CHRISTINE O'CONNELL BA Hons MSc

INTEGRATING PHYSICAL AND PSYCHOLOGICAL WELLBEING IN CHILD HEALTH

Section A: A systematic review of randomised controlled trials of psychological interventions for children and adolescents with medically unexplained symptoms: A focus on mental health outcomes.

Word Count
7748 (+229)

Section B: Factors influencing the referral of children and families to paediatric psychology: Using Theory of Planned Behaviour to develop a questionnaire of health care professionals' referral behaviour.

Word Count
7704 (+360)

Overall Word Count
15608 (+589)

A thesis submitted in partial fulfilment of the requirements of
Canterbury Christ Church University for the degree of
Doctor of Clinical Psychology

APRIL 2016

SALOMONS
CANTERBURY CHRIST CHURCH UNIVERSITY

Acknowledgements

I am very grateful for all of the support and guidance I have received during the completion of this project. I would like to thank the participants for their generosity and kindness in giving up their time to be part of this research. I would also like to express my thanks to my supervisors, Professor Roz Shafran and Professor Paul Camic, for their guidance and encouragement throughout the process. Thanks also to my manager, Dr Rachel Terry for her consistent support.

I would like to say a special thanks to Paul, my partner, for his understanding, love and care, and for being there throughout the process. I am also very grateful for the support I have received from Amanda, my friend, who has helped me more than she knows. A thank you to Carrie, and my other fellow trainees for company on the days, evenings and weekends in the library. My parents, sisters and friends outside of psychology have been very supportive throughout this time and I greatly appreciate their understanding and care.

Summary of Major Research Project

Section A

This review used a systematic methodology to investigate the effectiveness of psychological interventions for mental health difficulties in children and adolescents with medically unexplained symptoms. Randomised controlled intervention studies were eligible for inclusion. Fifteen studies and one follow up study were identified. Psychological interventions show effectiveness in improving mental health outcomes in this population, although further research is necessary to establish efficacy. Clinical and research recommendations are discussed.

Section B

This study used Theory of Planned Behaviour to develop a questionnaire which explores factors influencing the referral of children and families to paediatric psychology. Psychometric properties of the questionnaire were examined. Findings indicate that the questionnaire holds good reliability and validity and that the main constructs of Theory of Planned Behaviour are useful in predicting intention to refer to paediatric psychology. Specific beliefs about referral were also shown to influence intention to refer. Findings that individual factors such as attitudes and beliefs can impact healthcare professionals' referral behaviour indicate that multidisciplinary interventions and inter-professional education relating to the psychological aspects of illness are required.

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MAJOR RESEARCH PROJECT

SECTION A

A systematic review of randomised controlled trials of psychological interventions for children and adolescents with medically unexplained physical symptoms: A focus on mental health outcomes.

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Abstract

Background. Medically unexplained symptoms affect between 4 and 20 percent of children and adolescents. Between 30 and 60 percent of these children also experience mental health difficulties such as anxiety and low mood. Trials and reviews have focussed on physical and functional gains in this population, often overlooking mental health outcomes.

Objectives. To use a systematic review methodology to:

(i) Investigate the effectiveness of psychological interventions for mental health difficulties in children and adolescents with medically unexplained symptoms.

(ii) Identify significant aspects of interventions associated with their success.

Methods. Randomised controlled intervention studies investigating the impact of psychological interventions on mental health symptoms for children and adolescents with medically unexplained symptoms were eligible for inclusion. Systematic searches of PsycINFO, MEDLINE and CINAHL databases were undertaken from inception to November 2015, using key search terms. Studies were appraised using the quality appraisal checklist for quantitative evaluative studies (National Institute for Health and Clinical Excellence, 2012). A qualitative synthesis of studies was carried out.

Results. Fifteen studies and one follow up study were identified.

Conclusions. Psychological interventions show effectiveness in improving mental health outcomes in this population, although further research is necessary to establish efficacy. Clinical and research recommendations are discussed.

Introduction

Defining Medically Unexplained Physical Symptoms

There is a historical and current lack of clarity around the definition of medically unexplained physical symptoms, which is generally acknowledged within the research community (Eminson, 2007; Lieb, Pfister, Mastaler, & Wittchen, 2000) and within clinical practice, with different interpretations of symptoms between medical specialities (Hinton and Kirk, 2016). Common terminology and classification of symptoms has changed multiple times over the past century, as have explanations for the causes of these symptoms. The term *medically unexplained symptoms*, which will be used in the current review, applies to physical symptoms that cannot be accounted for by an organic or disease-specific pathology (Geist, Weinstein, Walker & Campo, 2008). The advantage of using medically unexplained symptoms as opposed to historical terms such as "functional" or "psychosomatic" is to, respectively, avoid dualism and attribution of cause to symptoms, the aetiology of which are largely unknown (Sharpe & Mayou, 2004).

Medically unexplained symptoms are commonly associated with significant functional impairment and distress and can be long-standing or recurring (olde Hartman et al., 2009). Common medically unexplained symptoms and syndromes include chronic headache, pain or fatigue and gastroenterological or irritable bowel symptoms (Mobini, 2015). There is some evidence of particular medically unexplained symptoms clustering together (Walker, Garber & Green, 1991), giving credence to syndromes such as chronic fatigue syndrome (CFS; fatigue and pain), irritable bowel syndrome (IBS; gastroenterological symptoms) and conversion disorder (neurological symptoms). However, research suggests significant overlay across these syndromes in relation to case description, gender, symptomology and treatment response (Wessely, Nimnuan & Sharpe, 1999). Brown (2007) suggests that medically unexplained symptoms are experienced on a spectrum ranging from

solitary, fleeting and mild symptoms to multiple chronic symptoms. According to the Diagnostic Statistics Manual of Mental Disorders (5th ed.; DSM-5), if a collection of significantly debilitating symptoms are experienced over a long period of time and accompanied by disproportionate thoughts, emotions or behaviours, these symptoms may be categorised by a psychiatric classification such as somatic symptom disorder (American Psychiatric Association [APA], 2013). Further complicating definitions, medically explained and unexplained symptoms often occur together. Additionally, as of the DSM-5, symptoms can be medically explained or unexplained and still fall under the somatic symptom disorder category if thoughts, emotions or behaviours relating to the illness are deemed excessive (APA, 2013). Similarly, criteria for classification of medically unexplained symptoms within the ICD-10 (World Health Organisation [WHO], 1992) apply across the lifespan and examples are often more relevant to adults than children and adolescents (Eminson, 2007; Nunn, Nicholls, & Lask, 2000) with the exception of separate classification of neurological symptoms in children.

By their very nature, medically unexplained symptoms are diverse and difficult to define and individual experiences are reported in different ways. This is especially true for children, whose stage of development impacts how symptoms are expressed and communicated (Eminson, 2007). Additionally, children's propensity to seek help from medical professionals is largely dependent on their parent's beliefs and attitudes, rather than symptom experience (Eminson, 2007). Therefore, the execution of robust epidemiological studies of child medically unexplained symptoms which indicate accurate prevalence has proven highly challenging (Hinton & Kirk, 2016).

Prevalence and Comorbidity

Several longitudinal and cross-sectional studies have used a variety of methodologies and criteria to estimate numbers of children experiencing medically unexplained symptoms.

Aro (1987) and Offord et al., (1987) found that between 4 and 11 percent of adolescents and their parents reported recurrent child medically unexplained symptoms. In a longitudinal study of children followed from age 9 to 13 in the USA, headaches were found to be the most common medically unexplained symptoms (10 percent) followed by abdominal pain (2.8 percent) and musculoskeletal pain (2.2 percent; Costello, et al, 1996; Egger, Costello, Erkanli, & Angold, 1999). Studies involving younger children have found medically unexplained symptoms are experienced by approximately 20% of pre-schoolers (Domènech-Llaberia et al., 2004) and that abdominal pain is more common than headache in this group (Zuckerman, Stevenson, & Bailey, 1987). Medically unexplained symptoms are reported more often and to a higher degree in females than in males throughout childhood and adolescence (Berntsson, Kohler, and Gustafsson, 2001; Eminson, Benjamin, Shortall, Woods, & Faragher, 1996). Co-occurrence of different symptoms is frequently described and reporting has been found to increase with age (Eminson et al., 1996; Garber, Walker, Walker, & Zeman, 1991).

Several studies have found strong associations between medically unexplained symptoms and symptoms of mental ill-health (e.g. Henningsen, Zimmermann, & Sattel, 2003). Between 30 percent and 60 percent of children and adolescents reporting medically unexplained symptoms are thought to also be experiencing mental health difficulties meeting criteria for psychiatric diagnosis (Lieb et al., 2000; Husain, Browne & Chalder., 2007). Research illustrates that anxiety, depression and behavioural problems are the most common of these difficulties within this population (Zuckerman et al., 1987; Wasserman, Whitington, & Riviara, 1988; Garber, Zeman, Zeman, & Walker, 1990). Although their aetiology remains unclear, there are several psychological models of the causes and maintenance of medically unexplained symptoms, often linking physical symptoms with emotional distress.

Psychological Theories

Husain et al. (2007) carried out a review of explanatory psychological models of medically unexplained symptoms in children, categorising them into behavioural, systemic, psychodynamic, cognitive, and cognitive-behavioural models. Behavioural models argue that illness behaviours are reinforced by an experience of pleasant or avoidance of unpleasant consequences. Comparably, social learning theory (Bandura, 1977) assumes that illness behaviours are learned, however, this theory emphasises the social context in which learning takes place. Indeed, according to Levy et al. (2004) parents who continuously respond attentively to the child's illness tend to have children reporting a higher rate of medically unexplained symptoms. Social learning theory also holds that modelling influences children's learning about illness behaviours. Research shows children are more likely to report medically unexplained symptoms if their parents report medically unexplained symptoms themselves (Zuckerman et al., 1987). Parent's behaviours are also said to influence children's beliefs about the seriousness of illness (Levy et al., 2004).

Systemic models also emphasise ways in which families relate to and communicate with each other to maintain illness behaviours (Minuchin, 1974). Griffiths and Griffiths (1994) posit that these children are in an environment where emotional distress cannot be expressed verbally and so use their body to communicate (Griffin & Christie, 2008). Similarly, psychodynamic theory suggests that medically unexplained symptoms are the result of difficulty expressing emotional distress.

There is growing acknowledgement of the role of attachment theory medically unexplained symptoms (Adshead & Guthrie, 2015). This theory emphasises the importance of the quality of the relationship between an infant and their primary care-giver for emotional management throughout the lifespan (Bowlby, 1988). In a large population study, Davies, Macfarlane, McBeth, Morriss & Dickens (2009) showed that insecure early attachment may be a risk factor for chronic pain experience that does not respond to medication.

Cognitive theories tend to focus on cognitions and beliefs about illness (Barsky & Wyshak, 1990; Beck, Rush, Shaw & Emery, 1979). In these models, normal bodily sensations are catastrophised and misattributed to illness. Cognitive behavioural models account for medically unexplained symptoms by bringing cognitions, emotions, behaviours, and physiology together, postulating that the experience of a bodily sensation may cause thoughts about weakness or fainting, leading to anxiety which may lead to avoidance of activity, therefore maintaining the amplification and experience of the bodily sensation (e.g. Clark, 1986). Severeijns, Vlaeyen, and van den Hout (2004) illustrated that pain catastrophising and anxiety about physical illness symptoms was an independent predictor of health-care use in adults experiencing chronic pain symptoms, indicating that anxiety may interact with physical sensations to produce medically unexplained symptoms.

Randomised controlled intervention trials have supported social learning and cognitive-behavioural theories of medically unexplained symptoms (e.g. Levy et al., 2010; Nijhof, Bleijenberg, Uiterwaal, Kimpen, & van de Putte, 2012). There is less evidence supporting psychodynamic, attachment-based and systemic theories, although this may be due to a lack of research into the efficacy of interventions based on these theories. Several case studies and articles detailing practice-based evidence have illustrated the potential usefulness of these theories in relation to intervention (e.g. Alvarez, 2009; Griffin & Christie, 2008; Logan & Meltzer, 2006; Maunder & Hunter, 2004; Shapiro, 2003).

Intervention: Current Evidence

There have been several recent systematic reviews and meta-analyses of the evidence base for psychological interventions in adults and children with medically unexplained symptoms including headache, pain, abdominal pain and IBS and CFS (e.g. Huertas-Ceballos, et al., 2008; Eccleston, Morley, Williams, Yorke, & Mastroyannopoulou, 2002; Fisher et al., 2014, Smith et al., 2015). Generally, these reviews have illustrated benefits of

psychological therapies in terms of improvements of physical symptoms including reduction of pain, fatigue and IBS symptoms and functional outcome including improvements in school attendance and daily activities. However, review and synthesis of mental health outcomes has been scarce and largely unclear (Fisher et al., 2014). Trials and reviews have generally prioritised physical and practical gains, and where mental health results are included, they have mainly been measured as secondary outcomes. In a systematic review of psychological interventions for pain in children, Fisher et al. (2014) found insufficient evidence to draw conclusions relating to mental health outcomes, including anxiety. A review of how recovery is defined relating to CFS was carried out in 2014 and found that improvements generally related to fatigue and physical functioning. The authors concluded that more emphasis should be placed on the person's perception of their own wellbeing (Adamowicz, Caikauskaite, & Friedberg, 2014). Given the co-morbidity of mental ill-health and medically unexplained symptoms, it was argued that more focus should be placed on reducing distress including depression and anxiety in these populations (Fisher et al., 2014).

The aims of the current review are to:

(i) Investigate the effectiveness of psychological interventions for mental health difficulties in children and adolescents with medically unexplained symptoms.

(ii) Identify significant aspects of interventions associated with their success e.g., presence/absence of parents, mode of delivery.

A systematic review methodology was used in order that the review could be repeated as the evidence base in relation to mental health symptoms and medically unexplained symptoms in children and adolescents increases.

Methodology

Search Methods

Systematic review methods were carried out using Cochrane guidelines (Higgins & Green, 2008). Electronic, reference list and citation searches were carried out in PsycINFO, MEDLINE and CINAHL databases were searched from inception to November 2015. Searches of other sources including Google and Google Scholar were also undertaken. The search terms were largely categorised into three main areas; (1) medically unexplained symptoms, (2) mental health difficulties, (3) psychological intervention (See Appendix A for lists of terms by database). Reference and citation lists of identified studies were examined for additional studies.

Inclusion Criteria

Types of studies. Only randomised controlled trials published in a peer-reviewed journal in the English language were examined.

Types of interventions. Psychological interventions which were specifically designed to alter psychological processes thought to underlie or significantly contribute to distress and suffering (Fisher et al., 2014), were included in the review.

Types of participants. Study participants were children and adolescents up to the age of 18 years with medically unexplained symptoms. These were defined as any physical symptom causing distress or impeding function which was not accounted for through medical explanation. This definition was used in the absence of an agreed upon description that is consistently used by researchers and clinicians with regards to children and adolescents (Nunn, et al., 2000; Eminson, 2007). It has been acknowledged that the ICD-10 and DSM-V (APA, 2013; WHO, 1992) diagnostic classifications are inadequate for symptoms experienced by children and adolescents (Eminson, 2007). Therefore, in order to be inclusive, a wide range of search terms were applied. These were taken from various sources including adult functional symptoms by speciality (Wessely et al., 1999), the child somatisation

inventory (Walker et al. 1991) and the somatic symptom checklist adapted for adolescents (Eminson et al., 1996).

Outcome measures. Child mental health measures relating to common mental health problems including depression, anxiety and child behaviour were included. Parent-reports of child mental health (but not parent mental health) were also included.

Exclusion Criteria

Studies not reporting a mental health outcome measure, including studies using quality of life measures as opposed to measuring mental health outcomes (depression, anxiety and behavioural difficulties) were excluded. Studies lacking a clear psychological intervention, for example, a multi-disciplinary intervention with a psychological element, were excluded. Grey literature and unpublished dissertations were also excluded.

Data Collection and Analysis

Study selection. Studies were selected based on the above criteria. Abstracts were read and full text articles were accessed to examine articles for eligibility where necessary.

Data extraction. Data extraction was carried out using the Cochrane data extraction checklist (see Appendix B; Higgins & Green, 2008) and transferred to a table of characteristics of included studies (see Appendix C) and a table of treatment and outcomes (see Appendix D).

Methodological quality assessment. The review employed the quality appraisal checklist for quantitative evaluative studies (National Institute for Health and Clinical Excellence [NICE], 2012; See Appendix E). This tool was chosen for its ability to appraise controlled intervention studies of varying quality within the area of public health research. Both internal and external validity are rated as strong (++), adequate (+) or weak (-) using pre-defined criteria from within the following domains: population and sampling, allocation, blinding, outcome measurement treatment delivery, attrition and analysis. Strong external

and internal validity are related to a low risk of bias while adequate or weak validity is related to a moderate or high risk of bias, respectively. Assessor blindness was included in the review, however, the blinding of participants and investigators in relation to treatment was excluded as this is rarely applicable to delivery or receipt of psychological treatments (Fisher et al., 2014).

Results

The initial search identified 1611 articles, following removal of duplicates. A total of 15 studies, and one follow-up study, were eligible for inclusion (see Appendix F for a table of excluded studies). Figure 1 shows the study selection process using the PRISMA flow diagram (Liberati, Altman, Tetzlaff, Mulrow, Gøtzsche, Ioannidis,... & Moher., 2009) suggested by the Cochrane handbook (Higgins & Green, 2008).

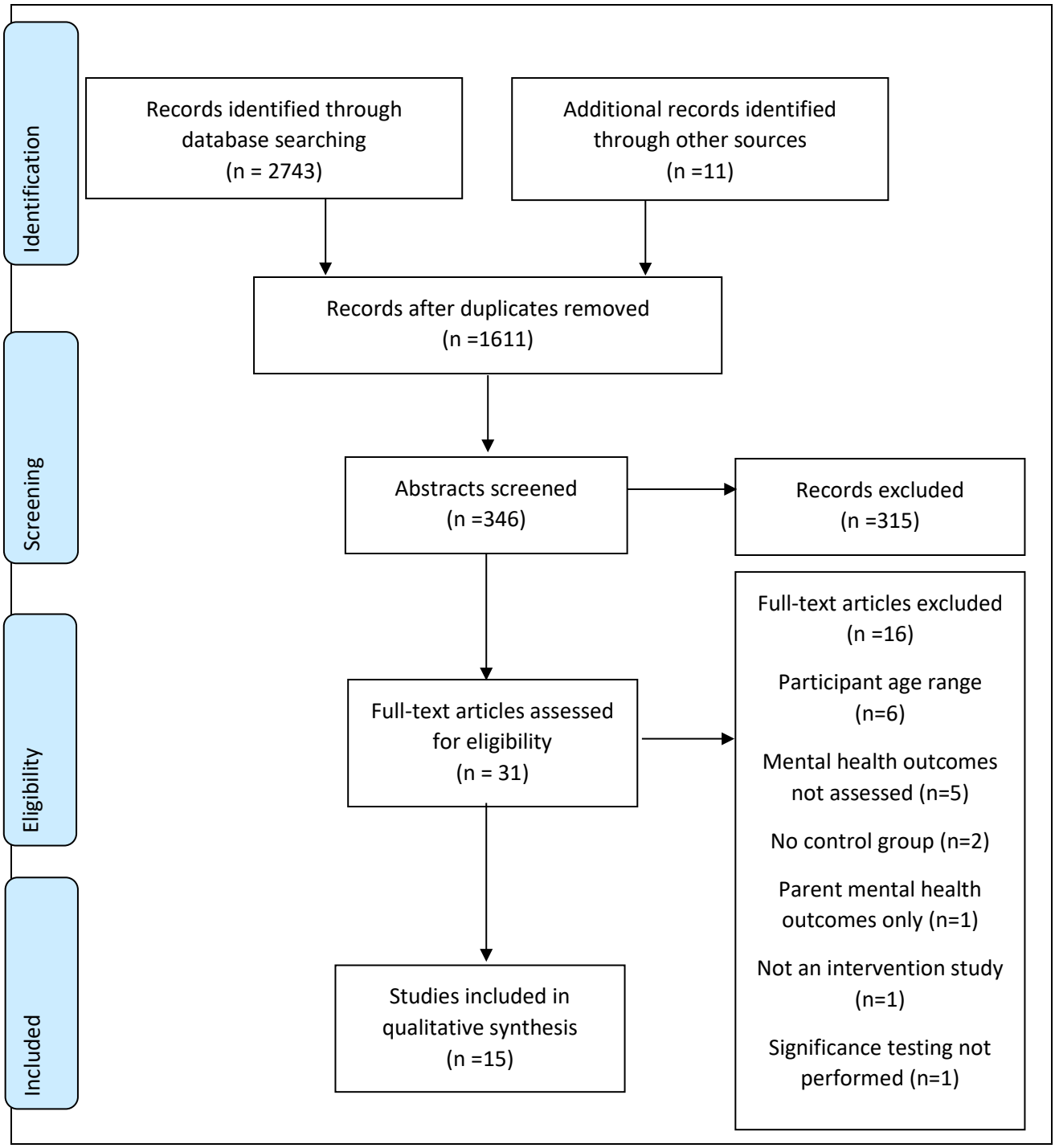

Figure 1: *PRISMA flow diagram of study selection process (Liberati et al., 2009)*

Description of Included Studies

Design and participants. All 15 studies reviewed were randomised controlled trials of relatively simple design. Twelve studies employed two treatment arms, while three used

three arms. A total of 940 participants, all experiencing medically unexplained symptoms took part in the studies. Participant age range and average age were reported in 14 and 12 studies, respectively (Range = 7-18, M = 13.46, SD = 2.75). Thirteen of the studies had participants who were predominantly female and two studies had equal numbers of male and female participants (Range = 50% - 100%; M = 67%). Eight studies included participants with headache or migraine; seven of the studies' participants were treated for gastroenterological complaints including recurrent abdominal pain, nausea, irritable bowel syndrome (IBS) and functional dyspepsia; four studies provided treatment for participants with muscular-skeletal pain, and one study treated participants with chronic fatigue syndrome (CFS).

Setting. Of the 15 included trials, ten were carried out in the USA and Canada, one in Australia, three in Northwest Europe and one (plus follow-up) in the UK. Four studies employed treatments, which were self-administered (online with therapist email contact, or via cassette tape or treatment manuals with therapist phone contact), while 12 studies involved treatment delivered by a therapist in a specialist, community or outpatient's hospital clinic.

Intervention. Interventions included Cognitive Behavioural Therapy (CBT; 12 studies) and biofeedback (three studies). Reportedly, all of the CBT studies incorporated a behavioural element for the children; eleven employed stress management or relaxation strategies, three included 'graded exposure' to avoided situations and three reported including activity pacing or sleep strategies. Additionally all CBT interventions included cognitive aspects; ten reported involving 'cognitive restructuring' or cognitive skills training, five included psycho-education, five incorporated problem-solving strategies, two employed self-esteem building strategies and one incorporated acceptance and commitment values.

Seven of the 12 CBT studies reported assigning homework as part of the therapy and three included relapse management strategies. Of the twelve studies employing CBT, four involved children only in the treatment (one of these employed child 'group' therapy) and eight involved parents as well as children. Of these eight studies, seven involved behaviour therapy directed at the parent (adapting parent responses to illness behaviours) and two involved strategies aimed at increasing parent-child communication.

For the biofeedback studies, two studies used autogenic hand-warming and the other used electromyography (EMG) biofeedback. All three studies employed progressive muscle relaxation while the hand-warming studies also reported incorporating abdominal breathing and imagery techniques. The hand-warming studies also used home practice to aid mastery of skills but the EMG study explicitly instructed participants not to practice at home in order to maximise treatment fidelity.

Regarding control conditions, five trials used education, three used standard medical care, one employed intensive medical care, two employed relaxation control groups, and one used therapist monitoring and support. Four studies used a wait-list control group (usually with self-monitoring of symptoms). One of the studies which used a wait-list control group also employed a hand-cooling biofeedback control group.

Outcome measures. In total, 28 outcome measures were used to examine treatment outcomes across a number of domains including physical health, functional disability and mental health. All trials measured self-report physical health experience, while four included objectively measured physical health status (tender point examination and EMG). Eight studies assessed functional disability or illness interference, with just one of these studies measuring school attendance. All studies assessed self-report mental health outcomes with thirteen measuring mood, eleven assessing child anxiety, which included stress and illness related catastrophising, and three studies evaluating child behaviour. One study assessed

anxiety through a diagnostic interview. Additionally, three studies assessed child social adjustment, two studies explored parent behaviour changes and four studies assessed quality of life. Three studies assessed global improvement. Coping and self-efficacy was assessed by four studies.

