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Sarcomas other than Kaposi sarcoma occurring in immunodeficiency: interpretations from a systematic literature review

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Abstract

Purpose of review—In immunodeficiency, an increased sarcoma risk is confirmed for Kaposi's sarcoma. Whether rates of other sarcoma subtypes are elevated in the setting of immunodeficiency is not known. We therefore reviewed published case reports on HIV/AIDS patients and organ transplant recipients with sarcomas. For comparison, we assessed sarcomas in the U.S. general population using Surveillance Epidemiology End Results (SEER) data.

Findings—One hundred seventy-six non-KS sarcomas were identified, 75 in people with HIV/AIDS and 101 in transplant recipients. Leiomyosarcomas (n=101) were the most frequently reported sarcomas, followed by angiosarcomas (n=23) and fibrohistiocytic tumors (n=17). Leiomyosarcomas were reported with two age peaks, in children and young adults. Epstein-Barr virus (EBV) was detected in the tumor cells in 85% and 88% of leiomyosarcomas in HIV-infected people and transplant recipients, respectively. Angiosaromas and fibrohistiocytic tumors were most frequently reported in males. Among kidney transplant recipients, 20% of sarcomas arose at the site of an arteriovenous fistula. In comparison, leiomyoscarcomas, angiosarcomas, and fibrohistiocytic tumors comprised 16.9%, 3.8%, and 18.7% of sarcomas in the U.S. general population.

Summary—Leiomyosarcoma and angiosarcoma may occur disproportionately in immunodeficiency. Leiomyosarcomas appear etiologically linked to EBV while angiosarcomas might be correlated with an arteriovenous fistula. Additional studies are necessary to understand the contribution of immunodeficiency to the etiology of these sarcomas.

Keywords

HIV; AIDS; organ transplant; cancer; EBV; HIV; sarcoma. HHV-8

Introduction

A dramatic eruption of an otherwise rare cancer, Kaposi sarcoma (KS), in young men in San Francisco heralded the HIV epidemic in the US. The incidence of KS rose from approximately 0.5 per 100 000 people in 1975 to a peak of 33.3 in the late 1980s to the early 1990s [1]. Reports describing KS in solid organ transplant recipients also linked this

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sarcoma with immunosuppression [2]. Later, systematic analyses of population-based registry data clearly demonstrated a several thousand-fold increased KS risk in people with AIDS compared to the general population [3].

The incidence of as many as twenty distinct cancers is elevated in people with HIV infection and in solid organ transplant recipients [4], and the commonality in the spectrum of malignancies in these two immunosuppressed conditions is striking. Risk is most increased for cancers with an infectious etiology, including those caused by oncogenic viruses such as human herpesvirus 8 (HHV-8, linked to KS), human papillomavirus (HPV, linked to anogenital cancers), Epstein-Barr virus (EBV, linked to non-Hodgkin and Hodgkin lymphomas), hepatitis B and hepatitis C viruses (HBV and HCV, linked to liver cancer), and Merkel cell polyomavirus (MCPyV, linked to Merkel cell carcinoma) [4].

Sarcomas are diverse and include more than fifty distinct histological subtypes originating from connective tissues, including fibrous tissue, muscle, bone, cartilage, fat, blood vessels, and nerves [5]. However, the entire group of sarcomas together account for less than 1% of all malignancies. While sarcomas occur across all ages, there are age-associated peaks in incidence specific for distinct entities. Rhabdomyosarcoma and Ewing sarcoma are both pediatric cancers, the former occurring in the first decade of life and the latter in the second decade. In contrast, liposarcomas and leiomyosarcomas most often occur in adults [6].

The rarity, diversity, and age-dependency of specific sarcomas make it difficult to single out histological subcategories that are increased in people with immunosuppression, particularly if these increases are much smaller than observed for KS. In a prior analysis of population-based HIV/AIDS and cancer registry data, Biggar and colleagues demonstrated an excess risk of leiomyosarcomas in children with AIDS [7]. Over the past decade, other reports have described an excess of leiomyosarcomas in children with HIV or following organ transplantation [8–10].

