants who either received CBT, client-centred, or eclectic therapy. While the authors reported that psychological therapy generally was better than treatment as usual (TAU) or waiting list controls at improving symptom severity, the quality of the evidence was low to moderate and effect sizes were small. Considering particular therapy types more closely, only CBT had been studied enough to enable conclusions to be drawn. It was found that CBT was better than waiting list controls or usual care at reducing somatic symptoms and that this improvement was maintained at one-year follow-up. In contrast, CBT was found to be no more effective than enhanced care (where structured appointments were given but no therapy for MUS), although initial effect sizes were small and varied across studies. Again, due to the small number of studies, it was not possible to make comparisons between CBT's efficacy compared to other psychological therapies. It was also not possible to draw conclusions about improvements to depression and anxiety scores and there were inconclusive findings about whether healthcare use reduced as a result of CBT interventions.

In contrast to this latter finding, an earlier review of 34 RCTs and 3922 participants by Kroenke (2007) did demonstrate that CBT generally improved psychological wellbeing relative to waiting list or TAU control groups. Moreover, that review also indicated a possible reduction in healthcare usage and, to a lesser extent, cost among recipients of CBT, although a reduction in cost was also seen in some studies comparing a psychiatric consultation letter to the GP with TAU. A recent Cochrane review by Martlew, Pullman and Marson (2014) considered the efficacy of psychological and behavioural therapies for unexplained non-epileptic attack disorder. They examined 12 studies with 343 participants and while they included RCTs, they also reviewed pre- and post- non-controlled trials. Treatments being offered were CBT, psychotherapy, hypnosis, paradoxical intention, and mixed interventions. CBT again appeared to be the most favourable intervention, based on one particular RCT which stood out in the review as being of especially high quality. Here, CBT was found to significantly reduce seizure frequency at the end of treatment, although an improvement at follow-up was no longer significant. Other examples of specific unexplained symptoms for which CBT's usefulness has been demonstrated include in irritable bowel syndrome (IBS, Moss-Morris, McAlpine, Didsbury, & Spence, 2010) and chronic fatigue syndrome (CFS, Malouff, Thorsteinsson, Rooke, Bhullar, & Schutte, 2008). In sum, CBT for unexplained symptoms has been investigated more than other psychological interventions. While it appears to hold the most promise for a range of unexplained symptoms in terms of physical, psychological and, to a lesser extent, economic outcomes, the effect sizes across the literature are small. Moreover, the long-term benefits of CBT for this client group are less clear.

2.1.3 Cognitive analytic therapy for MUS

Cognitive Analytic Therapy (CAT) is a time-limited therapy, typically of around 16 sessions, which integrates ideas from cognitive and analytic therapies (Denman, 2001). It looks for patterns in how a person thinks, feels and behaves and puts these in the context of earlier experiences and relationships. It also considers how people enter into roles with others and how these roles may have developed years earlier, sometimes in childhood, but cause difficulty in the present day (Calvert & Kellett, 2014). The relevance of attachment theory is therefore particularly pertinent when considering the role of CAT for MUS. Surprisingly however, a search of the literature revealed no published studies considering the efficacy of CAT for MUS or its related conditions. The therapy also does not appear in any of the reviews already mentioned. Anecdotally, Jenaway (2011) reports her own observations of a small sample of seven patients who received 16 sessions of CAT for a variety of MUS. While one of those showed no change on an assessment of wellbeing, symptoms, functioning, and risk, the remaining six patients demonstrated a reliable reduction in their scores at the end of therapy. However, this analysis did not report the statistical tests used or effect sizes.

The paucity of research into CAT is surprising considering its earlier application to long-term (explained) conditions such as type 1 diabetes (Fosbury, Bosley, Ryle, Sönksen, & Judd, 1997). CAT has also been found to be better than TAU in clients with personality disorder (Clarke, Thomas, & James, 2013) and anecdotally there has been the suggestion that it may be useful for clients who have experienced childhood abuse or trauma (Jenaway, 2011). The lack of research is further surprising considering Hyphantis, Guthrie, Tomenson, and Creed's (2009) findings that the improvement in health status of IBS patients following psychodynamic interpersonal therapy was most likely due to an improvement in their interpersonal problems. Moreover, van Dessel et al. (2014) in their review of psychological therapies for medically unexplained symptoms concluded that more research into therapies other than CBT was urgently needed. As CAT is a time-limited therapy based on relationship patterns (to ourselves, to others, to our illness, to those treating us) it would be prudent to investigate its effectiveness further. Indeed, greater interpersonal change was seen in the diabetes group receiving CAT than in the control group seen by diabetes nurses in Fosbury et al.'s study (1997), who also found that CAT offered more enduring change. The need for greater depth and breadth of research into CAT generally was also concluded in a recent review by Calvert and Kellett (2014). They included twenty-five outcome studies for CAT (five of which were RCTs) and reported that CAT was mostly used in complex clinical populations, notably for those with a personality disorder, and that while just over half of the studies were rated as high quality, they were mostly practice-based. In sum, while CAT has been applied to various mental and physical health conditions, it has been under-researched in the area of MUS. This, despite it being seen to effect interpersonal change; highly significant for unexplained symptoms that result in a great deal of reassurance-seeking.

2.1.4 Variables involved in therapeutic outcomes

While the literature on MUS documents CBT's usefulness and suggests a theoretical application for CAT, it would be useful to understand the mechanisms and possible mediators for change, so that individuals who are more or less likely to benefit from therapy may be identified and therapies better targeted. Some research has indicated that the various medically unexplained and functional somatic complaints, despite affecting different systems in the body, in fact have much in common with each other, for instance in aetiology, psychological mechanisms, and treatment response (Christensen, Frostholm, Ørnbøl, & Schröder, 2015). It is therefore possible that psychological interventions used in MUS and its related conditions may be working through some similar mechanisms. A 2007 study by Lackner et al. indicated that CBT was more effective at improving quality of life in patients with more severe initial levels of IBS symptoms and very tentatively suggested that CBT had a greater effect on those with more long-standing symptoms. However, this latter finding was no longer seen once other covariates were controlled. In contrast, Speckens, Spinhoven, van Hemert, and Bolk (1997) examined the impact of chronicity of symptoms and found that it did not correlate with outcomes after therapy. There is a clear need for further research in this area, which was also the conclusion of van Dessel et al.'s review (2014). Further investigation is also needed into whether the number of MUS that a person has influences how well they respond to therapy, with the literature appearing to suggest that a greater number may be linked to better outcomes (Bleichhardt, Timmer, & Rief, 2005 and Heider & Zaby, 2011).

In terms of participant variables potentially affecting outcomes, Lackner et al.'s (2007) mediation analysis did not find an interaction by gender when CBT improved symptom outcomes for IBS patients. Heider and Zaby (2011) as well as Speckens et al. (1997) similarly identified men and women as benefitting equally from CBT for their MUS. These findings are further in line with the findings for some other mental health conditions such as depression, with a review by Cuijpers et al. (2014) finding no evidence that either gender responds better or worse to CBT than to medication. Ethnicity is another variable of interest, as it has been identified that black and minority ethnic groups experience poorer outcomes with mental health services (Joint Commissioning Panel for Mental Health (2014).

2.1.5 Research aims

Primarily, this study aims to contribute to the evidence base for CBT and CAT's effectiveness at improving psychosocial, physical and economic outcomes for adults with MUS and its related conditions. It also aims to examine additional variables that may be involved in the therapeutic process. At present, the evidence for CBT with this population is still emerging, often limited by small sample sizes and small effect sizes, while for CAT it is very under-researched. The present study also aims to increase the one-year follow-up time seen in the existing literature, by following participants' healthcare cost up at 6 months, 12 months, and 18 months after the end of therapy.

2.1.5.1 Primary hypotheses. It is hypothesised that both CBT and CAT would significantly reduce psychosocial and physical distress (depression, generalised anxiety, health anxiety, somatic symptoms, impact on work and social adjustment) immediately after therapy compared to at the start. That both CBT and CAT would significantly reduce healthcare cost at the end of therapy, at 6 months, 12 months, and 18 months post-completion compared to a TAU group.

2.1.5.2 Secondary hypotheses. It is hypothesised that gender would not predict improvements after therapy, while ethnic minorities would show less improvement after psychological therapy than White British individuals. It is also predicted that the more MUS and the longer the duration of MUS that an individual has, the more improvement will be shown after psychological therapy.

2.2 Methods

2.2.1 Participants

Participants were recruited from an outpatient Medically Unexplained Symptoms service in the UK, which offered CBT and CAT. Patients were referred to this service by their General Practitioners (GPs) if they had at least one MUS and had presented with these symptoms to their GP at least ten times in the last year. Patients were excluded from the service if they were under 18 years old, or if they had chronic fatigue syndrome, fibromyalgia, or chronic pain, as separate services existed for these syndromes. All patients seen in the service who received at least 4 sessions of therapy were included in the study, although only data for those participants with complete pre- and post-data sets were analysed. A comparison group, the treatment as usual (TAU) group, consisted of patients whose referrals into the service were accepted who then declined to go ahead with therapy. As the study was retrospective and used information obtained from a working service, drop-out and withdrawal rates were not available. The final sample of 104 participants consisted of 68 females and 36 males, while information on age was not obtained.

2.2.2 Design

In this retrospective, non-randomised observational study, participants either received CBT or CAT and this decision was made by the clinicians running the service and was based on clinical need. CBT was delivered by qualified CBT therapists or clinical psychologists, while CAT was delivered by a consultant clinical psychologist who specialised in this therapy. All sessions were weekly individual face to face sessions lasting one hour. Mean number of CBT sessions were 11.7 (*SD* = 4.78), while CAT participants received a mean number of 15.2 sessions (*SD* = 5.4). For psychosocial and physical outcomes, the independent variable was group (CBT or CAT, between subjects), while the dependent variables were psychosocial and physical outcome measures (described in section 2.2.2 below), measured at two time points (pre- and post-therapy). For economic outcomes, the independent variable was group (CBT, CAT, or TAU, between subjects), while the dependent variables were inpatient, outpatient, and A&E costs, measured longitudinally at four time points.

