

**A review of patient and public  
involvement at the British Paediatric  
Surveillance Unit**

**Final Report  
June 2011**

**TwoCan Associates**

# Contents

	<b>Page</b>
<b>Thanks</b>	<b>3</b>
<b>Executive Summary</b>	<b>4</b>
<b>1. Introduction</b>	<b>10</b>
1.1 Aim of this review	10
1.2 Authors of this review	10
1.3 About the BPSU	10
1.4 Structure of this report	10
<b>2. Methods</b>	<b>12</b>
2.1 Overview	12
2.2 Semi-structured interviews with key stakeholders	12
2.3 Data analysis	13
2.4 Developing recommendations and action planning	13
2.5 Management of the review process	14
2.6 Next steps	14
<b>3. Lay membership of the Executive Committee</b>	<b>15</b>
3.1 Background	15
3.2 The current role of the lay members and their impact on the work of the committee	16
3.3 Suggestions for improvements/ future work	17
3.4 Future work – recruiting new members	18
3.5 Conclusions and recommendations	21
<b>4. Promoting PPI in BPSU studies</b>	<b>23</b>
4.1 Background	23
4.2 The importance of PPI in BPSU studies	24
4.3 The value of PPI at different stages of a BPSU study	25
4.4 Suggestions for improvements / future work	29
4.5 Conclusions and recommendations	35
<b>5. Expanding into new areas of PPI</b>	<b>36</b>
<b>6. Promoting PPI and supporting researchers</b>	<b>38</b>
<b>7. The challenges to taking forward PPI at the BPSU</b>	<b>40</b>
<b>8. Recommendations</b>	<b>41</b>
<b>9. Next steps</b>	<b>43</b>

## Thanks

We would like to thank all the people who agreed to be interviewed for this project. We are very grateful for their time.

**We would also like to thank the members of the Steering Group for their help with this review:**

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# EXECUTIVE SUMMARY

## 1. Introduction

The British Paediatric Surveillance Unit (BPSU – see [www.rcpch.ac.uk/bpsu](http://www.rcpch.ac.uk/bpsu)) supports research into rare childhood conditions and disorders. This research takes the form of national epidemiological surveillance – finding out how many children are affected, where and when cases have been presented and how they have been followed up.

This is a report of a review to assess the effectiveness of the patient and public involvement (PPI) processes at the BPSU. The review included an examination of how lay members have contributed to the work of the BPSU's Executive Committee as well as the processes by which the BPSU encourages PPI in its research studies.

The review was carried out by Bec Hanley, Carolyn Morris and Kristina Staley from TwoCan Associates ([www.twocanassociates.co.uk](http://www.twocanassociates.co.uk)). It has involved a review of relevant documents, BPSU application processes and the requirements for PPI in BPSU studies. We have also conducted a series of semi-structured interviews with selected members of the BPSU Executive Committee, lead researchers on selected BPSU studies and with the patients/ patient representatives who were involved in these studies. All data were analysed using framework analysis.

## 2. Lay membership of the Executive Committee

The BPSU Executive Committee oversees the strategic development of the organisation. Their other major role is to approve studies for inclusion on the BPSU system. Two lay members were invited to join the Committee in 2006.

### **The current role of the lay members and their impact on the work of the Executive Committee**

The role of the lay members was not well-defined at first and has evolved over time, with the input of the lay members and the support of past and present Chairs, other members of the Executive Committee and the staff at the BPSU. All of the clinical and scientific Executive Committee members who were interviewed said that they had found working with the lay members a very positive experience and that they had learnt a lot about PPI. The lay members are now viewed as full and equal members of the Committee.

The lay members play a crucial role in the evaluation of applications sent to the BPSU. They provide a lay perspective on potential studies which is separate from and independent of the clinical and scientific viewpoints. Clinical and scientific members of the Executive Committee said they found the lay members' input to be of particular value in assessing:

- the likely public perceptions of a topic
- how well public information sheets have been written
- the quality of the PPI in an application
- the ethical issues raised by a study
- potential links with relevant patient organisations

The lay members also help with advising the researchers about the PPI in their studies.

Although the involvement of lay members can be challenging and adds time to the approval process, Executive Committee members recognise this is necessary and appropriate. It is also recognised that the BPSU needs to 'practise what it preaches' and model best practice to outside researchers.

All of the interviewees were happy with the role lay members are now playing on the committee. There were no suggestions for changes or improvements, except perhaps for greater recognition and acknowledgment of the lay members' work.

### **Future work – recruiting new members**

The current lay members will soon reach the end of their term of office and therefore all interviewees were asked about their views on recruiting new members. The issues raised included:

- ***Broadening the lay membership:*** All of the interviewees recognised the importance of recruiting at least two lay members so that they can work together and support each other as a team.
- ***Running a formal recruitment process:*** There was broad agreement that new members should be recruited via an open and more formal recruitment process. This was thought to be important for the recruitment of high calibre individuals and to enable applicants to be sure the role was right for them.
- ***Providing training and support to new members:*** All the interviewees were conscious of not making assumptions about the training and support lay members might need and the importance of asking them what they would like help with, before developing any form of training or support.
- ***Paying lay members:*** Many interviewees felt that the payment of lay members for their time would demonstrate that their input was being valued – particularly as their role has become more time consuming. Although the payment of lay members' *expenses* is working well, this may need to be revisited with new members, in case their travel costs are greater and/or they cannot afford to be out of pocket for any length of time.
- ***Ensuring a successful handover:*** A number of the interviewees suggested that new members should be able to shadow existing members for one or two meetings, before the old members leave, so that they have time to build their understanding of the BPSU methodology.

### **3. Promoting PPI in BPSU studies**

The BPSU promotes PPI in its studies by making PPI a requirement for researchers wishing to use their approach. The lay members of the Executive Committee have had a major influence on the application process by developing the forms to include questions about PPI and making evaluation of PPI an integral part of the review process.

#### **The importance of PPI in BPSU studies**

All of the interviewees thought it important to involve patients and the public in BPSU studies because of the nature of this type of research. BPSU studies use patient data but do not have any patient participants. No one is asked to give consent for the

use of the information in their medical records. The data is anonymised and may be used without the patients' knowledge. This makes it all the more important to ensure openness, transparency and accountability to the public. PPI enables researchers to check out the acceptability of their approach with patients and the public. It is also critical to identifying studies that are controversial or of a sensitive nature that will need to be communicated carefully, particularly if they are likely to be picked up by the media.

### **The value of PPI at different stages of a BPSU study**

Since all BPSU studies follow much the same format it is possible to map PPI across the different stages and consider the value at each stage. The stages include:

- **Identifying and prioritising a research question:** It is valuable to involve patients at this very early stage because they are the only people who can confirm that a study is relevant and important to people affected by the condition or topic under investigation. PPI at this stage might also help researchers to realise that a study is of little public interest and so abandon unsuitable projects. PPI can also help to identify important research questions.
- **Developing the design of the study:** Most of the interviewees concluded that because BPSU studies use a standard approach that relies solely on retrospective data collection from paediatricians' notes, there is very limited scope for PPI to have an influence on the study design. No evidence was found of this type of involvement having had an impact.
- **Writing the BPSU application:** None of the interviewees were aware of PPI influencing the development of a BPSU application or were certain of its value.
- **Writing the public information leaflet (PIL):** PPI has been of value in the development of PILs by contributing additional content and by making PILs more accessible and more relevant to people affected by the condition / topic. PILs were felt to be a valuable communication tool, but many interviewees raised concerns about whether and how PILs were reaching the target audience and how often they are accessed, particularly when a rare condition is being studied.
- **Conducting the study:** There was no evidence of PPI taking place or having an impact at this stage.
- **Writing up and disseminating the findings:** In the same way that PPI helps with the production of PILs, PPI could help with producing lay-friendly reports of the findings of BPSU studies. PPI could ensure these reports were accessible and relevant to a lay-audience. As yet there is no evidence of this kind of involvement, mostly because few studies have been completed since PPI at the BPSU began. Patient organisations could also help with the dissemination of findings.

## **4. Expanding into new areas of PPI**

A number of suggestions for future work around PPI for the BPSU involved expanding current activity. These included:

## **Getting more involved in influencing public health policy**

A number of the interviewees thought that the BPSU could do more to ensure that the findings from BPSU studies are used to influence public health policy. They thought that if the BPSU were to develop activity in this area, then PPI would be very important. This was for a number of reasons:

- Policymakers are keen to know about public involvement and can be influenced by the human story.
- Patient organisations/ support groups often make helpful collaborators in influencing policy and practice.
- It also important to influence public opinion via the media – as this will in turn influence the policymakers. PPI can help with developing the media story and again adding the human dimension.

