



Evidence-based recommendations for the organization of long-term follow-up care for childhood and adolescent cancer survivors: a report from the PanCareSurFup Guidelines Working Group

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Abstract

Purpose To facilitate the implementation of long-term follow-up (LTFU) care and improve equality of care for childhood, adolescent, and young adult (CAYA) cancer survivors, the PanCareSurFup Guidelines Working Group developed evidence-based recommendations for the organization of LTFU.

Methods We established an international multidisciplinary guideline panel. A systematic review of the literature published from 1999 to 2017 was completed to answer six clinical questions. The guideline panel reviewed the identified studies, developed evidence summaries, appraised the quality of the body of evidence, and formulated recommendations based on the evidence, expert opinions, and the need to maintain flexibility of application across different healthcare systems.

Results We provide strong recommendations based on low level evidence and expert opinions, regarding organization of LTFU care, personnel involved in LTFU care, components of LTFU care and start of LTFU care. We recommend that risk-adapted LTFU care provided under the guidance of a cancer survivorship expert service or cancer centre should be available and accessible for all CAYA cancer survivors throughout their lifespan.

Conclusion Despite the weak levels of evidence, successful and effective implementation of these recommendations should improve LTFU, thereby leading to better access to appropriate healthcare services and an improvement in health outcomes for CAYA cancer survivors.

Gisela Michel and Renée L. Mulder have joint first authorship.
Leontien C. M. Kremer and Gill Levitt have joint last authorship.

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Implications for Cancer Survivors To improve health outcomes and quality of survivorship of current and future survivors, continued age-adapted education of survivors about the cancer, its treatment, risk of late effects, importance of health behaviours, and necessity of LTFU is important along the cancer and survivorship trajectory.

Keywords Evidence-based guideline · Recommendations · Follow-up care · Oncology · Survivor · Child · Adolescent

Introduction

Long-term follow-up (LTFU) care is important for childhood, adolescent, and young adult (CAYA) cancer survivors due to their high risk of late effects [1–5]. Prevention, early detection, and management of late effects are recommended to improve the quality of survivorship. Additionally, follow-up provides an opportunity to offer age-appropriate information about disease, treatment, and a healthy lifestyle, as well as practical advice about insurance, education or work, and psychosocial support [6–8]. With increasing age, survivors need to increase the personal responsibility for the management of their own health [9–11]. This is particularly relevant for survivors who are transitioned to adult care. Several guidelines recommend risk-stratified LTFU for CAYA cancer survivors [12–15]. However, there is a lack of LTFU care for paediatric and especially adult survivors of CAYA cancer in many countries [16]. Also, there is currently no evidence-based guideline on how to organize follow-up care to deliver the recommended content of LTFU effectively (e.g. surveillance and education). In a recent survey, we found that paediatric oncologists/haematologists and late-effect specialists across Europe agreed that guidelines and recommendations for LTFU are needed [17].

To facilitate the development of evidence-based LTFU services and improve quality of care for CAYA cancer survivors, the PanCareSurFup (PCSF) Guidelines Working Group aimed to develop a pan-European guideline for the organization of LTFU.

Methods

We used evidence-based methods to systematically search and review the literature, extract the evidence, and formulate recommendations. We followed the methods reported by the International Guideline Harmonization Group (www.IGHG.org) [18, 19]. The working group comprised 11 experts from paediatric and medical oncology, survivorship care, guideline methodology, psychology, and epidemiology (see author list).

Scope of the guideline

The scope of this guideline is to provide healthcare providers, patients, survivors, and parents/caregivers with recommendations for the provision of LTFU care for CAYA cancer survivors diagnosed with cancer up to 25 years of age and at least 2 years after the completion of treatment.

Systematic literature review

First, we defined four key issues that should be addressed: organization of LTFU care, personnel involved in LTFU care, components of LTFU care, and start of LTFU care. For each key issue, we assessed the concordance and discordance among five existing guidelines for CAYA cancer survivors: Scottish Intercollegiate Guidelines Network [20], Dutch Childhood Oncology Group (DCOG) [13], United Kingdom Children's Cancer and Leukaemia Group (CCLG) [14], North American Children's Oncology Group (COG) [15], and Australian Cancer Survivorship Centre [21](Online Resource 1).

Second, based on the identified discordances, we defined six clinical questions covering the formulated key issues about LTFU care for CAYA cancer survivors: (1) What are the *opinions of survivors and healthcare providers* about LTFU? (specifically, issues such as who should be involved in LTFU? What are the key requirements of LTFU? What are the barriers to LTFU? What are the facilitators of LTFU?) (2) What *theoretical models* of LTFU care exist? (3) What models of survivorship care are *used in practice*? (4) What is the *effectiveness* of survivorship care models in practice? (5) What is the *effectiveness of risk stratification*? (6) What is the *effectiveness of childhood cancer survivorship care plans*?

