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Title: Periosteal osteosarcoma; The University of Florida Experience

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Level of Evidence: IV

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Abstract:

Background:

Periosteal osteosarcoma is a rare tumor. It is estimated to represent less than 2% of all osteosarcomas. It is an intermediate grade malignancy, with a lower propensity for metastasis compared to conventional, high-grade medullary osteosarcoma. While resection for local control of this malignancy is accepted as the cornerstone of treatment, strategies differ as to whether adjuvant chemotherapy is administered in patients with this diagnosis. Survival is relatively high compared to other commoner subtypes of osteosarcoma, and the use of adjuvant chemotherapy has been found in several studies not to be associated with increased survival.

Questions/Purposes:

The purpose of this descriptive study is to describe the experience of the University of Florida with management of patients with this rare malignancy with respect to the characteristics of this patient group, and their survival outcomes. We also sought to assess if any patient or treatment related factors were associated with improved survival.

Patients and Methods:

This descriptive study is a retrospective case series. The prospectively collected musculoskeletal oncology database of the University of Florida was queried to identify patients who had been diagnosed with and managed for periosteal osteosarcoma. We identified 19 patients who had been managed during the period 1970 to 2007. Of the 19 patients, 1 had less than one year follow up and was excluded from statistical analysis The study population comprised 6 males and 13 females, with a mean follow up of 11.2 years (±7.5). Kaplan-Meier survival analysis was performed and the log-rank test was used to compare survival between patient subgroups. Multivariate analysis using Cox proportional-hazards regression was used to assess patient and treatment related factors for an effect on survival.

Results:

The 18 patients analyzed presented at a mean of 20.2 years of age (±10.1) and were followed for a mean of 10.4 years (±7.4). 10 year overall survival was 84% (Figure 1). The commonest site involved was the distal femur (5), followed by the proximal tibia (4) and the pelvis (3). 13 of 18 patients had tumor that were high grade, and 11 of the 18 patients received chemotherapy. 17 of 18 patients underwent wide resection of the primary tumor for local control, while the last patient's resection was read as a marginal resection. Medullary involvement was noted

in 9 patients. 2 patients (11.1%) developed metastasis (lung at 19 months and proximal humerus at 94 months), and they were treated with chemotherapy and resection of metastasis.

Univariate analysis (ie: log-rank test) comparing overall survival of patient subgroups did not reveal any significant differences when the following factors were assessed (Figure 2): Use of chemotherapy, p=0.47, Presence of medullary involvement, p=0.66, Tumor grade, p=0.13. Multivariate analysis using Cox regression analysis did not identify any of the factors assessed to be significant predictors.

One patient was diagnosed with mesenchymal chondrosarcoma at the same site of her periosteal osteosarcoma 81 months later, and died 8 months after that diagnosis. Interestingly, in the 33 patient series from The Rizzoli Institute[1], one patient died from mesenchymal chondrosarcoma at 128 months but at a site different from the primary periosteal osteosarcoma.

Conclusions:

Making strong recommendations regarding the management of periosteal osteosarcoma is challenging owing to its rarity and the limited and varied data. The 10-year survival of 84% in this study is comparable to that in the literature[1-5]. No significant difference was found in survival between patients who did and did not receive chemotherapy also echoes the findings of the 2 largest studies on the topic by Grimer et al[2] and Cesari et al[1]. Wide resection remains the preferred mode of surgical treatment. Studies on this disease neither support nor detract from the use of chemotherapy and patients should be informed regarding the silence of the literature regarding this issue. In our study, all patients with high-grade tumors were recommended to receive chemotherapy. The main limitations of this study are its retrospective nature and the small population size, and the according inability to detect differences in survival.

