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The acceptability of primary care or community-based behavioural interventions for persistent physical symptoms: Qualitative systematic review

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Purpose. Persistent physical symptoms (PPS) are often associated with profound physical disability and psychological distress. Interventions for PPS that promote behavioural change aim to reduce levels of symptoms and improve overall functioning in patients. The evidence for these interventions is mixed, with effective relationships between patients and health practitioners (HPs) reported as the key to the success of primary care interventions. The objectives of this systematic review were to synthesize the qualitative evidence and to evaluate the acceptability of behavioural interventions for PPS in primary care, from the perspective of both patients and HPs.

Methods. A comprehensive literature search was conducted in seven major electronic bibliographic databases, to February 2019. The aim was to identify a broad range of literature including, qualitative research, mixed methods research, and qualitative data embedded in trial reports or process evaluations. Fifty-eight full papers were screened against the inclusion criteria. Nine studies were included and quality-assessed. A qualitative evidence synthesis was conducted using thematic synthesis.

Results. Some patients and HPs reported positive gains from taking part in or delivering interventions, with appropriate support and explanation of their symptoms important for patients. Barriers appeared to be underpinned by the relationship between the patients and HPs, and by beliefs and attitudes held by both parties.

Conclusions. Patients should be provided with adequate information to make an informed decision about whether an intervention is appropriate for them, and interventions should not end suddenly or without adequate follow-up. HPs should receive training and supervision to address their lack of confidence, and improve their knowledge of PPS.

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Statement of contribution

What is already known on this subject?

- Evidence for interventions for PPS that aim to promote behavioural change is mixed, with some showing positive results and others unable to draw conclusions on effectiveness.
- The relationship between patients and service providers is reported as key to the success of interventions, with poor communication between parties, and lack of emotional and practical support suggested barriers to effective intervention.

What does this study add?

- Patients valued being supported and being provided with explanations for their symptoms.
- Training for health professionals was thought to be helpful and may address barriers.
- Interventions should not end suddenly or without adequate follow-up or booster sessions.

Background

Persistent physical symptoms ‘PPS’ is a portfolio term covering a wide range of presentations in referring to persistent bodily complaints, including pain and discomfort. PPS are often associated with psychological distress and functional impairment (Dirkzwager & Verhaak, 2007). The term has also been applied to patients presenting with chronic fatigue syndrome, also termed myalgic encephalomyelitis (CFS/ME); irritable bowel syndrome (IBS); or fibromyalgia, which are usually referred to as functional somatic syndromes (FSS) (Wessely, Nimnuan, & Sharpe, 1999). The term PPS is used in this paper rather than medically unexplained symptoms (MUS). The term MUS is controversial, and debate regarding its use is ongoing. For many patients with symptoms that are not, in the current state of medical knowledge, readily explicable by organic disease, a diagnostic label is important. However, the label ‘medically unexplained symptoms’ can be regarded as offensive (Stone et al., 2002), and it has been suggested that the use of the term is a barrier to improved care (Creed et al., 2010). Results of an online survey of healthy adults demonstrated that the most popular term to refer to persistent bodily complaints, including pain and discomfort, was PPS and was more acceptable than the term MUS (Marks & Hunter, 2015). A further study (Picariello, Ali, Moss-Morris, & Chalder, 2015) demonstrated that patients with CFS preferred the term PPS to describe their own condition and also illnesses such as IBS and non-cardiac chest pain. Whilst we have used the term PPS in this paper, previous academic literature has generally used the term MUS; therefore, it was necessary to use that term whilst conducting our literature searches, and we have kept references to MUS in this paper where appropriate in order to accurately report these previous findings.

Diagnostic criteria for PPS are varied, and patients visiting their GP frequently with unexplained symptoms are not necessarily offered a formal diagnosis. In the United Kingdom, Taylor, Marshall, Mann, and Goldberg (2012) report a prevalence rate of 18% in consecutive attenders to UK GP practices. Worldwide prevalence rates of primary care patients presenting with PPS have been reported as between 25 and 50% (Edwards, Stern, Clarke, Ivbijaro, & Kasney, 2010). It has been estimated that 50-75% of patients with PPS will improve, whilst between 10 and 30% will see their condition deteriorate (Olde Hartman et al., 2009). However, a recent study has reported that only 11% of UK primary care patients had recovered at 6 months and 55% were still having persistent symptoms (Lamahewa, Buszewicz, Walters, Marston, & Nazareth, 2019).

Generally, interventions offered for PPS, across a variety of health settings, are based around pharmacological, psychological, or physical therapeutic models. Pharmacological interventions have been shown to produce some improvement in patients in terms of symptom severity and functioning (Ford, Talley, Schoenfeld, Quigley, & Moayyedi, 2009; Hoedeman, Blankenstein, Krol, Koopmans, & Groothoff, 2010; Kroenke, 2007), although with significant heterogeneity of efficacy between different FSS.

The evidence for interventions that aim to promote cognitive, emotional, and/or behavioural change is mixed, with cognitive behaviour therapy (CBT) appearing to have the most consistent evidence. CBT for PPS is based on the broader model which suggests an acute event such as injury or infection precipitates the symptoms which are then perpetuated through an interaction of cognitive, behavioural, emotional, and physiological factors. CBT and behavioural-based therapies aim to alter one or more of these factors to reduce the severity and/or impact of the symptoms. For instance, worry about symptoms can heighten autonomic nervous responses and sensitization to symptoms. Prolonged periods of inactivity due to the fear that activity is harmful may impact sleep and muscle integrity and exacerbate symptoms such as pain and fatigue. For fibromyalgia, CBT has been shown to improve physical symptoms and functioning (Arnold et al., 2012), as have exercise therapies (Busch, Barber, Overend, Peloso, & Schachter, 2007; Marcus, 2009). A recent network meta-analysis of the psychological interventions for IBS concluded CBT and gut-directed hypnotherapy had the largest evidence base and were the most effective at reducing the severity of IBS long term (Black et al., 2020). However, the authors noted that there was significant heterogeneity between studies and issues regarding trial design, including lack of blinding, which may mean treatment effects were overestimated. A 24-month follow-up of the largest CBT trial for IBS to date showed that effects were largely at sustained at 24-month follow-up compared to usual care (Everitt et al., 2019). A Cochrane review of CBT for CFS (Price et al., 2008) concluded CBT was more effective at reducing fatigue at the end of treatment than usual care and wait list control as well as other therapies such as relational counselling. The data for longer term follow-up were more mixed. The review also concluded that there was a paucity of studies on the acceptability and possible side effects of CBT.

