PEER REVIEW HISTORY

ARTICLE DETAILS

TITLE (PROVISIONAL)	Priority setting partnership to identify the top ten research priorities for the management of Parkinson's disease
AUTHORS	Deane, Katherine; Flaherty, Helen; Daley, David; Pascoe, Roland; Penhale, Bridget; Clarke, Carl; Sakley, Catherine; Storey, Stacey

VERSION 1 - REVIEW

REVIEWER	Alex Pollock Nursing, Midwifery and Allied Health Professions Research Unit, Glasgow Caledonian University, UK I led the JLA life after stroke PSP (completed 2012). No other competing interests.
REVIEW RETURNED	03-Sep-2014

GENERAL COMMENTS	While the methods are fairly clearly described, a number of different stages were completed and readers who are not already familiar with these stages may find this difficult to follow. The addition of a simple figure to illustrate the key stages involved in a JLA PSP would be very beneficial and aid clarity. Currently the methods section titled "prioritisation" describes a number of different stages (i.e. checking of uncertainties, interim prioritisation stage, consensus meeting), and it would be helpful to have these more clearly illustrated. The strength of this process is in the systematic nature of prioritisation, and this might come across more clearly with less text and an illustrative figure. The one stage of the process where I feel transparency is currently lacking is at the stage of identifying the top 26 priorities. I do not understand how you got from the top 26 priorities for each of the 4 sets of participants to the "top 26 for all participants". Similarly there is a lack of information on this stage in the results section. Did the final top 26 priorities include a certain number of questions from each of the 4 sets of participants? How many of the top 26 questions represented questions submitted by PwP, carers etc? I would also like more information about WHO dealt with the collation, coding/labelling, PICO transformation. Although in places a number of authors are identified - was this done by a single person, or was duplicate checking performed. This information is important to enable me to assess the potential risk of bias at this stage in the process. I would like to congratulate the authors on a really thorough and comprehensive JLA prioritisation project; the level and amount of consultation is particularly impressive. I believe that the findings of this project are highly important to decisions around the prioritisation

	and planning of future research relating to the clinical managment of PD. However I do not understand the decision of the steering group to exclude the (18) submitted uncertainties which were "deemed to be unlikely to be important enough to reach the top 10 priorities". This appears to be a decision which is potentially open to the biases of the steering group members, and undermines the efforts to ensure that the prioritisation process was carried out by people affected by PD (and not researchers). In practice it is likely that the steering group will have been correct in their opinion that these questions may not have reached the final top 10, but it would have been more inclusive and transparent (and would not have added particularly to the onerousness of the task) to have allowed this decision to be made by the respondents at the interim prioritisation stage. Perhaps you would like to recommend that future PSPs ensure that these decisions are made by respondents and not by steering group members, which introduces a potential risk of bias? I think this is a clearly written and important paper, which would benefit from some very minor additions/clarifications around the methods.

REVIEWER	Huw Morris UCL UK I have served on Parkinson's UK Research Advisory Panel and received funding from Parkinson's UK for research. I am chair of the Dendron Parkinson's UK study group which has encouraged participation in the PSP survey.
REVIEW RETURNED	07-Oct-2014

GENERAL COMMENTS	I have a few comments on aspects of this paper that could be clarified: - clarifying the differences between prevention, cure and management in terms of framing the questions and "ground rules" for participants - I was surprised by the comments page 22-30 on evidence available to refute some suggestions , a further description of this evidence would be helpful - are all the priorities of equal standing? - A really interesting aspect of this is whether there are any differences between patient priorities, carer and health care professional priorities this would benefit from discussion and clarification - "Future Priority Setting Partnerships should consider making the provision of contact details mandatory for participants so that issues like this can be followed up and support offered" - I would tend to disagree with this and I think people can be offered blanket guidance on discussing issues of concern with HCP rather than trying to contact individual patients in this exercise - How the questions were asked is important - I would suggest that more information on the framing of the study and questions asked are include as supplementary material

REVIEWER	Professor Richard Walker Northumbria Healthcare NHS Foundation Trust, UK