## **Involving young people**

Several of the interviewees recognised the importance of the *principle* of involving children and young people, because their views are often distinct to those of adults. However, no one was certain of where children and young people could *practically* be involved in a way that would bring added value. It was agreed that it would be inappropriate to try to engage children and young people as lay members of the Executive Committee. It was also recognised that this type of PPI requires a specialist approach. It would therefore require investment of resources and careful planning to ensure the process was meaningful and relevant to all involved.

## **5. Promoting PPI and supporting researchers**

Several of the interviewees thought that researchers would benefit from more practical advice on *how* to involve patients/ patient representatives in their work. The lay members were keen to ensure that PPI is a positive experience for researchers and an activity that has value for them.

## **6. Main challenge to taking forward PPI at the BPSU**

Many of the interviewees thought that the main challenge to taking forward any of the recommendations for future work is a lack of resources – both of funding and time, and in particular the limited capacity of Executive Committee members and BPSU staff to carry out the work required.

## **7. Recommendations for future work**

### ***Recognition of existing lay members and promotion of PPI***

1. Include information about the lay members and the work they do on the BPSU website, in newsletters and the annual report.

### ***Recruitment of new lay members***

2. The Executive Committee should work together as a whole to decide:
  - how many new lay members it requires and why
  - the essential/ desirable and skills and experience they will require of new members

- whether it wishes to pay lay members in future and how funding might be obtained for this purpose
  - the process for recruiting new people
3. Evidence of best practice should be sought from other organisations to help shape any new developments in the lay membership (e.g. payment policies)
  4. Recruitment needs to be planned and timed carefully to allow a handover from existing lay members and for new members to receive an induction before they start.
  5. Any new scientific or clinical members of the committee should also be given an induction and importantly an explanation of the role of lay members.
  6. New lay members should be asked about their ongoing training/ information and support needs on a regular basis. Structures/ processes should be put in place to ensure that the necessary training and support is provided or sourced externally if more appropriate.

### ***Clarity about the BPSU's expectations of PPI in BPSU studies***

7. The Executive Committee should develop clear and explicit guidance on when and why it expects to see PPI at different stages of BPSU studies. This should consider:
  - a. the evidence available as to where PPI has been shown to bring added value and where it has not made a difference
  - b. the limits of BPSU influence on researchers and whether the BPSU wants to make PPI a condition for approving studies
  - c. whether patient or public involvement is most appropriate

### ***Guidance for researchers***

8. The BPSU should produce clear and explicit guidance for researchers on when and why PPI is expected in their study and provide a definition of what they mean by PPI. This should help researchers understand 'how' to involve people and also convince them of the value of involvement in BPSU studies.

### ***Refining the application forms and review processes***

9. The section on PPI in the phase 2 application form should be refined to reflect the context of BPSU studies and the expectations of PPI (see above).
10. The review process should ask for more explicit evidence of involvement that has already taken place and clear plans for future involvement.
11. Researchers should be given clearer and more explicit feedback on their PPI proposals so they know 'how' to respond.

### ***Focus on dissemination***

12. PPI in dissemination seems to offer great potential for added value from PPI but has so far been underdeveloped. The Executive Committee should consider:
  - a. How best to encourage and support researchers to involve people at this stage

- b. Creating the infrastructure to support this work e.g. establishing a network of relevant organisations, dedicating a section of the BPSU website to disseminating lay-friendly reports of findings from completed studies

### ***Expanding PPI activity***

13. The Executive Committee should consider whether it wishes to expand its PPI activity into new areas – in particular getting more involved in influencing public health policy or involving children and young people.

### ***Promoting PPI and supporting researchers***

14. The Executive Committee should promote the PPI work it has carried out to date and facilitate sharing of the lessons learnt through conferences, websites and publications. This should help researchers to understand how PPI will be of value to them and how best to go about it.

## **8. Next steps**

A draft of this report was discussed with the Steering Group and the BPSU Executive Committee. They agreed with the recommendations and in addition recommended that the BPSU should incorporate a statement of principle in support of PPI within the terms of reference for the organisation. This will provide a public demonstration of their commitment to PPI.

The Executive Committee have agreed to establish a PPI working group who will develop an action plan to implement the recommendations from this review.

# 1. Introduction

## 1.1 Aim of this review

The aim of this review was to assess the effectiveness of the patient and public involvement (PPI) processes at the British Paediatric Surveillance Unit (BPSU) and to work with a range of stakeholders to develop recommendations for future work in this area. This included a review of how lay members have contributed to the work of the BPSU's Executive Committee as well as the processes by which the BPSU encourages PPI in its research studies.

## 1.2 Authors of this review

This review was carried out by Bec Hanley, Carolyn Morris and Kristina Staley from TwoCan Associates. TwoCan Associates carry out research and evaluation as well as providing training and support to help voluntary and statutory organisations involve people who use services in their work. Almost all of our work is undertaken in partnership with service users or carers. For further information please see [www.twocanassociates.co.uk](http://www.twocanassociates.co.uk)

## 1.3 About the BPSU

The BPSU supports research into rare childhood conditions and disorders. This research takes the form of national epidemiological surveillance – finding out how many children are affected, where and when cases have been presented and how they have been followed up. The research process follows a standard format as follows:

- Once a study is accepted by the BPSU Executive and receives ethical approval, it is included on the BPSU 'orange card'. This card lists all of the disorders that the BPSU wishes to study. Most studies are listed in the card for 13 months.
- Every month, the orange card is sent to over 3200 consultant paediatricians and other specialists.
- Clinicians mark the card to notify the BPSU of any cases they have seen or if they have nothing to report.
- Any clinician who has seen a relevant case is sent a short questionnaire by the researcher conducting one of the current studies.
- The data from the questionnaires (which may take years to collect depending on the follow-up time for a specific condition) is analysed, and in most cases written up and published

BPSU studies have been important in informing public health policy e.g. determining the content of national screening programmes. Successfully applying for and running a BPSU study is widely held in high esteem by the paediatric community.

Further information about the BPSU and its work can be found at:  
[www.rcpch.ac.uk/bpsu](http://www.rcpch.ac.uk/bpsu)

## 1.4 Structure of this report

The remainder of this report is structured as follows:

- Section 2: Methods
- Section 3: Lay membership of the Executive Committee
- Section 4: Promoting PPI in BPSU research studies
- Section 5: Expanding into new areas of PPI
- Section 6: Promoting PPI and supporting researchers
- Section 7: The challenges to taking forward PPI at the BPSU
- Section 8: Recommendations
- Section 9: Next steps

In this report, quotes from interviewees are in *italic text*. These have not been attributed to particular individuals. However, because it is often helpful to know if a comment has been made by a member of the BPSU Executive Committee, a representative of a patient organisation/ support group, a researcher or clinician, quotes have been attributed to:

- a 'lay member of the Executive Committee'
- an 'Executive Committee member' (referring to clinical and scientific members of the committee)
- a 'researcher' (lead researcher on a BPSU study), or
- a 'patient / patient representative' (a patient or representative from a patient organisation/ support group who has been actively involved in a BPSU study).

## 2. Methods

### 2.1 Overview

The first part of this review was carried out between January and April 2011 and has included:

- ⇒ Reading reports and documents from an initial internal review of PPI (led by the lay members of the Executive Committee) as well as background information on the BPSU
- ⇒ Reviewing the BPSU application processes and the requirements for PPI in studies
- ⇒ Interviewing selected members of the BPSU Executive Committee – the Chair, lay members and scientific and clinical members
- ⇒ Interviewing the lead researchers on selected BPSU studies that involved patients/ or members of the public
- ⇒ Interviewing patients/ patient representatives who were involved in BPSU studies

### 2.2 Semi-structured interviews with key stakeholders

A purposive sample of interviewees was selected to obtain the views of a range of stakeholders with direct experience of PPI within the BPSU and with diverse perspectives on the processes. Within the time and budget available, it was not feasible to aim for complete coverage.

The interview schedules were developed with input from the steering group (see section 2.5 below) and were informed by:

- The aims and objectives of the review
- Background knowledge of PPI in the BPSU and the ways in which the BPSU encourages PPI in its studies
- Background knowledge of good practice in PPI and the kinds of impacts PPI has on research<sup>1, 2, 3, 4</sup>
- Background knowledge of effective methods of assessing the quality and effectiveness of PPI

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<sup>1</sup> R. Telford, J. Boote and C. Cooper. (2004) What does it mean to involve consumers successfully in NHS research? A consensus study. *Health Expectations* vol 7, pp209-220.

