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Case Report

Recurrent radiation-induced osteosarcoma of the sternum in a patient with remote radiation therapy for breast cancer

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ABSTRACT

Background: Recurrent radiation-induced osteosarcoma of the sternum following remote radiation therapy for breast cancer and sternal reconstruction.
Case: A 51-year-old woman presents with recurrent, radiation-induced sternal osteosarcoma 11 years after receiving both radiation therapy for breast cancer and sternal reconstruction. The case details both her work-up and subsequent course, including reconstructive and curative efforts.
Discussion: An uncommon side effect of radiation therapy for breast cancer is development of osteosarcoma of the chest wall. Even rarer is recurrence of the osteosarcoma following sternal reconstruction.

Introduction

Osteosarcoma is the second most common primary malignant bone tumor, typically occurring in the long bones of the pediatric population. Occurrence in the adult population is less common, and occurrence in the geriatric population is uncommon, with tumors of these populations usually related to radiation exposure [1]. Primary osteosarcoma of the sternum is exceedingly rare, with a reported median age of 42 years old, commonly causing symptoms of pain and swelling [2,3]. The best characterization of these lesions is by computed tomography (CT) and magnetic resonance imaging (MRI), with little gained from radiography. MR is the most useful examination for staging, as positron emission tomography imaging may overestimate the actual bony involvement [4,5].

We present the case of a 51-year-old woman with a remote history of invasive ductal carcinoma, treated with unilateral radical mastectomy, chemotherapy, and radiation therapy. While undergoing CT imaging to evaluate for a painful sternum 11 years after the initial treatment, a sternal mass was discovered, which was biopsied and shown to be a sternal osteosarcoma. Subsequent treatment was with chemotherapy, surgical resection, and reconstruction of the sternum. Two years later, the patient had recurrence of the osteosarcoma, which was found to be penetrating through the sternal reconstructive hardware.

Competing Interests: The authors have declared that no competing interests exist.

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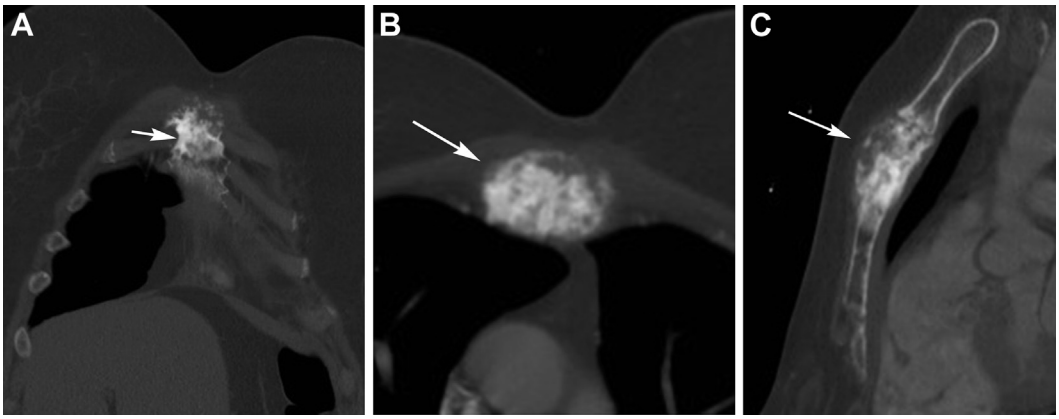

Fig. 1 – (A-C): (A) Coronal, (B) axial, and (C) sagittal CT images show an irregularly shaped, sclerotic (osteoid matrix containing) mass (arrows) arising from the superior sternal body, with extension into both the inferior margin of the manubrium and the bilateral costochondral cartilages of ribs 2-4.