General practitioners (GPs) play a major role in identifying and managing patients with PPS. Most patients with PPS will be seen initially in primary care, often for many consultations, prior to referral to secondary care, or less often specialist services. However, the majority of the research in this patient group has been conducted in secondary care with less evidence specific to the primary care setting. The relationship between patients and service providers has been reported as key to the success of primary care interventions in PPS in a number of studies (Gask, Dowrick, Salmon, Peters, & Morriss, 2011; Heijmans et al., 2011; Smith et al. 2003). Poor communication between GPs and patients during medical consultations, and lack of emotional and practical support are suggested barriers to effective recognition and treatment of PPS (Murray, Toussaint, Althaus, & Löwe, 2016). These barriers may limit opportunities to explore contributing factors and appropriate interventions. Creating a safe, therapeutic environment and offering effective reassurance are important enabling factors (Heijmans et al., 2011), thus highlighting the need for acceptability of interventions for both patients and health care practitioners.

This is the first qualitative systematic review, to our knowledge, to specifically explore the acceptability of behavioural interventions for diverse PPS populations based in primary care, including patients meeting the criteria for MUS, medically unexplained physical symptoms (MUPS), and somatoform disorders, and including populations with

specific FSS, for example, IBS, CFS, and fibromyalgia. Previous qualitative systematic reviews have only examined patients' experiences more generally, in terms of how patients, family, and the medical community interpret the illness, not relating to experiences of undertaking behavioural interventions (Anderson, Jason, Hlavaty, Porter, & Cudia, 2012). The objectives of this study were to synthesize evidence relating to the barriers and facilitators of the acceptability of primary care or community-based behavioural interventions for Persistent Physical Symptoms, in the United Kingdom, from the perspective of both patients and service providers, by undertaking a comprehensive systematic review of the available qualitative research literature, using rigorous methods for review and evidence synthesis.

Methods

This systematic review was pre-registered at PROSPERO.

Search methods

The search methods sought to identify evidence in the form of qualitative research, qualitative data reported in mixed methods research, and qualitative data embedded in trial reports or process evaluations. A systematic search strategy was developed by a library information specialist (ASu) to identify this evidence relating to a primary care or community-based population; population terms were combined with terms that define the setting. Published methodological search filters to limit results to qualitative studies were used where available (ISSG, 2008) and were combined with a geographic filter to identify UK studies only. No other search limits were applied. Initial search results therefore covered the period from 1980 to July 2016, updated in February 2019, in seven electronic databases including MEDLINE, Embase, CINAHL, PsycINFO, Science Citation Index, and Social Sciences Citation Index. Citation searching and reference list searching were also undertaken to identify any further citations that may not have appeared in electronic databases (see Supporting Information for further information).

Screening and eligibility

Titles and abstracts of all citations retrieved through the searches were scrutinized by one systematic reviewer (ASc) according to the inclusion and exclusion criteria (Table 1). Agreement on inclusion at title/abstract sift was checked by a second systematic reviewer (AB) for 20% of the total electronic search results. Agreement was calculated using the Kappa statistic. Given a Kappa statistic of 0.77 (i.e., above recognized acceptable levels [0.7]), double sifting was not deemed necessary. All full texts were considered for inclusion by two reviewers (ASc and AB). Disagreements on study inclusion were resolved with reference to a third reviewer (JL).

Quality assessment strategy

Inclusion criteria for study design were broad, with any study presenting qualitative evidence being eligible. Therefore, a study quality checklist for qualitative studies was developed prior to study screening (using an adapted form of the CASP checklist for qualitative research), to allow flexibility in accommodating findings from studies with

Table 1. Inclusion and exclusion criteria

| | Included | Excluded |
|------------------|---|--|
| P – Population | Patients meeting the criteria for PPS, MUS, MUPS, and somatoform disorders. Populations with FSS were included, for example, IBS, CFS, and fibromyalgia. Health care providers who had delivered behavioural interventions designed for these patients were also included | Subacute patients. Patients with intermittent pain (where current episode was less than 3 months – or this information was not available from the paper / or they cannot be disentangled from the rest of the sample.) |
| I – Intervention | Behavioural interventions delivered in a primary care or community-based setting, in the United Kingdom (<i>only UK studies were eligible for inclusion as this study aimed to synthesis evidence relating specifically to the UK primary care setting</i>) | Studies of management of PPS where evidence relating to a treatment of interest was not separately identifiable |
| C – Comparator | N/A | N/A |
| O – Outcomes | Qualitative data | N/A |
| S – Study design | Qualitative research, mixed methods research, and qualitative data embedded in trial reports or process evaluations | Quantitative reports without qualitative evidence |

research designs that were not primarily qualitative in nature (see Table 2). No studies were excluded on the basis of quality (Carroll & Booth, 2015).

Quality constituted one aspect of the assessment of the confidence of findings from the included studies, using CERQual as described below in the methods of synthesis section.

Data extraction strategy

Data extraction was undertaken by one reviewer (ASc) using a data extraction tool tailored for the purpose of this review. All data extractions were checked by a second reviewer (AB), with discrepancies discussed by both reviewers. Given the anticipated paucity of relevant evidence, an inclusive approach to data extraction was employed in which all qualitative data identified in the primary studies and relevant to the review question were extracted (Noyes & Lewin, 2011). The framework for extraction allowed data from the primary studies to be extracted verbatim into general categories relating to the review question, directly from the papers, as illustrated in Table 3 below. This allowed familiarization with the data and was akin to assigning preliminary codes to the data, which described the content.

