

PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	Testing a newly developed activity pacing framework for chronic pain/fatigue: a feasibility study
AUTHORS	Antcliff, Deborah; Keenan, Anne-Maree; Keeley, Philip; Woby, Steve; McGowan, Linda

VERSION 1 – REVIEW

REVIEWER	Cane, Douglas Nova Scotia Health Authority, Pain Management
REVIEW RETURNED	06-Nov-2020

GENERAL COMMENTS	<p>This study provides an initial evaluation of the feasibility and acceptability of a standardized intervention to increase activity pacing in individuals with ongoing pain and/or chronic fatigue. The intervention was based on a previously developed framework for activity pacing and was delivered as part of a larger, group rehabilitation program. Activity pacing, physical, and psychological functioning were assessed using standardized self-report measures at three times (pre-treatment, post-treatment, three-month follow-up). The treatment was well received by participants and delivered in a standardized manner. Increased activity pacing and improved functioning was observed at post-treatment with some improvements maintained at follow-up. Increased use of activity pacing was associated with improved psychological functioning at both post-treatment and at follow-up.</p> <p>This study is of interest to clinicians and researchers interested in the relationship between activity pacing and functioning. The study's objectives are clearly described and the design and analyses are appropriate to address the objectives. Overall, the paper is well written and the study's limitations are properly noted.</p> <p>Consideration of the following suggestions may further strengthen the manuscript.</p> <ol style="list-style-type: none">1. A key aspect of the study is the evaluation of an intervention to increase activity pacing. While the underlying conceptual framework is described, little information is provided about the actual intervention. It would be helpful to include more details about the actual treatment provided to participants.2. The APQ is used to evaluate activity pacing and changes in activity pacing. Although this measure is described in other
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	<p>papers, it would be helpful to provide more details here about the APS themes and sample items to illustrate the measure.</p> <p>3. There is a previously published study (Racine et al., 2019) of two forms of activity pacing (operant versus energy conservation). It would be useful to compare/contrast findings from this study with the present study and, if possible, discuss differences with respect to the interventions used to increase activity pacing.</p> <p>4. As expected, there is considerable attrition from baseline to follow-up. The authors do not provide a comparison of the end of treatment functioning (t2) for participants who complete treatment and provide t3 data with those who complete treatment but do not provide t3 data. The sample of participants who provided data at t3 may be biased by their response to treatment (i.e. participants who benefitted more from treatment and were functioning better after treatment may be more motivated to provide follow-up data). It would be helpful to explore this possibility.</p> <p>5. On page 34 of the discussion (line 49) the authors contrast a rehabilitative approach versus an adaptive approach. Could the authors clarify what they mean by these terms.</p>
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REVIEWER	Meijer, O VU University, Amsterdam, Human Movement Sciences
REVIEW RETURNED	18-Nov-2020

GENERAL COMMENTS	<p>Review of: BMJopen-2020-045398 "Testing a newly developed activity pacing framework for chronic pain / fatigue: A feasibility study".</p> <p>The authors study the feasibility of using a newly developed pacing framework for chronic pain and/or chronic fatigue in an uncontrolled study. Moreover, the authors explore if measures of pacing / symptoms detect changes after the treatment. Out of 139 eligible patients, 107 patients with chronic low back pain, chronic widespread pain, fibromyalgia and chronic fatigue consented to undergo six weeks of rehabilitation that used the standardised pacing framework. Patients' adherence and satisfaction were registered, as was caretakers' fidelity. Questionnaires included measures of pacing, and symptoms of the above afflictions. 65 patients (61%) completed the programme, and 52 (49%) were available for follow-up. At the end of the programme, satisfaction averaged 9 out of 10, and pacing as well as all symptoms had improved, but the improvement was smaller at follow-up. Between the beginning and the end of the programme, changes in pacing correlated with current pain, self-efficacy, and mental function. Between the beginning of the programme and follow-up, changes in pacing were also correlated with fatigue, depression, anxiety and quality of life. Healthcare professionals reported 100% fidelity to the framework. The authors conclude that the activity pacing framework was feasible.</p> <p>General evaluation</p>
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The submitted manuscript is clearly part of a research programme. The topic is relevant to the rehabilitation of a number of afflictions that are difficult to treat, and the results appear to warrant publication. Although we found no major error in the text, we were puzzled by our total lack of enthusiasm for the manuscript. Both may be due to the fact that the text lacks precision, tends to describe relevant decisions in meta-terms, rather than focus on the nature of those decisions, in particular: What was decided not to use or not to do. We will illustrate our dissatisfaction in a § by § discussion of the manuscript.

Introduction

"At present, there remains confusion regarding how activity pacing is defined or interpreted There is no widely used guide to standardise how healthcare professionals instruct pacing to patients"

We agree, and given the great importance of these statements, the reader needs some guidance on what has to be decided how. What are the dimensions of the decisions to be taken? For instance, will the framework follow time or persistence / worsening of symptoms, and whatever was chosen: Why was it chosen?

"mixed methods were implemented to encompass theoretical and stakeholder standpoints."

This is very much meta-language. What are "mixed methods"? Who are the stakeholders? What is the role of patients' preferences? An athlete with chronic fatigue may want to reengage in sports, and may then find that the fatigue hits again. What does the framework propose to do in such situations? One of our students met a cancer patient who was too tired to go to the hospital to undergo outpatient treatment. "What would you like to do again most?" "Go to the casino!" And the visit to the casino was cut into small pieces, such as washing, putting his clothes on, walking to the taxi, etc., each exercised, and soon the patient went to the casino for a short visit, was able again to go to the hospital, and started to engage in other pleasant activities.

