

Childhood cancer survivorship: barriers and preferences

Christina Signorelli ^{1,2}, Claire Wakefield,^{1,2} Jordana K McLoone,^{1,2} Joanna Fardell,^{1,2} Janelle M Jones,^{3,4} Kate H Turpin,⁵ Jon Emery ⁶, Gisela Michel,⁷ Peter Downie,⁸ Jane E Skeen,⁹ Richard Cohn,^{1,2} ANZCHOG Survivorship Study Group

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For numbered affiliations see end of article.

Correspondence to

Dr Christina Signorelli, Kids Cancer Centre, Sydney Children's Hospital Randwick, Randwick, NSW 2031, Australia; c.signorelli@unsw.edu.au

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ABSTRACT

Objective Many survivors are disengaged from follow-up, mandating alternative models of survivorship-focused care for late effects surveillance. We explored survivors' barriers to accessing, and preferences for survivorship care.

Methods We invited Australian and New Zealand survivors of childhood cancer from three age groups: <16 years (represented by parents), 16–25 years (adolescent and young adults (AYAs)) and >25 years ('older survivors'). Participants completed questionnaires and optional interviews.

Results 633 survivors/parents completed questionnaires: 187 parents of young survivors (mean age: 12.4 years), 251 AYAs (mean age: 20.6 years) and 195 older survivors (mean age: 32.5 years). Quantitative data were complemented by 151 in-depth interviews. Most participants, across all age groups, preferred specialised follow-up (ie, involving oncologists, nurses or a multidisciplinary team; 86%–97%). Many (36%–58%) were unwilling to receive community-based follow-up. More parents (75%) than AYAs (58%) and older survivors (30%) were engaged in specialised follow-up. While follow-up engagement was significantly lower in older survivors, survivors' prevalence of late effects increased. Of those attending a follow-up clinic, 34%–56% were satisfied with their care, compared with 14%–15% of those not receiving cancer-focused care ($p<0.001$). Commonly reported barriers included lack of awareness about follow-up availability (67%), followed by logistical (65%), care-related beliefs (59%) and financial reasons (57%). Older survivors ($p<0.001$), living outside major cities ($p=0.008$), and who were further from diagnosis ($p=0.014$) reported a higher number of barriers.

Conclusions Understanding patient-reported barriers, and tailoring care to survivors' follow-up preferences, may improve engagement with care and ensure that the survivorship needs of this population are met.

What is known about this topic

- Many survivors are disengaged from survivorship care, mandating alternative models of follow-up care to ensure survivors receive appropriate late effects surveillance and management.
- Barriers to accessing follow-up highlight the need for innovative solutions to promote follow-up engagement and reduce the long-term burden of cancer.
- Comprehensive data on childhood cancer survivors' preferences for follow-up to further promote engagement, and potentially changing preferences across the survivorship trajectory, remains unknown.

What this study adds

- Our findings offer insight into the optimal delivery of survivorship care from survivors'/parents' perspectives.
- Given many high-risk survivors are disengaged from cancer-related care, identifying patient preferences can be used to re-engage survivors who should be receiving cancer-related follow-up.

INTRODUCTION

The long-term impact of childhood cancer is substantial. Survival rates have improved, however reports of late treatment-related morbidities affecting childhood cancer survivors have also proliferated. Up to 88% of survivors experience 'late effects', often decades after completing treatment for childhood cancer.¹ Late effects include second neoplasms, potentially life-threatening organ dysfunction, delayed growth/development, neurocognitive impairment and psychosocial difficulties.¹ Survivors can have poor knowledge about their diagnosis and treatment, and often underestimate

their risk of late effects.² Ongoing long-term follow-up care for survivors is therefore strongly recommended to monitor for, and where possible prevent, secondary complications by educating survivors about a healthy lifestyle after treatment for cancer.

It is concerning that as few as 23% of survivors attend cancer-related follow-up care.^{3 4} Barriers to accessing follow-up care may relate to patient-related (eg, low perceived risk of late effects), provider-related (eg, poor physician knowledge about childhood cancer) and system-related factors (eg, limited health insurance).⁵ Few facilities offer follow-up beyond the paediatric age group, adding to the complexity of meeting survivors' unique follow-up needs.⁶ Survivors usually transition from paediatric to adult care as adolescents, or young adults, at which point many become disengaged.⁷ With time, survivors are more likely to become disengaged from follow-up care, despite the fact that late effects increasingly emerge as they age.⁸

Research suggests that survivors who attend long-term follow-up care have better long-term physical and psychosocial health outcomes than non-attendees.⁹ Poor follow-up engagement rates necessitate the development and implementation of improved models of survivorship care. Various models are recommended in the literature, including oncologist-led, nurse-led or primary care physician-led (PCP) or a shared-care model.¹⁰ The success of any model of survivorship care critically depends on establishing stakeholders' preferences.¹¹ Few data are available about the preferences of childhood cancer survivors and parents, a crucial gap in informing the development of new models of survivorship care.

Our study had two primary aims. The first was to characterise the nature and prevalence of patient-reported barriers to accessing follow-up care, and assess any clinical/demographic factors associated with survivor-reported barriers. The second was to explore survivors' and parents' preferences for the delivery of survivorship care, including their preferred health professional to lead their care, mode of delivery, location, timing and reminder methods and which professionals they would like to access for follow-up in a multidisciplinary team. We focused on these aspects because they are modifiable in considering future models of care, and research suggests that organisational aspects (eg, appointment times) are a key priority for survivors, significantly influencing their satisfaction with care.¹²

To gain a more balanced and comprehensive understanding of consumers' preferences for care, our study uniquely involved parents of young survivors, and young and older survivors, in both early and late survivorship. We purposely recruited survivors who were regular attendees at follow-up, and survivors who were not engaged in any cancer-related follow-up.

