Patient and Public Involvement in Research
a Journey to Co-Production

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The public and patients can be powerful sensors for shaping and powering healthcare research. They are joining research teams as investigators and collaborators to co-produce evidence for the practical use of interventions in clinical practice.

While clinicians and researchers are encouraged by funders and policymakers to involve the public and patients as partners in research, knowledge on what involvement consists of is limited, and the continuum between consultation, collaboration and co-production are not clearly defined. In this article, we explore patient and public involvement (PPI) and introduce greater involvement through research co-production. Co-production describes ways that research partnership can work through public and patient involvement and we outline the similarities of co-production to “The Commons”, a strategy utilized by economists to increase effective use of resources. We share examples of how Public and Patient Involvement have used co-production, to demonstrate financial and health
benefits. We then outline practical challenges at system, social and cultural levels and consider how others have worked to resolve them.

1. Background

The term Patient and Public involvement (PPI) is applied to research that is conducted ‘with’ or ‘by’ members of the public and patients rather than ‘on,’ ‘to,’ ‘about’ or ‘for’ them.[1,2] PPI is not serving as a research participant, answering surveys, being an interviewee or focus group participant providing a patient opinion.[3] PPI takes place when patients and members of the public work directly with researchers and health professionals to serve as informants, innovators, and distributors of health information.

PPI and Co-production of Healthcare Research

“Co-Production builds relationship beyond hierarchy. The relationship’s success is not in striving to make each other equal. The success of the relationship builds qualities that are necessary for people to work in safety outside of the hierarchy. Because then the rules, so to speak, are internalized within the relationship through mutual respect to strengthen the partnership. Hierarchy is external and is rule-driven to enforce boundaries and to minimize change. Co-production through relationship retains order as it extends boundaries”. (Amy Price 2020) [4]

Co-production is part of the spectrum of Patient and Public Involvement (PPI) in healthcare research and provides the opportunity for researchers, practitioners and the public to work together, sharing power and responsibility from the start to the end of the project, to create, redesign and generate research knowledge. [5] Other parts of the spectrum might include consultation, protocol and manuscript review, feedback on research questions, participant materials, editing survey questions, consent forms, information sheets and summaries for readability and providing assistance with recruitment and public engagement and dissemination [1,6]. Patients and the public can also initiate research co-production as they seek out researchers and clinicians to partner with them in research. Baker and colleagues articulate this well; “When patients are consulted, they are given information but have limited impact on decisions; when patients partner, they share power and are active participants in defining agendas and making decisions”. [7] See Box-1.

Box-1 INVOLVE Co-Production Values
1.1 The Healthcare Co-Production Commons

In the co-production of research, roles within research teams are assigned according to skill competencies, rather than by traditional power hierarchies based on professional backgrounds and educational degrees. Elinor Ostrom and Oliver Williamson are economists and Nobel laureates, whose research about co-production in communities demonstrates that ordinary citizens are capable of managing and sustaining resources without outside control to maximize the use of scarce resources. Ostrom et al. assert co-production occurs by combining professional expertise with the energy expenditure, wisdom, experience, and skills of end-users [8] nurtured through the core standards of love, empathy, watchfulness, care, reciprocity and willing instruction. [9]

Batalden et al. cite Victor Fuchs and Eugene Nelson [10] as they describe the co-production of healthcare services as ‘The interdependent work of users and professionals to design, create, develop, deliver, assess and improve the relationships and actions that contribute to the health of individuals and populations to create value’. In healthcare research, co-production could create value as resources for opportunity, training and implementation become available.

Co-production does not imply equality in skills or ownership but equal respect, voice, and access to shared resources. In health research, as in life, partnerships are seldom equal, likewise with the distribution of skills. Shared access to skills and knowledge in healthcare can multiply influence and scaffold community members to enrich common resources. Defining mutually beneficial roles can help everyone work together, productively as a team. Figure 1 illustrates potential research co-production tasks through multiple phases of the research process[11].
1.2 Freely Available Resources

Nasser and colleagues report the UK’s National Institute for Health Research (NIHR) as the leading funder worldwide for PPI. [12] The NIHR generates considerable growth in the understanding and breadth of research co-production.[13] The 2018 standards incorporate inclusive opportunities, working together, support and learning, communication, impact and governance. NIHR INVOLVE has recently produced guidance on research co-production and outlines where this differs from active involvement as a participant.[14,15] See Table 1 for helpful resources.

