Research Involvement and Engagement

The extent, quality and impact of patient and public involvement in primary care research: a mixed methods study

--Manuscript Draft--

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Full Title:	The extent, quality and impact of patient and public involvement in primary care research: a mixed methods study
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Abstract:	Background To improve the lives of patients in primary care requires the involvement of service users in primary care research. We aimed to explore the extent, quality and impact of patient and public involvement (PPI) in primary care research. Methods We extracted information about PPI from grant applications, reports and an electronic survey of researchers of studies funded by the NIHR School for Primary Care Research (SPCR). We applied recognised quality indicators to assess the quality of PPI and assessed its impact on research. Results We examined 200 grant applications and reports of 181 projects. PPI was evident in the development of 47 (24%) grant applications. 113 (57%) grant applications included plans for PPI during the study, mostly in study design, oversight, and dissemination. PPI during projects was reported for 83 (46%) projects, including designing study materials and managing the research. We identified inconsistencies between planned and reported PPI. PPI varied by study design, health condition and study population. Of 46 (24%) of 191 questionnaires completed, 15 reported PPI activity. Several projects showed best practice according to guidelines, in terms of having a PPI budget, supporting PPI contributors, and PPI informing recruitment issues. However few projects offered PPI contributors training, used PPI to develop information for participants about study progress, and had PPI in advising on dissemination. Beneficial impacts of PPI in designing studies and writing participant information was frequently reported. Less impact was reported on developing funding applications, managing or carrying out the research. The main cost of PPI for researchers was their time. Many researchers found it difficult to provide information about PPI activities. Our findings informed: *a new Cost and Consequences Framework for PPI in primary care research highlighting financial and non-financial costs, plus the benefits and harms of PPI *15 co-produced recommendations to improve PPI in research and within the SPCR. Conclusions The extent, quality and impact of PPI in primary care research is inconsistent across research design and topics. Pockets of good practice were identified making a positive impact on research. The new Cost and Consequences Framework may help others assess the impact of PPI.
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Response to Reviewers:	25th March 2018 Dear Editor-in-Chief, MS: RIAE-D-17-00050 The extent, quality and impact of patient and public involvement in primary care research: a mixed methods study Response to reviewers' comments On behalf of the authors, please pass on our gratitude to the reviewers for their fair and constructive critiques of our manuscript. We have responded to each one of the reviewers' points below and revised the manuscript accordingly. The revisions have been highlighted in red text. We hope that we have addressed the reviewers' concerns satisfactorily and improved the manuscript. We look forward to hearing from you. Kind regards Steven Blackburn, PhD Research Institute for Primary Care Health Science, Keele University # Reviewers Comment Response 1. #1: The article discussion section is the clearest account of the study's interest to the reader and I wondered if some of that language could be brought to the front of the paper. I was anxious to reach the commentary as to the co-produced recommendations to improve PPI in primary care research. There is no account of what those are in the plain English summary for example. Thank you for this kind comment. For the plain English summary, we have now included on how we used recognised quality indicators to ass the quality of PPI in studies and its impact. We have also include a general description of the types of recommendations that were co-produced. Unfortunately, a more detailed account of all 15 recommendations is difficult to achieve within the 350 word limit for this section 2. #1: The importance of the study is in the materials it has used to conduct the scrutiny and the recommendations it has developed from these materials and their responses. Thank you 3. #1: Interestingly, there was no exposure of the study's own PPI 'costs and consequences' quality assurance score. The article does not tell us about, for example, remuneration of the involved public contributors. An additional table could well bring the article to some more lively consideration if it disclosed its own self -scored costs and consequences matrix?

	This is a great suggestion. We have added our own self-assessment score against the quality indicators, and identified provided a cost and consequences framework for this study (supplementary file). We have summarised these on pages 21-22, line 592-607. 4. #1:The article's written presentation - no doubt tied to editorial convention - could nonetheless be more enthusiastic I suggest in its opening sections. There was a sense of over-formalised writing - until the discussion section We have followed reporting convention. However, we have reviewed the writing style in the opening sections and revised as appropriate. 5. #1: Although the GRIPP2 checklist was mentioned, along with INVOLVE's new standards, it is not within the compass of this review to comment on the usefulness or connectedness of this study's costs and consequences matrix to other PPI quality assurance approaches. It would be good, nonetheless to see those brought together into a single or shared document. Thank you for making this point. At the time of writing, we did bring together existing frameworks and resources (e.g. Going the Extra Mile recommendations, INVOLVE Values and Principles Framework, and SCPR PPI Strategy) as we were formulating our recommendations, in light of the costs and consequences identified during the study (p.18, lines 466-470). We also highlighted that it's too early to see the effect of recent development like the National Standards and the GRIPP2 on PPI quality. However, we have now added commentary on how future studies should consider how the National Standards for Public Involvement and GRIPP2 reporting checklist complement and facilitate each other (in the context of the costs and consequences of PPI highlighted in this study), to drive forward improvements in this field (page 20, lines 525-30) 6. #2: I did wonder if the cost and consequences framework could be made bold: 'Cost and Consequence Framework' to show this is something new and by adding a sentence in the abstract about this might be good for the reader, at the moment it lacks confidence as a framework. In the body of the text you could add a line about how this is to complement the INVOLVE Cost Calculator. Thank you for this suggestion. We have highlighted the novelty and importance of the Framework in the abstract, along with a short description of it. (line 58-9) We have added a sentence to highlight how the Cost and Consequences Framework should be considered when using INVOLVE's Involvement Cost Calculator [30]. (page 20, Line 528-30) 7. #2: The tables and figures are labelled incorrectly on the actual Table titles and Figure titles throughout, which made it hard to see which figure or table reflected the content being described, but with a bit of searching I was eventually able to find which table or figure fitted where. We have checked that the tables and figures are now all labelled correctly 8. #2: I would have liked a few more lines in the Conclusion about The Cost and Consequence Framework and in the body of the text. We have now added some more text about the Cost and Consequences Framework in the Abstract, the Plain English Summary and in the Discussion (see my response to comment #5) 9. #2: Although in most places the authors offer the number of applications to accompany the percentage, I wondered if this could be adopted throughout, this would help the reader to understand in the context of the 200 application, 'x' number had specified … so and so. We have now included the number as well as a percentage throughout the paper 10. #2: Some acronyms are introduced and then not followed through, e.g. 'PI' on p14, similarly 'FR' is in the list of abbreviations but then not in the text (unless I missed this).Keyword: should 'Framework' be added? Thank you for spotting this. We have now addressed the erroneous acronyms 11. #2: Sometimes n= is used to indicate number of applications, sometimes just the number of applications, sometimes just a percentage is offered. For consistency, it will

help to standardise this. On the following pages percentages offered without the specific number of applications was noted: pages: 2, 12 (line 309),
We have now reported frequencies and percentages consistently throughout the article.
12.
#2: I was unclear whether this work received specific funding? This needs clarity under either Setting on p7 or on Funding section on p22.
To clarify the funding of this study, the statement on p.25 has been changed to "This paper presents independent research funded by the National Institute for Health Research School for Primary Care Research (NIHR SPCR)." We have also included a disclaimer about the views expressed in the paper.
13.
#2: Tables and figures are all labelled incorrectly, e.g. there are five figure 1s at the back. Table 3 is currently table 1 etc. These figures and tables are listed on pages: 7,10,12,13,14,15,16,17,21
We have addressed the labelling of the figures and tables throughout the article.
14.
#2: On p9 researcher 'SS' is listed, this person is not listed in the team information
Thank you for spotting this error. We have now changed it to the correct initials of the author who conducted this piece of the study.
15.
#2: P10 what was the rationale for not doing PPI on the 26 projects? Was it the same as the rationale provided on page 11? If so this needs explaining clearer.
Reasons provided for not reporting PPI were similar across completed and uncompleted projects. We have inserted a new paragraph (p.11, line 270-275) to describe this and provided some example rationales.
16.
#2: P11 what was the average length of projects?
This proposed duration of projects was not extracted in the analyses of grant applications, and at the time of analysis, the start and finish dates of projects was not available in the project documentation. Most research studies take approximately 2 to 3 years. However, it is likely that some of the SPCR studies may have been shorter, and other studies may have taken longer to complete due delays, for example in project set up or participant recruitment. However without this data, it would be difficult for us to calculate or comment on the duration of studies.
17.
#2: P13 on line 336, was there a justification offered as to why 20 projects did more PPI than planned?
Information on the discrepancies between planned and reported PPI activities was not available in the project documentation. We have provided opinions to why the discrepancies may have occurred (page 14, lines 354-362)
18.
#2: P14 line 371 is there any literature to show this finding (of homogeneity in PPI) being congruent with literature. Similar point about literature and finding congruence about PPI being less common in methodological design and Systematic review (page 12)
The negative impact of PPI reported by one of the Principal Investigators was related to the homogeneity of the young people recruited into the study, not the PPI contributors. On page 15, line 395 we have clarified this sentence by stating that, "young PPI contributors encouraged their friends to participate in a study on reproductive health in young people." We are not aware of any literature that reports similar findings.
In terms of the comment about PPI being less common in methodological design and systematic reviews, we have stated that our finding that PPI is more common in study design such as clinical trials and mixed methods was also reported by Mathie et al (2014) and Gamble et al (2014) (page 21, line 546). We had added that Mathie et al also found that PPI was less common in observational and cohort studies (page 21, 547)
19.
#2: P17, can you offer a brief description about how your team understood 'co-production' this is a contested term at the moment so clarity will help readers your starting point.
We have revised section 6 [p18] which describes how the recommendations were co-produced, giving additional detail to how researchers and PPI contributors worked

together to develop and agree on the study’s recommendations

20.

#2: P18 line 468-469 text is larger than the rest of the document.

Thank you for spotting this. We have corrected.

21.

#2: In the discussion could you add a sentence about the 'Framework for public involvement at the design stage of NHS, Health and Social Care research' when you discuss some guidelines and frameworks?

We have now included a reference to this recently published framework (page 18, line 489-90).

22.

#2: Where you discuss the quality being indicators of good practice could you bring the point back that people spent on average 0-30 hours on PPI (those with fewer hours are less likely to have meaningfully engaged the public)

Thank you for making this insightful point. As suggested we have caveated our statement that no specific example of poor practice were found with:
a) the finding that time spent on PPI was variable and, b) the suggestion that those who spent fewer or minimal hours on PPI may not have spent sufficient time have meaningfully engaged with the public (page 19, line 517-519)

23.

#2: P20 line 519 does bureaucracy reflect in the literature for PPI processes?

We have revised this paragraph to state reasons for researchers difficulty to provide information on PPI and inability to pass on the PPI contributor survey are unclear and not explained in the literature elsewhere (page 20, line 535-541)

24.

#2: Numbering error page 20. Number 3 featured twice in the list (line 520 and 525)

This has been corrected

25.

#2: Future impact of this study, p21. Can you add some more to about the cost and consequence aspect.

We have included a sentence about the potential of the new Cost and Consequences Framework to help other consider the wider impacts of PPI (page 23, line 624-626)

26.

#2: Table 3 under 'Other' Category the 'f' label description is later referred to as '$' in notes form

Thank you. We have now corrected this error

27.

#3: I wondered if you might consider balancing a quote "Young people can be unreliable - it's sometimes difficult to know whether they will turn up or not, and to plan accordingly." which seems to generalise the behaviour of all young people, with some discussion of how attendance at PPI groups can vary for all age groups, depending on health conditions and other commitments

To address your valid comment, we have added the following sentence, “While the respondent in the above quote has commented on young people, it should be noted, however, that this is not generalizable of all young PPI contributors. The participation of all PPI contributors can be impacted by many factors, such as availability on scheduled meeting dates, changes in health status and other commitments.” (page 16, line 419-422)

28.

#3: I also wondered if there had been any discussion as to whether provision of admin support, or lack of it, had contributed to the cases where there was no record of costs and benefits of PPI activity. If so, perhaps this could be included in the article.

While we could not identify specific reasons for poor record keeping for PPI, other than simple statements from respondent that it was “due to a lack of information”, we have included the possibility of insufficient administrative supports as a reason (page 20, lines 535-541)

29.

#3: Line 41: Is the 47 (24%) of projects out of the 181 reports? If so, for consistency, should a number be given for where there was PPI from the 200 of grant applications? Not just 57% - but give a precise number too?

We have clarified the denominator and included the count of grant applications or projects as well as the percentages (page 2, line 43)

30.

#3: Line 42: delete semi-colon after study ... during the study; mostly in study oversight

This has been deleted.

31.

#3: Line 43: delete extra f in Ffrequent activities (This typo does not occur on the opening page abstract - only on the internal abstract

This spelling mistake has been corrected

32.