Whether other sarcoma subtypes are affected by immunosuppression is unclear. Even with large population-based registries [11], the sensitivity of identifying an elevated risk of uncommon cancers depends upon the magnitude of the association, the incidence of these cancers in the general population, and the categorization of cancer types. Linkage-based studies of HIV and cancer have typically categorized cancers by site. As sarcomas can arise within multiple organs, this approach could have masked an elevated risk of sarcomas.

For these reasons, case reports remain an important source for understanding the spectrum of cancers in immunocompromised populations and ascertaining clues into cancer etiology. To identify sarcomas that occur frequently in immunosuppressed populations and describe the characteristics of these sarcomas, we systematically reviewed published case reports and case series to catalog sarcomas reported in people with HIV infection and recipients of solid organ transplants.

Literature review approach

We conducted a literature review using PubMed and references cited within published articles to compile case reports of all sarcomas, excluding KS, in people with HIV or a solid organ transplant. Our PubMed search strategy included combinations of the following keywords: organ transplantation, malignancy, de-novo malignancy, immunosuppression, immunodeficiency, HIV, AIDS, and sarcoma. We also searched with names of specific sarcoma subtypes (e.g., leiomyosarcoma, osteosarcoma). We excluded studies of recipients of bone marrow transplants or congenital immunodeficiency syndromes, and articles not published in English.

Sarcomas were categorized into widely accepted histologic subtypes. To identify over- or under-representation of specific sarcoma subtypes in people with immunodeficiency, we compared the relative proportions of these subtypes reported in people with HIV or solid organ transplants with the distribution in the general population. General population counts for sarcoma subtypes were obtained from the U.S. Surveillance, Epidemiology, and End Results database (SEER 17 regions, 1974–2008) [12] using *International Classification of Diseases for Oncology* morphology codes [13].

In cataloging leiomyosarcoma cases, case reports with a diagnosis of EBV-associated smooth muscle tumor were categorized as leiomyosarcomas. We also compared the age distribution of case reports of leiomyosarcomas in people with immunodeficiency with leiomyosarcomas in SEER (histology code 8890, regardless of site). Finally, for comparison, we also captured case reports of leiomyomas in HIV-infected people and transplant recipients, which were analyzed separately.

Sarcomas in Immunodeficiency

A total of 176 cases of sarcoma (other than KS) were reported in people with HIV (n=75) or recipients of solid organ transplants (n=101) (Table 1). In both immunodeficient populations, more than half of the reported sarcomas were leiomyosarcoma. Fifty-four leiomyosarcomas occurred in solid organ transplant recipients [10, 14–57], and 47 occurred in people with HIV [58–86]. Thus, 57.8% of all sarcomas reported in immunodeficient hosts were leiomyosarcomas. In comparison, in the U.S. general population (SEER data), only 16.9% of non-KS sarcomas were leiomyosarcomas (Table 1).

The sex distribution of leiomyosarcoma cases was similar in people with HIV and transplant recipients. Of the 81 leiomyosarcomas for which EBV status was reported, EBV was detected in the tumor cells in 85% and 88% of cases in people with HIV and transplant recipients, respectively. EBV positive leiomyosarcomas were detected across all age groups. Among leiomyosarcoma cases with HIV infection, 94% had a prior AIDS diagnosis. Sixty-three percent of transplant-related leiomyosarcomas were reported among kidney recipients [22–47].

Figure 1a is a representation of the age distribution of case reports of leiomyosarcomas in HIV-infected individuals, transplant recipients, and the U.S. general population. In the general population, the number of leiomyosarcoma cases rises steeply with age, with 72% of cases occurring in people aged 50 years and older. In contrast, in people with HIV or transplant-associated immunodeficiency, leiomyosarcomas were reported across all age groups. Thirty-six percent of reports in HIV-infected individuals and 24% in transplant recipients were in children aged 0–9 years. In HIV-infected cases this proportion diminished between the ages 10–29 years and peaked again, with 34% of cases occurring in 30–39 year olds. In transplant recipients, this second peak (19%) occurred in people aged 50 years or older.

