



REVIEW

The psychological impact of sarcoma on affected patients

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Abstract

Objective: Sarcoma diagnosis and its treatment trajectory may deeply affect the somatopsychic balance of patients and their caregivers. This systematic review aimed at deepening the understanding of sarcoma's impact on the entire family unit involved in the illness experience on a physical (e.g. fatigue), psychological (e.g. mental health, affective regulation, defense mechanisms), and interpersonal (e.g. social isolation, loneliness) level.

Methods: The systematic review was conducted using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses. The literature search led to the identification and subsequent inclusion of 44 articles focused on sarcoma patients. Results were classified into seven categories: Quality of Life, worries and distress, anxiety and depression, suicide ideation, financial and occupational consequences, unmet needs, and coping strategies. Our search identified only one study focusing on informal caregivers, thus we could not perform a systematic review on these results.

Results: Our findings underlined the traumatic impact of the sarcoma diagnosis. Patients can experience an impoverished emotional life, somatization, social withdrawal, difficulty in decision-making, increased feelings of discouragement and demoralization, and profound experiences of helplessness and vulnerability. Moreover, they seemed to display anxiety and depression and might present a higher suicide incidence than the general population.

Conclusion: Our review highlighted that the psychosocial aftermath of sarcoma patients should guide institutions and healthcare professionals toward the design of assessment and intervention models that could contemplate the different dimensions of their suffering. Furthermore, it points out that there is still a lack of evidence regarding the psychosocial impact affecting sarcoma patients' caregivers.

KEYWORDS

cancer, oncology, psycho-oncology, psychosocial issues, quality of life, sarcoma, unmet needs

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1 | INTRODUCTION

Cancer represents a complex and difficult experience for patients, their families, and the health service organizations responsible for providing treatment and care.¹ Both oncological disease and its treatment exert a deep impact on patients' life: literature shows Quality of Life (QoL) impairment,^{2–4} anxious and depressive symptoms^{5,6} and preoccupation about the somatic Self.^{7,8} The traumatic experience of receiving a diagnosis of cancer and having to face treatment can also cause fears about the recurrence of the disease^{9,10} and shatter the subject's ability to make sense of the event, impairing those functions of the mind that are fundamental to giving meaning to life.¹¹

Cancer produces a conspicuous impact also on informal caregivers, whose tasks involve both a "practical" dimension concerning medical appointments and management of side-effects of treatments, and an "emotional" dimension, which entails the relationship with the patient and other family members.^{12,13} Frequently, caregivers' psychological distress is similar to the one experienced by patients.¹⁴ In addition to the social, health, and caregiving roles attributed to them, caregivers are exposed to intense emotions that are difficult to regulate and can last over time: they are often burdened with the responsibility of "thinking for the patient" and may have trouble handling their personal needs and developing a psychological representation of their experience and anticipatory fears (e.g., what is happening to them and what they are going to face in the course of the disease). Often, caregivers are faced with a traumatic reality in which their own need for support often remains undetected.¹²

Both during and after cancer treatments, caregivers may experience pain, fatigue, interrupted sleep and appetite, financial difficulties, work problems, isolation, anxiety, depression, fear, and avoidance tendencies.^{14–16}

The experience of cancer can affect both patients' and caregivers' physiological balance and their ability to regulate and symbolize their emotional experiences, which can be experienced at a bodily level, leading to mentalization deficits.^{11,17,18}

Considering these aspects is important, particularly for patients with rare forms of cancer. These patients are more often confronted with delayed or false diagnoses and late (or no) referral to a Center of Expertise and report higher levels of distress, insecurity, loneliness, social isolation, anxiety, and overall lower QoL compared to patients with more common forms of cancer.^{19,20} Rare cancer represent 1% of all malignancies.²¹ This heterogeneous malignancy tumor accounts for 1% of adult and 15% of pediatric tumors and predominantly (80%) affects soft tissues and only one in five affects the bones.²² Although multidisciplinary approaches in the care of sarcoma patients have led to a great improvement in oncological and functional outcomes, the prognosis of metastatic and refractory sarcomas remains unfavorable as the median survival is only 12–18 months.²³ Furthermore, the occurrence of nonspecific symptoms can cause delayed diagnosis and care resulting in a poorer prognosis.²⁴

Sarcoma and its treatment have physical side effects such as disfigurement and functional impairment, which can be disabling.^{25,26}

Furthermore, sarcoma has a conspicuous psychosocial impact on patients. Patients can suffer from anxiety, depression, impaired QoL, relational problems, and financial strain^{2,27}; undergo consistent physical changes which alter body image, leading to emotional distress²⁸; and have unmet needs such as the desire for more information and social support.²⁴ However, evidence on psychosocial distress in sarcoma patients can be quite heterogeneous, probably due to the influence of other variables, such as levels of impairment according to the phase of the disease, age, sex, and sarcoma type.²⁷

Caregivers of sarcoma patients can suffer from a high caregiver burden.²⁹ They may feel guilty about under-estimating the patients' symptoms, believing this may have contributed to the delay in diagnosis.³⁰ Caregivers may also worry about the urgency of the prescribed treatment^{24,29}; and the unexpected responsibility to provide caregiving tasks that will require substantial emotional and physical resources. In addition, the risks of caregivers' burden often go unnoticed because of their reluctance to seek support.²⁴

Thus, the purpose of this study is to present the results of a systematic review of published research focused on sarcoma's impact on both patients and caregivers involved in the illness experience on a physical (e.g. fatigue), psychological (e.g. mental health, affective regulation, defense mechanisms), and interpersonal (e.g. social isolation, loneliness) level.

2 | MATERIALS AND METHODS

The systematic review was conducted using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) 2020 guidelines³¹ and was registered in the International prospective register of systematic reviews PROSPERO (Registration number CRD42021249511), available at https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42021249511.

2.1 | Search strategies

Studies were identified by searching the following electronic databases: the Cochrane Library, Google Scholar, PsycInfo and PsycArticles, PubMed, Scopus, and Web of Science. We used a combination of the following keywords: "sarcoma" AND psych* OR mental* OR depress* OR anxi* OR alexithymi* OR distress OR emoti* OR bereave* OR grief OR coping OR "defense mechanism*" OR burden OR fatigue OR "social isolation" OR lonel* OR relationship* OR "quality of life" AND patient* OR inpatient* OR outpatient* OR caregiv* OR caretak* OR carer* OR famil* OR spous* OR partner* OR relative* OR parent* OR offspring* OR sibling*. Keywords were searched into (1) abstract for what concerns the Cochrane Library and Scopus; (2) topic for what concerns Web of Science; (3) all fields for what concerns all the other databases. To collect the most recent data, we chose to include only journal articles published since January 2010. Moreover, since we do not have staff who can review papers in all European languages, we decided for consistency to

include only those written in English. This limitation could reduce the number of research studies and data to corroborate the reported data. Articles were retrieved on 8 May 2021, and a new search was run on 5 July 2022 to update results.

2.2 | Selection criteria

The progressive exclusion was performed by the three authors of this paper (CG, MDS, IV) who read the title, the abstract, and the full text. In case of disagreement, a fourth author (IGF) was consulted. The inclusion criteria were:

- 1) Quantitative original research (e.g., cohort, case-control, cross-sectional, or longitudinal studies).
- 2) Research studies with explicit references to the psychological impact of sarcoma on affected patients and their informal caregivers.
- 3) Research on the adult population.
- 4) Articles limited to the English language.
- 5) Publications within the given time interval (2010–2022).

The exclusion criteria were:

- 1) Qualitative studies.
- 2) Studies not reporting original results (reviews, letters, editorials, and comments).
- 3) Studies on children and adolescents.
- 4) Dissertations.
- 5) Studies on animals.

