Author's response to reviews

Title: Evaluation of a group based cognitive behavioural therapy programme for menstrual pain management in young women with intellectual disabilities: Protocol for a mixed methods controlled clinical trial.

Authors:

Susan Kennedy (susankennedy2007@gmail.com)
Siobhan O'Higgins (siobhan.ohiggins@nuigalway.ie)
Kiran Sarma (kiran.sarma@nuigalway.ie)
Carla Willig (C.Willig@city.ac.uk)
Brian E McGuire (brian.mcguire@nuigalway.ie)

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Author's response to reviews: see over
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RE: Response to Reviewer’s Reports

Title: Evaluation of a group based cognitive behavioural therapy programme for menstrual pain management in young women with intellectual disabilities: Protocol for a mixed methods controlled clinical trial.

Dear Sir / Madam,

Thank you for taking the time to consider our manuscript for publication. We very much welcome the detailed and informative comments and recommendations received and outline our response below and in the amended manuscript, which is attached:

Reviewer’s Report 1 – Angela Hassiotis

1. Very good introduction! However, some caution about reports of pain and comparisons with peers of normal intelligence is required, given that the study of parents of women with autism or Down syndrome had only 24 participants.

This paragraph in the introduction section of the manuscript has been amended. It now states that due to the small sample size used in the study referred to, due caution is required in the interpretation of the findings.

2. I am uncertain as to whether the study refers to a protocol or a completed study as the text oscillates between what is to be done and what has been done, e.g. see measures/instruments section.
The manuscript refers to a study protocol which at this point is ongoing. For this reason, the text of the original manuscript varied between tenses to reflect the different elements of the research study which were completed, ongoing and yet to be completed. The text of the manuscript has now been amended throughout and consistently written in one tense (past tense), to address the confusion which this approach generated.

3. The study has misinterpreted the reported sample size in Hassiotis et al, 2011. This was a feasibility study and was not intended as a suggestion of what an appropriate sample size for such a trial ought to be. Currently, there is discussion that feasibility trials ought to include up to 60 individuals. As the study is not clear about its aims being those of a feasibility trial, the sample size calculation is not argued for persuasively. It gives the inaccurate perception that it is definitive which it most certainly is not.

The abstract and description sections of the manuscript have been amended to reflect that this is a pilot study, rather than a feasibility study. The study by Hassiotis et al., 2011, was used as a guideline in calculating a sample size due to the lack of treatment studies available to generate a power calculation. The Authors were reliant on the few previous studies available regarding this area of research.

4. The trial does not include randomisation. I am not convinced by the Authors rationale but probably nothing can be done about it at this point. They may wish to look at the Beeken et al, 2013 (Trials) protocol.

The Authors have reviewed with interest, the Beeken et al., 2013 (Trials) protocol. A number of key differences were noted between the two studies, which impacted on the ability of the researchers in this study, to include randomisation:

- The Beeken et al., 2013 study had a population of at least 450 service users to recruit from, at least 180 of whom were likely to be eligible to participate in the study. In our study, both the overall population of young women attending school and number eligible to participate in the study were much smaller. This was also the case with regard to young women, under age 30 years, attending an adult day service.

- The geographical location in which the two studies were completed were very different. The Beeken et al. study was completed with service users from two inner London
community services where a good public transport system is in operation. Our study was completed in an area which includes a significantly rural region, and where public transport is very limited.

- Whilst participants in the Beeken et al. study were all adult service users, more than two thirds of those in our study were adolescents (24/32). As such, the majority of participants were dependent on their parents for transportation. The Authors have prior knowledge and experience of the poor attendance rates achieved within this geographical area when training is delivered in the evenings at a “central” location. Such arrangements are often unsuitable and inconvenient for families, who must drive their son/daughter long distances to attend events and/or arrange child care for siblings. For this reason, training was delivered at schools and training centres as part of participants’ daily programme of activities. The Researchers were keenly aware that the absence of randomisation was a disadvantage to the study, however, it was determined that this was necessary to address anticipated problems with recruitment and attendance associated with transport. As randomisation was not deemed to be feasible in order to pilot the intervention programme, the intervention and control groups were matched on key demographic variables.

