SUSTAINING PATIENT AND PUBLIC INVOLVEMENT IN RESEARCH: A CASE STUDY OF A RESEARCH CENTRE.

Clare Jinks[1], Pam Carter[2], Carol Rhodes[1], Roger Beech[1], Krysia Dziedzic[1], Rhian Hughes[1], Steven Blackburn[1], Bie Nio Ong[1].

1 Arthritis Research UK Primary Care Centre, Institute of Primary Care and Health Sciences, Keele University, Keele ST5 5BG, UK

2 Social Science Applied to Healthcare Improvement Research (SAPPHIRE) Group & East Midlands Research Design Service, Department of Health Sciences, University of Leicester, 28 Princess Road West, Leicester LE1 6TP.

Corresponding author: BN Ong.
Tel 01782-734708
Fax 01782-733911
Email b.n.ong@keele.ac.uk

Abstract

The literature on Patient and Public Involvement (PPI) in research covers a wide range of topics. However, one area of investigation that appears underdeveloped is the sustainability and impact of PPI beyond involvement in time-limited research projects. This paper presents a case study of PPI development in one primary care research centre in England, and its approach to making this sustainable using documentary sources and material from a formal evaluation.

We provide narrative accounts of the set-up, operation and main processes of PPI, and its perceived impact. PPI requires a long-term perspective with participation and trust growing over time, and both users and researchers learning what approaches work best. PPI is a complex interplay of clarity of purpose, defined roles and relationships, organised support (paid PPI staff) and a well-funded infrastructure. 'Soft systems' are equally important such as flexible and informal approaches to meetings, adapting timetables and environments to meet the needs of lay members and to create spaces for relationships to develop between researchers and lay members that are based on mutual trust and respect. This case study highlights that the right combination of ethos, flexible working practices, leadership and secure funding goes a long way to embedding PPI beyond ad hoc involvement. This allows PPI in research to be integrated in the infrastructure and sustainable.

Introduction.

The burgeoning international literature on patient and public involvement (PPI) in research covers a wide range of issues: active participation as distinguished from participation as a research subject;[1,2] PPI in research design[3,4] and throughout the research cycle;[5,6] the various roles that patients can play in research;[7] assessing the impact of PPI[8,9] and formulating recommendations for good PPI practice.[10-13] Conceptual and ideological tensions have been identified, with the democratic, principled or ideological rationale for PPI presenting challenges to an instrumental or consequentialist rationale.[14,15] One area of investigation that appears to be underdeveloped is the sustainability and impact of PPI beyond involvement in time-limited research projects. Beresford[3] mentions the importance of

planning for sustainable involvement, but provides little further detail. A small number of longer-term involvement examples have been reported, such as OMERACT (Outcome Measures in Rheumatology) where rheumatology patients have worked with clinical researchers for over ten years;[7] or the long-term resourcing and embedding of PPI in joint university and NHS research.[16]

This paper focuses on the issues of sustainability and the importance of institutional leadership and the creation of a robust infrastructure in order to achieve long-term and wide-ranging PPI in research strategy and programmes. We start with providing a historical account of the evolution of PPI in the Primary Care Research Centre (the Centre)(anonymised), and follow this with drawing out a number of key conceptual issues regarding infrastructure, resource allocation, working methods, roles and relationships. The paper also addresses ongoing dilemmas and potential tensions before formulating conclusions about the more general applicability of the Centre's model.

A brief history of PPI in the Centre.

The Centre had involved patients in a few research studies during the 1990s[17,18] which highlighted the value of including the patient's voice from the outset in formulating objectives, design and methods. The involvement operated at the level of discrete projects, but in 2006 it was decided that a Centre-wide approach would be more appropriate as its research programme was expanding. The lessons learned from the projects were felt to be important in informing a broader and systematic approach to PPI across the whole of the Centre's research portfolio. This required a shift towards an organisational and structural way of thinking and considering issues around embedding and sustainability, and it would also respond to the emerging drive for PPI from funders.

