Behavioural and Cognitive Psychotherapy, 2002, 30, 451–458
Printed in the United Kingdom DOI: 10.1017/S135246580200406X

Clinical Section

SELF-HELP MANUAL-ASSISTED COGNITIVE BEHAVIOURAL THERAPY FOR SICKLE CELL DISEASE

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Abstract. The feasibility of using a self-help treatment manual to assist a cognitive behavioural therapy programme developed with the aim of improving pain coping ability and quality of life, and reducing psychological distress in sickle cell patients was evaluated. Adult patients attending a London hospital outpatient sickle cell clinic were invited to 12-week cognitive behavioural therapy. Outcome was assessed with measures of pain status, health service utilization, psychological coping (Coping Strategies Questionnaire – revised for Sickle Cell Disease), quality of life (Medical Outcomes Survey Short Form 36), and mood (Hospital Anxiety and Depression Scale). Baseline disease severity data were also collected. No significant differences in baseline data were observed between patients who completed cognitive behavioural therapy and ''dropouts''. Improvements over baseline at 12 weeks in patients who completed cognitive behavioural therapy were significant. Active coping and use of physician advised methods for pain increased. Emotional responses decreased, general health and vitality improved, while anxiety reduced. This demonstrates that a brief manual-assisted cognitive behavioural therapy programme is feasible in sickle cell disease, and may help to improve coping, quality of life and mood.

Keywords: Sickle cell disease, pain, cognitive behavioural therapy, self-help manual.

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Introduction

Self-help materials are now commonly employed to assist and enhance cognitive behavioural therapy (CBT) programmes, and have been shown to contribute to positive clinical changes in various interventions (Keeley, Williams, & Shapiro, 2002). However, the utilization of self-help adjuncts in psychological interventions for patients with sickle cell disease (SCD) has not been evaluated. We therefore developed a self-help manual-assisted CBT programme aimed at reducing physical and psychological problems associated with SCD.

SCD causes vascular occlusion with consequent pain and organ damage. Pain is the most prominent symptom causing considerable distress to patients and carers. Importantly, the majority of patients are not afflicted with frequent severe painful episodes requiring hospitalizations but live with a chronic illness that interferes with their daily lives. Pain accounts for over 90% of acute hospital admissions for SCD in Britain, with 6% of patients making up more than 40% of hospital admissions (Brosovic, Davies, & Brownell, 1987). Recent research in SCD adults attending London hospitals revealed that about 50% of accident and emergency visits and 45% of hospitalizations were by 10% of patients, while pain experience accounted for 12% of health service utilization independently of demographic and clinical severity factors (Anie, Steptoe, & Bevan, 2002). SCD involves not only severe recurrent pain, but also other impairments in quality of life; psychological coping patterns were shown to be relevant both to pain experience, and to broader adjustment (Anie et al., 2002). Contrary to previous studies, coping and quality of life were both unrelated to health service utilization suggesting that patients who make less frequent contact with health services are not necessarily free from daily problems relating to SCD (Anie et al., 2002). Therefore, interventions that enhance the use of appropriate coping techniques should not only focus on pain and utilization of health services, but also on quality of life.

Self-management interventions provide information to the patient, and also encourage the acquisition or improvement of effective coping skills to alleviate symptoms and achieve better quality of life. This approach is attractive for SCD because patients have to learn to cope daily with a chronic disabling condition. Also, studies on psychological therapies for managing sickle cell disease have shown encouraging results, but there are difficulties in generalizing these approaches (Anie & Green, 2002). We developed a structured self-help manual[1] to assist a CBT programme for patients with SCD, which comprises patient education (including pain control and general management), cognitive therapy, behaviour therapy including communication skills training, and relaxation. We report the outcome of a brief individual intervention by assessing changes in pain, health service utilization, psychological coping, health-related quality of life, and mood following manual-assisted CBT.

