High-grade Intramedullary Osteosarcoma of the Talus: Case Report

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This is a presentation of a rare malignant tumor that uncommonly affects the bones of the foot. The following case involves a malignancy which usually presents during childhood or adolescence presenting much later in life. Physicians treating the foot and ankle must include osteosarcoma in their list of differential diagnoses to exclude when vague symptoms involving deep pain and edema present. It is critical to prevent misdiagnosis and delayed treatment with this form of tumor. (The Journal of Foot & Ankle Surgery 46(6):480–483, 2007)

Osteosarcoma is rarely associated with the bones of the foot (1–4). Presenting symptoms most frequently involve vague, deep-seated tenderness and edema (3–5). Frequently, the patient’s tendency to ignore symptoms and the lack of clinical suspicion by the physician result in a delayed diagnosis. Because of the natural history of this tumor, expedient diagnosis and treatment are recommended.

Case Report

An 81-year-old white woman presented on May 3, 2005, with a history of approximately 4 years of unexplained ankle pain and edema. Within the past 8 to 10 weeks, she noted an increase in her symptoms. She described her ankle pain as being “deep in the joint,” with nothing in particular aggravating the condition. She could not recall any inciting events, and nothing seemed to relieve her pain. Up to this time, she had been an active individual who denied any trauma to the area.

Her medical and surgical history included diagnosis and treatment of squamous cell carcinoma of the skin over the left thigh. This was removed by excisional biopsy 4 years before presentation. She had a history of spinal degenerative joint disease, and she underwent spinal fusion for this condition approximately 4 years before presentation.

The patient was currently taking adendronate sodium for osteoporosis. She admitted that regional steroid injections were used for postoperative pain control for 2 months after her spinal fusion, but she denied the use of oral corticosteroids. She had no known drug allergies.

The patient’s family history was not significant for cancer or any other disease. The patient was an active woman who was an avid golfer and denied the use of alcohol, tobacco, or recreational drugs.

The review of systems was positive for generalized “aching” of bilateral lower extremities “for years.” She related infrequent numbness and tingling of the lower extremities as being associated with back pain she had developed after spinal fusion. All other systems displayed unremarkable findings.

Physical examination showed a palpable mass at the anterior aspect of the right ankle joint and minimal associated edema localized to the area. There was a decrease in dorsiflexion. Neurovascularly, she was intact, and, despite the restriction of the range of motion at the ankle, there were no functional deficits at the foot or ankle.

Obtained radiographs revealed complete sclerosis of the talus with calcification extending from the neck interiorly (Figure 1). There was no periosteal reaction noted. There was no articular involvement or pathology of the joints of the foot.

A magnetic resonance imaging (MRI) of the foot and ankle was obtained to further evaluate this lesion (Figures 2 and 3). There was a complete void of signal at the talus in the T1, T2, and short time inversion-recovery series. The soft tissue extension at the anterior ankle was also void of signal. The joints remained intact, and there was no collapse of the articular surfaces. There were no cystic changes noted.

During the 2 months before her presentation at Grant Medical Center, the patient had been evaluated by 2 other podiatrists, and a diagnosis of avascular necrosis had been made. A Technetium-99 bone scan had been obtained, which revealed increased tracer uptake only in the right ankle.
Given this unusual presentation, a percutaneous bone biopsy was taken from the talus through an incision in the sinus tarsi region on May 9, 2005. Differential diagnosis included avascular necrosis, metastatic tumor, transient bone marrow edema, and osteitis.

The biopsy was pathologically evaluated at Grant Medical Center on May 17, 2005, and the results were sent to The Mayo Clinic, Rochester, MN, for confirmation (Figures 4 and 5). The evaluation revealed necrotic bone with apparent focal proliferation of osteoblasts. There were no pathological changes seen in the soft tissues sent for evaluation. This osteoblastic lesion was thought to be permeative. On May 23, 2005, the diagnosis of osteoblastic sarcoma of the talus was made.

The patient was then referred to an orthopedic surgeon who specialized in oncology. On June 13, 2005, the patient underwent a below-the-knee amputation. The specimen was evaluated by the pathology department and was consistent with an intramedullary osteosarcoma, osteoblastic type. The tumor displayed some areas with well-differentiated features; however, other sites had characteristics of high-grade osteosarcoma including necrosis. The maximum size of the talus was 5.2 cm, and it involved nearly the entire talus. The final diagnosis was high-grade intramedullary osteosarcoma of the talus.

Several nodular lesions were identified on the patient’s chest radiographs during her preoperative work-up. These pulmonary lesions were feared to be metastasis. A positron emission tomography scan and a spiral computed tomography (CT) scan with intravenous contrast were obtained for further evaluation in May 2005, and the lesions showed no abnormal uptake of fluorodeoxyglucose. Repeat CT scans in August and December of 2005 showed stability of these nodules. The patient’s serum lactate dehydrogenase level was normal. Again, multiple enlarged nodules were noted. The patient’s most recent CT was done in December of 2005, and, at that time, there was no progression of the nodules.