#3: Line 44: State how many projects in numbers?

We have included the count of grant applications or projects as well as the percentages (page 2, line 44)

33.

#3: Line 52: Suggest delete PPI activities which was more common. Suggest insert: and the PPI activities which were more common were..

We have revised this sentence to: PPI during projects was reported for 83 (46%) projects, including designing study materials, advising on methods, and managing the research (page 2, line 44-45)

34.

#3: Line 52: Suggest sentence reads: Beneficial impacts of PPI and the PPI activities which were more common were frequently reported in designing studies and writing participant information

To simplify the sentence, we have deleted end of the sentence (“PPI activities which was more common”) to leave: “Beneficial impacts of PPI was frequently reported in designing studies and writing participant information.” (page 2, line 52)

35.

#3: Line 55: suggest deleting semi-colon ; and rewording ... suggesting record-keeping was poor. SUGGESTION: to insert mention of the workshop somewhere in this section.

As we have included additional text into the abstract and plain English summary, we have simplified the sentence by removing our suggestion as to why researchers found it difficult to provide information about PPI activities

36.

#3: Page 3

Keywords:

Line 66:   suggest inserting Framework

Thank you. We have included ‘framework’ in the keywords (page 3, line 68)

37.

#3: Page 4

Plain English Summary

Line 69 NOT ENTIRELY CLEAR. Suggest deleting...who receive primary care services in the research process. ? insert instead... insights and experiences of patients of the research process which they receive in primary care.   (If that's what you mean?)

We have revised this sentence to read: “Therefore it is important for research into primary care to be informed by the insights and experiences of patients who receive these services.” (page 3. Lines 72-72)

38.

#3: Line 75 NOT ENTIRELY CLEAR.   "did not have PPI as they planned to initially." Does this mean that the PPI got abandoned, or does this mean that the form of PPI input changed as it was going along? Sort of 'Let's not do that...' 'Instead, let's do ...' Could do with clarification

We have revised the sentence to highlight the important finding that “some studies did not undertake the PPI activities initially planned and funded for” (page 3, lines 77-8)

39.

#3: Line 77: I wondered if inserting the word 'not' would help emphasise the lack of training offered.... such as not offering PPI contributors training

Thank you for this suggestion, but we were highlighting that these good practice were not followed. Inserting ‘not’ might suggest that ‘not offering PPI contributors training’ was good practice. Nevertheless we have revised this sentence to emphasise that few studies offered these good practices. (page 4, lines 80-1)

40.

#3 Line 78 .. Not... using PPI to develop information... and ... not ... having PPI to advise on publishing findings

As above

41.

#3 Line 81 NEEDS EXPLANATION: I suggest that you explain what Higher Quality

means in practical terms. It seems obvious that higher quality will have most impact. However lay readers may not all understand what goes into making the approach of 'higher quality'. Even talking about 'a higher number of quality indicators met for a single project' (as on page 9, line 207) is not that easily understandable to lay people
We have revised this sentence to clarify the issue on quality. It now: ‘Most impact was reported when the approach to PPI included more indicators of good practice.’ (page 4, lines 83-4)

We have also made a similar change to the sentence on page 9, line 212-3
42.
#3 Line 82: delete suggesting and insert this suggested that... so the sentence will read Many researchers found it difficult to provide information about PPI activities; this suggested that record-keeping was poor
As we have included additional text into the abstract and plain English summary, we have simplified the sentence by removing our suggestion as to why researchers found it difficult to provide information about PPI activities (page 4, line 84-5)
43.
#3: Page 5
Background:
Line 97: ...[5,6]. Delete i and insert capital I so that it reads Its positive impacts are...
Thank you for spotting the error. We have now corrected this.
44.
#3: Line 111: insert comma after limited
Thank you for spotting the error. We have now corrected this.
45.
#3: Line 116: Delete ninety percent and insert 90%
We have now changed this to 90% (page 5, line 119)
46.
#3: Line 117: NOT ENTIRELY CLEAR Suggest the sentence is amended to read: Therefore it is important to gather the views, insights and experiences of patients about the research process which they receive in primary care. (If that's what you meant)
As per previous comment, we have revised this sentence to read: “Therefore it is important for research into primary care to be informed by the insights and perspectives of patients who receive these services.” (page 6, line 120-121)
47.
#3: Line 120: insert are after [20-22
We have now added this
48.
#3: Line 121: insert which have so that it reads ....description of the PPI activities which have taken place
We have now added this.
49.
#3: Line 129: No 3 EDITORIAL QUERY: Should patients (having the health research done on them) be referred to here in this article?
The impact of PPI on the patients having health research done on them (study participants) was not in the scope of the aims of this project. Nevertheless this is an interesting research question on its own
50.
#3 Line 137: Does the lack of ability need specifying? maybe ... lack of records... lack of staff to look this up? ... or whatever
We have now clarified this sentence by stating that a cost and consequences analysis was chosen due to the lack of available data and recorded information about PPI (page 6, line 141-2)
51.
#3 Line 143: Would it be useful to say how may PPI contributors attended the workshop? I was wondering as I was reading.
We have clarified that eleven PPI contributors were involved in the workshop (page 6, line 148)
52.
#3 Page 8
Researcher and PPI contributor surveys:
Line 199: Delete er from sentence beginning To examine the quality of PPI in primary care researchER so that it reads ... in primary care research.
Thank you for spotting this. We have now corrected it.

53.
#3 Page 10
Results:
Researcher and PPI contributor survey
1.2 PPI during the projects
Line 252: delete s from ... (plus one projects whose...)
Thank you for spotting this. We have now corrected it
54.
#3 Page 12
1.8
Line 299 I think it should be: delete were insert was so that it reads ...Though the study population of two-thirds of SPCR funded projects was patients only...
Thank you for spotting this. We have now corrected it
55.
#3 Page 13
1.9 The type of PPI
Line 333 ? Font size? Figure 3 looks inconsistent in size
Thank you for spotting this. We have now corrected it
56.
#3: Page 14
3. The impact of PPI from the researchers' perspective
Line 371: NOT ENTIRELY CLEAR about this sentence. Did the authors mean that 'The only negative impact reported was the view that a more homogenous (rather than diverse) study ended up being recruited, since PPI contributors encouraged their own friends to participate. ???
We have clarified this sentence by stating that, "young PPI contributors encouraged their friends to participate in a study on reproductive health in young people." (page 15, line 395-6)
57.
#3 p15
3.1 Quality-Impact Index scores
Line 399: insert in so that it reads ... activities in which the PI reported a perceived impact)...
This has now been added
58.
#3 Page 16
4. The financial costs of PPI
Line 410: insert a space before and after -
We have removed the '-' (page 17, line 438)
59.
#3 Line 414: QUERY Would it be relevant to make some reference here about help with transport (not costs, but booking, on behalf of PPI participants, taxis to make it possible for them to attend if they had a condition which made it difficult to access public transport)??
We have provided examples of the travel costs that might have been reimbursed: e.g. car mileage, public transport, parking page 17, (line 442)
60.
#3 Line 424: delete as so that it reads: Table 4 presents a framework ..
We have removed 'as' (page 17, line 453)
61.
#3 Page 18
Discussion
Line 466: insert ly so that it reads frequently
We have inserted 'ly' to frequent (page 18, line 500)
62.
#3 Line 467 EDITORIAL QUERY Line 505 says that the most commonly stated PPI activity was ... reviewing patient information leaflets ... Do not 'Reviewing' and 'Developing' go hand in hand? Maybe it would be worth addressing that point. On Line 467 it says 'less frequently for other aspects of the research process (eg developing Participation Information Leaflets)
This statement relates to a finding from Gamble et al's publication, which simply referred to PPI in reviewing patient information. We have not made any assumptions or interpretations with this. PPI may have helped to develop patient information, or simply reviewed documents produced by the researchers. In our study we specifically asked if

PPI was involved in ‘developing’ participant information.
63.
#3 Line 468: instert t conducting the research
We have now correcting this spelling error (line 502)
64.
#3 Line 468: some of the font size looks inconsistent - ? too large
We have checked that all font sizes are consistent
65.
#3 Line 470: query whether re-wording would help - maybe something like: ... with PPI semingly less prevalent in study designs where there was less direct contact with the study patient/participants
Thank you for this suggestion. We have reworded the sentence. (line 504)
66.
#3 Line 472: Is some of the font size on this line too large?
We have checked that all font sizes are consistent
67.
#3 Line 481: insert s terms
We have corrected this (line 515)
68.
#3 Line 483: Typo: should be Boote et (not Boote at)
We have corrected this (line 520)
69.
#3 Line 483: some of this line is in a different size font
We have checked that all font sizes are consistent
70.
#3 Page 19
Line 505: - is this at odds with line 467?
See our response to comment #62
71.
#3 Page 21
Future Impact of the Study
Line 557: IMPORTANT
IS THIS REFERENCE THE WRONG NUMBER? No. 30 seems to be Boote, so what should the correct number here be for NIHR School for Primary Care Research...?
Thank you for highlighting this. We have now amended the reference. (line 598)
72.
#3 Line 565: To make it clearer to read, would it be possible to have the bracketed numbers on separate lines:
... in terms of:
1) establishing..
2) recording and reporting...
3) promoting and..
Then a new paragraph for
This Study did identify...
We have now put this into a numbered list, on separate lines (lines 633-636)
73.
#3 Line 567: If you don't do separate lines as suggested above, then need to insert a comma after PPI,
We agree that a numbered list, on separate lines is clearer
74.
#3 Page 22
Line: 591 ? Amend sentence to that it reads: The work presented in this paper was independent research commissioned by...
We have now checked that we have the correct wording of the funding acknowledgement for the SPCR (line 661)
75.
#3 Page 23
Line 596: Important: Insert the word Health so that it reads: Collaborations for Leadership in Applied Health Research and Care West Midlands
We have now corrected this (line 665)
76.
#3 Line 610: Insert k so that it reads thank
We have now corrected this (line 680)
77.

#3 Line 614: Does superscript no. 1 need to be deleted, or where does it refer to? I couldn't find out what it referred to.
We use the endnote to define ‘PPI contributors’ on page 5 (line 110)
78.
#3 Page 24
Line: 624: Some punctuation needed in title of Going the Extra Mile. Suggest it reads: Going the Extra Mile - a strategic review ...
We have corrected the reference as suggested (line 694)
79.
#3 Line 640: Is some of the font size in this line too large? longitudinal study ???
We have checked that all font sizes are consistent
80.
#3 Page 25:
Line 649: insert apostrophe after researchers so that it reads researchers' attitude
We have corrected the reference as suggested (line 699)
81.
#3 Page 26
Line 672: insert a space before dash and after dash ... research - an example...
We have corrected the reference as suggested (line 739)
82.
#3 Page 30 - 34
IMPORTANT
What is the correct Table number? Is it meant to be Table 3 ??
Another question: would it help the reader to stay looking on the correct line if the table where in some sort of a grid?
We have checked the numbering of the tables and corrected accordingly

Thank you for this comment. We have checked the author guidelines on tables and have used the conventional format of tables for journal publications. Therefore, we have not reformatted the table as suggested
83.
#3 Page 36 Line 706 Table 4
EDITORIAL
Would it be easier for the reader if the internal boxes were re-formatted? There is a lot of indentation going on which makes it distracting for the reader.
Thank you for this comment. We have removed the unnecessary indentation.
84.
#3 Page 37 Individual page Line 21
Conducting and managing
EDITORIAL QUERY/EXPLANATION?
It would be informative to give the age range of the young.
Would it be possible to give, either here or somewhere in the article, an idea of the facilitation process (if any) specifically designed to engage young people?
Dissemination

It was not always possible to determine the ages or age range of children and young adults from the study documentation. Sometimes, ages were provided, sometimes the documentation simply referred to children. So we have assumed children and young adults to be 17 and under.
We have included this age range (0 – 17 years) for the children and young adults category in the table and added an explanation in the table footnotes (‘g’) (Line 774).
85.
EXPLANATION requested: Why is PPI contributors attending conferences and external events considered to be a Minus?
Thank you for this comment and appreciate that the statement could be interpreted in this way. We were referring to the financial costs of PPI contributors attending conferences and external events. We revised the statement to make this clearer. (table 4, page 38)
86.
#3 Page 39 Footnote
EDITORIAL QUERY Sorry, but this Footnote says Sometimes included within the direct payment. But what is it which is sometimes included within the direct payment.
The previous reference says: Direct payment of PPI contributors for attending meetings

Is there another aspect which is sometimes included within direct payments which needs a link to this number? Or should this footnote be deleted?
The footnote number was linked to the wrong statement. It should be linked to the travel costs of PPI contributors attending meetings (in the row below) – as reimbursement of travel expenses is sometimes included in the direct payment made to PPI contributors.
87.
#3 Page 40 Table 5
Individual page line no. 45: Delete ing All the other items say 'Improve' 'Stimulate' 'Create' (not improving...)
We have revised this for recommendation number 8.
88.
#3 page 41
Individual page line no. 27: Insert full stop after B
We have included the full stop (bottom of page 41)
89.
#3 Page 42
Line 716 Should this be Table 6
We have checked the numbering of the tables and corrected accordingly
90.
#3 Line 717
Individual Page Line no. 7: Does there need to be a reference to Primary Care somewhere here
Individual Page Line No. 7: Typo: delete second o so that it reads very aware of the varied approaches to PPI being undertaken ...
To clarify the types of research projects, we have included the reference so it now reads: As a lay coordinator of a growing group of research users involved in a variety of primary care research projects across a clinical trials unit… (line 789)

91.
Line 717
Individual Page Line No. 7: Typo: delete second o so that it reads very aware of the varied approaches to PPI being undertaken
We have corrected the spelling mistake in the quote (p 43, line 7)
92.
#3 Individual Page Line No. 20: Insert apostrophe after patients' patients' daily care..
We have corrected this
93.
#3 Individual Page Line no. 22: I believe it should be spelt practice with a c in this case
We have corrected this
94.
#3 IMPORTANT
Correct numbers for the Figures need to be typed onto the tops of the relevant tables. At the moment they all say Figure 1 apart from on the Download button
We have checked the numbering of the tables and corrected accordingly. I think this is an error with how the Figures are labelled during the uploading process
95.
#3 VERY IMPORTANT! within Figure No. 4
The fourth from the top quality indicator very much needs an l inserted so that it will read Public
Last line within the table: delete c and insert s to that it reads: Public involvement advised on informing..
Yes, very important. Thank you for spotting this. We have now corrected.

