

**Children's Cancer Priority Setting Partnership
PROTOCOL
24th February 2020, Version 2**

1. Purpose of the PSP and background

The purpose of this protocol is to set out clearly the aims, objectives and commitments of the Children's Cancer Priority Setting Partnership (PSP) in line with James Lind Alliance (JLA) principles. The Protocol is a JLA requirement and will be published on the PSP's page of the JLA website. The Steering Group will review the Protocol regularly and any updated version will be sent to the JLA.

The JLA is a non-profit making initiative, established in 2004. It brings patients, carers and professionals together in PSPs. These PSPs identify and prioritise the evidence uncertainties, or 'unanswered questions', that are the most important for research in their topic area. Traditionally PSPs have focused on unanswered questions about the effects of treatments, but some PSPs have chosen to broaden their scope beyond that. The aim of a PSP is to help ensure that those who fund health research are aware of what really matters to patients, carers and professionals. The National Institute for Health Research (NIHR – www.nihr.ac.uk) coordinates the infrastructure of the JLA to oversee the processes for PSPs, based at the NIHR Evaluation, Trials and Studies Coordinating Centre (NETSCC), University of Southampton.

In this PSP, the focus is on children's cancer. There have been several previous PSPs about cancer; the coordinating team have already completed a PSP focusing on Teenage and Young Adult Cancer (a population aged 13 to 24), there has also been a 'Living With and Beyond Cancer' PSP focusing on adults, and other PSPs focusing on particular cancer diagnoses (e.g. brain tumours). The conditions that children develop are often different to those of adults or young people; their physical, psychological and social responses to these life-threatening diagnoses are also different. Involving children who have experienced cancer and their families in guiding future research is crucial if the questions that are important to them are to be addressed. To encourage more research which is focussed on the priorities set by children with cancer, families/carers and the professionals that work with them, the Children's Cancer and Leukaemia Group (CCLG) and Little Princess Trust have funded this Children's Cancer PSP. The first Steering Group meeting took place on 11th October 2019. We propose that the final priority setting workshop takes place in September 2021.

2. Aims, objectives and scope of the PSP

The aim of the Children's Cancer PSP is to identify gaps and unanswered questions in research about Children's Cancer from patients, carers and professionals' perspectives and then prioritise those that these groups agree are the most important for research to address.

The objectives of the PSP are to:

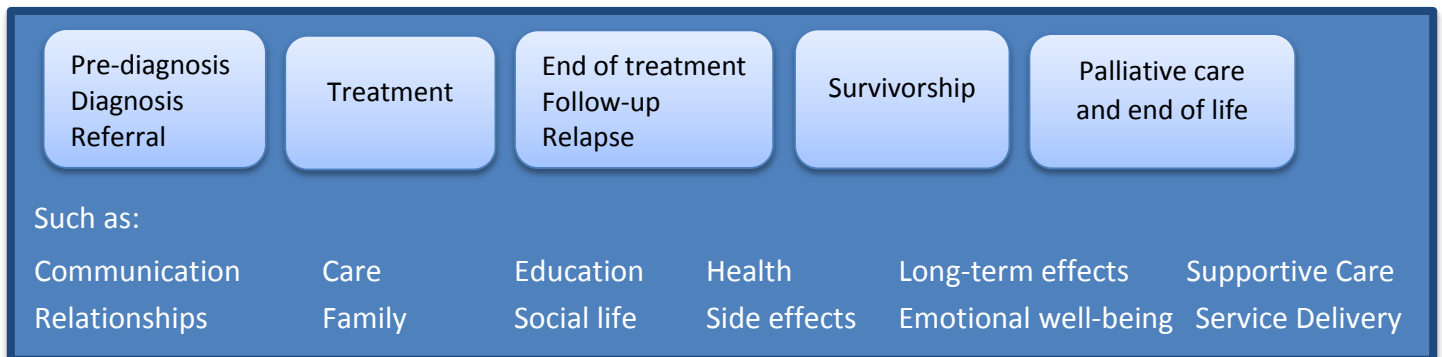
- Work with patients, carers and professionals to identify what matters to them and what they think future research on children's cancer should address.

