Abstract. Giant cell tumor (GCT) is a relatively common and locally aggressive benign bone tumor. The sternum is a rare location. To date, only four cases have been reported with detailed information. Here, we present a case with sternal GCT and discuss the clinical manifestation and treatment, in addition to reviewing the literature. A 53-year-old female noted pain in the sternum after a trauma. Radiographic findings revealed an expanding osteolytic lesion in the body of the sternum. Extended curettage of the tumor was performed followed by polymethylmethacrylate (PMMA) filling. No recurrence was found over a 7-year follow-up period. In conclusion, even though sternal benign tumors are rare, GCT should be considered as one of the differential diagnoses in a patient presenting with a sternal lesion in the 5th or 6th decade of life. The initial treatment should be extended curettage followed by filling with PMMA.

Giant cell tumor (GCT) is a relatively common benign bone tumor which represents approximately 25% of benign bone tumors (1). GCTs have strong predilection at the ends (epiphyses) of long bones in young adults (1, 2). In particular, 46-63% of the lesions occur around the knee (1, 3). The sternum is the least common site of this condition (1, 2). According to the series of the Mayo Clinic, benign sternal tumors were found in only 3 out of 2,334 cases of all benign bone tumors (0.1%). Two of these three benign cases were sternal GCTs (1). However, detailed information of these two cases is not mentioned in the literature. To date, only four cases have been reported with detailed information (4-7).

Here, we present long-term follow-up of a rare case with GCT developing from the sternum. We discuss the clinical manifestation and treatment, in addition to reviewing the literature for comparison.

Case Report

A 53-year-old female noted pain after a ball hit her in the anterior chest region while she was playing handball. The pain developed gradually over one month. The patient was diagnosed with a sternal tumor by a lateral X-ray at the regional hospital before being referred to the bone and soft tumor section of our department. On physical examination, a tender mass was found with slight local heat.

A lateral X-ray revealed an expanding osteolytic lesion in the body of the sternum (Figure 1). Computerized tomography (CT) clearly demonstrated the lesion in the body of the sternum with an expanded and thin cortical wall, in addition to pathological fractures due to tumor expansion (Figures 2 and 3). Magnetic resonance imaging (MRI) revealed an expansive mass with heterogeneous low to intermediate signal intensity area on the T2-weighted images.

An incisional biopsy of the sternal lesion was performed. Histological examination revealed that the lesion was composed of osteoclast-like giant cells interspersed between a vascular, spindle cell stroma, which indicated a GCT or a blown tumor due to hyperparathyroidism (Figure 4). A blood test was performed to differentiate a blown tumor from a GCT. Calcium, phosphate and parathyroid hormone were normal. Thus, a GCT of the sternum was diagnosed.

The tumor was resected in a piece-by-piece manner by the use of graspers and curettes. A Black Max cutting burr (Anschach, FL, USA) was applied to remove the remnants of the tumor attached to the cortical wall. After the tumor resection, the bone void was filled with polymethylmethacrylate (PMMA; Surgical Simplex P; Stryker Howmedica Osteonics, Allendale, NJ, USA).

The gross appearance of the resected specimen consisted of fibro-osseous fragments with small pieces of soft yellow tissue. Histology showed the same as the results of the incisional biopsy. Immediately after surgery pain relief was
obtained. No recurrence or metastasis has been found 7 years after surgery (Figure 5).

Discussion

Primary tumors located in the sternum account for approximately 0.9% of all primary bone tumors (1), and almost all are malignant (8). Chondrosarcoma is the most common primary malignant tumor of the sternum, followed by osteosarcoma, myeloma and malignant lymphoma (1, 8). However, benign primary sternal tumors are extremely rare. GCT (1, 4-7), chondroma (1), osteoblastoma (9), and hemangioma (10) have been described.

GCTs typically affect the ends of long bones. The distal femur is the most common site followed by the proximal tibia, distal radius and proximal tibia. According to the series of the Mayo Clinic, only 2 cases of sternal GCT were found out of 568 GCT cases accounting for 0.3% (1). Anatomically the sternum is divided into 3 regions: the manubrium, body and xiphoid process. Of GCTs in the sternum, 3 cases including the present case were found in the body (4, 5) and 2 in the manubrium (6, 7). There has been no reported case of GCT developing from the xiphoid process.

With regards to age, the large majority of the patients with GCT in bones other than the sternum are between 20 and 45 years old, with peak incidence in the third decade of life (3, 9). It is rare after the age of 50 years (2). In contrast, the

Figure 1. A lateral X-ray of the patient showing an expanding lesion in the body of the sternum (arrow).

