

Reconstruction Using a Frozen Bone Method for Osteosarcoma of the Talus. A Case Report and Review of the Literature*

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Abstract. *A case of osteosarcoma of the talus is reported. Osteosarcoma of the talus is very rare. The patient is alive and she has been continuously disease free for five years after surgery. This is the first case of osteosarcoma of the talus with reconstruction using a frozen bone method, an autograft containing tumor treated with liquid nitrogen. This is a rare case report of osteosarcoma of the talus without extrainvasion of the talus.*

Osteosarcoma of the talus is extremely rare (1-10). Malignant bone tumors of the talus is sometimes misdiagnosed as aseptic necrosis of the talus or a benign bone tumor at initial visits to a clinic (1-12). Osteosarcoma of the talus has been frequently misdiagnosed as another disease in many patients (1-10). In almost all previously reported cases of osteosarcoma, amputation was necessary (1-3, 5-8), excluding three cases where prognosis was unspecified (3, 4, 9), and the prognosis for cases where amputation was necessary was relatively poor (1, 2, 5, 6, 8, 9). It is essential to diagnose osteosarcoma at an early stage in order to avoid amputation and to achieve a better prognosis.

Case Report

A 15-year-old girl visited our hospital with a two-month history of pain in her right ankle during exercise, without any cause. The range of motion in her right ankle joint was normal and there was no instability. Laboratory investigations were

unremarkable. A plain radiograph revealed ill-defined lucent areas with surrounding sclerotic change in the right talus. Avascular necrosis of the talus was suspected and she was therefore referred to our Department (Figure 1, A and B). Computed tomography (CT) showed geographic osteolytic changes with osteosclerosis in the talus. Partial destruction of the bone cortex was suspected, which suggested inflammatory lesions such as osteomyelitis or posttraumatic change (Figure 2). On T1-weighted magnetic resonance imaging (MRI) (T1WI: TR=300-361 ms; TE=12 ms), the lesion showed a low intensity compared with muscle (Figure 3, A and D). On T2-weighted MRI (T2WI: TR=3000-3176 ms; TE=100 ms), the lesion showed heterogeneous high and low intensities (Figure 3, B and E). On gadolinium-diethylenetriamine pentaacetic acid (Gd-DTPA)-enhanced T1-weighted images, the lesion showed inhomogeneous contrast enhancement (Figure 3, C and F). The low-intensity areas on both T1WI and T2WI in the medial part of the talus appeared to suggest osteosclerotic change. Tc-99m hydroxymethylene diphosphonate (HMDP) bone scintigraphy showed a focal abnormal accumulation in the right talus (Figure 4, A). Thallium-201 (Tl-201) scintigraphy also showed a substantial accumulation in the right talus in the early scan (Figure 4, B). The accumulation persisted in the delayed scan (Figure 4, C). Therefore a differential diagnosis including a tumor and inflammation was considered.

From the findings described above, we first thought that this patient might have osteomyelitis, and then later suspected avascular necrosis, even though the patient showed atypical symptoms. Though it was very unlikely, the possibility of a tumorous lesion could not be ruled out. Fibroblastic osteosarcoma was diagnosed based on pathological examination of an open biopsy specimen (Figure 5). A histopathological examination of the lesion manifested a dense proliferation of short-spindle tumor cells with a fibroblastic profile, sparse osteoid formation, and destructive infiltration into cancellous bone tissue. We immediately conducted neoadjuvant chemotherapy and concomitant chemotherapy using cisplatin, doxorubicin, VP-16 and methotrexate.

*The patient was treated by Dr. Kenshi Sakayama at Ehime University Hospital, Ehime, Japan.

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Figure 1. Radiographs at the initial consultation. A, Frontal view; B; lateral view. Ill-defined osteolytic changes were observed in the talus.

Lung CT revealed no metastasis, and the patient therefore underwent surgery. We performed reconstruction using a frozen bone method (FBM), an autograft containing tumor treated by liquid nitrogen (13-15), resulting in successful salvage of the limb. Acrylic cement was used for mechanical support. We evaluated the effect of neoadjuvant chemotherapy using pathological specimens obtained at surgery. The tumor was graded 2 based on the FNCLCC (Fédération Nationale des Centres de Lutte Contre le Cancer) grading system (16).

Function five years after the surgery was evaluated as 70% (17), and the patient has been in a continuous disease-free (CDF) state for five years. A plain radiograph shows good alignment, although some slight bone atrophy and secondary osteoarthritic change have been observed (Figure 6). The patient can now walk without a crutch.

