Title: Complex Regional Pain Syndrome: An international survey of clinical practice

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Significance

- Health professionals expressed some level of difficulty recognising the signs and symptoms of CRPS despite the majority of health professionals having had clinical experience exceeding six years in the field of CRPS.
- More work is required to raise awareness amongst clinicians of the Budapest CRPS diagnostic criteria so as to promote early diagnosis and intervention
- Health professionals' treatment aims reflected the current clinical guidelines however, a lack of resources and fragmented care were frequently cited as barriers to achieving these.

Abstract

Background

Published guidelines promote best practice in Complex Regional Pain Syndrome (CRPS) treatment and management; however, these recommendations are not always applied in clinical practice. Understanding existing care internationally will help inform future patient and health professional service delivery, education initiatives, and content of clinical guidelines.

Methods

An e-survey was conducted in order to gain an insight into routine CRPS clinical practice. Health professionals and academics, from the field of CRPS, were recruited from an international population. Quantitative and qualitative data were elicited. Data were mapped onto a framework to identify macro-regional factors.

Results

Of the 260 survey respondents, 96% (n=241) provided clinical care for people with CRPS, with academics not involved in patient care also responding. Half of respondents expressed difficulty in recognising the symptoms of CRPS but treatment aims corresponded with published guidelines. However, a lack of resources and fragmented care were reported as barriers to early intervention. Service constraints were most frequently reported by European respondents. Five themes emerged from the qualitative data: the benefit of inter-disciplinary working; the importance of symptom management; need for early diagnosis and intervention; establishing a collaborative partnership with patients; the value of education for patients and health professionals.

Conclusions

Our data suggests that more work is required to raise awareness of the Budapest CRPS diagnostic criteria so as to promote early diagnosis and intervention. Future work to optimise clinical effectiveness should consider enhancing inter-disciplinary service delivery that encourages a collaborative patient/clinician partnership; includes excellent patient education; and addresses modifiable patient-related factors.

1. Introduction

Complex Regional Pain Syndrome (CRPS) is a distressing, persistent pain condition with unknown aetiology that occurs in a limb predominantly after trauma. Features include unremitting pain, changes to limb temperature, colour, nail and hair growth, and impaired limb function. CRPS has a significant impact on health related quality of life. Half of people with CRPS report anxiety or depression and over 90% experience difficulty in performing usual activities (Kemler & de Vet., 2000). There is no cure but early intervention should significantly improve outcomes (Birklein et al.,2015).

There are a number of published country-specific and generic treatment guidelines, which promote best practice in CRPS treatment and management (Goebel A, Barker CH, Turner-Stokes L et al., 2018; Harden et al., 2013; Perez et al., 2014.). In addition, a CRPS European Pain Federation Task Force has recently published the European standards for the diagnosis and management of CRPS (Goebel et al., 2019). However, research indicates that clinical guidelines are often not adopted (Fischer et al ., 2016), with many factors influencing implementation, including those related to the accessibility of the guideline and the awareness and attitudes of potential users (Fischer et al., 2016; Franke et al., 2008). We know, from our own clinical experience and that of clinical colleagues, that CRPS clinical practice varies widely, at a local, national and international level. Indeed the European Task Force was convened to find ways to address this anecdotal variation in care, but we do not currently have a good understanding of what existing care actually comprises. Furthermore, in order to promote the implementation of CRPS guidelines, there needs to be an understanding of the current barriers to achieving guideline recommendations. Establishing what current CRPS clinical practice looks like internationally will help inform future patient and health professional education initiatives, content of clinical guidelines, and future service design.

To gain an insight into routine CRPS clinical practice and better understand what care is being delivered globally, we conducted an international e-survey of clinical practice. Previous surveys investigating CRPS clinical practice have focused primarily on identifying current interventions used within specialist groups, such as health practitioners involved in rehabilitation (Miller et al., 2017), American interventional pain specialists (Burton et al., 2004), and delegates at a British Hand Therapists annual conference (Ramsey., 2008). CRPS

clinical practice has not been investigated more extensively in relation to the characteristics of the health professionals providing the care, the care pathway, and treatment aims. Specifically, we wished to understand how care was being provided including access to resources and expertise, to establish the aims of treatment as defined by survey respondents, and to identify the barriers and facilitators they perceive may impact on achieving these treatment goals. Through identification of the barriers, and using geographical data to detect macro-regional variation, we wished to highlight areas which may be targeted to improve service provision. This is the first survey to be undertaken in such a diverse population of health professionals, academics and researchers, with the aim to better understand the delivery of care.