If the reviewed articles focused on mixed samples, results were included only if data on sarcoma patients and caregivers were reported separately.

Any discrepancy concerning the inclusion/exclusion of articles was discussed by the four researchers until an agreement was reached. A list of excluded studies, including the level and reasons for exclusion, was maintained. The studies identified in this step underwent the same screening process as the papers retrieved by the database search. The entire procedure is displayed in Figure 1.

2.3 | Data analysis

Data analysis was conducted using a standardized data extraction form and included (a) general study details (e.g., authors, title, publication source, year of publication); (b) type of study; (c) sample characteristics (e.g., age, gender, country, patients vs. caregivers); (d) measures; and (e) results.

All papers included in the systematic review were subjected to rigorous appraisal using the Critical Appraisal Checklist for Case Control, Cohort, and Cross-Sectional studies.³² The Joanna Briggs Institute offers well-established reliability and validity tests to assess

the risk of bias in studies. Three authors of this paper (CG, MDS, IV) independently evaluated the quality of each study. In case of disagreement, a fourth author (IGF) was consulted. Articles were evaluated based on the following criteria: LOW risk of bias studies with more than 70% “yes” score; MODERATE risk of bias studies with 50%–69% “yes” score; and HIGH risk of bias studies with less than 49% “yes” score. As recommended by the JBI reviewers' manual, all decisions regarding the scoring system and cut-off points were approved by all reviewers before the start of the critical appraisal process.

3 | RESULTS

The electronic database search identified 13,241 articles. After duplicates were removed, 3762 articles were identified. Of these, 3603 articles were excluded based on title and abstract, and 114 articles were excluded based on full-text evaluation (Figure 1).

The 45 remaining articles from the electronic literature search underwent data extraction and qualitative analysis. Articles were mainly focused on sarcoma patients (44 papers), and only 1 article was focused on caregivers, thus we could not include data on caregivers in our final review. Articles focused on sarcoma patients were classified into seven categories: (1) QoL, (2) worries and distress, (3) anxiety and depression, (4) suicide, (5) financial and occupational consequences, (6) unmet needs, and (7) coping strategies. Table S1 summarizes the results. The assessment of the quality of the included articles is shown in Table S2. Evaluation using The Joanna Briggs Institute checklist found that all articles received an acceptable quality appraisal for inclusion in the current review. Of the 34 evaluated cross-sectional studies, 24 had a low risk of bias, nine had a moderate risk of bias, and one had a high risk of bias. Of the eight analyzed cohort studies, five articles had a moderate risk of bias scores and three articles had a high risk of bias scores. Out of the three assessed case-control studies, one exhibited a low risk of bias, and two articles exhibited a moderate risk of bias.

4 | SARCOMA PATIENTS

4.1 | Quality of life

QoL is a well-explored aspect of the experience of illness in sarcoma patients. Some studies show that the QoL of sarcoma patients appears to be stable.^{2,33–38} However, other research found more compromised levels of QoL.³⁹

Sarcoma patients experience lower QoL compared to the general population,^{2,4,33,35,40} in different age groups,⁴¹ even if other research detected poorer physical functioning but fewer symptoms such as fatigue and sleep disturbances in sarcoma patients than in the normative population.⁴² Moreover, in a cohort of epithelioid hemangioendothelioma patients, the group affected by a high

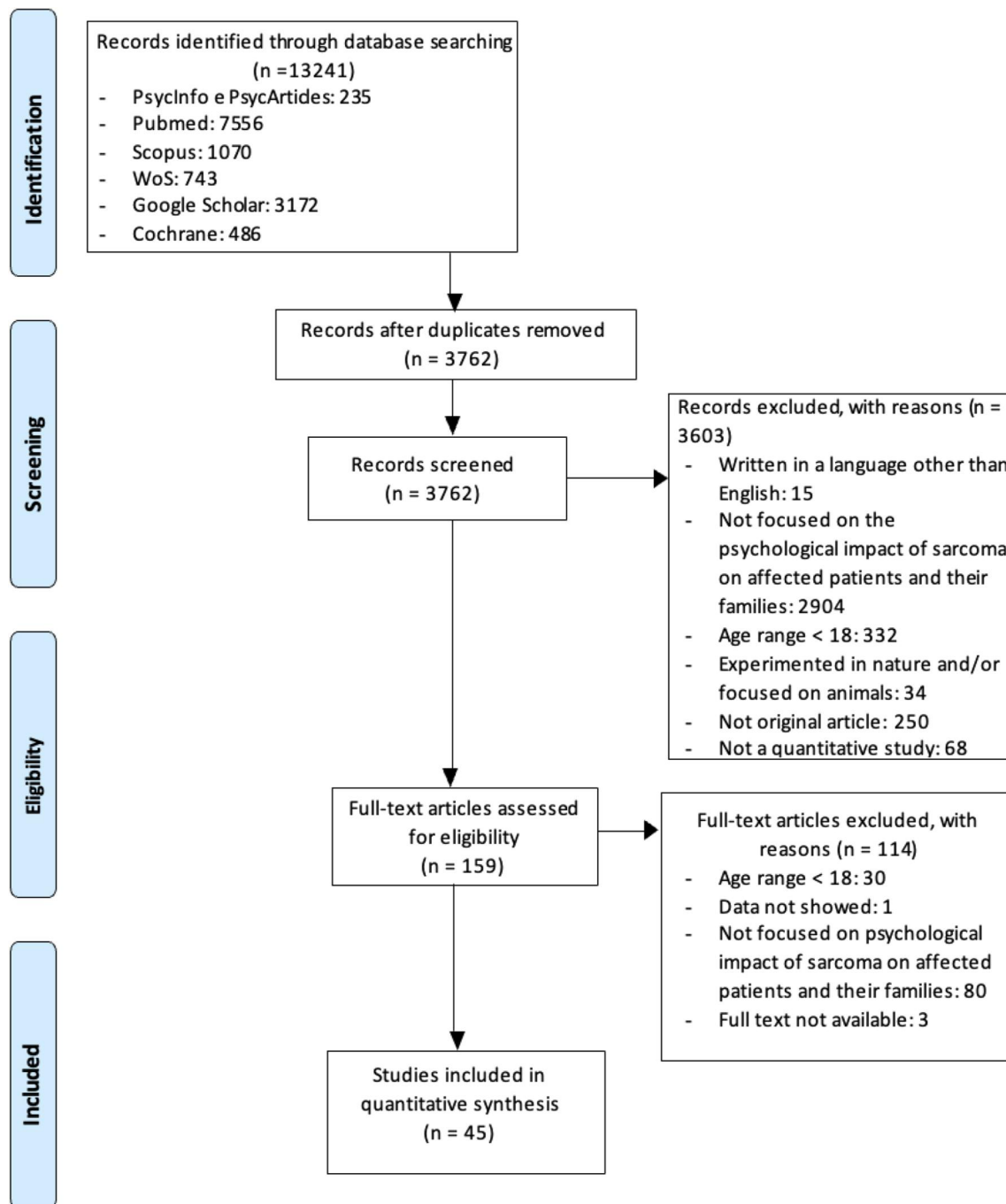


FIGURE 1 Preferred reporting items for systematic reviews illustrates the study selection process.

symptom burden had a lower QoL in comparison to the general population; patients with a low symptoms burden had a lower QoL than the general population; patients affected by an intermediate symptom burden reported a similar level of QoL compared to the general population.⁴³ Sarcoma patients also show lower social well-being than patients with other malignancies.⁴⁴ Young adults with sarcoma had higher levels of treatment-related distress compared with young adults with breast and hematological cancer. Moreover, their QoL was lower than that of patients with testicular cancer, but better than that of patients with gynecologic tumors.⁴⁵ However,

another study revealed a lower symptom burden in sarcoma patients compared with patients affected by skin cancer.⁴⁶