2.2.3 Questionnaires

The following measures were completed at the start and at the end of therapy for the treatment group (see appendices B-F):

2.2.3.1 Patient Health Questionnaire – 9 (PHQ-9). This self-report measure of depression is widely used in primary care in the UK and has been shown to be valid and reliable (Kroenke, Spitzer, & Williams, 2001). Its 9 questions are rated from 0 (“not at all”) to 3 (“nearly every day”) with a maximum possible score of 27. A total score of 5-9 denotes mild depression, 10-14 moderate depression, 15-19 moderately severe depression, and 20+ severe depression

2.2.3.2 Generalised Anxiety Disorder Questionnaire (GAD-7). This self-report measure of generalised anxiety is widely used in primary care in the UK and has been found to be reliable and valid as a screening tool (Spitzer, Kroenke, Williams, Löwe, 2006). Its 7 questions ask the person to consider the last two weeks and rate their difficulties from 0 ("not at all") to 3 ("nearly every day"), with a maximum possible score of 21. A score of 5-10 denotes mild anxiety, 11-15 moderate anxiety, and 16+ severe anxiety.

2.2.3.3 Patient Health Questionnaire – 15 (PHQ-15). This self-report measure of somatic symptom severity has been shown to be reliable and valid (Kroenke, Spitzer, & Williams, 2002) and covers 15 different somatic symptoms with each rated by the patient from 0 ("not bothered at all") to 2 ("bothered a lot"). The maximum possible score is 30, with a total score of 5-10 representing low somatic symptom severity, 11-15 medium severity, and 16+ high severity.

2.2.3.4 Work and Social Adjustment Scale (WSAS). This self-report measure of perceived impaired functioning is a reliable and valid measure (Mundt, Marks, Shear, & Greist, 2002), widely used in primary care in the UK. Patients are asked to rate their difficulties from 0 ("not at all") to 8 ("very severely"). The maximum possible score is 40 and a total score below 10 denotes subclinical levels of functional impairment, while 10-20 denotes significant impairment, and 20+ indicates moderately severe impairment.

2.2.3.5 Short Health Anxiety Inventory (SHAI). This self-report questionnaire measures health anxiety independent of actual physical health and is a reliable and valid measure (Salkovskis, Rimes, Warwick, & Clark, 2002). Its 18 questions are rated as 0 (no concerns) to 3 (most concerns), with a maximum possible score of 54. A total score of 15-17 denotes subclinical levels of health anxiety and 18+ denotes clinical levels.

2.2.4 Economic outcomes - healthcare cost

Information about healthcare cost was collected by the relevant NHS Trust from both the treatment and TAU group in the 6 month before therapy started and in the 6 months, 12 months, and 18 months after therapy ended. This included inpatient, outpatient and A&E attendance and cost.

2.2.5 Ethical approval

Full NHS ethical approval was not required, as this study was considered to be a service evaluation rather than research. However, the relevant NHS Trust Research and Development department approved the project and granted access to the NHS site where needed. University of Southampton ethical approval was sought for secondary data analysis (ERGO number 18829, see appendix A). All data were anonymised before analysis.

2.2.6 Statistical methods and data preparation

Independent t-tests were carried out to compare whether the CAT and CBT treatment groups differed significantly at any time point across the PHQ-9, GAD-7, PHQ-15, WSAS, SHAI, inpatient cost, outpatient cost, and A&E cost (see table 2.1). As they were not significantly different on any variable at any time point, the two therapies could therefore be combined into ‘Cognitive Therapy’ for the purpose of analysis. This was done because t-tests are more robust when sample sizes between groups are equal, so pooling the two samples in this way served to make the sample size of the treatment group (N = 48, when combined into Cognitive Therapy) comparable to that of the TAU group (N = 56) when analysing economic outcomes. Pooling the samples also increased the sample size and therefore the statistical power, increasing the reliability of the analyses. Histograms revealed the presence of two distinct groups for ethnicity (White British and Other), number of MUS (one MUS and two or more MUS) symptoms, and duration of MUS symptoms (less than 50 months’ duration and 50 months or longer). These variables were therefore recoded into binary variables.

Paired samples t-tests evaluated treatment effects of the psychosocial and physical distress outcome variables and effect sizes were calculated using Cohen’s *d.* To be conservative, the PHQ15, WSAS and SHAI were bootstrapped with 1000 samples due to a small number of outliers and slight kurtosis. For the secondary hypotheses, 2 x 2 repeated measures ANOVAs searched for possible interactions between gender, ethnicity, number of MUS, and MUS duration on pre- and post-therapy outcome measures. As Mauchly’s test of sphericity was not significant for any variable, corrections were not required in the main analysis. Effect sizes were given as partial eta squared. Independent t-tests compared the healthcare costs for TAU and Cognitive Therapy at 6 months, 12 months, and 18 months post- therapy completion. These comparisons were bootstrapped with 1000 samples due to the presence of outliers. Where effect sizes indicated the need, multiple regressions were used to compare TAU and Cognitive Therapy across the different time points, adjusting for baseline cost.

Outliers were not excluded from the analysis of healthcare cost, as they were felt to be a valid reflection of clinical experience and it did not seem appropriate to exclude these. A smaller number of outliers also existed among psychosocial outcomes, however these were also included as the outcome measures have pre-determined maximum possible scores. Instead, analyses of variables containing outliers were bootstrapped with 1000 samples.

2.3 Results

2.3.1 Descriptive statistics

There were no significant differences between CBT, CAT and TAU on any of the baseline characteristics or measures or between the CBT and CAT group on psychological and physical health measures (table 2.1). As stated in section 2.2.5, CBT and CAT were therefore grouped together as the ‘Cognitive Therapy’ group for future analyses, in order to increase the sample size and statistical power. The CAT group received an average of 15.20 sessions of therapy (*SD* = 5.40), while the CBT group received 11.70 sessions on average (*SD* = 4.78) and while this difference was significant (*p* = 0.03), it carried no effect, *d* = 0.06.

Table 2.1.

Baseline characteristics of participants in two intervention groups and control group

Characteristic	CBT $N = 31$	CAT $N = 17$	TAU $N = 56$	CBT vs CAT	Cognitive Therapy vs TAU
Gender					
Female, no. (%)	21 (68)	10 (59)	37 (66)	$\chi^2(1) = 0.38$, $p = 0.54$, $v = 0.09$	$\chi^2(1) = 0.03$, $p = 0.87$, $v = 0.02$
Male, no. (%)	10 (32)	7 (41)	19 (34)		
Ethnicity					
White British, no. (%)	21 (68)	10 (59)	37 (66)	$\chi^2(1) = 0.38$, $p = 0.54$, $v = 0.09$	$\chi^2(1) = 6.18$, $p = 0.05$, $v = 0.24$
Other ethnicity, no. (%)	10 (32)	7 (41)	19 (34)		
MUS no.					
One MUS, no. (%)	25 (81)	9 (53)	40 (75)	$\chi^2(1) = 4.08$, $p = 0.10$, $v = 0.21$	$\chi^2(1) = 0.28$, $p = 0.6$, $v = 0.05$
Two or more MUS, no. (%)	6 (19)	8 (47)	13 (25)		
MUS duration					
Less than 50 months’, no. (%)	11 (35)	9 (53)	22 (39)	$\chi^2(1) = 1.38$, $p = 0.24$, $v = 0.17$	$\chi^2(1) = 0.06$, $p = 0.81$, $v = 0.02$
50 months or longer, no. (%)	20 (65)	8 (47)	34 (61)		

Table 2.1.

Continued

Characteristic	CBT *N* = 31	CAT *N* = 17	TAU *N* = 56	CBT vs CAT	Cognitive Therapy vs TAU
Psychosocial and physical health					
PHQ-9, *M* (*SD*)	13.00 (7.24)	12.65 (6.18)		*t*(46) = 0.17, *p* = 0.87, *d* = 0.05	
GAD-7, *M* (*SD*)	9.68 (6.45)	11.53 (6.12)		*t*(46) = -0.97, *p* = 0.34, *d* = -0.29	
PHQ-15, *M* (*SD*)	12.00 (4.97)	14.24 (4.89)		*t*(46) = -1.5, *p* = 0.14, *d* = -0.45, 95% CI [-5.15, 0.53]	
WSAS, *M* (*SD*)	16.07 (11.02)	17.24 (9.62)		*t*(46) = -0.37, *p* = 0.72, *d* = -0.11, 95% CI [-7.31, 4.68]	
SHAI, *M* (*SD*)	24.10 (11.19)	25.41 (10.21)		*t*(45) = -0.4, *p* = 0.69, *d* = -0.12, 95% CI [-7.64, 4.71]	
Inpatient cost, *M* (*SD*)	444.91 (1432.74)	237.44 (469.38)	344.61 (997.68)	*t*(36) = 0.56, *p* = 0.58, 95% CI [-306.42, 928.33], *d* = 0.18	*t*(92) = 0.59, *p* = 0.96, 95% CI [-354.69, 500.82], *d* = 0.12
Outpatient cost, *M* (*SD*)	237.55 (484.06)	92.19 (112.71)	233.53 (566.51)	*t*(36) = 1.17, *p* = 0.25, 95% CI [-25.05, 410.01], *d* = 0.39	*t*(91) = -0.54, *p* = 0.57, 95% CI [-255.06, 130.52], *d* = -0.11
A&E cost, *M* (*SD*)	30.71 (90.84)	37.38 (80.25)	89.95 (193.65)	*t*(36) = -0.23, *p* = 0.82, 95% CI [-63.44, 45.85], *d* = -0.08	*t*(92) = -1.69, *p* = 0.09, 95% CI [-117.09, -3.15], *d* = -0.35

Note. Number of MUS *N* = 53
* < 0.05 ** <0.01 *** <0.001

2.3.2 Primary hypotheses

2.3.2.1 Within-group differences in psychosocial and physical distress. Table 2.2 shows that depression and generalised anxiety both reduced significantly in the cognitive therapy group at the end of treatment compared to at the start, both with intermediate effects. Work and social functioning, somatic symptoms, and health anxiety also improved significantly after therapy compared to at the start, although effects here were smaller.

Table 2.2.

