CASE PRESENTATION

History and Physical Examination

The patient is a 21-year-old male high school custodian who suffered acute onset of right knee pain one Friday in December after a long day of work driving his pickup truck and clearing snow. The pain persisted over the weekend becoming severe enough on Sunday night that it woke him from sleep. He was evaluated by his primary care physician the following morning and found on exam to have medial joint line tenderness. A possible intra-articular injury was suspected and the patient was referred to a local orthopedic surgeon for evaluation. Examination revealed pain with flexion of the knee beyond 90 degrees. Careful evaluation of the medial aspect of the knee revealed pain with palpation of the proximal tibia just adjacent to the medial joint line, but no actual joint line tenderness. Due to this discrepancy, radiographs were obtained, which showed a subtle radiolucent lesion in the proximal anteromedial tibial metaphysis. This was recognized as an abnormality and an MRI was ordered to more clearly define the lesion.

Imaging

Based on the imaging studies, what is your diagnosis and how would you proceed with the evaluation of this patient?

Differential Diagnosis

Benign lesions:
- Giant Cell Tumor
- Chondroblastoma
- Aneurysmal Bone Cyst

Malignant lesions:
- Osteosarcoma
- Lymphoma
- Ewing Sarcoma

Histology

A CT guided needle biopsy was obtained. Based on the histology, what is your diagnosis?

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**Pathology and Further Workup**

Histologic evaluation of CT guided core biopsies of the lesion showed a spindled and epithelioid neoplasm with marked cytologic atypia and atypical mitotic forms (Figure 6). Close examination of the tumor showed, a pale-pink extracellular matrix that is diagnostic of unmineralized bone or osteoid (Figure 7). The combination of the malignant cytology and the presence of osteoid are diagnostic of high-grade osteosarcoma. MRI showed the mass to involve the medial portion of the proximal tibial metaphysis and epiphysis. Staging studies, including computed tomography scanning of the chest and whole body bone scan, were done which did not show any evidence of metastatic disease to the lungs or other bony sites. The patient was referred to medical oncology for induction of chemotherapy for osteosarcoma. The patient underwent three months of neo-adjuvant chemotherapy and then underwent complete restaging examinations followed by operative resection for local control of the tumor. (Figures 8 and 9)

**Surgical Treatment**

**Resection**

The tumor was approached through a medial incision extending from the medial femoral condyle to the mid portion of the tibia. The biopsy site was kept in continuity with the resected specimen. The saphenous vein was identified, dissected and protected. The medial head of the gastrocnemius and soleus were retracted posteriorly. The soleus arch was divided while protecting the popliteal vessels. The pes anserine tendons and semimembranosus were divided, and the medial meniscotibial ligament was divided anteriorly as far as the patellar tendon. The patellar tendon was preserved and retracted anteriorly. The arthrotomy was extended posteriorly to the midline where the popliteal vessels were again identified and protected. Care was taken to preserve the entire medial meniscus. The anterolateral tibia was then exposed by raising a large anterolateral flap. Then, using C-arm control, K-wires were placed outlining the length and extent of our osteotomy. (Figures 10 and 11) An oscillating saw and osteotomes were used to make the osteotomy. We had direct visualization of the articular surface at this point and placed the osteotome just medial to the tibial spines, preserving the insertion of the cruciates by undercutting of the tibial spines. The osteotomies were completed, the
specimen was passed off, and curettings were taken for a deep margin. (Figures 12 and 13)

**Reconstruction**

An osteoarticular allograft had been thawed and cultured. After getting confirmation of negative margins, we sculpted the graft to fit the resection defect. (Figure 14) The meniscus from the allograft was trimmed, leaving a small rim peripherally. Sutures were then used to reattach the patient’s native menisco-tibial ligament to the remaining rim of allograft meniscus. An 8-hole narrow large fragment combi-plate was used to hold the graft in place with two screws in compression across the allograft-host junction. (Figure 15) The medial arthroscopy was then closed, and the pes anserine tendons, MCL and medial retinaculum were repaired. The subcutaneous tissues were then closed in layers, the wounds were dressed, and the patient was placed in a long posterior splint.

**Results**

The patient was placed into a long leg cast and was given instructions of touchdown weight bearing. He was allowed to begin range of motion exercises approximately 6 weeks after surgery.