Search strategy and selection criteria

To update a previous systematic search developed by the DCOG guideline (January 1999–2009) [13], we conducted a systematic literature search in PubMed in 2014 and updated the search in February 2017 (the updated search resulted in more evidence but no change of previous conclusions and recommendations). The search terms used are listed in Table 1. In addition, we searched the reference lists of the existing guidelines and existing reviews and contacted survivorship care experts to determine if any additional evidence was available. We included papers if the majority of participants were CAYA cancer survivors (i.e. $\geq 55\%$ of sample diagnosed before the age of 25 years) and they were published in English. For clinical questions 1–3, we included all types of study designs (cohort studies, reviews, qualitative studies, expert papers). For clinical questions 4–6, we initially aimed to include randomized controlled trials (RCTs) or cohort studies that compared different models of care or with pre- and post-measurements. However, due to a lack of such studies, we also included other types of study designs.

Table 1 Search strategy. The Dutch Childhood Oncology Group (DCOG) has performed a PubMed literature search for papers published between 1999 and 2009 for questions 1, 2, and 3. This search was updated for the current guideline until 8 February 2017.

Search 1	infant OR infan* OR newborn OR newborn* OR new-born* OR baby OR baby* OR babies OR neonat* OR perinat* OR postnat* OR child OR child* OR schoolchild* OR schoolchild OR school child OR school child* OR kid OR kids OR toddler* OR adolescent OR adoles* OR teen* OR boy* OR girl* OR minors OR minors* OR underag* OR under ag* OR juvenil* OR youth* OR kindergar* OR puberty OR puber* OR pubescen* OR prepubescent* OR prepuberty* OR paediatrics OR paediatric* OR paediatric* OR peadiatric* OR schools OR nursery school* OR preschool* OR pre school* OR primary school* OR secondary school* OR elementary school* OR elementary school OR high school* OR highschool* OR school age OR schoolage OR school age* OR schoolage* OR infancy OR schools, nursery OR infant, newborn
Search 2	((leukemia OR leukemi* OR leukaemi* OR (childhood ALL) OR AML OR lymphoma OR lymphom* OR hodgkin OR hodgkin* OR T cell OR B cell OR non-hodgkin OR sarcoma OR sarcom* OR sarcoma, Ewing's OR Ewing* OR osteosarcoma OR osteosarcom* OR wilms tumor OR wilms* OR nephroblastom* OR neuroblastoma OR neuroblastom* OR rhabdomyosarcoma OR rhabdomyosarcom* OR teratoma OR teratom* OR hepatoma OR hepatom* OR hepatoblastoma OR hepatoblastom* OR PNET OR medulloblastoma OR medulloblastom* OR PNET* OR neuroectodermal tumors, primitive OR retinoblastoma OR retinoblastom* OR meningioma OR meningiom* OR glioma OR gliom*) OR (paediatric oncology OR paediatric oncology)) OR (childhood cancer OR childhood tumor OR childhood tumors)) OR (brain tumor* OR brain tumour* OR brain neoplasms OR central nervous system neoplasm OR central nervous system neoplasms OR central nervous system tumor* OR central nervous system tumour* OR brain cancer* OR brain neoplasm* OR intracranial neoplasm*) OR (leukemia lymphocytic acute) OR (leukemia, lymphocytic, acute[mh]) OR cancer OR cancers OR cancer* OR oncology OR oncolog* OR neoplasm OR neoplasms OR neoplasm* OR carcinoma OR carcinom* OR tumor OR tumour OR tumor* OR tumour* OR tumors OR tumours OR malignan* OR malignant OR hematooncological OR hemato oncological OR hemato-oncological OR hematologic neoplasms OR hematolo*
Search 3	Survivor OR survivors OR Long-Term Survivors OR Long Term Survivors OR Long-Term Survivor OR Survivor, Long-Term OR Survivors, Long-Term OR survivo* OR survivi*
Search 4	continuity of patient care OR quality assurance, health care OR aftercare OR delivery of health care OR home care OR models of care OR shared care OR health care OR delivery of care OR follow-up care OR models, organisational OR models, organizational OR referral and consultation OR health services need and demand OR attitude of health personnel OR long-term care OR transition of care OR “transfer of care” OR transitional model OR transition-based model OR nurse[tiab] OR nursing[sh] OR nursing[tiab] OR nurse* OR nurses OR general practitioner OR family practice OR oncologist[All Fields] OR oncologists OR oncologist* OR health personnel OR “health care provider” OR “primary care provider” OR “secondary care provider” OR “care provider” OR physicians, family OR primary care physician OR family physician OR “health care professional”
Search 1 AND 2 AND 3 AND 4	2823 hits

Title/abstract and then full text of each paper were reviewed by two independent members of the working group for inclusion/exclusion in the development of recommendations, and it was decided for which clinical questions the study should be included. This work was shared among all authors. Where disagreement occurred, the paper was discussed between the two reviewers, and if disagreement persisted, the paper was discussed between the remaining members of the working group. We generated evidence summaries using standardized data abstraction forms to answer the clinical questions. We graded the quality of the evidence according to evidence-based methods (Online Resource) [18].

From evidence to recommendations

The working group reached consensus on final recommendations based upon the evidence, expert opinions (via a panel of

international experts and CAYA cancer survivors at a PanCare meeting in Erice in November 2016 [22]), and the need to maintain flexibility of application across different healthcare systems. Decisions were made through group discussion and consensus, and final recommendations had to be supported unanimously. The strength of the recommendations was graded according to published evidence-based methods (Online Resource 2) [18].