METHODS

We obtained written consent from all participants. We implemented a sequential explanatory mixed-methods study design, collecting data in two stages. We invited parents/survivors from 11 hospitals in Australia/New Zealand to complete a questionnaire as part of the Australian and New Zealand Children's Haematology/Oncology Group Survivorship Study.⁶ Questionnaire respondents could opt-in for an optional interview.

Participants

In order to capture potentially differing preferences across age groups, we recruited: (a) parents of young survivors aged <16 years ('parents'), (b) adolescent and young adult (AYA) survivors of childhood cancer aged 16–25 years ('AYA survivors') and (c) survivors of childhood cancer aged over 25 years ('older survivors'). Survivors were eligible if they were diagnosed with cancer before the age of 16, were at least 5 years from diagnosis, were in remission and were treated at an Australian/New Zealand hospital.

Data collection

We searched for eligible survivors in participating hospitals' electronic medical records. We posted invitations, questionnaires and reply-paid envelopes and conducted telephone follow-up 4 weeks after mail-out. For survivors aged <16 years, invitations were addressed to parents. We posted a second information package to questionnaire respondents who opted in for interviews, containing a consent form and reply-paid envelope. We telephoned interested survivors/parents to arrange an interview time. Experienced researchers who had no prior relationship with the participants (CS, JKMcL, JF) conducted the one-on-one interviews. We did not offer interviewees the opportunity to review their transcripts.

Measures

We collected participants' demographic and clinical information (table 1), as well as the number of late effects survivors reported experiencing since treatment (see online supplementary files 1 and 2 for details). We measured barriers to follow-up engagement and participants' satisfaction with care. Participants indicated their preferred health professional (eg, oncologist, PCP), location (eg, non-hospital facility), delivery mode (eg, phone, online), appointment reminders (eg, text), timing and access to services.

Statistical analysis

We analysed quantitative data using SPSS V.24.0, considering results statistically significant when $p < 0.05$ (two-tailed). To describe sample characteristics and compare barriers and preferences for follow-up across groups, we used descriptive statistics, χ^2 tests and analysis of variance. We used multivariate linear regressions to analyse demographic and clinical

Table 1 Participants' clinical and demographic characteristics

Characteristic	Parents of survivors* <16 years (n=187)	AYA survivors 16–25 years (n=251)	Older survivors >25 years (n=195)
Male survivors	101 (54%)	119 (48%)	85 (44%)
Fathers of survivors	25 (14%)	NA	NA
Self-identified ethnicity			
Australian/New Zealand	133 (74%)	197 (81%)	143 (77%)
Not Australian/New Zealand	48 (26%)	45 (18%)	42 (23%)
Area of residence†			
Major city	127 (76%)	166 (74%)	144 (81%)
Regional/remote	40 (24%)	57 (26%)	33 (19%)
Education			
High school only	49 (26%)	147 (60%)	51 (27%)
Postschool (eg, university)	137 (74%)	97 (40%)	139 (73%)
Employed	134 (72%)	152 (62%)	137 (73%)
Income			
<\$A60 000	41 (24%)	145 (60%)	74 (46%)
>\$A60 000	128 (76%)	62 (49%)	85 (54%)
Diagnosis			
Leukaemia	86 (46%)	105 (46%)	79 (41%)
Lymphoma	13 (7%)	24 (10%)	38 (20%)
Brain cancer	9 (5%)	30 (13%)	24 (13%)
Other	59 (31%)	71 (31%)	50 (26%)
Treatment(s) received			
Surgery	77 (44%)	118 (50%)	78 (46%)
Chemotherapy	176 (95%)	231 (94%)	168 (89%)
Radiation	47 (28%)	91 (40%)	110 (61%)
Bone marrow transplant	34 (20%)	46 (20%)	30 (18%)
Engaged in hospital-based follow-up	146 (78%)	143 (58%)	58 (30%)
Regular primary care physician	122 (65%)	169 (68%)	121 (64%)
Satisfied with care	107 (58%)	190 (78%)	107 (58%)
	Mean (SD)	Mean (SD)	Mean (SD)
Survivor age	12.4 (2.2)	20.6 (2.6)	32.5 (6.2)
Range	7–15	16–23	26–61
Years since diagnosis	9.7 (2.1)	14.3 (4.4)	24.6 (8.0)
Range	5–15	5–24	5–59
Years since treatment completion	8.1 (2.6)	12.3 (4.4)	22.3 (7.8)
Range	0–15	0–23	5–59
Total number of late effects	3.2 (3.1)	3.5 (3.4)	4.1 (3.8)
Range	0–15	0–17	0–19
Total number of barriers to follow-up	4.2 (4.0)	6.3 (4.4)	6.5 (4.7)
Range	0–19	0–18	0–19

*Parents' demographic data are reported, while child survivors' clinical data are reported (parent proxy).

†According to Area of Remoteness Index Australia classifications. We manually categorised New Zealand postcodes according to the Statistics New Zealand Urban/Rural Profile Classifications. Numbers and percentages may not add up due to missing values and rounding errors.

AYA, adolescent and young adult; N, number of participants; NA, not assessed or not applicable.

factors associated with the total number of barriers to accessing follow-up care. We included variables in the multivariate regression that were statistically significant at $p < 0.2$ in univariate models.

