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Prevalence of childhood cancer survivors in Europe: a scoping review

Neimar de Paula Silva [a,*], Andrea Gini [a], Anastasia Dolya [a], Murielle Colombet [a], Isabelle Soerjomataram [a], Danny Youlden [b], Charles Stiller [c], Eva Steliarova-Foucher [a], the CRICCS consortium, Joanne Aitken [d], Freddie Bray [a], Murielle Colombet [a], Neimar de Paula Silva [a], Anastasia Dolya [a], Friederike Erdmann [e], Jeanette Falck Winther [f], Andrea Gini [a], Delphine Heenen [g], Lars Hjorth [h], Claudia E. Kuehni [i], Kathy Pritchard-Jones [j], Marion Piñeros [a], Isabelle Soerjomataram [a], Eva Steliarova-Foucher [a], Charles Stiller [k], Zuzana Tomášiková [l], Danny Youlden [b]

[a] *Cancer Surveillance Branch, International Agency for Research on Cancer (IARC), Lyon, France*
[b] *Cancer Council Queensland and Griffith University, Australia*
[c] *National Cancer Registration and Analysis Service, NHS England, United Kingdom*
[d] *Cancer Council Queensland and University of Queensland, Australia*
[e] *Research group Aetiology and Inequalities in Childhood Cancer, Division of Childhood Cancer Epidemiology Institute of Medical Biostatistics, Epidemiology and Informatics (IMBEI), University Medical Center of the Johannes Gutenberg University Mainz, Germany*
[f] *Danish Cancer Society Research Center, Aarhus University and University Hospital, Denmark*
[g] *Childhood Cancer International – Europe, KickCancer, Belgium*
[h] *Lund University, Skane University Hospital, Department of Clinical Sciences Lund, Paediatrics, Sweden*
[i] *University of Bern, Institute of Social and Preventive Medicine (ISPM), Switzerland and Paediatric Haematology/Oncology, University Children's Hospital, University of Bern, Switzerland*
[j] *University College London, United Kingdom*
[k] *National Cancer Registration & Analysis Service, NHS England, United Kingdom*
[l] *Childhood Cancer International – Europe, Childhood Cancer Switzerland, Switzerland*

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ABSTRACT

Childhood cancer survivors (CCS) require specialized follow-up throughout their lifespan to prevent or manage late effects of cancer treatment. Knowing the size and structure of the population of CCS is crucial to plan interventions. In this scoping review we reviewed studies that reported prevalence of CCS in Europe. We searched Medline, Web of Science, and Embase using permutations of terms referring to childhood, cancer, survivors, prevalence, registries, and Europe. We followed PRISMA-ScR guidelines to select studies and The Joanna Briggs Institute Prevalence Critical Appraisal Tool to evaluate their quality. From 979 unique studies published between 1989 and 2022, 12 were included. Limited-duration prevalence (LDP) for all childhood cancers, assessed in three studies using counting method, varied between 450 and 1240 persons per million. Complete prevalence (CP) of survivors of any childhood cancer except skin carcinomas, reported in three studies using observed data complemented with modelled data for the unobserved period, varied between 730 and 1110 persons per million. CP of survivors of an embryonal tumour was estimated by completeness index method in six studies. In four of them CP ranged from 48 to 95 persons per million for all embryonal tumours, while CP for those occurring in central nervous system was 43 per million in one study and CP for rhabdomyosarcoma was 17 per million in another. Information on prevalence of CCS in Europe is fragmented and inconsistent. The large variations in LDP and CP estimates were linked to differences in data availability, the selection of populations, prevalence measure,

Abbreviations: CCS, childhood cancer survivors; CHILDPREV, method to compute prevalence of disease in childhood; CNS, central nervous system; CP, complete prevalence; CRICCS, Cancer Risk in Childhood Cancer Survivors; EU27, European Union 27 countries; GDPR, General Data Protection Regulation; LDP, limited-duration prevalence; HIC, high-income countries; IARC, International Agency for Research on Cancer; PIAMOD, Prevalence and Incidence Analysis Model; POCCS, Prevalence Of Childhood Cancer Survivors; PRISMA-ScR, Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews.

* Correspondence to: Cancer Surveillance Branch, International Agency for Research on Cancer, 25 avenue Tony Garnier, CS 90627, LYON CEDEX 07 69366, France.