External Validity

Population: Sampling Bias. Generally, external validity was rated as adequate to weak. Just two studies described source population demographics and no study made reference to differences between the source population, eligible population or study population with respect to demographics. Recruitment was mainly from specialist centres, indicating that generalisability of results may be limited. Eleven studies reported recruiting from specialist rheumatology, pain, headache, CFS or gastroenterological centres or clinics, while two studies report recruitment from a general children's hospital and another from primary care. Three studies reported recruitment via newspaper and online advertisement or community flyers.

No study reported random sampling, with twelve studies describing recruitment through consecutive admissions, two studies describing convenience sampling and one study not reporting a sampling method. Studies usually had suitable inclusion criteria, specifying an age range and a diagnosis of medically unexplained symptoms. Trials also mostly had appropriate exclusion criteria, for example, three studies excluded participants with psychological issues which would require treatment before participation in the study, including suicidality or psychosis. However, several studies employed stringent criteria, limiting participant numbers and generalisability. Four studies excluded participants with common mental health diagnoses, including depression and panic disorder and one study excluded all participants not meeting criteria for an anxiety disorder.

Internal Validity

Allocation: selection bias. Group allocation was generally of adequate quality. Fourteen out of the 15 studies employed 'pseudo-randomisation' of participants to a condition due to consecutive admissions or stratification of participants based on age, and one study used true randomisation. In relation to method of randomisation, eight concealed allocation of participants through a computerised system, three created a pre-determined list or table of conditions and one study had a system of sealed envelopes which were consecutively allocated to participants. The randomisation method was not reported for three studies.

In terms of other treatments, two studies specified that neither group were receiving other interventions at the time of the study, three studies described that participants continued with standard medical care and four studies reported the use of non-prescription, stable or usual medications. One study reported that both groups included participants on anti-depressant medication and five studies failed to report on other interventions at the time of treatment.

Contamination and crossover of treatments between groups was mainly of an acceptable standard with 11 studies reporting no cross-over between groups during the study phase. Two studies reported that control groups received the treatment after one month. These studies did not follow up on control participants' outcome scores following treatment, but continued to follow the treatment group at this time. However, one study was rated as weak on this criterion as a cross-over of groups immediately following the first treatment phase was reported, with group differences being compared at the end of the second treatment phase.

Blinding: Detection Bias. As outlined above, blinding was usually not applicable for participants receiving or investigators delivering an intervention. However for one study, both groups received another intervention (standard medical care) alongside the treatment in

question. Physician's delivering this other treatment were blind to participant treatment condition. Investigator blindness to treatment condition for baseline or outcome assessment was unclear for the majority of studies. Although most measures were self-report and therefore would not necessarily require an assessor, the manner in which the measures were administered to children was usually not mentioned. Just one study reported that a nurse blind to treatment condition administered the self-report measures to children over the phone. For eight studies, non-self-report baseline or outcome measures were also reported. Five of these studies reported that assessors were blind to participant condition, while for the other three studies, assessor blindness was not reported.

Measuring Outcomes. Outcome measures employed were generally adequate. All studies were deemed to have employed relevant outcomes, although seven studies did not examine functional improvement, four studies did not measure anxiety and two studies did not measure mood. All studies employed self-report measures. Acceptable reliability and validity was reported for the majority of instruments. For nine studies, a headache or pain diary was used, although only one study reported on the validity of its diary. Headache or pain diary reliability was not reported for any of the studies.

Additionally, six studies used non-self-report outcome measures. Five of these studies used measures which required the evaluator to make a subjective decision about the wellbeing of participants, for example, a tender point examination or interview about physical or mental health symptoms. No study reported testing inter-rater reliability of these measures. Three of the six studies using objective outcomes also employed measures which did not require a subjective judgement on the part of the rater, for example, electronic equipment measuring physiological changes and school attendance records.

Eleven studies had similar follow-up times across groups, while four studies followed the treatment group for longer due to offering treatment to the control group. All studies

followed up treatment and control groups at post-treatment or one month. Six studies followed up to 4 – 7 months, a further four studies followed up to 12 months and one study carried out a 24 month follow-up of treatment and control groups.

Treatment Delivery. All fifteen interventions were manualised, with just one intervention tailoring specific modules to children's needs, based on clinical judgement and just one trial reporting catering for participant age. Studies that employed self-help strategies used a combination of online modules, manuals and audio taped guidance. Treatment was predominantly delivered by doctoral level psychologists or trainee psychologists specifically trained in providing the treatment. Two trials employed nurses trained to deliver the structured interventions and one trial employed CBT therapists. The level of therapist training was not stated for the remaining three trials.

Generally, there was an unclear or high risk of bias in relation to treatment fidelity and therapist supervision. Just three trials reported independent evaluation of treatment fidelity and a further three reported monitoring of fidelity through supervision. Additionally, two interventions comprised online modules and therefore did not require fidelity monitoring, although these studies failed to report on supervision of therapists providing email contact. The remaining seven studies either failed to incorporate or did not make reference to supervision or treatment fidelity.

With regard to condition equivalence within trials, eight studies controlled for participant time, nine controlled for participant attention and eight controlled for therapist time spent with patient. Just one study assessed the credibility of the control versus the treatment condition.

Attrition Bias. Attrition bias was largely unclear. Attrition levels were acceptable (<20%) for eleven studies and high (≤ 30%) for the remaining three studies. However, just four studies reported that attrition rate did not differ as a function of group and the remaining

studies did not report whether there were significant differences between groups' attrition rates (although two of these studies reported an attrition rate difference of ≥20% between groups). Differences between treatment completers and non-completers were reported for just one study.

Bias Relating to Analysis. All studies performed appropriate formal statistical tests of interaction assessing whether intervention effect differed according to group. Accounting for group differences in the analyses was mostly adequate with seven studies reporting no group differences on demographics or outcome measures at baseline and one study describing no group differences on primary outcome variables at baseline. Five studies report group differences at baseline which are adjusted for at analysis. However, one study was rated as 'weak' in this domain as group differences on depression scores at baseline are reported, but are not taken into account at analysis.

Power across studies was generally insufficient. Five studies reported having adequate power (≥80%) to detect between groups differences. An additional study reports having sufficient power to detect differences in primary, but not secondary outcomes. Three studies reported power calculations illustrating insufficient power (<80%) and the remainder of studies did not report power calculations. Nine studies acknowledge small sample size and insufficient power as a limitation.

Intent to treat analysis, which is said to decrease the chance of Type I errors was carried out for just five studies, with ten studies excluding drop-outs post group allocation from the analysis and one study using completer analysis. Only four studies out of fifteen applied an alpha level adjustment to correct for the number of statistical tests performed, increasing the chance of a Type I error.

Reporting bias. The risk of selective reporting was high as data were not fully reported for the majority of studies. Five studies failed to report p values, seven studies did

not report treatment effect size estimates and four studies failed to report results relating to between groups differences for some measures and subscales scores. Additionally, one study failed to report results of outcomes taken at certain time-points. Appendix G contains a risk of bias table, pertaining to categories in quality appraisal checklist (NICE, 2012) and Appendix H contains a summary of the quality appraisal, also pertaining to the categories of the NICEB (2012) quality appraisal checklist.

Effects of Interventions

Generally, studies demonstrated mixed findings relating to treatment and control group differences on mental health outcomes. Of the three studies evaluating the effect of individual CBT approaches on mental health outcomes including child anxiety, depression and relationships, no study reported a significant difference between treatment and control groups. With regard to self-administered or internet delivered CBT with limited therapeutic contact via email or phone, a similar pattern was observed. These studies examined treatment versus control group effects on mental health outcomes including child depression, anxiety, behaviour, relationships, emotional wellbeing and illness coping. No significant differences between treatment and control groups were reported on these outcomes.

On the other hand, significant between groups differences on mental health outcomes are reported for the seven studies employing therapist-delivered CBT involving parents. Four of these studies measured child anxiety and all four reported significantly greater improvements in the treatment group compared to the control group post-treatment. This difference was no longer significant for two of the studies at follow-up, although treatment gains tended to be maintained.

A similar pattern was seen for depression scores, with three out of the five studies which measured child depression reporting a significantly greater improvement in the treatment compared with the control group at the post-intervention phase. Again, while

treatment improvements were usually upheld, between groups differences were observed in only two of the five studies at follow-up. The lack of significant group differences at follow-up on child anxiety and depression scores was often due to improvements control group scores or unavailable data due to control groups not being followed up. One study involving parents also measured children's emotional and behavioural adjustment and found significantly greater improvement in the treatment group compared with the control group from pre-treatment to 24 month follow-up, but not from pre-treatment to 12 month follow-up.

For the three biofeedback studies, one measured the effect of an intervention on depression another on anxiety and another on both depression and anxiety. Two of the studies reported between groups' differences from pre-to post-intervention or pre-intervention to follow-up on their respective mental health outcomes. However, for both studies once baseline levels were adjusted for, these effects disappeared. The third study found no effects of their intervention on mental health outcome.

Where studies failed to demonstrate a between groups effect, the majority of studies illustrated a statistically significant improvement for the intervention group on at least one mental health outcome measure.

Meta-analysis

As the current data did not meet recommendations for meta-analyses outlined in the Cochrane guidelines, including a low risk of bias within studies and homogeneity across studies, it was concluded that a meta-analysis of these data would not be meaningful (Higgins & Green, 2008). The studies comprise a clinically diverse population (headache, gastroenterological complaints, pain and CFS), diverse treatment delivery (family, individual and self-administered CBT and biofeedback) and diverse mental health outcome measures. Furthermore, the presence of bias was noted in the majority of the studies with nine studies judged to have low internal or external validity and one studies judged to have low internal

and external validity. According to the Cochrane guidelines, if bias is present in all or a percentage of the individual studies, meta-analysis is likely to compound biases and yield a misleading result (Higgins & Green, 2008). Furthermore, several studies failed to report treatment effect sizes necessary for collation of data.

Consideration was given to separating symptoms out and performing discrete quantitative synthesis on each symptom group, however the largest discrete group in the current review was pain. A recent meta-analysis of chronic pain in children included depression and anxiety outcomes and found that there was insufficient evidence to draw conclusions from the meta-analytic data regarding anxiety and could only draw conclusions from a pain subgroup (musculoskeletal) with regard to depression (Fisher et al., 2014). Taking each of these factors into account, it was decided that a meta-analysis would not currently add useful information to the evidence base.

Discussion

Fifteen trials met inclusion criteria for the current review. The main findings in relation to the two study aims were that;

(i) evidence for the effectiveness of psychological interventions for common mental health difficulties in children and adolescents with medically unexplained symptoms is limited due to study numbers and a high or unclear risk of bias within the majority of these studies. The most rigorous studies showing the lowest risk of bias included Levy et al. (2010), Palermo, Wilson, Peters, Lewandowski, & Somhegyi (2009), and van der Veek, Derkx, Benninga, Boer, & de Haan (2013), followed by Scharff, Marcus & Masek (2002), and Trautmann and Kroner-Herwig (2010). The remaining ten studies showed a high risk of bias in at least one of the overall domains (see Appendix

H for a table summarising study bias). The study with the highest risk of bias was Kashikar-Zuck, Swain, Jones, and Graham (2005).

(ii) Treatments which include parents and which are delivered by a therapist (as opposed to online modules) appear to yield better mental health outcomes compared to control groups.

Study design was generally simple, involving one or two treatment groups evaluated against a waiting list, treatment as usual or placebo control group. Randomisation procedures were generally acceptable, however blindness in relation to measurement of outcome was usually unclear, with just one study making reference to the manner in which self-report measures were administered by investigators at baseline and outcome (Levy et al., 2007).

Although, due to the psychological nature of treatments, investigators and participants were not blind to intervention delivery for any study, it is imperative that careful attention is paid to condition equivalence in the design of psychological treatment studies. This is in order to ensure control condition credibility is maximised and in order that active treatment components can be identified (Eccleston et al., 2002). For the studies included in the current review, condition equivalence within trials was poor, with just half of the trials employing control groups controlling for participant time or therapist time spent with participant and five trials (Kashikar-Zuck, et al., 2005; Palermo et al., 2009; Schurman, Wu, Grayson, & Friesen, 2010; Warner et al., 2011; Wicksell, Melin, Lekander, & Olsson, 2009) failing to control for participant attention altogether. Furthermore, treatment and control condition credibility, allocation of therapist to control or treatment group, and supervision of therapists were cited infrequently. In addition to this, although manualisation of treatment was high, adherence to manuals was assessed by an independent evaluator in only three trials (Kashikar-Zuck et al., 2012; Levy et al., 2010; Wicksell et al., 2009).

While accounting for group differences at baseline and through analyses was acceptable for the most part and attrition was acceptably low for most trials, attrition bias was generally unclear, with studies rarely referring to differences between groups' attrition rates or differences between completers and non-completers. Power within trials was largely weak, with just five studies reporting adequate power (≥ 80%) to detect between groups differences (Chalder, Deary, Husain, & Walwyn, 2010; Kashikar-Zuck et al., 2012; Levy et al., 2010; Palermo et al., 2009; van der Veek, et al., 2013) increasing the possibility of Type II errors in the remainder of studies. Additionally, intent to treat analysis was not carried out for the majority or studies, increasing the likelihood of a Type I error. Furthermore, results from analyses conducted were not fully reported for the majority of trials, with just five trials fully reporting results (Hickman, Jacobson, & Melnyk, 2015; Kashikar-Zuck et al., 2006; Palermo et al., 2009; Trautmann & Kroner-Herwig, 2010; Wicksell et al., 2009), suggesting a high degree of reporting bias and undermining the usefulness of pooling information about the effectiveness of these interventions.

Studies generally had weak external validity due to poor description and comparison of source population characteristics with study participants, high numbers of recruitment from specialist centres and poor sampling methods. Additionally, research shows that mental health conditions including depression and anxiety are experienced between 30 and 60 percent of those suffering with medically unexplained symptoms. However, several studies employed stringent criteria which excluded participants based on their diagnostic status in relation to these common mental health conditions (Chalder et al., 2010; Hickman et al., 2015; Kashikar-Zuck et al., 2005; Kashikar-Zuck et al., 2012; McGrath et al., 1992; Warner et al., 2011). Taken together, these concerns limit the generalisability of the studies included in the current review.

Findings

Overall, the CBT studies generated some interesting and informative findings in relation to mental health in children and adolescents with medically unexplained symptoms, although inferences should be considered with the caveat that condition equivalence was largely poor or unclear. Trials which involved parents demonstrated the most positive outcomes in mental health. Additionally, treatment gains were generally maintained in this group. In particular, those therapist-delivered interventions including components addressing parental responses to child illness behaviours consistently illustrated significant improvements in child anxiety (Levy et al., 2010; van der Veek et al., 2013; Warner et al., 2011; Wicksell et al., 2009) and also showed reductions in child depression (Kashikar-Zuck et al., 2012; Levy et al., 2010; van der Veek et al., 2013). Interventions including elements aimed at encouraging family communication about illness also demonstrated effectiveness in relation to child mental health when compared to an active control group (Chalder et al., 2010; Lloyd et al., 2012). This is consistent with research linking parental modelling of illness behaviour and parental solicitousness of child illness behaviour with higher rates of anxiety, depression and medically unexplained symptoms (e.g. Zuckerman et al., 1987) and with research illustrating that parental solicitousness influences aspects of child anxiety (children's catastrophisation of symptoms; Levy et al., 2004). Proportion of sessions involving parents did not appear to be a factor, with some interventions consisting of joint parent-child sessions only and others including parents for only a few sessions. Age did not appear to impact intervention effectiveness either, with younger children and older adolescents showing reduced anxiety and depression.

Delivery of CBT by a therapist in person or over the phone also demonstrated some value. Although no differences between groups were observed, there were consistent significant within groups differences in treatment groups from pre- to post intervention across

trials. One trial found that while therapist-delivered individual CBT showed a significant improvement in anxiety scores from baseline to post-treatment, neither the self-administered control group nor the waitlist control showed improvements (Griffiths & Martin, 1996). A parallel pattern was seen for depression scores, although this did not reach statistical significance. However, a trial with a similar format, but with an active control group found that all three groups improved significantly on depression scores (McGrath et al., 1992). Similarly, a third trial evaluating individual CBT (Hickman et al., 2015) found that both the treatment and active control group improved on measures of anxiety and depression, however, effect sizes were larger in the treatment group, indicating that the lack of power in these studies due to small sample sizes may have impacted results. In a systematic review of the literature, Chorpita et al. (2011) found strong evidence for the effectiveness of individual CBT for children with anxiety and depression. There is also evidence to suggest that individual CBT interventions are effective in reducing mental health symptoms for children who have a chronic illness with a known pathology (Bennett et al., 2015).

Two studies assessed online treatment plus weekly email contact with an active control group. One study (Trautmann & Kröner-Herwig, 2010) was individual and another (Palermo et al., 2009) addressed issues relating to parental responses to child illness. Neither of these studies demonstrated between groups differences on any mental health measure at any time point. The study which included components addressing parental responses to illness behaviour showed benefits at 3 month follow-up on depression scores, although the control group was not followed up. The individual online CBT course showed within group differences in pain catastrophising (a measure of anxiety) in the treatment group and in both of the active control groups. However, effect size was larger in the CBT group, again showing that power may have been an issue in detecting significant differences. Indeed, for internet-delivered interventions, current CBT evidence for improvement in mental health

symptoms for children who do not experience medically unexplained symptoms is strong (Ebert et al., 2015).

Generally, while biofeedback trials tended to be somewhat effective for physical symptoms, evidence for their effectiveness in the treatment of mental health difficulties is less promising. None of the three trials (Bussone Grazzi, D'Amico, Leone, & Andrasik, 1998; Scharff, et al., 2002; Schurman et al., 2010) demonstrated significant gains in the treatment group compared with the control group on any measure of mental health, indicating that there is currently no evidence to suggest biofeedback is a suitable treatment for those experiencing mental health difficulties along with medically unexplained symptoms.

Improvement in mental health symptoms did not appear to be related to improvement in physical health symptoms at the post-treatment phase. Several studies illustrated reduction in medically unexplained symptoms, but not improvement in depression or anxiety (e.g. Bussone et al. 1998, Griffiths et al., 1996; McGrath et al., 1992). Other studies showed the reverse pattern (e.g. Chalder et al., 2010; Lloyd et al., 2012; van der Veek et al., 2013). This is consistent with previous reviews' findings indicating similar patterns (e.g. Kroenke & Swindle, 2000).

However, given the strong relationship between mental health symptoms and medically unexplained symptoms, it is possible that mental and physical health symptoms interact and impact one another between the post-treatment and follow-up phase, or thereafter. Indeed, although there is good evidence for the effectiveness of psychological treatments for physical health outcomes for children with chronic pain, there is an overwhelming lack of evidence for lasting effectiveness on physical outcomes at follow-up (Fisher et al., 2014). This suggests that other factors may prevent maintenance of physical health gains. According to cognitive-behavioural models of medically unexplained symptoms, psychological processes can maintain physical symptoms. Indeed in a review on

psychological risk factors of unexplained pain, Keefe, Rumble, Scipio, Giordano and Perri (2004) found associations between persistent pain, pain catastrophisation, and other anxiety related beliefs, indicating that maintenance of physical symptom improvement following therapy may be impacted by mental health.

Research Implications

The current review highlights several areas for further research. There was a lack of power to detect statistically significant differences between groups, particularly in relation to studies measuring the effectiveness of individual CBT and internet-delivered CBT. Additionally, condition equivalence was poor. Therefore further research including large-scale randomised control trials (RCTs) with active control groups, controlling for participant time and attention are necessary in order to gauge the effectiveness of these interventions in relation to mental and physical health outcomes in children with medically unexplained symptoms.

While there are numerous large scale trials which have explored the effects of psychological interventions on medically unexplained symptoms, despite the relationship between medically unexplained symptoms and mental health symptoms, the majority of these trials fail to include outcomes focussed on mental health (e.g. Kroener-Herwig, & Denecke, 2002; McGrath et al., 1998; Passchier et al., 1990). Griffin and Christie (2008) argue that the boundaries between physical and mental health are blurred where children and adolescents present with medically unexplained symptoms. Regardless of the widely recognised usefulness of the biopsychosocial model of health (e.g. Salt & Season, 2000; Williams & Erskine, 1995) the concept of the mind-body split (dualism) proposed by Descartes in the 17th century continues to direct western language, culture and perhaps even healthcare and research.

With regards to medically unexplained symptoms, physical health gains are prioritised and mental health outcomes are often viewed as an after-thought. This is reflected in the reviewed studies, where mental health measures were usually secondary outcomes and often not fully reported. It is also reflected in large-scale randomised controlled trials of psychological interventions for this population, where mental health outcomes have not been included (e.g. Nijhof et al., 2012). Trials are often deemed successful by authors if physical health outcomes improved, regardless of effectiveness in relation to mental health. Future trials should assess the interaction between physical and mental health outcomes at various follow-up time points in order to examine the impact of these outcomes on each other over time. Data on this is currently sparse and could inform clinical intervention in addition to explanatory theories of medically unexplained symptoms.

There is a similar pattern for children with chronic health conditions of a known pathology. Physical health is often prioritised with mental health difficulties often going untreated, despite rates of mental illness being up to four times higher in this population compared with those children who are physically well (Pinquart & Shen, 2011). Ott et al. (2003) found that only 33% of children presenting with epilepsy plus a psychiatric disorder had received a mental health intervention, despite attending frequent medical appointments for epilepsy. There are a number of possible reasons for the lack of integration of mental and physical health services for children with medically unexplained symptoms and chronic physical health conditions. Children with medically unexplained symptoms and chronic physical illness, along with mental health conditions are seen as complex. Where this is the case, physical healthcare is often prioritised by healthcare professionals, unintentionally leading to neglect of mental healthcare (Bennett et al., 2015).

Additionally, due to limited research about mental health outcomes for children with chronic physical symptoms and conditions, healthcare professionals may not have well

established guidance and information they feel can be relied upon to assist them in decision-making processes involving interventions with established efficacy for this population (Fisher, et al., 2014; Bennett et al, 2015). Thus children and adolescents are unable to gain access to timely, suitable interventions which support mental health difficulties in the context of physical health symptoms and conditions (Katon et al., 2006). For both medically unexplained symptoms and chronic illness in children, there is a paucity of research about which children get referred for psychological input, and the reasons for a referral. According to Grol, Bosch, Hulscher, Eccles & Wensing (2007) most clinical practice choices are based on an individual's professional decision. In line with this, research into factors which facilitate and hinder health-care professionals' referral of children with chronic and medically unexplained illness to psychology is indicated.

Clinical Implications

Identifying key aspects leading to positive treatment outcomes is challenging for a number of reasons. According to Fisher et al., (2014), components of interventions are intended to interact and combine with one another to produce an effect, meaning that removing certain aspects of interventions or separating elements from one another may not be useful as one component may need another component in order to be effective. For the current review, while interventions were manualised, study protocols and available treatment manuals did not contain sufficient detail to thoroughly examine specific elements of treatment, for example, how much of the intervention focusses on which aspects, whether this differs across therapists and participants, and whether therapist skill, therapeutic alliance or extra-therapeutic factors have also influenced results. According to Fisher et al. (2014), comparable concerns are apparent with regards to maintenance of improvements over time.

Therefore, it is not possible to conclude based on these data whether one aspect of treatment is active, while another is not.

However, several studies within this review showed the benefits of including parents. A focus on response to illness behaviour or family communication in relation to illness for at least some of the sessions was a salient factor of these interventions. While further randomised controlled trials of sound quality with clear *a priori* hypotheses are necessary to support the significance of this treatment component, these findings may be clinically valuable. They illustrate the usefulness of including parents in sessions and are supported by theory (Bandura, 1977) and empirical evidence in the literature (Levy et al., 2004). They also demonstrate the usefulness of including family interactions within psychological formulation, even if it is not possible to include parents in interventions.

Limitations of the review

The limited scope of the review (RCTs only) meant that the main interventions reviewed were CBT-based and that evidence for less well-researched interventions was excluded. Furthermore, while the search criteria were inclusive in terms of symptomology, broadening the intervention-type and outcome search terms may have provided a wider picture (e.g. attachment-based interventions were not included in the search criteria).