It is our impression that a consensus meeting with patients after collecting expert opinions may underemphasize the patient. Which is OK, but it should be stated, reasons for it should be explicit, and it should come back in the discussion.

Note that Figure 1 does not help to answer our questions as to the nature of the pacing framework.

What is "quota-contingency"? If that is: so much activity today, and so much tomorrow, in how far is your approach different from Cognitive Behavioural Therapy, as, for instance, in the work of Hans Knoop?

The text of p. 6, 45 through p. 7, 8 is so vague and general that it is impossible to be wrong. In our mind, it is the hallmark of science to be so clear that you can be shown to be wrong.

Methods

"Activity pacing was formally instructed on two sessions (weeks 2 -3), but also referenced throughout the programme in relation to other coping strategies."

Hard to understand what exactly was instructed, and this "in relation to other coping strategies" is puzzling. In other words, we do not know what you did (and reported the feasibility of). Also the " participants chose whether to complete the study questionnaires" is insufficiently clear.

As to the feasibility outcomes, did you set minimum requirements for the framework to be "feasible"? Or did you just collect data, and then decided if these suggested feasibility or not. Note that this is an important point.

The five dimensions of the Activity Pacing Questionnaire are insufficiently clear, and require some examples.

What do you mean by "depression"? Are there not at least two different dimensions to depression: The being poorly motivated as in Dantzer's sickness behaviour, and dark mood (up to having suicidal thoughts)? And if people were depressed, did they receive psychopharmaca?

As to the clinical outcomes, would some "effect size" not be best to decide if these changes were potentially relevant?

What is a "nominal group technique"?

What is "accessibility" of participation? Assuming that the staff were enthusiastic because this was a new programme, and that such enthusiasm is usually infectious, did you control for that possible confounder? Should it be mentioned in the Discussion?

Results

Table 1 may be a bit overcomplete.

Table 2 will gain readability if you give the range from worst to best possible scores for all variables mentioned.

Recruitment and attrition: The paper would be much more readable if you first give some cut-off values for good/bad recruitment / attrition.

To us, it is not clear what are 'activity pacing' sessions versus 'pacing specific' sessions.

Table 3: Maybe some Figures will enhance readability.

As to health care worker 'fidelity', the abstract states this as a goal, but gives no results.

Again, mean changes in activity pacing and symptoms would contain more valuable information for the reader if more quantitative information were given. This is important for the reader who wants to decide if these changes are sufficiently 'important'.

Given the relatively modest aims of the study, we invite the authors to contemplate the possibility that the Tables give too much information.

Discussion

The §1 "demonstrated improvements in pacing strategies and reductions in symptoms." suggested that you performed an intervention study, which you did not. Please, reformulate.

The §2 "demonstrated feasibility" is a bit of a puzzle because it seems to assume that health care worker fidelity plus patient satisfaction are sufficient criteria to decide on feasibility. Is it? If so, we should have known that earlier. If not, reformulate.

	<p>"the study protocol" was published, right? If so, give a reference. And we need some arguments why reference 44 is adequate as a source for judging recruitment "good".</p> <p>The fact that "many participants" dropped out after the first session suggest that modesty would be in order. The "excellent" attendance of those who completed T2 needs some downplaying because of the initial dropouts.</p> <p>We do not understand your analysis of the difference between the present study and that of White et al. (47). What does it mean that the activity pacing framework 'encourages a rehabilitative rather than an adaptive approach', and that a "causative investigation" is required. Do you mean in terms of inflammation, fibrosis, etc.? If (not) so, that should be stated.</p> <p>As to the clinical outcomes, again, we would like to know how important to the patient the observed differences are, particularly since the greater than minimally clinically important change was lost at T3</p> <p>Note that "much this improvement was maintained" contains a typo.</p> <p>What is the status of "pacing was not associated with improved physical function among patients with chronic conditions (5)"? Are you implying that psychological well-being should be the main goal of your pacing framework. If so, and why not?, this should be stated more explicitly.</p> <p>Conclusion</p> <p>The readability of the paper needs to be improved. We saw that another study was attached to the submission, but we reviewed the present manuscript as a stand-alone publication. If you derive cut-off values from an earlier paper, our problems can be easily solved, such as in "the minimum xxx was set at yyy (reference zzz)", which will help a lot. Remains the problem that we do not fully understand what the pacing framework consists of. These are the two major problems we have. If the Editor agrees that this manuscript should be reviewed as a stand-alone paper (with references to earlier publications), we would expect that you clarify the contents of the pacing framework with examples, that you clearly describe what the differences are with cognitive behavioural therapy, and that you know the health care worker should do if the symptoms worsen. Good luck</p> <p>Onno G. Meijer</p> <p>Maarten R. Prins</p>
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VERSION 1 – AUTHOR RESPONSE

Comments from Reviewer 1	
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This study provides an initial evaluation of the feasibility and acceptability of a standardized intervention to increase activity pacing in individuals with ongoing pain and/or chronic fatigue. The intervention was based on a previously developed framework for activity pacing and was delivered as part of a larger, group rehabilitation program. Activity pacing, physical, and psychological functioning were assessed using standardized self-report measures at three times (pre-treatment, post-treatment, three-month follow-up). The treatment was well received by participants and delivered in a standardized manner. Increased activity pacing and improved functioning was observed at post-treatment with some improvements maintained at follow-up. Increased use of activity pacing was associated with improved psychological functioning at both post-treatment and at follow-up.

This study is of interest to clinicians and researchers interested in the relationship between activity pacing and functioning. The study's objectives are clearly described and the design and analyses are appropriate to address the objectives. Overall, the paper is well written and the study's limitations are properly noted.

