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Title: Primary Osseous Leiomyosarcoma: A Comparison with its Somatic Soft-Tissue Counterpart

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ABSTRACT BODY:

Background: Leiomyosarcoma is a rare malignant tumor with two orthopedic variants: osseous and somatic soft-tissue (SST). While the SST variant has been well-described, there are less than 150 reported cases of osseous leiomyosarcoma, and no studies to date have directly compared these two tumors.

Questions/Purposes: This study hopes to elucidate differences in tumor characteristics and patient outcomes between these two sarcomas. The secondary objective is to evaluate metastasis of osseous and SST leiomyosarcoma, including the sites most commonly effected.

Patients and Methods: All patients of the senior author diagnosed with either an osseous or SST leiomyosarcoma between 2004 and 2014 were retrospectively reviewed. Twenty-six patients (10 osseous, 16 SST) with at least one-year follow-up were identified. All 26 patients underwent en bloc resection, and the mean time to latest follow-up was 3.2 ± 1.9 years. Demographic, diagnostic, histologic, treatment, and outcomes details were collected for all patients. Paired student t-tests and Fisher's exact tests were used to compare variables between the two variants.

Results: Patients with primary osseous leiomyosarcomas were significantly younger at the time of diagnosis than the SST group ($p = 0.04$) (Table). Mitotic rate was significantly lower in the osseous group ($p = 0.04$), but metastatic disease was present in 70% of the osseous cases compared to 31% of the SST group. Using a log-rank test, differences in metastatic-free survival curves were significant ($p = 0.03$) (Figure). Of those with metastases, the lung was the most common site (71% osseous, 60% SST, $p = 1.0$). Distant bone involvement was present in 71% of metastatic cases in the osseous group but none in the SST group ($p = 0.03$). Metastases to the liver were not present in any osseous patients but were present in three SST cases ($p = 0.04$). No statistically significant differences were found in mean tumor size, Charleston Comorbidity Index, or time to metastasis, local recurrence, or death. The primary lesion was more commonly located in the lower extremity ($n = 20$) than upper extremity ($n = 6$). Eighty-eight percent of all patients (23/26) had received radiation and/or chemotherapy, and the chemotherapy regimens were similar between groups.

Conclusion: In our series, osseous leiomyosarcomas had a significantly lower mitotic rate than SST variants but were paradoxically more likely to metastasize. Only the osseous variant demonstrated distant bone metastasis and did so at an abnormally high 71% rate. This indicates a distinct form of biologic activity, which may be attributable to a different cell of origin, biomarkers, or response to therapeutic regimens. The main limitation of this paper is the small sample size due to the rarity of these two tumors.

Table. Demographic, diagnostic, histologic, treatment, and outcomes details

	Osseous (n = 10)	SST (n = 19)	P values
Demographics			
Gender			
Male	5 (50)	6 (38)	0.69
Female	5 (50)	10 (62)	
Age	47 ± 13	59 ± 16	**0.04***
Diagnostic			
Charleston Comorbidity Index	5.0 ± 3.9	3.2 ± 3.3	0.22
Tumor size (cm^3)	213 ± 298	308 ± 440	0.50
Mitotic rate (per 10 high-power fields)	9.6 ± 5.8	17.8 ± 8.3	**0.04***
Treatment			
Chemotherapy (%)	9 (90)	6 (38)	**0.01***
Neoadjuvant (%)	4 (40)	5 (31)	0.69
Adjuvant (%)	7 (70)	5 (31)	0.11
Radiation (%)	5 (50)	13 (81)	0.19
Combined chemo- and radiation (%)	5 (50)	5 (31)	0.43
Outcomes			
Re-operations (%)	3 (30)	6 (38)	1.0
Local recurrences (%)	2 (20)	3 (19)	1.0
Mean time to local recurrence (years)	2.1 ± 0.8	4.1 ± 4.0	0.56
Metastatic involvement (%)	7 (70)	5 (31)	0.11
Lungs (%)	5 (71)	3 (60)	1.0
Distant bone (%)	5 (71)	0 (0)	**0.03***
Liver	0 (0)	3 (60)	**0.04***
Mean time to metastasis (years)	1.0 ± 1.1	2.6 ± 3.1	0.27
Deaths (%)	3 (30)	3 (19)	0.64
Mean time to death (years)	3.5 ± 1.4	4.0 ± 4.1	0.86

*** = statistically significant**

SST = somatic soft-tissue

Figure. Kaplan-Meier curve depicting metastatic-free survival for osseous and somatic soft-tissue (SST) leiomyosarcoma variants