Can pain-related fear be reduced? The application of cognitive-behavioural exposure in vivo

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Although cognitive-behavioural treatments of patients with chronic pain generally are reported to be effective, customization might increase their effectiveness. One possible way to customize treatment is to focus the intervention on the supposed mechanism underlying the transition from acute to chronic pain disability. Evidence is accumulating in support of the conjecture that pain-related fear and associated avoidance behaviours are crucial in the development and maintenance of chronic pain disability. It seems timely to apply this knowledge to the cognitive-behavioural management of chronic pain. Two studies are presented here. Study 1 concerns a secondary analysis of data gathered in a clinical trial that was aimed at the examination of the supplementary value of coping skills training when added to an operant-behavioural treatment in patients with chronic back pain. The results show that, compared with a waiting list control, an operant-behavioural treatment with or without pain-coping skills training produced very modest and clinically negligible decreases in pain-related fear. Study 2 presents the effects of more systematic exposure in vivo treatment with behavioural experiments in two single patients reporting substantial pain-related fear. Randomization tests for AB designs revealed dramatic changes in pain-related fear and pain catastrophizing. In both cases, pain intensity also decreased significantly, but at a slower pace. Differences before and after treatment revealed clinically significant improvements in pain vigilance and pain disability.

Key Words: *Back pain; Cognitive-behavioural treatment; Exposure; Fear*

Est-il possible de réduire la peur reliée à la douleur ? L'application de l'exposition cognitivocomportementale in vivo

RÉSUMÉ : Bien que les traitements cognitivocomportementaux des patients souffrant de douleurs chroniques sont généralement déclarés efficaces, la personnalisation accroîtrait peut-être l'étendue de leurs effets. Un moyen possible de personnaliser le traitement consiste à axer l'intervention sur le mécanisme qu'on suppose sous-jacent à la transition de la

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douleur aiguë à l'incapacité par douleur chronique. Les observations s'accumulent à l'appui de la conjecture selon laquelle la peur reliée à la douleur et les comportements d'évitement connexes sont essentiels pour que se développe et que persiste une incapacité par douleur chronique. Le temps semble venu d'appliquer cette constatation à la prise en charge cognitivo-comportementale de la douleur chronique. Deux études sont présentées dans le présent article. La première porte sur une analyse secondaire de données colligées dans un essai clinique visant à examiner la valeur complémentaire de la formation aux habiletés d'adaptation ajoutée à un traitement par comportement opérant chez des patients souffrant de douleurs dorsales chroniques. Les résultats démontrent que, par rapport à un contrôle par des listes d'attente, un traitement par comportement opérant accompagné ou non de formation aux habiletés d'adaptation à la douleur a entraîné une diminution très modeste et cliniquement négligeable de la peur reliée à la douleur. La deuxième étude présente les effets d'un traitement par exposition in vivo plus systématique à des expériences comportementales chez deux patients célibataires faisant état d'une importante peur reliée à la douleur. Des essais aléatoires de type AB ont révélé des modifications marquées de la peur reliée à la douleur et de la catastrophisation causée par la douleur. Dans les deux cas, l'intensité de la douleur diminuait aussi de manière significative, mais à un rythme plus lent. Les différences avant et après le traitement ont révélé des améliorations importantes d'un point de vue clinique, en matière de vigilance à la douleur et d'incapacité causée par la douleur.

Chronic musculoskeletal pain syndromes may disrupt the lives of those affected and are responsible for considerable costs to health care and society (1). When traditional and biomedical treatments are unsuccessful and disability due to back pain grows, patients with chronic pain are often referred to rehabilitation centres for interdisciplinary treatment. These centres often adopt a biopsychosocial or behavioural rehabilitation model that assumes that disability is not only determined by the underlying pathology, but also, and perhaps more, by social, cognitive, emotional and behavioural factors (2). Operant conditioning principles, psychophysiological concepts and concepts from cognitive psychology are applied to both the assessment and treatment of chronic pain (3). Operant-behavioural treatments aim to increase activity levels and decrease pain behaviours by changing behaviour consequences. Activity levels are increased using time-contingent quota systems, including baseline determination, treatment contract, positive self-reinforcement for activity increments, and workplace or home visits to generalize new behaviour in daily life environments (4). Respondent treatments, often using applied relaxation and biofeedback, are developed to decrease sympathetic arousal levels and to apply relaxation responses in personally stressful situations (5). In the literature, the term 'cognitive treatment' in patients with pain is used for divergent forms of treatment, such as education, hypnosis, challenging maladaptive beliefs and cognitive pain-coping skills training. Cognitive treatment is often used in combination with other forms of therapy (6), and can address either the reduction of pain (7) or the reduction of stress (8). Most of these treatment programs are offered in combination or as adjuncts to physical approaches (6). Typically, the basic goal of these approaches is to increase quality of life (ie, to care for the sufferer), rather than remove the pain (ie, to cure the pain).