**Discussion**

Patients with tumors about the knee present with a variety of complaints that may initially be attributed to injuries or other causes. A careful history and physical exam can often lead to the proper diagnosis. Malignant lesions generally have pain that is present at rest or at night, while benign lesions and
traumatic injuries are more likely to be painful with activity. Identification of a palpable mass is a key finding on physical exam and pain at the site of the mass or bony lesion must be distinguished from joint line tenderness, tendonitis, or apophysitis pain.

Sports related injuries are increasingly managed with arthroscopy of the knee. Recent reports have described tumors about the knee that were not initially recognized and were treated as sports injuries – sometimes with arthroscopy. One report showed that ultimate oncologic therapy was altered in 60% of these cases. The most common cause of these erroneous diagnoses and treatments were insufficient or poor quality radiographs.a

In this report we describe a case of a proximal tibial osteosarcoma presenting with complaints suspicious for an intra-articular injury. Careful and appropriate management by the referring primary care physician and orthopaedic surgeon led to timely diagnosis and treatment. A novel approach to resection and reconstruction of this tumor is also described.

**General Principles of Treatment**

Current management of osteosarcoma requires a multidisciplinary approach including a team of orthopedic oncologists, pediatric oncologists, radiologists and pathologists. The goal of treatment is to achieve local control of the tumor and systemic control of the disease. Chemotherapy has been the main factor in the increased survival rates seen over the last several decades for patients with osteosarcoma. It is imperative to think of high-grade osteosarcoma as a systemic disease. For example, in patients with apparently isolated osteosarcoma treated with amputation alone, metastatic disease developed in 80-90% within the first two years following surgery. This implies that the vast majority of patients have undetectable micrometastatic disease at the time of initial diagnosis. The addition of adjuvant chemotherapy to treat this micrometastatic disease has led to dramatic improvements in disease free survival rates. Prior to the use of chemotherapy the probability of remaining disease free after amputation for osteosarcoma was less than 20%, the current probability is estimated to be between 65 and 80%.b

**Local Control**

Amputation was once the standard of care for all patients with high-grade osteosarcoma and remains an important tool in the surgeon’s armamentarium. Currently however, the majority of patients who present with osteosarcoma of all grades are treated with limb-salvage surgery. Although randomized trials have not been performed comparing survival rates of patients treated with amputation versus limb salvage, non-randomized retrospective studies do not show a survival advantage to treating with amputation. Furthermore, an increase in local recurrence with limb-salvage surgery has not been shown. With amputation in some cases having poorer functional result than limb salvage surgeryc d, and with the addition of chemotherapeutic agents that are moderately to highly effective against this tumor, limb salvage surgery has become the current standard of care for treatment of most patients who present with osteosarcoma.

**Reconstruction**

The large majority of these tumors present in young patients who are still growing or have recently completed growth, as in this case presentation. The knee area (distal femur and proximal tibia) is the most common site for osteosarcoma, and several options have been developed for reconstruction after resection of a malignancy from these sites. Reconstruction options for various sites must be carefully considered in order to maximize the patient’s result with respect to function and durability. Particularly in the growing child, reconstruction of a defect from a tumor resection can be difficult and fraught with subsequent mechanical, wear-related, and growth-related complications. Osteosarcoma about the knee most commonly does not involve the joint itself, and an intra-articular resection can usually be performed.e

Reconstruction can then be carried out with an osteoarticular allograft, allograft prosthetic composite (APC), or a metallic endoprosthesis. Metallic prostheses are more stable initially and generally will return a patient to function earlier than an allograft. However, concerns about loosening, particle disease and overall implant longevity are a concern in this young patient population.e Allografts require a longer post-operative recovery period but have the advantage of restoring bone stock. The first reported human osteoarticular allograft may have been as early as 1902.e The longevity of articular cartilage in osteoarticular allografts is indefinite, and some patients will ultimately require traditional joint replacement surgery. Other issues with allograft use include malunion or non-union at the osteosynthesis site, allograft fracture, joint laxity and infection.f

In the case of the proximal tibia, allograft reconstructions have the significant advantage of allowing restoration of the extensor mechanism by reattachment of the patellar tendon to the allograft tendon. The resulting extensor mechanism is considered to be superior to the metallic prosthesis, which relies on soft tissue ingrowth into a porous surface on the implant. This type of reconstruction results in an extensor lag that generally does not improve over time.