The authors are thankful for the reviewers' comments

<p>Consideration of the following suggestions may further strengthen the manuscript.</p> <p>1. A key aspect of the study is the evaluation of an intervention to increase activity pacing. While the underlying conceptual framework is described, little information is provided about the actual intervention. It would be helpful to include more details about the actual treatment provided to participants</p>	<p>Thank you for highlighting that further information regarding the intervention would strengthen the manuscript. To address this, the authors have added Supplementary Figure 1 to show the content of the Activity Pacing Framework: Theory and Overview, and Appendices and Teaching Guide booklets; and Supplementary Figure 2 to show the content of the two sessions on activity pacing contained within the rehabilitation programme.</p>
<p>2. The APQ is used to evaluate activity pacing and changes in activity pacing. Although this measure is described in other papers, it would be helpful to provide more details here about the APS themes and sample items to illustrate the measure</p>	<p>The authors have added Supplementary Table 2 to show the five themes of the activity pacing questionnaire, with examples of items for each theme.</p>

<p>3. There is a previously published study (Racine et al., 2019) of two forms of activity pacing (operant versus energy conservation). It would be useful to compare/contrast findings from this study with the present study and, if possible, discuss differences with respect to the interventions used to increase activity pacing</p>	<p>3. Thank you for this suggestion of a highly relevant paper. Reference to how the present findings relate to this paper, and methods of increasing activity pacing have been added to the Discussion (amendments highlighted in blue font) on pages 37-39.</p> <p>Related to this, methods to increase uptake of activity pacing in the current study included using handouts, together with goal setting. This has been added (in blue font) to the Methods section (page10).</p>
<p>4. As expected, there is considerable attrition from baseline to follow-up. The authors do not provide a comparison of the end of treatment functioning (t2) for participants who complete treatment and provide t3 data with those who complete treatment but do not provide t3 data. The sample of participants who provided data at t3 may be biased by their response to treatment (i.e. participants who benefitted more from treatment and were functioning better after treatment may be more motivated to provide follow-up data). It would be helpful to explore this possibility.</p>	<p>4. Thank you for raising this interesting point regarding differences between patients who complete T2 and those who do not complete T3. Due to the uneven subgroup sizes between the small number of patients who completed T2 but not T3 (n=13) compared to the larger group who completed T2 and T3 (n=52), we did not estimate statistical differences since it is difficult to find statistical differences when group sizes are so different. However, we agree that this is a valuable point and as such, we have added this to the Discussion, page 39 (in blue font).</p> <p>In addition, the original closing paragraph of the 'Strengths and limitations' (page 40) returns to this concept of potential bias from attrition.</p>

<p>5. On page 34 of the discussion (line 49) the authors contrast a rehabilitative approach versus an adaptive approach. Could the authors clarify what they mean by these terms</p>	<p>Thank you for this comment. The authors felt it would be suitable to expand on their meaning of the rehabilitative pacing approach of the activity pacing framework in the Introduction. The original paragraph in the Introduction (page7) has been expanded (in blue font)</p> <p>Furthermore, within Figure 1: Conceptual model of the activity pacing framework, the use of the term rehabilitative compared to adaptive is stated.</p>
<p>Comments from Reviewer 2</p>	<p>Authors' response</p>

The authors study the feasibility of using a newly developed pacing framework for chronic pain and/or chronic fatigue in an uncontrolled study. Moreover, the authors explore if measures of pacing / symptoms detect changes after the treatment. Out of 139 eligible patients, 107 patients with chronic low back pain, chronic widespread pain, fibromyalgia and chronic fatigue consented to undergo six weeks of rehabilitation that used the standardised pacing framework. Patients' adherence and satisfaction were registered, as was caretakers' fidelity. Questionnaires included measures of pacing, and symptoms of the above afflictions. 65 patients (61%) completed the programme, and 52 (49%) were available for follow-up. At the end of the programme, satisfaction averaged 9 out of 10, and pacing as well as all symptoms had improved, but the improvement was smaller at follow-up. Between the beginning and the end of the programme, changes in pacing correlated with current pain, self-efficacy, and mental function. Between the beginning of the programme and follow-up, changes in pacing were also correlated with fatigue, depression, anxiety and quality of life. Healthcare professionals reported 100% fidelity to the framework. The authors conclude that the activity pacing framework was feasible.

The authors are thankful that Reviewer 2 believes the research findings are worthy of publication. We will endeavour to address each of the reviewers' concerns in turn.

<p>1. Introduction "At present, there remains confusion regarding how activity pacing is defined or interpreted There is no widely used guide to standardise how healthcare professionals instruct pacing to patients"</p> <p>We agree, and given the great importance of these statements, the reader needs some guidance on what has to be decided how. What are the dimensions of the decisions to be taken? For instance, will the framework follow time or persistence / worsening of symptoms, and whatever was chosen: Why was it chosen?</p>	<p>1. The decisions on the contents of the activity pacing framework were the product of the stages of development: the online healthcare professionals' survey and nominal group technique (please see the Introduction, page 6).</p> <p>The activity pacing framework follows quota-contingency and an operant approach, that encourage flexibility around patients' choice and symptoms that can vary. Flexibility was identified as an important component of the framework during its development, since historical pacing approaches have been criticised for being overly-prescriptive or punitive. This is discussed in the two papers that are referenced in the current manuscript that report the online survey and nominal group technique (Antcliff et al. 2019a&b).</p> <p>The conceptual model of the activity pacing framework is provided in Figure 1. This states the model is underpinned by the operant approach and quota contingency, together with other key elements of the activity pacing framework approach to pacing. Furthermore, the introduction (page 7) has been expanded to provide more detail regarding the conceptual model of the activity pacing framework (amendments highlighted in blue font).</p>
<p>2. "mixed methods were implemented to encompass theoretical and stakeholder standpoints." This is very much meta-language. What are "mixed methods"? Who are the stakeholders? What is the role of patients' preferences?</p>	<p>2. In the Introduction (page 6) we have added that mixed methods incorporate both quantitative and qualitative research methods and included a relevant reference to Creswell and Plano-Clark (2011).</p> <p>The paragraph reporting the development of the activity pacing framework aims to provide an overview of the stages of development since they are reported elsewhere (as referenced to Antcliff et al. 2019a&b). It is beyond the scope of this manuscript to report on these stages in full. However, the authors have added information that the stakeholders included healthcare professionals and patients with the aim of increasing the clinical utility and acceptability of the framework in the Introduction, page 6 (in blue font):</p>