- Work with patients, carers and professionals to identify important areas for research, and unanswered questions about cancer in children.
- Agree by consensus how to rank in importance a list of priority areas and unanswered questions for research.
- Publicise the results of the PSP and process.
- Take the results to research commissioning bodies to be considered for funding.

The scope of the Children’s Cancer PSP includes questions about:

- All types of cancer and cancer-like conditions.
- Cancer in children aged 0 to 15 years at initial diagnosis (up to their 16th birthday).
- Any aspect of the prevention or diagnosis of cancer in children.
- Any aspect of the referral, treatment and management of childhood cancer, and the care of children who have or have had cancer.
- Childhood cancer survivorship, including follow-up and late effects.
- Questions relating to the families/carers of children with cancer.
- Psychological, emotional and social aspects of childhood cancer.
- Palliative and end of life care.

Questions can be about any aspect of Children’s Cancer along the care pathway:



It is recognised that this is a very broad remit, and decisions may need to be made to focus the scope of the PSP depending on what questions are submitted. Although the Teenage and Young Adult Cancer PSP included young people who had cancer aged 13 and over, very few responses were received from those aged 13 to 15 years old; therefore we would like to include this group in the Children’s Cancer PSP.

The PSP will exclude from its scope questions about:

Cancer in people who are 16 years and older at the time of initial diagnosis.

The Steering Group is responsible for discussing what implications the scope of the PSP will have for the evidence-checking stage of the process. Resources and expertise will be put in place to do this evidence checking.

3. The Steering Group

Steering Group membership includes patients, carers and professionals, as individuals or representatives from a relevant group. The role of the Steering Group is to manage the PSP, ensuring that it is delivered in a timely way and adheres to the JLA's key principles of transparency, inclusion/exclusion, equality of voice and use of the existing evidence base. The Steering Group will ensure that the process from the initial survey to the identification of the top 10 priorities is documented, open and transparent and entirely determined by the responses received from patients, carers and professionals and the review of the existing evidence base; members of the Steering Group have no influence on setting priorities either as individuals or representatives of organisations.

The Children's Cancer PSP will be led and managed by a Steering Group involving the following people:

Patient and parent representatives:

- Scott Crowther
- Angela Polanco
- Loveday Langton
- Alex Brownsdon

Professional representatives:

- Rachel Hollis, PSP Lead, Leeds Children's Hospital
- Ashley Gamble, Chief Executive Officer, CCLG
- Wendy Tarplee-Morris, Research Manager and Co-founder, The Little Princess Trust
- Penelope Hart-Spencer, Health Play Specialist, The Christie, Manchester
- Jess Morgan, Trainee Paediatric Oncologist & NIHR Clinical Lecturer in Paediatric Oncology, Leeds Children's Hospital and University of York
- Julia Chisholm, Consultant in Paediatric and TYA Oncology, The Royal Marsden NHS Foundation Trust
- Simon Parke, Consultant Paediatrician, Royal Devon and Exeter NHS Foundation Trust
- Jenni Hatton, Children's Cancer Network Pharmacist, Queen's Medical Centre, Nottingham
- Helen Morris, Matron, Lead Nurse Southwest Paediatric Oncology, Bristol Royal Hospital for Children
- Louise Henry, Senior Specialist Dietitian (Paediatrics and TYA), The Royal Marsden NHS Foundation Trust
- Rosa Reed-Berendt, Clinical Psychologist, Great Ormond Street Hospital for Children NHS Foundation Trust
- Sue Picton, Consultant Paediatric and Adolescent Oncologist, Leeds Children's' Hospital/ Martin House Hospice
- Rachel Dommett, Consultant in Paediatric Leukaemia and Aftercare, Bristol Royal Hospital for Children
- Martin English, Consultant Paediatric Oncologist, Birmingham Women's and Children's NHS Foundation Trust
- Charmaine Jagger, Speech and Language Therapist, The Royal Marsden NHS Foundation Trust
- Helen Hartley, Specialist Paediatric Physiotherapist - Acute Neurosciences/ Oncology, Alder Hey Children's NHS Foundation Trust
- Helen Gravestock/Sonia Malik, CLIC Sargent Representative, CLIC Sargent, London
- Nigel Hall, Consultant Paediatric Surgeon, University of Southampton
- Dan Saunders, Consultant in Clinical Oncology, The Christie, Manchester
- Keeley McEvoy, Assistant Headteacher, Medical Needs Teaching Service, Leeds Children's Hospital
- David Weller, Professor of General Practice, University of Edinburgh

