Journal of Psychosomatic Research 94 (2017) 73–81

Contents lists available at ScienceDirect

Journal of Psychosomatic Research

Long-term economic evaluation of cognitive-behavioural group treatment versus enhanced usual care for functional somatic syndromes☆,☆☆

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ARTICLE INFO

Article history:
Received 23 June 2016
Received in revised form 5 January 2017
Accepted 7 January 2017

Keywords:
Economic evaluation
Cost-effectiveness
Cognitive-behavioural therapy
Functional somatic syndromes
Bodily distress syndrome
Fibromyalgia
Chronic fatigue syndrome

ABSTRACT

Objective: Patients with functional somatic syndromes (FSS) such as fibromyalgia and chronic fatigue syndrome have a poor outcome and can incur high healthcare and societal costs. We aimed to compare the medium-term (16 months) cost-effectiveness and the long-term (40 months) economic outcomes of a bespoke cognitive-behavioural group treatment (STreSS) with that of enhanced usual care (EUC).
Methods: We obtained complete data on healthcare and indirect costs (i.e. labour marked-related and health-related benefits) from public registries for 120 participants from a randomised controlled trial. Costs were calculated as per capita public expenses in 2010 €. QALYs gained were estimated from the SF-6D. We conducted a medium-term cost-effectiveness analysis and a long-term cost-minimization analysis from both a healthcare (i.e. direct cost) and a societal (i.e. total cost) perspective.
Results: In the medium term, the probability that STreSS was cost-effective at thresholds of 25,000 to 35,000 € per QALY was 93–95% from a healthcare perspective, but only 50–55% from a societal perspective. In the long term, however, STreSS was associated with increasing savings in indirect costs, mainly due to a greater number of patients self-supporting. When combined with stable long-term reductions in healthcare expenditures, there were total cost savings of 7184 € (95% CI 2271 to 12,096, $p = 0.004$) during the third year after treatment.
Conclusion: STreSS treatment costs an average of 1545 €. This cost was more than offset by subsequent savings in direct and indirect costs. Implementation could both improve patient outcomes and reduce costs.

1. Background

Functional somatic syndromes (FSS) such as fibromyalgia, irritable bowel and chronic fatigue syndrome are a major public health issue. FSS are prevalent worldwide in all medical settings, and when severe pose a major burden on sufferers, health services, and on society. They incur considerable direct and even greater indirect costs [1–7]. The direct costs mainly reflect repeated referrals to secondary medical care in order to exclude physical disease [5,8]. Indirect costs are consequences of reduced productivity at work, sick leave, dependence on social benefits and, in the most severe cases, a permanent loss of the ability to support oneself [6,8–11]. While psychological treatments such as cognitive-behavioural therapy (CBT) may reduce symptom severity and improve health-related quality of life in patients with various FSS [12–16], knowledge about their long-term effects on direct and indirect costs in these patients is limited [8,17].

Potentially effective psychological treatments for FSS are currently not routinely delivered, even in severe cases, because of organisational and other barriers [18,19]. We have addressed these barriers with a group CBT programme (Specialised Treatment for Severe Bodily Distress Syndromes, STreSS) designed as a common treatment for patients with a range of severe and impairing FSS, and suitable for delivery in a general University hospital setting [20]. In a recent trial, we found STreSS to be superior to enhanced usual care (EUC) both on the primary outcome (self-rated physical health) and also on most secondary outcomes, including somatic symptoms, illness worrying and social functioning [21]. The specific FSS diagnosis had no differential effect on treatment response [19].

☆ **Author contribution:** AS, MS and PF are co-investigators of the STreSS-1 trial. AS, EØ and PF conceived and designed the study. AS and EØ obtained and combined data from public registries and calculated individual direct and indirect costs. EØ and JSJ conducted statistical analysis. AS drafted the paper. All authors participated in the interpretation of the findings, were involved in critically revising the paper and approved the final manuscript.
☆☆ **Conflict of interest:** All authors have completed the Unified Competing Interest form at http://www.icmje.org/coi_disclosure.pdf. The authors have no competing interests to report.

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http://dx.doi.org/10.1016/j.jpsychores.2017.01.005
0022-3999/

In our trial, patients were sampled using the severe multi-organ subtype of the newly proposed diagnosis bodily distress syndrome (BDS), the criteria for which have been included in the current draft of the World Health Organization's International Classification of Diseases, 11th Revision, with some adaptations [22]. Recent studies have confirmed the high healthcare costs and unfavourable prognosis of untreated multi-organ BDS, especially as regards a high risk of new disability pension awards [23,24]. Although the trial results show the effectiveness of STreSS, the long-term economic consequence of the treatment is not known.

We therefore conducted a long-term economic evaluation. We did this within the context of the Danish healthcare and welfare system, which is tax-financed, and where access to both direct (i.e. healthcare expenditures) and indirect (i.e. public expenditures associated with occupational status and social benefits) cost data is possible through public registries [25–29]. This allowed us to do analyses from both a healthcare (direct costs) and a societal (total costs, i.e. direct plus indirect) perspective.

The aims of this study were to: (1) compare healthcare and total costs during and following STreSS and EUC up to 40 months after randomisation, and (2) compare the cost-effectiveness of STreSS and EUC in terms of QALYs gained and percentage of patients achieving clinically significant improvement for the trial period of 16 months.

