



University of Nottingham
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James Lind Alliance
Priority Setting Partnerships

Lichen Sclerosus Priority Setting Partnership

PROTOCOL August 2017

1. Purpose of the PSP and background

The purpose of this protocol is to set out the aims, objectives and commitments of the Lichen Sclerosus Priority Setting Partnership (PSP) and the basic roles and responsibilities of the partners therein. It is recommended that the Protocol is reviewed by the Steering Group and updated on at least a quarterly basis.

The James Lind Alliance (JLA) is a non-profit making initiative, established in 2004. It brings patients, carers and clinicians together in Priority Setting Partnerships (PSPs). These partnerships identify and prioritise uncertainties, or 'unanswered questions', about the condition that they agree are the most important. The aim of this is to help ensure that those who fund health research are aware of what really matters to both patients and clinicians. The National Institute for Health Research (NIHR – www.nihr.ac.uk) funds the infrastructure of the JLA to oversee the processes for priority setting partnerships, based at the NIHR Evaluation, Trials and Studies Coordinating Centre (NETSCC), University of Southampton.

Lichen sclerosus is a chronic, inflammatory skin condition. Women, men and children can be affected. Information about incidence and prevalence in each of these groups is not known. It was previously thought to be most common in adult women, but anecdotal evidence suggests it is just as common in men and boys. The condition is believed to occur in all ethnic groups, but data to confirm this are not available.

In women and pre pubertal girls, it primarily affects the vulva. A variety of symptoms, for example, intense itch, pain and splitting occur. The physical appearance of the vulva is affected. These symptoms of lichen sclerosus impact upon daily function. Delay or poor response to treatment leads to ongoing inflammation. Subsequent scarring can cause labial fusion, narrowing of the vaginal opening and burying of the clitoris. In girls lichen sclerosus can result in an itchy vulva and often pain with defecation, medical treatment helps but cure is not expected in most and the disease will continue in adult life.

In men and boys, lichen sclerosus tends to develop on the glans penis and sometimes on the foreskin. Symptoms in men may be difficulty urinating due to narrowing of the urethra, difficulty in retracting the foreskin due to scarring and painful sexual intercourse. In boys lichen sclerosus will usually lead to a tight foreskin (phimosis). Circumcision is often required, especially when symptoms are poorly controlled with topical treatment. It is not known if this procedure is curative or not.

The anatomical changes that occur in lichen sclerosus are usually irreversible and can have a detrimental effect upon day to day function and psychological health of those affected. Men are at risk of serious urethral disease. People with lichen sclerosus also appear to have an increased risk of genital cancer.

There are many uncertainties about the causes, diagnosis and management of lichen sclerosus due to lack of published high quality evidence. A Cochrane review in 2011 concluded that current evidence in the field of lichen sclerosus was limited and that gaps in knowledge needed to be filled (1).

In 2015 a European Dermatology Forum S3 guideline on lichen sclerosus was published (2) which also highlighted a number of gaps in knowledge of the condition.

In an attempt to address the lack of evidence, the NIHR recently released a Health Technology Assessment (HTA) commissioned call for a trial to investigate topical treatments for lichen sclerosus of the vulva. The funding call asked for proposals for RCTs to compare the use of topical steroids and topical calcineurin inhibitors in the initial treatment and long-term maintenance of remission for patients with lichen sclerosus.

Researchers at the Centre of Evidence Based Dermatology at the University of Nottingham responded to this call by coordinating a multidisciplinary Steering Group of healthcare professionals, researchers and patients. However, despite enthusiasm for the topic area of lichen sclerosus, the group did not feel that the research question as outlined in the NIHR HTA brief was the most important research priority in this area. This was confirmed by preliminary work. Following a survey of 184 healthcare professionals (including dermatologists, gynaecologists, genitourinary medicine specialists, GPs and nurses), considerable variation was expressed in terms of willingness to randomise patients to a trial that included long-term calcineurin inhibitors as one of the intervention arms.