E-mail address: DePaulaSilvaN@iarc.who.int (N. de Paula Silva).

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statistical method, incidence period, index date, age at diagnosis and prevalence, cancer types, sex, and, for LDP, also the length of follow-up. Standardisation of methodology and reporting are needed to systematically monitor and compare CCS prevalence in Europe and provide data to help address survivors' needs.

1. Introduction

The proportion of childhood cancer survivors (CCS) in populations of high-income countries (HIC) is steadily increasing [1,2]. This proportion, expressed as prevalence, is determined by other measures of disease burden: incidence, and survival or mortality. The mild increase in incidence rates [3], partly sustained by improving diagnostic techniques, mainly in HIC [4,5], and reduced mortality [6] contribute to growing prevalence. Mortality is decreasing due to the improved survival [1,2,7], as seen in Europe [8], the USA [1,9], Canada, and Australia [9,10]. Better survival has been attributed to advances in tumour diagnostics, improved treatment effectiveness, advanced provision and organization of health care, supported by improving socioeconomic conditions, and general health status [8].

Survivors of childhood cancer have a greater risk of acquiring various chronic health conditions in comparison with their peers, who have not had cancer during childhood [11,12]. Findings from the PanCare studies show that CCS have higher risk of developing cardiac events [13,14], fertility impairments [15], and second cancers [16–19]. Prevalence of chronic health conditions was estimated to range from 66% (prevalent at age 5–19 years) to 88% (prevalent at age 40–49 years) among CCS in the USA. One third of these 5-year survivors were affected by severe chronic conditions, including second cancers, and the absolute risk of these adverse effects increased with time since diagnosis [1].

To meet specific needs of the growing population of CCS, it is essential to know its size. Population-based cancer registries collect information on all cancers occurring in populations they cover, and most of them also follow-up the registered patients and record date of death or emigration, as they occur [20]. These data can be used to count the number of CCS and thus estimate their prevalence at any given index date. To provide accurate and complete prevalence, the whole population should be monitored for the whole life span of all its members. However, most cancer registries have not yet attained such a long surveillance period and may miss information on migrants. Therefore, prevalence of CCS is estimated using incidence data provided by registries and vital statistics of the general population.

Limited-duration prevalence (LDP) describes the number of survivors diagnosed during a specified registration period and alive on an index date and complete prevalence (CP) is the number of all CCS alive on an index date, regardless of date of diagnosis. The counting method [21] provides precise prevalence estimates [22], by counting the number of CCS alive on an index date and accounting for loss to follow-up. The Prevalence and Incidence Analysis MODel (PIAMOD) [23] is a parametric method that estimates CP, even in the absence of the complete surveillance period, using age period cohort models to project incidence and relative survival estimates to substitute the unobserved data for the past and future parts of a study period. The flexibility of the incidence model implemented in PIAMOD [23] makes it appropriate for future projections of prevalence [24]. The completeness index method [25] corrects the LDP estimated with observed data by quantifying the completeness of the estimate and complements it for the period preceding the start of the registration to account for all cancer patients living in a population. To estimate CP of CCS specifically, the CHILDPREV [24] method is the most appropriate because it is less reliant on past survival trends, which are difficult to impute because childhood cancer survival has improved dramatically in Europe during 1950 s and 1960 s [26], prior to the start of most cancer registries [27]. CHILDPREV is a semi-parametric method that corrects the LDP estimate [28] with completeness index [25]. Implementation of any of the methods described above requires individual cancer records with follow-up information for the entire population, long-term mortality estimates, and assumptions on patterns of incidence and survival for the unobserved period.

Five-year prevalence was estimated systematically for populations of any age in all countries, for total and for selected cancers in the Global Cancer Observatory (GCO, gco.iarc.fr). The GCO estimate of the overall 5-year LDP in the age range 0–19 years for Europe in 2020 shows 605 per million [29]. These estimates were computed using the country-specific incidence and the ratio of 5-year LDP prevalence to incidence observed in Nordic countries in 2006–2015, which was scaled by the ratio between the Human Development Index (HDI) of the country of interest and the Nordic region [29]. The GCO methodology allows comparisons of 5-year LDP across countries without a distinction of age at diagnosis, as a useful indicator of health care needs in the population diagnosed with cancers mostly occurring in mid or older age. Although GCO prevalence estimates serve the purpose for which they were designed, they do not estimate prevalence of CCS. Recently, a systematic evaluation of CP of cancer patients irrespective of age, using data from 61 population-based cancer registries in 27 European countries (EU27), showed an increase in prevalence in the region, however no prevalence of CCS was reported [30].

