



# Research priorities in children's cancer

Final report of the James Lind Alliance Children's  
Cancer Priority Setting Partnership



# Executive summary

The Top 10 research priorities for children's cancer as agreed at the final workshop of the Children's Cancer Priority Setting Partnership (PSP) are detailed on the right. Fifteen professionals, four young adults who had cancer as a child, five parents and one grandparent prioritised the final 23 questions. These questions had been identified by children, young people, adult survivors of childhood cancer, families, and professionals during the national consultation. Questions were considered, debated, and deliberated throughout the day as the final list was agreed.

The final questions reflect the breadth of the cancer experience for children and families, including diagnosis, relapse, experience in hospital, support during and after treatment and the long-term impact of a cancer diagnosis. A workshop had previously been held with children and young people who identified their Top 5 priorities – these priorities are all reflected in the Top 10.

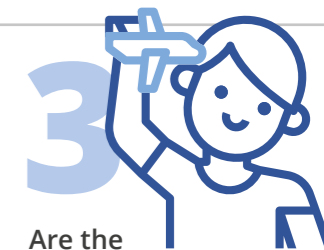
## Top 10 research priorities



**1** Can we find effective and kinder (less burdensome, more tolerable, with fewer short and long-term effects) treatments for children with cancer, including relapsed cancer?



**2** Why do children develop cancer (including the role that genetics plays) and could it be prevented? \*



**3** Are the psychological, practical, and financial support needs of children with cancer, survivors, and their families being met during treatment and beyond? How can access to this support be improved and what further support would they like?



**4** How can we speed up the process of getting diagnosed and starting treatment in the right place? \*



**5** Why do children relapse, how can it be prevented, and what are the best ways to identify relapse earlier? \*



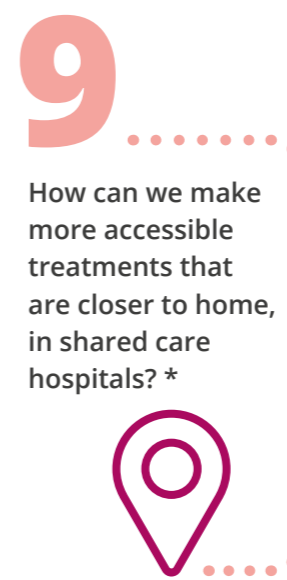
**6** How can we make being in hospital a better experience for children and young people? (Like having better food, internet, toys, and open visiting so other family members can be more involved in the child's care.) \*



**7** What are the best ways to ensure children and families get and understand the information they need, in order to make informed decisions, around the time of diagnosis, during treatment, at the end of treatment and after treatment has finished?



**8** What impact does cancer and treatment have on the lives of children and families after treatment, and in the long-term; what are the best ways to help them to overcome these impacts to thrive and not just survive?



**9** How can we make more accessible treatments that are closer to home, in shared care hospitals? \*



**10** What is the relationship between chronic fatigue syndrome, fibromyalgia, chronic pain and treatment for childhood cancer? (Fibromyalgia is a long-term condition that causes pain all over the body).



\*These questions were in the Top 5 research priorities identified by children and young people.

# Foreword

Every week, more than 30 families in the UK will receive the devastating news that their child has cancer. Since the 1970s, there has been huge progress in the care and treatment of children with cancer, driven by research. Five-year survival for all childhood cancers has increased to 84%. But that headline figure, rightly celebrated, doesn't give the whole picture.

Cancer is still the biggest killer, by disease, of children aged 1-15. There are many different types of cancer diagnosed in children, and while great progress has been made in some diseases, for some types of cancer survival rate hasn't improved so rapidly – if at all. Where treatment is successful, survivors can expect lifelong health consequences as a result of their disease, many of which can be debilitating, disabling, or life threatening.

So, despite progress, there is still much we don't know. The answers will come from research. The field of childhood cancer research is broad, with many disciplines, and many questions to be answered. Often, researchers decide which of those questions to investigate, and don't always reflect what is important to patients and their families, or the professionals involved in their treatment or care.

As a charity, Children's Cancer and Leukaemia Group (CCLG) strives to ensure that the needs of young cancer patients and their families are at the heart of what we do. The well-established process of a James Lind Alliance (JLA) Priority Setting Partnership

(PSP) seemed like the perfect way to ensure those voices, along with the insight of professionals, were at the centre of our research strategy, and could inform researchers and other research funders.

The Children's Cancer PSP, funded by ourselves and the Little Princess Trust, brought together young patients, childhood cancer survivors, their families and a wide range of healthcare professionals who treat and care for children with cancer. Together, they submitted and prioritised 1299 potential research questions about childhood cancer. It has been particularly pleasing that we have been able to ensure we heard the views of children too, and ensure they were reflected in the PSP. This process has given us 10 priority areas on which to focus research, ranked in order of importance.

Research is how we'll continue to make improvements for children diagnosed with cancer: increasing survival; improving survivorship and quality of life; getting better at diagnosis; understanding the experiences of patients and families, and driving forward

change. We've committed that our future research strategy will focus on the priorities highlighted through this process, and we're calling on funders, researchers and decision-makers to increase investment in research to address these priority areas.

I'd like to say a huge thank you to everyone who has contributed to the Children's Cancer PSP – the coordinating team and steering group members, our co-funders Little Princess Trust, our charity and professional association partners, and in particular to the childhood cancer patients, survivors, family members and professionals who shared their views.

While the publication of this report marks the end of the PSP process, I really hope it is just a new beginning, and that the research priorities we've identified here will go on to shape future research, for the benefit of all children diagnosed with cancer, now and in the future.

**Ashley Ball-Gamble**  
Chief Executive, Children's Cancer and Leukaemia Group



“ The Little Princess Trust is proud to have jointly funded this very important piece of work and to have been involved throughout the process.

The partnership has provided an invaluable source of information from the people who matter most: the children and young people whose lives are so cruelly affected by cancer.

While we are pleased to see that many of the priorities identified by the children and young people closely align with our charity's own aims and objectives, we can also see that there is still so much work that needs to be done.

This rewarding and yet sobering feedback therefore offers an essential insight to guide and motivate us all to carry on our vital work funding childhood cancer research.

**Phil Brace**, CEO of The Little Princess Trust



“ As a parent, I found the opportunity to be involved in the PSP a positive, tangible thing I could do following my son's death. Not having any kind of clinical or scientific background, when Rory was diagnosed with cancer, I found myself propelled into a world in which I felt I had no knowledge, context, or control. It is so important to me that parents feeling this way have a means by which they can realistically influence the future of childhood cancer care; to feel like they are being listened to.

**Amy Walsh**, Parent Representative on the Children's Cancer PSP Steering Group



“ I wanted to be involved with the PSP because of the exciting opportunity to contribute towards future research topics in childhood cancer, bringing the voice of childhood cancer survivors from a service user perspective and advocating for the cohort. I have found the experience to be extremely positive and engaging. I feel that my presence is valued, and my contributions have been acknowledged and implemented throughout the process. I am very grateful to be part of a group that is striving to improve the shape of future childhood cancer research.

**Alex Brownsdon**, Patient Representative on the Children's Cancer PSP Steering Group



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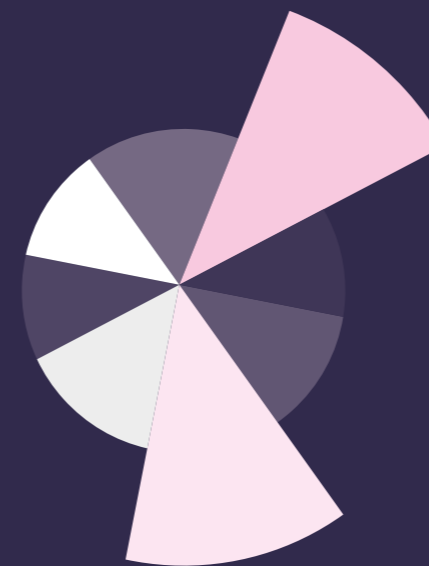
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## Abbreviations

CCLG:	Children’s Cancer and Leukaemia Group
GP:	General Practitioner
JLA:	James Lind Alliance
PSP:	Priority Setting Partnership

# Background

While research over the last four decades has dramatically increased the overall five-year-survival rate for all childhood cancers to around 84%<sup>1</sup> further research is needed to not only improve outcomes for all types of children's cancer, but to ensure all children and young people go on to live long, healthy and happy lives.

Topics of healthcare research in children's cancer are often driven by the interests of researchers and the pharmaceutical industry, meaning what is most important to children, their families and the professionals who care for them, may sometimes be overlooked.

In 2019, Children's Cancer and Leukaemia Group (CCLG) and The Little Princess Trust partnered with the James Lind Alliance (JLA) on a Priority Setting Partnership (PSP) to identify the research questions that are most in need of answering, according to those they matter to the most.

The JLA is a non-profit making initiative bringing together patients, carers and professionals in PSPs. The JLA PSPs identify and prioritise unanswered questions that they agree are the most important, so that researchers and research funders are aware of the issues that matter most to the people who could benefit from the research.

## Management and scope

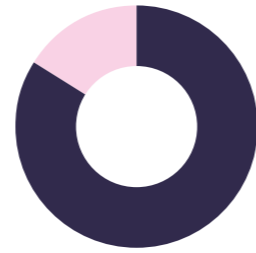
### Establishing the partnership

The project was funded by Children's Cancer and Leukaemia Group (CCLG) and the Little Princess Trust. Guidance on the costs involved in undertaking a PSP is available from the James Lind Alliance<sup>2</sup>. In this project, some of the funds were held by CCLG (relating to travel for steering group meetings and the final workshop); CCLG provided administrative support for the PSP, for example supporting minute-taking at the steering group meetings and dealing with travel claims. Other funds were held by the university at which members of the coordinating team were based. The coordinating team oversaw and directed the project to completion.

The coordinating team identified the range of professionals involved in the multidisciplinary treatment and care of children with cancer and aimed to reflect this breadth in the steering group, by including not just health professionals, but also professionals from education and the third sector. Parent representatives were invited to participate through CCLG social media channels. A patient representative was invited through the coordinating team. A few steering group members who were involved at the start of the project left midway. This is expected for projects which are carried out over a few years, as people's circumstances and time available to be involved changes. One parent representative left the steering group and three others joined.

<sup>1</sup> <https://www.cancerresearchuk.org/health-professional/cancer-statistics/childrens-cancers#heading-Two>

<sup>2</sup> <https://www.jla.nihr.ac.uk/jla-guidebook/chapter-3-/what-are-the-costs-involved-in-running-a-ssp.htm>



**84%**  
overall five-year-  
survival rate for all  
childhood cancers

# Steering group

Parent  
and patient  
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**Alex Brownsdon**



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**Loveday Langton**



**Andy Stewart**



**Amy Walsh**



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**Wendy Tarpsee-Morris**  
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**Martin English**  
Consultant Paediatric Oncologist, Birmingham Women's and Children's NHS Foundation Trust

**Penelope Hart-Spencer**  
Health Play Specialist, The Christie, Manchester

**Angela Polanco**  
Parent representative

# Involving children and young people

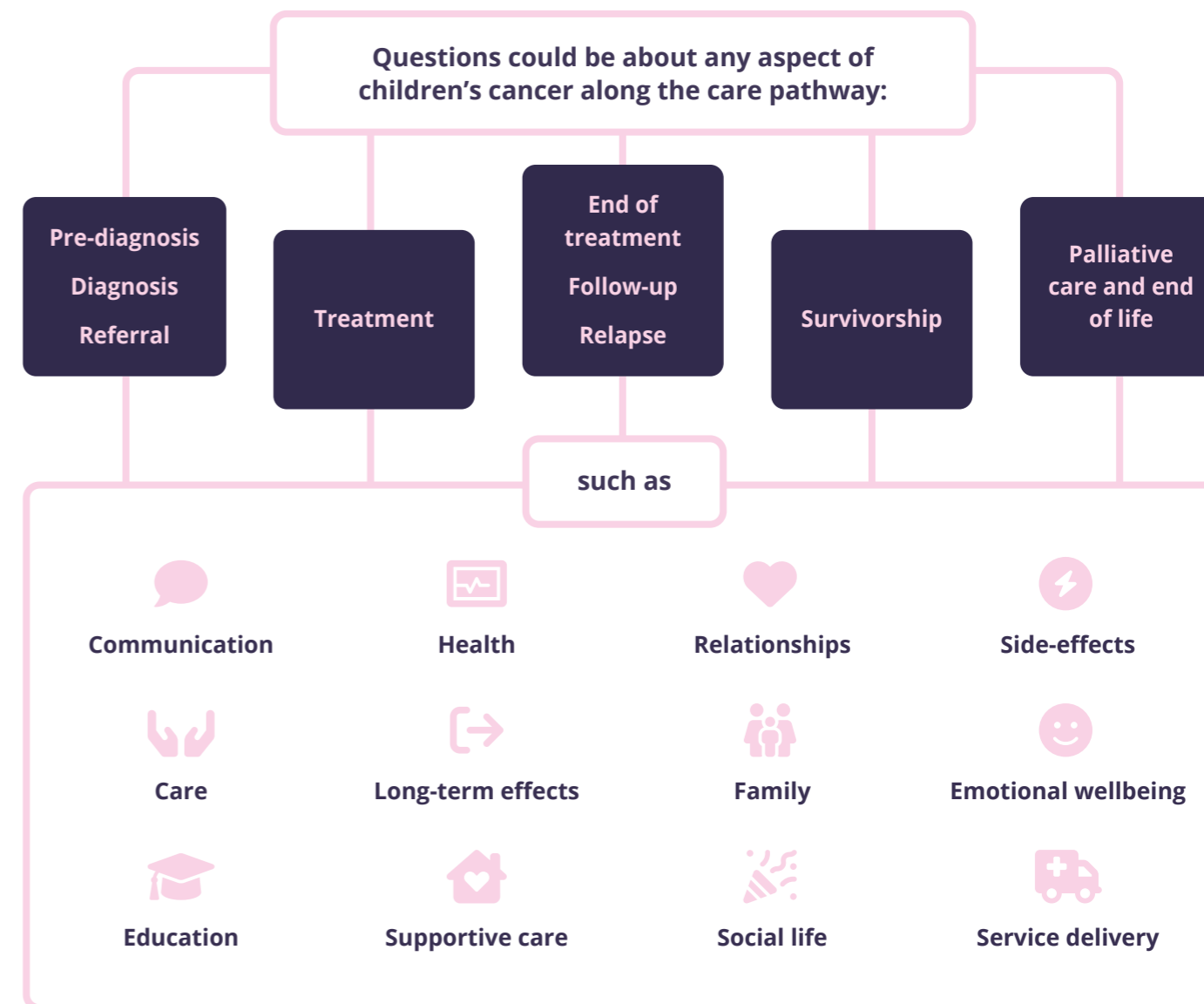
One of our primary goals in the Children's Cancer PSP was to find out from children, those under 16 years, what research we should do. In our Teenage and Young Adult Cancer PSP<sup>3</sup> we received very few responses from young people aged 13-15, and few from those who were still on treatment. Previous PSPs have sought to involve children and young people, but in the final reporting it is evident that few children, especially young children, had been engaged through the process (Postma et al. 2022). We recognised the challenges of engaging with these populations, in terms of reach and accessibility of the information. At the outset of the Children's Cancer PSP, we wanted to invest time, resources, and energy in anticipating and resolving any challenges that could impact on participation.

## The Scope

The aim of the Children's Cancer PSP was 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 scope of the Children's Cancer PSP was intentionally broad and included questions about:

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



As noted opposite, the previous Teenage and Young Adult Cancer PSP (Aldiss et al. 2019) included young people who had cancer aged 13 and over, but very few responses were received from those aged 13 to 15; therefore, the steering group decided to include this age group in the Children's Cancer PSP.