We have corrected this
96.
#3 Figure No. 5
? EDITORIAL
Association between the Quality Score (number of quality indicators met by a project)
Do we need a link to the Quality Score/Impact Score to see what the activities were
Also: Typo in heading: insert in so that it reads .. (number of PPI activities in which the PI reported a perceived impact)
Thank you for this comment.

	We have not linked impacts of PPI with the individual Quality Indicators met in the Quality-Impact Index Score. In the 13 projects included in this analysis, the researchers perceived impact on different types of PPI activities, so it would be difficult to separate these activities out to provide anything meaningful. We have discussed in the main text that the most impact was perceived on the PPI activities most commonly conducted (shown in Figure 2). In Figure 5, we have summed the number of PPI activities in which the Principal Investigators perceived an impact. We have inserted the word ‘in’ in the heading 97. #3 SUPPLEMENTARY MATERIAL - If possible, on Patient Survey of the Costs and Effects of Patient and Public Involvement (PPI) in Primary Care Research, P6. No 9. amend Typo: Delete ing and insert e so that it will say: Leave lines blank where you did not receive any help. We have corrected this simple spelling mistake 98. #3 Patient and Public Involvement in your research project questionnaire: For publication, would it be possible to align the questions which are most obviously semi-indented, to aid the reader? A9, A11, A12, A18; B3, B6, B9, B10 We are not sure about what the reviewer is asking to change. The style and format of the questionnaire was developed and agreed with our PPI group (as well as being ethically approved) so we would prefer not to change them. 99. Reviewer #4: This is a model of how to write a clear, compelling article with a defined outcome. It shows the value of ppi ; it outlines the issues which cause concern; it identifies the weaknesses in the research it is doing; it offers ideas for future research. Thank you for this kind comment. The authors are greatly appreciative.
Additional Information:	
Question	**Response**

TITLE

The extent, quality and impact of patient and public involvement in primary care research: a mixed methods study

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ABSTRACT

Background

To improve the lives of patients in primary care requires the involvement of service users in primary care research. We aimed to explore the extent, quality and impact of patient and public involvement (PPI) in primary care research.

Methods

We extracted information about PPI from grant applications, reports and an electronic survey of researchers of studies funded by the NIHR School for Primary Care Research (SPCR). We applied recognised quality indicators to assess the quality of PPI and assessed its impact on research.

Results

We examined 200 grant applications and reports of 181 projects. PPI was evident in the development of 47 (24%) grant applications. 113 (57%) grant applications included plans for PPI during the study, mostly in study design, oversight, and dissemination. PPI during projects was reported for 83 (46%) projects, including designing study materials and managing the research. We identified inconsistencies between planned and reported PPI. PPI varied by study design, health condition and study population.

Of 46 (24%) of 191 questionnaires completed, 15 reported PPI activity. Several projects showed best practice according to guidelines, in terms of having a PPI budget, supporting PPI contributors, and PPI informing recruitment issues. However few projects offered PPI contributors training, used PPI to develop information for participants about study progress, and had PPI in advising on dissemination.

Beneficial impacts of PPI in designing studies and writing participant information was frequently reported. Less impact was reported on developing funding applications, managing or carrying out the research. The main cost of PPI for researchers was their time. Many researchers found it difficult to provide information about PPI activities.

Our findings informed:

- a new **Cost and Consequences Framework for PPI** in primary care research highlighting financial and non-financial costs, plus the benefits and harms of PPI
- **15 co-produced recommendations** to improve PPI in research and within the SPCR.

Conclusions

The extent, quality and impact of PPI in primary care research is inconsistent across research design and topics. Pockets of good practice were identified making a positive impact on research. The new Cost and Consequences Framework may help others assess the impact of PPI.

Keywords

Patient and public involvement; quality; impact; mixed methods, primary care research; Cost and Consequences Framework

Plain English Summary

In the UK, more patients go to primary care than other parts of the health service. Therefore it is important for research into primary care to include the insights and views of people who receive these services. To explore the extent, quality and impact of patient and public involvement (PPI) in primary care research, we examined documents of 200 projects and surveyed 191 researchers.

We found that about half of studies included PPI to develop research ideas and during the study itself. Common activities included designing study materials, advising on methods, and managing the research. Some studies did not undertake the PPI activities initially planned and funded for. PPI varied by study design, health condition and study population. We found pockets of good practice: having a PPI budget, supporting PPI contributors, and PPI informing recruitment issues. However, good practice was lacking in other areas. Few projects offered PPI contributors training, used PPI to develop information for participants about study progress and included PPI to advise on publishing findings.

Researchers reported beneficial impacts of PPI. Most impact was reported when the approach to PPI included more indicators of good practice. The main cost of PPI for researchers was their time. Many reported difficulties providing information about PPI.

In partnership with PPI contributors, we have used these findings to develop:

- a new **Cost and Consequences Framework for PPI** highlighting financial and non-financial costs, benefits and harms of PPI
- **15 co-produced recommendations** to improve the practice and delivery of PPI.

BACKGROUND

Actively involving patients and the public in research is seen as a marker of good research practice because it leads to research that is relevant, better designed, with clearer outcomes, and a faster uptake of new evidence [1]. Now a requirement and priority of many research funding bodies, patient and public involvement (PPI) is promoted at all stages throughout the research cycle [2]. The recent National Institute for Health Research (NIHR) *Going the Extra Mile* report highlighted the need to improve the quality and practice of PPI in health and social care research [3]. In response to NIHR recommendations by the NIHR, INVOLVE has published their Values and principle's Framework for best practice in PPI [4].

PPI has been documented in a number of research areas [5, 6]. Positive impacts are reported for all stages of research, including enhancing its quality and appropriateness, an increased understanding and insight of researchers into their research field, and the increased sense of self-worth, confidence and skills gained by PPI contributors[i] as a result of their involvement [7, 8]. PPI has also been associated with success in achieving participant recruitment targets in studies, securing funding, designing study protocols and choosing relevant outcomes [9, 10]. Recent research has identified the characteristics of effective PPI [11] and the mechanisms required to sustain it [12].

In contrast, reported negative impacts are PPI contributors' frustration with the lengthy process and lack of feedback, the extra time needed to complete research, time constraints of patients and researchers, and the increased financial costs [10]. Moreover, PPI can be tokenistic due to negative attitudes of researchers and the requirements of research funders [10, 13]. Variation in the context of, and approaches to, PPI, combined with lack of validated tools to assess its quality, causes challenges to identify best practice of PPI and its impact [10, 14].

Though PPI in research is a clear priority for the government, the NIHR and other research organisations, there is growing, though limited, evidence relating to the costs (financial and non-financial) and consequences (benefits and harms) of PPI in research . This seems to be driven by the lack of and poor quality of reporting [7, 15, 16].

Our study is set in the context of primary care research. That is, research conducted within health services providing first-contact care for patients (e.g. general practices, district nursing, and community-based health services) [17]. 90% of all NHS interactions occur in primary care [18], with the management of chronic illnesses a key component. Therefore it is important for research into primary care to be informed by the insights and perspectives of patients who receive these services.

However, little is known about the extent, quality and impact of PPI across the whole range of primary care research. To date, the small number of primary care studies reporting on PPI [19-21] are largely limited to a description of the PPI activities which have taken place. More recently, a case study of a primary care research centre reported how dedicated infrastructure and resourcing for PPI, flexible working practices, leadership, and secure funding has enabled the fostering of sustained long term PPI across all of its research [12, 22].

The literature in this area is limited and the benefits and costs of PPI for both researchers and the public is unclear. Our study therefore set out to gain a broader understanding of PPI in primary care research. Specific research questions were:

1. What is the extent of PPI in primary care research?
2. What is the quality of PPI in primary care research?
3. What is the impact of involvement on PPI contributors, researchers and research institutions involved?
4. What are the costs associated with PPI in health research?

The four research questions were addressed through use of surveys, analysis of relevant research documents and a workshop. Results from research questions three and four were analysed and structured as a cost-consequence framework, a largely qualitative way of summarising key costs (financial costs as well as negative impacts on individuals or institutions) and key benefits (financial savings and positive impacts). Cost-consequence analysis is typically used by economists when it is not feasible to conduct a standard economic evaluation. In this case, cost-consequence analysis was adopted due to a lack of available data and recording information about PPI to accurately quantify all monetary costs and the lack of an appropriate single metric for summarising non-monetary consequences.

METHODS

The study used a mixed methods approach consisting of 1) analysis of documents relating to research projects such as grant applications, annual reports, final reports; 2) a survey of researchers and PPI contributors; and 3) a workshop with eleven PPI contributors and the research team to discuss the findings and co-produce recommendations. The analysis of project documents was used to provide evidence of the scope of PPI in primary care research. The surveys were used to provide evidence on the quality, experiences, and impact of PPI. The findings informed the recommendations workshop and development of the cost-consequences framework.

Setting

This study focused on research projects funded by and taking place within the NIHR School of Primary Care Research (SPCR) since its inception in 2008 to 2014. This included all projects funded on each of the SPCR's annual funding rounds (FR1 to FR8). The SPCR is a partnership between the leading academic centres for primary care research in England. Through dedicated funding, its remit is to increase the evidence base for primary care practice through high quality research and strategic leadership.

Patient and public Involvement in this study

The study embraced PPI throughout every stage of the research cycle, from developing the initial idea, designing the study and being lay co-applicants (AH, CR) on the grant application, through to working with the researchers to understand the findings and writing the recommendations. Full details of the involvement of the PPI contributors of our study team are published elsewhere [23]; however the contribution of our PPI contributors are embedded throughout this article.

Analysis of project documents

We requested all documentation relevant to all projects from the SPCR. This included grant applications, along with annual and final reports provided by grant holders to the SPCR. We also collected other documents containing data on PPI in projects from the SPCR and from researchers who had included PPI within their projects (e.g. posters presented at the annual SPCR Showcase event and articles).

Data from the documents were recorded using a data extraction form and compiled in an electronic spread sheet. The type of data extracted from each document is shown in Table 1. Two members of the research team (SB, SM) completed the data extraction. To examine the scope of PPI in primary care research, the data from the project documents were summarised using descriptive statistics. We examined the change in the extent of PPI activities over time, using each annual funding round as a proxy measure of time. Subgroup analyses were conducted to examine the frequency and type of PPI activity by study design, disease/condition (categorised using the Health Research Classification System [24]), study population, and the age profile of the study population. Descriptions and explanations relating to PPI were analysed qualitatively to provide further insight to the activities reported.

[TABLE 1 Types of data extracted from the project documentation HERE]

Researcher and PPI contributor surveys

A cross-sectional survey design was employed using self-completed questionnaires. The researcher and PPI contributor questionnaires were developed through a review of the literature and a search for existing questionnaire items on the costs and consequences of PPI. The contribution of our three PPI contributors to develop the questionnaires ensured that items reflected the range of costs and consequences experienced by PPI contributors. The questionnaires included items aligning with Boote et al's [25] quality indicators to enable the assessment of PPI activity against best practice (Table 2). Also, the questionnaire included items relating to the type of PPI activities and the perceived impact of these activities on the research study and the respondent. Most items included a free text box to allow the respondent to explain their answer or give further insights. The researcher and PPI contributor questionnaires are provided in Supplementary Files 1 and 2, respectively.