2. Methods

2.1 Survey design

An e-survey was designed with survey content informed by published United Kingdom (UK) treatment guidelines (Goebel A, Barker CH, Turner-Stokes L et al., 2012), our own experience of clinical practice as a team delivering the National NHS England CRPS service, and anecdotal reports of variations in practice from the international clinical community, including from the International Association for the Study of Pain (IASP) Special Interest Group for CRPS, and CRPSUK Network (http://www.crpsnetworkuk.org). Quantitative and qualitative data were elicited.

The e-survey comprised a total of 32 questions. Twenty one questions captured the professional characteristics and geographical location of the respondents and their experience of the provision of CRPS care via polar questions (yes/no), multiple choice and free text questions (Table 1). Five further qualitative questions, specifically for those respondents involved in the clinical care of patients with CRPS, explored the focus of the treatments they provided for this patient group, including the barriers and facilitators to achieving their treatment aims (Table 2). In addition, six questions were asked about CRPS research practice as part of a separate nested project, which are not reported here. Throughout the survey, respondents were directed to subsequent questions dependent on their previous response. This was to ensure that the questions were applicable to participants' roles. What constituted persistent CRPS was not defined within the survey. No

distinction was made between CRPS I and II as this does not have an implication on clinical care.

INSERT TABLE 1 HERE

INSERT TABLE 2 HERE

The survey was created using the Qualtrics Insight Platform. This was piloted and refined with clinical and academic colleagues. Revisions were made following feedback, and included revising the wording of questions and the option to navigate in a forward and backward direction. As the survey was to be distributed to an international audience, the readability was confirmed by three colleagues whose first language was not English.

2.2 Ethical approvals and funding

Ethical approval was granted by the Research Ethics Committee at the Faculty of Health and Applied Sciences, University of the West of England (UWE REC REF number: HAS 16.07.185). Higher Education Innovation Funding was received from the University of the West of England. Participants were required to indicate their consent to participate in the survey prior to being able to view the questionnaire. All responses were anonymous.

2.3 Recruitment

The survey recruited health professionals, academics and researchers from across an international population. Survey dissemination methods included:

i) Advertisements on the web pages of clinical networks with an embedded link to the survey

ii) Tweeting a survey link to professional special interest groups

iii) Advertisements on the webpages of professional bodies, alerting members to the survey

iv) Snowballing; where the link might be sent to those who had an interest in CRPS

v) Disseminating of survey information by professional bodies to their members, with a reminder issued after two weeks.

Recruitment, via health professional national/international organisations, including the International Association for the Study of Pain (IASP) Special Interest Group for CRPS (see Appendix S1), was targeted at those with a current interest in CRPS, either clinically or through research. Respondents were not identified by professional discipline.

The survey was open during September 1st - October 31st 2016.

2.4. Analysis

All data were stored within the Qualtrics website. Data reports were exported for analysis. Not all individuals completed all questions in the survey, however, the responses they did provide were included in the analysis.

2.4.1 Quantitative analysis

Data were analysed to identify the characteristics of the respondents, their experience of working with CRPS patients and their local CRPS service provision. Data analysis was conducted by three researchers (VG, LJ, SG) and frequency counts and percentages were used to describe and summarise the data using Microsoft Excel. Results were calculated as a percentage of the number of respondents answering each question except for Fig.3 and Fig. 4; where referral destinations were calculated as a percentage of total number of responses to the question, as respondents could list as many referral destinations as they wished. Free text responses to the qualitative, open ended questions were categorised in Microsoft Excel by LJ and agreed with SG and AL. Unintelligible answers were excluded from the analysis. For a small proportion of respondents, we noted that data were missing from a number of questions however, as there was no pattern to this, we retained all cases within our analysis and have reported data as provided. A frequency count of the reported barriers to achieving the treatment aims was elicited from the qualitative data. Fischer et al's (2016) framework was applied, to identify and categorise barriers to clinical guideline adoption.