Results

The search in PubMed yielded 2823 abstracts, of which 273 were included for full-text search. In total, 84 papers were included in this guideline (Fig. 1). The conclusions of evidence for the six questions are presented in Table 2 and in more detail in Online Resource 3, and the evidence tables for each paper are available in Online Resource 4.

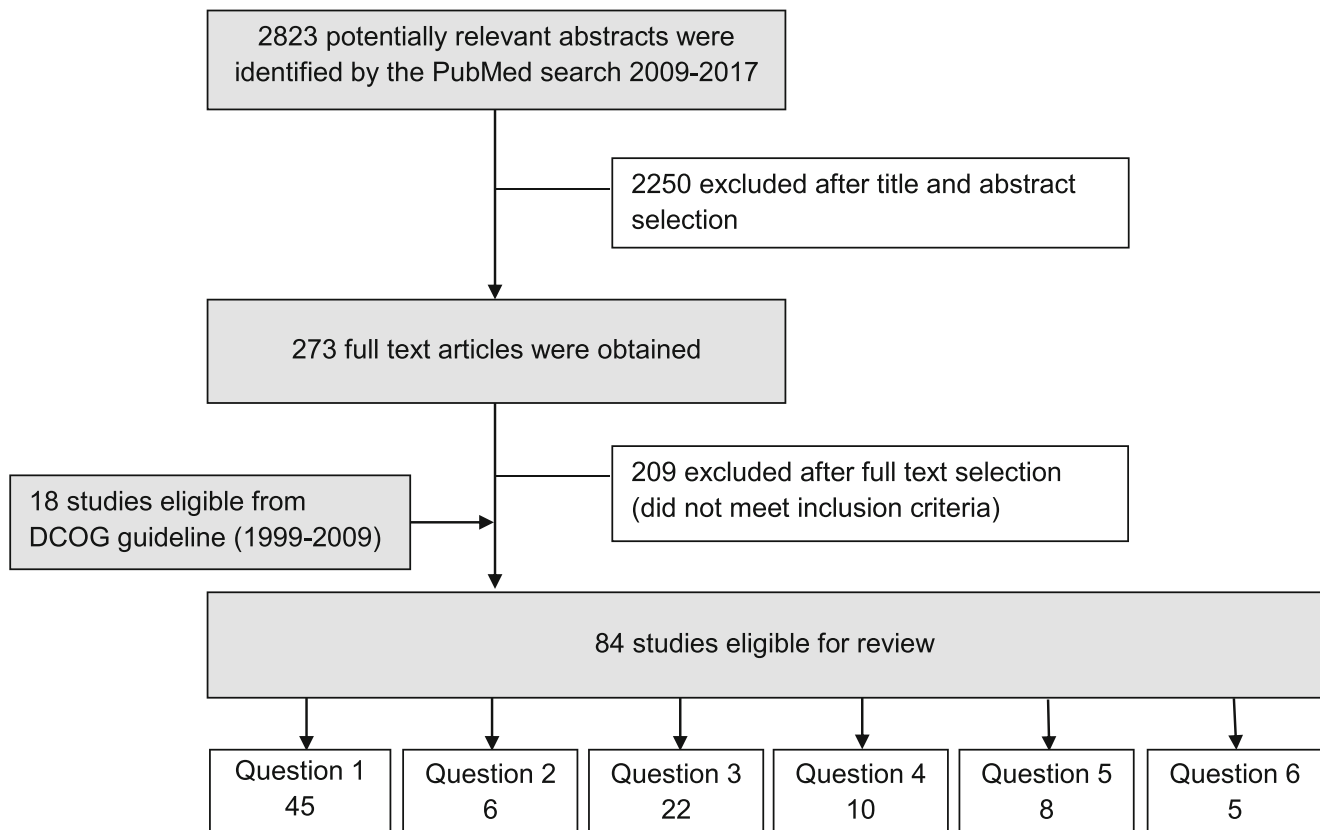


Fig. 1 Flowchart of literature search

The final recommendations are shown in Table 3. All recommendations were based on a combination of low-quality evidence (level C) and expert opinions because no high-level (A/B) evidence was identified. Below we describe the recommendations and their rationale.

Organization of LTFU care

Evidence

Different models of survivorship care are described in the literature, of which the cancer centre-delivered care in the paediatric oncology clinic, medical oncology clinic, or LTFU clinic is the most common model [16, 23–38]. Two other described models are the primary care physician-led LTFU care [27, 37, 39–41, 98] and shared care between treatment hospital and local hospital or primary care [37, 98]. Risk stratification is seen as an important component of LTFU [42–44] with a variety of models described in CAYA cancer survivors [34, 42, 43, 79–81]. Risk stratification is based on cancer treatment, diagnosis, and current health status. There is no evidence that one stratification model is better than the other. Studies have shown that it is possible to both identify [82, 83] and categorize survivors correctly [34, 42, 43, 79–81]. Frobisher et al. showed that survivors classified into a higher risk group according to their cancer diagnosis and

treatment were more likely to be diagnosed with subsequent primary neoplasms, as well as fatal and non-fatal non-neoplastic outcomes than the lower risk groups [83].