Several reviews and meta-analyses concluded that cognitive-behavioural treatment programs are more effective than no treatment at all, waiting list and monodisciplinary treatments (9). More precisely, cognitive-behavioural treatments produced significantly greater changes in pain experience, cognitive coping and appraisal, and reduced behavioural expression of pain (10). Despite these advances, there continues to be a substantial proportion of patients who do not seem to benefit from available interventions. As a result, pain researchers have argued in favour of matching treatments to patient characteristics to increase the effectiveness of treatments (11). Customization is not an easy task because there is very little empirical evidence for so-called 'Aptitude × Treatment' interactions in chronic pain research. To develop effective customized treatments, theoretically derived, a priori explanations as to why particular interactions occur need to be developed (12).

One of the recent developments in behavioural pain research concerns the mechanisms by which patients with acute pain become chronically disabled. The early notion that the lowered ability to accomplish tasks of daily living in patients with chronic pain is merely the consequence of pain severity has now been reconsidered. Indeed, a steadily increasing number of studies are showing that observable physical performance and self-reported disability levels in patients with subacute and chronic pain are associated with cognitive and behavioural, rather than sensory and biomedical, aspects of pain (13-15). Fear-avoidance models have been proposed, and an increasing number of studies have successfully tested their major assumptions (15-19). A number of studies have reported that pain-related fear is one of the strongest predictors of variation in physical performance in terms of spinal isometric strength measured by a lumbar extension machine (20), lifting capacity (17,21,22), and trunk and leg flexion and extension (22,23). Avoidance of daily activities may result in functional disability (18,22,24) and the so-called 'disuse syndrome' (25), in terms of both physical deconditioning (26) and guarded movements (27). Avoidance also means the withdrawal from essential reinforcers, leading to mood disturbances, such as irritability, frustration and depression. Both depression and disuse are known to be associated with decreased pain tolerance (28,29) and, hence, might promote the painful experience. Prospective studies have indeed revealed that, in patients with acute back pain, pain-related fear is predictive of future disability and work status (30-32).

If pain-related fear is indeed one of the important mechanisms responsible for the development and maintenance of chronic pain disability, we must be able to show that decreasing these fears leads to decreased disability. Current treatments of excessive fears and anxiety are based

on the experimental systematic desensitization work of Wolpe (33). In this keystone treatment method, individuals progress through increasingly more anxiety-provoking encounters with phobic stimuli while using relaxation as a reciprocal inhibitor of rising anxiety. Because relaxation was intended to compete with the anxiety response, a graded format was chosen to keep anxiety levels as weak as possible. Later studies revealed that exposure to the feared stimuli appeared to be an essential component of the systematic desensitization, and when applied without relaxation produced comparable effects (34). Which of the currently available pain management programs would be good candidates for fear reduction through exposure to feared stimuli? Although operant-behavioural treatments (sometimes referred to as graded activity [35]) were initially designed to modify contingencies between pain behaviours and their direct consequences, they also provide ample exposure to physical movements. Surprisingly, no studies have specifically examined the effects of operant-behavioural treatment on pain-related fear.

On the other hand, operant-behavioural methods may not be specific enough. Philips (36) was one of the first to argue for the systematic application of graded exposure in vivo to produce disconfirmations of expected consequences (pain, reinjury) of physical activity. Experimental support was provided by Crombez et al (23) in a sample of patients with chronic low back pain who were requested to perform four exercise bouts (two with each leg) at maximal force. During each exercise bout, the baseline pain, the expected pain and experienced pain were recorded. As predicted, the patients with chronic low back pain initially overpredicted pain, but after repetition of the exercise bout, the overprediction was readily corrected. The clinical applications of exposure in vivo have been reported more recently. Using a replicated single-case experimental design, Vlaeyen et al (37) provided preliminary evidence showing that a tailored graded exposure in vivo is superior to a no-treatment baseline period and a graded activity. Only patients who reported substantial fear of movement/(re)injury (Tampa Scale for Kinesiophobia [TSK] score greater than 40) and who were referred for outpatient behavioural rehabilitation were included. After a no-treatment baseline measurement period, the patients were randomly assigned to one of two interventions. In intervention A, patients received the exposure first, followed by graded activity. In intervention B, the sequence of treatment modules was reversed. Daily measures of pain-related cognitions and fears were recorded with visual analogue scales. The following measures were taken before and after treatment: pain-related fear, pain catastrophizing, pain control and pain disability. Although dramatic improvements were observed when the graded exposure in vivo was introduced, both the exposure and the graded activity were imbedded in a comprehensive behavioural rehabilitation program provided by an interdisciplinary treatment staff, following the operant treatment principles, and including pacing techniques, relaxation and group education about ergonomics. This may have caused contamination in the sense that ingredients of the background program may have selectively moderated the effectiveness of the exposure in vivo.