2.4.2 Qualitative analysis

Applying inductive thematic analysis, the data were analysed by three researchers (SM, AL & SG) who were blinded to the country of origin of each response. For the small proportion of answers that were not in English, Google translate (https://translate.google.com/) was used to understand the meaning of the response. Non-identifiable abbreviations were excluded from the data. Each question was read and re-read by a first researcher, in order to gain familiarisation with the data and initial ideas for codes or themes were noted (Braun and Clarke., 2006). The researcher then coded the response to each question in NVIVO 10 or 11 using words or short phrases which captured their meaning. The codes were sorted into potential themes and data extracts relevant to each theme collated. A second researcher

independently coded the data for each question by hand. For each question, a collaborative approach was adopted where two or more researchers discussed and shared insights into the emergent themes, reviewing the collated extracts that sat within each theme and confirming their appropriateness. This gave an opportunity to discuss any codes which lacked clarity and to develop a shared understanding across the team of data analysts. The value of having more than a single coder is contested within qualitative research (Braun and Clark., 2013); however, the researchers had different epistemologies (SG-clinical nurse researcher; AL- research psychologist; SM-qualitative health researcher), and drawing on these different perspectives maximised the contribution of differing knowledge, interests and approach when interpreting the data (Green and Thorogood., 2011). Finally, the themes per question were considered across the qualitative dataset and supra- themes were agreed by all three researchers, defined and named.

3. Results

We firstly present the quantitative data, followed by the findings of the qualitative analysis.

3.1 Quantitative data

The number of people who responded to each question is reported in Table 3.

3.1.1 Characteristics of respondents

Complete or partially complete surveys were received from n=260 health professionals or academics working in 35 different countries, across six continents (Figure 1). 254 respondents reported the country in which they worked, with the highest number of returns from Europe (126/254, 50%), Australasia and Oceania (41/254, 16%) and North America (47/254, 19%). Highest country-specific returns were from (Canada (32/254, 13%), the United Kingdom (UK) (30/254, 12%) and New Zealand (27/254, 11%).

INSERT FIGURE 1 HERE

The majority of respondents were clinicians providing patient care (193/252, 77%), with clinical academics who provided patient care (48/252, 19%) and academics not involved with patient care (11/252, 4%) also responding (Table 4). Respondents worked within

hospitals (164/257, 64%), community health organisations (85/257, 33%) and academic institutions (49/257, 19%).

INSERT TABLE 4 HERE

A range of clinical specialties were represented with respondents working most frequently in rehabilitation (119/251, 47%), orthopaedic (104/251, 41%), and pain (101/251, 40%) services (Figure 2). Half of the respondents to this question (125/251, 50%) worked in more than one specialism.

INSERT FIGURE 2 HERE

Respondents were asked to list any CRPS related diagnostic criteria of which they were aware. The Budapest criteria (Harden et al., 2010) were most frequently cited (112/166, 67%), followed by criteria from the International Association for the Study of Pain (IASP) (26/166, 16%). Other responses included criteria from professional societies and international guidelines (17/166, 10%) and many respondents listed a range of signs and symptoms of CRPS as diagnostic criteria (29/166, 17%). Three respondents (3/166, 2%) stated they were not aware of any diagnostic criteria. A large number of the total survey respondents did not respond to this question (94/260, 36%).

3.1.2 Respondents' experience of working with CRPS patients (Table 5)

The duration of experience of clinicians providing care to CRPS patients ranged from 0-5 years (49/170, 29%) to > 20 years (23/170, 14%). Respondents had most commonly seen between one and five CRPS patients over the previous 12 months (99/213, 46%) with 23/213 (11%) reporting to have seen >20 CRPS patients. Half of respondents (107/216, 50%) reported difficulty recognising CRPS signs and symptoms, with 21% of these (22/216) reporting at least some or much difficulty.