This preliminary work did, however, demonstrate significant interest from the clinical community for research to be carried out in the field of lichen sclerosus. Our Steering Group firmly believe that the most appropriate starting point in this regard is to take a 'step back' and conduct a James Lind Alliance PSP for lichen sclerosus in women, men and children. A formal PSP is a well-established method to identify and prioritise research uncertainties of importance to patients and healthcare providers. Conducting a PSP also helps to raise awareness of the condition amongst funding bodies and NHS providers.

The NIHR HTA have been consulted regarding this and are supportive of the concept, as well as remaining committed to funding future research into lichen sclerosus. However, funding a PSP is currently outside of the NIHR HTA remit. This has led to the need for our Steering Group to seek an appropriate source of funding to carry out this important work.

We have chosen to focus on the condition lichen sclerosus, as opposed to all genital skin diseases, as we felt that the scope of the latter would be too broad making it difficult to engage with patients and relevant stakeholder organisations. This PSP will include both sexes affected by lichen sclerosus. The final scope of the Lichen Sclerosus PSP has been clarified by the Steering Group following the first meeting.

2. Aims and objectives of the Lichen Sclerosus PSP

The aim of the Lichen Sclerosus PSP is to identify the unanswered questions about the causes, diagnosis, treatment and prevention of lichen sclerosus from the perspectives of those with the disorder, their parents/carers/relatives/friends and health professionals, and then prioritise those that participants agree are the most important. Questions relating to service provision and provision of care will be excluded.

This is an initiative led from the U.K, but we will include all responses regardless of geographical location. International entries will be welcomed and existing international connections will be approached to raise awareness of the LSPSP. However, the survey will only be made available in the English language. The organisations approached will be acknowledged on study documentation for transparency.

The objectives of the Lichen Sclerosus PSP are to:

- work with people with lichen sclerosus, their parents/carers/relatives/friends and health professionals, to identify uncertainties about the effects of lichen sclerosus causes, diagnosis, treatment and prevention in men, women and children.
- to agree by consensus a prioritised list of those uncertainties, for research

- to publicise the results of the PSP and process
- to take the results to research commissioning bodies to be considered for funding.
- Via an additional research question, to establish the most important outcomes of lichen sclerosis treatment from the perspective of people with lichen sclerosis, their parents/carers/relatives/friends and health professionals.

3. The Steering Group

The Lichen Sclerosis PSP will be led and managed by the following:

- Patient representative/s (initials used for confidentiality):
 - MD
 - HB
 - JS
 - LH
 - Suzanne Larsen (Representative of the Lichen Sclerosis Association Denmark)
 - SR
- Researchers (non-clinical)
 - Kim Thomas, Professor of Applied Dermatology Research

Clinical representative/s:

- Dermatologists
 - Dr Rosalind Simpson, Research Fellow and Dermatology Trainee
 - Dr Ruth Murphy, Consultant Dermatologist
 - Dr Susan Cooper, Consultant Dermatologist
 - Dr Gudula Kirtschig, Consultant Dermatologist
 - Professor Chris Bunker, expert in male genital lichen sclerosis
- Nurses
 - Mrs Sandra Lawton, Nurse Consultant
- Gynaecologists
 - Mr David Nunns, Consultant Gynaecologist

The Partnership and the priority setting process will be supported and guided by:

- The James Lind Alliance (JLA)
 - Maryrose Tarpey, Independent Advisor

The project co-ordinating team are from the Centre of Evidence Based Dermatology, University of Nottingham

- Rosalind Simpson, Clinical Research Fellow in Dermatology
- Kim Thomas, Professor of Applied Dermatology Research
- Margaret McPhee, Research co-ordinator and administrator

The Steering Group includes representation of patient/carer groups and health professionals¹.

The Steering Group will agree the resources, including time and expertise that they will be able to contribute to each stage of the process. The JLA will advise on this.

4. The wider Partners

Organisations and individuals will be invited to be involved with the PSP as partners. Partners are groups or individuals who will commit to supporting the PSP by disseminating the PSP survey and helping the PSP to gather questions and uncertainties of practical clinical importance relating to the treatment and management of the health problem in question. Partners represent the following groups:

- people who have lichen sclerosis
- carers, relatives and friends of people who have lichen sclerosis
- health professionals with clinical experience of lichen sclerosis.

