

## Juvenile Idiopathic Arthritis (JIA) Priority Setting Partnership

### PROTOCOL 19-09-2018<sup>1</sup>

## 1. Purpose of the PSP and background

The purpose of this protocol is to set out the aims, objectives and commitments of the JIA 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 effects of treatments 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](http://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.

The Dutch JIA PSP was initiated by C. Schoemaker, a carer of a patient with JIA, senior health researcher and patient representative, to bring patients and clinicians together in formulating research questions. Juvenile Idiopathic Arthritis is the most common rheumatic disease in children and affects 1 in 1000 children under the age of 16 years. Although a vast amount of research has been done in this field, patients have only minimally been involved in research priority setting. This process is hindered by the fact that it is more difficult for children to voice their opinions on their specific research priorities and needs.

The JIA PSP is a joint collaboration of patient organisations and clinician organisations (see below) and is supported by all treatment centres in The Netherlands where patients with JIA are treated. This PSP aims to identify unanswered questions (uncertainties) about the cause, diagnosis, treatment (management or self-management) and outcome of JIA, and for patients, carers and clinicians to prioritise these unanswered questions for research.

## 2. Aims and objectives of the JIA PSP

The aim of the Juvenile Idiopathic Arthritis (JIA) PSP is to identify the unanswered questions about JIA treatment from patient and clinical perspectives and then prioritise those that patients and clinicians agree are the most important.

The objectives of the JIA PSP are to:

- Work with patients and clinicians to identify uncertainties about the effects of JIA treatments. Our goal is to be as inclusive as possible and engage patients from all socioeconomic backgrounds.

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<sup>1</sup> This is a generic protocol which should be updated to include the names and details of the Steering Group members. The document may be modified with agreement from the JLA to reflect the make-up of different PSPs and the organisations driving them.

- 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.

### 3. The Steering Group

The JIA PSP will be led and managed by the following:

Name	Role	Organisation	City and academic centre
<i>Core</i>			
Casper Schoemaker (Lead)	Carer,	JVN	Utrecht, WKZ
Wineke Armbrust (Lead)	Pediatric rheumatologist	NVKR	Groningen, UMCG
Anouk Verwoerd (coordinator, information specialist)	MD, PhD student		Utrecht, WKZ
Katherine Cowan (JLA adviser)	JLA adviser	JLA	London
<i>Steering group</i>			
<u>Doctors</u>			
Joost Swart	Paediatric rheumatologist	NVKR	Utrecht, WKZ
Petra Hissink Muller	Paediatric rheumatologist	NVKR	Leiden, LUMC
Ellen Schatorjé	Paediatric rheumatologist	NVKR	Nijmegen, Radboud & St. Maartens kliniek
Joke Boer	Ophthalmologist	NOG	Utrecht, UMCU
<u>Paramedics</u>			
Otto Lelieveld	Physical therapist	NHPKR	Groningen, UMCG
Jeannette Capon	Physical therapist	NHPKR	Amsterdam, Reade
Margot Walter	Nurse practitioner		Rotterdam, SKZ
<u>Patients</u>			
Wendy Olsder	Patient	Youth-R-Well	
Johanna Rocha	Patient	Youth-R-Well	
Sanne Bookelman	Patient	JVN	
Lotte van den Berg	Patient	JVN	
<u>Carers (parents)</u>			
Karin van den Haspel	Parent	JVN	
Marjan Britstra	Parent	JVN	
Natasja Schouten	Parent	JVN	
<i>Focus groups</i>			
Christine Dedding	Researcher		Amsterdam, VUMC
<i>Process evaluation</i>			

Karin Jongsma	Medical ethicist		Utrecht, UMCU
Annemiek van Rensen	Patient participation adviser	PGO Support	

Abbreviations:

JVN (jeugdreumavereniging Nederland), LUMC (Leiden University Medical Centre), NHKPR (Nederlandse Health Professionals Kinderreumatologie), NOG (Nederlands Oogheelkundig Gezelschap), NVKR (Nederlandse Vereniging voor Kinderreumatologie), SKZ (Sophia Kinderziekenhuis), UMCG (University Medical Centre Groningen), UMCU (University Medical Centre Utrecht), VUMC (Vrije Universiteit Medisch Centrum), WKZ (Wilhelmina Kinderziekenhuis)

The Steering Group includes representation of patient/carers groups and clinicians<sup>2</sup>.

