Author’s response to reviews

Title: PPI in research: a reflection from Early Stage Researchers

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Author’s response to reviews:

Dear editors,
We thank you for the opportunity to review our manuscript and to resubmit it to the BMC Journal of Research Engagement and Involvement. Below you will find a point-by-point response to the issues raised by the four reviewers. Their comments helped us to clarify our messages. The edits within the manuscript are inserted as track changes to signal what has been edited.
We would also like to inform you that our second affiliation changed due to a restructuring at our university in Paris.
We look forward to your editorial decision,
Kind regards,

Alice Biggane
On behalf of the three authors

Reviewer #1:

1. I make the following comments which are intended to be helpful. I trust that they will help in promoting the paper's central message of the importance of PPI in research. General.
Response: Thank you very much for taking the time to review our manuscript and providing your insight and comments. We have addressed your comments below and made the appropriate changes within the manuscript.

2. The background meetings / training sessions at Liverpool and Split are an important part of developing the key message of the paper. A bit more background information about these two events, their purpose, agenda, attendees, conference outcome/resolutions/ would set a contextual scene for the discussion/conclusion.
Response: Thank you for your comment. This have now been further elaborated as suggested and in the context of other edits (made in response to other comments) from line 232-250.

3. ESR. Some more background information about the learning journey of these people would be useful. Are there commonalities/differences across different countries/learning centres?
Response: Thank you for the opportunity to elaborate on this. We have now moved the short introduction of the 15 ERS from the “Journal club” paragraph to the “Background” and further elaborated how the MiRoR training has contributed the learning journey for both groups, as for the ESRs individually, in the context of their different backgrounds and research topics (covered from line 204-228).

4. Naming of researchers within the paper. Best if adopt a common policy. Some people are referenced with/without title, sometimes first name is given / sometimes not. It appears that women are first name surname and men title, first name, surname. Could be 'misinterpreted' as issues of inequality. [??] Equality of gender and equality of role [Researcher/ PPI]. Unless formal protocol dictates, I would simply go for first name, second name [i.e. no title]
Response: We have followed standard naming procedures throughout the article. Using title, first name and surname when introducing the individual. Thereafter we use title and surname only. Professor Paula Williamson is mentioned in the “journal club” section, but as she is an author of this paper we use her initials (PRW), following standard convention. There are no other women mentioned within this article. The authors (all women) are in the “Declaration” section, again standard author naming procedures were followed here by using first name and surname or initials.

Background.
5. Some more information about 'waste in clinical research' at this stage would be helpful. More information is given later on but a bit more at this stage would aid initial understanding. May be a reference to greater detail 'follows'.
Response: Thank you for the opportunity to clarify this. We have now added two paragraphs in the Background section to address this and to further clarify the difference, but context, of research waste, -integrity, and PPI (line 99-140).

6. Para. 3. PPI can occur … etc… Suggest that you add, PPI being involved in 'data analysis' to the list of potential PPI input. I have worked on a project where this was 'invaluable'. [The SHARED Project.]
Response: Thank you for your suggestion. The sentence now reads (line 164-166);
“PPI can occur at multiple stages of the research cycle, from identifying the research question, funding application, design, conduct and analysis, to dissemination and translation of findings into daily health services.”

7. Para. 5 It would be worth mentioning support and training for PPI members in this section. Not to mould them into compliant PPIs, but to enable them to make the most/best of the potential to express their view and be part of the research 'team'.
Response: Thank you for your suggestion. This has now been included and the sentence reads (line 183-187);
“This includes threads such as who to involve as research partners, when to involve PPI contributors, how to access and keep people involved in project, training and support mechanisms for PPI members, follow-up plans and dissemination approaches.”

Actions and Initiatives.
8. Para. 2. End of para. There is mention of 'conversation' … Nice terminology .. But what does it
mean? And how would it 'happen'/ be conducted?
Response: Thank you for your comment. Based on this we have now edited the sentence, it reads as (line 280-284);
“Concerns were raised that patients might not be willing to review items that did not align with their priorities, beliefs and values and conversation ensued about whether the peer review process should be a collaborative process between patients and researchers. This would allow all perspectives to be discussed collectively.”

9. Para. 3. I wonder if the concept of PPI as being part of a research 'team' could be introduced here. On the basis that all the 'players' have their own roles … professional researchers alongside lay members who each bring an equally valid perspective to the table. Together the 'team' produces research which is 'better' than the contributions of the individual parts.
Response: Thank you for your comment. While we agree with what you are saying, this paragraph serves to summarise what Mr Stephens spoke about during his interactions with the consortium and the ESRs. As this specific point was not recorded during these interactions we have not included it in our summary.

Prioritisation of hot topics.
10. Differences between patients and researchers leading to waste. Need to clarify what is meant here. Is it differences in the research priorities which lead to waste or an inability to work together? I guess what is meant, but the script leaves some doubt!
Response: Thank you for pointing this out. The sentence now reads as (line 302-305);
“These differences can lead to irrelevant and redundant research studies and evidence, leading to waste in research, if the outcomes that truly matter to patients are omitted.”

