

BMJ Open Quality of life, functioning and participation of adult patients with an amputation following complex regional pain syndrome I or brachial plexus injury: a scoping review protocol

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ABSTRACT

Introduction This planned scoping review aims to provide insight into current literature regarding perceived quality of life (QoL), functioning and participation of patients with upper limb amputations (ULA) because of therapy-resistant debilitating complex regional pain syndrome type I (CRPS-I) or brachial plexus injury (BPI). It is important to gain insight into these outcomes, so we can properly inform and select patients eligible for amputation.

Methods and analysis Joanna Briggs Institute methodology for scoping reviews, Systematic Reviews and Meta-Analyses Scoping Reviews guidelines and Arksey and O'Malley's framework will be used. Studies regarding adult patients with either BPI or CRPS-I who underwent ULA will be considered for inclusion. Studies should include one or more of the following topics: QoL, functioning or participation and should be written in English, German or Dutch. Searches will be conducted in the Cochrane database, PubMed, EMBASE and Google Scholar. Search strings will be provided by a licenced librarian. All relevant literatures will be considered for inclusion, regardless of published date, in order to give a full scope of available literature. Studies will be selected first by title, then abstract and finally by full article by two reviewers who will discuss after every round. A third reviewer will make final decisions to reach consensus if needed. Data will be presented as brief summaries and in tables using a modified data extraction table.

Ethics and dissemination No ethical approval is required since no original data will be collected. Results will be disseminated through publication in a peer-reviewed journal and presentations at (inter)national conferences.

INTRODUCTION

Complex regional pain syndrome type I (CRPS-I) is a pain syndrome with debilitating pain and loss of function in a limb, for which a limited number of treatment options exist.^{1–4} Not all patients recover from CRPS-I after treatment, and for therapy-resistant cases, amputation might be the only remaining treatment option.^{5–7} Similarly, some patients

STRENGTHS AND LIMITATIONS OF THIS STUDY

- ⇒ The quality of our scoping review will be assured by adhering to the guidelines provided by the Joanna Briggs Institute, Preferred Reporting Items for Systematic Reviews and Meta-Analyses Scoping Reviews reporting guidelines and using Arksey and O'Malley's framework.
- ⇒ Using search algorithms provided by a certified medical librarian will contribute to methodological quality.
- ⇒ The search algorithms cover three major databases as well as Google Scholar.
- ⇒ Dutch, German and English articles are considered for this review, meaning that there is a possibility of excluding potentially interesting articles written in different languages.
- ⇒ There are limited studies available concerning amputation after complex regional pain syndrome type I or brachial plexus injury, and the quality of these studies varies widely.

with a complete brachial plexus injury (BPI) can experience therapy-resistant severe pain due to traction on the glenohumeral joint or on the neck and upper back because of the flail arm's weight. Another important reason is hindrance of their afunctional arm. Amputation can be considered a last resort treatment for these patients as well.^{8,9} To our knowledge, no data regarding incidence of amputation in these patient categories are available.

Since amputation is an irreversible and last resort treatment, it is important to select those patients who will benefit from this treatment, and to properly inform them about the implications of amputation. Can patients expect a higher quality of life (QoL), will their functionality and participation improve? Literature answers some of these questions, but

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a clear overview of current literature is, to our knowledge, absent.^{10–14} To properly inform clinicians who treat patients considering amputation, an overview of current literature is needed, so that they can select and inform patients eligible for amputation better.

A preliminary search in the Cochrane database, PubMed, EMBASE, Open Science Framework (OSF), The International Prospective Register of Systematic Reviews (PROSPERO) and Google Scholar showed no current or planned reviews (either systematic or scoping) regarding this topic. There is one systematic review by Ayyaswamy *et al*¹² discussing QoL after amputation in patients with advanced CRPS-I.¹² A combined scoping review of amputation because of either CRPS-I or BPI has, to our knowledge, not been conducted yet. Furthermore, Ayyaswamy's review only includes studies up until 2017, and patients with CRPS-I who were diagnosed following the standard diagnostic CRPS-I criteria. Only QoL outcomes that were reported using descriptive analyses and/or standard tools were included. Our intended scoping review will complement what can be learnt from the review of Ayyaswamy *et al*, as we will look at both quantitative and qualitative reports of QoL, functionality and participation for both CRPS-I and BPI. Furthermore, we will also look at synonyms for CRPS-I (such as Sudeck's dystrophy). We will consider all articles that mention either BPI or CRPS-I and amputation concerning QoL, functioning and/or participation, regardless of publication date. Finally, our goal is not to provide the reader with a summarised set of overarching statistics, but rather an overview of what is currently written about QoL, functioning and participation following amputation due to CRPS-I or BPI.