Pre- and post-therapy psychological and physical distress in Cognitive Therapy group

	Pre	Post			
	M (*SD*)	*M* (*SD*)	*t* (*df*)	95% CI	*d*
PHQ-9 *N* = 48	12.88 (6.82)	8.52 (7.11)	5.57 (47) ***	[2.78, 5.93]	0.63
GAD-7 *N* = 48	10.33 (6.33)	7.19 (6.09)	4.60 (47) ***	[1.77, 4.52]	0.51
PHQ-15 *N* = 48	12.79 (5.01)	11.00 (5.6)	3.26 (47) **	[0.69, 2.85]	0.33
WSAS *N* = 48	16.48 (10.45)	13.04 (10.8)	2.94 (47) **	[1.13, 5.63]	0.32
SHAI *N* = 43	24.28 (10.56)	20.67 (10.86)	3.96 (42) **	[1.92, 5.36]	0.34

Note. PHQ-15, WSAS, and SHAI confidence intervals based on 1000 bootstrapped samples
* < 0.05 ** <0.01 *** <0.001

2.3.2.2 Between-group differences in inpatient cost at 6, 12, and 18 months. There was no significant difference between the Cognitive Therapy group and TAU on inpatient cost at 6 months, 12 months, and 18 months after treatment ended (see table 2.3). However, small trends in the data indicated that the Cognitive Therapy group on average had greater inpatient costs at 6 months and at 12 months, while the TAU group had greater average costs at 18 months after treatment ended. These differences were therefore controlled for in further analyses, however group still did not predict outcomes (see tables 2.4-2.6).

Table 2.3.

Between-group differences in inpatient cost at 6 months, 12 months, and 18 months

	Cognitive Therapy	TAU			
	M (SD)	*M (SD)*	*t* (*df*)	95% CI	*d*
Inpatient cost at 6 months	826.16 (2375.16)	511.20 (1440.24)	0.73 (78)	[-583.18, 1518.48]	0.18
Inpatient cost at 12 months	273.72 (944.23)	206.34 (457.21)	0.37 (57)	[-280.36, 617.39]	0.11
Inpatient cost at 18 months	426.71 (1369.91)	740.82 (2225.72)	-0.54 (54)	[-1209.75, 674.94]	-0.16

Note. At 6 months Cognitive Therapy *N* = 25, TAU *N* = 55. At 12 months Cognitive Therapy *N* = 18, TAU *N* = 41. At 18 months Cognitive Therapy *N* = 17, TAU *N* = 39
* < 0.05 ** <0.01 *** <0.001

Table 2.4.

Multiple regression of predictors of inpatient cost at 6 months after treatment end

	b	95% CI	*SE B*	*β*	*p*
Step 1					
Baseline IP cost	0.56	[-0.03, 0.75]	0.25	0.35	0.02*
Step 2					
Baseline IP cost	0.56	[-0.002, 0.75]	0.25	0.35	0.02*
Group	-299.19	[-1420.45, 554.97]	481.89	-0.08	0.59

Note. R^2 = 0.12 for Step 1, p = 0.001**; ΔR^2 = 0.01 for Step 2, p = 0.46
95% bias corrected and accelerated confidence intervals in parentheses. Confidence intervals and standard errors based on 1000 bootstrapped samples

Table 2.5.

Multiple regression of predictors of inpatient cost at 12 months after treatment end

	b	95% CI	*SE B*	*β*	*p*
Step 1					
Baseline IP cost	0.91	[-0.14, 1.27]	0.43	0.49	0.03*
Step 2					
Baseline IP cost	0.90	[-0.12, 1.28]	0.43	0.48	0.03*
Group	-285.52	[-2168.99, 1111.64]	652.27	-0.06	0.68

Note. R^2 = 0.24 for Step 1, p = 0.00**; ΔR^2 = 0.003 for Step 2, p = 0.63
95% bias corrected and accelerated confidence intervals in parentheses. Confidence intervals and standard errors based on 1000 bootstrapped samples

Table 2.6.

Multiple regression of predictors of inpatient cost at 18 months after treatment end

	b	95% CI	*SE B*	*β*	*p*
Step 1					
Baseline IP cost	1.17	[-0.35, 1.84]	0.76	0.45	0.05*
Step 2					
Baseline IP cost	1.17	[-0.39, 1.87]	0.77	0.45	0.05*
Group	5.32	[-2177.49, 2337.97]	848.14	0.001	0.99

Note. R^2 = 1.2 for Step 1, p = 0.000**; ΔR^2 = 0.000 for Step 2, p = 0.1
95% bias corrected and accelerated confidence intervals in parentheses. Confidence intervals and standard errors based on 1000 bootstrapped samples

2.3.2.3 Between-group differences in outpatient cost at 6, 12, and 18 months. There was no significant difference between the Cognitive Therapy group and TAU on outpatient cost at 6 months, 12 months, and 18 months after treatment ended (see table 2.7). However, small trends in the data indicated that the TAU group on average had greater outpatient costs at 6 months and at 12 months. These differences were therefore controlled for in further analyses, however group still did not predict outcomes (see tables 2.8-2.9).

Table 2.7.

Between-group differences in outpatient cost at 6 months, 12 months, and 18 months

	Cognitive Therapy	TAU			
	M (SD)	*M (SD)*	*t* (*df*)	95% CI	*d*
Outpatient cost at 6 months	89.74 (128.91)	150.26 (291.96)	-0.99 (77)	[-160.17, 20.94]	-0.24
Outpatient cost at 12 months	128.83 (189.09)	196.64 (333.71)	-0.80 (55)	[-212.70, 69.69]	-0.23
Outpatient cost at 18 months	187.39 (228.82)	159.44 (301.27)	0.35 (52)	[-104.16, 183.42]	-0.10

Note. At 6 months Cognitive Therapy *N* = 25, TAU *N* = 54. At 12 months Cognitive Therapy *N* = 18, TAU *N* = 39. At 18 months Cognitive Therapy *N* = 18, TAU *N* = 36
* < 0.05 ** <0.01 *** <0.001

Table 2.8.

Multiple regression of predictors of outpatient cost at 6 months after treatment end

	b	95% CI	*SE B*	*β*	*p*
Step 1					
Baseline outpatient cost	0.37	[0.14, 0.44]	0.11	0.78	0.14
Step 2					
Baseline outpatient cost	0.37	[0.13, 0.44]	0.10	0.77	0.001**
Group	49.44	[-6.71, 100.92]	32.83	0.09	0.15

Note. R^2 = 0.6 for Step 1, *p* = 0.000**; ΔR^2 = 0.008 for Step 2, *p* = 0.21
95% bias corrected and accelerated confidence intervals in parentheses. Confidence intervals and standard errors based on 1000 bootstrapped samples

Table 2.9.

Multiple regression of predictors of outpatient cost at 12 months after treatment end

	b	95% CI	*SE B*	*β*	*p*
Step 1					
Baseline outpatient cost	0.63	[0.26, 0.73]	0.15	0.76	0.001**
Step 2					
Baseline outpatient cost	0.63	[0.23, 0.72]	0.15	0.76	0.001**
Group	117.21	[-20.91, 258.49]	76.25	0.11	0.15

Note. R^2 = 0.58 for Step 1, p = 0.000**; ΔR^2 = 0.01 for Step 2, p = 0.21
95% bias corrected and accelerated confidence intervals in parentheses. Confidence intervals and standard errors based on 1000 bootstrapped samples

2.3.2.4 Between-group differences in A&E cost at 6, 12, and 18 months

There was no significant difference between the Cognitive Therapy group and TAU on A&E cost at 6 months, 12 months, and 18 months after treatment ended (see table 2.10). However, small trends in the data indicated that the TAU group on average had greater A&E costs at 12 months. This difference was therefore controlled for in further analysis, however group still did not predict outcomes (see table 2.11).

Table 2.10.

Between-group differences in A&E cost at 6 months, 12 months, and 18 months

	Cognitive Therapy	TAU			
	M (SD)	*M (SD)*	*t* (*df*)	95% CI	*d*
A&E cost at 6 months	40.20 (101.44)	55.06 (112.15)	-0.56 (77)	[-60.69, 35.09]	-0.14
A&E cost at 12 months	22.50 (76.01)	37.80 (86.97)	-0.64 (56)	[-56.97, 35.06]	-0.18
A&E cost at 18 months	51.44 (82.22)	70.62 (191.44)	-0.41 (53)	[-94.90, 47.67]	-0.12

Note. At 6 months Cognitive Therapy *N* = 25, TAU *N* = 54. At 12 months Cognitive Therapy *N* = 18, TAU *N* = 40. At 18 months Cognitive Therapy *N* = 18, TAU *N* = 37
* < 0.05 ** <0.01 *** <0.001

Table 2.11.

Multiple regression of predictors of A&E cost at 12 months after treatment end

	b	95% CI	*SE B*	*β*	*p*
Step 1					
Baseline A&E cost	0.35	[-0.01, 1.79]	0.38	0.33	0.18
Step 2					
Baseline A&E cost	0.35	[-0.04, 1.81]	0.38	0.33	0.18
Group	6.31	[-99.51, 97.49]	50.77	0.02	0.91

Note. $R^2 = 0.11$ for Step 1, $p = 0.01$**; $\Delta R^2 = 0.000$ for Step 2, $p = 0.91$
95% bias corrected and accelerated confidence intervals in parentheses. Confidence intervals and standard errors based on 1000 bootstrapped samples

2.3.3 Secondary hypotheses

2.3.3.1 Impact of participant variables on psychosocial and physical distress. 2 x 2 repeated measures ANOVAs revealed that neither gender nor ethnicity significantly predicted the improvements seen to depression, generalised anxiety, health anxiety, somatic symptoms, or work and social functioning (see tables 2.12 and 2.13). However, some trends did emerge; small effects were found for both gender and ethnicity on depression and health anxiety, such that females showed slightly larger improvements on both scales, while White British participants showed slightly larger improvements in depression and ethnic minorities showed larger improvements in health anxiety. There was also a small effect for gender on work and social functioning, with females showing a slightly larger improvement than males. A small effect was also found for ethnicity on generalised anxiety, with ethnic minorities showing larger improvements here than their White British counterparts.

Table 2.12.