11. Determine, Is it … James Lind Alliance, James Lind Alliance [JLA] or JLA?
Response: Thank you for pointing this out. The abbreviation has now been included directly after the first time the James Lind Alliance is introduced. The abbreviation (JLA) is used thereafter.

Journal Club
12. Clarification is needed about the 15 ESRs. Suggest something like … 'There are 15 ESRs known to be working on this topic. For this project/paper each ESR is given a 'working identifier', ESR1 thorough to ESR 15.'
Response: Thank you for your comment. This paper specifically relates to the 15 ESRs within the MiRoR framework. We have signalled more clearly throughout the manuscript that we are writing specifically about this group, for example by introducing the ESRs earlier in the Background.

Closing the gap; Challenges and Concerns
13. Reference to the quote from ESR 5. Need to clarify that PPI involvement is not about researchers telling the PPIs what to do, but researchers engaging with PPIs in discussing what needs to/could be done. Perhaps this clarification needs to come after the quote.
Response: Thank you for your comment. We asked ESR 5 to provide further comment and to clarify their stance. The section now reads (line 487-496)
“For example, ESR 5 is exploring the impact of mobilising collective intelligence in clinical research planning. They hope to include patients and members of the public in their work, however in a topic that is currently quite new and complex for the researchers involved they struggle to find the appropriate language and explanation to do so: “I plan to involve patients in research planning; however as researchers we struggle to translate the complex process of research planning into understandable language that other researchers and patients and public can contribute to. It is challenging to define the task, and then find a way to describe and visualise the problem to help contributors understand what they have to do”.”
Discussion.
14. ESR training: Part of this needs to be training for the ESRs to develop the skills of 'active listening' to what the patients / public are saying. Sometimes it's not the actual words that people say but the thinking behind the words and what's driving the person to make a comment, which is the important bit. As it stands the text currently implies that the ESRs need to simplify / be clear in what they are saying so that the PPIs can understand. This is 'good' but on its own could be misinterpreted as 'talking down' to PPIs [!].
Response: We agree that the understanding of patients/public is an important aspect of the communication and interaction. We have now added a sentence to emphasize this, which reads as (line 540-543):
“Future training should also stress the importance of researchers “actively listening” to PPI contributors. Understanding the motivations and logic behind a contributor’s comments will enable researchers to ensure a more meaningful collaboration”

Conclusion.
15. First sentence .... The rationale for the importance of PPI as and when necessary is indisputable, .... I would argue that the section underlined is not required, as it implies that 'you' don't always want it [!] .. and that's [I assume] not what you intended to say.
Response: We fully agree and this has been removed. In accordance with other edits, the sentence now read as (line 569-570):
“The rationale for the importance of PPI is indisputable, for both research value, quality and integrity”

Competing Interests.
16. No other authors … ? There must be a better/ different way of expressing what's meant here? .. or is it that? What about,
* There are no other competing interests. ?
* All the competing interests of the authors have been declared. ?
Response: We have removed “other”. The line now reads “No authors have any competing interests”

RICHARD J GRANT
20 MARCH 2019

Reviewer #2:
This paper is a reflective piece, from early stage researchers, on a PPI training event. Overall I thought the piece was well written and would be of interest to, in particular, other early stage researchers. I would though like to see the authors give some consideration to the points below. The major points are points 1 to 4
Response: Thank you very much for taking the time to review our manuscript and providing your insight and comments. We have addressed your comments below and made the appropriate changes within the manuscript.

1. The abstract highlights the importance of PPI to research integrity and ethical reasons. Yet the Background section seems to focus on 'waste in clinical research' (and without explaining what is meant by 'waste'). I'm not sure how waste relates to research integrity and ethical reasons - is 'waste' a third issue or is it something that is linked to research integrity or ethics?
Response: We agree that concepts of research waste, integrity and ethics were not entirely clear. We have now elaborated what “research waste” is and how this is related to research value, integrity and PPI in the background section (covered over line 99-158). We have furthermore limited the commentary to include issues only related to research waste, value and integrity by excluding sentences regarding ethics because we, during the revisions, realized that we think of this as another separate topic. Thus, to address ethics, would be beyond the scope of this commentary.

2. Related to point 1 above - I’m not quite sure what the authors mean when they speak of PPI being 'important for both research integrity and ethical reasons' (abstract). When they do cover research integrity (p7-8) they don't fully explain how PPI can contribute to research integrity and they focus more on 'waste in research'. I'm not clear at all on what the authors mean by 'ethical reasons' - do they mean that 'waste' is an ethical issue?

Response: We agree that this was not clear. We have now elaborated what is meant by 'important for both research integrity and ethical reasons' in the abstract conclusion. This now reads as (line 91-92)

“As with intervention research, PPI is also important for methodological research since this will help to increase both the value, integrity and quality of research.”