As CCS may have increased health care needs for long periods of life, estimating their prevalence over variable follow-up periods is of interest. As part of the Cancer Risk in Childhood Cancer Survivors (CRICCS) study (criccs.iarc.who.int), we conducted a scoping review of literature to map existing knowledge and identify gaps in information on prevalence of CCS in Europe.

2. Methods

We performed a scoping review using methodology recommended by Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) [31]. We did not register the study protocol. Our research question was: "What is the prevalence of childhood cancer survivors in Europe?" We selected three bibliographic databases as sources of evidence: Medline, Web of Science, and Embase, to identify all publications estimating prevalence of CCS in Europe. The key search terms were related to childhood, cancer, survivors, prevalence, registries, and Europe (Supplement, Panels S1-S3). Europe was defined according to the UN definition [32]. The search strategies were implemented on 23 September 2022 without limits on language and date of publication (Supplement, Panels S1-S3).

The identified publications were imported into EndNote X9.3.3 [33]. After removal of duplicates, two reviewers (NdPS and AG) assessed independently the relevance of all studies. A study was included in the review, if it: was based on data from a population-based cancer registry; was conducted in one or more European countries; and reported on at least 10-year prevalence of individuals who were diagnosed before the age of 20 years with any malignancy or non-malignant tumors of the central nervous system (CNS). During the search, we identified several studies reporting prevalence estimates for patients diagnosed with embryonal tumours at any age. As more than 90% of embryonal tumours occur in young children [34], we have also included these studies because they implicitly complied with the selection criteria.

Fig. 1 shows the selection of studies. In the first step, we selected studies based on title and abstract. In the second step, we reviewed the full text of the identified publications. All disagreements between the two reviewers were resolved by consensus. Studies excluded in the second step are listed in Supplement, Panel S4.

Two reviewers (NdPS and AG) extracted the following information

from the included studies: authors' names, publication year, population, prevalence measure, statistical method, incidence period, index date, age at diagnosis, age at prevalence, cancer type, sex, and prevalence estimate. Extracted information was recorded in an Excel file [35].

The same reviewers evaluated independently the methodological quality of the included studies according to The Joanna Briggs Institute Prevalence Critical Appraisal Tool [36,37]. The 10 assessment criteria are shown in Supplement, Table S1. All disagreements on the assessment were resolved by consensus. No a-priori limit was set to exclude studies from the review (Supplement, Panel S5).

3. Results

Overall, 1237 studies were identified and 979 were retrieved after the removal of duplicates. Based on title and abstract, 928 studies were excluded. Full text screening was thus performed on 51 studies and resulted in the exclusion of a further 39 studies. The remaining 12 studies were included in this review (Fig. 1). All selected studies were published in English.

An overview of the 12 included studies is shown in Table 1. The studies varied with respect to the age at cancer diagnosis (any age including adults or only childhood), age at prevalence (<1 year up to any age), cancers considered (all or selected types), statistical method used (counting, completeness index, PIAMOD or CHILDPREV), the type of the prevalence measure (CP or LDP), etc. Complete results for all included studies are shown in Supplement, Table S2. No data quality concerns were identified using the standard quality assessment criteria [36,37] (Supplement, Table S1).

3.1. Prevalence of survivors diagnosed with any neoplasm during childhood

Both the lowest and the highest prevalence estimates were found in the study of 26-year LDP in Sweden in 1984 after cancers diagnosed during the period 1958–1984 [38]. They ranged from 450 per million children for the age group 0–4 year at prevalence to 1240 per million females aged 15–19 years at prevalence [38]. Two studies [39,40] used comparable set of parameters with overlapping incidence periods, the same index date, age at diagnosis under 15 years and any age at prevalence for all cancers except skin carcinomas. However in the Swiss study [40] the estimates were obtained using the PIAMOD [23] method, while the Italian study reported results obtained using the CHILDPREV [24] method. The resulting overall estimate for both sexes combined was somewhat higher in Switzerland (897 per million) [40] than in Italy (730 per million) [39], Table 1.