The present paper consists of two studies focusing on different approaches that have the potential to reduce pain-related fear in patients with chronic low back pain. The first study presents a secondary analysis of data on pain-related fear based on a randomized, clinical trial that evaluated the supplementary value of cognitive coping strategies when added to an operant graded activity approach. The second study is a replicated single-case study in which the novel-exposure, in vivo treatment is applied in two patients reporting substantial fear of movement/(re)injury.

STUDY 1

The present study is a secondary analysis based on an attention-controlled, randomized, clinical trial aimed at examining the supplementary value of a cognitive program with relaxation when added to an operant treatment for patients with chronic low back pain (7). After completing the pre-treatment assessment, patients received operant treatment with cognitive coping skills training (OPCO) or operant treatment with a group discussion (OPDI), or they were assigned to the waiting list control condition (WLC). Compared with patients in the WLC, patients receiving either OPCO or OPDI reported less negative affect, higher activity tolerance, less pain behaviour, and higher pain coping and pain control. Although pain-related fear measures were included in the study, they were not reported, given that the intervention was not primarily aimed at modifying pain-related fear. The aim of the current secondary analysis is to test whether both interventions, which included a graded activity approach as described by Fordyce (4), also produced reductions in pain-related fear by using available pain-related fear measures.

Subjects

Of the 148 patients who participated in the three conditions, valid pre- and post-treatment data on the TSK were available for 114 patients (OPCO 45, OPDI 45, WLC 24). The sample consisted of 43 men and 71 women with a mean age of 39.8 years (SD=8.8, range 21 to 64 years), who had a mean pain duration of 9.5 years (SD=8.2, range 10 months to 39 years). Of the total sample, 87% received financial disability compensation, with a mean duration of 3.6 years (SD=4.6, range 0.4 to 28 years).

Treatments

The operant-behavioural treatment, based on program descriptions by Fordyce (4) and Roberts (38), was aimed at increasing healthy behaviours and decreasing pain behaviours. It was provided by the whole rehabilitation team using a treatment protocol manual. A part of the operant-behavioural treatment was spouse group training, consisting of seven 90 min weekly sessions. Baseline levels of activities and pain behaviours were registered during the first two

weeks. Patients were asked to engage in activities until pain or other physical discomfort prevented them from continuing. Subsequently, a treatment contract was made with patients in which the concrete goals and quota were registered. Patients agreed to follow the quota according to the activity-rest contingency principle. If necessary, medication use was managed in a time-contingent fashion. Throughout the treatment program, physical therapists provided 50 h of individual treatment (with the strict exclusion of passive treatment modalities) and 38 h of group treatment. Occupational therapy consisted of 12 h of individual contact and 26 h of group training. Patients were taught to increase their sitting and standing tolerance, and developed a daily activity schedule, according to the operant principles, to be used at home. In weekly 15 min meetings between patients and the rehabilitation team, progress was discussed and ample reinforcement was provided. The psychologist saw patients individually in weekly 30 min sessions.

The cognitive coping skills training was aimed at increasing sense of pain control through self-management. The program consisted of 12 group sessions each of 90 min duration according to a treatment manual given by a skilled behaviour therapist. It consisted of three phases: a reconceptualization phase, a skills acquisition phase and a generalization phase. The goal of the reconceptualization phase was to modify the pain experience in terms that imply self-control and resourcefulness. Patients learned that 'hurt' does not necessarily mean 'harm' and that pain is influenced by multiple factors. In the skills acquisition phase, patients practised two types of imagery: imaginative transformation of the pain sensation and pain-incompatible sensory imagery. These techniques were drawn from Diamond (39) and Fernandez (40). In addition, applied relaxation (41) was used to teach patients to relax and to use relaxation in situations reported as personally stressful. Electromyography (EMG) biofeedback was used to help patients recognize changes in tension and relaxation. Each session ended with homework assignments consisting of texts and tapes with the imagery or relaxation exercises. To compare with the cognitive treatment, a group discussion program was developed as an attention control during which patients were requested to read parts of a book about pain, and to share information and thoughts with the other group members. In addition, participants listened to audio taped music fragments.