All Principal Investigators (lead researchers) of projects funded by the SPCR received an electronic survey via email for each project that they were leading or had led. Project details were provided by the SPCR. As the details of PPI contributors involved in SPCR projects were not available, an item was included in the researcher survey to determine Principal Investigators' willingness to pass on a paper-based questionnaire to the public members involved in their projects. Our PPI contributors were consulted about this recruitment strategy. They felt that, while possibly not ideal, particularly as this relied on good record keeping of public members' involvement, this approach was pragmatic and acceptable.

Descriptive statistics were calculated for all quantitative items in the survey: types of PPI activities, costs (financial and non-financial), and impacts. To examine the quality of PPI in primary care research, we compared PPI activity reported by Principal Investigators with Boote et al's quality indicators of best practice [25]. The analysis focused on how many projects met each quality indicator. Two of Boote et al's [25] quality indicators related to the description and acknowledgement of PPI contributors' involvement in publications. Therefore for the projects which the Principal Investigator in the survey reported PPI activities, we searched for related publications via the PubMed online search engine, using the Principal Investigator's name and key words from project title as a search strategy. Retrieved publications were scrutinized for information relating to PPI.

To explore whether projects with higher quality PPI (as defined by achieving a higher number of indicators of good practice, using Boote et al's Quality Indicators [25]) was associated with a higher level of perceived impact, a *quality-impact index* score was also calculated for each project. A Pearson correlation coefficient was calculated between the number of quality indicators met (the *quality score*) and the number of PPI activities where Principal Investigators reported a perceived positive impact (the *impact score*).

[TABLE 2 The principles and indicators of successful consumer involvement in NHS research. Adapted from Boote *et al* [25] HERE]

A cost and consequences framework of PPI in primary care research

Two researchers (PK, SJ) independently categorised survey items relating to the time spent on involvement activities and associated costs, impacts and related free text comments as either costs (financial and non-financial) and consequences (benefits and harms). An overall framework of all potential financial and non-financial costs and consequences of PPI was therefore constructed.

Recommendations workshop

Following completion of the data analysis, public members who had been previously involved with the project (AH, CR) plus seven other members of a Research User Group at Keele attended a workshop with the research team to discuss key findings of the study. The aim was to co-develop recommendations to improve PPI practice within the SPCR.

Ethical Approval

Ethical approval was obtained from Keele University's Research Ethics Committee (21st March 2014).

RESULTS

Documentary analysis

A total of 200 full project proposals, 233 annual reports and 39 final reports were provided by the SPCR for the documentary analysis. The annual and final reports provided data on 180 projects; reports for the remaining 20 projects were unavailable from the SPCR. However, for one project for which reports were not available to the research team, data on PPI were extracted from a poster presented at an SPCR Showcase Event. Therefore, the PPI activities reported in 181 projects were included in the analysis.

Researcher and PPI contributor survey

191 questionnaires were emailed to Principal Investigators, of which 46 were completed and returned (response rate 24%). The Principal Investigators who responded to the survey were unable to pass on a survey to the PPI contributors involved in their projects, so we did not collect any data from PPI contributors at this stage. Of the 46 responses received from Principal Investigators, 15 (33%) reported PPI activity, most commonly in designing methods (8 out of 15) and developing participant information (7 out of 15).

1. Scope of PPI in primary care research

1.1 PPI during the development of grant applications

Of the 200 funded projects for which full grant applications were available, there was evidence of PPI in the development of the application for 47 (24%) projects. Just over half of the applications (113, 57%) provided evidence of plans to conduct PPI during the study. Table 3 provides a summary of these projects by research design and health conditions under study.

1.2 PPI during the projects

Of the 181 projects for which annual and/or final project reports were available (plus one project whose information on PPI was extracted from a SPCR poster), 69 (38%) projects had been completed, 108 (60%) were uncompleted and this data was missing for three projects.

For all 181 projects (completed and uncompleted), PPI activities was reported for 84 projects (46%), not reported in 74 projects (41%), and for 23 projects (13%) there was insufficient data available to determine whether PPI had taken place or not. Where PPI had not been reported in the project, a rationale for the absence of PPI was provided for 26 projects (14%).

In the case of the 108 uncompleted projects, PPI activities were planned for 36 projects (33% of uncompleted projects), there were no plans for PPI in 42 projects (39% of uncompleted projects), and there was insufficient information available to determine whether PPI was planned for the remaining 30 projects (18% of uncompleted projects). Where there were no plans for PPI, a rationale for this decision was provided for seven projects (7% of uncompleted projects).

Rationales provided for 26 projects (completed and uncompleted) which did not report on PPI were similar, referring mostly to the applicability and relevance of PPI for the project. They included "user involvement was integrated into the original main trial, in which this project is nested. No additional user involvement was needed for the purposes of this project"; "this has been a database study and as a result there has been no direct involvement of patients or the public in this work"; "being a straightforward questionnaire study, PPI input to the project has been minimal"; or simply "not applicable".

1.3 Change in the scope of PPI over time

There was no clear trend for an increase in PPI in the development of grant applications or the reporting of PPI in annual/final reports over time, using the SPCR funding rounds as a proxy measure of time. However, there was a trend for an increase over time in the percentage of project proposals which provided details of plans for PPI for the delivery of the research (R^2=0.62) (Figure 1).

[FIGURE 1 HERE]

1.4 Budgeting for PPI

Of the 113 grant applications that included plans for PPI during the study, 57 (50%) made reference to a budget for this and 32 (28%) referred to rewards and/or recognition for those who would be involved. There were no references to a budget for PPI in any of the annual/final reports or posters obtained from the SPCR, and only one reference to rewards and recognition for those involved. However, the research team did not have access to the full costings for each project and it is therefore possible that this represents an underestimation of the number of projects for which PPI was budgeted.

1.6 The scope of PPI by study design

The extent of PPI varied across study design. Across both grant applications and project reports, PPI was relatively more common for mixed methods, qualitative and interventional trial designs study designs compared to other study types (Table 3). Relatively, PPI was most prevalent in the development of grant applications for projects with a cross sectional design (4 of 7 projects, 57%). Evidence of PPI was relatively least frequent in cohort (longitudinal and retrospective), methodological (study of research methods or development of data collection systems), systematic reviews and analysis of secondary data study designs.

When examining individual types of project documents, the data suggest a certain degree of inconsistency between planned and reported PPI. Plans for PPI within grant applications were relatively frequent for methodological and longitudinal cohort designs compared to other study types. However, reported PPI within annual/final reports was more common for cross sectional designs but less common for methodological design (as well as retrospective cohort and systematic reviews).

1.7 The scope of PPI by health condition

In terms of health condition researched, PPI in the development of grant applications and reported involvement during the study was relatively more common for projects focusing on cancer, renal and urogenital, reproductive health and childbirth (Table 3). PPI was most frequently planned for studies in the fields of neurology and other types of health conditions not listed in the Health Research Classification System [24] (labelled 'Other' in Table 3). However, evidence of PPI was relatively least frequent for studies on cardiovascular, metabolic and endocrine, stroke, infection and multimorbid (multiple, co-existing) health conditions.

1.8 The scope of PPI by study population and age

Though the study population of two-thirds of SPCR funded projects was patients only, projects focused on the general public, health care professionals only, and both patients and health care professionals, tended to have more PPI described in grant applications (both to develop the application and plans during the projects) (Table 3). Except for the carers category, reported PPI during the study ranged from 38% (3 out of 15 projects on healthcare staff) to 65% of projects (20 out of 37 projects on patients and health care professionals) within each population category. PPI was not reported in the annual and/or final reports of the single project involving carers.

Inconsistencies were noted again between planned and reported PPI during the study across population categories. For example, for general public or health care professional study populations, the relative proportion of projects reporting PPI activities during the study in their annual/final reports was 30% lower than the proportion of projects with plans for PPI described in the grant applications.

Projects focused on children tended to have less PPI described in proposals and annual/final reports compared to projects focusing on other age groups. For all other age groups PPI was similar: 20%-36% in the development of grant applications; 56%-64% in plans for PPI; 43%-56% reported PPI during the study. However, for nearly half of all projects (93, 47%), the age group of the study population was unspecified or difficult to ascertain from the project documents.

[TABLE 3 PPI in SPCR projects, by study design, health condition, population and population age HERE]

1.9 The type of PPI

Of the 200 grant applications, PPI activities reported during the development of projects related to consulting with patients and members of the public and gaining their comments and feedback on plans for research (24 projects, 12%) and contributing to the grant application (20 projects, 10%). Advising on study methods, such as outcomes and recruitment methods, were specifically reported in 14 grant applications (7%). A range of planned PPI activities were outlined in the grant applications and reported in annual/final reports, relating to different stages of the research cycle (Figure 2). Plans within grant applications for involvement in managing research through membership of a project steering committee or management group were most common (51 projects, 26%), followed by involvement in dissemination of project findings (41 projects, 21%). Designing study methods, analysing/interpreting data and designing study materials (such as questionnaires) were also frequently planned involvement activities. Planned PPI in conducting the research and recruiting participants were the lowest areas of activity.

However, the proportions of the PPI activities reported during the study were considerably lower than the planned activities described in grant applications. The most frequent activities reported during the project were designing study materials (33 projects, 18%), designing methods (25 projects, 14%) and managing the research (17 projects, 9%). To explore this inconsistency further, reports of PPI activities in project annual and/or final reports (either already conducted or plans to conduct for uncompleted projects) were compared with plans (and non-plans) for PPI outlined in their associated grant applications. This was done for 179 projects where both the grant application and annual and/or final reports were available (Figure 3). Over a third of these 179 projects (65, 36%) reported the PPI activities as proposed in the grant applications, including 27 projects (15%) which had no plans to do PPI anyway.

However there was inconsistency for 70 projects (39%): 20 projects (11%) reported more PPI than originally planned; 50 projects (28%) reported less than originally planned (including 23 projects (13%) which did not do any of the PPI planned). Information on the discrepancies between planned and reported PPI activities was not available in the project documentation. In most cases where more PPI was reported that originally planned, the annual and/or final reports documented PPI activities that was not part of a project's grant application. It is speculated that any additional PPI was conducted as the project evolved and new opportunities for involvement were created. For 64 projects (35%), the annual and/or final reports made either no reference to the PPI activities planned in the grant application, or made references to a few PPI activities only, but not all that were planned. It is possible that for some of the uncompleted projects, plans for future PPI activities outlined in the grant application but not yet done were not reported. For six projects (3%), however, it was explained in the reports that '[the] PPI member no longer want[ed] to be involved in the study'; there was less PPI than planned because 'it was a highly technical study and utilised anonymous clinical data with no direct patient contact'; there has been "no real PPI in the project…and the PPI section is not applicable since the project involved a secondary analysis of a database with specific policy relevant questions"; and "not applicable" or "none" was provided in the PPI section of the final report (3 projects, 2%).

There was insufficient information to make a judgement on the consistency of planned versus reported PPI for 44 other projects (25%). In most of these, the nature of the PPI could not be determined due to the insufficient information about PPI provided in either the grant application or the annual and/or final reports.

[FIGURE 2 HERE]

[FIGURE 3 HERE]

2. Quality of PPI in primary care research

The quality of PPI was assessed using the data from the 15 Principal Investigators who responded to our survey and reported PPI in their project. Overall, there was variation in how best practice, according to the quality indicators reported by Boote et al [25], was met across studies (Figure 4). Best practice was more frequently achieved in terms of offering PPI contributors personal and technical support (13 out of 15 projects, 87%); involving PPI contributors in advising on recruitment issues (11 out of 15 projects, 73%) and having a specific budget for PPI (9 out of 15 projects, 60%). Fewer studies met best practice for PPI in terms of PPI contributors advising on informing participants about study progress (1 out of 15 projects, 7%); advising on dissemination methods (1 out of 15 projects, 7%) or having to access to training (3 out of 15 projects, 20%). We could not provide evidence towards the endorsement of the quality indicators: ‘PPI training needs are agreed’ (this was to be captured via the patient survey) and ‘Distribution of research findings to relevant patient groups was in appropriate formats and easily understandable language’.