Interactions between gender and time on psychosocial and physical distress in Cognitive Therapy group

	Females		Males				
	Pre	Post	Pre	Post			
	M (*SD*)	*M* (*SD*)	*M* (*SD*)	*M* (*SD*)	*F* (*df*)	*p*	η_p^2
PHQ-9 *N* = 48	11.26 (6.11)	6.39 (5.00)	15.82 (7.23)	12.41 (8.77)	0.79 (1, 46)	0.38	0.02
GAD-7 *N* = 48	9.06 (6.07)	5.71 (5.18)	12.65 (6.31)	9.88 (6.83)	0.17 (1, 46)	0.68	0.004
PHQ-15 *N* = 48	11.65 (3.95)	9.94 (4.34)	14.88 (6.09)	12.94 (7.10)	0.04 (1, 46)	0.84	0.001
WSAS *N* = 48	15.16 (10.12)	10.77 (9.13)	18.88 (10.93)	17.18 (12.57)	1.21 (1, 46)	0.28	0.03
SHAI *N* = 43	23.31 (10.10)	18.96 (10.34)	25.76 (11.38)	23.29 (11.43)	1.01 (1, 41)	0.32	0.02

* < 0.05 ** <0.01 *** <0.001

Table 2.13.

Interactions between ethnicity and time on psychosocial and physical distress in Cognitive Therapy group

	White British		Ethnic Minorities				
	Pre	Post	Pre	Post			
	M (*SD*)	*M* (*SD*)	*M* (*SD*)	*M* (*SD*)	*F* (*df*)	*p*	η_p^2
PHQ-9 *N* = 47	13.11 (6.19)	9.03 (7.00)	13.27 (8.22)	7.64 (7.58)	0.68 (1, 45)	0.41	0.02
GAD-7 *N* = 47	10.50 (6.25)	7.53 (6.32)	10.64 (6.54)	6.45 (5.63)	0.55 (1, 45)	0.46	0.01
PHQ-15 *N* = 47	12.94 (4.79)	11.28 (5.38)	12.27 (6.10)	10.45 (6.60)	0.01 (1, 45)	0.91	0.00
WSAS *N* = 47	17.47 (10.35)	14.44 (10.25)	14.00 (11.00)	9.55 (12.04)	0.25 (1, 45)	0.62	0.006
SHAI *N* = 42	24.61 (10.21)	21.55 (10.31)	24.27 (12.03)	19.18 (12.67)	0.91 (1, 40)	0.35	0.02

* < 0.05 ** <0.01 *** <0.001

As can be seen from tables 2.14 and 2.15, neither the number nor the duration of MUS at baseline significantly predicted the above improvements to psychosocial and physical distress. Once again, some trends did emerge however; small effects were found for the number of MUS on both depression and somatic symptoms. Specifically, those with just one unexplained symptom at baseline generally showed larger improvements to depression and somatic symptoms. Duration of MUS also showed a small effect on health anxiety outcomes, such as that those whose symptoms had been present for less than 50 months at the time of referral showed slightly larger improvements in health anxiety than those whose symptoms had been present for more than 50 months.

Table 2.14.

Interactions between number of MUS and time on psychosocial and physical distress in Cognitive Therapy group

	One MUS		Two or more MUS				
	Pre	Post	Pre	Post			
	M (*SD*)	*M* (*SD*)	*M* (*SD*)	*M* (*SD*)	*F* (*df*)	*p*	η_p^2
PHQ-9 *N* = 48	13.26 (6.67)	8.44 (7.40)	11.93 (7.34)	8.71 (6.62)	0.87 (1, 46)	0.36	0.02
GAD-7 *N* = 48	10.76 (6.18)	7.50 (6.31)	9.29 (6.79)	6.43 (5.67)	0.07 (1, 46)	0.79	0.002
PHQ-15 *N* = 48	12.47 (5.20)	10.21 (5.23)	13.57 (4.59)	12.93 (6.17)	1.84 (1, 46)	0.18	0.04
WSAS *N* = 48	16.88 (11.54)	13.74 (11.68)	15.50 (7.46)	11.36 (8.41)	0.15 (1, 46)	0.70	0.003
SHAI *N* = 43	23.90 (11.23)	20.50 (11.36)	26.15 (9.19)	21.08 (10.03)	0.11 (1, 41)	0.74	0.003

* < 0.05 ** <0.01 *** <0.001

Table 2.15.

Interactions between duration of MUS and time on psychosocial and physical distress in Cognitive Therapy group

	Less than 50 months		50 months or longer				
	Pre	Post	Pre	Post			
	M (*SD*)	*M* (*SD*)	*M* (*SD*)	*M* (*SD*)	*F* (*df*)	*p*	η_p^2
PHQ-9 *N* = 48	13.40 (6.40)	9.45 (7.78)	12.50 (7.19)	7.86 (6.66)	0.19 (1, 46)	0.67	0.004
GAD-7 *N* = 48	11.55 (6.16)	8.80 (6.39)	9.46 (6.41)	6.04 (5.71)	0.24 (1, 46)	0.63	0.005
PHQ-15 *N* = 48	12.90 (3.97)	11.0 (5.76)	12.71 (5.70	10.93 (5.58)	0.00 (1, 46)	0.99	0.00
WSAS *N* = 48	15.95 (11.50)	13.25 (11.86)	16.86 (9.84)	12.89 (10.20)	0.28 (1, 46)	0.60	0.006
SHAI *N* = 43	25.29 (9.67)	20.82 (11.55)	23.62 (11.24)	20.58 (10.62)	0.58 (1, 41)	0.45	0.01

Note. PHQ-15, WSAS, and SHAI confidence intervals based on 1000 bootstrapped samples
* < 0.05 ** <0.01 *** <0.001

2.4 Discussion

The main aims of the study were to demonstrate that both CBT and CAT would significantly reduce psychosocial and physical distress in patients with MUS immediately after therapy compared to at the start. The study also sought to demonstrate that both therapies would reduce healthcare costs compared to a TAU group. As expected, both CBT and CAT significantly reduced levels of depression, generalised anxiety, health anxiety, somatic symptom severity, and perceived impaired work and social functioning, at the end of therapy compared to at the start, with small to intermediate effect sizes. Moreover, there was no notable difference between the two treatments in terms of these improvements while controlling for baseline levels. The effects identified in this study for the effectiveness of cognitive therapy are in line with the existing literature into CBT's effectiveness for MUS and its related conditions (see for instance the 2014 Cochrane review by Van Dessel et al.), where reductions in distress have come with generally small effects. However, the finding that CAT provided comparable outcomes had not been demonstrated until now and adds to the earlier literature into its efficacy for explained physical health conditions such as type 1 diabetes (Forsbury et al., 1997).

That both treatments appear to be equally effective with this client group also lends strength to the possibility that the two interventions may be working through similar mechanisms. Alternatively, the finding may be due to the matching of participants to treatment type based on clinical need in the present study. It is therefore unclear whether clients who were offered CAT would have done as well with CBT and vice versa. In particular, there appears to be a subset of MUS clients who have interpersonal problems as a result of insecure attachments (Delfstra & van Rooij, 2015). Indeed, in a study into IBS by Hyphantis et al. (2009), they found that an improvement in interpersonal problems after psychodynamic interpersonal therapy was associated with reduced psychological distress and in turn, improved health status. Likewise, the study by Forsbury et al. (1997) observed greater interpersonal change among the group of diabetes patients receiving CAT compared to controls receiving no psychological therapy. Therefore, one reason why CAT may have been equally as effective as CBT in the present sample is because it was offered to clients who early on exhibited interpersonal problems and where the more relational focus of CAT was expected to be more effective (Ryle, 2010). In the present study, while interpersonal problems were observed clinically, it may have been useful to administer an interpersonal questionnaire, such as the Inventory of Interpersonal Problems (Barkham, Hardy, & Startup, 1996), at baseline. This would have determined whether CBT and CAT clients indeed differed significantly at baseline on interpersonal features. Such a measure could then be used as an objective tool to supplement clinical decision making, when considering whether a client ought to receive CBT or CAT for their MUS. This presents a crucial avenue of further research, so that services may consider whether both treatments ought to be available in order to adequately treat these symptoms. However, despite the research limitations that the lack of randomisation carries in the present study, this is nonetheless a pragmatic and accurate reflection of what happens in applied settings, where a therapy may be offered to a client based on their particular presentation.

Contrary to expectations, the present study was unable to demonstrate a significant reduction in inpatient, outpatient or A&E costs at any time point between those who received cognitive therapy and those who received treatment as usual. Indeed, even when considering instances where small effects were seen for either the treatment or TAU group costing more, there was no clear trend that emerged. This is most likely to be due to the fact that information was not collected about the reasons for participants using primary healthcare services, that is to say whether they were attending because of unexplained physical symptoms or for other, explained complaints. Interestingly, reductions in healthcare costs have also not been demonstrated consistently in the literature and it is notable that in many cases, no mention is made as to whether the different reasons for attendance were taken into account.

Secondary aims of the study were to demonstrate that participants would improve regardless of gender, while ethnic minorities would show less improvement. A greater number and duration of symptoms at baseline was also expected to predict greater improvement after psychological therapy. As expected, regardless of gender both males and females improved significantly across measures of depression, generalised anxiety, health anxiety, somatic symptom severity, and perceived impaired work and social functioning. It is noteworthy that males entered therapy with higher scores on their baseline psychosocial and physical measures than females. It is possible that, while MUS and somatoform disorders are more common in females (Van Dessel et al., 2014), males might be taking longer to access services, at which point their symptoms and difficulties have worsened. The present study also found that having symptoms that had been around for more than 50 months made no significant difference to one’s chances of improving in depression, generalised anxiety, health anxiety, somatic symptom severity, and perceived impaired work and social functioning. While this is in contrast to work by Lackner et al., (2007), who found that more long-standing symptoms of IBS appeared to respond better to CBT, their finding disappeared once covariates were controlled for. The findings are in line with research by Speckens et al. (1997), who found that neither number nor duration of symptoms at the start correlated with outcomes at the end, or one year later. The present study additionally demonstrated that having more than two unexplained symptoms did not make a significant difference to improvements after cognitive therapy. This finding is at odds with the rest of the literature, which appears to suggest that a greater number of symptoms is correlated with improved outcomes (Bleichhardt et al., 2005 and Heider & Zaby, 2011). Finally, it was hypothesised that ethnic minorities would show less improvement after cognitive therapy than their White British counterparts. This also did not turn out to be the case, with no significant difference in psychosocial and physical outcomes between the White British and ethnic minority group. It has been documented that many black and minority ethnic (BME) communities will seek mental health support from religious leaders in the first instance, due to the belief that their symptoms are caused by bad spirits. This is less true for younger BME individuals, who are more likely to ascribe to a Western medical model and will instead visit their GP first (Glasgow Anti Stigma Partnership, 2007). It is possible that the BME participants in the present study were part of this younger generation who were (a) more open to a bio-psychosocial way of treating their symptoms (and therefore responded just as well as their White British counterparts to cognitive therapy) and (b) were willing to report symptoms in the first instance (and therefore had similar symptoms at baseline as their White British counterparts). However, without having been able to control for the variable of age in the present study, it is difficult to know for sure whether this is the reason for a lack of significant difference between White British and BME participants' outcomes.