Statistical analyses

Normality was tested with the Kolmogorov-Smirnov test. To test the effects of the treatment on changes in the TSK, ANCOVA with post-TSK score as the dependent variable, pre-TSK score as the covariate and group as the independent variable (OPCO, OPDI and WLC) was carried out with the reversed Helmert contrast for groups.

Results

Both pre- and post-TSK scores were normally distributed (Kolmogorov-Smirnov Z=0.684 and 0.804 respectively, P>0.50). Table 1 displays means and standard deviations of the TSK for the three groups before and after measurement. Reductions in pain-related fear were rather modest, with the largest reduction in the OPDI group. For the WLC group, a slight increase was evident. ANCOVA revealed no significant overall group effect (F=2.87, P=0.061), but the second reversed Helmert contrast (mean of both treatment conditions versus the WLC) was significant (P=0.045), with an overall difference of 2.5. Based on a large data pool, the mean post-treatment TSK score corresponds with the 30th percentile (42). Even when patients with an elevated score are selected (TSK greater than 39), the same pattern occurs. In this subgroup, the TSK score drops from a mean score of 44.7 to 41.4 (corresponding with the 70th percentile). Again, only the second reversed Helmert contrast was significant (P=0.043).

TABLE 1
Mean (SD) pre- and post-treatment Tampa Scale for Kinesiophobia scores for patients in three conditions

	Pre-treatment	Post-treatment
OPCO	38.7 (8.0)	37.0 (7.3)
OPDI	38.6 (6.7)	35.5 (6.4)
WLC	37.8 (8.0)	38.3 (8.2)

OPCO Operant-cognitive treatment; OPDI Operant-discussion treatment; WLC Waiting list condition

Discussion

The results of the present study suggest that a graded activity approach with or without pain coping skills training may reduce pain-related fear, and fear of movement/(re)injury in particular. However, the reductions are quite modest. What might be the reason for this? A plausible explanation is that the operant treatment and graded activity program, although not aimed at reducing pain-related fears, allow too many escape possibilities for patients. In addition, it is not clear whether patients are exposed to the situations that they actually fear the most. Philips (36) was one of the first to argue for the systematic application of graded exposure to produce disconfirmations among expectations of pain and harm, the actual pain, and the other consequences of the activity. She further suggested:

> *These disconfirmations can be made more obvious to the sufferer by helping to clarify the expectations he/she is working with, and by delineating the conditions or stimuli which he feels are likely to fulfill his expectations. Repeated, graded, and controlled exposures to such situations under optimal conditions are likely to produce the largest and most powerful disconfirmations (36)*

In general, graded exposure in vivo may appear to be quite similar to the usual graded activity programs (35,43), in that it gradually increases activity levels despite pain. However, both conceptually and practically, exposure in vivo is quite different from graded activity. First, graded

activity is based on instrumental learning principles, and selected health behaviours are shaped through positively reinforcing predefined quota of activities. Exposure in vivo, originally based on extinction of Pavlovian conditioning (44), is currently viewed as a cognitive process during which fear is activated, and catastrophic expectations are challenged and disconfirmed, thus resulting in reductions of the threat value of the originally fearful stimuli. Second, during graded activity, special attention goes to the identification of positive reinforcers that can be provided when the individual quotas are met, whereas graded exposure pays special attention to the establishment of an individual hierarchy of the pain-related fear stimuli. Third, graded activity programs typically include individual exercises according to functional capacity and observed individual physical work demands, while graded exposure includes activities that are selected based on the fear hierarchy and the idiosyncratic aspects of the fear stimuli. For example, if the patient fears repetitive spinal compression produced by riding a bicycle on a bumpy road, then the graded exposure should include an activity that mimics that specific activity, in that specific context, and not just a stationary bicycle.