Survivors at low risk can be adequately followed up by a primary care physician-led LTFU (community-based care) model [43–46]. Some evidence suggests that primary care physicians are both willing and able to provide LTFU care for adult survivors of childhood cancer [27, 39, 41, 47], but they sometimes may feel uncomfortable [48–50, 56]. Primary care providers reported several barriers to successful LTFU care: lack of communication between primary care physician and paediatric oncologists [41, 67] (which was also mentioned by oncologists [68, 72]), limited access to refer survivors to specialist care (i.e. oncology, mental health, and non-cancer care) [51], and their overall workload [41, 67]. Adequate access to cancer survivor specialists [48, 51], support services such as social work and psychology [48], the ability to contact specialists for advice by telephone or email [48], and more medical/support staff in the primary care office [48] can facilitate the involvement of primary care providers in LTFU.

Some evidence suggests that for survivors at moderate risk of late effects, LTFU care can be provided by a primary care physician [43–46], or a nurse experienced in management of late effects [42, 44], followed by supported self-management [42–44]. Follow-up at a cancer survivorship expert centre is an option as well [42, 44–46]. In addition, shared care

Table 2 Conclusions of evidence from the systematic literature search for models of LTFU care for childhood, adolescent, and young adult cancer survivors

Organisation of LTFU	
Theoretical LTFU care models and LTFU care models used in practice	
Risk-based models for LTFU care: - Cancer centre-delivered care in the paediatric oncology clinic, medical oncology clinic or LTFU clinic - Shared care between treatment hospital and local hospital or primary care - Primary care physician-led LTFU care (community-based care) [16, 23-39, 41-45, 51-53]	Level C
- Specialist nurse-led supported LTFU care - Self-management supported by health professionals within a shared care or primary care model - Certain groups might need specialised clinics such as survivors with HSCT and CNS tumour survivors [43-45, 51, 52]	Expert opinion
LTFU care is provided in a multidisciplinary setting [16, 24-30, 32, 34-36, 38, 41, 42, 53]	Level C
Risk stratification is an important component of LTFU care to assign the level and model of LTFU care [43-45]	Level C
Opinions of survivors and healthcare providers about models of LTFU care	
Primary care physicians are willing to care for childhood cancer survivors in collaboration with a LTFU clinic / cancer centre within a shared care model [42, 54-56, 61, 62, 69, 85, 93]	Level C
Primary care physicians may feel uncomfortable in caring for adult survivors of childhood cancer [54-57]	Level C
Paediatric oncologists preferably provide care in a multidisciplinary team [16, 70, 71]	Level C
Paediatric oncologists feel comfortable in caring for younger childhood cancer survivors, but less comfortable in caring for older childhood cancer survivors [70, 71]	Level C
Survivors are satisfied with a shared-care model [37, 62]	Level C
Survivors want to be able to make their own choice of care provider [67]	Level C
Survivors rate consultant-led follow-up higher than general practitioner-led follow-up, nurse-led follow-up and postal/telephone follow-up [73-75]	Level C
Survivors see the paediatric/adolescent clinic as appropriate location for their LTFU, and do not see a need to change [81]	Level C
Parents prefer the paediatric oncologist model and multidisciplinary team model above the medical oncologist model and general practitioner model [72]	Level C
Opinions of survivors and healthcare providers about barriers to LTFU care	
- Lack of a LTFU programme [16, 80, 87] - Lack of communication between primary care physician and paediatric oncologists [42, 58, 59, 68] - Confusion about role of survivorship programs, oncologists and primary care provider [16, 25, 68] - Lack of support and staff to provide LTFU care [16, 59, 66, 68] - Limited access to refer survivors to specialist care [61] - Lack of adequate insurance or funding for LTFU care [16, 23, 25, 59, 65-68] - Lack of time/ high workload [16, 25, 42, 58, 59, 61, 65, 68, 69] - Distance to clinic for survivors [16, 68] - Inability to locate adult survivors [16, 23]	Level C
Opinions of survivors and healthcare providers about facilitators to LTFU care	
- Access to LTFU care, including access to cancer survivor specialists, access to support services, like social work and psychology, ability to telephone or email specialist for advice, and more medical/support staff in primary care office [53-55, 58, 60, 61, 69, 85, 90, 91, 93] - Adequate insurance [66]	Level C
Effectiveness and feasibility of LTFU care models in practice	
Shared-care for childhood cancer survivors by the family doctor and paediatric oncologist is feasible [62]	Level C
Childhood cancer survivors are satisfied with LTFU care at a paediatric or adult LTFU clinic or shared-care between primary care provider and LTFU clinic [37, 62, 76, 77]	Level C