A letter was sent to a random sample of people who had participated in the Centre's studies, (mostly on musculoskeletal conditions) and who had given permission to be contacted again. They were invited to a meeting to discuss how patients could be involved in the Centre's research. A dozen people came to the session where short presentations were given about the work of the Centre, but the largest part of the meeting was devoted to exploring ideas about involvement. The result was the creation of a Research User Group (RUG) that adopted a definition of PPI that was later confirmed in

the INVOLVE approach: 'public involvement in research' is 'research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them'.[6] The remit of the group was to discuss research proposals with researchers in terms of 'patient benefit', to advise on feasibility and acceptability, and to review research design, methods and materials in order to recommend changes or improvements. These would be based on their lay and direct experience of being a patient, service user or carer.[19] The group had face-to-face meetings and met on average four times throughout the year chaired by a senior researcher. The RUG could be described as a standing panel whose members did not have a stated time of office. This allowed relationships to be built over time, and of the original twelve members half were still active six years later. Working methods evolved over time and broadened out from researchers delivering presentations and consulting RUG members, followed by discussion of issues to receiving regular updates and results, and helping to design dissemination methods.

In 2006 the first Centre PPI strategic framework was formulated which was a necessary requirement to secure the support from the North Staffordshire Research Consortium (the 'Consortium') that funded the PPI activities. Creating a budget caused considerable debate about realistic costing and in particular payment to RUG members (in recognition of their investment of time and expertise) was a major issue. After seeking advice from various sources, such as INVOLVE, the Citizens' Advice Bureau and HM Revenue and Customs, and consulting literature[20] a rewards policy was devised that took into account the diverse circumstances of research users. Some RUG members were unable to claim payment for their time because of the impact on their welfare benefits. Some claimed, while others chose not to. Irrespective of financial recompense, it was important to members to receive feedback and acknowledgement and researchers provided this in different ways: one member received a bouquet of flowers; another member was sent a letter explaining the difference that her contribution had made to the researcher's thinking about a particular study. Having an established PPI budget enabled senior academics to help setting up the RUG, making it work and forging links with the Centre's researchers. They played an important role in raising awareness of PPI internally and externally to funders and partner organisations.

Increasingly, the RUG members were invited to speak at meetings about their experiences, for example, at the Chartered Society of Physiotherapists, UK. They were part of a research team presenting a proposal to the Health Foundation for funding. These activities increased their confidence and led the RUG to organise and run a 'PPI in Research' conference in October 2008 which attracted a national audience. All presentations and workshops were led jointly by patients and researchers.

In 2008 the Centre became a Centre of Excellence, qualifying for infrastructure funding from Arthritis Research UK (ARUK) of £2,500,000 over five years. This award was a major achievement for the Centre and PPI played a key role. Assessors, including international researchers, spoke to three of the RUG members as part of their assessment. Earlier sessions with Centre staff to prepare for the important visit helped RUG members feel confident as a group and they talked with passion about their involvement at the Centre to assessors. The assessors were impressed by the evidence of PPI at the Centre and a report documents that reviewers described the three RUG members as 'inspirational'.

In 2009 it became clear that the RUG was becoming over-burdened because of the growth in the Centre's portfolio and it was decided to broaden the range of people who lived with a chronic musculoskeletal condition. In response, the Virtual Panel (VP) was set up with people who could be involved via e-mail, telephone and post. Letters were sent to 250 patients of one of the Centre's research practices asking whether they would be interested in working with researchers. Twenty one members of the public attended an information meeting where researchers and RUG members discussed how PPI operated at the Centre, and 19 people joined the VP. In practice, VP members preferred to be involved mainly in face-to-face meetings. Occasionally telephone conferences or postal communication have been used. At this stage, the RUG and VP together included 27 people. Later, following a team building day in 2012 with RUG, some VP members and senior academics to review the Centre's PPI activities and structure, the VP was merged into a re-launched RUG which became the single and more cohesive Centre-wide group to support PPI activities, with several working parties aligned to the Centre's main programmes (Figure 1).