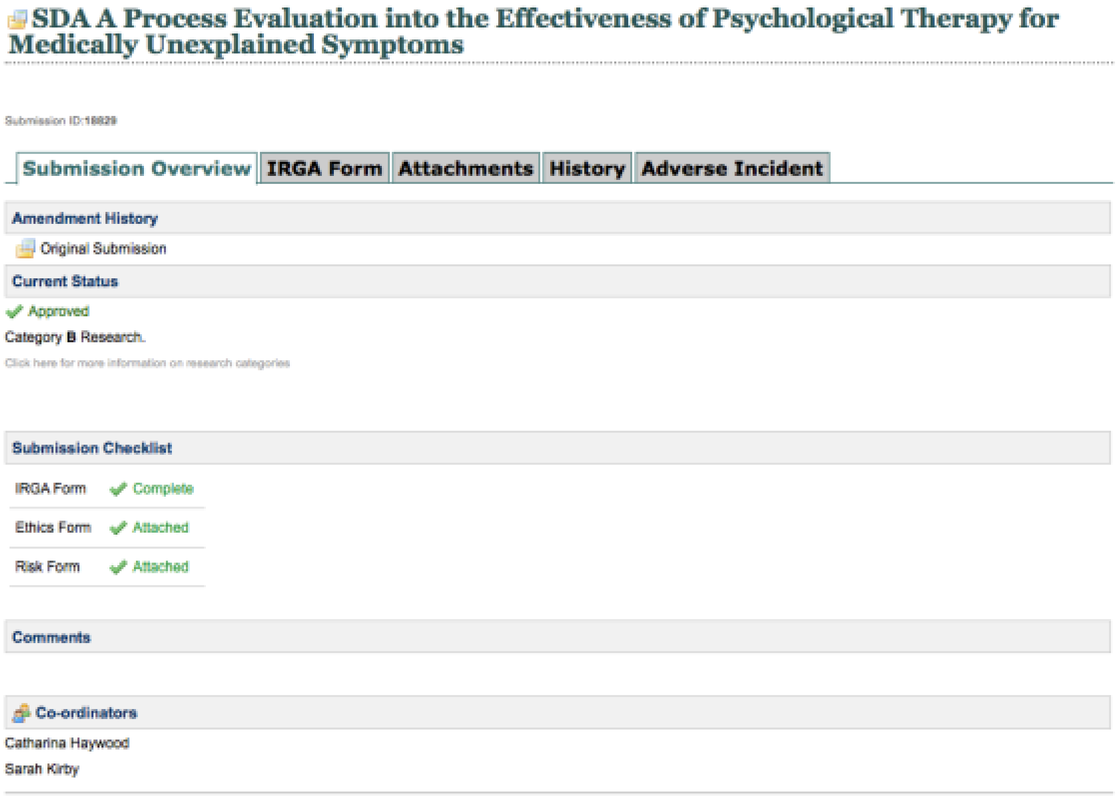
2.4.1 Limitations and directions for future research

There are some important limitations to the present study, most notably that participants were not randomly allocated to either treatment or TAU groups, but were instead matched to either therapy based on clinical presentation. However, while randomised control trials are arguably the gold standard of research, randomisation may not always be feasible or ethical and it is important to strike a balance between practice-based evidence and evidence-based practice (Calvert & Kellett, 2014), at least to inform future directions of research. Secondly, on measures of generalised anxiety, depression, health anxiety, somatic symptoms, and work and social impairment, there was no control group with which to compare the treatment group, as well as a lack of follow up beyond the end of therapy. Related to this, there is also a risk of positive selection bias, as those who did not engage with treatment (despite being offered it) were classed as the control group, however because they never completed the psychosocial and physical outcome measures, it is unclear whether they would have had less favourable outcomes. Sample sizes were also generally somewhat small, particularly the initial CAT group and these methodological limitations should be improved upon in future research designs. Moreover, drop-out rates were not compared between the two psychological therapies and it is possible that other factors such as levels of motivation may have differed between the two therapy samples. Future research would benefit from comparing the two treatments in terms of their acceptability with this patient group, such as by investigating drop-out rates, while investigating specific mechanisms of change within the two therapies will enable researchers to understand how it is that they might bring about similar effects in this client group. A further question that remains unanswered is that, if patients were reporting significant improvements to their generalised anxiety, depression, health anxiety, somatic symptoms, and work and social impairment, why is it that they continued to incur healthcare costs as much as before therapy and as much as a control group that did not receive therapy. As reported earlier, the present study was not able to differentiate between MUS and non-MUS appointments and it is possible that many appointments were routine visits. Future cost analyses should include larger samples and differentiate between MUS-related healthcare costs and non-MUS related costs before drawing firm conclusions. Secondly, qualitative interview methods might go some way to explaining why patients continued to use services, while longer follow-up periods for psychosocial outcome measures, as well as some measure of illness beliefs and attitudes would help to assess for cognitive change.

2.4.2 Implications for clinical practice

Promisingly, the present study suggests that, regardless of gender or ethnicity, a person stands a good chance of recovery in terms of their reported mental and physical health and social functioning. As men might be waiting for their symptoms to worsen before accessing services, psychoeducation aimed at men might encourage them to seek help sooner, particularly considering the finding that women are more likely to seek support for mental health difficulties (Department of Health, 2008). Moreover, it may be motivating for services to know that neither the number nor the duration of symptoms are likely to impact on how well a person can recover after receiving either CAT or CBT. This is particularly relevant considering how, for many, such unexplained symptoms may have been a feature of their lives for some time (Rief & Hiller, 1992). Finally, being the first paper to demonstrate the efficacy of CAT with this client group can further the evidence base and provide clinicians with an additional therapeutic tool for those clients where relational difficulties are more prominent. It is hoped that this research will contribute to future National Institute for Health and Care Excellence (NICE) guidance, which currently makes no recommendations for MUS or somatoform disorders other than it seems IBS (NICE, 2017) and CFS (NICE, 2007).

Appendix A Southampton University School of Psychology Ethics Committee and Research Governance Approval



SDA A Process Evaluation into the Effectiveness of Psychological Therapy for Medically Unexplained Symptoms

Submission ID:18829

Amendment History

Original Submission

Current Status

✓ Approved

Category **B** Research.

Click here for more information on research categories

Submission Checklist

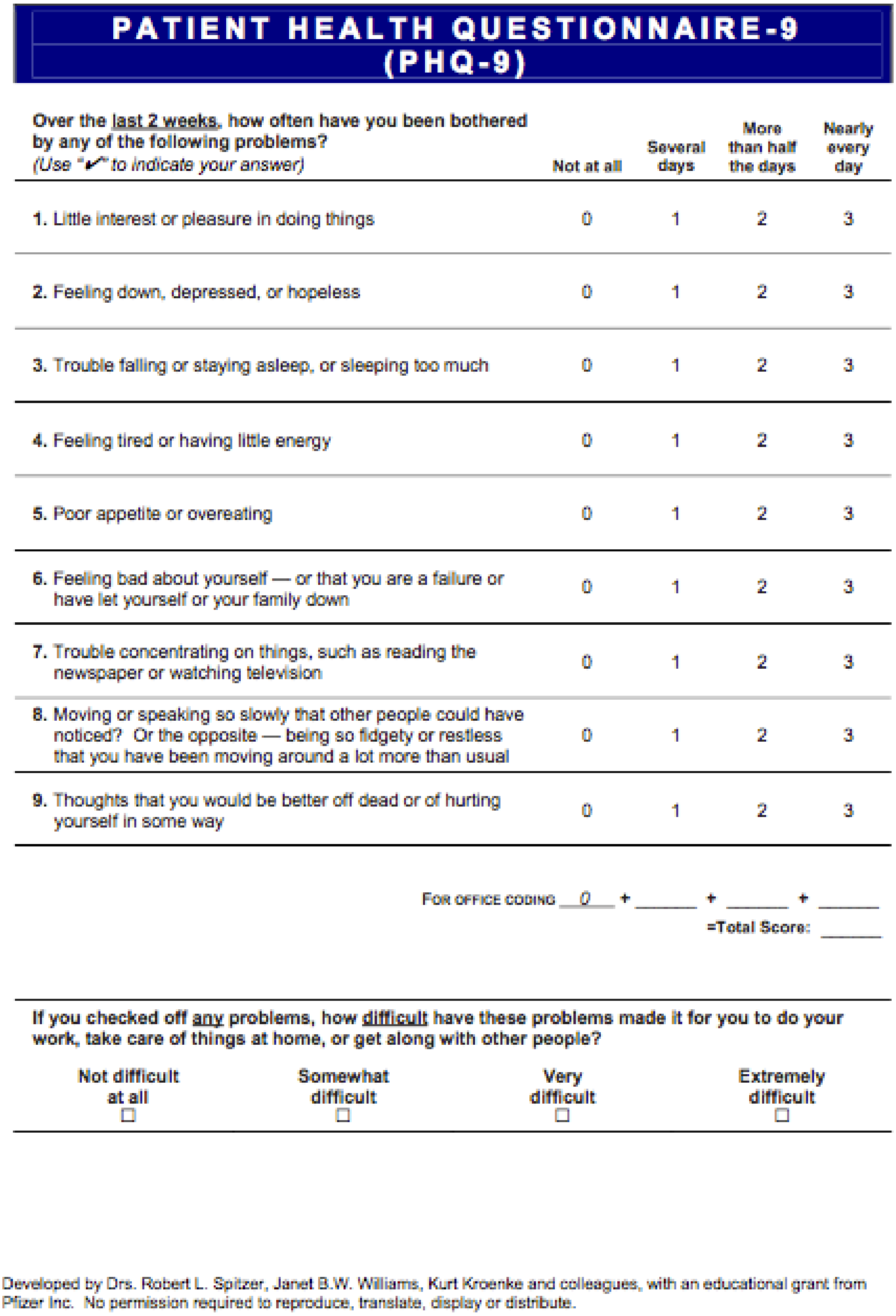
IRGA Form	✓ Complete
Ethics Form	✓ Attached
Risk Form	✓ Attached