Variables such as gender, age, other demographic variables, sarcoma type, surgical procedure, site of cancer, or phase of the disease may also influence QoL and lead to different levels of impairment in sarcoma patients.^{35,47–50} The symptoms most reported as disabling were pain^{2,39,43,51,52} and fatigue.^{2,36,43,52}

Survivors of skin sarcomas had the highest scores on global health status, physical functioning, and social functioning; survivors with sarcoma of the axial skeleton reported impairment on all functioning

scales except for emotional and cognitive functioning and had the highest scores on the symptom scales except for nausea/vomiting, dyspnea, diarrhea, and financial impact. Emotional functioning and dyspnea seem particularly impaired in patients with breast sarcoma, which was the only sarcoma showing levels of pain comparable to those of axial skeleton sarcoma.⁵³ Patients with upper extremity sarcomas reported higher QoL than patients with lower-extremity sarcomas.^{2,50,54} Moreover, patients with extremity tumors scored higher in global QoL and cognitive functioning and lower in physical functioning compared to patients with non-extremity tumors.⁵⁵

Surgically treated patients showed overall good QoL concerning physical functioning in activities of daily living.^{34,35} Amputation seems to be associated with poorer QoL outcomes,^{42,50,54} whereas patients treated with limb-sparing surgery had good QoL.³⁷ Patients with more proximal amputations had higher levels of impairment than those with below-knee or minor amputations, particularly concerning daily activities and sexual activity.³⁹ Moreover, wound complications increase the risk of impaired physical functioning and anxiety.⁵⁶ Non-surgical treatments and radiotherapy predicted good social functioning during treatment,⁴ whereas chemotherapy was associated with several impairment outcomes in older patients.⁴¹

QoL seems to improve as the time from the beginning of the treatment and/or surgery increases.^{42,57,58} Patients who had not received treatment or who had already completed treatment had higher overall QoL compared to patients currently in treatment³³ or who had recently been treated with surgery.⁵⁶ However, a longitudinal study showed that elderly sarcoma patients (>50 years) did not return to QoL baseline values even after the last follow-up visit.⁴⁸

The diagnostic phase, treatment phase, and palliative treatment seem to have different impacts based on the aspect of QoL measured. For example, in patients with indications for chemotherapy or radiotherapy, global QoL was higher in the treatment phase than in the diagnostic phase, whereas physical functioning and fatigue showed better outcomes in the diagnostic phase.^{4,51} Results on pain are diverging. A study found that pain was higher in the diagnostic phase compared to the treatment phase,⁴ whereas another research found that pain was higher during chemotherapy.⁵¹ Patients receiving palliative care seem to have a higher symptom burden,^{2,33,51} even if they do better in social functioning, physical functioning, and role functioning.²

Concerning gender differences, females seem to have poorer QoL outcomes than males,^{2,55,57,59} but other studies show different results.^{36,44} No differences emerged for what concerned pain.⁶⁰

Several studies found lower levels of overall QoL in older patients.^{4,34,36,39,41,56,57,59,61–63} On the other hand, pain⁶³ and social functioning⁶⁴ seem poorer in younger patients who showed remarkably poorer physical functioning, role functioning, emotional functioning, and cognitive functioning.⁴¹

Higher socioeconomic status is related to higher physical functioning.⁵⁵ Regarding the impact of other socio-economic aspects on QoL, sarcoma patients who still had their job showed higher QoL than non-working patients.^{4,35,59} Retired patients showed better functioning compared to non-retired patients, although they may still

feel excluded from the social environment compared to working patients.⁵⁹ Married patients not only show poorer QoL outcomes,^{4,48} but they also show a lower reduction in psychological distress over time than single patients.⁴⁸

For what concerns the impact of health-related QoL on 1-year survival, global health had the biggest impact. Functioning scales (except for cognitive functioning), fatigue, appetite loss, pain nausea/vomiting, dyspnea, constipation, and financial difficulties were significant prognostic factors for survival.⁶⁵

4.2 | Worries and distress

Psychological distress was identified in 32% of patients affected by malignant bone and soft tissue tumors. Sarcoma types that were more associated with the presence of distress were osteosarcoma (50%) and chondrosarcoma (40%), followed by undifferentiated sarcoma (32%) and liposarcoma (27%).⁶⁶

Higher distress seems to be experienced by females.^{47,66} Concerning age, while the distress level of younger patients seems to decrease over time, it remains stable in elderly patients, even if they seem less affected by occupational worries.^{48,67}

The diagnosis seems to be the most burdensome and stressful stage of the disease,^{48,67} but surgery and chemotherapy are also sources of distress.^{47,59,66}

The comparison between distress in sarcoma patients and other cancer patients showed contrasting results.^{51,68}

4.3 | Anxiety and depression

Anxiety varied from 21.3% to 100% in a sample of patients who had undergone secondary amputation,⁶ while depression varied from 6.6% to 88% in a sample of patients who had undergone extended resection.⁵⁴

Anxiety seems to reach higher levels in the early phases of the disease.⁶ Similarly, the anxiety score decreased over time in patients with surgically treated soft tissue sarcoma^{37,56} and in patients with retroperitoneal sarcoma who had been treated with radiotherapy.⁵⁷

Contrary to anxiety, depression showed the highest prevalence during the treatment phase, followed by the diagnostic and follow-up phases. However, a longer period since the completion of treatment was correlated with fewer depressive symptoms.⁶

In a sample of sarcoma patients in the treatment phase, only female patients had an increased depression score, while older sarcoma patients showed higher levels of depression and poorer adjustment during the follow-up phase.⁶ Consistently, in patients with retroperitoneal sarcoma, age was positively correlated with high depression and anxiety scores.⁵⁷ Moreover, in a sample of young sarcoma patients, they showed a higher risk for antidepressant purchases compared to their sibling controls.⁶⁹

Among sarcoma survivors who had undergone surgery, both the patients who received limb-sparing surgery and patients who had an

amputation showed similar levels of anxiety and depression.⁴² Patients with major wound complications reported higher anxiety than patients without wound complications.⁵⁶ Interestingly, patients treated with surgery reported depression levels comparable to that of the normative population³⁵; limb-salvage patients reported a lower level of depression than the US general population.^{42,50} Another research found that sarcoma patients showed higher levels of depression and lower levels of anxiety compared to patients with common cancers, even if such results seemed influenced by gender: in females, sarcoma patients showed lower anxiety than other oncological patients, while males showed a slightly opposite trend.⁶¹

Anxiety and depression levels seem to account for the differences in functional outcome and overall QoL⁵⁰ and appear to be related to higher levels of symptom burden.⁴³ Coping strategies (e.g., denial and use of humor) used by sarcoma patients explained 60.5% of the variance in anxiety and 47.2% of the variance in depression.⁷⁰

4.4 | Suicide

Sarcoma patients showed an overall higher suicide incidence than the general population, but this outcome appears to be influenced by other variables such as marital status, ethnicity, gender, and the length of time since diagnosis. Sarcoma patients showed a higher prevalence of suicide than the US general population,^{71,72} even if suicide incidence was significantly higher in sarcoma patients only within the first 5 years of a cancer diagnosis.⁷² Suicide was higher in sarcoma patients who are single, divorced, separated, or widowed, white, and male than in the general population.^{71,72}

African-American patients showed lower risks of suicide than white American patients, while older patients displayed a lower incidence of suicide than younger patients,⁷¹ even if in another study sarcoma patients aged 21–30 years had the highest incidence of suicide, followed by patients aged 61–70.⁷²

The highest incidence of suicide was recorded among patients with sarcoma of the vertebral column, sarcoma of the pelvic bones, and sarcoma of the spinal cord.⁷² Studies that compared suicide rates in different phases of the disease showed contrasting results. A study found that patients who have been diagnosed less than a year ago showed a lower risk of suicide,⁷¹ while another study found that suicide incidence in sarcoma patients was only significantly higher than in the US general population within the first 5 years of a cancer diagnosis.⁷²

Concerning treatment, chemotherapy seems associated with a lower risk of suicide,⁷¹ whereas patients who received neither radiation nor surgery had the highest suicide incidence of all other patients with known treatment regimens.⁷²

4.5 | Financial and occupational consequences

The type of surgery performed (limb-sparing vs. amputation) appears to have a significant impact on re-employment.⁵⁰ Most of the

surgically treated sarcoma patients were able to return to work.^{35,59} Patients who returned to work after receiving a diagnosis of sarcoma were younger and had a lower proportion of high tumor grades than those who did not.³⁵

Sarcoma patients showed statistically higher financial difficulties than the general population.⁷³ A longer time frame between diagnosis and remission seemed to be a protective factor against the financial toxicity of cancer (e.g., payments and income losses), and the material consequences and psychological effects of the financial burden.⁷³ Moreover, sarcoma patients aged between 40 and 52.5 years had higher odds to report a financial burden compared to younger (18–27.5 years) and older patients (52.5–65 years).³⁵ Research showed that a higher level of education and a higher income also reduced the odds of financial strain among sarcoma patients. Conversely, patients receiving a disability pension and currently on sick leave suffered from higher financial toxicity.⁷³

4.6 | Unmet needs

Sarcoma patients reported a need for more psychological support.^{47,55,59} Moreover, they underlined insufficient medical guidance (32%) and a desire for contact with fellow survivors (27%).⁷⁴ Adolescents (18+) and young adults reported more unmet needs than older adults (40–69 years old) and elderly patients (>70 years old). They also reported more difficulty in getting the final diagnosis, complained more about a lack of non-medical guidance, and indicated a desire for contact with fellow survivors.⁷⁴

4.7 | Coping strategies

Sarcoma patients who underwent amputation reported a major attitude of resigned passivity to events in comparison to patients who underwent limb-sparing surgery.⁵⁰ Sarcoma survivors with a lower global QoL score showed maladaptive coping strategies, reporting helplessness and a difficulty in accepting their illness and in perceiving it as a possible opportunity of growth.⁵⁵ Furthermore, differences in coping strategies used to face cancer (fatalism, fighting spirit, anxious preoccupation, helpless-hopelessness, positive avoidance) seemed influenced by anxiety, depression, and functional outcomes.⁵⁰

5 | DISCUSSION

The results of this systematic review confirm that cancer can disrupt patients' somatopsychic balance. Sarcoma patients may display impaired QoL,^{2,4,33,35,40,44} as well as high levels of distress and worries.^{47,51,59,66} They seem to display anxiety and depression,^{6,52,54,61} and might present a higher suicide incidence than the general population.^{71,72} Moreover, that can resort to maladaptive coping strategies to face the experience of the disease.⁵⁰

Situations that represent a threat to somatopsychic integrity, such as an oncological disease, may cause the loss of several aspects of the Self and alterations in a sense of personal coherence. They need to be elaborated through the process of grief. Nevertheless, when patients and caregivers lack the psychological resources that sustain the representation and symbolization of the affects, they are not able to process their own experience. The reality of the disease, where suffering cannot be represented, may lead to an impoverished emotional life, somatization, social withdrawal, difficulty in decision-making, increased feelings of discouragement and demoralization, and profound experiences of helplessness and vulnerability. People may no longer be able to access their internal resources, which are frozen and dissociated from the impact of trauma. Patients and family members can experience the feeling of having failed in one's life story and having insufficient resources to cope with the circumstances of everyday life.^{11,75}

Our review sheds light on the fact that sarcoma patients often report unmet psychological and social needs, in particular the desire for more psychological support^{47,55,59} and for contact with other survivors.⁷⁴ Cancer patients may feel the urge to meet other people living the same experience and search for group occasions where they can share their feelings and reactions, feel their emotions mirrored, and build new meanings.^{76,77} This need becomes even more meaningful and specific in sarcoma patients due to the rarity of the disease and the difficulty in finding and meeting with other patients.²⁶

Our review highlights that sarcoma patients can experience different degrees of psychosocial impairment and confirms that other variables, such as the type of treatment, the phase of the disease, the sarcoma type, patients' gender, age, and other demographic variables, should be considered when measuring the levels of impairment.²⁹

For instance, surgically treated patients, especially those who underwent limb-sparing surgery,³⁷ show good overall QoL, even managing to return to work.^{34,35,59} On the other hand, although surgery and chemotherapy are considered elective treatment regimens, they can represent a source of distress^{47,59,66} and QoL impairment.⁴¹ In particular, undergoing an amputation seems to be a risk factor for maladaptive coping strategies and poorer QoL.⁵⁰

Despite the potential impairment caused by treatment, patients who received neither radiation nor surgery show a higher incidence of suicide than all other patients with known treatment regimens.⁷² This result might be interpreted as follows: patients who do not undergo treatment have a poorer prognosis, which can lead to overwhelming death anxieties that cannot be represented and regulated^{11,75,78} and might result in higher rates of suicide. It would be interesting to investigate the psychological impact of different treatments in future studies. For example, undergoing radiotherapy or chemotherapy may lead to different psychological consequences for cancer patients and can result in different fears and fantasies.

Psychosocial outcomes (anxiety and depression, distress, QoL, financial toxicity) of sarcoma patients appear to improve with time from the diagnostic phase,^{4,33,42,51,57,58,67,73} similar to the outcomes

for other cancer populations.¹⁹ Anxiety appears to be higher in the diagnostic phase, coherently with results obtained in patients affected by other cancers,⁷⁹ whereas depression shows higher rates during the treatment phase,⁶ as confirmed by other research.²⁷

We could assume that soon after the diagnosis when the patient starts to feel a threat to his/her own life, affects such fear and anxiety prevail, eliciting the use of defense mechanisms such as splitting and dissociation. Thus, affects can be perceived and experienced only at the bodily level, and the subject cannot symbolize and integrate them into a coherent discourse. Moreover, dissociation prevents the traumatic experience from being processed and may contribute to the development of depressive symptoms.^{11,75,77}

Concerning gender, female patients show higher level of depression,⁶ more distress,⁴⁷ and lower QoL.^{33,55,57,59} However, some contrasting evidence indicates that male sarcoma patients can suffer from a greater psychological impact⁴⁴ and symptoms.³⁶ Moreover, male patients show a higher suicide risk.^{71,72} These results are worthy of further investigation. A possible interpretation of this outcome relies on the different ways men and women acknowledge and handle their emotional suffering. Men are thought to regulate their emotions by suppressing them, so that their distress remains more hampered than women's distress and may lead to suicidal behavior.⁸⁰ On the contrary, female patients are freer in expressing their emotions, even the most dysregulated ones, but may suffer more from psychosocial issues, particularly those connected to an impairment of body image, which is essential in the perception of female identity.^{8,30,53}

Although older age in sarcoma patients seems to be associated with poorer psychosocial outcomes,^{4,34,36,39,41,48,57,61-63,69} adolescent and young adults show higher impairment in social and emotional functioning,^{64,67,74} higher rates of suicide,⁷¹ and a higher number of unmet needs.⁷⁴ Suicidal ideation in young adults is complex and multidimensional. Regression toward more primitive modes of thought, defenses, and modes of relating is common during this phase.⁸¹ As the body plays an important role in the evolution of one's identity, developing an organic disease such as sarcoma at a young age might seriously affect the perception of the self and compromise the capability of mentalizing the necessity to live for life with a modified body and a compromised state of health and to deal with limitations connected to the perspective of death.¹³

Sarcoma can variously affect patients depending on the type of tumor. Patients receiving diagnoses of axial skeleton sarcoma,⁵³ bone sarcoma,⁴¹ and non-extremity sarcomas⁵⁵ report higher psychosocial impairment and have poorer prognosis, more complicated resections, and more invalidating symptoms. Receiving a poor prognosis might disrupt the possibility of creating meanings and the patient can start living a routine that becomes less and less connected to his affective life, with a resulting inability to invest in his/her existence and relationships.^{11,75} Breast sarcoma patients report substantial emotional problems, supposedly because breast localization involves specific body image issues that increase emotional burden.⁵³

Furthermore, being single appears to be a protective factor for QoL^{4,48} and psychological distress.⁴⁸ This result might be related to the

interpersonal problems that frequently occur between cancer patients and their partners, which could enhance psychological distress.^{14,77}

The fact that African-American sarcoma patients show a lower risk of suicide⁷¹ may be related to religious and cultural influences. Religion is deeply embedded in African-American culture and can be considered a protective factor against mental health problems, because relying on religion can help patients to give meaning to their experience from a transcendent perspective, increasing patients' hope and the ability to tolerate the difficulties induced by cancer.⁸²

Although sarcoma patients display lower psychosocial outcomes than the general population,^{2,4,40,41,61,71,72} two studies highlight the presence of comparable or lower levels of depression in sarcoma patients than in general population.^{35,42} However, a potential explanation may be connected to patients' illness phase and their altered expectations they are thought to be cancer-free.^{35,42}

Another research revealed poorer social well-being in the sarcoma sample compared to other cancers.⁴⁴ This outcome is consistent with the social aftermath of sarcoma, whose esthetic impact can deeply affect patients' QoL and social behavior. As self-esteem sources such as appearance, independence, and social roles may be compromised, cancer can trigger narcissistic vulnerabilities and lead to social isolation because of patients' feelings of shame and desire to avoid pity and compassion from other people. Furthermore, the affective dysregulation characterizing the experience of cancer might induce or exacerbate mentalization deficits, undermining supportive interpersonal relationships.^{11,14,77}

For what concerns caregivers, it is extremely relevant and alarming that we could not perform a systematic review, since we could find only one paper.¹⁶ Indeed, caregivers of patients with rare tumors such as sarcoma present a higher risk of developing caregiver burden when compared to other cancers.³⁰ Furthermore, they can be reluctant to seek support because they worry about stealing time from their loved one's care, and their suffering risks going unnoticed.²⁴ Distress and suffering related to the caregiving experience, especially when they are not addressed, can also negatively affect the grief process that caregivers have to face after the patient's death.^{11-14,75,77}

6 | LIMITATIONS

This review has several limitations. Results may be affected by the heterogeneity of the instruments employed and by the exclusion of qualitative studies, which might have resulted in our bypassing significant outcomes. It is also plausible that many important results may have been missed, particularly because this study only considered articles in English, allegedly restricting the inclusion of significant outcomes. Moreover, although the importance of caregivers' well-being is fully recognized, our review points out that there is still a paucity of data. We strongly suggest deepening the understanding of the psychological and social burden of this specific caregiver population to provide them with targeted interventions that could support them during the illness trajectory.

7 | CLINICAL IMPLICATIONS

Our review sheds light on the impact of sarcoma on patients' somatopsychic balance. The physical, psychological, and social aftermath of sarcoma patients should guide institutions and health-care professionals to designing assessment and intervention models that measure (a) the different facets of patients' suffering and (b) the influence of variables such as gender, age, other demographic variables, sarcoma type, surgical procedure, site of cancer, and phase of the disease. From this perspective, patients' and caregivers' needs must be screened and addressed by a multidisciplinary team, so that each professional expert can handle a specific aspect of patient treatment and guarantee integrated care.

The psychological impact of diagnosis, at various stages of the disease, can have significant repercussions in terms of treatment adherence and health care costs. To cope with this complexity, health services need to move toward the implementation of multidisciplinary care protocols that pay adequate attention to the interrelationship between the bodily, psychological, and environmental components of the experience of illness and loss.^{11,75,77}

8 | CONCLUSIONS

This systematic review confirms that the cancer experience involves the physical, psychological, and social dimensions of life, and that it is important to consider a wide number of these dimension when exploring the experience of sarcoma patients and their caregivers, without focusing solely on QoL, but rather relating it to other factors, which may account for the discrepant results that emerge.^{1-10,27} In particular, our results highlight the need to investigate the experience of caregivers, considering the lack of research on this important issue. Sarcoma potentially exposes caregivers to several losses such as loss of autonomy, health, vitality, and personal time. We believe structuring targeted and multidisciplinary interventions can help both patients and caregivers improve their psychophysiological balance and enhance their ability to regulate and symbolize emotions and mentalize experience.

AUTHOR CONTRIBUTIONS

Isabella Giulia Franzoi, Alessandro Comandone, Marco Gonella and Antonella Granieri conceived and designed the study; Maria Domenica Sauta, Carola Grimaldi and Irene Vallauri screened the articles and analyzed the data; Isabella Giulia Franzoi participated in the exclusion process and analyzed the data; Isabella Giulia Franzoi, Maria Domenica Sauta, Carola Grimaldi and Irene Vallauri wrote the original draft of the article; Isabella Giulia Franzoi, Antonella Granieri, Alessandro Comandone, Maria Domenica Sauta, Marco Gonella, Monica Agnesone, Antonella Bogleione, Federica Vana, and Paola Bergnolo critically revised the manuscript and contributed to important intellectual content. All authors approved the final version of the paper to be published and agree to be accountable for all aspects of the work in ensuring that questions related to the

accuracy or integrity of any part of the work are appropriately investigated and resolved.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

The data that supports the findings of this study are available in the supplementary material of this article.

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REFERENCES

- Downe-Wamboldt B, Butler L, Coulter L. The relationship between meaning of illness, social support, coping strategies, and quality of life for lung cancer patients and their family members. *Cancer Nurs*. 2006;29(2):111-119. PMID: 16565620. <https://doi.org/10.1097/00002820-200603000-00006>
- Eichler M, Hentschel L, Richter S, et al. The PROSa study group. The health-related quality of life of sarcoma patients and survivors in Germany-cross-sectional results of a nationwide observational study (PROSa). *Cancers*. 2020;12(12):3590. PMID: PMC7759994. <https://doi.org/10.3390/cancers12123590>
- Nayak MG, George A, Vidyasagar MS, et al. Quality of life among cancer patients. *Indian J Palliat Care*. 2017;23(4):445-450. PMID: 29123353; PMID: PMC5661349. https://doi.org/10.4103/IJPC.IJPC_82_17
- Paredes T, Pereira M, Moreira H, Simões MR, Canavarró MC. Quality of life of sarcoma patients from diagnosis to treatments: predictors and longitudinal trajectories. *Eur J Oncol Nurs*. 2011;15(5):492-499. Epub 2011 Feb 8. PMID: 21306951. <https://doi.org/10.1016/j.ejon.2011.01.001>
- Hammermüller C, Hinz A, Dietz A, et al. Depression, anxiety, fatigue, and QoL in a large sample of patients suffering from head and neck cancer in comparison with the general population. *BMC Cancer*. 2021;21(1):94. PMID: 33482771; PMID: PMC7825198. <https://doi.org/10.1186/s12885-020-07773-6>
- Paredes T, Canavarró MC, Simões MR. Anxiety and depression in sarcoma patients: emotional adjustment and its determinants in the different phases of disease. *Eur J Oncol Nurs*. 2011;15(1):73-79. Epub 2010 Jul 29. PMID: 20667777. <https://doi.org/10.1016/j.ejon.2010.06.004>
- Liu J, Griva K, Lim HA, Tan JY, Mahendran R. Longitudinal study of the protective effect of hope on reducing body image distress in cancer patients. *J Psychosoc Oncol*. 2017;35(1):77-89. <https://doi.org/10.1080/07347332.2016.1221490>
- Miaja M, Platas A, Martínez-Cannon BA. Psychological impact of alterations in sexuality, fertility, and body image in young breast cancer patients and their partners. *Rev Invest Clin*. 2017;69(4):204-209. PMID: 28776605. <https://doi.org/10.24875/ric.17002279>
- Ghazali N, Cadwallader E, Lowe D, Humphris G, Ozakinci G, Rogers SN. Fear of recurrence among head and neck cancer survivors: longitudinal trends. *Psycho Oncol*. 2013;22(4):807-813. Epub 2012 Mar 27. PMID: 22451036. <https://doi.org/10.1002/pon.3069>
- Koch L, Jansen L, Brenner H, Arndt V. Fear of recurrence and disease progression in long-term (≥ 5 years) cancer survivors--a systematic review of quantitative studies. *Psycho Oncol*. 2013;22(1):1-11. Epub 2012 Jan 10. PMID: 22232030. <https://doi.org/10.1002/pon.3022>
- Granieri A. Extreme trauma in a polluted area: bonds and relational transformations in an Italian community. *Int Forum Psychoanal*. 2016;25(2):94-103. <https://doi.org/10.1080/0803706x.2015.1101488>
- Bonafede M, Chiorri C, Azzolina D, et al. Preliminary validation of a questionnaire assessing psychological distress in caregivers of patients with malignant mesothelioma: mesothelioma psychological distress tool--caregivers. *Psycho Oncol*. 2022;31(1):122-129. <https://doi.org/10.1002/pon.5789>
- Guglielmucci F, Franzoi IG, Bonafede M, Borgogno FV, Grosso F, Granieri A. "The less I think about it, the better I feel!": a thematic analysis of the subjective experience of malignant mesothelioma patients and their caregivers. *Front Psychol*. 2018;20(9):205. PMID: 29515509; PMID: PMC5826294. <https://doi.org/10.3389/fpsyg.2018.00205>
- Bonafede M, Granieri A, Binazzi A, et al. Psychological distress after a diagnosis of malignant mesothelioma in a group of patients and caregivers at the national priority contaminated site of Casale Monferrato. *Int J Environ Res Public Health*. 2020;17(12):4353. PMID: 32560553; PMID: PMC7345082. <https://doi.org/10.3390/ijerph17124353>
- Lee Wong C, Choi KC, Lau MN, Lam KL, Wei So WK. Caregiving burden and sleep quality amongst family caregivers of Chinese male patients with advanced cancer: a cross-sectional study. *Eur J Oncol Nurs*. 2020;46:101774. Epub ahead of print. PMID: 32460141. <https://doi.org/10.1016/j.ejon.2020.101774>
- Priya SS, Shavi GR, Sanga R, et al. Assessment of the perceived stress and burden of family caregivers of the head-and-neck cancer patients at a tertiary care cancer center: a cross-sectional study. *J Cancer Res Ther*. 2021;17(4):1039-1046. PMID: 34528561. https://doi.org/10.4103/jcrt.JCRT_309_19
- De Couck M, Mravec B, Gidron Y. You may need the vagus nerve to understand pathophysiology and to treat diseases. *Clin Sci*. 2012;122(7):323-328. PMID: 22150254. <https://doi.org/10.1042/CS20110299>
- Fonagy P, Luyten P. A developmental, mentalization-based approach to the understanding and treatment of borderline personality disorder. *Dev Psychopathol*. 2009;21(4):1355-1381. <https://doi.org/10.1017/s0954579409990198>
- Bergerot CD, Bergerot PG, Philip EJ, et al. Assessment of distress and quality of life in rare cancers. *Psycho Oncol*. 2018;27(12):2740-2746. Epub 2018 Sep 25. PMID: 30171792. <https://doi.org/10.1002/pon.4873>
- de Heus E, van der Zwan JM, Husson O, et al. Unmet supportive care needs of patients with rare cancer: a systematic review. *Eur J Cancer Care*. 2021;30(6):e13502. Epub 2021 Aug 18. PMID: 34409667. <https://doi.org/10.1111/ecc.13502>
- Jo VY, Fletcher CD. WHO classification of soft tissue tumours: an update based on the 2013 (4th) edition. *Pathology*. 2014;46(2):95-104. <https://doi.org/10.1097/PAT.000000000000050>
- Miller KD, Siegel RL, Lin CC, et al. Cancer treatment and survivorship statistics, 2016. *CA Cancer J Clin*. 2016;66(4):271-289. Epub 2016 Jun 2. PMID: 27253694. <https://doi.org/10.3322/caac.21349>
- Italiano A, Di Mauro I, Rapp J, et al. Clinical effect of molecular methods in sarcoma diagnosis (GENSARC): a prospective, multi-centre, observational study. *Lancet Oncol*. 2016;17(4):532-538. Epub

