Aggressive intraosseous lipoma of the scapula: A case report

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Abstract. The details of the pathogenesis of intraosseous lipomas are not fully elucidated, although most cases do not require surgical treatment. The present report describes the case of a 79-year-old female patient diagnosed with intracapsular lipoma who also exhibited an extraosseous extension. Chest computed tomography revealed an abnormal shadow or a mass in the right scapula and destruction of the glenoid bone. Magnetic resonance imaging revealed a high-intensity mass on T1-weighted and T2-weighted images in the same area. Marginal resection of the mass was performed. The histopathology confirmed that the mass was a lipoma. No postoperative recurrence was observed. Oncologists must be aware that lipoma arising within the scapula may extend outside the bone.

Introduction

Intraosseous lipoma is an extremely rare neoplasm that constitutes no more than 0.1% of all the bone tumors (1). The first case of intraosseous lipoma was described in 1955, with Milgram having published the largest series of these lesions (2,3). Lipoma is common in adults, being slightly more common in males (4). The recurrence rate of lipoma is <5%. Because it is benign, the mortality rate is 0%. The treatment strategy is based on complete resection. Although pain is the major symptom reported, more than 30% of the cases have been found incidentally through imaging studies performed for other reasons (3,5). Intraosseous lipomas occur mainly in the lower limbs with calcaneus and long tubular bones as common lesion sites. Nevertheless, these lesions may occur anywhere in the skeleton (3,5,6) making them difficult to diagnose. Intraosseous lipoma of the scapula is extremely rare and, so far, only case has been reported (7). Here, we report the case of an aggressive intraosseous lipoma that extended outside of the scapula.

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Case report

The patient was a 79-year-old woman with no previous medical history. The patient had no complaints of localized pain in the scapula prior to the visit. When the patient underwent chest radiography and computed tomography (CT) scan at the Department of General Medicine in our hospital (Kushimoto Hospital) in January 2019, a lytic lesion was noticed in the right scapula (Fig. 1). The pulmonologist advised the patient to undergo a detailed examination and further tests. However, owing to personal reasons, the patient did not visit the hospital for 2 years. Additionally, no significant changes were observed in blood test parameters (Table I). The inflammatory response was also negative (CRP=0.06 mg/dl, WBC=5300/μl).

The patient visited the hospital again and was referred to our clinic for further examination in January 2021. Radiographs of the right scapula showed a lytic lesion in the glenoid fossa (Fig. 2). Later, in February 2021, a CT scan of the right scapula also revealed a low-density, homogeneous lytic lesion in the same region (Fig. 3). Specifically, the destruction of dorsal cortical bone of the glenoid fossa was observed. Magnetic resonance imaging (MRI) also indicated a high-intensity mass on both T1 and T2-weighted images in February 2021 (Fig. 4). No septa were detected inside the tumor mass. Since the lesion was deep and had spread outside the skeleton, the possibility of malignancy was considered. Therefore, in March 2021, we performed a tumor resection. The final observational visit took place in June 2023 and no recurrence was observed.

The tumor appeared yellow and shiny. Hematoxylin and eosin staining was performed by our laboratory technician (M.T.) per standard protocol. Tumor tissue samples were fixed in 4% paraformaldehyde at 4˚C for 12 h. After washing with phosphate-buffered saline (PBS), the samples were decalcified in 10% ethylenediaminetetraacetic acid solution at 4˚C for 2 weeks and then embedded in paraffin. Coronary sections (5-μm) were cut and mounted onto slides. The sections were deparaffinized in xylene and dehydrated using an ethanol gradient, and then immersed in hematoxylin solution (Agilent Technologies) for 10 min. The sections were washed in PBS for 5 min, and then immersed in eosin solution (Agilent Technologies) for 5 min. The histopathological examination by our pathologist (T.I.) revealed that the tumor was a lipoma (Fig. 5). No lipoblasts were observed. The patient was

followed up for 2 years. Patients with lipoma are followed up postoperatively every 6 months with CT examination. Neither recurrence nor shoulder joint range of motion restriction were seen thereafter.

The patient provided written informed consent for the publication of this information. Ethical approval was not necessary because the patient was conventionally treated.