Case history

A 51-year-old woman with a history of treated, left-sided, invasive ductal carcinoma of the breast presented with an enlarging, painful sternal mass. She previously underwent unilateral radical mastectomy, chemotherapy, and radiation therapy for her breast cancer 11 years before her presentation. At the time of presentation, it was felt that this mass was suspicious for recurrent breast carcinoma and CT of the neck, chest, abdomen, and pelvis was ordered to restage the patient. Initial laboratory work included only a complete blood count, which was within normal limits. The follow-up CT of the chest showed an expansile, sclerotic, osteoid matrix containing lesion (Fig. 1) of the manubrium and superior body of the sternum, measuring 4.5 × 3.8 × 2.5 cm, extending from the superior sternal body into the inferior margin of the manubrium. The lateral extent of the lesion extended to the bilateral 2-4 ribs costocartilaginous junctions. For further characterization of the mass, an MRI examination was performed (Fig. 2) showing a hypointense mass arising from the superior sternal body, with extension into the inferior margin of the manubrium, with perilesional edema.

The initial differential considerations were broad and included metastatic disease and radiation-induced osteosarcoma. After initial imaging evaluation, the patient underwent open biopsy of the lesion, performed by the general surgery department. Pathologic review of the biopsy specimen showed highly atypical cells and was initially felt to show

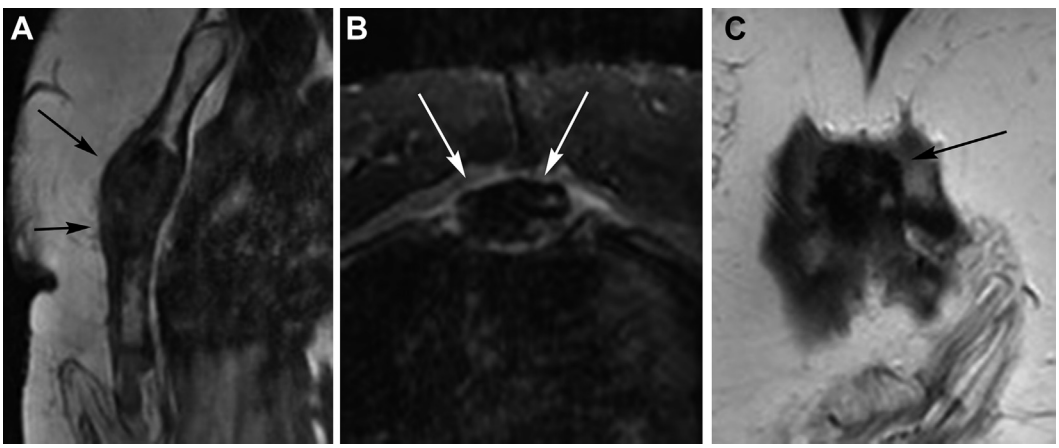

Fig. 2 – (A-C): (A) Sagittal T2 weighted, (B) axial T2FS, and (C) coronal T2-weighted MR images show a hypointense mass (arrows) arising from the superior sternal body, with extension into the inferior margin of the manubrium, with perilesional edema.

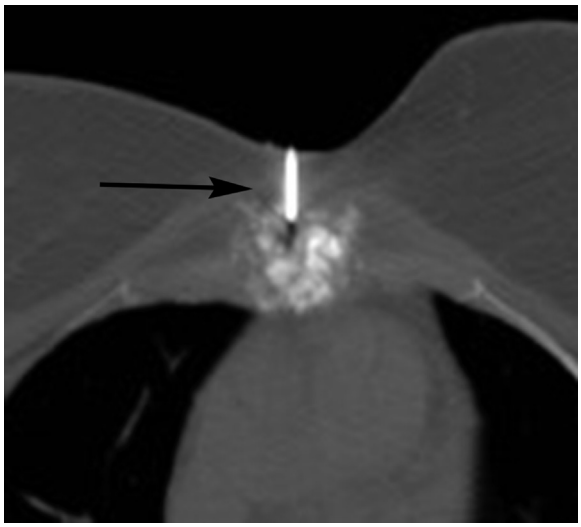

Fig. 3 – Axial CT image used for needle localization during a CT-guided core biopsy, using a 13-ga biopsy needle (arrow).