INSERT TABLE 5 HERE

3.1.3 CRPS service provision: at the first clinical consultation

An overview of CRPS service provision at the first clinical consultation with the respondent (clinician or clinical academic) is summarised in Table 6

INSERT TABLE 6 HERE

At the first clinical consultation, 90% (191/212) of respondents reported that, on average, the patient's signs and symptoms were present for less than a year. Patients typically waited 1-4 weeks for the onward referral appointment (109/180, 61%), however, longer waits of 4-6 months (16/180, 9%) and 7-9 months (3/180, 2%) were also reported.

INSERT FIGURE 3 HERE

Over half of patients were referred on to other professions/services after their first clinical consultation (109/180, 55%). Figure 3 displays the professions and services to which they were referred. The most common professions or services for referral were pain services (96/380, 25%), psychological services (83/380, 22%) and physiotherapy (55/380, 14%) (Figure 3).

3.1.4 CRPS service provision: for patients with persistent symptoms

INSERT TABLE 7 HERE

As shown in Table 7, referral to other services or professions due to patients' persistent symptoms was reported by 77% (153/198) of respondents. Respondents reported that the majority of onward referrals were seen within 0-3 months (134/179, 75%). The most common reasons reported for referring a patient with persistent symptoms were the patient's pain (136/173, 79%), function (135/173, 78%) or psychological well-being (133/173, 77%).

INSERT FIGURE 4 HERE

Figure 4 displays the professions and services to whom respondents reported they refer patients with persistent CRPS symptoms. The most common professions or services to be referred to for persistent symptoms were pain services (84/280, 30%), psychological services (36/280, 13%) and physiotherapy (29/280, 10%) (Figure 4). Also of note, 9/280 (3%) of respondents referred patients to a specialist CRPS service.

3.1.5 Mapping to the Fischer et al (2016) framework: barriers in clinical guideline implementation

In the Fischer et al (2016) model, barriers to guideline implementation are categorised into personal factors, guide-line related factors and external factors. Table 8 shows how our data mapped onto this framework and the variations in reported macro-regional barriers. In response to the question asking what made it difficult to achieve the aims they had outlined, a lack of resources was cited by some respondents. This included limited consultation time and therapy resources, limited access to specialist review and psychological input, and service constraints. Service constraints were most frequently reported by health professionals in Europe (n=27), and not cited by those working in Asia or Central and South America. A lack of awareness of CRPS by health professionals was reported by respondents from Europe (n=12), North America (n=7) and Australasia and Oceania (n=7). A few respondents from Europe (n=3) reported that a lack of collaboration presented difficulties with achieving their aims.

INSERT TABLE 8 HERE

3.2 Qualitative data

182/260 people responded to the qualitative questions. Five over-arching themes emerged from the data. Only those involved in CRPS clinical care responded to the qualitative questions and indicative quotes are represented verbatim.

1. The benefit of inter-disciplinary team working

Inter- and multi-disciplinary working was repeatedly reported by health professionals as key to achieving their treatment aims. The terms were perceived to be used interchangeably. Responses included: '*We have a very good interdisciplinary work environment*' (Denmark); *'Multi-disciplinary team approach, both to diagnosis and management*' (UK).

Inter-team communication appears fundamental to this collaborative approach, in particular the importance of being able to access CRPS expertise, both from colleagues and through onward referral: *'Working in a large team with support for second opinions,*

discussion of treatment plans' (UK); *'Consultation with more experienced colleagues'* (Canada).

One respondent identified '*working alone and not with a team'* (Israel) as a barrier to achieving their aims and another recognised the challenges that maintaining dialogue between colleagues can present, stating that a ' *Team approach is vital...but this can also have its challenges due to effort/time required for communication'* (New Zealand).
A system that supports an integrative approach helped respondents to meet their aims: *'A health system that allows a personalized and integrated attention to an interdisciplinary team'* (translated- Colombia).

2. The importance of symptom management

Symptom management and the subsidiary theme of patient self-management are addressed within this theme.

