Socioeconomic and Health Care Coverage Disparities in Children, Adolescents and Young Adults with Sarcoma

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Abstract

Background: Socioeconomic and health care coverage disparities are established as poor prognostic markers in adults with sarcoma, but few studies examine these differences among pediatric, adolescent and young adults (AYA). This study examines the association between socioeconomic status (SES), insurance status, and disease presentation among children and AYA patients with sarcoma. Methods: This is a retrospective cohort study of patients aged 0-25 years with bone or soft tissue sarcoma from the National Cancer Database. SES assignments were based on estimated median income and education-level. Patient demographics and clinical factors were compared by SES and insurance status. Multivariate logistic regression models were fitted to determine adjusted odds ratios of SES and insurance status on metastatic disease or tumor size [?]5cm at time of presentation. Results: In a cohort of 9112 patients, 2932 (32.1%) had low, 2084 (22.8%) middle, and 4096 (44.9%) high SES. For insurance status, 5864 (64.3%) had private, 2737 (30.0%) public, and 511 (5.6%) were uninsured. Compared to high SES, patients with low SES were more likely to have metastatic disease (OR=1.16, p=0.03) and tumors [?]5cm (OR=1.29, p<0.01). Compared to private insurance, public and no insurance were associated with metastatic disease (OR=1.35, p<0.01 and OR=1.32, p=0.02) and increased tumors [?]5cm (OR=1.28, p<0.01 and OR=1.67, p<0.01). Conclusions: SES disparities exist among children and AYA patients with sarcoma. Low SES and public or no insurance are associated with advanced disease at presentation. Further studies are needed to identify interventions to improve earlier detection of sarcomas in at-risk children and young adults.

Introduction:

Sarcomas comprise a heterogenous group of soft tissue and bony mesenchymal tumors and account for 12-15% of pediatric cancer diagnoses.[1,2] These include soft tissue sarcomas such as rhabdomyosarcoma (RMS) and non-rhabdomyosarcoma soft tissue sarcoma (NRSTS), as well as bone tumors including osteosarcoma and Ewing sarcoma. Sarcomas typically present with a mass or pain, and can arise anywhere in the body, and as a result, can present with a wide range of symptoms.[3] Successful treatment of pediatric sarcomas requires prompt diagnosis, multimodal therapy, multidisciplinary teams, and specialized care centers[4–7], however some patients do not have access to high-quality care, or may not be brought to the health system until the cancer has progressed to an advanced stage.

Racial, ethnic, and socioeconomic status (SES) disparities in cancer care are well-documented within the adult population.[8–12] Previous research also suggests that racial and ethnic differences exist regarding delays in presentation and outcomes for sarcomas in the pediatric population.[13–15] While differences in tumor biology may exist among certain ethnic groups[16,17], social and cultural barriers also affect access to care and patient outcomes. Insurance status, for example, is associated with survival disparities, where uninsured

and Medicaid patients are more likely to present with advanced-staged cancer.[18] This disparity has been demonstrated across multiple cancer types, including Hodgkin[18] and non-Hodgkin lymphoma, and bladder and thyroid cancers.[19] This trend is even more prominent in cancer types detectable with early screening such as breast cancer[20] and melanoma[21], where lack of regular medical care and preventive screening can delay diagnosis and treatment.

Children and many AYA patients are dependent on parents and guardians functionally and financially, increasing the complexity of studying health disparities in this population. In addition, young adults are often still dependent on their parents' insurance. As a result, the impact of SES and insurance status on the pediatric and AYA population is less clear. The current study looks to examine the national associations of SES and insurance status on the degree of advanced disease at diagnosis in children and young adults with sarcoma using the National Cancer Database.

Methods:

Data Source and Study Population:

The National Cancer Database (NCDB) is a clinical database for adult and pediatric oncology patients that is sponsored by the American Cancer Society and American College of Surgeons.[22,23] Over 1500 centers contribute hospital-level data, capturing approximately 70% of the annual cancer incidence nationally. The NCDB contains information on patient demographics, insurance status, staging, treatment, and patient outcomes. This study was deemed exempt from IRB review due to the de-identified nature of the dataset.

