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What do we know about survivorship after treatment for extremity sarcoma? A systematic review

T. Ng Kee Kwong [a], S. Furtado [b], C. Gerrand [c,*]

[a] University Hospital of North Tees, TS19 8PE, United Kingdom
[b] Department of Orthopaedics, Freeman Hospital, Newcastle-upon-Tyne NE7 7DN, United Kingdom
[c] North of England Bone and Soft Tissue Tumour Service, Department of Orthopaedics, Freeman Hospital, Newcastle-upon-Tyne NE7 7DN, United Kingdom

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Abstract

Objective: The varied presentations and treatments of extremity bone and soft tissue sarcoma mean that the issues faced by survivors are diverse and complex. The aim of this systematic review was to investigate what is known about this topic with a view to identifying areas for further research or service development.
Methods: This was a review of the English language literature identified from Medline and Ovid and hand searches published between January 2000 and September 2012. Results were compiled according to physical, psychological and social domains of survivorship.
Results: Of 182 studies identified, 22 met the inclusion criteria. There is a wide range of outcome measures used and a need for more objective measures. Unsurprisingly, survivors of extremity sarcoma typically demonstrate lower levels of physical functioning than healthy controls. In addition, survivors demonstrate a substantial psychological morbidity.
Conclusions: Services for survivors of extremity sarcoma should include rehabilitation and psychological support, sexual health services, expert pain management, and support to return to work.

Keywords: Sarcoma; Bone cancer; Soft tissue sarcoma; Survivorship; Extremity; Quality of life

Introduction

Sarcomas are a diverse group of rare cancers arising in connective tissue; they include bone sarcomas, such as osteosarcoma (OS) and Ewing's sarcoma (ES), and soft tissue sarcomas (STS), such as myxofibrosarcoma and liposarcoma. While the combined prevalence of bone and soft tissue sarcomas make them only the 21st most common type of cancer worldwide,[1] the need for multimodality treatment (ie chemotherapy, major surgery to an extremity and radiotherapy) and the young age of many patients means that the impact of these tumours on patients and society is significant but also highly variable.[2] Although 85% of patients with an extremity sarcoma can have limb sparing surgery, they are frequently left with permanent physical limitations, require long-term follow up and need further surgical intervention.

As survival has improved, an understanding of survivorship and the impact of treatment has become more important. 5-year survival rates for all types of bone cancer improved from 40% in 1979 to 54% in 1998[3] and remain around 59% for soft tissue sarcomas.[4] Bone sarcomas predominantly affect children and young adults whereas patients with soft tissue sarcomas tend to be older. There is therefore considerable variation in the survivorship experience and a need for a range of strategies to improve outcomes.[5] Furthermore sarcoma patients tend to differ from other cancer survivors because of the effect of limb surgery on appearance and physical functioning.[6] A recent systematic review of quality of life in survivors of adult extremity sarcoma found that patients undergoing limb sparing

* Corresponding author.
E-mail address: craig.gerrand@nuth.nhs.uk (C. Gerrand).

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surgery had superior physical functioning to those who had amputation, but quality of life scores comparable to the general population. However, the authors did not consider patients aged 15 years or younger, and focussed on quality of life outcomes rather than survivorship, which is a concept more allied to assessing the health needs of those who have been treated for cancer.[7]

The aim of this paper was to review the literature about survivorship after extremity bone and soft tissue sarcoma to establish what is known and to suggest topics for further research or service provision. Our specific objectives were to address the questions: (1) How do sarcoma survivors compare with the general population on measures of survivorship? (2) Do age and gender influence survivorship in extremity sarcoma patients?

Methods

This was a systematic review of the literature.

Identification of the literature and article selection

Databases used included Medline and Ovid. Basic searches were performed using the search terms: "sarcoma", "bone tumour", "cancer", "survivorship", "quality of life", "physical function", "pain", "sexual", "cognitive", "employment", "emotional", "psychological", "depression", "anxiety", "psychosocial" and "social". Survivorship-specific terms such as "quality of life", "employment" and "psychological" were combined with the terms "sarcoma", "cancer" and "bone tumours" with the Boolean operator "AND" to obtain more relevant results. Articles under the same Medical Subheading (MeSH) terms were also examined and appropriate studies selected. Furthermore, the reference lists of related papers were reviewed to identify articles. Limits were set for the date of publication (January 2000 to September 2012) and only English language and peer reviewed publications were included.

Because we were interested in how sarcoma patients compared to population norms, only papers reporting aspects of survivorship in extremity sarcoma patients after surgery and which compared sarcoma patients to population norms, control groups or reference values were selected. In keeping with other authors, patients were defined as survivors from the time of diagnosis.[8,9]

Data relating to the methodology of selected papers were collected and included study design, number of participants, assessments used and time period over which patients were assessed. The nine domains of survivorship defined by the National Cancer Research Institute (Fatigue and physical functioning; Pain; Sexual function; Cognitive functioning; Employment, finance and return to work; Emotional distress; Depression; Anxiety; and Social needs) were combined into a biopsychosocial model containing three domains for this analysis. These were: physical (fatigue, physical functioning and pain); psychological (emotional distress, cognitive functioning, depression and anxiety); and social (sexual function, employment and social needs).[10]

Methodological quality assessment

Quality was assessed using a checklist adapted from Mols et al.[11], Borghouts et al.[12] and Kuijpers et al.[13] The checklist included items for study design, response rate, comparison groups and participant information and papers were scored against this (Table 1). The maximum score achievable was 13: a score of 10 or more was used to define a study with a high quality methodology (Table 2).

Results

Initially, 158 papers were identified: a further 24 were selected after hand searching reference lists. Of these 182 papers, 142 did not meet the selection criteria. The remaining 40 were fully reviewed. 18 of these were rejected either because they lacked comparison group/reference values or there was inadequate representation of extremity sarcoma survivors (Fig. 1).

The final 22 papers comprised fifteen cross-sectional, three longitudinal and four cohort studies.[2,14–34] Although there were no control groups in the cross-sectional or longitudinal studies, previously published norms were used to draw comparisons. In contrast, two of the cohort studies from the Childhood Cancer Survivorship Study (CCSS) used siblings as controls.[25,31] The remaining cohort study used age-matched controls from the general population.[23]

Of the 22 papers, seventeen examined quality of life and/or physical functioning,[2,16–22,24,26–30,32–34] two focused on health status and late effects (including organ function and symptom complaints),[14,23] and the remaining three explored sexual function,[15] level of physical activity[31] and education, employment, insurance and marital status.[25] Three studies used clinical assessments,[14,20,23] the

Table 1
Criteria for assessing quality of studies.

A.Prospective study design (also positive in studies where previously unknown outcomes are measured in a historical cohort)
B.Types of sarcoma are described
C.Socio-demographic data are provided
D.Participant inclusion and exclusion criteria outlined
E.Data collection process described
F.Type of sarcoma treatment mentioned
G.Presence of a control group (no score was given when population norms were used in the absence of a comparison group)
H.Time since diagnosis mentioned
I.Participation rate (positive if participation rate was greater than 75%)
J.Use of a standardised and valid assessment tool
K.Use of mean, median, standard deviation or percentage for outcomes
L.Explicit mention that consent form was signed by patient
M.Analysis techniques (positive if correlation or significance are tested using appropriate tests)