

Recovery from CRPS. How do patients define recovery, and what would be their treatment priorities to achieve recovery?

Deciding when someone has recovered from Complex Regional Pain Syndrome (CRPS) can be difficult due to a number of different factors. Firstly, the common signs and symptoms of CRPS change over time. This can mean that although someone met the Budapest Diagnostic criteria for CRPS at one time point, they may not meet it at a later date. However, the patient may still experience some of the symptoms of CRPS or have some long-term disability as a result of having CRPS, even though they no longer meet all the diagnostic criteria. Are these people recovered from CRPS or not?

Secondly, it is common for people who live with a long-term condition to adjust the way that they think about their health status over time. As the day to day signs and symptoms of their condition become 'normal' to them they may start to consider that, although they are not back to their pre-CRPS self, they are actually much better than they were in the early stage of the condition, and therefore think of themselves as 'better'. Are these people recovered or not?

If we do not know what 'recovered' looks like in CRPS then it is very difficult to decide if treatments are effective or not, and to also know what the natural course of CRPS is.

In 2013 RSDSA and the Dutch National CRPS Patient Organization provided funding for an international team of academics, clinicians and people with CRPS to conduct research to answer the questions:

- What defines recovery from CRPS from the patients' perspective?
- What are the priorities for recovery from the patients' perspective?

What did we do?

The research team invited people to participate in the study if they had been diagnosed with CRPS and were either already enrolled on a country specific CRPS database, or were attending a CRPS service in one of the 10 participating centres. These were located across 8 different countries -USA, Canada (2 sites), UK, Netherlands, Germany (2 sites), Denmark, Switzerland and Poland.

Once people had agreed to be involved in the study they received two packs of questionnaires in the post, over approximately a one year period. Both packs included questionnaires that asked them to tell the research team about different aspects of their health. For example: their pain, level of function, and psychological well-being. In addition, the first pack of questionnaires asked them to answer one of two questions:

1. 'I **would** consider myself recovered from CRPS if...' or
2. 'I **do** consider myself recovered from CRPS because...'

The participants wrote their answers to one of the above questions in a free text box that allowed up to about 100 words. These responses were then analysed by members of the research team, including some of the research patient partners to make sure that we had the patients' viewpoint. In the analysis the responses were grouped under themes that described the data within them. For example, some of the themes were 'carrying out daily routine', 'self-care', and 'CRPS symptoms'.

The themes that were reported most commonly by the participants were then formed into 62 different statements. These statements were sent out with the second questionnaire pack to all of the people who had responded to the first round questionnaire. Participants were asked to select the 10 items they felt were most relevant to their view of recovery and to rank these items in order of importance.

What did we find?

347 people participated in the first round of questionnaires, and 252 of these people also completed the second round of questionnaires. In the first round, 80.4% of participants were women; the average age of all participants was 53yrs; more than half of the participants had lived with CRPS for 3yrs or more. Of the 310 people who told us their recovery status, 280 (90.3%) considered themselves to be “not recovered”.

From the first questionnaire pack, the most frequently reported themes for patient-defined recovery were: activities of daily living; bodily functions (including CRPS symptoms and pain); external factors (including medication use) and participation (e.g. housework, shopping). Personal factors (anxiety, depression) were least represented.

From the second round of questionnaires, our participants told us that their top five priorities for recovery were-

To no longer have:

1. CRPS-related pain,
2. Generalised pain and discomfort
3. Restricted range of movement
4. Need for medication
5. Stiffness in the affected limb.

The top three statements of most subgroups (males, females, recovered, non-recovered, age groups 30-50 and 50+, paid employment, non-employed, all disease durations, upper and lower limb CRPS) were all within the overall top five priorities.

From the questionnaire that asked about different aspects of participants' health, the data showed that people with CRPS were more likely to report themselves as recovered if they had: fewer symptoms, knew their type of CRPS, and had caring responsibilities.

There was no relationship found between self-reported recovery and demographic factors (age, gender etc), disease duration, disease trigger, or which limb was affected.