Discussion

Osteosarcoma of the talus has rarely been reported (1-10). Histopathologically, patients with this disease have been diagnosed with conventional fibroblastic osteosarcoma, and



Figure 2. Transaxial CT image showing geographic osteolytic change in the lateral part, and osteosclerotic changes in the medial part of the talus.

there has been one report of telangiectatic osteosarcoma (5), two reports of parosteal osteosarcoma (3, 4), two reports of fibroblastic osteosarcoma (1, 10), and other reports of osteoblastic osteosarcoma (1, 2, 5-9). In the previous reports, there are 12 cases of osteosarcoma of the talus reported in 10 papers (Table I) (1-10). They include nine cases in males and three cases in females, with ages ranging from 2 to 81 years (mean age 31.8 years). Except for three cases that were not fully described and thus for which sufficient information was not available (3, 4, 9), there were five who had died of their disease, three with no evidence of disease, and one alive with disease (Table I). However, there were lung metastases in many cases; thus it can be said that the prognosis for patients with osteosarcoma of the talus is poor. Almost all the cases of osteosarcoma in the previous reports were treated by amputation (1-3, 5-8).

Malignant bone tumor of the talus is frequently misdiagnosed as another disease (1-12). Jee *et al.* (4) reported a case where the pathological diagnosis of the biopsy specimen was inaccurate and the patient was misdiagnosed as having a benign tumor, and this hindered their treatment. The incidence of bone tumor including osteosarcoma occurring primarily in the talus is extremely low (1-12, 18-21). In the talus, avascular necrosis is often difficult to accurately distinguish from a tumor (1-4, 7, 10). Weissman *et al.* (12) reported a case of Ewing's sarcoma in the talus that was initially misdiagnosed as avascular necrosis. They advised an unusual location of spontaneous avascular necrosis therefore leading to a suspected unusual

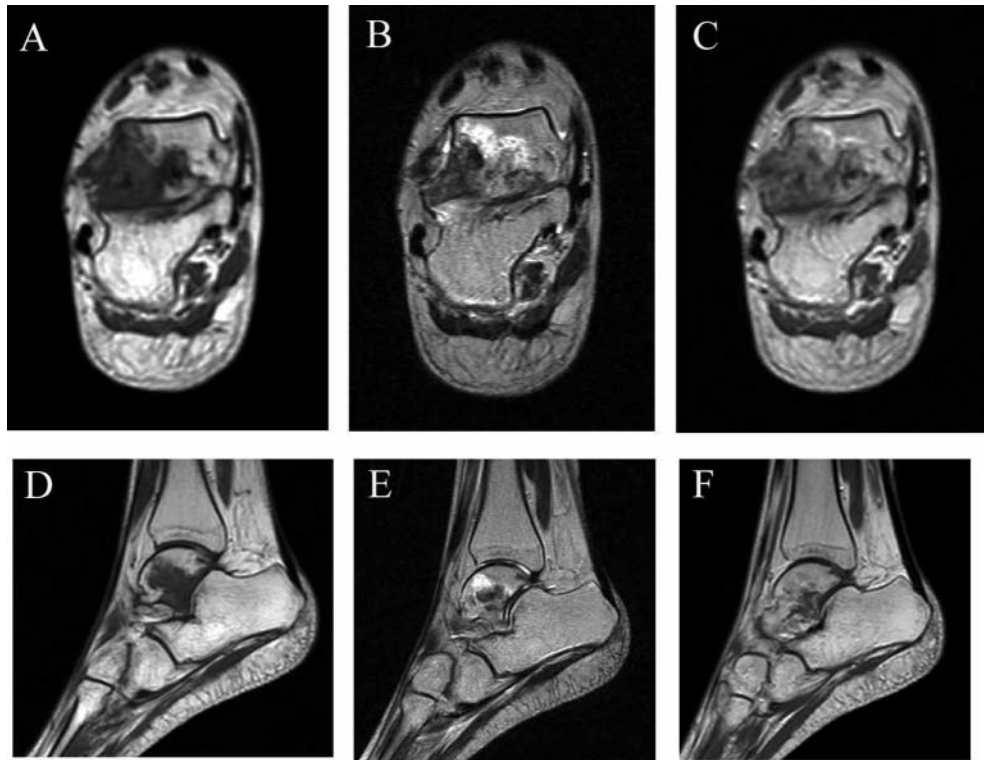


Figure 3. MRI. A-C Coronal images, D-F sagittal images. T1-weighted images (T1WI; TR/TE=300-361/12) show a low intensity (A and D). T2-weighted images (T2WI; TR/TE=3000-3176/100) show heterogeneous intensities (B and E). Gadolinium-enhanced T1-weighted images show inhomogeneous enhancement (C and F). The low-intensity areas on T1WI and T2WI images without Gd enhancement suggest osteosclerotic change.