[FIGURE 4 HERE]

3. The impact of PPI from the researcher’s perspective

Principal Investigators most commonly reported impact for study processes with the most PPI activity (i.e. designing methods and developing participant information). Perceived impact of PPI on the research process and individual Principal Investigators was largely positive and included benefits such as improving the clarity of information, increased recruitment and follow-up rates, validation of findings and more useful outputs for clinicians and patients. The only negative impact reported was the view that a more homogenous study sample may have been recruited, since the young PPI contributors encouraged their friends to participate in a study on reproductive health in young people. Despite reported PPI activity in developing the grant application (3 out of 15, 20%), managing the research (3 out of 15, 20%), and conducting the research (2 out of 15, 13%), Principal Investigators reported minimal perceived impact in these areas.

No Principal Investigators reported a negative impact of PPI on them personally but most (10 out of 15, 67%) believed that it had little impact on the reputation of their institution. However, most researchers (13 out of 15, 87%) reported that they would engage with PPI in their research again, regardless of whether or not it was a requirement set down by funders. From the free text responses in the questionnaire, some researchers expressed a positive impact of PPI:

> *"Very helpful in helping me gain a better understanding of the issues involved with [disease]"* (PI119)
>
> *"Feedback from patient representatives raised some key concerns which were important to address in our branding and overall presentation from the outset"* (PI116)
>
> *"[PPI] provide a reality check on patient benefit of research, broaden perspectives and focus on the lived experience"* (PI89)

A few principal investigators offered some alternative experiences and less positive viewpoints of PPI in research:

> *"Sometimes patients are really helpful and give good ideas and have good contacts. I am sorry to be cynical but it is also a requirement for funders so you HAVE to do it"* (PI70)
>
> *"Young people can be unreliable – it's sometimes difficult to know whether they will turn up or not, and to plan accordingly*." (PI90)

While the respondent in the above quote has commented on young people, it should be noted, however, that this is not generalizable of all young PPI contributors. The participation of all PPI contributors can be impacted by many factors, such as availability on scheduled meeting dates, changes in health status and other commitments.

3.1 Quality-Impact Index scores

Figure 5 shows the Quality-Impact Index scores based on the Principle Investigators' responses relating to the quality and impact of PPI activities for the 15 projects included in the research survey. There was a moderate positive correlation between the Quality Score (number of quality indicators met by a project) and Impact Score (number of PPI activities in which the PI reported a perceived impact) (Pearson correlation coefficient, r = .50, p = .056). Though statistically insignificant, this results suggests a greater perceived impact of PPI activity for projects where a higher number of quality indicators for PPI were met.

[FIGURE 5 HERE]

4. The financial costs of PPI

The most significant cost from a researcher perspective appeared to be the researcher's time. However, researchers reported a variable numbers of hours related to PPI, ranging from 0 to 30 hours as a total across all activities.

Half of Principal Investigators (8 out of 15, 53%) reported that they 'Always' or 'Sometimes' offered some form of payment to PPI contributors and a third (5 out of 15, 30%) reimbursed expenses. Principal Investigators reported that public members received payment for attending meetings (6 out of 15, 40%), reviewing documents in their own time (2 out of 15, 13%) and attending events (1 out of 15, 7%). Payment for other activities (e.g. conducting the research, responding to letters and emails from researchers) was not reported. Travel costs (e.g. car mileage, public transport, parking) and food and drink were the only expenses reimbursed.

However, few studies were able to confirm the actual financial costs associated with PPI. A third of the respondents reported difficulty in providing general information about PPI in their project(s) (5 out of 15, 30%) and almost half (7 out of 15, 47%) found it difficult to give information relating to the costs of PPI. Free text responses indicated that the researchers did not keep records of the costs associated with PPI activity in their projects. Due to the overall lack of systematic recording of resourcing PPI activity and the time lag for some of the older projects in the sample, the responses of the Principal Investigators on the costs and time commitment of PPI are likely to be underestimated.

5. A new Cost and Consequences Framework of PPI in research

Table 4 presents a framework of the individual costs and consequences for both the research/researcher and patient. Responses from the researcher survey provided information for the costs and consequences framework under sub-headings of researcher, research project, research institution and funder. As no responses were received for the patient survey, costs and consequences identified from the literature are included in the framework.

[TABLE 4 Costs and Consequences Framework HERE]

6. Recommendations workshop

Following the data analysis by the research team, public members who had been previously involved with the project plus other members of a Research User Group at Keele attended a workshop. The aim was to co-produce recommendations to improve PPI practice within the SPCR. Three PPI contributors of the study, eight other members of the Research User Group at Keele and the research team came together to discuss as a group the key findings of the study and consider recommendations to address the findings, build upon existing good practice and improve PPI in research. During the workshop consensus was achieved on the general content of the recommendations. Following the workshop, the research team mapped the recommendations to those in the NIHR 'Going the Extra Mile' report [3], INVOLVE's "Values and Principles Framework' [4] and the SPCR strategy for PPI [26]. To ensure consistency with these national priorities and directions, the research team refined the final wording of our recommendations, which were reviewed and agreed upon by our PPI contributors (Table 5).

[TABLE 5 Recommendations for improving the practice and delivery of PPI in research HERE]

DISCUSSION

This is the first study to systematically investigate the quality and impact of PPI across a wide cohort of primary care research studies. Furthermore, we have applied recognised quality indicators to assess the quality of PPI and linked the level of quality with its perceived impact on the research process. We have also identified and developed a framework of the costs (financial and non-financial) and consequences of PPI in primary care research. This should enable others to assess the impact of different approaches to PPI on key research outcomes and the people involved.

Previous studies have tended to focus on *scope* and *impact*, i.e. *what* PPI has taken place and *how* this may or may not have made a difference to the research process. However, knowing the *quality* of PPI (or how well it has been undertaken) is just as essential. New Standards for Public Involvement are expected in 2018 [27]. INVOLVE have published resources on good practice and approaches to PPI, including a *Values and Principles Framework* [4]. There are a few appraisal guidelines and frameworks for assessing the quality of PPI [25, 28], including a recently published framework designed to help researchers to recognise the ethical issues when involving the public during the design of research studies [29]. In particular, Boote et al. [25] produced eight principles of successful PPI in NHS research, with each principle having at least one clear and valid indicator (or measure) of good practice (Table 2). Furthermore, the GRIPP2 (Guidance for Reporting Involvement of Patients and Public) checklist has been developed to enhance the quality of PPI reporting [16]. Nevertheless, we are not aware of any studies that have formally evaluated the quality of PPI in research.

Our study has shown that PPI has not been routinely undertaken across SPCR-funded research studies. While some have included PPI at different stages of research, most projects have not in either developing the grant application, and/or whilst conducting the research, or both. This does not seem to have improved over time, despite becoming a requirement of funding. PPI was reported most frequently in the management of studies (e.g. steering group membership), and designing study materials (e.g. questionnaires) and methods (e.g. recruitment strategy, intervention design), but less frequently for other aspects of the research process (e.g. developing and reviewing participant information leaflets, commenting on the study protocol, conducting the research, developing future research areas). Similar studies on the extent and type of PPI have reported similar findings [5, 6]. Furthermore, the extent of PPI in primary care research was inconsistent across research design, with PPI seemingly less prevalent in study designs where there was less direct contact with patient/participants during the study. The wide variability of PPI across health research topics identified in this study is difficult to interpret or provide reasons for but our findings suggest that the level of PPI in the research of some health conditions is markedly lower. These findings indicate that greater awareness of the value of PPI throughout the research cycle, across research designs and in different health conditions is required.

The quality of PPI reported by Principal Investigators did not always meet guidelines for best practice. Though there were a few projects which conducted good quality PPI, findings from our researcher survey highlighted particular areas where best practice was not being followed. For example, in a number of projects PPI contributors were not offered payment for their time or reimbursement of expenses; and few projects documented the role of PPI contributors or engaged with them regarding the dissemination of research findings. We assessed quality in terms of meeting indicators of good practice. While, we were not able to identify specific examples of *poor* practice in either the analysis of project documents or the researcher survey, we did find that researchers spent variable amounts of time on PPI activities during a study (ranging 0 to 30 hours). This suggests that those who spent fewer or minimal hours on PPI may not have taken sufficient time to have meaningfully engaged with the public.

Furthermore, whilst we acknowledge that not all of Boote et al's quality indicators may be relevant for all study types (e.g. obtaining advice from PPI contributors on recruitment issues may not be relevant for studies where there is no participant recruitment, such as systematic reviews or some cohort studies) they provide a benchmark of quality that ought to be achieved if a study involved members of the public. This study was conducted before INVOLVE's *Values and Principles Framework* was published [4]. However, most of Boote et al quality indicators are incorporated in some form or another within this new framework and the soon to be launched Standards for Public Involvement [27] . Yet it is too early at this stage to tell how INVOLVE's *Values and Principles Framework* will be used in practice and/or how well the National Standards for Public Involvement might be used to assess and improve the quality of PPI. Nevertheless, future studies should consider how the National Standards for Public Involvement, GRIPP2 reporting checklist and other PPI resources and tools complement each other, in the context of the costs and consequences of PPI highlighted in this study. This should help drive forward improvements in this field in a coherent and consistent way. For example the financial and non-financial costs of PPI highlighted in this study should be considered when using INVOLVE's Budgeting for Involvement Cost Calculator.

Our survey highlighted that researchers found it difficult to provide information on PPI and its costs. We have also shown that it is difficult to contact public members who have been involved in research, as researchers were unable to pass the PPI contributor survey to those involved in their research. Reasons for are not entirely clear and we are not aware of similar findings in the literature. Some researchers reported that they did not have this information. So it is possible that the researchers and/or their organisations did not systematically and routinely keep records of PPI activity (or at least were not able to readily access these records at the time of completing the survey). This could be due, in part, to a possible lack of administrative support in some organisations.

A key finding of this study was the inconsistency between the plans to conduct PPI during a study and the reported delivery of that activity. The fact that PPI activities were often different to those described in research proposals - and sometimes planned PPI was not conducted at all - is problematic. Mathie et al also reported a lack of documentation providing evidence of monitoring or how the PPI strategy within a study may have changed as the research develops [5]. This suggests a need for research funders to keep a check on PPI activity within research projects and to help researchers to make realistic plans for PPI at the outset.

This study complements the results of similar studies:

- Mixed methods design and interventional trials tended to have the most PPI compared to other research designs [5, 6]. PPI was less common in observational and cohort studies [5].
- The most commonly stated PPI activity was membership of steering committees and reviewing patient information leaflets [5, 6]
- Increased time in building relationship with PPI contributors and planning and managing PPI is a major consideration for researchers [6, 10, 11, 12]
- There is limited amount of available information about PPI in publicly accessible research documents [5]

The limitations to this study include:

1. A low response rate to the researcher survey (24%). While this is in line with similar studies [5], the length of questionnaire and the approximate 45 minute completion time may have been a barrier to participation. Nevertheless, the level of detail was necessary to obtain a comprehensive understanding of PPI in primary care research. Secondly, some researchers commented that it was difficult to recall details of the PPI in studies that may have begun as early as 2008.

2. Although it is not unreasonable to suggest that the direct costs of PPI (e.g. payment to individuals, reimbursement of expenses, room hire, etc.) could and should be recorded, it is likely that financial systems differ across universities, and there may also be problems, particularly in terms of workload, obtaining access to that level of detail once a project has been completed.

3. Data from the documentary analysis was inconsistent due to changes in the SPCR application and reporting forms over the funding rounds. Nevertheless, many of the annual/final reports contained very little information, and were incomplete or ambiguous. This highlights a problem with recording and reporting of PPI activity. This made extracting data difficult and the research team sometimes made a judgement by consensus as to the meaning of the information.

4. We were not able to conduct the PPI contributor survey as we experienced difficulties with accessing PPI contributors to invite them to participate. As the contact details of PPI contributors involved with SPCR work were not available, we decided to ask Principal Investigators to pass the survey to PPI contributors who had been involved in their projects. Unfortunately, all Principal Investigators who responded to the survey were either unable (due to lack of recorded contact information) to pass on the postal survey to PPI contributors.

This meant that we were unable to gather data on the costs and consequences of involvement from PPI contributors. However, members of the Research User Group at Keele were involved in the analysis of the data and the development of recommendations to ensure some representation of the patient and public perspective.

4. We originally planned to observe PPI in research studies. In the final section of the survey, Principal Investigators were asked to indicate whether they had any forthcoming meetings with PPI planned, and if so whether they were happy for two researchers to observe the meeting. Unfortunately, most respondents did not have any meetings with PPI planned, and one respondent was not willing for us to observe their meeting.

Role of PPI in the study

Public members have played a fundamental role in shaping the project, from the initial development of the research idea to the dissemination and implementation of findings. The role of PPI has been described and embedded through this article. In addition to the activity already described, there has also been involvement in the dissemination of early project findings with a PPI contributor co-presenting with a researcher at the INVOLVE Conference 2014. Two lay co-applicants were invited to comment on and contribute to the plain English summary of this article and the final project report to the SPCR. They also commented on their experience of the research study and their views of its findings (Table 6).