## Partners

The steering group identified potential partner organisations. This was achieved through peer knowledge, consultations, steering group member's networks and the project funder's networks. Potential partners were contacted about the Children's Cancer PSP and asked to join the partnership and contribute to disseminating surveys and results through their contacts and networks. The partner list can be seen in Appendix 1 on page 44. Partners included charities working with children with cancer and their families and organisations representing the professionals who work with them.

## Process

The Children's Cancer PSP followed the methodology described in the JLA Guidebook<sup>4</sup>. The full protocol is available here: [www.jla.nihr.ac.uk/priority-setting-partnerships/childrens-cancer/](https://www.jla.nihr.ac.uk/priority-setting-partnerships/childrens-cancer/) An overview of the process is shown in Appendix 2 on page 45.

<sup>3</sup> <https://www.jla.nihr.ac.uk/priority-setting-partnerships/teenage-and-young-adult-cancer/downloads/Teenage-and-Young-Adult-Cancer-PSP-final-report.pdf>

<sup>4</sup> <https://www.jla.nihr.ac.uk/jla-guidebook/>

## Stage 1a

# Gathering the questions

Questions were gathered in an online public survey<sup>5</sup> which was launched on 9th September 2020 and closed on 8th January 2021. The following groups of people were invited to participate:

- People diagnosed with cancer before their 16th birthday;
- Relatives/friends/partners/carers of someone who has been diagnosed with cancer before their 16th birthday;
- Professionals involved in diagnosing or treating children who have cancer or had cancer under the age of 16;
- Professionals involved in the care of children who have cancer or had cancer under the age of 16 and/or their families.

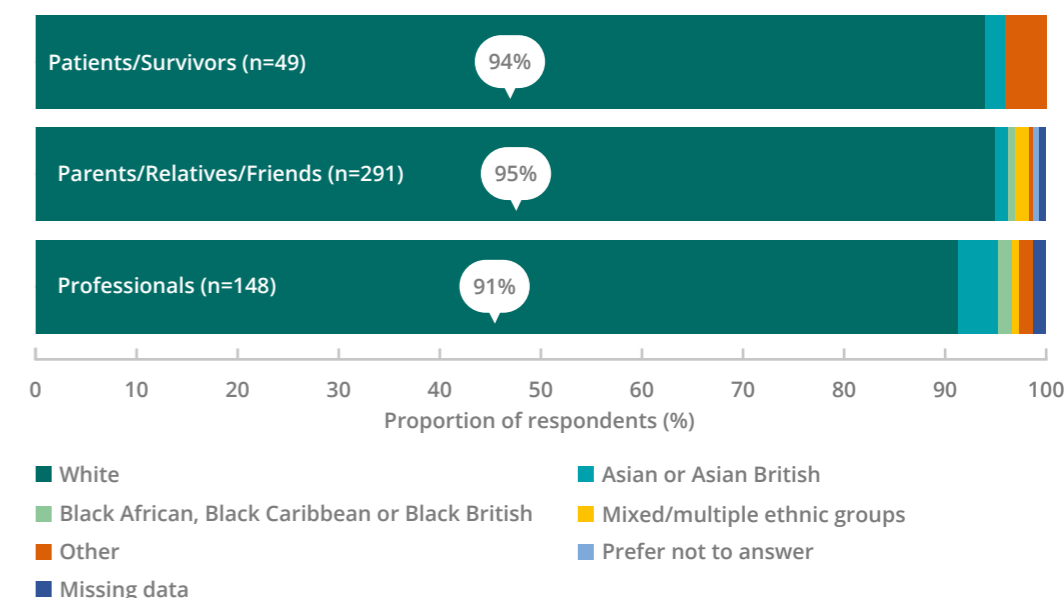
The survey was built using Qualtrics online software. The wording and design of the survey was piloted with eight adult survivors of childhood cancer, nine parents and two professionals outside the steering group and adapted to incorporate their feedback. On the day of the survey launch, a press release was issued and the partner organisations were notified that the survey was open. Some of these partner organisations added a link to the survey on their website, mentioned the project in their newsletter/on social media or sent an email to their members to alert them. Social media was used throughout the four-month period to promote the survey through CCLG's channels.

Respondents were invited to submit up to eight questions about any aspect of children's cancer they considered to be important and unanswered. Basic demographic data were requested.

5 <https://www.jla.nihr.ac.uk/priority-setting-partnerships/childrens-cancer/>

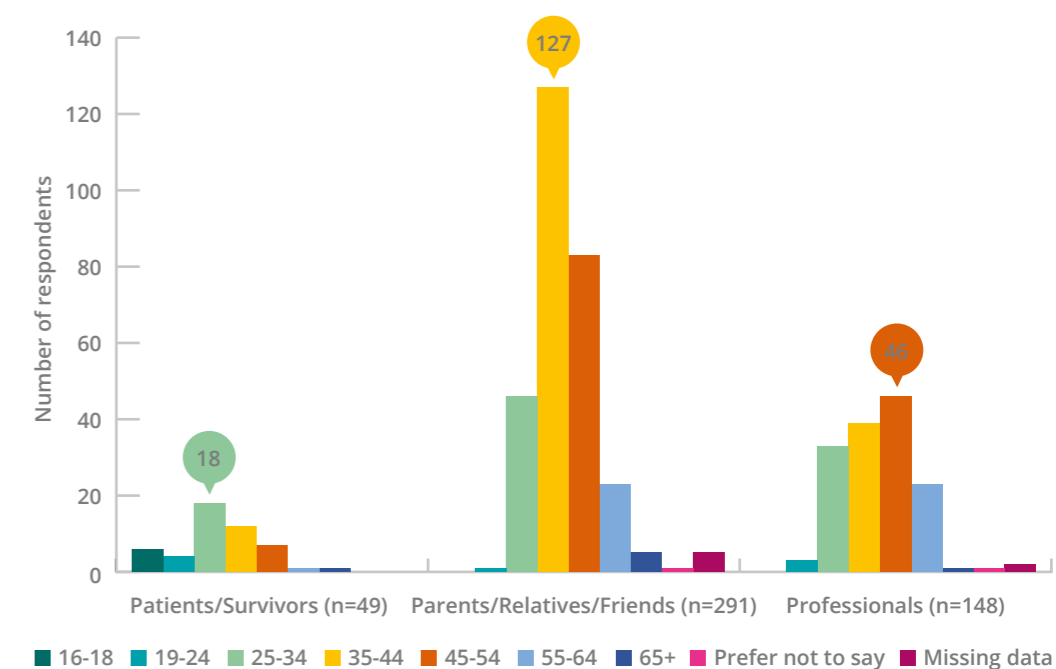
**488**  
responded to  
the survey

Figure 1  
Ethnicity of  
respondents  
(initial survey)



The majority of patients/survivors answering the survey were aged 25-34 (37%, n=18), followed by those aged 35-44 (24%, n=12). For parents/relatives/friends the majority were aged 35-44 (44%, n=127), followed by 45-54 (29%, n=83). Most professionals were aged 45-54 (31%, n=46). See Figure 2.

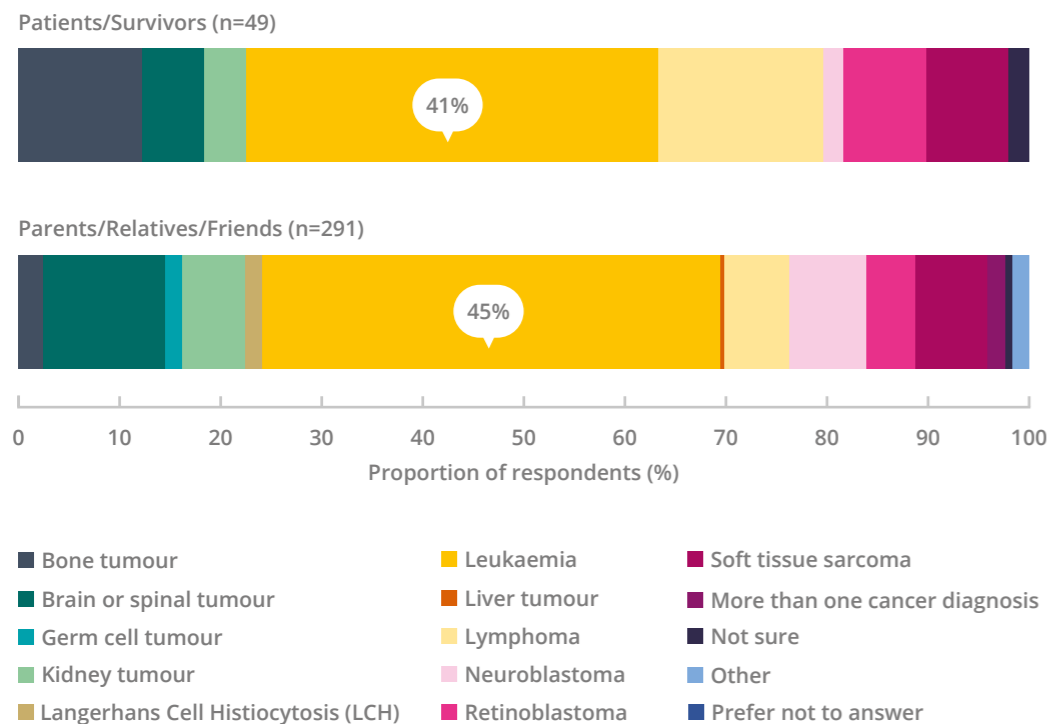
Figure 2  
Age, in years,  
of respondents  
(initial survey)





The geographical distribution of respondents was broadly similar to the proportion of incidence cases from the three devolved nations in the United Kingdom: the majority of children are diagnosed in England, followed by Scotland, Wales and Northern Ireland<sup>6</sup>. Most respondents in each group were from England (patients/survivors 73%, n=36; parents/relatives/friends 83%, n=241; professionals, 83%, n=123). For patients/survivors 20% (n=10) selected 'Other' for their country of residence, 4% (n=2) were from Scotland and 2% (n=1) selected 'Prefer not to answer'. Some survivors left a comment stating that they lived in the UK when they were diagnosed/treated and had since moved elsewhere. Seven percent (n=20) of parents/relatives/friends selected 'Other' for their country of residence, 4% (n=13) were from Wales, 4% (n=12) from Scotland, 1% (n=4) from Northern Ireland and 0.3% (n=1) selected 'Prefer not to answer'. For professionals, 5% (n=8) selected 'Other' as their country of work, 5% (n=7) worked in Scotland, 4% (n=6) in Wales and 2% (n=3) in Northern Ireland (missing data 1%; n=1).

The spread of respondents with different cancer types was broadly similar to those occurring in this age group (Figure 3). The largest group of respondents for patients/survivors and parents/relatives/friends were those who had leukaemia/had a child with leukaemia (41%, n=20 and 45%, n=132); leukaemia represents around a third of all newly diagnosed cases in this age group<sup>7</sup>. Brain and spinal tumours were underrepresented, 6% (n=3) of patients/survivors and 12% (n=35) of parents/relatives/friends; the incidence rate for brain and spinal tumours is 26%<sup>7</sup>; this may reflect the lower survival rates in this group and the long-term cognitive effects of these tumours and their treatments.

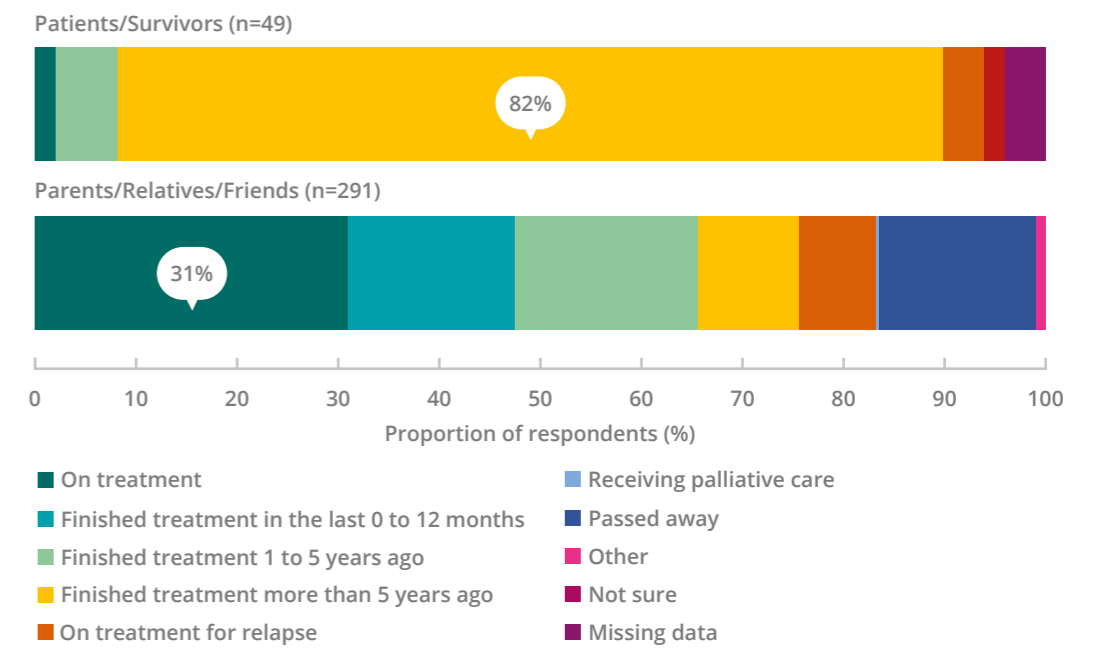


**Most respondents in each group were from England, followed by Scotland, Wales and Northern Ireland**

**Figure 3**  
Cancer distribution in patients/survivors and parents, friends and relatives (initial survey)

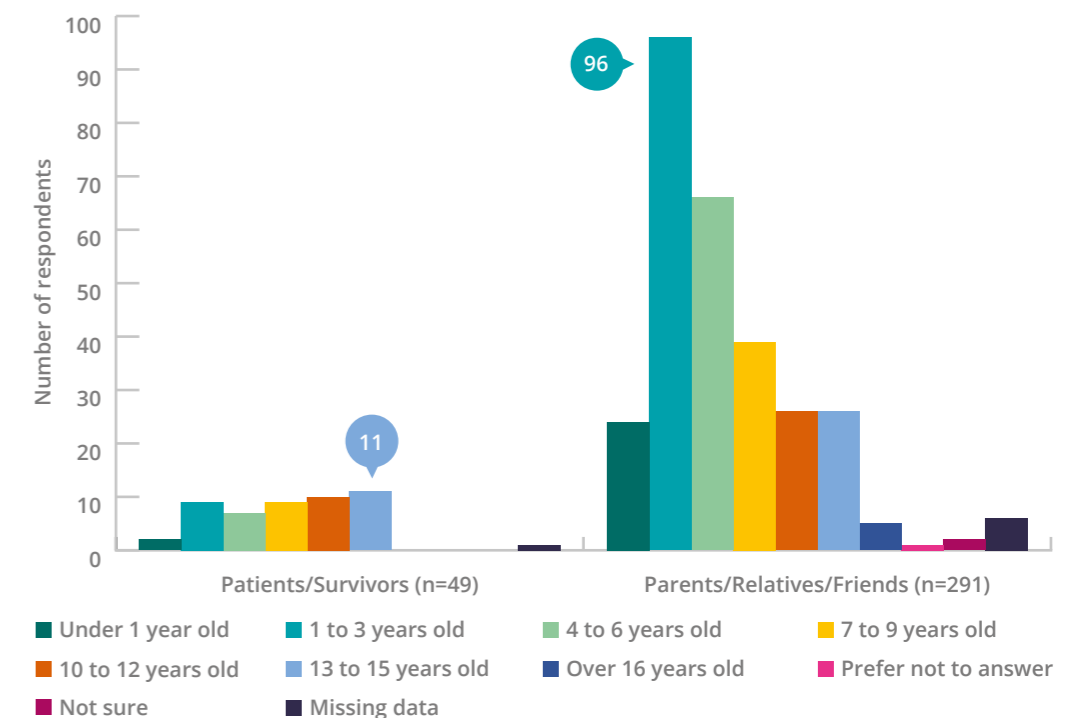
**Figure 4**  
Cancer timeline of patients/survivors and parents, friends and relatives (initial survey)

The steering group identified several time points for people to indicate what best described themselves/the child (Figure 4). Differences were observed between the two groups. Most patients/survivors, 82% (n=40), described themselves as 'Finished treatment more than 5 years ago'. For just under one third (31%, n=90) of parents/relatives/friends who responded, the child was 'On treatment'.