Data synthesis strategy

Thematic synthesis was used to aggregate the data (Thomas & Harden, 2008). Themes were developed within the data extraction elements (Table 3). Specifically, one reviewer (ASc) went through the data assigning preliminary codes line-by-line according to its meaning and content. These codes were then organized and grouped together to become sub-themes, which were largely descriptive, and which were later grouped into over-

Table 2. The Quality Assessment Questions applied to included studies

| Question | |
|----------|---|
| 1 | Is the study qualitative research or does it provide qualitative data |
| 2 | Is the study context and aims clearly described? |
| 3 | Is there evidence of research reflexivity? |
| 4 | Are the sampling methods clearly described and appropriate for the research question? |
| 5 | Are the methods of data collection clearly described and appropriate to the research question? |
| 6 | Is the method of analysis clearly described and appropriate to the research question? |
| 7 | Are the claims made supported by sufficient evidence (i.e., Did the data provide sufficient depth detail and richness)? |

Table 3. Framework for data extraction

| | |
|--------------------------------|---|
| Data from patients | Positive factors relating to behavioural interventions as reported by patients / What did you gain from being referred to a behavioural intervention Factors reported as important, wanted or expected in behavioural interventions What patients didn't like about (being referred to) behavioural interventions Barriers – why patients didn't want behavioural interventions / or couldn't engage with them Neutral effects (neither benefits or harms) |
| Data from health professionals | Positive factors relating to behavioural interventions as reported by health professionals / What did you gain from being trained to and/or delivering a behavioural intervention Factors reported as important, wanted or expected for training in and delivery of behavioural interventions What did not help or was detrimental to the patients or delivery of the intervention Barriers – from the perspective of health professionals as to why patients didn't want behavioural interventions/or couldn't engage with them, or barriers to delivery of the intervention Neutral effects (neither benefits or harms) |

arching themes, which were more 'analytical'. Line-by-line coding allowed the translation of concepts from one study to another; as each study was coded, new data were added to existing codes, and new codes were developed as necessary. These sub-themes remained very close to the original findings, whilst the over-arching themes went beyond the findings of the original study to suggest new understandings, based on the judgement and insights of the reviewers.

The Confidence in the Evidence from Reviews of Qualitative research (GRADE-CERQual) (Lewin et al., 2015) approach was used to summarize confidence in findings from included studies. CERQual assesses confidence in synthesized review findings based on the four key components of methodological limitations, adequacy, coherence, and relevance. Confidence in each review finding was judged as high, moderate, low, or very low, as defined according to the CERQual approach (see Supporting Information for further definition).

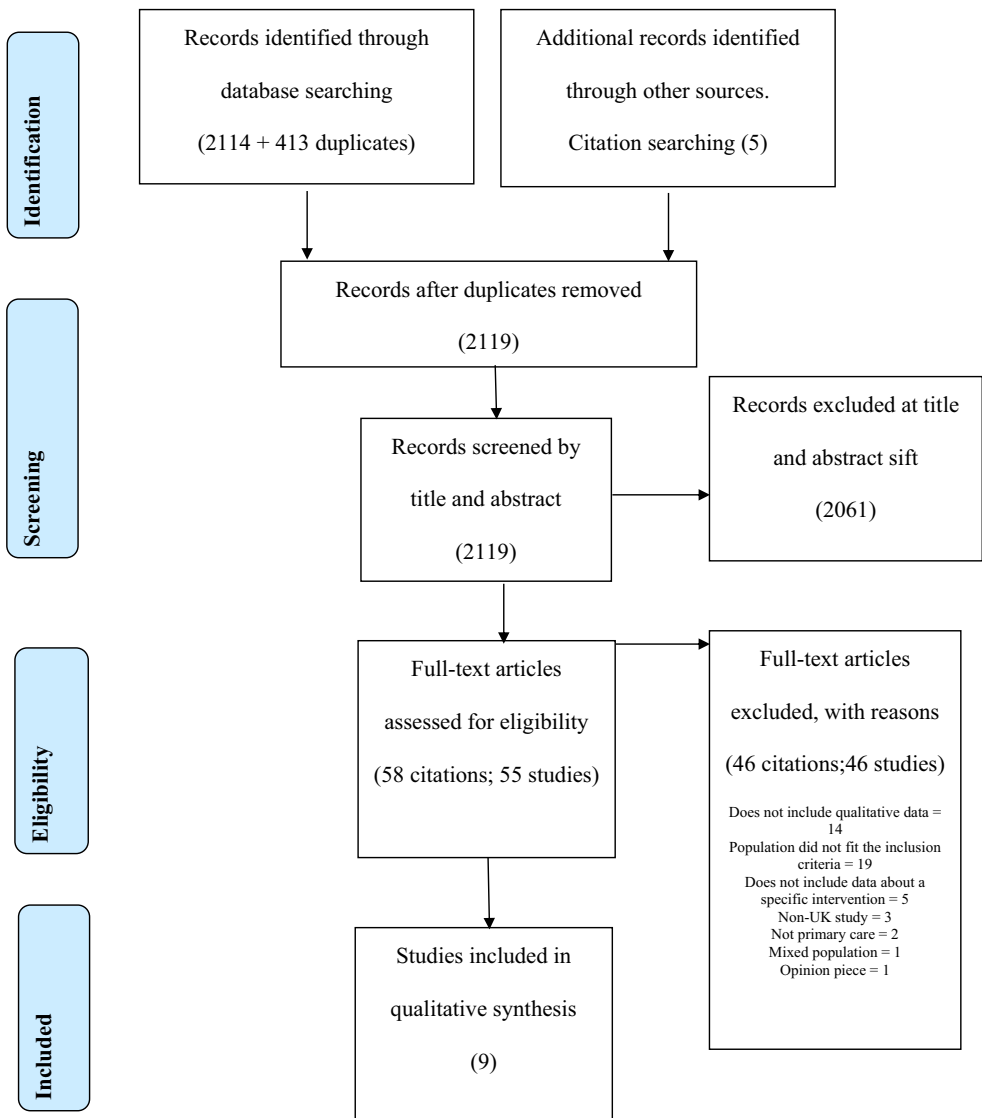


Figure 1. PRISMA flow diagram.

Results

From the 2,119 citations identified from the searches, 58 remained after title and abstract sift, and were considered at full paper sift. Twelve full papers reporting evidence from nine studies were included in the review. Figure 1 shows the PRISMA flow chart of studies included in the qualitative review. All included full papers were published between 2007 and 2018.