Figure 1. Centre PPI structure 2014
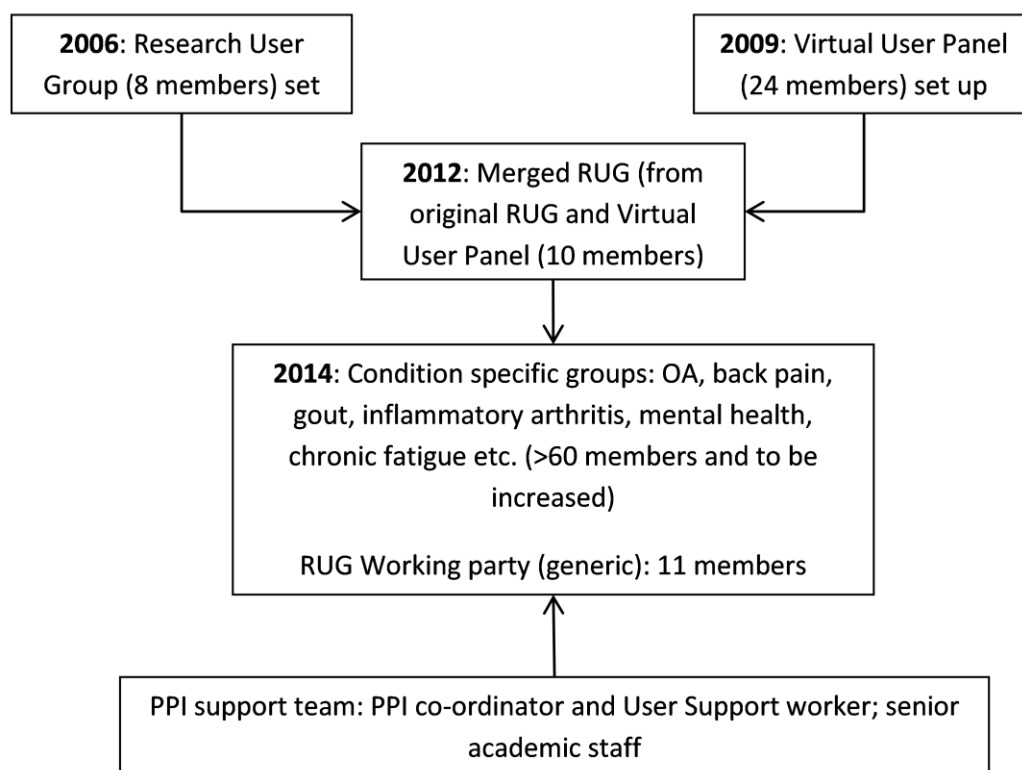
Over the years, the involvement of RUG members has evolved and broadened (Table 1, Figure 2). Two large National Institute of Health Research (NIHR) programmes on which RUG members were co-applicants and sat on their Steering Groups, were funded in 2008 and 2009. This led to the formation of specialist back pain and osteoarthritis (OA) PPI groups (Figure 3) which allowed them to influence the direction and content of the two programmes (e.g. the original idea for the OA programme came from users; delivering training for professionals) and working alongside researchers in the constituent projects that drew on their specific interest and lived experience.

Table 1. Selected milestones of the Research User Group (RUG)

Year	Milestone

Year	Event
2006	Set up of original Research User Group
2007	RUG member interviewed as part of team for Health Foundation grant
2007	RUG members as Co-applicants to two National Institute for Health Research programmes
2007	First RUG member on Research Consortium board
2007	First RUG member to present at a national conference for physiotherapists
2008	User-led national conference
2008	Two members participated in INVOLVE conference
2008	Three RUG members interviewed as part of bid for ARUK Centre of Excellence status
2009	Appointment of User Support Worker
2009	Launch of Virtual User panel
2010	First of formal training sessions for RUG
2010	Two RUG members presented at INVOLVE conference
2011	Formal evaluation of PPI in the Centre
2011	First exchange visit with the Netherlands
2011	First RUG annual meeting
2012	PPI co-ordinator presented at training event of patient research partners (EULAR)
2012	PPI co-ordinator appointed to the INVOLVE Advisory group
2012	Appointment of PPI co-ordinator, RUG member appointed as User Support worker
2012	Three RUG members presented as part of the assessment for renewal of ARUK Centre of Excellence
2012	RUG re-launch (Virtual User Panel merged with RUG)
2012	Production of guides for RUG members and researchers
2012	Article about the Centre's PPI in "Arthritis Today"
2013	Expansion of PPI into areas such as gout, inflammatory arthritis, chronic fatigue/ME and mental health (increase to 60 RUG members)
2013	RUG members and PPI co-ordinator teach at BSR fellowship for Rheumatologists

2014	Clinical Trials Unit status awarded to Centre, and PPI restructured to be aligned to condition-specific work streams.