LTFU long-term follow-up

There were no significant differences in engagement in LTFU care between primary care providers and subspecialty survivorship care providers [37]	Level C
LTFU care for childhood cancer survivors in a paediatric LTFU clinic is feasible [64]	Level C
LTFU care for childhood cancer survivors by a multidisciplinary transition programme in an adult setting is feasible [98]	Level C
LTFU care for childhood cancer survivors by a joint adult/paediatric LTFU care at a paediatric hospital is feasible [29]	Level C
Effectiveness and feasibility risk stratification	
It is feasible to identify and appropriately categorize childhood cancer survivors by the severity (grade) of the expected late effects[34, 43, 44, 46-50]	Level C
Personnel involved in LTFU care	
Theoretical LTFU care models and LTFU care models used in practice	
LTFU care is provided in a multidisciplinary setting, including a paediatric oncology physician, (paediatric oncology) nurse practitioner, primary care physicians (paediatrician, internist, general practitioner), radiation oncologist, medical oncologist, (neuro)psychologist, (paediatric) social worker, dietician, clinical research associate, physician assistants, clinical pharmacist, physiotherapist, (paediatric) endocrinologist, non-clinical staff (program manager, clinical research associate, database manager, administrative support) [16, 24-30, 32, 34-36, 38, 41, 42, 53]	Level C
LTFU care should be coordinated by a key worker [43, 44, 86]	Expert opinion
Opinions of survivors and healthcare providers about who should be involved LTFU care	
Childhood cancer survivors want to be able to make their own choice of care provider [67]	Level C
Childhood cancer survivors prefer hospital-based LTFU, while others would rather attend LTFU at their primary care provider [72-75]	Level C
Nurses can have important roles in the care for childhood cancer survivors [78]	Level C
Paediatric oncologists feel comfortable in caring for younger childhood cancer survivors, but less comfortable in caring for older childhood cancer survivors [70, 71]	Level C
Opinions of survivors and healthcare providers about barriers to LTFU care	
Lack of staff to provide LTFU care [16, 59]	Level C
Effectiveness and feasibility of LTFU care models in practice	
Childhood cancer survivors who attend a paediatric clinic are significantly more [76] or equally [77] satisfied with LTFU care than survivors attending an adult clinic	Level C
Components of LTFU care	
Theoretical LTFU care models and LTFU care models used in practice	
Important components of LTFU care include a survivorship care plan, patient/survivor and parent education, and planned transition to assign the level and model of LTFU care [16, 25-30, 33-36, 39, 41-45, 86]	Level C
Training, development and education programmes for professionals caring for survivors are important [30, 34, 42, 43]	Level C
Audits to test and review if the clinical level and model of care is still appropriate are important [43]	Expert opinion
Opinions of survivors and healthcare providers about requirements of LTFU care	
Survivors and parents have wide ranging requirements for follow-up care, including information and counselling about late effects and preventive care, psychosocial support, parenting issues, and education for primary care providers and parents [66, 67, 72-75, 79-84]	Level C
Survivors are more satisfied with LTFU care when discussing topics they want [73]	Level C
Survivors are willing to transition to adult survivorship care [37, 97]	Level C
Opinions of survivors and healthcare providers about barriers to LTFU care	
<ul style="list-style-type: none"> - Lack of a LTFU programme [16, 80, 87] - Lack of knowledge or awareness about late effects, survivorship issues and needs [10, 16, 23, 25, 37, 42, 54, 55, 57-59, 61, 62, 65, 66, 69, 70, 73, 87, 92] - Lack of knowledge about late effects among survivors and parents [10, 79, 84, 88] - Survivor-related psychosocial barriers (fear, avoidance) [68, 96] - Lack of knowledge and familiarity of LTFU guidelines [54, 55, 61, 65, 66, 70] - Low confidence in managing their survivorship care among survivors [96] - Difficulties organising an appointment (time, distance, scheduling) [68] or finding the right place to go [80] - Lack of a transition program from paediatric to adult healthcare [87] - Inadequate access to survivors' cancer treatment history [61, 65] 	Level C

Table 2 (Continued)

- Lack of experience and inadequate preparation/formal training about survivorship [10, 16, 23, 25, 37, 42, 54, 55, 57-62, 65-70, 73, 79, 80, 87, 88, 92]	
Opinions of survivors and healthcare providers about facilitators to LTFU care	
- Evidence-based LTFU guidelines [54, 55, 70, 85, 103] - Survivorship care plan [53, 55, 61, 70, 90, 91, 93, 103] - Access to support information, medical education seminars, courses or online tools regarding LTFU care [53-55, 58, 60, 61, 66, 68-70, 85, 90, 91, 93]	Level C
Effectiveness and feasibility of LTFU care models in practice	
A survivorship clinic, including a survivorship care plan and patient/survivor education, improved survivors' knowledge of their treatment history and risk of late effects [33]	Level C
Risk-based education in a survivorship clinic increased awareness of personal health risk; the change in awareness did not significantly differ by clinician type (physician vs. nurse practitioner) [31]	Level C
Attendance at a specialized survivor clinic decreases emergency department visits among childhood cancer survivors [89]	Level C
Effectiveness and feasibility of LTFU childhood cancer survivorship care plans	
A survivorship care plan increased compliance with recommended medical surveillance among adult survivors of childhood Hodgkin lymphoma [94]	Level C
A survivorship care plan increased family doctors' and childhood cancer survivors' knowledge of late effects and awareness of the benefits of LTFU [33, 39]	Level C
Primary care providers and childhood and young adult cancer survivors are satisfied with a survivorship care plan; the survivorship care plan did not cause worry or concern in the majority of survivors [91, 93]	Level C
A survivorship care plan supported healthcare providers' adherence to LTFU guidelines and improved communication with childhood cancer survivors about late effects, screening or health behaviour [95]	Level C
Start of LTFU care	
Experts agree that LTFU care should start not later than 5 years after treatment or 5 years from diagnosis, depending on the individual healthcare systems.	Expert opinion