- The Beeken et al. study was funded by the National Institute for Health research (NIHR) and involved the delivery of the intervention programme by multiple trained professionals. This study is being completed by one professional, without funding, in partial fulfilment of the requirements for a Post-Qualification Doctorate in Clinical Psychology, whilst working full-time as a Senior Clinical Psychologist. As such, the scope to deliver the intervention to randomised small groups and/or pairs of service users (as in the Beeken study) was limited.

5. In my view, the protocol needs revision prior to being published and should include the aims and objectives based on the definition of feasibility or pilot trials as per NIHR HTA guidelines. In this context, it is also important to use a unified format throughout the manuscript of what the processes of the study will be.

As suggested, the protocol has been revised to include the aims and objectives of a pilot study as per NIHR HTA guidelines. As suggested, a more unified format has also been adopted throughout the manuscript with regard to the processes of the study e.g. recruitment, randomisation, treatment, follow-up.
Reviewer’s Report 2 – Lynn Breau

1. Background

The background is easy to read and helpful, but does not provide a great deal of information to support the use of CBT with this group. The authors should add discussion of research indicating those with mild to moderate ID can participate in CBT delivered in a group format, whether for pain or other issues.

Additional references have been included in the background section of the manuscript which support the use of CBT in a group format, with individuals with mild and moderate intellectual disabilities.

2. Research Hypotheses:

The rationale for Hypothesis 3 is not provided. Why is it expected that parent catastrophizing will affect the participants’ behaviour? Which behaviour is it expected to affect? Please describe relevant studies that support this hypothesis in the Background section and/or this section.

Catastrophizing – experiencing extremely negative thoughts about one’s plight and interpreting even minor problems as major catastrophes – appears to be a powerful way of thinking that greatly influences pain and disability and is important in determining one’s reaction to pain (Sullivan et al., 2001). Goubert, Eccleston, Vervoort, Jordan and Crombez (2006) found that parents’ catastrophic thinking about their child’s pain made a significant contribution in explaining the child’s disability and school attendance. For this reason, parent catastrophizing is expected to affect participant’s ratings of pain intensity, pain interference and pain coping strategies used. Additional references have been included in the background/research hypotheses section of the manuscript to support this hypothesis.

The rationale for Hypothesis 4 is also not described. Please provide a discussion of the relevant literature that supports the expectation that the participants will adopt more behavioural than cognitive strategies in the Background section and/or this section.
Burkitt et al. (2011) found that greater developmental level (rather than chronological age) was associated with the use of more cognitively demanding strategies as may be used in cognitive behavioural therapy treatment approaches. On this basis it was hypothesised that participants in the menstrual pain management group would adopt more behavioural than cognitive coping strategies to manage their menstrual pain. Our own previous work (McManus & McGuire, JIDR 2014) also showed participants were least likely to use the cognitive coping strategies compared with behavioural strategies. This rationale has now been included in the background/hypotheses section of the manuscript and references included to support this hypothesis.

3. Recruitment and Eligibility:

We are told that the participants will provide consent “via a visual participant information sheet and consent form”. It is very likely that most participants will not be able to read. Please explain if the information will also be provided verbally.

Copies of the visual participant information sheet and consent form were provided to each potential participant and read out to them by the Researcher, at the same time.

Inclusion Criteria:

Please describe in more detail what the level of speech will be of participants. Is this being formally assessed? It would be appropriate to know both their Receptive and Expressive verbal abilities because they will be expected to learn during the CBT and to provide verbal responses to questionnaires.

The level of speech of all participants was consistent with their level of intellectual ability, or greater. Part of the rationale for the initial meeting with potential participants was to enable the Researcher to informally assess if participants’ speech and language skills were sufficiently developed to enable them to participate in the group intervention. Although
formal assessment was not completed by a Speech & Language Therapist, formal speech and language therapy data was available for some participants, but not for all of them.

Some of the participants may be as young as 12 years, suggesting they may only have begun to menstruate recently. This means they may have had very few experiences with menstrual pain. This may impact their pain perception, as we know that past pain affects our perception of pain now. It may also impact their ability to take in the information provided during the CBT because they will have had less experience with the situation. How will this be accounted for in the data analyses?