Figure 2. Types of PPI activity in the Centre's research
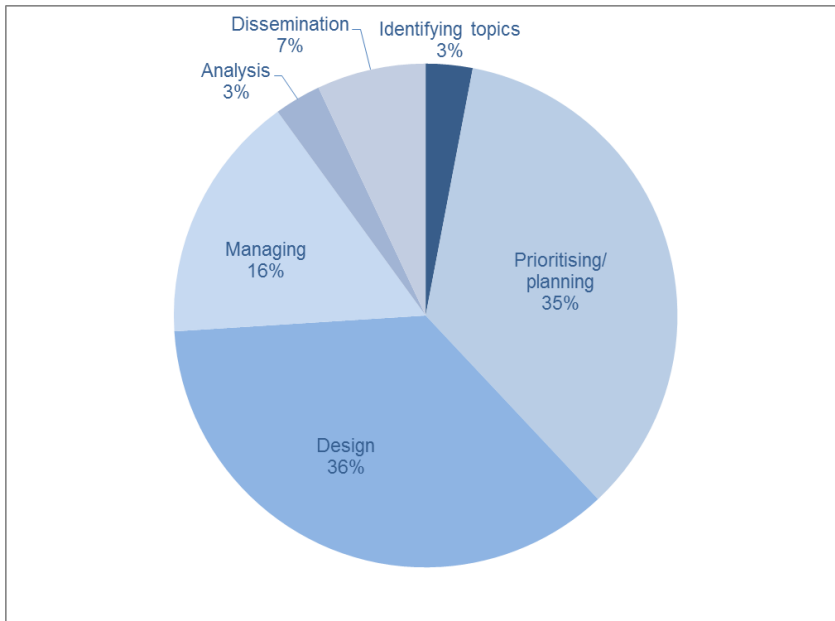
Figure 3. PPI in the NIHR Osteoarthritis Programme.

- Initiated topics for study through discussions with research team

Identifying and prioritising

Commissioning

- Discussion of overall study outline and possible research methods
- OA Guidebook designed in collaboration with RUG.
- Development of consensus statements for a Delphi exercise/ RUG involved in Delphi for sub studies.
- Prioritised content of questionnaire for cluster trial
- Developed quality indicators for osteoarthritis care.
- Delivered training to Health Care Practitioners delivering

- Help plan and took part in the evaluation of the Centre's PPI activities

Evaluating impact

Research Cycle

Designing and managing

Implementing

Undertaking

- OA Group formulate proposals for their implementation and dissemination.
- Presented at INVOLVE conference on the RUG activities.
- Work of RUG and the development of OA guidebook published in INVOLVE newsletter
- Participated in Consensus conference to discuss and disseminate study findings

Disseminating

- Members of project steering committees for sub studies and two members sit on the overarching OA Management group.
- Co-applicant on NIHR OA programme grant.
- Patient advisor on a research assistant recruitment panel
- Involved in interpreting early data and findings

Support provided to RUG members:
- Plain English glossary of research terminology
- Plain English summaries of each research theme
- A series of guidebooks about different aspects of research
- Training event: Contributing Assertively in Meetings
- Annual RUG event to celebrate their contribution

PPI involvement increased beyond the Centre into ARUK Clinical Studies Groups (CSGs). These were fora for supporting researchers and funders to establish research priorities for funding, and to help shape the design and improve the quality of grant applications. Two RUG members sat on the Consortium board, one on the Comprehensive Local Research Network and several on their GP practice groups.

The expansion of activities and numbers meant that it became increasingly difficult for the supporting senior researchers to run PPI alongside their own research. The Research Institute manager who provided key logistical support in terms of financial issues (processing payments and expenses) and liaison with the Human Resources department (honorary contracts) could not also absorb all the extra work. As a result a new paid post of User Support Worker was created (Supplementary Material 1). The main purpose of this post was to act as a bridge between patients and researchers and coordinate all activities.[21] Importantly, the person had to live with a chronic musculoskeletal condition him/herself so that they could bring an experiential perspective. In 2009 this worker was appointed

and started to design systems for researchers to request PPI in their research, organise training and support for patients, develop information, network with local and national PPI organisations and much more. The PPI infrastructure was underpinned by a budget earmarked by the Deputy Director of the Centre, who also was the Director of the Research Consortium. Stable Centre infrastructure funding, rather than project-dependent funding, allowed for continuous engagement of PPI and for this to become embedded in the Centre's work. Explicit rules of engagement between researchers and RUG/VP members were developed, clarifying respective roles, expectations and responsibilities. These were defined for the different types of activities such as participation in projects and programmes, sitting on boards or Steering Groups. With the Centre's success in attracting large grants PPI had to expand again and by 2012 more than 35 people were involved in 30 projects across the Centre's portfolio. This led to re-profiling the User Support Worker to become the PPI Co-ordinator, taking a more pro-active and outwards facing role, and the recruitment of a new User Support Worker. One of the longest-standing RUG members was appointed to this position in 2012.