Table-1
2 Financial Value and Practical Value

A prognostic model built to assess the financial value of trials found potential for a 500-fold return on investment.[16] The model targets reducing the costs of under-recruitment and protocol amendments by improving clinical trial design through co-production. The estimates the authors used are based on historical cost models and not active case studies. The estimates do not estimate the considerable cost of unsuccessful research co-production. [17] For this reason, we share examples of successes and failures of research co-production.

2.1 The Protec Trial

In the “Prostate testing for cancer treatment trial" (ProtecT)[18,19] total funding inclusive of follow-up exceeds £39,722,444.52.[18] Co-production of research was reported as beneficial early in the project after investigators reported rebuilding consent forms with patients as co-production partners. In addition, the patients recommended renaming the “watchful waiting" arm to “active monitoring". These changes improved recruitment by 40-70% with significant gains in participant satisfaction and retention.[19,20]

2.2 The IMPROVDENT Trial

The “Improving dentures for patient benefit" (IMPROVDENT) trial applied research co-production to make the intervention “user-friendly" by following their patient partner recommendations. This action resulted in fewer appointment cancellations and timely recruitment. In a study by Vogsen et al. [21], revisions to participant information materials that were suggested by patient partners increased the expected recruitment by 146% and this benefit remained robust through follow-up, with 86% of participants completing follow-up. There was initial resistance to inviting patients as team partners but as the research continued even the most reluctant researchers applauded the collaboration with patient representatives [19],

2.3 The SWITCH RCT

In a less successful trial, the “Alternative tumor necrosis factor inhibitors (TNFi) or abatacept or rituximab following the failure of initial TNFi in rheumatoid arthritis: the SWITCH RCT" [22,23] Pavitt, one of the investigators shares how the co-production of research improved ethical and practical aspects of the study design, by changing trial conditions to eliminate unneeded procedures, forms, and appointments and by ensuring that the burden of participation was similar for intervention and control groups [20]. Pavitt projected that co-production of research could multiply participant retention,
and reduce trial costs by an estimated £2.5 million.[20] Sadly, the trial recruited slowly, with only 122 participants due to third-party contractual and commissioning difficulties. Recruitment was slow and resulted in funders terminating the trial while leaving research co-production benefits unrealized. This outcome may have been avoided by adopting QuinteT Recruitment Intervention (QRI) methods for recruiting. This method audio-records recruitment appointments, interviews recruiters and trial management staff and then maps eligibility, recruiting strategies and other trial documents. The results are triangulated, shared and discussed with the research team to build an improved plan of action. [24]

We note that co-production benefits are not limited to financial gains. A recent systematic review and meta-analysis of the impact of PPI found benefits for enrolment in clinical trials.[25] Others report substantial improvements and implementations of services;[26] numerous improvements were documented in social care,[27] enhancements in research design and end-user experience can be attributed to co-production. [28] Despite the evidence of the advantages of research co-production, doubts linger. [29]

3 Challenges

There are multiple reporting challenges for research co-production and this means benefits may occur but remain unreported. The consequences are that methods used in research co-production are disconnected from research, leaving researchers and patient partners to navigate shared power, role confusion, misplaced assumptions, and unmet expectations with minimal guidance and unclear impact.[1] In the following section, we address these points in detail along with recommendations for better reporting of research co-production using an “Everyone Included” no one left behind [30] philosophy.

3.1 Gaps in Reporting Research Co-Production

While the evidence base of research co-production has expanded over the past decade, the reporting of co-production in research papers is still inconsistent and partial, with minimal information about the context and process of public involvement.[31] One challenge is that co-production of research might remain hidden, but reported in a separate journal that is not linked to the primary paper. Inconsistent or absent reporting contributes to an evidence base where the collective understanding of what works, for whom, why, and in what context is fragmented.[32] The GRIPP-2 long and short-form research reporting guidelines can improve the quality of co-production research planning and reporting. We recommend that authors and researchers use this guidance to inform research
protocols and in reporting and writing up research.[31] While an account of research co-production is a requirement in most final reports for NIHR funded research, the publishing of these methods and results with the primary paper is not required. In publishing, since 2017, The BMJ requires authors to report PPI and research co-production. Since 2019 The BMJ asks authors to outline any barriers and challenges to initiating PPI and to state how they plan to share their work with others including patients and members of the public. [33] Multiple journals including CMAJ and 30 of the BMJ sister journals now follow this reporting practice.[34]