It is important that all organisations which can reach and advocate for these groups should be invited to become involved in the PSP. For this Lichen Sclerosis PSP this includes international connections. We will approach our existing international links to disseminate as widely as possible within the available resources. A JLA Adviser will take responsibility for ensuring the various stakeholder groups are able to contribute equally to the process.

Exclusion criteria

Some organisations may be judged by the JLA or the Steering Group to have conflicts of interest. These may be perceived to adversely affect those organisations' views, causing unacceptable bias. As this is likely to affect the ultimate findings of the PSP, those organisations will not be invited to participate. It is possible, however, that interested parties may participate in a purely observational capacity when the Steering Group considers it may be helpful.

¹ In some cases, it has been suggested that researchers are represented at this level, to advise on the shaping of research questions. However, researchers cannot participate in the prioritisation exercise. This is to ensure that the final prioritised research questions are those agreed by patients, carers and clinicians only, in line with the JLA's mission.
James Lind Alliance: Lichen Sclerosis Priority Setting Partnership Protocol – August 2017

5. The methods the PSP will use

This section describes a schedule of proposed stages through which the PSP aims to fulfil its objectives. The process is iterative and dependent on the active participation and contribution of different groups. The methods adopted in any stage will be agreed through consultation between the Steering Group members, guided by the PSP's aims and objectives. More details can be found in the Guidebook section of the JLA website at www.jla.nihr.ac.uk where examples of the work of other JLA PSPs can also be seen.

Step 1: Identification and invitation of potential partners

Potential partner organisations will be identified through a process of peer knowledge and consultation, through the Steering Group members' networks. Potential partners will be contacted and informed of the establishment and aims of the Lichen Sclerosus PSP.

Potential partner organisations that have been identified so far are as follows:

- Medical organisations/societies
 - British Society for the Study of Vulval Disease (BSSVD)
 - International Society for the study of Vulvovaginal Disease (ISSVD)
 - UK Dermatology Clinical Trials Network (UKDCTN)
 - British Association of Dermatologists (BAD)
 - British Association for Sexual Health and HIV (BASHH)
 - British Male Genital Dermatology group
 - Royal College of Obstetricians and Gynaecologists (RCOG)
 - Royal College of Pathologists
 - Primary Care Dermatology Society (PCDS)
 - British Society of Academic General Practitioners
 - British Society for Medical Dermatology
 - Royal College of General Practitioners
 - British Association of Urological Surgeons
 - British Society of Paediatric Dermatology (BSPD)
 - British Dermatological Nursing Group
- Patient organisations
 - Dermnet.nz
 - Facebook groups
 - Local patient support groups who do not have internet presence
 - Male support groups if identified
 - UK Association for Lichen Sclerosus and Vulval Health
 - Danish support group
 - [Dutch support group](#)
 - Swiss Lichen Sclerosus organization
 - Dutch support group
 - Vulval pain society
 - Manchester vulval support network

Step 3: Identifying uncertainties

A self-completion on-line survey will be used. The survey will be open for up to eight weeks depending on the response rate and the need to chase under-represented groups.

The methods to be adopted to publicise the survey and encourage participation include but are not necessarily limited to:

- Advertisements and links within stakeholder web sites
- Social media
- PSP members' personal networks

- Patient and professional bloggers
- Newsletters distributed by stakeholders to members
- Leaflets, postcards and posters for display in specialist treatment centres

The methods may be designed according to the nature and membership of each organisation, but must be as transparent, inclusive and representative as practicable. Methods may include membership meetings, email consultation, postal or web-based questionnaires, internet message boards and focus group work. The participant information and survey text will be designed to be easy to understand and provide all the relevant information for self-completion.

Existing sources of information about treatment uncertainties for patients and clinicians will be searched. These can include question-answering services for patients (e.g. healthtalk.org) and carers and for clinicians; research recommendations in systematic reviews and clinical guidelines; protocols for systematic reviews being prepared and registers of ongoing research.

The starting point for identifying sources of uncertainties and research recommendations is NHS Evidence: www.evidence.nhs.uk.

Step 4: Refining questions and uncertainties

The JLA will participate in this process as an observer, to ensure accountability and transparency.

