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Review Article

Patient and Public Involvement in Research: Lessons for Inflammatory Bowel Disease

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Abstract

Participatory research, also referred to as patient and public involvement, is an approach that involves collaborating with patients affected by the focus of the research, on the design, development and delivery of research to improve outcomes. There are two broad justifications for this: first, that it enhances the quality and relevance of research, and second, that it satisfies the ethical argument for patient inclusion in decisions about them. This synergistic and collaborative effort, which bridges the divide between researchers and participants with the lived condition, is now a mainstream activity and widely accepted as best practice. Although there has been a substantial increase in the literature over the past two decades, little has been published on how participatory research has been used in inflammatory bowel disease [IBD] research and little guidance as to how researchers should go about this. With an increasing incidence and prevalence worldwide, combined with declining study enrolment in an era of perennial unmet need, there are a multitude of benefits of participatory research to IBD patients and investigators, including research output that is informed and relevant to the real world. A key example of participatory research in IBD is the I-CARE study, a large-scale, pan-European observational study assessing the safety of advanced therapies, which had significant patient involvement throughout the study. In this review, we provide a comprehensive overview of the benefits and challenges of participatory research and discuss opportunities of building strategic alliances between IBD patients, healthcare providers and academics to strengthen research outcomes.

Key Words: Patient and public involvement; participatory research; inflammatory bowel disease

1. Introduction: What is Participatory Research?

Participatory research is used to describe research that is conducted 'with' patients rather than 'to', 'on' or 'about' them.[1] It is an umbrella term in the field of qualitative research methodology that encompasses the utilization of various research designs, methods and frameworks, performed in direct collaboration with those affected.[2,3] Numerous definitions and sub-definitions are used interchangeably in the literature, and nomenclature can be confusing, but all share the common denominator of actively involving patients.[4] We use 'participatory research' and 'patient and public involvement' [PPI], two of the more commonly used terms, interchangeably throughout.

Although all research requires participation of some description, participatory research and PPI enable patients, carers or lay public members to be involved at the core stages of design, development and delivery of research. Participatory research is distinct from simple recruitment to research studies or patient completion of a survey; the methodology in the former permits reciprocity and relinquishes a degree of control to the research participant who can influence wider decisions. Table 1 summarizes the key differences between conventional and participatory research. The degree of patient involvement, contribution and shared decision-making is highly variable ranging from generating research questions and designing study protocols, to analysing/interpreting results and co-authoring manuscripts. Table 2 shows the continua of participation in research studies from the passive role of being a data point, to the deeper involvement of an active co-researcher.[2,5] The International Collaboration for Participatory Health Research [ICPHR] does not recommend a specific model for defining the levels of participation in a research process as this is dependent on resources and context.[6,7]

Participatory research was first described in the 1940s in the context of performing research on society's most marginalized

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Table 1. Overview of differences between conventional and participatory research.

		Conventional research	Participatory research
Research focus	What is the research for?	Understanding—action later	Meaningful change/action
	Who is the research for?	Institutional, personal and professional interests	Participants
	Whose knowledge counts?	Clinicians/researchers	Shared
	Who influences choice of topic?	Funding priorities, institutional agendas, professional interests	Shared
	What is the role of the researcher?	Director/driver/investigator	Facilitator/catalyst
	What is the methodology chosen for?	Disciplinary conventions, 'objectivity' and 'truth'	Empowerment, mutual learning
Roles in stages of research process	Problem identification	Clinicians/researchers	Shared
	Data collection	Clinicians/researchers	Shared
	Interpretation	Clinicians/researchers	Local concepts and frameworks
	Analysis	Clinicians/researchers	Shared
	Presentation of findings	By researcher to other academics or funding body	Locally accessible and useful
	Action on findings	Separate and may not happen	Integral to the process
	Who takes action?	Researcher, external agencies	Shared
	Who owns the results?	Clinicians/researchers	Shared
	What is emphasized?	Outcomes	Process

Adapted from: Cornwall et al. [1995][2] and Amaya et al. [2014].[5]

populations.[8] The concept that 'individuals should not be seen as empty vessels and objects of inquiry, but as full participants in inquiry, able to determine their own needs in order to improve their own lives' gained significant traction in the late 1990s/early 2000s.[9] There is now an increasing awareness of the importance of PPI in health-related research, but the implementation of this is uneven and not routinely embedded into healthcare systems, particularly across Europe.[10] In areas where this is well established, such as the UK and North America, reporting PPI is increasingly mandated on the application forms of funding bodies and ethical committees. Nonetheless, the past 15 years has seen a proliferation of studies investigating its utilization, particularly in the fields of diabetes, mental health and social care research.[11,12] However, there is a dearth of literature about how participatory research can be of benefit to patients with chronic diseases, such as inflammatory bowel disease [IBD]. Here, we outline the potential benefits and challenges of PPI and then examine how this can be and has been of benefit in IBD research, the principles of which can be widely applied to other medical disciplines.

2. What are the Potential Benefits to IBD Patients?

There are several potential benefits to patients with IBD involved in participatory research. Conventionally, the principal investigator assumes responsibility for setting the research agenda and driving the research processes, creating a power imbalance and division on multiple levels between researcher and the researched. IBD patients report they often feel like 'guinea pigs' being 'experimented on' and have a poor understanding of the research process, thus disincentivizing them from partaking.[13,14] A fundamental tenet of participatory research is that these differences are minimized or eradicated to place patients on an equal footing with researchers and removing negative stereotypes.[15] Involving patients not only increases understanding of their condition, but also provides knowledge and skills in research methodology so that they feel more valued and part of the solution rather than part of the problem.