<sup>2</sup> InterAct. Working Paper: Evaluating participatory, deliberative and co-operative ways of working (2001). ([www.sharedpractice.org.uk/Downloads/Interact Working Paper.pdf](http://www.sharedpractice.org.uk/Downloads/Interact_Working_Paper.pdf)) accessed 26 Jan 2011

<sup>3</sup> INVOLVE: Involving the public in NHS, public health, and social care research: Briefing Notes for Researchers (2004)

<sup>4</sup> Staley, K. (2009) *Exploring Impact*. Eastleigh: INVOLVE

- Good practice in developing interview schedules e.g. using warm-up questions, using open and non-directive questions with prompts, and allowing opportunities for interviewees to introduce relevant topics

Although the review did not require formal ethical approval, the principles of good ethical practice were still applied, for example, by addressing issues of confidentiality and ensuring interviewees were fully informed about the project before agreeing to take part. An information sheet was produced for potential interviewees, which explained the aims of the review as well as describing what interviewees could expect from their participation. Everyone who was invited to take part in the review agreed to be interviewed.

A copy of the final report was sent to all the interviewees.

### **2.3 Data analysis**

Preliminary analysis of the data was carried out alongside data collection (via regular telephone/ email communication between the review team members) to allow questions to be refined and new avenues of inquiry to develop.

The analysis of the qualitative data took the form of framework analysis<sup>5</sup>. The interviewers read all of the transcripts and listed key ideas and recurrent themes. These initial reflections were used to develop a thematic framework based on:

- previous knowledge of good practice in PPI and the kinds of impacts PPI has on research (see above)
- questions derived from the aims and objectives of the review
- issues raised by the interviewees.

The framework was used to index all the data, by annotating the transcripts. The data was then collated and arranged into key themes or topics. In addition, key observations, interpretations and summaries of the team's views were noted. This material was used to write a draft report.

Where appropriate and helpful, verbatim quotes were included to illustrate important themes. Experience has shown this is a powerful way of communicating key messages and creating ownership of the findings.

By mapping and drawing associations across themes, a series of draft recommendations were developed for the BPSU, to help improve its PPI practice and identify priorities for PPI in the future.

### **2.4 Developing recommendations**

A draft report summarising the key findings and the draft recommendations was discussed with the steering group and with the BPSU Executive Committee. This allowed BPSU members to:

- reflect on whether they agreed with the reported findings

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<sup>5</sup> Bryman, A. & Burgess, R. Eds. (1993) *Analysing qualitative data*. London: Routledge.

- reflect on whether they supported the draft recommendations
- amend or add further recommendations based on the findings

## **2.5 Management of the review process**

A Steering Group from the BPSU (see Thanks) commissioned this project and were involved at all stages of the review. One of the lay Executive Committee members, Ann Seymour, was the Chair of this group.

The Steering Group met twice during the project. They helped to shape the review, design the interview questions and select the interviewees. They also commented on an early draft of the report of the findings and helped develop priorities for the recommendations and suggestions for next steps.

### 3. Lay membership of the Executive Committee

#### 3.1 Background

The BPSU Executive Committee oversees the strategic development of the organisation, addressing issues of governance, direction and funding. Their other major role is to approve studies for inclusion on the BPSU system (see section 4.1). The BPSU does not provide funding or commission research, but ensures that studies using the BPSU methodology are scientifically robust and of high quality.

Two lay members were invited to join the BPSU Executive Committee in 2006. As is often the case with PPI work, the role of the lay members was not well-defined at first and has evolved over time with the input of the lay members:

*It's never really been specified, as I think these things often aren't. You're put on as a lay member and sometimes you have to find your role... I think it's evolved a lot because now I've got a better handle on what our role should be and I feel more confident now of contributing.* **Lay member Executive Committee**

*I don't feel as a committee we had very clear ideas about what the role of a lay rep should be, other than we recognised we needed better lay review of the studies... and to a certain extent we thought that they [the lay members] might actually help us ascertain what they could do...* **Executive Committee member**

This process of developing the lay member role has been made easier by the support offered by past and present Chairs, other members of the Executive Committee and the staff at the BPSU:

*Both Chairs have always been extremely supportive of the role. I think that's made a huge difference... One or two of the other committee members might have been sceptical at the beginning... but that's fair enough... it's a question of what difference is it going to make?* **Lay member Executive Committee**

All of the clinical and scientific Executive Committee members who were interviewed said that they had found working with the lay members a very positive experience and that they had learnt a lot about PPI:

*Obviously the two lay members we've got are very senior, experienced and confident... I have learnt about INVOLVE and some of the bigger issues and found them to be very supportive.* **Executive Committee member**

*What they've done is opened up the range of possibilities... Now when I respond to investigators I can think more about that [PPI] aspect and ask them more about it than I would have in the past.* **Executive Committee member**

The lay members are now viewed as full and equal members of the Executive Committee:

*They should be and they are full members of the Exec. That's the most important thing. We are a multi-disciplinary committee, with different backgrounds that we bring.* **Executive Committee member**

*I think we are valued. We're absolutely part of that team now, we're not a bolt on. At the beginning I felt a bit of a bolt on...I think that is quite a significant change.*

**Lay member Executive Committee**

### **3.2 The current role of the lay members and their impact on the work of the committee**

The lay members play a crucial role in the evaluation of applications sent to the BPSU. They provide a lay perspective on potential studies which is separate from and independent of the clinical and scientific viewpoints:

*In the broadest sense it's that different perspective that's really really important... You are the nearest thing to having a beneficiary of that research there... Other times we're the voice of common sense.*

**Lay member Executive Committee**

Clinical and scientific members of the Executive Committee said they found the lay members' input to be of particular value in assessing:

⇒ the likely public perceptions of a topic

*They give advice about what public responses might be to studies. They might say this is important because such and such pressure group is interested. They offer a perspective that a lot of the clinical and scientific members of the committee may not have.*

**Executive Committee member**

*They have helped us understand that we have a wide audience that we hadn't realised we had - that there are more groups out there that would be interested to hear about us.*

**Executive Committee member**

⇒ how well public information sheets have been written - as well as any other information for the public (See also section 4.3)

*There may things we assume everyone understands – but we're stuck in a scientific framework and the lay members are good at pointing out not everyone will understand what you've just said.*

**Executive Committee member**

⇒ the quality of the PPI in an application (see also section 4.3)

*The presence of the lay members helps to raise the importance of PPI in the applications – because people tend to pay lip service to it – but lay members insist they are specific about exactly what they've done.*

**Executive Committee member**

*They're there to hold us all to account. To make sure that we don't forget about public involvement in studies. And they do that very well and conscientiously.*

**Executive Committee member**

⇒ the ethical issues raised by a study

*Where there's a process that might identify individuals, although there may be a need to do that from a scientific perspective, there are issues about*

*confidentiality that the researchers lose track of...the lay members are good in contributing to that discussion.*

**Executive Committee member**

*One of the issues about BPSU studies is that it's unconsented data. I think there was some hesitancy early on...about how acceptable that would be. Having lay reps on the committee who can understand the detail of the studies is very helpful for us to understand what is and what isn't acceptable to the public.*

**Executive Committee member**

⇒ links with relevant patient organisations

*They can give advice to the researchers about where they might go for PPI. Because you get the impression sometimes – that the applicant has done rather a superficial search to find a support group for a very rare condition and haven't thought outside the box. They [lay members] can signpost researchers to other sources of good PPI.*

**Executive Committee member**

The lay members also help with advising the researchers about the PPI in their studies:

*They're very good about being contacted directly or speaking directly with the investigators, which is helpful to me because I often have to write the letters back and give guidance. It would be difficult for me to give good, detailed guidance about [PPI].*

**Executive Committee member**

Although the involvement of lay members can be challenging and adds time to the approval process, Executive Committee members recognise this is necessary and appropriate:

*Sometimes they make life a bit uncomfortable for us which is fine.*

**Executive Committee member**

*It adds another dimension– and it means we have to make sure we do an appraisal of projects in a structured way – but that obviously improves the quality. We could maybe rattle through it quicker [without involvement] but I'm not sure that that would be better.*

**Executive Committee member**

It is also recognised that the BPSU needs to 'practice what it preaches' and model best practice to outside researchers:

*Good standards [of PPI] within the BPSU enable us to raise standards on the outside.*

**Executive Committee member**

### **3.3 Suggestions for improvements**

All of the interviewees were happy with the role lay members are now playing on the committee:

*The way in which it is working is the best way.* **Executive Committee member**

No one had any suggestions for changes or improvements, except perhaps for greater recognition and acknowledgment of the lay members' work:

*I'm very impressed with how well they grasp some of the obscure conditions – so they've done a lot of work – we may not appreciate how much work they have to do before the meetings.*

**Executive Committee member**

### **3.4 Future work – recruiting new members**

The current lay members will soon reach the end of their term and therefore all interviewees were asked about their views on recruiting new members, both in terms of the attributes of new recruits and the process of recruitment. The issues raised included:

- ⇒ broadening the lay membership
- ⇒ running a formal recruitment process
- ⇒ providing training and support to new members
- ⇒ paying lay members
- ⇒ ensuring a successful handover

These will now be discussed in turn.