Project coordinators:

- Bob Phillips, Paediatric Oncology Consultant, Leeds Children's Hospital and University of York
- Susie Aldiss, Research Fellow, University of Surrey
- Faith Gibson, Professor of Child Health and Cancer Care, University of Surrey and Great Ormond Street Hospital for Children NHS Foundation Trust

Administrative support for the PSP:

- Angela Stewart, Project Manager, CCLG

James Lind Alliance Adviser and Chair of the Steering Group:

- Jonathan Gower, James Lind Alliance

The Steering Group will agree the resources, including time and expertise that they will be able to contribute to each stage of the process, with input and advice from the JLA.

A children's group will be set up that involves 10-12 children who have cancer or had cancer and siblings. This group will help to guide the PSP, particularly regarding how best to engage with children to collect and prioritise research questions. They will also advise on dissemination of findings to children, to ensure any materials produced are relevant and accessible for children. We will hold separate face-to-face meetings for the children's group which will be led by Faith Gibson and Susie Aldiss. Children will be recruited through the steering group and the charities involved in the project/social media (via their parents).

4. Partners

Organisations and individuals will be invited to be involved with the PSP as partners. Partners are organisations or groups who will commit to supporting the PSP, promoting the process and encouraging their represented groups or members to participate in the surveys. Organisations which can reach and advocate for these groups will be invited to become involved in the PSP. Partners represent the following three groups:

1. People who have been diagnosed with childhood cancer.
2. Family, friends and carers of children who have been diagnosed with cancer.
3. Health and social care professionals who work with children who have cancer.

Exclusion criteria

Some organisations may be judged by the JLA or the Steering Group to have a conflict of interest and may be perceived to potentially cause 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 steps through which the PSP aims to meet its objectives. The process is iterative and dependent on the active participation and contribution of different groups. The methods used in any step will be agreed through consultation between the Steering Group members, guided by the PSP's aims and objectives. More details of the method are in the Guidebook section of the JLA website at www.jla.nihr.ac.uk where examples of the work of other JLA PSPs can 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 Children's Cancer PSP.

Step 2: Awareness raising

The PSP will raise awareness of their proposed activity among their patient, carer and professional communities, in order to secure support and participation. This will be through contacting Partners, via social media and information on the CCLG website or through contact from Steering Group members.

Awareness raising has several key objectives:

- To present the proposed plan for the PSP.
- To generate support for the process.
- To encourage participation in the process.
- To initiate discussion, answer questions and address concerns.

Step 3: Gathering unanswered questions

The Children's Cancer PSP will carry out a consultation to gather questions from patients, carers and professionals. Most PSPs collect questions using an online or postal survey. It is important that this PSP engages with children in identifying questions; it is recognised that this will require the use of additional innovative methods. A subgroup of the Steering Group will focus on the engagement of children in suggesting and prioritising unanswered questions. Methods will be piloted with children and their feedback sought about the best ways of engaging. The Steering Group plan to use the following methods to reach different groups:

- Online survey – for older children/young people (including survivors of childhood cancer), families and professionals.
- Face-to-face workshops – for younger children, younger siblings and friends.
- Paper survey or surveys on a 'tablet' or notebook in the Principal Treatment Centres – for younger children and families to complete together.

