PEER REVIEW HISTORY

ARTICLE DETAILS

TITLE (PROVISIONAL)	Developing and implementing a culturally informed FAmily Motivational Engagement Strategy (FAMES) to increase family engagement in first episode psychosis programs: Mixed methods pilot study protocol
AUTHORS	Oluwoye, Oladunni; Dyck, Dennis; McPherson, Sterling; Lewis-Fernández, Roberto; Compton, Michael; McDonell, Michael; Cabassa, Leopoldo

VERSION 1 – REVIEW

REVIEWER	Vyv Huddy University of Sheffield, UK
REVIEW RETURNED	09-Mar-2020

GENERAL COMMENTS	The protocol describes a useful and worthwhile study. The three-phase method is realistic and appropriate. There are a number of areas where the protocol where further consideration might be useful. Self-determination theory is an appropriate choice but other perspectives might have been useful. Other interventions addressing poor engagement have used motivational interviewing techniques (e.g. Lundahl, B. W., Kunz, C., Brownell, C., Tollefson, D., & Burke, B. L. (2010). A Meta-Analysis of Motivational Interviewing: Twenty-Five Years of Empirical Studies. Research on Social Work Practice, 20(2), 137–160). It is not clear how SDT to arrive at the use of “motivational statements”. The justification of monthly motivation sessions is not clear, as these sessions may not be necessary for all families. Some families are already very keen to attend making these redundant. If authors are suggesting low uptake is a factor affecting all families this should be more explicitly stated and justified. Further detail on how the research team will select family members on the advisory committee would be useful. The proposed convenience sample gathered by circulation of a survey link may not garner the most useful information. People who answer a voluntary survey are likely to be much more motivated than the “hard to reach” people the intervention is intended for. Additional purposive sampling my help identify people who are not engaging in the services and then get feedback from them. Identifying people who have previously opted out (dropped out) from previous similar intervention may achieve this. The qualitative section needs further justification of the choice of content analysis over other approaches (e.g. framework analysis). Further consideration to acceptability including unpacking the dimensions of acceptability (e.g. burden, ethicality, opportunity costs). Some of these relate to my earlier point on potential ambivalence that participants may hold about whether to engage.

	A recent paper on the topic might be useful (e.g. Sekhon, M., Cartwright, M. & Francis, J.J. Acceptability of healthcare interventions: an overview of reviews and development of a theoretical framework. BMC Health Serv Res 17, 88 (2017). https://doi.org/10.1186/s12913-017-2031-8) Sample size in phase 2 and 3 should be further justified. The team will recruit five family members in phase 2 but it is unclear why this number is necessary. Given this is a pilot trial; presumably one aim of this is to determine the sample size of a definitive trial. For this reason, further justification for sample size is required. It is also not clear how the research team will use this information to inform the subsequent larger trial. The use of explicit cut off scores for acceptability / feasibility could help judge whether the study has met its feasibility / acceptability objectives. For example, one recent pilot trial used a traffic light system (e.g. Bryant, M., Burton, W., Collinson, M. et al. Cluster randomised controlled feasibility study of HENRY: a community-based intervention aimed at reducing obesity rates in preschool children. Pilot Feasibility Stud 4, 118 (2018). https://doi.org/10.1186/s40814-018-0309-1)

REVIEWER	Geoffrey Dickens Western Sydney University, Australia
REVIEW RETURNED	21-Apr-2020

GENERAL COMMENTS	A very well organised and well communicated protocol. Two minor comments and one request for clarification 1. The brief review pp 6-7 of reasons for low treatment engagement is informative but seems to skirt around the key issue of differential engagement across cultural/race lines. Given these differences, what strategies will you use to ensure adequate representation from these groups in the study? 2. I missed any reference to intention to treat analysis, potentially important given difficulty in retention. Clarification: p6 line 36/7 "non Hispanic black and hispanic family members" is confusing. I have checked NA to replicability above given that the study cannot be replicated until the intervention is developed)

VERSION 1 – AUTHOR RESPONSE

Reviewer: 1
Reviewer Name: Vyv Huddy
Institution and Country: University of Sheffield, UK
Please state any competing interests or state ‘None declared’: None declared

1. Self-determination theory is an appropriate choice but other perspectives might have been useful. Other interventions addressing poor engagement have used motivational interviewing techniques (e.g. Lundahl, B. W., Kunz, C., Brownell, C., Tollefson, D., & Burke, B. L. (2010). A Meta-Analysis of Motivational Interviewing: Twenty-Five Years of Empirical Studies. Research on Social Work Practice, 20(2), 137–160). It is not clear how SDT to arrive at the use of “motivational statements”.