<p>An athlete with chronic fatigue may want to reengage in sports, and may then find that the fatigue hits again. What does the framework propose to do in such situations? One of our students met a cancer patient who was too tired to go to the hospital to undergo outpatient treatment. "What would you like to do again most?" "Go to the casino!" And the visit to the casino was cut into small pieces, such as washing, putting his clothes on, walking to the taxi, etc., each exercised, and soon the patient went to the casino for a short visit, was able again to go to the hospital, and started to engage in other pleasant activities.</p>	<p>The framework is designed to be used by a range of patients with varying abilities. The conceptual model (Figure 1) states: "Different facets of activity pacing are tailored to individuals' needs, aims and activity behaviours." Therefore, each individual patient will present differently and will benefit from some themes of pacing more than others, and this may vary over time, for example, during a flare up. Examples of activity pacing are provided in the framework as short exemplar case studies. We have added supplementary figure 1 which shows the contents of the activity pacing framework, including the use of these exemplars in the Appendices and Teaching Guide.</p>
<p>It is our impression that a consensus meeting with patients after collecting expert opinions may underemphasize the patient. Which is OK, but it should be stated, reasons for it should be explicit, and it should come back in the discussion.</p>	<p>The framework was developed as a guide for healthcare professionals. Therefore, Stage 1 of the framework development sought to seek a wide range of healthcare professionals' opinions to develop the first draft of the framework. To increase clinical utility and relevance to patients, we invited a group of patients and clinicians to participate in the nominal group technique to refine the framework. The methods of development in these two stages, the justification and strengths and weaknesses of these stages are reported as referenced (Antcliff et al. 2019a&b). Future work is planned to develop a patient friendly version of the activity pacing framework, and as such, this development will involve a greater number of patients.</p>

<p>Note that Figure 1 does not help to answer our questions as to the nature of the pacing framework.</p>	<p>Figure 1 shows the conceptual model as taken from the activity pacing framework. This model provides the type of pacing: operant approach driven by quota-contingency, but with important components of flexibility (which contrasts previous opinions that pacing is rigid). The model provides the aim of activity pacing and examples of the multidimensional facets of pacing.</p> <p>To further demonstrate how the activity pacing framework is structured, we have added Supplementary Figure 1 which shows the content of the Theory and Overview, and Appendices and Teaching Guide.</p>
<p>What is "quota-contingency"? If that is: so much activity today, and so much tomorrow, in how far is your approach different from Cognitive Behavioural Therapy, as, for instance, in the work of Hans Knoop?</p> <p>The text of p. 6, 45 through p. 7, 8 is so vague and general that it is impossible to be wrong. In our mind, it is the hallmark of science to be so clear that you can be shown to be wrong.</p>	<p>Quota-contingency involves undertaking a task or goal according to time/distance/activity), as stated in the Introduction (page 7). The activity pacing framework does align with some principles of CBT, however, this model encourages active decision making and flexibility according to individuals' goals and priorities which may be more similar to principles of acceptance and commitment therapy (ACT) and psychological flexibility. The activity pacing framework has not sought to mimic a particular psychological approach, rather, modify and clarify an attainable approach to pacing. It is the intention that the framework can be used alongside psychological therapies in pain management programmes.</p> <p>The authors have added information about the conceptual model of the framework to the Introduction and the contents of the framework to the supplementary material. The activity pacing framework has been developed based on learning (from literature and stakeholder's opinions) how previous pacing approaches have been helpful and also unhelpful. Some previous pacing approaches have been unidimensional, rigid and overly-prescriptive. Therefore, this pacing framework is multidimensional and flexible. We do not consider this to be vague, rather an approach to activity pacing that has developed as our knowledge of living with long-term conditions has developed; and a pacing approach that can be tailored to individuals' needs and allows for life's changes.</p>

<p>3. Methods</p> <p>"Activity pacing was formally instructed on two sessions (weeks 2 -3), but also referenced throughout the programme in relation to other coping strategies."</p> <p>Hard to understand what exactly was instructed, and this "in relation to other coping strategies" is puzzling. In other words, we do not know what you did (and reported the feasibility of).</p>	<p>The authors have clarified this meaning in the Methods, page 10 (in blue font), to explain that activity pacing specifically instructed on weeks 2-3, but has relevance to other strategies used on varying weeks of the programme, e.g. graded exercise and set-back management.</p> <p>In addition, supplementary Figure 1 shows the contents of the activity pacing framework: Theory and Overview, including section 12: "How pacing relates to other coping strategies". Supplementary Figure 2 shows the full content of the programme to provide context to when activity pacing is instructed.</p> <p>Of note, we tested the feasibility of using the pacing framework to underpin a rehabilitation programme that includes activity pacing, together with other strategies such as graded exercise, relaxation etc. This decision was made since activity pacing is often an integral part of other coping strategies. In addition, activity pacing is often used clinically as one component of a holistic rehabilitation programme. We acknowledge that we tested feasibility of the activity pacing framework within a rehabilitation programme, and not activity pacing as a lone strategy in the Discussion (page 40).</p>
<p>Also the " participants chose whether to complete the study questionnaires" is insufficiently clear.</p>	<p>The authors have clarified that patients chose whether or not to participate in the study through their completion of questionnaires in the Methods, page 10 (in blue font).</p>