Discussion

Intraosseous lipoma is one of the rarest tumors with an incidence of 0.1-2.5% (1,8,9). Most intraosseous lipomas occur in the lower extremities (71%). The most common site is the calcaneus (32%), followed by the subtrochanteric femur, proximal tibia, distal femur, and proximal and distal fibula (10). Lesions of the upper extremities usually involve the proximal and distal humerus and the radial shaft, although they have also been observed in the mandible, pelvis, and ribs (11). To date, only one case of intraosseous lipoma (early phase) has been reported in a 20-year-old man, but without any sign of destruction in the body of the scapula (7). Compared to this previously reported case, the present case is characterized by the absence of pain. Moreover, for the present case, a relatively detailed treatment course has also been provided.

Intraosseous lipomas are highly prevalent in the calcaneus and femur intertrochanteric regions, where trabecular bone is scarce. This has led to the hypothesis that these lipomas are an 'overshoot' phenomenon that occurs during the transition from hematopoietic to fatty bone marrow (12). Further, it was suggested that the intraosseous lipomas developing in these regions can likely be considered hematomas rather than neoplasms (1,8). In the current case, the tumor was present in the scapula, which is a flat bone. In addition, the patient did not face any trauma in the past according to her medical record. Therefore, the tumor cannot be considered a hematoma.

In general, intraosseous lipomas are painless (3,5,11). If painful, they are thought to be due to coexisting bone expansion, remodeling, and ischemic changes in the bone. Also, pathological fractures are uncommon (1,3,5). The current case is an extremely rare one in which the patient, inexplicably, did not complain of any pain, despite pathological fracture. However, it is important to keep in mind that lipomas in the scapula can cause pathological fractures.

Milgram classified intraosseous lipomas into three types. In the first type, the tumor contains viable fat cells; in the second type, the viable fat cells are partially replaced by necrosis and calcification; and in the third type, necrosis, calcification, and lipid cyst formation are seen (3). The current case is a rare and aggressive case of extraosseous extension in spite of classification I. Furthermore, the differential diagnosis, in this case, included fibrous dysplasia, inflammation, hematoma, malignant soft tissue tumor, and bone tumor. MRI findings of a typical acute hematoma include a hypointense mass on T1-weighted images and a hyperintense mass on T2-weighted images (13). The presence of fresh and preexisting hematomas may present as a mosaic pattern (13). In this case, there were no MRI findings suggestive of hematoma. Histopathology was performed for the excised specimen to confirm the diagnosis. Notably, the intraosseous lipoma lesions may be indistinguishable from normal adipose tissue in the xanthoma, making the pathological interpretation difficult (1,8,9).

Table I. Blood test results.

Item	Value (normal value)
Total protein, g/dl	6.5 (6.5-7.9)
Albumin, g/dl	3.8 (3.8-4.5)
BUN, mg/dl	20.1 (8.0-20.0)
Creatinine, mg/dl	0.59 (0.5-0.9)
eGFR, ml/min/1.73 m^2	73 (>60)
Urea acid, mg/dl	4.5 (2.5-5.8)
Total cholesterol, mg/dl	208 (140-199)
Total bilirubin, mg/dl	0.7 (0.2-1.2)
AST, U/l	22 (7-23)
ALT, U/l	25 (7-23)
ALP, U/l	209 (38-113)
CK, U/l	90 (20-150)
Na, mEq/l	143 (135-150)
Cl, mEq/l	107 (101-108)
K, mEq/l	4.4 (3.5-5.0)
Ca, mg/dl	8.9 (8.8-10.4)
CRP, mg/dl	0.06 (<0.03)
WBC, /μl	5,300 (4,000-8,000)
Hb, g/dl	12 (11.4-14.6)
PLT, /μl	$21.4x10^4$ ($25x10^4$-$40x10^4$)

BUN, blood urea nitrogen; eGFR, estimated glomerular filtration rate; AST, aspartate aminotransferase; ALT, alanine aminotransferase; ALP, alkali-phosphatase; CK, creatine kinase; CRP, C-reactive protein; WBC, white blood cell; Hb, hemoglobin; PLT, platelet.