RESPONSE: We have cited work throughout the introduction and methods of previous engagement interventions. Specifically, we present a summary of engagement interventions used in other settings and among other populations in mental health on page 5. “Several of these studies have used techniques that enhance motivation and family engagement.38,43 For example, Nock and Kazdin developed the Participation Enhancement Intervention composed of three major components: 1) describing the importance of treatment engagement, 2) motivational statements about engagement, and 3) addressing engagement barriers.” While we agree that other perspectives such as MI may be useful, the self-determination theory for FAMES was also chosen because it also aligns with the underlining theory for FEP treatment models (such as NAVIGATE) in the US. On page 7 we have clarified the use of motivational statements which have been successful in previous engagement strategies and on page 8/9 we have addressed how motivational statements address the intrinsic motivation construct of the SDT.

2. The justification of monthly motivation sessions is not clear, as these sessions may not be necessary for all families. Some families are already very keen to attend making these redundant. If authors are suggesting low uptake is a factor affecting all families this should be more explicitly stated and justified.
RESPONSE: We appreciate the reviewers concerns related to the monthly motivation component. In a recent study of mine (Oluwoye et al 2020), we found that monthly family psychoeducation appointments are scheduled once per month. We have highlighted on page 7 that the motivational component that occurs monthly, aligns with the frequency of in-person family psychoeducation appointments for CSC programs and is intertwined with existing appointment to promote continued family engagement. While we agree with the reviewer that some families are already engaged in treatment and continuous participate, some families disengage after attending a few family appointments. For example, Glynn and colleagues (2018) that only 29% of families attended 5 or more family psychoeducation appointments during a two-year period, which is referenced in the introduction. FAMES intends to address the challenges with engaging families in family psychoeducation while also addressing barriers and family member concerns by using strategies in-between appointments and during appointments.

3. Further detail on how the research team will select family members on the advisory committee would be useful.
RESPONSE: We have included additional details with how family members were selected on page 8.

4. The proposed convenience sample gathered by circulation of a survey link may not garner the most useful information. People who answer a voluntary survey are likely to be much more motivated than the “hard to reach” people the intervention is intended for. Additional purposive sampling my help identify people who are not engaging in the services and then get feedback from them. Identifying people who have previously opted out (dropped out) from previous similar intervention may achieve this.
RESPONSE: We thank the reviewer for their thoughtful insight and recommendation. We have included additional information on page 10 detailing that how we conducted purposeful sampling strategy to recruit family members who discontinued services.

5. The qualitative section needs further justification of the choice of content analysis over other approaches (e.g. framework analysis).
RESPONSE: We appreciate the reviewers’ recommendation and rather than comparing content analysis to other equally acceptable approaches to qualitative coding and to stay within word count limits, we have now included the following sentence on page 13 that provides a justification for using a directed approach to content analysis. “Due to its flexibility and ability to build on previous research identified in IM step 1 and phase 1, a directed content approach was selected for qualitive data analysis. Using a directed approach to content analysis the open-ended targeted questions will focus

on satisfaction and areas for improvement will serve as initial pre-determined coding categories." The following reference was also used to highlight support the use of directed content analysis: Hsieh H, Shannon SE. Three approaches to qualitative content analysis. Qual Health Res. 2005;15(9):1277-1288

6. Further consideration to acceptability including unpacking the dimensions of acceptability (e.g. burden, ethicality, opportunity costs). Some of these relate to my earlier point on potential ambivalence that participants may hold about whether to engage. A recent paper on the topic might be useful (e.g. Sekhon, M., Cartwright, M. & Francis, J.J. Acceptability of healthcare interventions: an overview of reviews and development of a theoretical framework. BMC Health Serv Res 17, 88 (2017). https://urldefense.com/v3/__https://doi.org/10.1186/s12913-017-2031-8__;!!JmPEgBY0HMszNaDT!6g5iyJfxqjlnrhsK2o_RvN81cp6CA6EVAvUht1Ps7PQHD-m8UZAKIdxgHkb7fh6yjfkSpQ$)
RESPONSE: We agree with this excellent recommendation and agree unpacking the multifaceted layers of acceptability are important. As outlined in the methods section for Phase 2 and 3, we explore family members and clinicians' opinions (attitudes) toward the intervention and experiences (satisfaction) through qualitative and quantitative methods. Based on the reviewers' recommendation and provided reference, and keeping within the current scope of the project, we have included additional acceptability components (e.g., satisfaction, influence of intervention on engagement, intervention burden) to in the qualitative section of Phase 3 on page 15.