The patient chose to refuse adjunctive chemotherapy. The patient ambulates with a below-the-knee prosthesis and requires regular CT scans to monitor the pulmonary nodules. There is currently no plan to remove these nodules, which are believed to be benign.

In May of 2006, the patient was evaluated for left lower extremity radicular symptoms. A bone scan and MRI revealed a metastasis to the lumbar spine. To date, the patient is being treated with a round of palliative radiation.
Discussion

Osteosarcoma is a rare neoplasm encompassing approximately 2% of all diagnosed tumors (6). Osteosarcoma is the most common malignant, primary bone tumor in pediatric and adolescent patients (5–7). The tumor usually arises from long bones during the second and third decades of life, and diagnosis during the first decade and after the third decade is uncommon. Osteosarcoma reportedly involves the bones of the foot .17% to 2.08% of the time (1, 2).

It is rare for primary osteosarcoma to be associated with the talus. More often, when the bones of the foot are involved, the calcaneus and metatarsal bones are the origin of this tumor. Few cases involving the talus are reported in the literature. Amini and Colacecchi first reported such a case in 1980 (4). Another case was reported in 1988 (8). In a series of 1929 cases of osteosarcoma, the bones of the foot were primarily affected .6% of the time and the talus specifically .15% (1). The authors noted that primary osteosarcoma of the foot seemed to present and behave differently in comparison with osteosarcomas originating from other locations. They found the mean age of these patients to be 33 years old. It was noted that low-grade osteosarcoma of the foot had a better prognosis than osteosarcoma at other sites. High-grade osteosarcoma of the foot had a prognosis and mortality rate similar to those associated with this tumor found at other sites.

This case involved a high-grade osteosarcoma that was diagnosed in the patient’s ninth decade. The location and time of presentation are highly unusual for this tumor. The literature reports that diagnosis can be delayed an average of 28 months after presentation of symptoms (1). The diagnosis for this patient was made approximately 48 months after her initial symptoms developed.

The presentation of osteosarcoma, histologically and radiographically, can be varied (6). Some authors have noted difficulty differentiating osteosarcoma from benign tumors such as fibrous dysplasia and desmoplastic fibroma (9). Macroscopically, this tumor is typically a large (5–10 cm), intraosseous tumor with frequent soft-tissue extension. There are associated mineralized regions of osteoid and cartilage as well as hemorrhagic foci. The diagnosis is usually made from the presence of irregular deposits of osteoid surrounded by bizarre, pleomorphic cells and necrotic debris (10). A subclassification based on histology is frequently used, which includes osteoblastic, chondroblastic, and fibroblastic classes depending on the dominant cell type. Osteoblastic is the most dominant form of osteosarcoma and was diagnosed in this patient after being confirmed by a consulting institution.

Osteosarcoma is difficult to diagnose radiographically because of its unpredictable appearance. It is characterized by being a large lesion because of its rapid doubling rate (20–30 days). Osteosarcoma can be blastic or lytic but usually presents as a combination of these features. Some experts suggest that the predominant feature is fluffy, cloud-like opacities within the lesion. Others characterize this tumor as having ill-defined osteolytic lesions without typical periosteal reactions such as Codman’s triangles or sunburst patterns (3). Joint involvement is thought to be uncommon, and it has been postulated that hyaline cartilage acts as a barrier (11). The radiographic finding for this patient displayed involvement of the entire talus. The appearance could be described as sclerotic from the blastic nature of this tumor. There is soft tissue extension noted at the anterior aspect of the talus.

MRI is reportedly a reliable means by which to quantify the extent of the tumor (12). MRI confirmed that the lesion in this case consumed the entire talus. The plain films and MRI led treating physicians to suspect that the underlying pathology of these vague symptoms was spontaneous avas-
cular necrosis. Osteitis was the misdiagnosis of all 3 talar osteosarcomas in a previous study (1).

The diagnosis of this unsuspected tumor was made because of the bone biopsy that was taken from the talus. The patient was then referred to an orthopedic surgeon who specialized in oncological cases.

Treatment consisted solely of an amputation because the patient refused chemotherapeutic treatment. Currently, the most accepted treatment for osteosarcoma is amputation or wide resection (1, 5, 13). The use of chemotherapy as adjunctive therapy for osteosarcoma is well accepted and encouraged in cases of a high-grade tumor (14–17). The patient refused chemotherapy because of the fear of toxicity.

Typical 5-year survival rates reported for osteosarcoma range from 60% to 80% (18, 19). Metastases most frequently present in the lungs, followed by bones and distal nodes (5).

Conclusion

Osteosarcoma is an uncommon tumor that rarely presents in the bones of the foot. A physician treating vague pain and edema without an inciting episode must include primary osteosarcoma in his or her differential diagnoses. The appropriate course must be taken to promptly rule out this malignant tumor.

References