#### **(i) Broadening the lay membership**

All of the interviewees recognised the importance of recruiting at least two lay members so that they can work together and support each other as a team. However, there was also a suggestion to increase the number of lay members and to recruit a mix of people of different ages, gender and socio-economic backgrounds. This was thought to be important to ensure a greater diversity of opinion amongst lay members:

*Maybe having two from a pool of three or four might be better. It's important to always have two there, but it's also important to have a diversity of views...In these positions people's own personal views and experience are quite important - the input of the lay member is influenced by their personal values and experiences and is a bit more subjective.*

**Executive Committee member**

This is a common issue in PPI and may need to be addressed through the recruitment process and perhaps offering payment to lay members (see (d) below) to make it possible for different groups of people to be involved:

*There's a lot of criticism of not getting people in from more diverse backgrounds. This is the same with a lot of committees. You sometimes get criticised as 'a lot of middle class women' – we have husbands who support us, we have time to go to these things. Of course we are able to do it.*

**Lay member Executive Committee**

#### **(ii) Running a formal recruitment process**

There was broad agreement that new members should be recruited via an open and more formal recruitment process: developing a role description, advertising widely, shortlisting and interviewing potential candidates. This was thought to be important for the recruitment of high calibre individuals and to ensure people could be sure the role was right for them:

*Something that lets both sides know whether this is a good match.*

**Executive Committee member**

It was suggested that a job description be developed around the current role of the lay members (see Section 3.2). The lay members thought it important that some aspects were highlighted and made very clear, particularly the demands on people's time, probable workloads and issues around confidentiality:

*[Sometimes you're asked to] "get something back by Friday". That might not be possible, if this role is just a tiny bit of your working life... If you really want lay representatives... to be able to respond that quickly... you should tell them, be upfront about it.*

**Lay Executive Committee member**

*The only negative is it takes up a lot of time - I don't think anyone asks in advance if you're prepared to do it.*

**Lay Executive Committee member**

The essential qualities of for new lay members were thought to include:

- ⇒ an understanding of research and PPI
- ⇒ an ability to represent broad interests – not focused on narrow agendas
- ⇒ experience of working with professionals
- ⇒ able to offer genuinely lay perspectives i.e. not anyone with a professional role in the NHS nor academics
- ⇒ being good at communication e.g. writing about research for the public, and communicating well with other committee members

### **(iii) Providing training and support to new members**

Some interviewees expected new members to be recruited with previous experience of being involved in scientific or academic groups. They therefore assumed that new members would not need basic training in research.

Others thought that new members might not have this background knowledge, and should therefore be offered some training in the basics of research, particularly ethics and confidentiality issues, methodology and funding processes. Some also thought that any training should reinforce the value and importance of their lay perspective and should avoid 'turning them into researchers':

*One of the general difficulties is that lay members become professionalised – that's fine in some cases – but there's a downside in that they move away from being lay.*

**Executive Committee member**

It was also thought that new members would benefit from an introduction to the BPSU methodology through written guidance or training:

*A handbook of terminology would have been useful - something about what epidemiological research is, a little bit about the workings of it... something that lies between doing a professional course and the very brief public information leaflet.*

**Lay Executive Committee member**

*Because it's such a technical endeavour and all the studies follow more or less the same approach – once you know about that, life's a lot easier and maybe there needs to be some kind of formal induction that the BPSU could set up.*

**Executive Committee member**

As part of an induction, some interviewees also suggested that new members should meet with key people (e.g. the Chair, BPSU staff and the current lay members) before their first committee meeting. The existing lay members thought an introduction to the academic culture would also be important:

*Part of the induction should be about the different approaches of different professionals, what you'll get and what to expect. It's a shock because we're not academics, we don't come from that background, you have none of that preparedness.*

**Lay Executive Committee member**

In terms of providing ongoing support, there were suggestions of offering new lay members:

⇒ support from mentors

*I would like to see one specific committee member named as a mentor for six months to a year. Just to be on the end of that phone, to stay behind for 10 minutes after a meeting, saying how did you feel that went? It's not an onerous task.*

**Lay Executive Committee member**

⇒ briefings before committee meetings

*Sometimes we have to explain the medical issues or dilemmas or treatment problems - I sometimes wonder whether we should do more of that – could we perhaps give them better briefing – we should certainly encourage them to say when they don't understand something.*

**Executive Committee member**

⇒ links to lay members in other surveillance units

*In a way the BPSU is leading the way, but there could be cross-representation or communication at the least with lay members of other units... Cross fertilisation of ideas as well.*

**Executive Committee member**

⇒ access to further sources of information e.g. journal articles via membership of a medical library

All the interviewees were conscious of not making assumptions about the training and support lay members might need and the importance of asking them what they would like help with, before developing any form of training or support.

#### **(iv) Paying lay members**

From the perspective of the existing lay members, payment for their time would demonstrate that their input was being valued and has become more of an issue as greater demands have been placed on them:

*It's perhaps a matter of valuing your time. Clinicians give a lot of free time for such roles but of course it goes on their CV's: there are benefits to them... Sometimes you wonder if you're being contaminated by being paid. Which you shouldn't really be... quite honestly for me it's fine, but then I sometimes think when I put a lot of work into it... perhaps I should get something.*

**Lay Executive Committee member**

*It's not just what I'm prepared to do but how much I can easily do... I think that's a really important issue to be addressed, because as you become more involved and hopefully PPI becomes embedded in the whole process of what the BPSU does, the burden on the lay reps tends to be greater.*

**Lay Executive Committee member**

This issue was also recognised by some of the other members of the Executive Committee:

*Maybe one of the problems is they are unpaid. It's quite problematic because you come to a point at which the commitment you're putting into the committee is quite significant. That's the thing that hampers, or that now is beginning to limit further involvement.*

**Executive Committee member**

Some interviewees suggested that payment for lay members' time be included in any future bids for BPSU funding.

In terms of paying lay members' expenses, the process is working well - people are being reimbursed quite promptly. However, this issue may need to be revisited with new members, in case their travel costs are greater and/or they cannot afford to be out of pocket for any length of time.

#### **(v) Ensuring a successful handover**

A number of the interviewees suggested that new members should be able to shadow existing members for one or two meetings, before the old members leave:

*I would like to see one or two meetings where you've got one or other of us there. A continuation. You can't expect someone to come and sit on a committee and be able to contribute fully immediately... you've got not only to understand the work but also understand the dynamics... the first couple of meetings are more about observation than contribution. It's looking at the dynamics, what's going on here?*

**Lay Executive Committee member**

*It takes quite a long time to understand the BPSU methodology. Now it seems obvious to me, but it is quite complex.*

**Lay Executive Committee member**

### **3.5 Conclusions and recommendations**

Lay members have been pioneers in developing completely new areas of PPI work at the BPSU. Although the process of their recruitment and induction may not have followed best practice, they have been welcomed and well supported by the committee and BPSU staff. They have developed and strengthened their role and have also helped to develop mechanisms to promote PPI in BPSU studies (see Section 4). Having successfully broken new ground for PPI, they have opened the way for new lay members to take their place and continue the good work.

More could be done to acknowledge their achievements e.g. through including information about them and the work they do on the BPSU website, in newsletters and the annual report. Raising their profile would also help to encourage others to consider involvement.

The key issue for the future is the recruitment and induction of new lay members. It will be important for the Executive committee to decide:

- how many new lay members it requires and why
- the essential/ desirable and skills and experience they will require of new members
- whether it wishes to pay lay members in future and how funding might be obtained for this purpose
- how it will recruit members through a more formal process

If the committee does decide to develop the current model of lay membership (e.g. by expanding the number of lay members, paying people), it would be helpful to talk to other organisations about different models and examples of good practice, to help shape future plans.

It is important that the whole committee is involved in these decisions and helps to design the recruitment process and any job descriptions for lay members or terms of reference for the committee. This is important to ensure ownership of the process and a shared understanding of the role and purpose of lay membership.

Recruitment of new members needs to be planned and timed carefully so as to allow a handover from existing lay members and for new members to receive an induction before they start. The induction needs to include a clear explanation of the BPSU methodology and workings of the committee. Any new scientific or clinical member of the committee might also benefit from an induction and importantly this should include an explanation of the role of lay members.

New lay members should be asked about their ongoing training/ information and support needs on a regular basis, as these needs may change over time or may only become apparent after some time in post. Structures/ processes should be put in place to ensure that the necessary training and support is provided or sourced externally if more appropriate.

## 4. Promoting PPI in BPSU studies

### 4.1 Background

The BPSU promotes PPI in its studies by making PPI a requirement for researchers wishing to use their approach. Researchers are required to go through a two stage application process. The first stage asks for an outline proposal which is reviewed by the Executive Committee. If the researcher is successful at this stage, he or she is then asked to complete a full application describing their proposal in more detail. This full application is also assessed by the Executive Committee and researchers are invited to attend an interview with the Executive Committee. The committee gives feedback to applicants outlining any necessary changes or suggestions for improvements, before giving their final approval for the study to go ahead. The application process therefore often involves committee members doing a considerable amount of work to develop the proposal, with the end result often looking quite different from the original application.