Comments

Co-ordinators

Catharina Haywood
Sarah Kirby

Appendix B Patient Health Questionnaire – 9 (PHQ-9)



PATIENT HEALTH QUESTIONNAIRE-9 (PHQ-9)

Over the last 2 weeks, how often have you been bothered by any of the following problems? *(Use "✔" to indicate your answer)*	Not at all	Several days	More than half the days	Nearly every day
1. Little interest or pleasure in doing things	0	1	2	3
2. Feeling down, depressed, or hopeless	0	1	2	3
3. Trouble falling or staying asleep, or sleeping too much	0	1	2	3
4. Feeling tired or having little energy	0	1	2	3
5. Poor appetite or overeating	0	1	2	3
6. Feeling bad about yourself — or that you are a failure or have let yourself or your family down	0	1	2	3
7. Trouble concentrating on things, such as reading the newspaper or watching television	0	1	2	3
8. Moving or speaking so slowly that other people could have noticed? Or the opposite — being so fidgety or restless that you have been moving around a lot more than usual	0	1	2	3
9. Thoughts that you would be better off dead or of hurting yourself in some way	0	1	2	3

FOR OFFICE CODING __0__ + ______ + ______ + ______

=Total Score: ______

If you checked off any problems, how difficult have these problems made it for you to do your work, take care of things at home, or get along with other people?

Not difficult at all	Somewhat difficult	Very difficult	Extremely difficult
☐	☐	☐	☐

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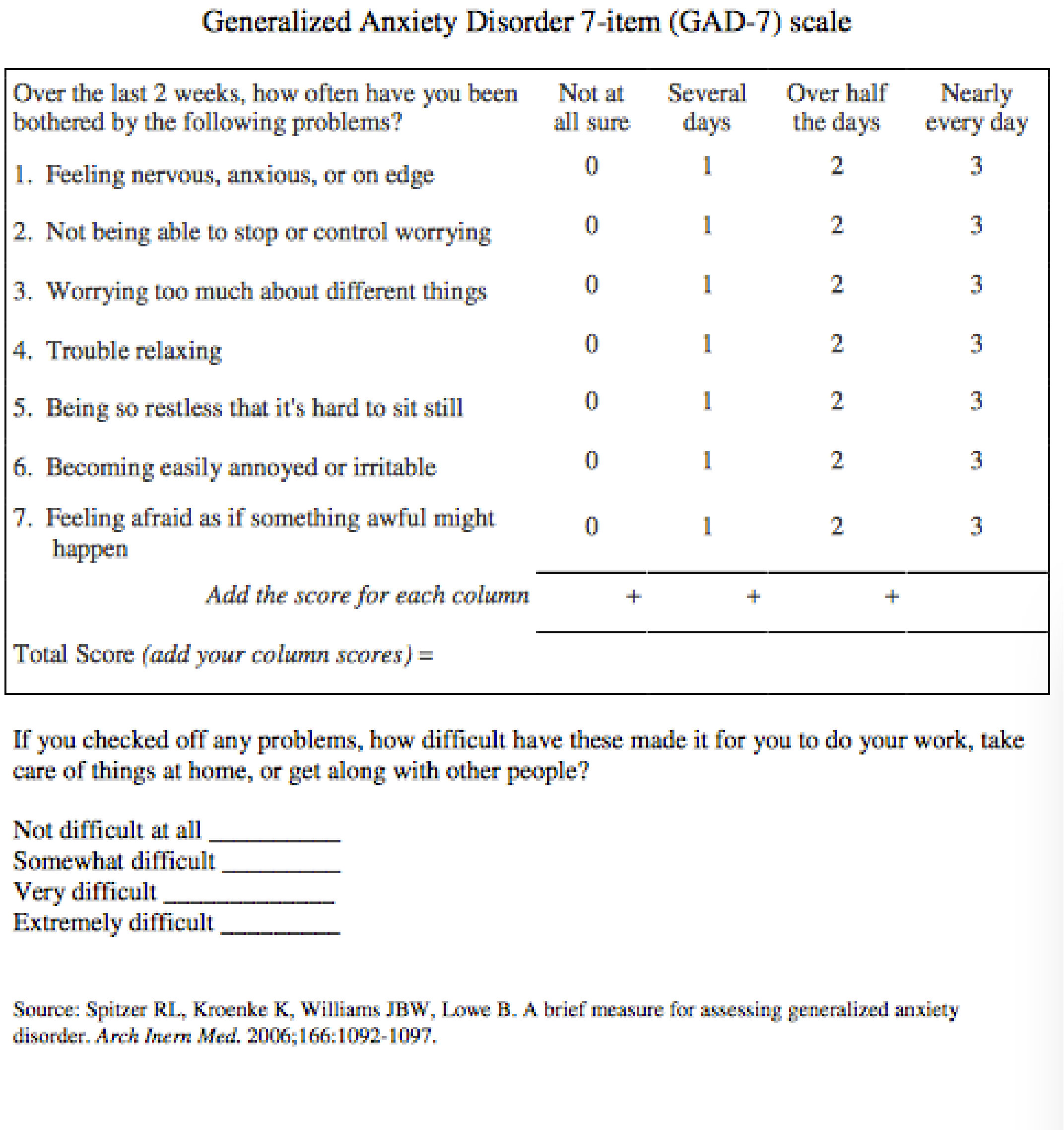
Appendix C Generalized Anxiety Disorder Questionnaire - 7 (GAD-7)



Generalized Anxiety Disorder 7-item (GAD-7) scale

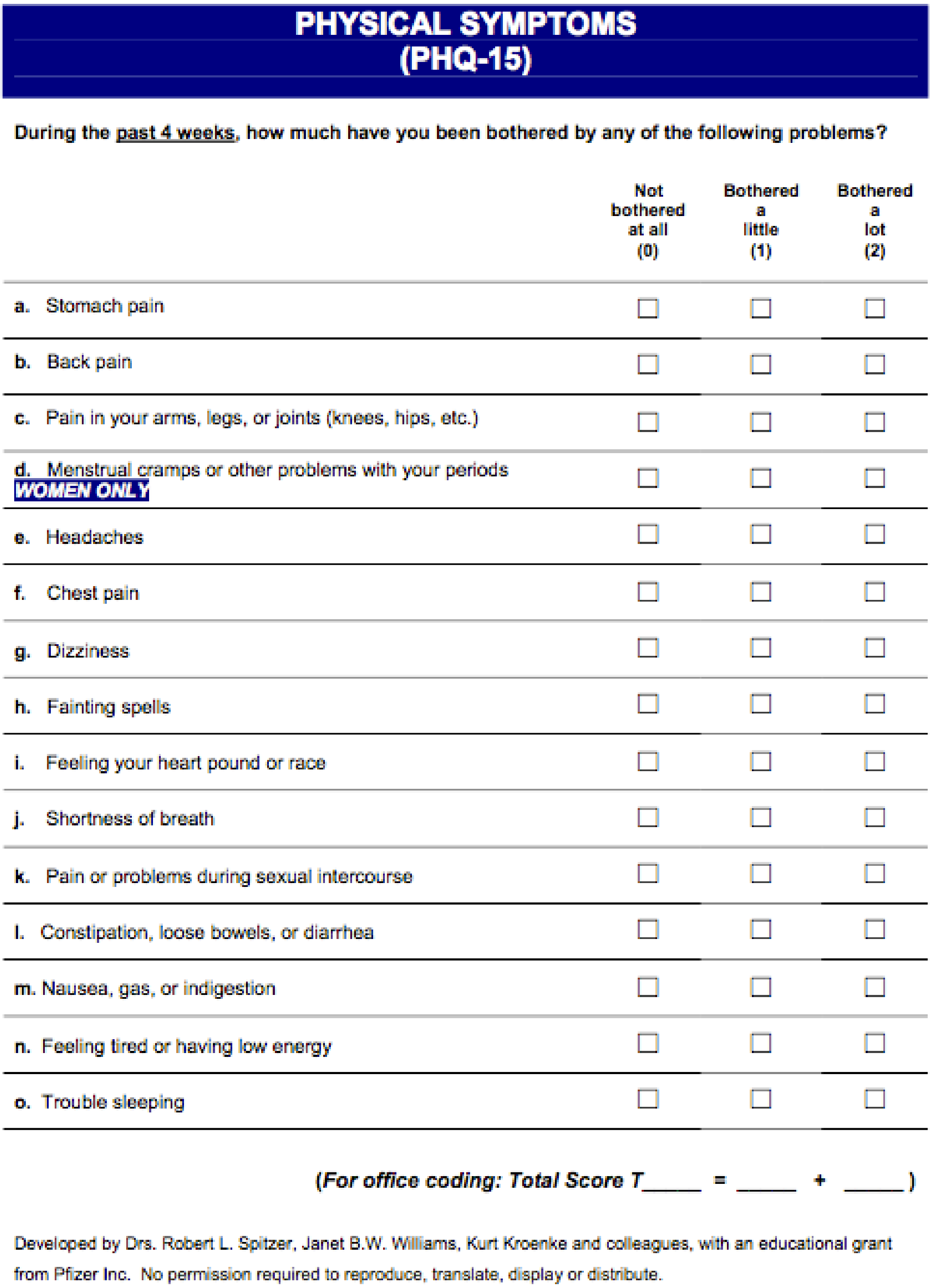
Over the last 2 weeks, how often have you been bothered by the following problems?	Not at all sure	Several days	Over half the days	Nearly every day
1. Feeling nervous, anxious, or on edge	0	1	2	3
2. Not being able to stop or control worrying	0	1	2	3
3. Worrying too much about different things	0	1	2	3
4. Trouble relaxing	0	1	2	3
5. Being so restless that it's hard to sit still	0	1	2	3
6. Becoming easily annoyed or irritable	0	1	2	3
7. Feeling afraid as if something awful might happen	0	1	2	3
Add the score for each column	+	+	+	
Total Score *(add your column scores)* =				

If you checked off any problems, how difficult have these made it for you to do your work, take care of things at home, or get along with other people?