Conclusions

Psychological interventions are effective in improving mental health outcomes for children and adolescents with medically unexplained symptoms. Interventions including aspects relating to parental responses to illness behaviour and family communication appeared to have the best outcomes. Biofeedback studies were effective in reducing physical but not mental health symptoms, while further research is necessary in order to ascertain the

effectiveness of individual and internet-based CBT for mental health outcomes in this population. Furthermore, given explanatory theories and research about the processes maintaining medically unexplained symptoms which emphasise the relationship of medically unexplained physical symptoms with psychological processes such as pain catastrophisation, researchers and clinicians should prioritise both physical and mental health outcomes. Related to this, while physical health gains did not appear to be associated with mental health improvement (or vice versa), further investigation should assess the interaction between physical and mental health outcomes at various time points in order to examine potential impacts of these outcomes on each other over time.

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MAJOR RESEARCH PROJECT

SECTION B

Factors influencing the referral of children and families to paediatric psychology: Using Theory of Planned Behaviour to develop a questionnaire of health care professionals' referral behaviour.

WORD COUNT

7704 (+360)

Abstract

Background. Research shows that the integration of physical and mental healthcare in paediatric settings is beneficial in terms of clinical and cost effectiveness. Due to the high rates of mental health problems within this population, several studies have shown that referral to paediatric psychology should be increased. However, there are few studies investigating factors influencing healthcare professionals' referral behaviour.

Methods. The current study used Theory of Planned Behaviour to develop a questionnaire which explores factors influencing the referral of children and families to paediatric psychology. Psychometric properties of the questionnaire were examined.

Results. Findings indicate that the questionnaire holds good reliability and validity and that the main constructs of Theory of Planned Behaviour are useful in predicting intention to refer to paediatric psychology. Specific beliefs about referral were also shown to influence intention to refer.

Conclusions. Findings that individual referrer factors such as attitudes and beliefs can impact healthcare professionals' referral behaviour indicates that multidisciplinary interventions and inter-professional education relating to the psychological aspects of illness are required. Recommendations for future research are discussed.

Introduction

Children with physical illness can experience high amounts of pain, distress and maladjustment (Duff & Bryon, 2005). For these children, rates of mental health problems are up to four times higher than children in the general population (Meltzer, Gatward, Goodman, & Ford, 2004; Pinquart & Shen, 2011). Psychological therapies are effective for reducing mental and physical health problems in children and adolescents with chronic physical health conditions and medically unexplained physical symptoms (Bennett, Shafran, Coughtry, Walker & Heyman, 2015; Fisher et al., 2014; Warner et al., 2011). However, carrying out interventions on co-morbid mental health problems in isolation is not always effective, sometimes only reducing either physical or mental health problems (Chalder, Deary, Husain & Walwyn, 2010; Hains, Davies, Behrens & Biller 1997; Mcgrady & Hood, 2013; Van der Veek, Derkx, Benninga, Boer, & de Haan, 2013). According to Naylor et al. (2012), more substantial clinical and cost effectiveness may be obtained through integration of physical and mental health treatments, instead of adding mental health interventions to existing physical treatment practices.

The biopsychosocial model of health suggests that in order to respond effectively, it is necessary for healthcare professionals to hold biological, psychological, and social dimensions of illness in mind (Engel, 1977). Indeed research demonstrates that developing a framework of support that integrates physical and mental health services for children with chronic health conditions and their families can improve physical and mental health outcomes (Griffin & Christie, 2008; Kazak, 2005; Spirito & Kazak, 2006). Psychological integration can improve psychological adjustment, medical treatment adherence and self-management for children with physical health conditions (Kahana, et al., 2008; Wysocki, et al., 2006). Furthermore, integrated treatment can increase communication between healthcare professionals, leading to improved patient outcomes (Zwarenstein, Goldman & Reeves, 2009;

Strasser et al., 2005). Integrated treatment can combine the similarities between physical and mental health care, for example, between self-management protocols for long-term conditions and behavioural activation approaches for low mood (Egede & Ellis 2010). Tensions between different treatments can be avoided, decreasing cost and improving the treatment experiences of children and families (Griffin & Christie, 2008; Naylor et al., 2012).

In addition to clinical benefits, adult literature relating to the integration of physical and mental health services illustrates cost-effectiveness. In a meta-analysis, Chiles et al. (1999) demonstrated that psychological interventions in hospitals and primary care settings decreased overall health care costs. Howard, Dupont, Haselden, Lynch and Wills (2010) found that incorporating a psychological component into a chronic obstructive pulmonary disease hospital clinic meant fewer emergency service visits and reduced hospital bed days. While child literature relating to the cost of service integration is sparse, qualitative research suggests that cost-effectiveness may be another advantage of integrating paediatric physical and mental healthcare (Douglas & Benson, 2015; Kaplan & Groessl, 2002). Despite evidence for the clinical and cost effectiveness of integration, UK health care services are not structured and organised to support an integrated response to physical and mental healthcare needs (Naylor et al., 2012).

Paediatric psychology is a relatively new branch of clinical psychology which aims to integrate physical and mental healthcare through incorporating psychological principles and approaches in medical settings (Kazak et al., 2007; Kessler et al., 2005). Paediatric psychologists work together with other healthcare professionals to understand and address the high rates of mental health difficulty amongst children with physical health conditions and their families, reduce emotional consequences of physical ill-health and improve quality of life for young people (Duff & Bryon, 2005). While a proactive (rather than referral based) integrative approach to physical and mental health care has been proposed as the ideal model

for paediatric services, it is necessary for physical healthcare professionals in community and hospital settings to make a referral for psychological services in the UK, Europe and USA, if they feel this is indicated (Kazak et al., 2007).

Several studies have shown limited interaction with and referral to psychological services within paediatric settings. Glazebrook, Hollis, Heussler, Goodman, and Coates (2003) demonstrated that in a UK paediatric outpatient hospital setting, paediatricians identified just a quarter of cases where emotional and behavioural difficulties were indicated in addition to a physical health condition. Furthermore, paediatricians said they would refer just 14 percent of children to an on-site mental health service, although 21 percent were identified as having psychological difficulties. Wagner and Smith (2007) tracked referral patterns to an on-site psychology service in a paediatric epilepsy clinic. Referral rates were significantly lower than the prevalence of mental health difficulties in children with epilepsy. Authors emphasised the necessity of facilitating increased psychology referrals within the paediatric population.

However, children with physical illness along with mental health difficulties are often seen as complex. Where this is the case, physical healthcare may be given precedence by health care professionals, meaning that mental health needs can be overlooked (Bennett et al., 2015). Thus, children are unable to access services which support mental health in the context of physical health difficulties (Katon et al., 2006). In line with this, research into factors which facilitate and hinder health-care professionals' referral of children with physical illness to psychology is indicated. Given that referral guidelines have been shown to have had little impact on the referral behaviour of healthcare professionals (e.g. O'Donnell, 2000), considering factors influencing referral behaviour may be a first step to integrating physical and mental health services more effectively.

While factors influencing referral behaviours are currently under-researched (Beacham, Herbst, Steitweiser, Scheu & Sieber, 2012; Reeves, Perrier, Goldman, Freith & Zwarenstein, 2013), several studies have found associations between mental health referral rates and referrer characteristics, attitudes and beliefs (Hugo Kendrick, Reid, & Lacey, 2000; Green Johnston, Cabrini, Fornai, & Kendrick, 2008). Kainz (2002) divided doctors who work in multi-specialist services, which included psychologists, into two groups: high and low psychology referrers. Through qualitative observations, she noted that doctors with high rates of referral to psychology tended to be more positive about psychological services than low referrers. Green et al., (2008) found that much of the variance in GP referral rates to specialist eating disorder services was based on GP cognitive attitudes and subjective norms.

Consistent with this, several authors have hypothesised that attitudes, prejudice and specific beliefs between different professional groups have the potential to improve or dilute integration of separate health disciplines (Mitchell, Parker, Giles, & Whilte, 2009; Parsell and Bligh, 1999). Despite the importance of integration of psychological services in paediatric settings, studies suggesting variation in psychology referral rates and research demonstrating that referrers' beliefs and attitudes may play a role in successful integration of services, there is currently no research regarding factors influencing healthcare professionals' referral of children and families to paediatric psychology.

Theory of Planned behaviour

In a systematic review of the literature Godin, Bélanger-Gravel, Eccles and Grimshaw (2008), found that Theory of Planned Behaviour (Ajzen, 1988, 1991) appears to be an appropriate means of understanding factors predicting behavioural intention in healthcare professionals. According to this theory, an individual's intentional behaviour can be predicted based on attitudes, subjective norms and perceived behavioural control, all of which are influenced by beliefs. Behavioural beliefs create a positive or negative attitude toward a

behaviour; normative beliefs produce subjective norm (perceived social obligation); and control beliefs lead to perceived behavioural control. Together, these constructs lead to a behavioural intention, which people usually carry out (Ajzen, 2002).

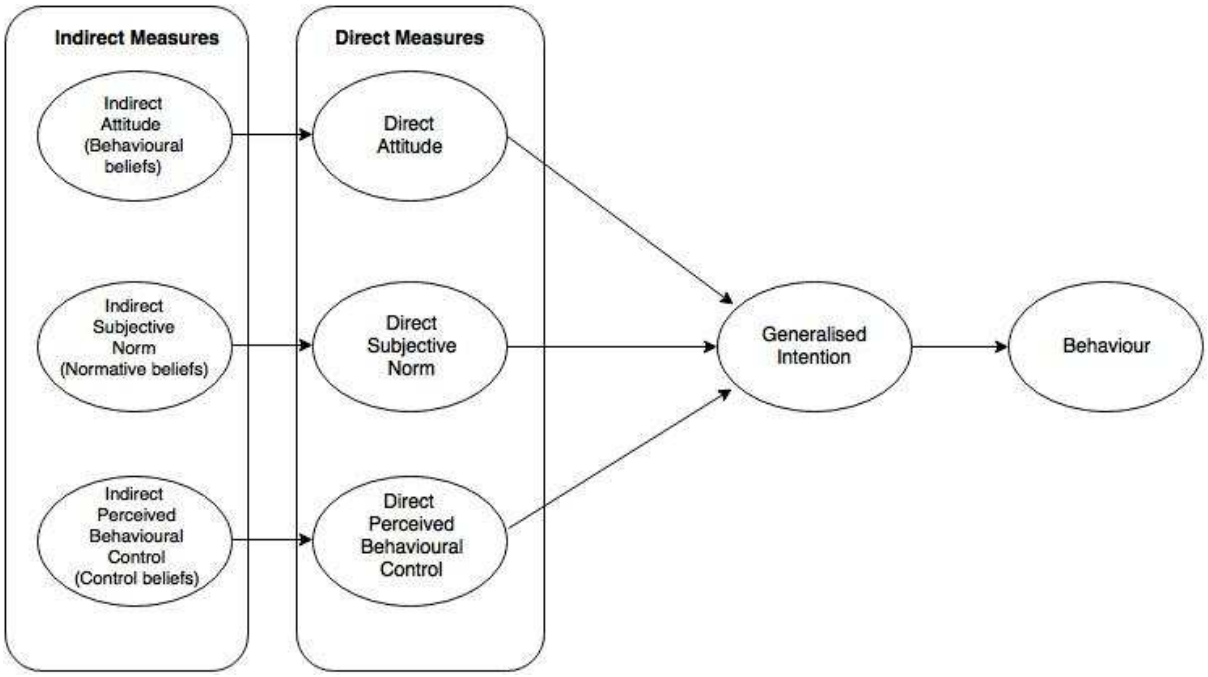

Figure 2: *The Theory of Planned Behaviour (Ajzen, 2002; p1.)*

Questionnaire Development

In order to determine beliefs, attitudes, subjective norm and perceived behavioural control relating to the behaviour of healthcare professionals, Francis et al. (2004) recommend the development of a questionnaire using Theory of Planned Behaviour. To identify beliefs related to the behaviour, a belief elicitation study involving the collection of qualitative data through open ended questions relating to the behaviour is recommended. The beliefs inferred can then be used to develop a quantitative questionnaire which allows the researcher to assess which specific beliefs significantly predict behavioural intention. Direct measures of the Theory of Planned Behaviour constructs are also included in the quantitative questionnaire.

Similarly, these data can be used to find whether direct measures of attitude, subjective norm and perceived behavioural control significantly predict the behaviour in question.

To assess the feasibility of using a questionnaire to examine Theory of Planned Behaviour constructs, it is important to monitor response rate and missing data (Bouwmans et al., 2013; Varni, Burwinkle, Seid, & Skarr, 2003). Response rates are often lower in medical professionals compared with other populations, due to demanding work schedules (Flanigan, McFarlane & Cook, 2008). Therefore using email rather than paper based questionnaires may be of benefit (Cummings, Savitz & Conrad, 2001). Similarly, keeping the questionnaire brief is necessary in order to maximise response rate (Flanigan et al., 2008).

In order to evaluate whether the questionnaire is reliable and valid, Francis et al. (2004) recommend assessing the psychometric properties of the questionnaire. Two types of reliability; internal consistency and test-retest reliability are suggested. Internal consistency aims to assess the degree to which different items which are intended to investigate the same construct produce similar results. Test-retest reliability endeavours to evaluate the degree to which the questionnaire produces stable and consistent results. It is also important to test validity to determine whether the questionnaire is measuring the constructs it intends to measure. Concurrent validity is illustrated when two scales aiming to measure the same construct are related, while construct validity is established when theoretical constructs of cause and effect accurately represent the actual constructs they are intended to model.

Aims of the current study

This study has the following aims and hypotheses:

Stage 1. (1) To identify specific beliefs relating to paediatric healthcare professionals' referral of children and families to psychology. This was investigated through collection and analysis of qualitative data of healthcare professionals' beliefs about referral to paediatric psychology.

Stage 2. (2) To assess whether Theory of Planned Behaviour constructs (attitudes, subjective norm and perceived behavioural control) can be used to predict healthcare professionals' intention to refer to paediatric psychology. This was investigated through developing and testing the feasibility, reliability and validity of a questionnaire based on Theory of Planned Behaviour.

In order to assess feasibility, questionnaire response rate and missing data were examined. It was expected that response rate would be lower than email surveys of the general population (approximately 36%; Sheehan, 2001) due to the profession of the sample.

In order to test the reliability of the questionnaire, the following hypotheses were tested:

(i) Items within composite subscales of direct measures are significantly correlated with one another.

(ii) Indirect (belief based) composite subscales at Time 1 are correlated with the same subscale at Time 2.

In order to test the validity of the questionnaire with respect to Theory of Planned Behaviour, the following hypotheses were tested:

(iii) Indirect (belief based) measures are associated with direct measures of the same construct.

(iv) Referrer attitude, subjective norm and perceived behavioural control influence intention to refer to paediatric psychology.

(3) In order to gauge specific beliefs influencing referral to paediatric psychology, the following hypothesis was tested:

(v) Specific beliefs regarding attitude, subjective norm and perceived behavioural control influence intention to refer to paediatric psychology.

Methods

Design

A two stage qualitative and quantitative study with an observational design was used to create a cross-sectional online questionnaire aiming to investigate factors influencing referral to paediatric psychology by healthcare professionals. The questionnaire was developed in accordance with a Theory of Planned Behaviour questionnaire manual (Francis et al., 2004) which outlines the steps involved in constructing a Theory of Planned Behaviour questionnaire. Therefore, the questionnaire was developed in two stages. Stage one involved eliciting beliefs held by healthcare professionals about referring children and families to paediatric psychology through a series of open-ended questions (see Appendix I). The main questionnaire was then developed using themes derived from analysis of this material (indirect beliefs-based measures), in addition to the development of items aiming to measure direct predictors (core Theory of Planned Behaviour constructs) of referral to paediatric psychology. The resultant questionnaire was piloted on six healthcare professionals prior to stage two. Stage two involved administration of the main questionnaire to healthcare professionals and re-testing the questionnaire approximately four weeks later.

Ethics

The study received full approval from the Salomons Ethics Panel at Canterbury Christ Church University and from the NHS Research and Development offices at both study sites through the Integrated Research Application System (IRAS) prior to study commencement (see Appendices J, K, and L for Ethics and R&D letters). All study participants were provided with a study information sheet and gave their consent to participate (see Appendix M).

Recruitment sites

Data were collected from two UK hospital sites. For both sites, referrals are received from three sources; from inpatients, from outpatients and as part of a multi-disciplinary package of care. Site one manages approximately 70,000 peadiatric cases per year, with approximately 1500 to 3000 referrals to paediatric psychology per annum. Site two manages approximately 10,000 paediatric cases per year and the paediatric psychology service receives referrals for approximately 200- 250 cases per annum. These figures exclude those presenting to accident and emergency only. Therefore, for both sites, referrals are made to paediatric psychology for an estimated 2 to 5 percent of cases.

Stage 1: Questionnaire Development

Participants. Participants were consultant medical doctors who work within medical teams which have access to input from paediatric psychology (n = 23). Recruitment was carried out through convenience sampling from two UK hospital sites.

Procedure. All eligible participants (n = 99) were emailed as a group by the lead paediatric psychologists within the two hospital sites and subsequently emailed individually by the researcher with an online link to a set of open ended questions (see Appendix I) which were accessed through Qualtrics online survey software. The questions, taken from the questionnaire manual (Francis et al., 2004), aimed to elicit beliefs related to referral to paediatric psychology. Core factors of Theory of Planned Behaviour, on which the set of open-ended questions were based, included healthcare professionals' attitudes towards, normative beliefs about and perceived control over referral to paediatric psychology. Francis et al. (2004) recommends obtaining responses from approximately 25 individuals in order to elicit a range of beliefs to the point where saturation of themes is likely. Twenty-five per cent of those contacted (n=23) responded to the questions.

Questionnaire development. The main questionnaire was developed using Ajzen's (2002) model of Theory of Planned Behaviour which included direct and indirect predictors of behavioural intention (Figure 2). Each direct and indirect measure (statement) was presented on a 7-point Likert scale. Twelve questionnaire items which aimed to directly measure predictors of referring to paediatric psychology were developed in accordance with the manual (Francis et al., 2004). Three of these items aimed to measure attitude to referral, three were intended to measure subjective norm, three targeted perceived control and three aimed to measure intention.

Direct attitude was measured using pairs of semantic descriptors (e.g. "*useful/worthless*") which were attached to the sentence stem, "Overall, I think referring to paediatric psychology is…" Items relating to others' views were used to measure direct subjective norm, for example: "most people whose opinions are important to me think that I should refer to paediatric psychology". Direct perceived behavioural control was examined using items such as: "Whether I refer children/families to paediatric psychology is within my control". Three items were designed to measure intention to refer, for example, "I expect to refer children/families to paediatric psychology".

Indirect (belief-based) measures were also developed based on Ajzen's (2002) model of Theory of Planned Behaviour. According to this model, beliefs provide the emotional and cognitive basis for attitude, subjective norm and perceptions of behavioural control. Therefore, beliefs can provide indirect measures of these key constructs. Development of the indirect items of the main questionnaire are presented in the next section as their development involves analysis of qualitative data.

Data Analysis. Responses to the belief-eliciting questions were downloaded to Microsoft Word. Content analysis, a method of analysing written or verbal data, was employed to analyse the data (Cole, 1988). Content analysis allows for frequency of themes

to be considered, permitting the researcher to exclude the least frequently cited themes from the main questionnaire. This is necessary in order to develop a suitably brief questionnaire. According to Francis et al. (2004), including 75 percent of themes should give adequate coverage of commonly held beliefs within a population.

Deductive content analysis was used as it also allows for the researcher to code data according to a pre-existing model (Elo & Kyngas, 2007). A coding frame was developed according to Theory of Planned Behaviour constructs (see Appendix N for coding frame), and themes were ranked from most to least frequently occurring. The data from a randomly selected participant was analysed independently by a second rater, who was supplied with the pre-prepared coding frame. The Kappa statistic indicated very good inter-rater agreement (Pallant, 2007), Kappa = 0.8 ($p = .005$).

Each Theory of Planned Behaviour construct contained four themes. The least frequently occurring theme within each category was excluded, resulting in a final total of nine themes (three for each construct). Subsequently, themes were transformed into statements. For each behavioural belief statement, a corresponding statement was developed which aimed to evaluate the outcome of that belief. Similarly, for each normative belief statement there is a corresponding statement assessing motivation to comply with the normative belief and for each statement of control belief there is a statement about the power of the control belief over the behaviour. Therefore, 18 items representing indirect measures were developed from the belief elicitation data, making a total of 30 direct and indirect measures of referral behaviour (see Appendix O for the main questionnaire).

Consultation with relevant population and quality assurance checks. A large multi-disciplinary team (MDT) meeting at one of the research sites was attended by the researcher who requested that MDT members (healthcare professionals) stay behind to complete and give feedback on a questionnaire about referral to paediatric psychology. Six

healthcare professionals (four nurses, one consultant doctor and one social worker) agreed to contribute to this task. Healthcare professionals were asked to complete the questionnaire individually and were subsequently asked a series of questions as a group. These questions, taken from the questionnaire manual (Francis et al., 2004) related to the wording and format of items. Minor changes were suggested and incorporated into the final version.

Stage 2: Administering the Questionnaire

Participants. Participants (n = 93) were recruited through convenience sampling from the same two hospital sites as stage one. For stage two, any healthcare professional who had authority to initiate a referral process and who was working within a team which had access to paediatric psychology was eligible for recruitment. Participants who worked as part of a paediatric psychology team were excluded. Given limited research in this area, it was not possible to carry out *a priori* power analyses. However, Tabachnick and Fidell (2007, p123) give a formula for calculating sample size requirements for regression analyses: $n > 50 + 8m$ (where m is the number of predictor variables in a given analysis). Therefore 75 participants were required for analyses with three predictor variables ($50 + 8 (3) = 74$).

Procedure. All eligible participants (n = 868) were emailed as a group by the lead paediatric psychologists within the two hospital sites with an online link to the main questionnaire, which was accessed through Qualtrics online survey software. Eleven per cent of those contacted (n = 93) completed and returned the main questionnaire. Data from 26 participants who clicked on the link but did not begin the questionnaire were discarded from subsequent analysis. Following questionnaire completion, participants were asked to enter their email address if they were happy to be contacted to fill in the questionnaire for a second time. Twenty-eight participants agreed, all of whom were emailed with link to the same questionnaire four weeks later. Twelve participants (43 percent) returned the questionnaire within five weeks of completing the questionnaire for the first time.

Data Analysis. Data were downloaded to IBM SPSS Statistics Data Editor, Version 22 and screened for missing data, of which there were none.

Composite variables. Negatively worded variables were reversed and the following composite variables were computed from the questionnaire items; direct attitude, indirect attitude, direct subjective norm, indirect subjective norm, direct perceived behavioural control, indirect perceived behavioural control and intention.

Reliability. Internal consistency analyses were carried out for each of the direct composite variables; direct attitude, direct subjective norm, direct perceived behavioural control and intention. According to Francis et al. (2004), it is not appropriate to assess the internal consistency of indirect subscales as individuals often have both positive and negative beliefs about the same behaviour. It is appropriate to use test-retest reliability for this purpose. Test-retest reliability analyses were carried out for all indirect composite variables, but not direct measures as, unlike beliefs, attitude is changeable by nature and is therefore less likely to remain stable over a number of weeks. (Francis et al., 2004; Pallant 2007).

Validity. In order to test the concurrent validity of indirect composite measures, a series of bivariate correlations were carried out between direct and indirect measures of the same construct. It was planned that construct validity of the measure would be assessed by testing the following hypotheses based on Ajzen's (2002) Theory of Planned Behaviour model using a series of cross sectional linear regression analyses:

- Indirect measures will predict direct measures of the same construct
- Direct measures will predict healthcare professionals' intention to refer to paediatric psychology.

Specific beliefs influencing intention. In order to determine which specific beliefs had the greatest influence over intention to refer, specific beliefs were entered into a series of linear regressions, using intention as the outcome variable.

Statistical assumptions for linear regression analyses. Preliminary analyses and inspection of the data using histograms, box plots and the Kolmogorov-Smirnov, skewness and kurtosis values suggested that while the assumption of normality was met for four of the seven composite variables, it was not met for direct attitude, direct perceived behavioural control or intention. A square root transformation of the data normalised the intention distribution only. According to Field (2009), if some predictor variables are not normally distributed, it is appropriate to carry out a regression analysis as long as the outcome variable has a normal distribution. Therefore the transformed intention variable was used in all subsequent analyses. It was not possible to carry out linear regression using the direct measures as outcome variables for the indirect measures due to violation of the assumption of normality and inability to transform the data to create a normal distribution.