Study respondents

Eight studies provided evidence about patients' attitudes and four studies provided evidence about HP attitudes to the intervention. Overall, the studies contained qualitative

Table 4. Summary of the included studies and their sample and study characteristics

| Author (date) | Sample (contributing qualitative data) | Population being treated as described in the study | Data collection | Intervention |
|--|---|--|--|---|
| Burton et al. (2012) | 11 patients | MUS | Interview | GP with special interest 'symptoms' clinic |
| Chew-Graham et al. (2011) | Three nurse therapists, three supervisors (psychiatry, clinical psychology, counselling), 46 patients | CFS/ME | Semi-structured interviews | Pragmatic rehabilitation and supportive listening |
| Peters et al. (2011) | | | | |
| Cowell et al. (2018a, 2018b) | 10 physiotherapists | Non-specific chronic low back pain | Semi-structured interview | Cognitive functional therapy |
| Dowrick et al. (2008); Peters et al. (2009) (report different data from the study above) | 24 general practitioners 23 patients | MUS | Semi-structured interviews | Reattribution (by trained GPs) |
| Gerskowitch et al. (2015) | 11 patients | MUPS | Semi-structured interviews | CBT and mindfulness-based stress reduction |
| Graham et al. (2007) | Six patients | MUS | Self-reports – a series of open questions put to each patient in writing | Group counselling (humanistic) |
| Lewis (2013) | Eight CBT therapists | MUS | Semi-structured interviews | Individual CBT |
| Morton et al. (2016) | 17 patients | MUS | Semi-structured interviews | GP with special interest 'symptoms' clinic |
| Payne (2015) | 16 patients | MUS | Case studies; qualitative data from a survey | The BodyMind Approach (TBMA) group intervention |

data from 130 patients and from 48 HPs. HP data were derived specifically from 24 general practitioners (GPs), 10 physiotherapists, eight CBT therapists, three nurse therapists, and three nurse therapist supervisors (see Table 4).

Respondent characteristics

Six studies focused on interventions for MUS/MUPS, one for CFS/ME, one for non-specific chronic low back pain, and one defined as for somatization, where all patients included in

the study were described as presenting with physical symptoms that were medically unexplained. Of the 130 patients taking part in the included studies, most patient participants met the criteria for MUS or MUPS ($n = 84$) with the remaining 46 patients diagnosed with CFS/ME. Reporting of participant characteristics was limited and incomplete in several studies. Details where reported are shown in Tables 5 and 6 below.

Intervention description and facilitators

Three interventions were delivered by GPs (Burton, Weller, Marsden, Worth, & Sharpe, 2012; Dowrick et al., 2008; Morton et al., 2016; Peters et al., 2009), one by nurse therapists in a single study (Chew-Graham et al., 2011; Peters et al., 2011), and one by physiotherapists in a single study (Cowell et al., 2018a, 2018b). Trained CBT therapists delivered one intervention investigated in two studies (Gerskowitch, Norman, & Rimes, 2015; Lewis, 2013). A psychological well-being practitioner (PWP) delivered a low-intensity CBT intervention, and a trained facilitator delivered a mindfulness-based stress reduction (MBSR) intervention in one of these studies (Gerskowitch et al., 2015). Group counselling (humanistic) was facilitated by a 'group counsellor' (Graham et al., 2007), and the BodyMind Approach intervention was delivered by clinical psychologists together with facilitators who were psychotherapists or art therapists (Payne, 2015). Most interventions were delivered individually to patients; however, three interventions were delivered in a group setting, that is, MBSR, humanistic counselling, and the BodyMind approach (Gerskowitch et al., 2015; Graham et al., 2007; Payne, 2015) (see Table 7).

Quality of the included studies

Results of the quality assessment for each study are presented in Table 8. The quality levels to be applied to the evidence, high, moderate, or low were chosen to be conservative. The reviewing team made a judgement that a study must meet all criteria to

Table 5. Summary of patient characteristics

| Author (date) | Sample (contributing qualitative evidence) | Population being treated as described in the study | Gender | Age |
|---|--|--|--------------------------|---|
| Burton et al. (2012) | 11 patients | MUS | NR | NR |
| Chew-Graham et al. (2011); Peters et al. (2011) | 46 patients. | CFS/ME | 33 female; 13 male | Patients = Mean age 46.11 years (range 20–73) |
| Gerskowitch et al. (2015) | 11 patients | MUPS | Eight female; three male | Median age 50 years (range 19–60) |
| Graham et al. (2007) | Six patients | MUPS | NR | NR |
| Morton et al. (2016) | 17 patients | MUS | NR | NR |
| Payne (2015) | 16 patients | MUS | 10 female; six male | Range 19–80 years |
| Peters et al. (2009) | 23 patients | MUS | 20 female; three male | Mean age = 53 years (range 32–84 years) |

Table 6. Summary of health practitioner characteristics

| Author (date) | Sample | Population being treated as described in the study | Gender | Age |
|---|---|--|--------------------------------------|--|
| Cowell et al. (2018a); Cowell et al. (2018b) | 10 physiotherapists | Non-specific chronic low back pain | Seven male; three female | Not reported (years working in MSK range 3–14 years) |
| Peters et al. (2011) | Three nurse therapists, three supervisors | CFS/ME | Nurses = 3 women; NR for supervisors | NR for nurses and supervisors |
| Dowrick et al. (2008) | 24 general practitioners | MUS | 16 female; eight male | Three aged under 35 years; 14 between 35 and 50 years, and 7 over 50 years |
| Lewis (2013) | Eight CBT therapists | MUS | Five female; three male | Mean age 43.5 (SD = 9.04) years (range 30–60 years) |

be regarded as high quality. If any criterion was not fulfilled, the study was regarded as moderate quality. However if more than one criterion had not been achieved, it was concluded that it was not possible to assess the study as high or moderate quality.

Certainty of the review findings – CERQual assessment

The CERQual approach utilized the methodological quality of each study contributing to the review finding. Relevance was assessed by considering the format of the intervention (whether individual or group based) and the facilitator (whether a GP, nurse therapist, or psychological therapist). Given that the inclusion criteria specified a primary care- or community-based setting, this criterion was not considered in the analysis of relevance. Coherence was assessed by considering if all the data contributing to a finding supported that finding and the presence of ambiguities or plausible alternative explanations. Finally, adequacy was assessed by considering the richness and quantity of data supporting each review finding. The synthesis of evidence from patients yielded 22 review findings. Only one finding was assessed as high confidence, six findings were assessed as moderate confidence, 13 were assessed as low confidence, and two were assessed as very low confidence. The evidence from HPs yielded 16 findings overall. Four findings were assessed as high confidence, eight as moderate confidence, and four as low confidence. (The results of the CERQual assessment are set out alongside each sub-theme in Tables S1 and S2).