3.2. Prevalence of survivors of embryonal tumours at any age

Four studies [34,41–43], based on the same RARECARE data (rarecarenet.istitutotumori.mi.it), estimated CP of embryonal tumours at any age at diagnosis and prevalence, for both sexes combined, on the index date of 1st Jan 2003 for 27 countries of the European Union (EU27), considering the incidence period of 1988–2002. The settings were the same in two of these studies in which CP of all embryonal tumours were 79.6 [42] versus 77.9 [34] per million persons. CP for embryonal tumours of CNS was 43 per million [41] and CP for embryonal rhabdomyosarcoma was 17 per million [43]. In an Italian study [44], CP of survivors of all embryonal tumours for the same period in a population of five regions in Italy was 48 per million. In another Italian study [45], based on pooled data from 11 Italian regions including four of the five regions from the previous study [44], with the incidence period and index date shifted to four years later (1992–2006 and 1st January 2007, respectively), CP for all embryonal tumours was 95 per million. All the six studies [34,41–45] used the completeness index method [25].

4. Discussion

In this scoping review, we summarised estimates of CCS prevalence in Europe published in international journals catalogued in three

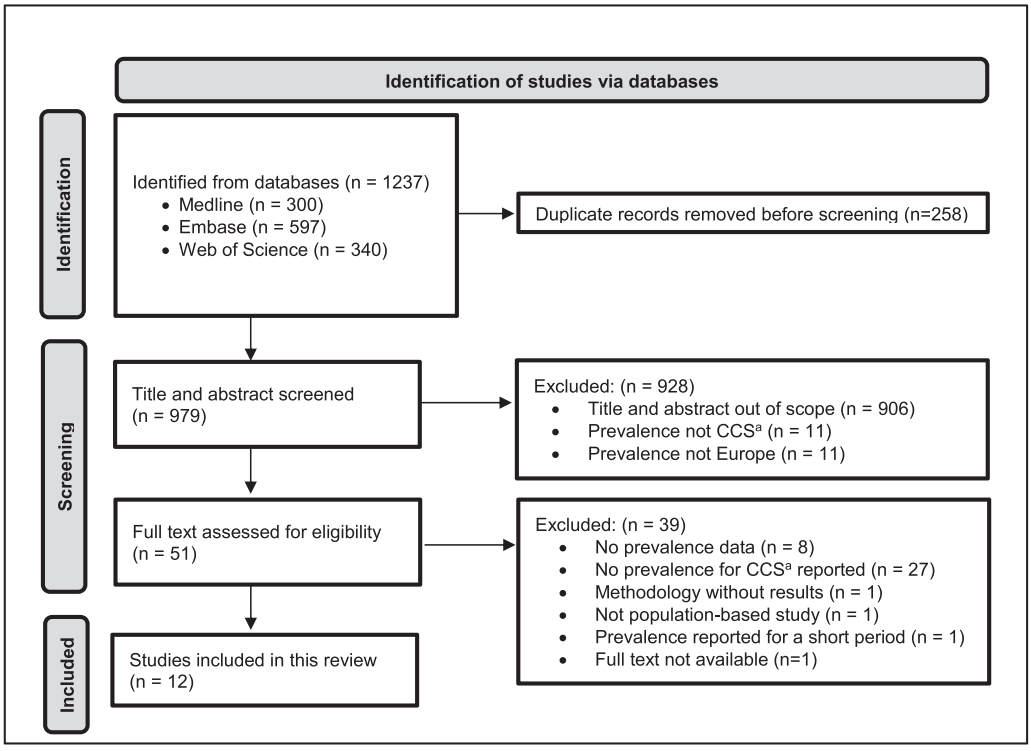

Fig. 1. Selection process of studies of prevalence of childhood cancer survivors in Europe in the scoping review, according to the Preferential Reporting Items in Systematic review and Meta-Analyses Extension for Scoping Reviews (PRISMA-ScR) guidelines [31]. [a]CCS, childhood cancer survivors.

Table 1
Studies included in this scoping review of publications reporting on prevalence estimates of childhood cancer survivors in Europe.