- 2016 Mar 10. PMID: 26970672. [https://doi.org/10.1016/S1470-2045\(15\)00583-5](https://doi.org/10.1016/S1470-2045(15)00583-5)
24. Weaver R, O'Connor M, Halkett GK, Carey Smith R. The unmet needs of carers of patients diagnosed with sarcoma: a qualitative study. *Psycho Oncol.* 2021;30(7):1095-1103. Epub 2021 Mar 1. PMID: 33544399. <https://doi.org/10.1002/pon.5651>
 25. Fauske L, Bruland OS, Grov EK, Bondevik H. Cured of primary bone cancer, but at what cost: a qualitative study of functional impairment and lost opportunities. *Sarcoma.* 2015;2015:484196. <https://doi.org/10.1155/2015/484196>
 26. Fauske L, Lorem G, Grov EK, Bondevik H. Changes in the body image of bone sarcoma survivors following surgical treatment--A qualitative study. *J Surg Oncol.* 2016;113(2):229-234. <https://doi.org/10.1002/jso.24138>
 27. McDonough J, Elliott J, Neuhaus S, Reid J, Butow P. Health-related quality of life, psychosocial functioning, and unmet health needs in patients with sarcoma: a systematic review. *Psycho Oncol.* 2019;28(4):653-664. Epub 2019 Feb 12. PMID: 30680843. <https://doi.org/10.1002/pon.5007>
 28. Fingeret MC, Teo I, Epner DE. Managing body image difficulties of adult cancer patients: lessons from available research. *Cancer.* 2014;120(5):633-641. <https://doi.org/10.1002/cncr.28469>
 29. Martins A, Alvarez-Galvez J, Fern LA, et al. The BRIGHTLIGHT national survey of the impact of specialist teenage and young adult cancer care on caregivers' information and support needs. *Cancer Nurs.* 2021;44(3):235-243. <https://doi.org/10.1097/NCC.0000000000000771>
 30. Applebaum AJ, Polacek LC, Walsh L, et al. The unique burden of rare cancer caregiving: caregivers of patients with Erdheim-Chester disease. *Leuk Lymphoma.* 2020;61(6):1406-1417. <https://doi.org/10.1080/10428194.2020.1719090>
 31. Page MJ, McKenzie JE, Bossuyt PM, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *Int J Surg (London, England).* 2021;88:105906. <https://doi.org/10.1016/j.ijsu.2021.105906>
 32. Moola S, Munn Z, Tufanaru C, et al. *Chapter 7: Systematic Reviews of Etiology and Risk. Joanna Briggs Institute Reviewer's Manual.* The Joanna Briggs Institute; 2017:5. <https://synthesismanual.jbi.global>
 33. Eichler M, Pink D, Menge F, et al. Quality of life of GIST patients with and without current tyrosine kinase inhibitor treatment: cross-sectional results of a German multicentre observational study (PROSa). *Eur J Cancer Care.* 2021;30(6):e13484. <https://doi.org/10.1111/ecc.13484>
 34. Kask G, Repo JP, Tukiainen EJ, Blomqvist C, Barner-Rasmussen I. Soft tissue sarcoma of lower extremity: functional outcome and quality of life. *Ann Surg Oncol.* 2021;28(11):6892-6905. <https://doi.org/10.1245/s10434-021-09774-6>
 35. Kollár A, Müller S, Limacher A, Briner I, Klenke F, Bernhard J. Return to work and quality of life in disease-free adult patients with soft-tissue and bone sarcoma of the extremity. *Praxis.* 2021;110(1):22-29. <https://doi.org/10.1024/1661-8157/a003592>
 36. Lim HJ, Ong CJ, Skanthakumar T, et al. Retrospective quality of life study in patients with retroperitoneal sarcoma in an Asian population. *Health Qual Life Outcomes.* 2020;18(1):270. PMID: 32762683; PMCID: PMC7409714. <https://doi.org/10.1186/s12955-020-01491-0>
 37. Saebye C, Fugløe HM, Nymark T, et al. Factors associated with reduced functional outcome and quality of life in patients having limb-sparing surgery for soft tissue sarcomas - a national multicenter study of 128 patients. *Acta Oncol.* 2017;56(2):239-244. <https://doi.org/10.1080/0284186X.2016.1268267>
 38. Younger E, Jones RL, den Hollander D, et al. Priorities and preferences of advanced soft tissue sarcoma patients starting palliative chemotherapy: baseline results from the HOLISTIC study. *ESMO Open.* 2021;6(5):100258. <https://doi.org/10.1016/j.esmoop.2021.100258>
 39. Furtado S, Grimer RJ, Cool P, et al. Physical functioning, pain and quality of life after amputation for musculoskeletal tumours: a national survey. *Bone Joint J.* 2015;97-B(9):1284-1290. <https://doi.org/10.1302/0301-620X.97B9.35192>
 40. Peiper M, Matthaei H, Bölke E, et al. Compartmental resection for subfascial extremity soft tissue sarcoma and quality of life in long-term survivors. *Wien Klin Wochenschr.* 2011;123(15-16):488-495. <https://doi.org/10.1007/s00508-011-1592-5>
 41. Drabbe C, Van der Graaf WTA, De Rooij BH, et al. The age-related impact of surviving sarcoma on health-related quality of life: data from the SURVSARC study. *ESMO Open.* 2021;6(1):100047. <https://doi.org/10.1016/j.esmoop.2021.100047>
 42. Wilke B, Cooper A, Scarborough M, Gibbs P, Spiguel A. A comparison of limb salvage versus amputation for nonmetastatic sarcomas using patient-reported outcomes measurement information system outcomes. *J Am Acad Orthop Surg.* 2019;27(8):e381-e389. <https://doi.org/10.5435/JAAOS-D-17-00758>
 43. Weidema ME, Husson O, van der Graaf WTA, et al. Health-related quality of life and symptom burden of epithelioid hemangioendothelioma patients: a global patient-driven Facebook study in a very rare malignancy. *Acta Oncol.* 2020;59(8):975-982. <https://doi.org/10.1080/0284186X.2020.1766696>
 44. Üstündağ S, Zencirci AD. Factors affecting the quality of life of cancer patients undergoing chemotherapy: a questionnaire study. *Asia Pac J Oncol Nurs.* 2015;2(1):17-25. <https://doi.org/10.4103/2347-5625.152402>
 45. Kaal SEJ, Lidington EK, Prins JB, et al. Health-related quality of life issues in adolescents and young adults with cancer: discrepancies with the perceptions of health care professionals. *J Clin Med.* 2021;10(9):1833. PMID: 33922382; PMCID: PMC8122798. <https://doi.org/10.3390/jcm10091833>
 46. Bulls HW, Chang PH, Brownstein NC, et al. Patient-reported symptom burden in routine oncology care: examining racial and ethnic disparities. *Cancer Rep.* 2022;5(3):e1478. <https://doi.org/10.1002/cnr2.1478>
 47. Lenze F, Pohlig F, Knebel C, et al. Psychosocial distress in follow-up care - results of a tablet-based routine screening in 202 patients with sarcoma. *Anticancer Res.* 2019;39(6):3159-3165. <https://doi.org/10.21873/anticancer.13453>
 48. Maggi G, Terrenato I, Giacomelli L, et al. Sarcoma patients' quality of life from diagnosis to yearly follow-up: experience from an Italian tertiary care center. *Future Oncol.* 2019;15(27):3125-3134. <https://doi.org/10.2217/fon-2019-0237>
 49. van Eck I, den Hollander D, Lidington E, et al. Health-related quality of life issues experienced by thoracic and breast sarcoma patients: a rare and understudied group. *J Clin Med.* 2021;10(22):5334. <https://doi.org/10.3390/jcm10225334>
 50. Weschenfelder W, Gast-Froehlich S, Spiegel C, Vogt M, Hofmann GO. Factors influencing quality of life, function, reintegration and participation after musculoskeletal tumour operations. *BMC Cancer.* 2020;20(1):351. <https://doi.org/10.1186/s12885-020-06837-x>
 51. Gough N, Koffman J, Ross JR, Riley J, Judson I. Symptom burden in advanced soft-tissue sarcoma. *J Pain Symptom Manage.* 2017;53(3):588-597. <https://doi.org/10.1016/j.jpainsymman.2016.10.357>
 52. Kokkali S, Boukovinas I, Samantas E, et al. A multicenter, prospective, observational study to assess the clinical activity and impact on symptom burden and patients' quality of life in patients with advanced soft tissue sarcomas treated with trabectedin in a real-world setting in Greece. *Cancers.* 2022;14(8):1879. <https://doi.org/10.3390/cancers14081879>
 53. van Eck I, den Hollander D, Desar IME, et al. Unraveling the heterogeneity of sarcoma survivors' health-related quality of life regarding primary sarcoma location: results from the SURVSARC study. *Cancers.* 2020;12(11):3083. <https://doi.org/10.3390/cancers12113083>

