

## **Outcome measures in research and in practice: have you ever thought how these are developed?**

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How clinicians measure the health status of their patients is extremely important. We must be able to determine if treatment is effective and helping patients. Outcome measures, usually in the form of questionnaires, are becoming part of routine care, but have you ever thought about how these were developed in the first place?

Here at WWL it is my pleasure to tell the story of how our patients have helped to develop a measure which has taken many years and a huge amount of hard work and dedication. It truly isn't an exercise to be undertaken by the faint hearted.

WWL have an established Patient Research Advisory group, [WWL Teaching Hospitals NHS Foundation Trust | Patient Research Advisory Group](#), and to cut a long story short, my clinical and research carer, had links which allowed myself and our Patient and Public Involvement (PPI) group to become an integral part of the research team who have helped to develop an outcome measure.

The focus of the research was to answer the question: so, what is it like to live with fatigue when you have axSpA (formerly known as Ankylosing Spondylitis) and how can we measure it? As a research team we applied for research funding and had two failed bids before we were successful. Two of our PPI group members became Patient Partners and had extra tasks and responsibilities in advising on the grant applications and on the way that the research would be conducted. The research was led by my close colleague, Dr Kirstie Haywood, at Warwick University and for anyone who has connections with the research world, it is this collaborative working that leads to success! As a result, we were able to employ a Research Associate (Nathan Pearson) who delivered the research and achieved his PhD.

The research process begins when you check what is out there already. As expected, Nathan's review of outcome measures for fatigue showed that the evidence was both incomplete and limited.

We had no confidence that the current questionnaires could capture *what* matters and there was only a single question in the assessment toolkit in both research and clinical practice for axSpA.

Nathan began his research by asking the PPI group: 'So what is fatigue and what does it mean to you?' We helped him to develop what is known as an 'interview schedule' so that the questions that he asked our participants would cover all aspects that we needed to hear about and were written in a suitable way for correct interpretation. He conducted interviews with 17 patients in the north, midlands and south of England.

When all the interviews had been analysed, we then had to check that we had captured everything and that nothing was missing. This process was completed by holding four group meetings: two with patients and two with healthcare professionals. The hard work continued when the fatigue outcome measure, which was informed by the 17 semi-structured interviews, was developed. Our PPI involvement, further interviews and focus groups were held with patients and clinicians from WWL and University Hospitals of North Midlands NHS Trust leading to the development of the Warwick Axial Spondyloarthritis faTigue and Energy Description scale (WASTEd) questionnaire.

The first version had 30 questions and formed the first Axial SpA fatigue and energy-specific questionnaire which had two sections: Fatigue and Energy. There then followed two rounds

of five pre-testing interviews conducted with patients. This was important because we had to check that the questionnaire would be clear, understandable, and measured what is important.

To test this, a further study took place which asked as many participants as possible throughout the country to complete the questionnaire. WWL were one of the recruitment sites and within a three-month recruitment window, 106 patients from WWL gave their permission for the questionnaire to be sent to them. We had a remarkable response rate of 89% and became the top recruiter in the country.

Our research is the first to illustrate the importance of energy as a related, but distinct concept in axSpA-fatigue experience. As a research team we feel that this distinction is likely to have important implications for patient self-management and may identify unmet needs which include service provision, service evaluation and has the potential for individualised care.

Being involved in this research as a clinician and an academic has been so impactful. It has allowed me to contribute on a national level by working with the main charity for the condition National Axial Spondyloarthritis Society (NASS) who helped to fund the research, to rewrite their Fatigue literature and to help to share our findings to the rheumatology community. Research can be so powerful when you consider that people who took part in the study told us that 'making sense of fatigue is difficult and hard to talk about: "it's the closest thing to pain that does not hurt"'.