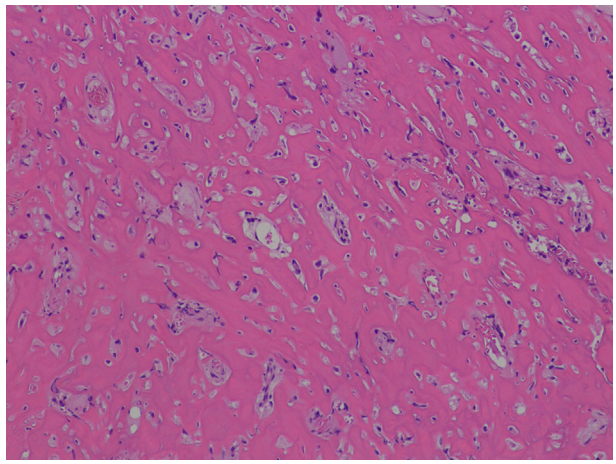

Fig. 4 – A 100× magnified pathology slide showing clusters of malignant epithelioid and spindle cells in a background of dense, pink, amorphous material consistent with osteoid.

recurrent breast disease. However, on immunohistochemical work-up, it was revealed that this was likely osteoblastic in origin. Unfortunately, a definitive diagnosis was not reached with the initial surgical specimen. Therefore, a CT-guided sternal biopsy was performed (Fig. 3) and subsequent review yielded a diagnosis of radiation-induced osteosarcoma (Fig. 4).

The initial treatment included high-dose methotrexate therapy, hemisternectomy, and bilateral resection of costal cartilages of ribs 2-4, with reconstruction, performed by the plastic surgery department. The reconstruction was performed using bilateral pectoral flaps over 3 contoured Stryker reconstruction plates (Stryker Corporation, Kalamazoo, MI) fixated to the second, third, and fourth ribs bilaterally (Fig. 5).

Follow-up MRI performed 2 months after sternal reconstruction showed a new metastatic disease to the patient's cerebellar vermis (Fig. 6), for which the patient elected to undergo surgical resection. Pathologic review of the resection showed recurrent breast carcinoma. Subsequently, the patient had CSF dissemination of tumor requiring a burr hole and ventricular catheter placement.

These palliative efforts proved successful in relieving the patient's symptoms; however, the most recent follow-up CT imaging of the chest at 2 years poststernal resection and/or reconstruction showed a lower sternal, osteoid matrix containing mass measuring 4.1 × 3 × 2.1 cm, extending from the level of the fourth-sixth ribs costochondral junctions laterally, superiorly, and inferiorly. The mass anteriorly

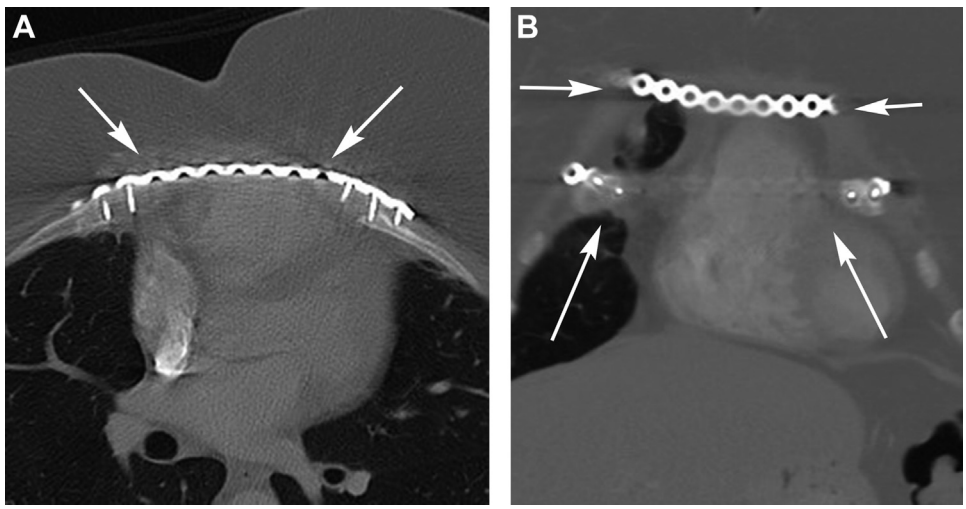

Fig. 5 – (A, B): (A) Axial and (B) coronal CT images show reconstruction of the chest wall with 3 Stryker plates and screw fixation device (arrows), with an overlying pectoral flap.