For patients/survivors, there was an even spread across the ages at which they had been diagnosed (Figure 5). For parents/relatives/friends, most of the children had been diagnosed at a younger age, between 1 and 6 years of age. The highest incidence rates for all children's cancers combined are in the under-fives, with almost half (45%) of all cases in children being diagnosed in this age group (UK, 2016-2018)<sup>8</sup>. Five parents (2%) indicated that their child was over 16 at diagnosis, the questions they submitted were later removed as 'out of scope' as this PSP focuses on children diagnosed before their 16th birthday.

**Figure 5**  
Age at diagnosis, in years, of survivors and parents, friends and relatives (initial survey)

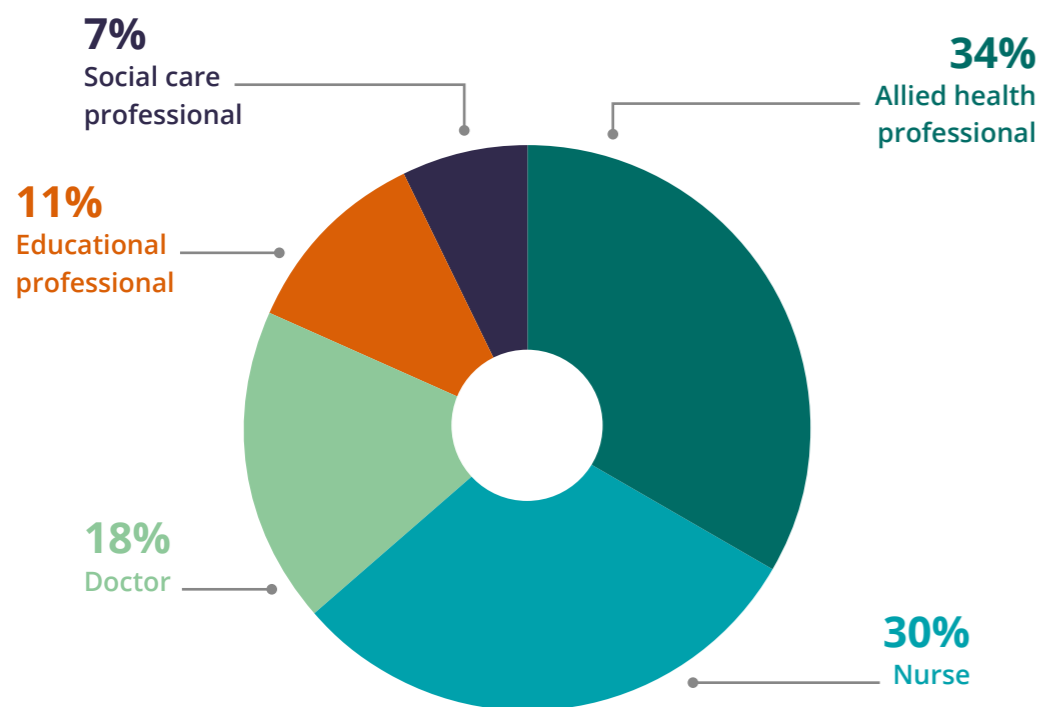


<sup>6</sup> [http://ncin.org.uk/cancer\\_information\\_tools/eatlas/links\\_to\\_useful\\_data\\_sources](http://ncin.org.uk/cancer_information_tools/eatlas/links_to_useful_data_sources)  
<sup>7</sup> <https://www.cclg.org.uk/types-of-childhood-cancer>

<sup>8</sup> <https://www.cancerresearchuk.org/health-professional/cancer-statistics/childrens-cancers/incidence#heading-One>



A broad range of professionals responded to the initial survey, as would be expected considering the multidisciplinary care of children with cancer. Figure 6 illustrates the distribution between medical, nursing, allied health professionals, social care professionals and educational professionals. Allied health professionals included physiotherapists, dietitians, clinical psychologists, occupational therapists, and health play specialists. The majority of doctors worked in tertiary care (78%, n=21), 19% (n=5) worked in secondary care and none in primary care (4%, n=1 doctor responded 'Other'). Most nurses were children's cancer nurses (67%, n=30), 16% (n=7) were children's community nurses and 11% (n=5) were children's nurses (7%, n=3 nurses responded 'Other').



◀ **Figure 6**  
Distribution of professionals (initial survey)

## Stage 1b

# Gathering questions from children and young people



A subgroup was set up which included members of the steering group who wanted to be involved in the PSP's engagement with children. The subgroup consisted of two researchers, a teacher, doctor, health play specialist, parent, clinical psychologist, and charity representative. At the start of the project, our plan was to run a series of face-to-face workshops with children to collect research questions and have each Principal Treatment Centre in the UK involved in publicising the project to families within their care and helping to collect questions. These plans changed with the pandemic which meant that in-person group work was not possible until the final workshop in the PSP process. Drawing on experiences from other PSPs that had sought to involve children, face-to-face methods seemed to have been most successful<sup>9,10</sup>. We learnt from the Learning Difficulties PSP<sup>10</sup>, that some children can find it difficult to understand what is meant by research or how to phrase a question.

It was clear that we needed to produce some engaging materials that would help children to understand the process. We planned for hospital school staff to work with children to complete the survey while they were in hospital or in the community. The teacher on our sub-group worked with other teachers to produce school lesson plans for children and young people at different key stages in the national curriculum. The lesson content focused on explaining to children about research, engaging them in thinking about what matters to them and what questions about children's cancer they would like to see answered by research, ending with completion of the survey. This approach was piloted in one cancer treatment centre, but it quickly became clear that this was not working. Although a few children did participate in the lessons and completed the survey, feedback from hospital school staff was that they were finding the lessons difficult to deliver, as there never seemed to be a good time. They felt that lessons in hospital were a time when children did not focus on their cancer and so asking them to think about their cancer experience and complete the survey did not feel appropriate. It was decided that the best way to reach children would be through their parents/carers, with some additional help from professionals to promote the survey to families. We wanted to help parents to explain the project and survey to their child(ren) and thought that animations would be a good way to do this.



9 <https://www.jla.nihr.ac.uk/priority-setting-partnerships/Mental-health-in-children-and-young-people/index>  
10 <https://www.jla.nihr.ac.uk/priority-setting-partnerships/learning-difficulties-scotland/>

We looked for an animator with previous experience of explaining research projects to children and young people. One of our steering group members had worked with an animator from ScienceSplained ([www.sciencesplained.com](http://www.sciencesplained.com)) to do this previously. We decided to make two different cartoons, one for younger<sup>11</sup> and one for older children<sup>12</sup> that would allow audiences to self-select what looked most applicable to them. There was already an animation about the PSP process on the JLA website that was appropriate for young people<sup>13</sup>. The ideas for the cartoons were worked up by the subgroup along with the animator and the scripts were checked by children and young people that the 'stories' made sense to them.

Three different versions of the surveys were built using Qualtrics online software, aimed at children and young people of different ages (4-7 years, 8-12 years and 13-15 years)<sup>14</sup>. Children and young people were invited to complete whichever survey version they preferred. The surveys varied in the complexity of language used in the introduction section and questions, and the surveys for older children and young people contained more questions seeking demographic information. After discussion with parents on the steering group, the word 'cancer' was not used in the survey or animation for younger children as they said that this would give flexibility for parents to use the words their child is familiar with when helping their child to complete the survey. The surveys were piloted with children and young people.

The surveys were launched on 6th September 2021 and closed on 16th November 2021. The following groups were invited to participate:

- **Children and young people diagnosed with cancer before their 16th birthday;**
- **Children and young people who have a brother or sister with cancer now or who had cancer when they were younger;**
- **Children and young people who have a friend with cancer now or who had cancer when they were younger.**

Respondents were invited to submit up to eight questions/topics about any aspect of children's cancer they considered to be important. The surveys were promoted through the PSP's Partner organisations, social media and posters were sent to all Principal Treatment Centres in the UK.

## Children and young people's survey results

74 children and young people answered the surveys, three did not submit any questions and were removed from the analysis. A total of 71 respondents submitted 252 questions/topics. 61 respondents were children and young people who had experienced cancer (aged 3-21) and ten were siblings (aged 4-19). No friends participated. 43 (61%) participants were female and 27 (38%) were male, one respondent selected, 'Prefer not to answer'.

11 <https://www.youtube.com/watch?v=O492QZ1myko&t=72s>

12 <https://www.youtube.com/watch?v=pRaRuMr7ol0>

13 The PSP Process – <https://www.jla.nihr.ac.uk/about-the-james-lind-alliance/>

14 <https://www.jla.nihr.ac.uk/priority-setting-partnerships/childrens-cancer/>

## 71 children and young people responded to the surveys

Children who completed the survey for those aged 8-12 were asked if they were on or off treatment, eight children were on treatment and 10 had finished treatment; for siblings, two children had a sibling who had finished treatment and one was on treatment.

Young people completing the survey for those aged 13-15 were asked what best described their current situation: four were currently on treatment, three had finished treatment in the last 12 months, five had finished treatment one to five years ago, five had finished treatment more than five years ago and one respondent selected 'Other'. Two siblings responded that their sibling was on treatment and two that their sibling had finished treatment over five years ago. This group were also asked their age at diagnosis – they were aged under one year to 14 years at diagnosis. For siblings, their sibling was aged two to four years at diagnosis.

The majority of children and young people responding to the survey were from England (68%, n=48). Fifteen percent (n=11) of children and young people were from Scotland, 11% (n=8) were from Wales, 3% (n=2) answered 'Other', 1% (n=1) was from Northern Ireland and 1% (n=1) selected 'Prefer not to say'.

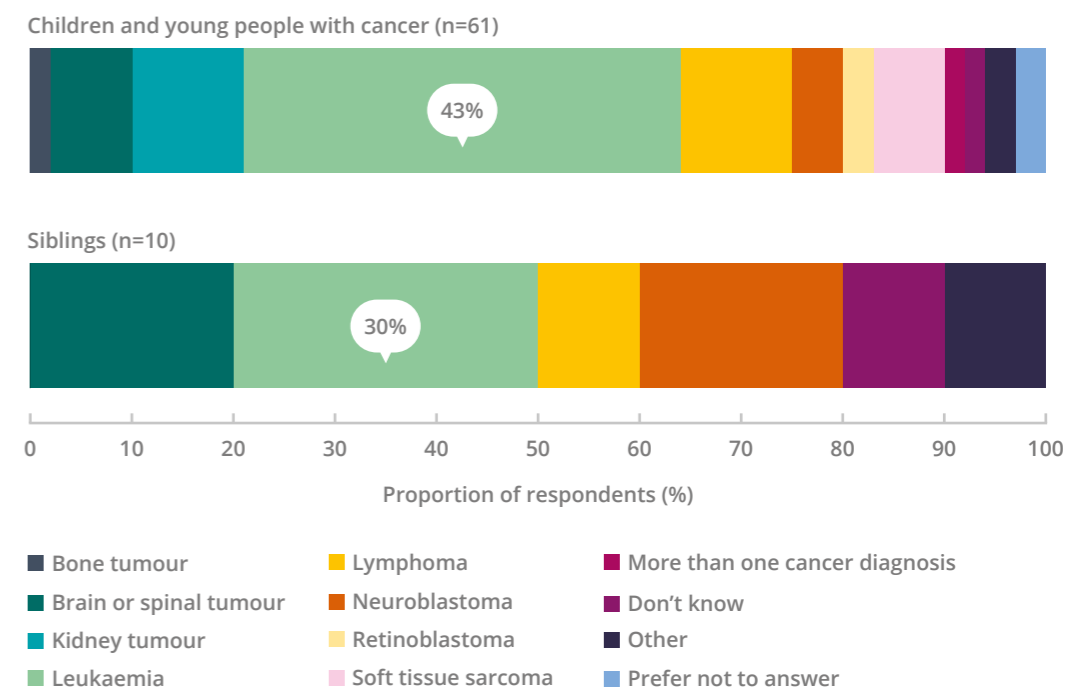
Children and young people answering the surveys aimed at those aged 8-12 and 13-15 were asked about their ethnicity (n=43). The majority of respondents were White (88%, n=38), with one respondent (2%) from each of the following ethnic groups: Asian or Asian British; Black African, Black Caribbean or Black British; Mixed/multiple ethnic groups. Two respondents (5%) selected 'Prefer not to say'.

Similar to the respondents in the initial survey, the most common diagnosis for children and young people and siblings responding was leukaemia (43%, n=26 and 30%, n=3; Figure 7).



Respondents were invited to submit up to eight questions /topics about any aspect of children's cancer they considered to be important.

Figure 7 ► Cancer distribution of respondents to the children and young people's surveys



## Stage 2a

# Refining the questions

All the submitted questions were extracted from Qualtrics into an Excel spreadsheet. Multiple questions written in the same box were separated. Comments sections were checked for further questions. In total, 1299 questions were submitted. For brevity, we refer to the submissions as 'questions'; in reality many submissions were not written as questions, some were broad topics for research and others were people's experiences indicating what was important to them.

## Organising the questions

An initial coding of the questions was carried out by coordinating team member Susie Aldiss (SA), with support from Faith Gibson (FG). Questions were grouped into themes to make them easier to review and discuss. The themes were:

- |                                    |                                        |                                           |
|------------------------------------|----------------------------------------|-------------------------------------------|
| 1. Causes of cancer and prevention | 9. Physio, exercise and rehab          | 17. Education                             |
| 2. Genetics                        | 10. Nutrition                          | 18. End of life care and bereavement      |
| 3. Research and funding issues     | 11. Covid-19                           | 19. Communication and information sharing |
| 4. Incidence and statistics        | 12. Service delivery                   | 20. End of treatment                      |
| 5. Tests and imaging               | 14. Side-effects and management        | 21. Survivorship                          |
| 6. Diagnosis and early detection   | 15. Treatment                          | 22. Long-term follow-up care              |
| 7. Environment of care             | 16. Psychosocial wellbeing and support | 23. Long-term effects                     |
| 8. Fertility                       |                                        |                                           |

Some questions were coded in more than one category. Once all questions had been coded, those in the same category were grouped together and categories separated into different tabs within the Excel spreadsheet to assist with data management.



**1299**  
questions were submitted



**139**  
were identified as out of scope

## Removing out of scope questions

During the coding, SA and FG identified questions that were potentially 'out of scope'. The following criteria were used to identify out of scope questions:

- 1. Ambiguous questions** – where what the person is asking is unclear, were interpreted in different ways by steering group members and the meaning could not be resolved following discussion (e.g. 'Remaining scar tissue', 'How research is going').
- 2. Questions not answerable by research** – (e.g. 'Why does paediatric cancer research receive so very little funding?', 'Who is present when you give the diagnosis').
- 3. Questions submitted by people outside our age range** – by someone whose experience was not of childhood cancer as defined by our project scope, there were a few parent respondents whose child was over 16 at diagnosis. These questions were checked to verify that all the themes within them had been covered by 'in scope' questions.