The lay members of the Executive Committee have had a major influence on the application process by developing the forms to include questions about PPI and making evaluation of PPI an integral part of the review process. Changes that have been made to the application forms include:

- The application form at Stage 1 asks a simple question about PPI with the aim of encouraging researchers to think about the possibilities. PPI is not required at this stage, so that patient groups are not contacted about studies that may not go ahead.
- In Stage 2, applicants are asked to provide a lay summary of their proposal. This has been essential to enable the lay members to effectively evaluate the proposed research.
- The Stage 2 application form also asks for details about how patients/ the public have been involved in the development of the application and any plans for further involvement in the study. This section includes a tick-box grid that asks researchers to indicate the level of involvement (consultation, collaboration or user-led) and the stage of involvement. There is also an open question which asks for a full description of the involvement.
- At the same time as submitting their stage 2 application, researchers are asked whether they intend to produce a public information leaflet (PIL). This leaflet describes the plans for the study in lay-friendly terms and is used to publicise the research.

As part of the Executive Committee's review process, lay members (and other committee members) assess:

- the level of PPI in the development of the application
- proposals for further PPI in the proposed study
- the quality of the PIL (if written at this stage)
- whether links have been made with appropriate patient/ support groups

As described in Section 3.2, this involvement of lay members has helped to improve the quality of PPI in BPSU studies and improved the accessibility of the PILs.

## 4.2 The importance of PPI in BPSU studies

All of the interviewees thought it important to involve patients and the public in BPSU studies because of the nature of this type of research. BPSU studies use patient data but do not have any patient participants. Questionnaires are sent only to clinicians, mostly paediatricians. Therefore no one is asked to give consent for the use of the information in their medical records. The data is anonymised and may be used without the patients' knowledge. This makes it all the more important to ensure openness, transparency and accountability to the public:

*It's a serious thing to do research without a patient's consent – so there has to be a good reason to do it. Asking patients whether they think that's acceptable – that's really important... Ask them if it's important to people with their condition.*

**Researcher**

*The research needs to be transparent – for scrutiny – to see exactly what's going on in these studies. It's about research governance. It has to be clear there is no covert aspect to what we are doing.*

**Executive Committee member**

The fact that BPSU studies involve children with rare conditions also raises ethical concerns that individuals may be identified even when the data is anonymised. PPI therefore also enables researchers to check out the acceptability of their approach with patients and the public:

*I think PPI is really important for these studies because they're done in a non-consensual way. So it's collecting albeit anonymous data but on sometimes very rare conditions where there might only be 50 in the whole country, sometimes less. Therefore I really think it's important that it's not just researchers that have an input to that process, for all sorts of ethical reasons.*

**Lay Executive Committee member**

Involving patients/ lay members is also critical to identifying studies that are controversial or of a sensitive nature that will need to be communicated carefully, particularly if they are likely to be picked up by the media:

*PPI makes them [the studies] possibly more acceptable, if they're fairly controversial things. Sometimes you can get so bogged down as a researcher being academically and clinically accurate, you don't realise what the impact might be on a wider audience. Those kinds of things get picked up on.*

**Lay Executive Committee member**

*You've always got to think how it will sound in the News of the World when you're publishing this study. I always say "News of the World, Daily Mail – how are they going to pick up on this, is that the message you want?" and then it becomes totally obvious.*

**Lay Executive Committee member**

Families affected by rare diseases also want to know about research on their condition. Therefore PILs are important for publicising BPSU studies. PPI in the development of PILS ensures they are relevant and accessible:

*There definitely needs to be more information given out which is easily accessible, to let them [parents] know this study is going on – so they can look it up and find out the results on completion of the project. That will be important to them as it relates to their children.*

**Patient/ patient rep**

### **4.3 The value of PPI at different stages of a BPSU study**

Since all BPSU studies follow much the same format it is possible to map PPI across the different stages and consider the value at each stage. The stages include:

- Identifying and prioritising a research question
- Developing the design of the study
- Writing the BPSU application
- Writing the PIL
- Conducting the study
- Writing up and disseminating the findings

These stages will now be discussed in turn.

#### **(i) Identifying and prioritising a research question**

It is valuable to involve patients at this very early stage because they are the only people who can confirm that a study is relevant and important to people affected by the condition or topic under investigation:

*It's important...because we want good quality studies that will make a difference to treatment or understanding of rare conditions.*

**Lay Executive Committee member**

*There is very little point in doing research unless you're going to help the actual users – I think children and families can tell us whether they think the research is important or will make a difference in their lives or potentially could help other people if not themselves directly – because they are in the position to tell us about these things.*

**Researcher**

PPI at this stage might also help researchers to realise that a study is of little public interest and so abandon unsuitable projects:

*It can help investigators realise that they're onto a loser or a winner – with some applications you can see that if they had actually genuinely engaged with the public and thought carefully about it, they'd have realised there's not a lot of public interest in it. But researchers are fairly myopic sometimes.*

**Executive Committee member**

*I think the whole process of having to engage with that does make you think – sometimes it's not what they say but the fact you've got to think about it beforehand.*

**Executive Committee member**

PPI can also help to identify important research questions that could be addressed using the BPSU method. One researcher described how they had worked with a

national patient organisation to run a study day with a wide range of stakeholders to help define research questions and identify the best approach:

*There were invited experts. There were families with [affected] children... One of the main things that came out was that research studies were needed to find out how common these conditions are, how they present at the early stages and what are the predictors... The main point from the parents was that there's a severe lack of awareness of this condition amongst clinicians. We discussed the BPSU approach and thought this would be a very good way of raising awareness because we would ask all paediatricians if they've seen a case and send information leaflets... We need everyone to know about this – rather than just do a study in the few specialist centres where these patients are being treated.*

**Researcher**

This early involvement therefore helped to identify the BPSU methodology as the most appropriate from the parents' perspective. As a consequence, the patient organisation also identified the topic as a priority within their research strategy, which eventually led to them funding this work.

## **(ii) Developing the design of the study**

Most of the interviewees concluded that because BPSU studies use a standard approach that relies solely on retrospective data collection from paediatricians' notes, there is very limited scope for PPI to have an influence on the study design (unlike other kinds of research studies):

*The questions that users come up with are not usually answerable by this kind of surveillance. They are usually much more specific about particular treatments... Often the sorts of questions the users want answering have to be answered by a different research design.*

**Executive Committee member**

*From our point of view we would like some of the social impact of these rare diseases to be included, but you're very restricted by what is in the case notes and this is what we have to understand all the time.*

**Lay Executive Committee member**

*It's about horses for courses. Some studies suit the context of PPI better.*

**Executive Committee member**

Some interviewees suggested that patients *might* be able to contribute to the design of the questionnaire for clinicians:

*There may be issues in the design where lay members could have input for example in studies of mental health. As clinicians we focus on the biomedical aspects – but lay members can consider psychological or other aspects.*

**Executive Committee member**

*Maybe we could ensure that everything was covered – if we could see an overview of what was sent – that would help. But I don't know if I could have suggested anything – because they know what they need to ask and what information they need...*

**Patient/ patient rep**

However no evidence was found of this type of involvement having had an impact. Patient / patient reps are not often asked to comment on questionnaires and where they have been consulted, they have not made any changes. It seems that the potential for influence at this stage is largely theoretical:

*There maybe aspects that questionnaires overlook – maybe that we’ve not seen because we’ve focused on the biomedical model – but I’ve not seen that happen in the last three years.*

**Executive Committee member**

*I have to be honest and say that I don’t think the PPI aspect contributed to the design at all.*

**Researcher**

*Unfortunately the things that they were interested in looking at weren’t methodologically possible to look at...it wasn’t possible from a scientific point of view to incorporate their comments and priorities. Had it been – then we would have.*

**Researcher**

Although lay members on the Executive Committee have commented on questionnaire design, these comments have been about issues of accessibility and ambiguity, which are not specific to a lay input:

*You can’t ask questions if... it’s a subjective judgement. We pull them up on that sometimes. You’re asking someone about non-accidental injury “did you think this could be NAI?” Think? You can’t collect data that’s going to go in an epidemiological study that says “think”. It has to be established .We pick up on things like that.*

**Lay Executive Committee member**

This lack of evidence of PPI at this stage has led to some people to question the merits of asking researchers to go through a process of consultation on study design:

*Actually, you think people are ticking boxes and jumping through hoops here rather than meaningfully engaging with the processes...It’s not that I don’t believe in the idea...I just don’t think it relates very well to the BPSU methodology.*

**Researcher**

### **(iii) Writing the BPSU application**

None of the interviewees were aware of PPI influencing the development of a BPSU application or were certain of its value:

*Sometimes it’s helpful to be able to ask specific questions of lay reps – it’s more obvious when you’re writing a grant application...but putting the BPSU application together is fairly formulaic – it sometimes hard to know what PPI can offer at that stage.*

**Researcher**

One researcher commented that involving patients can help with getting ethical approval for a BPSU study:

*If you can show the patients think this is the best design to do the study – then that helps with ethics review – as they will ask why are you doing it without consent.*

**Researcher**

#### (iv) Writing the PIL

PPI has been of value in the development of PILs in a number of ways including:

⇒ Contributing additional content

*It seemed to me an opportunity to contribute information and understanding about the area being studied which might otherwise not have been included. We were familiar with the statistics and case studies and it seemed helpful to contribute that – I wanted to make sure that issues were covered appropriately and that they had explained the issues clearly and correctly.* **Patient/ patient rep**

⇒ Making them more accessible

*The first initial leaflet that I received had lots of jargon – it's easy to forget as a researcher that lots of people don't have a scientific background so don't understand the words you're using – even the diagrams were very technical and scientific – so I tried to make them a bit easier to understand and follow.*

**Patient/ patient rep**

⇒ Making them more relevant to people affected by the condition / topic

*The main involvement was about what should or shouldn't be in the information leaflet. There were important issues about how much information to put in about [the condition] – because not all children with [initial symptoms] go on to develop the [condition]...*

**Researcher**

The PILs are thought to be a valuable way to let as many affected children/ parents as possible know that the research is taking place. Many commented on the need to ensure the public can access the PILs through more effective dissemination e.g. via the websites and newsletters of relevant organisations, project websites and the BPSU website:

*So anybody might say, my child has X, and you google it - we hope eventually that maybe the BPSU will come up.* **Lay Executive Committee member**

However many interviewees raised concerns about whether and how PILs were reaching the target audience and how often they are accessed, particularly when the number of affected families is low:

*It's quite difficult to imagine how a leaflet could be meaningful.* **Researcher**

#### (v) Conducting the study

There were no examples of involvement of patient or patient representatives in conducting a BPSU study. Some of the interviewees thought PPI could be of value in analysing and interpreting the data, though again this remains hypothetical. There was no evidence of involvement or its impact at this stage:

*When the results are in, then they ought to go back to their PPI contacts and share the results with them and get them to help interpret the results – as there may well be issues for example around delay in presentations – which a parent perspective of why that might be happening would be useful – because the doctor may have a particular view and the parent a different view.*

**Executive Committee member**

*Certain things might have been discussed with the paediatrician, but the parents might have relevant information or input that might be important and might completely change the look of the results – so there might be another angle on that...It could just be a minor thing that hasn't been mentioned to the paediatrician – but it's something that could be really important.*

**Patient/ patient rep**

#### **(vi) Writing up and disseminating the findings**

In the same way that PPI helps with the production of PILs, PPI could help with producing lay-friendly reports of the findings of BPSU studies. PPI could ensure these reports were accessible and relevant to a lay-audience. As yet there is no evidence of this kind of involvement, mostly because few studies have been completed since PPI at the BPSU began.

Patient organisations could also help with the dissemination of findings to the target audience (i.e. people affected by the condition/ topic) via newsletters, websites and conferences. The researchers and patients/ patient reps who were interviewed were keen to see this happen:

*We would like to see the findings, we'd obviously put it on our website, publish it in our magazine and send the relevant information out to families.*

**Patient/ patient rep**

*The final stages will involve giving feedback of the results to people affected by [the condition] – that's one of the important stages for involvement – so that you let people know what has actually come out of a project after you have told people that it's happening...I would like to help put together the information, to give an overview of the results...that is a place where maybe I could have an input...*

**Patient/ patient rep**

*Dissemination is very important...having the information more widely disseminated is something that the lay rep should encourage. Having it easily accessible in lay terms would be worthwhile – so that information is more widely read – not just by a select group of academics.*

**Researcher**

#### **4.4 Suggestions for improvements / future work**

The suggestions for improvements/ future work fell into the following categories:

- Clarity about the Executive Committee's expectations of PPI in BPSU studies
- Guidance for researchers
- Refining the application forms
- Refining the review process
- A focus on dissemination
- Creating links with relevant organisations

These will now be discussed in turn.

**(i) Clarity about the BPSU's expectations of PPI in BPSU studies**

Many of the interviewees thought that the Executive Committee needs to develop a clear and more explicit view of where they expect to see PPI in BPSU studies and also explain the rationale for this involvement. This needs to be based on the evidence of where PPI has already made an impact on BPSU studies, rather than on theoretical possibilities. This is important for researchers and involved patients/ patient representatives, who want to ensure that their efforts genuinely bring added value:

*In an ideal world we would want more input from patient organisations in the whole application process, study design etc – but unless you can see real benefits in that – will that just be a detrimental factor and slow the whole process down? We need to further explore what the pros and cons of involvement are.*

**Executive Committee member**

*PPI needs to be seen as an opportunity and not as a barrier. To me it felt like yet another thing they wanted me to do.*

**Researcher**

*Make it clear where they expect to see patient involvement - this makes it clear both to the researcher and the person that's actually involved, so they can see a clear job and the stages they need to be involved in – then they can prepare for that and work together.*

**Patient/ patient rep**

*A researcher could quite legitimately say 'why am I having to do this?' - And we've got to have the answers ready.*

**Lay Executive Committee member**

Some of the issues that will need to be addressed include:

⇒ ***Being clear about the limits of the Executive Committee's influence on PPI in BPSU studies***

There are some stages in the research process where it is easier for the BPSU to have an influence on the PPI than at other stages. For example, some of the interviewees commented on the fact that although early involvement in identifying a research question and prioritising topics is of great value, there is little that the BPSU can do to promote this, because:

- ⇒ This involvement needs to happen *before* developing the BPSU application
- ⇒ The BPSU does not commission or fund research studies and therefore cannot influence what topics researchers decide to address
- ⇒ Unlike funding organisations, the Executive Committee does not prioritise different study applications – so there is no process to promote studies addressing patients' priorities over and above any other study

The Executive Committee does make decisions about how much time to invest in helping to develop a proposal, which may be influenced by whether it is a high priority:

*Sometimes if a study is of bad quality, the committee will take extra effort to get it into shape - if it is an important priority ...*

**Researcher**

To have a genuine influence at this stage, the BPSU would need to insist on this early involvement, making it an essential requirement for all applications:

*It would be nice to think you've got ideas coming in from the public, or families to say "I would be very interested if you could find an investigator interested in having a study done [on this rare disease] because there seems to be not much known about treatment..." Got to think how you're going to get that contact in.*

**Lay Executive Committee member**

*Input into the study design really should have happened before the application was made. The applicant should have consulted widely about the sort of questions families with the conditions want answered. We need to insist on involvement before the applications comes in.*

**Executive Committee member**

Similar issues were raised in relation to promoting PPI in dissemination (see (v) below).

⇒ ***Being clear about when 'patient' or 'public' involvement is preferable***

Depending on the topic under investigation and the stage of involvement in a BPSU study it is sometimes preferable to involve a person with direct knowledge of a condition/ topic (either as a parent, or as a child/young person living with a condition) – a 'patient' – and sometimes preferable to involve a 'general member of the public' who can offer a more common sense view and a perspective on the 'public interest'. This may need to be considered for each study.

It is important that these issues are clarified so that researchers know who they need to engage and why, involved patients/ patient reps know what is expected of them and lay members of the Executive Committee are clear about the role they play. The role of lay Executive Committee members has sometimes seemed to be a bit blurred in the past, when they have made comments in the absence of patient/ patient rep involvement or in the absence of relevant patient organisations / support groups:

*We can and do sometimes comment on some of the questions that might be asked or some of the areas they might cover...in a general sense, but remember these are rare conditions so we don't know. We feel that would be very much the role of an individual who is much more au fait with what that particular study is asking. So we really strongly recommend that [additional involvement].*

**Lay Executive Committee member**

*It's a bit much to expect the lay reps to be familiar with all these different rare conditions – but they often make some quite useful / more generic comments...*

**Executive Committee member**

One area where there are two types of involvement is in writing the PIL. There may be benefits to having a 'two pronged attack'. Patients will know what language is familiar to other people with their condition, but can also sometimes slip into jargon because of their familiarity with the topic. A 'member of the public' can ensure that the PIL remains accessible to a more general audience. However, this also creates

potential for disagreements. It may be helpful to consider who the target audience is for the PIL – patients or the public – and ensure involvement of the most appropriate group.

As PPI becomes more embedded in BPSU studies, there is also a question as to whether the Executive Committee lay members are there to do the job that an involved patient/ patient rep might do, or whether they are there to check the quality of the PPI and help to promote greater involvement. This issue will need clarification for new lay members and may need to change over time. This raises the question of what 'lay' means to the Executive Committee and whether they are looking for a public or a patient input into the work of the committee, or both.