As also addressed in the previous comment, we have now limited the manuscript to concern only research waste, value and integrity. Together with above edits, we believe that the meaning of research waste and research integrity and the contribution of PPI is more clear now (covered over line 99-158).

3. The importance of PPI -I think the researchers could add in two more rationales re the importance of PPI. Firstly, that PPI can improve the quality of research. Secondly, the moral imperative - the public /patients should have a voice in research that will impact upon them. (See for example INVOLVE's Briefing notes for researchers' - available from their website)

Response: We fully agree that these aspects of PPI are important and were lacking. We have now added this to the Background section (line 144-150) and re-ordered the structure of the Background section to improve the context and clarity.

4. Conclusion - this needs strengthening. eg what is it that you would like to see happen re education and training in PPI for early stage researchers?

Response: We agree that ‘a way forward’ was missing in the conclusion. To address this, we have added some specific suggestions on what we believe could be done in the conclusion, and now reads as (line 576-580).

“To facilitate this opportunity for others ESRs outside of network such as MiRoR, we believe that PPI should be a fundamental educational topic in the academic graduate curriculum, supported by universities via courses and seminars on PPI alongside other scientific skills that are required of an early stage researcher.”

5. Background para 3 - 'PPI in research has hinged on ensuring patients are equal stakeholders etc'. I'm not sure that I would agree with this. 'hinged on' is the phrase that I'm struggling with. It either needs explaining more or changing slightly eg 'much of the focus of PPI has been on ensuring patients etc' - as long as this can be backed up.

Response: Thank you for your suggestion. We have replaced “hinged” with “mainly focussed” and it now reads as follows (line 167- 169);
“To date, PPI in research has mainly focussed on ensuring patients as equal stakeholders in an expert-dominated environment and integrating their lived experience and knowledge into clinically scientific studies.”

The references provided support this statement.

6. Background - para 4. 'There are numerous methods' and 'the most appropriate method'. I don't think 'method' is the right word here. Perhaps 'ways'? And rather than 'dependent' perhaps 'influenced' might be a better word.
Response: Thank you for your suggestion, we have changed the sentence and it now reads as follows (line 174-177);
“There are numerous ways of facilitating PPI including community involvement, presence on committees or management groups, patient research partners; the most appropriate of which is likely influenced by the specific research question, health condition, population, and the available resources.”

7. p5 - top of the page. 'threads and principles'. I would avoid using the word 'principles' - unless you are going to identify some principles. So either 'common threads' or 'considerations'. You then go on to call these 'items' - call them threads or considerations.
Response: Thank you for your suggestions. Principles and items have now been removed and replaced with threads (line 182-187).

8. p6 Actions and initiatives. First line. Lose 'within different health conditions.'
Response: We have now removed this as suggested.

9. Typo - line 30 I think that should be 'our own research' rather than 'their own research'
Response: Thank you for pointing this out. We have now changed it as suggested.

10. "research on research team" - this needs explaining more. ANd is there PPI involved in this?
Response: Thank you for pointing this out. The MiRoR team has now been described in more details and clarified that PPI was involved at several training events (covered over line 204-238).

11. p12 'may no longer be representative of the typical patient.' I'm not sure there is such a thing as a 'typical' patient. Furthermore it is unreasonable to expect a single patient to be 'representative - they are 'a' voice rather than 'the' voice - see the work of Kristina Staley. At best they can be asked what they think might be the view of other patients.
Response: Thank you for your comment. We agree with you that it not possible to ask patients to be representative of all. We have edited the sentence based on your comment as follows (line 507-510):
“However, while it was acknowledged that it is not possible to ask PPI contributors to represent all patients; concerns about being as inclusive as possible remained, in particular considering ways of reflecting the diversity of the patient community and including under-represented patient groups.”

Reviewer #3:
General Comments
I thought the paper was interesting and addresses an important issue: how to involve the public in methodology-focused research. Whilst the paper described discussions at the MiRoR meeting and subsequent discussions, I did feel there should be more clarity on what the next steps should be (e.g. what specific training/support could be developed/provided for this group and who could develop this; what topics should be incorporated in this training/support; how this can be rolled-out) and what further
research needs to be undertaken to better understand this area. An important consideration could be how to involve members of the public in designing and delivering this training/support.

Response: Thank you kindly for taking the time to review our manuscript. We are grateful for your valuable input. We agree that this aspect was missing from the manuscript. Also in accordance to other comments, we have now added more suggestions on the next steps in throughout the discussion and also added some specific suggestions to the conclusion (line 576-580).

I do think that the paper would benefit more from a more detailed explanation early on of methodological research and identifying how this research differs from clinical research and how this presents a challenge for researchers involving public contributors. I didn't feel that this was addressed properly until p.9 (in the Journal Club section). Moreover, whilst the focus is on early career researchers, discussions and strategies developed for involving the public in methodological research would be of benefit to researchers at any stage of their career.