Table 7. Description of the interventions delivered in the included studies

| Author (date) | Population being treated as described in the study | Intervention | Intervention provider | Intervention setting | Intervention duration | Individual or group |
|---|--|--|--|--------------------------------------|---|---------------------|
| Burton et al. (2012) | MUS | GP with special interest 'symptoms' clinic. The consultations were structured to first hear the patient's experience of illness then to propose and negotiate constructive explanations of physical symptoms. These explanations were used as the basis for simple cognitive and behavioural actions to modify symptoms and their impact | GP | GP practice | Four appointments; the first was of 1-hr duration and the subsequent three lasted 20 min | Individual |
| Chew-Graham et al. (2011); Peters et al. (2011) | CFS/ME | (1) Pragmatic rehabilitation – a therapist facilitated CFS specific self-management intervention, which shares features in common with CBT and GET, but which does not require a specialist CBT or physiotherapist to deliver it. (2) Supportive listening | Primary care nurses trained to deliver the interventions | Patients homes | 90-min session followed by 1-hr sessions on weeks 2, 4, 10, and 18. 30-min telephone calls on weeks 3, 6, 8, 12, and 15 | Individual |
| Cowell et al. (2018a, 2018b) | Non-specific chronic low back pain. | Cognitive functional therapy (CFT). A biopsychosocial orientated behavioural intervention for lower-back pain, underpinned by a multidimensional clinical reasoning framework (MDCRF) (O'Sullivan et al., 2018) | Physiotherapists | Not reported other than primary care | Not reported | Individual |
| Dowrick et al. (2008); Peters et al. (2009) | MUS | Reattribution – a structured intervention, designed to provide a simple explanation of the mechanism of a patient's MUS, through negotiation and other | GP | GP Practice | The time since the index consultation ranged from 8 to 55 weeks (mean = 32) | Individual |

Continued

Table 7. (Continued)

| Author (date) | Population being treated as described in the study | Intervention | Intervention provider | Intervention setting | Intervention duration | Individual or group |
|--------------------------|--|---|---|---|---|---------------------|
| Gerskowitz et al. (2015) | MUPS | features of patient-centred communication, and to be delivered during routine consultations CBT high intensity based on the cognitive-behavioural model of MUPS | HI-CBT therapist | Primary care psychological therapies service | Weekly 1-hr appointments. The median number of sessions attended was 17 (range 4–25) Fortnightly with homework set between meetings An 8-week programme for 2 hr per week 1 hr 30 min weekly sessions for half a year Not specified | Individual |
| Graham et al. (2007) | MUPS | CBT low intensity based on the cognitive-behavioural model of MUPS | Psychological well-being practitioner (PWP) | | | Individual |
| Lewis (2013) | MUS | Mindfulness-based stress reduction | Trained MBSR facilitator | | | Group |
| Morton et al. (2016) | MUS | Group counselling (humanistic) CBT – based on the cognitive-behavioural model of MUPS Symptoms clinic intervention – a structured series of consultations. The SCI is comprised of four key elements: recognition and validation of symptoms, explanation of symptoms, actions to manage symptoms, and learning | Group counsellor CBT therapists with a postgraduate diploma in CBT GP | GP practice Primary care mental health care trust GP Practice | | Individual |
| Payne (2015) | MUS | The BodyMind Approach (TBMA), based on a biopsychosocial model derived from dance movement psychotherapy | Clinical psychologist/facilitators (psychotherapists or art therapists) | Community setting | 2 hr for 12 sessions over eight weeks and other communication over a 12-month period. Total face-to-face contact is 27 hr | Group |

Table 8. Quality assessment results for the included studies

| | Chew-Graham et al. | | | | | | | | | |
|--|----------------------|-----------------------|---|-----------------------|--------------------------|------------------------------|--------------|-----------------------|---|----------------------|
| | Burton et al. (2012) | Peters et al. (2011) | Cowell et al. (2018a); (2018b) | Dowrick et al. (2008) | Gerskowitz et al. (2015) | Graham et al. (2007) | Lewis (2013) | Morton et al. (2016) | Payne (2015) | Peters et al. (2009) |
| Question (Yes/No/Can't tell) | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes |
| Is the study qualitative research or does it provide qualitative data | Yes | Yes | Yes | Yes | Yes | Somewhat | Yes | Yes | Somewhat, each element could have been clearer | Yes |
| Is the study context and aims clearly described? | No | No | Reflexivity mentioned in the methods but not obviously applied in the analysis. | No | No | Yes | Yes | No | Yes | No |
| Is there evidence of research reflexivity? | Yes | Yes – part of a trial | Yes | Yes – part of a trial | Yes | Yes | Yes | Yes – part of a trial | No, it is unclear how many patients contributed data/ took part in the intervention | Yes |
| Are the sampling methods clearly described and appropriate for the research question? | Yes | Yes | Yes | Yes | Yes | Somewhat – not very detailed | Yes | Yes | No not clearly described | Yes |
| Are the methods of data collection clearly described and appropriate to the research question? | Yes | Yes | Yes | Yes | Yes | Somewhat – not very detailed | Yes | Yes | No not clearly described | Yes |

Continued

Table 8. (Continued)

| | Burton et al. (2012) | Chew-Graham et al. (2011); Peters et al. (2011) | Cowell et al. (2018a); (2018b) | Dowrick et al. (2008) | Gerskowitz et al. (2015) | Graham et al. (2007) | Lewis (2013) | Morton et al. (2016) | Payne (2015) | Peters et al. (2009) |
|--|---|---|--------------------------------|-----------------------|--------------------------|--|--------------|---|--------------|----------------------|
| Is the method of analysis clearly described and appropriate to the research question? | No – qualitative methods not described | Yes | Yes | Yes | Yes | No – content analysis mentioned, details of how this was conducted not present | Yes | Yes – content analysis using a framework approach | No | Yes |
| Are the claims made supported by sufficient evidence (i.e., Did the data provide sufficient depth, detail and richness)? | No – very little qualitative data presented | Yes | Yes | Yes | Yes | No – data were not very detailed or rich | Yes | No – only examples of the data were presented | No | Yes |
| Assessment of quality | Low | Moderate | Moderate | Moderate | Moderate | Low | High | Moderate | Low | Moderate |

Synthesis of patient and health practitioner evidence

Data were synthesized across all intervention types and organized according to the questions outlined in the data extraction framework (Table 2). Each theme and sub-theme, together with evidence from the primary studies and an estimate of the strength of the evidence, is presented within the following narrative synthesis and also in Tables S1 and S2.