**Hemicortical resection**

In the case example presented here, a unicondylar wide resection and reconstruction of a high-grade intramedullary osteosarcoma of the proximal end of the tibia is described. This type of resection and reconstruction is not commonly performed for high-grade sarcomas because of the concern for local recurrence. Whereas intramedullary margins have been described for benign and low grade malignant tumors, this case presents the use of this type of resection for a high grade malignant osteosarcoma, because it was felt preoperatively that adequate margins could be obtained.

The question of what distance of normal marrow defines an adequate bony margin for a high grade osteosarcoma has not been adequately described in the literature. Traditionally, a resection of the entire segment of bone containing a 3-5cm margin has been considered adequate.e However, recent evidence from successful treatment of low grade parosteal osteosarcoma without resection of the entire segment (hemicortical resection) and with bony margins closer than previously
required has given credence to the thought that this type of resection may be useful in the treatment of high grade osteosarcoma. Several studies have shown that focal intramedullary involvement in the case of surface osteosarcoma does not increase the risk of local recurrence as long as the margin is tumor-free. Further follow up on this patient will be necessary to determine whether or not there is an impact on local recurrence rate in this case, but if the suggestion from current data on lower grade osteosarcoma holds true in the higher grade cases, there should be no impact. A lengthy discussion was had with the patient about the alternatives to this type of resection, including amputation, and he was made aware of the risks and benefits of this type of resection and reconstruction.

The difficulty in achieving a negative margin must be also be considered in this case. The hemicortical resection described here is technically demanding, and may significantly increase the risk of obtaining an intra-operative cut through bone containing tumor. Adequate and precise imaging both pre and post neo-adjuvant chemotherapy is therefore a prerequisite for any tumor for which this type of reconstruction is being considered. The lack of an intra-operative imaging modality that can identify the exact location of a tumor in the bone (such as MRI) makes this type of resection challenging. This type of resection is therefore not recommended for all tumors in this location, and may only apply to a select subset of tumors such as the one presented here where the anticipated benefit of hemicortical resection outweighs the presumed additional risk. In this case, the described resection allowed preservation of the native cruciate ligaments, which should significantly improve the patient’s functional outcome.

Current reports have demonstrated that proximal tibial osteoarticular allograft reconstruction compare favorably to metal endoprosthetic reconstruction in this location, and probably have improved long term function because of the ability to salvage the patellar tendon attachment. In the described resection, the majority of the functional components of the joint are preserved, including not only the anterior and posterior cruciate ligaments, but also the lateral collateral ligament and greater than 50% of the articular cartilage of the tibial plateau. Presumably this joint preservation will translate into further improvements in graft survival and long-term function. Although it remains to be seen whether there is additional risk in terms of local recurrence with this type of resection, these potential benefits must be considered.

In terms of allograft incorporation, multiple current studies suggest that allograft incorporation in the metaphyseal region is faster when compared to the diaphyseal region. Allograft reconstruction in metaphyseal osteotomies heat at an average of 6 months compared to a range of 9 to 18 months for diaphyseal osteotomies. It is not clear whether this is a function of surface area or some other phenomenon, as at least one study comparing oblique osteotomies, to transverse osteotomies, the former having greater surface area than the latter, failed to show a significant difference in healing times. Nonetheless, the broad metaphyseal surface area of the osteotomy described in this surgical technique may lead to an accelerated healing time in this hemicortical resection as compared to standard osteoarticular reconstructions in this region in which the allograft-host junction is entirely diaphyseal.

**SUMMARY AND FUTURE DIRECTIONS**

The case presented in this article represents an unusual circumstance in which a hemicortical intramedullary resection could be performed for a high-grade malignant osteosarcoma of the proximal tibia. This was possible in large measure by the astute, early diagnosis made by the referring orthopaedic surgeon and should remind all practicing orthopedic surgeons to have a high index of suspicion in evaluating young patients with atypical knee pain. It is our hope that with current modern chemotherapy resulting in 95% or greater tumor necrosis as in this case, that the resection described above with allow adequate local control without any increased risk of local recurrence, while offering significantly improved durability and function compared to other standard reconstruction options. Careful follow up over the next several years will be required to demonstrate this. The future direction for this research, of which this patient represents the first step, should be to set up a prospective trial for patients that could reasonably undergo this type of procedure to see if statistically significant differences exist between the described resection and standard resections.

**References**


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