Responses from people outside the UK were reviewed to check they were relevant to the UK setting; all these submissions were relevant and so were included. Also, for overseas responses from parents, relatives, and survivors it was not possible to determine whether they had been diagnosed/treated in the UK and now moved overseas, so the steering group agreed these responses should be kept.

Identification of out of scope questions was an iterative process. All out of scope questions were checked and agreed by the steering group. In total, 139 questions were identified as out of scope. There is commitment from the project funders to review and consider how to make the best use of these questions.

## Formatting questions

SA worked through the categories to further group similar questions together and form summary questions. The aim was to retain the sense of what the respondent meant, but in the form of a clear question. FG supported SA with this process.

Steering group members met online in small groups to review the summary questions, to confirm the grouping of questions together, and the wording of each summary question. Steering group members were given the list of categories of questions and asked to choose a group that they felt was most aligned with their experience and expertise. Each of the six groups was led by a member of the coordinating team/James Lind Advisor (SA, RH, FG, BP, JG). Each group met twice to review their question list, with further checking undertaken via email. Changes were made to the wording of summary questions, to the allocation of questions which formed each summary question, and some questions were moved to other categories (particularly where there was overlap, to avoid duplication). The steering group then reviewed the whole list of summary questions.

For example, 10 questions were submitted in the survey about supporting families when giving information about relapse and treatment options including:

- What do we do if there's a relapse?
  - How likely is relapse? What happens if it happens?
  - What happens if it comes back after treatment?
  - What do I need to be aware about regarding relapse and long-term side-effects of treatment?
  - How best to support parents through multiple relapses when there are
- increasing early phase research/treatment options available worldwide.
  - Supporting families and patients during relapse to ensure they feel fully supported and cared for. Understanding what families need to enable them to cope with the challenges they face during treatment. It is very difficult to support families as there are so many areas of concern.
  - How do you support a child/family at relapse.

These questions were all incorporated into the summary question:

**What are the best ways to support families when giving information about a) the possibility of relapse and b) treatment options when relapse has happened?**

This stage resulted in 108 summary questions.

## Searching for evidence

A search strategy was produced and agreed with the steering group. Searches were limited to evidence published in the last five years (since January 2017) to ensure evidence was up to date. Only publications which brought evidence from multiple studies together (such as systematic reviews and qualitative meta-synthesis) were considered. As the evidence may not be restricted to cancer in children aged 0 to 15 years at initial diagnosis, evidence which included participants between these ages along with older participants was also considered. When discussing this evidence, the steering group considered whether further work focusing specifically on children aged 0 to 15 years at initial diagnosis was needed. For questions where we were aware that older evidence had answered a question and further research or an update may be unnecessary, the steering group agreed that a discussion would take place with a consensus reached on whether the question has been answered. Searches were also carried out for ongoing studies. This involved personal communication with experts in the field and steering group knowledge of current research.

Searches were carried out by SA between January and May 2022. For many questions, no reviews were identified. In some cases, the identified reviews only partly answered the question; these questions were recorded as unanswered. When any evidence was found, it was initially reviewed by SA. SA sought second opinions from the steering group/coordinating team members within their area of expertise for any studies where it was unclear whether the review(s) fully answered the question. Some questions were too broad to conduct a comprehensive search (e.g. Why do treatment strategies differ between countries and what difference does this make to outcomes?). These questions were discussed with the steering group who agreed they were unanswered. Four questions were identified as already answered (see Appendix 3). All were discussed with the steering group to ensure agreement that the question had been answered. Three questions were the focus of studies currently underway (see Appendix 4). 101 unanswered questions remained.

## Stage 2b

# Refining the questions from children and young people



**252**  
questions were submitted

The same process was followed for refining the questions from the children and young people's surveys as had been followed for the initial survey. All the submitted questions were extracted from Qualtrics into an Excel spreadsheet. Multiple questions written in the same box were separated. In total, 252 questions were submitted. 34 questions were from siblings. For brevity, we refer to the submissions as 'questions'; nearly all of the submissions were not written as questions as children and young people were invited to write what was important to them.

## Organising the questions from the children and young people's surveys

An initial coding of the questions was carried out by coordinating team member SA, with support from FG. The questions were grouped into themes to make them easier to review and discuss. The themes were:

- |                                                     |                        |                                  |
|-----------------------------------------------------|------------------------|----------------------------------|
| 1. Impact on life                                   | 4. Hospital experience | 8. Friends                       |
| 2. Treatment                                        | 5. Emotional impact    | 9. Information and communication |
| 3. Being poorly, side-effects and long-term effects | 6. Education           | 10. Siblings                     |
|                                                     | 7. Family              |                                  |

Some questions were coded in more than one category. Once all the questions had been coded, questions in the same category were grouped together and categories separated into different tabs within the Excel spreadsheet to assist with data management.

13 questions were identified as out of scope and removed, as they were unrelated to cancer or were unclear, examples include, 'cost to hospital', 'wildlife' and 'meeting new people'. These submissions were checked and agreed by subgroup members.

## Formatting questions from the children and young people's surveys

SA worked through the categories to further group similar questions together and form summary questions. The aim was to retain the sense of what the respondent meant, but in the form of a clear question. FG supported SA with this process.

The subgroup met online to review the summary questions with further checking undertaken via email until agreement was reached. This stage resulted in 24 summary questions. Many children responded that their family, friends, and pets were important to them, but it was unclear what it was about these aspects that were important (several responses were one or a few words, such as 'family', 'mum and dad', 'seeing friends'). The subgroup decided that it would be wrong to guess or presume what children meant and further consultation with children and young people was planned. We tried to hold two online workshops for children and young people to ask them about what was important to them about family, friends and pets but were unable to recruit enough participants to do this. Consequently, this discussion took place as part of the final workshop with children and young people where the summary questions would be prioritised.



**24**   
summary  
questions  
were agreed on



### Stage 3

## Prioritising the questions

### Preparation for the shortlisting survey

The steering group discussed whether to take all 101 unanswered questions to the shortlisting survey or whether to shorten the list to make the survey quicker to complete, as some other PSPs have done. The consensus was that the group did not want to remove any questions at this stage and wanted them all to go out to be considered in the public vote.

To ensure that all questions going out to the public were in an understandable form, they were reviewed by patient and parent members of the steering group and a Health Information Executive from one of the funding charities. Questions were simplified following this review and definitions of words that may not be easy to understand were added.

### Shortlisting survey

The shortlisting survey was created using Qualtrics online survey software and launched on 3rd August 2022; it was open until 30th September 2022. Responses were invited from the same groups of people as the initial survey. The opportunity to take part was publicised through the same partner organisations as the initial survey and on social media. Everyone who had requested to stay involved in the project and provided their email address in the initial survey was sent the survey link directly.

Respondents were invited to read the 101 questions (Appendix 5) and select the questions that were important to them. The aim of this stage was to filter out some of the questions to shorten the list. The questions the respondent selected were added to their own personal 'shortlist' ready for them to make their final selection of up to 15 questions.

Questions on similar topics were grouped into sections. Each respondent was presented with the sections, and questions within each section, in a random order to minimise the chance of survey fatigue influencing choice. The sections were:

1. Side-effects and management
2. Treatment
3. Education
4. Physical activity, play and therapies
5. Long-term effects and follow-up care
6. Communication and information sharing
7. Psychological and social wellbeing
8. Food and nutrition
9. Healthcare delivery
10. Causes of cancer, diagnosis and research

Respondents  
were invited  
to read the  
**101**  
questions  
and select the  
questions that  
were important  
to them

## Shortlisting survey results

327 people participated. Demographics of those responding can be seen in Figures 8 to 13. Similar to the initial survey, the largest group of respondents was parents/relatives/friends (64%, n=210; made up of 197 parents, 10 relatives and three friends, no partners), followed by professionals (28%, n=90) and patients/survivors (8%, n=27).

Across all groups more females responded than males; 85% (n=23) of patients/survivors, 89% (n=186) of parents/relatives/friends and 83% (n=75) of professionals responding were female. An even lower proportion of males responded to the shortlisting survey compared to the initial survey in the patient/survivor (11%, n=3) and professional groups (16%, n=14). The proportion of males in the parents/relatives/friends group was the same across both surveys (10%), however taking into account the smaller number of respondents in the second round the actual number of males responding had decreased (n=21).

The ethnic distribution of those answering the shortlisting survey was similar to the initial survey, with under representation of Black and minority ethnic groups (Figure 8).

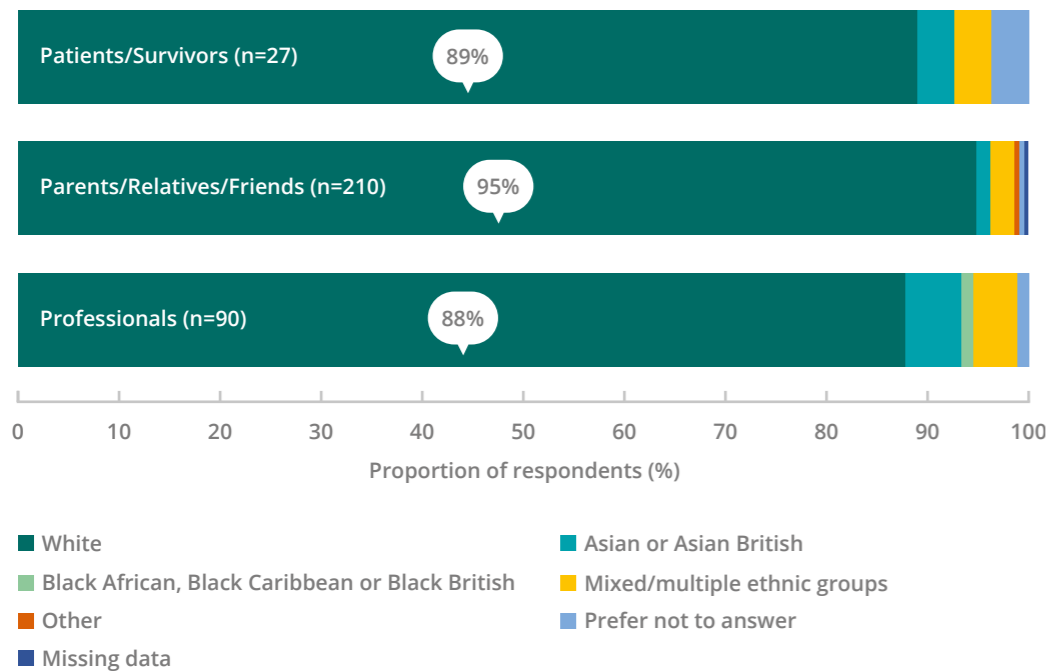


Figure 8 Ethnicity of respondents (shortlisting survey)

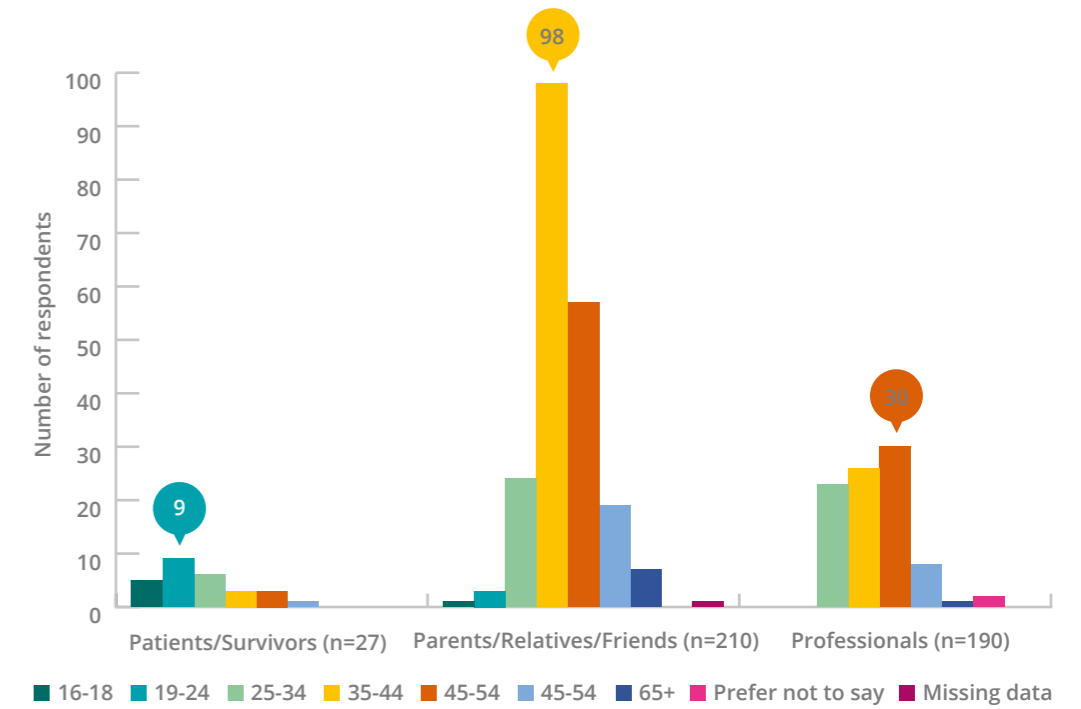
The age distribution of parents/relatives/friends answering the shortlisting survey was similar to that observed in the initial survey with the majority of respondents aged 35-44 (47%, n=98; Figure 9). Respondents in the patients/survivors' group for the shortlisting survey tended to be younger than those completing the initial survey, four young people aged 19-24 participated in the initial survey whereas nine in this age group completed the shortlisting survey.

The geographical distribution of respondents was similar to the initial survey, with most living/working in England (patients/survivors 93%, n=25; parents/relatives/friends 81%, n=170; professionals, 87%, n=78). For patients/survivors there were notably fewer selecting the 'Other' category for their country of residence in the shortlisting survey than in the initial survey, 4% (n=1) selected 'Other' for their

**327**  
people participated

Most respondents were living/working in England

Figure 9 Age, in years, of respondents (shortlisting survey)

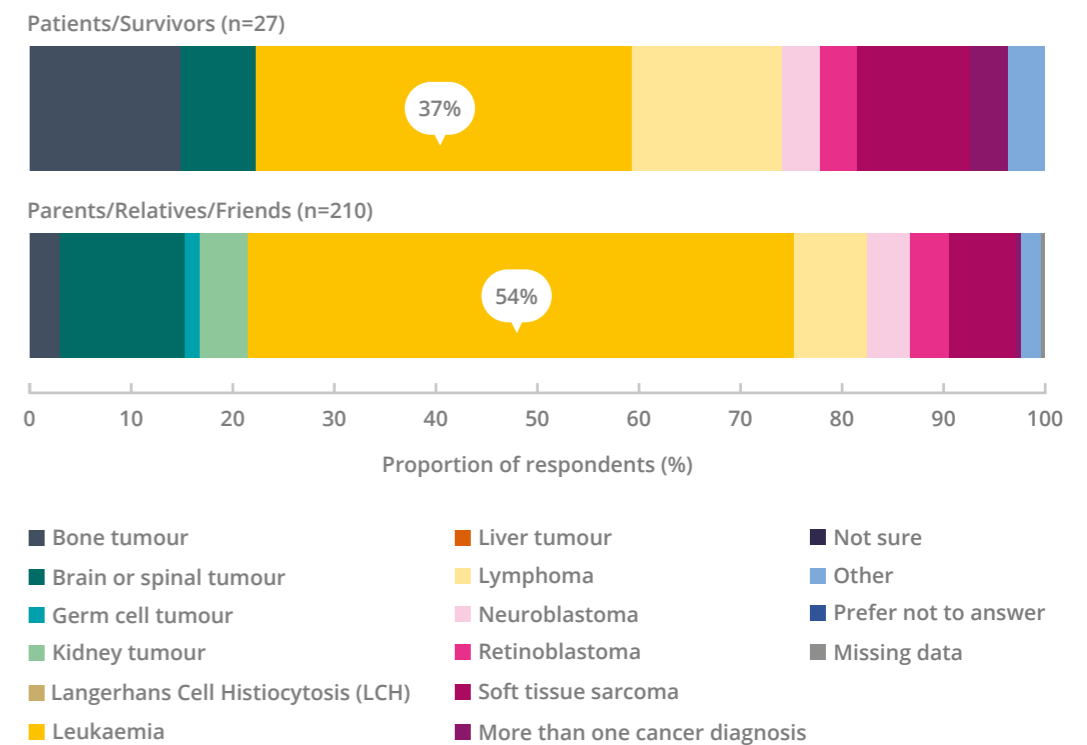


country of residence, and 4% (n=1) selected 'Prefer not to answer'. Eight percent (n=17) of parents/relatives/friends were from Scotland, 5% (n=10) were from Wales, 5% (n=10) selected 'Other' for their country of residence, 1% (n=2) were from Northern Ireland and 0.5% (n=1) selected 'Prefer not to answer'.