LTFU, Long-term follow-up

Table 2 (Continued)

between the cancer survivorship expert centre and primary care or paediatric centres has been proposed [42, 43, 46]. Some evidence suggests that LTFU within a shared-care model is feasible [52] and that the vast majority of survivors and primary care physicians were satisfied with the follow-up care [37, 52]. In addition, when primary care and subspecialty survivorship care providers were compared, there were no significant differences in survivors' satisfaction and engagement in LTFU [99]. The shared-care model has been successfully implemented, though less frequently than the cancer centre-delivered care model [41, 47].

Some evidence suggests that for high-risk survivors, LTFU can be provided in a cancer centre-delivered care model in the paediatric oncology clinic, medical oncology clinic, or LTFU clinic and many clinics around the world have already adopted this model [16, 23-30, 32, 34-36, 38, 77]. However, distance to the clinic for survivors [16] and the inability to locate adult survivors with increasing time since treatment [16, 23] have been recognized as potential barriers to attend LTFU. Other reported barriers to successful LTFU are lack of support, both from the institution [68] and from colleagues [16, 25], and

especially a lack of health insurance coverage for survivors [16, 23, 25, 59, 68-72]. In addition, lack of dedicated time was reported by primary care physicians [51, 71] and oncologists [16, 25, 53] as an important barrier.

Overarching is the necessity of the provision of multidisciplinary care [16, 24-30, 32, 34-36, 38, 40, 41, 47]. Studies reported that paediatric oncologists preferably provide LTFU care in a multidisciplinary team [16, 57, 58]. Parents of survivors prefer a multidisciplinary team model as well [64].

Recommendations

Based on expert opinion and low-quality evidence, we recommend that LTFU care should be available and accessible for all CAYA cancer survivors throughout their lifespan. Due to their diverse needs, CAYA survivorship care should be provided under the guidance of a cancer survivorship expert service or cancer centre, preferably in a multidisciplinary setting. The adult CAYA cancer survivor and healthcare provider should make the decision for the optimal model of LTFU care together based on previous cancer treatment, diagnosis and health

Table 3 Recommendations for the LTFU care of childhood, adolescent, and young adult cancer survivors based on low-quality evidence and expert opinion

General recommendation
We recommend that LTFU care should be available and accessible for all childhood, adolescent and young adult cancer survivors throughout their lifespan.
Organisation of LTFU care
We recommend that LTFU care for survivors of childhood, adolescent and young adult cancer should:
<ul style="list-style-type: none"> • Be provided in or under the guidance of a cancer survivorship expert service or cancer centre • Provide multidisciplinary care
We recommend that the survivor and healthcare provider make a joint decision for the optimal model of LTFU care,¹ based on previous cancer treatment, health conditions, survivor preferences and the healthcare system.
To provide LTFU care for survivors of childhood, adolescent and young adult cancer we recommend:
<ul style="list-style-type: none"> • To have commitment of the (national and local) healthcare providers (systems) and insurers • To have sufficient time for consultation
Personnel involved in LTFU care
We recommend that each survivor can make their own informed choice for a healthcare provider after informed discussion with the survivorship team.
We recommend that the cancer survivorship expert centre that will organize LTFU care includes:
<ul style="list-style-type: none"> • Key worker/coordinator • Lead doctor specialised in late effects • Nurse practitioner • Multidisciplinary expert team of specialists² • The possibility of consulting specific specialist³
Components of LTFU care
We recommend that LTFU care for survivors of childhood, adolescent and young adult cancer include:
<ul style="list-style-type: none"> • Surveillance and preventive strategies based on published evidence based guidelines • Coordination of care (particularly in shared care models) • Education for professionals • Education of survivors, families & carers • Coordination of scientific research
We recommend that the cancer survivorship expert centre provides:
<ul style="list-style-type: none"> • An individualized survivorship care plan <ul style="list-style-type: none"> - Including a treatment summary with risk stratification care plan • Patient/survivor and parent education to support effective self-management • A plan for transition of care: <ul style="list-style-type: none"> - From active treatment to LTFU - From survivorship expert centre to primary care (for low risk survivors) - From paediatric to adult health services
Start of LTFU care
We recommend that LTFU care should start not later than 5 years after treatment or 5 years from diagnosis, depending on the individual healthcare systems.

Green, class I = strong recommendations to do
LTFU, Long-term follow-up care

¹ Self-management with primary care support for adult survivors; follow-up at primary care level or by a nurse experienced in management of late effects, followed by supported self-management; follow-up at cancer survivorship expert centre; or shared care between survivorship expert centre and primary care or paediatric centres.