Angiosarcomas also comprised a higher proportion of sarcomas in immunocompromised populations compared to the general population (6.7% of sarcomas in HIV-infected people and 18.0% in transplant recipients, vs. 3.8% in SEER; Table 1). We identified 23 cases of angiosarcomas, of which 5 cases were in HIV-infected adults (4 males and 1 female) [87–90]. The remaining 18 cases were reported in kidney recipients [91–106], and the vast majority were in males (n=16) (Table 2). The median age at diagnosis of angiosarcomas in transplant recipients was 43 years (range 24–71 years). The time to diagnosis following transplant ranged from 2–18 years.

Eleven angiosarcomas in kidney transplant recipients (61%) arose at the site of an arteriovenous fistula. Of interest, an arteriovenous fistula was also the site of dermatofibrosarcoma protuberans (a subtype of fibrohistiocytic tumor) in three kidney recipients [107–109]. Thus, 14 sarcomas reported in kidney recipients occurred at an arteriovenous fistula, representing 20% of sarcomas in kidney recipients and 14% of sarcomas in transplant recipients overall.

Seventeen cases of fibrohistiocytic tumors were reported in immunodeficient individuals (Table 1) [107–120]. These include one case of angiomatoid fibrous histiocytoma in an HIV-infected person [107]; 5 cases of dermatofibrosarcoma protuberans [108–111]; 2 cases of atypical fibroxanthoma [112, 113]; and 9 cases of malignant fibrohistiocytoma [113–120], 8 of which were in transplant recipients. Fourteen of these seventeen cases occurred in males. In comparison, 18.7% of sarcomas in the U.S. general population were fibrohistiocytic tumors.

Eight cases of rhabdomyosarcoma were reported (Table 1), of which the majority (n=6) were in people with HIV [84; 121–124]. Three of these rhabdomyosarcoma cases occurred in children, but both cases in transplant recipients were adults, one a 21 year-old female liver recipient [125] and the other a 67 year-old male kidney recipient [126]. Only a limited number of other sarcomas were described in the case literature (Table 1) [127–152].

Fifty leiomyomas were reported in HIV-infected individuals [9, 60, 70, 73–75, 153–182] or transplant recipients [183–192], of which 80% were in HIV-infected individuals. Among HIV-infected people, 93% of leiomyomas occurred after an AIDS diagnosis. The male-to-female ratio of leiomyoma cases in HIV-infected individuals (2:1) differed from that in transplant recipients (1:1.5), and also differed from the ratio for leiomyosarcomas in HIV-infected individuals (1:1.5). Among transplant recipients, 80% of leiomyomas were reported in kidney recipients [183–190]. Among the 24 leiomyomas for which EBV status was reported, EBV was detected in 100% of transplant-related leiomyomas but only 53% of HIV-related leiomyomas (Table 3). We queried leiomyoma and leiomyosarcoma reports for CD4 counts as a measure of immunosuppression. As shown in Figure 1b, most cases were in people with CD4 counts below 50 cells/mm^3.

Interpretations from case literature review

The objective of the current study was to identify subtypes of sarcomas other than KS that occur in immune suppressed populations. Given the rarity and broad diversity of sarcomas, our hypothesis was that modestly elevated risks for rare sarcomas might have been missed in large registry-based studies of HIV-infected people and solid organ transplant recipients. However, unusual individual cases or series of cases occurring in such individuals might have prompted clinicians to publish descriptive reports. Our literature review identified three sarcoma subtypes that are often reported in people with immunosupression: leiomyosarcoma, angiosarcoma, and fibrohistiocytic tumors. Of these, only leiomyosarcoma and angiosarcoma were disproportionately represented in both HIV and transplant compared to the general population (Table 1).