The various iterations of the Centre's PPI model have been informed by comparisons with other institutions in the UK and abroad. The PPI team networked extensively, for example, with INVOLVE, Arthritis and Rheumatism Musculoskeletal Alliance and EULAR (European League Against Rheumatism) and went on a study visit to the Netherlands. A Dutch expert held a series of workshops for researchers and RUG members at the Centre. From these exchanges it became apparent that the Centre's approach was considered distinctive in its comprehensiveness: being properly funded, led by an academic lead for PPI (KD) who sits on the Institute's Management Board, supported by a dedicated PPI coordinator (CR) and support worker who together maintained good communication and feedback with RUG members. Informal workshops about various research topics and attendance at internal and external conferences increased the confidence and capability of the RUG.

The following discussion of conceptual issues how sustainability of PPI was addressed in the Centre will partly draw on the formal evaluation that was carried out by one of the co-authors (PC) in 2012.

PC analysed existing documents, interviewed researchers and RUG members and observed a number of meetings (internal report).

Key conceptual issues in sustainability.

Who gets involved and whose voice counts?

There is much debate in the PPI literature about the 'usual suspects',[11] user-led, participatory or consultative approaches[1] and the relationship or distinction between service users and the general public.[22] The Centre PPI Framework does not specify selection criteria apart from the key criterion that the individual must have 'expertise by experience' of the health condition being researched in the Centre, that is, experience of a relevant musculoskeletal condition. Carers have also been included but their role has not always been clear in the Centre's activities: as a (practical) support to the patient or providing a distinct perspective. The recruitment process influences who gets involved, and the majority of people have entered the RUG through participation in previous research studies which tend to be situated within the Centre's geographical area. This process is supplemented by targeted recruitment for new research areas such as gout (10 individuals), chronic fatigue (5), self harm (7), heart failure (3) and plantar fasciitis (2). Most are available to attend day time meetings so this might be regarded as unwritten, informal, selection criteria. In contrast to, for example, the Dutch Rheumatology organisation, educational attainment is not considered and the RUG members' background ranges from having left school at 15 years of age to holding a PhD. Apart from when people hold University casual employment contracts, monitoring of diversity is not carried out. However, the coordinator and support worker have consulted with an external group that has expertise in supporting Black and Minority Ethnic communities in research. They attended the RUG annual meeting in 2013 to discuss how diversity of the RUG could be increased as its members are predominantly white British. Targeting specific community groups, for example the Polish community in one of the catchment area's towns, will form part of the ongoing recruitment strategy.

The emphasis on the experience of illness that people bring to the table has had to be negotiated. For the RUG members with research or educational expertise it has not been easy to put that aside and

focus on their experiential contribution. Researchers had to navigate this carefully so as not to alienate these members, yet, be clear that the Centre's own professional expertise was sufficient. From the perspective of the RUG members, they felt that while they shared having a musculoskeletal condition, their experiences were very diverse and provided a rich source of information for researchers. Two issues are important here: first, in the early days of the RUG people wanted to talk extensively about their personal experiences and were less focused on discussing research. The Centre team considered this a natural part of team building and creating trust, and allowed meetings to be relatively informal. Over time, the agenda and discussions shifted more towards research matters, and group members themselves said that the 'RUG is not a mutual support group' stating that they could find that elsewhere. Second, the right balance needed to be found between the personal experience, and whether this pointed to more generic issues worth exploring. Some researchers worried about 'representativeness' of the RUG members, while in contrast, others could discern themes within the experiential accounts. Thus, ongoing discussions between the senior researchers involved with the RUG and colleagues had to clarify that representation was not the purpose of the RUG, rather how people's experiences could inform and enrich the research.

