

■ ONCOLOGY

Impact of racial disparities and insurance status in patients with bone sarcomas in the USA

A POPULATION-BASED COHORT STUDY



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Aims

Socioeconomic and racial disparities have been recognized as impacting the care of patients with cancer, however there are a lack of data examining the impact of these disparities on patients with bone sarcoma. The purpose of this study was to examine socioeconomic and racial disparities that impact the oncological outcomes of patients with bone sarcoma.

Methods

We reviewed 4,739 patients diagnosed with primary bone sarcomas from the Surveillance, Epidemiology and End Results (SEER) registry between 2007 and 2015. We examined the impact of race and insurance status associated with the presence of metastatic disease at diagnosis, treatment outcome, and overall survival (OS).

Results

Patients with Medicaid (odds ratio (OR) 1.41; 95% confidence interval (CI) 1.15 to 1.72) and uninsured patients (OR 1.90; 95% CI 1.26 to 2.86) had higher risks of metastatic disease at diagnosis compared to patients with health insurance. Compared to White patients, Black (OR 0.63, 95% CI 0.47 to 0.85) and Asian/Pacific Islander (OR 0.65, 95% CI 0.46 to 0.91) were less likely to undergo surgery. In addition, Black patients were less likely to receive chemotherapy (OR 0.67, 95% CI 0.49 to 0.91) compared to White patients. In patients with chondrosarcoma, those with Medicaid had worse OS compared to patients with insurance (hazard ratio (HR) 1.65, 95% CI 1.06 to 2.56).

Conclusion

In patients with a bone sarcoma, the cancer stage at diagnosis varied based on insurance status, and racial disparities were identified in treatment. Further studies are needed to identify modifiable factors which can mitigate socioeconomic and racial disparities found in patients with bone sarcomas.

Cite this article: *Bone Joint Res* 2022;11(5):278–291.

Keywords: Bone sarcoma, Cancer disparity, Racial disparities, Insurance, Health equality

Article focus

■ This study aimed to assess the associations of racial disparities and socioeconomic status with diagnosis, treatment, and survival outcomes in patients with a primary bone sarcoma.

Key messages

■ Black and Asian/Pacific Islander patients are less likely to undergo surgery

compared to White patients for primary bone sarcomas.

■ Patients with Medicaid, or those who are uninsured, were more likely to be diagnosed with metastatic disease diagnosis compared to patients with medical insurance.

■ Compared to patients with medical insurance, those with Medicaid had worse survival for chondrosarcoma of bone.

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doi: 10.1302/2046-3758.115.BJR-2021-0258.R2

Bone Joint Res 2022;11(5):278–291.

Strengths and limitations

- Using the population-based Surveillance, Epidemiology and End Results Program, our study included a large dataset of bone sarcoma, which is a rare form of cancer.
- Racial disparities in treatment and survival for patients with bone sarcoma were identified for all bone sarcoma subtypes, providing information to promote health equity for patients.
- Further work is needed to examine the association of racial disparities and insurance status in patients with bone sarcomas.

Introduction

Primary bone sarcomas are rare malignancies which arise from mesenchymal cells, with an annual incidence of 1.0 per 100,000 people, accounting for 3,600 new cases of cancer in the USA per year.^{1,2} Early diagnosis of a bone sarcoma depends on clinical and imaging examination, and is confirmed via biopsy.^{3,4} Multidisciplinary management is essential for patients diagnosed with a bone sarcoma, with treatment typically including surgical resection with negative margins and chemotherapy.⁵⁻⁷ Through advancements in medical and surgical management, the five-year survival of patients with a bone sarcoma has improved over time,² however there are likely socioeconomic factors which impact outcomes.

The American Society of Clinical Oncology (ASCO) endeavours to create awareness of disparities in cancer care and promote health equity.⁸ Racial disparities and insurance status are two factors that impact health equity. Previous studies have examined the impact of racial disparities and insurance status in patients with breast cancer,⁹ lung cancer,^{10,11} colorectal cancer,¹² and many other common cancers.^{13,14} Currently there are a lack of data examining the impact of racial disparities and insurance status in patients with bone sarcoma, which might be due to the rare nature of this disease. Therefore, the purpose of the current study was to examine the impact of insurance status and race on oncological outcomes of patients with primary bone sarcomas.

Methods

Patient selection. The Surveillance, Epidemiology and End Results (SEER)-18 registry was used to identify patients undergoing treatment for bone sarcoma between 2004 and 2015, using the International Classification of Diseases of the World Health Organization (ICD) codes C40.0-3, C40.8-9, C41.0-4, and C41.8-9.¹⁵ Exclusion criteria included patients diagnosed at the time of death, those without follow-up or a positive biopsy, patients with an unknown race, patients with an unknown metastatic stage, and lack of confirmation if they underwent surgery. To analyze the impact of insurance status, only patients with a diagnosis after 2007 were included, since insurance status was only available in SEER after 2007. In

addition, patients aged ≥ 65 years were excluded due to their ability to enrol in Medicare.¹⁴ The remaining group of 4,739 patients were included in the study (Figure 1).

Exposures, covariates, and outcomes. Race was divided into four categories: White; Hispanic; Black; and Asian/Pacific Islander.¹⁶ Insurance status was divided into three categories: insured (non-Medicaid); Medicaid; and uninsured.

The demographic data included age, sex, and marital status. Tumour type was extracted based on ICD codes and divided into five categories: osteosarcoma, chondrosarcoma, Ewing's sarcoma, chordoma, and other (unspecified malignant bone tumours, miscellaneous malignant bone tumours, odontogenic malignant tumours, and malignant fibrous neoplasms of bone). Tumour location was divided into six categories: lower limb; sacro-pelvic; scapular and upper limb; craniofacial; chest wall; and other (tumour site of others included short bones, mandible, vertebral column, overlap bones, joints, and cartilage, and bone not otherwise specified). In addition, the American Joint Committee on Cancer (AJCC) T, N, and M stages were obtained from the SEER database.¹⁷

Outcome data included metastatic stage at diagnosis, overall survival (OS), and the use of surgery, chemotherapy, and radiotherapy. OS was defined as the time from sarcoma diagnosis to all-cause mortality.

Statistical analysis. Analysis was performed using SPSS 24.0 (IBM, USA) and GraphPad Prism 7.0 (GraphPad, USA). Differences in patient characteristics according to the race and type of insurance were examined by the chi-squared test, the independent-samples *t*-test, or Z-test (compare column proportions and adjust p-values via the Bonferroni method in the chi-squared test). The association between race and insurance status with metastatic disease at the time of diagnosis, and the use of different treatments (surgery, chemotherapy, radiotherapy), were analyzed with multivariable logistic regression model and adjusted odds ratio (OR) with 95% confidence interval (CI). The Kaplan-Meier method was used to estimate survival outcomes between groups, and the comparisons were examined by the log-rank test. Multivariable Cox regression models with adjusted hazard ratio (HR) with 95% CIs were used to examine factors associated with survival. All tests were two-sided, and p-values < 0.05 were considered statistically significant.

Results

Patient characteristics stratified by race. The patient group comprised 4,739 patients, including 2,807 (59.2%) White, 1,064 (22.5%) Hispanic, 526 (11.1%) Black, and 342 (7.2%) Asian/Pacific Islander (Table I). Compared to the White group, minority patients (Hispanic, Black, and Asian/Pacific Islander) were younger at the time of diagnosis ($p < 0.001$, independent-samples *t*-test). There was no difference in the sex distribution between the racial groups ($p = 0.321$, chi-squared test). White and Asian/Pacific Islander patients were more likely to be married

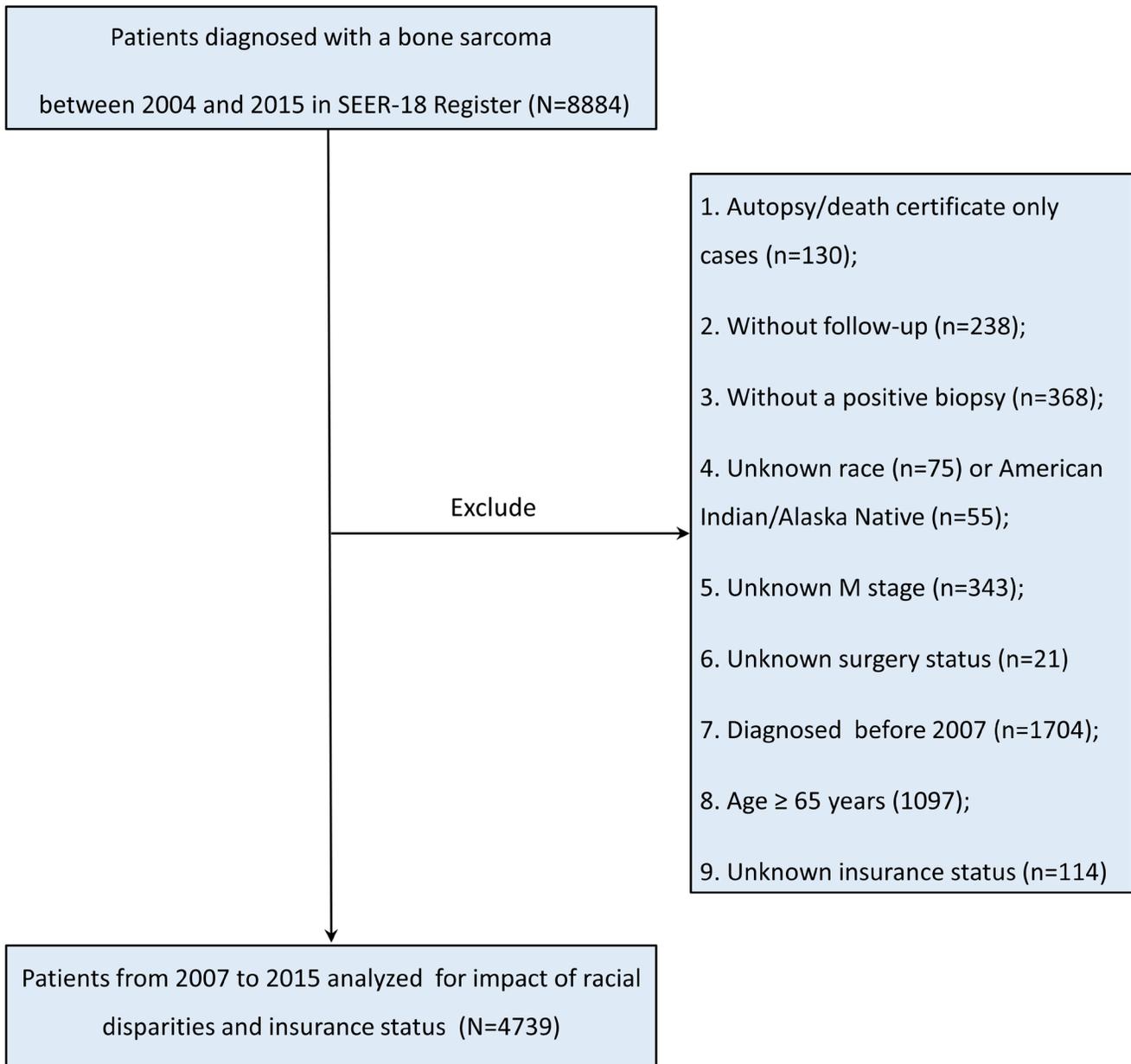


Fig. 1

Flowchart diagram showing patient enrolment. SEER, Surveillance, Epidemiology and End Results.

at the time of diagnosis compared to Hispanic and Black patients ($p < 0.001$, chi-squared test).

Osteosarcomas were more common in Black patients (305 (58.0%)) compared to White patients (937 (33.4%)). Black (260 (49.4%)) and Hispanic (469 (44.1%)) patients were more likely to have a bone sarcoma in the lower limb compared to Asian/Pacific Islander (136 (39.8%)) and White (1,023 (36.4%)) patients ($p < 0.001$, chi-squared test). White patients were more likely to have a sacropelvic tumour than black patients (454 (16.2%) vs 61 (11.6%)).

White patients were more likely to present with an AJCC T1 tumour (1,315 (46.8%)) compared to Hispanic (434 (40.8%)) and Black (213 (40.5%)) patients ($p = 0.003$). Hispanic patients were more likely to present with metastatic disease (M1) compared to White patients (19.1% vs 14.7%, $p = 0.003$, chi-squared test). There was no difference in the use of surgery between White, Hispanic, Black, and Asian/Pacific Islander patients ($p = 0.340$, chi-squared test). Black patients had a lower use of radiotherapy (47 (8.9%)) compared to White patients (472 (16.8%)) ($p < 0.001$, chi-squared test). Hispanic, Black, and Asian/Pacific Islander patients were more likely

Table 1. Demographic and clinical characteristics by race (n = 4,739).

Characteristic	White (n = 2,807)	Hispanic (n = 1,064)	Black (n = 526)	Asian/Pacific Islander (n = 342)	p-value
Mean age, yrs (SD)	33.7 (18.6)	26.4 (16.3)	28.8 (16.5)	29.7 (17.3)	< 0.001‡
< 18 yrs, n (%)	803 (28.6)	441 (41.4)	190 (36.1)	108 (31.6)	
18 to 59 yrs, n (%)	1,733 (61.7)	579 (54.4)	317 (60.3)	215 (62.9)	
≥ 60 yrs, n (%)	271 (9.7)	44 (4.1)	19 (3.6)	19 (5.6)	
Sex, n (%)					0.321§
Female	1,215 (43.3)	433 (40.7)	238 (45.2)	145 (42.4)	
Male	1,592 (56.7)	631 (59.3)	288 (54.8)	197 (57.6)	
Marital status, n (%)					< 0.001§
Married	1,019 (36.3)	262 (24.6)	94 (17.9)	115 (33.6)	
Not married	1,721 (61.3)	785 (73.8)	412 (78.3)	221 (64.6)	
Unknown	67 (2.4)	17 (1.6)	20 (3.8)	6 (1.8)	
Tumour type, n (%)					< 0.001§
Osteosarcoma	937 (33.4)	511 (48.0)	305 (58.0)	148 (43.3)	
Chondrosarcoma	864 (30.8)	184 (17.3)	99 (18.8)	68 (19.9)	
Ewing's sarcoma	582 (20.7)	196 (18.4)	35 (6.7)	56 (16.4)	
Chordoma	246 (8.8)	91 (8.6)	23 (4.4)	40 (11.7)	
Others*	178 (6.3)	82 (7.7)	64 (12.2)	30 (8.8)	
Tumour site, n (%)					< 0.001§
Lower limb	1,023 (36.4)	469 (44.1)	260 (49.4)	136 (39.8)	
Upper limb	367 (13.1)	113 (10.6)	73 (13.9)	41 (12.0)	
Sacropelvic	454 (16.2)	153 (14.4)	61 (11.6)	49 (14.3)	
Craniofacial	286 (10.2)	127 (11.9)	40 (7.6)	47 (13.7)	
Rib, chest wall	268 (9.5)	64 (6.0)	17 (3.2)	22 (6.4)	
Others†	409 (14.6)	138 (13.0)	75 (14.3)	47 (13.7)	
AJCC T stage, n (%)					0.003§
T1	1,315 (46.8)	434 (40.8)	213 (40.5)	162 (47.4)	
T2-3	1,026 (36.6)	457 (43.0)	222 (42.2)	128 (37.4)	
Unknown	466 (16.6)	173 (16.3)	91 (17.3)	52 (15.2)	
AJCC N stage, n (%)					0.008§
N0	2,641 (94.1)	969 (91.1)	477 (90.7)	317 (92.7)	
N1	69 (2.5)	34 (3.2)	18 (3.4)	7 (2.0)	
Unknown	97 (3.5)	61 (5.7)	31 (5.9)	18 (5.3)	
AJCC M stage, n (%)					0.003§
M0	2,393 (85.3)	861 (80.9)	441 (83.8)	299 (87.4)	
M1	414 (14.7)	203 (19.1)	85 (16.2)	43 (12.6)	
Surgery, n (%)					0.340§
No	450 (16.0)	185 (17.4)	97 (18.4)	64 (18.7)	
Yes	2,357 (84.0)	879 (82.6)	429 (81.6)	278 (81.3)	
Radiotherapy, n (%)					< 0.001§
No	2,335 (83.2)	900 (84.6)	479 (91.1)	280 (81.9)	
Yes	472 (16.8)	164 (15.4)	47 (8.9)	62 (18.1)	
Chemotherapy, n (%)					< 0.001§
No	1,294 (46.1)	372 (35.0)	215 (40.9)	135 (39.5)	
Yes	1,513 (53.9)	692 (65.0)	311 (59.1)	207 (60.5)	
Follow-up time, mths					< 0.001‡
Mean (95% CI)	42.9 (41.8 to 44.0)	37.0 (35.2 to 38.8)	39.8 (37.1 to 42.4)	37.6 (34.5 to 40.8)	
Median (IQR)	38 (16 to 67)	29 (12 to 58)	31 (13 to 65)	30 (10.75 to 58.25)	
OS rate					0.336§
Dead	693 (24.7)	252 (23.7)	145 (27.6)	79 (23.1)	
Alive	2,114 (75.3)	812 (76.3)	381 (72.4)	263 (76.9)	

*Tumour type of others included unspecified malignant bone tumours, miscellaneous malignant bone tumours, odontogenic malignant tumours, and malignant fibrous neoplasms of bone.

†Tumour site of others included short bones, mandible, vertebral column, overlap bones, joints, and cartilage, and bone not otherwise specified.

‡Independent-samples t-test.

§Chi-squared test.

AJCC, American Joint Committee on Cancer; CI, confidence interval; IQR, interquartile range; OS, overall survival; SD, standard deviation.

to receive chemotherapy compared to White patients ($p < 0.001$, chi-squared test).

Patient characteristics stratified by insurance status. Of the 4,739 patients, 3,401 (71.8%) were insured, 1,141 (24.1%) had Medicaid, and 197 (4.2%) were uninsured (Table II). Patients with Medicaid were younger than insured and uninsured patients ($p < 0.001$, chi-squared test). There was no difference in the sex portions based on insurance status ($p = 0.464$, chi-squared test). Patients with insurance were more likely to be married compared to patients with Medicaid and those without insurance ($p < 0.001$, chi-squared test).

Patients with Medicaid were more likely to present with an osteosarcoma (593 (52.0%)) and involving the lower limbs (526 (46.1%)). Patients with Medicaid and those without insurance were more likely to have a higher T stage (T2-3) and N stage (N1) at the time of diagnosis compared to patients with insurance. Patients with Medicaid (247 (21.6%)) and those without insurance (41 (20.8%)) were more likely to present with metastatic disease (M1) compared to patients with insurance (457 (13.4%)) ($p < 0.001$, chi-squared test).

Patients with Medicaid (910 (79.8%)) and those without insurance (155 (78.7%)) were less likely to undergo surgical resection compared to patients with insurance (2,878 (84.6%)) ($p < 0.001$, chi-squared test). There was no difference in the use of radiotherapy based on insurance status ($p = 0.112$, chi-squared test). Patients with Medicaid were more likely to receive chemotherapy (820 (71.9%)).

Associations of racial disparities and insurance status with metastasis at diagnosis. After adjusting for age, sex, marital status, tumour type, site, and stages (Table III), race was not associated with the presence of metastatic disease at the time of diagnosis ($p = 0.867$ (Hispanic vs White), $p = 0.472$ (Black vs White), and $p = 0.240$ (Asian/Pacific Islander vs White)). However, patients with Medicaid (adjusted OR = 1.41; 95% CI = 1.15 to 1.72) and those without insurance (adjusted OR = 1.90; 95% CI = 1.26 to 2.86) were at increased risk of presenting with metastatic disease compared to patients with health insurance. In addition, male sex (adjusted OR = 1.20; 95% CI = 1.00 to 1.43), patients with osteosarcoma (adjusted OR = 1.48; 95% CI = 1.03 to 2.13) or Ewing's sarcoma (adjusted OR = 2.52; 95% CI = 1.73 to 3.69), sacropelvic tumours (adjusted OR = 2.06; 95% CI = 1.61 to 2.63), and higher AJCC T stage (adjusted OR = 2.72; 95% CI = 2.18 to 3.40) were at increased risk of presenting with metastatic disease at diagnosis. Patients with a chondrosarcoma (adjusted OR = 0.43; 95% CI = 0.28 to 0.66) or chordoma (adjusted OR = 0.18; 95% CI = 0.08 to 0.38) and craniofacial sarcomas (adjusted OR = 0.35; 95% CI = 0.19 to 0.65) were at less risk of presenting with metastatic disease at diagnosis.

In subgroup analyses based on the histological diagnosis, patients with an osteosarcoma, with Medicaid, had an increased risk of presenting with metastatic disease (adjusted OR = 1.42, 95% CI = 1.08 to 1.88). By contrast, race and insurance status were not significantly associated

with a diagnosis of metastatic disease at presentation in patients with a chondrosarcoma, Ewing's sarcoma, or chordoma (Table III). Patients with sacropelvic osteosarcomas (adjusted OR = 2.57; 95% CI = 1.71 to 3.87) and Ewing's sarcomas (adjusted OR = 1.97; 95% CI = 1.30 to 2.97) were more likely to present with metastatic disease at the time of diagnosis.

Associations of racial disparities and insurance status with treatment. After adjusting for age, sex, marital status, tumour type, site, and stages, Black (adjusted OR = 0.63, 95% CI = 0.47 to 0.85) and Asian/Pacific Islander patients (adjusted OR = 0.65, 95% CI = 0.46 to 0.91) were less likely to undergo surgery compared to White patients (Table IV). Patients with Medicaid were less likely to undergo surgery compared to patients with insurance (adjusted OR = 0.80, 95% CI = 0.64 to 0.99). As expected, patients between the age of 18 and 59 years (adjusted OR = 0.59, 95% CI = 0.47 to 0.75), age ≥ 60 years (adjusted OR = 0.44, 95% CI = 0.29 to 0.66), with Ewing's sarcoma (adjusted OR = 0.33, 95% CI = 0.23 to 0.47), upper limb sarcoma (adjusted OR = 0.72, 95% CI = 0.53 to 0.97), sacropelvic sarcomas (adjusted OR = 0.19, 95% CI = 0.15 to 0.25), higher N stage (adjusted OR = 0.54, 95% CI = 0.35 to 0.83), and metastatic disease (adjusted OR = 0.21, 95% CI = 0.17 to 0.26) were less likely to undergo surgical resection.

These trends were confirmed in a subgroup analysis focusing on chondrosarcoma; Black (adjusted OR = 0.31, 95% CI = 0.15 to 0.63) and Asian/Pacific Islander patients (adjusted OR = 0.22, 95% CI = 0.10 to 0.47) were less likely to undergo surgery compared to Whites; patients with Medicaid (adjusted OR = 0.48, 95% CI = 0.27 to 0.88) and those without insurance (adjusted OR = 0.35, 95% CI = 0.14 to 0.85) were less likely to undergo surgical resection compared to patients with medical insurance. In patients with Ewing's sarcoma, Asian/Pacific Islander patients were less likely to undergo surgery compared to White patients (adjusted OR = 0.37, 95% CI = 0.20 to 0.69).

After adjusting for age, sex, marital status, tumour type, site, and stage, Black patients were less likely to receive chemotherapy compared to White patients (adjusted OR = 0.67, 95% CI = 0.49 to 0.91, Supplementary Table i). Patients with an osteosarcoma (adjusted OR = 5.85, 95% CI = 4.42 to 7.73), Ewing's sarcoma (adjusted OR = 27.5, 95% CI = 17.3 to 43.6), sacropelvic sarcomas (adjusted OR = 1.42, 95% CI = 1.03 to 1.97), higher T stage (adjusted OR = 2.42, 95% CI = 1.92 to 3.05), higher N stage (adjusted OR = 2.84, 95% CI = 1.36 to 5.91), and metastatic disease at the time of diagnosis (adjusted OR = 5.25, 95% CI = 3.67 to 7.50) were associated with the use of chemotherapy. In subgroup analyses based on histological subtypes, the use of chemotherapy was not associated with race or insurance status (Supplementary Table i).

Race and insurance status were not significantly associated with the use of radiotherapy (Supplementary Table ii). Patients with craniofacial sarcoma were more likely to

Table II. Demographic and clinical characteristics by insurance type (n = 4,739), number (%).

Characteristics	Insured (n = 3,401)	Medicaid (n = 1,141)	Uninsured (n = 197)	p-value
Mean age, yrs (SD)	33 (18)	24 (15)	34 (16)	< 0.001‡
< 18	988 (29.1)	529 (46.4)	25 (12.7)	
18 to 59	2,111 (62.1)	575 (50.4)	158 (80.2)	
≥ 60	302 (8.9)	37 (3.2)	14 (7.1)	
Sex				0.464§
Female	1,464 (43.0)	491 (43.0)	76 (38.6)	
Male	1,937 (57.0)	650 (57.0)	121 (61.4)	
Marital status				< 0.001§
Married	1,302 (38.3)	139 (12.2)	49 (24.9)	
Not married	2,013 (59.2)	982 (86.1)	144 (73.1)	
Unknown	86 (2.5)	20 (1.8)	4 (2.0)	
Tumour type				< 0.001§
Osteosarcoma	1,239 (36.4)	593 (52.0)	69 (35.0)	
Chondrosarcoma	994 (29.2)	164 (14.4)	57 (28.9)	
Ewing's sarcoma	602 (17.7)	240 (21.0)	27 (13.7)	
Chordoma	316 (9.3)	66 (5.8)	18 (9.1)	
Others*	250 (7.4)	78 (6.8)	26 (13.2)	
Tumour site				< 0.001§
Lower limb	1,290 (37.9)	526 (46.1)	72 (36.5)	
Upper limb	542 (15.9)	145 (12.7)	30 (15.2)	
Sacropelvic	416 (12.2)	158 (13.8)	20 (10.2)	
Craniofacial	370 (10.9)	106 (9.3)	24 (12.2)	
Rib, chest wall	291 (8.6)	66 (5.8)	14 (7.1)	
Others†	492 (14.5)	140 (12.3)	37 (18.8)	
AJCC T stage				< 0.001§
T1	1,616 (47.5)	425 (37.2)	83 (42.1)	
T2-3	1,241 (36.5)	514 (45.0)	78 (39.6)	
Unknown	544 (16.0)	202 (17.7)	36 (18.3)	
AJCC N stage				0.007§
N0	3,188 (93.7)	1,040 (91.1)	176 (89.3)	
N1	83 (2.4)	39 (3.4)	6 (3.0)	
Unknown	130 (3.8)	62 (5.4)	15 (7.6)	
AJCC M stage				< 0.001§
M0	2,944 (86.6)	894 (78.4)	156 (79.2)	
M1	457 (13.4)	247 (21.6)	41 (20.8)	
Surgery				< 0.001§
No	523 (15.4)	231 (20.2)	42 (21.3)	
Yes	2,878 (84.6)	910 (79.8)	155 (78.7)	
Radiotherapy				0.112§
No	2,846 (83.7)	984 (86.2)	164 (83.2)	
Yes	555 (16.3)	157 (13.8)	33 (16.8)	
Chemotherapy				< 0.001§
No	1,590 (46.8)	321 (28.1)	105 (53.3)	
Yes	1,811 (53.2)	820 (71.9)	92 (46.7)	
Follow-up time, mths				< 0.001‡
Mean (95% CI)	42.6 (41.6 to 43.6)	36.3 (34.6 to 38.0)	36.8 (32.8 to 40.7)	
Overall survival rate				0.146§
Dead	813 (23.9)	302 (26.5)	54 (27.4)	
Alive	2,588 (76.1)	839 (73.5)	143 (72.6)	

*Including unspecified malignant bone tumours, miscellaneous malignant bone tumours, odontogenic malignant tumours, and malignant fibrous neoplasms of bone.

†Including short bones, mandible, vertebral column, overlap bones, joints, and cartilage, and bone not otherwise specified.

‡Independent-samples *t*-test.

§Chi-squared test.

AJCC, American Joint Committee on Cancer; CI, confidence interval; SD, standard deviation.

Table III. Adjusted odds ratio for metastatic disease at the time of diagnosis in bone sarcomas.

Characteristics	Metastatic disease		Metastatic osteosarcoma		Metastatic chondrosarcoma		Metastatic Ewing's sarcoma		Metastatic chordoma	
	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value
Race										
White	1.00		1.00		1.00		1.00		1.00	
Hispanic	1.02 (0.82 to 1.27)	0.867	1.07 (0.79 to 1.45)	0.684	0.67 (0.30 to 1.51)	0.335	1.05 (0.71 to 1.55)	0.820	1.15 (0.10 to 13.8)	0.913
Black	0.90 (0.67 to 1.20)	0.472	0.90 (0.63 to 1.30)	0.575	0.92 (0.32 to 2.63)	0.880	1.49 (0.70 to 3.19)	0.303	N/A	
Asian/Pacific Islander	0.81 (0.56 to 1.16)	0.240	0.84 (0.51 to 1.40)	0.506	0.86 (0.25 to 2.98)	0.813	0.80 (0.42 to 1.56)	0.518	4.96 (0.56 to 43.7)	0.149
Insurance type										
Insured	1.00		1.00		1.00		1.00		1.00	
Medicaid	1.41 (1.15 to 1.72)	0.001	1.42 (1.08 to 1.88)	0.013	1.55 (0.69 to 3.50)	0.290	1.29 (0.90 to 1.86)	0.164	1.07 (0.09 to 13.1)	0.956
Uninsured	1.90 (1.26 to 2.86)	0.002	1.43 (0.73 to 2.79)	0.295	2.43 (0.89 to 6.64)	0.084	1.77 (0.76 to 4.15)	0.187	1.56 (0.07 to 32.8)	0.774
Age, yrs										
< 18	1.00		1.00		N/A		1.00		1.00	
18 to 59	0.82 (0.66 to 1.01)	0.058	0.62 (0.46 to 0.83)	0.002	N/A		1.27 (0.89 to 1.80)	0.185	0.07 (0.004 to 1.27)	0.073
≥ 60	1.33 (0.87 to 2.03)	1.190	1.72 (0.89 to 3.31)	0.106	N/A		0.90 (0.16 to 5.01)	0.900	0.07 (0.002 to 2.35)	0.136
Sex										
Female	1.00		1.00		1.00		1.00		1.00	
Male	1.20 (1.00 to 1.43)	0.048	1.26 (0.98 to 1.62)	0.074	1.46 (0.82 to 2.59)	0.199	1.05 (0.76 to 1.45)	0.758	0.38 (0.07 to 2.05)	0.259
Marital status										
Married	1.00		1.00		1.00		1.00		1.00	
Not married	0.94 (0.73 to 1.21)	0.627	1.07 (0.72 to 1.60)	0.736	0.47 (0.26 to 0.87)	0.016	1.35 (0.79 to 2.30)	0.277	3.00 (0.39 to 23.4)	0.294
Unknown	1.30 (0.69 to 2.45)	0.409	2.42 (0.94 to 6.25)	0.068	0.89 (0.19 to 4.05)	0.876	1.67 (0.39 to 7.05)	0.488	N/A	
Tumour type										
Others*	1.00		N/A		N/A		N/A		N/A	
Osteosarcoma	1.48 (1.03 to 2.13)	0.033	N/A		N/A		N/A		N/A	
Chondrosarcoma	0.43 (0.28 to 0.66)	< 0.001	N/A		N/A		N/A		N/A	
Ewing's sarcoma	2.52 (1.73 to 3.69)	< 0.001	N/A		N/A		N/A		N/A	
Chordoma	0.18 (0.08 to 0.38)	< 0.001	N/A		N/A		N/A		N/A	
Tumour site										
Lower limb	1.00		1.00		1.00		1.00		N/A	
Upper limb	0.95 (0.71 to 1.25)	0.692	1.21 (0.84 to 1.74)	0.303	0.35 (0.13 to 0.97)	0.043	0.71 (0.40 to 1.29)	0.262	N/A	
Sacropelvic	2.06 (1.61 to 2.63)	< 0.001	2.57 (1.71 to 3.87)	< 0.001	1.34 (0.69 to 2.60)	0.386	1.97 (1.30 to 2.97)	0.001	1.00	
Craniofacial	0.35 (0.19 to 0.65)	0.001	0.45 (0.17 to 1.16)	0.097	0.36 (0.08 to 1.68)	0.194	0.46 (0.15 to 1.40)	0.172	0.05 (0.002 to 1.03)	0.052
Rib, chest wall	1.02 (0.70 to 1.49)	0.905	1.64 (0.76 to 3.54)	0.212	1.05 (0.48 to 2.33)	0.902	0.78 (0.45 to 1.36)	0.380	N/A	
Others†	0.95 (0.71 to 1.27)	0.714	0.77 (0.45 to 1.32)	0.347	0.63 (0.22 to 1.81)	0.394	1.22 (0.77 to 1.94)	0.406	0.35 (0.03 to 3.66)	0.382
AJCC T stage										
T1	1.00		1.00		1.00		1.00		1.00	
T2-3	2.72 (2.18 to 3.40)	< 0.001	2.26 (1.63 to 3.12)	< 0.001	6.15 (3.06 to 12.4)	< 0.001	2.48 (1.69 to 3.65)	< 0.001	3.14 (0.32 to 31.1)	0.328

Continued

Table III. Continued

Characteristics	Metastatic disease		Metastatic osteosarcoma		Metastatic chondrosarcoma		Metastatic Ewing's sarcoma		Metastatic chordoma	
	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value
Unknown	3.04 (2.33 to 3.95)	< 0.001	2.92 (1.94 to 4.40)	< 0.001	4.23 (1.76 to 10.2)	0.001	2.29 (1.48 to 3.54)	< 0.001	0.87 (0.06 to 13.1)	0.923
AJCC N stage										
N0	1.00		1.00		1.00		1.00		1.00	
N1	5.21 (3.52 to 7.71)	< 0.001	7.64 (3.91 to 14.9)	< 0.001	2.98 (0.71 to 12.5)	0.135	4.80 (2.63 to 8.78)	< 0.001	5.66 (0.20 to 159.3)	0.308
Unknown	2.83 (2.03 to 3.96)	< 0.001	3.53 (2.15 to 5.80)	< 0.001	3.59 (1.03 to 12.6)	0.046	2.03 (1.14 to 3.63)	0.017	13.0 (1.00 to 169.5)	0.050

p-values were calculated using multivariable logistic regression.

*Including unspecified malignant bone tumours, miscellaneous malignant bone tumours, odontogenic malignant tumours, and malignant fibrous neoplasms of bone.

†Including short bones, mandible, vertebral column, overlap bones, joints, and cartilage, and bone not otherwise specified.

AJCC, American Joint Committee on Cancer; CI, confidence interval; N/A, not available; OR, odds ratio.

receive radiotherapy as part of their care (adjusted OR = 16.7, 95% CI 11.9 to 23.5). In patients with Ewing's sarcoma, Asian/Pacific Islander patients were less likely to receive radiotherapy compared to Hispanic patients (adjusted OR = 0.40, 95% CI 0.17 to 0.93).

Associations of racial disparities and insurance status with survival. Overall survival was not significantly different based on race in patients with bone sarcomas (Figure 2a). The five-year survival rate for White, Hispanic, Black, and Asian/Pacific Islander patients was 70.3%, 67.3%, 66.6%, and 67.8%, respectively ($p = 0.210$, log-rank). In patients with Ewing's sarcoma, Black patients had worse OS than other racial groups ($p = 0.049$, log-rank test; Supplementary Figure a). In contrast, OS was significantly different based on insurance status (Figure 2b); the five-year survival rates for insured, Medicaid, and uninsured patients were 70.6%, 64.6%, and 67.1%, respectively ($p = 0.001$, log-rank test). Specifically, patients with Medicaid and uninsured patients had worse OS than insured patients with chondrosarcoma ($p = 0.048$, log-rank test; Supplementary Figure b).

After adjusting for age, sex, marital status, tumour type, site, stage, and surgery, we found that race and insurance status were not independently associated with OS in patients with bone sarcoma (Table V). In patients with chondrosarcoma, patients with Medicaid had worse survival compared to those with health insurance (adjusted HR = 1.65, 95% CI = 1.06 to 2.56).

Discussion

Treatment for primary bone sarcomas varies based on patient and tumour factors, and requires a multidisciplinary team for success. Although the National Comprehensive Cancer Network (NCCN) has formulated guidelines for the diagnosis and treatment for patients with a bone sarcoma,⁵ the results of the current study highlight the impact of racial disparities and insurance status on the presentation, treatment, and survival in patients presenting with common bone sarcomas.

Patient characteristics varied according to their race and type of insurance. Chondrosarcomas and Ewing's sarcomas were more common in White patients, whereas osteosarcomas were more common in Hispanic and Black patients. Although previous studies described racial differences in patients with Ewing's sarcoma,^{18–20} differences in patients with osteosarcoma and chondrosarcoma have not been previously described. The higher incidence rate of osteosarcomas in Hispanic and Black patients may account for the younger age at presentation in these racial groups compared to White.

Surgery has become a key component in the treatment of patients with bone sarcoma. The results of the current study show that Black patients were less likely to undergo surgery compared to White patients. Racial disparities have been known to exist in the use of common surgical procedures when comparing Black and White patients. To address this, the USA Department of Health and Human services initiated an action to reduce racial health disparities.²¹ A recent study by Best et al²² showed that racial disparities still exist, and in some cases have even worsened over time. Although there are likely multiple factors which impacted the use of surgery in the treatment of patients with a bone sarcoma in the current study, one factor could be stage at presentation for patients, as previous studies have shown the time from diagnosis to treatment initiation (TTI) is associated with poorer survival patients with these tumours.²³ This could be due to a lack of healthcare access for these patients due to various socioeconomic and geographical factors which were not captured in this study, and may play a role in these healthcare disparities.

Although Hispanic and Black patients were more likely to have metastases at diagnosis in the univariate analysis (Table I), race did not independently affect the likelihood of advanced stage at diagnosis in the adjusted analysis with the type of insurance (Table III). Indeed, higher proportions of Hispanic (456 (42.9%)) and Black patients (196, 37.3%) hold Medicaid than White (422 (15.0%)) and

Table IV. Adjusted odds ratio for surgical treatment in bone sarcomas.

Characteristics	Surgical treatment		Surgical treatment for osteosarcoma		Surgical treatment for chondrosarcoma		Surgical treatment for Ewing's sarcoma		Surgical treatment for chordoma	
	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value
Race										
White	1.00		1.00	-	1.00	-	1.00	-	1.00	-
Hispanic	0.96 (0.76 to 1.22)	0.747	0.82 (0.55 to 1.22)	0.326	0.78 (0.39 to 1.53)	0.467	1.11 (0.74 to 1.66)	0.629	1.13 (0.36 to 3.53)	0.840
Black	0.63 (0.47 to 0.85)	0.002	0.80 (0.51 to 1.25)	0.316	0.31 (0.15 to 0.63)	0.001	0.61 (0.28 to 1.34)	0.220	2.08 (0.24 to 17.9)	0.505
Asian/Pacific Islander	0.65 (0.46 to 0.91)	0.012	1.29 (0.63 to 2.63)	0.484	0.22 (0.10 to 0.47)	< 0.001	0.37 (0.20 to 0.69)	0.002	0.88 (0.21 to 3.64)	0.857
Insurance type										
Insured	1.00		1.00		1.00		1.00		1.00	
Medicaid	0.80 (0.64 to 0.99)	0.041	0.80 (0.55 to 1.15)	0.220	0.48 (0.27 to 0.88)	0.018	0.94 (0.65 to 1.37)	0.749	4.54 (0.72 to 28.8)	0.109
Uninsured	0.77 (0.50 to 1.20)	0.245	0.73 (0.33 to 1.57)	0.417	0.35 (0.14 to 0.85)	0.021	1.44 (0.57 to 3.61)	0.441	13.1 (0.31 to 551)	0.177
Age, yrs										
< 18	1.00		1.00	-	1.00	-	1.00	-	1.00	-
18 to 59	0.59 (0.47 to 0.75)	< 0.001	0.60 (0.41 to 0.88)	0.009	0.75 (0.20 to 2.76)	0.663	0.74 (0.52 to 1.06)	0.100	1.42 (0.21 to 9.87)	0.722
≥ 60	0.44 (0.29 to 0.66)	< 0.001	0.31 (0.15 to 0.65)	0.002	0.75 (0.17 to 3.22)	0.693	0.53 (0.12 to 2.35)	0.404	2.17 (0.25 to 19.0)	0.484
Sex										
Female	1.00	-	1.00	-	1.00	-	1.00	-	1.00	-
Male	1.11 (0.92 to 1.33)	0.287	1.13 (0.82 to 1.56)	0.454	1.19 (0.74 to 1.92)	0.464	1.03 (0.74 to 1.42)	0.879	0.78 (0.33 to 1.85)	0.570
Marital status										
Married	1.00	-	1.00	-	1.00	-	1.00	-	1.00	-
Not married	0.99 (0.78 to 1.25)	0.920	1.07 (0.70 to 1.65)	0.750	1.09 (0.64 to 1.83)	0.760	1.26 (0.75 to 2.13)	0.380	0.98 (0.38 to 2.54)	0.971
Unknown	1.44 (0.75 to 2.77)	0.275	1.79 (0.48 to 6.69)	0.389	1.31 (0.36 to 4.74)	0.678	3.51 (0.73 to 16.8)	0.116	0.23 (0.03 to 1.56)	0.131
Tumour type										
Others*	1.00	-	N/A	-	N/A	-	N/A	-	N/A	-
Osteosarcoma	1.46 (1.04 to 2.06)	0.030	N/A	-	N/A	-	N/A	-	N/A	-
Chondrosarcoma	2.32 (1.60 to 3.36)	< 0.001	N/A	-	N/A	-	N/A	-	N/A	-
Ewing's sarcoma	0.33 (0.23 to 0.47)	< 0.001	N/A	-	N/A	-	N/A	-	N/A	-
Chordoma	2.72 (1.67 to 4.42)	< 0.001	N/A	-	N/A	-	N/A	-	N/A	-
Tumour site										
Lower limb	1.00	-	1.00	-	1.00	-	1.00	-	N/A	-
Upper limb	0.72 (0.53 to 0.97)	0.032	0.94 (0.56 to 1.58)	0.825	0.57 (0.30 to 1.09)	0.090	0.73 (0.42 to 1.24)	0.243	N/A	-
Sacropelvic	0.19 (0.15 to 0.25)	< 0.001	0.13 (0.08 to 0.20)	< 0.001	0.70 (0.36 to 1.39)	0.308	0.17 (0.11 to 0.26)	< 0.001	1.00	-
Craniofacial	0.89 (0.59 to 1.33)	0.566	1.24 (0.52 to 2.97)	0.629	0.99 (0.42 to 2.31)	0.975	0.86 (0.38 to 1.98)	0.728	5.17 (1.55 to 17.2)	0.008
Rib, chest wall	1.01 (0.68 to 1.51)	0.955	0.48 (0.20 to 1.15)	0.099	1.87 (0.77 to 4.56)	0.169	1.28 (0.73 to 2.23)	0.390	N/A	-
Others†	0.87 (0.65 to 1.17)	0.361	0.82 (0.47 to 1.42)	0.472	1.39 (0.55 to 3.50)	0.487	0.70 (0.44 to 1.10)	0.122	6.91 (1.73 to 27.6)	0.006
AJCC T stage										
T1	1.00	-	1.00	-	1.00	-	1.00	-	1.00	-
T2-3	0.90 (0.72 to 1.12)	0.345	1.19 (0.79 to 1.78)	0.408	0.95 (0.52 to 1.75)	0.878	0.76 (0.52 to 1.12)	0.164	0.61 (0.18 to 2.07)	0.429

Continued

Table IV. Continued

Characteristics	Surgical treatment		Surgical treatment for osteosarcoma		Surgical treatment for chondrosarcoma		Surgical treatment for Ewing's sarcoma		Surgical treatment for chordoma	
	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value	Adjusted OR (95% CI)	p-value
Unknown	0.35 (0.28 to 0.45)	< 0.001	0.26 (0.17 to 0.41)	< 0.001	0.23 (0.13 to 0.41)	< 0.001	0.76 (0.49 to 1.18)	0.217	0.12 (0.04 to 0.38)	< 0.001
AJCC N stage										
N0	1.00	-	1.00	-	1.00	-	1.00	-	1.00	-
N1	0.54 (0.35 to 0.83)	0.005	0.24 (0.12 to 0.52)	< 0.001	0.28 (0.07 to 1.21)	0.089	0.97 (0.52 to 1.81)	0.933	0.02 (0.01 to 0.23)	0.003
Unknown	0.63 (0.44 to 0.91)	0.012	0.56 (0.31 to 1.01)	0.054	1.08 (0.31 to 3.77)	0.903	0.77 (0.41 to 1.44)	0.416	0.34 (0.08 to 1.38)	0.129
AJCC M stage										
M0	1.00	-	1.00	-	1.00	-	1.00	-	1.00	-
M1	0.21 (0.17 to 0.26)	< 0.001	0.26 (0.18 to 0.36)	< 0.001	0.05 (0.03 to 0.10)	< 0.001	0.28 (0.20 to 0.39)	< 0.001	0.19 (0.02 to 1.56)	0.123

*Including unspecified malignant bone tumours, miscellaneous malignant bone tumours, odontogenic malignant tumours, and malignant fibrous neoplasms of bone.

†Including short bones, mandible, vertebral column, overlap bones, joints, and cartilage, and bone not otherwise specified.

AJCC, American Joint Committee on Cancer; CI, confidence interval; N/A, not available; OR, odds ratio.

Asian/Pacific Islander patients (67 (19.6%)) in our dataset ($p < 0.001$; Supplementary Table iii). These data indicate that the type of insurance has a major impact on cancer screening; the insured individuals are more likely to go for a cancer screening, which may contribute to the detection of bone sarcoma at an earlier stage. As expected, larger tumours and tumours of pelvis, sacrum, and coccyx were independent predictors of advanced stage at diagnosis, which was consistent with the published reports.^{24–26}

The impact of insurance status and presentation with metastatic disease has been reported by previous studies.^{18,27} In our study, patients with Medicaid and those without medical insurance were more likely to present with metastatic disease at the time of diagnosis. However, if patients are able to receive adequate health insurance, the risk of presenting with advanced stage disease could be mitigated, as highlighted by a study by Ko et al.⁹ In that study, the authors noted that non-Hispanic Black, Hispanic, and American-Indian or Alaskan Natives had a higher risk of presenting with locally advanced breast cancer, however if these patients had health insurance, the risk of presenting with locally advanced disease was cut in half. Smartt et al²⁷ identified the impact of insurance disparities on patients with bone and soft-tissue sarcomas and found results similar to our study, however they did not consider the tumour type and location when analyzing the patients' outcome. In the current study, we found that patients with chondrosarcoma covered by Medicaid had a worse outcome when compared to patients with health insurance. In addition, these patients were less likely to undergo surgery. Since the receipt of surgery strongly affects the prognosis of chondrosarcoma due to its refractoriness to chemotherapy and radiotherapy,^{28–31} worse prognosis in patients with Medicaid should be attributed to lower likelihood of receipt of surgery. However, having Medicaid remained an

independent predictor of worse survival after adjusting with receipt of surgery. Further investigation with other factors including surgical quality (i.e. resection margins) and hospital-related factors (i.e. academic or non-academic) may provide better understanding of survival impact of insurance type.³² When using Medicaid insurance status as a marker for other socioeconomic status, Medicaid is a 'safety-net' programme and allows patients to enrol in coverage after a diagnosis of cancer. As such, these patients are likely presenting with advanced stage of disease, with patients with Medicaid insurance known to have delays in care.^{33–36}

In addition to racial disparities in the use of surgery, previous studies have shown the impact of race on the outcome of patients with head and neck cancer,³⁷ lung cancer,³⁸ as well as brain cancer.³⁹ Previous studies had shown that Black patients had worse survival compared to White patients, however if the patients underwent the same treatment protocols, these survival differences were mitigated.^{37–39} However, this may not be the case for all types of cancer.^{40,41} In our current study, we did not find an association between overall survival and race in patients with a bone sarcoma. However, instead of racial disparities, the key determinant in survival in patients with bone sarcoma is the interplay between multiple socioeconomic factors not captured in the current study.

The results of the current study should be interpreted considering certain limitations. Although this was a large study of patients with a bone sarcoma, a large proportion of the patients in the SEER database needed to be excluded due to a lack of data. Although we were able to focus on racial disparities and insurance status, we are unable to account for socioeconomic factors that were not captured by the SEER database, which likely have an impact on the outcome of the study. In addition, SEER does not provide information on when the patient

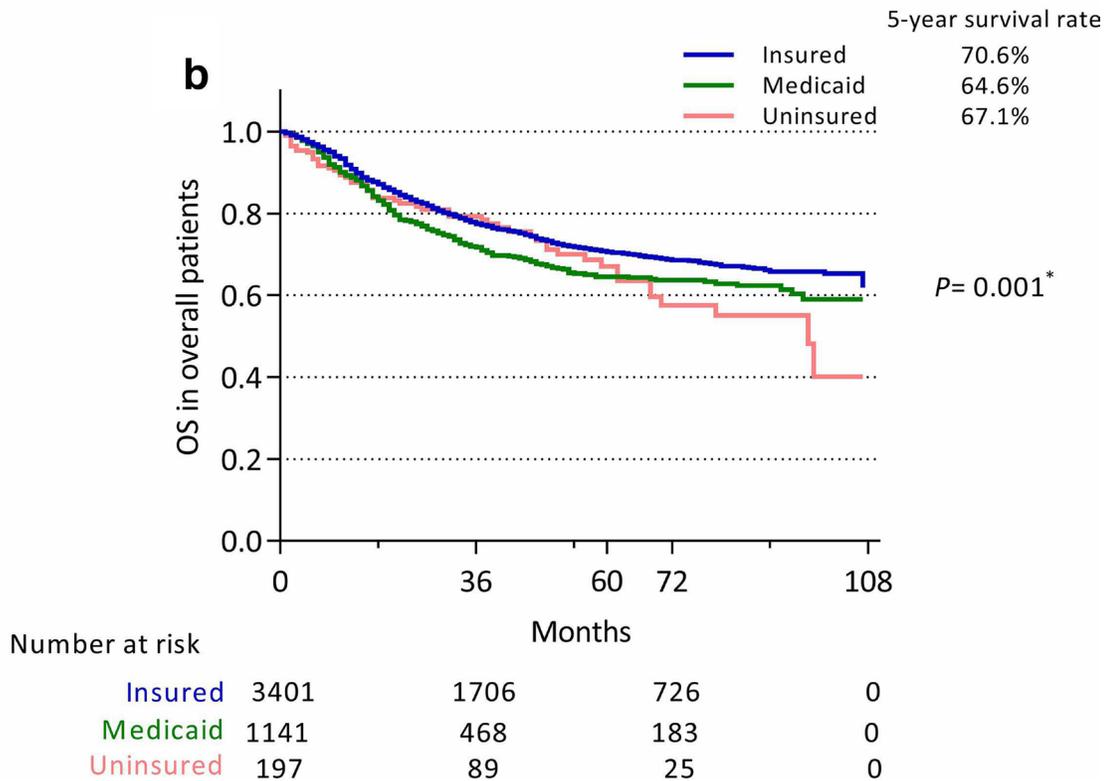
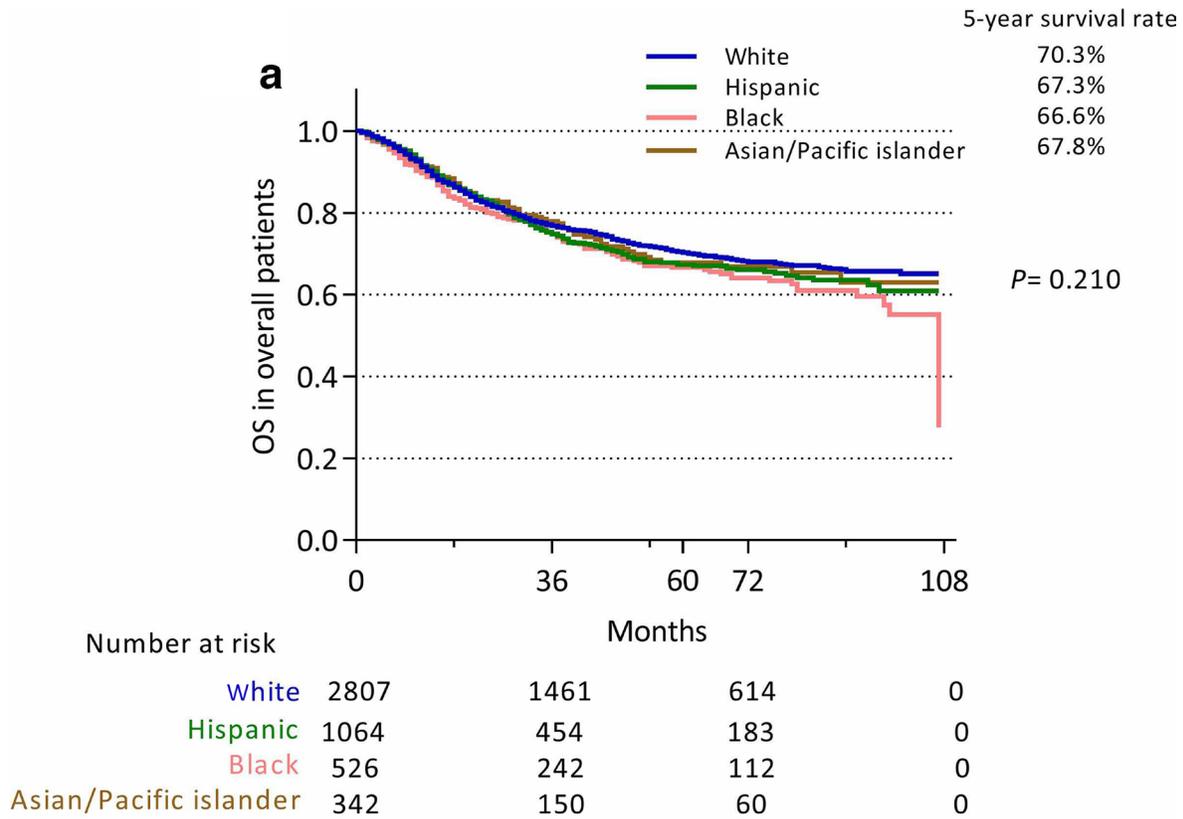


Fig. 2

Kaplan-Meier curves comparing overall survival (OS) by a) race and b) insurance status. p-values were calculated using log-rank test.

Table V. Adjusted hazard ratio for overall survival in bone sarcomas.

Characteristics	Overall survival		Overall survival for osteosarcoma		Overall survival for chondrosarcoma		Overall survival for Ewing's sarcoma		Overall survival for chordoma	
	Adjusted HR (95% CI)	p-value	Adjusted HR (95% CI)	p-value	Adjusted HR (95% CI)	p-value	Adjusted HR (95% CI)	p-value	Adjusted HR (95% CI)	p-value
Race										
White	1.00		1.00		1.00		1.00		1.00	
Hispanic	0.88 (0.75 to 1.02)	0.098	0.93 (0.76 to 1.15)	0.494	0.90 (0.56 to 1.46)	0.675	0.81 (0.57 to 1.16)	0.253	0.86 (0.37 to 2.00)	0.719
Black	0.98 (0.81 to 1.18)	0.840	1.00 (0.79 to 1.27)	0.987	0.91 (0.52 to 1.61)	0.750	1.47 (0.85 to 2.55)	0.171	0.64 (0.17 to 2.44)	0.518
Asian/Pacific islander	1.05 (0.83 to 1.33)	0.685	1.24 (0.91 to 1.70)	0.176	1.48 (0.80 to 2.72)	0.209	0.82 (0.46 to 1.50)	0.525	0.84 (0.25 to 2.82)	0.773
Insurance type										
Insured	1.00		1.00		1.00		1.00		1.00	
Medicaid	1.08 (0.94 to 1.25)	0.279	1.06 (0.87 to 1.28)	0.568	1.65 (1.06 to 2.56)	0.025	1.08 (0.78 to 1.50)	0.636	0.67 (0.26 to 1.74)	0.411
Uninsured	1.08 (0.81 to 1.43)	0.619	0.98 (0.64 to 1.51)	0.935	1.83 (1.00 to 3.34)	0.05	0.76 (0.33 to 1.77)	0.525	1.87 (0.52 to 6.74)	0.340
Age, yrs										
< 18	1.00		1.00		1.00		1.00		1.00	
18 to 59	1.81 (1.56 to 2.10)	< 0.001	1.69 (1.39 to 2.06)	< 0.001	1.65 (0.51 to 5.31)	0.404	1.85 (1.38 to 2.48)	< 0.001	0.39 (0.11 to 1.37)	0.140
≥ 60	2.69 (2.09 to 3.47)	< 0.001	3.10 (2.11 to 4.54)	< 0.001	2.76 (0.82 to 9.34)	0.102	N/A		0.50 (0.12 to 1.99)	0.322
Sex										
Female	1.00		1.00		1.00		1.00		1.00	
Male	1.09 (0.97 to 1.23)	0.140	1.13 (0.96 to 1.34)	0.144	1.26 (0.90 to 1.75)	0.178	1.05 (0.79 to 1.38)	0.748	0.77 (0.43 to 1.38)	0.380
Marital status										
Married	1.00		1.00		1.00		1.00		1.00	
Not married	0.85 (0.73 to 0.98)	0.028	0.83 (0.67 to 1.04)	0.106	1.01 (0.71 to 1.43)	0.968	0.54 (0.38 to 0.79)	0.001	1.31 (0.66 to 2.63)	0.444
Unknown	1.07 (0.75 to 1.52)	0.725	0.84 (0.48 to 1.47)	0.546	1.46 (0.73 to 2.93)	0.290	0.82 (0.28 to 2.37)	0.711	2.37 (0.68 to 8.28)	0.175
Tumour type										
Others*	1.00		N/A		N/A		N/A		N/A	
Osteosarcoma	1.56 (1.25 to 1.95)	< 0.001	N/A		N/A		N/A		N/A	
Chondrosarcoma	0.53 (0.41 to 0.69)	< 0.001	N/A		N/A		N/A		N/A	
Ewing's sarcoma	0.67 (0.52 to 0.86)	0.002	N/A		N/A		N/A		N/A	
Chordoma	0.41 (0.29 to 0.59)	< 0.001	N/A		N/A		N/A		N/A	
Tumour site										
Lower limb	1.00		1.00		1.00		1.00		N/A	
Upper limb	0.86 (0.70 to 1.06)	0.163	1.07 (0.83 to 1.38)	0.601	0.65 (0.39 to 1.09)	0.102	0.58 (0.32 to 1.04)	0.065	N/A	
Sacropelvic	1.56 (1.32 to 1.86)	< 0.001	1.98 (1.53 to 2.57)	< 0.001	1.40 (0.94 to 2.08)	0.097	1.12 (0.78 to 1.62)	0.534	1.00	
Craniofacial	1.26 (0.96 to 1.65)	0.090	1.68 (1.16 to 2.44)	0.006	0.67 (0.32 to 1.38)	0.274	1.17 (0.54 to 2.51)	0.697	0.99 (0.39 to 2.48)	0.979
Rib, chest wall	1.20 (0.92 to 1.56)	0.176	1.21 (0.73 to 2.00)	0.457	0.77 (0.47 to 1.27)	0.312	1.01 (0.63 to 1.61)	0.985	N/A	
Others†	1.23 (1.01 to 1.50)	0.039	1.16 (0.86 to 1.57)	0.328	1.01 (0.58 to 1.79)	0.964	1.12 (0.74 to 1.69)	0.588	1.74 (0.70 to 4.35)	0.235
AJCC T stage										
T1	1.00		1.00		1.00		1.00		1.00	
T2-3	1.58 (1.36 to 1.84)	< 0.001	1.38 (1.12 to 1.70)	0.002	2.09 (1.46 to 3.01)	< 0.001	1.40 (1.00 to 1.97)	0.053	3.26 (1.37 to 7.77)	0.008

Continued

Table V. Continued

Characteristics	Overall survival		Overall survival for osteosarcoma		Overall survival for chondrosarcoma		Overall survival for Ewing's sarcoma		Overall survival for chordoma	
	Adjusted HR (95% CI)	p-value	Adjusted HR (95% CI)	p-value	Adjusted HR (95% CI)	p-value	Adjusted HR (95% CI)	p-value	Adjusted HR (95% CI)	p-value
Unknown	1.34 (1.12 to 1.60)	0.001	1.47 (1.13 to 1.90)	0.004	1.42 (0.86 to 2.33)	0.169	1.23 (0.86 to 1.78)	0.259	0.94 (0.39 to 2.29)	0.895
AJCC N stage										
N0	1.00		1.00		1.00		1.00		1.00	
N1	1.78 (1.37 to 2.31)	< 0.001	2.74 (1.84 to 4.09)	< 0.001	2.47 (1.05 to 5.84)	0.039	1.07 (0.68 to 1.71)	0.764	0.42 (0.06 to 2.95)	0.380
Unknown	1.30 (1.04 to 1.63)	0.022	1.01 (0.72 to 1.41)	0.955	1.03 (0.39 to 2.75)	0.949	1.34 (0.88 to 2.05)	0.177	3.33 (1.32 to 8.42)	0.011
AJCC M stage										
M0	1.00		1.00		1.00		1.00		1.00	
M1	3.47 (3.01 to 4.00)	< 0.001	3.51 (2.89 to 4.25)	< 0.001	7.39 (4.81 to 11.3)	< 0.001	2.51 (1.89 to 3.34)	< 0.001	3.79 (1.17 to 12.3)	0.026
Surgery										
No	1.00		1.00		1.00		1.00		1.00	
Yes	0.46 (0.39 to 0.53)	< 0.001	0.43 (0.34 to 0.54)	< 0.001	0.66 (0.42 to 1.05)	0.079	0.58 (0.43 to 0.79)	< 0.001	0.28 (0.13 to 0.58)	< 0.001

*Including unspecified malignant bone tumours, miscellaneous malignant bone tumours, odontogenic malignant tumours, and malignant fibrous neoplasms of bone.

†Tumour site of others included short bones, mandible, vertebral column, overlap bones, joints, and cartilage, and bone not otherwise specified. AJCC, American Joint Committee on Cancer; CI, confidence interval; HR, hazard ratio; N/A, not available.

obtained insurance and divide the type of insurance that patients have. Likewise, the data in SEER rely on coding of data, and as such this adds a possibility of coding error in these patients.

Overall, racial disparities and insurance status were found to impact the outcome of patients with primary bone sarcomas. Our analysis attempted to control for tumour variables within the confines of the analysis performed, and there are likely factors which are not accounted for in the SEER database which impact overall survival and metastatic disease at the time of presentation. Further work is needed to reduce socioeconomic disparities in the care of patients with bone sarcomas.

Supplementary material



Kaplan-Meier graphs comparing overall survival stratified by racial groups and insurance status, and tables displaying adjusted odds ratios for chemotherapy in bone sarcomas, radiotherapy in bone sarcomas, and association between race and insurance status.