We audio-recorded and transcribed interviews verbatim. We conducted directed qualitative content analysis using NVivo11. Two researchers (CS,

MEB) read transcripts alongside data collection and discussed emergent categories to develop a coding tree, based on predetermined themes, informed by the study aims and guided by the Miles and Huberman methodology.¹³ Two coders (CS, MEB) coded 20% of randomly selected interviews. Discrepancies were resolved through discussion and amendments made to

the coding tree as needed. Given that concordance was high (96.8%) and the scale of the study, one coder (CS) analysed the remainder of the data. We used qualitative data to further explain the quantitative findings, stopping data collection once we achieved data saturation.

RESULTS

Of 900 eligible and contactable parents/survivors who we invited, 633 completed questionnaires (71% response rate): 187 parents of young survivors (<16 years; survivors' mean age: 12.4 at study participation; 9.7 years on average from diagnosis), 251 AYAs (16–25 years; mean age: 20.6; 14.3 years from diagnosis) and 195 older survivors (>25 years; mean age: 32.5; 24.6 years from diagnosis). Table 1 details participant characteristics.

Questionnaire respondents were more likely to be female (51.7% vs 43.9%, $p=0.009$); and older (mean age: 21.5 vs 20.1, $t(1150)=-2.827$, $p=0.005$) than non-respondents. Of the questionnaire respondents, 151 also completed optional telephone interviews. There was no significant difference between interviewee respondents and non-respondents in sex ($p=0.645$), however interview respondents were

younger than non-respondents (mean age: 19.7 vs 22.0, $t(624)=2.870$, $p=0.004$).

More parents (75%) and AYAs (58%) than older survivors (30%) were engaged in follow-up care ($p<0.001$). Sixty-five per cent of parents, 68% of AYAs and 64% of older survivors reported having a regular PCP. Figure 1 demonstrates that older survivors' follow-up attendance was lower, while the mean prevalence of late effects was higher. Participants in all age groups who were not currently receiving specialised follow-up (ie, hospital-based, oncologist-led or multi-disciplinary team follow-up) reported significantly greater dissatisfaction with their care (34%–56%) than those who were engaged (14%–15%, $p<0.001$; figure 2). Online supplementary file 3 contains illustrative quotations further explaining key barriers and preferences for care.

Barriers to care

The most commonly endorsed barriers related to lack of awareness of follow-up availability (67%), followed by logistical (65%), belief-related (59%) and financial (57%) reasons. On average, parents endorsed fewer barriers to accessing care (mean: 4.2)

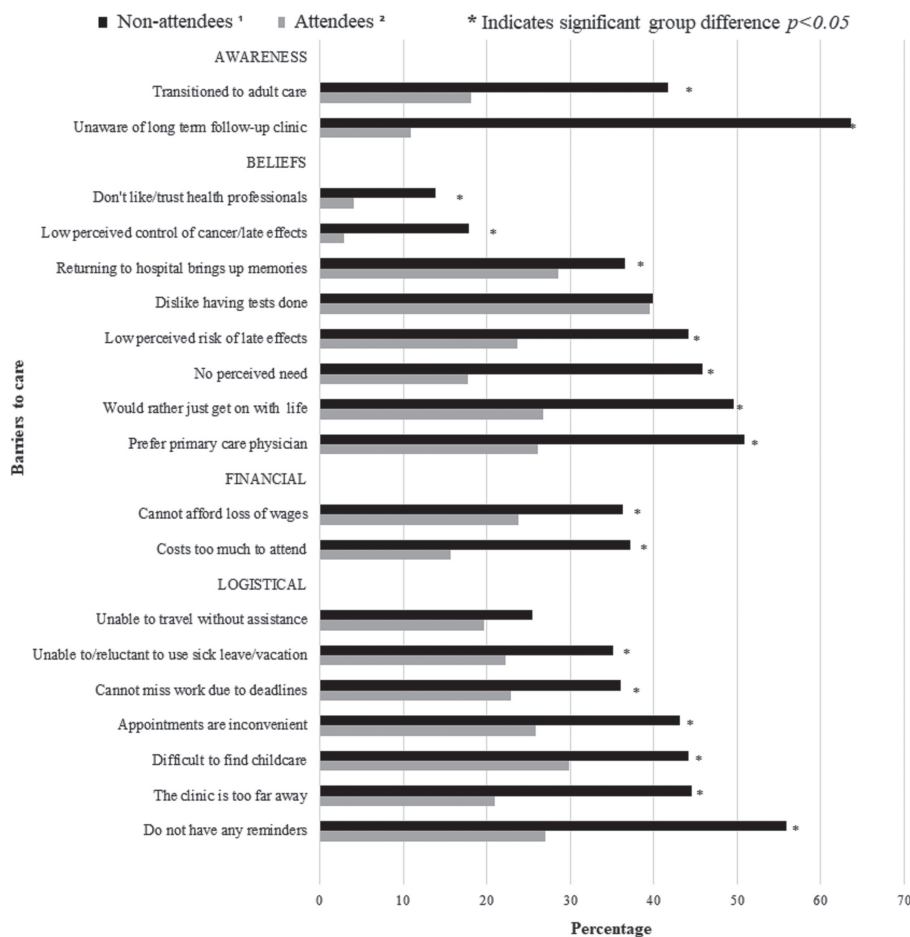


Figure 1 (A) Proportion of survivors engaged in cancer-related follow-up and (B) survivors' mean number of reported late effects by age group.