<p>As to the feasibility outcomes, did you set minimum requirements for the framework to be "feasible'? Or did you just collect data, and then decided if these suggested feasibility or not. Note that this is an important point.</p>	<p>Thank you for raising the point regarding minimum requirements that can be used in feasibility trials. As per guidance on feasibility and pilot trials from Eldridge et al. 2016, using minimum requirements can be a component of pilot and feasibility studies as appropriate. However, feasibility studies occur in many different formats to ask questions whether something can be done and how to proceed, including both quantitative and qualitative work. According to the MRC framework, feasibility seeks to explore whether an intervention is acceptable, and likely recruitment/retention rates.</p> <p>Accordingly, the aim of the current feasibility study was to undertake the first exploration of using the newly developed activity pacing framework in the clinical setting to see whether it was usable and acceptable for healthcare professionals and patients. Since this was the first stage in this exploratory work, we did not set <i>a priori</i> requirements, rather we sought to see whether the activity pacing framework could be used, ahead of planning towards a future effectiveness trial.</p>
<p>The five dimensions of the Activity Pacing Questionnaire are insufficiently clear, and require some examples.</p>	<p>We have added Supplementary Table 2 to show the APQ themes with examples of scale items</p>

<p>What do you mean by "depression"? Are there not at least two different dimensions to depression: The being poorly motivated as in Dantzer's sickness behaviour, and dark mood (up to having suicidal thoughts)?</p>	<p>Within this context, depression is defined and measured according to the Patient Health Questionnaire (PHQ-9), the items of which are based on the based on the Diagnostic and Statistical Manual of Mental Disorders 4th Edition (DSM-IV). The PHQ-9 contains nine items that screen for and measure the severity of depression in the clinical setting and one item does refer to suicidal thoughts.</p> <p>All patients attending the rehabilitation programme were assessed prior to attending the rehabilitation programme to check their eligibility and suitability to attend the programme (as per normal practice). This does include assessing for motivation to attend the programme and all patients are screened for suicide risk.</p> <p>Within the Methods (page 9), the participant eligibility criteria are stated to exclude those with severe mental health or cognitive functioning issues.</p> <p>Following the reviewer's comments, the authors have added a sentence to the description of the PHQ-9 is based on the DSM-IV definition of depression (Methods, page12, in blue font).</p>
<p>And if people were depressed, did they receive psychopharmaca?</p>	<p>Patients are prescribed appropriate medication as part of their holistic and multidimensional approach to pain management. However, the authors believe that discussing varying medication use of patients attending the programme is beyond the scope of this manuscript.</p>
<p>As to the clinical outcomes, would some "effect size" not be best to decide if these changes were potentially relevant?</p>	<p>In keeping with a single arm feasibility study, mean changes and 95% confidence intervals of change in symptoms were initially reported (Eldridge et al., 2016).</p> <p>To address the comment by the reviewer, the authors have added effect sizes to Table 3. However, due to the small sample size, clinical implications cannot be inferred (as advised by Eldridge et al 2016).</p>

<p>What is a "nominal group technique"?</p>	<p>The Nominal group technique (NGT) is a well reported and widely used consensus method (Jones & Hunter, 1995; Wainwright, Boichat, & McCracken, 2014). Within this manuscript, the nominal group technique is stated to be a consensus meeting between healthcare professionals and patients (Methods, page 6). The reference to the methods and findings of this study are provided for further information (Antcliff et al. 2019b). The authors believe this provides sufficient information about the nominal group technique within the context of this manuscript to maintain focus on the current feasibility study.</p>
<p>What is "accessibility" of participation? Assuming that the staff were enthusiastic because this was a new programme, and that such enthusiasm is usually infectious, did you control for that possible confounder? Should it be mentioned in the Discussion?</p>	<p>Accessibility of participation relates to improving patients' accessibility to attend the research meetings in terms of practicality, for example, considering location, stairs, comfort breaks, length of meetings. The authors have added clarification to this in the Methods, page 14 (in blue font)</p>
<p>4. Results Table 1 may be a bit overcomplete.</p>	<p>The authors believe the level of information in Table 1 presents the demographics of the sample to enable the reader to appreciate the generalisability of the sample.</p>
<p>Table 2 will gain readability if you give the range from worst to best possible scores for all variables mentioned.</p>	<p>Best-to-worst scores for each scale are explained in the Methods: Clinical Measures (pages 11-13). Following the reviewer's comment, the authors have added the range of scores for each scale to Table 2.</p>