The last hurdle and probably the hardest to overcome is to finalise the testing of the WASTEd questionnaire, and then supporting its use in clinical practice. We plan to work with patients to refine and test the questionnaire, to ensure that it really helps patients in telling clinicians about how fatigue is affecting them. This knowledge is important to help clinicians to understand the impact of fatigue, introduce treatments and measure how effective this can be. To allow for easier completion of the questionnaire, we plan to develop an electronic questionnaire (or ePROM) that can be completed using an online system supported by the British Society of Rheumatology ePROMs platform. This will be accessible via laptop or computer.

As a result of collaborative working – researchers, clinicians, and our Patient Partners - we have successfully secured funding from the National Institute of Health Research, Research for Patient Benefit (RfPB) scheme to support the final stage of evaluation and adoption into practice. Moreover, two publications from the research are in press and under review, respectively: the first, "Living with axial spondyloarthritis and fatigue: difficult, demanding and draining" has been accepted for publication in British Medical Journal Open; the second, "Development and pretesting of the Axial Spondyloarthritis faTigue and Energy questionnaire (WASTEd) – a new patient-reported outcome measure" is under review with the Rheumatology (Oxford) Journal.

We are entering the sixth year of our collaboration and I hope that sharing our journey has helped you to see how outcome measures are developed – it's through hard work, perseverance, patience and, perhaps most importantly, continued collaborative engagement with patients and clinicians. A strength of our endeavour has been the long-term commitment of our amazing WWL Patient Partners and members of the study's Research Advisory Group, in addition to the many patients participated in the various stages of the study. I cannot thank them enough for their involvement and it is truly humbling to have been a part of such a successful partnership.

## **Sharing the thoughts of our Patient Partner, Mrs Jean Thompson**

I was a member of the Patient Research Advisory Group when Dr Kirstie Haywood from Warwick University first came to discuss her proposed research with us. She particularly wanted to conduct research into the impact of fatigue on the lives of patients with axSpA. This meeting left a significant impression on me because it was the first time that I'd heard fatigue discussed as a major issue, as opposed to an occasional side-effect of the condition.

I had first been diagnosed with Ankylosing Spondylitis over 35 years previously (although early symptoms had begun to develop almost ten years prior to that). Pain and stiffness and ways to alleviate them had been discussed at length, but little mention, if any, was made of fatigue. In my experience, the pain was initially my primary concern until gradually it started to recede, and I was left with the stiffness and a sense of frustration caused by the struggle I often experienced when undertaking a normal day's activities. I now recognise this struggle was being caused by fatigue, which has affected me for much of my adult life.

It was explained that to deliver the project a full-time Research Associate would need to be employed and that funding would be required for the project to proceed. I had been asked if I would be willing to be a Patient Partner and was very excited and proud to be involved in this way. I remember the disappointment when two bids for funding were rejected, but also the elation when funding from NASS was obtained and we knew the research could proceed.

Our PPI group has had several meetings with Nathan, the Research Associate (a doctoral student) appointed to the task, over the course of the project. The first meeting was very wide-ranging in scope as we described our experiences of fatigue, which often varied from person-to-person. I watched Nathan taking copious notes and I remember thinking what a huge task he was facing. At a later meeting he shared the results of the interviews he had conducted with 17 patients, and it was striking how familiar many of the responses were, as we compared them with our own experiences.

After much hard work and doggedness on Nathan's part, a questionnaire was developed and refined, and we were asked for our thoughts on whether the questions were clear and understandable and covered all the things we thought were important. No doubt further refinement will take place before the questionnaire is implemented in practice. However, the benefits achieved so far, in opening up this subject for discussion and creating much greater awareness and understanding of its significance, will be much welcomed by patients.

I hope to continue to be involved as a Patient Partner in the future development of the WASTed and look forward to the time when it can be used in practice to help patients and clinicians to measure the impact of fatigue and treat it accordingly. If it can help patients to lead more fulfilling and productive lives that would be a wonderful achievement. I have thoroughly enjoyed being involved in this research and want to place on record my admiration and respect for the members of the research team who committed so much to ensure the success achieved so far, most particularly Nathan, Kirstie and Jane.