² paediatric oncologist/haematologist, (neuro-) psychologist, cardiologist, endocrinologist, medical oncologist, haematologist, rehabilitation physician, occupational worker, radiotherapist, social worker

³ pulmonologist, nephrologist, neurologist, neurosurgeon, ear nose and throat specialist, ophthalmologist, gynaecologist, dermatologist, insurance worker, urologists, general internal medicine

Green, class I = strong recommendations to do

LTFU long-term follow-up care

¹ Self-management with primary care support for adult survivors; follow-up at primary care level or by a nurse experienced in the management of late effects, followed by supported self-management; follow-up at a cancer survivorship expert centre; or shared care between survivorship expert centre and primary care or paediatric centres

² Paediatric oncologist/haematologist, (neuro-) psychologist, cardiologist, endocrinologist, medical oncologist, haematologist, rehabilitation physician, occupational worker, radiotherapist, social worker

³ Pulmonologist, nephrologist, neurologist, neurosurgeon, ear nose and throat specialist, ophthalmologist, gynaecologist, dermatologist, insurance worker, urologists, general internal medicine

status, and survivor preferences and taking into account the available healthcare system. The model of care for children will depend on local policies.

Regular review of the level of care and flexibility herein is important, especially the possibility for quick referral back into the cancer survivorship care system. Moreover, we recommend that (national and local) healthcare providers (systems) and insurers have commitment to provide LTFU care for survivors and allocate sufficient time (strong recommendations).

Personnel involved in LTFU care

Evidence

Some evidence suggests that despite the various models recommended for survivors at different risks for late effects, experts and survivors agree that survivors should make their own choice for their LTFU provider [22, 59]. A successful model of LTFU needs the flexibility to adapt to the individual survivor. This is especially important because many survivors prefer hospital-based LTFU, while others would rather attend LTFU at their primary care provider [60–62, 64]. Also in contrast to expectations, many childhood cancer survivors who attend a paediatric clinic are significantly more [75] or equally [76] satisfied with LTFU care than survivors attending an adult clinic. This is important because in many clinics, LTFU for adult CAYA cancer survivors is still provided at the paediatric hospital [77], although studies suggest that paediatric oncologists do not feel very comfortable caring for older adults [57, 58], and some healthcare systems do not allow adult survivors to be seen in paediatric settings. To empower survivors in making their choice of provider, the evidence and our expert panel in Erice [22] agreed on the importance of a key worker or coordinator of care [40]. Other important LTFU staff that should be available at a cancer survivorship expert centre as reported in the literature are a lead doctor specialized in late effects and a nurse practitioner [85]. Lack of staff to provide LTFU has been indicated to be a major barrier [16, 68].

Recommendations

Based on expert opinion and low-quality evidence, we recommend that each CAYA cancer survivor should make their own informed choice for a healthcare provider after discussion with the survivorship team. The cancer survivorship expert centre that will organize LTFU care should include a key worker/coordinator, lead doctor specialized in late effects, nurse practitioner, and a multidisciplinary expert team of specialists, like paediatric oncologist/haematologist, (neuro-) psychologist, cardiologist, endocrinologist, medical oncologist, haematologist, rehabilitation physician, occupational worker,

radiotherapist, and social worker. In addition, it is important that there is the possibility of consulting other specialist, like pulmonologist, nephrologist, neurologist, neurosurgeon, ear nose and throat specialist, ophthalmologist, gynaecologist, dermatologist, insurance worker, urologists, and general internal medicine. It is important to keep in mind that all aspects of LTFU might need to be adapted to the healthcare system in place (strong recommendations).

Components of LTFU care

Evidence

Survivors [59–63, 65, 70, 86–88] and their parents [64, 89] reported a wide-range of needs for LTFU. One of the main core components of LTFU care is the provision of information, counselling and surveillance for late effects, and preventive care based on evidence-based guidelines. Primary care physicians [48, 49, 53, 54, 67] and oncologists [53, 57] report the need for guidelines, and a lack thereof is seen as an important barrier to successful LTFU by primary care physicians [48, 49, 51, 71], oncologists [57], and survivors [70]. The ideal format may be one that can be instantly accessed during the follow-up appointment [54].

Another important core component is the coordination of care. There is potential confusion between the different roles of survivorship programmes, oncologists, and primary care providers [72]. Experts have highlighted the importance of well-coordinated care (particularly in shared-care models) and proposed the inclusion of a key worker [42, 43, 84].

Education of professionals involved is essential. Some evidence suggests that a lack of knowledge and experience is seen as a barrier to providing LTFU care by primary care physicians [41, 51, 67], oncologists [53], and policy experts [66], and the need for continued education is widely accepted [30, 34, 41, 42, 48, 53]. Online tools are seen as a suitable possibility to reach many professionals [48, 54]. Education of survivors, families, and carers to increase awareness and knowledge about cancer and treatment-related risks of late effects, and the importance of LTFU care is necessary as well. This has been highlighted by experts [42, 84] but is also shown in practice [16, 25, 28, 30, 31, 33, 39, 41]. Studies showed that the lack of knowledge is seen as a barrier to successful LTFU not only by oncologists [16, 23, 25, 68] but also by survivors [10, 86, 92] and parents themselves [89, 92]. In addition, some evidence suggests that a survivorship clinic, including survivor education, may improve survivors' knowledge of their treatment history and risk of late effects [33], may increase awareness of personal health risk [31], and may decrease emergency department visits [95].