<p>Recruitment and attrition: The paper would be much more readable if you first give some cut-off values for good/bad recruitment / attrition.</p>	<p>The authors provided references towards good/bad recruitment/attrition rates in the Discussion, page 35 as a comparison to other feasibility studies that have explored interventions for similar samples of people chronic pain/fatigue.</p>
<p>To us, it is not clear what are 'activity pacing' sessions versus 'pacing specific' sessions.</p>	<p>The authors are sorry for any confusion caused by using "pacing" as short hand for "activity pacing". The "pacing" sessions are the "activity pacing" sessions in this context. The authors have added "activity" to every "pacing" session to improve clarity.</p>
<p>Table 3: Maybe some Figures will enhance readability.</p>	<p>Since the study involves a number of clinical outcomes, the authors believe that these changes (measured on three time intervals) are most clearly presented as a table. We hope that the addition of effect sizes as requested by reviewer 2 will enhance the reader's interpretation of these findings further.</p>
<p>As to health care worker 'fidelity', the abstract states this as a goal, but gives no results.</p>	<p>Healthcare professional fidelity is reported in the Results, on page 21.</p>
<p>Again, mean changes in activity pacing and symptoms would contain more valuable information for the reader if more quantitative information were given. This is important for the reader who wants to decide if these changes are sufficiently 'important'.</p>	<p>To address the reviewer's comment, the authors have added effect sizes to the mean changes in activity pacing and symptoms in Table 3.</p>

<p>Given the relatively modest aims of the study, we invite the authors to contemplate the possibility that the Tables give too much information.</p>	<p>The main aim of the study is to test the feasibility of using the activity pacing framework in the clinical setting for chronic pain and fatigue. As such, the authors have reported on factors relating to the implementation, adherence and satisfaction of the activity pacing framework among patients with predominant symptoms of pain and those with predominant symptoms of fatigue. The authors state four specific objectives (on page 7-8 of the Introduction) as: (1) Exploring participant recruitment/attrition rates and adherence/acceptability (for both chronic pain and fatigue); (2) Exploring healthcare professionals' fidelity to the framework; (3) Exploring the suitability of the outcome measures, including the modified activity pacing questionnaire (APQ-28); and (4) Exploring associations between changes in activity pacing and self-reported symptoms. The authors believe that the results presented in this manuscript fulfil these aims accordingly.</p>
<p>5. Discussion The §1 "demonstrated improvements in pacing strategies and reductions in symptoms." suggested that you performed an intervention study, which you did not. Please, reformulate.</p>	<p>The Discussion (page 35) now reads as: "The study recruited to target and patients with chronic pain and chronic fatigue demonstrated both improvements in pacing strategies and reductions in symptoms."</p> <p>This statement is true since both improvements are seen in pacing and symptoms. The authors have not mentioned causality. Rather, the authors state in the limitations that causation cannot be inferred (on page 40).</p>

<p>The §2 "demonstrated feasibility' is a bit of a puzzle because it seems to assume that health care worker fidelity plus patient satisfaction are sufficient criteria to decide on feasibility. Is it? If so, we should have known that earlier. If not, reformulate.</p>	<p>The feasibility outcomes are stated in the Methods, p11 as: "Measures of feasibility included participant recruitment/attrition rates, adherence (number of sessions attended), acceptability (two satisfaction rating scales regarding the programme content and length where 0=dissatisfied and 10=fully satisfied), and missing data in the questionnaire. For every programme, healthcare professionals completed a 13-item fidelity checklist based on the conceptual model of the activity pacing framework to ensure their inclusion of key elements from the framework. Each clinician was observed once by the lead researcher."</p> <p>As stated above, the authors did not set a priori minimum requirements to demonstrate feasibility since this study served an initial exploratory work.</p>
<p>"the study protocol" was published, right? If so, give a reference.</p>	<p>The protocol was not published, but is available within the feasibility study registration with clinicaltrials.gov. The authors provided a reference to where the protocol can be found in the Methods, page 8.</p>
<p>And we need some arguments why reference 44 is adequate as a source for judging recruitment "good".</p>	<p>Reference 44 (Reference 47 in the resubmission) developed a 'mind-body physical activity program for patients with heterogeneous chronic pain'. Therefore, in the absence of previous recruitment rates for the activity pacing framework, we compared our recruitment to Reference 44 which developed a similar intervention for a similar patient group to the current study.</p>

<p>The fact that "many participants" dropped out after the first session suggest that modesty would be in order. The "excellent" attendance of those who completed T2 needs some downplaying because of the initial dropouts.</p>	<p>The rating of 'excellent' is that given by another research study and so this term cannot be changed by the present author. However, the authors have acknowledged that modesty may be required and have added this acknowledgement to the Discussion, page 36 (in blue font)</p>
<p>We do not understand your analysis of the difference between the present study and that of White et al. (47). What does it mean that the activity pacing framework 'encourages a rehabilitative rather than an adaptive approach", and that a "causative investigation" is required. Do you mean in terms of inflammation, fibrosis, etc.? If (not) so, that should be stated.</p>	<p>The authors have clarified the meaning of the rehabilitative approach to pacing in the Introduction. (<i>Please see response to Reviewer 1, comment 5</i>). By "causative investigation", the authors refer to undertaking effectiveness trials to decipher whether improvements in pacing lead to improvements in symptoms, rather than exploring correlations. The authors have re-worded this for clarity in the Discussion, page 36 (in blue font)</p>
<p>As to the clinical outcomes, again, we would like to know how important to the patient the observed differences are, particularly since the greater than minimally clinically important change was lost at T3</p>	<p>The authors have provided insight into some patients' opinions by the comments provided in the questionnaire booklet (please see Figure 3).</p> <p>The authors have explored patients' opinions on the activity pacing, their experience of attending this rehabilitation programme and changes in their symptoms/coping in more depth as qualitative acceptability interviews. This has been reported elsewhere (Antcliff et al. 2021, accepted for publication) and signposted towards in the Methods, page 8 (in blue font).</p>