Age 12 years was used as a cut-off point for inclusion in the study as this is the age at which students typically transfer to Secondary School. This enabled delivery of the intervention to participants in the school setting. Only 1 participant in the study was aged 12 years. She started her menstrual period at age 9 years and therefore had significant experience of menstrual pain before participating in the study. All other participants had been menstruating for at least one year. The 12 year old participant was in the control group and therefore her experience did not have implications for her ability to take in information provided during the CBT intervention.

4. Research Design:

Please indicate how the information regarding what is done for the control group will be monitored. It is very important to know if any will be taking analgesics, as this may reduce the difference found between groups.

All participants were asked about their usual menstrual pain care routine, at the start of the study. Usual care included medication for some participants in both the intervention and control groups. This can be accounted for in the data analysis.

5. Data Collection:

Primary Outcome Measures:
The authors report no psychometrics for the Pain Coping Strategies Questionnaire, the Pain Coping Scenarios Questionnaire and the Pain Knowledge Questionnaire and do not report any published analyses to indicate these are valid and reliable. This is a serious problem because they are the primary outcomes. Information should also be provided regarding how they are scored, etc.

The measures chosen were used in a previous study of CBT for chronic pain in people with intellectual disability (McManus and McGuire, JIDR 2014). It was felt that these measures were the most appropriate given the lack of measures available for use in this area. It is hoped that the use of other validated measures in this study will assist in providing data on the validity of the Pain Coping Strategies Questionnaire, the Pain Coping Scenarios Questionnaire and the Pain Knowledge Questionnaire. Information on the scoring of these measures has been included in Table 1 of the manuscript.

Secondary Outcome Measures:

A pain score should be a primary outcome, but is only included here as a secondary outcome. It is likely that a good number of the participants will not be able to use the VAS described reliably. De Knegt et al. (2013) found that only 32%-61% of adults with ID could use a 0 - 10 numerical scale and Fanurik et al. (1998) found that no children with moderate ID could use a 0 – 5 pain scale reliably. Although it is admirable that the authors are attempting to include self-report, and pain is the target of the intervention, they should consider also including a proxy measure by parents. Further, most adults without ID find it difficult to retrospectively report ‘average” pain. Asking about “worst pain” and “least pain” would be better. Also, are the authors sure that pain intensity is the most likely thing that will change? Could pain intensity remain high, but duration of pain reduce? It is advisable to re-evaluate how pain will be measured.

The Authors acknowledge and agree with the reviewer’s comment regarding the challenges of using numerical rating scales with individuals with intellectual disabilities. Proxy measures are also obtained from parents and the manuscript has been amended to emphasise this important information. The use of a visual analogue scale with both participants and their parents will provide information on the reliability and validity of this self-report measure for this population.
The Authors wish to clarify that although the word “average” was used in the manuscript, this term was not used in the administration of the Pain Rating Scale. This term has now been removed from the manuscript. Participants were asked to rate their pain during their last period. It was felt that a neutral statement would minimise potential response bias as it is not a leading question.

The authors concur with the Reviewer that pain intensity could remain high following intervention. Pain intensity was included as a secondary outcome measure as the researchers did not think that pain intensity was the most likely variable to change i.e. they did not think it was likely to be a primary outcome of the intervention. Pain interference was measured in terms of the impact of pain on various aspects of daily living as it was hypothesised that this could reduce but pain intensity could remain high. Pain duration was not measured in the study and has the same potential limitations in terms of reliability of measurement.

The Brief Pain Inventory – Short is a tool designed for people without ID. It is very probable that few of the participants in this study will be able to comprehend and respond on this questionnaire. The use of a 0 – 10 likert response format is also beyond the numerical understanding of most individuals with moderate ID.

A modified version of the Brief Pain Inventory – Short Form was developed for use with people with intellectual disabilities. Modifications included front and back female body outlines to enable participants to identify the areas of the body in which they experienced menstrual pain, changes to the questions and categories assessed and modifications to the response categories including the use of a visual analogue rating scale in conjunction with a numerical rating scale.