Another core component of LTFU care is the coordination of scientific research to improve evidence-based care and to

audit its effectiveness [41]. One US study showed that three quarters of 179 responding survivorship programmes had a research associate available [25].

An important tool for successful LTFU is the individualized survivorship care plan (SCP) including a treatment summary [16, 25, 28, 30, 33, 35, 39–43, 84]. A SCP has been reported as being of great importance by survivors [10, 37, 47, 52, 60, 73, 74, 91], oncologists [57], and also primary care providers [49, 51, 55, 67, 71]. Some evidence suggests that primary care providers and survivors are satisfied with the SCP and that it may increase knowledge of late effects and awareness of the benefits of LTFU [39, 55, 74]. In addition, a SCP may improve communication with survivors about late effects and the sharing of more comprehensive information [96, 97]. It may support healthcare providers' adherence to guidelines on late-effect screening and survivors' compliance with recommended medical surveillance [96, 97]. However training of providers is essential [56].

Effective self-management is an important component of survivorship care. To achieve this aim, survivor and parent education is vital, especially when survivors reach adulthood and take over responsibility for their health. Some evidence suggests that survivors reported low confidence in managing their survivorship care [93], including problems organizing appointments [69], or finding the right place to go [65].

Another aspect of high importance to successful LTFU of adult CAYA cancer survivors is their transition from paediatric to adult care [42, 43, 84]. Oncologist and healthcare policy experts agree that the lack of a transition programme is a major barrier [16, 66], despite evidence that survivors are willing to transition to adult survivorship care [37, 90]. Several institutions have already successfully implemented planned transition in their programmes [25, 26, 30, 35, 36, 41, 78].

Recommendations

We recommend that LTFU care for CAYA cancer survivors include (1) surveillance and preventive strategies based on published evidence-based guidelines, (2) coordination of care (particularly in shared-care models), (3) education for professionals, (4) education of survivors, families, and carers, and (5) coordination of scientific research. Based on expert opinion and low-quality evidence, we recommend that an individualized SCP is provided to every CAYA cancer survivor at the end of treatment, including a treatment summary with a care plan. In addition, the cancer survivorship expert centre should provide survivor and parent education to support effective self-management and a plan for transition of care from active treatment to LTFU, from a survivorship expert centre to primary care (for low-risk survivors) and from paediatric to adult health service (strong recommendations).

Start of LTFU care

Recommendations

Although there is currently no evidence available, our expert panel agreed that LTFU is recommended to start no later than 5 years after treatment or 5 years from diagnosis, depending on the healthcare system (strong recommendation).

Discussion

This systematic review of available evidence and expert panel discussions confirmed the importance of LTFU care for CAYA cancer survivors. We have made comprehensive recommendations for delivering effective LTFU care, relevant to healthcare providers, survivors and their families, and researchers.

Our recommendations describe the minimal requirements for LTFU. The exact organization of care will depend on the national healthcare system, regional characteristics, features of the hospital, and finally preferences and needs of survivors and healthcare providers. In the UK, for example, the National Cancer Survivorship Initiative (NCSI) has been developed by experts and stakeholders and resulted in similar recommendations as we concluded from our evidence [100]. A one-stop survivorship clinic, including access to a multidisciplinary team, when needed, within one appointment, would provide a suitable system decreasing organizational barriers for survivors. Another important aspect is easy accessibility for all survivors, especially those who might not have attended LTFU for a long time and who wish to return to follow-up care.

Despite the overall lack of clinical trials addressing the impact of LTFU on survivors' medical and psychosocial outcomes, we showed that there is some evidence for a positive effect: survivors attending LTFU demonstrated higher knowledge about their diagnosis and treatment and more accurate perception of their risk for late effects [31, 33], were engaged in regular surveillance [29, 96], and had fewer visits to the emergency department [95]. In addition, a recent review also showed that survivors attending LTFU care had more late effects detected [101]. This demonstrates the importance of LTFU.

We also provide evidence for the usefulness of an individualized SCP. However, clear evidence for the effectiveness of an SCP to improve LTFU and survivor's health in the long term is still lacking [102]. Furthermore, follow-up recommendations will change with new and updated guidelines, and survivors' follow-up care will therefore need to be reviewed regularly and adapted if necessary.

More than 10 years after the report "From Cancer Patient to Cancer Survivor: Lost in Transition" by the Institute of

Medicine [103], there has been a large amount of research and publications addressing LTFU of CAYA cancer survivors. However, our literature review still showed some important gaps in knowledge. While opinions of survivors, parents, providers, and experts have been assessed frequently and LTFU implemented in many places, there is still a lack of studies on effectiveness. The models themselves, the provision of care including risk stratification, and also specific interventions such as an SCP need to be evaluated for their effectiveness to detect late effects and improve survivors' quality of life and knowledge acquisition as well as their cost-effectiveness. An update of our guideline in 5 years will hopefully be able to include stronger evidence of effectiveness.

To improve health outcomes and quality of survivorship of current and future survivors, continued age-adapted education of survivors about the cancer, its treatment, risk for late effects, importance of health behaviours, and necessity of LTFU is important along the cancer and survivorship trajectory. Well-organized LTFU will help to reach this goal.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval This article does not contain any studies with human participants performed by any of the authors.

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