

# An investigation into self-reported foot and lower limb problems associated with systemic lupus erythematosus (SLE): A research proposal

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## Abstract

Systemic lupus erythematosus (SLE) can manifest in the lower limb with joint and muscle pains and in severe cases, disorganisation of the joints and tendon rupture. In the foot and lower limb, Raynaud's phenomenon and other circulatory problems have been reported and may be associated with loss of sensation or altered pain perception. Associated with impaired peripheral neurovascular function are changes in tissue viability leading to either callus formation or thinning of the skin and ulceration. Many medications used to manage SLE can impact on the person's resistance to infections and consequently fungal, bacterial or viral infections can spread rapidly and persist. Although there is some evidence for this range of problems occurring in the foot, it is not known how prevalent they are, what interaction there is between problems, and how these impact upon a person's health related quality of life (HRQoL). Thus, the main aim of this study is to determine the self-reported foot and lower limb problems experienced by people with SLE. A secondary aim is to explore the impact that the identified foot and lower complications may have upon patients with SLE. It is anticipated that this research will highlight areas of potential health care need and thus can be used to inform recommendations about foot health care for this patient group and the focus of future research.

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## Introduction

Systemic lupus erythematosus (SLE) is a complex, chronic, multi-system autoimmune disease which varies in prevalence and incidence depending on ethnicity and is diagnosed about nine times more frequently in women than men (1, 2). In the UK approximately 25,000 people have a diagnosis of SLE (3). The disease is highly heterogenous in that it varies in manifestations and severity between individuals. Disfigurement caused by the involvement of skin and joints (4) can affect the patient's perception of body image and sexuality (5), can have an impact on emotional health (6) and overall quality of life (7).

Involvement of the musculoskeletal system is common during the clinical course of SLE in up to 95% of patients, with joint pain being the first presenting symptom in up to 50% of cases (8). The extra articular manifestations of SLE include soft tissue pathology such as capsular swelling, synovial hypertrophy (thickening of joint lining) and tenosynovitis (9) which can lead to tendon rupture (10) or tendon contracture (11). Tendons are the force transmitting units of the musculoskeletal system, but due

to their low metabolic rate and slow healing, injury can result in considerable morbidity and prolonged disability. In relation to the symptoms associated with muscle involvement, these can range in severity from mild aches in up to 80% of cases to painful inflammatory myositis in up to 11% of cases (8).

An additional factor that has the potential to contribute to a lower health related quality of life (HRQoL) in this patient group is vascular involvement. Vascular involvement can be either a direct complication of SLE or develop as co-morbidity. Vascular involvement represents the most frequent cause of death in patients with SLE (12). Mathieu et al (13) identified that abnormal vascular reactivity and coagulopathy both contribute to an increased risk of atheroma and therefore recommend careful monitoring for any vascular change with the aim of preventing tissue necrosis and ultimately amputation or death. Thus regular review of lower limb vascular health may be a key factor in maintaining health related quality of life.

A recent narrative review (14) identified that there are some indications that SLE affects foot and lower limb morbidity and

that these have the potential to impact upon HRQoL. However, the scale of these problems is unclear and little research in this area has been completed to date. It could be speculated that the altered peripheral neurovascular and tissue health experienced by patient with SLE renders their feet particularly susceptible to pain, deformity, poor function, ulceration or infection. Further, the provision of foot health care for patients with SLE is inconsistent across the UK, and examination of the feet may be omitted from routine clinical examination (15). As such, the impact of such foot and lower limb problems, or the care thereof, upon HRQoL in this patient group also remains unclear to date.

One of the obstacles to achieving consistent and high quality foot and lower limb health review is the lack of a 'gold standard' model for assessment in this patient group. Arguably, early identification of foot and lower limb problems, would allow appropriate interventions to be provided in a 'window of opportunity' which may be similar to that experienced by patients with rheumatoid arthritis (16). However, before 'gold standard' foot assessment can be achieved, there is a need to determine the specific problems experienced by patients with SLE, how these may interact, and how they may affect HRQoL. An improved understanding of the range and impact of foot or lower limb problems experienced by patients with SLE could be used to inform recommendations regarding the type, frequency and nature of foot health care required to maintain HRQoL and provide the information to develop a future research strategy in this area.

## Research aim and objectives

The main aim of this study is to determine the self-reported foot and lower limb problems experienced by people with systemic lupus erythematosus. A secondary aim is to explore the impact that the identified foot and lower limb problems may have upon patients with SLE. The study objectives are therefore to:

1. Develop a survey questionnaire through a focus group of purposively sampled participants
2. Use the developed survey to determine the foot and lower limb problems

experienced by patients with SLE .

3. Explore the impact of reported problems upon health related quality of life through a series of conversational style patient interviews

4. Use the above objectives to inform recommendations regarding the type, frequency and nature of foot health care required to maintain health related quality of life for patients with SLE

## Proposed methodology

### Study design

The study utilises a mixed method design with quantitative survey data (stage one) and qualitative patient interview (stage two). Participants will be prospectively, purposively selected for both stages. Research ethical approval for the study has been sought and all activity will be conducted in accordance with the declaration of Helsinki guidelines for research practice (1975).