Authors, publication year	Population	Prevalence measure	Statistical method	Incidence period	Index Date[a]	Age at diagnosis (years)	Age at prevalence (years)	Cancer type	Sex	Prevalence[b] (N per million)
Adami et al., 1989 [38]	Sweden	26-year LDP[c]	Counting [21]	1958–1984	31/12/1984	all	0–4	all cancers[d]	male	450
									female	450
							5–9	all cancers[d]	male	920
									female	880
							10–14	all cancers[d]	male	1150
									female	1080
							15–19	all cancers[d]	male	1220
									female	1240
Johannesen et al., 2007 [54]	Norway	49-year LDP[c]	Counting [21]	1953–2002	31/12/2002	<15	all	all cancers[e]	both	651
Herrmann et al., 2013 [40]	Switzerland, 4 registries	CP[f]	PIAMOD [23]	1990–2010	01/01/2010	<15	all	all cancers excl. skin carcinoma	both	897
Pisani et al., 2013 [55]	Italy, 1 registry	33-year LDP[c]	Counting [21]	1976–2008	01/01/2009	<15	0–33	all cancers[g]	both	1166
Francisci et al., 2017 [39]	Italy, 15 registries	CP[f]	CHILDPREV [24]	1995–2009	01/01/2010	<15	all	all cancers excl. skin carcinoma	both	730
Guzzinati et al., 2018 [2]	Italy, 8 registries	CP[f]	C. index[h,25]	1976–2009	01/01/2010	all	<15	all cancers excl. skin carcinoma	male	1110
									female	930
Embryonal tumours										
Gatta et al., 2011 [42]	EU27[i,j]	CP[f]	C. index[h,25]	1988–2002[k]	01/01/2003	all	all	all embryonal tumours	both	80
Crocetti et al., 2012 [41]	EU27[i,j]	CP[f]	C. index[h,25]	1988–2002[k]	01/01/2003	all	all	embryonal tumours of CNS[l]	both	43
Gatta et al., 2012 [34]	EU27[i,j]	CP[f]	C. index[h,25]	1988–2002[k]	01/01/2003	all	all	all embryonal tumours	both	78
Trama et al., 2012 [44]	Italy, 5 registries	CP[f]	C. index[h,25]	1988–2002	01/01/2003	all	all	all embryonal tumours	both	48
Stiller et al., 2013 [43]	EU27[i,j]	CP[f]	C. index[h,25]	1988–2002[k]	01/01/2003	all	all	embryonal rhabdomyosarcoma of soft tissue	both	17
Busco et al., 2016 [45]	Italy, 11 registries	CP[f]	C. index[h,25]	1992–2006	01/01/2007	all	all	all embryonal tumours	both	95

[a] Date at which prevalence was estimated
[b] Rounded to a full number
[c] LDP, limited-duration prevalence
[d] ICD-7 code 140–209
[e] Including all malignant and benign tumours of central nervous system
[f] CP, complete prevalence
[g] According to ICCC-3; it is not clear if the authors included only malignant or all central nervous system tumours
[h] C. index, completeness index
[i] Estimates based on national and regional cancer registries covering 32% of European population
[j] EU27, European Union 27 countries
[k] Studies using the same dataset
[l] CNS, central nervous system

bibliographic databases. We found childhood cancer prevalence range from 450 to 1240 per million people, according to the choice of methods and study designs. The CP of patients diagnosed with embryonal tumours ranged from 48 to 95 per million people. The variations in the estimates depended on the study setting, underlying population, prevalence measure, statistical method, incidence period, index date, age at diagnosis, age at prevalence, cancer type, and, for LDP studies, also on the length of follow-up. Overall, CCS prevalence was estimated for four European countries and, prevalence of embryonal tumours only, for the EU27.