De Wit and colleagues[7] draw on a Habermasian framework by arguing that the 'life world' of the patient and the 'system world' of research have been considered as separate, but if PPI is to be incorporated in research these two world views have to learn to value each other. An example will highlight this: a new research proposal on carpal tunnel syndrome was presented to the RUG and a discussion ensued about the key questions that the research should address. A RUG member explained that she lost her job as a phlebotomist because the condition stopped her from doing fine finger movements. The researchers realised that they had not considered the importance of remaining in work and being economically active within their proposal. Thus, the one story raised awareness of a major area of investigation that was therefore included in the (successful) bid. Another example was one where RUG members wanted more attention to be paid to complementary medicine as they often used this. The researchers explained that at the time it was not a Centre priority even though that type

of research had been carried out previously. This again raised the question whose voice counts in prioritising research topics and that it can involve difficult negotiations.

Institutional and personal support.

In terms of support for PPI recognition of potential barriers to involvement have to be recognised. Attitudes of researchers can be negative or at best tokenistic, and consistent leadership in promoting PPI in the Centre has been important. This comes in different forms: first, providing infrastructure resources in the form of paid workers who can explain the value of PPI to researchers. They also set up a request system whereby researchers have to describe why they need RUG members, in what role, the time commitment required and the types of participation. In this way RUG members can be matched to research projects according to their condition, interests, abilities, circumstances and sometimes their personal characterstics. In turn, researchers need to think carefully about how to manage PPI engagement in the development of their research and account for the impact that it may have. It also enables researchers to have ready access to a PPI pool, removing the responsibility from them to find and recruit people. Second, recognition of PPI is established at institutional level through having a voice at the Institute Management Board, being included in Centre and Consortium strategies which are reinforced by national policy. Third, inclusion of fully costed PPI in projects and programmes emphasises to researchers that it is not an optional extra. This has evolved into many proposals now having RUG members as co-applicants and being part of interviews when bidding for funding. Finally, the support from the Centre's leadership in allocating a dedicated budget from the infrastructure funds (ARUK and Consortium) has provided a stable footing for all PPI activities and paid staff over a long period.

Where problems sometimes arose was when RUG members attended meetings chaired by academics from outside the Centre who had little experience in PPI. They reported feelings of being ignored or dismissed, leading them to decline further participation. Little and colleagues[23] used the term 'dys–empowerment' to describe such 'feelings of humiliation, anger, indignation and hostility' that might arise from negative experiences of involvement. These instances led the PPI coordinator to design a

clear approach to supporting RUG members, which included planning a preparatory meeting with Chief Investigators, and the Support Worker accompanying people to meetings.

Practical support has been important to facilitate RUG members' attendance at Centre and national/international meetings. The conduct of meetings had to change with RUG members feeling that they were given opportunities to contribute, which depended much on the chair and if they were explicitly asked for their views this was seen as helpful. Awareness that people who lived with discomfort related to their conditions needed to have regular breaks, letting people walk about, sorting accessible car parking or ensuring other appropriate ways of transport (e.g. accompanied) were essential. Again, infrastructure funding played a key role in that resources were available to make this possible.

Increasingly RUG members have been asked to speak at national and international conferences. They have the opportunity to prepare for these presentations together with the PPI team and relevant researchers, often 'rehearsing' their contribution. Again, they tend to be accompanied by researchers or the Support Worker.

Language can also be a barrier to involvement, especially when users are new and unused to scientific jargon and conventions. This is a two-way process with researchers speaking in plain English, or explaining terms and concepts either in the meetings or beforehand. The PPI team has produced a glossary of terms and a series of leaflets to outline different aspects of the research process. They also wrote leaflets for researchers as to how to make the most of PPI. For some time RUG members resisted any formal training worrying that they would lose their authenticity, but they increasingly realised that having relevant grounding in research designs and/or methods would make their involvement more effective. As a result they have increased their work in areas such as systematic reviews and analysing interview transcripts. An academic publication was co-authored by a researcher and RUG member.[24]

Roles and relationships.