<p>Note that "much this improvement was maintained" contains a typo.</p>	<p>Thank you-this typo has been corrected to "much of this improvement was maintained".</p>
<p>What is the status of "pacing was not associated with improved physical function among patients with chronic conditions (5)"? Are you implying that psychological well-being should be the main goal of your pacing framework. If so, and why not?, this should be stated more explicitly.</p>	<p>This statement suggests that at present, pacing appears to be more correlated with psychological wellbeing. We are not implying that psychological wellbeing should be the main goal of the framework. Rather, the authors suggest that this may warrant further investigation as stated in the Discussion, page 40.</p>
<p>6. Conclusion The readability of the paper needs to be improved. We saw that another study was attached to the submission, but we reviewed the present manuscript as a stand-alone publication. If you derive cut-off values from an earlier paper, our problems can be easily solved, such as in "the minimum xxx was set at yyy (reference zzz)", which will help a lot.</p>	<p>The authors attached only the protocol for the feasibility study, in which the methods for the acceptability study are also enclosed. The acceptability manuscript has now been accepted for publication and is cited within this manuscript.</p> <p>As per the protocol, this feasibility study did not include a priori minimum requirements.</p>

Remains the problem that we do not fully understand what the pacing framework consists of. These are the two major problems we have. If the Editor agrees that this manuscript should be reviewed as a stand-alone paper (with references to earlier publications), we would expect that you clarify the contents of the pacing framework with examples, that you clearly describe what the differences are with cognitive behavioural therapy, and that you know the health care worker should do if the symptoms worsen. Good luck

The authors have expanded the information on the conceptual model of the activity pacing framework in the Introduction, and the conceptual model taken from the framework is presented as Figure 1.

In addition, the authors have added supplementary Figure 1 to show the full contents of the activity pacing framework. We have also added Figure 2 to show the individual sessions of the rehabilitation programme.

The authors have referenced the two papers that report the development of the activity pacing framework, together with the reference of the newly published acceptability paper.

The aim of this work is not to compare the activity pacing framework with CBT, since there are overlaps between activity pacing and many psychological approaches such as CBT, acceptance and psychological flexibility. The aim of this work was to test whether the framework could be used in a clinical setting, ahead of future preparations towards testing effectiveness. It is intended that following further refinement ahead of an effectiveness trial, the activity pacing framework will be made widely available in different formats-one for healthcare professionals and one for patients. This feasibility study was the first essential step in gauging whether further research may be possible.

Regarding the worsening of symptoms, the activity pacing framework does contain multidimensional facets of pacing including adjusting activities, finding baselines, acceptance etc. Facets of pacing are used according to an individual's needs and tailored to their abilities. Supplementary Figure 1 shows the inclusion of facets of pacing that may be used during a flare up. The management of setbacks is specifically discussed in Week 6 of the rehabilitation programme (please see Supplementary Figure 2), and pacing forms part of this, together with medicines management, graded movement etc.

Thank you for your comments.

VERSION 2 – REVIEW

REVIEWER	Cane, Douglas Nova Scotia Health Authority, Pain Management
REVIEW RETURNED	18-May-2021

GENERAL COMMENTS	Thank you for the opportunity to review this revised manuscript. The revised manuscripts addresses my previous comments. Specifically, it provides additional details regarding the conceptual framework, pacing measure, and content of the sessions specific to activity pacing. I have no further suggestions.
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REVIEWER	Meijer, O VU University, Amsterdam, Human Movement Sciences
REVIEW RETURNED	02-Jun-2021

GENERAL COMMENTS	<p>Our main objection to the paper was that the language was too formal (and that much information needed to know was in papers that had to be downloaded, which is not user-friendly). The authors clearly tried to accomodate our problems, but their answer still containa lot of formal statements, which are essentially unreadable for the clinically interested. Wetherefore recommend rejection.</p> <p>We asked it the authors had some a priori criterion to decide if the study could be deemed successful or not, and the authors now respond that they didn't have such a criterion. This renders the paper 'unscientific' in the sense of Karl Popper. Sure, it is our impression that subjects liked to be confronted with (the extra attention of) this new approach, but that would be true for any new approach, and we find it impossible to interpret the 'feasibility' reported.</p> <p>Some of our questions were clearly answered, but the answer itself was regularly disappointing. One example may be sufficient. The authors do not problematize the notion of 'depression', and apparently use the DSM, which was regarded as'invalid' by the National Institute of Mental Health. So, when an effect size of 1 is reported for depression, we do not know what happened. Does this reflect that subjects became more active? Or that their mood improved? Surely, these are different dimensions.</p> <p>The Tables contain a tsunami of results (now with effect sizes, which improves readability), but there is a clear lack of focus (also in reporting relationships), and one cannot but wonder if any correction for multiple testing should have been used, or any other methods to focus on what is really relevant. The above is for us enough to recommend rejection of the paper. We are not willing to see another version.</p> <p>Sincerely, Onno G. Meijer and Maarten R. Prins</p>
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VERSION 2 – AUTHOR RESPONSE

Comments from Reviewer 1	Authors' response
<p>Thank you for the opportunity to review this revised manuscript. The revised manuscripts addresses my previous comments. Specifically, it provides additional details regarding the conceptual framework, pacing measure, and content of the sessions specific to activity pacing. I have no further suggestions.</p>	<p>The authors are thankful for Reviewer 1's comment and review of our resubmission.</p>
Comments from Reviewer 2	Authors' response

Our main objection to the paper was that the language was too formal (and that much information needed to know was in papers that had to be downloaded, which is not user-friendly). The authors clearly tried to accommodate our problems, but their answer still contain lot of formal statements, which are essentially unreadable for the clinically interested. We therefore recommend rejection.