## **(ii) Guidance for researchers**

With greater clarity about the BPSU's expectations of PPI, it should be possible to produce clear and more explicit guidance for researchers on when and why PPI is expected in their study:

*Fix milestones where people need to be involved.* **Patient/ patient rep**

*We need information about how you can involve users, why involve users, what stages, what they can add – how they can help – a document that clarifies that a bit more.* **Researcher**

*And perhaps also to be clear when PPI is not absolutely necessary...* **Researcher**

*If the BPSU knew lots about it and were also sufficiently confident to say, look you're never going get anybody to help you there in study questionnaire development... then everyone could be open and upfront about that. It would make it less of a barrier to investigators.* **Researcher**

It was also suggested that this guidance could include case studies and examples of good practice to encourage researchers and 'sell' PPI. It could also be promoted on the BPSU website:

*Have a tab that says user involvement – if that was on their website – it would make it very clear that user involvement was a cornerstone of the BPSU and is considered important.* **Researcher**

## **(iii) Refining the application forms**

Some interviewees suggested changing the phase 2 application form because the tick-box grid is difficult for researchers to understand and doesn't clearly relate to the ways in which people are involved in BPSU studies:

*It's quite difficult to know the differences [between the categories on the grid]. I think I put down consultation but I wasn't very sure. The process would be better if they were more explicit about what they're expecting of you.* **Researcher**

*[The difficulty in involving people in the design stage of BPSU studies] raises the question what would 'collaboration' involve? Nor could I see how a study could be user-led ...not sure I'd expect it, very difficult, with confidentiality.* **Researcher**

*It would be much more useful to talk about stages of the BPSU process – asking more explicit questions about how people have been involved in the application and plans for future involvement...*

**Researcher**

Greater clarity about the Executive Committee's requirements for PPI combined with these changes to the form were also expected to make the process of judging the quality of PPI in proposals a lot easier and more standardised.

There were no suggestions for changing the phase 1 form as the question about PPI is thought to fulfil its purpose in encouraging researchers to start thinking about PPI.

#### **(iv) Refining the review process**

It was also suggested that the application form and review process could ask researchers for more robust evidence of PPI:

*We haven't done any audit or proof of what they've done. If we asked them 'what difference did it [PPI] actually make?', I suspect if you challenged these applicants it might all be a bit fragile – they know it's expected so they just tick the box... We should raise our expectations and ask people to be a lot more specific and measurable about what they say. It's raising standards of communication and engagement – we can set the bar as high or low as we like – and I think the bar has got to go up.*

**Executive Committee member**

Some thought that feedback given to researchers about the PPI in their proposals should also be more specific:

*The feedback to investigators when going through the proposal stage – perhaps that could be a bit more explicit about what people should do – 'Contact this group and ask them this'.*

**Executive Committee member**

One researcher suggested inviting involved patients/ patient reps to the phase 2 interview, to provide more background on the topic and explain why the study is thought to be important from the patient perspective.

#### **(v) A focus on dissemination**

Dissemination of the findings of BPSU studies is one of the key stages where all of the interviewees believed there to be an important role for PPI. Involvement is useful at this stage to help write up the findings in a way that's relevant, useful and accessible to a lay audience and to find ways to reach patients and the public. However, many also thought that the BPSU could do more to promote PPI at this stage at the same time as recognising the limitations of their influence:

*The researchers own the data and have to decide what to do with it – all the BPSU can do is to say a bit more overtly what are our expectations are of good practice... When we approve a study we should ask for a dissemination strategy. If it's a good study, they'll have a clear research question and will know what they will do with the results... we could say that as part of the deal – we'd want to see a copy of what they've done.*

**Executive Committee member**

If the BPSU wanted to ensure this dissemination happened, they might need to do more of the work themselves:

*We could help them [researchers] turn a scientific paper into an article for a lay audience – our lay reps could do that.* **Executive Committee member**

Some researchers may also need help with PPI at this stage to understand how and why to do it. They also need to understand the importance of maintaining relationships with patients/ patient reps throughout a study, so that people are able and willing to contribute to dissemination at the end:

*Quite how a group who are supportive of the study, at least they were 18 months ago, quite how they can influence that [dissemination] I don't know. Again I struggle to find a meaningful way of doing it.* **Researcher**

The BPSU already strongly encourages researchers to publish their results and would look less favourably on an application from a researcher who had failed to publish previous work. The same principles might also be applied to dissemination of the findings to patients and the public i.e. if a researcher failed to do this, any future applications could be rejected until the work was completed.

#### **(vi) Links with patient organisations**

The BPSU has done a lot to develop links with patient organisations, as one of the patient/ patient reps commented:

*BPSU do an awful lot of work in this area. What they do, they do well.* **Patient/ patient rep**

However there were some suggestions as to how these links could be improved:

- Ensure early contact with relevant organisations to build the relationships necessary to support a study

*Ensure good communication with all agencies with an interest in the study as part of the scoping for running the study...* **Patient/ patient rep**

*For me a lot of it's about dissemination at the end. You have to think about that early.* **Executive Committee member**

- Provide feedback to organisations about how their involvement has made a difference

*When they come to the end of the study it's nice to get feedback of the family involvement...and any recommendations that families have made that have been taken forward. Because it's always nice for families to feel that they've been listened to... and to pass that back to the families through our website and magazine is always very useful.* **Patient/ patient rep**

- Publicly recognise the contributions made by patient organisations – including on the BPSU website

*Put the information out to the general public saying this study's gone on, they'd like to thank the families and the following recommendations that have been made by families have been taken on board and listened to. That's all it needs.* **Patient/ patient rep**

- Develop a network of organisations who have helped with studies or would be willing to help

*There are a number of groups now involved in studies with experience of what we do – maybe BPSU could be reaching out more to them and looking whether we can build wider group/ network of those who might contribute in future.*

**Executive Committee member**

#### **4.5 Conclusions and recommendations**

The Executive Committee and particularly the lay members have done great deal of work to promote PPI in BPSU studies, developing the mechanisms to assess the quality of proposals for PPI and providing advice on who to involve. Much valuable experience and evidence has thus been accumulated which could now be used to refine these processes. The lay members have played a vital role in breaking new ground for PPI and exploring the potential for involvement. The lessons learnt can now be used to pinpoint exactly where and when PPI brings the most added value.

To take this work forward, the Executive Committee needs to define and agree exactly where and why it expects to see PPI in BPSU studies and to develop clear guidance on this for researchers. The application forms and review process also need to be refined to reflect these more explicit expectations of PPI. They also need to be tailored to the specific context of epidemiological surveillance studies.

Given the standard format of BPSU studies and the evidence of impact gathered to date, it seems that the most valuable areas for involving patient/ patient representatives in BPSU studies have so far been:

- identifying the need for a research study and confirming its relevance, importance and acceptability to people affected by the condition or topic being studied
- helping to write and disseminate the PIL

There also seems to be strong potential for their involvement in disseminating the findings to a lay audience, although this has yet to be achieved.

There may also be potential for their involvement to add value at the stages of developing the study design/ questionnaire and analysing data, but it would seem advisable to test this out through piloting or by finding evidence of where it has worked in previous BPSU studies, before making this an essential requirement for researchers.

## 5. Expanding into new areas of PPI

A number of suggestions for future work around PPI for the BPSU involved expanding current activity. These included:

- Getting more involved in influencing public health policy
- Involving children and young people

These will now be discussed in turn.

### (i) Getting more involved in influencing public health policy

A number of the interviewees thought that the BPSU could do more to ensure that the findings from BPSU studies are used to influence public health policy:

*One of the things that the BPSU doesn't do very well is take things forward after the study has been done... one of the criteria for accepting a study is that it has some kind of policy implication – publishing the results is only an interim step, the next step is to get something done about it. There are other surveillance units across the world that do get very much more actively involved in that.*

**Executive Committee member**

*Because of the national nature of the studies it can have very important consequences on policy – much beyond other studies.*

**Executive Committee member**

They also thought that if the BPSU were to develop activity in this area, then PPI would be very important. This was for a number of reasons:

- Policymakers are keen to know about public involvement and can be influenced by the human story

*Being able to say to policymakers we have these connections and collaborations is quite important. It does flag up we're not working in isolation... We've actually consulted, we know these are the important issues [for patients/ the public]*

**Executive Committee member**

*We worked with a support group to develop a vignette for a report for the CMO that really flagged up the issues... It definitely had an impact. A lovely description put in a real sense of what it means: it would be difficult to get in any other way.*

**Executive Committee member**

- Patient organisations/ support groups often make helpful collaborators in influencing policy and practice – the BPSU may not then always have to lead on this but simply lend the weight of its evidence. Many patient organisations also have a lot of skills and experience in this area, which would be helpful to the BPSU.
- It is also important to influence public opinion via the media – as this will in turn influence the policymakers. PPI can help with developing the media story and again adding the human dimension:

*A lot of people I worked with wanted to tell me their stories. This was helpful as background at the beginning and may be helpful again later if we're thinking of going to the press to disseminate our findings.* **Researcher**

## **(ii) Involving young people**

Several of the interviewees recognised the importance of the *principle* of involving children and young people because their views are often distinct to those of adults:

*The work is really about them. The problem with adults is that we're all adults. A young person's perspective is very often completely different. It's useful sometimes to be reminded of that.* **Executive Committee member**

However, no one was certain of where children and young people could *practically* be involved in a way that would bring added value. It was agreed that it would be inappropriate to try to engage children and young people in the review process (i.e. as lay members of the Executive Committee). Careful thought would also need to be given as to where children and young people could make a difference given the constraints of BPSU studies (see section 4.2).