From the NCDB (2004-2015), patients aged 0-25 years diagnosed with sarcoma based on ICD-O-E codes were included in the study. This cohort included all sarcoma histological subtypes as detailed in Appendix A. Patients missing data on insurance status and median household income and educational status, which were used to determine SES, were excluded from this study.

Classification of Characteristics and Outcomes

Our exposures of interest were SES and insurance status. Insurance status was categorized as having private, public, or no insurance. The NCDB uses US Census Data to provide population-level estimates of median household income and educational status based on zip code of residence. The database adjusts for inflation and defines educational status as the percentage of adults in a zip code region that did not graduate from high school. Patients were grouped into three tiers of SES (highest, middle, and lowest) as determined by their geographical residence. The highest SES category reflected patients with an estimated median household income of [?]$48000 and living in a well-educated geographic region (<13% of adult residents not graduated from high school). The lowest category had an estimated median household income of < $48000 and lived in a less-educated geographic region ([?]13% of adult residents not graduated from high school). The middle category was composed of discordant income and educational status (e.g. high income in a less-educated geographic region).

Our primary outcomes were two binary variables for the presence metastatic disease and increased tumor size ([?] 5 cm) at diagnosis. Metastatic disease was defined as patients with clinical or pathological metastases according to American Joint Committee on Cancer (AJCC) clinical and pathologic staging.[24] Increased tumor size was defined as [?] 5cm at the time of a patient's diagnosis.

Statistical Analysis

Demographic and clinical characteristics were compared between SES tiers using descriptive statistics. SES was compared with patient demographic, social, and clinical factors using Chi-square test for categorical variables and Kruskal-Wallis for continuous variables. For the outcomes of interest, unadjusted odds ratios were obtained from logistic regression models to determine independent risk factors. Variables for multivariate logistic regression were chosen based on backwards stepwise logistic regression for a $p<0.05$ as well as clinically relevant confounders determined by author consensus. The final multivariate model was adjusted for sex, age, race, residential area, distance to facility, SES, and insurance status. Collinearity was checked

using variance inflation factor, and there was no evidence of covariance between independent variables in our final model. Significance was considered for p-values <0.05. All statistical analyses were performed using STATA SE 15.1 (Stata Corp., College Station, Texas).

Results

A total of 9112 pediatric and AYA sarcoma cases were included in this study. The demographic and clinical characteristics of the population are shown in Table 1. The majority of patients were white (75.9%) and non-Hispanic (80.1%) with a median age at diagnosis of 17 years. Primary tumors were most commonly in the extremities (39.1%). Of the total study population, 2932 (32.1%) patients were classified as having low SES, 2084 (22.8%) with middle SES, and 4096 (44.9%) with high SES. The low SES category had a higher proportion of black (26.4%) and Hispanic patients (21.7%) as compared to the high SES category.

There were significant differences in the proportion of patients in each SES category who presented with advanced disease at diagnosis. For patients with low SES, 18.6% had metastatic disease compared to 16.9% in the middle SES category and 15.7% in the high SES (p<0.01). A similar trend was found with increased tumor size, where 53.1% of low SES, 51.6% of middle SES, and 48.4% of high SES patients presented with a tumor that was [?]5cm (p<0.01). The median time to treatment was 5 days (IQR 0-20). Only low SES was associated with an increased median time to treatment of 6 days (IQR 0-22, p<0.01). Most patients received chemotherapy (60.1%), 44.4% received radiation therapy, and 71.8% underwent surgery. More patients in the highest SES category lived in a metropolitan area compared to low SES (90.4% vs. 74.9%, p<0.01) and less than 60 miles from the facility where they received treatment (82.5% vs. 74.4%, p<0.01).

The results of the univariate and multivariate analysis are presented in Table 2. In the univariate analysis, black race was significantly associated with an increased risk of tumor size [?]5cm compared to white (OR=1.36, p<0.01), but not for the presence of metastatic disease. Significant univariate predictors of metastatic disease included low SES (OR=1.23, p<0.01), public or no insurance (OR=1.34, p<0.01 and OR=1.42, p<0.01, respectively). Low and middle SES were significantly associated with an increased risk of a tumor [?]5 cm (OR=1.37, p<0.01 and OR=1.23, p<0.01, respectively) along with both public and no insurance (OR=1.34, p<0.01 and OR=1.92, p<0.01, respectively). Additionally, odds ratios increased with age, with patients in the 13-25-year category 1.96 times more likely to present with metastatic disease and 1.34 times more likely to present with a tumor [?]5 cm.