Response: We agree that an introduction to methodological research was missing. We have now elaborated what methodological research is and the difference to primary research in the Background section (line 133-140). Regarding the importance of PPI for all stages in research, we agree and this have now been added to the discussion section (line 564-566).

As the focus is on Early Career Researchers, I think there needs to be greater exploration of what specific needs this group have and how these can be addressed. You do state that these are the 'next generation' of researchers, which is a very valid point, but are there any other considerations about this group?

Response: We have now added a reflection on this in the discussion section (line 560-563):
“We also believe initiatives specific to ESRs, such as consideration of supervision from a senior with PPI experiences or that all ESRs and supervisors could consider whether having a Patient Research Partner would be beneficial to the research.”

Towards the end of the paper, you discuss engagement with the wider public with research. You describe the need for researchers to increase their skills and develop a range of strategies for engagement. Whilst this is important, I did feel there needed to be a stronger link to the main focus of the paper - public involvement rather than engagement - and I did feel that there was a great opportunity missed here. In research projects with a strong methodological focus, there are great benefits to working in partnership with patients and the public to communicate these messages effectively and disseminate findings.

Response: Thank you very much for pointing this out. It is indeed correct that our focus is on involvement rather than engagement. To avoid confusion and emphasise on the involvement, we have now replaced the “engagement” with “involvement.

Specific comments are detailed below.

Abstract:
- I think you need to be clear here that MiRoR has a focus on methodological research. You do spell the acronym out (in the Main Body), but it's not immediately obvious to an unfamiliar audience the nature of this research. I think some examples of the types of research researchers in this field undertake would be beneficial.

Response: Thank you for your comment. We now spell out the acronym earlier in the abstract and signal that it is methodological research. However, because of word limitations (max 250) we have not
added them to the abstract.

- In the conclusion, I think you need to highlight the fact this is training for early stage researchers in the field of methodology research. There is a wealth of training on involvement that is accessible to researchers, but as you highlight in the paper, it can be more challenging to conceptualise public involvement in some of these projects.
Response: We fully agree and have now added a sentence to the conclusion (line 570-573).

Plain English Summary
- 25-26 - I think this would benefit from more detail about 'Methods in Research on Research' with a few examples provided. This would give readers a better understanding, early on, about the nature of research projects this group focus on.
Response: Thank you for your suggestion we have now included the following sentence to the plain English summary (line 51-53);
The Methods in Research on Research (MiRoR) project is a consortium brought together by their interest in tackling waste in clinical research, by exploring topics for methodological improvement across a range of clinical research areas.
Due to word limitations (max 250) we have not added specific examples to the summary.
- 27 - suggest 'The outputs enabled the ESRs that attended the event to learn more about the value of PPI …'
- 28 - I found the phrase 'consider it within their own research' to be a bit weak. I think the statement could be stronger - rather than just consider whether to involve public contributors, the statement should reflect when and how public contributors can be involved in research to add value.
Response: Thank you for your recommendations 27 & 28 above. The section now reads as (line 54-57);
Drawing on a recent PPI training event held by the MiRoR project, we describe the learning outputs that we, the 15 MiRoR ESRs attending the event took home with us. The outputs enabled us as ESRs to learn more about the value of PPI and when and how public contributors can be involved in methodological research.
- 31 - I would remove 'in short'
- 43 - I would use 'enhance our partnerships with public contributors'
- 43 - For the plain English Summary, I would use 'share' rather than 'disseminate'
Response: Thank you for the wording suggestions in 31, 43, 43 above. We have made the changes as recommended.

Background (p.4)
- 5-6 - What about waste of public resource/charity funding that is used to finance the research?
Response: Thank you for your suggestion. In accordance with other comments, we have now elaborated on research waste as originally identified by Chalmers and Glasziou (2009) This section has now been amended to read as follows (line 99-105);
In 2009, Chalmers and Glasziou estimated that 85% of all clinical research is wasted despite large financial investments, including public funding [1]. They identified four stages within current research practices that lead to waste in research including: i) prioritising research questions that are irrelevant to health professionals and patients ii) conducting unnecessary or inappropriate studies or study designs, iii) failing to publish research findings and iv) selective reporting of research findings [1].
- 13-14 - I'm not sure that the word 'guiding' does justice to the contributions of public contributors. You could consider using 'shaping' perhaps?
-17 - I think you should use the correct definition of involvement from NIHR INVOLVE (INVOLVE defines public involvement in research as research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them.). This is the definition most people would be familiar with and it can be accessed here: (https://www.invo.org.uk/find-out-more/what-is-public-involvement-in-research-2/)

Response: Thank you for pointing this out and providing the reference. We have now cited INVOLVE’s definition (line 148-150) using the existing reference as this also included the definition.


Whilst i appreciate that this is an International audience, this guidance has been quite impactful.