Further analysis of the data showed no violation of the assumptions of multicollinearity, linearity or homoscedasticity. The presence of outliers was assessed by examination of the residuals and Mahalanobis distances. These were acceptable for the most part, however, one case was removed for one analysis (holistic care predicting intention) due to a Mahalanobis distance which exceeded the critical value by >20 (Pallant, 2007).

Results

Stage 1: Qualitative Data

The data from the qualitative questionnaire were analysed using deductive content analysis. Within the three main categories of behavioural, normative and control beliefs, twelve sub-categories were identified. These sub-categories were ranked according to frequency of occurrence in the data (see Table 1). All data were coded and where data fitted into more than one category, it was coded within all relevant categories (see Appendix P for a fully coded transcript).

1. Behavioural beliefs. Many participants described the benefits and challenges of referring to paediatric psychology.

***1a. Holistic approach*.** The majority of participants explained that referring to paediatric psychology provided scope for 'information sharing' between disciplines and a 'more holistic approach to care'. The introduction of different perspectives was said to give a 'view of the child as a whole'.

"This allows clinicians to look at not only the disease itself but the effect on the individual as well as family members."

1b. Stigmatised views of psychology. Many participants also referred to the 'taboos and stigma' attached to a psychology referral in the context of a physical health service.

"May disengage the family… Some people are open to this idea but others may be worried that people think they are 'mad.'"

How a referral to psychology is perceived by families was also spoken about in relation to medically unexplained physical symptoms.

"Some families whose children have functional disorders, e.g. functional abdominal pain, struggle to accept that there is no organic cause for their child's symptoms and feel that they are not taken seriously if a psychologist is consulted."

1c. Type of service offered. Other participants spoke about the type of paediatric psychology service offered, including psychological 'model' used, level of therapist (e.g. 'trainees') and how this may differ from what they would view as helpful for a family they refer to paediatric psychology.

"Sometimes the service may not be offering the type of support that you are looking for."

1d. Family support. Many participants explained the importance of psychology for family 'emotional support' for various conditions.

"Involving a paediatric psychologist can help with child and family to cope with the illness."

"Help the patient/family live with physical symptoms which are medically unexplained despite medical assessment and investigation."

2. Normative beliefs. Groups whose views about paediatric psychology were described as significant by participants were identified; families, colleagues, budget holders and support groups/charities.

2a. Families. The vast majority of participants referred to the importance of families' views about a referral. Many described families 'crying out' for psychology, while others describe 'parental resistance' to a referral.

"Our patients greatly value [paediatric psychology]."

"Sometimes they don't understand the relevance."

"Most people are willing if it's presented as a way for supporting ordinary families under stress."

2b. Colleagues. The views and approval of colleagues in relation to paediatric psychology referrals was also described as significant, with some participants describing approval of 'all colleagues' while others described some colleagues who may not view paediatric psychology as helpful.

"Some physicians feel that no intervention will change a patient / family from their longstanding viewpoint or beliefs."

"Psychologically minded colleagues [would approve]."

2c. Budget holders. Several participants expressed the potential disapproval of referral to psychology from budget holders, due to 'cost' and 'financial resources'.

"Hospital managers can view it an expensive and under recognise the benefits"

"CCGs [Clinical Commissioning Groups], budget holders!"

2d. Support groups/charities. Some participants acknowledged the approval of support groups and charities in relation to referral to paediatric psychology

"Support groups appreciate the MDT approach with psychology input"

3. Control Beliefs. Participants described several barriers to referral as well as factors enabling them to refer to paediatric psychology.

3a. Waiting lists. 'Long waiting times' and lack of psychology availability due to 'limited resources' was the most frequently cited barrier to referral.

"More psychology time should be allocated to the service"

3b. Referral process. Some participants described being 'not sure of the referral process' while others explained 'pedantic inflexible referral pathways' as a barrier.

"I don't understand the need for a formal referral for psychology input. Patients are automatically proffered consultations with all other members of multi-disciplinary teams"

3c. Healthcare professionals' time. Several participants noted that it is difficult to 'commit the time' for joint meetings and that having 'another person to keep in the loop' can be 'time consuming'.

"Joint consultations are really excellent and I really enjoy them but they can be very time consuming"

3d. Cost and resources. Some participants commented on the 'lack of financial resources' for psychology.

"Their role is essential and I am worried about long term funding"

Table 1

Categories and Sub-categories from Content Analysis

Category	Sub-category	No. of participants within sub-category
Behavioural Beliefs		
1a	Holistic care	21
1b	Stigmatised views of psychology	21
1c	Type of service offered	17
1d	Family support	11
Normative Beliefs		
2a	Family (dis)approval	18
2b	Colleague/profession (dis)approval	14
2c	Budget holder/Managerial (dis)approval	5
2d	Support groups/charities (dis)approval	2
Control beliefs		
3a	Waiting lists and psychology availability	16
3b	Referral Process	12
3c	Healthcare Professional Time	9
3d	Cost/Resources	6

Three questions per category were required for the main questionnaire (Francis et al, 2004). The least frequently cited sub-category within each category was removed.

Stage 2: Quantitative Data

Participant Characteristics**.** Participants (n = 93) included consultant doctors, doctors, nurses, therapists, social workers and 'other occupations' consisting of play therapists and an audiologist. The majority of participants were female and ages ranged between 24 and 65 years. See Table 2 for details of participant characteristics.

Table 2

Participant Characteristics (n = 93)

Demographic	Category	Frequency (%)
Professional Role	Consultant Doctor	30 (32%)
	Doctor	11 (12%)
	Nurse	25 (27%)
	Therapist (Occupational, Physio)	22 (24%)
	Social Worker	2 (2%)
	Other	3 (3%)
Gender	Female	70 (75%)
	Male	21 (23%)
	Prefer not to state	2 (2%)
Age range	24-35 years	23 (25%)
	36-45 years	34 (37%)
	46-55 years	25 (27%)
	56-65 years	11 (12%)
Number of years qualified	Under 5 years	2 (2%)
	5-10 years	17 (18%)
	11-20 years	35 (38%)
	Over 20 years	39 (42%)

Reliability. Reliability was assessed using internal consistency for direct measures and test-re-test reliability for indirect measures.

Internal consistency. Internal consistency was assessed using Cronbach's Alpha and mean inter-item correlation values. Each subscale was made up of three items. Where scales have fewer than 10 items, it is common to find low Cronbach's alpha values (< 0.7). Direct subjective norm was one such case. In these cases, it may be more appropriate to report mean inter-item correlation (Briggs & Cheek, 1986). All subscales demonstrated acceptable reliability using these conventions (see Table 3).

Table 3

Subscale Internal Consistency: Direct Measures (n = 93)

Composite variable (Subscale)	No. of items	Cronbach's Alpha*	Mean inter-item correlation**
Direct Attitude	3	.74	n/a
Direct Subjective Norm	3	.47	.21
Direct Perceived Behavioural Control	3	.70	n/a
Intention	3	.75	n/a

* Cronbach's alpha value of .7 or above indicates an acceptable level of reliability (Devellis (2003)

** Mean inter-item correlation values between .2 and .4 indicate acceptable reliability (Briggs & Cheek, 1986).

Test-retest reliability. Twelve participants completed the questionnaire for a second time. For indirect composite variables, temporal stability was assessed using Pearson's *r*. Cohen (1988) suggests the following conventions to measure the strength of the relationship between two variables; small (*r* = 0.1 to .29), medium (*r* = 0.3 - .49), large (*r* = 0.5 - 1.0). Each of the subscales at Time 1 demonstrated a medium or large correlation with the same

subscale at Time 2 (see Table 4). However, perhaps due to the small sample size (n = 12), only indirect perceived behavioural control reached statistical significance. The significance of Pearson's *r* is strongly influenced by sample size (Pallant, 2007).

Table 4

Subscale Test-Retest Reliability: Indirect Measures (n = 12)

Composite variable (Subscale)	Pearson's r
Indirect Attitude	.39
Indirect Subjective Norm	.52
Indirect Perceived Behavioural Control	.79*

* Correlation is significant at the .05 level (two tailed)

**Correlation is significant at the .01 level (two tailed)

Validity. Validity was assessed using concurrent validity for the indirect measures and construct validity for the direct measures.

Concurrent validity. According to Theory of Planned Behaviour, if indirect measures are valid, they should correlate with direct measures of the same construct (Francis et al., 2004). Again, conventions recommended by Cohen (1988) were used to measure the relationship between the direct and indirect subscales. All direct subscales were significantly and positively correlated with indirect subscales of the same construct (see Table 5).

Table 5

Correlation Coefficient: The Relationship Between Direct And Indirect Subscales Of The Same Construct (n = 93)

Direct-Indirect Subscale	Pearson's r
Attitude	0.23*
Subjective Norm	0.41**
Perceived Behavioural Control	0.32**

* Correlation is significant at the .05 level (two tailed)

**Correlation is significant at the .01 level (two tailed)

Construct validity. Three cross-sectional linear regression analyses were used to assess the ability of direct measures to predict intention to refer to paediatric psychology. Direct attitude significantly predicted intention, accounting for 7 percent of the variance, F (1, 91) = 6.91, p = 0.01. Direct subjective norm significantly predicted intention, accounting for 22 percent of the variance, F (1, 91) = 26.27, $p < 0.0001$. Finally, direct perceived behavioural control significantly predicted intention, accounting for 7 percent of the variance, F (1, 91) = 6.31, p = 0.01.

The standardised beta values represent the number of standard deviations that scores in the outcome variable (intention) would change if there was a one standard deviation unit change in the predictor variable.

Table 6

Individual Linear Regression Analyses: Direct Measures Predicting Intention (n = 93)

Predictor	R^2	β	Mean	Standard Deviation
Direct Attitude	.07*	- .27*	6.57	.59
Direct Subjective Norm	.22**	-.47**	5.14	.96
Direct Perceived Behavioural Control	.07*	- .26*	5.87	1.16

* Significant at the .05 level

**Significant at the .01 level

A cross-sectional multiple linear regression analysis was carried out in order to assess how much shared and individual variance in the outcome could be explained by the direct measures. Direct attitude, subjective norm and perceived behavioural control were entered as predictor variables and intention was the outcome variable. Together, direct measures significantly predicted intention to refer to paediatric psychology, accounting for 24 percent of the variance, F (3, 89) = 9.4, $p < 0.001$. When the contribution shared by the predictor variables was removed, only direct subjective norm made a significant unique contribution to the outcome, uniquely accounting for 14 percent of the variance in intention to refer to paediatric psychology ($p < 0.0001$; see Table 7).

Table 7

Multiple Linear Regression: Direct Measures Predicting Intention (n = 93)

Predictors		R^2	β
All Direct Measures		.24**	n/a
	Direct Attitude	.012	-.12
	Direct Subjective Norm	.14**	-.42**
	Direct Perceived Control	.002	-.05

* Significant at the .05 level

**Significant at the .01 level

Specific beliefs influencing intention. Three cross-sectional multiple linear regression analyses were carried out in order to assess which beliefs influenced intention to refer to paediatric psychology. The three behavioural beliefs were entered as predictor variables; intention was the outcome variable. Together, behavioural beliefs did not significantly predict intention to refer to paediatric psychology, accounting for 6.7 percent of the variance, F (3, 88) = 2.10, p = 0.106. On inspection of individual behavioural beliefs, only beliefs about holistic care made a significant contribution to the outcome, uniquely accounting for 6.5 percent of the variance in intention (p = .015).

Table 8

Multiple Linear Regression: Behavioural Beliefs Predicting Intention (n = 92)

Predictors		R^2	β	Mean	Standard Deviation
All		.067	n/a		
Behavioural	Behavioural Belief 1 (Holistic)	.065*	-.26*	16.43	3.72
Beliefs	Behavioural Belief 2 (Stigma)	.0009	-.030	-.47	7.6
	Behavioural Belief 3 (Service Type)	.004	.064	-.02	3.06

* Significant at the .05 level

**Significant at the .01 level

The three normative beliefs were entered as predictor variables and intention was the outcome variable. Together, normative beliefs significantly predicted intention to refer to paediatric psychology, accounting for 14 percent of the variance, F (3, 89) = 4.73, p = 0.004. On inspection of individual beliefs, only beliefs about families made a significant contribution to the outcome, uniquely accounting for 4 percent of the variance in intention (p = 0.015), although beliefs about colleagues approached significance (p = 0.067), uniquely accounting for 3 percent of the variance in intention.

Table 9

Multiple Linear Regression: Normative Beliefs Predicting Intention (n = 93)

Predictors		R^2	β	Mean	Standard Deviation
All		.14**	n/a		
Normative	Normative Belief 1 (Budget Holders)	.012	-.11	-.89	5.40
Beliefs	Normative Belief 2 (Families)	.04*	-.23*	6.9	7.35
	Normative Belief 3 (Colleagues)	.03	-.19	10.12	6.9

* Significant at the .05 level

**Significant at the .01 level

The three control beliefs were entered as predictor variables and intention was the outcome variable. Together, control beliefs did not significantly predict intention to refer, accounting for 1.1 percent of the variance, F (3, 89) = 0.34, p = 0.80. Additionally, none of the individual control beliefs made a significant contribution to the outcome.

Table 10

Multiple Linear Regression: Control Beliefs Predicting Intention (n = 93)

Predictors		R^2	β	Mean	Standard Deviation
All		.011	n/a		
Control	Control Belief 1 (HCP Time)	.002	-.044	-.32	5.04
Beliefs	Control Belief 2 (Referral Process)	.01	.12	-2.2	6.7
	Control Belief 3 (Waitlist)	.00005	.008	-1.27	8.41

* Significant at the .05 level

**Significant at the .01 level

Discussion

This study had the following aims: to assess the feasibility, reliability and validity of a questionnaire designed to measure healthcare professionals' beliefs and attitudes about referring children and families to paediatric psychology, to identify healthcare professionals' beliefs related to referral to paediatric psychology and lastly, to identify those beliefs significantly influencing intention to refer children and families to paediatric psychology.

The data support the feasibility of this questionnaire as a tool to measure healthcare professionals' beliefs and attitudes towards referring children and families to paediatric psychology. The questionnaire was completed by 11% of individuals who were given access. Although this percentage is low compared to reported response rates from email-based surveys (36 percent; Sheehan, 2001), this is unsurprising given that healthcare professionals have significantly lower questionnaire response rates than the general population, due to demanding work schedules and frequently being approached by researchers (Flanigan et al., 2008).

Additionally, more females than males completed the questionnaire. Again, this was not unexpected; female healthcare professionals have consistently been reported as more likely to fill in questionnaires than their male counterparts (e.g. Cull, O'Connor, Sharp, & Tang, 2005). In spite of this, response bias appears to be less of an issue within healthcare populations than in the general population, with responders and late responders (a standard proxy for non-responders used in survey research, Kellerman & Herold, 2001) not differing significantly in their responses (Flanigan et al., 2008). Returned questionnaires had no missing data, suggesting a willingness to answer questions relating to referral behaviour.

The questionnaire demonstrated good reliability. Internal consistency within all direct subscales was acceptable. Test-retest reliability was also acceptable; all indirect (belief-

based) subscales at Time 1 illustrated a medium or large correlation with the same subscale at Time 2.

Validity of the measure was illustrated. The indirect and direct measures showed concurrent validity; all direct subscales were significantly and positively correlated with indirect subscales of the same construct. Construct validity was also demonstrated. The direct measures significantly predicted healthcare professionals' intention to refer to paediatric psychology. When shared variance was taken out of the equation, only direct subjective norm made a significant unique contribution to intention to refer to paediatric psychology, indicating that others' beliefs and behaviours are the strongest predictor of healthcare professionals' intention to refer.

Green et al., (2008) also found that much of the variance in general practitioner (GP) referral rates to specialist eating disorder services was based on GP subjective norms, however, attitude was also a unique predictor of referral in their study. This may be because, according to Godin et al. (2008), the efficacy of Theory Of Planned Behaviour's predictive value can vary according to context, behaviour being measured and characteristics of the population performing the behaviour. Therefore, it was important to carry out a more in depth exploration of beliefs underlying the main Theory of Planned Behaviour constructs and their relationship with healthcare professional intention to refer to paediatric psychology.

In this study, decision making about referral to paediatric psychology was shaped by a wide range of beliefs, demonstrated through qualitative analysis. A further aim of the study was to illustrate which of these identified beliefs predicted intention to refer. This is recommended by Francis et al. (2004) to illustrate which beliefs to target in the context of an intervention designed to modify the behaviour. Only normative beliefs significantly predicted intention to refer. With regard to individual normative beliefs, beliefs about families' approval made a significant unique contribution to intention while the unique contribution of

beliefs about colleagues' referral behaviour approached statistical significance. Behavioural beliefs about the benefits of holistic care were also found to make a statistically significant contribution to intention. Beliefs significantly predicting intention are discussed under the relevant subheadings below, along with the other qualitatively identified beliefs about referral to paediatric psychology.

Behavioural Beliefs

In relation to behavioural beliefs, the type of psychology service received by the child and family post-referral, and opportunity for family support were factors relating to decision to refer. Indeed, Briggs-Gowan et al., (2000) found that in a paediatric outpatient setting, doctors' decision to refer was based on their own perception of the child's need for mental health services. Mental health stigma was frequently cited in the qualitative data as a disadvantage to referral, reflecting the findings of qualitative studies about referral to mental health services (Kainz, 2002; Stavrou, Cape & Barker, 2009).

Beliefs about the advantages of holistic care was the only behavioural belief significantly predicting intention to refer to paediatric psychology. The benefits of holistic care such as increased job satisfaction and improved outcomes have been consistently demonstrated for both healthcare professionals and patients within a variety of settings (Zwarenstein, et al., 2009; Boone, Minore, Katt, & Kinch, 1997). According to van Knippenberg et al. (2004), advantages for inter-professional teams are established through the joining up of knowledge which was previously separate. This is known as 'cognitive heterogeneity' (Mitchell, 2009. p7) and permits the construction of solutions to problems which are more innovative and inclusive (Ancona & Caldwell, 1992; DeDreu & West, 2001). According to van Knippenberg et al. (2004) cognitive heterogeneity mediates the relationship between team diversity and team effectiveness. This fits with the results of the current study, where beliefs about holistic care were shown to predict intention to refer to paediatric

psychology, indicating that where holistic care was seen as useful, a referral to paediatric psychology was more likely.

Normative Beliefs

With regard to normative beliefs, the approval of families, colleagues, budget holders and third sector organisations were cited in the qualitative data. Together, normative beliefs significantly predicted intention to refer. While research shows that cost is a large factor in U.S studies of mental health referral (e.g. Beacham et al., 2012; Kainz et al, 2002), perhaps due to the UK context of the study, where the finance burden lies with budget holders as opposed to families, this was not a uniquely significant predictor of intention to refer. Healthcare professionals' perception of their colleagues' (others within their profession) referral behaviour accounted for three percent of the variance in intention to refer and the unique contribution of this belief approached statistical significance. Similarly, Green (2008) found that GPs' referral to mental health services was related to their perception of other GPs' mental health referral behaviour. Indeed according to the model of similarity-attraction, members of the in-group trust and mirror each other's actions (Tajfel, 1982; Williams & O'Reilly, 1998).

Healthcare professionals' intention to refer was also predicted by approval of the referral by families. Research shows that patient interest in, request for and approval of referral are important factors in healthcare professionals' decision to refer to mental health services (Stavrou et al., 2009; Nandy et al., 2001). Encouragingly, this finding shows the growing role of patient involvement in decision making about their use of mental health services (Stavrou et al., 2009). On the other hand, examination of the qualitative comments relating to families' approval may indicate that the referrers' own perception of families' positions in relation to psychology and mental health may also play a role in referral decision. Several referrers described families as more or less 'psychologically minded' than others, and

some referred to families being 'put off' due to lack of understanding of what psychology could offer. It is therefore possible that beliefs about patient resistance to referral influences referral decisions. According to Kainz (2002), a healthcare professional's belief that patients often resist a psychology referral can be easily conveyed to the patient, potentially influencing referral discussions.

Control Beliefs

Practical constraints including healthcare professional availability, cost, waiting lists and long referral processes were cited within the qualitative data as control beliefs relating to referral. However, none of these control beliefs were found to influence intention to refer, indicating that although there are perceived barriers over which healthcare professionals have variable control, these do not influence referral decisions. Similarly, Green et al., (2008) found that control beliefs did not appear to influence decision to refer to mental health services. In the current study, perceived behavioural control was high, suggesting that healthcare professionals feel able to refer to psychology. This may be at odds with services in other countries, where due to insurance, patients have to initiate contact with psychology services following a referral (Kainz, 2002). Since the UK National Health Service offers the same service to all, healthcare professionals may feel that both they and families are more empowered to make or avail of psychology referrals.

Limitations and Implications

These results should be considered in light of several limitations. Firstly, since this is an exploratory study with no established effect sizes, it was not possible to perform *a priori* power calculations for sample size. Instead, a formula for calculating sample size requirements for regression analyses was utilised (Tabachnick & Fidell, 2007). According to this formula, the study sample size was sufficient to carry out linear regression analyses using

several predictor variables. However, previous studies have also found links between referral behaviour and referrer characteristics such as gender (e.g. Hugo et al., 2000), and therefore it may have been beneficial to adjust for these characteristics within the main analyses. In order to adjust for variables, a larger sample size is needed ($N \geq 104 + m$), particularly if the number of cases within specific categories are low (Green, 1991). Therefore, due to this study's limited sample size ($n = 93$) and low numbers of cases within demographic categories due to an uneven gender split (23% male), it was not possible to adjust for demographic variables. Future research would benefit from larger samples to test whether referrer characteristics as well as attitudes and beliefs play a role in referral.

Secondly, Cronbach's Alpha was low for the direct subjective norm subscale, perhaps due to the low number of subscale items (three). Therefore, inter-item correlation was used to demonstrate internal consistency (Briggs & Cheek, 1986). However, it may have been prudent to remove items lowering Cronbach's Alpha, as a low Cronbach's Alpha may indicate low internal consistency of the subscale. Unfortunately this was not possible owing to the small number of items within the subscale. Future research should consider including more items in the scale to begin with, or using one item to represent direct subjective norm.

Thirdly, for those beliefs which significantly predicted intention to refer, it is unknown whether the size of the effect (approximately 1-2 scale points for each of the beliefs significantly predicting intention to refer) is clinically significant. That is, whether changes in these beliefs would impact actual referral rates. Future research with an estimate of actual referral behaviour (rather than intention only) may be able to better interpret the clinical relevance of the effect sizes, and therefore whether it would be worthwhile to intervene on these beliefs.

Fourthly, referral is one means of integrating psychology into paediatric health settings. Future research should explore other means of integration such as psychological consultation and the effect of including psychologists in MDT meetings.

Additionally, for practical reasons, the eligible population for stage one and stage two varied. Stage one participants were consultant medical doctors while stage two were any healthcare professional. For stage one, participant demographic data were not collected. It is unknown whether the stage one sample were representative of the referrer population, or if the beliefs of stage one participants may have differed from the beliefs of other healthcare professionals. For future studies, eliciting beliefs from a wide range of healthcare professionals, which is representative of the population of referrers, will be important.

Furthermore, the sample for stage two of the current study may have over-represented the views of certain groups, namely females and nurses. Unfortunately, it was not possible to map participant demographic characteristics to the population of referrers as a whole. Future research should aim to collect data from a sample that is demographically representative of the population of referrers.

Finally, while research shows that the Theory of Planned Behaviour predicts behavioural intention to an extent (Godin et al., 2008), the theory has been heavily criticised for a lack of validity, utility and a failure to consider other factors which may influence behaviour, for example, unconscious processes and self-regulation (Sheeran, Gollwitzer, & Bargh, 2014; Sniehotta, Presseau & Araújo-Soares, 2014). Indeed, other processes which have the potential to impact behaviour are not considered in the Francis (2004) manual used to create the questionnaire for the current study. Therefore, perhaps unsurprisingly, in the current study, the theory of planned behaviour constructs accounted for 24 percent of the variance in intention to refer, meaning that a large percentage of variance was unexplained. According to Sniehotta et al. (2014) it is therefore necessary to replace Theory of Planned

Behaviour with a new, wider theoretical approach. However, presently, there does not appear to be a viable alternative that is superior in in its ability to predict behaviour (Ajzen, 2015).