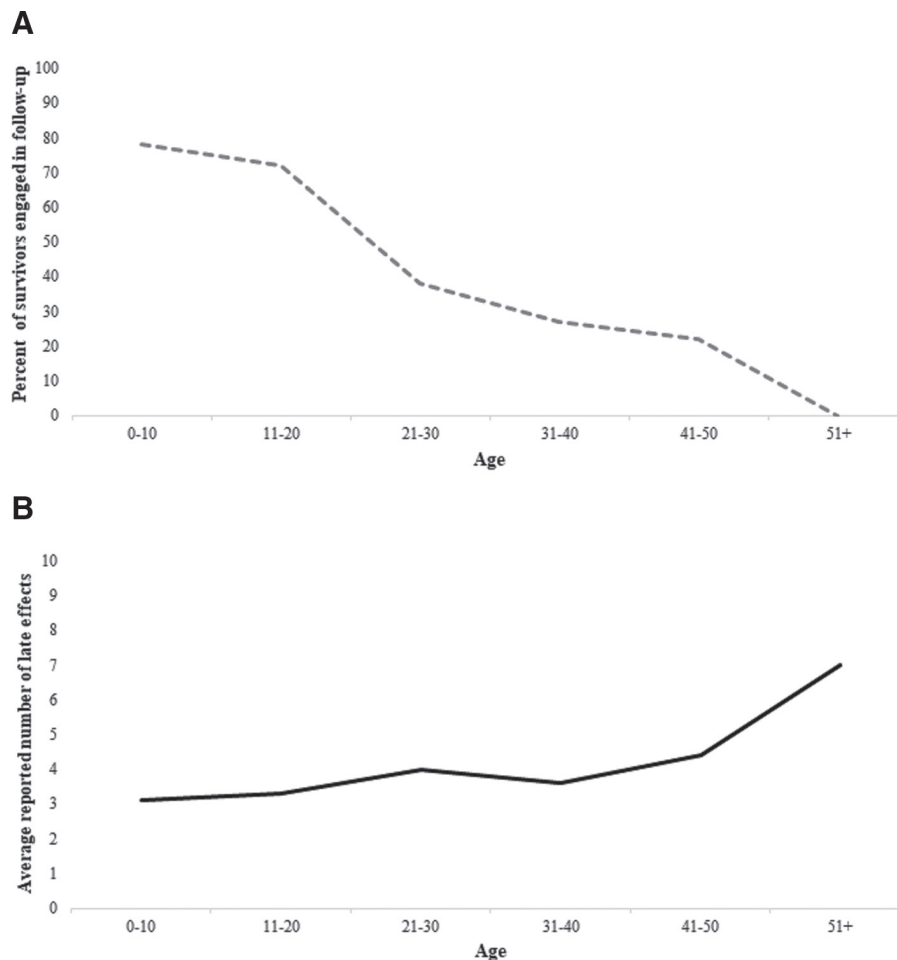


Figure 2 Proportion of participants engaged in hospital-based follow-up and dissatisfied with care. AYA, adolescent and young adult.

than AYAs (mean: 6.3) or older survivors (mean: 6.5; $F(2,625)=16.103$, $p<0.001$). We found no difference in the average number of barriers endorsed by parents ($p=0.410$) or AYAs ($p=0.333$) living in rural/remote areas compared with metropolitan areas. However, older survivors in rural/remote areas endorsed more barriers than survivors in metropolitan areas (mean: 5.2 vs 4.6; $t(172)=-2.781$, $p=0.006$).

Survivors who were disengaged from cancer-related care endorsed all barriers more frequently than those who attended follow-up (all $p<0.05$), with the exception of an 'inability to travel without assistance' and 'aversion to tests' (figure 3). Survivors who reported being dissatisfied with their follow-up endorsed significantly more barriers in total (mean=6.5) than those who were satisfied with their care (mean=5.3; $t(608)=3.152$, $p=0.002$).

Online supplementary file 4 details the univariate and multivariate regression results, assessing demographic and clinical factors associated with participants' total number of follow-up barriers. In multivariate regression analysis, survivors tended to report a greater number of barriers if they were older ($\beta=2.10$, 95% CI=1.24 to 2.95, $p<0.001$), lived outside a major city

($\beta=1.22$, 95% CI=0.32 to 2.12, $p=0.008$) and were further from their primary diagnosis ($\beta=0.06$, 95% CI=0.01 to 0.11, $p=0.014$). No other demographic (eg, sex, ethnicity) or clinical factors (eg, diagnosis, treatment) were associated with the number of barriers reported.

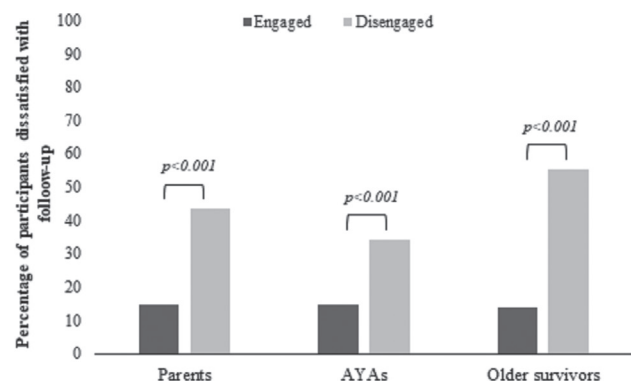


Figure 3 Percentage of participants who endorsed each barrier to accessing follow-up care. *Indicates significant group difference, $p<0.05$. ¹Attendees: engaged in cancer-related follow-up care; ²Non-attendees: not receiving any cancer-related follow-up care.