2. Methods

2.1. Study design and participants

The STreSS-1 trial (clinicaltrial.gov NCT00132197) was a two-arm, single-site, non-blinded, randomised controlled trial comparing a group CBT programme (STreSS) with usual care enhanced by a thorough clinical assessment (EUC) [20,21]. The trial was conducted at Aarhus University Hospital, Denmark from 2005 to 2008 within a general hospital setting. Most patients were referred by their primary care physician. Referred patients were included in the trial if they fulfilled criteria for the severe multi-organ subtype of bodily distress syndrome [30]. This unifying definition captures both patients with severe FSS diagnoses and also most patients with somatoform disorders [31]. Other inclusion criteria were: age 20–45 years and multiple symptoms for at least 2 years. Exclusion criteria have been reported previously [21].

A total of 54 patients were randomly assigned to STreSS and 66 to EUC since unequal patient attrition had been expected (but not observed). Self-report data were obtained immediately prior to randomisation and 4, 10 and 16 months after randomisation. Direct and indirect cost data were obtained from Danish registers for 12 months before and up to 40 months after randomisation.

2.2. Interventions

2.2.1. Enhanced usual care

Usual care was delivered by patients' primary care physicians and various specialists. There was no restriction on the psychological or pharmacological interventions that could be given to these patients, or on new referrals to secondary care services. Usual care was 'enhanced' by a thorough clinical assessment prior to randomisation that aimed to achieve a shift from diagnostic procedures to the management of somatic symptoms and comorbid mental illness. Details of the assessment are reported elsewhere [32,33].

2.2.2. STreSS

Patients allocated to STreSS received the same assessment as patients in the EUC group. Additionally, they received nine modules of manualised group CBT, each of 3.5 h duration and delivered to groups of nine patients by two psychiatrists over a 4-month period. Details about the STreSS treatment modules have been reported previously [18,20,21]. The STreSS treatment contained no module or specific interventions regarding patients' occupational situation, but individual goals to enhance one's work ability or solve specific problems at the working place could be set by participants. The STreSS treatment manual is freely available at www.functionaldisorders.dk.

As previously reported, 83% of allocated patients completed STreSS, while 11% did not receive any treatment module. Only 3 patients (6%) discontinued treatment. We did not find any differences regarding other psychological or psychiatric treatment between the EUC and the STreSS group [21].

2.3. Outcomes

2.3.1. QALYs and clinical improvement

Quality-adjusted life years (QALYs) were generated on the basis of eleven items of the 36-item Short Form Health Survey (SF-36) [34] converted into SF-6D utility scores based on weights of the general UK population according to the method of Brazier [35]. The accrual of QALYs during the 16 months after randomisation was calculated using the area under the curve, assuming a linear change between each available time point (0, 4, 10 and 16 months after randomisation).

In order to add a condition-specific evaluation of cost-effectiveness, we calculated costs per patient achieving clinically significant improvement from baseline to 16 months on two different measures: 1) Self-rated physical health (primary trial outcome), assessed with an aggregate score of the SF-36 scales physical functioning, bodily pain and vitality [36], and 2) distressing somatic symptoms (secondary outcome) measured with the SCL-90 R somatisation subscale [21]. Clinically significant improvement was defined conventionally as a 0.5 SD change [37], equalling 4 points increase on the SF-36 aggregate score and 0.35 points reduction on the SCL-90 R somatisation subscale.

Questionnaire data (SF-6D utilities, physical health and somatic symptoms) were available for all 120 patients at baseline and for 105, 96 and 94 patients at 4, 10 and 16 months, respectively [21].

2.3.2. Healthcare (i.e. direct) costs

Denmark runs a nationwide centralized register of personal information, the Civil Registration System, for which purpose every citizen is given a unique personal identification number. All public registries in Denmark use this unique number, which allows linkage of registers and of trial data to register data. Data on healthcare costs for each trial participant were obtained from four national health registers [25–28]. The cost data in these registries are based on DRG codes (i.e. average costs for specific procedures or hospital stays) for in-patient and out-patient treatment in Danish hospitals, on actual reimbursement for primary care and medical specialists, and on public expenses for prescription medication.

In a first step, all healthcare costs were calculated separately for each patient and each calendar month within each registry and inflated to 2010 prices. In a second step, these costs were allocated to study months (i.e. months before or after a patient's randomisation date), summed up across the four registers, and collapsed into the following sectors and domains: (1) primary care, covering family physicians including primary care based physiotherapists and chiropractors, (2) inpatient and outpatient general hospital care, covering both hospital based care and medical specialists, (3) inpatient and outpatient mental healthcare, covering both hospital-based care and psychiatrists and psychologists, and (4) medication. The specific costs for assessment and treatment within the trial for both EUC and STreSS were obtained from the same registries, and added under the domain outpatient mental healthcare costs. In a third step, costs within each domain and sector were summed to create annual costs with exception of the trial's 4-month treatment period for which costs were calculated separately. For the third year after treatment, only costs for medication and primary care were available when we obtained data, and estimated annual healthcare costs for the third year are therefore not comparable with the preceding years. For each domain, the percentage of patients with