Balancing patient needs alongside researcher needs by allowing patients to express what matters most to them can be an empowering experience. It ensures studies are investigating clinical outcomes that are important to them, such as pain, urgency and quality of life. It also allows patients to evaluate whether study designs, such as a placebo arm, or interventions such as repeated endoscopic evaluations, are acceptable to them. By involving patients in decision-making, the motivation for the work they are involved in increases.[16] It demonstrates that meaningful attempts are being made to research the condition afflicting them. The importance of patients then sharing these positive experiences with the wider patient community should not be underestimated.

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Table 2. Degree of participation in research projects with examples.

Contractual	Researchers consent/contract patients to participate in research studies	Patients agree to enrol into a clinical trial of a novel compound at the baseline visit and complete a written consent form/Patients agree to complete a quality of life or IBD symptom questionnaire
Consultative	Researchers consult patients for their opinions at various stages before interventions are made. Researchers need not act on them	Patients are asked for their opinion on the readability of the investigator-led study protocol, patient information leaflet and/or consent form for comment
Collaborative	Researchers and patients work together on projects designed, initiated and managed by researchers. Patients may or may not be able to influence important decisions	Patients have a position on the IBD research project board and develop the study protocol and discuss how best to recruit patients
Collegiate/co-production	Researchers and patients work together as colleagues with different skills to offer, in a process of mutual learning where patients have power to influence wider decisions and able to lead	Patients co-lead the design, data collection and analysis exploring the psychosocial relationship of IBD patients and food, with full academic support from patient and clinician researchers

Adapted from: Cornwall et al. [1995][2] and Amaya et al. [2014].[5]

Researcher-Controlled ⟷ Ownership-Devolved

Superficial ⟷ Deep

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3. What are the Potential Benefits to IBD Researchers?

Academics should seek to actively partner with patients in the research process rather than simply viewing them as research subjects enrolled in a study. Patients provide a unique insight into how IBD impacts them and can generate new research ideas based on their lived experiences and assess whether certain interventions and outcomes are likely to meet their needs. They can challenge assumptions and identify problems and solutions that researchers may not have otherwise detected, for example simplifying protocols or creating pragmatic study designs.[17] This input can be empowering and can strengthen the academic and patient-group relationship to maximize rapport, which improves the likelihood of future research collaborations. Furthermore, positive patient experiences mean they will not dissuade others in the IBD patient community from joining and provide enhanced access to study populations.

Despite the perennial unmet need for durably effective and safe IBD treatments, recruiting to phase IIb/III clinical trials for novel IBD agents has declined over the past decade.[18] There are multiple reasons for this but is in part due to patient misunderstandings about research processes and insufficient patient–physician communication.[13,14] The design and conduct of clinical trials often fail to take account of patients' experiences.[19] Participatory research has been shown to improve enrolment and retention rates in clinical trials and enhance recruitment to investigator-led studies.[20] Crocker et al. describe numerous PPI interventions that have previously been successfully utilized. These include improving the quality and readability of patient information sheets, leading patient events for recruitment, having a designated trial contact for lay research explanations, developing educational videos on research processes, having ethnic minority representation present to dispel stigma, and analysing reasons for refusal to participate to modify recruitment strategies.[20]

Early adoption of participatory research may have advantages for researchers. Accumulating evidence highlights that patient engagement improves the quality, relevance and research outcomes.[21] In view of this, evidence of PPI is increasingly being requested from funding organizations and ethical bodies. Furthermore, some journals such as the *British Medical Journal* have amended their policy and now require that the level of PPI is disclosed at submission and explain how authors plan to disseminate the results to the patient community.[19,22] Incorporating this approach earlier to gain experience may help satisfy future mandated requirements from funders and journals.

4. How can Participatory Research be Introduced into IBD?

To improve the quality and consistency of PPI in research, countries where the process of PPI is well established have set standards to aid researchers in this process.[23] For example, the UK has a national advisory group for supporting patient involvement in health research, which was first founded in 1996 and at the time was one of the very few government-funded programmes established for this purpose worldwide.[24] The National Institute for Health and Care Research [NIHR] in the UK sets standards for PPI in research and provides ideas and guidance to researchers. Similar government-funded bodies in the USA, the Patient Centred Outcomes Research Institute [PCORI], and Canada, Strategy for Patient-Oriented Research [SPOR], aim to achieve similar objectives.[25,26] Ethical approval is usually not needed where people are involved in planning or advising on research. Patients can make important contributions to the research at various stages of the cycle. Figure 1 outlines how patients and members of the public [important if healthy controls are needed] can be involved in research from identifying research topics to analysis and dissemination of results. Figure 2 details important considerations that researchers should take when taking patients into their research sphere.

For researchers, it is first important to select appropriate patient representatives and then to ensure they are adequately trained to fulfil the role. Patients can be selected by convenience sampling, such as at the time of an outpatient clinic consultation or endoscopy visit. Equally, patients already enrolled into trials/studies can be approached to see if they wish to be involved as a collaborator or co-researcher in future research. Inviting patients with the aid of volunteer-led charities, such as the Crohn's and Colitis Foundation [USA], Crohn's and Colitis [UK] or AFA Crohn RCH [France], may further help to ensure varied demographics and disease characteristics. This includes, for example, new diagnoses, patients on a specific treatment or indeed ethnic diversity, with the aim of recruiting a highly motivated and representative cohort.[27] However, no particularly strategy has been found to be more successful for recruiting patients.[28] Second, as outlined in Table 2, researchers should work with patients to establish the level of patient input, based on feasibility, resources and context, and decide which stages of the research cycle patients should be involved. Careful consideration should be given to how to engage with patients, that is whether this is in the form of a regular membership to an advisory or steering group or invitations to project-specific meetings. It is essential to foster a climate of open communication. Finally, evaluating PPI is an important activity to determine its impact on research outcomes, but this is poorly reported in the literature. Evaluation not only identifies whether the original aims and objectives defined during planning were achieved, but also improves the planning of future projects. Although no single framework for evaluating PPI in research has been unanimously adopted by researchers, the Guidance for Reporting Involvement of Patients and the Public 2 [GRIPP2] is the first international guidance for reporting of patient and public involvement in health and social care research.[29]

5. How has Participatory Research been used in IBD?

There are several ways in which IBD patients have been engaged in participatory IBD research, although the extent to which this is reported is minimal. A recent systematic review identified only 14 studies in which PPI in IBD research was reported.[30] However, these findings were published in abstract form only and details of the search strategy and methodology are unavailable. Most of the identified studies engaged patients in the form of focus groups and for the development of study materials, for example questionnaires and patient-friendly study tools.[30] In this section, we describe several examples of how patient participation has been incorporated into IBD research.