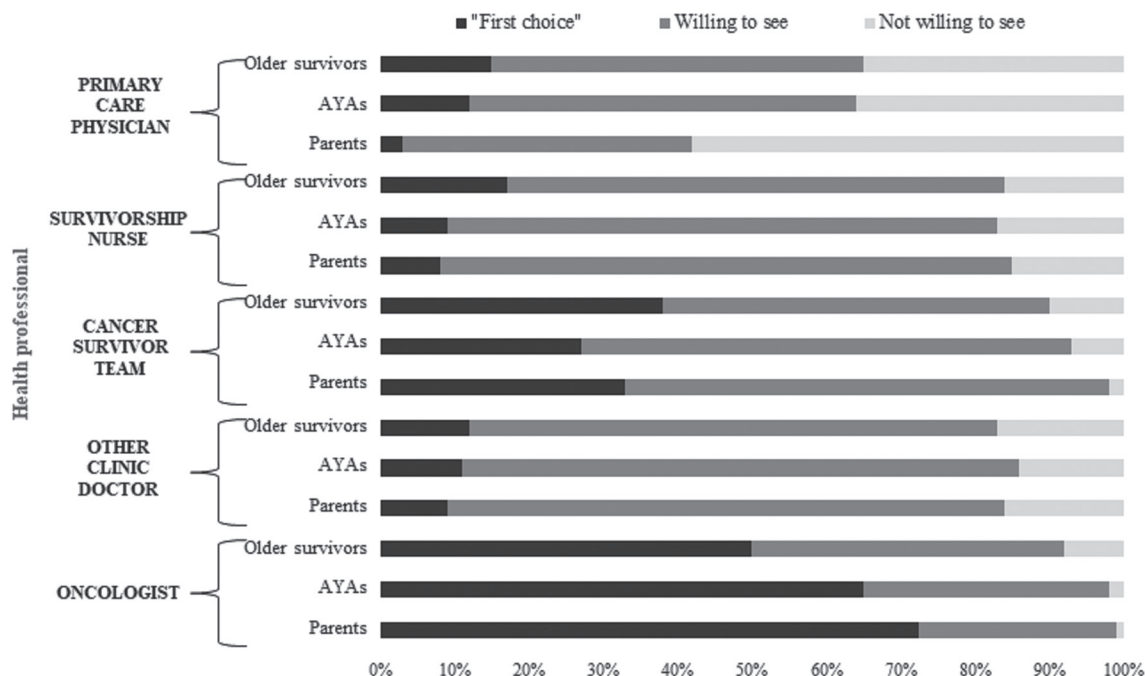


Figure 4 Participants' 'first choice' and willingness to see physicians for follow-up. AYA, adolescent and young adult.

Survivorship care preferences

Preferred follow-up professionals

Hospital-based survivorship care—involving an oncologist or other clinic doctor, cancer survivorship nurse or team—was most commonly endorsed as the 'first choice' across all age groups (97% parents, 88% AYAs, 86% older survivors; figure 4). Primary-based follow-up was least preferred, with many stating that they were unwilling to visit a PCP for cancer-related follow-up (parents: 58%, AYAs: 36%, older survivors: 35%). A reliance on oncologists, and reluctance to visit PCPs, was supported by interview data suggesting that participants had lower confidence in PCPs who were less involved during cancer treatment: "I would probably be more confident in seeing (my oncologist), because she was there...from when I was diagnosed to when I finished" (female neuroblastoma survivor, aged 25 years).

Besides oncologists, the healthcare professional most desired to be available in a follow-up clinic differed by age (online supplementary file 5). Among parents and AYAs, the two most requested professionals were psychologists (64% and 55%, respectively) and fertility specialists (60% and 54%). Parents' third request was for access to dentists (58%), while AYAs' third most requested professional was PCPs (53%). Among older survivors, fertility specialists were most requested (62%), followed by PCPs (56%) and psychologists (54%).

Preferred follow-up mode

The preferred method of delivery for follow-up consultations was face-to-face (81% parents, 75% AYAs, 64% older survivors), followed by telephone

follow-up (13%, 18% and 25%, respectively) or questionnaire assessment with a face-to-face appointment only if needed (6%, 7% and 11%, respectively). As one parent summarised "sometimes face-to-face is better. But at the same time I certainly think that when it's appropriate using (online) tools is a really good idea from an efficiency point of view" (father of hepatoblastoma survivor, aged 14 years).

Preferred follow-up location

Eighty-eight per cent of parents preferred follow-up to be delivered in a paediatric outpatient hospital setting, but fewer AYAs (54%) and older survivors (29%) identified this as their 'first choice'. An adult outpatient hospital setting was the most commonly endorsed location for older survivors (42%), who reported being uncomfortable in a paediatric setting: "I am now 30 years old, so attending clinics with this age group is uncomfortable. I would like to meet or attend with people my age" (female acute lymphoblastic leukaemia (ALL) survivor, aged 30 years). The least endorsed option among all groups was a stand-alone clinic at a university or other non-hospital facility (3%–15%).

Most participants reported feeling comfortable receiving follow-up care in a clinic with patients still receiving active treatment (68% parents, 74% AYAs, 66% older survivors). For the remainder of participants, clinics run in an acute treatment setting was a disincentive to attend. Qualitatively, participants receiving care in any hospital setting meant facing "the memories that my childhood treatment brings back" (male ALL survivor, aged 44 years).

Preferred follow-up timing

Most parents (66%) were willing to attend follow-up with their child during working hours. However, 49% of AYAs and older survivors requested follow-up clinic be held after-hours. Some families suggested '*making times more flexible*' (mother of neuroblastoma survivor, aged 9 years) would improve attendance to allow travel at more convenient times, for example avoiding school drop-off/pick-up times.

Preferred approach to reminders

Participants in all groups preferred reminders for follow-up appointments directly from the clinic, specifically letters (51%–60% of participants), emails (50%–60%) or text messages (34%–50%). Few indicated that their partners (2%–3%) or their parents/child (4%–16%) were their preferred source of reminders. When left to remember clinic appointments themselves this made some participants feel "*we are less important and a bit of a nuisance*" (mother of ALL survivor, aged 8 years). Participants also suggested calendar reminders through an online application/program that contained the patients' treatment plan/summary.

DISCUSSION

This is the first Australian/New Zealand study evaluating survivors'/parents' barriers to attending, and preferences for, survivorship care. Attendance in our young survivors was 75%, with lower attendance in AYAs (58%) and older survivors (30%). We found that older survivors, living outside major cities and survivors who were a longer time from diagnosis, were more likely to endorse a higher average number of barriers to engaging in follow-up. Our findings suggest that the survivorship care preferences of parents, AYAs and older survivors are mostly similar. Most participants indicated a preference for face-to-face hospital-based care. The most requested additional health professionals at survivorship clinics were psychologists and fertility specialists.

Follow-up engagement in our study was suboptimal (30%–75%), mirrored in long-term survivors globally.^{14–16} Commonly endorsed barriers related primarily to low follow-up awareness, low perceived risk and logistical factors, reinforcing research suggesting that non-financial barriers significantly hinder follow-up adherence.¹⁷ Low follow-up clinic awareness may indicate a group of survivors who have missed the educational opportunities on survivorship including navigating the healthcare system. Around two-thirds of survivors endorsed logistical barriers in our study, many of which are modifiable through simple intervention (eg, appointment reminders). Survivors with higher perceived barriers are less likely to attend follow-up, highlighting the need to address practical barriers.¹⁸ Survivors in our cohort who lived outside major cities reported a greater number of barriers.

Research shows that a greater distance travelled to follow-up is associated with diminished attendance rates,¹⁴ echoed in our qualitative findings. Distance-related barriers may be perpetuated by Australia/New Zealand's highly dispersed population, centralisation of clinics and clinic's broad catchment areas.¹⁹

Participants preferred hospital-based to community-based follow-up as their first choice, possibly reflecting early detachment from PCPs during treatment and lack of exposure to alternative models.^{20 21} However, a purely hospital-based model is not sustainable as the number of survivors increases.^{6 22} Risk-stratification may address the pitfalls of this model, and limited global resources, by triaging high-risk/complex survivors to multidisciplinary specialist-led care and lower risk survivors to shared or nurse/PCP-led care.^{23 24} However, the implementation of risk-based models requires that survivors and health professionals are both willing and confident participants. While PCPs seem willing to deliver childhood survivorship care, up to 58% of survivors in our study were unwilling to receive community-based follow-up, potentially reflecting survivors' emotional connection to their oncology team.^{21 25 26} Additional PCP education, support, communication²⁷ and a 'culture change' to establish expectations from diagnosis may be required to ensure the success of community-based models.²⁶

Our data echo existing literature showing that many survivors who are at high risk of developing late effects (eg, second malignancies, cardiovascular disease) are disengaged from cancer-related care.^{22 23} Our study highlights the alarming pattern of declining engagement in follow-up care over time, despite survivors' escalating risk of developing late effects, resulting in a significant gap in care. Additional research is therefore needed to evaluate interventions that promote follow-up adherence,²⁸ particularly in age cohorts where this disparity is greatest. To achieve this, it is critical to address patient-reported barriers and preferences to ensure older survivors remain engaged in care, and to mitigate poor patient outcomes and the associated healthcare system costs. A significant minority of survivors in our study (up to 25%) were open to non-traditional follow-up (eg, telephone/questionnaire). Innovative solutions, including postal health questionnaires,^{29 30} internet-based^{31 32} and telephone/video-conferencing interventions,³³ may help to promote follow-up engagement by overcoming reported barriers.

Future models should address the complex and varying needs of this population, indicated by the diversity of health professionals to whom our participants requested access. High demand for psychologists highlights the long-term challenges necessitating psychosocial support, emphasised in the release of recent guidelines.³⁴ Traditional risk-based models may not fully account for psychological late effects, for example, referring medically 'low-risk' survivors

to primary care where psycho-oncology care is not routine. Psychological support may also help to overcome barriers transitioning from paediatric to adult follow-up care.³⁵

Differing preferences across age groups may be explained by highly varying follow-up practices across Australia/New Zealand, for example, discharge age from paediatric follow-up care.⁶ Variations in the type and quality of care survivors have previously received,³⁶ or their prior/current engagement patterns,³⁷ could shape their preferences and openness to alternative models. This adds to the complexity of conceptualising future models of care that address key barriers and capture patient preferences.

This multicentre study includes quantitative and qualitative data of survivors' preferences. Our sample captured follow-up attendees and non-attendees, many of whom were several decades from treatment. As is common in this field,³⁸ most participating parents were mothers, leaving fathers' opinions under-represented. Our questionnaire omitted options such as AYA specialised clinics, reflecting local practices at the time of the study. Results nonetheless indicated that many AYAs were dissatisfied with their follow-up, echoing the need for age-appropriate models.³⁹ Our study included a wide age range of survivors. Grouping older survivors as >25 years may have masked unique preferences in much older survivors (eg, over 40s). Longitudinal studies are needed to better capture the nature of changes in barriers and preferences over time.

CONCLUSION

This study evaluated patient-reported barriers to accessing follow-up care, and survivors' and parents' preferences for the delivery of survivorship care. It is critical to establish practices which align with patient preferences to encourage engagement in care and better ensure patients' survivorship needs are met. Partnering with survivors and families will ensure we meet the survivors' complex needs through coordinated and well-designed care. Innovative solutions may be required to overcome patient-reported barriers and align with families' preferences, while meeting the needs of resource-limited healthcare environments.

Author affiliations

¹Kids Cancer Centre, Sydney Children's Hospital Randwick, Randwick, New South Wales, Australia

²School of Women's and Children's Health, University of New South Wales, Sydney, NSW, Australia

³Hudson Institute of Medical Research, Melbourne, Victoria, Australia

⁴Australian and New Zealand Children's Haematology/Oncology Group, Melbourne, Victoria, Australia

⁵Paediatric Haematology/Oncology, Women's and Children's Hospital Adelaide, North Adelaide, South Australia, Australia

⁶Centre for Cancer Research, University of Melbourne, Victorian Comprehensive Cancer Centre, Melbourne, Victoria, Australia

⁷Department of Health Sciences and Health Policy, University of Lucerne, Lucerne, Switzerland

⁸Children's Cancer Centre, Royal Children's Hospital Melbourne, Parkville, Victoria, Australia

⁹Starship Blood and Cancer Centre, Starship Children's Hospital, Newmarket, Auckland, New Zealand

Twitter Christina Signorelli @chrissigno

Collaborators The ANZCHOG Survivorship Study Group; Dr Frank Alvaro; Professor Richard Cohn; Dr Rob Corbett; Dr Peter Downie; Ms Karen Egan; Ms Sarah Ellis; Professor Jon Emery; Dr Joanna Fardell; Ms Tali Foreman; Dr Melissa Gabriel; Professor Afaf Girgis; Ms Kerrie Graham; Ms Karen Johnston; Dr Janelle Jones; Dr Liane Lockwood; Dr Ann Maguire; Dr Maria McCarthy; Dr Jordana McLoone; Dr Francoise Mechinaud; Ms Sinead Molloy; Ms Lyndal Moore; Dr Michael Osborn; Dr Christina Signorelli; Dr Jane Skeen; Dr Heather Tapp; Ms Tracy Till; Ms Jo Truscott; Ms Kate Turpin; Professor Claire Wakefield; Ms Jane Williamson; Dr Thomas Walwyn and Ms Kathy Yallop.

Contributors CS contributed to the study design and design of the data collection instruments, collected data, carried out the analyses, drafted the manuscript and coordinated revisions. CW and RC conceptualised and designed the study, designed the data collection instruments, coordinated and supervised data collection, reviewed and revised the manuscript. JKMcL conceptualised and designed the study, designed the data collection instruments, contributed to data collection, reviewed and revised the manuscript. JF contributed to data collection, the interpretation of the data, manuscript writing and critically reviewed and revised the manuscript. MJM, KHT, JE, GM, PD and JES contributed to the interpretation of the data, manuscript writing and critically reviewed and revised the manuscript. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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ORCID iDs

Christina Signorelli <http://orcid.org/0000-0002-7091-0347>

Jon Emery <http://orcid.org/0000-0002-5274-6336>

REFERENCES

- 1 Phillips SM, Padgett LS, Leisenring WM, *et al*. Survivors of childhood cancer in the United States: prevalence and burden of morbidity. *Cancer Epidemiology Biomarkers & Prevention* 2015;24:653–63.
- 2 Carretier J, Boyle H, Duval S, *et al*. A review of health behaviors in childhood and adolescent cancer survivors:

- toward prevention of second primary cancer. *J Adolesc Young Adult Oncol* 2016;5:78–90.
- 3 Miller KA, Wojcik KY, Ramirez CN, *et al.* Supporting long-term follow-up of young adult survivors of childhood cancer: correlates of healthcare self-efficacy. *Pediatr Blood Cancer* 2017;64:358–63.
 - 4 Wilson CL, Cohn RJ, Johnson KA, *et al.* Tracing survivors of childhood cancer in Australia. *Pediatr Blood Cancer* 2009;52:510–5.
 - 5 Essig S, Skinner R, von der Weid NX, *et al.* Follow-Up programs for childhood cancer survivors in Europe: a questionnaire survey. *PLoS One* 2012;7:e53201.
 - 6 Signorelli C, Wakefield CE, McLoone JK, *et al.* Models of childhood cancer survivorship care in Australia and New Zealand: strengths and challenges. *Asia Pac J Clin Oncol* 2017;13:407–15.
 - 7 Henderson TO, Friedman DL, Meadows AT. Childhood cancer survivors: transition to adult-focused risk-based care. *Pediatrics* 2010;126:129–36.
 - 8 Rokitka DA, Curtin C, Heffler JE, *et al.* Patterns of loss to follow-up care among childhood cancer survivors. *J Adolesc Young Adult Oncol* 2017;6:67–73.
 - 9 Signorelli C, Wakefield CE, Fardell JE, *et al.* The impact of long-term follow-up care for childhood cancer survivors: a systematic review. *Crit Rev Oncol Hematol* 2017;114:131–8.
 - 10 Oeffinger KC, McCabe MS. Models for delivering survivorship care. *J Clin Oncol* 2006;24:5117–24.
 - 11 Lawrence RA, McLoone JK, Wakefield CE, *et al.* Primary care physicians' perspectives of their role in cancer care: a systematic review. *J Gen Intern Med* 2016;31:1222–36.
 - 12 Absolom K, Greenfield D, Ross R, *et al.* Predictors of clinic satisfaction among adult survivors of childhood cancer. *Eur J Cancer* 2006;42:1421–7.
 - 13 Miles MB, Huberman AM. *Qualitative data analysis: an expanded sourcebook*. Sage, 1994.
 - 14 Barakat LP, Schwartz LA, Szabo MM, *et al.* Factors that contribute to post-treatment follow-up care for survivors of childhood cancer. *J Cancer Surviv* 2012;6:155–62.
 - 15 Casillas J, Oeffinger KC, Hudson MM, *et al.* Identifying predictors of longitudinal decline in the level of medical care received by adult survivors of childhood cancer: a report from the childhood cancer Survivor study. *Health Serv Res* 2015;50:1021–42.
 - 16 Nathan PC, Agha M, Pole JD, *et al.* Predictors of attendance at specialized survivor clinics in a population-based cohort of adult survivors of childhood cancer. *J Cancer Surviv* 2016;10:611–8.
 - 17 Smits-Seemann RR, Kaul S, Zamora ER, *et al.* Barriers to follow-up care among survivors of adolescent and young adult cancer. *J Cancer Surviv* 2017;11:126–32.
 - 18 Lupatsch JE, Wengenroth L, Rueegg CS, *et al.* Follow-Up care of adolescent survivors of childhood cancer: the role of health beliefs. *Pediatr Blood Cancer* 2016;63:318–25.
 - 19 Daniel G, Wakefield CE, Ryan B, *et al.* Accommodation in pediatric oncology: parental experiences, preferences and unmet needs. *Rural Remote Health* 2013;13:2005.
 - 20 Michel G, Gianinazzi ME, Eiser C, *et al.* Preferences for long-term follow-up care in childhood cancer survivors. *Eur J Cancer Care* 2016;25:1024–33.
 - 21 Signorelli C, Wakefield CE, Fardell JE, *et al.* The role of primary care physicians in childhood cancer survivorship care: Multiperspective interviews. *Oncologist* 2019;24:710–719.
 - 22 Warren JL, Mariotto AB, Meekins A, *et al.* Current and future utilization of services from medical oncologists. *J Clin Oncol* 2008;26:3242–7.
 - 23 Wallace WH, Blacklay A, Eiser C, *et al.* Developing strategies for long term follow up of survivors of childhood cancer. *BMJ* 2001;323:271–4.
 - 24 Frobisher C, Glaser A, Levitt GA, *et al.* Risk stratification of childhood cancer survivors necessary for evidence-based clinical long-term follow-up. *Br J Cancer* 2017;117:1723–31.
 - 25 Gray WN, Schaefer MR, Resmini-Rawlinson A, *et al.* Barriers to transition from pediatric to adult care: a systematic review. *J Pediatr Psychol* 2018;43:488–502.
 - 26 Mayer DK, Alfano CM. Personalized risk-stratified cancer follow-up care: its potential for healthier survivors, Happier clinicians, and lower costs. *J Natl Cancer Inst* 2019;111:442–8.
 - 27 Blaauwbroek R, Tuinier W, Meyboom-de Jong B, *et al.* Shared care by paediatric oncologists and family doctors for long-term follow-up of adult childhood cancer survivors: a pilot study. *Lancet Oncol* 2008;9:232–8.
 - 28 May L, Schwartz DD, Frugé E, *et al.* Predictors of suboptimal follow-up in pediatric cancer survivors. *J Pediatr Hematol Oncol* 2017;39:e143–9.
 - 29 Nathan PC, Ness KK, Mahoney MC, *et al.* Screening and surveillance for second malignant neoplasms in adult survivors of childhood cancer: a report from the childhood cancer Survivor study. *Ann Intern Med* 2010;153:442–51.
 - 30 Zheng DJ, Sint K, Mitchell H-R, *et al.* Patterns and predictors of survivorship clinic attendance in a population-based sample of pediatric and young adult childhood cancer survivors. *J Cancer Surviv* 2016;10:505–13.
 - 31 Mertens A, Williamson R, Gilleland-Marchak J, *et al.* Evaluation of the adoption, acceptance, and satisfaction of SurvivorLink as a communication tool for pediatric cancer survivors. *Journal of Clinical Oncology* 2016;34.
 - 32 Horowitz ME, Fordis M, Krause S, *et al.* Passport for care: implementing the survivorship care plan. *J Oncol Pract* 2009;5:110–2.
 - 33 Signorelli C, Wakefield CE, Johnston KA, *et al.* 'Re-engage' pilot study protocol: a nurse-led eHealth intervention to re-engage, educate and empower childhood cancer survivors. *BMJ Open*. In Press 2018;8:e022269.
 - 34 Wiener L, Kazak AE, Noll RB, *et al.* Standards for the psychosocial care of children with cancer and their families: an introduction to the special issue. *Pediatr Blood Cancer* 2015;62 Suppl 5:S419–24.
 - 35 Gray WN, Monaghan MC, Gilleland Marchak J, *et al.* Psychologists and the transition from pediatrics to adult health care. *J Adolesc Health* 2015;57:468–74.
 - 36 Lisy K, Emery JD, Jefford M. Provider roles in survivorship care: moving beyond surveys of patient preference. *J Clin Oncol* 2017;35.
 - 37 Vetsch J, Rueegg CS, Mader L, *et al.* Parents' preferences for the organisation of long-term follow-up of childhood cancer survivors. *Eur J Cancer Care* 2018;27:e12649.
 - 38 Wakefield CE, Fardell JE, Doolan EL, *et al.* Participation in psychosocial oncology and quality-of-life research: a systematic review. *Lancet Oncol* 2017;18:e153–65.
 - 39 Fardell JE, Patterson P, Wakefield CE, *et al.* A narrative review of models of care for adolescents and young adults with cancer: barriers and recommendations. *J Adolesc Young Adult Oncol*. In Press 2018;7:148–52.