We thank the reviewer for bringing this to our attention and agree that it is in important initiative. We have now added a sentence about this guideline (incl. its reference) in the context of future training resources as a future training resource (discussion section, line 543-546):

As a number of ESRs pointed to uncertainty about how to implement PPI in research future learning opportunities could look specifically at ways of implementation such as co-production, we note such guidance already exists for PPI in primary research

26-33 - In terms of what funders are looking for, presumably the relevance of a particular research project to members of the public is also important. It might also be worth mentioning at this point that many funding bodies have lay reviewers to review bids who will scrutinise how the public have been involved in shaping the design of the study and how they will be involved in the project moving forwards.

Response: Thank you for your comment. We agree with your suggestion, and believe the Actions and Initiatives section of Dr Westmore’s presentation covers this. In this section we say (line 339-347):

Every year the NIHR receives thousands of funding applications, however, it is not always a straightforward task to prioritise the research projects and decide which ones to fund. Thus, the NIHR work with a multidisciplinary panel of experts including the public, to consider applications. The feedback received leads to the creation of a short-list for further evaluation. In this process, one key factor for consideration is the impact of the research, i.e. what will the impact for patients be and how to maximise it. It is the hope of the NIHR that “every patient in the country has the opportunity to get involved in research”.

46 - You talk about the 'developing area' of PPI and you could mention here that there are attempts to improve the experience of involvement. For example, NIHR INVOLVE have published some Standards on Involvement (https://www.invo.org.uk/posttypepublication/national-standards-for-public-involvement/) and papers have been published recently on considerations for the ethics of involvement (e.g. Pandya Woods https://researchinvolvement.biomedcentral.com/articles/10.1186/s40900-017-0058-y)

I did also feel that there would be benefit to mentioning current efforts to better report public involvement in research (e.g. Staniszewska https://www.bmj.com/content/358/bmj.j3453) so people are aware of how public have been involved in research and what works/doesn't work in what contexts (thereby further reducing research waste).

Response: We are very thankful for all these references. We have now included the reference of National Standards for Public Involvement and the paper of Staniszewska (line 171-173). Regarding the ethics (paper of Pandya Woods), we think this is outside the scope of the research waste and
integrity and for this reason, we preferred not to include it. Please see response to reviewer 2’s comment 1 and 2.

53 - I wasn't clear on what the term 'participant involvement' meant. NIHR INVOLVE has a clear distinction between research participants and those involved in research. I wasn't sure if you meant user-researchers here?
Response: Thank you for pointing this out. We have now removed this from the sentence to avoid any confusion. It now reads as (line 182-177):
There are numerous ways of facilitating PPI including community involvement, PPI presence on committees or management groups, patient research partners; the most appropriate of which is likely influenced by the specific research question, health condition, population, and the available resources

56 - I would use 'one size fits all approach' rather than 'mechanism'. I also think that you need to emphasise that the approach to meaningful public involvement should be determined through ongoing discussions between the researchers and public contributors about what is most appropriate/relevant.
Response: Thank you for your suggestion. The sentence has been amended to read as follows (line 177-180);
However, despite no one-size-fits-all approach, we believe that involving public contributors, unless there is a good reason not to, should be the default position of all research teams.

(p.5)
4-7 - The language here is very 'research' focused and can be very confusing. Rather than 'what patients to recruit as research partners', why not say 'who to involve as research partners', 'when to involve people', 'how to get people involved', 'how to keep public contributors involved in the project'.
Response: Thank you for your suggestion. The sentence now reads as follows;
This includes threads such as who to involve as research partners, when to involve PPI contributors, how to access and keep people involved in project, training and support mechanisms for PPI members, follow-up plans and dissemination approaches.
Line 183-187

21- You introduce Early Stage Researchers at this point. A clear definition of this group would be welcomed.
Response: Thank you for your comment. We have added the following sentence (line 196-197).
ESR’s are typically researchers in the first four years of their research activity, including the period of research training.

27 - You talk about researchers producing 'more valid and relevant research evidence'. I feel a reference would be merited here. You have included Brett and Staniszewska (2014) in the references, but there are others that could be used (e.g. Wilson et al, 2015, RaPPORT study - https://core.ac.uk/download/pdf/30710021.pdf) I also think you could make a stronger point here: earlier training and skills development on value of public involvement, and adopting these practices early on, will help involving public contributors become a natural way of working rather than a 'tick box' exercise that needs to be done in order to secure research funding.
Response: We thank the reviewer for the reference and this is now included (line 203). We also fully agree that the practice needs to be normalised by for example, earlier introduction to researchers. We have now added a sentence about this, which reads as (line 197-203):
Hence, it is important that they develop and foster PPI-related awareness, skills and expertise. This will enable them to champion meaningful PPI, and to produce more valid and relevant research evidence
Moreover, if the skills and values of PPI are introduced earlier in their training, it will help them adopt these practices better and thus, the involvement of public contributors will become a natural way of working, rather than a ‘tick box’ exercise.