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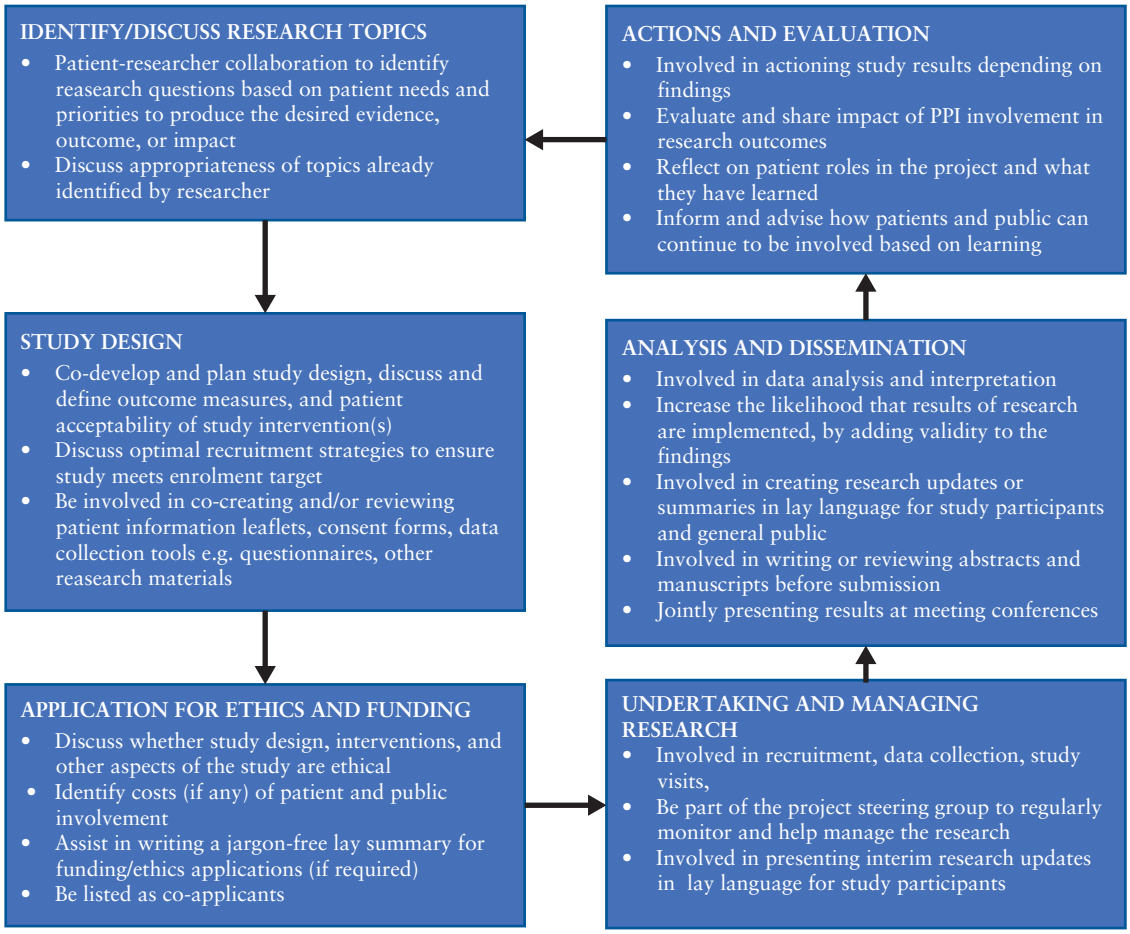

Figure 1. Incorporating participatory research into IBD studies—what can the patient and the public do?

The AFA Crohn RCH is an association of French IBD patients created in 1982 with ~35 000 members.[31] Although large-scale nationwide surveys on disease perception have been distributed through the AFA, more recently, studies have had significant PPI.[31–34] Perhaps one of the best examples of IBD participatory research is the I-CARE [IBD-Cancer And seRious infections in Europe] study and originates from France.[35] This Europe-wide, prospective observational study seeks to assess the long-term safety of advanced IBD therapies and has eventually enrolled 10 206 patients as active participants in the research project.[35,36] Here, the AFA was intimately involved in all aspects of study set up and delivery, including the co-creation of patient diaries and patient-reported outcome measures [PROMs]. The subsequent patient completion of ~350 000 diaries with PROMs has facilitated the development of a unique database of IBD patients with outcomes that are likely to shape future clinical practice.[36]

Another prominent example involving the AFA and patient-reported outcomes is the FLARE-IBD study, which developed a validated PROM tool to measure disease relapse in IBD and was constructed with patient perspectives throughout, including patient discussions in online forums.[34] An IBD patient, who represents and consulted the extensive AFA network, and is part of the study's scientific committee, was involved in all decisions at each stage of the research, and is a co-author on the resultant publication [including the present paper [A.B.]].[34] Similarly, Adegbola et al. developed and validated a PROM for perianal fistulas in Crohn's disease [CD] with PPI.[37] Here, four patients with fistulas were part of the study steering group, were involved in the PROM development and data analyses, were manuscript co-authors, and also later organized a PPI day to boost recruitment and response rates.[37] PROMs are increasingly used as endpoints in clinical trials and in real-world practice. However, very few PROMs in IBD have had patient involvement during their development.[38] PROMs are indeed 'patient-reported' but they are usually 'physician-devised' for patients to complete. The aforementioned studies demonstrate a meaningful shift towards a 'patient-devised' approach.