With these caveats in mind, some inferences can be made in relation to clinical intervention. Findings from the current study suggest that those healthcare professionals who value information sharing with psychology are more likely to intend to refer to paediatric psychology. Indeed, according to Mitchell et al. (2009, p. 7) successful 'cognitive heterogeneity' can increase integration of individuals from different healthcare professions, whereas misunderstanding between professions can lead to conflict between MDT members.

In a Cochrane review, Reeves et al. (2013) found that multi-disciplinary education (where healthcare professionals learn from professionals other of healthcare disciplines) can increase information sharing between disciplines and integration of different healthcare professions. Research has demonstrated that healthcare professionals would welcome more input and teaching from psychologists, to increase biopsychosocial perspectives (Grenier, Chomienne, Gaboury, Ritchie, & Hogg, 2008; Douglas & Benson, 2015). Psychology led MDT workshops which focus on the biopsychosocial model of healthcare (Engel, 1977) may be useful in order to increase cognitive heterogeneity, information-sharing and thus intention to refer to paediatric psychology.

Such workshops could include systemic models of discussion, which have been shown to increase information sharing within professional systems (Wynne, McDaniel & Weber, 1986). Douglas and Benson (2015) interviewed healthcare professional about psychology led meetings within a paediatric setting. Participants reported that these meetings helped to maintain focus on holistic care and improved their likelihood of identifying child and family psychological issues.

Additionally, findings from the current study suggest that healthcare professionals' intention to refer is based on their beliefs about families' ideas and knowledge in relation to

psychology. Psychology led workshops could focus on different ways in which healthcare professionals could engage families to think about psychology referrals. Workshops could also focus on other beliefs identified within this study, such as stigma or medically unexplained symptoms, depending on service context. Furthermore, although the healthcare professional is significant in the integration of physical and mental health, the need for a top down, public-policy led approach, for example, a government initiative, which emphasises the importance of psychological support for young people in paediatric settings is also necessary. This may influence higher rates of self-referral amongst young people and families.

Conclusions

Although integration of physical and mental healthcare by means of referral to psychological services is essential in order to maximise physical and mental health outcomes in the current UK health service, there are few studies investigating factors influencing referral behaviour. This is particularly important within the context of paediatrics, where research has illustrated the importance of integrating psychology and health. Therefore, this study is exploratory and theory-driven in assessing the factors influencing healthcare professionals' referral of children and families to paediatric psychology services.

Findings from this study suggest that the main constructs of Theory of Planned Behaviour are useful in predicting intention to refer to paediatric psychology, and that subjective norm (perceptions of others' views and behaviours) was the strongest predictor. Specific beliefs about referral were also shown to influence intention to refer. Findings that individual factors such as attitudes and beliefs influence referral to paediatric psychology suggest the need for MDT interventions and inter-professional education relating to the psychological aspects of illness. Future research would benefit from measuring actual rates of referral as well as intention to refer to paediatric psychology services.

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Appendix A: Search Terms by Database

Medline via Ovid search strategy

1. exp somatoform Disorders/ or exp symptoms/ or exp physical disorders/ or exp somatization/ or exp somatisation/ or medically unexplained.mp.
2. unexplained symptoms.mp.
3. exp Illness behaviour/ or exp illness behavior/ or unexplained physical symptoms.mp.
4. somat$.mp.
5. psychosomat$.mp.
6. psychogenic.mp.
7. functional illness.mp.
8. exp pain/
9. exp chronic pain/ or exp somatoform pain disorder/
10. functional pain.mp.
11. unexplained pain.mp.
12. exp Headache/
13. exp fibromyalgia/
14. exp back pain/
15. exp irritable bowel syndrome/
16. (non-ulcer dyspepsia or nonulcer dyspepsia).mp.
17. functional dyspepsia.mp.
18. Belly pain.mp.
19. stomach pain.mp.
20. Vomiting.mp. or exp Vomiting/
21. exp Diarrhea/ or diarrhoea.mp.
22. Bloating.mp.
23. swelling of the stomach.mp.
24. exp fatigue/ or exp chronic fatigue syndrome/ or exp chronic fatigue/
25. myalgic encephalomyelitis.mp.
26. pseudoneurological.mp.
27. exp conversion disorder/
28. (nonepileptic or non-epileptic).mp.
29. (pseudoseizure or pseudo-seizure).mp.
30. Globus Syndrome.mp.
31. exp blind$/ or blurred vision.mp. or double vision.mp.
32. (paralysis or weakness).mp.
33. deaf$.mp.
34. exp amnesia/
35. aphonia.mp.
36. Fainting.mp. or exp Syncope/
37. premenstrual syndrome.mp. or exp Premenstrual Syndrome/
38. chronic pelvic pain.mp.
39. exp Headache/ or exp Migraine Headache/ or migraine.mp.

40. hyperventilation syndrome.mp.
41. Postural tachycardia syndrome.mp.
42. Postural orthostatic tachycardia syndrome.mp.
43. multiple chemical sensitivity.mp.
44. Environmental illness.mp.
45. sick building syndrome.mp.
46. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45
47. exp Group Psychotherapy/ or exp Psychotherapy/ or psychotherap*.mp.
48. cognitive therap*.mp. or exp Cognitive Behavior Therapy/
49. cognitive behavior therap*.mp. or exp Cognitive Behavior Therapy/
50. ((CBT or psychol* or behavio*) adj (therap* or treat* or modif*)).mp.
51. (group adj (treat* or therap*)).mp.
52. ((dynamic or psychodynamic or analytic or psychoanalytic) adj (treat* or therap*)).m
53. ((counsel* or psycho-ed* or family) adj (therap* or treat* or intervene*)).mp.
54. (system* adj (therap* or treat*)).mp.
55. 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54
56. exp anxi$/ or exp depress*/ or exp conduct disorder$/ or exp panic$/ or exp obsess$/ or exp OCD/
57. (anx* or depress* or conduct disorder* or panic* or obsess* or OCD).mp.
58. 56 or 57
59. 55 and 58
60. 46 and 59
61. limit 60 to peer reviewed journal
62. limit 61 to (childhood or adolescence <13 to 17 years>)
63. exp child/
64. Infant/
65. Adolescent/
66. (child$ or adolescen$ or infant$ or juvenil$ or pediatric$ or paediatric$).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
67. ('young person$' or 'young people' or youth$ or 'young adult$').mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
68. 63 or 64 or 65 or 66 or 67
69. 60 and 68

PsycINFO via OVID

1. exp somatoform Disorders/ or exp symptoms/ or exp physical dis
orders/ or exp somatization/ or exp somatisation/ or medically

unexplained.mp.
2. unexplained symptoms.mp.
3. exp Illness behaviour/ or exp illness behavior/ or unexp
lained physical symptoms.mp.
4. somat$.mp.
5. psychosomat$.mp.
6. psychogenic.mp.
7. functional illness.mp.
8. exp pain/
9. exp chronic pain/ or exp somatoform pain disorder/
10. functional pain.mp.
11. unexplained pain.mp.
12. exp Headache/
13. exp fibromyalgia/
14. exp back pain/
15. exp irritable bowel syndrome/
16. (non-ulcer dyspepsia or nonulcer dyspepsia).mp.
17. functional dyspepsia.mp.
18. Belly pain.mp.
19. stomach pain.mp.
20. Vomiting.mp. or exp Vomiting/
21. exp Diarrhea/ or diarrhoea.mp.
22. Bloating.mp.
23. swelling of the stomach.mp.
24. exp fatigue/ or exp chronic fatigue syndrome/ or exp chroni
c fatigue/
25. myalgic encephalomyelitis.mp.
26. pseudoneurological.mp.
27. exp conversion disorder/
28. (nonepileptic or non-epileptic).mp.
29. (pseudoseizure or pseudo-seizure).mp.
30. Globus Syndrome.mp.
31. exp blind$/ or blurred vision.mp. or double vision.mp.
32. (paralysis or weakness).mp.
33. deaf$.mp.
34. exp amnesia/
35. aphonia.mp.
36. Fainting.mp. or exp Syncope/
37. premenstrual syndrome.mp. or exp Premenstrual Syndrome/
38. chronic pelvic pain.mp.
39. exp Headache/ or exp Migraine Headache/ or migraine.mp.
40. hyperventilation syndrome.mp.
41. Postural tachycardia syndrome.mp.

42. Postural orthostatic tachycardia syndrome.mp.
43. multiple chemical sensitivity.mp.
44. Environmental illness.mp.
45. sick building syndrome.mp.
46. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45
47. exp Group Psychotherapy/ or exp Psychotherapy/ or psych otherap*.mp.
48. cognitive therap*.mp. or exp Cognitive Behavior Therapy/
49. cognitive behavior therap*.mp. or exp Cognitive Behavior T herapy/
50. ((CBT or psychol* or behavio*) adj (therap* or treat* or mod if*)).mp.
51. (group adj (treat* or therap*)).mp.
52. ((dynamic or psychodynamic or analytic or psychoanalytic) adj (treat* or therap*)).mp.
53. ((counsel* or psycho-ed* or family) adj (therap* or treat * or intervene*)).mp.
54. (system* adj (therap* or treat*)).mp.
55. 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54
56. exp anxi$/ or exp depress*/ or exp conduct disorder$/ or e xp panic$/ or exp obsess$/ or exp OCD/
57. (anx* or depress* or conduct disorder* or panic* or obses s* or OCD).mp.
58. 56 or 57
59. 55 and 58
60. 46 and 59
61. limit 60 to peer reviewed journal
62. limit 61 to (childhood or adolescence <13 to 17 years>)
63. limit 61 to (100 childhood or 120 neonatal or 140 infancy <2 to 23 mo> or 160 preschool age or 180 school age or 200 adolescence or 320 young adulthood)
64. limit 24 to (childhood or adolescence <13 to 17 years>)
65. limit 64 to peer reviewed journal
66. limit 55 to ((childhood or adolescence <13 to 17 years>) a nd "0110 peer-reviewed journal")
67. exp Intervention/
68. 55 or 67
69. 46 and 68
70. limit 69 to (childhood or adolescence <13 to 17 years>)

71. limit 70 to peer reviewed journal

Cumulative Index to Nursing and Allied Health Literature (CINAHL)

1. (MH "Somatoform Disorders+") OR "somatoform disorders" OR (MH "Psychophysiologic Disorders+")
2. (MH "Medically Unexplained Symptoms") OR "medically unexplained"
3. (MH "Factitious Disorders")
4. "somatization"
5. "unexplained symptoms"
6. "Illness behavior"
7. "Illness behaviour"
8. "functional illness"
9. "functional symptoms"
10. (MH "Pain+") OR (MH "Chronic Pain")
11. (MH "Headache+") OR "Headache" OR (MH "Tension Headache")
12. (MH "Fibromyalgia") OR (MH "Fatigue Syndrome, Chronic")
13. (MH "Irritable Bowel Syndrome")
14. (MH "Dyspepsia") OR "functional dyspepsia"
15. (MH "Abdominal Pain+")
16. "myalgic encephalomyelitis"
17. "pseudoneurological"
18. "conversion disorder"
19. "nonepileptic seizures"
20. "non-epileptic seizures"
21. "pseudoseizure"
22. "Globus Syndrome"
23. (MH "Muscle Weakness")
24. (MH "Syncope+") OR "Fainting"
25. (MH "Premenstrual Syndrome+") OR (MH "Premenstrual Dysphoric Disorder")
26. (MH "Pelvic Pain") OR "chronic pelvic pain"
27. (MH "Pelvic Pain") OR "chronic pelvic pain"
28. (MH "Migraine") OR (MH "Tension Headache")
29. (MH "Postural Orthostatic Tachycardia Syndrome")
30. (MH "Multiple Chemical Sensitivity")
31. (MH "Environmental Illness")
32. (MH "Sick Building Syndrome")
33. S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12 OR S13 OR S14 OR S15 OR S16 OR S17 OR S18 OR S19 OR S20 OR S21 OR S22 OR S23 OR S24 OR S25 OR S26 OR S27 OR S28 OR S29 OR S30 OR S31 OR S32
34. (MH "Psychotherapy+") OR "psychotherapy" OR (MH "Psychotherapy, Brief") OR (MH "Psychotherapy, Psychodynamic") OR (MH "Psychotherapy, Group") OR (MH "Cognitive Therapy")

35. "cognitive behavior* therapy"
36. ""cognitive behaviour* therapy""
37. (MH "Behavior Therapy")
38. (MH "Behavior Therapy+")
39. "Behavior* Therapy"
40. "Behaviour* Therapy"
41. (MH "Counseling+") OR "counselling"
42. (MH "Family Therapy")
43. (MH "Behavior Modification+")
44. "psychoanalytic therapy"
45. S34 OR S35 OR S36 OR S37 OR S38 OR S39 OR S40 OR S41 OR S42 OR S43 OR S44
46. (MH "Anxiety+")
47. "anx*" OR (MH "Anxiety Disorders+")
48. (MH "Depression+") OR "depress*"
49. "Panic*"
50. (MH "Panic Disorder")
51. (MH "Child Behavior Disorders+") OR "conduct disorder"
52. (MH "Obsessive-Compulsive Disorder+")
53. (S46 OR S47 OR S48 OR S49 OR S50 OR S51 OR S52
54. (S46 OR S47 OR S48 OR S49 OR S50 OR S51 OR S52) AND (S33 AND S45 AND S53)
55. (S46 OR S47 OR S48 OR S49 OR S50 OR S51 OR S52) AND (S33 AND S45 AND S53)
56. (MH "Child")
57. "child*"
58. (MH "Adolescence") OR "adolesce*"
59. "pediatric"
60. "paediatric"
61. (MH "Infant")
62. "juvenile"
63. (MH "Young Adult") OR "young person"
64. "youth"
65. S56 OR S57 OR S58 OR S59 OR S60 OR S61 OR S62 OR S63 OR S64
66. (S56 OR S57 OR S58 OR S59 OR S60 OR S61 OR S62 OR S63 OR S64) AND (S54 AND S65)
67. (S56 OR S57 OR S58 OR S59 OR S60 OR S61 OR S62 OR S63 OR S64) AND (S54 AND S65)
68. MM "Treatment Outcomes"
69. (S56 OR S57 OR S58 OR S59 OR S60 OR S61 OR S62 OR S63 OR S64) AND (S54 AND S65)

Appendix B: Data Extraction Checklist (Higgins & Green, 2008)

Source
- Study ID (created by review author).
- Citation

Eligibility
- Confirm eligibility for review.
- Reason for exclusion.

Methods
- Study design.
- Total study duration.

Participants
- Total number.
- Age.
- Sex.
- Country.

Interventions
- Total number of intervention groups.

For each intervention and comparison group of interest:
- Specific intervention.
- Intervention details (sufficient for replication, if feasible).
- Integrity of intervention.

Outcomes
- Outcomes and time points

Results
- Summary data for each intervention group

Appendix C: Table of Characteristics of Included Studies

	Study	Physical Symptom	Therapists delivering interventions	Study Type	Control group: controlling for (time, attention, therapist time)	Primary Outcome Measure(s) *(Mental health measures boded and italicised)*	Secondary Outcome Measure (s) *(Mental health measures boded and italicised)*	n (% female)	Participant Age (M; SD)	Outcome Time points	Country
1	Bussone et al. (1988)	Tension-type headache	NR	RCT 2 arms	Relaxation placebo (Y,Y,Y)	1.Headache diary (PTI)	***1. STAIC***	30 (50%)	Treat (M=11.1, SD=2.6) Ctrl (M=13, SD=1.5)	Baseline 1month 3month 6month 12month	USA
2	Chalder, Deary, Husain & Walwyn, 2010 ***2012 Follow up study included**	Chronic Fatigue Syndrome	Trained Cognitive behavioural therapists	RCT 2 arms	Psycho-education (N,Y,N)	1. School attendance	1.Chalder fatigue scale 2. SF-36 (physical functioning subscale) 3. WSAS ***4. SDQ*** 5. GOS	63 (68%)	11-18	Baseline 6 month 24 month for ***SDQ*** Baseline 3month 6month 12month for primary outcomes.	UK
3	Griffiths et al. (1996)	Chronic Headache	Postgraduate clinical psychology student in her final year of studies	RCT 3 arms	Self-monitoring (N, N, N)	1. Headache diary	***1.CMAS; 2. CDS*** 3. SEQ-C 4. Expectancy for change 5. CHAS	32 (50%)	Clinic 11.4 (0.58) Home 11.5 (0.58) WL 11.1 (0.58)	Baseline Post-treatment 9 week groups 1 and 2, not for SM group	Australia

4	Hickman et al. 2015	Chronic Daily Headache	Nurse Practitioner with cognitive behavioural training	RCT 2 arms	Headache Education (YYY)	***1. BYI-II*** 2. Healthy Lifestyle Beliefs Scale ***3. PSS*** 4. PedMIDAS 5. PPPI		32 (72%)	13-17 (M=15.09; SD=1.1)	Baseline Post Intervention	USA
5	Kashikar-Zuck et al. (2005)	Juvenile Primary Fibromyalgia	Doctoral level psychologists trained in the intervention	RCT 2 arms	Self-monitoring (N,N,N)	1.FDI 2.Pain VAS ***3.CDI***	1. PCQ 2. Tender point examination	30 (100%)	13-17 (M=15.83, SD=1.26	Baseline Post-treatment Post-crossover of treatment	USA
6	Kashikar-Zuck, Ting, Arnold, Bean, Powers, Graham, et al. (2012)	Juvenile Fibromyalgia	Doctoral level psychologists in the intervention	RCT 2 arms	FM Education (YYY)	1. FDI	1. Pain Severity (VAS) ***2. CDI*** 3. Tender point exam 4. PedsQL 5. Sleep quality VAS	114 (92%)	11-18 (M=15; SD=1.8)	Baseline Post-treatment 6 month	USA
7	Levy et al (2010)	Functional Abdominal Pain	Trained therapists (Master's degree)	RCT 2 arms	Educational Intervention (Y,Y,Y)	1. FPS-R 2. CSI 3. FDI	***1. CDI*** ***2. MASC*** 3. ARCS (Protectiveness)	200 (94%)	7-17 (M=NR; SD=2.5)	Baseline 1 month 3 month 6 month	USA
8	McGrath et al. (1992)	Migraine	'Trained therapist'	RCT 3 arms	CBT (Y,Y,Y) Assessment & monitoring (Y,Y,Y)	Total Headache Index	***Poznanski Depression Scale (1979)***	87 (72%)	11-18 (M=NR; SD=1.75)	Baseline 1 month 3 month 12 month	Canada

9	Palermo et al. (2009)	Chronic Pain	postdoctoral psychologist with CBT experience	RCT 2 arms	Wait list with medical care (N,N,N)	1. CALI 2. Pain Diary 3. ARCS 4. TEI	**1. *RCADS (Depression subscale only*)**	48 (73%)	11-17 (M=14.8, SD=2.0)	Baseline Post treatment 3 month	USA
10	Scharff et al. (2002)	Migraine	NR	RCT 3 arms	1. Hand-cooling (Y,Y,Y) 2. Waitlist control (N,N,N)	1. Headache Pain	***1. Child Depression Inventory (CDI)*** ***2. STAIC*** 3. BDI (parent) 4. STAI (parent)	36 (67%)	7-17 (M=12.8; SD= 2.4)	Post treatment 3 month 6 month 12 month	USA
11	Schurman et al (2010)	Functional Dyspepsia Associated with Duodenal Eosinophilia	Registered nurses with professional biofeedback certification	RCT 2 arms	SMC (N,N,N)	1. Pain Diary 2. FPS-R 3. Global Response Assessment	***1. BASC (somatisation, depression & Anxiety subscales)*** 2. PedsQL 4.0	20 (65%)	8-17 (M=12.2; SD=2.8)	Baseline Post-treatment	USA
12	Trautman n & Kroner-Herwig (2010)	Recurrent Headache	Graduate students of clinical psychology.	RCT 3 arms	1. Internet based relaxation: (Y,Y,N) 2. Education: (Y,Y,Y)	1. Headache diary	1. ***PCS-C*** ***2. CDI*** 3. KINDL (QoL) ***4. SDQ*** 5.Patient-therapist Alliance	66 (55%)	Age range: NR (M=12.7; SD=2.2)	Baseline Post-treatment 6 month	Germany
13	van der Veek, Derkx, Benninga, Boer, de Haan (2013)	functional ab dominal pain	CBT: masters students in psychology / psychologists IMC: Paediatrician or Paediatric Gastroentero logist	RCT 2 arms	Intensive (6 sessions) medical treatment (IMC) (Y, Y, Y)	1. API 2. Pain Diary	1. CSI 2. FDI ***3. RCADS*** 4. KIDSCREEN-27 (QoL)	104 (72.2)	7-18 (M=11.9; SD 2.77)	Baseline Post-treatment 6 month 12 month	Netherlan -ds

14	Warner et al., (2011)	Functional Somatic Complaints (nausea, IBS, RAP; Chest Pain)	PhD level Psychologists trained in CBT	RCT 2 arms	Wait list control (N,N,N)	**1. *ADIS-IV-CP*** **2. *CSR – independent evaluator*** 3. Pain diary 4. CSI	1. CGAS 2. CGI-I	40 (65%)	8-16 M= 12.4 (SD= 2.6)	Baseline Post-treatment 3 month (intervention group only)	USA
15	Wicksell et al. (2009)	Longstanding Pain	Two psychologists trained in CBT	RCT 2 arms	MDT care with amitriptyline (N,N,N)	1. FDI 2. MPI 3. PAIRS 4. Short Form Health Survey (SF-36)	***1. CES-DC*** 2. VAS: Pain Intensity ***3. PCS*** 4. Pain-related Discomfort	32 (78%) 10.8 and 18.1 years	10.8 - 18.1 years (M=14.8, SD= 2.4).	Baseline Post-treatment 3.5 month 6.8 month	Sweden

NR: Not reported
N: No
Y: Yes
FDI: Functional Disability Inventory
API: Abdominal Pain Index
CSI: Child Somatisation Inventory
ARCS: Adult Responses to Children Symptoms
MPI: Multidisciplinary pain Inventory
CSR: Clinical Severity Ratings
PAIRS: Pain and Impairment Relationship Scale
CMAS: Children's Manifest Anxiety Scale (Castaneda, McCandless 8c Palermo, 1956), revised by Reynolds & Richmond (1978)
CES-DC: Centre for Epidemiological studies Depression Scale for Children
STAIC: The State-Trait Anxiety Inventory for Children
PTI: Pain Total Index
CDS: Children's Depression Scale (Lang & Tisher, 1983
SEQ-C: Self Efficacy Questionnaire for children
BYI-II: Beck Youth Inventory, 2nd Edition
PSS: Perceived Stress Scale
CHAS: Children's Headache Assessment Scale (assesses coping responses Budd 8c Kedesdy, 1989)
PCS-C: Pain Catastrophizing Scale for Children
PPPI: Parent Perception of Pain Interference
Chalder fatigue scale (Chalder et al. 1993).
WSAS: Work and Social Adjustment Scale (Mundt et al. 2002).
PCQ: Pain Coping Questionnaire
CALI: Child Activity Limitations Interview
TEI: Treatment Evaluation Inventory
FPS-R: Faces Pain Scale-Revised
GOS: Global Outcome Scale
VAS: Visual Analog Scale
PedsQL: Pediatric Quality of Life Scale

Appendix D: Table of Treatment and Outcomes

	Intervention	**Length of treatment**	**Treatment Fidelity/ manualised?**	**Home practice**	**Main Mental Health Outcomes**	**Main Physical Health Outcomes**
1	1.Biofeedback and progressive muscle relaxation involving: - Progressive muscle relaxation training - EMG Biofeedback: baseline, auditory feedback and self-control (feedback signal turned off, while subject was instructed to continue attempting to relax). 2. Relaxation placebo	7 hours	Manualised: Yes Treatment Fidelity independently rated: NR Supervision: NR	No home practice	Significantly greater reductions in the biofeedback group compared to the control group from pre-treatment to follow-up (post treatment, 3,6, and 12 month) in • trait anxiety** No significant difference between groups on state anxiety	Significantly greater reductions in the biofeedback group compared to the control group from baseline to follow-up (post treatment, 3,6, and 12 month) in • pain total index • EMG ratings (reduction of muscle tension).
2	1.Family focussed CBT: - activity pacing - sleep routine	13 hours	Manualised: Yes Fidelity independently rated: No	Families given treatment manual and homework	Significantly greater reductions in the FFCBT group compared to the	There were no significant differences between groups on post-

	- cognitive re-structuring - encouraging family members to communicate about the illness - relapse prevention. 2.Psycho-ed: dyadic teaching about CFS - pre-disposing and maintaining factors symptom management)		Treatment Fidelity monitored through supervision: Yes		control group from pre-treatment to 24 month follow-up on • emotional • behavioural and • relationship difficulties No significant differences between groups from post-treatment to follow-up (6 and 12 month) on depression scores. Depression scores not reported at 24 month follow-up.	treatment, 6, 12 or 24 month follow-up school attendance or fatigue scores.
3	1. Clinic, group based 2. Home, individual Protocol for both groups: • Education • progressive relaxation and breathing exercises • Cognitive skills I (monitoring positive and negative self-	12 hours	Manualised: Yes Treatment Fidelity independently rated: NR Supervision: NR		Between groups differences were not reported. Statistically significant within groups improvements from baseline to post-treatment for • coping skills for the home and clinic treatment groups but not the control group	There was a significantly greater reduction in • headache scores for groups 1 and 2 compared with group 3 from pre- to post-treatment. There were no significant

	statements about headaches) • Cognitive skills II (generating positive self-statements to replace negative self-statements about exercises headaches) • Mental imagery • relapse management				Statistically significant within groups improvements from baseline to post-treatment for • anxiety for the clinic group, but not the home or the control groups Non-significant reduction in depression scores for the clinic group, but not the home or the control groups Psychological measures were not collected at follow-up (9weeks).	differences between groups 1 and 2 on headache index scores. Non-significant within groups reduction for groups 1 and 2 but not the control group from baseline to post-treatment in • medication usage Between groups differences were not reported at follow-up (9weeks).
4	1.COPE-HEP: • Thinking, feeling, behaving headache triggers • Self-esteem • Stress and coping • Problem solving	3 hours	Manualised:Yes Treatment Fidelity independently rated: No Treatment Fidelity monitored through supervision: Yes		There were significant improvements in both groups from pre- to post- intervention in • anxiety scores**** • depression scores • coping beliefs	There was a significant reduction in headache disability for both groups*** No group differences were observed.