The estimates of prevalence of survivors of embryonal tumours in the EU27 [34,42] were the most comprehensive. These studies were based on a selection of RARECARE data, which covered almost 30% of the EU27 population during the incidence period 1988–2002 with the index date end of 2003. Data from five Italian registries, likely participants of the RARECARE study [34,42], were also analysed separately [44]. The EU27 prevalence estimate of 78 per million [34] was 63% higher than the estimate for the Italian subset (48 per million) [44]. This striking difference can be explained by the lower crude incidence (Italy 2.7 per million, EU27 4.0 per million) and the 5-year relative survival (Italy 76.8%, EU27 80.2%) [34,44]. in Italy compared to the EU27. In addition, the CP of embryonal tumours, estimated in a more recent study [45] targeting an enlarged Italian population of 11 registries, doubled in 2007 to 95 per million for a slightly later incidence period 1992–2006, surpassed the earlier European estimates [34,42]. Although incidence and survival of embryonal tumours have been increasing slightly in Southern Europe [8,46], these changes alone are unlikely to explain such a large difference in CP. Instead, we suggest that data used in the later study [45] contained information on a bigger number of survivors, due to the longer registration history of the contributing registries, and included a larger number of those diagnosed more recently with likely better survival, consequently producing more reliable and higher estimates. It would be possible to test this hypothesis, for example, by comparing CP based on incidence period 1988–2002 with CP based on period 1992–2006 for the same population of five registries [44]. The findings from the two Italian studies [44,45] thus demonstrate the considerable sensitivity of prevalence estimates to the selection of population covered, period of incidence, and index date.

The selection of age at prevalence also affects prevalence estimates. Reporting LDP for selected age groups at prevalence implies including only cases incident in specific periods of registration. This is evident from the Swedish study [38], where 26-year LDP is reported by 5-year age groups at prevalence. Cancer prevalence of 450 per million, reported for the age group 0–4 years at prevalence in 1984 refers to CCS diagnosed between 1980 and 1984 and corresponds to 5-year LDP. Prevalence of 1240 per million for age group 15–19 years at prevalence concerns CCS diagnosed in 1965–1984 for age at incidence of 0–19 years and implies 20-year LDP. Both these LDP estimates are also CP for these two cohorts, and they differ markedly because of differences in the age range of incident cases. The shown differences highlight the importance of interpreting prevalence estimates in terms of the corresponding parameters.

Similarly, age at diagnosis influences the resulting prevalence estimates. For example, the CP of 730 per million in 2010 was estimated for CCS aged 0–14 years at diagnosis in Italy [39], while CP estimated for the USA in 2011 corresponded to 1260 per million people for CCS aged 0–19 years at first cancer diagnosis [1,47]. In addition to differences in the underlying populations and cancer burden indicators, the much higher CP in the USA also reflects the inclusion of the age group 15–19, in which the incidence rates are much higher than in age 0–14 years [46]. Therefore, prevalence estimates for different age groups at diagnosis are not directly comparable.

Differences in the prevalence estimates are further affected by the prevalence measure used. While LDP is the number of CCS diagnosed over a limited time period, CP augments the LDP by the unobserved prevalence of survivors diagnosed before the registry started, which is estimated in statistical models using assumptions on incidence and survival in the pre-registration period. CP provides a higher number of prevalent CCS than LDP because LDP is a part of CP. In CHILDPREV [24], the standard method used to estimate CP of CCS, age-, sex-, and period-specific incidence and survival estimates are employed to model the unobserved prevalence. CHILDPREV [24] was applied to estimate CP of CCS in Italy [39] and the USA [1]. In both settings, part of the incidence and survival patterns, prior to the start of cancer registration, were unobserved and the length of the unobserved period therefore influenced the resulting estimates.

The length of the unobserved period requires modelling of incidence and survival, which impacts the precision and validity of CP estimates. Many European registries started in the 1970 s or 1980 s, so that survival estimates for the preceding years are mostly based on scarce data, possibly observed in other populations, and these may not represent patterns experienced within the populations to which extraneous data were imputed. In addition, childhood cancer survival changed dramatically during the 1950 s and 1960 s [26], but there were very few registries to report on childhood cancer survival for that period [27]. Therefore, when the unobserved period is very long or when cancer burden patterns are not known, using LDP instead of CP may be preferable.

Prevalence estimates may be sensitive to the use of statistical methods. For example, a similar set of parameters (e.g, prevalence measure, period of incidence, age at diagnosis, age at prevalence, and the index date), used in the Swiss [40] and Italian [39] studies, yielded higher CP in Switzerland [40]. The authors of the Swiss study [40] used PIAMOD [23], a fully parametric method reliant on the assumptions about incidence and survival to a larger extent than CHILDPREV [24], therefore the Swiss estimates [40] are more likely to have been influenced by changes in incidence and survival trends in the past which may have contributed to the observed differences.