There are several examples of where PPI has been used to guide trial design and intervention. The Medical Rehabilitation in Chronic Inflammatory Bowel Disease [MERCED] randomized controlled trial [RCT] evaluated the effectiveness of an intense 12-month rehabilitation programme for IBD patients. Rehabilitation included, but was not limited to, stress management counselling and dietetic advice, and the effect on health-related quality of life and psychological well-being was measured. The project was supported throughout by an eight-person patient panel who reviewed all project materials including flyers, cover letters, study information and questionnaires. The rehabilitation interventions and outcome measures were jointly devised by patients and researchers.[39] The second example is the ongoing Optimisation before Crohn's surgery using Exclusive enteral Nutrition [OCEaN] RCT, which compares pre-operative exclusive enteral nutrition to usual diet in patients undergoing CD-related surgery. A panel of eight

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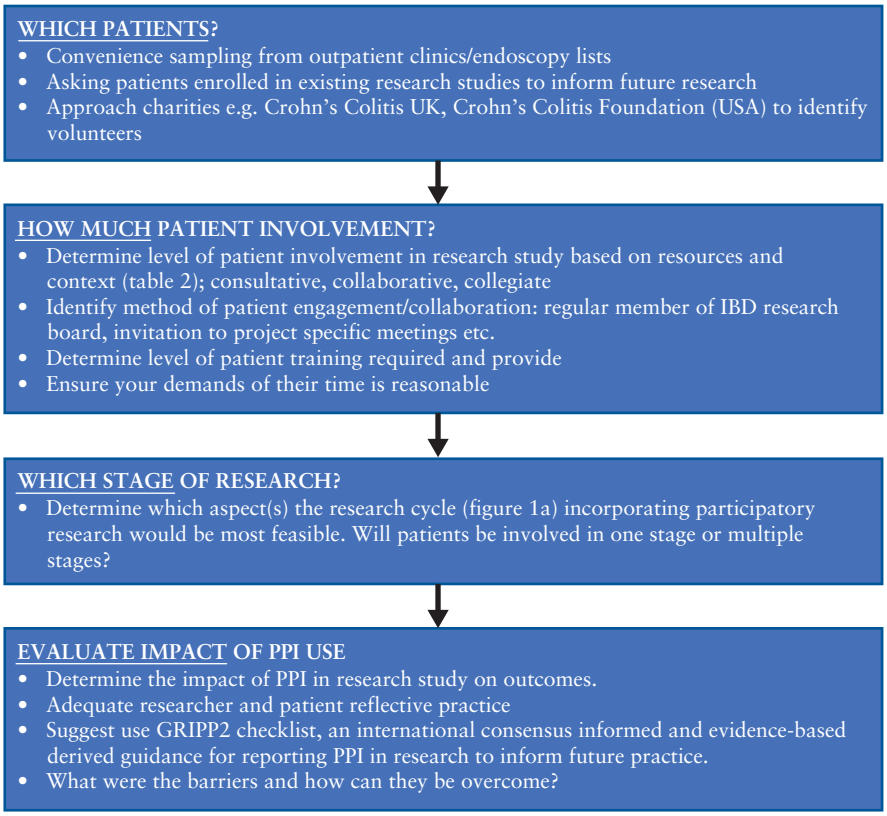

Figure 2. Incorporating participatory research into IBD studies—what can researchers do?

IBD patients, who received training in research terminology, was established to inform the design and conduct of the OCEaN study. PPI in this study was integral to determining the trial duration, mode of delivery and outcome measures, which were all chosen by patients.[40] The final example is of AlphaBiomics, a precision medicine company investigating the stool microbiome and therapeutic response to advanced therapies. Their project panel consisted of patient representatives who advised on study design and patient recruitment, reviewed the stool collection kit, and drafted patient-friendly instructions for use to improve acceptability.[41] Patients will also be invited to co-author research updates and help communicate research findings.[41] These engagements exemplify the importance of PPI in studies where the acceptability of interventions/investigations are in question.

Examples described so far are those of a collaborative rather than a collegiate approach to participatory research, with study ownership only partially devolved [Table 2]. There are very few examples in the literature where patients take the lead in decision-making. Dames et al. published the first study entirely conceptualized and delivered by patients with some input from clinicians/academics.[42] The study, which investigated the impact of colorectal and pelvic surgery on sexual function, identified significant unmet physical and psychosexual needs pre- and post-IBD surgery.[42] Patients led this research voluntarily without pay and the proposal did not require ethical approval as this was driven by non-institution-affiliated patients. While patients steered decision-making in all stages of the research without prior training in research methodology, there was guidance from clinicians/academics, but only after invitation. Similarly, Zelinsky et al. conducted a patient-led study assessing motivations and barriers to a concurrent longitudinal environmental factor IBD study from the same institution.[43] Patient representatives had some input from clinician academics when required, but were already trained in research methodology and led at every stage of the research, with findings shared by patients at international IBD conferences.

6. Can we Learn from other Medical Specialties?

Several medical fields have adopted participatory research from which the IBD community can gain key insights. Two aspects of direct relevance to IBD include the use of PPI to boost recruitment and retention to clinical trials and the use of PPI in drug development and regulation. A systematic review and meta-analysis including 26 UK/North American trials across a broad range of medical disciplines demonstrated significant increases in trial enrolment rates.[20] This was particularly the case if patients involved had lived experience of the condition in question. While the impact on retention was less clear due to the paucity of eligible studies, the positive findings with varying degrees and types of PPI interventions, as already described above, add further weight to the argument for PPI in trial design and conduct.[17,20] Regulatory agencies, such as the Food and Drug Administration and the European Medicines Agency, have started to introduce PPI initiatives.[44] These include series of PPI workshops, patient-focused drug development documents and long-term strategies that place patient involvement at the heart of their delivery plans.[44] PPI in drug development is particularly important following the high levels of distrust, misinformation and anti-vaxxer campaigns in the Covid-19 pandemic.