### Patient engagement in the study advisory group

Two patients with SLE will be identified from a rheumatology outpatient clinic as volunteers to join the study advisory group. A participant information sheet will be provided prior to their inclusion in the advisory group and consent obtained. The advisory group will comprise the 2 patients, the principal investigator, the co-investigator, the research associate, and two consultant rheumatologists. Patient advisors will contribute feedback in relation to questionnaire design, participant recruitment and involvement, survey completion rates, potential interview questions, and results dissemination.

## STAGE ONE: Survey of patient experience

### Survey development

Survey development will take the form of a focus group with the aim of generating an item pool to develop the questionnaire. The focus group will review existing literature on foot problems associated with Lupus, information from other foot health related questionnaires such as The Manchester pain and disability questionnaire (17), a SLE specific health related quality of life measure ('LupusQoL') (18, 19), a disease activity measure ('BILAG-2004') (19), a damage measure ('SLICC-ACR-Damage Index')

(20) and consider the experiences of patients and practitioners involved in foot and lower limb health care. From this consensus approach to item generation, the survey questionnaire will be formulated based on the agreed themes, categories, question format and overall structure (21). The survey will be checked for face and content validity prior to cognitive debriefing in order to ensure usability, understanding of the process of completing the survey and understanding of the questions, prior to its launch (21).

### Survey sample size

Audit data from the proposed clinical sites suggests that approximately 500 patients may be eligible for participation; given a 50% response rate, a sample size of approximately 250 participants is feasible. This sample size and response rate is in-line with similar surveys of this kind.

### Survey completion

Patients will be recruited from University Hospital Southampton NHS Foundation Trust, Southampton; The Royal Bournemouth and Christchurch hospitals NHS Foundation Trust, Bournemouth; Royal Blackburn Hospital, Lancashire; Chapel Allerton Hospital, Leeds; Manchester Royal Infirmary and Salford Royal Hospitals Foundation Trust, Salford. Patients will be invited by their consultant/specialist nurse to consider completing the questionnaire. Posters will be displayed in the waiting areas of each respective clinic to publicise the survey and provide an overview of the aims and objectives of the study. Patients will be reassured by their consultant/specialist nurse that a decision to take part in the survey or not, will have no impact on their ongoing care provision. Patients will be included who are: diagnosed with definite SLE according to ACR criteria (22, 23), aged 18 years or older and are able and willing to participate.

## STAGE TWO: Patient interviews

### Participant interviews

Potential participants for this stage (n=12) will be identified as a purposive sample by their consultant or specialist nurse. The sample size is pragmatically derived and in accordance with similar published work of this kind. Patients will be included who: are diagnosed with

definite SLE according to ACR criteria (22, 23), have self-reported foot or lower limb problems, are aged 18 years or above and are willing and able to participate in an interview. Conversational style interviews are proposed using an underpinning interpretivistic, phenomenological approach to both data collection and analysis (24). The interviews will be digitally recorded and complemented by field notes. It is intended for the researcher to only ask trigger questions for all participants and then dependent on responses, further questions will be asked (24). An opening question will be used for all participants; “tell me about your experiences of having lower limb and foot problems?”

## Data Analysis

### Survey Data Analysis

All participants completing the survey will be identified by the location of the centre and participant project number and all forms will be identified by the number only. Once all data forms collected from questionnaire stage of the study have been entered into SPSS the original forms will be destroyed. The descriptive data obtained from the survey will be analysed using SPSS software. Continuous data will be summarised by means ( $\pm$ standard deviations), frequency counts and percentages. The Student's t-test, analysis of variance (with appropriate post hoc tests), Chi square and correlation coefficients will be used to test for significant associations between lower limb/foot problems and reported symptoms; disease duration and reported foot problems, with significance set at the  $p < 0.05\%$  confidence level. All participants will be described in terms of gender, age, disease duration and current medical management.

### Interview Data Analysis

Data will be transcribed verbatim and analysed using a thematic framework approach (25) by the research team. Themes that have emerged from the analysis of the qualitative data will be supported with exemplars from the transcripts in order to support the truthfulness of the data and illuminate the themes. Also the results will be analysed by a second researcher in order to add to the credibility of the analyses. Further the results will be viewed by the participants to confirm a true reflection

of what they meant and to ensure correct interpretation of the dialogue.

The results of both stages will be discussed by the advisory group and a consensus achieved on the conclusions, recommendations for practice and for future research.

## Intended outcomes and benefits for clinical practice

It is anticipated that this research project will lead to new knowledge about the type, frequency and impact that foot and lower limb problems may have upon HRQoL for patients with systemic lupus erythematosus (SLE). An improved understanding of the range and impact of foot or lower limb problems experienced by patients with SLE could be used to inform recommendations regarding the frequency and nature of foot health care required to maintain HRQoL. Specifically, the results of the study will be used to:

- Influence the consultation practice of health professionals involved in the management of patients with SLE. This will be implemented through clinical forums such as the NW Rheumatology Club meetings, British health Professionals in Rheumatology, Arthritis Research UK, and through the Arthritis and Musculoskeletal Alliance standards committee.
- Develop guidelines for the assessment and management of foot problems through the Podiatric Rheumatology Clinical Effectiveness Group network
- Influence the undergraduate and postgraduate training of practitioners involved in the assessment and provision of foot care to patients with SLE

Findings from this study will also be disseminated to patients via various patient groups (e.g. Lupus UK) and as advised by the study advisory group. Overall, it is anticipated that the findings of this study will inform clinical guidelines in this area of practice thereby potentially improving the consistency and quality of foot health care for this patient group.

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