Not difficult at all __________
Somewhat difficult _________
Very difficult _____________
Extremely difficult _________

Source: Spitzer RL, Kroenke K, Williams JBW, Lowe B. A brief measure for assessing generalized anxiety disorder. *Arch Inern Med.* 2006;166:1092-1097.

Appendix D Patient Health Questionnaire – 15 (PHQ-15)



PHYSICAL SYMPTOMS (PHQ-15)

During the <u>past 4 weeks</u>, how much have you been bothered by any of the following problems?

	Not bothered at all (0)	Bothered a little (1)	Bothered a lot (2)
a. Stomach pain	☐	☐	☐
b. Back pain	☐	☐	☐
c. Pain in your arms, legs, or joints (knees, hips, etc.)	☐	☐	☐
d. Menstrual cramps or other problems with your periods ***WOMEN ONLY***	☐	☐	☐
e. Headaches	☐	☐	☐
f. Chest pain	☐	☐	☐
g. Dizziness	☐	☐	☐
h. Fainting spells	☐	☐	☐
i. Feeling your heart pound or race	☐	☐	☐
j. Shortness of breath	☐	☐	☐
k. Pain or problems during sexual intercourse	☐	☐	☐
l. Constipation, loose bowels, or diarrhea	☐	☐	☐
m. Nausea, gas, or indigestion	☐	☐	☐
n. Feeling tired or having low energy	☐	☐	☐
o. Trouble sleeping	☐	☐	☐

(For office coding: Total Score T_____ = _____ + _____)

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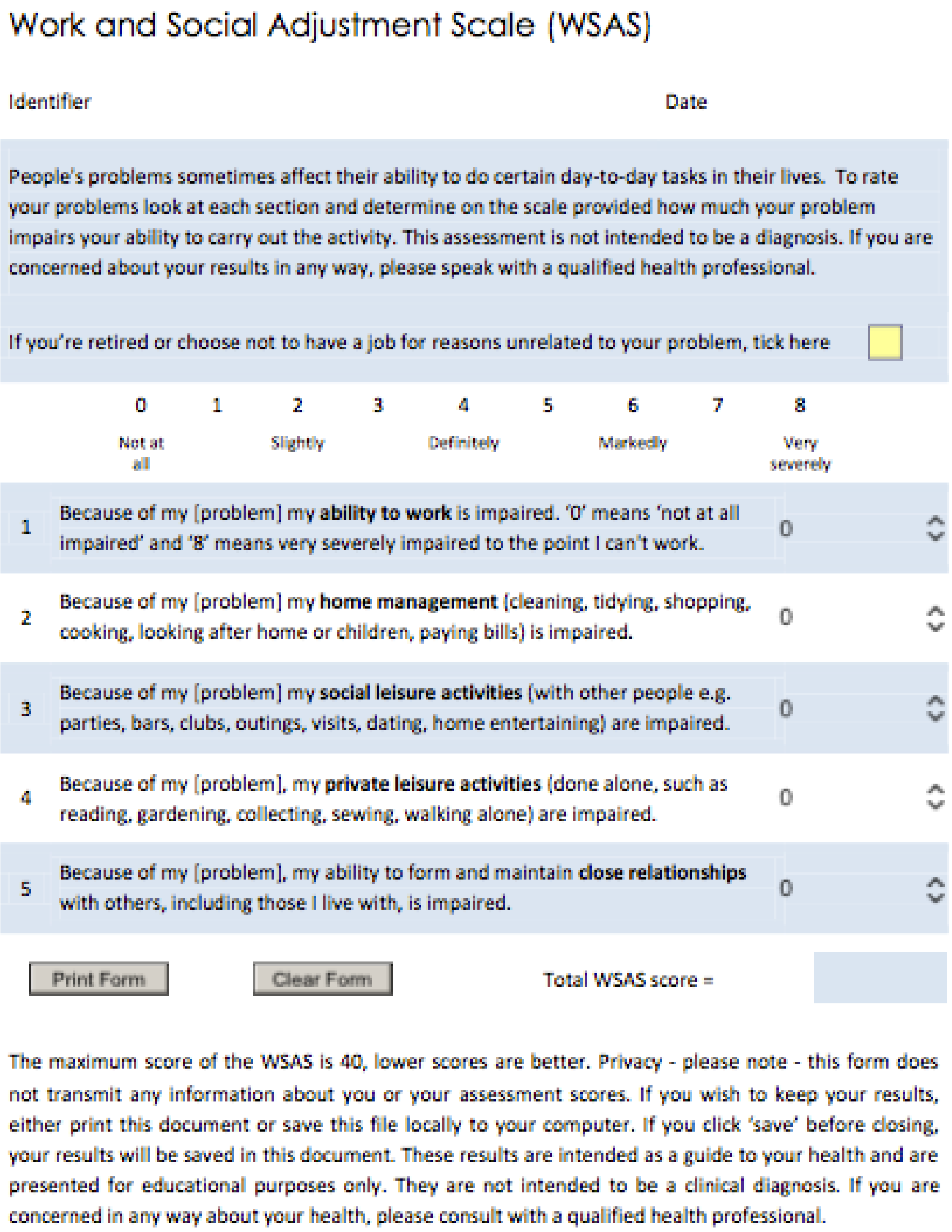
Appendix E Work and Social Adjustment Scale (WSAS)



Work and Social Adjustment Scale (WSAS)

Identifier

Date

People's problems sometimes affect their ability to do certain day-to-day tasks in their lives. To rate your problems look at each section and determine on the scale provided how much your problem impairs your ability to carry out the activity. This assessment is not intended to be a diagnosis. If you are concerned about your results in any way, please speak with a qualified health professional.

If you're retired or choose not to have a job for reasons unrelated to your problem, tick here ☐

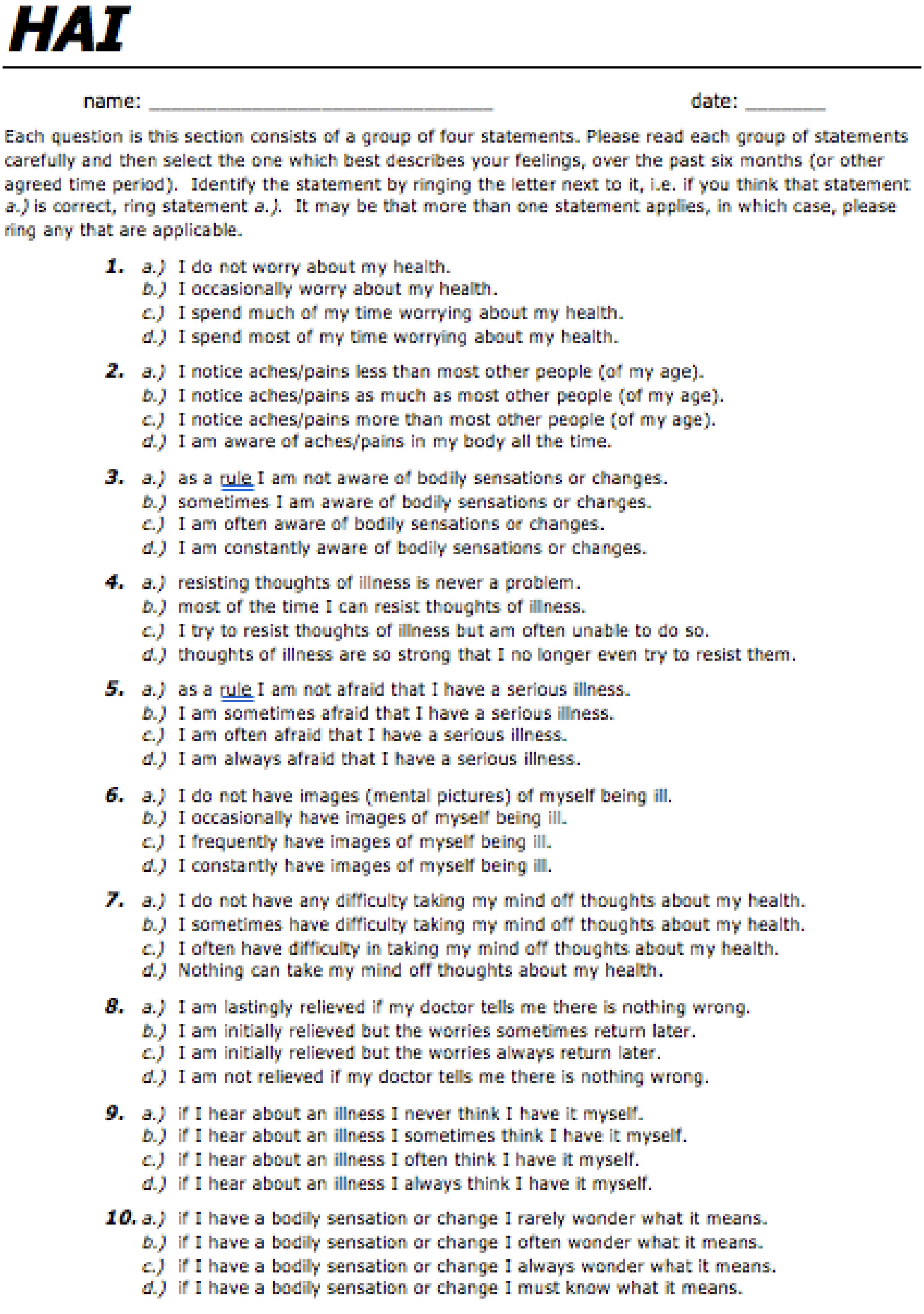
0	1	2	3	4	5	6	7	8
Not at all		Slightly		Definitely		Markedly		Very severely

1	Because of my [problem] my **ability to work** is impaired. '0' means 'not at all impaired' and '8' means very severely impaired to the point I can't work.	0
2	Because of my [problem] my **home management** (cleaning, tidying, shopping, cooking, looking after home or children, paying bills) is impaired.	0
3	Because of my [problem] my **social leisure activities** (with other people e.g. parties, bars, clubs, outings, visits, dating, home entertaining) are impaired.	0
4	Because of my [problem], my **private leisure activities** (done alone, such as reading, gardening, collecting, sewing, walking alone) are impaired.	0
5	Because of my [problem], my ability to form and maintain **close relationships** with others, including those I live with, is impaired.	0

Total WSAS score =

The maximum score of the WSAS is 40, lower scores are better. Privacy - please note - this form does not transmit any information about you or your assessment scores. If you wish to keep your results, either print this document or save this file locally to your computer. If you click 'save' before closing, your results will be saved in this document. These results are intended as a guide to your health and are presented for educational purposes only. They are not intended to be a clinical diagnosis. If you are concerned in any way about your health, please consult with a qualified health professional.