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 JIA
- carers of people who have JIA
- medical doctors, nurses and professionals allied to medicine with clinical experience of JIA

It is important that all organisations which can reach and advocate for these groups should be invited to become involved in the PSP. The 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.

## 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](http://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

<sup>2</sup> 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: Priority Setting Partnership Protocol – January 2016

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 JIA PSP and may be invited to attend and participate in an initial stakeholder meeting if this is being arranged.

### **Step 2: Initial stakeholder meeting / awareness raising<sup>3</sup>**

The initial stakeholder meeting / awareness raising will have several key objectives:

- to welcome and introduce potential members of the JIA PSP
- to present the proposed plan for the PSP
- to initiate discussion, answer questions and address concerns
- to identify those potential partner organisations which will commit to the PSP and identify individuals who will be those organisations' representatives and the PSP's principal contacts
- to establish principles upon which an open, inclusive and transparent mechanism can be based for contributing to, reporting and recording the work and progress of the PSP.

### **Step 3: Identifying treatment uncertainties**

All members of the steering group together will identify a method for soliciting from its members questions and uncertainties of practical clinical importance relating to the treatment and management of JIA. A period of three to four months will be given to complete this. Methods include web-based questionnaires as well as focus group work to include the voices of younger children. Karin Jongsma will also perform a medical ethical evaluation of the process.

Existing sources of information about treatment uncertainties for patients and clinicians will be searched. These can include question-answering services for patients 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 PubMed: <https://www.ncbi.nlm.nih.gov> and management guidelines of JIA.

### **Step 4: Refining questions and uncertainties**

The Steering Group will need to have agreed exactly who will be responsible for this stage – the JLA can advise on the amount of time likely to be required for its execution. The JLA will participate in this process as an observer, to ensure accountability and transparency.

The consultation process will produce “raw” unanswered questions about diagnosis and the effects of treatments. These raw questions will be assembled and categorised and refined by Anouk Verwoerd 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 Anouk Verwoerd (information specialist) 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

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<sup>3</sup> PSPs will need to raise awareness of their proposed activity among their patient and clinician communities, in order to secure support and participation. Depending on budget this may be done by way of a face-to-face meeting, or there may be other mechanisms by which the process can be launched.

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.<sup>4</sup>

Uncertainties which are not adequately addressed by previous research will be collated and recorded on a template supplied by the JLA) by Anouk Verwoerd (information specialist). 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 JIA. This will be carried out by members of the Steering Group and the wider partnership that represents patients and clinicians.

- 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), may be carried out over email or online, whereby the partner organisations consult their membership and choose and rank their top 10 most important uncertainties.
- The final stage, to reach, for example, 10 prioritised uncertainties, will be conducted in a face-to-face meeting, using group discussions and plenary sessions.
- The methods used for this prioritisation process will be determined by consultation with the partner organisations and with the advice of the JLA Adviser. Methods which have been identified as potentially useful in this process include: adapted Delphi techniques; expert panels or nominal group techniques; consensus development conference; electronic nominal group and online voting; interactive research agenda setting and focus groups.

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 JIA PSP will be reported to funding and research agenda setting organisations such as the NIHR and the major 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.<sup>5</sup>

### **Publicity**

As well as alerting funders, partners and Steering Group members are encouraged to publish the findings of the JIA 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

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<sup>4</sup> Steering Group members should insert information on how they intend to do this.

<sup>5</sup> Add further detail here about how and where the priorities will be developed and researched.

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. Agreement of the Steering Group**

**The steering group approved this protocol on their first meeting on October 29<sup>th</sup>, in Utrecht, The Netherlands.**