The most commonly reported sarcoma in HIV-infected people and transplant recipients was leiomyosarcoma, with distinct age peaks in very young children, in young adults (for HIV), and older individuals >50 (for transplant). EBV is not required for transformation, since leiomyomas and leiomyosarcomas that arise in immunocompetent individuals are not associated with EBV [193], but our review suggests that immunosuppression-related leiomyosarcomas are highly associated with EBV across all ages. Of interest, EBV is also detected in muscle tumors in individuals with congenital immunodeficiency syndromes [194]. When present in leiomyosarcoma, EBV infection occurs prior to the clonal

proliferation of tumor cells [15], but most case reports indicate that EBV LMP-1, an important oncoprotein [195], is not detectable [15,5,192,]. EBV infection requires the expression of the cell surface receptor CD21. While some studies have demonstrated CD21 expression in smooth muscle tumors [9, 60], other studies have not [188]. EBV infection of rare mesenchymal cells has been reported in immunocompetent hosts [196], and high circulating levels of EBV during immunosuppression may more readily allow such infections.

The vast majority of HIV-infected individuals reported with leiomyosarcoma had AIDS, and CD4 cell counts were typically low among both leiomyosarcoma and leiomyoma patients. These observations highlight the importance of immunosuppression in facilitating transformation of mesenchymal cells. EBV appears to be less often detected in leiomyomas than in leiomyosarcomas in the setting of HIV infection. While the proportion of leiomyomas with missing data for EBV was large, the available reports point to a need for a systematic study of the molecular features of leiomyomas and leiomyosarcomas arising in immunosuppressed people.

Angiosarcomas are also frequently reported in people with immunodeficiency, raising the possibility of a viral etiology. Early studies, based solely upon amplification of DNA by polymerase chain reaction PCR, suggested the presence of HHV-8 genome in angiosarcomas [197], but more recent studies that have utilized *in situ* methods to detect HHV-8 RNA or proteins have been mixed [87, 198–200]. Our review found a male predominance of angiosarcomas (20 male vs. 3 female reported cases). These sarcomas were also more frequently reported following kidney transplant than other organ transplants. While this predominance may merely reflect that kidneys are the most commonly transplanted organ, it may instead be a clue to intrinsic factors that affect the development of angiosarcoma.

Along these lines, an interesting feature of the angiosarcomas reported in kidney recipients is their apparent predilection to occur at the site of an arteriovenous fistula used for hemodialysis. Angiosarcomas have also been reported in hemodialysis patients who have not received a transplant [201*]. Inert foreign material can induce inflammation that might lead to the development of sarcomas, including angiosarcomas [202]. The milieu around an arteriovenous fistula is characterized by chronic venous stasis, altered capillary pressures, and tissue hypoxia, which may induce neovascularisation and cellular proliferation. Angiosarcomas have also been documented within areas of lymphedema, as occur in the Stewart-Treves syndrome [203], and they are a known complication of radiation exposure [204].

Fibrohistocytic sarcomas reported in case literature in people with HIV and organ transplants included dermatosfibrosarcoma protuberans, atypical fibroxanthomas and malignant fibrous histocytomas. Our review identified 17 fibrohistocytic sarcomas, the majority (n=14) of which occurred in males. However, these sarcomas represented only 5% and 13% of all sarcomas in people with HIV and in transplant recipients, respectively (Table 1).

An important limitation of our study is that it is solely based upon case reports. There is an inherent publication bias in these reports, in that investigators are more likely to report novel cases, such as those with unusual features, involvement of EBV, or occurrence in children. Therefore, our results should not be interpreted as a direct measure of risk associated with immunodeficiency or a complete characterization of the sarcomas that arise in immunosuppressed people. Nonetheless, the disproportionate representation of leiomyosarcomas that we observed is consistent with an increased risk for this sarcoma

subtype observed in previous studies of immunosuppressed populations [7,10]. Case report literature reviews also cannot incorporate a central pathology review or common standards for pathology diagnosis, which might introduce biases in the classification of sarcomas. The major strength of this report is our systematic review of the published literature, which provided a unique approach to assess rare sarcoma subtypes occurring in people with HIV and transplant recipients.