STUDY 2

Based on the above suggestions, we decided to develop an exposure in vivo treatment consisting of three components: development of a fear hierarchy, education, and graded and repeated exposure to feared situations (2,22). The first session of the graded exposure (GEXP) always consists of unambiguously educating patients to view their pain as a common condition that can be self-managed, rather than as a serious disease or a condition that needs careful protection. Patients are also given a careful explanation of the fear-avoidance model, using their individual symptoms, beliefs and behaviours to illustrate how vicious circles (pain leads to catastrophic thought leads to fear leads to avoidance leads to disability leads to pain) maintain the pain problem. Subsequently, individually tailored practice tasks are developed based on the graded hierarchy of fear-eliciting situations. These tasks take the form of a series of behavioural tests during which irrational expectations are challenged. Further, the general principles for exposure are followed. Patients agree to perform certain activities or movements that they used to avoid. Patients are also encouraged to engage in these fearful activities as much as possible until anxiety levels decrease. The therapist, who demonstrates how the activity can be done efficiently, first models each activity or movement. Hereby the therapist takes care to convey the message that activities are only safe when performed in an ergonomically 'justified' way. Finally, the GEXP is presented as a start and patients are encouraged to continue exposing themselves to more activities in everyday life after termination of treatment. A more detailed description of the exposure in vivo treatment can be found in the *Handbook of Clinical Pain Management* (45). The effects of the exposure in vivo treatment are illustrated by two single-case studies of patients with chronic low back pain who were referred to the Department of Rehabilitation Medicine of the University Hospital Maastricht, the Netherlands.

Subject 1

Subject 1 was a 45-year-old cleaner with a one-year history of low back pain for which she received disability pension. A neurologist examined her, and a magnetic resonance imaging scan revealed degeneration of several lumbar discs. Based on this information and what she had heard from other people, she believed that her back was slowly crumbling away and that certain movements, such as lifting, bending over, and too much compression on the disc might promote the degeneration process. An epidural at L5, chiropractic, physical therapy (including massage and specific exercises) and analgesics did not produce any substantial improvement. The psychological assessment revealed that she was excessively fearful of physical movements and might benefit from an exposure in vivo treatment.

Subject 2

Subject 2 was a 47-year-old married woman who worked as a nurse, but had been on sick leave since the sudden onset of her back pain complaints 12 years ago. Her pain started during defecation on the toilet and has been present ever since. She was convinced that some structure, possibly nerves in the back, were damaged. Because no one had ever given her a plausible explanation for her complaints, she felt very insecure about what movements and activities she might or might not do. One year before undergoing the exposure in vivo, she had been discharged early from a rehabilitation program with exercise treatment because she supposedly was not "motivated enough". Because of her elevated TSK score of 43, it was decided that an exposure in vivo might be the best treatment option.

Design

For both patients, a single-case experimental AB design was chosen, consisting of a one-week baseline period followed by a five-week treatment period. The exposure in vivo treatment was carried out by a physical therapist and a movement scientist. It consisted of 15 sessions each of 90 min duration. In contrast to previous studies (37,46,47), there were no other treatment components included in the rehabilitation program.

Measurements

Manipulation check: To check whether the exposure indeed modified the fear appraisals, a short instrument was developed using 11 items representing main factors of existing questionnaires for pain-related fear and catastrophizing. These included Dutch versions of the TSK (17,48), the Pain Catastrophizing Scale (PCS [49]) and the Pain Anxiety Symptoms Scale (PASS [50]). Visual analogue scales (VAS) consisting of 10 cm horizontal lines followed the items derived from these questionnaires. These VASs

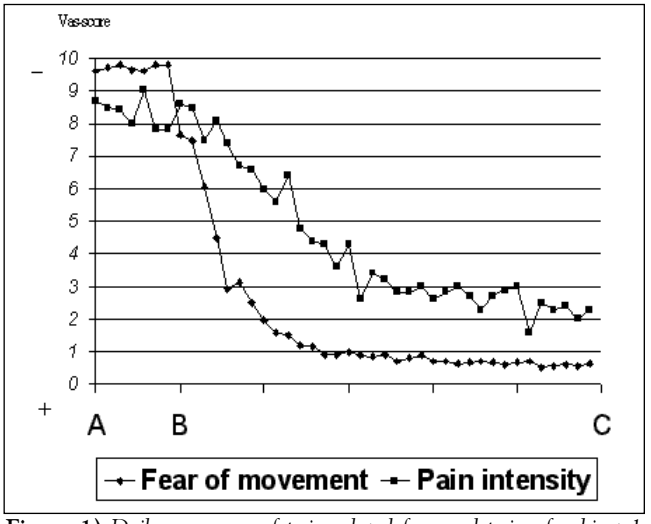

Figure 1) *Daily measures of pain-related fear and pain of subject 1 during baseline (A-B) and exposure treatment (B-C)*

