Cancer Epidemiology 39 (2015) 440–446

Contents lists available at ScienceDirect

Cancer Epidemiology

The International Journal of Cancer Epidemiology, Detection, and Prevention

journal homepage: www.cancerepidemiology.net

Burden of soft-tissue and bone sarcoma in routine care Estimation of incidence, prevalence and survival for health services research

Freya Trautmann [a,*], Markus Schuler [b], Jochen Schmitt [a]

[a] University Hospital Carl Gustav Carus Dresden, Center for Evidence-Based Healthcare, Fetscherstraße 74, 01307 Dresden, Germany
[b] University Hospital Carl Gustav Carus Dresden, Medizinische Klinik und Poliklinik 1, Fetscherstraße 74, 01307 Dresden, Germany

ARTICLE INFO

Article history:
Received 13 September 2014
Received in revised form 19 February 2015
Accepted 3 March 2015
Available online 21 March 2015

Keywords:
Soft-tissue sarcoma
Bone sarcoma
Routine data
Incidence
Prevalence
Survival
Outcomes research
Health service research
Epidemiology

ABSTRACT

Background: Sarcomas constitute a rare group of malignant tumors which can originate from any organ, tissue, bone or cartilage. Due to their heterogeneity, estimates of sarcoma incidence, prevalence and survival are rare. We estimated the burden of sarcoma in Germany from a large unselected cohort of patients from routine healthcare.

Methods: We utilized the AOK PLUS health services research database covering complete medical information on 2,615,865 individuals from the German federal state of Saxony from 2005 to 2012. Persons were defined as sarcoma cases if they had ≥4 medical accounts with respective ICD-10 code C49 (soft-tissue sarcoma) or C40/C41 (bone sarcoma). We assessed sarcoma burden by calculating five-year prevalences, cumulative incidences, and one- and five-year relative survival rates.

Results: Overall 1,468 persons with soft-tissue sarcoma and 671 persons with bone sarcoma were identified. Age-standardized cumulative incidence was 4.5/100,000 persons for soft-tissue and 2.1/100,000 persons for bone sarcoma (European Standard). One- and five-year relative survival was 87.8% and 66.4% for soft-tissue and 91.8% and 52.9% for bone sarcoma, respectively.

Conclusion: This is the first estimation of the burden of sarcoma based on an unselected sample of routine care data and the first estimation of the burden of sarcoma in Germany. We believe that the proposed methods offer a valuable approach for further outcomes research on cancer.

1. Introduction

Sarcomas are a heterogeneous group of solid malignant tumors arising from mesenchymal cells. They can occur in almost any anatomic site and are defined by diverse histopathologies of the affected organ, tissue, bone or cartilage [1–3]. This rare entity accounts for approximately 1% of all malignant neoplasms in adults [4]. Due to their heterogeneity in terms of origin and histology estimates of sarcoma incidence, prevalence and survival are rare. A large epidemiologic study [1] recently estimated an incidence of soft-tissue and bone sarcoma of 4.9/100,000 and 0.8/100,000, respectively, in the EU27 countries based on data from 64 epidemiologic cancer registries. There was substantial regional variation in sarcoma incidence between Northern and Eastern Europe with higher incidence in Northern Europe. However, the completeness of the underlying registries is not completely clear so that these numbers may underestimate the true incidence [5–7].

Although epidemiological cancer registries are generally suitable to estimate the disease burden, they are unsuitable for health services research and outcomes research since no detailed information regarding treatments, medical doses, involved medical professions or comorbidities can be provided. Also cancer registries in Germany may be incomplete to a certain extent [6,8]. Therefore administrative healthcare databases are being increasingly recognized as a valuable resource for both, epidemiological research and for outcomes research [9,10]. Health insurance data provide large datasets which are free of recall and selection bias, low-cost and can be available for a relatively long time period.

Abbreviations: ICD-10-GM, International Classification of Diseases, 10th Revision, German Modification; SD, standard deviation; 95%CI, 95% confidence intervals; Prop, proportion.

* Corresponding author. Tel.: +49 3514584269.
E-mail addresses: freya.trautmann@uniklinikum-dresden.de (F. Trautmann), markus.schuler@uniklinikum-dresden.de (M. Schuler), jochen.schmitt@uniklinikum-dresden.de (J. Schmitt).

http://dx.doi.org/10.1016/j.canep.2015.03.002
1877-7821/

Because the data were gathered for billing purposes the provided information regarding pharmacological therapies as well as medical procedures can be considered complete. Therefore administrative healthcare data are suitable for descriptive analysis and health services research on rare diseases such as sarcoma for which information regarding course of disease and treatments as well as involved medical professions are missing [9].

Up to now no generalizable data on incidence, prevalence and medical care of sarcoma in Germany have been published so far. In general knowledge regarding medical care and treatment outcomes of sarcoma patients in Germany is very limited.

The aim of our study is to describe the burden of sarcoma in routine care by estimating incidence, prevalence as well as one- and five-year survival. We describe methodologies for identifying cases with soft-tissue and bone-sarcoma from outpatient data of a large German statutory health insurance.