	Headache education • Headache triggers Headache management				Perceived stress scores were not significantly reduced in either group. No between groups differences were observed.	
5	CBT (CST) • muscle relaxation techniques • activity pacing • cognitive techniques (negative thoughts/ mood difficulties) • problem-solving • sleep hygiene • Parent training in behavioural management techniques (changing response to illness) 2. SM Self-monitoring headaches with diary	6 hours	Manualised:Yes Treatment Fidelity independently monitored: No Treatment Fidelity monitored through supervision : Yes	Home practice was assigned for each of these techniques at the end of each training session.	Significant within subjects improvement in both groups from pre-to post-treatment for • depressive symptoms There were no significant differences between groups. Non-significant greater reduction in the SM to CST group compared to the CST to SM group in • depressive symptoms from pre-treatment to follow-up (after cross-over)	Significantly greater reduction in the CST compared with the SM group from pre- to post-treatment in • pain levels Significant within groups differences from pre-to post-intervention in • pain-coping efficacy No significant change in • tender point or • pressure-pain threshold.

						Non-significant greater reduction in the SM to CST group compared to the CST to SM group in • overall functional disability from pre-treatment to follow-up (after cross-over)
6	1.CBT • muscle relaxation techniques • activity pacing • cognitive techniques (negative thoughts/ mood difficulties) • problem-solving • sleep hygiene • Parent training in behavioural management techniques (changing response to illness) 2. FM Education	7 hours 30 minutes	Manualised:Yes Treatment Fidelity independently monitored: Yes Treatment Fidelity monitored through supervision : NR	H/w for both groups on content of previous session	Significantly greater reduction in the CBT group compared with the FME group from pre- to post-treatment in • depression scores. Treatment gains, were maintained at follow-up, however the between groups difference did not reach significance at 6 month follow-up.	Significantly greater reduction in the CBT group compared with the FME group from pre- to post-treatment in • Functional disability Treatment gains were maintained at 6 month follow-up. Significant within groups reduction from baseline to follow-up in • pain severity

	• Information about medication Discussion of lifestyle issues (diet, sleep, exercise)					No significant between group differences.
7	1.SLCBT: • relaxation training • modification of family responses to illness and wellness behaviours • cognitive restructuring 2.Education (ES): • education about GI system anatomy and function • nutrition guidelines additional food-related information.	4 hours	Manualised:Yes Treatment Fidelity independently monitored: Yes Treatment Fidelity monitored through supervision: NR		Significantly greater improvement in the SLCBT group, compared with the ES group in • parent report depression scores • child report catastrophising (anxiety measure) Treatment gains were maintained at follow-up, however the between groups difference was not significant at 3 and 6 month follow-up as the ES group had also improved. Significantly greater improvement in the SLCBT group,	Significantly greater reduction in the SLCBT group, compared with the ES group in • parent report pain from pre-treatment to follow-up (time points: post-treatment, 3 and 6 months) Significantly greater improvement in the SLCBT group, compared with the ES group in parent report • parent and child report symptom severity • parent report child functional disability

					compared with the ES group in parent report • Parental solicitousness (protectiveness in relation to pain) • Perceived pain threat • Problem focussed coping • Emotion focussed coping And child report • Pain coping skills from pre-treatment to follow-up (time points: post-treatment, 3 and 6 months)	from pre-treatment to post-treatment, but not for follow-ups, where the ES group had also improved.
8	1.Self-Administered (SA) at home 2.Clinic based (CB): Protocol for both groups: • Relaxation exercises • Cognitive restructuring	8 hours	Manualised: Yes Treatment Fidelity independently rated: NR Treatment Fidelity monitored through supervision : NR	Yes for both groups (not ctrl)	No between group differences were observed. A significant reduction in • depression scores	Significantly greater reduction for the SA group compared with the CB and control groups from pre- to post-treatment in • headache index scores

	• Distraction and coping strategies • Problem-solving 3.Control: • Provided with list of common triggers for headache, asked to avoid Taught a brainstorming technique for stressful situations				from pre- to post-treatment for all 3 groups. *Treatment gains were maintained for the clinic and self-administer groups at 3 and 12 month follow-up.	Significant within groups improvements from pre- to post-treatment in • headache index scores for clinic and self-administer groups but not the control group. Treatment gains were maintained at follow-up
9	1.Online Family SLCBT: Children: • psycho-education • cognitive restructuring • relaxation techniques Parents: • psychoeducation • operant skills training (response to illness) • communication	9 hours	Manualised: Yes Treatment Fidelity independently rated: NA (online modules)	Yes (assignments between logins)	No significant within group or between group differences on pre- to post-treatment • depression scores. *A significant within groups reduction in the treatment group in • depressive symptoms at 3 month follow-up (control group not followed up).	Significantly greater reduction in the SLCBT group compared with the control group in • activity limitations • pain intensity *Within groups treatment gains in these domains were maintained for the SLCBT group at follow-up (control group not followed up).

	• modelling 2.Wait list control group					
10	1.Hand-warming group: • Thermal biofeedback training • Progressive muscle relaxation • Imagery • Vasodilation • Breathing techniques • Stress management techniques 2.Hand-cooling group: • Thermal biofeedback training • Imagery (cold places) • Peripheral vasoconstriction • Therapist support 3. Wait list control group	4 hours (hand-warming and hand-cooling groups)	Manualised: Yes Treatment Fidelity independently rated: No Treatment Fidelity monitored through supervision : NR	Yes	No significant between or within groups differences reported at any time point on • Depression scores • Anxiety scores	Significantly greater reduction in the hand-warming group than the hand-cooling and waitlist control groups from pre- to post-treatment in • Headache index No between group differences at 3,6 or 12 month follow-up

11	1.BART: • standard medical treatment plus • relaxation methods such as abdominal breathing progressive muscle relaxation imagery and autogenic handwarming • integration and maintenance of these skills. • Participants were provided with a practice CD and a temperature trainer for use at home 2.SMT: Standard medical treatment alone	8 hours and 20 minutes	Manualised: Yes Treatment Fidelity independently rated: NR Treatment Fidelity monitored through supervision : NR	Homework, focused on mastery of relaxation skills	Significantly greater reduction in • parent report depressive scores for the SMC group than the BART+SMC group from pre-treatment to 6 month follow-up. No significant reduction in in either group for anxiety scores	Significantly greater reduction in the BART+SMC group compared with the SMC group from pre-treatment to 6 month follow in • highest level of pain intensity pain duration up
12	1.Internet based CBT: • education on headaches	Unknown (6 modules)	Manualised: Yes Treatment Fidelity independently	Yes	No significant between group differences in	No significant between group differences in

	• stress management • progressive relaxation techniques • cognitive restructuring • self-confidence strategies • problem-solving 2.Applied Relaxation (AR): • progressive relaxation • cue-controlled relaxation • differential relaxation 3.Education (control): headache education		rated: NA (online modules)		• pain catastrophizing (measure of anxiety) Significant reduction in all three groups from pre-treatment to follow-up (time-points at post-treatment and 6 months) in • pain catastrophizing No significant within or between group differences in • depression • emotional adjustment • behavioural adjustment relationship factors	• headache frequency • headache intensity • headache duration Significant reduction in all three groups from pre-treatment to follow-up (time-points at post-treatment and 6 months) in • headache frequency headache duration
13	1.CBT: Tailored to child's needs • relaxation exercises • cognitive re-structuring • graded exposure	4 hours 30 minutes	Manualised: No Tailored to child needs, including the therapists choice of 1 standard and 3 optional modules.	Not reported	Significantly greater reduction in the CBT group compared with the IMC group from pre- to post-intervention and at 6 month follow-up in • depression scores	No significant between group differences on • Pain intensity • pain duration • functional disability

	• behaviour therapy directed at parent (response to illness behaviours) 2.Intensive Medical Treatment (IMT) • prescription of medication • education about complaints • advice about continuing school and daily activities dietary advice		Treatment Fidelity independently rated: NA Treatment Fidelity monitored through supervision : Yes		• anxiety scores. These differences did not reach significance at 12 month follow-up as the IMC group scores had improved.	• Somatisation Significant within groups reduction from pre- to post-treatment for both groups in • Pain intensity • pain duration • functional disability • Somatisation These treatment gains were maintained at 6 and 12 month follow-up.
14	1.CBT (Treatment of Anxiety and Physical Symptoms; TAPS) • relaxation • cognitive restructuring • exposure exercises to target fears related to	14 hours and 15 minutes	Manualised: Yes Treatment Fidelity independently rated: NR Treatment Fidelity monitored through supervision : NR	NR	Significantly greater reductions in TAPS group compared with control group from pre- to post-treatment in • those meeting criteria for Anxiety Disorders	Significantly greater reductions in TAPS compared with waitlist control group from pre- to post-treatment in • parent pain rating • child pain rating

	physical pain and anxiety inducing situations • change parental responses to illness (decrease avoidance related to pain and reinforce active coping) 2.Wait list control group				• clinical severity rating of anxiety *Treatment gains were maintained at 3 month follow-up	Significantly greater increases in TAPS compared with control group from pre- to post-treatment in • independent evaluator global functioning scores *Treatment gains were maintained at 3 month follow-up
15	1.ACT/CBT Child: • exposure to avoided situations • coping: emphasis of acceptance as an alternative to avoidance • cognitive shift in perspective from symptom reduction to valued living • behavioural activation	13 hours	Manualised: Yes Treatment Fidelity independently monitored: Yes Treatment Fidelity monitored through supervision : Yes	Yes	Significantly greater changes were seen in the ACT group as compared to the MDT group from pre- to post-treatment in • Kinesiophobia (pain related anxiety) • pain impairment beliefs This difference reached significance only for pain impairment beliefs when 3.5 and 6.5	Significantly greater changes were seen in the ACT group as compared to the MDT group from pre- to post-intervention in • health-related quality of life • pain interference • pain intensity • pain-related discomfort

	Parent: • exposure • operant mechanisms (adapting responses to illness) • shift perspective from symptom alleviation to valued life 2.Multidisciplinary treatment and amitriptyline (MDT) • The MDT was performed by a psychiatrist, a child psychologist, a physiotherapist and a pain physician Participants were seen by the different health care providers based on individual needs.				month follow-up assessments were included in the analysis. A non-significant greater reduction in the ACT group compared to the MDT group in • in depression scores from pre-treatment to 6.5 month follow-up	These differences reached significance only for Pain intensity and Pain-related discomfort when 3.5 and 6.5 month follow-up assessments were included in the analysis.

NR=Not Reported
*referring to pre-post treatment gains as follow-up comparison not carried out since control group had been given treatment at this stage
** Findings due to baseline differences and were not clinically significant.
***significant at the .10 level
**** Effects disappeared when analysis controlled for baseline levels

Appendix E: NICE Checklist

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Appendix F: Table of Excluded Studies

Author(s) and Year	Study name and publication details	Reason for exclusion
Dennison, L., Stanbrook, R., Moss‐Morris, R., Yardley, L., & Chalder, T. (2010).	Cognitive behavioural therapy and psycho‐education for chronic fatigue syndrome in young people: Reflections from the families' perspective. *British Journal of Health Psychology, 15,* 167-183.	Uncontrolled study
Gauntlett-Gilbert, J., Connell, H., Clinch, J., & McCracken, L. M. (2013).	Acceptance and values-based treatment of adolescents with chronic pain: Outcomes and their relationship to acceptance. *Journal of Pediatric Psychology, 38,* 72-81.	Uncontrolled study
Kennedy, T., Jones, R., Darnley, S., Seed, P., Wessely, S., & Chalder, T. (2005).	Cognitive behaviour therapy in addition to antispasmodic treatment for irritable bowel syndrome in primary care: randomised controlled trial. *British Medical Journal, 331,* 435	Participant age range 16 – 50 years
Labus, J., Gupta, A., Gill, H. K., Posserud, I., Mayer, M., Raeen, H., ... & Mayer, E. A. (2013).	Randomised clinical trial: symptoms of the irritable bowel syndrome are improved by a psycho‐education group intervention. Alimentary pharmacology & therapeutics, *37, 304-315.*	Participants outside age-range
Levy, R. L., Langer, S. L., Walker, L. S., Romano, J. M., Christie, D. L., Youssef, N., ... & Feld, L. D. (2013).	Twelve-month follow-up of cognitive behavioral therapy for children with functional abdominal pain. *JAMA pediatrics, 167,* 178-184.	Does not report mental health outcomes
Lloyd, S., Chalder, T., Sallis, H. M., & Rimes, K. A. (2012).	Telephone-based guided self-help for adolescents with chronic fatigue syndrome: A non-randomised cohort study. *Behaviour research and therapy, 50,* 304-312.	Mental health outcomes not reported
Kashikar-Zuck, S., Sil, S., Lynch-Jordan, A. M., Ting, T. V., Peugh, J., Schikler, K. N., ... & Powers, S. W. (2013).	Changes in pain coping, catastrophizing, and coping efficacy after cognitive-behavioral therapy in children and adolescents with juvenile fibromyalgia. *The Journal of Pain, 14,* 492-501.	Not an intervention study. Same sample as previous included study (Kashikar-Zuck et al., 2012)

Metin, S. Z., Ozmen, M., Metin, B., Talasman, S., Yeni, S. N., & Ozkara, C. (2013).	Treatment with group psychotherapy for chronic psychogenic nonepileptic seizures. *Epilepsy & Behavior, 28,* 91-94.	Participant age range
Nijhof, S. L., Bleijenberg, G., Uiterwaal, C. S., Kimpen, J. L., & van de Putte, E. M. (2012).	Effectiveness of internet-based cognitive behavioural treatment for adolescents with chronic fatigue syndrome (FITNET): a randomised controlled trial. *The Lancet, 379,* 1412-1418.	Mental health outcomes not reported
Osterhaus, S. O. L., Passchier, J., van der Helm-Hylkema, H., de Jong, K. T., Orlebeke, J. F., de Grauw, A. J. C., et al. (1993).	Effects of behavioral psychophysiological treatment on schoolchildren with migraine in a nonclinical setting: Predictors and process variables. *Journal of Pediatric Psychology, 18,* 697–715.	Participants outside age-range (up to and including age 19)
Osterhaus, S. O., Lange, A., Linssen, W. H., & Passchier, J. (1997)	A behavioral treatment of young migrainous and nonmigrainous headache patients: prediction of treatment success. *International Journal of Behavioral Medicine, 4,* 378-396.	Participants outside age-range (up to and including age 22)
Palermo, T. M., Law, E. F., Essner, B., Jessen-Fiddick, T., & Eccleston, C. (2014).	Adaptation of problem-solving skills training (PSST) for parent caregivers of youth with chronic pain. *Clinical Practice in Pediatric Psychology, 2,* 212.	Parent mental health outcomes only
Sieberg, C. B., Flannery-Schroeder, E., & Plante, W. (2011).	Children with co-morbid Recurrent Abdominal Pain and anxiety disorders: Results from a multiple-baseline intervention study. *Journal of Child Health Care, 15,* 126-139.	Significance testing not performed
Stulemeijer, M., de Jong, L. W., Fiselier, T. J., Hoogveld, S. W., & Bleijenberg, G. (2004).	Cognitive behaviour therapy for adolescents with chronic fatigue syndrome: randomised controlled trial. *British Medical Journal, 330,* 14.	Mental health outcomes not reported
Taghizadeh, Z., Shirmohammadi, M., Feizi, A., & Arbabi, M. (2013).	The effect of cognitive behavioural psycho‐education on premenstrual syndrome and related symptoms. *Journal Of Psychiatric And Mental Health Nursing, 20,* 705-713.	Participants outside age-range (up to and including age 19)

Wassom, M. C., Schurman, J. V., Friesen, C. A., & Rapoff, M. A. (2013).	A pilot study of "Gutstrong" for adolescents with functional gastrointestinal disorders. *Clinical Practice in Pediatric Psychology, 1*, 201-210.	Mental health outcomes not reported

Appendix G: Risk of Bias Table Based on the Categories of the Quality Appraisal Checklist (NICE, 2012)

Population and sampling

	1.1	1.2	1.3
1.	•Setting adequately described. •Source population demographics not reported	•Recruitment from specialist centre only •Method of referral to centre not reported (NR)	•Consecutive cases (all cases considered) •Well described appropriate inclusion/exclusion criteria •% participants excluded based on •Study criteria: 0% •Declined to participate: 0%
2.	•Setting adequately described. •Source population demographics not reported	•Recruitment from specialist centre only •Method of referral to centre: GP or paediatrician	•Consecutive cases (all cases considered) •Stringent inclusion/exclusion criteria. •Excluded participants with the following diagnoses: Major Depressive Disorder, Somatisation Disorder, Conversion Disorder •% participants excluded based on •Study criteria: 13% •Declined to participate: 4%
3.	•Setting adequately described. •Source population demographics not reported	•Recruitment from newspaper add (all interested assessed for inclusion)	•Convenience sampling •Well described appropriate inclusion/exclusion criteria •% participants excluded based on •Study criteria: not reported •Declined to participate: not reported
4.	•Setting adequately described. •Source population demographics not reported	•Recruitment from specialist centre only •Method of referral to centre not reported (NR)	•Convenience sample from consecutive cases •Stringent inclusion/exclusion criteria. •Included only participants who's score indicated 'mild to moderate depression' •Excluded participants with 'mental Health conditions'

			•% participants excluded based on •Study criteria: 12% •Declined to participate: 14%
5.	•Setting and source population adequately described. •	•Recruitment from specialist centre only •Method of referral to centre not reported (NR) •Males potentially under-represented (all participants were female although males eligible)	•Convenience sample from consecutive cases •Stringent inclusion/exclusion criteria. •Excluded participants with the following diagnoses: Major Depressive Disorder •% participants excluded based on •Study criteria: 14% •Declined to participate: 18%
6.	•Setting adequately described. •Source population demographics not reported	•Recruitment from 4 specialist centres •Method of referral to centre not reported (NR) •	•Method of sampling not reported •Stringent inclusion/exclusion criteria. •Excluded participants with the following diagnoses: Major Depressive Disorder, Panic Disorder, Bipolar Disorder, Psychosis •% participants excluded based on •Study criteria: 25% •Declined to participate: Not reported
7.	•Setting adequately described. •Source population demographics not reported	•Recruitment from specialist centre •Recruitment via community flyers (all interested assessed for inclusion)	•Consecutive cases •Well described appropriate inclusion/exclusion criteria •% participants excluded based on •Study criteria: 8% •Declined to participate: Not reported
8.	•Setting adequately described. •Source population demographics not reported	•Recruitment from specialist centre only •Method of referral to centre: Paediatrician	•Consecutive cases (all considered) •Excluded participants with ‘major psychological problems’ •% participants excluded based on •Study criteria: Not reported •Declined to participate: Not reported

9.	•Setting adequately described. •Source population demographics not reported	•Recruitment from specialist and general health centres •Method of referral to centre: 'speciality care physician'	•Consecutive cases (all considered) •Well described appropriate inclusion/exclusion criteria •% participants excluded based on •Study criteria: 26% Declined to participate: 19%
10.	•Setting adequately described. •Source population demographics not reported	•Recruitment from general children's hospital •Method of recruitment: contact details referred by neurologist to study investigators, investigators sent information to prospective participants and participants contacted investigators if they were interested.	•Convenience sampling •Well described appropriate inclusion/exclusion criteria •Declined to participate: Not reported
11.	• Setting adequately described. •Source population demographics not reported	•Recruitment from specialist centre only •Method of referral to centre: not reported •	•Consecutive cases (all considered) •Well described appropriate inclusion/exclusion criteria •% participants excluded based on •Study criteria: 0% •Declined to participate: 29%
12.	•Setting adequately described. •Source population demographics not reported	•Recruitment through newspaper adds, website adds, migraine society •People without access to a computer under-represented	•Convenience sample •Well described appropriate inclusion/exclusion criteria •% participants excluded based on •Study criteria: 10% •Declined to participate: 11%
13.	•Setting adequately described. •Source population demographics not reported	•Recruitment from a general children's hospital •Method of referral to hospital: not reported	•Consecutive cases (all considered) •Excluded participants with a 'psychiatric disorder requiring treatment prior to FAP treatment' (e.g. psychosis) •% participants excluded based on •Study criteria: 31% •Declined to participate: 11%

14.	•Setting and source population adequately described.	•Recruitment from primary care and specialist centres via mail out (families contacted research team following mail explaining project)	•Convenience sample •Participants were excluded if they did not meet criteria for an anxiety disorder •% participants excluded based on •Study criteria: 41% •Declined to participate: 29%
15.	•Setting adequately described. •Source population demographics not reported	•Recruitment from specialist centre only •Method of referral to centre: not reported	•Consecutive cases (all considered) •Excluded participants with a 'psychiatric disorder requiring treatment prior to pain treatment' (e.g. suicidality) •% participants excluded based on •Study criteria: not reported •Declined to participate: unable to calculate

Allocation to condition

	2.1	2.2	2.3	2.4	2.5 (reported in description)	2.6	2.7	2.8	2.9 (reported in description)	2.10 (reported in description)
1.	•Allocation: pseudo-randomisation – consecutive admissions	•Interventions were well described •Comparison conditions were appropriate	•Allocation concealment: NR	•Participant blind to condition: NA •Investigator blind to condition: Self report measures: NR (whether blind investigator aided children to fill in) Physiological measure: NR	•Control group controlled for: •Participant time: Yes •Participant attention: Yes •Therapist time with participant: Yes	•No cross-over or contamination reported	•Other interventions: Both groups were taking non-prescription medication	•Attrition was acceptable (<20%). •Attrition bias: 33% for the control group, but 0% for the treatment group.	•Treatment carried out in specialist headache centre	•Intervention delivered by: NR
2.	•Allocation: pseudo-randomisation – consecutive admissions	•Interventions were well described •Comparison conditions were appropriate	•Allocation concealment: A 'list of consecutive random treatment assignments	•Participant blind to condition: NA •Investigator blind to condition:	•Control group controlled for: •Participant time: No	•No cross-over or contamination reported	•Other interventions: Both groups included participant taking	• Attrition was acceptable (<20%) for 3, 6 and 12 month	•Treatment carried out in specialist CFS centre	•Intervention delivered by: Trained CBT therapists