While this study was funded by the SPCR, we did not include it as part of the analysis of documentation and surveys in order to remain independent. However, we worked with our PPI contributors (AH, CR) to conduct our own self-assessment of the quality of PPI in this study against Boote et al's quality indicators [25] as a separate exercise (Additional File 3). We achieved 10 out of the 11 quality indicators. This suggests the PPI in this study was of high quality. The single indicator not achieved was PPI in advising on informing participants about study progress. This might have been achieved if the survey of PPI contributors had been completed.

Furthermore, to highlight the benefits and challenges of PPI experienced, we produced our own Cost and Consequences Framework for this study (see Additional File 4). This provides an example of the use of the Cost and Consequences Framework in practice. We have included relevant items about the PPI activity that we experienced during the course of this study. We have not included items relating to 'researchers gaining a better understanding of the condition of interest' as this was not a study of a particular health condition. The exercise has identified areas that we need to be aware of and improve on in future studies involving PPI contributors (e.g. ensuring all PPI activities are fully costed and budgeted), and many benefits of PPI that need to be reported and shared (e.g. PPI as a motivating factor, with PPI contributors bringing an enthusiasm to the project, and a keenness to see results).

[TABLE 6 Experience of lay co-applicants and co-authors (CR, AH) regarding their involvement in this study and its findings HERE]

[Additional File 3 Self-assessment of the quality of PPI for this study HERE]

[Additional File 4 A Cost and Consequences Framework for this study HERE]

Future impact of this study

The results of this study have provided a detailed account of PPI within primary care research and have shown the variability of PPI in projects to date. In particular, findings have highlighted areas for improvement in PPI. This has led to the development of recommendations for good PPI practice, in collaboration with members of a Research User Group, to ensure that the patient perspective is represented. Implementation of these recommendations, which complement the NIHR 'Going the Extra Mile' report [3], INVOLVE's "Values and Principles Framework'[4], Standards for Public Involvement [27] and the NIHR School for Primary Care Research (SPCR) PPI Strategy [26], will ensure that PPI activities meet quality indicators and that standardised records of PPI activities are kept to facilitate the evaluation of impact. The new Costs and Consequences Framework considers many potential benefits, harms and costs (financial and non-financial) of PPI which will help others assess the wider impacts of PPI. Further, the surveys developed within the project can be used by the SPCR and others to collect detailed data on the costs and consequences of PPI in future projects and also alter grant application forms and project reports to improve reporting of PPI activities.

CONCLUSION

PPI in primary care research is inconsistent in terms of its extent, nature and quality across research design and topics. There is scope for improvement in terms of:

1) establishing the costs and consequences for researchers and PPI contributors of involvement in research
2) recording and reporting the contribution and impact of PPI

3) promoting and implementing best practice, and PPI.

This study did identify pockets of good practice and this tended to be reported as making a positive impact on researchers and research studies. We were unable to access PPI contributors to obtain their views and experiences. Nevertheless, the public perspective, through PPI involvement in our study, was instrumental in interpreting the findings and co-producing recommendations to improve PPI in primary care research. The findings of this study have informed a cost and consequences framework which may help others assess the impact of PPI.

LIST OF ABBREVIATIONS

PPI	Patient and public involvement
SPCR	School for Primary Care Research
NIHR	National Institute for Health Research
UK	United Kingdom
NHS	National Health Service

DECLARATIONS

Ethics approval and consent to participate

Ethical approval was obtained from Keele University's Research Ethics Committee (21st March 2014). Respondents to the survey were informed that a return of a completed questionnaire implied informed consent to participate in the study and share anonymous information publicly.

Consent for publication

Not applicable

Availability of data and materials

The datasets used and/or analysed during the current study are available in an anonymous format from the corresponding author on reasonable request. The questionnaires used in the survey are available in Additional Files 1 and 2.

Competing interests

The authors declare that they have no competing interests

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Authors' contributions

CJ, FS, SJ, AH, CR and PG conceived of the research. All authors contributed to collaborative decision making, design of the survey, analysis and planning. SB, SM, SJ and PK analysed the data obtained from the documentary analysis and the surveys. SB, SM and CJ wrote the first draft and assembled revisions based on the comments of co-authors. SB, SM, CJ, AH and CR, along with RUG members, interpreted the findings and co-wrote the recommendations. SJ and PK developed the Cost and Consequences framework. AH and CR contributed to the Plain English Summary and provided their experiences of being involved in the study. SB and CJ revised the manuscript. All authors read and approved the final manuscript.

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ENDNOTES

[i] We have used the term 'PPI contributors' to collectively describe members of the public actively involved in research projects, including patients, members of the public, service users and carers.

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TABLES

Table 1 Types of data extracted from the project documentation

- Study design
- Disease/condition studied
- Study population
- SPCR funding round
- Presence of a section dedicated to PPI within the document
- Presence of PPI in the development of the grant application (including a description of the type and number of public members involved)
- Description of plans for PPI (including a description of the type and number of public members involved)
- Details of PPI activities conducted
- Explanation for any lack of PPI
- References to a specific budget for PPI
- References to rewards and recognition for involvement.
- Level of consistency between planned and reported PPI activities was noted (including explanations for any discrepancies).

Table 2 The principles and indicators of successful consumer involvement in NHS research. Adapted from Boote *et al* [25]

	Principle	**Indicator(s)**
1	The roles of consumers are agreed between the researchers and consumers involved in the research	The roles of consumers in the research were documented
2	Researchers budget appropriately for the costs of consumer involvement in research	Researchers applied for funding to involve consumers in the research
		Consumers were reimbursed for their travel costs
		Consumers were reimbursed for their indirect costs (e.g. carer costs)
3	Researchers respect the differing skills, knowledge and experience of consumers	The contribution of consumers' skills, knowledge and experience were included in research reports and papers
4	Consumers are offered training and personal support, to enable them to be involved in research	Consumers' training needs related to their involvement in the research were agreed between consumers and researchers
		Consumers had access to training to facilitate their involvement in the research
		Mentors were available to provide personal and technical support to consumers
5	Researchers ensure that they have the necessary skills to involve consumers in the research process	Researchers ensured that their own training needs were met in relation to involving consumers in the research
6	Consumers are involved in decisions about how participants are both recruited and	Consumers gave advice to researchers on how to recruit participants to the research

	kept informed about the progress of the research	Consumers gave advice to researchers on how to keep participants informed about the progress of the research
7	Consumer involvement is described in research reports	The involvement of consumers in the research reports and publications was acknowledged
		Details were given in the research reports and publications of how consumers were involved in the research process
8	Research findings are available to consumers, in formats and in language they can easily understand	Research findings were disseminated to consumers involved in the research in appropriate formats (e.g. large print, translations, audio, Braille)
		The distribution of the research findings to relevant consumer groups was in appropriate formats and easily understandable language
		Consumers involved in the research gave their advice on the choice of methods used to distribute the research findings

Table 3 **PPI in SPCR projects, by study design, health condition, population and population age.**

	Projects (Grant applications)		Projects with evidence of PPI in developing the grant application (N=200)			Projects with evidence of plans for PPI during the study in the grant application (N=200)			Projects (Annual/ Final Reports)[a]		Projects with evidence of PPI reported in annual/final reports (N=181)		
	n	(%)	n	(%)	(Relative %)[b]	n	(%)	(Relative %)[b]	n	(%)	n	(%)	(Relative %)[c]
All Projects	**200**	**(100)**	**47**	**(23.5)**		**113**	**(56.5)**		**181**	**(181)**	**83**	**(46.1)**	
Study Design													
Mixed methods	47	(23.5)	15	(7.5)	**(31.9)**	30	(15.0)	**(63.8)**	39	(21.5)	24	(13.3)	**(61.5)**
Qualitative	36	(18.0)	9	(4.5)	**(25.0)**	23	(11.5)	**(63.9)**	30	(16.6)	17	(9.4)	**(56.7)**
Longitudinal cohort	29	(14.5)	5	(2.5)	**(17.2)**	18	(9.0)	**(62.1)**	29	(16.0)	11	(6.1)	**(37.9)**
Intervention trial	25	(12.5)	6	(3.0)	**(24.0)**	15	(7.5)	**(60.0)**	23	(12.7)	14	(7.7)	**(60.9)**
Systematic reviews	17	(8.5)	2	(1.0)	**(11.8)**	7	(3.5)	**(0.0)**	17	(9.4)	2	(1.1)	**(11.8)**
Retrospective cohort	13	(6.5)	2	(1.0)	**(15.4)**	4	(2.0)	**(30.8)**	13	(7.2)	3	(1.7)	**(23.1)**
Secondary analysis	8	(4.0)	0	(0.0)	**(0.0)**	4	(2.0)	**(50.0)**	8	(4.4)	3	(1.7)	**(37.5)**
Cross sectional	7	(3.5)	4	(2.0)	**(57.1)**	3	(1.5)	**(42.9)**	7	(3.9)	4	(2.2)	**(57.1)**

	Projects (Grant applications)		Projects with evidence of PPI in developing the grant application (N=200)			Projects with evidence of plans for PPI during the study in the grant application (N=200)			Projects (Annual/ Final Reports)[a]		Projects with evidence of PPI reported in annual/final reports (N=181)		
	n	(%)	n	(%)	(Relative %)[b]	n	(%)	(Relative %)[b]	n	(%)	n	(%)	(Relative %)[c]
Methodological	5	(2.5)	1	(0.5)	**(20.0)**	4	(2.0)	**(80.0)**	4	(2.2)	1	(0.6)	**(25.0)**
Case control	4	(2.0)	0	(0.0)	**(0.0)**	1	(0.5)	**(25.0)**	3	(1.7)	0	0	**(0.0)**
Multi-stage study[d]	4	(2.0)	2	(1.0)	**(50.0)**	2	(1.0)	**(50.0)**	2	(1.1)	2	(1.1)	**(100)**
Individual participant meta analysis	3	(1.5)	0	(0.0)	**(0.0)**	1	(0.5)	**(33.3)**	2	(1.1)	1	(0.6)	**(50.0)**
Other[e]	2	(1.0)	1	(0.5)	**(50.0)**	1	(0.5)	**(50.0)**	4	(2.2)	2	(1.1)	**(50.0)**
Health Condition Under Study													
General Health	28	(14.0)	9	(4.5)	**(32.1)**	14	(7.0)	**(50.0)**	22	(12.2)	9	(0.0)	**(40.9)**
Cardiovascular	27	(13.5)	4	(2.0)	**(14.8)**	16	(8.0)	**(59.3)**	28	(15.5)	10	(5.5)	**(35.7)**
Mental Health	21	(10.5)	5	(2.5)	**(23.8)**	12	(6.0)	**(57.1)**	17	(9.4)	11	(6.1)	**(64.7)**

	Projects (Grant applications)		**Projects with evidence of PPI in developing the grant application (N=200)**			**Projects with evidence of plans for PPI during the study in the grant application (N=200)**			**Projects (Annual/ Final Reports)[a]**		**Projects with evidence of PPI reported in annual/final reports (N=181)**		
	n	(%)	n	(%)	(Relative %)[b]	n	(%)	(Relative %)[b]	n	(%)	n	(%)	(Relative %)[c]
Cancer	16	(8.0)	7	(3.5)	**(43.8)**	8	(4.0)	**(50.0)**	16	(8.8)	10	(5.5)	**(62.5)**
Metabolic and Endocrine	14	(7.0)	1	(0.5)	**(7.1)**	6	(3.0)	**(42.9)**	16	(8.8)	9	(5.0)	**(56.3)**
Musculoskeletal	14	(7.0)	3	(1.5)	**(21.4)**	6	(3.0)	**(42.9)**	13	(7.2)	6	(3.3)	**(46.2)**
Respiratory	13	(6.5)	3	(1.5)	**(23.1)**	6	(3.0)	**(46.2)**	12	(6.6)	5	(2.8)	**(41.7)**
Multimorbidity	7	(3.5)	0	(0.0)	**(0.0)**	1	(0.5)	**(14.3)**	8	(4.4)	3	(1.7)	**(37.5)**
Stroke	7	(3.5)	1	(0.5)	**(14.3)**	3	(1.5)	**(42.9)**	5	(2.8)	2	(1.1)	**(40.0)**
Infection	5	(2.5)	1	(0.5)	**(20.0)**	3	(1.5)	**(60.0)**	5	(2.8)	1	(5.0)	**(20.0)**
Renal and Urogenital	5	(2.5)	2	(1.0)	**(40.0)**	5	(2.5)	**(100)**	5	(2.8)	3	(1.7)	**(60.0)**
Reproductive Health and Childbirth	5	(2.5)	2	(1.0)	**(40.0)**	5	(2.5)	**(100)**	5	(2.8)	4	(2.2)	**(80.0)**