Treating the specific signs and symptoms of CRPS were among the most common aims across the dataset. Within this, the treatment of pain (pain reduction/pain management) and, for a few, pain resolution, was by far the most frequently cited aim. Pain was also seen as the greatest barrier to achieving the treatment aims: *'the resistant nature of the pain*' (Australia). Delays in treatment were reported while the patient accessed appropriate pain relief: *'Sometimes patients have to wait to see a specialist and receive the right pain medication'* (UK). There was recognition that there was a lack of consistency in the individuals' response to pain relief and that to develop a plan for appropriate pain relief could take time : *'The response to analgesic drugs is inconsistent'* (Brazil); '*Cannot predict the response of the treatment provided*' (Thailand) .

Respondents also frequently cited the improvement, maintenance or restoration of function as an important treatment aim, including: *'improve everyday function'* (Israel)*;* '*to alleviate pain, encourage use of the affected limb and return to function*' (Kenya). The importance of normalising limb function was frequently reported: '*to establish a productive cycle of use, exercise and functional gains that will progress the patient back to normal use of the affected limb and re-establish functional roles' (*Canada). For many respondents improving function and managing, or improving pain were not reported as mutually exclusive

objectives, and aims included: *'Pain reduction and functional improvement*' (Australia) *and 'To manage pain to support participation in rehabilitation' (*Canada).

Patient self-management

Providing the patient with strategies to manage their pain was seen as a key role of the clinical team and this included a continuation of rehabilitation into the home environment: *'Providing a therapy package/ home programme that the patient can adopt to manage the pain'* (UK). A treatment aim was *'to help them improve their management of CRPS'* (New Zealand). This was reported to be facilitated by those patients who engaged with self-management: *'Self motivation from the client. Willingness to improve function or health. Client taking responsibility for their own health'* (South Africa).

3. Need for early diagnosis and intervention

The early diagnosis of CRPS was frequently cited as facilitating the aims of treatment: *'Early recognition of the signs and symptoms and coordinated management from medical and rehabilitation perspectives'* (Canada); '*When the patient is treated within the first month after the symptoms has started'* (South Africa). However, lack of agreement between health professionals made it difficult to achieve the aim; '*Lots of different diagnoses from other professionals*' (UK).

Initiating treatment early was also recognised as an important aim in CRPS, however, a delay in making a referral to a CRPS specialist or Pain Clinic was reported as a significant barrier to achieving treatment aims, '*The patients usually are referred too long after the symptoms has started'* (South Africa) and then confounded by an additional delay in attending the appointment. Several respondents suggested that early intervention had implications on the outcome of CRPS and its treatment, and this could *'reverse it if early enough or maintain passive range of motion and maximize function'* (Canada). However, a lack of resources was reported as a barrier to achieving early intervention. Predominately this focused on inadequate time for therapy or clinical consultation, limited access to therapies and support, but also included the consequence of limited resources from the patient's perspective: *'patient's access to services is often challenged (e.g. transport costs from rural areas to attend outpatient treatment)'*(South Africa).

Frustration with the inadequate resources available within health care systems was apparent and barriers to achieving treatment aims included: *'Lack of time'* (Germany) and *'Fragmentation of care'* (UK). Psychology support was reported by some as not readily available: *'Lack of good pain psychologists'* (Germany).
It was suggested that a *'streamlined access to services'* (Australia) would facilitate the achievement of treatment aims.

4. Establishing a collaborative partnership with patients

Establishing a collaborative partnership with patients was evidenced throughout the responses as an important aspect of care and essential to maintaining a therapeutic relationship: *'Positive therapeutic relationship is key to success' (*Canada)**.**

When asked about other aspects of patient care that are important, the benefit of a therapeutic relationship was cited frequently. Establishing trust appears to be fundamental to this: *'Trying to establish an atmosphere of trust with the patient'* (Argentina). One response illustrated the difficulties of maintaining this relationship when the treatment outcomes are slow to manifest; '*Patients cannot have long-term follow-up at my clinic. This is because they would go to other doctor after unsatisfied short-term treatment response*' (Taiwan).

In addition to health professionals requiring clinical skills, respondents identified the value of higher level attributes in meeting the needs of people with CRPS: *'Deep compassion and patience needed'* (South Africa); *'compassion, understanding, empowering the patient'* (New Zealand). Respondents described the importance of patients having time to tell their story: *'Open honest conversation that is not rushed to allow them to tell their story and express their needs'* (Canada). The need for managing expectations was also described as an important aspect of care: *'Realistic expectations of the patient and provider'* (USA) and a positive, therapeutic environment was also considered important: *'I believe they really need time and comfortable space to be, and the clinic can give that to them'* (Israel).