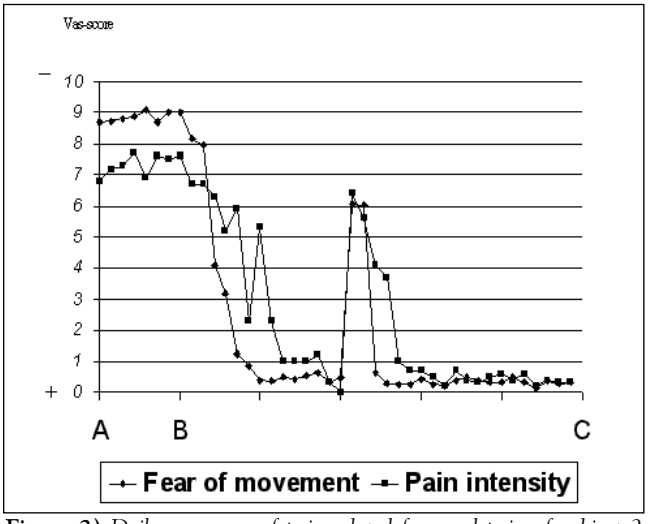

Figure 2) *Daily measures of pain-related fear and pain of subject 2 during baseline (A-B) and exposure treatment (B-C)*

were anchored at the left and right by the words "totally disagree" and "totally agree". This measure was administered on a daily basis for the study duration (42 days). The subjects were instructed to fill in the scales each morning and to send the completed form immediately to the researchers by means of prestamped envelopes. Three main scores were derived consisting of the mean scores of the items from TSK, PASS and PCS, respectively (range 0 to 10).

Pain intensity: One VAS measuring present pain intensity was added to the 11 scales that were completed daily. The scale was anchored with "no pain at all" at one extreme and "worst pain experienced" at the other.

Pain-related fear: The full TSK was taken during the initial screening, before and after the exposure in vivo treatment. In addition, the Photograph series of Daily Activities (PHODA – A CD-rom version of PHODA including the 98 pictures and a brief manual is available, and can be requested by e-mail at PHODA@hszuyd.nl [51]), a standardized method using 98 photographs representing various physical daily life activities including lifting, bending, walking, bicycling, etc, is used as a more direct measure of fear for the development of an individually tailored fear hierarchy to be used during the exposure. The patient was requested to place each photograph along a fear thermometer, consisting of a vertical line with 11 anchor points (ranging from 0 to 100) printed on a 60 × 40 cm hardboard. The fear-thermometer was placed on a table in front of each patient who was instructed to:

> *Please watch each photograph carefully, and try to imagine yourself performing the same movement. Place the photograph on the thermometer according to the extent to which you feel that this movement is harmful to your back.*

After completion of the test, each photograph was given a rating according to the position on the thermometer. A total score ranging from 0 to 100 was calculated as the sum of each rating divided by 9800 (the maximum total score). The PHODA was used to establish a hierarchy of fear stimuli, but was also repeated after the exposure in vivo treatment.

Pain disability: The Dutch version of the Roland Disability Questionnaire (RDQ, [52]) was used in this study. The RDQ is a widely used 24-item 2-point scale measuring the difficulty to perform daily activities (eg, "I only walk short distances because of my back pain").

Pain vigilance: The Pain Vigilance and Awareness Questionnaire (PVAQ, [53]) was used to assess changes in the degree to which patients focused their attention towards the pain.

Statistical analyses

Although the use of single-case experimental designs has a long history, there is still some debate concerning the analysis of the resulting data (54). At least two approaches have been described which might be particularly useful for AB designs: time series analysis and randomization tests (41,42). Because of the limited number of observations in phase A, the authors chose a randomization test using the rationale of Edgington (55). A difference of the usual application of randomization tests, however, is that the present study does not include preplanned random assignment of the intervention point but, instead, has a fixed number of baseline measurements. Nevertheless, the randomization test is still a powerful descriptive statistical tool in such cases. Given that there are 42 observations (7-A and 35-B), it could be assumed that the initiation of treatment B was randomly assigned to any of the second through 42nd observations. In this design, there are 41 possible assignments and the minimum P-value that can possibly be attained is 1/41=0.02439. However, if the number of observations in each phase is restricted to a minimum of five, there are only 33 possible assignments with a minimal P-value of 0.03030 (for the general formula and algorithms see [55]). Because treatment B is expected to be superior to baseline A, the null hypothesis that there is no differential

