

# Research letter

## Future research priorities for lichen sclerosis – results of a James Lind Alliance Priority Setting Partnership

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DEAR EDITOR, Lichen sclerosis (LS) is a chronic, inflammatory genital skin condition affecting men, women and children. Long-term complications include loss of normal anatomy from scarring, and malignant transformation. Uncertainties exist about the cause, diagnosis and management of LS. For example, the aetiology is contested; evidence suggests that chronic irritation by occluded urine is important in males;<sup>1</sup> however, autoimmune mechanisms are proposed in females.<sup>2</sup> Genetic associations are suggested in both sexes.<sup>2</sup> Existing evidence on which to make recommendations about management and the prevention of malignancy is generally poor quality.<sup>2–4</sup>

We conducted a Priority Setting Partnership (PSP) from June 2017 to July 2018 using James Lind Alliance (JLA) methodology.<sup>5</sup> The aim was to identify future research priorities about the causes, diagnosis, management and prevention of LS in men, women and children. This research letter details the key findings; a full detailed report is available in the public domain.<sup>6</sup>

The protocol was written prospectively and made publicly available.<sup>6</sup> The steering group, chaired by a JLA independent adviser, included seven patient representatives (five patients, one parent of a child with LS, one patient support organization representative), seven health professionals (five dermatologists, one gynaecologist and one nurse) and three researchers/administrators. From September to October 2017 we conducted the first of two online surveys using SurveyMonkey™. These surveys, available in English, were promoted by partner organizations and social media to reach patient and health professional

stakeholders internationally (see Acknowledgments), as well as posters in outpatient clinics.

The first survey gathered patients' and health professionals' views. Participants submitted up to five questions that they had about LS. This survey was supplemented by evidence gaps identified from three key systematic reviews and guidelines in the literature.<sup>2–4</sup>

There were 653 respondents from 28 countries. Of these, 64% were patients (92% females, 5% parents) and 35% were health professionals (29% gynaecologists, 26% dermatologists, 16% sexual health physicians, 10% general practitioners, 9% urologists). Of 2580 submitted questions, 660 were removed as they were out of scope, illegible/unintelligible, or too ill-defined. The remainder were grouped and refined, to produce 38 'unique uncertainties' that reflected the overarching themes of the individual submissions and had not already been answered by previous research. Input from all members of the Steering Group ensured that questions were worded to be understood by the general public.

An 'interim prioritization' online survey (March–April 2018) was completed by 954 participants, with the same patient/health professional ratios as in survey 1 (except for proportionally higher gynaecology respondents), from 35 countries. Participants were asked to select their 10 most important questions from the 38 uncertainties identified during stage 1. Uncertainties were presented to individuals in a random order to minimize selection bias.

The top-rated uncertainties overall, plus the most important uncertainties from each of the stakeholder groups (health professionals, females, males and children) were selected to form a shortlist of 23 questions.

These 23 questions were discussed at a face-to-face workshop (London, June 2018) involving three independent JLA

**Table 1** Results of the Lichen Sclerosis Priority Setting Partnership final workshop: summary of the Top 10 uncertainties to be addressed by future research (full wording available in the detailed report<sup>6</sup>)

- 1 What is the best way to prevent and manage anatomical changes caused by lichen sclerosis?
- 2 What is the best way to diagnose lichen sclerosis (diagnostic criteria)?
- 3 What surgical treatments should be offered for lichen sclerosis?
- 4 Are there effective topical treatments other than topical steroids in the treatment of lichen sclerosis?
- 5 What is the risk of developing cancer in patients with lichen sclerosis?
- 6 Which aspects of lichen sclerosis should be measured to assess response to treatment?
- 7 Can lichen sclerosis be prevented from occurring and what are the trigger factors?
- 8 Is it necessary to continue treatment for patients with lichen sclerosis who do not have any symptoms and/or signs of disease activity?
- 9 What is the impact on quality of life?
- 10 Does the disease course of lichen sclerosis differ in boys and girls, adult males and females?

facilitators, 14 patients (10 female, two male, two parents) and 15 health professionals. Using nominal group technique, after two rounds of small-group discussions and a final whole-group discussion, consensus on the 'Top 10' priorities was agreed by all workshop participants (Table 1). There was agreement for further merging and rewording of some of the questions which reduced the shortlist of questions from 23 to 20. Increasing knowledge and awareness of LS was felt to be a key overarching theme for any future research in this field.

The strength of this project is the global reach, large number of respondents and transparent methodology.

The main challenge was in ensuring adequate representation of male patients. Possible reasons are twofold. Firstly, males are less willing than women to engage with health-related surveys.<sup>7</sup> Secondly, LS treatment in males differs from women as circumcision potentially offers a cure. Therefore, the proportion of males with 'unanswered questions' about LS may be far fewer than women. Similarly, representation of children with LS was low, possibly because LS is less common in children and may go undiagnosed. As a result, the Steering Group took care to ensure that the uncertainties were worded to be applicable to all patient groups (where possible), and workshop participants representing men and children were specifically asked to confirm that the final Top 10 reflected priorities relevant to them.

Results of this PSP will now be widely publicized. Due to the high level of stakeholder engagement, these Top 10 questions will be attractive to both researchers and funders, leading to future studies that will inform clinical practice and minimize research waste.<sup>8</sup>

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<sup>1</sup>Centre of Evidence Based Dermatology,  
University of Nottingham, King's Meadow  
Campus, Lenton Lane, Nottingham NG7  
2NR, U.K.

<sup>2</sup>Department of Dermatology, Oxford  
University Hospitals, Oxford, U.K.

<sup>3</sup>University of Marburg, Germany

<sup>4</sup>Patient representative

<sup>5</sup>Rotherham NHS Foundation Trust,  
Rotherham, U.K.

<sup>6</sup>Department of Dermatology, Sheffield  
Teaching Hospitals NHS Foundation Trust,  
Sheffield, U.K.

<sup>7</sup>Department of Gynaecology, Nottingham  
University Hospitals NHS Trust,  
Nottingham, U.K.

<sup>8</sup>University of Warwick, Warwick, U.K.

<sup>9</sup>James Lind Alliance, National Institute for  
Health Research Evaluation, Trials and

R. C. SIMPSON<sup>1</sup> 

S. M. COOPER<sup>2</sup>

G. KIRTSCHIG<sup>3</sup>

S. LARSEN<sup>4</sup>

S. LAWTON<sup>5</sup>

M. MCPHEE<sup>1</sup>

R. MURPHY<sup>6</sup>

D. NUNNS<sup>7</sup>

S. REES<sup>8</sup> 

M. TARPEY<sup>9</sup>

K. S. THOMAS<sup>1</sup> 

on behalf of the Lichen  
Sclerosis Priority  
Setting Partnership  
Steering Group

Studies Coordinating Centre (NETSCC),  
University of Southampton, Southampton,  
U.K.

E-mail: [rosalind.simpson@nottingham.ac.uk](mailto:rosalind.simpson@nottingham.ac.uk)

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