However, also noted within the data was the frequency of reference to patient compliance with rehabilitation and treatment. Patient compliance was reported as both a barrier and facilitator to achieve treatment aims: *'If the patient do not comply with the simplest of*

indication I give' (Uruguay); *'Patient noncompliance is, by far, the most common impediment to treatment success'* (USA), however, *'Patient compliance through understanding'* (New Zealand) helped respondents achieve their treatment aims. Much of the responsibility was directed at the patient highlighting '*attitude', 'commitment', and 'drive'* and *'motivation' as* key to achieving treatment aims. It is suggested that compliance is achieved through the patient understanding CRPS and the mechanisms behind it.

5. The value of education

This theme encompasses the education of health professionals and patients. These will be addressed as subsidiary themes.

Educating the patient

Respondents reported that they wanted to give their individual patients an understanding of the mechanisms of CRPS and the rationale for treatment approaches, so that they were more likely to engage with therapies and self-manage their condition effectively; *'Good education from day one!!!! A person will only benefit from management modalities such as graded motor imagery or graded exposure if they are on board with the treatment they are engaging in'* (New Zealand); *'As with all chronic non-specific pain, a treatment plan will usually only be successful if it involves some combination of scientifically credible patient education'* (USA).

Respondents reported their belief that the responsibility to educate lay firmly with the health professional working directly with the patient, although there was some reference to sign-posting to online materials which were considered to be from a reputable source: *'Having good patient information - it unifies the language and key messages used amongst therapists as well as giving the patient something to take away'* (UK).

'Poor understanding of his (sic) condition by the patient' (Canada) and those unable to accept CRPS as a diagnosis, were identified as presenting a barrier to treatment success. In particular; *'The patient's lack of understanding (of) the seriousness of the condition'* (Denmark) was considered a barrier to achieving treatment aims.

Several respondents commented that it was important to be aware that increased time was needed to meet the needs of people with CRPS, in addition to that required for other conditions suggesting that they may be a different kind of patient: *'Time consuming patients'* (Germany); *'To realize that the CRPS patients need different approach'* (Norway).

Education of Health Professionals

The need for a greater awareness of CRPS was frequently reported and one respondent commented that: '*among health professionals CRPS does not seem to be recognised as a condition to all parties'* (Ireland). Lack of knowledge and lack of understanding were some of the most frequently cited reasons for not achieving the treatment aims; by other health professionals, in the general community and by the patient.

The complexity of the condition was reported as providing health professionals with the challenge of imparting difficult concepts in an accessible way and this requires a higher level of skills: *'The complex nature of CRPS and the relation between the brain and pain - and how to explain this to patients in a way that makes it easy to understand'* (UK).

The importance of consistency between health professionals resulting in the *'Patient receiving conflicting information from different professionals/internet'* (Ireland) was described as a barrier to achieving the treatment aims. In addition, frustration was expressed in relation to '*The mountain of mis-information available on the internet and propagated by clinicians who also are not current'* (Canada). Ways in which educational opportunities were sought included networking, peer support and accessing CRPS-focused education via IASP and local meetings.

Discussion

It is apparent from this survey of clinical practice that the clinical care of those with CRPS was reported as being provided by health professionals from a range of specialism's, with the majority working in rehabilitation, orthopaedics and pain. This is not surprising for a condition which manifests in the limb as unremitting pain, sensory impairment and reduced function. Although data were not collected on the number of individuals who received an invitation to complete the survey, the number of completed responses was high in comparison to a recent, methodologically similar, survey of CRPS health professionals

(Miller et al., 2017). This may be as a result of the professional diversity of those approached to participate. Similarly, whilst recruitment targeted to those with a current interest in CRPS, anyone with access to the survey was able to respond. As we expected, higher returns were from countries where English is the first language however, a breadth of countries was represented across many continents. The geographical spread of responses, weighted heavily in favour of Europe (50%), with 16% to include Central and South America, Africa and Asia.