Our rationale for combining BPI and CRPS-I is that despite the differences in cause and (types of) pain, both diseases share that in some therapy-resistant cases, amputation is considered a last resort treatment and is always performed as an elective surgery. Furthermore, both diseases share a peripheral origin, meaning that (not taking comorbidities into account) both patients with BPI and CRPS-I have a clear view of their life before and after their amputation with regard to QoL, functioning and participation. Finally, both CRPS-I and BPI have a great impact on the functionality of an upper limb.

METHODS AND ANALYSIS

The proposed scoping review will be conducted in accordance with the Joanna Briggs Institute (JBI) methodology for scoping reviews and reported as per Systematic Reviews and Meta-Analyses Scoping Reviews (PRISMA-ScR).^{15 16} Furthermore, we will follow the steps as described in Arksey & O'Malley's framework.¹⁷ Finally, we have registered the scoping review on OSF under 'Quality of life, functioning and participation of patients with an amputation following Complex Regional Pain Syndrome I or Brachial Plexus Injury; A scoping review' (registration number: <https://doi.org/10.17605/OSF.IO/JMBGK>).

Inclusion criteria

Studies have included adults with either a BPI (including obstetric BPI) or therapy-resistant CRPS-I who underwent amputation. Patients should be 18 years or older. For CRPS-I specifically, we will also include studies that use (older) synonyms for CRPS-I, such as post-traumatic dystrophy and Sudeck's dystrophy. All studies that describe either QoL, functioning and/or participation will be considered for inclusion. Studies published in either English, German or Dutch will be included. All published articles that are available at the time of conducting our search will be considered. Since the goal of this scoping review is to provide the reader with a summary of all available studies regarding this topic, we felt no earlier date limit should be set.

Exclusion criteria

Studies about limb amputation due to other diagnoses than CRPS-I or BPI (acquired or by birth) and studies that do not mention either QoL, functioning and/or participation after amputation, studies that have no full text in English, German or Dutch will not be taken into account.

Concept

The WHO defines QoL as 'an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns'.¹⁸ Indicators that qualify QoL are employment status, mental and emotional health, physical health, education, freedom, recreation and leisure and the ability to perform activities of daily living. The International Classification of Functioning, Disability and Health (ICF) defines functioning as: 'an umbrella term for body function, body structures, activities and participation. It denotes the positive or neutral aspects of the interaction between a person's health condition(s) and that individual's contextual factors (environmental and personal factors)'.¹⁹ The ICF defines participation as involvement in a life situation.¹⁹ We will include all studies that discuss or measure (aspects of) the above definitions. This includes, but is not limited to, questionnaires, qualitative studies, experts' opinions and case reports, as per Arksey and O'Malley's framework.¹⁷

Context

All studies will be included if they are written in either English, German or Dutch. Since we are aware that culture and perhaps even geographic location may play a role in perceived QoL and possibilities of participating in society, we will also include articles that are not from 'western countries' as they will provide valuable insight as to what encompasses perceived QoL in these cultures and/or countries.

Types of sources

This scoping review will consider both experimental and quasi-experimental study designs, including randomised controlled trials, non-randomised controlled trials, before

and after studies and interrupted time-series studies. In addition, analytical observational studies including prospective and retrospective cohort studies, case-control studies and analytical cross-sectional studies will be considered for inclusion. This review will also consider descriptive observational study designs including case series, individual case reports and descriptive cross-sectional studies for inclusion.

Studies that focus on qualitative data will also be considered including, but not limited to, designs such as phenomenology, grounded theory, ethnography, qualitative description, action research and feminist research.

In addition, systematic reviews that meet the inclusion criteria will also be considered, depending on the research question; we will use the relevant original articles from these reviews.