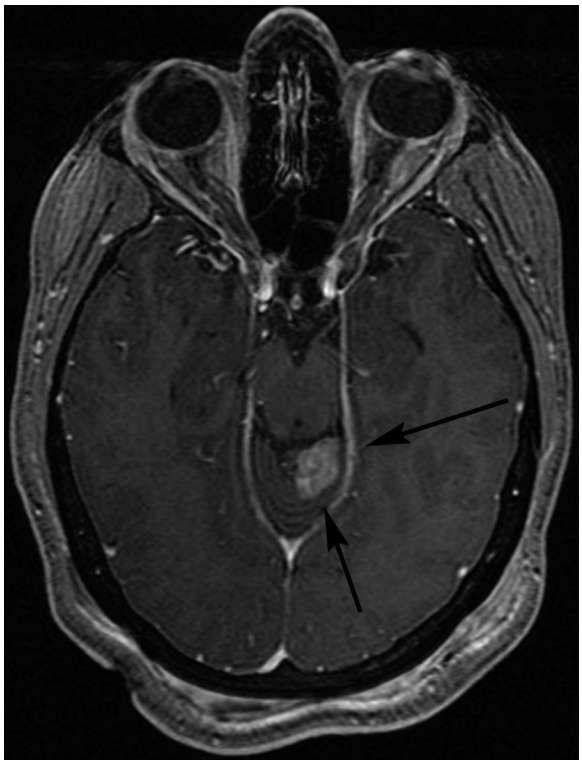

Fig. 6 – Axial T1-weighted post–gadolinium contrast MR image shows a hyperintense mass (arrows) along the left lateral border of the cerebellar vermis.

extended through the inferior most reconstructive hardware plate (Fig. 7). The lesion fully encompassed the hardware. The mass was biopsied by the thoracic surgery department and found to be recurrent radiation-induced osteosarcoma.

Discussion

Primary osteosarcoma has a relatively low incidence, with a bimodal age distribution. Common presentation is a patient with a painful bone mass, which leads imaging work-up. CT has been identified as useful in characterization of the mass, while MRI has benefit in evaluating the extent of the mass [4]. Although our case did not yield a definitive diagnosis with the initial surgical biopsy, open biopsy is the recommended method of obtaining a tissue diagnosis [1].

Primary malignancies of the sternum make up only 1% of sternal masses. Of the primary malignancies of the sternum, chondroblastoma is the most common, making primary osteosarcoma of the sternum a very rare entity [6]. Often, patients will have a history of prior radiation treatment of the chest wall [7]. Therapy often involves preoperative chemotherapy with high-dose methotrexate and resection with wide margins, and additional postoperative chemotherapy if the lesion demonstrated response to treatment [8]. Overall, prognosis is poor at approximately 15% 5-year survival, which decreases to 0% 5-year survival if there is metastatic disease [3].

Reconstructive surgery of the sternum in our case was accomplished with a muscle flap over an implantable hardware plate, while other reconstructive techniques include muscle flap alone, muscle flap and mesh, muscle flap and rigid prosthesis, or use of homologous iliac bone graft. Muscle flap-only reconstruction is generally only used if only the upper sternum and/or ribs 1-3 are resected. Resection of the lower sternum requires hardware or more rigid reconstruction to protect the thoracic contents and stabilize the chest wall [9,10]. There are no devices specifically made for reconstruction of the sternum, although some devices can be tailored to fit the need. With the advent of 3-dimensional printed implantable hardware, and the ubiquity of volumetric scans, there have also been case reports of reconstructing a sternum with the help of 3-dimensional printing technology [11].

Fig. 7 – (A-C): (A) Axial, (B) coronal, and (C) sagittal CT images showing a recurrent sclerotic, osteoid matrix containing mass (arrows), which extends anterior through the Stryker plate hardware.

Conclusions

Ultimately, our case was a demonstration of a rare complication of radiation therapy to the chest wall during treatment of a breast cancer, with detail on our approach to treatment, based on the current literature.

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