After accounting for significant variables of interest, our multivariate analysis showed that low SES and public and no insurance remained significant predictors of advanced disease at diagnosis. Compared to high SES, patients with low SES had a 16% increase in the odds of presenting with metastatic disease (p=0.03), and 29% increased odds of presenting with tumor size [?]5cm (p<0.01). Middle SES showed a 17% increase in odds of tumor size [?]5cm compared to high SES (p=0.01), but no difference in metastatic disease (p=0.37). Compared to private insurance, public insurance was associated with metastatic disease (OR= 1.35, p<0.01) and increased tumor size (OR=1.28, p<0.01). Additionally, lack of insurance was associated with increased odds ratios of presenting with metastatic disease (OR=1.32, p=0.02) and larger tumor size (OR=1.67, p<0.01).

Discussion

The results of this study demonstrate that SES and insurance-related disparities in cancer burden exist among children and AYA patients diagnosed with sarcoma. After adjusting for confounders, our multivariate analysis revealed that SES and insurance status were independently associated with advanced disease at presentation. The presence of distant metastasis and larger tumors upon diagnosis are poor prognostic factors for adult and pediatric sarcoma patients.[3,25] Therefore, the socioeconomic disparities in disease presentation found in our study have important implications for patient prognosis and our results highlight the need to improve access to medical care and early detection.

These findings are consistent with previous studies that have found that low SES, participation in public health insurance programs, and lack of insurance are associated with staging disparities and inferior

survival across many pediatric cancers including thyroid cancer[26], retinoblastoma[27], leukemia[28,29], and sarcoma.[30]However, these studies utilize different approaches in evaluation of SES and insurance status. In a study by Penumarthy et al., public insurance was found to be a risk factor for advanced disease at diagnosis and worse survival for pediatric and AYA patients with bone and soft tissue sarcomas in the University of San Francisco California Cancer Registry. In their study, public insurance was used as a proxy for low income as, under California state law, public insurance is only available to individuals with low-income.[30] Given that public insurance eligibility varies by state, our findings add to the existing evidence by examining trends nationally and separating other markers of SES from insurance status.

Our results support the need for improved access to medical care and early detection in higher risk socioeconomic populations. Researchers have hypothesized that presentation with later stage disease is partially responsible for the association between SES and insurance-related survival disparities.[31,32] In studies of adult cancer, low SES and poor insurance have been found to result in delays in diagnosis and time to treatment, which may account for the associated advanced disease and poorer outcomes.[33,34] Our data suggest that a similar pattern may be occurring in the pediatric and AYA population. Successful sarcoma management may be challenging and requires a coordinated multidisciplinary approach at a center with expertise in the management of these rare cancers. Another possible mechanism for inferior survival may be related to limited access to cancer care centers, resulting in inferior treatment.

Limitations of our study include those inherent from a large centralized database, including its retrospective nature, missing data, and errors in coding. The NCDB does not provide individual level estimates of income and education level, and our measure of SES is an approximation based on zip codes of residence. In addition, other demographic variables not measured in this database may be surrogates for SES. In pediatric populations, SES and insurance status are dependent on a parent or guardian rather than the individual. This may present unmeasured confounders that we are unable to account for.

Conclusion

Our findings add to the growing evidence that SES and health care coverage are strong prognostic factors that influence the stage of presentation for children and AYA with sarcoma, potentially reflecting decreased access to care for these patients and families. Further studies are needed in order to evaluate mechanisms that underlie these disparities so that interventions may be designed to improve outcomes in this population.

Conflict of Interest

The authors have no conflicts of interest.

Data Availability Statement

The data that support the findings of this study are available from the National Cancer Database at https://www.facs.org/quality-programs/cancer/ncdb, reference number [22]. These data were derived from the following resources available in the public domain:https://www.facs.org/quality-programs/cancer/ncdb/publicaccess.

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