Methods

Patients and procedures

Thirty-five adult patients with SCD (aged 18 years and over) attending Central Middlesex Hospital were consecutively invited during their routine outpatient clinic appointment. Two psychologists aimed to offer six manual-assisted individual CBT sessions of one hour over

[1] Website for SCD Self-Help Manual: www.sickle-psychology.com

https://doi.org/10.1017/S135246580200406X Published online by Cambridge University Press

12 weeks. The number and timing of sessions was negotiated with the patient. At the end of each session, individuals were given the corresponding section of the self-manual to take home. Those who had interruptions in their CBT programme were posted the relevant sections of the manual to use at home. Patients were supported and encouraged through frequent telephone contact. We planned to assess patients at baseline and immediately at the end of 12 weeks. Patients who were not assessed after 12 weeks were classified as ''dropouts''.

Outcome measures

Pain status and health service utilization. Pain and health service utilization were assessed using a Pain Interview (Anie et al., 2002) that records patients' reports of frequency, intensity and duration of painful episodes and health care utilization within a 12-month period preceding the interview. Visits to the Accident and Emergency department, the number and duration of hospital admissions, and consultations with general practitioners for pain were ascertained.

Psychological coping. The Coping Strategies Questionnaire revised for SCD (CSQ-SCD – Gil, Abrams, Phillips, & Keefe, 1989) was used to assess behavioural responses and cognitive strategies used by patients to manage and control sickle cell pain. The questionnaire covers: Diverting attention, Reinterpreting pain sensations, Calming self-statements, Ignoring pain sensations, Praying and hoping, Catastrophizing, and Increasing activity level; together with six scales specifically relevant to SCD: Fear self-statements, Anger self-statements, Isolation, Taking fluids, Resting, and Heat/cold/massage. Patients rated their use of each response from 0 (never) to 6 (always), and scores were averaged to produce a mean for each scale.

Quality of life. Health related quality of life was measured with the Medical Outcomes Survey Short Form 36 (SF-36 – Ware & Sherbourne, 1992). The SF-36 is a generic measure of health concepts relevant to functional status and well-being, and it is not age, disease or treatment specific. Scores on each of the eight SF-36 dimensions were obtained with the aid of the scoring algorithm (Jenkinson, Wright, & Coulter, 1993), transforming raw scores into scales from 0 (worst possible health state) to 100 (best possible health state).

Mood. Anxiety and depression was measured on the Hospital Anxiety and Depression Scale (HADS – Zigmond & Snaith, 1983). The HADS consists of 2 subscales each with 7 items for measuring anxiety, and 7 items for depression rated from 0–3. Anxiety and depression scores were obtained by adding up the ratings of the items on each subscale. A score above 8 in any of the subscales indicates clinically significant anxiety or depression.

Medical records. Medical records of patients were examined for complications associated with SCD (e.g. chest syndromes, leg ulcers) steady state haemoglobin (Hb) level recorded as grams per decilitre (g/dl) over the 12 months preceding CBT. These are markers of disease severity.

Results

Thirty-five patients in the CBT programme were treated a mean of 4.3 (*SD* 2.8) sessions. Fourteen (40%) of 35 patients assessed at baseline were classified as dropouts. Each participant received a complete copy of the self-hep manual within the 12-week period.

Demographic and clinical characteristics

Table 1 shows the characteristics of dropouts versus those who completed CBT. There were no significant differences between the two groups on demographic factors or the two markers of disease severity (HB level/number of complications).

Pain status and health service utilization

No significant differences were reported in measures of pain and health service utilization before and after CBT (Table 2). Also, there were no significant differences between patients who completed CBT and dropouts on pain parameters or health service utilization indices in the 12-months prior to CBT (Table 2).

Psychological coping: baseline and post CBT

Pain coping strategies were collapsed into three main categories consistent with previous factor analyses (Anie et al., 2002). 1. Active coping – strategies such as increasing activity and diverting attention. 2. Affective coping – emotional responses such as catastrophic thoughts, fear and anger. 3. Passive adherence coping – adherence to methods, such as resting, and fluid intake, commonly recommended by clinicians. Affective coping is inappropriate, passive adherence coping is usually helpful for medical reasons, but active coping is psychologically most important.