	Projects (Grant applications)		Projects with evidence of PPI in developing the grant application (N=200)			Projects with evidence of plans for PPI during the study in the grant application (N=200)			Projects (Annual/ Final Reports)[a]		Projects with evidence of PPI reported in annual/final reports (N=181)		
	n	(%)	n	(%)	(Relative %)[b]	n	(%)	(Relative %)[b]	n	(%)	n	(%)	(Relative %)[c]
Neurological	3	(1.5)	0	(0.0)	**(0.0)**	3	(1.5)	**(100)**	1	(0.6)	1	(0.6)	**(100)**
Cancer, Mental Health	1	(0.5)	1	(0.5)	**(100)**	0	(0.0)	**(0.0)**	1	(0.6)	1	(0.6)	**(100)**
Inflammatory and Immune System	1	(0.5)	0	(0.0)	**(0.0)**	0	(0.0)	**(0.0)**	0	(0.0)	0	(0.0)	
Oral and Gastrointestinal	1	(0.5)	0	(0.0)	**(0.0)**	0	(0.0)	**(0.0)**	1	(0.6)	0	(0.0)	**(0.0)**
Skin	1	(0.5)	1	(0.5)	**(100)**	1	(0.5)	**(100)**	1	(0.6)	1	(0.6)	**(100)**
Other[f]	31	(15.5)	7	(3.5)	**(22.6)**	24	(12.0)	**(77.4)**	23	(12.7)	8	(4.4)	**(34.8)**
Study population													
Patients	134	(67.0)	26	(13.0)	**(19.4)**	68	(34.0)	**(50.7)**	123	(68.0)	54	(29.8)	**(43.9)**
Patients & HCPs	37	(18.5)	11	(5.5)	**(29.7)**	24	(12.0)	**(64.9)**	31	(17.1)	20	(11.0)	**(64.5)**

	Projects (Grant applications)		**Projects with evidence of PPI in developing the grant application (N=200)**			**Projects with evidence of plans for PPI during the study in the grant application (N=200)**			**Projects (Annual/ Final Reports)[a]**		**Projects with evidence of PPI reported in annual/final reports (N=181)**		
	n	(%)	n	(%)	(Relative %)[b]	n	(%)	(Relative %)[b]	n	(%)	n	(%)	(Relative %)[c]
Healthcare professionals (HCPs)	15	(7.5)	5	(2.5)	**(33.3)**	10	(5.0)	**(66.7)**	8	(4.4)	3	(1.7)	**(37.5)**
General public	13	(6.5)	5	(2.5)	**(38.5)**	10	(5.0)	**(76.9)**	15	(8.3)	7	(3.9)	**(46.7)**
Carers	1	(0.5)	0	(0.0)	**(0.0)**	1	(0.5)	**(100)**	1	(0.6)	0	(0.0)	**(0.0)**
Study population age													
Unspecified	93	(46.5)	19	(9.5)	**(20.4)**	54	(27.0)	**(58.1)**	83	(45.9)	35	(19.9)	**(43.4)**
Adult (18+ years)	89	(44.5)	24	(12.0)	**(27.0)**	50	(25.0)	**(56.2)**	83	(45.9)	42	(23.2)	**(50.6)**
Adult and Children	11	(5.5)	4	(2.0)	**(36.4)**	7	(3.5)	**(63.6)**	9	(5.0)	5	(2.8)	**(55.6)**
Children and young adults (0 – 17 years)[g]	7	(3.5)	0	(0.0)	**(0.0)**	2	(1.0)	**(28.6)**	6	(3.3)	1	(0.6)	**(16.7)**

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[a] Included one project whose data on PPI was obtained from an SPCR poster; [b] Percentage relative to the number of projects in each category in grant applications ; [c] Percentage relative to the number of projects in each category in annual/final reports; [d] Multi-stage studies included case control and intervention trial (1), cross sectional

and longitudinal cohort (1), systematic review and longitudinal cohort (1), systematic review and secondary analysis; [e] Included projects to set up and maintain a SPCR PPI group (1) and a preliminary descriptive study (1); [f] Conditions not classified under the Health Research Classification System [24]; [g] It was not always possible to determine the ages or age range of children from the study documentation. Sometimes, ages were provided, sometimes the documentation referred to 'children'. So we have assumed children and young adults to be 17 and under.

Table 4 Costs and Consequences Framework

Impact upon		**Costs (-)**	**Benefits (+)**
Researcher		- Time (recruiting PPI contributors; travelling to meet with PPI contributors; meetings; electronic communication; preparing newsletters) - *Increased pressure/stress* - *Sensitivity to criticism*	+ A motivating factor, with PPI contributors bringing an enthusiasm to the project, a keenness to see results + PPI contributors supportive of the project + Researchers gaining a better understanding of the condition of interest
Research Project	Shaping the research question and maintaining focus		+ Setting and maintaining focus on the research question + Addressing important issues but also ensuring a degree of realism
	Research methods/design	- Can result in duplication of effort (PPI involvement and qualitative work)	+ Helping to make surveys and processes relevant, accessible and acceptable + Ensuring research is beneficial to patient group
	Recruitment & recruitment materials	- Potentially homogenous sample	+ Relevance, clarity & accessibility of recruitment materials + Making useful contacts, increasing recruitment rates
	Conducting & managing research	- PPI contributors can be unreliable (this was reported in the case of young people)	+ Validity and safety of research + Improved follow-up rates

		- Direct payment of PPI contributors for attending meetings - Travel costs (either the researcher visiting the PPI representative or the PPI representative attending meetings[1]) - Food and refreshment costs - External venues	
	Commenting on results		+ Opportunities to gain feedback and to validate the results. + PPI contributors helping to interpret the data.
	Dissemination	*- Financial cost of PPI contributors attending conferences and external events*	+ Promotion of outputs when these take the form of training modules or tool kits + Guidance in terms of presenting results in a format useful to non-researchers.
	Generating new research questions (expanding upon current research)		+ Generating new/future research questions

Research Institution	**-** Diversion of research funds to PPI (opportunity cost in terms of funded researcher time, etc.) **-** IT and other support infrastructures/resources (including printing & internal room bookings)	**+** Increased impact of research **+** Recognition as a centre with expertise and experience of involving patients and public in research (raising the institution's profile)
Funder		**+** Avoiding devoting resources to a topic which is not important (e.g. exploring an intervention which is not appealing to service users)
PPI contributors	**-** *Opportunity cost (paid work, child care, informal care & leisure time)* **-** *Monetary costs not reimbursed (travel, formal child care)* **-** *Negative impact on health associated with stress, anxiety or frustration* **-** *Complications in terms of state provided welfare payments*	**+** *Increased understanding & knowledge of one's own condition* **+** *Increased awareness of treatment options and how to access services* **+** *Developing or enhancing skills (e.g. public speaking, team work, IT) – possibly through formal training* **+** *Understanding of research and research processes* **+** *Positive emotional impact associated with meeting new people, feeling as though one is doing something worthwhile and generally being active*

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[1] Sometimes included within the direct payment

Entries in italics were identified from the literature but not verified by respondents

Table 5 Recommendations for improving the practice and delivery of PPI in research

Key Findings	Recommendations for improving PPI in research
	Best Practice
A. Overall PPI in research was low and inconsistent across research design and topics	1. Promote PPI as a core research function in all research by raising awareness of its value and impact
B. PPI was mostly limited to a few activities in the research cycle	2. Identify and share good examples of PPI activity across the research cycle to improve range and quality of PPI in future funded projects
	3. Raise awareness of and promoting the role of PPI in the lowest areas of activity, where appropriate and justified
C. 'Best Practice' was inconsistent	4. Create dedicated champion(s) for PPI within research institutions to promote best practice
	5. Establish and implement a best practice framework to enable appropriate and meaningful PPI
	6. Stimulate sharing of best practice and resources for PPI across research organisations and institutions
	7. Improve the skills of researchers and member of the public for PPI

	8. Establish a culture in which a) rewards and reimbursement of expenses are offered to PPI contributors as a matter of routine practice and b) PPI is appropriately costed in research
	9. Improve and support the recording and reporting of PPI
	10. Improve the accountability of public funded research to the general public
D. Time to do PPI is the biggest consequence to researchers	11. Raise awareness of time commitment for meaningful PPI so researchers can plan for it effectively
E. PPI is good for research and researchers	12. Continue to showcase and celebrate the impact of PPI in research
	SPCR Systems and Processes
A. Overall PPI activity in research was low	13. Increase the overall PPI activity in SPCR projects, by developing networks for PPI groups and researchers, and encouraging sustainable processes and infrastructure for PPI
B. PPI was mostly limited to a few activities in the research cycle	14. Increase the range of appropriate PPI in SPCR funded research, by providing more guidance and support to researchers and grant reviewers

C. PPI is poorly recorded and reported	15. Improve the recording and reporting of PPI in SPCR to promote transparency, support diversity and enable the evaluation of impact by improving reporting form templates and better monitoring of PPI in SPCR activities and funded research

Table 6 Experience of lay co-applicants and co-authors (CR, AH) regarding their involvement in this study and its findings

CR: "As a lay coordinator of a growing group of research users involved in a variety of primary care research projects across a clinical trials unit, I was very aware of the varied approaches to PPI being undertaken both regionally and nationally. So I was very interested in being involved in a project looking at PPI within a group of projects across one funder, looking particularly at the costs and benefits of PPI to the patients and the researchers, as not all costs are quantifiable and those that are, are not routinely recorded. Yet in my experience many patients and researchers go above and beyond what is asked of them, because they sincerely believe that patient involvement is an absolute must for good rigorous primary care research that can go on to be implemented to improve patients' daily care. I was also keen to be involved in looking at the results and how they could be used to inform PPI practice for the future.

However, it was disappointing that no opportunities for observations of meetings were forthcoming and quite worrying that no details of patients involved in the studies were available, so no real patient perspective could be obtained of what the costs and benefits to the patients were throughout the studies. So this highlights for me a gap in the literature where more research needs to be undertaken to fully understand the costs and benefits for the patients involved in primary care research.

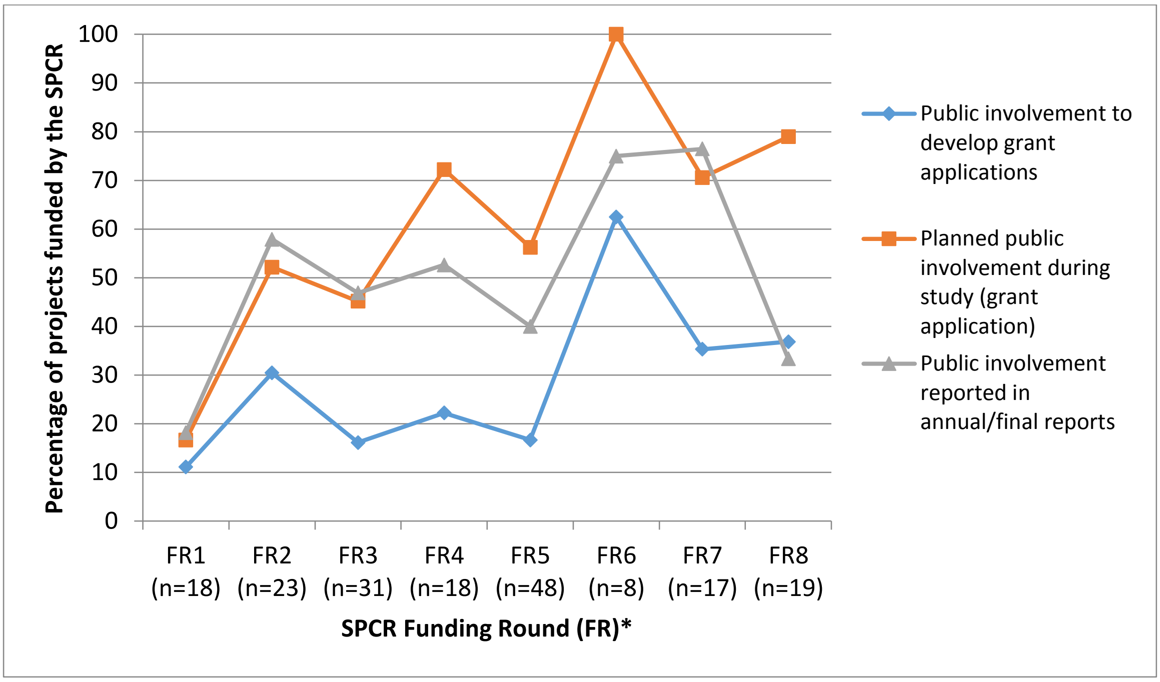
However I was impressed with the further specific recommendations on systems and processes compiled to fully integrate PPI into any future SPCR projects, which showed a real commitment from the SPCR to learn from the study findings."