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While significant advances in participatory research have been made in the field of social care research, lessons may be learned from specialties managing immune-mediated inflammatory diseases using advanced therapies and research structures not too dissimilar from IBD. In 2011, the European League Against Rheumatism [EULAR] recommended that at least two patient representatives are included in all scientific projects at every stage of the research.[45] Such recommendations from an international IBD body are not yet in place. Resultant patient partnerships over the past decade have positively influenced the scope and conduct of outcomes research in rheumatology, with previously neglected domains such as fatigue, insomnia and effects of disease relapse on quality of life now at the fore.[46] Patient involvement has also been shown to be beneficial in basic or translational research by motivating young scientists performing laboratory research who hear the patient perspective.[47] It also allows patients to better understand research processes and the complexities of their own disease, which in turn can inspire them to become involved in further projects. Examples of established PPI models in rheumatology that facilitate ongoing collaboration include the Oxford-based OPEN ARMS initiative, a large patient advisory group that co-creates study recruitment material with an overarching aim of promoting diversity and involving patients from hard-to-reach community groups.[48] Another example comes from the NIHR Leeds-based rheumatology group of 250 patient/public volunteers with ten core members who meet bi-monthly to help prioritize research topics and help academics focus on inclusivity.[48] Both PPI groups regularly apply frameworks to monitor and evaluate the success of PPI in all stages of research at their institutions, an approach that should be adopted by all involved in participatory research.

Similar strides for patient inclusion in research have been taken in dermatology, where the physical and psychological burdens of inflammatory diseases remain at the core of its unmet needs. Although there are numerous examples of patient involvement at the study design and outcome-defining stages, some attention should be given to how PPI has been incorporated at other research stages.[49] Recruitment is a significant challenge for many academics but one way this can be enhanced is through broadcast and social media. For example, in the CLOTHES trial, Thomas et al. identified significant expressions of interest from eligible participants within 3 months after patient representatives discussed the impact of atopic dermatitis on local and national news.[50] In the SAFA trial of spironolactone in acne, patient and public representatives of the trial management group harnessed the power of social media to boost recruitment.[51] Similar strategies may also be used for dissemination of study findings and conclusions. Although patients are less involved during the data analysis stages, patient representatives of the qualitative RECAP study of atopic eczema were involved in deciding the variables for the multivariate regression analyses.[52] Finally, in a recent vitiligo RCT, patients' representatives were involved in all aspects of the research but were also enabled to analyse treatment success of study participants by scoring clinical photographs.[53]

While these examples provide important lessons, it is important to remember that uptake of participatory research is not universal and is at different stages within medicine specialties. For example, geriatrics falls short of achieving ideals related to collaboration and co-researching. Synthesis of existing literature identified that older patients were rarely positioned as prominent research partners or were rarely meaningfully involved in all stages of research.[54] The suboptimal involvement, engagement and participation may relate to a number of challenges that we outline next.

7. Challenges and Future Considerations

One of the major hurdles of adopting participatory research is that healthcare professionals lack the practical knowledge and experience of how best to design and deliver it.[55,56] While detailed guidance is available from government-funded bodies such as the NIHR in the UK, and the PCORI in the USA, tailoring this to specific projects may be challenging.[23,25] There are many indications to suggest that participatory research is beneficial, but there are currently insufficient data demonstrating substantial improvement in research outcomes.[5,57] However, better evidence of the impact of patient involvement in research can be achieved by using established frameworks from those who are committed to co-produce research with patients.[29,58]

There may be a degree of apprehension and uncertainty from researchers as to what a lay-person may bring to the table and whether contributions could affect the methodological rigour, robustness and validity of studies.[17] It may be difficult for academics to relinquish control and direction of the study, and participatory research has always been a source of contention with academics, with the qualitative or 'softer' aspects of this approach looked upon less favourably.[16] Research partnerships can be complex and time-consuming and require commitment from both parties. Additionally, there is a possibility for tensions to arise from disagreements between researchers and patients, which may create ethical dilemmas, and lead to delays in grant funding, study set up and subsequent publication.[7]

Forsythe et al. highlight that the most commonly reported problems with participatory research are the lack of time from researchers and participants, lack of participant training and research experience to allow them to engage, and difficulty finding appropriate patient or public representatives.[59] Appropriately remunerating patients for time spent, particularly those financially affected by active disease, may be considered if funding permits and should be incorporated into grant proposals. These are some of the hurdles that should be overcome at an organizational level to improve acceptability and acceleration of this research approach.

Despite the increasing requirement to document the extent of PPI in research proposals for funding bodies and ethics applications, the frequency of PPI reporting remains low and patient contributors are mostly unacknowledged.[17,21,22] A recent study highlighted that of 148 research publications identified [all NIHR-funded], only 16 [14%] reported some aspect of PPI activity; the UK's NIHR actively promotes the inclusion of PPI.[21] It was largely unclear who was involved, what they contributed and how this eventually impacted research. Journals and peer reviewers do not usually request PPI information within manuscripts, so an absence of information about PPI in research papers could be attributable to either a lack of reporting or a lack of PPI activity. Further education is required to ensure PPI does not become a tick box exercise to satisfy what may perceived to be unnecessary requirements.

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8. Discussion and Conclusions

In this article, we describe some of the benefits and challenges participatory research can bring to the IBD community, although these concepts are transferable across many disciplines of healthcare research. Participatory research, which should be viewed as an approach rather than a specific methodology, offers an important complement to the more traditional investigator-driven research. There are two broad justifications for PPI: first, that it enhances the quality and relevance of research, and second, that it satisfies the ethical argument of patient inclusion for decisions about them.