7-8 - More detail about the nature of training to public contributors would be welcome (or a link to further information).
Response: We have now added to the discussion that future initiatives should include research on training to PPI contributors in methodological research and reads as (line 540-548):

Future training should also stress the importance of researchers “actively listening” to PPI contributors. Understanding the motivations and logic behind a contributor’s comments will enable researchers to ensure a more meaningful collaboration. Yet, more research on training to PPI contributors in methodological research is also needed to improve the current practice.

11-12 - In addition to exploring how the role of the public contributor is different to the role of the researcher, was there exploration of how the knowledge of the public contributor complements the knowledge of the researcher and how both contributions are necessary to co-produce the research?
Response: Thank you for your question. We agree that both contributions are necessary to co-production of research. In methodological research, the researcher knows about the methodology whilst the relevant knowledge of the public contributor may vary and the aims of PPI in the methodological research may vary more. We explored this in journal club in which discussed the varying needs across the ESR projects (covered over line 387--483).

15-16 'There was conversation about what are the most appropriate studies and time points in which PPI should be utilised'… I think that there are some people that would take issue with this statement. I think that there is a case to be made for involvement in all projects, but detail on how and when public contributors are involved is very much dependent on the nature of the project and where the public contributors and researchers feel public involvement can add most value.
Response: We agree that PPI should be the standard also for methodological research, unless there are is good reason not to. The key question is at what stage and how. We have now added a sentence about this, which now reads as (line 147-181):

There are numerous ways of facilitating PPI including community involvement, PPI presence on committees or management groups, patient research partners; the most appropriate of which is likely influenced by the specific research question, health condition, population, and the available resources [16]. However, despite no one-size-fits-all approach, involving public contributors should be the default unless there is a good reason not to. Thus, ensuring PPI in research is standard practice, yet not forcing a wrongful fit.

Prioritisation of hot topics
36 -48 - I think there needs to be more discussion/explanation about the James Lind Alliance and the Priority Setting Partnerships. You could provide more detail on the thorough process of reaching consensus on the priorities and the range of conditions. However, it's important to be aware that this is an international audience and not all conditions have had a Priority Setting partnership, so other methods of identifying research priorities should be considered. This doesn't, necessarily, have to be on a grand scale: it is important, however, for researchers to establish that the research is of relevance to patients/public and that the research outcomes will benefit patients/public.
Response: We agree that elaborating more on JLA would be interesting. However, we think that such details are outside the scope of this commentary. Also because this section aims to summarize the talk
of Mr Stephens and we would like to avoid adding content that was not included in his talk. For these reasons, we have preferred to leave out further details.

Actions and Initiatives (p.8)

36 - 37 - When you talk about public involvement aims of the NIHR, you could mention initiatives such as 'People in Research' (a platform for people to find out about opportunities for involvement) or perhaps 'Patient Research Ambassadors' (organised by the Clinical Research Network). You could also highlight NIHR campaigns such as 'I am Research' https://www.nihr.ac.uk/news-and-events/support-our-campaigns/i-am-research/ that are part of the strategy to increase awareness of research.
Response: Thank you for raising this. We have considered your suggestion and decided to include it as follows (line 349-352);

While not a part of Dr Westmore’s presentation it is worth noting that the NIHR also has initiatives and strategies to support PPI contributors in becoming involved in research studies and raising research awareness, including the 2018 “I am Research” campaign.

38 - Whilst the RDS do support researchers in applications, which includes supporting researchers to do meaningful public involvement, I would remove the words 'to help meet this goal' at the start of the sentence because this might be confusing.
Response: Thank you for pointing this out, we have followed your suggestion and removed this.

Mind the Gap

54 - You mention 'JLA' but you haven't mentioned the acronym earlier when you discuss the James Lind Alliance.
Response: Thank you for pointing this out. The abbreviation has now been included directly after the first time the James Lind Alliance is introduced. The abbreviation (JLA) is used thereafter.

Journal Club (p.9)

21-22 - You mention here the challenges of involving public contributors in the projects the MiRoR group undertake and how these projects differ from clinical research projects. I think this is very important and needs to be brought out much earlier in the paper.
Response: We agree that this is important and have now addressed this difference between primary and methodological research in the Background (line 133-140), also as response to one of reviewer 3’s general comments.