In the reviewed studies, prevalence was reported as number of survivors or as a proportion of the total population. As countries differ in the demographic structure of their populations, prevalence proportions should be compared by age group or standardized for international comparisons. However, for planning and allocating health resources for CCS on a national level, the crude number of prevalent cases of CCS may be more useful.

The principal strength of our study is its large scope. Although we identified 979 unique studies in the initial literature search, only 12 fulfilled selection criteria to describe prevalence of CCS in Europe. In addition, prevalence of CCS in Europe was systematically assessed only for embryonal tumours. Another strength of our review is the quality assessment of the reviewed studies, according to the standard criteria [36,37], although such assessment is not mandatory in scoping reviews [48,49].

We admit that the omission of grey literature, such as registries' periodic reports, which might have included estimates of CCS prevalence, might be considered as a limitation. However, such reports might be not widely available and their documentation not exhaustive. Therefore, we assume that searching in widely accessible databases likely covers most estimates and methodological approaches. This approach is in line with the purpose of scoping reviews, which are designed to capture an overview of easily accessible evidence [48,49]. Although our search strategies were not limited by language or time, all included studies were published in English and the period of incidence was limited to 2–3 decades. We believe that if a study existed that reported prevalence of CCS based on data from a registry covering the full lifespan of a cohort it would be very likely to have been published as a peer-reviewed publication and found in the searched databases. Therefore, the prevalence estimates gathered in this review might be considered as a reasonable proxy to what is known on CCS prevalence in Europe.

Most registries operating in Europe collect or have access to all data required to monitor childhood cancer prevalence. National registries

with a long history of registration of cancers diagnosed at any age provide the best setting to achieve complete ascertainment of childhood cancers and patients' follow-up needed to estimate prevalence of CCS. Coverage of the childhood population by existing registries may attain 52–83%, depending on the definition of Europe [50]; however, part of this coverage is ensured only by paediatric registries, which are less likely to contain all the data needed to generate prevalence estimates at any age. Most countries thus do not have enough data to construct stable models and apply standard prevalence methodology (e.g., CHILDPREV [24]) to their data.

Another obstacle to a systematic assessment of CCS prevalence in Europe is the General Data Protection Regulation (GDPR) of the EU, introduced in 2018 [51]. Sharing individual records, needed to estimate prevalence by CHILDPREV [24] in international studies is currently impossible, or highly restricted to only few European countries. This serious impediment [52] to collaboration is particularly regrettable when rare conditions, such as cancer in childhood, require multi-national teams and databases. Information on prevalence of CCS in Europe is thus deferred to alternative methods or, hopefully, to a later time.

In conclusion, this review highlights the importance of CCS prevalence as a measure of cancer burden on one hand, and as an inventory of gaps in knowledge and data availability in Europe on the other. The occasionally published estimates are mostly neither systematic, nor comparable. The results of our review call for a consistent assessment of CCS prevalence, using standard and comparable parameters, and methodology. Such systematic evaluation of prevalence of CCS in Europe is one of the objectives of the CRICCS study (criccs.iarc.who.int), coordinated by the IARC and conducted in collaboration with cancer registries. A new method, estimating prevalence of CCS using aggregated data and named Prevalence of Childhood Cancer Survivors (POCCS) [53], is one contribution towards attaining this objective. Information on prevalence of CCS will increase awareness of the size of this growing population and facilitate planning of health care for their ongoing needs.

Disclaimer

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CRediT authorship contribution statement

Neimar de Paula Silva: Conceptualization, Methodology, Formal analysis, Investigation, Data Curation, Writing - Original Draft, Visualization **Andrea Gini:** Conceptualization, Methodology, Formal analysis, Investigation, Writing - Review & Editing **Anastasia Dolya:** Data Curation, Writing - Review & Editing **Murielle Colombet:** Data Curation, Writing - Review & Editing **Isabelle Soerjomataram:** Conceptualization, Writing - Review & Editing **Danny Youlden:** Conceptualization, Writing - Review & Editing **Charles Stiller:** Conceptualization, Writing - Review & Editing **Eva Steliarova-Foucher:** Conceptualization, Writing - Review & Editing, Supervision, Project administration, Funding acquisition **the CRICCS consortium:** Conceptualisation, Writing - Review & Editing

All authors approved the final version of the manuscript and agree to be accountable for all aspects of the work.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at doi:10.1016/j.ejcped.2024.100155.

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