Appendix F Short Health Anxiety Inventory (SHAI)



HAI

name: ______________________________ date: _______

Each question is this section consists of a group of four statements. Please read each group of statements carefully and then select the one which best describes your feelings, over the past six months (or other agreed time period). Identify the statement by ringing the letter next to it, i.e. if you think that statement *a.)* is correct, ring statement *a.)*. It may be that more than one statement applies, in which case, please ring any that are applicable.

1. *a.)* I do not worry about my health.
b.) I occasionally worry about my health.
c.) I spend much of my time worrying about my health.
d.) I spend most of my time worrying about my health.

2. *a.)* I notice aches/pains less than most other people (of my age).
b.) I notice aches/pains as much as most other people (of my age).
c.) I notice aches/pains more than most other people (of my age).
d.) I am aware of aches/pains in my body all the time.

3. *a.)* as a rule I am not aware of bodily sensations or changes.
b.) sometimes I am aware of bodily sensations or changes.
c.) I am often aware of bodily sensations or changes.
d.) I am constantly aware of bodily sensations or changes.

4. *a.)* resisting thoughts of illness is never a problem.
b.) most of the time I can resist thoughts of illness.
c.) I try to resist thoughts of illness but am often unable to do so.
d.) thoughts of illness are so strong that I no longer even try to resist them.

5. *a.)* as a rule I am not afraid that I have a serious illness.
b.) I am sometimes afraid that I have a serious illness.
c.) I am often afraid that I have a serious illness.
d.) I am always afraid that I have a serious illness.

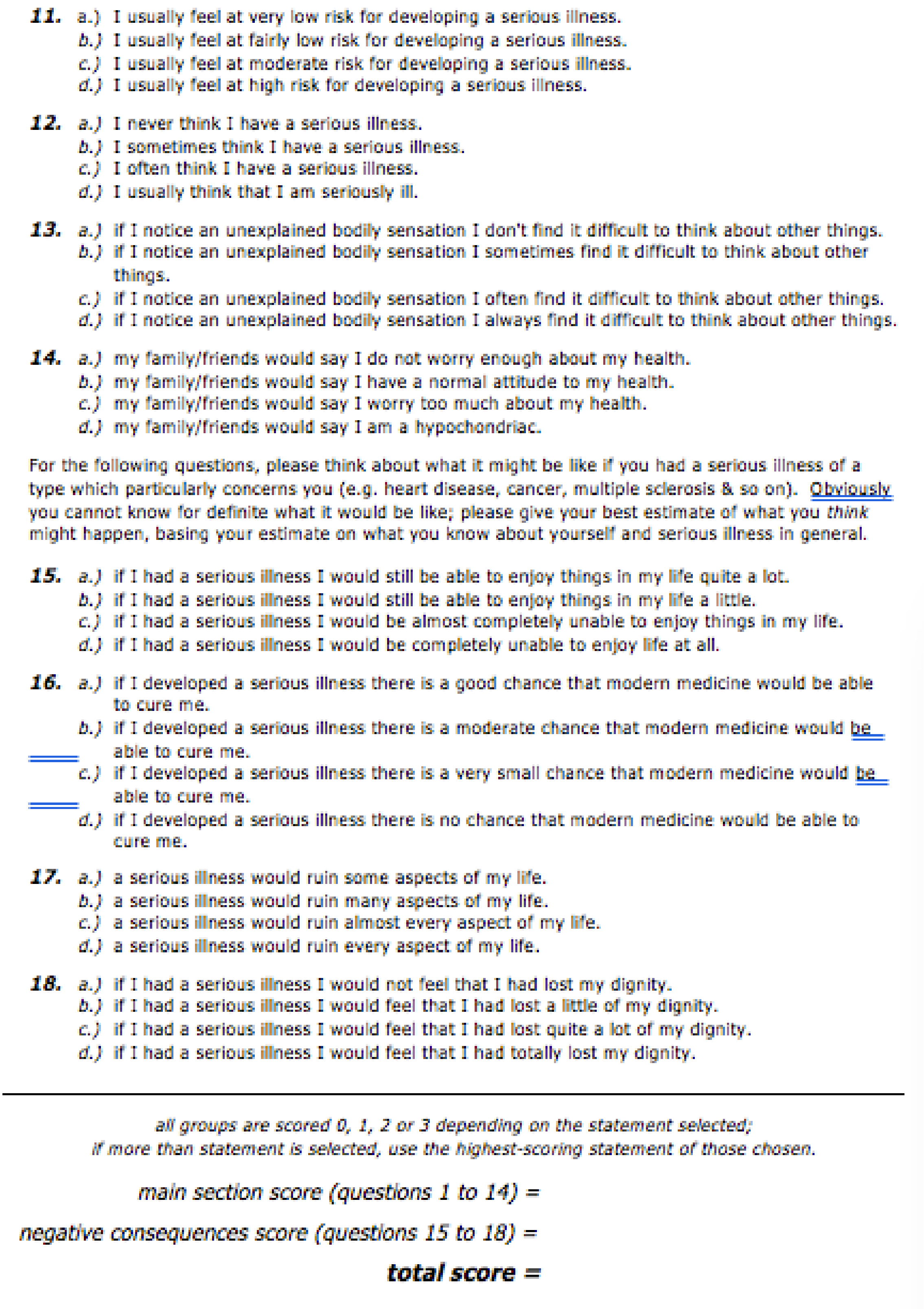
6. *a.)* I do not have images (mental pictures) of myself being ill.
b.) I occasionally have images of myself being ill.
c.) I frequently have images of myself being ill.
d.) I constantly have images of myself being ill.

7. *a.)* I do not have any difficulty taking my mind off thoughts about my health.
b.) I sometimes have difficulty taking my mind off thoughts about my health.
c.) I often have difficulty in taking my mind off thoughts about my health.
d.) Nothing can take my mind off thoughts about my health.

8. *a.)* I am lastingly relieved if my doctor tells me there is nothing wrong.
b.) I am initially relieved but the worries sometimes return later.
c.) I am initially relieved but the worries always return later.
d.) I am not relieved if my doctor tells me there is nothing wrong.

9. *a.)* if I hear about an illness I never think I have it myself.
b.) if I hear about an illness I sometimes think I have it myself.
c.) if I hear about an illness I often think I have it myself.
d.) if I hear about an illness I always think I have it myself.

10. *a.)* if I have a bodily sensation or change I rarely wonder what it means.
b.) if I have a bodily sensation or change I often wonder what it means.
c.) if I have a bodily sensation or change I always wonder what it means.
d.) if I have a bodily sensation or change I must know what it means.



11. a.) I usually feel at very low risk for developing a serious illness.
b.) I usually feel at fairly low risk for developing a serious illness.
c.) I usually feel at moderate risk for developing a serious illness.
d.) I usually feel at high risk for developing a serious illness.

12. *a.)* I never think I have a serious illness.
b.) I sometimes think I have a serious illness.
c.) I often think I have a serious illness.
d.) I usually think that I am seriously ill.

13. *a.)* if I notice an unexplained bodily sensation I don't find it difficult to think about other things.
b.) if I notice an unexplained bodily sensation I sometimes find it difficult to think about other things.
c.) if I notice an unexplained bodily sensation I often find it difficult to think about other things.
d.) if I notice an unexplained bodily sensation I always find it difficult to think about other things.

14. *a.)* my family/friends would say I do not worry enough about my health.
b.) my family/friends would say I have a normal attitude to my health.
c.) my family/friends would say I worry too much about my health.
d.) my family/friends would say I am a hypochondriac.

For the following questions, please think about what it might be like if you had a serious illness of a type which particularly concerns you (e.g. heart disease, cancer, multiple sclerosis & so on). Obviously you cannot know for definite what it would be like; please give your best estimate of what you *think* might happen, basing your estimate on what you know about yourself and serious illness in general.

15. *a.)* if I had a serious illness I would still be able to enjoy things in my life quite a lot.
b.) if I had a serious illness I would still be able to enjoy things in my life a little.
c.) if I had a serious illness I would be almost completely unable to enjoy things in my life.
d.) if I had a serious illness I would be completely unable to enjoy life at all.

16. *a.)* if I developed a serious illness there is a good chance that modern medicine would be able to cure me.
b.) if I developed a serious illness there is a moderate chance that modern medicine would be able to cure me.
c.) if I developed a serious illness there is a very small chance that modern medicine would be able to cure me.
d.) if I developed a serious illness there is no chance that modern medicine would be able to cure me.

17. *a.)* a serious illness would ruin some aspects of my life.
b.) a serious illness would ruin many aspects of my life.
c.) a serious illness would ruin almost every aspect of my life.
d.) a serious illness would ruin every aspect of my life.

18. *a.)* if I had a serious illness I would not feel that I had lost my dignity.
b.) if I had a serious illness I would feel that I had lost a little of my dignity.
c.) if I had a serious illness I would feel that I had lost quite a lot of my dignity.
d.) if I had a serious illness I would feel that I had totally lost my dignity.

all groups are scored 0, 1, 2 or 3 depending on the statement selected;
if more than statement is selected, use the highest-scoring statement of those chosen.

main section score (questions 1 to 14) =

negative consequences score (questions 15 to 18) =

total score =

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