Patients should be seen as important partners to facilitate effective and accessible research for those living with IBD. Patients may not wish to be involved in all stages of research, but they should be offered the opportunity to do so. While the expectation is not that there should be a sudden abundance of patient-led IBD studies, there is certainly room for cultural change to increase the consultative and collaborating aspects of participatory research to strengthen the status quo. In the UK's National Health Service, this engagement is commonly referred to as PPI rather than participatory research, and has been accepted and acknowledged for the past 30 years.[24] Despite general optimism and accruing data of the positive impact for incorporating this approach, the uptake has thus far been variable.[28,59] Collaboration should not just be between patients and academics but also include early involvement of wider members of the research team, such as study practitioners and IBD nurses. Significant change is only likely if adopted and driven by senior academics, who may still prefer the more linear models of research.

The concept of shared decision-making with patients is not new in IBD.[60] The number of treatments options and paradigms entering the arena for IBD has expanded over the past decade, making decision-making increasingly more complicated. Shared decisions to ensure a degree of personalized care are now integral to the way we manage IBD, for the patients who wish to be involved in decisions about their care. A similar approach may be applied to participatory research. While co-researching is encouraged in IBD, not all patients are right for collaborating or co-researching with and not all research decisions should be shared.

Long-term investments to nurture the academic–patient partnership is important in IBD given the lifelong and life-changing aspect of the disease. These partnerships have been shown to have a positive impact on research to improve its quality and relevance, but there are challenges that need to be considered. Some of these can be avoided by careful planning in the early stages of research. Others, such as substantiating the benefit–risk and economic justification with high-quality evidence, are awaited. Further work in due course should also seek to identify the most optimum method of patient engagement and to establish the effectiveness of participatory research in achieving satisfactory health outcomes.

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Conflict of Interest

SH served as a speaker, a consultant and/or an advisory board member for Pfizer, Janssen, AbbVie and Takeda, and in addition has received travel grants from Ferring and Pharmacosmos. AB declares no conflicts. SD has served as a speaker, consultant and/or advisory board member for Schering-Plough, AbbVie, Actelion, Alphawasserman, AstraZeneca, Cellerix, Cosmo Pharmaceuticals, Ferring, Genentech, Grunenthal, Johnson and Johnson, Millenium Takeda, MSD, Nikkiso Europe GmbH, Novo Nordisk, Nycomed, Pfizer, Pharmacosmos, UCB Pharma and Vifor. LB received consulting fees from Janssen, Pfizer and Allergan; lecture fees from AbbVie, Janssen, Merck Sharp & Dohme, Ferring Pharmaceuticals, Mayoly-Spendler, Takeda and Tillots; and research support from Abbott, Ferring Pharmaceuticals, Hospira-Pfizer, Janssen, Merck Sharp & Dohme, Takeda and Tillots for unrelated studies. LPB reports the following disclosures: Lecture: Galapagos, AbbVie, Janssen, Genentech, Ferring, Tillots, Celltrion, Takeda, Pfizer, Sandoz, Biogen, MSD, Amgen, Vifor, Arena, Lilly, Gilead, Viatris, Medac. Consulting: AbbVie, Alimentiv, Alma Bio Therapeutics, Amgen, Applied Molecular Transport, Arena, Biogen, BMS, Celltrion, CONNECT Biopharm, Cytoki Pharma, Enthera, Ferring, Fresenius Kabi, Galapagos, Genentech, Gilead, Gossamer Bio, GSK, HAC-Pharma, IAG Image Analysis, Index Pharmaceuticals, Inotrem, Janssen, Lilly, Medac, Mopac, Morphic, MSD, Norgine, Novartis, OM Pharma, ONO Pharma, OSE Immunotherapeutics, Pandion Therapeutics, Par'Immune, Pfizer, Prometheus, Protagonist, Roche, Sanofi, Sandoz, Takeda, Theravance, Thermo Fisher, Tigenix, Tillots, Viatris, Vifor, Ysopia, Abivax. Grants: Takeda, Fresenius Kabi, Celltrion.

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Author Contributions

SH drafted the manuscript. AB, SD, LB and LPB critically reviewed and revised the manuscript. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

Data Availability

Data sharing not applicable—no new data were generated.

References

1. Health Research Authority. *What is public involvement in research? [Internet]*. [cited March 25, 2023]. https://www.hra.nhs.uk/planning-and-improving-research/best-practice/public-involvement/
2. Cornwall A, Jewkes R. What is participatory research? *Social Sci Med* 1995;**41**:1667–76.
3. Cargo M, Mercer SL. The value and challenges of participatory research: strengthening its practice. *Annu Rev Public Health* 2008;**29**:325–50.
4. Vaughn LM, Jacquez F. Participatory research methods – choice points in the research process. *JPRM* 2020;**1**. https://jprm.scholasticahq.com/article/13244-participatory-research-methods-choice-points-in-the-research-process.
5. Amaya A, Yeates N. Participatory action research: new users, new contexts, new challenges. In: *Participatory Action Research [Internet]*. Milton Keynes, UK: Open University; 2015 [cited March 26, 2023]. p. 23. https://www.gov.uk/research-for-development-outputs/participatory-action-research-new-users-new-contexts-new-challenges