P 10

In highlighting the examples from ESR 13 and ESR 15, you describe the ways that data collected from patient participants would enhance the studies. Whilst I don't disagree, the focus of the paper is public involvement and you could explore whether discussions in the journal club explored how more meaningful public involvement at an earlier stage of the research cycle may have prevented these groups from being excluded from the initial data collection.
Response: We fully agree with the reviewer, that this distinction seemed to be confused for some. We have now added a paragraph to JC section to highlight this point and that it argues for earlier introduction. This reads as (line 469-476):
ESRs appeared to confuse the juncture between involvement and participation, when thinking about PPI in their own projects. For those who had not considered PPI in their projects prior to this session they talked mainly in terms of how they could ask patients to participate in their research, rather in terms of involvement. This in particular points to the need for ESRs and other researchers to be aware that PPI can and should start much earlier in the research process, and is not solely reliant on whether the research will include direct patient participation.
50 - In discussing ESR 2, is it worth exploring why the group felt that there was 'no scope' for PPI. You highlight in the paper the importance of public in prioritising research and identifying research relevance and also in sharing research. Were these opportunities for involvement explored at all in the discussions in the journal club?
Response: We have now clarified this the argument of the ESR2, which now reads as (line 432-441):
To date the project has focused on documenting what study designs have been used in previously conducted studies. “As this was a literature study, I don’t think there was a meaningful scope for PPI, as I find it unlikely that the study’s impact on the public/patients and vice versa, would have been different by including PPI. Mainly due to the descriptive nature of this study”. However, I think that other stages of research in medical test evaluation should consider including PPI. E.g., for defining a “decision” (risk/benefit) threshold for when a patient would be willing to receive a diagnostic or screening test. Because, in contrast to our initial literature study, this will most likely have implications for the studies that directly evaluate a test/biomarker”.

p.11 - 1 -4 'For some projects however, the purpose of PPI in the design and conduct of the research is not evident, particularly those with a strong statistical focus'. I. think that there. are some that would challenge this statement and, as mentioned above, there is scope for public involvement in identifying the relevance of a research project and in sharing the findings. Hannigan (2018) has published an interesting paper on public involvement in quantitative research, that can be accessed here: https://onlinelibrary.wiley.com/doi/full/10.1111/hex.12800
Response: While we acknowledge that there are some who will challenge this statement what we are conveying in this section is this the reflections and attitudes of ESRs, in particular the ESRs responsible for the project. However in light of your comment we have added the following sentence (line 450-454).
“However, after the training and discussion during the journal club the ESRs do acknowledge that various research projects have included PPI contributors in quantitative projects such as Hannigan et al (ref provided), and that with the better planning, training and collaboration of both researchers and PPI contributors there is potential for successful collaborations.”

Closing the gap - Challenges and concerns p. 12

20-21 - Whilst there are ongoing debates about 'professional patients' and representativeness of public contributors, I do feel that you also need to highlight the importance of public involvement reflecting the diversity of communities and challenges of involving those under-represented in research.
Response: Thank you for your comments. We have now added the following sentence to the section (line 507-510);
However, while it was acknowledged that it is not possible to ask PPI contributors to represent all patients; concerns about being as inclusive as possible remained, in particular considering ways of reflecting the diversity of the patient community and including under-represented patient groups.

29-30 - In exploring the various skills required at different stages of research, you could also consider the different roles of public contributors. A useful paper to consider is Crocker (2016), which explores different 'types' of public contributors and how they can add value to the research process: https://onlinelibrary.wiley.com/doi/full/10.1111/hex.12479
Response: We agree that this is a relevant point. However, we feel that the perspective of the PPI’s point of view is outside the scope of this commentary. For this reason, we did not elaborate further on this, but instead inserted the reference in a paragraph concerning some examples of efforts that has
been made to improve PPI (line 169-173)
Efforts to improve PPI, such as the experience and reporting of involvement and how different types of public contributors can add value, have been made [21-23], yet it is still a developing area and with much debate about its definitions, methods, operations, research integrity and ethical standards [24].

Discussion

52 - Consider using 'impact on patients' rather than 'patient impact'
Response: Thank you for the suggestion. We have amended accordingly

54 onwards - The sharing of research is an important point and I would feel that this would benefit from more detailed exploration in the main body of the paper rather than in the discussion. Moreover, I think the role of public contributors in working in partnership with researchers to communicate research effectively needs much more development.

It is not clear whether the flyers and videos about the ESR research are co-produced with public contributors. If not, isn't this a missed opportunity?

Response: Thank you for your response. We have considered this suggestion and forwarded to the other ESRs in MiRoR who will try to co-produce these items moving forward. One definite line of co-production based on your suggestion is ESR3 and we have added the following to the discussion section to reflect this (line 552-557):

Further examples of this include ESR 3’s qualitative interview study with patient participants. ESR3 will co-produce a plain language summary of the results for the use of patients and the public with the People and Patient Participation, Involvement and Engagement (PoPPIE). Further, PoPPIE will be able to assist in disseminating this summary to appropriate patient organisations and groups.

Reviewer #4:
Thank you for submitting your commentary which I enjoyed reading. I particularly welcomed your clear structure and writing style and examples to illuminate points made. The topic is of interest to a wide readership including those who like me, are interested in gaining an appreciation of the benefits an initiative such as MiRoR can bring to the Early Stage Researcher community. The particular focus on your recent workshop in Croatia where wasteful research was considered, deserves a whole commentary on it as it affects many researchers and the communities they serve. For me a key strength of a paper is whether it makes the reader think or act differently and it has heightened my awareness of my own need to minimise wasteful research. In my opinion the commentary is close to publication standard and there are very few changes I would suggest.