As mentioned before RUG members were recruited as 'experts in experience'. Role clarity is crucial to successful PPI[7] and the Centre has taken this on board. While formal roles can be described, the practice may differ and in particular as a result of evolving relationships between researchers and RUG members. On occasion, Centre staff have felt that RUG members went 'off script' or undermined professional sensibilities. This illustrates the tensions between empowerment, collaboration and organisational support for PPI.[25] The RUG members place considerable emphasis on trust and respect, and want encounters with researchers to be informal, personal and inclusive.[26]

Researchers understood this need, yet, focused more on effective ways of working and clarity of purpose. The balance between these differing needs remained something to be continually negotiated, especially when people came together who had not previously collaborated. The time investment required to create and maintain relationships was considerable for both parties, but in general was considered as an integral part of ensuring the positive impact of PPI on research. One of the 'unintended effects' has been that several RUG members gained so much confidence from their roles in research that they could extrapolate this to other parts of their lives. The User Support Worker changed from someone who had not been in paid employment for years as a result of her condition to being able to take on this position. Another RUG member became the Chair of the local Arthritis Rheumatism Musculoskeletal Alliance branch. They felt that their Centre involvement conferred benefits to them personally beyond what they had expected.

Further considerations.

Ensuring that PPI is consistently followed through from the start to the end of research continues to be a challenge. The co-ordinator now reminds researchers that the RUG appreciates a discussion of results and dissemination plans, and in particular the identification of the impact that PPI has had on the design, methods and outcome of research. Reporting changes to consent procedures, redesigning interview schedules or questionnaires are examples of PPI influence as well as 'big occasion' examples where RUG members positively contributed to the external assessment of the Centre by

funders. RUG members have been invited to actively take part in developing a dissemination strategy for the Centre's work.

The PPI co-ordinator and support worker developed a communications strategy with RUG members that can respond flexibly to individual needs. This ranges from personal telephone conversations to prepare for meetings, to sending information electronically. In this way they maintain individualised contact and the result has been that many RUG members stay with the Centre for many years. This is key to creating a sustainable PPI structure as individuals build up their knowledge and expertise and feel that they increase their effective involvement.

Enhancing the profile of PPI within and beyond the Centre continues to be important. The PPI team and senior academics supporting them ensure that PPI remains an organisational priority, for example, with the Centre gaining Clinical Trials Unit status PPI has been formally included within its systems and operating procedures. Furthermore, it informs the work of the Research Design Service and under the umbrella of PILAR (Public Involvement and Lay Accountability in Research) provides a linked strategy for PPI in health-related research and implementation across the West Midlands. The Centre may be regarded at the forefront of embedding PPI through combining 'hard' (structural) and 'soft' (cultural) approaches, and reflects much of the latest thinking.[27]

Conclusion.

The case of the (anonymised) Centre highlights that sustaining PPI in research is a complex interplay of clarity of purpose, defined roles and relationships, organised support and a robust infrastructure that is well-funded. At the same time 'soft systems' are equally important such as flexible and informal approaches to meetings, adapting timetables and environments to meet the needs of current and new RUG members and most importantly, to create spaces for relationships to develop between researchers and RUG members that are based on mutual trust and respect. This requires researchers to be able to cope with a certain degree of uncertainty in planning and conducting research. At the institutional level it has been clear that learning from mistakes was important, and regular review of processes and impact had to take place. The formal evaluation (internal report) was a key element in

this ongoing journey, and helped to refocus both the PPI strategy and the PPI voice at decision-making levels. The benefits that RUG members reported were increased skills and confidence, access to social support and feeling useful in retirement. Researchers suggested that PPI impacted on research in the form of successful funding applications, improved team communication and enhanced validity of research instruments.[19] A key factor throughout had been the commitment of the Centre's leadership to allocate financial resources so that a robust infrastructure could be established.

The general principles that underpin the Centre's approach are a result of learning from others (literature and exchanges) and reflecting on our own mistakes. Tensions will remain in terms of a potential conflict between genuine involvement versus the demands and timetables of funders and researchers. Many other research centres may not have the financial capability or the leadership commitment required to create a sustainable infrastructure for PPI. However, we argue that the relevance of our case study is that the right combination of ethos, flexible working practices, leadership and secure funding goes a long way to embedding PPI beyond ad hoc involvement. The expertise that can be built up by both researchers and lay members will be incremental and enhance current and future research in meaningful ways.

Authors' contributions.

PC and BNO wrote the first draft of the paper; CJ and CR provided key data for the paper and all the

figures; CJ, CR, RB, RH, KD and SB gave detailed comments for the final submission. No conflicts

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