Table 1. Baseline demographic and clinical characteristics: CBT and dropout patients

Variable	CBT patients (n = 21)	Dropouts (n = 14)
	Means (standard deviations)	
Age	30.1 (7.8)	31.2 (12.5)
Haemoglobin level (g/dl)	9.9 (2.4)	8.2 (0.6)
Complications	1.4 (1.4)	1.4 (1.4)
No of CBT sessions	5.5 (2.7)	2.6 (2.0)*
	n	*n*
Gender		
Female	15 (71%)	12 (86%)
Male	6 (29%)	2 (14%)
Phenotype		
SS	12 (57%)	9 (64%)
SC	6 (29%)	2 (14%)
S Beta-Thalassaemia	3 (14%)	2 (14%)
Ethnic origin		
African	10 (48%)	10 (72%)
Caribbean	9 (43%)	2 (14%)
Indian	0 (0%)	1 (7%)
Other	2 (9%)	1 (7%)

*Significant difference from dropout patients (p<.01).

https://doi.org/10.1017/S135246580200406X Published online by Cambridge University Press

Table 2. Baseline pain and health service utilization indices: comparison between CBT and dropout patients

	Pre CBT (n = 21)	Post CBT (n = 21)	Dropouts (n = 14)
	Means (standard deviations)		
Number of pain episodes (12 months)	4.3 (5.3)†	2.7 (0.6)†	3.0 (2.3)
Duration of pain episodes (Hours)	114.7 (112.4)	90.0 (56.7)	108.9 (92.0)
Pain intensity rating	6.8 (3.2)	6.5 (1.3)	7.3 (2.8)
Accident & Emergency visits (12 months)	2.2 (2.6)	1.2 (1.3)	2.4 (2.7)
Hospital admissions (12 months)	2.0 (2.7)	1.0 (1.4)	1.9 (2.3)
Duration of admissions (Days)	3.7 (4.3)	2.6 (3.0)	4.9 (3.6)
General Practitioner visits (12 months)	3.1 (5.6)	2.4 (5.4)	4.6 (7.9)

†n=20

Significant differences were shown between the pain coping responses used before and after CBT (Table 3). There was a decrease in affective coping (t = 2.12, df = 20, p = .048), and an increase in both passive adherence coping (t = 3.06, df = 20, p = .006) and active coping (t = 2.67, df = 20, p = .015) indicating that CBT improves the ability of sickle cell patients to cope appropriately with their pain.

Quality of life: baseline and post CBT

Table 3 also shows overall quality of life improvements in the sickle cell patients after CBT. Significant improvements in general health (t = 3.41, df = 18, p = .003) and vitality (t = 2.15, df = 17, p = .046) are apparent over baseline.

Anxiety and depression: baseline and post CBT

Anxiety scores were significantly reduced in patients who completed CBT (t = 2.26, df = 18, p = .036). However, the change in their depression scores was not significant (Table 3).

Discussion

The ability to engage patients in any psychological treatment programme is crucial. Sickle cell patients who dropped out of treatment are thus of particular interest, since the CBT programme as provided did not meet their needs. All dropouts received a complete manual by the end of their allocated 12-week treatment period. Analyses of prospective data on outcome measures in all the patients are currently underway. We found no significant differ-

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Table 3. Average scores for coping, quality of life, and mood scales: pre and post CBT