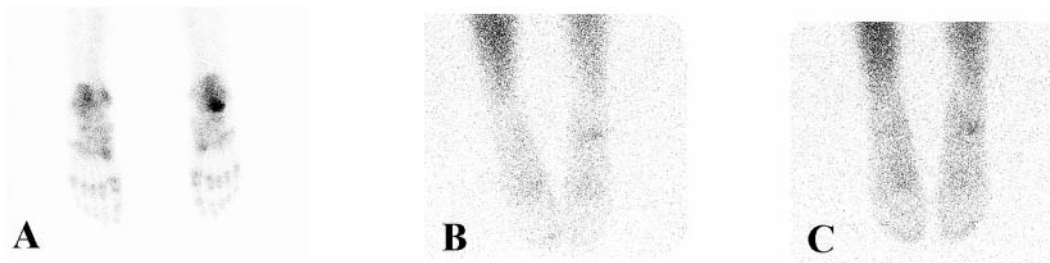


Figure 4. Tc-99m HMDP bone scintigraphy showing a focal intense uptake in the right talus (A). Tl-201 scintigraphy showing abnormal focal uptake in the right talus in the early scan (B), and which persisted in the delayed scan (C).

etiology. In this case, however, we suspected not only avascular necrosis, but also inflammatory lesions such as osteomyelitis because of the osteosclerotic change suggested on CT and MRI (Figures 2 and 3). In Tl-201 scintigraphy, we could not determine whether the findings were because of a tumor or inflammation (Figure 4, B and C). That is to say, we obtained imaging from which we first suspected osteomyelitis and avascular necrosis, although the case showed atypical symptoms. Tl-201 scintigraphy has been reported to be helpful for differentiating malignant from

benign lesions and for evaluating the treatment response in various musculoskeletal tumors (22-24). In this case, although MRI was inconclusive for indicating whether the residual lesion was a viable tumor or an inflammatory reaction and fibroblastic change after chemotherapy, Tl-201 scintigraphy showed a substantial isotope uptake suggesting the presence of residual viable tumor cells.

In this case, we succeeded in limb salvage using FBM (13-15) because the tumor was confined to the bone and it had fortunately been detected at an early stage. To the best of our

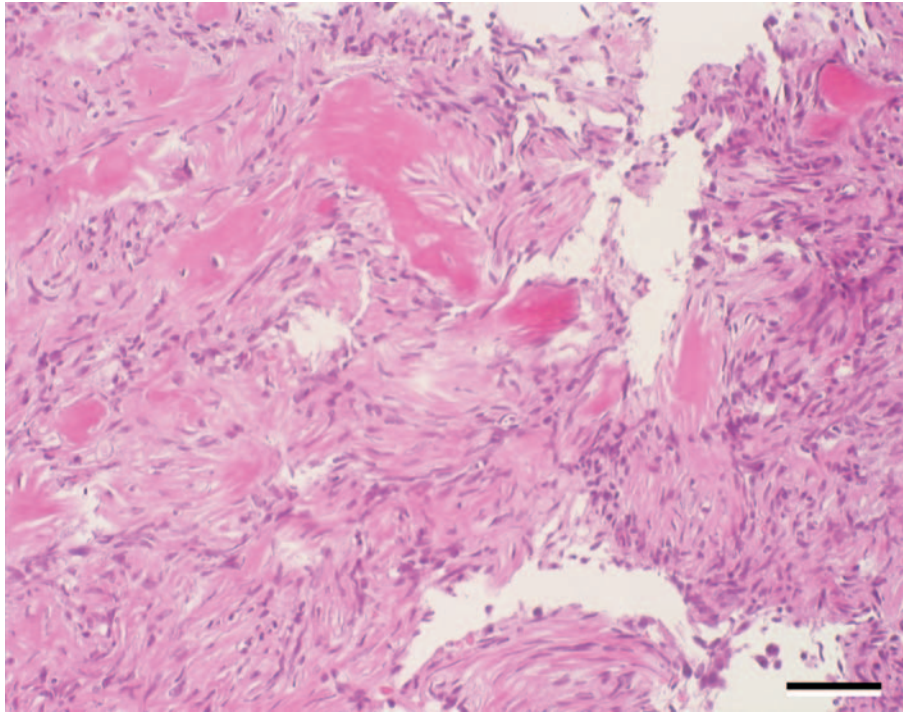


Figure 5. Histopathological examination of an open biopsy specimen demonstrated a dense proliferation of short-spindle tumor cells with a fibroblastic profile, sparse osteoid formation, and destructive infiltration into cancellous bone tissue. Bar=200 μ m.