It is seldom possible to remove an intraosseous lipoma completely. Usually, these lipomas do not show any symptoms, have a slow course, and can be managed with a 'wait and see' approach (1,5). The exception to this is when the lipoma results in a pathological fracture and extraskeletal extension (11). In the present case, surgical resection was performed because of extraskeletal extension and the possibility of malignancy. Intraosseous lipomas have a favorable prognosis with no recurrences following lesion curettage and grafting (6,7) and complete involution (14,15). These tumors are considered benign and patients are usually asymptomatic. These facts have prompted other authors to propose clinical and radiological observations only, rather than active treatment (15,16). Thus, the prognosis of intraosseous lipoma has been reported to be excellent; however, a careful follow-up is necessary for intraosseous lipoma particularly in cases with extraosseous extension.

The current case presentation has a limitation. We failed to demonstrate negative immunohistochemical findings for MDM2 or CDK4, which are specific for liposarcoma (17). However, we were able to confirm the benign nature of the tumor by demonstrating the absence of nuclear atypia on hematoxylin and eosin imaging. Additional immunostaining for MDM2 and CDK4 should be performed in future similar cases.

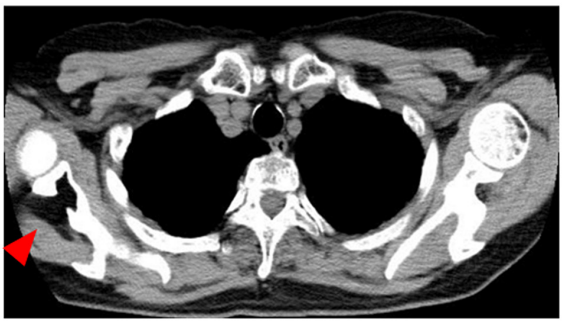

Figure 1. Computed tomography image of the right shoulder at the first visit in January 2019. The red arrowhead shows the tumor site.

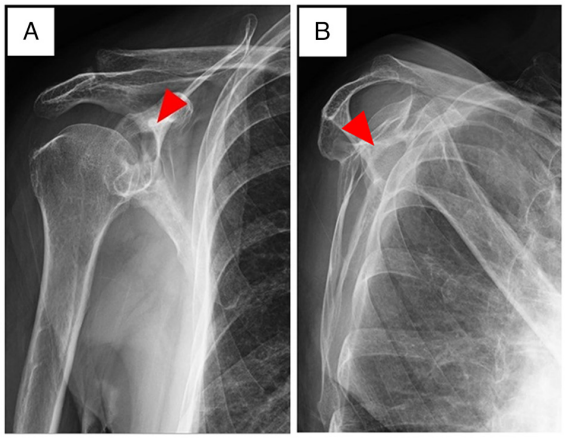

Figure 2. (A) Radiograph of the right shoulder with coronal view at the second visit (2 years after the first visit) in February 2021 (red arrowhead, tumor and lytic lesion in the glenoid of the scapula). (B) Radiograph of the scapula-Y view of the right shoulder. The lytic lesion was observed near the glenoid of the scapula (red arrowhead, tumor and lytic lesion in the glenoid of the scapula). (A and B) The red arrows all point to the same lesion.

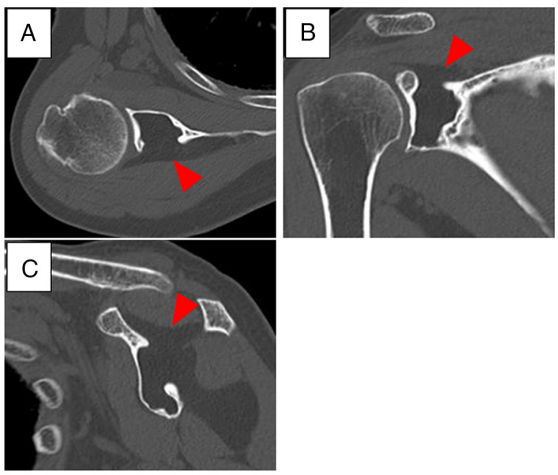

Figure 3. (A) Axial, (B) coronal and (C) sagittal computed tomography image of the right shoulder at the second visit in February 2021 (2 years after the first visit). The tumor mass existed adjacent to the glenoid, and some tumor mass extended to the upper side of the glenoid. The glenoid wall was thin. The red arrowhead shows the tumor site.

Oncologists must consider that intraosseous lipoma might occur in the scapula with cortical bone destruction.