Response: Thank you very much for taking the time to review our commentary. We are grateful for your input and comments.

On page 9 line 54, you say the example given is obvious. To who? Either explain why it is obvious or better still say it is a good example and justify.
Response: Thank you for your comment. The sentence now reads as (line 396-399);

This project is the most prominent example of where there is a role for meaningful PPI in its conduct, as the research directly investigates the perspectives of patients and members of the public and explores methodological aspects in an attempt to improve their experiences and participation in COS development.

On page 10 lines 30-38 did make me wonder if the project had captured attitudes and beliefs of the Early Stage Researchers before the course for comparison later? No problem if not. Their reflections
are important nonetheless. Just check that the quote you use best illuminates the point you are making. 
Response: Thank you for this suggestion. Unfortunately, we did not capture this, thus, we have no comparison.

On page 11 lines 1-12 I would reverse the ordering as you talk about 'the latter' before talking about 'the former'.
Response: Thank you for this point, we have now reversed the order (445-450).

On page 11 lines 15-30 it is suggested that this research type makes PPI unfeasible. Some studies lend themselves more readily than others for sure. Some patients are interested in hearing about reporting of research and its occasional distortion and some read research reports directly or access a plain language digest of a study or institution's programme of research. As you say yourself on page 11 line 45, there are dissemination opportunities as a minimum. Therefore if a patient is affected somewhere down the line there is an opportunity for PPI. What I would add is that just like we have seen assistive technology developed then a use for it sought/forced, we don't want to force a fit of PPI where it is not likely to be beneficial. There is a risk of wasteful PPI just like wasteful research perhaps?
Response: We agree that PPI should be the standard also for methodological research, unless there are is good reason not to, as this would risk wasteful PPI. Then, the key question is at what stage and how. We have now added a sentence about this, also in the context of response to Reviewer #3 above (line 177-181).

However, despite no one-size-fits-all approach, we believe that involving public contributors, unless there is a good reason not to, should be the default position of all research teams. Thus, ensuring PPI in research is standard practice, yet not forcing a wrongful fit.

On page 12 lines 1-19 you note that some research can be less accessible for public members wishing to be involved and this is where experienced PPI representatives can be helpful. On line 19 being 'representative' is not the goal and there is argument against seeking a 'typical patient'. There are times when a highly trained or experienced PPI representative is what is needed and I would always steer away from the term 'usual suspect'. It is good that the group are aware that tokenism exists and can hopefully steer away from this, instead thinking carefully about who to involve and why (what experience do they have, what characteristics, what role are they needed for etc). Also i'd like to flag how family carer's perspectives are often overlooked - they are often supporting the patient and so have valuable insights or there are implications for themselves. They shouldn't usually speak for a patient either.
Response: Thank you for your comment about a section that created a lot of discussion during our meeting. We believe further information and training to guide ESRs in this area is of great importance and are hopeful that this manuscript will communicate that.

On page 12 lines 31-35 I am unclear what you mean so perhaps rephrase? I understand funders may have advice on use of lay language but i do not know what guidance and support they could give to researchers with regards to recruitment and engagement in research and peer review.
Response: Thank you for your comment. We have now clarified this sentence and it reads as follows (line 517-521);
The group also discussed the need for guidance and support strategies for researchers from relevant organisations such patient representative groups and funders. The ESRs discussed the importance of receiving support and guidance specifically on lay language and communication, recruitment and engagement for a wide representation of patients in research and peer review.
Your Discussion section is appropriate. You rightly point out the challenges and merits of PPI in methodological work (page 12 line 44). It is good that awareness has clearly been raised amongst workshop participants and influenced them earlier in their careers to think about PPI and to do this at an early stage in their research planning.
Response: Thank you for this positive feedback on our discussion section.
The question I am left with is what is the likelihood of many future researchers being able to undertake development around PPI in such an intensive and possibly costly way as the MiRoR project? This makes it imperative that you disseminate well so that as many others as possible can learn through your experiences. This commentary will go some way to achieving this. Thank you.

Response: Thank you for your encouraging comments about our manuscript. We agree that it might be difficult for other researchers to receive the same opportunities as MiRoR. However, we hope this commentary will help raise awareness of the importance of this education and training for ESRs, leading to a conversation about how best to address this and provide opportunity.

We have further amended our conclusion to reflect this and enforce what we believe are vital learning opportunities for researchers. (line 574-580).

Nonetheless, education in PPI for ESRs such as the training provided by MiRoR is essential to increase understanding and enhance skills for its proper implementation and needs further prioritisation. To facilitate this opportunity for others ESRs outside of network such as MiRoR, we believe that PPI should be a fundamental educational topic in the academic graduate curriculum, supported by universities via courses and seminars on PPI alongside other scientific skills that are required of an early stage researcher.