Factors identified as important in interventions for PPS from the perspective of patients who had received the interventions

Support

Feeling supported was defined by patients as being accepted and validated by the HPs, receiving empathy, and being listened to. This need for support was identified across diverse intervention types with high confidence in this finding (Chew-Graham, Brooks, Wearden, Dowrick, & Peters, 2011; Gerskowitch et al., 2015; Graham et al., 2007; Morton et al., 2016; Payne, 2015; Peters et al., 2009; Peters et al., 2011;). Such support was described as the most positive part of the pragmatic rehabilitation intervention in one study, with 'being believed and feeling understood by the therapist' identified as a key part of the intervention (Chew-Graham et al., 2011). Patients across various intervention types reported being validated by the HP as important (Chew-Graham et al., 2011; Gerskowitch et al., 2015). This feeling was sometimes due to the knowledge the HP had of the patient's symptoms giving them a sense of having someone 'on their side' (Gerskowitch et al., 2015; Morton et al., 2016; Peters et al., 2009). In some instances, it appeared that no one else in their lives was able to provide such support, 'It was just an understanding from her that I didn't, haven't had from anybody else' (Patient, Chew-Graham et al., 2011). One patient reporting that the empathetic nature of the nurse delivering the supportive listening intervention was their most valued attribute (Peters et al., 2011).

The feeling of being understood also came from fellow participants with similar symptoms within group interventions (Gerskowitch et al., 2015; Graham, Manor, & Wiseman, 2007; Payne, 2015). These factors were reported as key to patient engagement and contributed to whether the interventions were reported as acceptable to the participants (Chew-Graham et al., 2011).

Explanation

Patients valued explanations of their symptoms which they had been provided with as part of the interventions. They reported that this led to their acceptance and understanding of the treatment model, both a CBT for MUPS study (Gerskowitch et al., 2015) and a pragmatic rehabilitation for CFS study (Chew-Graham et al., 2011), with moderate confidence assessed in this finding. They also felt that the pragmatic rehabilitation intervention helped them to come to terms with and accept a diagnosis (of CFS) and that having an explanation and understanding of their symptoms was key to this. 'She explained all about CFS and the physiology of it really, which was the first time really that I understood why my energy was so low, so that made a lot of sense.' (Patient, Chew-Graham et al., 2011). Gaining new knowledge about their symptoms was reported to be reassuring to patients enabling them to make an initial judgement about whether the intervention might be appropriate for them (Chew-Graham et al., 2011). Patients reported that they consulted their GP in order to seek an explanation for their symptoms (Peters et al., 2009), and took part in interventions to see someone who had specialist knowledge and information about their symptoms (Gerskowitch et al., 2015).

Important factors for the delivery and success of interventions from the perspective of patients

With interventions where behavioural self-management techniques were taught, some patients reported they had benefited from these. This was evidenced across four of the studies which included the following interventions: CBT for MUPS, mindfulness, group counselling, GP with a special interest 'symptoms' clinic, and the BodyMind approach intervention. Participants commented on their improved confidence (Gerskowitch et al., 2015; Graham et al., 2007; Morton et al., 2016; Payne, 2015) and improved communication with significant others, 'I have spoken to friends about my feelings and hold fewer grudges.' (Patient, Graham et al., 2007). Specific to group interventions patients found sharing their experiences with other patients (Gerskowitch et al., 2015) and learning skills from other patients (Gerskowitch et al., 2015; Graham et al., 2007) valuable as part of the intervention.

Patients reported needing reassurance and a good relationship with the person delivering the intervention (Peters et al., 2009). Patients involved in a reattribution intervention liked having a longstanding relationship with the GP, and this gave giving them a feeling that the HP would know what type of treatment, if any, would be suitable for them, 'He knows I like to keep myself to myself, knows I believe in self-help. . . knows I'm somebody that likes to work it out for myself'. (Patient, Peters et al., 2009). Others appreciated the understanding and reassurance they received from the HP and just wanted an opportunity to share their difficulties (Peters et al., 2009).

Important factors for the delivery and success of interventions from the perspective of health practitioners

Across four studies, and with high confidence in this finding, HPs reported positive factors around extending their own knowledge and skill. They reported training and supervision in applying behavioural interventions as helpful, and developed their knowledge and skills in the area of PPS (Cowell et al., 2018a, 2018b; Dowrick et al., 2008; Peters et al., 2011). Training helped them to learn about the boundaries of their own role (Peters et al., 2011) and to be flexible when delivering interventions (Peters et al., 2011; Lewis, 2013). In one study, some GPs reported that reattribution training increased their confidence in discussing PPS, and to reflect on management decisions, 'I'd like to think that I do go a little bit more into other agendas, other issues that might be fuelling the symptoms that they've got and try and approach those other problems rather than just focusing on a prescription for something for pain' (GP, Dowrick et al., 2008).

In three studies, contributing to a finding of high confidence (Lewis, 2013; Peters et al., 2011; Cowell et al., 2018a, 2018b), HPs reported that it can be rewarding when it is evident that patients have gained benefit from the intervention. 'And it was like watering a flower it was really lovely for me personally. . . it was lovely watching her just blossom, you know what I mean, because she finally took on board the physical stuff and the sleep.' (Nurse, Peters et al., 2011).

The primary care or community setting was reported to be a helpful factor by the HPs in two studies. In one study, the reason for this was that the primary care setting allowed for a more tailored approach, or that the setting, such as being in patients homes, was useful when trying to develop a therapeutic relationship (Peters et al., 2011). In another case, this contrasted with the perceived potential failing of the secondary care setting (Dowrick et al., 2008). 'You see these people getting referred to the hospital with back pain and the next thing you know some bright spark is going to operate on them and you

think 'What!'. . . Maybe we're here in a way as a gateway to try and prevent harm as well as anything else.' (GP; Dowrick et al., 2008). This appears to indicate that primary care practitioners see part of their role as protecting their patient from entering secondary care where they believed this was unnecessary.

Factors identified as barriers to intervention success by patients

Patients reported a feeling of scepticism towards HPs, with data contributing to this finding across four studies with moderate confidence. This included the concern that mentioning psychosocial problems would divert GP attention away from other physical problems associated with their symptoms (Peters et al., 2009), and in one study, patients had the feeling that the aim of the GP was to assert their problems were psychological (Burton et al., 2012). Patients also reported being concerned that they would only be provided with simplistic explanations for their symptoms, as in a study of CBT for MUPS delivered by CBT therapists (Gerskowitch et al., 2015).