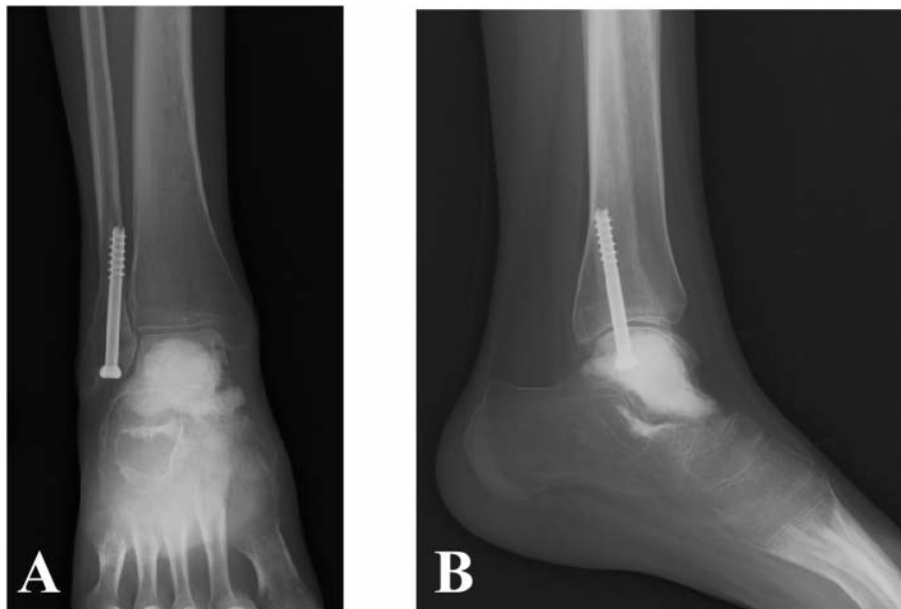


Figure 6. Radiographs four years after the operation show a good alignment. A, Frontal view; B, lateral view.

knowledge, there have been no previous reports of osteosarcoma of the talus treated using FBM reconstruction, nor have there been reports of osteosarcoma of the talus without extrainvasion of the talus (Table I). We believe that

FBM is an excellent therapeutic strategy, even though we realize that many surgeons may nevertheless choose amputation as the treatment method (1-3, 5-8). The advantages of FBM treatment include its high

Table I. *Osteosarcoma of the talus cases in the literature.*

Series	Patient No.	Age/ Gender	Extrainsvasion*	Metastasis	Management	Type	Local recurrence	Outcome	Reference no.
Amini M <i>et al.</i> (1979)	1	21/M	+	-	Amputation	Fibroblastic	-	9 m DOD	1
Matsumoto K <i>et al.</i> (1993)	2	20/M	+	-	Amputation	Osteoblastic	-	20 m DOD	2
Kerjes W <i>et al.</i> (1995)	3	19/M	+	-	Amputation	Parosteal	-	?	3
Jee WH <i>et al.</i> (1998)	4	2/F	+	-	Partial excision	Parosteal	+	?	4
Biscaglia R <i>et al.</i> (1998)	5	64/F	+	-	Amputation	Osteoblastic	-	18 m DOD	5
	6	30/M	+	-	Amputation	Telabiectatic	-	24 m DOD	5
	7	37/M	?	-	Amputation	Osteoblastic	-	120 m NED	5
Choong pfm <i>et al.</i> (1999)	8	8/M	+	-	Amputation	Osteoblastic	-	23 m DOD	6
Seijas R <i>et al.</i> (2006)	9	20/M	+	-	Amputation	Osteoblastic	-	10 m NED	7
Ellison BS <i>et al.</i> (2008)	10	81/F	+	+	Amputation	Osteoblastic	?	4 m AWD	8
Moghazy KM <i>et al.</i> (2008)	11	33/M	+	+	?	Osteoblastic	?	?	9
Katagiri H <i>et al.</i> (2008)	12	47/M	+	-	Wide excision and arthrodesis	Fibroblastic	-	47 m NED	10
Our case (2009)	13	15/F	-	-	Wide excision and FBM	Fibroblastic	-	65 m NED	

*Extrainsvasion multifocal lesion and extraosseous invasion. ?, insufficient information provided; DOD, died of disease; AWD, alive with disease; NED, no evidence of disease; FBM, frozen bone method; m, months.

histocompatibility, and soft tissues, especially many ligaments, can be reused with the patient's autogenous bone. Our patient graduated from high school and is now leading an ordinary life as a college student. Although she needs a brace and has been developing a slight osteoarthritic change in her ankle joint, she has not suffered from pain or swelling.

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