			' was prepared in advance by a non-clinical research assistant'	Self report measures: NR (whether blind investigator aided children to fill in) •Fatigue improvement interview: Assessor blind to condition	•Participant Attention: Yes •Therapist time with participant: No		anti-depressants (stable >3 months n=NR;)	follow-up (<20%) •Attrition bias: 2% in treatment group and 3% in control group. •Attrition was significant at 24 month follow up (30%). This percentage was split across groups: 25% in treatment group and 34% in control group.		
3.	• Allocation: 'true	•Interventions were well described	•Allocation concealment: NR	•Participant blind to condition: NA	•3 groups – clinic, home, self-monitoring.	•Control group 2 (self-monitorin	•Other interventions: It was 'ensured'	•Attrition was acceptable (<20%).	•Treatment carried out in 'clinic' (type of	•Intervention delivered by: A

	randomization'	•Comparison conditions were appropriate		•Investigator blind to condition: Self report measures: NR (whether blind investigator aided children to fill in)	Control group 1 (home based) controlled for: • Participant time: Yes •Participant Attention: Yes •Therapist time with participant: No •Control group 2 (selfmonitoring) controlled for: • Participant time: No •Participant Attention: No •Therapist time with participant: No	g) received intervention at 'post-intervention' phase of study for ethical reasons. These participants were not followed up after receiving intervention	that study participants were not receiving other interventions	•Attrition bias: 12% for each of the home and clinic based treatments and 29% for the self-monitoring group.	clinic: NR) or home	postgraduate clinical psychology student in her final year of studies

4.	•Allocation: Pseudo-randomisation - consecutive admissions	•Interventions were well described •Comparison conditions were appropriate	•Allocation concealment: Online randomisation system'	•Participant blind to condition: NA •Investigator blind to condition: Self report measures: NR (whether blind investigator aided children to fill in)	•Control group (headache education) controlled for: • Participant time: Yes •Participant Attention: Yes •Therapist time with participant: Yes	•No cross-over or contamination reported	•Other interventions:NR	•Attrition was acceptable (<20%). •Attrition bias: Spread across groups (11% in each group)	•Treatment carried out in specialist headache centre	•Intervention carried out by: Nurse Practitioner with cognitive behavioural training
5.	•Allocation: pseudo-randomisation – consecutive admissions	•Interventions were well described •Comparison conditions were appropriate (meds continued: TAU)	•Allocation concealment: Computer generated list	•Participant blind to condition: NA •Investigator blind to condition: Self report measures: NR (whether blind investigator aided	•Control group (self-monitoring with TAU) controlled for: • Participant time: No •Participant Attention: No •Therapist time with participant: No	•Cross-over trial: there was no time left in between the two interventions **(-)**	•Other interventions: Both groups were taking usual medications	•Attrition was acceptable (<20%) Attrition bias: 7% of the treatment to control group and 3% of the control to treatment group	•Treatment carried out in specialist rheumatology clinics	•Intervention carried out by: doctoral level psychology resident and psychology fellow trained in cognitive skills training

				children to fill in) Tender point examination assessor: blind to condition						
6.	•Allocation: pseudo-randomisation – stratified by site	•Interventions were well described •Comparison conditions were appropriate	•Allocation concealment: Computerised allocation	•Participant blind to condition: NA •Investigator blind to condition: Self report measures: NR (whether blind investigator aided children to fill in) •Tender point examination and physician global assessment scale -	•Control group (FM education) controlled for: • Participant time: Yes •Participant Attention: Yes •Therapist time with participant: Yes	•No cross-over or contamination reported	•Other interventions: Both groups 'receiving stable pain medications'	•Attrition was acceptable (<20%) •Attrition bias: spread over the two groups (12% in each group	•Treatment carried out in specialist rheumatology clinics	•Intervention carried out by: post-doctoral CBT trained therapists

				assessor: blind to condition						
7.	•Allocation: pseudo-randomisation – consecutive admissions	•Interventions were well described •Comparison conditions were appropriate	•Allocation concealment: Computerised allocation	•Participant blind to condition: NA •Investigator blind to condition: Self report measures (parents): NA •Investigator blind to condition: Self report measures (children): Nurse blind to condition interviewed children over the phone	•Control group (education) controlled for: • Participant time: Yes •Participant Attention: Yes •Therapist time with participant: Yes	•No cross-over or contamination reported	•Other interventions: Both groups continued their standard medical care	•Attrition was high at end of study (23%). •Attrition bias: 22% in the treatment group and 24% in the control group	•Treatment carried out in specialist GI centre	•Intervention delivered by: trained therapists (Master's degree or higher
8.	•Allocation: pseudo-randomisation – consecutive	•Interventions were well described •Comparison conditions	•Allocation concealment: NR	•Participant blind to condition: NA •Investigator blind to	•3 groups – therapist administered, self-administered and	•Control group received intervention 1 month 'post-	•Other interventions: NR	•Attrition was acceptable (<20%) •Attrition bias: 20%	•Treatment carried out at specialist migraine	•Intervention delivered by: 'Trained therapist'

	admissions and stratified by headache	were appropriate		condition: Self report measures: NR (whether blind investigator aided children to fill in)	control. Control controlled for: • Participant time: No •Participant Attention: Yes •Therapist time with participant: Yes	intervention' phase of study for ethical reasons. These participants were not followed up after receiving intervention		for each of the treatment groups and 7% for the control group	centre or at home	
9.	•Allocation: pseudo-randomisation – consecutive admissions	•Interventions were well described •Comparison conditions were appropriate (TAU)	•Allocation concealment: Computerised allocation	•Participant blind to condition: NA •Investigator blind to condition: Self report measures: NR (whether blind investigator aided children to fill in)	•Control group (TAU) controlled for: • Participant time: No •Participant Attention: No •Therapist time with participant: No	•No cross-over or contamination reported	•Other interventions: Both groups continued medical treatment as usual	•Attrition was acceptable (<20%) •Attrition bias: 4% for the treatment group and 2% for the control group)	•Treatment carried out online	•Intervention delivered by: CBT trained Ph.D. level psychology postdoctoral fellow
10	•Allocation: pseudo-randomisat	•Interventions were well described	•Allocation concealment: NR	•Participant blind to condition:	•Control group (Hand-	•No cross-over or contamina	•Other interventions: All	•Attrition was	•Treatment location: NR	•Treatment delivered by: NR

	ion – stratified by age	•Comparison conditions were appropriate	'randomisation table'	Yes – treatment credibility assessments carried out and found no differences between the treatment and active control group. •Investigator blind to condition: Self report measures: NR (whether blind investigator aided children to fill in) •Assessor (anxiety interview at baseline) blind to condition: NR	cooling) controlled for: • Participant time: Yes •Participant Attention: Yes •Therapist time with participant: Yes •Control group (Waitlist) controlled for: • Participant time: No •Participant Attention: No •Therapist time with participant: No	tion reported	groups continued taking non-prescription and prescription medication	acceptable (<20%) •Attrition bias: 8% of hand-cooling control group, 8% of the waitlist control group and 0% for the treatment group)		

11.	•Allocation: pseudo-randomisation – consecutive admissions	•Interventions were well described •Comparison conditions were appropriate	•Allocation concealment: Computerised allocation	•Participant blind to condition: NA •Investigator blind to condition: Self report measures: NR (whether blind investigator aided children to fill in) •Investigator blind to condition: physician global improvement measure: Blind • •Investigator blind to condition: physicians administering standard	•Control group (SMC) controlled for: • Participant time: No •Participant Attention: No •Therapist time with participant: No	•No cross-over or contamination reported	• Other interventions: Both groups received standard medical care	•Attrition was acceptable (<20%) •Attrition bias: 10% for the treatment group and 10% for the control group	•Treatment carried out at specialist pain clinic	•Intervention delivered by: registered nurses with professional biofeedback certification

				medical care: Blind						
12.	•Allocation: pseudo-randomisation – consecutive admissions	•Interventions were well described •Comparison conditions were appropriate	•Allocation concealment: Computerised allocation	•Participant blind to condition: NR (whether blind investigator aided children to fill in) •Investigator blind to condition: Self report measures: NA	•Control group 1 (AR) controlled for: • Participant time: Yes •Participant Attention: Yes •Therapist time with participant: Yes •Control group 2 (EDU) controlled for: • Participant time: Yes •Participant Attention: Yes •Therapist time with participant: Yes	•No cross-over or contamination reported	•Other interventions: Neither group was receiving psychological therapies or prophylactic mediation	•Attrition was acceptable (<20%) •Attrition bias: drop-out rates did not differ significantly as a function of group (reported by authors)	•Treatment was internet based	•Treatment delivered by: online modules and by graduate clinical psychology therapists by email

13.	•Allocation: pseudo-randomisation – consecutive admissions	•Interventions were well described •Comparison conditions were appropriate	•Allocation concealment: Computerised allocation	•Participant blind to condition: NA •Investigator blind to condition: Self report measures: NR (whether blind investigator aided children to fill in) •Investigator delivering anxiety assessment blind to condition: NR	•Control group (IMC) controlled for: • Participant time: Yes •Participant Attention: Yes •Therapist time with participant: Yes	•No cross-over or contamination reported	•Other interventions: NR	•Attrition was acceptable (<20%) •Attrition bias: 12% for the treatment group and 19% for the control group	• Treatment carried out in a Dutch academic centre of adolescent psychiatry	•Treatment delivered by: CBT: master's students in psychology or psychologists with a master's degree IMC: Paediatrician or Paediatric Gastro-enteologist
14.	•Allocation: pseudo-randomisation – consecutive admissions	•Interventions were well described •Comparison conditions were appropriate	•Allocation concealment: 'pre-determined table of random numbers'	•Participant blind to condition: NA •Investigator blind to condition: Self report measures:	•Control group (WL) controlled for: • Participant time: No •Participant Attention: No	•No cross-over or contamination reported	•Other interventions: NR	•Attrition was acceptable (<20%) •Attrition bias: 3% in WL group and 0% in	•Treatment carried out in hospital outpatients	•Treatment delivered by: CBT trained PhD level Psychologists

				NR (whether blind investigator aided children to fill in) •Investigator delivering assessment blind to condition: blind	•Therapist time with participant: No			treatment group		
15.	•Allocation: pseudo-randomisation – consecutive admissions	•Interventions were well described •Comparison conditions were appropriate	•Allocation concealment: 'sealed envelope prepared by secretary blind to study purpose'	•Participant blind to condition: NA •Investigator blind to condition: Self report measures: NR (whether blind investigator aided children to fill in)	•Control group (MDT care with amitriptyline) Participant time: No •Participant Attention: No •Therapist time with participant: No •Prolonged treatment in the MDT	•No contamination reported. All participants were offered the other treatment following the study follow-up phase	•Other interventions: NR	•Attrition was high (25%) •Attrition bias: 19% in control group and 31% in treatment group	•Treatment carried out at specialist pain clinic	•Treatment delivered by: CBT and ACT trained psychologists

					group complicated comparisons between groups at follow-up assessments					

Outcomes

	3.1	3.2	3.3	3.4	3.5	3.6
1.	•Objective (physiological) measure: Inter-rater reliability or assessor fidelity: NR •Subjective validated, reliable self-report measures •Subjective measure not tested for reliability (headache diary)	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Mood not assessed	•Outcomes relevant: Yes	•Follow-up times were similar for groups	•Followed up at: 1month 3month 6month 12month
2.	• Objective (school attendance; fatigue improvement interview) measure: Inter-rater reliability or assessor fidelity: NR •Subjective validated, reliable self-report measures	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Anxiety not assessed.	•Outcomes relevant: Yes	•Follow-up times were similar for groups	•Followed up at: 3month 6month 12month 24 month

3.	•Subjective validated, reliable self-report measures •Subjective measure not tested for reliability (headache diary)	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Yes	•Outcomes relevant: Yes	•Follow-up times were similar for home and clinic based treatment groups. Self-monitoring group did not receive 9 week follow-up as they had been offered treatment at this time	•Followed up at: Post treatment Nine weeks (not self-monitoring group)
4.	•Subjective validated, reliable self-report measures only	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Yes	•Outcomes relevant: Yes	•Follow-up times were similar for groups	•Followed up at: Post treatment only
5.	•Subjective validated, reliable self-report measures •Objective (tender point exam) measure: Inter-rater reliability or assessor fidelity: NR	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Yes	•Outcomes relevant: Yes	•Follow-up times were similar for groups	•Followed up at: Post treatment 1 (8 weeks) Post treatment 2 (16 weeks)
6.	•Subjective validated, reliable self-report measures •Objective (tender point exam and physician global assessment) measures: Inter-rater reliability or assessor fidelity: NR	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Anxiety not assessed	•Outcomes relevant: Yes	•Follow-up times were similar for groups	•Followed up at: Post treatment 6 months
7.	•Subjective validated, reliable self-report measures	•Identification of participants meeting study outcome	•Important outcomes assessed: Yes	•Outcomes relevant: Yes	•Follow-up times were similar for groups	•Followed up at: 1 month 3 months

		definition: see 2.8 and 4.2				6 months
8.	•Subjective validated, reliable self-report measures Subjective measure not tested for reliability (headache diary)	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Anxiety and functional changes not assessed	•Outcomes relevant: Yes	•Follow-up times were similar for therapist- and self-administered treatment groups. Control group did not receive 3 or 12 month follow-up as they had been offered treatment at this time	•Followed up at: 1 month 3 months (not control group) 12 months (not control group)
9.	•Subjective validated, reliable self-report measures •Subjective measure not tested for reliability (pain diary)	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Anxiety not assessed	•Outcomes relevant: Yes	•Both groups assessed post-treatment. Treatment group only tested at 3 month follow-up	•Followed up at: Post-intervention 3 months (not control group)
10	•Subjective validated, reliable self-report measures •Subjective measure not tested for reliability (head-ache diary)	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Yes	•Outcomes relevant: Yes	• Follow-up times were similar across groups	•Followed up at: Post-intervention 3 months 6 months 12 months
11.	•Subjective validated, reliable self-report measures •Subjective measure not tested for reliability (pain diary) •Objective (physician global assessment)	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Yes	•Outcomes relevant: Yes	•Follow-up times were similar for groups	•Followed up at: Post-intervention 6 months

	measures: Inter-rater reliability or assessor fidelity: NR •Objective: Electronic equipment					
12.	•Subjective validated, reliable self-report measures •Subjective measure not tested for reliability (pain diary)	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Yes	•Outcomes relevant: Yes	•Follow-up times were similar for groups	•Followed up at: Post-intervention 6 months
13.	•Subjective validated, reliable self-report measures •Subjective measure not tested for reliability (pain diary)	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Mood not assessed	•Outcomes relevant: Yes	•Follow-up times were similar for groups	•Followed up at: Post-intervention 6 months 12months
14.	•Subjective validated, reliable self-report measures •Subjective measure not tested for reliability (pain diary) •Objective reliable validated (ADIS and CSR) measures Inter-rater reliability or assessor fidelity: NR	•Identification of participants meeting study outcome definition: see 2.8 and 4.2	•Important outcomes assessed: Yes	•Outcomes relevant: Yes	•Both groups assessed post-treatment. Treatment group only tested at 3 month follow-up	•Followed up at: Post-intervention 3 month follow-up (Treatment group only)
15.	•Subjective validated, reliable self-report measures	•Identification of participants meeting study outcome	•Important outcomes assessed: Yes	•Outcomes relevant: Yes	•Follow-up times were similar for groups	•Followed up at: Post-intervention 3.5 months

		definition: see 2.8 and 4.2				6.8 months

Analysis

	4.1	4.2	4.3	4.4	4.5	4.6
1.	•Between groups differences at baseline: No	•Dropouts (14%) were excluded from the analysis	•A priori power analysis: NR •Expected effect size: NR •Authors acknowledge that the sample size was small which 'raises questions about representativeness and maximises the impact of outliers'.	•Effect size estimates: NR	•Analyses appropriate	•Precision of intervention effects: P values were reported •Alpha level adjustment for multiple testing: Not applied •Data were not fully reported (Effect size estimates: NR)
2.	•Between groups differences at baseline: Yes •Differences adjusted for within analysis: Yes	•Dropouts (6% at 3,6 and 12 month and 30% at 24 months) were excluded from the analysis	•A priori power analysis: acceptable power calculation of 80% based on n=58. • Power calculations at 24 month follow up where n=44: NR	• Effect size estimates: reported	•Analyses appropriate	•Precision of intervention effects: P values were reported •Alpha level adjustment for multiple testing: Not applied •Data were not fully reported (12 months - between groups longitudinal analyses not reported for secondary outcomes;) •Data were not fully reported (Post-intervention results not reported for some

						variables; differences between groups' subscale scores not reported at 24 months)
3.	•Between groups differences at baseline: Not for headache scores •Between groups differences at baseline for other variables: NR	•Dropouts (18%) excluded from analysis	•A priori power analysis: NR •Expected effect size: NR Authors acknowledge 'small' sample and yield that results would need to be replicated with a 'larger sample'.	•Effect size estimates: NR	•Analyses appropriate	•Precision of intervention effects: P values were not always reported (reported for non-parametric tests only) •Alpha level adjustment for multiple testing: Alpha level was reduced to .01 across all analyses •Data were not fully reported – effect size estimates not reported. between groups differences not reported for MH measures (just within groups)
4.	•Between groups differences at baseline: Yes •Differences adjusted for within analysis: Yes	•Dropouts (11%) excluded from analysis	•A priori power analysis: NR •Expected effect size: NR Authors acknowledge 'small' sample	•Effect size estimates: reported	•Analyses appropriate	•P values were reported •Alpha level adjustment for multiple testing: Not applied, alpha level was increased to .10 across all analyses which authors state is 'common practice' for pilot studies with small samples

						•Data were fully reported
5.	•Between groups differences at baseline: No	•Dropouts (10%) excluded from analysis	•Power analysis: power of 69% (below acceptable standard of 80%) •Authors acknowledge 'small' sample	•Effect size estimates: reported	•Analyses appropriate •Medication use not adjusted for	•P values reported •Alpha level adjustment for multiple testing: Applied •Data were not fully reported (differences between subscales not reported)
6.	•Between groups differences at baseline: No	•Dropouts (12%) excluded from analysis	•Power analysis: power of 80% to detect between groups differences	•Effect size estimates: reported	•Analyses appropriate	•P values reported •Alpha level adjustment for multiple testing: Not applied •Data were fully reported
7.	•Between groups differences at baseline: Yes •Differences adjusted for within analysis: Yes	•Intention to treat analysis was carried out	•Power analysis: power at adequate level (>80%)	•Effect size estimates: NR	•Analyses appropriate	•P values reported •Alpha level adjustment for multiple testing: Not applied Data were not fully reported (Effect size estimates not reported)
8.	•Between groups differences at baseline: No	•Dropouts (15%) excluded from analysis	•A priori power analysis: NR •Expected effect size: NR	•Effect size estimates: NR	•Analyses appropriate	•P values not reported •Alpha level adjustment for multiple testing: Applied •Data were not fully reported (P values, effect size estimates)

9.	•Between groups differences at baseline: Yes •Differences adjusted for within analysis: Yes	•Intention to treat analysis carried out	•A priori power analysis: study has adequate power based on sample size achieved	•Effect size estimates: reported	•Analyses appropriate	•P values reported •Alpha level adjustment for multiple testing: Applied •Data were fully reported
10	•Between groups differences at baseline: No	•Drop-outs (6%) excluded from analysis	•Power reported as inadequate (<80%)	•Effect size estimates: NR	•Analyses appropriate	•P values not reported •Alpha level adjustment for multiple testing: Not applied •Data were not fully reported
11.	•Between groups differences at baseline: reported for depression. Differences adjusted for within analysis: NR •Between groups differences at baseline for other variables: NR •Differences adjusted for within analysis: NR	•Intention to treat analysis carried out	A priori power analysis: •Study has adequate power to detect differences based on sample size achieved (primary outcome measures) •Study may not have adequate power to detect differences based on sample size achieved (secondary outcome measures: depression, anxiety)	•Effect size estimates: reported	•Analyses appropriate (mixed linear regression)	•P values not reported •Alpha level adjustment for multiple testing: Not applied •Data not fully reported
12.	•Between groups differences at baseline: No	•Dropouts (12%) excluded from analysis	•Post-hoc power analysis: Study may not have adequate power to detect differences based	•Effect size estimates: reported	•Analyses appropriate (ANOVA with	•P values reported •Alpha level adjustment for multiple testing: not applied

			on sample size (power: 41%)		repeated measures)	•Data were fully reported
13.	•Between groups differences at baseline: reported for anxiety diagnosis (not for self-report anxiety). Differences adjusted for within analysis: NR •Between groups differences at baseline: No	•Intention to treat analysis carried out	A priori power analysis: •Study has adequate power (80%) to detect differences based on sample size achieved	•Effect size estimates: NR	•Analyses appropriate (linear mixed models)	•P values reported •Alpha level adjustment for multiple testing: not applied •Data were not fully reported (effect size estimates)
14.	•Between groups differences at baseline: Yes •Differences adjusted for within analysis: Yes	•Completer analysis carried out	A priori power analysis: NR Authors acknowledge sample 'small'	•Effect size estimates: reported	•Analyses appropriate (chi square tests for dichotomous variables and ANCOVAS for continuous variables)	•P values not reported •Alpha level adjustment for multiple testing: not applied •Data not fully reported
15.	•Between groups differences at baseline: No	•Intention to treat analysis carried out	A priori power analysis: NR	•Effect size estimates: reported	•Analyses appropriate	•P values reported •Alpha level adjustment not applied •Data were fully reported

Appendix H: Summary of Quality Appraisal Based on the Categories of the NICE (2012) Quality Appraisal Checklist

	1.1	1.2	1.3	2.1	2.2	2.3	2.4	2.5	2.6	2.7	2.8	2.9	2.10	3.1	3.2	3.3	3.4	3.5	3.6	4.1	4.2	4.3	4.4	4.5	4.6	**EV**	**IV**	
1.	+	-	+	+	++	NR	NR	++	++	++	+	++	++	+	NA	+	++	++	++	+	+	-	NR	++	-	-	+	1
2.	+	-	-	+	++	++	NR	+	++	++	+	++	++	+	NA	+	++	++	++	++	+	+	++	++	-	-	+	2
3.	+	++	+	++	++	NR	NR	+	++	++	+	++	++	+	NA	++	++	+	+	NR	+	-	NR	++	-	-	+	3
4.	+	-	-	+	++	++	NR	++	++	NR	++	++	++	+	NA	++	++	++	+	++	+	-	++	++	+	-	+	4
5.	++	-	-	+	++	++	NR	-	-	++	+	++	++	+	NA	++	++	++	+	+	+	-	++	++	-	-	-	5
6.	+	-	-	+	++	++	NR	++	++	++	++	++	++	+	NA	+	++	++	++	+	+	++	++	++	+	-	++	6
7.	+	++	+	+	++	++	++	++	++	++	+	++	++	+	NA	++	++	++	++	++	++	++	NR	++	-	+	++	7
8.	+	-	+	+	++	NR	NR	+	++	NR	+	++	++	+	NA	+	++	+	++	+	+	NR	NR	++	-	-	+	8
9.	+	+	+	+	++	++	NR	-	++	++	+	++	++	+	NA	+	++	+	+	++	++	++	++	++	++	+	++	9
10	+	+	+	+	++	NR	NR	+	++	++	+	NR	NR	+	NA	++	++	++	++	+	+	++	NR	++	-	+	+	10
11.	+	-	+	+	++	++	NR	-	++	++	++	++	++	++	NA	++	++	++	++	-	++	+	++	++	-	-	+	11
12.	+	+	+	+	++	++	NR	++	++	++	++	++	++	+	NA	++	++	++	++	+	+	-	++	++	+	+	+	12
13.	+	+	+	+	++	++	NR	++	++	NR	+	++	++	+	NA	+	++	++	++	+	++	++	++	++	-	+	++	13
14.	++	+	-	+	+	++	NR	-	++	NR	+	++	++	+	NA	++	++	+	+	++	+	-	++	++	-	-	+	14
15	+	-	+	+	++	++	NR	-	++	NR	+	++	++	+	NA	++	++	++	++	+	++	NR	++	++	+	-	+	15

++ Strong validity
+ Adequate
- Weak
EV: External Validity
IV: Internal Validity

Appendix I: Qualitative Questions from the Belief Elicitation Study

1. What do you believe are the advantages of referring a patient/family to paediatric psychology?

2. What do you believe are the disadvantages of referring a patient/family to paediatric psychology?

3. Please give details of any other views you have about referring a patient/family to paediatric psychology.

4. Are there any individual(s) or group(s) that you know of who you think would approve of your referring a patient/family to paediatric psychology? If so, please give details.

5. Are there any individual(s) or group(s) that you know of who you think would disapprove of your referring a patient/family to paediatric psychology? If so, please give details.

6. Is there anything else you associate with other people's views about referring a patient/family to paediatric psychology?