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6. International Collaboration For Participatory Research. *Position Paper No. 1 [Internet]*. 2013 [cited March 24, 2023]. http://www.icphr.org/1/post/2013/05/position-paper-no-1.html
7. International Collaboration For Participatory Research. *Position Paper 2: Participatory Health Research - A Guide to Ethical Principles and Practice [Internet]*. 2022 [cited March 24, 2023]. http://www.icphr.org/1/post/2022/07/position-paper-2-2e-participatory-health-research-a-guide-to-ethical-principles-and-practice.html
8. Macaulay AC. Participatory research: What is the history? Has the purpose changed? *Fam Pract* 2017;**34**:256–8.
9. Freire P. *Pedagogy of the oppressed*. New York: Herder and Herder; 1970.
10. Biddle MSY, Gibson A, Evans D. Attitudes and approaches to patient and public involvement across Europe: a systematic review. *Health Soc Care Community* 2021;**29**:18–27.
11. Bindels J, Baur V, Cox K, Heijing S, Abma T. Older people as co-researchers: a collaborative journey. *Ageing Soc* 2014;**34**:951–73.
12. Campbell JA, Yan A, Egede LE. Community-based participatory research interventions to improve diabetes outcomes: a systematic review. *Diabetes Educ* 2020;**46**:527–39.
13. Sharp D, Ringer S, Park KT, Tole S, Rubin DT, Regueiro M. Listening to the patient: improving the design and conduct of clinical trials in inflammatory bowel diseases. *Crohns Colitis 360* 2020;**2**:otaa011.
14. Rubin DT, Peyrin-Biroulet L, Reinisch W, *et al*. Inflammatory bowel disease patients' perspectives of clinical trials: a global quantitative and qualitative analysis. *Crohns Colitis 360* 2021;**3**:otab079.
15. Rose D. Participatory research: real or imagined. *Soc Psychiatry Psychiatr Epidemiol* 2018;**53**:765–71.
16. MacDonald C. Understanding participatory action research: a qualitative research methodology option. *Can J Action Res* 2012;**13**:34–50.
17. Price A, Albarqouni L, Kirkpatrick J, *et al*. Patient and public involvement in the design of clinical trials: an overview of systematic reviews. *J Eval Clin Pract* 2018;**24**:240–53.
18. Johnson C, Barnes EL, Zhang X, Long MD. Trends and characteristics of clinical trials participation for inflammatory bowel disease in the United States: a report from IBD partners. *Crohns Colitis 360*. 2020;**2**:otaa023.
19. Wicks P, Richards T, Denegri S, Godlee F. Patients' roles and rights in research. *BMJ* 2018;**362**:k3193.
20. Crocker JC, Ricci-Cabello I, Parker A, *et al*. Impact of patient and public involvement on enrolment and retention in clinical trials: systematic review and meta-analysis. *BMJ* 2018;**363**:k4738. https://www.bmj.com/content/363/bmj.k4738.
21. Jones J, Cowe M, Marks S, *et al*. Reporting on patient and public involvement (PPI) in research publications: using the GRIPP2 checklists with lay co-researchers. *Res Involv Engagem* 2021;**7**:1–13.
22. Price A, Schroter S, Snow R, *et al*. Frequency of reporting on patient and public involvement (PPI) in research studies published in a general medical journal: a descriptive study. *BMJ Open* 2018;**8**:e020452.
23. *UK Standards for Public Involvement [Internet]*. 2023 [cited March 26, 2023]. https://sites.google.com/nihr.ac.uk/pi-standards/home
24. Research Design Service South Central. *Patient and Public Involvement (PPI) [Internet]*. [cited March 26, 2023]. https://www.rds-sc.nihr.ac.uk/ppi-information-resources/
25. Patient Centred Outcomes Research Institute (PCORI) [Internet]. 2023. https://www.pcori.org/
26. Canadian Institutes of Health Research. *Strategy for Patient-Oriented Research - Patient Engagement Framework - CIHR [Internet]*. 2014. https://cihr-irsc.gc.ca/e/48413.html#a6
27. Schöpf-Lazzarino AC, Böhm P, Garske U, *et al*. Involving patients as research partners exemplified by the development and evaluation of a communication-skills training programme (KOKOS-Rheuma). *Z Rheumatol* 2021;**80**:132–9.
28. Domecq JP, Prutsky G, Elraiyah T, *et al*. Patient engagement in research: a systematic review. *BMC Health Serv Res* 2014;**14**:89.
29. Staniszewska S, Brett J, Simera I, *et al*. GRIPP2 reporting checklists: tools to improve reporting of patient and public involvement in research. *BMJ* 2017;**358**:j3453.
30. Elsolh K, Neary E, Seleq S, *et al*. Patient and public involvement (PPIn) in IBD research – a scoping review. *J Can Assoc Gastroenterol* 2022;**5**[Supplement_1]:120–1.
31. AFA. *Bienvenue sur la page d'accueil de notre site internet: Afa Crohn RCH [Internet]*. [cited April 8, 2023]. https://www.afa.asso.fr/
32. Le Berre C, Peyrin-Biroulet L, Buisson A, *et al*. Impact of inflammatory bowel diseases on working life: a French nationwide survey. *Dig Liver Dis* 2019;**51**:961–6.
33. Williet N, Sarter H, Gower-Rousseau C, *et al*. Patient-reported outcomes in a French Nationwide survey of inflammatory bowel disease patients. *J Crohns Colitis* 2017;**11**:165–74.
34. Ricci L, Epstein J, Buisson A, *et al*. Flare-IBD: development and validation of a questionnaire based on patients' messages on an internet forum for early detection of flare in inflammatory bowel disease: study protocol. *BMJ Open* 2020;**10**:e037211.
35. Peyrin-Biroulet L, Rahier JF, Kirchgesner J, *et al*; I-CARE Collaborator Group. I-CARE, a European prospective cohort study assessing safety and effectiveness of biologics in inflammatory bowel disease. *Clin Gastroenterol Hepatol* 2023;**21**:771–788.e10.
36. *I-CARE: Ibd CAncer and seRious infection in Europe [Internet]*. 2022 [cited April 9, 2023]. https://www.icare-ibd.com/
37. Adegbola SO, Dibley L, Sahnan K, *et al*. Development and initial psychometric validation of a patient-reported outcome measure for Crohn's perianal fistula: the Crohn's Anal Fistula Quality of Life (CAF-QoL) scale. *Gut* 2021;**70**:1649–56.
38. de Jong MJ, Huibregtse R, Masclee AAM, Jonkers DMAE, Pierik MJ. Patient-reported outcome measures for use in clinical trials and clinical practice in inflammatory bowel diseases: a systematic review. *Clin Gastroenterol Hepatol* 2018;**16**:648–663.e3.
39. Hüppe A, Langbrandtner J, Lill C, Raspe H. The effectiveness of actively induced medical rehabilitation in chronic inflammatory bowel disease. *Dtsch Arztebl Int* 2020;**117**:89–96.
40. Cooney R, Grimes C, Mathers J, Chapman L, Brown F. Patients know best- How patients designed a crohn's disease study using exclusive enteral nutrition. *Gut* 2022;**71**[Suppl 1]:A145–6.
41. *Developing a test to predict whether patients with inflammatory bowel disease [IBD] will respond positively to biologic drugs [Internet]*. https://arc-sl.nihr.ac.uk/research-and-implementation/our-research-areas/patient-and-public-involvement-research/developing
42. Dames NB, Squire SE, Devlin AB, Fish R, Bisset CN, Tozer P; Respondents to the Sex After Colorectal Surgery Survey. 'Let's talk about sex': a patient-led survey on sexual function after colorectal and pelvic floor surgery. *Colorectal Dis* 2021;**23**:1524–51.
43. Zelinsky S, Daley K, Neary E, *et al*. Improving patient participation in longitudinal research: an innovative patient-led patient-oriented qualitative research project to understand the motivations and barriers to getting and staying involved in the IMAGINE SPOR study. *Inflamm Bowel Dis* 2021;**27**[Supplement_1]:S50.
44. Aiyegbusi OL, Cruz Rivera S, Oliver K, *et al*. The opportunity for greater patient and public involvement and engagement in drug development and regulation. *Nat Rev Drug Discov* 2023;**22**:337–8.
45. de Wit MPT, Berlo SE, Aanerud GJ, *et al*. European League Against Rheumatism recommendations for the inclusion of patient representatives in scientific projects. *Ann Rheum Dis* 2011;**70**:722–6.
46. Wit M de, Abma T, Loon MK van, Collins S, Kirwan J. Involving patient research partners has a significant impact on outcomes research: a responsive evaluation of the international OMERACT conferences. *BMJ Open* 2013;**3**:e002241.
47. de Wit MPT, Koenders MI, Neijland Y, *et al*. Patient involvement in basic rheumatology research at Nijmegen: a three year's responsive evaluation of added value, pitfalls and conditions for success. *BMC Rheumatol* 2022;**6**:66.