Half of respondents expressed some level of difficulty recognising the signs and symptoms of CRPS, with 21% of these reporting at least some or much difficulty. This is despite the majority of clinicians having had clinical experience of CRPS exceeding six years. Whilst this may appear surprising, this finding may reflect the infrequency with which our respondents were presented with the condition, as nearly half reported seeing 5 people, or fewer, in the past 12 months. In contrast, respondents to a previous survey of US pain specialists reported treating, on average, fifteen CRPS patients a month (Burton et al., 2004). Based on the European incidence rate of CRPS (de Mos et al., 2007), in the UK around 17,000 people will experience CRPS each year and many of these may not receive a formal diagnosis. In real terms, this means many clinicians, even those working in rehabilitation, orthopaedics and pain will rarely see a patient with CRPS and this is particularly true of those working outside pain or musculoskeletal specialties. This may contribute to the reports within our data of 'a [personal] lack of knowledge' being a barrier to achieving the treatment aims.

In line with international guidelines (Goebel A, Barker CH, Turner-Stokes L et al., 2018; Harden et al., 2013; Perez et al., 2014) and best practice, our qualitative data evidenced a consensus of opinion that early diagnosis and treatment is crucial. A recent study demonstrated that, for many symptoms, improvements were most likely in the first 6 months after onset and therefore suggested early intervention at this stage may prevent long term disability (Bean et al., 2016). Healthcare systems have many competing demands. Despite being a relatively rare condition, CRPS is associated with significant economic consequences (Kemler et al., 2010; Scholz-Odermatt et al., 2019). This survey provides support for healthcare resources to be directed towards the early diagnosis and intervention of CRPS. This is further supported by a recent retrospective analysis conducted in a Swiss post-accident population, which reported that average treatment costs, after an

accident resulting in CRPS, were 13 times higher, and the number of days lost at work were 20 times higher, than those in patients without CPRS (Scholz-Odermatt et al., 2019).

At initial presentation, 90% of our survey respondents reported that, on average, the patient's signs and symptoms were present for less than a year. These figures indicate that, for the patients seen by our respondents at least, the majority are reviewed early in their CRPS care-pathway. This is an encouraging finding as previous studies have demonstrated that a delay, of months or years, in obtaining a diagnosis is commonly experienced (de Mos et al., 2007; Lewis et al., 2007; Lunden et al., 2016, Shenker et al.,2015). It is possible, however, that our survey was completed by those who are familiar with CRPS and therefore were more able to recognise it early in the disease course; 50% of respondents reported no difficulty recognising CRPS. In contrast to our quantitative data, our qualitative data offered contradictory findings. Comments provided by some respondents suggested a delay in clinical review, a lack of resources and the fragmentation of services present barriers to early intervention. These factors are consistent with the key barriers to clinical guideline implementation reported in the framework by Fischer et al. (2016). It is likely that clinical care provision and access to therapeutic interventions are influenced by the availability of local resources, based on the location and health care system. The data indicated that, for some, a lack of resources or lack of awareness of CRPS by health professionals was a significant barrier to guideline implementation. It is however possible that these respondents were more willing to share this information or had higher expectations of service provision. Many factors were cited including inadequate clinical consultation time with no capacity to focus on patient education, limited therapy resources and lengthy waits for referral appointments.

Similar to the findings of another recent survey (Miller et al., 2017), two-thirds of our respondents reported being aware of the widely recognised Budapest diagnostic criteria (Harden et al., 2010). As these criteria have been published and utilised for nearly a decade, we had anticipated a larger proportion would have cited these. However, many of those citing the "IASP criteria" did not specify if this included the Budapest criteria, the 1994 IASP criteria (Stanton-Hicks et al., 1995) or country-specific criteria (Sumitani et al., 2010). Some respondents reported using a range of signs and symptoms of CRPS as a diagnostic tool but did not identify formal diagnostic criteria. A limitation of these findings is the reliance on