The authors believe that the manuscript is presented in a professional language that is appropriate and consistent with other articles included BMJ. This feasibility study reports the final stage of a multi-staged programme of developmental work. We have been unable to include the full details of all of the methods and findings from previous stages since that would distract from the purpose and findings of the current feasibility study, and also exceed a suitable word count. Instead, to produce a succinct report on the current feasibility study, we have included the important details of our earlier work, with clear referencing to our other publications if further details are required.

<p>We asked if the authors had some a priori criterion to decide if the study could be deemed successful or not, and the authors now respond that they didn't have such a criterion. This renders the paper 'unscientific' in the sense of Karl Popper. Sure, it is our impression that subjects liked to be confronted with (the extra attention of) this new approach, but that would be true for any new approach, and we find it impossible to interpret the 'feasibility' reported.</p>	<p>The authors have undertaken early feasibility work, the purpose of which was to ascertain if using the activity pacing framework in the clinical setting was 'do-able'. The authors appreciate that not having <i>a priori</i> criteria is a limitation of this study and this has been acknowledged in the Discussion (p34). However, we believe this early feasibility study has great interest and importance in the field of activity pacing and chronic pain/fatigue management. Furthermore, this study lays the foundation for our future pilot RCT which will include a control arm and specific progression criteria to determine whether to progress to the full trial.</p> <p>We have also signposted towards using the findings from this feasibility work to inform the progression criteria for a future pilot RCT on page 31.</p>
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<p>Some of our questions were clearly answered, but the answer itself was regularly disappointing. One example may be sufficient. The authors do not problematize the notion of 'depression', and apparently use the DSM, which was regarded as 'invalid' by the National Institute of Mental Health. So, when an effect size of 1 is reported for depression, we do not know what happened. Does this reflect that subjects became more active? Or that their mood improved? Surely, these are different dimensions.</p>	<p>Thank you for your comments. The authors have explored this, and we were unable to find a statement that the National Institute of Mental Health (NIMH) rejected the DSM. Instead, we noted that on the NIMH website, there is a signpost towards the DSM-5 for a further understanding of depressive disorders: https://www.nimh.nih.gov/health/topics/depression</p> <p>Relating to the NIMH regarding the DSM as “invalid”, we could only find press/bloggers articles (circa 2013) from health magazines, such as ‘GoodTherapy’, but even here the article reads: <i>“..many mental health experts were surprised when the National Institute of Mental Health, which is the largest organization of mental health research, and a significant source of funding for mental health researchers, issued a statement that was harshly critical of the updated manual, the DSM-V. Headlines in the popular press have treated this development as a shock to the mental health world and a complete torpedoing of the DSM. The reality, however, turns out to be a lot more nuanced. The NIMH is not withdrawing support for the DSM-V. Instead, it is developing its own mental health diagnostic system...”</i> https://www.goodtherapy.org/blog/national-institute-of-mental-health-will-not-support-dsm-v-0509137)</p> <p>The NIMH website reports of the development of the Research Domain Criteria (RDoC). This is a “research framework for investigating mental disorders”. It is further stated that “RDoC is not meant to serve as a diagnostic guide, nor is it intended to replace current diagnostic systems.” Since the RDoC comprises of genetic and molecular factors, the use in regular clinical settings may be limited. https://www.nimh.nih.gov/research/research-funded-by-nimh/rdoc/about-rdoc)</p> <p>The authors selected the PHQ-9 as a measure of depression due to its value of being widely used, validated and a clinically relevant measure that has shown sensitivity and specificity (Levis et al. BMJ 2019; 365 doi: https://doi.org/10.1136/bmj.l1476). In the current feasibility study, depression formed one aspect of the complex biopsychosocial factors that may impact on participants' wellbeing, ability to implement activity pacing and other strategies, and a symptom that may/may not improve following treatment. The main focus of this study was not to explore the concept of depression per se. Due to the complex interplay between components of mental and physical health among people living with chronic pain/fatigue, we implemented multiple measures regarding mental health, cognitive function and physical function: anxiety, mental/physical fatigue, self-efficacy and quality of life. The aim at this stage was to explore if any changes might be detected. However, due to the limited sample size in keeping with the feasibility design, further conclusions about treatment effects were not made. Effect sizes were</p>
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included after Reviewer 2's previous suggestion to include these calculations.

<p>The Tables contain a tsunami of results (now with effect sizes, which improves readability), but there is a clear lack of focus (also in reporting relationships), and one cannot but wonder if any correction for multiple testing should have been used, or any other methods to focus on what is really relevant.</p>	<p>Due to the nature of this early feasibility study, exploratory statistics have been used to highlight any changes in activity pacing and symptoms. Table 3 shows these changes between pre-treatment to post-treatment (T1-T2), post-treatment to three months' follow-up (T2-T3) and pre-treatment to three months' follow-up (T1-T3) since this has research and clinical interest to demonstrate changes after treatment and if any changes are maintained at three months. The tables are reported as mean changes with confidence intervals as per the recommendation for feasibility/pilot studies.</p> <p>Following Reviewer 2's comment, Tables 4 and 5 have now been removed from the manuscript. We appreciate Reviewer 2's comment that these tables distracted from the main aim of the study to explore the feasibility of implementing the activity pacing framework in the clinical setting. Future work will explore the effectiveness of activity pacing in a randomised controlled trial.</p> <p>The Abstract and manuscript (Aims, Methods, Results, Discussion) have been amended accordingly to remove any reference to the correlation analyses previously shown in Tables 4 and 5.</p>
<p>The above is for us enough to recommend rejection of the paper. We are not willing to see another version.</p>	<p>The authors thank Reviewer 2 for their time in commenting on this manuscript.</p>