TABLE 2
Subject 1: P-values for the randomization tests for AB designs with a minimum of one or a minimum of five observations per phase, and with delayed effects

	Fear of movement		Pain catastrophizing		Fear of pain		Pain intensity	
Effect lag	Min 1	Min 5	Min 1	Min 5	Min 1	Min 5	Min 1	Min 5
0	0.12195	0.15152	0.14634	0.18182	0.17073	0.21212	0.21951	0.27273
1	0.07317	0.09091	0.14634	0.15152	0.12195	0.15152	0.14634	0.18182
2	0.04878	0.06061	0.07317	0.09091	0.07317	0.09091	0.12195	0.15152
3	0.02439	0.03030	0.02439	0.03030	0.02439	0.03030	0.04878	0.06061
4							0.02439	0.03030

TABLE 3
Subject 2: P-values for the randomization tests for AB designs with a minimum of one or a minimum of five observations per phase, and with delayed effects

	Fear of movement		Pain catastrophizing		Fear of pain		Pain intensity	
Effect lag	Min 1	Min 5	Min 1	Min 5	Min 1	Min 5	Min 1	Min 5
0	0.02439	0.03030	0.09756	0.12121	0.14634	0.18182	0.41463	0.51515
1			0.02439	0.03030	0.09756	0.12121	0.34146	0.39394
2					0.02439	0.03030	0.29268	0.30303
3							0.26829	0.24242
4							0.07317	0.06061
5							0.02439	0.03030

effect for any of the measurement times is tested using a randomization test on the difference between B and A. The analysis is performed using the SCRT software (Single-Case Randomization Tests, version 1.1 [56]). The program also allows the calculation of a combined P-value when both cases are considered simultaneously in a meta-analysis according to Edgington's additive method (55). Finally, the test is repeated assuming delayed effects until the minimal P-value has been reached.

Results

Because the patterns of change for fear of movement/(re)injury, pain catastrophizing and fear of pain are quite similar, Figures 1 and 2 show the observations across time for fear of movement/(re)injury and pain intensity only. Visual inspection of the data suggests that the introduction of the exposure in vivo treatment produced substantial decreases in pain-related fear and, although more slowly, in pain report. Table 2 and 3 display the P-values for the phase design randomization tests on the raw data. In concordance with the graphical display, the change in pain-related fear occurred more rapidly in subject 2 than in subject 1, except for pain intensity, where significance was reached at lags 4 and 5, respectively. When a combined P-value was calculated, in which no postponed effects were allowed, significance was reached for fear of movement/(re)injury and pain catastrophizing, but not for fear of pain and pain intensity (Table 4). Finally, the changes before and after treatment displayed in Table 5 reveal clinically significant improvements. For example, a TSK score of 22 and 19 approach the lowest possible score of 17 and are far below the 10th percentile (42). For the RDQ, a dramatic decrease, even larger than in previous studies (37), can be seen.

Discussion

When applying a more systematic exposure in vivo treatment with behavioural experiments in patients with chronic low back pain reporting substantial fear of movement/(re)injury, a dramatic decrease in pain-related fear occurs. Also of interest is that, in both patients, pain-intensity decreased, but lagged behind the reduction of pain-intensity. This suggests that the reduction of fear is not the consequence of pain reduction. On the contrary, the data suggest the opposite direction of action, namely that decreases in fear produce a decrease in pain. Given the reduction in PVAQ scores after the exposure in vivo treatment, it is interesting to speculate that pain reduction is mediated by a decreased need of these patients to scan their bodies for threatening pain sensations that previously were considered to be cues of impending harm. Finally, the effects of the fear reduction appear to generalize toward a significant improvement of daily functional status as measured with the RDQ.

GENERAL DISCUSSION

The aim of the present study was to examine the effectiveness of two potentially useful pain management approaches in reducing pain-related fear in patients reporting substantial fear of movement/(re)injury. Although the existing

TABLE 4
Meta-analysis: Combined P-value for the two patients (without delayed effects)

Minimum observation	Fear of movement	Pain catastrophizing	Fear of pain	Pain intensity
1	0.0107	0.0297	0.0503	0.2011
5	0.0165	0.0459	0.0776	0.3104

operant-behavioural treatment was not explicitly designed to reduce pain-related fears, it generally provides ample exposures to physical movements and activities. Its effects on pain-related fear, however, have not yet been studied. Recently, a more systematic application of exposure in vivo principles in combination with behavioural experiments has been developed. The systematic evaluation of the effectiveness of the exposure in vivo treatment has just started and needs further replication.