respondents being able to recall the specific diagnostic criteria they used. However, a decision was made not to provide a list of criteria options for selection, as it was considered this risked 'priming' responses, thereby leading respondents to indicate awareness simply because they recognised a name. Recent Standards state that the recognised and well-established Budapest criteria must be used for a European population (Goebel et al., 2019). Failure to do so risks inappropriate management of CRPS (Mailis-Gagnon et al., 2014). This may result in misdiagnosis of CRPS, which could have long term consequences for the patient in relation to receiving appropriate and targeted treatments. This also presents a challenge when reporting the care pathway for CRPS internationally as there may be inconsistency in diagnoses. UK, USA and European recommendations for the diagnosis and management of CRPS, all advocate the use of the Budapest critieria. Continued efforts to disseminate these standards will help to raise awareness and encourage their adoption (Goebel et al 2019; Goebel A, Barker CH, Turner-Stokes L et al., 2018; Harden et al., 2013; Perez et al., 2014).

Reported treatment aims in this survey population are consistent with current international CRPS clinical guidelines: pain management, functional improvement, psychological support and patient education. Moreover, our qualitative data demonstrated that an inter-disciplinary team approach was recognised by clinicians as fundamental to the achievement of these treatment goals. The multifactorial nature of CRPS necessitates input from a range of health disciplines, and this was reported in practice by our respondents.

The qualitative data presented insight into the benefits of inter-disciplinary working in the field of CRPS. Respondents highlighted the importance of being able to discuss treatment plans, seek second opinions, and consult with more experienced colleagues, which may not be afforded to those working in isolation. The complex nature of CRPS and its multifactorial presentation may instigate this more readily than in other pain and non-pain health conditions. It is conceivable that the increased use of digital technology as a means to accessing CRPS expertise and interdisciplinary support could provide wider access to clinical guidance for health professionals in low resource settings (Orton et al., 2018).

Although a collaborative relationship between patients and healthcare professionals was reported as fundamental to achieving the respondents' treatment aims, the data indicated a more paternalistic approach. Respondents reported that 'patient compliance' influences the

likelihood of achieving the treatment aims, perhaps suggesting that shared decision making and patient-centred care is not adopted by all. A move towards patient–professional concordance, with a more equal relationship between health professional and patient has been shown to be beneficial (Treharne et al., 2006). Respondents emphasised the importance of their patients having an understanding of the mechanisms underlying CRPS, and the potential seriousness of the condition, in order to fully engage with treatment. Balancing optimism with an uncertainty of outcome is a particular challenge for health professionals, in a condition where the prognostic factors are currently not defined (Wertli et al., 2013).

There are limitations of this survey. It was only available in English and this will have limited the responses by non-English speaking participants. Where free text was required, this may have also deterred those non-fluent in English. Future research would be helpful to replicate the survey in multiple languages. Although we achieved a broad geographical representation, responses from some continents were low and therefore, we were unable to draw firm conclusions based on the location of respondents. This provides an opportunity for future research to resolve this geographical gap.

It is possible that those respondents, who had been engaged in CRPS clinical practice for long periods of time, and more confident in their abilities, were more likely to respond. Those less familiar with CRPS may have been deterred. Data were not collected on respondents' professional background and therefore responses could not be attributed to this.

It is acknowledged that respondents may have interpreted some of the questions differently from what was intended. For example, it was evident that some respondents listed treatment modalities in response to a question about what CRPS information was useful in everyday practice, which is not what we anticipated. The survey was designed from the perspective of UK researchers and their experience of UK healthcare and, inadvertently, assumptions may have been made when formatting the questions.

Conclusion

This international survey provides new insights into routine clinical practice and the challenges faced by those working in the field of CRPS. Raising international awareness of the widely recognised Budapest diagnostic criteria (Harden el al., 2010) may have an impact on ensuring patients receive an early diagnosis and timely treatment. Although the treatment aims often reflected the current published clinical guidelines, perceived service delivery limitations, a reported lack of resources and patient related factors have the potential to reduce clinical effectiveness and therefore may be target areas for future interventions. Future work should consider macro-regional variations to optimise clinical effectiveness.

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Author contributions

All authors made substantial contributions either to the study design and methodology, data acquisition, or data analysis and interpretation. All authors discussed the study findings, have been consulted in the drafting of the final article, and have given their approval for publication.

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