Patients reported that the lack of information given to them by those that referred them to the study meant that they did not know the remit of the intervention, and that the referring GP also often did not know what the intervention entailed. Patients were therefore surprised by the content of the intervention and found it difficult to see why they had been referred to a psychological therapy-based intervention when they perceived their problems as physical (Gerskowitch et al., 2015). There was also a feeling from patients in two studies, that some HPs, GPs, and nurse therapists were unskilled or lacked the knowledge to deal with their symptoms or provide an appropriate intervention (Peters et al., 2009, 2011).

In one study, data suggested that some patients may hold the belief that they should deal with their symptoms themselves, considering it inappropriate to discuss psychosocial problems (with the GP), with the idea of there being a stigma related to reporting psychosocial problems (Peters et al., 2009).

Factors identified unhelpful or barriers to intervention success by health practitioners

Across four studies, a finding with high confidence emerged around HPs' perceptions about patient beliefs. HPs reported that they considered patient beliefs (such as not being able to take part in aspects of interventions that required physical activity) could be significant barriers to engagement with behavioural interventions (Dowrick et al., 2008; Lewis, 2013; Peters et al., 2011). In one study, HPs described, and some appeared to expect, patients to hold a belief in a physical cause for their symptoms and suggested this could be a barrier to engagement with the intervention, 'Well it's their mindset isn't it? It's their belief that there is a physical cause.' (GP, Dowrick et al., 2008).

Health practitioners reported that their own emotions could be a source of difficulty in delivering PPS interventions in two studies (Lewis, 2013; Peters et al., 2011). Nurse therapists found at times that they did not deal well with perceived failure, when patients felt the intervention was not suitable or working for them, 'One common theme I think with has come up is the difficult of accepting that you can't get it right all the time.' (Nurse Therapist Supervisor, Peters et al., 2011).

In two separate studies, nurse therapists and CBT therapists who had been trained to deliver PPS specific interventions felt that they were novices and lacked the experience to deliver the interventions (Lewis, 2013; Peters et al., 2011). CBT therapists experienced anxiety due to this lack of training and experience in PPS (Lewis, 2013), whereas nurse

therapists were not comfortable dealing with the mental health aspects as compared with the physical health aspects of the interventions they were delivering (Peters et al., 2011). This also emerged as a barrier to service provision, with GPs acknowledging that the successful application of an intervention may be affected by variations in the communication skills of the practitioner delivering the intervention (Dowrick et al., 2008).

Time constraints and impositions about what data needed to be collected during a consultation presented barriers to HPs delivering interventions in three studies (Cowell et al., 2018a, 2018b; Dowrick et al., 2008; Peters et al., 2011). Concerns regarding time constraints were associated with each individual consultation (Dowrick et al., 2008) as well as the length of the course of therapy, with reservations expressed that too few sessions were available to deal with deep-seated issues (Peters et al., 2011). GPs were concerned about medico-legal issues, such as over-diagnosing and over-treating patients in one study (Dowrick et al., 2008).

Potential unwanted or adverse effects on patients

Lack of understanding of their situation from HPs was reported by patients in two studies (Morton et al., 2016; Peters et al., 2009), with low confidence in this finding. Sometimes this extended to patients feeling that they were being blamed by the GP (Morton et al., 2016). 'Well that's what I'm saying . . . how could I put it . . . I don't know, I felt as if Dr. D was maybe thinking I was getting a bit lazy or something like that, you know, which definitely I'm not that type of person, I'm not a lazy person, you know, I felt quite uncomfortable with that. He never actually said that, you know what I mean, but I just felt that within myself, you know.' (Patient, Morton et al., 2016).

Conflicts between HPs and patients were reported by patients in one study (Chew-Graham et al., 2011; Peters et al., 2011). This occurred when patients felt the HPs were indicating that the intervention (in this case pragmatic rehabilitation for CFS/ME) was the only appropriate intervention, and the patients disagreed with this, 'I think my main reason is the fundamental theory behind it [the treatment model offered] just disregards it as illness'. (Patient, Chew-Graham et al., 2011).

In one study, patients reported finding it difficult when the intervention came to an end, experiencing a feeling of loss when support was withdrawn (Chew-Graham et al., 2011; Peters et al., 2011). HPs in two other studies reported similar concerns, with those in one study worrying patients might form a dependent relationship with the GP, and become reliant on them (Dowrick et al., 2008), and a concern about withdrawing support in the other, 'I just hope she doesn't get a divorce. . . I am frightened in case I open up a can of worms. . . I don't want to leave an aftermath.' (Nurse, Peters et al., 2011). Key findings from the qualitative synthesis are shown in Table 9.

Discussion

This review presents patient and health practitioner (HP) perspectives about the acceptability, relative benefits, and potential harms of primary care- or community-based behavioural interventions for persistent physical symptoms (PPS). Our findings offer insight into which aspects of the interventions described patients and HPs found helpful, what was unhelpful, and what were the potential barriers to patient participation or a successful intervention. These could all contribute towards an understanding of what might make a more successful intervention. Patients valued support, coherent

Table 9. Summary of the key findings

| | Patients | Health practitioners |
|--|--|---|
| Valuable results from intervention participation or training | Gaining support being validated Explanation Support for self-management | Developing knowledge and skills around PPS Rewarding when patients engaged |
| Facilitators | Good relationships between patients and HPs | Training and supervision Primary care or community setting |
| Barriers | Patient attitudes and beliefs Poor relationship with HPs | Lack of confidence in their own skills and abilities to deal with PPS Patient attitudes and beliefs HP attitudes and beliefs Resources constraints |
| Adverse effects | It ended Conflicts between HPs and patients, and HP lack of understanding | Detrimental intervention consequences for patients Patient and health practitioner emotions |

explanations of their symptoms, and good relationships with HPs, whilst HPs felt extending their own knowledge and developing a therapeutic relationship would facilitate a successful intervention. Patient barriers included problematic relationships with HPs and a lack of knowledge regarding the intervention and its remit at the time of referral.