Conclusions

In summary, leiomyosarcomas and leiomyomas were frequently reported in HIV-infected people and organ transplant recipients, supporting an increased risk of this sarcoma in immunosuppressed people and highlighting the importance of EBV. The disproportionate occurrence of angiosarcomas may likewise reflect increased risks associated with immunodeficiency. Furthermore, among transplant recipients, angiosarcomas are most commonly reported in people with kidney transplants and might be correlated with an arteriovenous fistula. Additional studies are necessary to understand the contribution of immunodeficiency to the etiology of these sarcomas.

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Key Points

- Other than Kaposi sarcoma, a limited number of other sarcomas, including leiomyosarcoma and angiosarcoma, may occur disproportionately in immunodeficient populations.
- EBV is commonly associated with leiomyosarcoma in people with HIV and transplant recipients. EBV is also frequently detected in leiomyomas from these populations. In HIV-infected people, leiomyosarcomas are reported in two age peaks, for children and young adults.
- Among transplant recipients, angiosarcomas are reported exclusively in kidney transplants and have a high predilection for the site of an arteriovenous fistula.
- Angiosarcomas and fibrohistocytic tumors in transplant patients show a notable male predominance.

Figure 1a:

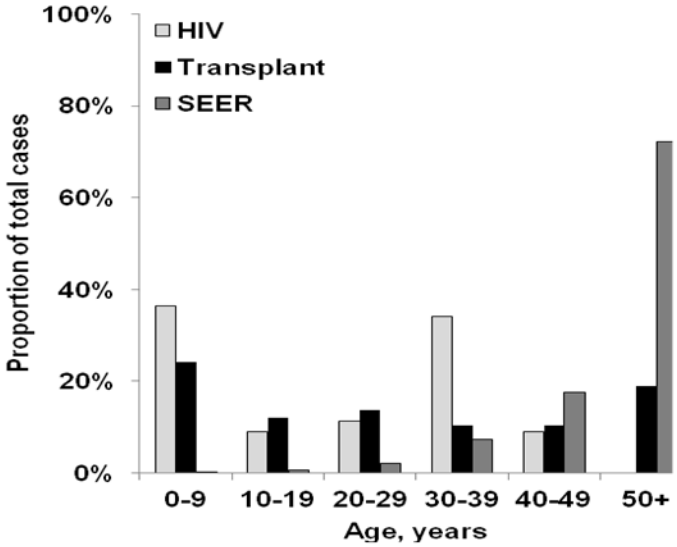

Figure 1b:

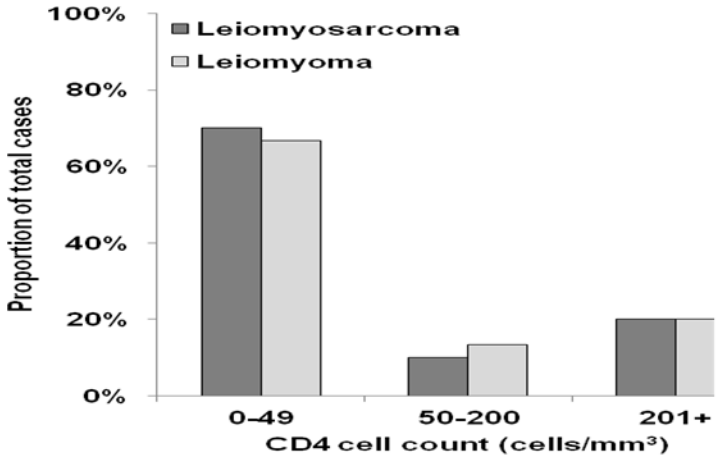

Figure 1.