References

- Ferguson JL, Turner SP. Bone cancer: diagnosis and treatment principles. *Am Fam Physician*. 2018;98(4):205–213.
- National Cancer Institute. SEER Cancer Stat Facts: Bone and Joint Cancer. Bethesda, MD. <https://seer.cancer.gov/statfacts/html/bones.html>
- Bandyopadhyay O, Biswas A, Bhattacharya BB. Bone-cancer assessment and destruction pattern analysis in long-bone X-ray image. *J Digit Imaging*. 2019;32(2):300–313.
- Hameed M, Dorfman H. Primary malignant bone tumors—recent developments. *Semin Diagn Pathol*. 2011;28(1):86–101.
- Biermann JS, Chow W, Reed DR, et al. NCCN Guidelines Insights: Bone Cancer, Version 2.2017. *J Natl Compr Canc Netw*. 2017;15(2):155–167.
- Redondo A, Bagué S, Bernabeu D, et al. Malignant bone tumors (other than Ewing's): clinical practice guidelines for diagnosis, treatment and follow-up by Spanish Group for Research on Sarcomas (GEIS). *Cancer Chemother Pharmacol*. 2017;80(6):1113–1131.
- Biermann JS, Adkins DR, Agulnik M, et al. Bone and joint cancer. *J Natl Compr Canc Netw*. 2013;11(6):688–723.
- Patel MI, Lopez AM, Blackstock W, et al. Cancer disparities and health equity: a policy statement from the American Society of Clinical Oncology. *J Clin Oncol*. 2020;38(29):3439–3448.
- Ko NY, Hong S, Winn RA, Calip GS. Association of insurance status and racial disparities with the detection of early-stage breast cancer. *JAMA Oncol*. 2020;6(3):385–392.
- Stram DO, Park SL, Haiman CA, et al. Racial/ethnic differences in lung cancer incidence in the multiethnic cohort study: an update. *J Natl Cancer Inst*. 2019;111(8):811–819.
- Jones CC, Mercaldo SF, Blume JD, et al. Racial disparities in lung cancer survival: the contribution of stage, treatment, and ancestry. *J Thorac Oncol*. 2018;13(10):1464–1473.
- Murphy CC, Wallace K, Sandler RS, Baron JA. Racial disparities in incidence of young-onset colorectal cancer and patient survival. *Gastroenterology*. 2019;156(4):958–965.
- Ellis L, Canchola AJ, Spiegel D, Ladabaum U, Haile R, Gomez SL. Racial and ethnic disparities in cancer survival: the contribution of tumor, sociodemographic, institutional, and neighborhood characteristics. *J Clin Oncol*. 2018;36(1):25–33.
- Cole AP, Lu C, Krimphove MJ, et al. Comparing the association between insurance and mortality in ovarian, pancreatic, lung, colorectal, prostate, and breast cancers. *J Natl Compr Canc Netw*. 2019;17(9):1049–1058.
- No authors listed. ICD-0-3 Coding Materials. National Cancer Institute. <https://seer.cancer.gov/icd-o-3/> (date last accessed 27 April 2022).
- Kong X, Liu Z, Cheng R, et al. Variation in breast cancer subtype incidence and distribution by race/ethnicity in the United States from 2010 to 2015. *JAMA Netw Open*. 2020;3(10):e2020303.

17. **No authors listed.** SEER Combined/AJCC Cancer Staging. National Cancer Institute. <https://seer.cancer.gov/seerstat/variables/seer/ajcc-stage> (date last accessed 27 April 2022).
18. **Jawad MU, Cheung MC, Min ES, Schneiderbauer MM, Koniaris LG, Scully SP.** Ewing sarcoma demonstrates racial disparities in incidence-related and sex-related differences in outcome: an analysis of 1631 cases from the SEER database, 1973-2005. *Cancer*. 2009;115(15):3526–3536.
19. **Lee J, Hoang BH, Ziogas A, Zell JA.** Analysis of prognostic factors in Ewing sarcoma using a population-based cancer registry. *Cancer*. 2010;116(8):1964–1973.
20. **Worch J, Matthay KK, Neuhaus J, Goldsby R, DuBois SG.** Ethnic and racial differences in patients with Ewing sarcoma. *Cancer*. 2010;116(4):983–988.
21. **Koh HK, Graham G, Glied SA.** Reducing racial and ethnic disparities: the action plan from the department of health and human services. *Health Aff (Millwood)*. 2011;30(10):1822–1829.
22. **Best MJ, McFarland EG, Thakkar SC, Srikumaran U.** Racial disparities in the use of surgical procedures in the US. *JAMA Surg*. 2021;156(3):274–281.
23. **Ogura K, Fujiwara T, Healey JH.** Patients with an increased time to treatment initiation have a poorer overall survival after definitive surgery for localized high-grade soft-tissue sarcoma in the extremity or trunk: report from the National Cancer Database. *Bone Joint J*. 2021;103-B(6):1142–1149.
24. **Bielack SS, Wulff B, Dellling G, et al.** Osteosarcoma of the trunk treated by multimodal therapy: experience of the Cooperative Osteosarcoma study group (COSS). *Med Pediatr Oncol*. 1995;24(1):6–12.
25. **Bielack SS, Kempf-Bielack B, Dellling G, et al.** Prognostic factors in high-grade osteosarcoma of the extremities or trunk: an analysis of 1,702 patients treated on neoadjuvant cooperative osteosarcoma study group protocols. *J Clin Oncol*. 2002;20(3):776–790.
26. **Matsunobu A, Imai R, Kamada T, et al.** Impact of carbon ion radiotherapy for unresectable osteosarcoma of the trunk. *Cancer*. 2012;118(18):4555–4563.
27. **Smartt AA, Jang ES, Tyler WK.** Is there an association between insurance status and survival and treatment of primary bone and extremity soft-tissue sarcomas? A SEER Database Study. *Clin Orthop Relat Res*. 2020;478(3):527–536.
28. **Healey JH, Lane JM.** Chondrosarcoma. *Clin Orthop Relat Res*. 1986;204:119–129.
29. **Gelderblom H, Hogendoorn PCW, Dijkstra SD, et al.** The clinical approach towards chondrosarcoma. *Oncologist*. 2008;13(3):320–329.
30. **Nazeri E, Gouran Savadkoobi M, Majidzadeh-AK, Esmaeili R.** Chondrosarcoma: An overview of clinical behavior, molecular mechanisms mediated drug resistance and potential therapeutic targets. *Crit Rev Oncol Hematol*. 2018;131:102–109.
31. **Fujiwara T, Kaneuchi Y, Stevenson J, et al.** Navigation-assisted pelvic resections and reconstructions for periacetabular chondrosarcomas. *Eur J Surg Oncol*. 2021;47(2):416–423.
32. **Fujiwara T, Ogura K, Healey J.** Greater travel distance to specialized facilities is associated with higher survival for patients with soft-tissue sarcoma: US nationwide patterns. *PLoS One*. 2021;16(6):e0252381.
33. **Bradley CJ, Given CW, Roberts C.** Late stage cancers in a Medicaid-insured population. *Med Care*. 2003;41(6):722–728.
34. **Koroukian SM, Bakaki PM, Raghavan D.** Survival disparities by Medicaid status. *Cancer*. 2012;118(17):4271–4279.
35. **Liederbach E, Sisco M, Wang C, et al.** Wait times for breast surgical operations, 2003-2011: A report from the National Cancer Data Base. *Ann Surg Oncol*. 2015;22(3):899–907.
36. **Naghavi AO, Echevarria MI, Grass GD, et al.** Having Medicaid insurance negatively impacts outcomes in patients with head and neck malignancies. *Cancer*. 2016;122(22):3529–3537.
37. **McDermott JD, Eguchi M, Morgan R, et al.** Elderly Black non-Hispanic patients with head and neck squamous cell cancer have the worst survival outcomes. *J Natl Compr Canc Netw*. 2020;19(1):57–67.
38. **Wolf A, Alpert N, Tran BV, Liu B, Flores R, Taioli E.** Persistence of racial disparities in early-stage lung cancer treatment. *J Thorac Cardiovasc Surg*. 2019;157(4):1670–1679.
39. **Bytnar JA, Lin J, Shriver CD, Zhu K.** Racial differences in brain cancer characteristics and survival: an analysis of SEER data. *Cancer Causes Control*. 2019;30(12):1283–1291.
40. **Hardy D, Du DY.** Socioeconomic and racial disparities in cancer stage at diagnosis, tumor size, and clinical outcomes in a large cohort of women with breast cancer, 2007-2016. *J Racial Ethn Health Disparities*. 2021;8(4):990–1001.
41. **Acuna-Villaorduna AR, Lin J, Kim M, Goel S.** Racial/ethnic disparities in early-onset colorectal cancer: implications for a racial/ethnic-specific screening strategy. *Cancer Med*. 2021;10(6):2080–2087.

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Funding statement:

- The authors disclose receipt of the following financial or material support for the research, authorship, and/or publication of this article: the National Natural Science Foundation of China (Grant number: 81872179; Recipient: Wangjun Yan).

ICMJE COI statement:

- M. T. Houdek is a board member of the Musculoskeletal Tumor Society Fellowship Committee.

Data sharing:

- All data were extracted from the Surveillance, Epidemiology, and End Results Program (<https://seer.cancer.gov/>).

Acknowledgements:

- We thank Dr. Jianrong Zhang in the Department of General Practice, Faculty of Medicine, Dentistry and Health Sciences, University of Melbourne, Melbourne, Australia and Dr. Kai Deng in Southwest Medical University, Luzhou, China, for providing their professional viewpoints on this paper.

Ethical review statement:

- Ethical approval was waived for this study because of the de-identified information of the patients in the public Surveillance, Epidemiology, and End Results Program.

Open access funding

- The open access funding for this study was from the National Natural Science Foundation of China (Grant number: 81872179).

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