Step 4: Refining questions

The consultation process will produce 'raw' questions and comments indicating patients', carers' and professionals' areas of uncertainty and what is important to them. The PSP acknowledges that not all respondents will submit responses in question format; these responses will be included in the refining process. These raw questions will be categorised and refined by Susie Aldiss and Faith Gibson in collaboration with the Steering Group into summary questions which are clear, addressable by research, and understandable to all. Similar or duplicate questions will be combined where appropriate. Out-of-scope submissions will be compiled separately. The Steering Group will have oversight of this process to ensure that the raw data is being interpreted appropriately and that the summary questions are being worded in a way that is understandable to all audiences. Both the 'raw questions' and the 'summary questions' will be published on the JLA Website. The JLA Adviser will observe to ensure accountability and transparency.

This will result in a long list of in-scope summary questions. These are not research questions and to try and word them as such may make them too technical for a non-research audience. They will be framed as questions that capture the themes and topics people have suggested and may then be developed into research questions at the end of the priority setting exercise.

The summary questions will then be checked against evidence to determine whether they have already been adequately answered by research. This will be done by Susie Aldiss and Bob Phillips with input from Steering Group members on their area of expertise. The PSP will complete the JLA Question Verification Form, which clearly describes the process used to verify whether questions have already been answered or are the focus of an ongoing study, before starting prioritisation. The Question Verification Form includes details of the types and sources of evidence used to check the questions. The Question Verification Form will be published on the

JLA website as soon as it has been agreed to enable researchers and other stakeholders to understand how the PSP has decided that its questions are unanswered, and any limitations of this.

Questions that are not adequately addressed by previous research will be collated and recorded on a standard JLA template by Susie Aldiss. This will show the checking undertaken to make sure that the questions have not already been answered. The data will 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.

The Steering Group will also consider how it will deal with submitted questions that have been answered, and questions that are out of scope. In some cases this will require 'sign-posting' respondents to available sources of information or support.

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 unanswered questions about Children's Cancer. This will involve input from patients, carers and professionals. The JLA encourages PSPs to involve as wide a range of people as possible, including those who did and did not contribute to the first consultation. There are usually two stages of prioritisation:

1. Interim prioritisation is the stage where the long list of questions is reduced to a shorter list that can be taken to the final priority setting workshop. This is aimed at a wide audience, and will be done using similar methods to the first consultation. With the JLA's guidance, the Steering Group will agree the method and consider how best to reach and engage children, carers and professionals in the process. The most highly ranked questions (around 25) will be taken to a final priority setting workshop. Where the interim prioritisation does not produce a clear ranking or cut off point, the Steering Group will decide which questions are taken forward to the final prioritisation.

2. The final priority setting stage is generally a one-day workshop facilitated by the JLA. With guidance from the JLA and input from the Steering Group, up to 30 patients, carers and professionals will be recruited to participate in a day of discussion and ranking, to determine the top 10 questions for research. All participants will declare their interests. The Steering Group will advise on any adaptations needed to ensure that the process is inclusive and accessible. Children will be involved in the final priority setting; a workshop will take place for children, which occur either alongside the workshop for carers and professionals or beforehand.

6. Dissemination of results

The Steering Group will identify key audiences that it will be important to engage when disseminating the results of the priority setting process, such as researchers, funders and patient and clinical communities. Outputs will include: academic papers, final report, summary for children, infographic, conference presentations and dissemination via social media.

It should be noted that the priorities are not worded as research questions. The Steering Group will discuss how they will work with researchers and funders to establish how to address the priorities and to work out what the research questions are that will address the issues that people have prioritised. The dissemination of the results of the PSP will be led by Faith Gibson, Bob Phillips and Susie Aldiss in collaboration with the CCLG and Little Princess Trust. A meeting to engage with funders is planned following publication of the Top 10 priorities.

The PSP will report back to the JLA, partners and stakeholders about any activities that have come about because of the PSP, including funded research.

7. Agreement of the Steering Group

The Children's Cancer PSP Steering Group agreed the content and direction of this Protocol on 24th February 2020.