AH: "I have enjoyed being a co-applicant on this study. I feel that I have been involved in all areas of the study. I think that the study is essential as it shows the inconsistency of reporting PPI.

I feel very disappointed about the response rate for the questionnaire, as no patient data was collected due to researchers being unwilling or unable to do this. This proves that there is a large gap here that needs to be addressed. I have also been surprised that in a lot of cases there were no plans for PPI, and for many researchers they held insufficient if any information. On the positive side - this paper will highlight areas for improvement and hopefully that will help to change attitudes and perspectives in the future."

Figure 1 Evidence of PPI in SPCR funded projects

Figure 1 Evidence of PPI in SPCR funded project grant applications and annual/final reports by funding round (N=200)



* Details of the respective funding rounds was unavailable for 18 projects

Figure 2 The nature of PPI in proposals and final reports

Figure 2 The nature of PPI planned in SPCR project proposals and reported in annual/final reports

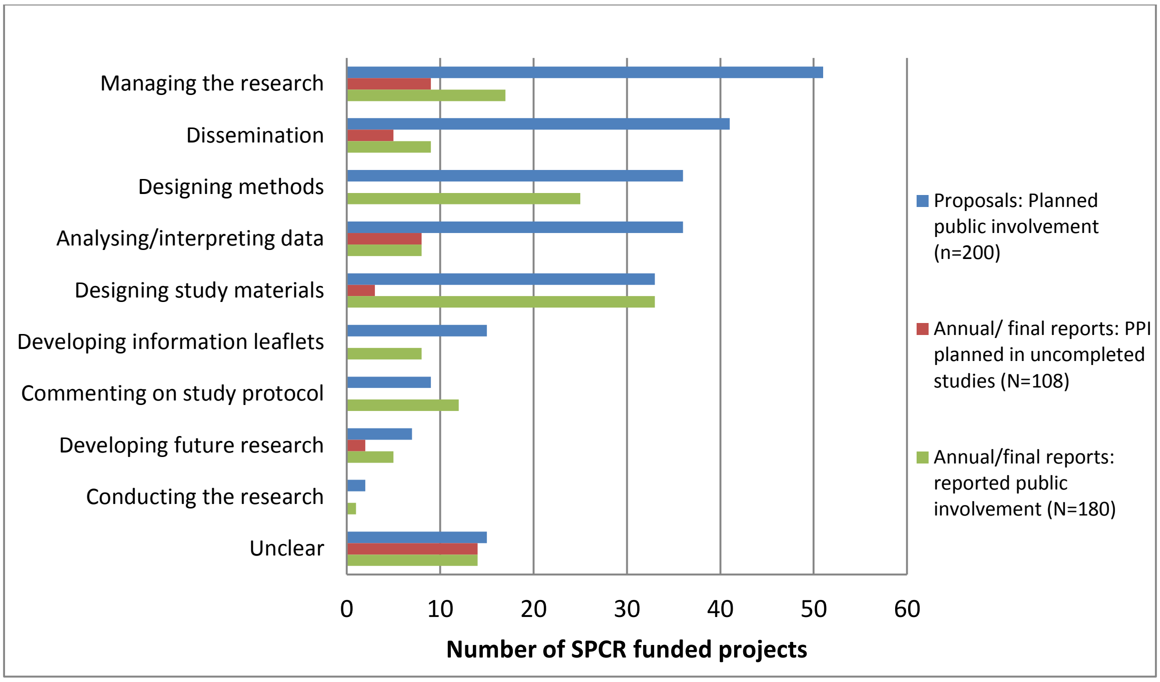


Figure 3 Consistency of PPI activities reported in annual/final reports compared to the plans for PPI within the project proposal (N=179)

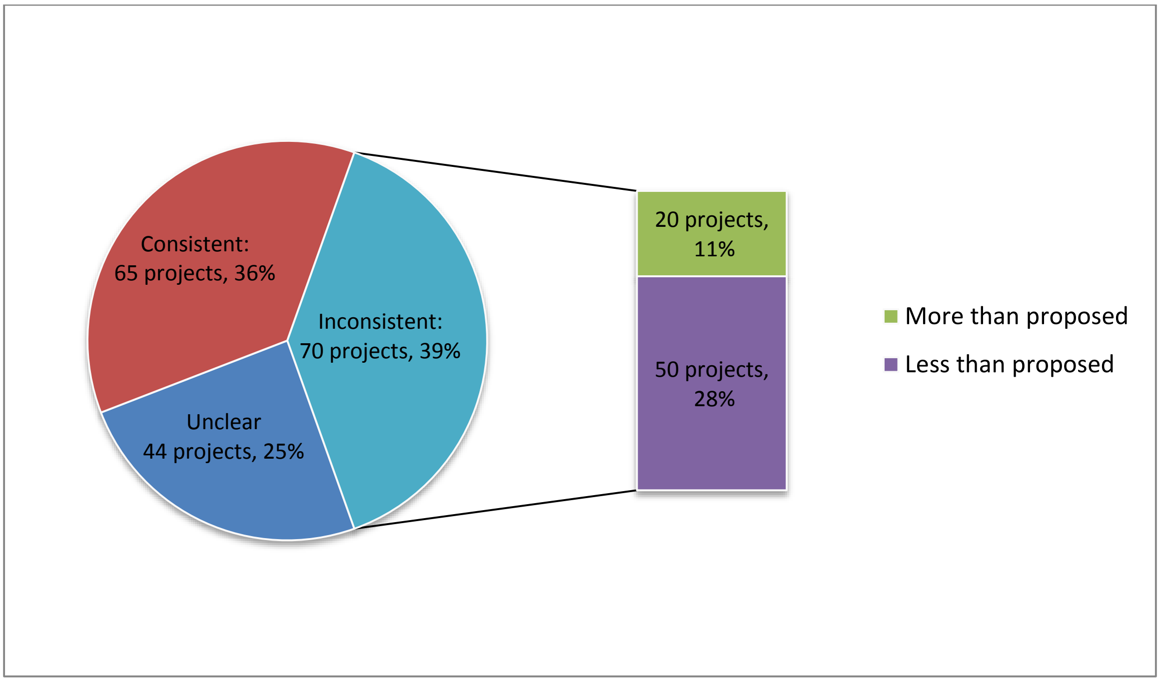
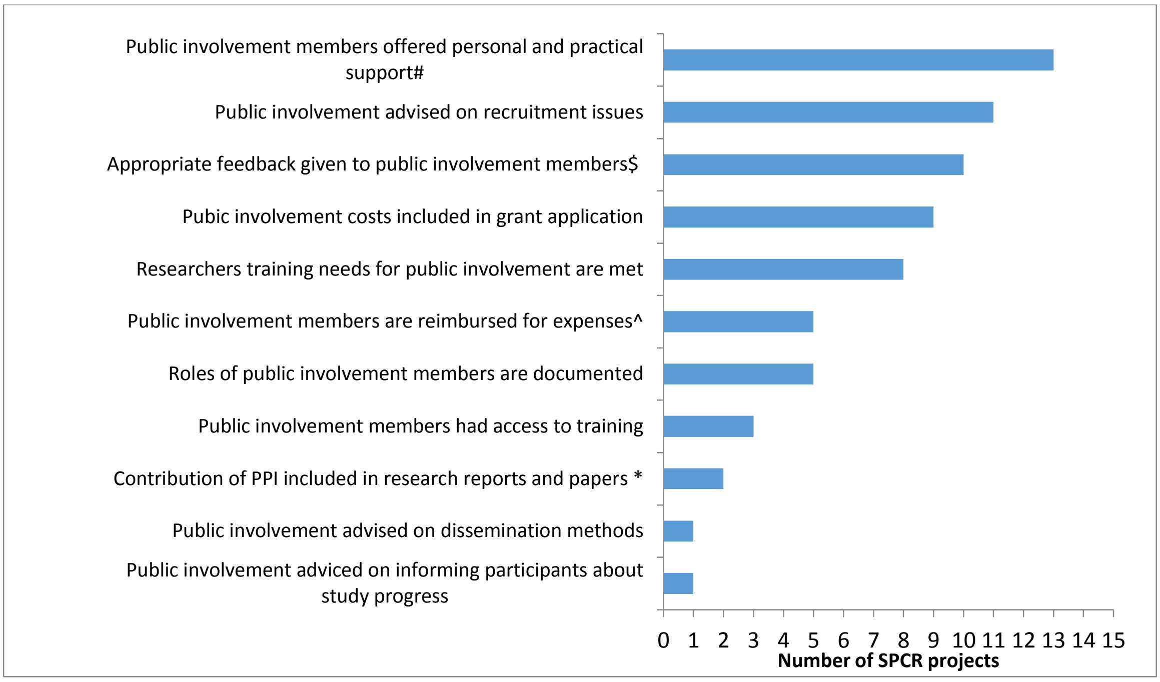
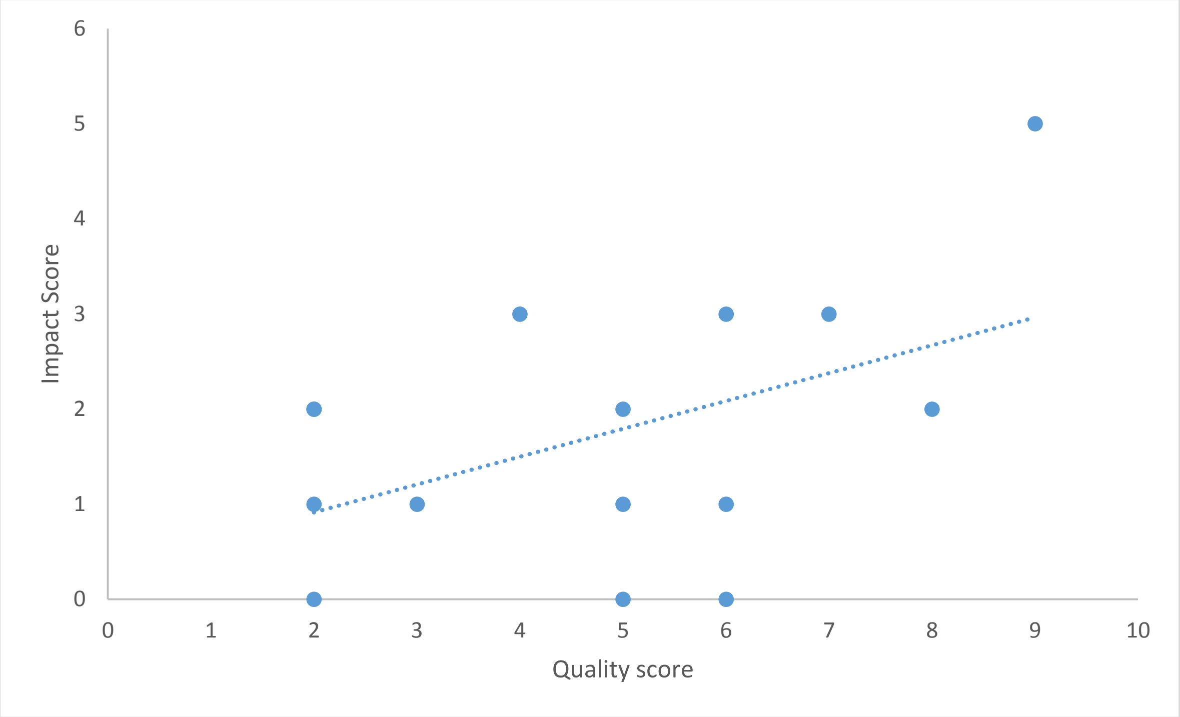


Figure 4 Levels of best practice for PPI in SPCR projects, according to Boote et al's quality indicators [25]



^ Combination of two quality indicators linked with expenses: travel costs and indirect costs (e.g. carer costs); * Combines three quality indicators: ‘contribution of PPI included in research reports and papers’, ‘PPI acknowledged in research reports and papers’ and ‘details of PPI reported in research reports and publications’. Data obtained from a PubMed search for articles associated with the 15 projects included in the analysis; # Adaptation of the quality indicator: ‘PPI offered mentors for personal and technical support’; $ Adaptation of the quality indicator: ‘Research findings were distributed to patients involved in the research in an appropriate format’

Figure 5 Quality-Impact Index scores: The association between the Quality Score (number of quality indicators met by a project) and the Impact Score (number of PPI activities which the PI reported a perceived impact)



Additional File 1 Researcher Survey_v3.0_17.03.14

Additional File 2 PPI Contributors Survey_v2.0_21.04.14