REVIEW RETURNED	30-Oct-2014

GENERAL COMMENTS	I think this is an excellent piece of work which is worthy of publication and will help researchers in the field of PD. This is an interesting, and important, paper which describes research priority setting for Parkinson's disease (PD) by patients, carers, their families, Parkinson's UK and health professionals involved with the care of people with Parkinson's (PwP). The authors, including the first author, have been involved in several systematic reviews in relation to treatment for PD, both drug related and non-drug related. They are therefore well placed to assess "evidence that has already been established". In conjunction with Parkinson's UK they had input from 1000 participants (60% PwP) about ideas on research uncertainties. These were then prioritised by 475 (72% PwP) before 27 (37% PwP) stakeholders agreed a final top 10. The methods utilised seem entirely appropriate. NHS ethical approval was not required but ethical approval was obtained from University of East Anglia's ethics committee. Specific comments: Strengths and limitations of this study (page 4) A lot of the text here relates to the results of the study. One bullet point relates to the lack of participants from minority ethnic populations or living in care homes which is an important limitation. The authors might want to consider putting further strengths and limitations in this section. The actions taken to gain a consensus from all relevant individuals for this survey were entirely appropriate. For health professionals this involved neurologists, PD nurse specialists, geriatricians with a special interest in PD, physiotherapists, occupational therapists and speech and language therapists and also involved the research network, DeNDRoN. Most of the patients and carers were identified via Parkinson's UK and, as acknowledged by the authors, these are not necessarily representative of all PwP. There is likely to be over representation of people from white, middle class backgrounds with higher levels of education and under representation of ethnic minorities and people in care homes. In fact, only 1% of responses were from individuals in care homes. Only 5% of PwP were black or Asian. In view of this, the authors suggest under the study limitations section that it might be worth looking at research priorities in the ethnic minority and care home groups separately. This aside, the mix of respondents seems appropriate thought it is unfortunate, for example, only 1% were social workers. This group might have had some interesting, and different, priorities. The survey was generally completed via a website but there were opportunities to complete it by phone, and also a translation service was provided if needed. It would be interesting to know how many people took advantage of the translation service? The 1000 original participants were asked to provide 4 priorities so it is not quite clear why there are 4100 responses rather than 4000?

	Of the 112 unique uncertainties 18 uncertainties with low duplication rates were excluded from prioritisation leaving 94 uncertainties for prioritisation. These were further cut down to 26 for the consensus meeting. It is not quite clear why 26 was chosen as a number. At the consensus meeting the top 10 research priorities were chosen. These are shown in the paper but it would be helpful for readers to be able to see at least the top 26. The authors expressed concern in the risk management section about individuals being identified at risk, eg suicidality. They suggest that in future it might be worth making provision of contact details mandatory. However, this would probably mean less people would be willing to take part. Within the top 10 I note that "What treatments are helpful for reducing dyskinesias in people with Parkinson's?" is number 3. As a clinician, the impression is that this is more of a concern to carers rather than PwP so it would be interesting to look at this in the responses to see if there was a difference between these 2 groups in relation to this item.

VERSION 1 – AUTHOR RESPONSE

Dr Pollock recommended that we create a flow chart to clarify the method's stages – this is provided in the new Figure 1.
He also recommended greater transparency of the generation of the top 26 priorities. We have provided an additional Table 2 which summarises the process and expanded our explanation in the methods page 9.
We have provided the information on who conducted the coding and checking processes page 9.
We have discussed the reasoning behind the exclusion of 18 uncertainties by the steering group more on page 9 methods, page 13 results, and in particular the risk of bias this could have introduced in the discussion page 17.

Dr Morris recommended we clarify definitions of prevention and cure which we have done on page 7.
The ground rules for participants in the consensus meeting have been clarified on page 10.
We have clarified where the evidence to refute the suggested uncertainties came from, references 22, 27, 34, page 13.
The priorities are ranked and so do not have equal standing.
The new Table 2 provides information on the differences in rankings between the four groups of respondents for the top 26 uncertainties.
We have amended our discussion of risks to include the option to provide blanket guidance page 16
We have provided all of the surveys used to be provided in supplementary materials.

Prof Walker recommended that we put in further strengths and limitations of the study in the summary on page 4. We have clarified these points on page 4 and enhanced our discussion of the studies limitations on page 17.
We have highlighted that the translation service was not used (pg11) but a representative of Parkinson's UK did help some respondents in clinics where their English literacy was poor.
We have clarified how the numbers of responses were generated on page 9.
We have clarified why 26 priorities were selected on page 9 (prioritisation).
The top 26 are shown in the new table 2
We have discussed the risk to recruitment in methods of managing e.g. suicidality on page 16
The new Table 2 provides the relative prioritisations of the four groups. There was no substantial

differences in the ratings for the statement relating to dyskinesia. We have a separate article being prepared to discuss these issues in more detail.