7. What factors or circumstances would enable you to refer a patient/family to paediatric psychology?

8. What factors or circumstances would make it difficult or impossible for you to refer a patient/family to paediatric psychology?

9. Are there any other issues that come to mind regarding the barriers to and facilitators of your referring a patient/family to paediatric psychology?

Appendix J: Ethics approval letter

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Appendix K: R&D Approval Letter from Site 1

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Appendix L: R&D Approval Letter from Site 2

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Appendix M: Participant information sheet and consent form for stage 1 and stage 2

Study title: Factors influencing the referral of children and families to paediatric psychology: Using theory of planned behaviour to develop a questionnaire of health care professionals' referral behaviour.

We would like to invite you to take part in our research study. Before you decide we would like you to understand why the research is being done and what it would involve for you. Please take time to read the following information carefully and discuss it with other members of staff from your team if you wish. Please contact me if anything is unclear or if you would like more information. Please take time to decide whether or not you wish to take part.

What is the purpose of the study?
Paediatric psychology is a relatively new branch of clinical psychology in which psychologists work closely with medical professionals to reduce emotional consequences of physical ill-health and improve quality of life for young people with medical problems. Recent research demonstrates the importance of inter-professional care in improving health outcomes. Research shows the extent to which different healthcare professionals work well together can affect the quality of the health care that they provide. Despite the benefits of inter-professional collaboration, the mechanisms underlying its effectiveness are under-researched and poorly understood. It has been hypothesised that values, attitudes, prejudice and stereotypical views between different professional groups may theoretically improve or dilute the success of inter-professional care. However, there is currently no tool which measures clinicians' attitudes towards and beliefs about paediatric psychology. Furthermore, the impact of these attitudes and beliefs on the amount of paediatric psychology input to medical teams and the rate of referral to paediatric psychology remain unknown. The current study aims to develop a questionnaire measure to explore clinicians' attitudes towards paediatric psychology so that we can begin to understand mechanisms impacting the success of inter-professional collaboration. We anticipate that understanding these mechanisms through the development of this measure will lead to future research and in the long-term, we hope that this will lead to improved inter-professional collaboration between paediatric psychologists and the wider paediatric care team and hence improved services for paediatric patients and their families.

Why have I been invited?
You are being invited to take part in this study as you are a clinician who works within a medical team that has access to support from paediatric psychology.

Do I have to take part?
It is up to you to decide whether to take part in the study. If you agree to take part, we will ask you to tick the consent box below. You are free to withdraw at any time without giving a reason. This will not impact your work.

What will I need to do if I decide to take part?
You will be asked to fill in a questionnaire about your attitudes towards paediatric psychology. Completing this questionnaire should take approximately 5 - 15 minutes.

What are the possible benefits of taking part?
There are no personal benefits to taking part in this study. We hope that in the long term, research such as this will lead to improved inter-professional collaboration between paediatric

psychologists and the wider paediatric care team, hence improving services for paediatric patients and their families.

What if there is a problem?
Any complaint about the way you have been treated during the study or any possible harm you might suffer will be addressed. If you have any concerns or complaints you should contact the researcher in the first instance.

What will happen if I don't want to carry on with the study?
If you decide to take part you are still free to withdraw at any time without giving a reason. If you withdraw consent from the study when some data has already been collected, unless you object, the data we collect may be included in the final study analysis.

Complaints
If you have a concern about any aspect of this study, you should email the researcher who will do their best to answer your questions. The researcher will be supervised by XXXXX of XXXXX and XXXXX of XXXXX, both of whom will be made aware of any complaints.

Will my taking part in this study be kept confidential?
If you consent to take part in this study, all data will remain strictly confidential at all times. However, in the very unlikely event that the data we receive indicates malpractice, the proper procedures for managing this will be put into place. The information will be held securely on paper and electronically under the provisions of the 1998 Data Protection Act. You will be allocated an ID number, which will be used as a code to identify your data.

In line with the 1998 Data Protection Act, at the end of the study your data may be securely archived for 10 years. Arrangements for confidential destruction will then be made.

What will happen to the results of the research study?
The results of the study will be available after it finishes and will usually be published in a medical journal or be presented at a scientific conference. The data will be anonymous and none of the participants involved will be identified in any report or publication. The results of the study will be written up as part a Doctoral Thesis. A letter outlining the results of the study (on a group level) will be sent to all participants and a presentation will be delivered to relevant groups (e.g. paediatric team meetings, paediatric psychology meetings). The results may also be published in a journal and be presented at relevant conferences.

Who is organising and funding the research?
Canterbury Christ Church University

Who has reviewed the study?
All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by Canterbury Christ Church University Research Ethics Committee.

If you decide you would like to take part then please read and sign the consent form (or tick the box below if you are completing this online). You can have more time to think this over if you are at all unsure.

Thank you for taking the time to read this information sheet and to consider this study.

Contact Details

If you would like more information or this study has harmed you in any way you can contact Canterbury Christ Church University using the details below for further advice and information.

Researcher
Christine O'Connell, Trainee Clinical Psychologist
Salomons Centre for Applied Psychology
Canterbury Christ Church University
Runcie Court, David Salomon Estate
Broomhill Road
Tunbridge Wells TN3 0TF
c.oconnell148@canterbury.ac.uk

Consent

I confirm that I have read and understand the information on this study presented above

I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care, work or legal rights being affected

I agree to take part in the above study.

Please select

Yes
No

Appendix N: Coding Frame Based on Theory of Planned Behaviour (with Coded Beliefs)

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Appendix O: Main Questionnaire

No.	Question	Response							
1	If I refer a child/the family of a child with chronic illness to psychology, they will receive a holistic approach to care	Not at all Likely	1	2	3	4	5	6	7 Extremely Likely
2	Children/families will be concerned about mental health stigma if I refer them to psychology	Not at all Likely	1	2	3	4	5	6	7 Extremely Likely
3	If I refer a child/family to psychology, the type of service they receive (e.g. therapy type or therapist profession/level) will be different to what I had intended	Not at all Likely	1	2	3	4	5	6	7 Extremely Likely
4	Referring children/families to psychology means that my time will be taken up by lengthy meetings and joint appointments	Not at all Likely	1	2	3	4	5	6	7 Extremely Likely
5	The process of referring a child/family to psychology is long and arduous	Strongly Agree	1	2	3	4	5	6	7 Strongly Disagree
6	If I refer children/families to psychology, they will have to wait a long time before they see a psychologist	Not at all Likely	1	2	3	4	5	6	7 Extremely Likely
7	Children/families receiving a different type of psychology service (e.g. therapy type or therapist profession/level) than I had intended at referral is	Extremely Desirable	1	2	3	4	5	6	7 Extremely Undesirable
8	Concern experienced by children/families about mental health stigma due to a psychology								

	referral is extremely undesirable for me	Strongly Agree (Undesirable)	1	2	3	4	5	6	7 Strongly Disagree
9	Children/families receiving a holistic approach to care is	Extremely Desirable	1	2	3	4	5	6	7 Extremely Undesirable
10	Budget holders think I should refer children/families to psychology	Strongly Agree	1	2	3	4	5	6	7 Strongly Disagree
11	Families/children would approve of my referring them to psychology	Strongly Agree	1	2	3	4	5	6	7 Strongly Disagree
12	My colleagues (those within my profession and other members of the MDT) refer children/families to psychology	Strongly Agree	1	2	3	4	5	6	7 Strongly Disagree
13	Overall, I think referring a child/family to psychology is	Harmful	1	2	3	4	5	6	7 Beneficial
14	Overall, I think referring a child/family to psychology is	Bad Practice	1	2	3	4	5	6	7 Good Practice
15	Overall, I think referring a child/family to psychology is	Useful	1	2	3	4	5	6	7 Worthless
16	Doing what my colleagues (those within my profession and other members of the MDT) do is important to me	Not at all	1	2	3	4	5	6	7 Extremely
17	The approval of families/children with whom I work is important to me	Not at all	1	2	3	4	5	6	7 Extremely
18	Doing what budget holders think I should do is important to me	Not at all	1	2	3	4	5	6	7 Extremely
19	I am less likely to refer children/families to psychology if the referral process is long and arduous	Strongly Agree (Less likely)	1	2	3	4	5	6	7 Strongly Disagree

20	I am less likely to refer children/families to psychology if it means that my time will be taken up by lengthy meetings and joint appointments	Strongly Agree (Less likely)	1	2	3	4	5	6	7	Strongly Disagree
21	I am less likely to refer children/families to psychology if they will have to wait a long time before they see a psychologist	Strongly Agree (Less likely)	1	2	3	4	5	6	7	Strongly Disagree
22	Most people whose opinions are important to me think that I should refer children/families to psychology	Strongly Agree	1	2	3	4	5	6	7	Strongly Disagree
23	I expect to refer children/families I see to psychology	Strongly Agree	1	2	3	4	5	6	7	Strongly Disagree
24	It is expected of me that I refer children/families to psychology	Strongly Agree	1	2	3	4	5	6	7	Strongly Disagree
25	I am confident that I could refer children/families to psychology if I wanted to	Strongly Agree	1	2	3	4	5	6	7	Strongly Disagree
26	For me, referring children/families to psychology is	Easy	1	2	3	4	5	6	7	Difficult
27	Whether I refer children/families to psychology is within my own control	Strongly Agree	1	2	3	4	5	6	7	Strongly Disagree
28	In general, I want to refer children/families I see to psychology	Strongly Agree	1	2	3	4	5	6	7	Strongly Disagree
29	I feel under social pressure to refer children/families to psychology	Strongly Agree	1	2	3	4	5	6	7	Strongly Disagree

Appendix P: Coded Transcript from a Randomly Selected Participant

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Appendix Q: Author Guideline Notes for the Journal of Paediatric Psychology

Instructions to Authors

The Journal of Pediatric Psychology is an official publication of the Society of Pediatric Psychology, Division 54 of the American Psychological Association. JPP publishes articles related to theory, research, and professional practice in pediatric psychology.

Types of Manuscripts:

- Original research, including case studies
- Review articles
- Commentaries

Manuscript preparation: General Instructions

Full instructions for uploading data and files etc. are given on Manuscript Central at the website under Instructions for online submission:

http://www.oxfordjournals.org/our_journals/jpepsy/for_authors/submission_online.html

Organization of manuscripts

Manuscript Central will guide authors through the submission steps, including: Abstract, Keyword selection, and the Manuscript. The manuscript must contain an Introduction, Methods, Results, Discussion, Acknowledgements and Reference List.

Length of manuscript: Original research articles should not exceed 25 pages, in total, including title page, references, figures, tables, etc. In the case of papers that report on multiple studies or those with methodologies that necessitate detailed explanation, the authors should justify longer manuscript length to the Editor in the cover letter. Case reports should

not exceed 20 pages. Review articles should not exceed 30 pages. Commentaries should not exceed 4 pages. The Journal of Pediatric Psychology no longer accepts brief reports but will accept manuscripts that are shorter in length than the 25 page manuscripts.

Manuscripts (text, references, tables, figures, etc.) should be prepared in detailed accord with the Publication Manual of the American Psychological Association (6th ed.). There are two exceptions:

(a) The academic degrees of authors should be placed on the title page following their names, and

(b) a structured abstract of not more than 150 words should be included. The abstract should include the following parts:

(1) Objective (brief statement of the purpose of the study);
(2) Methods (summary of the participants, design, measures, procedure);
(3) Results (the primary findings of this work); and
(4) Conclusions (statement of implications of these data).

Key words should be included, consistent with APA style. Submissions should be double-spaced throughout, with margins of at least 1 inch and font size of 12 points (or 26 lines per page, 12-15 characters per inch). Authors should remove all identifying information from the body of the manuscript so that peer reviewers will be unable to recognize the authors and their affiliations. E-mail addresses, whenever possible, should be included in the author note.

Informed consent and ethical treatment of study participants. Authors should indicate in the Method section of relevant manuscripts how informed consent was obtained and report the approval of the study by the appropriate Institutional Review Board(s). Authors will also be asked to sign a statement, provided by the Editor that they have complied with the American Psychological Association Ethical Principles with regard to the treatment of their sample.

Clinical relevance of the research should be incorporated into the manuscripts. There is no special section on clinical implications, but authors should integrate implications for practice, as appropriate, into papers.

Terminology should be sensitive to the individual who has a disease or disability. The Editors endorse the concept of "people first, not their disability." Terminology should reflect the "person with a disability" (e.g., children with diabetes, persons with HIV infection, families of children with cancer) rather than the condition as an adjective (e.g., diabetic children, HIV patients, cancer families). Nonsexist language should be used.

Special instructions for types of manuscripts

(1) Treatment studies/Randomized controlled trials: If you are submitting a manuscript of a randomized clinical trial to JPP, you are required to submit a flowchart of your research showing the steps found in the Consort E-Flowchart. This should be submitted as a figure. The Consort E-Flowchart and a checklist of items to be included when reporting a randomized trial can both be found on http://www.consort-statement.org Please clearly indicate the page numbers where each checklist item is reported in the manuscript. Please upload this checklist as supplementary material when you submit your manuscript for consideration.

(2) Case Studies: Although there may be some exceptions, most case studies should be sent to Clinical Practice in Pediatric Psychology (CPPP). Single-subject studies that employ rigorous A-B-A-B designs and/or statistical strategies can be sent to JPP. All others will probably fit better with CPPP. Case reports should not exceed 20 pages. Case reports are appropriate to document the efficacy of new treatment applications; to describe new clinical phenomena; to develop hypotheses; to illustrate methodological issues, difficult diagnoses, and novel treatment approaches; and to identify unmet clinical or research needs. Guidelines for case study submissions can be found in Drotar, D. (2009). Editorial: Case Studies and Series: A Call for Action and Invitation for Submissions, Journal of Pediatric Psychology, 34, 795-802; Drotar, D. (2011). Editorial: Guidance for Submitting and Reviewing Case Reports and Series in the Journal of Pediatric Psychology, 36, 951-958.

Guidelines for Single Subject Studies: Please read Rapoff, M. & Stark, L. (2008). Editorial: Journal of Pediatric Psychology Statement of Purpose: Section on Single-Subject Studies.

(3) Measurement development and validation articles: For additional guidance please read, Holmbeck, G. & Devine, K. (2009) Editorial: An Author's Checklist for Measure Development and Validation Manuscripts.

(4) Review articles: Please consult the recent editorial (New Guidelines for Publishing Review Articles in JPP) which describes new guidelines for review articles, and the Checklist for Preparing and Evaluating Review Articles.

a) Topical reviews: Topical reviews summarize contemporary findings, suggest new conceptual models, or highlight noteworthy or controversial issues in pediatric psychology.

They are limited to 2,000 words, contain no more than 2 tables or figures, and have an upper limit of 30 references. Supplementary online material (e.g., additional tables) may be considered on a case by case basis.

b) Systematic reviews: Systematic reviews should not exceed 30 pages. Authors are required to attach the PRISMA checklist and flow diagram as supplementary material for each submission. Authors can find the PRISMA checklist and flow diagram in downloadable templates that can be re-used at this URL, http://www.prisma-statement.org/statement.htm. Authors of systematic reviews that do not include a meta-analysis must provide a clear statement in the manuscript explaining why such an analysis is not included for all or relevant portions of the report.

(5) Commentaries: Commentaries are invited on all topics of interest in pediatric psychology, and should not exceed 4 pages, including references.

(6) Historical Analysis in Pediatric Psychology is a special series of papers devoted to the history of pediatric psychology. Authors interested in submitting a paper for this series should contact the Editor of JPP to discuss potential papers prior to submission. There is no deadline for these papers (they may be submitted anytime). All submissions will be peer reviewed and should comply fully with the JPP Instructions to Authors. Papers in this series should be tightly focused contributions that expand our understanding of the roots, evolution, and/or impact of pediatric psychology as a discipline. Manuscripts may focus on the influence of individuals, published works, organizations, conceptualizations, philosophies or approaches, or clinical and professional activities. Successful papers should articulate a clear purpose/question and develop a compelling argument for the topic. Contributions should

include a breadth of coverage, such that contradictory data are included and potential biases acknowledged. Historical analysis is more than a recounting of the “facts” and should include a thoughtful and scholarly interpretation of the subject matter. Papers should rely on primary sources and must be clearly and appropriately referenced. Supplemental materials to accompany the article may be posted online.

Additional Guidance:

The following links provide additional guidance for authors and reviewers. Editorial Policy, Authors' Checklist, Guidelines for Reviews, Suggestions for Mentored Reviews, "People First,", NIH policy, Replication of research, Duplicate and redundant policies Conflict of interest

See the following articles for detailed guidance concerning preparation of manuscripts: Editorial: Thoughts in Improving the Quality of Manuscripts Submitted to the Journal of Pediatric Psychology: How to Write a Convincing Introduction. ; Methods: Editorial: How to Report Methods in the Journal of Pediatric Psychology; Results and Discussion: Editorial: How to Write an Effective Results and Discussion Section for the Journal of Pediatric Psychology.

Funding

Details of all funding sources for the work in question should be given in a separate section entitled 'Funding'. This should appear before the 'Acknowledgements' section.

The following rules should be followed:

- The sentence should begin: ‘This work was supported by …’

- The full official funding agency name should be given, i.e. ‘the National Cancer Institute at the National Institutes of Health’ or simply 'National Institutes of Health', not 'NCI' (one of the 27 subinstitutions) or ‘NCI at NIH’ (full RIN-approved list of UK funding agencies)
- Grant numbers should be complete and accurate and provided in parentheses as follows: ‘(grant number xxxx)’
- Multiple grant numbers should be separated by a comma as follows: ‘(grant numbers xxxx, yyyy)’
- Agencies should be separated by a semi-colon (plus ‘and’ before the last funding agency)
- Where individuals need to be specified for certain sources of funding the following text should be added after the relevant agency or grant number 'to [author initials]'.

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Appendix R: Extracts from Research Diary

Jan 2014

I am interested in carrying out a project to do with paediatric psychology, an interest which stems from my work as an assistant psychologist at a children's hospital. I contact Professor Roz Shafran, with whom I used to work. We discuss ideas about physical illness and mental health in children. I'm interested in medically unexplained symptoms, because as a clinician I have observed that in order to work with these symptoms, integration of psychology and medicine is required.

March 2014

Met with Roz to discuss the feasibility of recruiting a sample of children experiencing medially unexplained symptoms. This would prove difficult due to service re-structuring at the hospital. Discuss alternatives, such as interviewing healthcare professionals about their experiences of working with psychologists in relation to medically unexplained symptoms.

April 2014

Work on research proposal, due in May. Decide to broaden research question to include doctors working with paediatric psychology across all illness categories. Through research, study aim evolves to creating a questionnaire to identify factors associated with referral to paediatric psychology.

June 2014

MRP Proposal review at Salomons. Reviewers expressed interest in the project but suggested to think carefully about the process of creating the questionnaire and also necessary participant numbers. It was decided to use theory of planned behaviour to create the questionnaire because of its efficacy in predicting behavioural intention in healthcare professionals. I also decided to add a second research site to maximise participants.

November 2014 – January 2015

Ethics submitted and approved without conditions. Putting time in to changing the project following the reviewers comments seemed to have paid off. I had two separate R&D

processes at the two hospital sites. This took a lot of time and it felt like I was writing the same information over and over in several different forms. Once I submitted the applications the approval came quickly.

January 2014 – April 2014

Stage 1 recruitment involved meetings with the heads of psychology at the two hospital sites, presenting my project. At the first site meeting the head psychologist suggested recruitment could be maximised if she sent emails with the questionnaire links on my behalf. I accepted this offer gratefully and subsequently requested the same at a meeting with the head psychologist at the 2nd site. For the first site during stage 1, it was only possible to recruit consultant medical doctors as opposed to all the healthcare professionals in the hospital due to service restructuring. This is disappointing but seemed to be important in relation to how the service re-structuring may impact healthcare professionals and psychology services. The outcome of the service re-structure was unknown at the time. To maintain consistency between sites, and because I required only 25 participants for stage 1, I decided to recruit only consultant medical doctors from both sites at this stage.

May 2014 – July 2014

Stage 1 recruitment from both sites. I emailed each doctor individually with a survey link, after they had received an email from the head of psychology at each site which described the project. I received qualitative data from 23 consultants, which felt like a reasonable number, given that I was aiming for 25. I decided not to send out reminders as I was aware I would be asking this population to be filling in a second questionnaire in a few months and did not want to continually bother them with emails.

August 2015 – Sept 2015

Analysis of qualitative data. I began by reading and re-reading the data with the theory of planned behaviour constructs in mind. I first grouped statements according to the appropriate construct(s). I then began to make subgroups of themes. Stigma stood out as an interesting theme, as I had been interested in the impact of this (if any) on the mental healthcare children and families receive. I was also interested to read about the perception of some participants - that a child would more likely benefit from psychology by being ‘psychologically minded’.

Another interesting idea was that psychology referrals were helpful for 'abnormal' emotional reactions in children and adolescents, as opposed to a natural response to a challenge. Some participants expressed annoyance at the type of psychology models used within the service. One participant remarked that a 'narrative approach has been adopted without any discussion'. This made me think about the opportunities psychologists have to share knowledge with other medical professionals about why particular approaches are offered in particular contexts for particular families.

September - October 2015

I missed the formative deadline for section A draft (Sept 28th 2015). This is due to working on analysing data for section B. I am also in a very busy and demanding placement at this time (paediatric psychology) and feeling stressed about the size of this project and the challenges of carrying it out alongside clinical work and other course assignments.

November – December 2015

Stage 2 recruitment. I email the participants (all healthcare professionals with access to paediatric psychology) in both sites, following emails from the heads of psychology asking participants to read my email and respond if they have time.

February 2016

First draft section A completed. I received helpful and constructive feedback from my supervisors which was welcomed. Meeting with Paul Camic to discuss section B write up. Feeling time pressured.

March – April 2016

Quantitative data analysis and write up. Analysis seem to suggest decent psychometric properties for the questionnaire and some interesting findings relating to which beliefs impact intention to refer. I am encouraged to find that holistic care and the families views are important factors relating to psychology referral.

Appendix S: End of Study Letter to Ethics Panel

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Appendix T: End of Study Report to Research Ethics Committee form Site 1

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Appendix U: End of Study Report to Research Ethics Committee form Site 2

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Appendix V: End of Study Report for Participants

13/04/2016

Dear Research participants

Thank you for taking part in a research study about factors influencing healthcare professionals' referral of children and families to paediatric psychology. Due to the high rates of distress and mental health difficulties in children and adolescents with physical health problems, several studies have shown that referral of these children and their families to paediatric psychology should be increased. Additionally, research has shown the benefits of integrating physical and mental health services for those with physical health difficulties both in terms of clinical and cost effectiveness.

However, there are few studies investigating factors influencing healthcare professionals' referral behaviour. Following stage 1 of your participation (qualitative questions about views on paediatric psychology), a questionnaire was developed which aimed to examine how these views impact paediatric healthcare professionals' referral behaviour (specifically referral to paediatric psychology).

The study used theory of planned behaviour to develop the questionnaire. According to this theory, an individual's behaviour can be predicted based on attitudes, subjective norms and perceived behavioural control, all of which are influenced by beliefs. Behavioural beliefs create a positive or negative attitude toward a behaviour; normative beliefs produce subjective norm (perceived social obligation); and control beliefs lead to perceived behavioural control. Together, these constructs lead to a behavioural intention, which people usually carry out.

Findings indicate that the questionnaire holds good reliability and validity and that the main constructs of theory of planned behaviour (attitudes, subjective norm and perceived behavioural control) are useful in predicting intention to refer to paediatric psychology. Subjective norm (others' ideas standards and behaviours) was the strongest predictor of

intention to refer to paediatric psychology. The views and ideas of colleagues within the healthcare professional's own profession was a salient factor.

Specific beliefs about referral, including valuing a holistic approach to care were also shown to influence intention to refer to paediatric psychology. This shows that where information sharing between disciplines is valued, healthcare professionals are more likely to refer to paediatric psychology. Beliefs about families' approval of referral were also shown to influence intention to refer to paediatric psychology. Encouragingly, this finding shows the growing role of children and family involvement in decision making about their use of mental health services.

Findings that individual referrer factors such as attitudes and beliefs can impact healthcare professionals' referral behaviour indicates that multidisciplinary interventions and inter-professional education relating to the psychological aspects of illness may be useful. Recommendations for future research are recommended, including controlling for other characteristics such as gender and level of healthcare professional experience.

With best wishes

Christine O'Connell

Trainee Clinical Psychologist