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48. Yeoh SA, Burke B, Castelino M, *et al*. Patient and public involvement in rheumatology research: embracing the wave of change. *Lancet Rheumatol* 2021;**3**:e540–2.
49. Heague M, Ray C, Bowers J, Guckian J, Arents BWM, Layton A. Patient and public involvement in dermatology research: a review. *Am J Clin Dermatol* 2022;**23**:319–29.
50. Thomas KS, Bradshaw LE, Sach TH, *et al*. Randomised controlled trial of silk therapeutic garments for the management of atopic eczema in children: the CLOTHES trial. *Health Technol Assess* 2017 ;**21**:1–260.
51. Renz S, Chinnery F, Stuart B, *et al*. Spironolactone for adult female acne (SAFA): protocol for a double-blind, placebo-controlled, phase III randomised study of spironolactone as systemic therapy for acne in adult women. *BMJ Open* 2021;**11**:e053876.
52. Howells LM, Chalmers JR, Gran S, *et al*. Development and initial testing of a new instrument to measure the experience of eczema control in adults and children: Recap of atopic eczema (RECAP). *Br J Dermatol* 2020;**183**:524–36.
53. Thomas KS, Batchelor JM, Akram P, *et al*; UK Dermatology Clinical Trials Network's HI-Light Vitiligo Trial Team. Randomized controlled trial of topical corticosteroid and home-based narrowband ultraviolet B for active and limited vitiligo: results of the HI-Light Vitiligo Trial. *Br J Dermatol* 2021;**184**:828–39.
54. Corrado AM, Benjamin-Thomas TE, McGrath C, Hand C, Laliberte Rudman D. Participatory action research with older adults: a critical interpretive synthesis. *Gerontologist* 2020;**60**:e413–27.
55. Wicks P, Richards T, Denegri S, Godlee F. Patients' roles and rights in research. *BMJ* 2018;**362**:k3193.
56. Abma TA, Nierse CJ, Widdershoven GAM. Patients as partners in responsive research: methodological notions for collaborations in mixed research teams. *Qual Health Res* 2009;**19**:401–15.
57. Mockford C, Staniszewska S, Griffiths F, Herron-Marx S. The impact of patient and public involvement on UK NHS health care: a systematic review. *Int J Qual Health Care* 2012;**24**:28–38. https://pubmed.ncbi.nlm.nih.gov/22109631/.
58. *Evaluating patient and public involvement – NIHR Oxford Health Biomedical Research Centre [Internet]*. https://oxfordhealthbrc.nihr.ac.uk/patient-and-public-involvement/ppi-strategy/evaluating-patient-and-public-involvement/
59. Forsythe L, Ellis L, Edmundson L, *et al*. Patient and stakeholder engagement in the PCORI Pilot Projects: description and lessons learned. *J Gen Intern Med* 2016;**31**:13–21. https://pubmed.ncbi.nlm.nih.gov/26160480/.
60. Siegel CA. Shared decision making in inflammatory bowel disease: helping patients understand the tradeoffs between treatment options. *Gut* 2012;**61**:459–65.

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