Figure 2. A sagittal multiplanar reconstruction image shows the lesion in the anterior and upper region of the sternum. The cortical wall is expanded, thin and partly destroyed due to tumor expansion.
average age of the 5 patients with sternal GCTs was 51 years old, ranging from 45 to 55 years old (Table I). Thus, the mean age of patients with sternal GCTs tends to be higher than that of patients with other GCTs (7).

The primary symptom of all sternal GCTs except one was moderate pain in relation to the affected region (Table I). Pain might be caused by pathological fracture of the thin cortical bone due to the locally aggressive behavior of this condition. This aggressive behavior can be seen in relation to local recurrence and when the affected location is the radius (11). In some cases, GCT can metastasize, even though the tumor is cytologically benign (1). No metastasis of sternal GCTs has been reported so far.

Treatment of GCT is the most challenging of all benign tumors of bone due to a high local recurrent rate (1, 12). Even though bisphosphonate administration (13), and radiotherapy (14) were found as effective treatment in some cases, removal of the GCT is the main treatment choice (1). Surgery has ranged from simple curettage to wide resection. The results of simple curettage were poor since the local recurrence rate reached approximately 50% due to the locally aggressive behavior of GCTs. This led to a more aggressive form of surgery such as marginal or wide resection. However, such resection requires reconstruction, which may be associated with significantly higher infection rate. Recently, extended curettage and PMMA have been used as an alternative to resection since this technique is less invasive, with a lower infection rate, and the local recurrence rate is approximately 25%, which is low compared to simple curettage (1, 12).

In cases of sternal GCT, the surgical options are either extended curettage followed by filling with PMMA or subtotal sternectomy. After subtotal sternectomy, a prosthetic replacement is needed to protect the lungs, heart and main vessels, and to restore functional thoracic movement to prevent paradoxical respiration (4, 15). However, the potential risk of infection has to be taken into account. Among the sternal GCTs in the literatures, subtotal sternectomy was performed in 3 cases, and curettage and cementing in 2 cases, including the present case (Table I).

Table 1. Patient demographics.

<table>
<thead>
<tr>
<th>Authors</th>
<th>Number of cases</th>
<th>Gender</th>
<th>Age (years)</th>
<th>Symptom</th>
<th>Location</th>
<th>Size (cm)</th>
<th>Surgery</th>
<th>Reconstruction</th>
<th>Follow-up (months)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Imai et al. (4)</td>
<td>1</td>
<td>M</td>
<td>45</td>
<td>Pain</td>
<td>Body</td>
<td>8.5x4.5x2.5</td>
<td>Subtotal sternectomy</td>
<td>Prosthesis</td>
<td>12</td>
</tr>
<tr>
<td>Segawa et al. (5)</td>
<td>1</td>
<td>M</td>
<td>55</td>
<td>Pain</td>
<td>Body</td>
<td>3.5x3.0</td>
<td>Curettage</td>
<td>Polymethylmethacrylate</td>
<td>12</td>
</tr>
<tr>
<td>Bay et al. (6)</td>
<td>1</td>
<td>F</td>
<td>49</td>
<td>Pain</td>
<td>Manubrium</td>
<td>3.9x3.2</td>
<td>Subtotal sternectomy</td>
<td>Prosthesis</td>
<td>60</td>
</tr>
<tr>
<td>Sundaram et al. (7)</td>
<td>1</td>
<td>M</td>
<td>55</td>
<td>Painless swelling</td>
<td>Manubrium</td>
<td>NA</td>
<td>Subtotal sternectomy</td>
<td>None</td>
<td>NA</td>
</tr>
<tr>
<td>Present case</td>
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<td>F</td>
<td>53</td>
<td>Pain</td>
<td>Body</td>
<td>8x4x2.5</td>
<td>Curettage</td>
<td>Polymethylmethacrylate</td>
<td>84</td>
</tr>
</tbody>
</table>

F, female; M, male; NA, not available.

Figure 3. An axial CT image demonstrates pathological fractures through the cortical wall.

Figure 4. Histology of the tumor consists of osteoclast-like giant cells interspersed between a vascular, spindle cell stroma. (Bar equals 100 μm).
With either surgical procedure, no recurrence has been reported in sternal GCTs during the 12 to 84 months’ follow-up. In the present case, no recurrence was found 7 years after surgery, which is the longest follow-up among the sternal GCTs.

In conclusion, even though sternal benign tumors are rare, GCT should be considered as one of the differential diagnoses in a patient presenting with a sternal lesion in the 5th or 6th decade of life. The initial treatment should be extended curettage followed by filling with PMMA, unless reconstruction with PMMA is impossible.

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References


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