3.2 Disparity and Representativeness

There are worries about who qualifies as a patient and whether patients can be too “expert”. [35] For study investigators, the “patient state” may be critical to test interventions or define the sample population. However, with research partners or members of the public, these constraints may not apply. Besides, healthcare professionals and researchers can be patients too, and it would be discriminatory to disallow their input.[35] Others argue that research systems favor educated, middle-class patient advocates who are emotionally controlled and not very ill, they signpost the need for genuine partnership with psychological safety for research and in clinical practice.[36] On the other hand, there is a misplaced emphasis on having only “representative.” patients. Their role is not to represent other patients in the data sense of representativeness. It is to provide a range of perspectives about an experience. In fact, the range of perspectives, methods and practice can increase value. We see this in other research fields where experiences can lead to conducting a case study and further lead to a hypothesis that when tested, may change practice. [37,38]

Research co-production and implementation require adequate funding and full system support that is co-produced with a clear strategy underpinned by mutually agreed values. Lay persons on the team need to contribute to the goals and methods of the research and be aware of the power of their attitudes to build or discourage the team. Words can be devastating, and respect must be mutual. Researchers and decision-makers need to feel safe and supported. Consider that while organisations wield power, the researchers and decision-makers within them are as vulnerable and fragile as any layperson. There may be the need for all parties to debrief about past hurts in research relationships or health care mistreatment and to build trust before entering into beneficial research co-production.[39] Furthermore, without mindful leadership, patient and public chosen priorities and outcomes may be neglected and not converted into the language or context where someone will hear them.[40] Mutually agreed values can reduce unintentional harm in vulnerable populations, including in areas such as forensic psychiatry,[34] research in sick children [41] and sexual health. [42] Poor
PPI experiences can alienate, stigmatize and isolate patients and the public.[29] Professional researchers, often employed on short-term grants with little or no job security, also have needs for respect and support to enable effective co-production, which requires significant time and resources to facilitate relationship building with involved patients and the public.[43]

The ReseArch with Patient and Public InvOlvement: a RealisT evaluation (RAPPORT) study 2015 [44], reports that 51% of (n = 92) studies contained evidence of PPI, although transparency in reporting the PPI was minimal. The authors identify funder requirements and study design as influencers and “clear purpose, role and structure for PPI; ensuring diversity; whole research team engagement with PPI; mutual understanding and trust between the researchers and lay representatives; ensuring opportunities for PPI throughout the research process; and reflecting on, appraising and evaluating PPI within a research study” as the six salient actions for effectiveness. Box 2 offers words of experience from PPI co-production partners.

Box- 2 Words of experience from our PPI Co-production Partners

Strategies that work in one research study may need to be adapted to gain maximum benefit in a future project. Research projects are unique, and uncertainty is a constant factor, even for those with experience and training. [45]

3.3 Adopting an Everyone Included Philosophy

Early adoption of co-production research by using an Everyone Included philosophy to “provide the stage from which even the hardest and most difficult stories can be told” could be a constructive solution [11]. Diverse expertise and shared power within a health research team can lead to creative and innovative solutions.[11] Stanford Medicine X launched The Everyone included philosophy to provide a co-production platform. In this atmosphere, PCORI launched reviewer training and The Apple Research kit was born [46]. The Everyone Included initiative provided the standard for public and patient partnership guidelines for the Precision medicine initiative [47] and the National Institutes of Health program All of Us, [48]. One group of clinicians explains, “Having a patient in the room has changed the way we look at serious incidents”. [49] Benefits can work both ways. Having a clinician or researcher in the room can also expand patients’ and public perspectives in patient-initiated research. Confidence and equality can be reinforced by asserting that best outcomes do not always come by agreement without discussion but that mutual respect and resilience for change are prerequisites to being heard.[6] Greenhalgh and colleagues published a review of 65 PPI frameworks and they
suggest local relationship based ideals might be strengthened by adapting local frameworks using existing evidence-based resources rather than depending on a one-size-fits-all framework [50].