54. Reijers SJM, Husson O, Soomers VLMN, et al. Health-related quality of life after isolated limb perfusion compared to extended resection, or amputation for locally advanced extremity sarcoma: is a limb salvage strategy worth the effort? *Eur J Surg Oncol*. 2022;48(3):500-507. <https://doi.org/10.1016/j.ejso.2021.08.007>
55. Soomers VLMN, Desar IME, van de Poll-Franse LV, et al. The perceived impact of length of the diagnostic pathway is associated with health-related quality of life of sarcoma survivors: results from the Dutch nationwide SURVSARC study. *Cancers*. 2020;12(8):2088. <https://doi.org/10.3390/cancers12082088>
56. Moon TM, Furdock R, Rhea L, Pergolotti M, Cipriano C, Spraker MB. PROMIS scores of patients undergoing neoadjuvant and adjuvant radiation therapy for surgically excised soft tissue sarcoma. *Clin Transl Radiat Oncol*. 2021;31:42-49. PMID: 34622039. <https://doi.org/10.1016/j.ctro.2021.08.008>
57. Palm RF, Jim HSL, Boulware D, Johnstone PAS, Naghavi AO. Using the revised Edmonton symptom assessment scale during neoadjuvant radiotherapy for retroperitoneal sarcoma. *Clin Transl Radiat Oncol*. 2020;22:22-28. <https://doi.org/10.1016/j.ctro.2020.02.007>
58. Saebye C, Amidi A, Keller J, Andersen H, Baad-Hansen T. Changes in functional outcome and quality of life in soft tissue sarcoma patients within the first year after surgery: a prospective observational study. *Cancers*. 2020;12(2):463. <https://doi.org/10.3390/cancers12020463>
59. Sachsenmaier SM, Ipach I, Kluba T. Quality of life, physical and mental status and contentment of patients with localized soft tissue or bone sarcoma: a questionnaire analysis. *Orthop Rev*. 2015;7(2):5920. <https://doi.org/10.4081/or.2015.5920>
60. Pinto E, Gonçalves F, Sacarlal J, Castro L, Rego G. Pain management in cancer patients in the main hospitals in Mozambique. *Ann Palliat Med*. 2021;10(4):4069-4079. <https://doi.org/10.21037/apm-20-2009>
61. Ostacoli L, Saini A, Zuffranieri M, et al. Quality of life, anxiety and depression in soft tissue sarcomas as compared to more common tumours: an observational study. *App Res Qual Life*. 2014;9(1):123-131. <https://doi.org/10.1007/s11482-013-9213-2>
62. Ranft A, Seidel C, Hoffmann C, et al. Quality of survivorship in a rare disease: clinicofunctional outcome and physical activity in an observational cohort study of 618 long-term survivors of Ewing sarcoma. *J Clin Oncol*. 2017;35(15):1704-1712. <https://doi.org/10.1200/JCO.2016.70.6226>
63. van Houdt WJ, Griffin AM, Wunder JS, Ferguson PC. Oncologic outcome and quality of life after hindquarter amputation for sarcoma: is it worth it? *Ann Surg Oncol*. 2018;25(2):378-386. <https://doi.org/10.1245/s10434-017-5806-6>
64. Stish BJ, Ahmed SK, Rose PS, Arndt CA, Laack NN. Patient-reported functional and quality of life outcomes in a large cohort of long-term survivors of Ewing sarcoma. *Pediatr Blood Cancer*. 2015;62(12):2189-2196. <https://doi.org/10.1002/pbc.25710>
65. Eichler M, Singer S, Hentschel L, et al. The association of health-related QoL and 1-year-survival in sarcoma patients-results of a nationwide observational study (PROSa). *Br J Cancer*. 2022;126(9):1346-1354. <https://doi.org/10.1038/s41416-022-01702-z>
66. Ise M, Nakata E, Katayama Y, et al. Prevalence of psychological distress and its risk factors in patients with primary bone and soft tissue tumors. *Healthcare*. 2021;9(5):566. <https://doi.org/10.3390/healthcare9050566>
67. Srikanthan A, Leung B, Shokoohi A, Smrke A, Bates A, Ho C. Psychosocial distress scores and needs among newly diagnosed sarcoma patients: a provincial experience. *Sarcoma*. 2019;2019:5302639. <https://doi.org/10.1155/2019/5302639>
68. Horick NK, Manful A, Lowery J, et al. Physical and psychological health in rare cancer survivors. *J Cancer Surviv*. 2017;11(1):158-165. <https://doi.org/10.1007/s11764-016-0573-0>
69. Ahomäki R, Kero A, Koivisto M, Madanat-Harjuoja L, Malila N, Lähteenmäki PM. Purchases of antidepressants after cancer at a young age in Finland. *Int J Cancer*. 2019;144(6):1227-1233. <https://doi.org/10.1002/ijc.31942>
70. Paredes T, Pereira M, Simões MR, Canavarró MC. A longitudinal study on emotional adjustment of sarcoma patients: the determinant role of demographic, clinical and coping variables. *Eur J Cancer Care*. 2012;21(1):41-51. <https://doi.org/10.1111/j.1365-2354.2011.01269.x>
71. Shen J, Zhu M, Li S, et al. Incidence and risk factors for suicide death among Kaposi's sarcoma patients: a surveillance, epidemiology, and end results analysis. *Med Sci Monit*. 2020;26:e920711. <https://doi.org/10.12659/MSM.920711>
72. Siracuse BL, Gorgy G, Ruskin J, Beebe KS. What is the incidence of suicide in patients with bone and soft tissue cancer?: suicide and sarcoma. *Clin Orthop Relat Res*. 2017;475(5):1439-1445. <https://doi.org/10.1007/s11999-016-5171-y>
73. Büttner M, Singer S, Hentschel L, et al. Financial toxicity in sarcoma patients and survivors in Germany: results from the multicenter PROSa study. *Support Care Cancer*. 2022;30(1):187-196. <https://doi.org/10.1007/s00520-021-06406-3>
74. Drabbe C, Grünhagen DJ, Van Houdt WJ, et al. Diagnosed with a rare cancer: experiences of adult sarcoma survivors with the healthcare system—results from the SURVSARC study. *Cancers*. 2021;13(4):679. <https://doi.org/10.3390/cancers13040679>
75. Guglielmucci F, Franzoi IG, Barbasio CP, Borgogno FV, Granieri A. Helping traumatized people survive: a psychoanalytic intervention in a contaminated site. *Front Psychol*. 2014;5:1419. <https://doi.org/10.3389/fpsyg.2014.01419>
76. Spiegel D. Effects of psychosocial support on patients with metastatic breast cancer. *J Psychosoc Oncol*. 1992;10(2):113-120. https://doi.org/10.1300/j077v10n02_08
77. Granieri A, Borgogno FV, Franzoi IG, Gonella M, Guglielmucci F. Development of a brief psychoanalytic group therapy (BPG) and its application in an asbestos national priority contaminated site. *Ann I Super Sanità*. 2018;54(2):161-167. https://doi.org/10.4415/ANN_18_02_12
78. Straker N, ed. *Facing cancer and the fear of death: A psychoanalytic perspective on treatment*. Rowman & Littlefield; 2013.
79. Bronner MB, Nguyen MH, Smets EMA, van de Ven AWH, van Weert JCM. Anxiety during cancer diagnosis: examining the influence of monitoring coping style and treatment plan. *Psycho Oncol*. 2018;27(2):661-667. <https://doi.org/10.1002/pon.4560>
80. Bardeen JR, Stevens EN, Murdock KW, Lovejoy MC. A preliminary investigation of sex differences in associations between emotion regulation difficulties and higher-order cognitive abilities. *Pers Individ Dif*. 2013;55(1):70-75. <https://doi.org/10.1016/j.paid.2013.02.003>
81. Schechter M, Herbstman B, Ronningstam E, Goldblatt MJ. Emerging adults, identity development, and suicidality: implications for psychoanalytic psychotherapy. *Psychoanal Study Child*. 2018;71(1):20-39. <https://doi.org/10.1080/00797308.2017.1415596>
82. Nguyen AW. Religion and mental health in racial and ethnic minority populations: a review of the literature. *Innov Aging*. 2020;4(5):igaa035. <https://doi.org/10.1093/geron/igaa035>

SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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