*I think we've got to be very, very careful that it's not just superficial the way we involve children and young people. There's got to be a real reason that is relevant to them about this work.* **Executive Committee member**

Specific suggestions for their involvement included:

- Asking children and young people a general question about the acceptability of the BPSU approach – although recognising that anonymisation and data protection are complex concepts that young people may not understand nor be interested in

*Perhaps understanding what kind of views children are putting forward about the work generally, would be useful for us to know more about and to take on board.* **Executive Committee member**

*It's an ethical thing. It should be there and it is missing.*

**Lay Executive Committee member**

- Getting their help in writing child-friendly summaries of the findings from BPSU studies

*We never look at whether the information we disseminate is of any relevance to children themselves. As more websites come up where children can find out about their health – perhaps that would be useful to look at.*

**Executive Committee member**

It was also recognised that involving children and young people requires a specialist approach - skills and expertise beyond the capacity of current Executive Committee members. It would therefore require investment of resources and careful planning to ensure the process was meaningful and relevant to all involved. Some interviewees were aware that the Royal College has a member of staff who is responsible for involving children and young people and suggested this person be approached for specialist advice.

## 6. Promoting PPI and supporting researchers

Several of the interviewees thought that researchers would benefit from more practical advice on *how* to involve patients/ patient representatives in their work:

*Everybody's very keen to support PPI but it's knowing what it means... we're all in favour of it and think there should be more including me – but if you ask us how we go about it - we scratch our heads – there's not so much practical advice about this is what you do and this is what you want to get out of it in the end.*

**Researcher**

Suggestions for the training and support that the BPSU might offer included:

- Advice from the lay members of the Executive Committee

*That could be an extension of the [lay members'] role - a more active start, quite early to help researchers understand what they need to do and give them some of the skills that they need, point them in the right direction.*

**Executive Committee member**

*If a rare condition doesn't have an established charity/ patient group then advice on how to approach families or lay reps – practicalities of that would be useful.*

**Researcher**

- Promoting shared learning amongst researchers i.e. facilitating sharing of experiences and examples of good practice

*Ultimately the biggest drivers for change aren't organisations like us, but colleagues who've had a good experience. If someone else says, it worked really well for me. It is quite a small community; some of those networks are already there and working.*

**Lay Executive Committee member**

*Giving them concrete examples really helps them think, understand what it is they're trying to aim for and how it could benefit them. Examples of studies that were successful.*

**Executive Committee member**

*We're good at communicating – paediatrics is a small world – having some kind of formal mechanism [to share PPI experiences] could be useful.*

**Researcher**

- Signposting researchers to organisations who can offer advice and support including patient organisations

*They [researchers] could come to organisations supporting people with the condition and find out about how to involve people with that condition – for example they could arrange a meeting to talk about what they may need to consider when involving people...*

**Patient/ patient rep**

- Better promotion of PPI for example through talks at conferences/ sessions about PPI at the Royal College of Paediatricians conference/ a PPI leaflet / dissemination of this report/ more information on the BPSU website

The lay members were conscious of making PPI a positive experience for researchers and an activity that has value for them:

*What we don't want to happen is people to go away and think 'The burden of PPI is so great for me I just never want to do it again'. Some are experienced clinicians, some much more junior, on a learning curve. They're the champions of the future. It's got to be a positive experience. I'm very conscious of my role in that.*

**Lay Executive Committee member**

Researchers were also aware of the huge cultural shift that PPI represents and the importance of engaging people who are new to PPI and may not appreciate its value:

*You're talking to someone who's converted – so you need to find ways to reach out to the people who are not converted.*

**Researcher**

## 7. The challenges to taking forward PPI at the BPSU

Many of the interviewees thought that the main challenge to taking forward any of the recommendations for future work is a lack of resources – both of funding and time, and in particular the limited capacity of Executive Committee members and BPSU staff to carry out the work required:

*We've got to look at how PPI will be resourced in the future. Not just the individuals, but pieces of work, support, mentoring...*

**Lay Executive Committee member**

Some suggestions for overcoming these difficulties were to:

- Seek funding to recruit more BPSU staff
- Seek funding to pay lay members for their involvement
- Increase the numbers of lay members
- Identify individuals to lead on certain projects, supported by sub-groups of Executive Committee members – thus spreading the workload

With some of the recommendations for the way forward, it has been assumed that patient organisations/ groups will be able to offer help e.g. in writing PILs and lay summaries of findings. However, there are also capacity issues here, because some patient organisations are small and overstretched, and may also be receiving similar requests for help from other researchers and research institutions:

*Who would do it? This is always the question. People always look to the lay person. But how much time have they got available? And do they have the ability to convert information into succinct lay-friendly reports?*

**Lay Executive Committee member**

These limits on resources re-emphasise the importance of prioritising different recommendations.

## **8. Recommendations for future work**

The Steering Group and Executive Committee agreed that the recommendations fell into the following order of priority:

### ***Recognition of existing lay members and promotion of PPI***

1. Include information about the lay members and the work they do on the BPSU website, in newsletters and the annual report.
2. The Executive Committee should promote the PPI work it has carried out to date and facilitate sharing of the lessons learnt through: conferences, websites and publications. This should help researchers to understand how PPI will be of value to them and how best to go about it.

### ***Recruitment of new lay members***

3. The Executive Committee should work together as a whole to decide:
  - how many new lay members it requires and why
  - the essential/ desirable and skills and experience they will require of new members
  - whether it wishes to pay lay members in future and how funding might be obtained for this purpose
  - the process for recruiting new people
4. Evidence of best practice should be sought from other organisations to help shape any new developments in the lay membership (e.g. payment policies)
5. Recruitment needs to be planned and timed carefully to allow a handover from existing lay members and for new members to receive an induction before they start.
6. Any new scientific or clinical members of the committee should also be given an induction and importantly an explanation of the role of lay members.
7. New lay members should be asked about their ongoing training/ information and support needs on a regular basis. Structures/ processes should be put in place to ensure that the necessary training and support is provided or sourced externally if more appropriate.

### ***Clarity about the BPSU's expectations of PPI in BPSU studies***

8. The Executive Committee should develop clear and explicit guidance on when and why it expects to see PPI a different stages of BPSU studies. This should consider:
  - a. the evidence available of where PPI has been shown to bring added value and where it has not made a difference
  - b. the limits of BPSU influence on researchers and whether the BPSU wants to make PPI a condition for approving studies
  - c. where patient versus public involvement is most appropriate

### ***Guidance for researchers***

9. The BPSU should produce clear and explicit guidance for researchers on when and why PPI is expected in their study and provide a definition of what they mean by PPI. This should help researchers understand 'how' to involve people and also convince them of the value of involvement in BPSU studies.

### ***Refining the application forms and review processes***

10. The section on PPI in the phase 2 application form should be refined to reflect the context of BPSU studies and the expectations of PPI (see above).
11. The review process should ask for more explicit evidence of involvement that has already taken place and clear plans for future involvement.
12. Researchers should be given clearer and more explicit feedback on their PPI proposals so they know 'how' to respond.

### ***Focus on dissemination***

13. PPI in dissemination seems to offer great potential for added value from PPI but has so far been underdeveloped. The Executive Committee should consider:
  - a. How best to encourage and support researchers to involve people at this stage
  - b. Creating the infrastructure to support this work e.g. establishing a network of relevant organisations, dedicating a section of the BPSU website to disseminating lay-friendly reports of findings from completed studies

### ***Expanding PPI activity***

14. The Executive Committee should consider whether it wishes to expand its PPI activity into new areas – in particular getting more involved in influencing public health policy or involving children and young people.

## **9. Next steps**

The Steering Group and Executive Committee agreed with the recommendations from this review and in addition recommended that the BPSU should incorporate a statement of principle in support of PPI within the terms of reference for the organisation. This will provide a public demonstration of their commitment to PPI.

The Executive Committee also agreed to establish a PPI working group who will develop an action plan to implement the recommendations from this review.