4 Discussion

PPI is an integral component of research methods however power sharing through co-production requires a fundamental change in academic mindset and practice. The right to equality and co-production conversations between researchers, clinicians and patients tends to overlook the realities of hierarchical academic life and the uneven distribution of power amongst researchers; between research disciplines; and the administrations which shape their futures, such as employers, funders, planning boards, and publishing obligations [51]. We note through the article examples that co-production has improved health services and brought considerable gains for the research enterprise. These examples are few as the co-production is inadequately reported in the literature. Co-production is an emergent concept and the guidance on how to proceed is limited. Newly minted researchers and clinicians may feel powerless to ensure quality and stability in co-production. Senior personnel may not yet see co-production as a priority and may be uncomfortable with the uncertainties, the time and the targeted focus and emotional labor that co-production can evoke. Nevertheless, researchers, clinicians, and patients working in co-production hold the potential to transform healthcare research and build the kind of respect and solutions that transcend hierarchies and build partnerships.

4.1 Conclusions

We are on the brink of moving from a model of researcher-driven action to research co-production, with patient, public and clinician partners identifying research needs and actively involved in the production of knowledge and its implementation.[52] The time is ripe for the effective implementation and reporting of PPI in research that grows beyond logging patient experience to the real power shift of co-created research and social policy aimed to refine clinical practice and deliver community benefit.[53] Co-production matters because it generates personalized solutions that can take research from the lab to implementation and minimize the burden of illness, through the shared management of co-created care. [54] The American Board of Internal Medicine summarized work in this area in a publication titled “The Value of Co-Creation in Health Care” [55]. Researchers, clinicians and patients will continue to negotiate effective partnerships and identify shared interests.[5] Victor Fuchs noted as early as 1968 that measuring productivity in the service economy, was challenging because consumers and providers of services “always work together to create value”. [10]
Co-production can make “Nothing about me without me” the accepted policy in research, health and social care including a partnership in health technology, policy, clinical guidelines, shared decision making, industry, medical training and quality standards. Co-production is collaboration in action to create value by embracing diversity as we aim for service improvement through effective co-production partnerships and implementation.

DECLARATIONS

Ethics approval and consent to participate

The research reported here used existing published publicly available literature where ethics clearance was not required.

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Co-Production

Marjorie Kirkpatrick and Yazmin Nelken were our co-production partners and co-authors, they worked side by side with us to conceive and write this article. Their perspectives, examples, and encouragement made this a better paper and strengthened our support for co-production.

Acknowledgments

Marjorie Kirkpatrick (Jo) one of our co-authors died Friday March 13, 2021. She was Amy Price’s (AP) first university friend at Open University, where Dr Price attended after sustaining a significant brain injury. We are sharing Amy’s words in memorial “She showed me how to make friends, included me in her study groups, fixed my references and the one side of my papers that suffered from the unilateral blindsight, she introduced me to the Disabled Students Association and celebrated that I went from crippled and brain injured to completing a doctorate at Oxford. She was kind, funny, smart, opinionated, imperfect and curious. I loved her. She was one of my closest friends and one of our patient authors.” We are grateful for the helpful feedback we received from Navjoyt Ladher MD and Dr Sara Schroter of the BMJ. Special thanks to Professor Glyn Elwyn who urged AP to write a paper to help researchers navigate struggles with research co-production. We appreciate the detailed help of the editors and reviewers in structuring our content.
Competing Interest Disclosures

AP is the Research Editor (Patient and Public Partnership) at The BMJ and is a senior research advisor with Medicine X. LC is on editorial boards at the BMJ and is the Executive Director of Stanford Medicine X, MC is an academic researcher involved in many clinical trials and systematic reviews who seeks funding for these trials and reviews, as well as for research into methodology, including dissemination and accessibility. SS is the Co-editor in chief of Research Involvement and Engagement and is part funded by NIHR ARC-West Midlands, NIHR HPRU in Gastrointestinal Infections and NIHR HPRU in Genomics and Enabling Data. DT is the Senior Research Manager, NIHR Evaluation Trials and Studies Coordinating Centre, MK and YN have no competing interests to declare.

Authors’ contributions

AP conceived the paper, wrote the initial draft and worked through the edits suggested by co-authors. LC contributed the graphics for (Figure 1), All authors contributed to the writing of this manuscript and approved its final version.

Availability of data and materials

All data used and analysed are present within the manuscript.

