Changing priorities for pulmonary fibrosis: the patient will see you now!

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Over the last century, the doctor–patient relationship has gradually evolved from a paternalistic to a more patient-centred approach, with a marked change in the last 20 years. This cultural shift is reflected in health research, where patients and the public have become increasingly engaged and empowered. The UK pioneered these changes, with patient and public involvement becoming, in 2006, a founding principle of the National Institute of Health Research (NIHR). The NIHR aims 'to conduct leading edge research focussing on the needs of patients and the public', with research being undertaken with or by members of the public, rather than on or to them.[1] A basic premise is that involving lay people and bringing in different perspectives and experiences can improve research quality, while also empowering patients to influence change on issues they consider important. There are many different frameworks but one of the most useful is described by NIHR INVOLVE, the English national advisory group on public involvement, shown in table 1.[2]

Some other frameworks consider 'coproduction' as the highest level, which means collaboration of researchers and patients throughout the research process—design, implementation and dissemination. A systematic review of patient and public involvement in Europe found more examples of collaboration and coproduction than consultation, which is encouraging, but the extent of the involvement of lay people is uneven across countries. In northern European countries, patient and public involvement is well developed, while in southern and eastern Europe, it has proved more difficult to translate into practice, due to barriers such as professional resistance, lack of guidelines and best practice examples.[3]

In the UK, an important role is played by the James Lind Alliance (JLA), a nonprofit initiative founded in 2004 and a partner organisation of NIHR since 2013. The JLA aims to bring together patients, carers and healthcare professionals in priority setting partnerships. In a priority setting partnership, stakeholders work jointly and systematically to identify priority topics and to inform funders and researchers of the key questions of people living with the disease. This innovative approach led to successful identification of patient priorities across a range of diseases and has evolved over time in response to several challenges. For example, questions related to basic science and translational medicine were sometimes difficult to address because the patients, carers and clinicians involved lacked necessary specialist knowledge. Also, it occasionally proved difficult to ensure that the patient's voice was heard because patient organisations, rather than actual patients took part or because clinicians dominated. Finally, it has sometimes been difficult to get funders and researchers to engage with the new priorities, because JLA outputs are more likely to be used when they support a project that is already planned.[4] Despite these possible limitations, the JLA remains one of the best examples of patient and public involvement, with over 100 projects completed so far in the UK and also Canada, Europe, Australia and Africa.

In this issue of Thorax, Tikellis *et al* present an interesting study identifying research priorities on pulmonary fibrosis.[5] It is a good example of patient and public involvement. Drawing on JLA experience, the authors designed a study to investigate the unmet needs of people living with pulmonary fibrosis, caregivers and healthcare professionals. It differed from the JLA approach in additionally involving researchers. There was good consensus across the four groups on the main unanswered questions, with an emphasis on the need for effective drugs. The top shared priorities were to reverse the fibrotic process, improve lung function, reduce symptoms, improve well-being, halt disease progression and identify the causes of pulmonary fibrosis and how to prevent it. Subtle differences were noted between stakeholders on the other priorities. Patients highlighted the side effects of antifibrotic therapy and the importance of exercise programmes. Carers were interested in symptom management, improved well-being and speeding up diagnosis by raising awareness of GPs. Healthcare professionals and researchers wanted to explore the causes of acute exacerbation and the chance of improving survival with early diagnosis. Despite the differences between the two healthcare systems, the research priorities identified by the Australian study are similar to those which emerge during discussions with patients in the UK, such as those facilitated by the patient-led organisation Action for Pulmonary Fibrosis. From our experience, people living with pulmonary fibrosis in the UK are mainly concerned with finding a cure for the disease, access to effective medication, delays in diagnosis, lack of reliable information, management of symptoms and access to pulmonary rehabilitation and mental health support. An official JLA project has been launched in late 2020 to systematically assess the unmet pulmonary fibrosis research needs across the UK. The results will be released in early 2022.[6]

Over the last 10 years, research on pulmonary fibrosis has focused mainly on drug development, financed by industry. Of 127 studies registered on

Table 1 Types of patient involvement

Level of patient involvement	Example
Consultation Patients' views inform research.	Pharmaceutical firms' patient advisory groups, where elected patients are consulted on clinical trial design.
Collaboration Patients share in decision-making on research.	National Institute of Health Research (UK) projects, where patients are ideally involved in the design, implementation and dissemination of research as co-researchers.
User-led research Patients, the public, or organisations representing them, actively control, direct and manage the research.	Patient-led research funded by patient organisations. For example, Asthma UK Centre for Applied Research.

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ClinicalTrials.gov, which were recruiting in January 2021, 47 are related to medications and other medical treatments; 36 are national registries or observational studies and the remaining 44 are equally divided into basic science, development of new diagnostic instrument and primarily addressing symptoms and quality of life.[7] Despite intense research efforts, only two antifibrotic molecules have so far been approved, though a number of clinical trials are underway.[8] Antifibrotics can slow down the fibrosis progression and their introduction represented a milestone, but this is not enough. While slowing disease progression is a reasonable medium-term objective, as the study by Tikellis *et al* shows, stakeholders want to find a cure for pulmonary fibrosis. The research community should strive to achieve this ambitious target. On the other hand, it is a shame that even with effective drugs available, patients do not have universal access to them. In fact, some countries have strict prescribing criteria while others lack a public healthcare system. Policymakers should focus on what people most want and do more to guarantee universal access to antifibrotic medications and supportive care.

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Contributors LF and SJ contributed equally to this paper.

Funding The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

Competing interests None declared.

Patient consent for publication Not required.

Provenance and peer review Commissioned; externally peer reviewed.

To cite Fabbri L, Jones S. *Thorax* Epub ahead of print: [*please include* Day Month Year]. doi:10.1136/thoraxjnl-2020-216616

Accepted 10 March 2021

Linked

► http://dx.doi.org/10.1136/thoraxjnl-2020-215731

Thorax 2021;**0**:1–2. doi:10.1136/thoraxjnl-2020-216616

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