For professionals, 7% (n=6) worked in Scotland, 3% (n=3) in Wales and 1% (n=1) in Northern Ireland, 1% (n=1) selected 'Other' as their country of work and 1% (n=1) selected, 'Prefer not to answer'.

The types of cancer that respondents in the shortlisting survey had experienced was similar to those in the initial survey (Figure 10). Again, the largest group of respondents for patients/survivors and parents/relatives/friends were those who had leukaemia/had a child with leukaemia (37%, n=10 and 54%, n=113).

Figure 10 Cancer distribution in patients/survivors and parents, friends and relatives (shortlisting survey)



Where parents/relatives/friends reported the child to be on the cancer timeline was also similar to the initial survey. For patients/survivors, a higher proportion of respondents were on treatment or finished treatment more recently than the initial survey respondents (Figure 11).

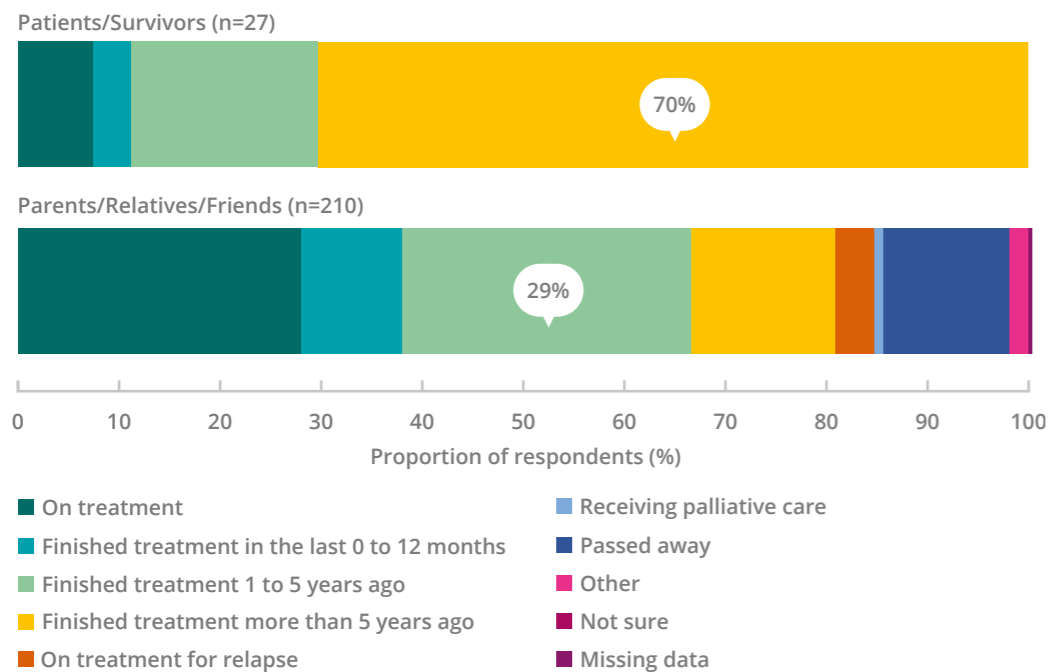


Figure 11  
Cancer timeline of patients/survivors and parents, friends and relatives (shortlisting survey)

For patients/survivors, the largest group of participants were diagnosed between age 13 and 15 (37%, n=10). For parents/relatives/friends, as in the initial survey, most of the children had been diagnosed at a younger age, between 1 and 6 years (Figure 12). One parent indicated that their child was over 16 at diagnosis, their response was removed from the analysis. Two patients/survivors were aged 16 at diagnosis, their responses were included due to the low numbers of patients/survivors responding.

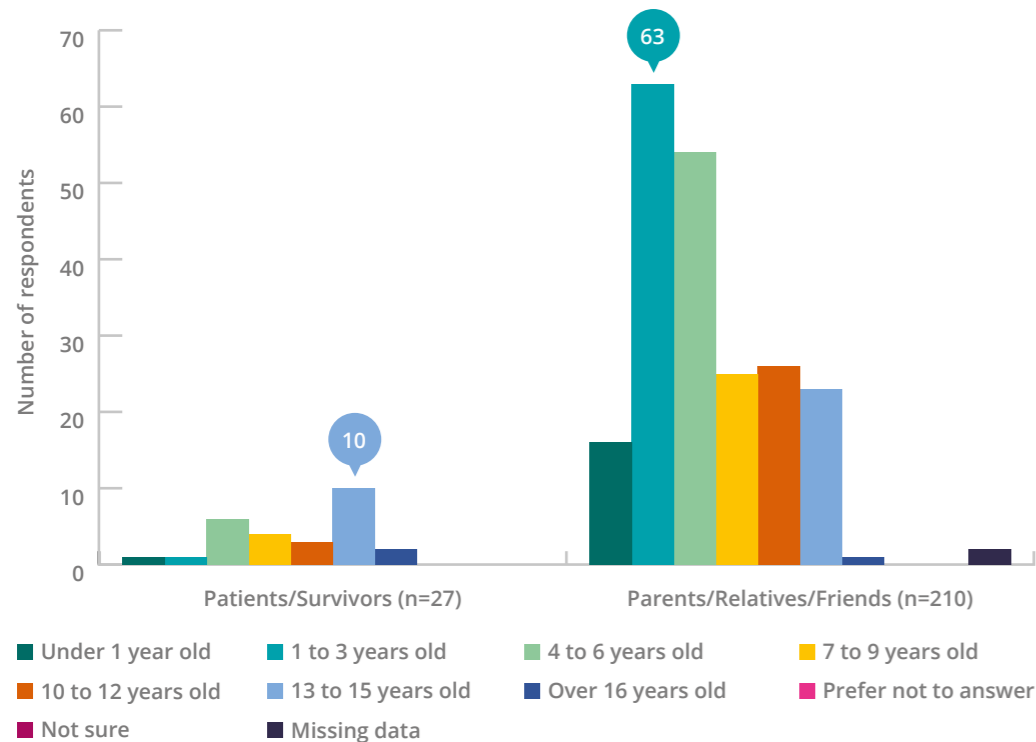
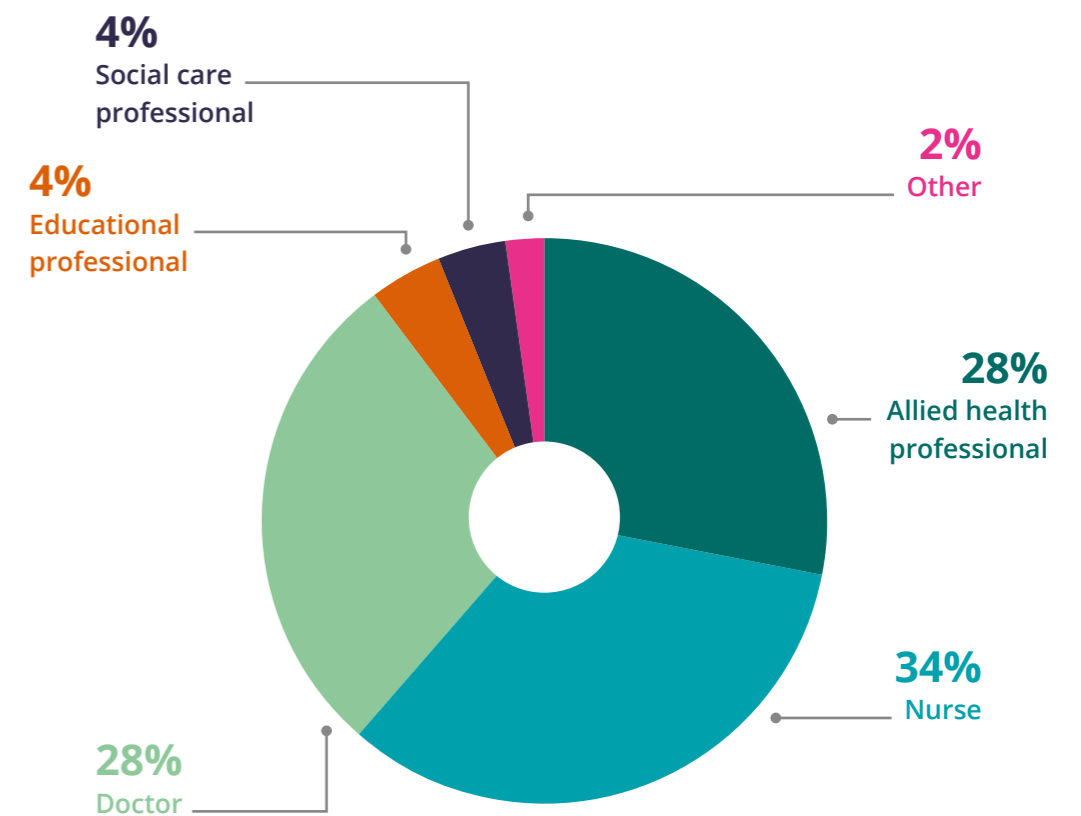


Figure 12  
Age at diagnosis, in years, of survivors and parents, friends and relatives (shortlisting survey)

A broad range of professionals responded to the shortlisting survey. Figure 13 illustrates the distribution between medical, nursing, allied health professionals, social care and educational professionals. The main professions represented in the allied health professionals' group were dietitians (28%, n=7), physiotherapists (24%, n=6), and clinical psychologists (20%, n=5). The majority of doctors worked in tertiary care (76%, n=19), 12% (n=3) worked in secondary care and 4% (n=1) in primary care, 8% (n=2) answered 'Other' and indicated that they were academics. Most nurses were children's cancer nurses (63%, n=19), 13% (n=4) were children's nurses, 7% (n=2) were children's community nurses and 17% (n=5) responded, 'Other'.

Figure 13  
Distribution of professionals (shortlisting survey)

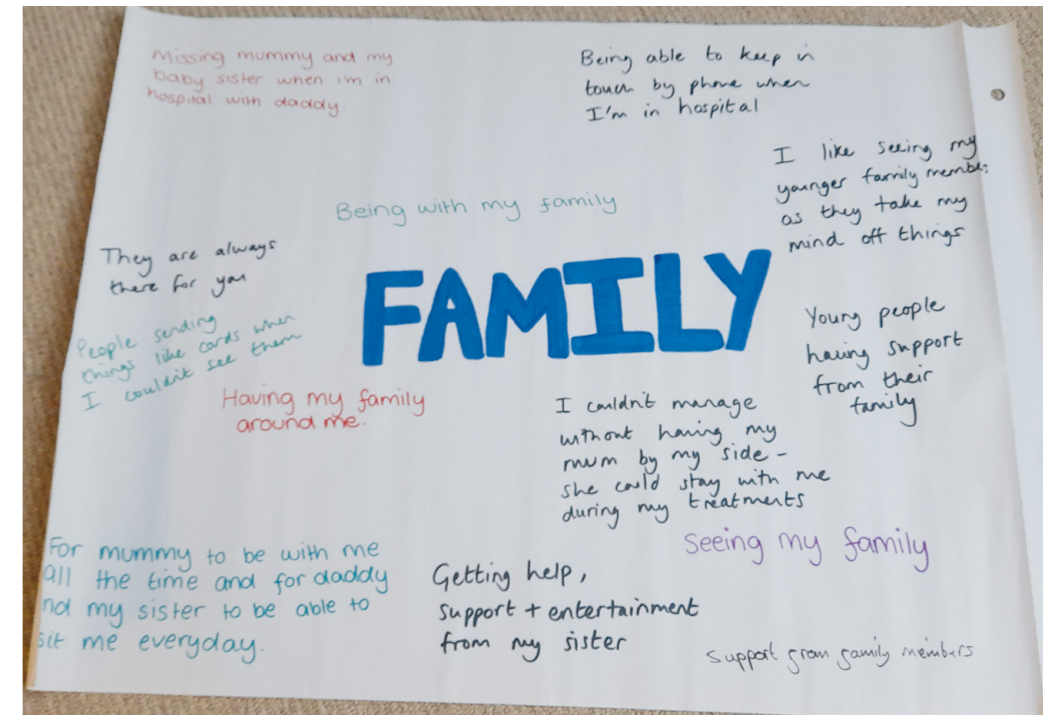


The results were analysed in three groups: 1) patients/survivors, 2) parents/friends/relatives, 3) professionals. This gave equal weight to each group's choices as more parents/friends/relatives took part than the other groups. Questions were each given a rank depending on the number of votes (rank 1 was the question with the highest number of votes). Questions were then ordered from highest to lowest rank for each group. The steering group reviewed and compared respondent groups, they decided to take the Top 10 questions for each of the three groups to the workshop. This ensured that what is important to each group would be considered and gave 21 questions, as there were some shared priorities between the groups.





**Figure 14** ▶  
Photo from the discussion on what is important to children and young people about 'Family'



## Stage 4a

# Workshop with children and young people

The children and young people's workshop took place on Sunday 23rd October 2022 from 11am to 3pm. It was held in a community centre in central London and was facilitated by FG and SA. Eight children and young people aged 8-16 attended; three were siblings. Their diagnoses included lymphoma and leukaemia. Parents were able to wait in the venue in a separate room if they wished, parents did not contribute to the discussion.

The workshop began with an 'ice-breaker' activity. We played 'People Bingo' to help everyone to get to know each other. Each participant (including the facilitators) was given a card with a 3x3 grid containing statements such as, 'Someone who likes reading', 'Someone who has a pet'. The task was to go around the group and find someone who each statement applied to and write down their name against it.

Once this activity was complete, the facilitators gave a brief introduction to the day, outlining the purpose of the workshop and what was going to happen. We then moved on to a discussion about 'family, friends, and pets', to make some summary questions on these topics as the meaning of the submissions to the survey about these aspects had been unclear. The words 'family', 'friends' and 'pets' were each written in the centre of a sheet of A1 size flip-chart paper. The submissions from the surveys about each topic were written around the word. Each topic was discussed in turn, this focused on what was important to the participants about family, friends, and pets – their responses were added to the paper by one of the facilitators (Figure 14).



**Eight children and young people aged 8-16 attended**

Each topic was then revisited; the facilitators gave a verbal summary of what was on each sheet and worked with the children and young people to support them to make summary questions for each topic. Seven summary questions were created:

### Family:

1. How can we make the most of open visiting so other family members can be more involved?
2. How can we make sure all children and young people can see all family members when they are hospital?

### Friends:

3. What are the best ways to spread awareness to help friends and classmates understand the reality of cancer?
4. How can we help children and young people to stay connected with friends and keep their relationships strong during treatment and afterwards?

### Family and friends:

5. What are the best ways to help children and young people to keep in contact with family and friends when they are in hospital?

### Pets:

6. How can we make it so children can meet and interact with their pets when they are in hospital?
7. How can we help more children to see therapy animals when they are in hospital?

Each question was written onto a card in preparation for the next discussion.

We then followed the methodology used by the Children's Arthritis PSP in the Netherlands (Aussems et al. 2021). We had seven envelopes, each containing questions on a different topic. In total there were 31 questions – 24 summary questions from the children and young people's surveys (Appendix 6), plus the seven new questions on family, friends, and pets. The topics were:

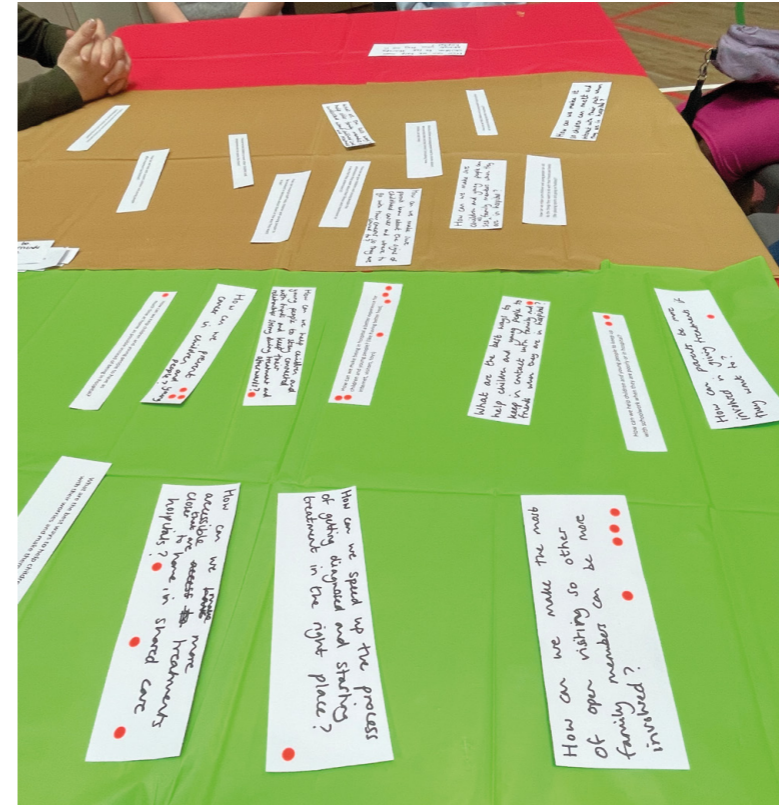


Each participant chose an envelope that corresponded to a topic that was important to them. They could share envelopes if they wished; six participants worked in pairs and two worked individually. The topics that were not picked were 'Being poorly, side-effects and long-term effects' and 'Being in hospital'. The table was covered in red, amber, and green tablecloths (Figure 15). These colours represented the importance of the question; green was most important, red was least important, with amber being moderate importance. The envelopes were opened, and the participants placed the questions on the table in the colour that reflected the importance of the question to them. Only one question was placed in the red area (How can we help more children to see therapy animals when they are in hospital?). Participants looked at the questions on the table and were invited to add more questions if there was anything missing that was important to them. Six questions were added and were written onto cards:

1. How can we prevent cancer in children and young people?
2. How can we make more accessible treatments that are closer to home, in shared care hospitals?
3. How can we speed up the process of getting diagnosed and starting treatment in the right place?
4. How can we make sure parents know about the signs of childhood cancer and where to go with their concerns, so they are listened to?
5. What are the best ways to help older family members to understand about childhood cancer and treatments?
6. How can parents be more involved in giving treatments if they want to?

The participants decided together which theme each of these new questions should be placed in.

Improving the experience of being in hospital was also discussed, and they wanted to create a question about this. There was already a question on this topic in the envelope on 'Being in hospital' which had not been opened. The participants were shown the question, 'How can we make being in hospital a better experience for children and young people? (like having better food, internet, visitors, toys)', they decided that it reflected what they wanted to say and placed it in the green area.



▲ **Figure 15**  
Photo from the children and young people's workshop showing the ranking system with coloured tablecloths

Each participant was given three stickers which they could use to vote for their Top 3 questions in the green area. Before the voting took place, they could move any questions from the amber/red areas up if they wished, so that they could be included in the vote. The questions were then placed in order of most to least votes and a discussion followed to agree the 'Top 5'. The question, 'How can we make being in hospital a better experience for children and young people? (like having better food, internet, visitors, toys)' had received six votes and was placed as top priority. Underneath this was, 'How can we make the most of open visiting so other family members can be more involved?', which had four votes. The children and young people spoke about how important it was for family members to be able to visit them in hospital as this helped them to understand more about the treatment. They could also

offer support to the child and their parents/family and be involved in the child's care. As the question about improving hospital experience already mentioned 'visitors', the participants asked if they could expand on this to combine it with the question about open visiting which would then leave room for another question on a different topic in the Top 5. The question was changed to: 'How can we make being in hospital a better experience for children and young people? (like having better food, internet, toys, and open visiting so other family members can be more involved in the child's care)'.

The question, 'How can we prevent cancer in children and young people?' was placed at number 2 as the participants said that if cancer could be prevented, then answering the other questions would not be necessary. 'How can we make more accessible treatments that are closer to home, in shared care hospitals?' was placed at number 3 as some participants had to travel a long way for treatment and they wanted treatments to be available closer to home. This question, 'How can we help children and young people to keep up with schoolwork when they are poorly or in hospital?' was initially in the Top 5 but was then moved out as the participants discussed that it was possible to catch up with schoolwork later and they did not always feel like doing schoolwork when they were very unwell; the questions about getting a timely diagnosis and being supported emotionally were considered more important.

The Top 5 are shown in Table 1. Three of the questions were closely aligned to the questions already going to the final workshop from the shortlisting survey (priorities 2, 4 and 5). For priority 4, the children and young people's version of the question had an extra part on the end about starting treatment in the right place, therefore their version of the question was taken to the final workshop rather than the version from the shortlisting survey. Priorities 1 and 3 from children and young people were new and were added into the list of questions for the workshop, making 23 questions for the final workshop in total.

Rank	Top 5 questions from the children and young people's workshop	Question going to the final workshop from the shortlisting survey
1	How can we make being in hospital a better experience for children and young people? (like having better food, internet, toys, and open visiting so other family members can be more involved in the child's care)	
2	How can we prevent cancer in children and young people?	Why do children develop cancer (including the role that genetics plays) and could it be prevented?
3	How can we make more accessible treatments that are closer to home, in shared care hospitals?	
4	How can we speed up the process of getting diagnosed and starting treatment in the right place?	How can time to diagnosis be improved for children with suspected cancer?
5	What are the best ways to help children and young people with their worries and make them feel happier?	What are the best ways to provide emotional support for children and their families 1) around the time of diagnosis, 2) during treatment and 3) after treatment (including survivors who are now adults)?

◀ **Table 1**  
Children and young people's Top 5 and questions taken to the final workshop



## Stage 4b

# Agreeing the Top 10 – final workshop

A final prioritisation workshop took place in central London on 1st November 2022 to identify the Top 10 unanswered questions for future research on children's cancer. The workshop was attended by 25 participants: four young adults who had experienced childhood cancer, five parents and one grandparent of a child who had experienced cancer, and 15 professionals who work with children who have cancer and their families. The professionals were from a wide range of backgrounds including nurses, doctors, a social worker, health play specialist, dietitian, clinical psychologist, physiotherapist and chaplain. One participant was a member of the steering group. The other participants were invited as they had indicated they would like to attend when completing the shortlisting survey or they were suggested by steering group members due to their professional role. Participants were asked to individually rank the 23 questions in order of importance prior to the workshop; this was used as a starting point for discussion. With permission, biographies of participants were also circulated before the day.

The workshop was chaired and facilitated by Jonathan Gower with support from two co-facilitators from the JLA, Tricia Ellis and Toto Gronlund. Prior to the workshop, participants were divided into three groups to ensure a balance of professionals from different disciplines, young adults and parents/relatives. Each group had a set

of the 23 questions on A4 cards, which were laid out on a table. For the first step, each person was asked to tell their group the three questions they had ranked highest and lowest in their individual ranking. The participants were told which of the questions were in the children's Top 5. On the back of each card was a list of the rankings from the shortlisting survey for patients/survivors, parents/friends/relatives and professionals. Discussion followed and the groups were asked to place the 23 questions in a collective order of importance. Each participant was encouraged to share their views and consider other people's opinions. At lunchtime, the ranking of the 23 questions from the three groups were combined. In the afternoon session, in new group compositions, the consensus ranking was the starting point for discussion. Following this second round of discussion, the group rankings were again collated, and the participants came together as one group to agree the Top 10 and debate the order.

## Decision making: prioritising the Top 10

In this section we aim to give an overview of the discussions in the workshop and how the Top 10 were decided upon. What was striking was the similarities between the three groups within the workshop who independently developed very similar strategies to decide which questions should be in the Top 10.

All agreed that this was extremely difficult. Their strategies involved:



### Ensuring that children's views were represented

All three groups wanted to ensure the Top 10 questions included most, if not all, of the questions that were in the children's Top 5. When the groups were told which questions were important to children, those question cards were picked out and moved high up the ranking and their placement discussed. Most of these questions remained ranked in the Top 10, or just outside, for the duration of the discussions. There was a lot of discussion about whether the question, 'What are the best ways to provide emotional support for children and their families: 1) around the time of diagnosis, 2) during treatment and 3) after treatment (including survivors who are now adults)?' should be in the Top 10 as this could be covered by other, broader questions such as, 'Are the psychological, practical, and financial support needs of children with cancer, survivors, and their families being met during treatment and beyond? How can access to this support be improved and what further support would they like?'

### Opting for questions that could include other questions /overlap

As previously mentioned, the groups considered which questions overlapped and could cover other questions. For example, 'Can we find effective and kinder (less burdensome, more tolerable, with fewer short and long-term effects) treatments for children with cancer, including relapsed cancer?' mentions side-effects and so could include, 'What are the best ways to reduce, predict and manage the side-effects of treatment for children (including life threatening side-effects)?' The question, 'What impact does cancer and treatment have on the lives of children and families after treatment, and in the long-term; what are the best ways to help them to overcome these impacts to thrive and not just survive?' was selected rather than, 'What are children's and survivors' experiences of the side-effects and long-term effects of cancer treatment?' as the former question is broader.

### Opting for questions focussed on intervention rather than description

The groups were clear that although it is useful to describe a problem, it is action through intervention that is required to improve children's and families' experiences. Therefore, 'Are the psychological, practical, and financial support needs of children with cancer, survivors, and their families being met during treatment and beyond? How can access to this support be improved and what further support would they like?' and 'What are the best ways to provide emotional support for children and their families 1) around the time of diagnosis, 2) during treatment and 3) after treatment (including survivors who are now adults)?' were placed higher in the rankings than 'What is the psychological and social impact of cancer and treatment on children and their families during treatment and in the long-term; what factors affect these impacts?' as the latter question involves description, rather than action.

### Opting for questions that could have wider impact for children and families

Initially, most participants selected their top three questions based on what was relevant to their personal experience or focused on the area they worked within. However, during the discussions that followed, their opinions changed, and the groups decided that the Top 10 questions should be generic and have the potential to have an impact on as many children and families as possible, and that this might also mean increased likelihood of research being funded. For example, 'How can experiences of having a Hickman line be improved for children with cancer?' was considered to be too specific and did not apply to all children. The question, 'What impact does cancer and treatment have on the lives of children and families after treatment, and in the long-term; what are the best ways to help them to overcome these impacts to thrive and not just survive?' was moved further down the Top 10 in recognition that not all children survive their cancer. 'During and after treatment, what issues prevent or encourage physical activity, which interventions are most effective and what should be measured to assess effectiveness?', had been a priority for an allied healthcare professional because of their role, they voiced the need to relinquish this when considered alongside other priorities.

Professionals had ranked the question about supporting their own wellbeing as low in their

individual ranking, stating that the priorities should focus on families, and that there are other factors that impact their wellbeing, such as working hours. The young adults and family members wanted this question placed higher up (although not necessarily in the Top 10) and argued that it was important that the wellbeing of professionals is supported as families are reliant upon them for support and want continuity of care (so do not want professionals to be off sick or to leave/move roles). Highlighted specifically, were professionals working in palliative care, that they would need targeted support. Across all groups, there was a lot of discussion about this topic, and like other areas it was seen as important, but not as important as other questions to be prioritised.

### Ensuring all themes within the questions were represented

The groups tried to cluster questions into similar themes, such as support, treatment, care, side-effects and improving experiences for survivors as well as children on treatment, their aim being to include each 'theme' in the Top 10. For example, the question about relapse ('Why do children relapse, how can it be prevented, and what are the best ways to identify relapse earlier?') was moved up during the discussions as this was not covered by any other question.

From the very start of the workshop when the participants were asked to give their top three questions, there were some questions that were clearly high priority for many and stayed high in the Top 10 throughout the workshop. The question ranked as top priority, 'Can we find effective and kinder (less burdensome, more tolerable, with fewer short and long-term effects) treatments for children with cancer, including relapsed cancer?' was the top priority for all three groups after the first group discussion. This question had also ranked at number one in the shortlisting survey for all three respondent groups. After the second group discussion, all three groups had the same questions ranked at one to five, which remained in the same positions in the final Top 10.

### Group discussion and decision making

When everyone came together for the final group discussion, the focus of the discussion was around the inclusion of, 'What is the relationship between chronic fatigue syndrome, fibromyalgia, chronic

pain and treatment for childhood cancer?' in the Top 10. This push for inclusion came from a couple of the young adults present who said that these long-term effects had a huge impact on their lives and there was a lack of recognition and support available. Several participants felt that a question about side-effects/long-term effects should be included in the Top 10 and perhaps it would better to include, 'What are the best ways to reduce, predict and manage the side-effects of treatment for children (including life threatening side-effects)?' as it is not so specific. The young adults argued that it was important to have these specific side-effects identified in a targeted question as then they would be more likely to become the focus of research, rather than being ignored. Some professionals commented that they had not realised these long-term effects were such a problem for survivors and felt it was important that the views of those present were heard and reflected in the Top 10. It was also noted that two broader questions that include reducing/managing short and long-term effects were already in the Top 10, 'Can we find effective and kinder (less burdensome, more tolerable, with fewer short and long-term effects) treatments for children with cancer, including relapsed cancer?' and 'What impact does cancer and treatment have on the lives of children and families after treatment, and in the long-term; what are the best ways to help them to overcome these impacts to thrive and not just survive?' There was a group vote and the decision was made to move the question about fatigue and pain up to number 10 and move 'What are the best ways to provide emotional support for children and their families 1) around the time of diagnosis, 2) during treatment and 3) after treatment (including survivors who are now adults)?' down to number 11 as it was agreed that this was covered by the broader question, 'Are the psychological, practical, and financial support needs of children with cancer, survivors, and their families being met during treatment and beyond? How can access to this support be improved and what further support would they like?' which was at number 3.

The positions of a few other questions were moved in the final discussion, for example 'How can transition (moving) from child into adult services be improved for young people who had cancer as a child?' was moved up a rank ahead of 'What is the psychological and social impact of cancer and treatment on children and their families during treatment and in the long-term; what factors affect these impacts?'

# Top 10 research priorities



1

Can we find effective and kinder (less burdensome, more tolerable, with fewer short and long-term effects) treatments for children with cancer, including relapsed cancer?

2

Why do children develop cancer (including the role that genetics plays) and could it be prevented?



3

Are the psychological, practical, and financial support needs of children with cancer, survivors, and their families being met during treatment and beyond? How can access to this support be improved and what further support would they like?



4

How can we speed up the process of getting diagnosed and starting treatment in the right place?



5

Why do children relapse, how can it be prevented, and what are the best ways to identify relapse earlier?



6

How can we make being in hospital a better experience for children and young people? (Like having better food, internet, toys, and open visiting so other family members can be more involved in the child's care).



7

What are the best ways to ensure children and families get and understand the information they need, in order to make informed decisions, around the time of diagnosis, during treatment, at the end of treatment and after treatment has finished?



8

What impact does cancer and treatment have on the lives of children and families after treatment, and in the long-term; what are the best ways to help them to overcome these impacts to thrive and not just survive?



9

How can we make more accessible treatments that are closer to home, in shared care hospitals?



10

What is the relationship between chronic fatigue syndrome, fibromyalgia, chronic pain and treatment for childhood cancer? (Fibromyalgia is a long-term condition that causes pain all over the body).



## All 23 questions were discussed and put in order of priority at the workshop:

11. What are the best ways to provide emotional support for children and their families 1) around the time of diagnosis, 2) during treatment and 3) after treatment (including survivors who are now adults)?
12. What are the best ways to reduce, predict and manage the side-effects of treatment for children (including life threatening side-effects)?
13. How can transition (moving) from child into adult services be improved for young people who had cancer as a child?
14. What is the psychological and social impact of cancer and treatment on children and their families during treatment and in the long-term; what factors affect these impacts?
15. How common are the different long-term effects of childhood cancer treatment, how do they change across the lifespan, can we predict them and how can they best be prevented, detected and/or treated?
16. What are the best ways to support the emotional wellbeing of professionals who care for children with cancer and their families?
17. During and after treatment, what issues prevent or encourage physical activity, which interventions are most effective and what should be measured to assess effectiveness?
18. What are the best ways of making sure people who had cancer as a child receive the information they need about the long-term effects of cancer and treatment?
19. What fertility preservation options work best for children and teenagers with cancer?
20. What are the long-term effects of additional medications children with cancer may receive (such as antibiotics, pain killers, laxatives) and how can these effects be reduced?
21. What are children's and survivors' experiences of the side-effects and long-term effects of cancer treatment?
22. How can experiences of having a Hickman line be improved for children with cancer? (A Hickman line is a small tube which is inserted into a vein so that treatments can be given, and blood taken without the repeated need to access veins with a needle. The Hickman line can stay in place for several months).
23. What are the best ways to support children as they get older, and their needs change, to understand and take responsibility for their health, and to live with the long-term effects of cancer and treatment?

# Next steps



## For research funders

The Top 10 list provides major national research funders such as the National Institute for Health Research (NIHR), Medical Research Council (MRC) and Economic and Social Research Council (ESRC) with clear guidance on the frontline priorities for future research, voted on by end-users of health research. Any research funders, large or small, working independently or in collaboration, can use this list to target funds effectively, and to inform future fundraising to prioritise what matters most to all those involved in the care of children and young people with cancer. Funders could also learn from this PSP that where sufficient expertise and resource are available, patient involvement of children as young as three and four years can be achieved. Therefore, funding guidance should encourage applicants to undertake such work.

## For researchers

A common question in reviewers' comments and applications for funding include whether it is: a) an important question; b) whether it has already been answered; c) have patients/caregivers/the public been involved in the process. This Top 10 will allow researchers to tailor their research questions and strategies to develop a portfolio of studies relevant to children and young people with cancer based on priorities agreed by multiple stakeholders. The long list of questions will be made openly available via the James Lind Alliance website.

## For charities

Charitable funders within their research and policy teams can refer to the Top 10 and the detailed priority list to demonstrate need for patient-orientated research funding to improve the evidence base. Such charitable bodies are uniquely placed to operationalise these priorities, through studies focused on service delivery and patient/family experience, to improve the care experienced by children and young people with cancer.

## For the Children's Cancer PSP

The initial survey returned 1299 potential research questions, some of which included comments and questions which did not fit within our scope, for example 'Treatment time scale' and 'Why are there placebo treatment arms?', which were removed. Many of these questions suggested a knowledge gap. We will look at how these questions, statements and service enquiries can be best used to improve information giving and influence outcomes.

Submitted to the survey were questions on research funding, for example, 'Will low grade tumours be given the same priority in researching treatments as high grade tumours?' and 'Why isn't more money spent on research into children's cancer and better treatments?', these were also removed. We will consider these questions separately and share more widely through a commentary piece on the funding of children's cancer research, as these reflected strong opinions and perceptions that would benefit from further exploration and articulation.

Absent voices must also be considered. Of particular note, the majority of our patients/survivors and family/friends respondents described themselves as White (95%, n=322 in the initial survey and 94%, n=223 in the shortlisting survey). The priorities therefore represent the views of the majority, White population. Inequalities exist, this might be because of both visible and invisible disabilities, and how far the views of those experiencing such effects of cancer and treatment were represented, and as a result impacted on the prioritisation of these research questions are unknown. In addition, although there was a primary care representative on the steering group and at the final workshop, only one response to the shortlisting survey was received from a primary care professional and none responded to the initial survey. Primary care has an important role in the care of children with cancer from diagnosis into survivorship (Jain et al. 2019), but the voices of these professionals are absent from the questions collected.

## Acknowledgements

This project was supported and funded by Children's Cancer and Leukaemia Group (CCLG) and Little Princess Trust.

The Children's Cancer PSP would like to thank everyone who took the time to send in their questions and vote on the importance of them. Thank you also to the children, young people, parents, relatives and professionals who attended the workshops.

## References

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- Jain, J., Qorri, B., & Szewczuk, M. R. (2019). The crucial role of primary care providers in the long-term follow-up of adult survivors of childhood cancer. *Cancer management and research*, 11, 3411–3418. <https://doi.org/10.2147/CMAR.S197644>
- Postma L, Luchtenberg ML, Verhagen AAE, et al. Involving children and young people in paediatric research priority setting: a narrative review. *BMJ Paediatrics Open* 2022;6:e001610. <https://bmjpaedsopen.bmj.com/content/6/1/e001610>

## Appendix 1 Partners



The Children's Cancer PSP is grateful for the support of all our Partners who helped to distribute the surveys and Top 10 priorities.

Charity partners:			
Abby's Heroes	Childhood Eye Cancer Trust (CHECT)	MOVE Charity	
Alice's Arc	Children with Cancer UK	Rainbow Trust	
Anthony Nolan	Chris Lucas Trust	SimPal	
Beads of Courage UK	Ellen MacArthur Cancer Trust	Solving Kids Cancer	
Blood Cancer UK	Grace Kelly Childhood Cancer Trust	Teenage Cancer Trust	
brainstrust	Grandparents of Kids with Cancer	Teenagers and Young Adults with Cancer (TYAC)	
The Brain Tumour Charity	The Joshua Tree	World Child Cancer	
Brain Tumour Research	Milly's Smiles	Young Lives vs Cancer	
Candlelighters			

Professional organisation partners:			
Association of Paediatric Chartered Physiotherapists	National Association for Hospital Education	Royal College of Nursing	
British Dietetic Association	National Association of Health Play Specialists	Royal College of Paediatrics and Child Health	
Clinical Psychology Special Interest Group in Paediatric Oncology	National Cancer Research Institute (NCRI) Children's Group	Royal College of Radiologists	

## Appendix 2

# Overview of Children's Cancer PSP methodology and results



# Answered questions

**Q. D2:** Is it possible to detect neuroblastoma cells in a baby or before a child is born?

## Evidence

1. Cass DL. Fetal abdominal tumors and cysts. *Transl Pediatr* 2021;10(5):1530-1541. <http://dx.doi.org/10.21037/tp-20-440>
2. Shinagawa, T., Kitamura, T., Katanoda, K., Matsuda, T., Ito, Y., & Sobue, T. (2017). The incidence and mortality rates of neuroblastoma cases before and after the cessation of the mass screening program in Japan: A descriptive study. *International Journal of Cancer*, 140(3), 618–625. <https://doi.org/10.1002/ijc.30482>

## Comments

The Cass article reviews the diagnosis and management of the most common abdominal tumours and cystic lesions diagnosed in the foetus, including neuroblastoma.

The Shinagawa article is about screening in babies at six months old.

Correspondence with an expert in this field indicated that this question did not warrant further investigation. There are some large population-based studies which have looked at detecting neuroblastoma at birth – and it leads to an increase in the number of cases diagnosed (approx. double), but no change in mortality. The cases picked up are likely those which would have spontaneously regressed and not required any treatment, and it does not prevent cases of later more aggressive disease. Furthermore, picking up cases early may lead to unnecessary investigation, procedures, and anxiety. We also see cases picked up incidentally on maternal antennal scans – and the vast majority of these are observed and do not require any treatment. (See Frank Berthold, MD, Claudia Spix, PhD, Rudolf Erttmann, MD, Barbara Hero, MD, Joerg Michaelis, MD, Joern Treuner, MD, Angela Ernst, MSc, Freimut H Schilling, MD, Neuroblastoma Screening at 1 Year of Age: The Final Results of a Controlled Trial, *JNCI Cancer Spectrum*, Volume 5, Issue 4, August 2021, pkab041, <https://doi.org/10.1093/jncics/pkab041>)

## Submitted by

 1 parent/carer

**Q. D4:** For children with leukaemia, what is the relationship between white blood cell count at diagnosis and survival?

## Evidence

Henze, G., Langermann, H. J., Lampert, F., Neidhardt, M., & Riehm, H. (1979) ALL therapy study 1971-1974 of the German working group for leukemia research and therapy in childhood: prognostic significance of initial features and different therapeutic modalities *Klinische Padiatrie*, 191(2), 114–126. <https://pubmed.ncbi.nlm.nih.gov/286842/>

## Comments

Evidence is older than 2017; this became embedded into risk stratification and modelling from the early 80s.

Steering group agreed this is answered and there is ongoing work.

## Submitted by

 1 parent/carer

**Q. LTFU1:** What are the best methods and timing of long-term follow-up for childhood cancer survivors to ensure personalised and coordinated care into adulthood?


## Evidence

Michel, G., Mulder, R. L., van der Pal, H. J. H., Skinner, R., Bárdi, E., Brown, M. C., Vetsch, J., Frey, E., Windsor, R., Kremer, L. C. M., & Levitt, G. (2019). Evidence-based recommendations for the organization of long-term follow-up care for childhood and adolescent cancer survivors: a report from the PanCareSurFup Guidelines Working Group. *Journal of Cancer Survivorship*, 13(5), 759–772. <https://doi.org/10.1007/s11764-019-00795-5>

## Comments

To facilitate the implementation of long-term follow-up (LTFU) care and improve equality of care for childhood, adolescent, and young adult cancer survivors, the PanCareSurFup Guidelines Working Group developed evidence-based recommendations for the organization of LTFU. They provide strong recommendations based on low level evidence and expert opinions, regarding organisation of LTFU care, personnel involved in LTFU care, components of LTFU care and start of LTFU care.

## Submitted by

 11 patients/survivors

 2 professionals

 2 parents/carers

**Q. SD11:** How can medication errors be prevented in children's cancer treatment?

## Evidence

1. Koeck, J. A., Young, N. J., Kontny, U., Orlikowsky, T., Bassler, D., & Eisert, A. (2021). Interventions to Reduce Medication Dispensing, Administration, and Monitoring Errors in Pediatric Professional Healthcare Settings: A Systematic Review. *Frontiers in Pediatrics*, 9, 633064. <https://doi.org/10.3389/fped.2021.633064>
2. Marufu, T. C., Bower, R., Hendron, E., & Manning, J. C. (2022). Nursing interventions to reduce medication errors in paediatrics and neonates: Systematic review and meta-analysis. *Journal of Pediatric Nursing*, 62, e139–e147. <https://doi.org/10.1016/j.pedn.2021.08.024>
3. Naserallah, Lina Mohammad; Hussain, Tarteel Ali; Jaam, Myriam; Pawluk, Shane Ashley (2020) Impact of pharmacist interventions on medication errors in hospitalized pediatric patients: a systematic review and meta-analysis, *International Journal of Clinical Pharmacy*, Vol. 42 Issue 4, pp. 979–994.

## Comments

These reviews focus on prevention of medication errors for children in hospital generally – they do not focus on children with cancer.

The reviews focus on general issues around dispensing, administration etc. and interventions that are effective are identified. The interventions referred to in all these papers would apply to children with cancer.

## Submitted by

 1 parent/carer



## Appendix 4

# Ongoing studies

**Q. N5:** What factors or tools are most effective in monitoring a child's nutrition during cancer treatment and the identification (screening) of those most at risk? How could this be standardised across the UK?

### Evidence

Ongoing work by the NIHR nutrition group:  
<https://cancerandnutrition.nihr.ac.uk/work-streams/nutrition-in-children-teenagers-and-young-adults-with-cancer/>

### Comments


Ongoing work on this question by the NIHR Cancer and Nutrition Collaboration - Children, Teenagers and Young Adults work stream (Three PSP steering group members are involved in this ongoing work: Bob Phillips, Faith Gibson and Louise Henry).

The first part of this work was a survey of current practice in the UK regarding dietetic resource and nutritional assessment. A paper on this has been published:

Henry, L., Aldiss, S., Gibson, F., Pugh, G., Stevens, M., & on behalf of the Children, Teenagers and Young Adults (CTYA) Workstream of the NIHR Cancer and Nutrition Collaboration (2022). Nutritional assessment and dietetic resource for children and young people with cancer in the United Kingdom. *Pediatric blood & cancer*, 69(9), e29743. <https://doi.org/10.1002/psc.29743>

This survey was undertaken to inform the development of guidance to harmonise assessment and management of nutritional care for the future.

### Submitted by

 6 professionals

**Q. RF1:** What are the best strategies to accelerate the development and testing of new drugs and implement clinical trials for childhood cancer?

### Evidence

Ongoing work by ACCELERATE (SIOP Europe)  
<https://www.accelerate-platform.org/why-accelerate>

Pearson, A., Weiner, S. L., Adamson, P. C., Karres, D., Reaman, G., Rousseau, R., Blanc, P., Norga, K., Skolnik, J., Kearns, P., Scobie, N., Barry, E., Marshall, L. V., Knox, L., Caron, H., Wariabharaj, D., Pappo, A., DuBois, S. G., Gore, L., Kieran, M., ... Vassal, G. (2022). ACCELERATE - Five years accelerating cancer drug development for children and adolescents. *European journal of cancer* 166, 145–164. <https://doi.org/10.1016/j.ejca.2022.01.033>

### Comments

Aim is to ACCELERATE innovation in drug development for children and adolescents with cancer. ACCELERATE is a transparent discussion forum that tackles overarching issues in the development of new anticancer medicines for children and adolescents. ACCELERATE brings together academia, industry, advocacy and regulators to find solutions for more and better innovative therapies for children and adolescents with cancer.

### Submitted by

 21 parents/  
carers

 9 professionals

 2 relatives

**Q. IS1:** For children being treated for cancer, what is the survival rate and likelihood of relapse or developing another cancer?


### Evidence


The NCIN (<http://www.ncin.org.uk/home>) in the UK and SEER (<https://seer.cancer.gov/>) in the USA publish data on this.

### Comments

We know the 'now' answers but different therapies can change those answers so we need to keep updating our knowledge, there is ongoing data collection on this.

### Submitted by

 18 parents/  
carers

 3 patients/  
survivors

 3 relatives

 2 professionals

## Appendix 5

# Unanswered questions included in the shortlisting survey

### Side-effects and management

- How can infections in children with cancer be prevented?



- What are the risks of participating in activities (such as swimming, attending birthday parties, soft play) for children who are on treatment?