Figure 1a: Age distribution of leiomyosarcoma cases reported in HIV-infected individuals and transplant recipients, and in the US general population

Figure 1b: Distribution of CD4 counts in HIV-infected individuals with leiomyosarcoma (data based upon 20 cases) or leiomyoma (data based upon 15 cases)

Table 1

Distribution of sarcoma subtypes in cases reported among HIV-infected individuals, transplant recipients, and the general U.S. population

	HIV		Transplant		SEER	
Sarcoma (ICD-O-3 codes)	**N**	**%**	**N**	**%**	**N**	**%**
Osteosarcomas(9180-9187, 9191-9195, 9200)	1	1.3	0	0	4,510	5.1
Chondrosarcomas(9210, 9220-9221, 9230, 9240-9243)	0	0	0	0	3,707	4.2
Ewing tumor and related sarcomas of bone(9260, 9363-9365)	1	1.3	1	1.0	2,456	2.8
Malignant chordomas(9370-9372)	0	0	0	0	1,191	1.3
Rhabdomyosarcomas(8900-8905, 8910, 8912, 8920, 8991)	6	8.0	2	2.0	3,283	3.7
Fibrosarcomas(8810, 8811, 8813-8815, 8820-8827, 8834-8835, 9150, 9160)	0	0	3	3.0	3,693	4.2
Nerve sheath tumor(9540-9571)	1	1.3	1	1.0	2,516	2.9
Other fibromatous neoplasia (9491-9580)	0	0	0	0	103	0.1
Extrarenal rhabdoid tumor(8963)	0	0	0	0	210	0.2
Liposarcomas(8850-8858, 8860-8862, 8870, 8880, 8881)	2	2.7	2	2.0	8,831	10.0
Fibrohistiocytic tumors(8830-8833, 8836, 9251, 9252)	4	5.3	13	12.8	16,481	18.7
Leiomyosarcoma(8890-8898)	47	62.7	54	53.5	14,943	16.9
Synovial sarcomas(9040-9044)	1	1.3	0	0	2,183	2.5
Alveolar sort parts sarcoma(9581)	0	0	0	0	182	0.2
Angiosarcomas and other blood vessel tumors (9120-9125, 9130-9133, 9135, 9136, 9141, 9142, 9161, 9170-9175)	5	6.7	18	17.8	3,320	3.8
Gastrointestinal stromal tumor(8936)	2	2.7	4	0	4,624	5.2
Interdigitating dendritic cell sarcoma(9757)	2	2.7	1	1.0	20	0
Carcinosarcoma, NOS(8980-8982)	0	0	1	1.0	5,369	6.1
Miscellaneous sarcomas(8587, 8710-8713, 8806, 8840-8842, 8921, 8990, 9373)	2	2.7	0	0	701	0.8
Unspecified sarcomas(8800-8805)	1	1.3	1	1.0	9,928	11.2

Abbreviations: ICD-O-3 International Classification of Diseases for Oncology third edition, NOS not otherwise specified, SEER Survival, Epidemiology, and End Results

Table 2

Characteristics of angiosarcoma cases occurring in kidney transplant recipients.

Characteristic	N (%) or median (range)
Total	18 (100)
Sex , n (%)	
Female	2 (11)
Male	16 (89)
Age in years, median (range)	43 (24–71)
Geographic location of study, n (%)	
North America	6 (33)
Europe	9 (50)
Australia	1 (6)
Asia	2 (11)
Located at the arteriovenous fistula, n (%)	
Yes	11 (61)
No	7 (39)
Months from transplant to angiosarcoma, median (range)	84 (24–216)

Table 3

Characteristics of leiomyoma cases occurring in HIV-infected individuals and transplant recipients.

	HIV/AIDS N (%)	Transplant N (%)
Total	40 (100)	10 (100)
Sex		
Female	13 (33)	6 (60)
Male	26 (67)	4 (40)
Missing	1	0
Age, years		
0–9	15 (38)	1 (10)
10–19	8 (20)	0 (0)
20–29	3 (8)	0 (0)
30–39	12 (30)	2 (20)
40–49	1 (3)	3 (30)
50+	1 (3)	4 (40)
EBV status of tumor		
Positive	9 (53)	7 (100)
Negative	8 (47)	0 (0)
Missing	23	3
AIDS status		
Present	28 (93)	-
Absent	2 (7)	-
Missing	10	-
Organ transplanted		
Liver	-	0 (0)
Lung	-	1 (10)
Kidney	-	8 (80)
Heart	-	1 (10)
Multiple	-	0 (0)