2. Methods and materials

2.1. Data source

We conducted a retrospective cohort study based on data of the AOK PLUS, a large Saxon statutory health insurance covering approximately 60% of all inhabitants of the German federal state of Saxony. The data are available from 2005 to 2012 and include anonymized information regarding diagnosis, drug prescriptions and medical procedures of 2,615,865 insured persons from outpatient care. The study sample consisted of all individuals who were insured as of January 1st, 2005 until death or December 31st, 2012.

This study is based on the Declaration of Helsinki [11] and adheres to the principles of Good Epidemiological Practice and Good Practice in Secondary Data Analysis [12].

Case definition: One prerequisite to conduct outcomes research on cancer care based on administrative healthcare data is the adequate case definition. Patients with sarcoma were identified via ICD-10-GM codes (International Classification of Diseases, 10th Revision, German Modification [13]) using C40 and C41 for bone sarcoma and C49 for soft-tissue sarcoma. Internal case validation was based on respective diagnostic confidence. In Germany physicians working in the outpatient sector account their efforts by charging the patients' health insurance per patient visit per quarter. Billable performances and procedures have to be linked to at least one specific ICD-10 code, which has to be named on the account before it can be reimbursed by the insurance. According to the German Social Insurance Code, Volume V, §295 [14], physicians in the German statutory healthcare system have to indicate the degree of diagnostic confidence as "confirmed" (condition diagnosed and confirmed), "suspicion" (condition suspected but not confirmed), "exclusion" (condition excluded), or "aftercare" (condition confirmed in the past and patient treated for aftercare reasons) for each ICD-10 diagnosis [15].

Persons were identified as *prevalent* sarcoma cases if they fulfilled the following criteria:

- ≥4 accounts with ICD-10 code C49 (soft-tissue sarcoma) or C40/C41 (bone sarcoma) within the entire period 2005–2012, given that the last account had no diagnostic confidence "exclusion",
- ≥1 account including a diagnostic confidence of "confirmed" or "aftercare".

For *incident* sarcoma cases the following criteria were defined:

- ≥4 accounts with ICD-code C49 (soft-tissue sarcoma) or C40/C41 (bone sarcoma) within the entire period under observation, given that the last account had no diagnostic confidence "exclusion",
- ≥1 account with a diagnostic confidence of "confirmed", to exclude patients which were treated for aftercare reasons only,
- No respective medical account for sarcoma for at least two previous years.

The decision to use a minimum of four medical accounts as identification of sarcoma patients was based on the fact that patients are seeing several physicians during the process of diagnostic identification, treatment and aftercare in which each of them generates a separate medical account including ICD-10 code and diagnostic confidence. The typical course of diagnosis and treatment is depicted in Fig. 1. Treatments conducted in the inpatient sector (hospitals) are accounted by a different coding system and are therefore not included in the health services research database utilized for this study. The diagnostic process of sarcoma typically requires at least four medical outpatient visits.

To define incident cases, sarcoma-free lead-time was set to two years, since incidence rates remained stable when increasing the cancer-free lead time to three or more years.

2.2. Statistical analysis

Firstly, cumulative incidence and corresponding 95% confidence intervals (95%CI) for soft-tissue sarcoma and bone sarcoma were calculated for each year 2007 through 2011 separately. Secondly, the overall cumulative incidence with corresponding 95%CI for the period 2007–2011 was calculated as the weighted mean of the annual risks. For better comparability incidence was age-standardized according to the European Standard Population provided by Eurostat [16]. Cumulative incidence was calculated separately for the years 2007–2011. Respective calculations for the year 2012 were considerably lower compared to previous years for

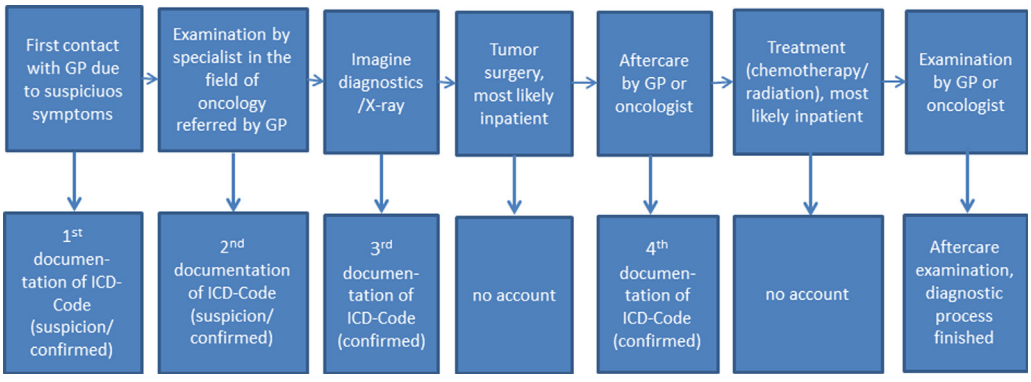

Fig. 1. Typical course of diagnosis and treatment for cancer patients in Germany.