6. Supplementary Research Methodologies:

Moderator Analyses:

It is unclear whether the self-efficacy scale will be completed by the participant or the parent. Please provide more detail regarding the questionnaire, and evidence that it has good psychometric properties with adults with moderate ID, or even with individuals who would have a similar mental age equivalent.
A modified version of the self-efficacy scale for child functioning despite chronic pain was used in the study and completed with the participants. Further information on this questionnaire has been included in the manuscript. This questionnaire was used by McManus & McGuire (2014) in their study on the use of CBT for pain management in people with intellectual disabilities and chronic pain. No other suitable self-efficacy scale was available to compare this one to, it was used as an indicator of this construct. Further information on the psychometric properties of the Self-Efficacy Scale for Child Functioning despite Chronic Pain scale was provided by Bursch, Tsao, Meldrum & Zelter (2006). This relates to its’ use with children (who would have a similar mental age equivalent to research participants).

The reference given for the Pain Catastrophizing Scale – Parent refers to the original self-report scale developed by Sullivan. Please provide a reference and describe the psychometrics of the parent version. As mentioned regarding the hypotheses, no rationale is provided to support the idea that parent catastrophizing may predict child pain or child pain behaviour. Please describe some here. Also, please specify what exactly parent catastrophizing is expected to predict (e.g. pain severity, pain coping, self-efficacy?).

Psychometric data for the Pain Catastrophizing Scale – Parent is outlined in Goubert, Eccleston, Vervoort, Jordan and Crombez (2006). In that study, parents’ catastrophic thinking about their child’s pain had a significant contribution in explaining the child’s disability and school attendance. As such, it is hypothesised that parent catastrophizing regarding menstrual pain will impact on the participants pain impact scores as well as their ratings of pain interference, pain self-efficacy and pain coping strategies.

7. Data Analyses:

Quantitative Analyses:

The MANOVA should include baseline scores as a covariate to allow for regression to the mean. Please indicate what type of regression will be used. Please provide a discussion of power / how many predictors will be included. Please outline the specific analyses that will be conducted; what are the dependent variables of interest, Will predictors be entered blockwise/stepwise? What order will be used? Will they be forward/backward entry? Will demographic characteristics (e.g. age, ID level) be included?
Baseline scores will be included as a covariate and a multiple regression model will be used. Predictors will include the demographic characteristics of age and degree of intellectual disability as well as time since onset of menstruation; frequency and duration of menstruation; number and frequency of menstrual symptoms experienced and history, treatment and use of medication to manage gynaecological problems and other medical conditions. A stepwise method of regression analysis will be conducted with predictors entered using the backward method. This method will be used for exploratory model building and will be cross-validated by then splitting the data. The dependent variables of interest will be strategies used to cope with pain and pain knowledge. This information has now been added to the manuscript.

The description of counter balancing is difficult to understand. Is this across the three treatment groups? Please clarify.

This section has been reworded for clarity. Counterbalancing will be applied across the intervention programme to the three treatment groups.

Will the process analyses be conducted by comparison to baseline or to the previous assessment?

Process analyses will be conducted to assess change over time and therefore both comparison to baseline and previous assessment will be completed.

8. Minor Essential Revisions (not for publication):

There are several places where the tense switches from future to present, or, in some cases, to the past tense. This is confusing and should be addressed. If some parts of the data have already been collected, please state so specifically.

The past tense has been used throughout the manuscript.

9. Background:

The discussion of differences in brain structure due to dysmenorrhea is difficult to follow. It is unclear which cited reference contains the results described and it is not clear what the
differences were or if these are long-term or only occur during at specific points in the menstrual cycle. Please re-write this section.

This section has been re-written as suggested.

10. Recruitment and Eligibility:

Please indicate what framework the diagnoses of ID will be based on (DSM vs. ICD). Please indicate the IQ ranges that will be included.

The DSM framework was used for the diagnoses of intellectual disability as this is the system used in Irish services. Participant IQ scores were in the range 35 - 70.

In this section, it is stated that the participants will provide ‘consent’, but later, this is described as ‘assent’. Please be consistent.

Revised to “assent” as requested, to ensure consistency.

Table 1: It would be helpful to include the times of the “time points” (e.g. “baseline, 5 weeks, etc.”).

As suggested, the times of the “time-points” have now been included in the table.

We thank the reviewers again for their careful and expert commentary on our manuscript. We hope that we have adequately addressed the queries and comments of the reviewers and look forward to your response.

Kind Regards,

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Susan Kennedy