The survey will produce “raw” unanswered questions about the causes, diagnosis treatment and prevention of lichen sclerosis. These raw questions will be assembled and categorised and refined by Rosalind Simpson, a research assistant and members of the Steering Group where relevant. Those which are out of scope i.e. relate to delivery of care or are not uncertainties will be excluded from the list and kept separately.

The included questions will be categorised into “collated indicative questions” which are clear, addressable by research and understandable to all. Similar or duplicate questions will be combined where appropriate.

Systematic reviews and guidelines will be identified and checked by Rosalind Simpson, an Information Specialist and a research assistant, to see to what extent these refined questions have, or have not, been answered by previous research. Sometimes, uncertainties are expressed that can in fact be resolved with reference to existing research evidence - ie they are "unrecognised knowns" and not uncertainties. If a question about treatment effects can be answered with existing information but this is not known, it suggests that information is not being communicated effectively to those who need it. Accordingly, the JLA recommends strongly that PSPs keep a record of these 'answerable questions' and deal with them separately from the 'true uncertainties' considered during the research priority setting process.²

Uncertainties which are not adequately addressed by previous research will be collated and recorded on a template (supplied by the JLA) by Rosalind Simpson. This will demonstrate the checking undertaken to make sure that the uncertainties have not already been answered. This is the responsibility of the Steering Group, which will need to have agreed personnel and resources to carry this accountability. The data should be submitted to the JLA for publication on its website on completion of the priority setting exercise, taking into account any changes made at the final workshop, in order to ensure that PSP results are publicly available.

Step 5: Prioritisation – interim and final stages

The aim of the final stage of the priority setting process is to prioritise through consensus the identified uncertainties relating to the treatment or management of lichen sclerosis. This will be carried out by members of the Steering Group and the wider partnership that represents patients and clinicians.

² Steering Group members should insert information on how they intend to do this.

- The interim stage, to proceed from a long list of uncertainties to a shorter list to be discussed at the final priority setting workshop (e.g. up to 30), will be carried out over email or online. This is via a second survey whereby individuals who responded to Survey 1 choose and rank their top 10 most important uncertainties. Partner organisations are also asked to consult their membership to complete the survey.
- The final stage, to reach, a top 10 prioritised uncertainties, will be conducted in a face-to-face meeting, using group discussions and plenary sessions.

The JLA will facilitate this process and ensure transparency, accountability and fairness. Participants will be expected to declare their interests in advance of this meeting.

6. Dissemination of findings and research

Findings and research

It is anticipated that the findings of the Lichen Sclerosus PSP will be reported to funding and research agenda setting organisations such as the NIHR and the major UK research funding charities. Steering Group members and partners are expected to develop the prioritised uncertainties into research questions, and to work to establish the research needs of those unanswered questions to use when approaching potential funders, or when allocating funding for research themselves, if applicable.³

Publicity

As well as alerting funders, partners and Steering Group members are encouraged to publish the findings of the Lichen Sclerosus PSP using both internal and external communication mechanisms. The Steering Group may capture and publicise the results through descriptive reports of the process itself in Plain English. This exercise will be distinct from the production of an academic paper, which the partners are also encouraged to do. However, production of an academic paper should not take precedence over publicising of the final results.

7. Additional research element of this project

Whilst the basic processes for the conduct of a priority setting exercise are fixed, there is scope and opportunity to collect additional information that will be of future benefit to research informed by the results of the PSP. It is not currently clear which outcomes are considered most important by those who have lichen sclerosus, or healthcare professionals that treat the condition. We will therefore collect this information from participants via an open question in the survey.

The analysis of this supplementary question will be conducted as time and resources permit. Where sufficiently rigorous, results will be submitted for publication in relevant journals.

8. Agreement of the Steering Group

Signed by the Steering Group

The undersigned agree to follow the Lichen Sclerosus Priority Setting Protocol.

[Insert name or initials and organisation]

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³ Add further detail here about how and where the priorities will be developed and researched.
James Lind Alliance: Lichen Sclerosus Priority Setting Partnership Protocol – August 2017

Date:

[Insert name and organisation]

.....

Date:

[Insert name], The James Lind Alliance

.....

Date: