



National Cancer Survivorship Needs Assessment:

# Survivorship after Childhood Cancer

Health needs assessment, 2018



**Cancer survivorship is a distinct period in the continuum of cancer care which begins at diagnosis and continues until the end of life.**

Providing optimal survivorship care can empower patients to achieve their best possible health outcomes while living with and beyond a diagnosis of cancer.

Survivorship after childhood cancer: health needs assessment 2018

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# Contents

|  |           |
|--|-----------|
| <b>Summary</b>                             | <b>3</b>  |
| <b>1. Introduction</b>                     | <b>6</b>  |
| 1.1 Context                                | 6         |
| 1.2 Scope                                  | 7         |
| 1.3 Process                                | 7         |
| 1.4 Structure                              | 8         |
| <b>2. Epidemiology</b>                     | <b>9</b>  |
| 2.1 Incidence of childhood cancer          | 9         |
| 2.2 Survival rates                         | 9         |
| 2.3 International comparison               | 11        |
| <b>3. Literature Review</b>                | <b>12</b> |
| 3.1 Medical & psychosocial needs           | 12        |
| 3.2 Information needs                      | 14        |
| 3.3 Model of care                          | 14        |
| 3.4 Summary                                | 16        |
| <b>4. National qualitative study</b>       | <b>18</b> |
| 4.1 Rationale                              | 18        |
| 4.2 Methods                                | 18        |
| 4.3 Results                                | 20        |
| 4.4 Strengths and limitations of the study | 27        |
| <b>5. Stakeholder consultation</b>         | <b>29</b> |
| 5.1 Key findings                           | 29        |
| 5.2 Summary of findings                    | 32        |
| <b>6. Recommendations</b>                  | <b>33</b> |
| References                                 | 36        |

# Summary

**Cancer survivorship is a distinct period in the continuum of cancer care which begins at diagnosis and continues until the end of life. Providing optimal survivorship care can empower patients to achieve their best possible health outcomes while living with and beyond a diagnosis of cancer. The National Cancer Strategy for Ireland 2017-2026 outlined the need to undertake a Cancer Survivorship Needs Assessment to ascertain the most suitable model of survivorship healthcare for Ireland.**

One of the distinct groups of cancer survivors in Ireland is those who have lived through cancer in childhood or adolescence. Although there are several thousand survivors of childhood cancer living in Ireland, little is known about the physical, psychological and social needs of this unique group. This report aims to assess and document some of the major needs of these individuals.

There are limited epidemiological data available on survivorship after childhood cancer in Ireland, and there is no comprehensive database of survivors in the country. The National Cancer Registry of Ireland estimates that there are approximately 218 new cases of cancer diagnosed annually among those aged under 19 years, and the most common diseases are leukaemia, brain or central nervous system (CNS) tumours, and lymphoma. In recent decades, survival rates have consistently improved, and currently, overall five-year survival for childhood cancer is 81%. Therefore, although the overall number of survivors of childhood cancer in Ireland is unknown, the total number is known to be increasing.

Adult survivors of childhood cancer experience a broad range of late effects from their cancer treatment, including (but not limited to) cardiac disease, infertility, hearing problems, thyroid disease, osteopaenia/osteoporosis, and secondary cancers.

They are more likely to be hospitalised during follow-up than the general population, and they may need long-term monitoring of their risks of late effects. Furthermore, they are more vulnerable to adverse mental health effects from the experience and treatment of cancer; they often have higher levels of psychological distress, as well as a greater burden of cognitive problems in later life. From a social perspective, some survivors experience peer-related challenges, and may underperform academically due to missed schooling and treatment effects. This predisposes them to employment barriers and financial difficulties in later life.

International research has shown that many survivors, and their parents, have unmet information needs, particularly with regard to potential late effects or complications of their treatment. In some settings, survivors receive a summary treatment record (STR) which can help to address information gaps, and can help to coordinate their care. Several countries struggle to provide universal, streamlined follow-up care for adult survivors of childhood cancer; where services exist, they are often under-resourced or access is inequitable. Attendance at survivorship care can depend on multiple factors, including health insurance status, individual experience of complications or recurrence, time since cancer treatment, distance from follow-up services, and cultural factors.

There has been a dearth of research on survivorship after childhood cancer in Ireland to date, and this has left a knowledge gap which needs to be addressed. The National Cancer Control Programme undertook a national qualitative study in April 2018 in order to fill some of this knowledge gap, and to elucidate the major needs of this group. Seven focus groups were held in Dublin, Cork and Galway with 33 participants (15 survivors, 18 parents) who had lived through a range of different cancers in childhood. Five overarching themes were generated from the data:

**(1) Information for empowerment:**

Participants identified substantial gaps in their knowledge of late effects from the treatment they had received in childhood. They sought personalised, age-appropriate information about late effects which they could use to take more control of their own health as adults. Provision of a Summary Treatment Record was perceived as an essential tool for this.

**(2) Communication and coordination of care:**

Survivors typically experienced excellent clinical care from individual healthcare professionals, but poor overall coordination of follow-up care, particularly after transition to adult services. Many survivors had no single point of contact within secondary or tertiary care who had responsibility for their overall needs. This resulted in multiple inefficiencies, including duplication of tests and fragmentation of care.

**(3) Psychological supports:**

There was a strong need for improved psychological support expressed. Many survivors and parents experienced psychological distress, often several years after completing treatment, which they attributed to cancer, and in some cases necessitated crisis intervention. They sought improved access to psychological services through the public system, and enhanced peer support networks and interventions.

**(4) Social supports and adjustment:**

Survivors commonly experienced academic challenges upon return to school. Access to educational support was perceived as unequal and school-dependent. This compounded employment challenges which were experienced by some survivors. Financially, families faced a large burden of expenditure related to cancer treatment and supports, and some described catastrophic levels of health expenditure.

**(5) Navigating the system:**

Survivors encounter multiple challenges trying to access follow-up services, particularly after transition out of paediatric services. Care pathways are not always streamlined, and some individuals have considerable difficulty trying to navigate the system. Participants described the need to constantly fight for clinical and social supports, and some survivors disengage from follow-up care as a consequence.

Multiple stakeholders were consulted as part of this needs assessment, including paediatric oncologists, specialist nurses, voluntary organisation representatives, and survivors, and their views were consistent with those of focus group participants. They emphasised the need to gauge each individual's desire to receive information about late effects, and to consider the optimal timing of this. The Summary Treatment Record was regarded as a necessary component of standard care, and the need for a more standardised approach to provision of fertility-related information

was highlighted. Transition from paediatric services was regarded as a particularly challenging time for survivors and their families, and current resources to support psychological wellbeing were described as inadequate. There was no consensus around the optimal model of survivorship care, and stakeholders suggested that this requires dedicated consideration and consultation going forward.

Survivors of childhood cancer face an array of physical, psychological and social needs. Survival rates have improved for childhood cancer in Ireland, but there is now, more than ever, a responsibility to ensure that these individuals achieve the best possible health outcomes throughout their adult lives.



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# 1 Introduction

## 1.1 Context

Cancer survivorship is a distinct period in the continuum of cancer care which begins at diagnosis and continues until the end of life. Survivorship includes prevention, diagnosis, treatment and post-treatment care and life beyond cancer. A focus on high quality survivorship care can empower patients to achieve their best possible health outcomes while living with and beyond a diagnosis of cancer. There are currently more than 150,000 cancer survivors living in Ireland, and the National Cancer Strategy for Ireland 2017-2026 highlights that there are gaps in current understanding of the needs of this distinct group (1).

There are several thousand survivors of childhood cancer living in Ireland, but the exact number is unknown. Little is known about the perceived physical, psychological and social needs of this group, particularly as they transition from childhood and adolescence in to adulthood. One of the recommendations of the National Cancer Strategy for Ireland is that the HSE National Cancer Control Programme (NCCP) should undertake a Cancer Survivorship Needs Assessment to ascertain the most suitable model of survivorship healthcare for Ireland.

Childhood cancer is unique in Ireland in that the large majority of treatment is centralised in one hospital, Our Lady's Children's Hospital, Crumlin. Some children and adolescents receive active treatment in other institutions, depending on their age or specific diagnosis, and some shared care is also provided by peripheral hospitals with paediatric services, using national treatment guidelines. However, most children receive active treatment in Crumlin at some point.

Survivors of childhood cancer are recognised as a distinct group with their own unique set of needs throughout the lifecourse. The aim of this report is to identify and document the major needs of adult survivors of childhood cancer from the perspective of patients, parents, and healthcare professionals, in accordance with Recommendation 41 of the National Cancer Strategy. It is expected that the findings will help to guide the development of services for survivors of childhood cancer in the future.

### National Cancer Strategy 2017-2026, Recommendation 41:

**The NCCP, in conjunction with the ICGP, cancer centres, the Irish Cancer Society and cancer support centres, will conduct a Cancer Survivorship Needs Assessment to ascertain the most suitable model of survivorship healthcare. The Needs Assessment will be completed by the end of 2018.**

**Figure 1.** Continuum of cancer care. Source: National Cancer Strategy, 2017-2026

## 1.2 Scope

This report is focused on the needs of adult survivors of childhood cancer and their immediate families. For the purposes of this report, “survivors” are individuals who were diagnosed with cancer in childhood or adolescence, before age 18 years, and who have finished their active treatment. It is acknowledged that not all individuals who live through a diagnosis of cancer in childhood identify with the “survivor” label, particularly since the definition of successful treatment has now shifted towards minimising late effects and maximising long-term quality of life.

This report is intended to identify the principal physical and psychological challenges faced by survivors, together with their major social needs. The report uses evidence-informed approaches to describe and quantify the needs of survivors, using epidemiological data where available. Original qualitative data and information obtained through stakeholder consultation has been used to fill information gaps where no data exist in the Irish context.

## 1.3 Process

This work was undertaken over a six month period from mid-January to mid-July 2018. This began with a scoping review of the literature, and exploration of the epidemiological data on this topic in Ireland. It was recognised from the outset that there were limited data on survivorship after childhood cancer in Ireland. This was discussed with the NCCP Executive Team and with the National Cancer Registry Ireland (NCRI), and a decision was taken to complement existing epidemiological data with original qualitative research. Ethical approval was sought (February 2018), and granted (March 2018) to conduct focus groups with survivors of childhood cancer and their parents around Ireland as part of a national qualitative study to fill some of these gaps. The qualitative study was widely promoted in March 2018, and seven focus groups were held in Dublin, Cork, and Galway during April 2018. The findings were analysed during May 2018, and communicated back to participants and stakeholders in June/July 2018. Stakeholder consultation was held with several healthcare professionals concurrently throughout this process.

**Figure 2.** Flow chart to describe the process used for this health needs assessment

|                 |   |
|-----------------|---|
| <b>January</b>  | <ul style="list-style-type: none"> <li>&gt; Scoping review of literature</li> <li>&gt; Review of epidemiological data in Ireland</li> </ul>   |
| <b>February</b> | <ul style="list-style-type: none"> <li>&gt; Scoping review of literature</li> <li>&gt; Discussion with NCCP and NCRI around data limitations</li> <li>&gt; Ethical approval sought for qualitative study</li> </ul> |
| <b>March</b>    | <ul style="list-style-type: none"> <li>&gt; Ethical approval sought for qualitative study</li> <li>&gt; Widespread promotion of qualitative study</li> <li>&gt; Stakeholder consultation</li> </ul>                 |
| <b>April</b>    | <ul style="list-style-type: none"> <li>&gt; Focus groups held in Dublin, Cork and Galway</li> <li>&gt; Stakeholder consultation</li> </ul>  |
| <b>May</b>      | <ul style="list-style-type: none"> <li>&gt; Transcription of focus groups completed</li> <li>&gt; Analysis of qualitative study findings</li> </ul>   |
| <b>June</b>     | <ul style="list-style-type: none"> <li>&gt; Analysis of qualitative study findings</li> <li>&gt; Communication of preliminary qualitative study findings to key stakeholders</li> </ul>                             |
| <b>July</b>     | <ul style="list-style-type: none"> <li>&gt; Synthesis of results</li> <li>&gt; Writing of report</li> </ul>   |

## 1.4 Structure

This report begins with an overview of the epidemiological data in Ireland on survivorship after childhood cancer. This is followed by a summary of the international research on perceived needs in this group, as noted from the scoping literature review. A summary of key results from the national qualitative study follows, together with findings from the stakeholder consultation. Finally, the main messages are summarised, and a suite of recommendations are listed.

## 2 | Epidemiology

There are limited epidemiological data on childhood cancer available in Ireland. Cancer registration has only been in place in Ireland since 1994, and there is no single database for childhood cancer survivors in the country. The total number of survivors of childhood cancer in Ireland is unknown. In 2017, the NCRI published a report on trends in childhood cancer between 1994 and 2014 (2). This contains the best, and most contemporary, epidemiological data on childhood cancer in the country. Key findings of this report are summarised here.

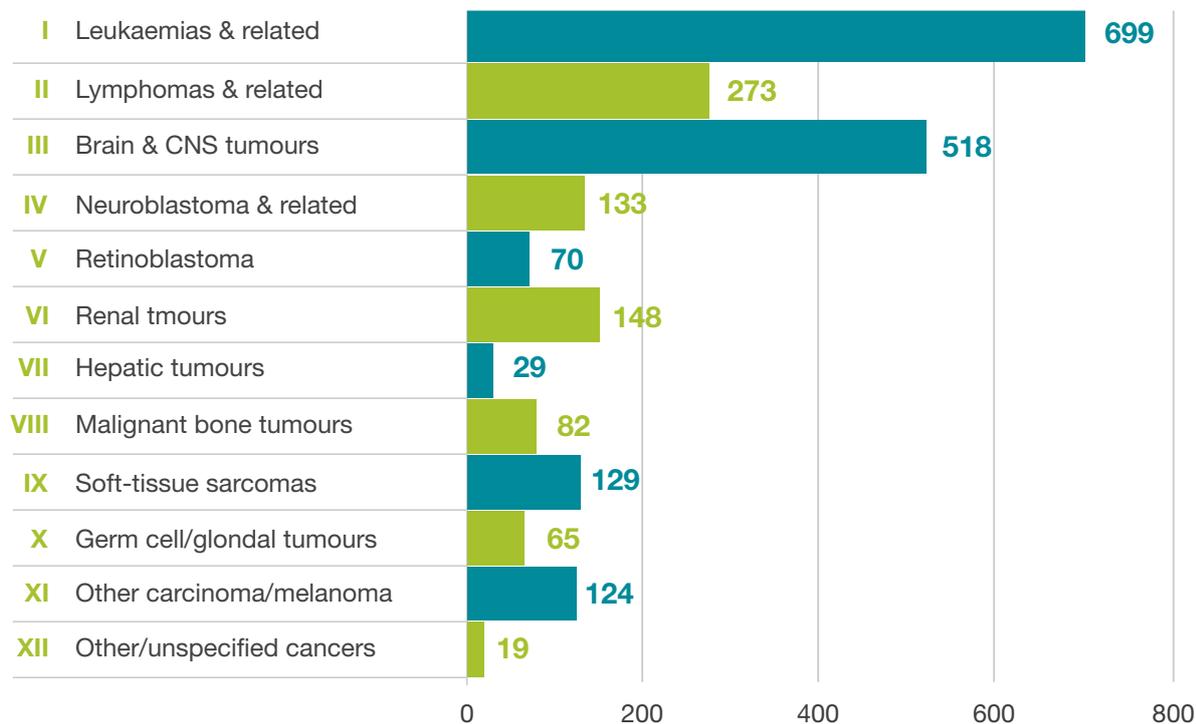
### 2.1 Incidence of childhood cancer

- > On average, there are 160 cases/year of childhood cancer in Ireland in individuals age 0 to 15 years and a further 58 cases/year among the 16-19 year olds. Thus on average, there are 218 cases of cancer diagnosed annually among children and teenagers.
- > About two thirds of all cancers are either haematological (i.e. leukaemia/lymphoma) or tumours of the brain or central nervous system (CNS).
- > Incidence rates of childhood cancer are 8% higher in boys than in girls, but this is not a statistically significant difference.
- > The overall incidence rate of all childhood cancers increased significantly, by 27%, between the time periods 1994-2000 and 2008-2014. Since 1994, incidence rates have been significantly increasing for girls, but not for boys. In recent years there has been a significant increase in the incidence rates of brain/CNS tumours in children.
- > Some cancers are more common among children aged under 6: acute myeloid leukaemia, neuroblastoma, retinoblastoma, ependymoma, hepatoblastoma.
- > Some cancers are more common among children aged over 10: osteosarcoma, Hodgkin's lymphoma.
- > Between 2008 and 2013, two thirds of children with cancer received chemotherapy, and 20% received radiotherapy (with or without chemotherapy).

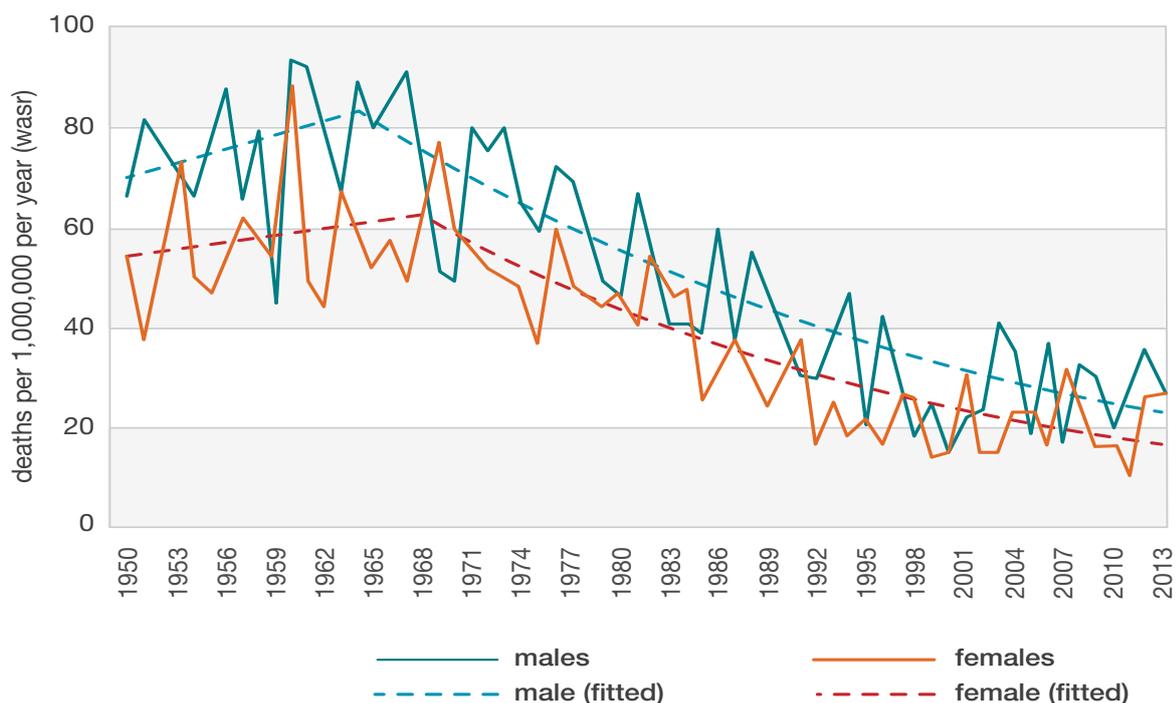
### 2.2 Survival rates

- > Five year survival for all childhood cancers in Ireland is 81% for those diagnosed between 2004 and 2013.
- > Five year survival rates are higher for retinoblastomas (98%), lymphomas (96%), hepatic tumours (95%), germ cell and gonadal tumours (92%), renal tumours (89%), and leukaemias (85%).
- > Five year survival rates are lower for soft tissue sarcomas (76%), brain & CNS tumours (71%), neuroblastomas (69%), and bone tumours (66%).
- > There has been a statistically significant improvement in survival rates for leukaemias between 1994 and 2014 (from 78% to 85%). Other improvements have also occurred, but have not been statistically significant.
- > Of the 2,873 patients diagnosed with cancer before age 15 years between 1994 and 2014, 2,289 (80%) were still alive at the end of 2014 (Figure 3).
- > Of those diagnosed between 1994 and 2014, the majority had been diagnosed with leukaemias (31%), or brain/CNS tumours (23%), or lymphomas (12%) (Figure 3).
- > There has been an ongoing decline in mortality from childhood cancer since the mid/late 1960s by, on average, 2.6% per year for boys and 2.9% per year for girls. Large reductions in mortality have been achieved for leukaemia and lymphoma, but smaller reductions in mortality have been achieved for brain/CNS tumours (Figure 4).
- > A registry-based study indicated that for the time periods 1994-1999 and 2000-2005, there was no clear evidence of regional or deprivation-related variation in childhood cancer survival in Ireland. This may be due to the application of standard treatment protocols nationally, although this study was based on relatively small numbers of incident cases (3).

**Figure 3.** Childhood cancer survivors diagnosed between 1994 and 2014, alive at 31/12/2014, broken down by cancer type. Source: National Cancer Registry Ireland



**Figure 4.** Childhood cancer mortality rate in Ireland (deaths per million per year, world age standardised (wasr)), based on all deaths from cancer at ages 0-14, 1950-2013. Source: National Cancer Registry Ireland



Most common types of childhood cancers in Ireland

1. **Leukaemia**
2. **Brain or CNS tumour**
3. **Lymphoma**

# 218

Approximate number of new cases of cancer diagnosed annually in Ireland in 0-19 year olds



Five-year survival from childhood cancer in Ireland, 2004-2013

**Mortality from childhood cancer has been declining since mid/late 1960s in Ireland**

- ♀ 2.6% per year for boys
- ♂ 2.9% per year for girls

## 2.3 International comparison

- > It is estimated that there are between 300,000 and 500,000 survivors of childhood cancer in Europe. Survival rates have improved substantially in recent decades, and overall five-year survival rates now exceed 80% in high-income European countries (4).
- > Incidence rates of childhood cancer vary, depending on whether CNS tumours are included, particularly because of difficulties distinguishing benign and malignant brain tumours. However, incidence rates of childhood cancer in Ireland are very similar to the European average.
- > Survival and mortality rates from childhood cancer in Ireland compare favourably with other European countries. For the period 2000-2007, 5-year survival for all European countries was 78.2%; in Ireland it was 78.8% (rank 12th of 27 countries) (5).
- > For the period 1999-2005, mortality rates were 21% lower in Ireland than the European average; only Switzerland and Austria had lower mortality rates during this period.

# 3

# Literature Review

A scoping review of the literature was undertaken to identify the major needs of survivors of childhood cancer. Searches were conducted between January and March 2018 using PubMed, Google, and through searching reference lists of relevant articles. Search terms included, but were not limited to, the following: cancer, child\*, adolescen\*, teen\*, surviv\*, follow-up, “follow-up”, Ireland.

It was clear from the outset that the Irish literature on survivorship after childhood cancer was very limited. Given the paucity of research done in this area at a national level, any relevant research from other high-income countries was considered for review.

## 3.1 Medical & psychosocial needs

### Medical needs

Childhood cancer comprises a wide range of diverse diseases. Survivors of childhood cancer are at risk of a myriad of late effects associated with their diagnosis and treatment. Each individual’s risk profile for late effects depends on multiple factors including their age of diagnosis, stage of disease at diagnosis, treatment modality, treatment response, and comorbidities. It was beyond the scope of this review to discuss all of the late effects of childhood cancer in detail. However, it is clear from the literature that survivors experience a considerable burden of late effects.

Some common late effects of childhood cancer include: secondary breast cancer after chest radiation; cardiomyopathy after treatment with anthracyclines, or chest radiation; premature ovarian insufficiency after treatment with alkylating agents, or radiotherapy to ovaries; impaired sperm production after gonadotoxic chemotherapy, or radiotherapy to testes; secondary thyroid cancer after radiation therapy involving the thyroid; ototoxicity after platinum-based chemotherapy, or cranial irradiation (6-10). Some late effects are more likely to manifest in later adulthood. Those treated with steroids or cranial radiotherapy are at greater risk of reduced bone mineral density, and subsequent osteopaenia and osteoporosis

(11, 12). Cardiac mortality is also increased among survivors, and remains higher beyond 50 years of age, several decades after finishing treatment (13, 14). One of the challenges in estimating the incidence and prevalence of late effects after childhood cancer is the international variation in guidelines. An International Guideline Harmonization Group has been established to achieve greater worldwide consensus on the surveillance of late effects and secondary cancers for survivors of childhood cancer (9).

A large Scandinavian cohort study followed 21,297 survivors of childhood cancer over several decades, and reported that survivors had a 94% increased risk of hospitalisation compared with the general population. The risk of hospitalisation was highest for survivors of neuroblastoma, hepatic tumours, CNS tumours, and Hodgkin lymphoma. Survivors spent on average five times as many days in hospital as other individuals during the follow-up period (15). Similar findings were observed among 5,229 childhood cancer survivors in Scotland followed for over five years, where the risk of admission to an acute hospital was significantly elevated relative to the general population, and survivors spent significantly longer in hospital when admitted (16). Survivors remained at higher risk of all-cause mortality than the general population many years after finishing treatment (17). In a Canadian cohort of 2,354 survivors who completed treatment at least five years previously, an increase in relative mortality was observed for cancer-related deaths, and for deaths from circulatory diseases and respiratory diseases (18).

A recent retrospective cohort study of 23,601 adult survivors in North America reported that those treated for childhood cancer more recently had improved health outcomes relative to those treated in previous decades, consistent with efforts to modify childhood cancer treatment regimens to maximise overall survival, while reducing risk of long-term adverse events. However, achieving long-term survival continues to come at a disproportionate cost of adverse physical health outcomes for many survivors (10).

## Psychological needs

The long-term needs of survivors include psycho-social needs which may be related back to their diagnosis and treatment. In the UK, the mental health of 10,488 survivors of childhood cancer was assessed using the Short-Form 36 survey. Compared with the general population, childhood cancer survivors reported significantly higher levels of mental health dysfunction, particularly among CNS tumour and osteosarcoma survivors. Survivors who were older, unemployed, or had lower educational attainment were more likely to experience poorer mental health (19). In the USA, the Childhood Cancer Survivor Study has identified survivors of brain tumours as being particularly vulnerable to adverse mental health effects, with higher levels of psychological distress, a greater burden of cognitive problems, and diminished life satisfaction compared with other childhood cancer survivors (20).

A Danish cohort study of 7,085 survivors indicated that survivors of childhood cancer were at significantly increased risk of hospital contact for mental disorders compared with the general population. The risk was especially high among males and children diagnosed before age 10 years (21). Similarly, a registry-based study in Denmark indicated that survivors had a 40% increased risk of having antidepressants prescribed during follow-up compared with the general population. In that study, the risk of antidepressant use was highest among survivors who had undergone haematopoietic stem cell transplants (22). Increased rates of antidepressant use have also been observed among survivors in Canada (23) and Norway (24).

The long-term physical effects of childhood cancer are likely to impact on the psychological wellbeing of some survivors.

However, not all survivors are concerned about their future health risks. In the USA, a large survey of 15,620 adult survivors of childhood cancer reported that a substantial minority were unconcerned about their health (31%) or future risk of cancer (40%).

Those who had received higher doses of radiation tended to be more concerned about their future health, or their risk of cancer (25).

## Social needs

Survivors of childhood cancer face a range of social challenges at the end of treatment. They are significantly more likely to experience learning difficulties upon return to school, and they are more likely to underperform academically. A considerable minority of survivors also experience peer-related difficulties when they return to school, and survivors of brain tumours are at particularly high risk of both academic and social difficulties in school (26, 27).

In the UK, a cohort study of 10,183 survivors indicated that educational attainment was lower among those who had cancer in childhood, and the most vulnerable groups included those who were treated with cranial irradiation, diagnosed with a CNS tumour and those younger at diagnosis (28). Results from the Childhood Cancer Survivor Study in the USA indicate that childhood cancer survivors are less likely to attend third level education, and they are more likely to be unemployed than the general population (29).

Survivors are more likely to experience financial hardship for a range of different reasons. However, financial vulnerability depends largely on the healthcare and social protection systems which are in place. Thus, research findings may not be generalisable between countries. In the USA, survivors experience more difficulties affording healthcare, and pay higher out-of-pocket medical expenses. They are less likely to be in full-time employment, and they are more likely to report difficulties with obtaining insurance coverage (30, 31). No studies examining this issue among survivors of childhood cancer in Ireland were identified.

### 3.2 Information needs

Survivors commonly report information needs regarding their diagnosis, treatment, and individual risk of late effects. In Switzerland, a survey of 485 survivors and parents identified high levels of unmet information needs relating to late effects of childhood cancer treatment. Survivors were concerned about their knowledge gaps relating to late physical effects, whereas parents were particularly concerned about lack of information around fertility-related issues (32). The Swiss Childhood Cancer Survivor Study also surveyed parental perceptions of the information they received during and after their child's treatment. Relatively few participants reported receiving written information relating to treatment (46%), follow-up (27%) and late effects (19%), although parents were more likely to have received verbal information in each of these areas (33). The majority of parents reported unmet information needs, particularly in relation to late effects (71%).

In the USA, a survey of 523 adolescent and young adult (AYA) survivors recruited from seven population-based cancer registries reported unmet information needs among more than half of study participants, particularly relating to risk of recurrence and late effects. In multivariable analysis, unmet needs were significantly more likely among males, older participants, those from minority backgrounds, and among those with poorer self-reported health (34).

Some institutions provide survivors with a summary treatment record (STR) and/or a survivorship care plan (SCP) at the end of treatment, or at transition, in a bid to address perceived information gaps, and to coordinate overall care for the survivor. In Canada, a national survey of all paediatric oncology institutions indicated that a STR and/or SCP was provided to each survivor prior to transfer out of paediatric care in most institutions (13/17; 76%), but this was not universal (35). In the USA, 68% of institutions in the Children's Oncology Group provided survivors with a copy of their SCP (36).

A cross-sectional study of 1,395 American AYA survivors reported that only 30% had received a STR at the end of treatment. Receipt of a STR was associated with significantly lower odds of reporting unmet information needs regarding late effects, and fewer concerns about cancer recurrence (37). Similarly, a cross-sectional study of 376 survivors of childhood and AYA cancer reported that 48% of survivors had no STR, and 55% had no survivorship care plan (SCP). The majority of these survivors were already attending formal survivorship services, yet their confidence in managing survivorship care was low, particularly if they lacked a SCP (38).

### 3.3 Model of care

Long-term follow-up is warranted for many survivors of childhood cancer, to oversee their care and to prevent adverse outcomes. There is considerable variation in the different models of survivorship care available in different countries and in the services offered to individuals who have experienced childhood cancer. Most of the existing research has come from the USA, which has a predominantly privatised funding model for healthcare, or from Canada, which has a predominantly publicly-funded system. Although limited comparisons can be drawn between the Irish healthcare system and these models, it is nonetheless important to consider how services are configured elsewhere.

In 2012, a Europe-wide survey of paediatric oncology institutions was undertaken to identify the availability of long-term follow-up services for survivors of childhood cancer. Among 110 participating institutions, the availability and content of services varied widely.

Two thirds of institutions reported having follow-up services for paediatric survivors, and only 38% provided follow-up for adult survivors of childhood cancer. Services were typically located within paediatric hospitals and run by paediatric oncologists, but several services were challenged by a lack of dedicated personnel, time and resources (39).

In the Children's Oncology Group, a partnership of paediatric oncology centres predominantly in the USA, late effects services were provided for survivors of childhood cancer in the majority (87%) of institutions.

Specialised long-term follow-up was overseen by a designated provider for paediatric survivors in 59% of institutions, and for adult survivors in 47% of institutions (36).

In Canada, a national survey of 17 paediatric oncology centres indicated that 12 centres (71%) had a formal programme or clinic offering survivorship care, and only six centres (35%) had access to a formal programme for survivors once they reached adulthood (35). Another Canadian survey of paediatric oncology institutions indicated that most centres (87%) provided timely access to specialist services for paediatric survivors, although this information was reported by heads of paediatric services, and not based on patient feedback. Adult survivors of childhood cancer had more limited access to services, and these patients were frequently lost to follow-up (40).

In the USA, a survey of 12 paediatric oncology centres reported that 11 centres offered dedicated survivorship clinics after childhood cancer. All clinics were staffed by paediatric oncologists, but there was considerable variation in the availability of other healthcare staff for these clinics. Transition from children's to adults' services was a particular challenge, with median age at transition of 32 years for attendees (41). This survey was confined to New England, and thus may not be representative of the USA more widely.

The study did not indicate whether access to services was needs-based, or whether private health insurance was a determining factor.

A survey of 145 institutions across the USA providing care for children with brain tumours identified high levels of participation in follow-up clinics, but variation in the content of services.

One third of institutions offered dedicated neuro-oncology follow-up clinics for survivors, and they were significantly more likely to use neuro-psychology testing and continued surveillance imaging than general follow-up clinics. Perceived barriers to care for survivors included lack of health insurance, and lack of dedicated funding and clinical time for providers (42).

Engagement in long-term follow-up care may have benefits at a health service level. In Canada, those who attended at least one survivorship clinic appointment as an adult survivor were 19% less likely to attend an emergency department during follow-up. For each additional visit to a survivorship clinic, the rate of emergency department visits decreased (43).

### Attendance at follow-up

Where follow-up services exist after childhood cancer, a number of factors may influence the decision of the survivor, or the parent of the survivor, to attend. In Tennessee, USA, a cohort study of 941 survivors attending a single paediatric oncology centre reported that 15% of eligible patients did not attend for follow-up appointments. Non-attendance was significantly more likely among uninsured patients, those who had not experienced secondary cancer events, and among ethnic minorities.

In Switzerland, 189 parents of 11-17 year old survivors were surveyed about attendance at follow-up clinics, a mean 11 years after diagnosis. Non-attendance was significantly more likely among those with lower perceived control over late effects, and among those who were longer after finishing treatment.

Parents who reported higher unmet information needs were significantly more likely to continue to attend follow-up clinics (44).

In Ontario, Canada, attendance at survivorship clinics is free at the point of access. However, a cohort study of 3,912 adult survivors of childhood cancer reported that only 43% of survivors attended at least one follow-up clinic as an adult during a median 7.8 years of follow-up.

Attendance was particularly low (26%) among survivors of CNS tumours. Rates of attendance were significantly higher among females, those with higher socio-economic status, and among those who had higher treatment intensity, radiation, or alkylating agent exposure. Distance from the follow-up service was a significant predictor of attendance, with those living >50km away least likely to attend (45).

These findings suggest that barriers to providing appropriate care to survivors of childhood cancer are likely to include both a lack of specialised survivor clinics and, where such clinics do exist, suboptimal attendance among some survivors.

### Role of primary care

The existing literature suggests that survivorship care should be shared between the hospital and the community, but that there is a need to improve communication between paediatric oncology services and primary care. Few countries have existing national efforts to educate primary care physicians in this area, and the relative rarity of childhood cancer makes this more challenging to prioritise (46).

A systematic review of 26 articles found that GP involvement in survivorship care varies considerably between settings. There are two commonly-described models: GP-only care, and shared care between GP and paediatric oncology. The shared care model confers advantages over GP-only follow-up.

Essential components of successful follow-up include well-organised transition arrangements, provision of a STR to patients, provision of a SCP, updated clinical guidelines, and GP education on survivorship care (47).

In the UK, a survey of 65 clinicians working in paediatric oncology treatment centres was combined with a postal survey of 10,979 GPs. Only 45% of clinicians reported that they discharged patients after treatment for childhood cancer, but of those, 97% were discharged back to GP care. The majority of GPs (65%) reported that their patients were not receiving regular hospital follow-up.

This suggests variation in practice in discharging survivors back to primary care, and that in some cases, survivors may be exclusively followed up by their GP (48).

In the Netherlands, a questionnaire was administered to 233 GPs regarding their motivation to participate in the regular follow-up of childhood cancer survivors. Most respondents (97%) were willing to participate in a shared care model.

The main requirements cited for shared care were clear guidelines, sufficient information about the patient's history, and easy lines of communication with the hospital. However, these respondents were engaged in a postgraduate course on late effects of cancer treatment, and may not be representative of all GPs (49).

### 3.4 Summary

Survivors of childhood cancer experience multiple medical, psychological, and social needs after completing treatment. They are at risk of a range of medical complications including, but not limited to, secondary cancers, cardiac disease, reduced fertility, and bone disease. Survivors are more likely to be hospitalised in the years following treatment, and they tend to spend longer periods in hospital compared with the general population.

Survivors experience a disproportionate burden of anxiety and psychological distress following treatment, and those with brain or CNS tumours are particularly vulnerable to adverse mental health effects and cognitive problems. Several studies have demonstrated heightened rates of anti-depressant use among survivors of childhood cancer, although not all are equally impacted; a substantial minority remain unconcerned about their future health risks, and remain resilient to the adverse psychological effects.

Many survivors experience social challenges during follow-up, particularly upon return to school, and they face lower levels of educational attainment and peer-related difficulties. Financial hardship is common in adulthood, due to a combination of reduced educational and employment opportunities, and higher out-of-pocket healthcare expenditure in some settings.

Survivors and their parents commonly report unmet information needs, particularly with regard to the risk of cancer recurrence, late physical effects of treatment, and fertility-related effects. In some settings, the STR and SCP are used to address some of these gaps and to coordinate follow-up care. However, these are not universally available.

Provision of long-term follow-up services can confer benefits at a health service level, by reducing hospitalisation rates for survivors. The availability and content of follow-up services varies widely between institutions across Europe, USA and Canada. Common challenges include lack of dedicated time, resources and personnel within institutions to deliver this care, while perceived barriers to access for survivors include lack of healthcare insurance in countries with privatised funding models.

Transition to adult follow-up services is difficult in settings where formal survivorship services do not exist, and many of these adults are lost to follow-up. Where these services do exist, survivor attendance is impacted by their distance from the service, time since finishing treatment, and perceived control over late effects.

GP involvement in survivorship care should be as part of a shared care model with the hospital team. In some settings, GPs are willing to be involved in long-term follow-up of survivors, provided the essential components of shared care are put in place. These include streamlined transition arrangements to adult services, sufficient information about the patient's history, provision of a STR and SCP to the patient, updated clinical guidelines, clear lines of communication between primary care and the hospital team, and availability of education on survivorship care for GPs.

There is a paucity of research on survivorship after childhood cancer in Ireland, and thus, most of the literature in this review refers to settings elsewhere in Europe or in North America. Although not all findings may be generalised to the Irish context, there are common issues highlighted across these settings which point to some of the major needs experienced by survivors of childhood cancer.

# 4 National qualitative study

## 4.1 Rationale

At the outset of this needs assessment, it was recognised that there were limited data available on survivorship after childhood cancer in Ireland. This gap has also been highlighted in section 3 above. A qualitative study was planned to address some of the data gaps and to identify the major needs of this group.

## 4.2 Methods

### 4.2.1 Study design

A focus group methodology was chosen as this study design can provide detailed information on an area where there is insufficient prior data or knowledge (50). Adults (over age 18) were invited to participate if they were diagnosed with cancer before age 18, and were no longer under active treatment for cancer. Parents of children who had cancer within the past 20 years, and who were no longer under active treatment, were also invited to take part.

Seven separate focus groups were conducted over a three week period in April 2018, and each focus group comprised of survivors only, or parents of survivors. Focus groups were conducted in Dublin, Cork, and Galway, in four different locations including NCCP offices, a public health department, a hotel, and a voluntary organisation. Focus groups were intentionally scheduled to occur on a mixture of weekdays and weekends, as well as during working hours and evenings to accommodate a diverse range of participants. Participants were encouraged to attend in person, but teleconference facilities were provided for anyone unable to do so. The maximum number of participants in any single focus group was eight.

### 4.2.2 Study promotion

The study was widely promoted by the NCCP, Health Service Executive (HSE) Communications Division, and by voluntary organisations all around Ireland. The study was frequently advertised through social media during the month of April 2018 on the Twitter accounts of HSE (>28,000 followers), NCCP (>800 followers), Irish Cancer Society (>38,000 followers) and several other voluntary organisations (several thousand followers combined).

Email alerts were sent out to 45 voluntary organisations around Ireland working with cancer survivors to cascade the message. Individual meetings were held between NCCP and three of the leading voluntary organisations working in the area of childhood cancer to encourage promotion of the study. Professional networks of medical oncologists, haematologists, and specialist nurses were informed of the study and encouraged to invite participants. Email alerts were also sent to paediatric oncologists and haematologists to disseminate information on the study to interested participants.

A broadcast email was sent by the HSE Communications Division to all HSE staff members nationally (>115,000 members) to alert the wider public of the study and to encourage participation. The study was also promoted in a national broadsheet (Irish Times) and through local radio.

Fifty expressions of interest were subsequently received to participate in the study.

**Figure 5(a) and 5(b).** Advertisements used to promote survivorship study on social media.

#### 4.2.3 Data collection

Each focus group was facilitated by a trained researcher from NCCP and a scribe was present to take notes. Participants were first asked to write down or doodle any thoughts or ideas they had regarding the major needs of survivors of childhood cancer in Ireland. After five minutes, each participant was asked to feed back individually to the group. After each participant had the opportunity to speak, two or three common topics or themes were chosen for more in-depth discussion between all participants. These topics were agreed by consensus between the interviewer, scribe, and focus group participants.

An open-ended topic guide was developed, and this was designed to elicit free-flowing discussions between participants, while still identifying specific long-term needs of survivors. Where possible, probes were used to encourage elaboration on how gaps or deficiencies in services might be addressed or improved.

After each focus group, a debriefing process took place between the interviewer and scribe to identify areas where more details were required, to reflect on the predominant emotions expressed by participants, and to modify the topic guide as needed. Focus groups lasted between 35 and 120 minutes and were audio-recorded. The recordings were transcribed verbatim by an independent company.

#### 4.2.4 Ethical considerations

Ethical approval for this study was obtained from the Clinical Research Ethics Committee of the Cork Teaching Hospitals in Ireland in March 2018. All participants were reminded of the voluntary nature of the study, and gave written informed consent to participate. No identifiable details were collected.

#### 4.2.5 Analysis

Braun & Clarke's 6-step Framework for Thematic Analysis was used to identify semantic and latent themes using an inductive approach. Open coding was used to develop and modify initial codes. Short segments of the transcripts that consisted of comments, phrases and sentences from participants were first coded. Two researchers undertook manual line-by-line coding of the first three transcripts independently, and these were reviewed for consistency and face validity. The remaining transcripts were then coded by the lead researcher.

Related codes were grouped together to form categories. These categories were then continuously reviewed to identify and generate themes and subthemes. Two members of the study team audited the codes, categories, themes and subthemes to ensure they were objectively interpreted and to minimise potential for bias. This iterative process was repeated until there was consensus with the generated themes.

Member-check was undertaken with a number of participants after the study team had interpreted the findings, and completed a preliminary analysis. Informant feedback confirmed that the summaries reflected participants' views, feelings and experiences.

### 4.3 Results

In total there were 33 participants: 15 survivors and 18 parents. The majority of participants (n=27) were female. Of the six male participants, four were survivors and two were fathers of survivors. Twenty nine participants attended the focus groups in person, and four participated by teleconference. Survivors had experienced a diverse range of diseases, including leukaemia, lymphoma, brain/spinal tumours, and osteosarcoma, and their age at the time of diagnosis ranged from infancy to late adolescence. The majority of survivors were currently aged 18-32 years. Participants came from a range of counties in Ireland, including Cork, Dublin, Galway, Kildare, Leitrim, Louth, Mayo, and Meath.

The following five themes were generated from the analysis:

- Information for empowerment
- Communication and coordination of care
- Psychological supports
- Social supports and adjustment
- Navigating the system as a survivor

The results below are a shortened summary of the full results of the study. They reflect the consensus or majority viewpoint for each topic or theme. Where appropriate, relevant minority views, or clearly opposing views, are also presented. Quotes have been used to illustrate the point where appropriate.

The full results of the study are available in a separate document, accessible through the NCCP website.

#### 4.3.1 Information for empowerment

Participants identified substantial gaps in their own knowledge of long-term effects after treatment for childhood cancer. Some felt that they did not receive enough information about long-term side effects, and they felt unable to recognise the warning signs of treatment complications. Currently, many survivors and parents undertake independent enquiry to try to inform themselves of their individual risk profile, but this is challenging to do without specialist knowledge or advice. They do so using the internet, by approaching voluntary organisations, or by making direct contact with healthcare providers and hospitals.

“...it is all a fine thing to say this was my diagnosis and I was treated for X length of time in (the hospital) but that actually doesn't mean anything, I actually don't know any more beyond that”

Female survivor, Focus group 7 (Galway)

Fertility was a key area of concern, and participants regarded fertility information as a necessity for patients and families at the time of diagnosis. Although some participants had received fertility information at the time of diagnosis, this discussion often happened in an ad-hoc manner, and there was no standardised approach to this.

“The only reason that we kind of talked about (fertility) was because my Mam had brought it up, I wouldn't have brought it up otherwise and the hospital wouldn't have either”

Female survivor, Focus group 1 (Dublin)

Survivors sought personalised information so that each individual could overcome some of their own information gaps, and could be empowered to take more control over their health. They felt that this should be done in a way which is age-appropriate, and which acknowledges the varying levels of interest of survivors in receiving this information.

The timing of this information should also be sensitive to the needs of the individual at any given time in their continuum of care; some will want to know the potential late effects from the outset, whereas others may only want to receive information several years after finishing treatment.

“...some people just want to have nothing to do with it and leave that in the past but then there are others, but like even just to have that option there... they don't have to relive it, it just works out that (personalised information) is there if they like”

Male survivor, Focus group 7 (Galway)

Some participants had received a summary treatment record (STR) on completing treatment, on transfer out of paediatric services, or after directly requesting it from their healthcare team. Others had no concise summary of their previous treatment, and were unaware that the STR could be requested. Participants perceived that universal access to a STR could help to overcome many of their own information gaps. Their preference was that the STR should be prepared for all survivors at the end of treatment by default, and that it should be available for them if they ever needed it in the future.

“...some sort of summary that... when I go to a wedding in Poland in June and you know if something happens there that this information would be available”

Male survivor, Focus group 4 (Dublin)

Some participants had requested a STR, either independently or with the assistance of voluntary organisations. However, a number of barriers to obtaining this were mentioned by participants, including reluctance on the part of survivors to return to their treating hospital, logistical difficulties for the hospital retrieving the records, and the perception that their requests were being dismissed by some healthcare professionals.

### 4.3.2 Communication and coordination of care

Participants described receiving excellent care from individual clinicians, but poor overall coordination of their care, particularly after transfer out of paediatric services. Reasons for this included the lack of a unified information system across hospital sites and the lack of an electronic health record. Scans and other investigations were frequently duplicated across hospital sites if they could not be accessed or interpreted on a common information platform.

“...when she has MRIs in Galway or in Dublin there is no way that the guy in Dublin can read (the scan) in Galway and vice versa, so if there was a system that they could all be of the one...”

Mother of survivor, Focus group 5 (Galway)

“...my chart is about like four phone books, there is nothing electronic or anything, it's not something that you just email that over”

Male survivor, Focus group 4 (Dublin)

A minority of participants gave examples of coordinated appointments, joint survivorship clinics, and effective communication between hospital consultants in these shared clinics. However, survivors were regarded as being very fortunate to benefit from this model of care; this was perceived to be “luck of the draw” rather than a standard model. Typically, survivorship care was fragmented within individual hospitals; follow-up appointments were rarely coordinated, and members of the multidisciplinary team often worked independently of one another rather than making treatment decisions as a collective.

“...the renal doctor isn’t communicating with the cardiologist, the cardiologist isn’t communicating with the psychologist – her needs are multi-dimensional so her care needs to be multi-dimensional”

Mother of survivor, Focus group 2 (Dublin)

Survivors expressed frustration that they needed to repeat their full medical history multiple times to different healthcare professionals and, in some cases, poor communication between healthcare teams caused them to disengage from follow-up care.

“...they just have to keep telling different people the same story every single time and they just disengage”

Mother of survivor, Focus group 2 (Dublin)

Many participants felt that survivors were being “tossed around a lot” within the current system because they had no single point of contact (SPOC) for their follow-up care.

There was a prevailing perception that there was a lack of clinical ownership over their follow-up care, and participants perceived that the SPOC could play a vital role in coordinating their care, and in overcoming communication barriers between healthcare professionals. The SPOC was described as someone who could understand medical information, advocate for survivors, help them to navigate the system, and access services throughout follow-up. Participants also described the roles which the SPOC should fulfil (Table 1). When probed as to who should fill this role, most participants described a dedicated clinical nurse specialist or advanced nurse practitioner. Other suggestions were a dedicated medical Consultant, dedicated social worker, or specialised GP.

Separately from the SPOC, some participants suggested that a dedicated late effects clinic could provide more coordinated follow-up care for survivors. They described how this clinic should be a place where “you are not telling your story again and again every time”, and where appointments between different specialties were scheduled to coincide.

The ideal model for a late effects clinic was described as being in a single location, with members of the multidisciplinary team in separate clinic rooms but communicating directly with a coordinator of care or SPOC.

**Table 1.** Suggested roles of the Single Point of Contact

| Suggested roles of the Single Point of Contact  |
|---|
| Provide basic information to survivors relating to their personal treatment history and risk of late effects          |
| Provide answers to questions from survivors and families, and provide reassurance                                     |
| Enable survivors to better understand their own treatment effects, and long-term risks                                |
| Act as a central coordinator of multidisciplinary follow-up care for survivors  |
| Arrange and/or oversee follow-up investigations   |
| Receive the results of all investigations related to survivorship   |
| Act as a gatekeeper for other resources and services. Facilitate and/or administrate access to supports and services. |

### 4.3.3 Psychological support

Participants articulated a strong need for improved psychological support for survivors during treatment and follow-up. Those who had received psychological support spoke positively about it, but some had experienced considerable difficulties accessing services for this. Several survivors experienced a delayed trauma years after finishing treatment, which they attributed to their cancer diagnosis and treatment.

A small number of participants described very serious mental health effects which they attributed in part, or in full, to their diagnosis or treatment. One survivor described his need for crisis intervention for suicidality; another survivor suffered a “mental breakdown” and dropped out of university; and one parent described how her child had been hospitalised with repeated self-harm and attempted suicide after completing treatment.

“I ended up getting worse and worse, I went to Pieta House when it got to an intervention stage”

Male survivor, Focus group 1 (Dublin)

Participants were not routinely asked about their overall psychological wellbeing and coping during follow-up appointments, and their preference was that all survivors should be offered psychological support by default. Survivors who were able to avail of formal psychological support on the public system described those services as “gold dust”, whereas many others described how they were “left in the lurch”, because they had to search for psychological support independently, often at considerable financial expense. Participants viewed these gaps in services as short-sighted and a false economy; they felt that by having their psychological needs addressed, they would avoid the need for later treatment and be better able to participate in education and employment, and ultimately contribute more to society as a result.

“...they will feel more confident... just feel better about themselves and probably want to get back to work and feel more able with college or whatever”

Female survivor, Focus group 1 (Dublin)

There was no consensus on the optimal format psychological support should take, with some participants articulating a need for individual counselling, while others preferred group-based support. It was acknowledged that some survivors would not want to avail of psychological support, and for others, informal peer support networks provided a valuable alternative to this. There was no single service model suggested which could meet all perceived needs.

“...getting to see people who I perceived as normal who had gone through it was enough for me and that was the first time I was optimistic about the whole lot”

Male survivor, Focus group 4 (Dublin)

Parents expressed concern for the psychological wellbeing of other members of the family unit, and not just the survivors. They articulated a need for support for siblings who had sometimes been overlooked during the treatment phase. Parents also experienced considerable personal distress and readjustment challenges, particularly around the time of diagnosis or at the end of treatment. They perceived that there was a lack of psychological support available to them, except for cases where their distress had culminated in a mental health crisis. They did not describe the format which the optimal service model should take, but felt that it should be visible, accessible, and offered by default.

“...when you get go-ahead that things are going to be okay that is often when the parent collapses and she falls down ill and falls apart because it is a very very lonely road as a parent”

Mother of survivor, Focus group 5 (Galway)

“(My husband) took the psychological effect of it very very badly... and there was no talking, if he had felt it was an automatic thing... if only it was just there and it wasn't something you had to look for... (he would have spoken about it)”

Mother of survivor, Focus group 2 (Dublin)

#### 4.3.4 Social supports and adjustment

Social readjustment was a challenge for survivors for a myriad of reasons. Educational challenges were commonly experienced. Survivors had difficulties keeping up with their peers on return to school both academically and socially, and often experienced memory and cognitive challenges. Some survivors underestimated the challenges they would face when returning to school, particularly those diagnosed with cancer during adolescence. Often, their priority was to rejoin the same peer group despite missing long periods of school, and some survivors described how they underperformed in school-leaving exams as a consequence of this.

Some survivors received practical supports, including special needs assistants (SNAs) and disability supports, but participants perceived that allocation of these resources was not streamlined, and that eligibility criteria needed clarification.

“...we were very very fortunate that in the beginning he had an SNA for Junior Infants... Other children aren't as fortunate because the school systems differ in every way so I do think it needs to be streamlined and it's automatic”

Mother of survivor, Focus group 2 (Dublin)

Employment barriers were experienced as a result of educational challenges, and survivors expressed a need for more widespread employment access programmes and workplace supports, including improved opportunities for flexible working. Parents who had given up jobs around the time of diagnosis were also very vulnerable and had difficulties re-entering the workforce.

“I am very employable, well able to work, but I suppose I just need to maybe do a shift or two less than the average person just due to sometimes I just get a little more tired... it took a good year or two to kind of reduce just like a couple of hours in work”

Female survivor, Focus group 1 (Dublin)

Families affected by childhood cancer commonly experienced a considerable financial burden, and some described catastrophic health expenditure. There were multiple factors which contributed to this, including time away from work and associated loss of income, travel and accommodation costs during treatment and follow-up, out-of-pocket payments for multidisciplinary team supports, GP care and medication expenses, and education costs. These families felt “financially crushed” and described how they had needed to borrow money from relatives and friends, or in one case, they had to re-mortgage their house. One parent stressed the need for greater information for parents about financial supports.

Survivors also inherited a financial burden in adulthood, in part due to employment barriers, expiry of their medical card, and difficulties getting insurance.

“...it takes some families ten years to recover from a childhood cancer diagnosis and you have some cases where I’d say the families never recover because the parents lose jobs, don’t return to work for a myriad of reasons and particularly people who run their own businesses suffered big big time”

Mother of survivor, Focus group 2 (Dublin)

“...like I found I didn’t go to (my) GP to get my ninety euro inhalers and pay sixty quid to see the GP, and I wouldn’t go with chest infection, I would leave it until it was full on pneumonia and then requiring emergency admissions”

Female survivor, Focus group 1 (Dublin)

For many participants, the expiry of the medical card was a key issue of concern. Several participants expressed dissatisfaction over the “invisible deadline” five years after diagnosis when their medical card expired, and how their financial difficulties were compounded at this time. Some described frustration over the inclusion of leukaemia on the long-term illness scheme, but not lymphoma or other common childhood cancers.

“We found it extremely hard to get a medical card, she had to be diagnosed a second time with her brain tumour before we got that which was very very tough”

Mother of survivor, Focus group 5 (Galway)

Some survivors also struggled with their sense of identity at the end of treatment, and with the challenges of getting back to “normal life”. They felt like a “mismatch” or the “odd one out”, and experienced challenges fitting back in with their peers. This struggle was compounded by hair loss in some cases, which marked them out as different, and in some situations, inadvertently isolated the survivor. They perceived that the challenges of social readjustment were often underestimated, and in some cases, were just as difficult as the treatment itself.

#### 4.3.5 Navigating the system

Survivors of childhood cancer experience various challenges trying to navigate the health services after transfer out of paediatric care. At this time of transition, many of them feel unwanted by adult services, and there is a perceived lack of clinical ownership over their care.

“I was at the children’s hospital and... they wanted to outsource me to the adults, the adults said I was too young so I was left in limbo for two years and no one did the drill that could have saved the hip”

Female survivor, Focus group 1 (Dublin)

Some survivors began to disengage from their follow-up care, or became non-compliant with clinical advice or with medication around this time. Parents suggested that the reasons for non-compliance included poor communication from the health system, lack of ready access to the services the survivors wanted or needed, a sense of exclusion from the system, and a natural teenage phase of rebellion for some individuals.

Survivors and parents articulated a sense of fear around the time of transition, and this was compounded by the perceived challenges in accessing supports as an adult survivor outside of the paediatric system. For some, this came as “a big shock to the system”.

Three participants suggested that the introduction of an Adolescent and Young Adult (AYA) treatment unit would help to address some of the shortcomings in the current system at the time of transition. During follow-up, they felt that a dedicated AYA service could help to ensure a smoother transition to adult services was achieved.

Despite receiving excellent clinical care during treatment, some survivors expressed concern over gaps in their medical follow-up. Care pathways were not always streamlined, and gaps in follow-up were a source of concern. Several participants had experienced late effects, and some perceived that their medical complications could have been averted if there had been greater monitoring during follow-up. Two participants described severe cardiac complications, and that they had never realised that this was a risk for them. One survivor had experienced cardiac failure in late pregnancy, requiring ICU admission and needed to have an emergency delivery via Caesarean section. One parent described how her teenager developed cardiac failure years after the end of treatment, and ultimately required cardiac transplant for this. In both cases, participants felt that they had not been warned of the possibility of this late effect, and that this prevented them from being diagnosed in a timely manner.

“...she got transplanted in December which was amazing but horrific, she is doing brilliantly since but we are in a kind of another world you know so these are the very real consequences of cancer treatment and nobody told us, you know we weren't prepared for it”

Mother of survivor, Focus group 2 (Dublin)

Several participants had experienced dental complications after finishing their treatment. Some had tooth decay, shortened roots, or needed root canal treatment as a consequence of their chemotherapy, and they described a particularly high risk of dental problems among those who had received oral chemotherapy agents. Survivors articulated the need to attend multiple dental appointments, and they expressed high levels of concern related to the cost of this. Parents were also anxious about the need to fund all dental follow-up independently.

**Table 2.** Summary of participants' experiences of fighting for information or services

| Areas where participants described the need to fight                             |
|--|
| Fight for information relating to their own diagnosis or treatment               |
| Fight for summary treatment record   |
| Fight for clinical services and appointments                                     |
| Fight for surgical interventions, including joint replacements                   |
| Fight for education supports, such as Special Needs Assistants                   |
| Fight for multidisciplinary supports, such as physiotherapy/occupational therapy |
| Fight for medical card or long-term illness card                                 |
| Fight for rehabilitation supports after neurosurgery                             |
| Fight for psychology services, and specialist neuro-psychology services          |
| Fight for financial or other supports from the health service                    |

Participants experienced considerable difficulties accessing services, particularly after transition to adult services. They described the need to be “constantly fighting” for a wide range of supports, summarised in Table 2. Participants recounted high levels of stress associated with this, and they found it to be “exhausting emotionally”.

Some individuals recounted physical illnesses deteriorating considerably over time while awaiting appointments. This led to a sense of fear among some participants that they would be unable to access services when needed, and a perceived need to purchase health insurance to overcome this barrier.

“...an urgent appointment for occupational therapy took one year and that’s because I rang them every single week”

Mother of survivor, Focus group 2 (Dublin)

“...it always carries a sense of fear and concern around accessing the services”

Mother of survivor, Focus group 3 (Cork)

The challenges of navigating follow-up care impacted on family dynamics. In particular, survivors experienced guilt around involving their parents in follow-up care, whereas for some parents, their own guilt made them reluctant to withdraw from follow-up. Some survivors expressed their reluctance to divulge all information to their parents, whether this related to late clinical effects or psychological distress. Although this may have been symptomatic of a desire for increased independence, it may also have related to survivors’ strong sense of guilt about upsetting family members.

“I just sometimes think I am a burden and I know I shouldn’t but I do with everything that has happened”

Female survivor, Focus group 6 (Galway)

Some parents described how they strove to balance vigilance for symptoms or late effects without smothering their child. However, they perceived that they still needed to know what was happening with their child’s healthcare throughout survivorship. One mother described her sense that “you just can’t withdraw” around the time of transition when the young person is still very vulnerable, but that this is sometimes perceived as parents wanting to encroach on their child’s independence.

#### 4.4 Strengths and limitations of the study

This is the first detailed study of perceived needs conducted among survivors of childhood cancer in Ireland, and it fills a gap in the existing literature. A multi-pronged promotion strategy was used for this study, including social media, print media, radio, correspondence with key stakeholders, and email dissemination via the HSE and a range of voluntary organisations nationwide. This ensured that the study had a wide reach, and it gave survivors and parents multiple opportunities to take part.

Open-ended questions were asked in the focus groups to ensure that complex topics could be discussed, and to identify nuances within group perceptions and beliefs. These offered opportunities for clarification of information points, and allowed all participants to contribute individually, as well as in groups. By asking individuals to write down and/or share their own thoughts at the outset, efforts were made to include all participants in discussions, and to avoid disproportionate focus on the dominant view.

A robust coding process was used, whereby codes, categories, themes and subthemes were audited to enhance trustworthiness of the analysis. Member-check was also used to ensure the analysis aligned with participants’ views.

There are a number of limitations to this study. The sample was self-selected, and the majority of participants were female.

Those who took part were more likely to be interested and engaged in this area, and may not be representative of the wider population of childhood cancer survivors in the population.

This may have limited our ability to capture the full range of perceived needs which exist among survivors and their parents. Although no personal details were collected in this study, some participants disclosed this information unprompted. They had experienced several different diseases and came from at least eight different counties of Ireland, thus they are likely to represent a heterogeneous mix of perspectives.

Six of the seven focus groups were facilitated by the same researcher, a male medical doctor who was a similar age to some of the adult survivors (early thirties). This may have introduced medical preconceptions and assumptions in the research process, or it may have introduced power differences based on sex or education. However, a reflexive approach was taken; a female scribe was present in each of these focus groups, and debriefing was undertaken between the researcher and scribe after each focus group to foster dialogue, and to identify any divergent understandings of situations or overlooked perspectives. Furthermore, a female research psychologist was heavily involved in the analysis and interpretation of the data, and a reflexive dialogue was used to ensure reliability of the findings.

# 5 Stakeholder consultation

A process of stakeholder consultation was undertaken to complement the findings of the national qualitative study. This was done to ensure alignment between the views of study participants with other stakeholders working in the area of survivorship after childhood cancer. This allowed for a wide spectrum of views to be represented in the needs assessment, within the allotted time and resource constraints, and to identify whether volunteer bias had influenced the study findings.

As part of this consultation exercise, 12 individual interviews were held with three Consultant Paediatric Oncologists/ Haematologists, one Advanced Nurse Practitioner (ANP), two Clinical Nurse Specialists, three voluntary organisation representatives, two survivors, and one university professor with an interest in survivorship. We also sought to include the views of GPs who had experience of caring for survivors of childhood cancer, but none of those contacted agreed to participate.

## 5.1 Key findings

### 5.1.1 Information needs

- Each individual may have different information needs, depending on their own personality, age at diagnosis, age of completing treatment, and family dynamics.
- Some doctors, nurses, dedicated social workers, and others are contacted by survivors or their family members several years after completing treatment, seeking more information about their disease and treatment.
- Many survivors want a summary treatment record (STR), but do not receive this at the time of discharge or transition from paediatric services.

This is not a standardised part of care for all survivors. Some healthcare providers were aware of survivors who had relapsed while living abroad, and had no record of their previous treatment regimens.

- Although many survivors and parents want information relating to long-term effects, it is important not to overwhelm them at the time of completing their treatment. It is best to establish each individual's own information needs, and set up a dedicated appointment to address these.
- There is a need for a more standardised approach to the provision of information relating to fertility. Some fertility preservation options are only effective if done before cancer treatment, and others are only available through the public system prior to treatment.

### 5.1.2 Model of survivorship care

- There is no clear consensus around the optimal model of survivorship care. This needs more dedicated consideration and consultation of a wide range of stakeholders.
- One Consultant described how the Canadian style of survivorship care could work well in the Irish context:
  - Children are followed up by their treating clinician for 2-5 years after treatment, then undergo transition to a dedicated survivorship clinic with a comprehensive discharge letter from the Consultant Haematologist/ Oncologist.
  - This clinic may be led by a General Physician for older age groups, or by a General Paediatrician (with oncology experience) for younger age groups.

- This General Physician or Paediatrician could coordinate follow-up care between all clinicians and allied health professionals involved.
- An alternative suggestion was that ANPs would be more actively involved in coordinating follow-up care for survivors, even in the absence of a dedicated survivorship clinic. This would overcome the common problem whereby survivors have nobody within adult services to oversee their care.
- Not all survivors need intensive follow-up care. Some may be discharged back to GP care if they are at low risk of further complications and if their GP agrees. In such cases GPs would need to receive a detailed discharge letter, a copy of the patient's STR, and clear instructions of what late effects are likely to occur.
- Some GPs are reluctant to take over care of survivors, particularly if there is a need for ongoing contact with hospital-based specialties.

### 5.1.3 Challenges at transition

- Transition to adult services typically happens around age 16, but there is some variation in this. Some survivors remain in paediatric services for longer, particularly if they have special needs. Others transition at an earlier stage; for example, in Galway the transition to adult services occurs at age 14.
- One of the main differences for survivors after transition is that their care is not always coordinated in adult services. They often need to attend the same hospital multiple times on different days for medical imaging, blood tests, outpatient appointments etc. This is time-consuming, inefficient, and financially challenging.
- Some parents find the personal disconnect from paediatric services very difficult, and find it challenging to let go of control over their child's care.

- Some adult oncologists are reluctant to take over the care of survivors of childhood cancer. This is due to the increasing numbers of survivors, the need for lifelong follow-up (in some cases), and a lack of resources.

### 5.1.4 Psycho-social needs

- Some survivors experience residual anger relating to the experiences they may have missed out on while dealing with illness.
- Some survivors deal with a sense of guilt relating to their own survival if they have witnessed their friends or peers dying from the same disease.
- Some survivors turn to substance misuse as a means of relief or escapism from their distress.
- Psycho-oncology and Liaison psychiatry services in the community are inadequate to meet current needs. Many survivors need psycho-oncology support, but only a small minority ever need psychiatric support.
- Community-based organisations are helpful in the absence of other psycho-social support services, but more formal psycho-oncology services are needed, particularly in the public system.
- In many cases, the only way for survivors to access psycho-oncology services is through private funding or through the voluntary sector, due to a lack of resources in the public system.
- Some survivors have no interest in engaging with psycho-oncology support services during treatment, and only experience distress several years later when they can no longer access these services.
- Survivors who engage in peer support programmes appear to do well from a social perspective relative to those who remain disengaged from these services.

### 5.1.5 Educational challenges

- Children who spend prolonged periods of time in hospital may attend the hospital school (for example, in Our Lady's Children's Hospital, Crumlin). However, this may only be appropriate for the educational needs of younger children, and not for teenagers.
- Some schools provide excellent support to affected students/families when they return to formal education, and can provide good access to disability supports if required.
- Some survivors avail of the Disability Access Route to Education (DARE) scheme, or receive supports from the Association for Higher Education Access & Disability (AHEAD).
- Many survivors struggle with re-entry to a new peer group if they have missed large amounts of schooling. Some may be reluctant to return to school due to issues related to body image, or due to academic challenges after a long absence.
- Many schools seek guidance on how to facilitate survivors of childhood cancer. There appears to be no specific training or guidance available from the Department of Education to advise schools how they should accommodate or facilitate children who have been diagnosed and treated for cancer.

### 5.1.6 AYA services

- International experience suggests that AYA achieve better outcomes when they are treated for cancer in dedicated AYA centres.
- AYA centres are well established in other jurisdictions, including the UK and Netherlands. In Ireland, AYA centres may be needed in Dublin, Cork and Galway. Dedicated staffing is required for an AYA centre; medical and nursing teams could divide their time between AYA and adult haematology/oncology services if appropriate.
- Advantages of dedicated AYA services may include the smoother transition of teenagers and young adults to adult services, better engagement in follow-up care among this cohort, and improved health outcomes.

- It is anticipated that the new National Children's Hospital will have dedicated bed capacity to treat AYA, but follow-up services for AYA will need to be developed.

### 5.1.7 Lifestyle and preventive advice

- Some survivors get dedicated lifestyle advice from their healthcare providers at the time of transition to adult services. Although this does not happen in a standardised manner, preventive messages are commonly communicated regarding avoidance of smoking, excess alcohol use, staying physically active, and skin protection.
- Two of the nurses interviewed for this consultation felt that the provision of this lifestyle advice could be improved. They also emphasised the need to discuss the benefits of vaccination with survivors, because many families seek information about vaccines after completing treatment.

### 5.1.8 Survivors of brain and CNS tumours

- This group of survivors have very distinct needs. Some experience personality changes after treatment, and may become disinhibited or display uncharacteristic behaviour. There is a dedicated brain tumour support group for adults in Ireland, but less support is available for children/adolescents.
- There is a need for a dedicated Consultant Neuro-Oncologist in Ireland to deal with the specific needs of these individuals.
  - Many patients/survivors of brain and CNS tumours need specialised neuro-psychological assessment. This should ideally take place at baseline before treatment, and then again after completing treatment so that changes in cognition can be tracked. There is a need for more dedicated neuro-psychologists to provide this service.
  - Many survivors are unable to get life insurance and a mortgage, and this is especially challenging for those who have had brain tumours.

### 5.1.9 Living outside of Dublin

- The experience of cancer in childhood or adolescence can be particularly isolating for those in smaller towns or villages. It is harder for those outside of Dublin to engage in peer support groups such as CanTeen due to geographical distance.
- Social media plays a very important role in facilitating peer support for survivors and families outside Dublin.
- Although access to formal psychological support services is limited for all survivors, it is particularly difficult for those living outside of Dublin.

### 5.1.10 Other needs

- Individuals who have cancer predisposition syndromes have very specific needs, but there are no pre-determined clinical pathways for these groups. Some of the relevant predisposition syndromes include (but are not limited to) Von-Hippel-Lindau Syndrome, Li-Fraumeni Syndrome, Beckwith-Wiedemann Syndrome.
- There is a need for a specialised sarcoma service for patients/survivors.
- There is a need to strengthen data systems around survivorship. Most information held by hospitals on survivors is paper-based. A database of survivors and their medical needs should be established.
- Some parents of survivors have undertaken peer support training courses, and have found these to be a very helpful means of giving volunteers and advocates clear roles and defined boundaries when advising other parents/families during treatment and follow-up.

## 5.2 Summary of findings

It is clear that the views of the stakeholders consulted during this needs assessment were largely aligned with those of the survivors and parents who participated in focus groups. Many survivors have unmet information needs, and may want to learn more about the potential long-term effects of their disease or treatment. There is a need to gauge each individual's desire to receive this information, and to set aside protected time to discuss this. The provision of a STR to each survivor may help to empower survivors, and this should be a standard part of care. There is also a need for a more standardised approach to the provision of fertility-related information, so that survivors can avail of fertility-preserving interventions if required.

Transition from paediatric services is a challenging time for survivors and their families, and the perceived lack of coordination of this care is a source of dissatisfaction for some. Survivors have a range of needs with regard to psycho-social wellbeing, and currently the resources to support and address these needs are inadequate. Survivors who avail of formal psycho-oncology services and/or peer support appear to do better during follow-up. Although there are universal difficulties for survivors in accessing psychological support services, this is particularly difficult for survivors living further from Dublin. Survivors also experience a range of academic and social challenges when returning to education. Schools require additional guidance to facilitate survivors and to advise them on how to access disability support services, if needed.

There is no consensus around the optimal model of survivorship care, and this requires dedicated consideration and consultation going forward. This model may be predominantly medical or predominantly nursing-led, and the role of primary care in this model needs clarification. There is a desire for AYA centres to be established as part of the overall care for survivors of childhood/adolescent cancer, and these may help to overcome some of the challenges around the time of transition or discharge from paediatric services.

A number of groups of survivors have distinct, unmet clinical needs during survivorship. Their distinct needs arise from a combination of unique pathological processes and a lack of specialist expertise in the country. They include those who have experienced brain or CNS tumours, sarcomas, and cancer predisposition syndromes.

# 6 Recommendations

The following recommendations have arisen from this needs assessment.

## 1. Information needs

- The information needs of each individual survivor of childhood cancer should be gauged as a standard part of care. Each survivor's information needs with regard to long-term effects should be addressed in a dedicated appointment or clinic.
- Information relating to long-term treatment effects should be provided to survivors in a personalised and age-appropriate manner. The optimal timing of provision of information relating to long-term effects needs careful consideration, and may need to be tailored for each individual.
- A STR should be prepared for all survivors of childhood cancer by default. This should be offered at the time of finishing treatment, or upon discharge out of paediatric services. Survivors who do not wish to receive their STR at this time should still be able to rapidly access the STR from their treating hospital upon request.
- The content and format of the STR should be standardised, and should be decided upon through collaboration between healthcare professionals, survivors, and others who may use this document.
- A copy of the STR should be provided to the survivor's GP, as well as a copy of the SCP if available.
- Patients with childhood or adolescent cancer, and their parents/guardians, should receive information relating to fertility in a standardised manner, prior to commencing treatment wherever possible.

## 2. Coordination of care

- The coordination of follow-up appointments for survivors of childhood cancer needs to be improved and streamlined, within individual hospitals and across multiple hospital sites.
- Communication mechanisms need to be improved between those providing follow-up care (including clinical teams based in hospitals, GPs, allied healthcare professionals) and survivors and their families. Appropriate technology and digital supports need to be in place to enable improved communication between all parties.
- There needs to be greater clarification of clinical responsibility for survivors of childhood cancer, who often have no clinician overseeing their follow-up care after discharge from paediatric services. This includes their routine follow-up, surveillance for recurrence, and monitoring for late effects.
- There is a need for a dedicated single point of contact (SPOC) who can oversee the coordination of follow-up appointments for survivors of childhood cancer. This person should have an understanding of medical information, know how to navigate the healthcare system, and should be able to advocate on behalf of survivors to facilitate access to follow-up services. There is need for further consideration of the role of the SPOC, and whether this should be a medical, nursing, or other allied health professional.

- The benefits and disadvantages of establishing a dedicated survivorship clinic for survivors of childhood cancer should be formally explored. If established, this may provide improved coordination of follow-up care for survivors, and could bring together all members of the MDT in a single place with improved communication.

### 3. Psycho-social needs

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- Psychological support should be offered to all survivors of childhood cancer by default. The initial offer of support should be made at the outset of treatment, and with further offers of support and/or signposting to appropriate services at the end of treatment and during follow-up appointments.
- Psychological support should be offered to parents whose children have experienced cancer in childhood. The initial offer of support should be made at the outset of treatment, and with further offers of support and/or signposting to appropriate services for themselves and any affected siblings at the end of treatment.
- Public psycho-oncology support services are unable to meet current needs. Additional staffing and resourcing for psycho-oncology services are required, both within the hospital system and in the community.
- Patients who have brain/CNS tumours should have a dedicated neuro-psychological assessment at the time of diagnosis, and again at the time of finishing treatment. There is a need for greater public neuro-psychology services to meet current needs.
- Peer support activities should continue to be facilitated and enhanced for survivors of childhood cancer to encourage greater levels of engagement, particularly for those living outside of Dublin who may have difficulties identifying a peer network.

### 4. Educational needs

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- Access to educational supports needs to be streamlined for survivors of childhood cancer, with clarification of eligibility criteria for supports. This includes criteria for access to SNAs, and disability supports during school and upon entry to university.
- There is a need for a nominated person, in the Department of Education or other government agency, to act as a contact point for schools and parents who require advice regarding provision of educational and disability supports.

### 5. Financial and social protections

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- Families of children and adolescents with cancer should be protected from high levels of financial expenditure during treatment and follow-up. Supports need to be put in place for affected families to provide protection against excessive out-of-pocket payments.
- Practical supports should be taken to ease the financial burden among survivors of childhood cancer. These could include an extension of medical card eligibility for survivors of childhood cancer beyond the current five year threshold, extension of the current long-term illness scheme to cover more childhood cancers, and availability of free, accessible dental care for survivors.
- There is a need for improved employment access programmes and workplace supports for survivors of childhood cancer and parents trying to regain entry to the workplace.

### 6. Data and intelligence needs

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- There is a need for a single database of survivors of childhood cancer in Ireland. This will allow more accurate enumeration of survivors, estimation of incident cases of childhood cancer in the future (and corresponding survival rates), and will enhance health system planning.

- There is a need for a unified information system to enable exchange of information between hospital sites. This is particularly important for radiology, where scans are often duplicated, or difficult to share.
- An electronic health record should be developed for patients with childhood cancer, to enable rapid exchange and retrieval of health-related information, and improved coordination of care.

## **7. Other needs**

- Access to follow-up services needs to be improved for survivors of childhood cancer. At present, some clinical services have unacceptably long waiting lists, and access is inequitable, depending on the presence of private health insurance. There is a need for dedicated resources to ensure more timely access to services for all survivors.
- The establishment of an AYA treatment unit with its own dedicated staff would enable smoother transition from paediatric to adult services, and may facilitate greater engagement in follow-up services.
- Care pathways for survivors of childhood cancer should be streamlined to avoid gaps in follow-up and to minimise avoidable complications. This is particularly important after transition to adult services.
- There is a need for specialist expertise in neuro-oncology and in treatment of sarcoma in Ireland.
- There is a need for further dedicated consultation on the optimal model of survivorship care in Ireland. This requires input from all relevant stakeholders, including survivors, parents, hospital-based clinical teams, GPs, allied health professionals, voluntary organisations, community partners, and health service management.

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