7. Sample size in phase 2 and 3 should be further justified. The team will recruit five family members in phase 2 but it is unclear why this number is necessary. Given this is a pilot trial; presumably one aim of this is to determine the sample size of a definitive trial. For this reason, further justification for sample size is required. It is also not clear how the research team will use this information to inform the subsequent larger trial.
RESPONSE: A sample size justification has now been included for phase 3 in the section titled sample justification on page 15, while the justification for sample size in phase 2 is less warranted. Phase 2 is intended to be part of iterative development of FAMES which includes feedback from family members and clinicians that will be used by the 5-person advisory committee to inform modifications prior to the stepped-wedge trial, which is similar to other intervention development studies such as, Collom JR, Davidson J, Sweet D, Gillard S, Pinfold V, Henderson C. Development of a peer-led, network mapping intervention to improve the health of individuals with severe mental illnesses: protocol for a pilot study. BMJ open. 2019 Jun 1;9(6):e023768. OR Cabassa LJ, Druss B, Wang Y, Lewis-Fernández R. Collaborative planning approach to inform the implementation of a healthcare manager intervention for Hispanics with serious mental illness: a study protocol. Implementation Science. 2011 Dec;6(1):80.
8. The use of explicit cut off scores for acceptability / feasibility could help judge whether the study has met its feasibility / acceptability objectives. For example, one recent pilot trial used a traffic light system (e.g. Bryant, M., Burton, W., Collinson, M. et al. Cluster randomised controlled feasibility study of HENRY: a community-based intervention aimed at reducing obesity rates in preschool children. Pilot Feasibility Stud 4, 118 (2018). https://urldefense.com/v3/__https://doi.org/10.1186/s40814-018-0309-1__;!!JmPEgBY0HMszNaDT!6g5iyJfxqjlnrhsK2o_RvN81cp6CA6EVAvUht1Ps7PQHD-m8UZAKIdxgHkb7fh4Gx3686Q$)
RESPONSE: While we appreciate the reviewers' recommendation, cut-off scores for several measures including the Client Satisfaction Questionnaire and the Youth Services Survey-Families which assess acceptability are presented in table 1. These measures will be combined with qualitative interviews with family members and providers to assess acceptability and feasibility. Unlike the recommended paper pre-specified criteria such as the traffic light system have not been agreed to by the funder of this trial.
Reviewer: 2
Reviewer Name: Geoffrey Dickens

Institution and Country: Western Sydney University, Australia
Please state any competing interests or state 'None declared': None declared

1. The brief review pp 6-7 of reasons for low treatment engagement is informative but seems to skirt around the key issue of differential engagement across cultural/race lines. Given these differences, what strategies will you use to ensure adequate representation from these groups in the study?
RESPONSE: We thank the reviewer for their comment, we highlight on page 11 that 55% of families at CSC sites are racial and ethnic minorities. We will over recruit families from sites with larger numbers of racial and ethnic minorities. For instance, 80% of clients served in one of the sites identify as Hispanic and 60% of clients served in another site identify as non-Hispanic Black.

2. I missed any reference to intention to treat analysis, potentially important given difficulty in retention.
RESPONSE: We agree with the reviewer and all analyses will be conducted on the intent-to-treat sample in Phase 3, although it is most commonly associated with randomized trials. We have now included the following sentence on page 15, "At the completion of the stepped-wedge trial in phase 3, analyses on the intent-to-treat sample will be performed (N=50)."

3. Clarification: p6 line 36/7 "non Hispanic black and hispanic family members" is confusing.
RESPONSE: We have rephrased the sentence on page 4 of the manuscript to improve clarity for the reader.

VERSION 2 – REVIEW

REVIEWER	Geoffrey Dickens Western Sydney University Australia
REVIEW RETURNED	03-Jul-2020

GENERAL COMMENTS	Thank you for addressing my previous remarks, the responses are satisfactory.