Across all the interventions, patients' valued gaining support, being accepted and validated by the HP (and by other intervention participants in the case of group interventions), receiving empathy and being listened to. These findings are consistent with previous findings that have demonstrated the importance of the therapeutic relationship between patients and HPs in working with people with PPS (Gask et al., 2011; Heijmans et al., 2011; Smith et al., 2003). This HP validation and support may however often be lacking, and poor communication between GPs and patients can cause barriers to effective treatment of PPS. Picariello, Ali, Moss-Morris, & Chalder's (2015) survey of people with CFS reported that participants often felt a lack of empathy and understanding from HPs. Potential barriers include unempathic HP communication styles and behaviours during the consultation, patient health beliefs about treatment in the primary care setting, the extent of problem exploration, HPs attitudes towards patients, a biomedical disease model approach, HP knowledge of PPS, and level of confidence in treatment (Murray et al., 2016). Being less well informed about PPS may affect practitioner attitudes in a way which isn't always helpful for patients. GPs can find such presentations frustrating and difficult if they don't lead to very therapeutic consultations, resulting in a negative impact on the doctor-patient relationship (Wileman, May, & Chew-Graham, 2002). Patients are more likely to feel that they are being taken seriously when the GP is aware of their personal circumstances and has an open and empathic approach (McLeod, Budd, & McClelland, 1997; Peters, Stanley, Rose, Kaney, & Salmon, 2002; Smith et al., 2006), treats the patient as an equal (McLeod et al., 1997; Morriss et al., 2007), and explores their symptoms in depth and acts upon them (Morriss et al., 2007; Ryan & Gevirtz, 2004). Effective explanations of symptoms have been shown to be empowering for some patients (Sowinska & Czachowski, 2018). A recent metaanalysis of studies based

on Leventhal's common-sense model and outcomes in PPS showed patients who report a greater understanding of their condition report lower symptom severity, lower psychological distress, and better quality of life (McAndrew et al., 2019).

Clinical implications

The impact of not feeling believed or not having symptoms taken seriously may have a significant detrimental impact, leading to feelings of shame associated with a perceived invalidation of symptoms (Wearden et al., 2010). Validation has been shown to be critical for patients in a qualitative study comparing psychoeducation and CBT for CFS, regardless of the effectiveness of an intervention (Dennison, Stanbrook, Moss-Morris, Yardley, & Chalder, 2010). Therefore, the importance of a good therapeutic relationship and credible explanations of symptoms should be key to any future intervention. Being able to provide flexible explanations that take into account individual differences between patients and taking account of patients' own health beliefs may be important. In some cases, HPs felt the training they had received was not enough, highlighting a need for more specific training, which was consistent with patients finding a lack of knowledge and skills in HPs a barrier. Specifically, CBT therapists wanted to extend their knowledge of PPS, whilst GPs and nurse therapists wanted to improve their knowledge of psychological interventions and thus increase their confidence in discussions with patients.

Our findings as well as previous findings (Gerskowitch et al., 2015) show that if patients do not have the information required for an informed choice, it is less likely that the intervention will be suitable for them; therefore, it is important to provide accurate information and to support HPs to provide this information prior to referral. Some patients reported finding it difficult when the intervention came to an end. It is important therefore to consider when developing future interventions that some patients may experience such effects, and how this should be addressed.

As the review showed, there are a number of ways interventions can be delivered in primary care, and therefore, the complexity of the primary care setting should also be considered and acknowledged in the development of new interventions. The settings covered here included GPs delivering interventions to their own patients, or coordinating care of another HP, and where the intervention might take place away from a GP practice but not in secondary care. Although it is difficult to make specific recommendations about which type of primary care setting is optimal, there is a perceived importance to some involvement with primary care rather than being referred directly to secondary care, with patients valuing working with HPs that they know and are aware of their circumstances.

Limitations

Whilst the importance of the findings cannot be understated, some included studies were of moderate or low quality and the findings assessed as moderate or low confidence using the CERQual assessment (Lewin et al., 2015). Such limitations may result in a lack of rich data consistently across all studies, limiting the degree to which interpretations can be made for some of the findings (Booth et al., 2018). Variation across type of intervention, setting, the patient population, and health professionals, being from very different professions with different levels of understanding of PPS, will also have had an impact on how data were collected and the type of data yielded in the primary studies. Furthermore, not all intervention studies have nested qualitative studies; for example, in a report linked to this study (Leaviss et al., 2020), 59 studies were identified for the quantitative review,

compared to only nine studies in this qualitative review, and this difference will also bias what we know about the interventions. In addition, the findings are reflective of a patient population attending primary care. This does not necessarily generalize to PPS patients in secondary care. All of these factors limit our ability to generalize the findings when assessing wider acceptability.

Finally, it is important to note that qualitative analysis relies to some degree on the reviewer's understanding and interpretation of the data, with the potential for a range of ways of interpreting this. To ensure that the qualitative synthesis was reliable as possible, two reviewers were involved in selecting and data extracting studies, and a framework for data extraction was utilized to promote objectivity in the data extraction process.

Future directions

Persistent physical symptoms are common in primary care, with GPs playing a major role in identifying and managing such patients. The quality of this care and the importance of it being acceptable to patients cannot be understated. Future research, in the form of randomized controlled trials, to assess the feasibility, acceptability, and effectiveness of behavioural interventions for PPS delivered in primary care is important. These interventions should emphasize the validation of patients' symptoms and address the factors we have identified here as important, including patients gaining support, effective explanations of symptoms, and good relationships with their HPs, with adequate information about any proposed intervention provided to patients at the time of referral. These interventions should be delivered by HPs that have been trained to deliver the intervention with specific knowledge of PPS and should have the skills to develop a therapeutic relationship. As it is currently unclear what and how GPs learn about PPS, studies on the impact of specific educational initiatives with different groups of HPs should be undertaken, together with process research on what distinguishes a good versus a poor outcome in consultations. These studies should be co-designed with patients.

Conclusions

Patients valued being supported and being provided with explanations for their symptoms. Training for health professionals was thought to be helpful and may address barriers that were underpinned by the relationship between the patients and HPs delivering the intervention where this was not therapeutically effective. Interventions should not end suddenly or without adequate follow-up or booster sessions. Multiple key enabling factors to inform the development of future interventions were identified, and patients should be provided with adequate information to make an informed decision when offered such interventions. These findings, together with the findings of the wider report which also included a quantitative evidence synthesis and economic modelling, have identified research recommendations for primary research in this important area affecting a large number of patients and consuming considerable NHS resources.

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Conflicts of interest

Dr Andrew Booth is a member of the NIHR CRRSU Funding Board. Professor Rona Moss-Morris was previously an advisor to the NHS IAPT programme and is a scientific advisor to Mahana Therapeutics and a beneficiary of a licence agreement signed between King's College London and Mahana Therapeutics.

Author contributions

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Supporting Information

The following supporting information may be found in the online edition of the article:

Table S1. Thematic synthesis of evidence from patients.

Table S2. Thematic synthesis of evidence from health practitioners delivering the intervention.