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Table 1: Freely available resources for PPI and Research Co-production

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<tr>
<th>Resource</th>
<th>Description</th>
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<tbody>
<tr>
<td>Canadian Institutes of Health Research, Strategy for Patient-Oriented Research (SPOR)⁵⁹</td>
<td>Patient Engagement Framework <a href="http://www.cihr-irsc.gc.ca/e/48413.html#a11">http://www.cihr-irsc.gc.ca/e/48413.html#a11</a></td>
</tr>
<tr>
<td>European Patient Academy (EUPATI)⁶⁰ Network of European National Platforms</td>
<td>PPI and training in medicines research including pharmaceuticals and regulatory agencies. <a href="https://www.eupati.eu/">https://www.eupati.eu/</a></td>
</tr>
<tr>
<td><strong>Gordon and Betty Moore Foundation</strong> and The American Institutes for Research⁶¹</td>
<td>Roadmap for patient and family engagement in health and research: <a href="http://patientfamilyengagement.org/">http://patientfamilyengagement.org/</a></td>
</tr>
<tr>
<td>INVOLVE Funded by NIHR⁸</td>
<td>Support for PPI in NHS, public and social care research. <a href="http://www.invo.org.uk/">http://www.invo.org.uk/</a></td>
</tr>
<tr>
<td><strong>Everyone Included</strong>⁷⁰⁶¹ Stanford Medicine X, Stanford School of Medicine, USA³⁰</td>
<td>Co-production leadership principles with videos, coursework and training for health research redesign. <a href="http://everyoneincluded.org/">http://everyoneincluded.org/</a></td>
</tr>
<tr>
<td>Hatching Ideas Hub (International)⁶²</td>
<td><a href="https://www.hatchingideashub.com/serviceuser-oriented-research-resources">https://www.hatchingideashub.com/serviceuser-oriented-research-resources</a></td>
</tr>
<tr>
<td>National Co-ordinating Centre for Public Engagement (NCCPE) UK⁵³</td>
<td>Help to explore, support and plan public engagement. <a href="https://www.publicengagement.ac.uk/do-engagement/choose-method">https://www.publicengagement.ac.uk/do-engagement/choose-method</a></td>
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<tr>
<td>NIHR Research design service (RDS) UK⁴²</td>
<td>Support for grant application development (UK only) <a href="https://www.nihr.ac.uk/about-us/how-we-are-managed/our-structure/research/research-design-service/">https://www.nihr.ac.uk/about-us/how-we-are-managed/our-structure/research/research-design-service/</a></td>
</tr>
<tr>
<td>Patient-Centered Outcomes Research Institute (PCORI) USA⁴⁵</td>
<td>Engagement awards and conference support (USA only) <a href="https://www.pcori.org/funding-opportunities/announcement/engagement-award-conference-support">https://www.pcori.org/funding-opportunities/announcement/engagement-award-conference-support</a></td>
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<tr>
<td>People in research (UK only)⁶⁴</td>
<td>Connects stakeholders for public involvement in NHS, public health and social care research <a href="https://www.peopleinresearch.org/">https://www.peopleinresearch.org/</a></td>
</tr>
<tr>
<td>RAND Corporation⁶³</td>
<td>Patient and public involvement in research. Enabling meaningful contributions <a href="https://www.rand.org/pubs/research_reports/RR2678.html?dm_i=4V0Q,45NG,1R0SYT,DW13,1">https://www.rand.org/pubs/research_reports/RR2678.html?dm_i=4V0Q,45NG,1R0SYT,DW13,1</a></td>
</tr>
<tr>
<td>Public Involvement Resource Hub</td>
<td>Key steps in the public involvement process, with links to further resources, <a href="https://www.imperial.ac.uk/patient-experience-research-centre/ppi/ppi-resource-hub">https://www.imperial.ac.uk/patient-experience-research-centre/ppi/ppi-resource-hub</a></td>
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<tr>
<td><strong>James Lind Alliance (JLA) Priority Setting Partnerships</strong></td>
<td>Priority Setting Partnerships (PSPs) prioritize evidence uncertainties in particular areas of health and care that could be answered by research</td>
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Authors' contributions statement

AP conceived the paper, wrote the initial draft and worked through the edits suggested by co-authors. LC contributed the graphics for (Figure 2). All authors contributed to the writing of this manuscript and approved its final version.
Declaration of interests

☒ The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

☐ The authors declare the following financial interests/personal relationships which may be considered as potential competing interests:
HIGHLIGHTS

- Power sharing through co-production requires a fundamental change in academic mindset and practice
- Researchers, clinicians, and patients working in partnership have the potential to transform healthcare research
- Active collaboration and respect can sustain partnerships

Reporting highlights the strengths and barriers of Patient and Public Involvement while increasing replicability