People who did not consider themselves recovered from CRPS reported higher pain, lower quality of life and had more difficulty with coping psychologically with their pain, than those who were recovered.

Those people with lower-limb CRPS more frequently reported increased pain responses and pain to normally non-painful stimuli, than those with upper-limb CRPS. They also more frequently reported hair changes, and involuntary muscle movements and greater overall pain, poorer quality of life, lower levels of physical functioning and poorer energy/fatigue scores.

Function, psychological flexibility, and number of symptoms were found to be predictive of quality of life for non-recovered participants with upper-limb CRPS.

What can we conclude from this?

The data from this study suggests a very small number of themes are of highest importance to people with CRPS in their definition of recovery and these vary little across age groups, gender, nationality, employed or unemployed groups. People want their CRPS-related pain, generalised pain, movement difficulties, and medication reliance to be addressed, above all other factors, for them to consider themselves recovered.

Persistent CRPS has negative consequences for mental and physical well-being, and the poorest health outcomes were found for people with lower-limb CRPS.

How will this information help people with CRPS?

The conduct of this study and the new information gained from it has:

- Helped us to know which particular areas people with CRPS want clinicians and researchers to focus on when developing new treatments and designing future research studies
- Given us new information about the impact of CRPS in different populations and an insight into how people with CRPS define recovery when they have, or have not still got symptoms.
- Helped us to select questionnaires that ask about these areas that are important to people with CRPS when we have been designing a new set of questionnaires to be used in *all* future CRPS clinical trials. Having the same set of questionnaires that can be used across all countries will mean we can conduct much larger, and therefore more effective research studies and, hopefully, move ahead more quickly in helping people with CRPS.
- Given us an opportunity to translate a large number of questionnaires into different languages, which will make our future international studies much quicker to establish.
- Built a new CRPS international research community that includes patient researchers, and has demonstrated we can conduct research, that is important to people with CRPS, in an efficient and effective manner.

Acknowledgements and thanks

This study would not have been possible without the generous financial support from the Reflex Sympathetic Dystrophy Syndrome Association, and the Dutch National CRPS Patient Organization. We would like to thank our funders and the patients who kindly participated in this study.

Who were the research team?

McCabe CS^{1,2}, Hibberd Y¹, Llewellyn A^{1,2}, White P², Davies L¹ Marinus J³, Perez RSGM⁴, Thomassen, I⁵, Brunner, F⁶, Sontheim C⁶, Birklein F⁷, Goebel A⁸, Haigh R⁹, Connett R⁹, Maihöfner C¹⁰, Knudsen L¹¹, Harden N¹², Zyluk A¹³, Shulman D¹⁴, Small H¹⁵, Gobeil F¹⁶, Moskovitz P¹⁷

¹ Royal United Hospitals Bath, UK (lead centre)

² University of the West of England, Bristol, UK

³ Leiden University Medical Centre, Leiden, The Netherlands

⁴ VU University Medical Centre, Amsterdam, The Netherlands

⁵ CRPS Patient Society, The Netherlands

⁶ Balgrist University Hospital, Zurich, Switzerland

⁷ University Medical Centre Mainz, Mainz, Germany

⁸ The Walton Centre, Liverpool, UK

⁹ Royal Devon & Exeter Hospital, Exeter, UK

¹⁰ Klinikum Fürth, Fürth, Germany

¹¹ Danish Pain Research Center, Aarhus University Hospital, Aarhus, Denmark

¹² Rehabilitation Institute of Chicago, USA

¹³ Pomeranian Medical University, Szczecin, Poland

¹⁴ Markham-Stouffville Hospital, Markham, Canada

¹⁵ PARC (Promoting Awareness of RSD and CRPS in Canada)

¹⁶ CSSS Pierre Boucher, Longueuil, Canada

¹⁷ The George Washington University Hospital, Washington DC, USA

C McCabe was funded by a National Institute for Health Research Career Development fellowship. This paper presents independent research funded by the National Institute for Health Research (NIHR). The views expressed are those of the authors and not necessarily those of the NHS, the NIHR or the Department of Health.