Figure 1: Kaplan-Meier curve of overall survival of study population

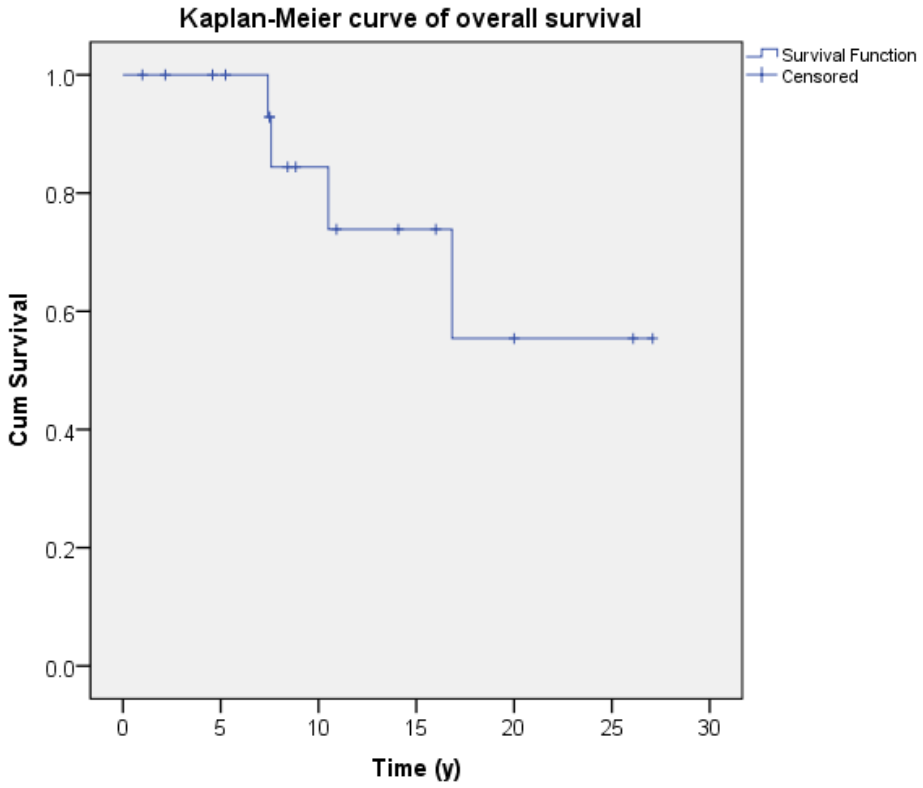
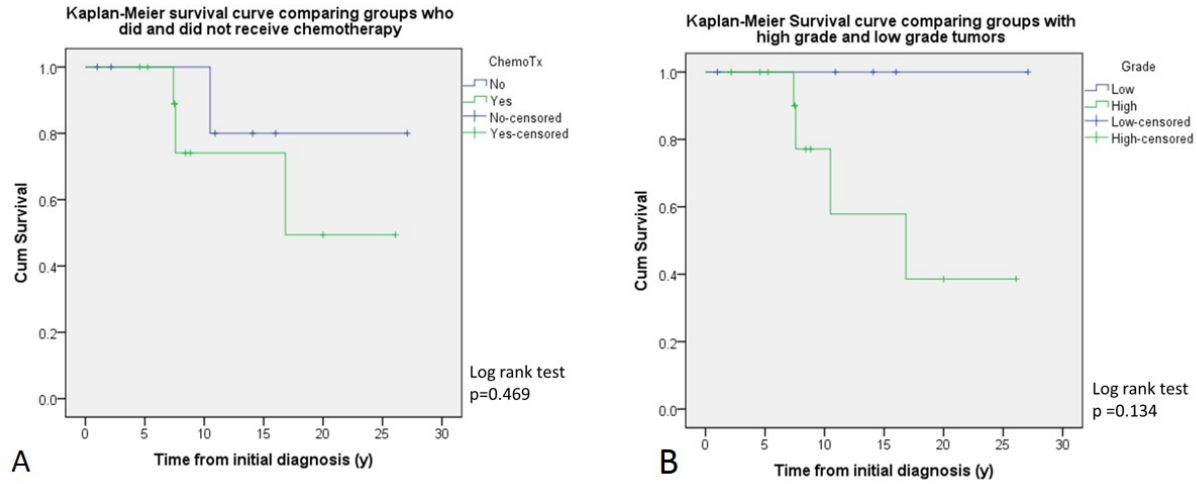


Figure 2: Kaplan-Meier curves comparing overall survival of (A) Patients who did and did not receive chemotherapy, and (B) Patients with high grade and low grade tumors



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