Text and opinion papers will also be considered for inclusion in this scoping review.

Search strategy

First a search on PROSPERO and OSF was carried out, which confirmed that no similar or identical reviews are currently being conducted. Subsequently, an initial limited search of the Cochrane database, PubMed, EMBASE and Google Scholar was undertaken to identify articles on the topic. The text words contained in the titles and abstracts of relevant articles, key words and the index terms used to describe the articles were employed to develop a full search strategy in the aforementioned databases (see online supplemental appendix 1). The search strings, including all identified keywords and index terms, will be adapted for each included database and/or information source. The reference list of all included studies will be screened for additional eligible studies.

To assess whether we would be missing valuable articles, we used the search string provided by our certified librarian in PubMed both with and without exclusively including articles that were written in English, German or Dutch. Including all languages, a total of 590 articles came up (search conducted on 16 November 2023). Excluding English, German and Dutch articles, 54 articles came up, none of which has titles or abstracts that seem relevant to the topic of our review.

Study/source of evidence selection

Following the search, all identified citations will be collected and uploaded into Mendeley Reference Manager (Elsevier, Mendeley Desktop version V.1.19.8). Data will then be transferred to Rayyan for removal of duplicates and to be able to make notes of included articles.²⁰ Titles and abstracts will then be screened by two independent reviewers for assessment against the review's inclusion criteria. An initial selection round will be conducted on title screening, followed by a second selection round after thorough reading of the abstract and finally a third selection round will take place after reading the entire article. After every selection round, the two reviewers will discuss any differences in

Box 1 Data extraction instrument

- ⇒ Author, year
- ⇒ Country
- ⇒ Aim
- ⇒ Study type/source
- ⇒ Population (age, inclusion criteria)
- ⇒ Sample size (total *n* and per group if applicable)
- ⇒ Country in which the study has been conducted
- ⇒ Gender
- ⇒ Other relevant demographics: level of amputation, side of amputation, years postamputation, prosthesis use, education, work, comorbidities
- ⇒ Complex regional pain syndrome type I or brachial plexus injury (or both)
- ⇒ Outcome measures (how did the author measure the outcomes; questionnaires, etc)
- ⇒ Results (statistical evidence, *p* values, CIs, effect sizes)
- ⇒ Strong points of this study
- ⇒ Weak points of this study

included articles. Reasons for exclusion of studies after full-text reading will be recorded and reported in the scoping review. Any disagreements that arise between the reviewers at each stage of the selection process will be resolved through discussion, or with an additional reviewer to reach consensus. The results of the search and the study inclusion process will be reported in full in the final scoping review and presented in a PRISMA-ScR flow diagram.¹⁵

Data extraction, analysis and presentation

Relevant data from the selected papers will be extracted using a data extraction form we have developed specifically for this scoping review, following the template provided by the JBI and the article by Pollock *et al* (box 1).^{20 21} This form has been discussed within the research group before finalisation. During the data extraction process conducted by two independent reviewers, this form will be modified and revised if necessary, and modifications will be mentioned in detail in the scoping review. Should disagreements occur between the reviewers regarding any modifications, a third reviewer will be asked to help resolve this and guide the reviewers to consensus. Relevant data will include details regarding type of study, year in which the study has been conducted, country in which the study has been conducted, number of patients included, relevant inclusion and exclusion criteria, primary and secondary outcome(s) and strong and weak (deemed as such by the reviewers) points of each study. This data will be presented both in a table, and as part of an overview of the articles in plain text. We will report data from CRPS-I and BPI studies separately in the Tables and in the Results section, so that the readers of our manuscript will be able to judge the outcomes of the limb amputations separately for both disorders. If any relevant data are missing from the article, the reviewers

will try to acquire this data by contacting the authors of the papers. As this is a scoping review, with the goal to inform what is currently known in literature, no critical appraisal of individual articles shall be conducted.

Patient and public involvement

No patients were involved in making this study protocol, nor will they be involved in making the actual review.

ETHICS AND DISSEMINATION

No ethics approval is required since no original data will be collected. Results will be disseminated through publication in a peer-reviewed journal and presentations will be given at (inter)national conferences.

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