The results can be summarized as follows. Although a statistical reduction of mean TSK scores are found after an operant-behavioural treatment approach, the decrease is quite modest and clinically not convincing. Much more dramatic changes are seen when a systematic exposure in vivo with behavioural experiments is applied. In line with previous studies (37,47), decreases in pain-related fear also concurred with decreases in pain catastrophizing, pain disability and pain vigilance. In addition, current pain intensity levels were also affected by the exposure in vivo treatment, although these changes occurred at a slower pace. Consistent with experimental studies on the role of attention and pain-related fear, successful exposure in vivo treatment also resulted in decreases of pain vigilance. This finding corroborates the idea that the most important function of anxiety is the early detection of potentially threatening situations. Our study provides preliminary evidence for a process in which the reduction of the threat value of previously fear-eliciting stimuli (in casual physical activity) also produced an attentional redirection away from pain and bodily sensations. It is likely that the decrease in pain intensity in the treatment group might have been mediated by this attentional redirection. The current findings also suggest that the effects of the previous studies (37,46,47) do not seem to be contaminated by the background rehabilitation program that was delivered along with the exposure in vivo treatment.

There are a number of caveats to be considered. First, study 2 is limited in that it included only two patients. On the other hand, a single-case experimental design was chosen with appropriate randomization tests. Second, although the results suggest that exposure in vivo with behavioural experiments are superior to operant-behavioural treatment, we did not compare the two approaches directly in one single study. Theoretically, it is possible that the effects of the exposure in vivo treatment are due to other features of both studies. Although the patients in study 2 met the criteria used in study 1, they still may differ on characteristics not measured because the subjects were not randomly assigned

TABLE 5
Changes in fear of movement/(re)injury (Tampa Scale for Kinesiophobia [TSK], Photo Series of Daily Activities [PHODA]), pain disability (RDQ) and pain vigilance (PVAQ) before and after treatment

		TSK (17-68)	PHODA (0-100)	RDQ (0-24)	PVAQ (0-85)
Subject 1	Before treatment	61	81	19	49
	After treatment	22	22	2	15
Subject 2	Before treatment	47	81	16	47
	After treatment	19	16	4	19

PVAQ Pain Vigilance and Awareness Questionnaire; RDQ Roland Disability Questionnaire. Theoretical ranges are marked in parenthesis

over both conditions. Also, the operant-behavioural treatment was delivered in groups of five patients, while the exposure in vivo was delivered individually, which may have potentiated its effectiveness. A replication in the form of a randomized, controlled trial comparing both treatments using larger samples and long term follow-up measurements are warranted. We are currently preparing such a study that also includes costeffectiveness analyses.

Because no long term follow-up measures were included in the exposure study, it is unclear whether the gains are maintained over time. There is some evidence that generalization to other activities and situations than those used during the exposure treatment are difficult to achieve in patients with chronic pain (57). Research findings on exposure in anxiety disorders suggest that generalization and maintenance can be enhanced by a number of means. First, providing exposures to the full variety of contexts and natural settings in which fear has been experienced may be effective (eg, [58]). The PHODA might be a useful tool for eliciting information about these contexts in patients with chronic pain. It is preferable to carry out an exposure session in the context in which the fear has been acquired. Second, varying the stimuli during the exposure may be useful (59). Activities to be included in the exposure procedure can be best extended from those derived from PHODA to other activities as well. For example, bicycling can be done in very different ways (eg, using a mountain bicycle rather than a city bicycle, bicycling uphill as well as downhill, bicycling on rough as well as even terrain). Third, an expanded-spaced rather than a massed exposure schedule may enhance generalization and maintenance (60). It is preferable to spread the treatment over a longer period of time, rather than concentrate the treatment to a limited number of weeks. More studies are needed to examine fully the effects of these measures in patients with 'kinesiophobic' pain.

What can be said about the generalizability of the results to patients other than those included in the single-case experimental design? Although within single-case demonstrations with one or a few subjects it is, by definition, not possible to assess generality across subjects, a few comments are warranted. First, interventions that produce dramatic effects are likely to be more generalizable than those with

weaker effects, as is the case here. Second, generalizability may be derived from the fact that replications of 12 different patients (including patients of previous studies) show consistently similar results. So far, it seems justifiable to generalize the results to other patients with back pain who report substantial fear of movement/(re)injury. It may be desirable in future studies to increase the number of differences between the experiments and to test the intervention in patients with other musculoskeletal pain problems, such as whiplash, fibromyalgia or shoulder pain.

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