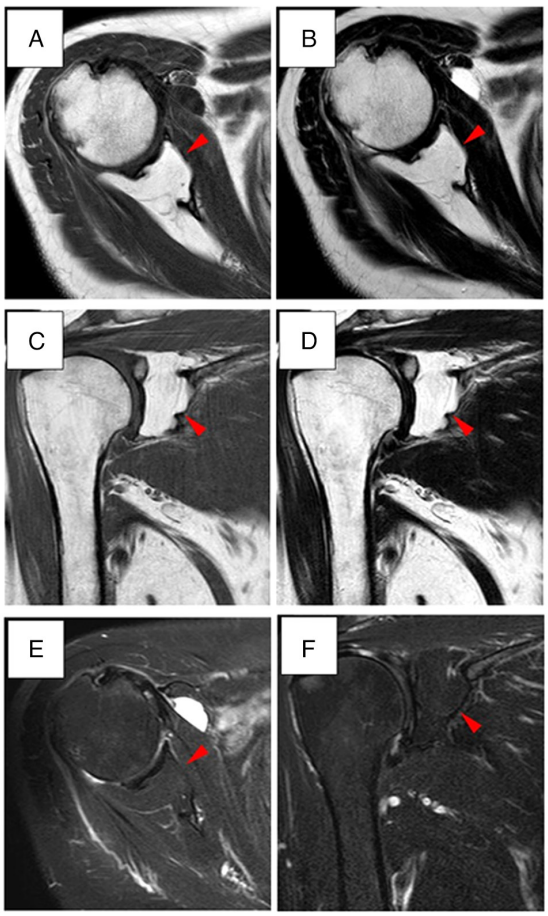

Figure 4. MRI of the right shoulder at the second visit in February 2021 (2 years after the first visit). (A) Axial view of the shoulder on T1-weighted MRI (red arrow, tumor and lytic lesion in the glenoid of the scapula). (B) Another axial slice view of the shoulder on T2-weighted MRI (red arrow, tumor and lytic lesion in the glenoid of the scapula). (C) Coronal view of the shoulder in which the tumor appears to be at its maximum on T1-weighted MRI (red arrow, tumor and lytic lesion in the glenoid of the scapula). (D) Coronal slice view of the shoulder on T2-weighted MRI (red arrow, tumor and lytic lesion in the glenoid of the scapula). (E) Axial view of the shoulder on T2-weighted fat suppression imaging (red arrow, tumor and lytic lesion in the glenoid of the scapula). (F) Another coronal view of the shoulder on T2-weighted fat suppression imaging (red arrow, tumor and lytic lesion in the glenoid of the scapula). The red arrowhead shows the tumor site. MRI, magnetic resonance imaging.

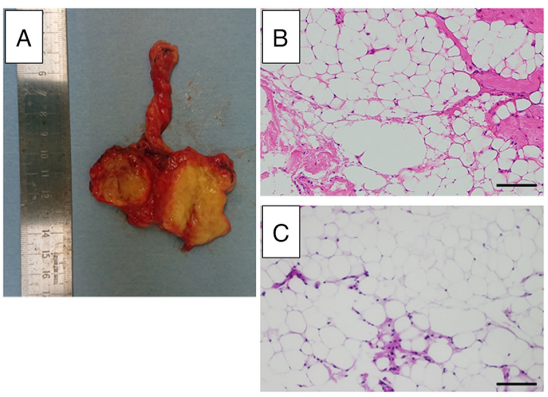

Figure 5. (A) Resected specimen showing that adipose tissue was yellow and shiny. (B) Hematoxylin and eosin-stained image of the excised specimen. (C) Another image of the hematoxylin and eosin-stained excised specimen (magnification, x40; scale bar, 100 μm). Adipose cells with no malignancy were observed.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Authors' contributions

NO, YS, SN, MA and KH participated in data collection, analysis and manuscript writing. YS, SN and MA participated in the study design. NO, KH, YS, SN and MA confirm the authenticity of all the raw data. All authors have read and approved the final manuscript.

Ethics approval and consent to participate

The case studies were conducted in accordance with the Declaration of Helsinki. Ethical approval was not necessary because the patient was conventionally treated.

Patient consent for publication

Written informed consent for publication was obtained from the patient.

Competing interests

The authors declare that they have no competing interests.

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