Scale	Pre CBT $n = 21$ Means (standard deviations)	Post CBT $n = 21$
Psychological coping (CSQ-SCD)		
Active coping	2.1 (0.8)	2.8 (1.3)*
Affective coping	2.9 (1.1)	2.5 (1.0)*
Passive adherence coping	3.6 (0.8)	4.2 (0.8)◆
Quality of life (SF-36)		
Physical functioning	62.4 (28.7)	67.9 (25.8)
Role limitations due to physical problems	47.2 (43.6)	69.4 (38.9)
Role limitations due to emotional problems	51.8 (41.6)	66.7 (36.2)
Social functioning	66.1 (27.3)	76.1 (21.1)
Mental health	65.9 (23.6)	77.3 (15.6)
Vitality	47.8 (22.4)	58.3 (19.8)*
Bodily pain	55.0 (31.3)	66.1 (25.1)
General health perceptions	41.8 (22.0)	58.3 (19.8)◆
Mood (HADS)		
Anxiety	7.9 (3.0)	5.5 (4.2)*
Depression	6.0 (4.7)	3.8 (4.0)

◆ Significant difference from Post CBT Scores ($p < .01$).
* Significant difference from Post CBT Scores ($p < .05$).

ences between patients who completed the programme as compared to dropouts on demographic and clinical characteristics, pain status or health service utilization at baseline. This suggests that those who completed the programme are representative of our patient population.

None of the patients identified the treatment procedure itself as leading to attrition. Nevertheless, patients may be less likely to identify such a cause to avoid upsetting those they perceive as trying to help them, or may have future contact with. Reasons given for attrition included severe pain, tiredness, cold weather (which increases pain frequency and severity), and being busy with domestic, work or social activities.

The manual-assisted approach was inexpensive in terms of psychologists' time utilized, an average of about 6 hours per patient. Although there appeared to be some reductions in SCD patients' report of pain experience and health service utilization, these were not statistically significant. Given the short period of intervention, this was not surprising. Nonetheless, compared with baseline there were worthwhile changes at 12 weeks. A reduction in the use of inappropriate affective coping responses for pain such as catastrophic thoughts, fear and anger was indicated. Utilization of psychological active coping strategies including distraction and behavioural activity, together with physician advised (passive adherence) methods of fluid intake and massage increased. Impairments in various aspects of quality of life showed changes with significantly better general health perceptions and energy/vitality following therapy. The assessment period was relatively short; a longer time scale may be required for significant changes to be made in other areas such as physical and social func-

https://doi.org/10.1017/S135246580200406X Published online by Cambridge University Press

tioning. It may be the case that limitations on the everyday lives of patients with sickle cell disease are not due to the incidence of symptomatic episodes, but may be the outcome of broader appraisals of their illness over time.

SCD causes much distress to patients. Mood is usually affected and patients reported reduced levels of anxiety, but not depression following CBT. Anxiety was of clinical importance before intervention, hence it is perhaps not surprising that CBT had an effect while depression was unaffected because it was not initially a clinical problem. Clinical levels of both anxiety and depression have been reported in earlier studies (Alao & Cooley, 2001), attesting that possible clinical differences between patient populations should be noted.

This brief CBT intervention using a self-help manual was therefore associated with improvements in pain coping strategies, quality of life, and mood. We must be cautious in attributing these changes to particular elements of the self-help manual and treatment programme. Teasing out the effective elements will require a well-designed randomized control trial. Previous studies have not provided sufficient evidence to demonstrate the efficacy of specific psychological interventions for sickle cell patients, because of their poor quality (Anie & Green, 2002). Nonetheless, this case series evaluation demonstrates the feasibility and applicability of a brief inexpensive manual-assisted treatment programme in SCD. Such programmes might readily be developed for other chronic diseases and show promise for delivering a service to the majority of patients for whom specialist service input is not currently easily accessible. However, the long term cost effectiveness of this approach in terms of patients' appropriate utilization of health services should be determined.

Acknowledgements

We are grateful to the Department of Health National Ethnic Health Projects Initiative (NT 65) for funding the development of the Self-Help Manual. We are also very thankful to all sickle cell patients attending Central Middlesex Hospital, Orla Cummins for data collection, and staff at the Brent Sickle Cell and Thalassaemia Centre. There are no conflicts of interest.

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