- What are the best ways to reduce, predict and manage the side-effects of treatment for children (including life threatening side-effects)?

- What are children's and survivors' experiences of the side-effects and long-term effects of cancer treatment?

- How can monitoring of children with cancer be improved when they are a baby and/or too young to communicate their symptoms, pain and side-effects?

- Is it possible to have a monitoring system for neutropenic fevers, (such as wearing a thermometer as a wrist band) to give an alert if a child has a high temperature?

- What are the experiences of families the first time their child with cancer has a spike in temperature?

- How do we best manage other conditions the child has alongside their cancer?

- What fertility preservation options work best for children and teenagers with cancer?

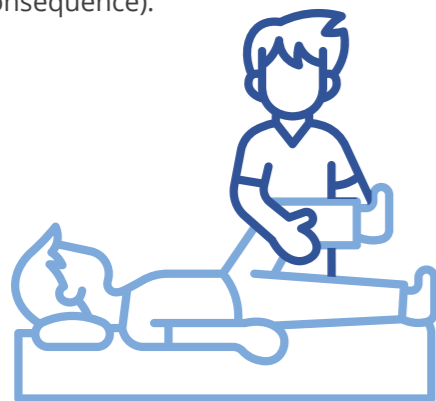
- Does discussing palliative care with families around the time of diagnosis improve the management of symptoms for children with cancer? (Palliative care involves providing support to children to live as actively as possible).

- What are the best ways of accurately recording children's symptoms during cancer treatment?

## Treatment

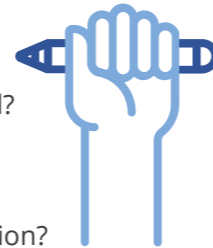


- Can we find effective and kinder (less burdensome, more tolerable, with fewer short and long-term effects) treatments for children with cancer, including relapsed cancer?
- Can we use individualised therapies for each child with cancer? (Such as using genetic, targeted and pharmacodynamic information).
- Which complementary therapies benefit children with cancer?
- How do delays or variations in 'planned' treatment affect outcomes for children? (Outcomes means something that follows as a result or consequence).
- Why do treatment strategies differ between countries and what difference does this make to outcomes? (Outcomes means something that follows as a result or consequence).
- How can experiences of having a Hickman line be improved for children with cancer? (A Hickman line is a small tube which is inserted into a vein so that treatments can be given, and blood taken without the repeated need to access veins with a needle. The Hickman line can stay in place for several months).
- What are the best ways to support professionals to decide when to stop active treatment and begin end of life care when a child cannot be cured?
- Can modern imaging approaches (such as PET or diffusion-weighted MRI) improve the way in which we plan, treat and monitor cancer during and after treatment to improve outcomes and minimise radiation risks? (Outcomes means something that follows as a result or consequence).



## Education

- What are the best ways to support children to continue with their education during treatment and return to education after treatment?
- What are the best ways to give education professionals information about the impact of cancer that then enables them to provide social and emotional support for children during and after treatment?
- What are the best ways to raise awareness amongst health professionals of the importance of education in hospital for children with cancer?
- What are the benefits of education on children's development and wellbeing whilst they are having cancer treatment?
- What are the views of children who have cancer on the importance of education while they are in hospital?
- How can parents of children with cancer be best supported to make decisions about their child's education?



## Physical activity, play and therapies

- During and after treatment, what issues prevent or encourage physical activity, which interventions are most effective and what should be measured to assess effectiveness?
- How can evidence-based rehabilitation and prehabilitation programmes for children with cancer be developed and put into practice to lead to better outcomes? (Prehabilitation aims to enhance health and wellbeing before treatment starts. Outcomes means something that follows as a result or consequence).
- How can occupational therapy be more included in care for children during and after treatment?
- What are the benefits of having communication therapy during treatment for children with cancer who have communication needs?
- What interventions/methods of help from play specialists are effective in the care of children with cancer and how can these services be consistently provided across treatment centres?
- How can access to physical therapy and rehabilitation be improved for children during and after treatment in the hospital and community?

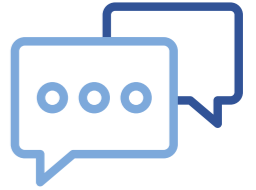
## Long-term effects and follow-up care

- What are the best methods and timing of follow-up for children with cancer after treatment?
- What impact does cancer and treatment have on the lives of children and families after treatment, and in the long-term; what are the best ways to help them to overcome these impacts to thrive and not just survive?
- What are the best ways to support children as they get older, and their needs change, to understand and take responsibility for their health, and to live with the long-term effects of cancer and treatment?
- How can long-term follow-up care be carried out by non-specialists (such as GPs and non-Principal Treatment Centres) and what knowledge and skills do they need?
- What are the best ways of making sure people who had cancer as a child receive the information they need about the long-term effects of cancer and treatment?
- How can transition (moving) from child into adult services be improved for young people who had cancer as a child?
- How common are the different long-term effects of childhood cancer treatment, how do they change across the lifespan, can we predict them and how can they best be prevented, detected and/or treated?
- What is the relationship between chronic fatigue syndrome, fibromyalgia, chronic pain and treatment for childhood cancer? (Fibromyalgia is a long-term condition that causes pain all over the body).
- What are the long-term effects of additional medications children with cancer may receive (such as antibiotics, pain killers, laxatives) and how can these effects be reduced?
- Do children with cancer who undergo bone marrow aspirations and lumbar puncture procedures experience long-term effects on their spine/back?
- What impact does having cancer and treatment as a child have on a person's life expectancy?
- How can plans for follow-up help children and families to be better prepared when treatment ends?



## Communication and information sharing

- What are the best ways to ensure children and families get and understand the information they need, in order to make informed decisions, around the time of diagnosis, during treatment, at the end of treatment and after treatment has finished?
- What are the best ways to support professionals to communicate effectively, sensitively, and promptly with children and their families, including at diagnosis, when sharing results and when giving distressing news?
- What are the best ways to support families to talk about their child's cancer?
- What are the best ways to make sure families get the information they need when their child has cancer and pre-existing additional needs (such as learning and/or physical disabilities, communication difficulties)?
- What are the best ways to support survivors to share information about their cancer with their own children, partners and friends?
- What are the best ways to support parents to speak up for their child with cancer?
- What are children's experiences of how they were given information at diagnosis, how did this affect their wellbeing and what would they like to be done differently?
- What are children's experiences of Beads of Courage and what is the impact on their understanding of, and conversations about, their treatment and personal journey? (Beads of Courage is a programme whereby at every part of treatment, children receive a new bead. The beads then show a physical representation of a cancer journey, keeping a record of every hospital visit and treatment).
- How can information about the ways in which different cancers can affect development best be shared with health and social care professionals?
- What are the best ways to support families when giving information about a) the possibility of relapse and b) treatment options when relapse has happened?
- What is the best approach to giving information to children and parents about fertility, including the timing of discussions, support with decision



making on fertility preservation, and support for those whose fertility is affected?

- What are the best ways to provide information and support to friends of children with cancer?

### Psychological and social wellbeing

- What is the psychological and social impact of cancer and treatment on children and their families during treatment and in the long-term; what factors affect these impacts?
- Are the psychological, practical, and financial support needs of children with cancer, survivors, and their families being met during treatment and beyond? How can access to this support be improved and what further support would they like?
- What are the best ways to provide emotional support for children and their families 1) around the time of diagnosis, 2) during treatment and 3) after treatment (including survivors who are now adults)?
- What strategies do children with cancer, survivors and families use to cope during and after treatment?
- How can peer support for children with cancer and their families be provided?
- How does the relationship with health professionals impact on emotional wellbeing for families of children with cancer?
- How does the emotional wellbeing of parents and other family members impact on the recovery and wellbeing of children with cancer?
- What are the best ways to raise awareness of the impact on siblings of children with cancer, to ensure they get the support they need?
- What are the best ways to support children with cancer and their families with their spiritual wellbeing?
- What interventions/methods of help are effective in supporting attachment and the relationship between parents and babies with cancer?
- What are the best ways for friends and relatives to support families of children with cancer?
- What are the best ways of supporting children with cancer and their families when the child is receiving end of life care?
- What are the best ways of supporting families following the death of a child with cancer?



### Food and nutrition

- How does a child's diet affect their response to cancer treatment, growth and development?
- How does the level of nutrients (including vitamins and minerals) in the body affect the response to treatment and treatment outcomes for children with cancer? (Outcomes means something that follows as a result or consequence).
- What impact does proton therapy have on nutrition and nutrition-related problems (such as mucositis and nausea) for children with cancer?
- How does the microbiome (gut micro-organisms/ bacteria) affect outcomes for children during treatment? (Outcomes means something that follows as a result or consequence).
- What is the best form of nutrition support for children (such as diet, dietary supplements, tube feeding) and what is the best way to deliver this?
- What are the best ways of giving information about diet to children and their families during and after treatment; are remote consultations appropriate?
- What nutritional or eating related issues do children experience after treatment; what are the best ways to help them with these issues and what is the best diet for improving their long-term health?
- What is the evidence for removing lipid (fat) from parenteral nutrition (TPN or PN) when a child has Sinusoidal Obstruction Syndrome (SOS)? (Parenteral nutrition is a way of getting nutrition into the body through a central line. SOS was known as VOD - Venous Occlusive Disease - and happens when the small blood vessels that lead into the liver and are inside the liver become blocked).
- What are the best ways to prepare children and their families when a child needs a nasogastric (feeding) tube and what are the best ways to manage the ongoing use of a feeding tube?



### Healthcare delivery

- How can services, (such as main treatment centre and local services), for children with cancer be more joined up?
- What are the best ways to support the emotional wellbeing of professionals who care for children with cancer and their families?
- Are services for children with cancer delivered to the same standard across the UK? If not, how can they be made more consistent, including onward referral and moving between services?
- What are families' experiences of care received in different settings (such as Principal Treatment Centres, Shared Care Units, at home); how can care be safely delivered in the place that a family chooses?
- What are the best ways to make sure children get the full range of supportive care services they need? (Supportive care includes the physical, social, educational and psychological aspects of cancer and its treatment).
- What are the benefits of having a nurse specialist in long-term effects to support children after treatment?
- What are the needs of families of children with cancer when they are preparing to leave hospital for the first time and/or just afterwards?
- For children with a brain or spinal tumour, what are the benefits of multi-professional clinics and at what time points during and after treatment are these of most benefit?
- How can the level of care needed by children with cancer in hospital inform staff workload?
- How can children's cancer services be delivered to support children to live as 'normal' life as possible?
- What targeted education is required for health care professionals across all settings (including newly registered nurses) to ensure: skills and knowledge acquisition; compliance with national and local guidelines; the ability to provide families with information and education?
- How can the experiences of children with cancer and their families in hospital be improved? (Including the availability of the facilities, resources, support and equipment they need.)



- How has COVID-19 impacted on care delivery, outcomes and the psychological and social wellbeing of children with cancer and their families and what additional support is required in a pandemic? (Outcomes means something that follows as a result or consequence).

### Causes of cancer, diagnosis and research

- Why do children develop cancer (including the role that genetics plays) and could it be prevented?
- What factors influence a child who is a gene carrier to develop cancer? ('Gene carrier' means you carry a gene that is linked to developing cancer).
- How can knowledge of genetics benefit children with cancer, and identify the risk of their siblings, and their own children in the future, developing cancer?
- What are the best ways to improve collaboration and share knowledge about childhood cancer and treatment between professionals within the UK and worldwide?
- What is the best way to collect information on the rates of childhood cancer across the world and how these vary by country?
- How can time to diagnosis be improved for children with suspected cancer?
- What other conditions are associated with Langerhans' cell histiocytosis (LCH)? (LCH is a rare cancer which occurs when Langerhans cells, part of the body's immune system, build up and form tumours).
- Why do children relapse, how can it be prevented, and what are the best ways to identify relapse earlier?
- What are the best ways to give families information about research and clinical trials on children's cancer, including research they have already taken part in?
- How can research into treatment side-effects and the less common complications children experience be enabled (including, for example, gathering information from online forums for parents/carers)?
- What are the best ways for children with cancer to give their views about their experiences and care to improve cancer services?



# Summary questions in the envelopes at the children and young people's workshop

## Family, friends and pets

1. How can we make sure children and young people can still do the things they want to do with their friends and family (like playing sports and going on holiday)?
2. What can hospital staff do to make sure children and young people feel involved when their brother or sister is in hospital?
3. How can we make the most of open visiting so other family members can be more involved?\*
4. How can we make sure all children and young people can see all family members when they are hospital?
5. What are the best ways to spread awareness to help friends and classmates understand the reality of cancer?
6. How can we help children and young people to stay connected with friends and keep their relationships strong during treatment and afterwards?
7. What are the best ways to help children and young people to keep in contact with family and friends when they are in hospital?
8. How can we make it so children can meet and interact with their pets when they are in hospital?
9. How can we help more children to see therapy animals when they are in hospital?

\*These questions were later combined to make: 'How can we make being in hospital a better experience for children and young people? (like having better food, internet, toys, and open visiting so other family members can be more involved in the child's care)'.

## Treatments and medicines

10. How can we make the experience of having injections (needles) better for children and young people?
11. How can we make the experience of taking medicines better for children and young people? (including having the choice of tablets or liquid medicine).
12. How can we help children and young people to have as much time at home as possible instead of being in hospital?
13. What treatments work the best for children and young people to make them better?

## Being poorly, side-effects and long-term effects

14. How can we help children and young people when treatment changes the way they look? (like when they lose their hair).
15. What helps children and young people when they are feeling poorly?
16. Can we find ways to use treatments that make children and young people feel less poorly? (like feeling less sick, not feeling weak).

## Being in hospital

17. How can we make being in hospital a better experience for children and young people? (like having better food, internet, visitors, toys).\*
18. How can play and distraction help children and young people with procedures? (like having something else to think about or do when having scans and cannulas).
19. What can hospital staff do that helps children and young people to feel well looked after in hospital?

## Emotions, worries and getting help or support

20. What are the best ways to help children and young people with their worries and make them feel happier?
21. How can we help and support children and young people after treatment has finished?
22. What are the best ways to help families with their worries when a child or young person is poorly? (including brothers, sisters, parents and grandparents).
23. How can we help children to meet other children and young people who are poorly like them?

## School and education

24. How can we help children and young people to go to school or nursery during and after treatment?
25. How can we help children and young people to keep up with schoolwork when they are poorly or in hospital?

## Getting the information you need

26. How can we give children and young people the information they want about their illness and treatment in a way that they understand it?
27. What do children and young people need to know about who can help them and their family? (including how charities can help them).
28. How can hospital staff help children and young people to be involved in decisions about them in the way they want to be?
29. How can we help children and young people to talk with their friends and family about their illness?
30. How can we help people to understand about disabilities that you can't see?
31. How can we give children and young people the information they want about their brother or sister's illness and treatment in a way that they understand it?

## Additional questions added by children and young people at the workshop:

1. How can we prevent cancer in children and young people?
2. How can we make more accessible treatments that are closer to home, in shared care hospitals?
3. How can we speed up the process of getting diagnosed and starting treatment in the right place?
4. How can we make sure parents know about the signs of childhood cancer and where to go with their concerns, so they are listened to?
5. What are the best ways to help older family members to understand about childhood cancer and treatments?
6. How can parents be more involved in giving treatments if they want to?

## **This report should be cited as:**

Aldiss S., Hollis R., Phillips B., Gibson F., on behalf of the Children's Cancer Priority Setting Partnership Steering Group (2023) Research priorities in children's cancer. Final report of the James Lind Alliance Priority Setting Partnership.

[www.jla.nihr.ac.uk/priority-setting-partnerships/childrens-cancer](http://www.jla.nihr.ac.uk/priority-setting-partnerships/childrens-cancer)

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