

Case Report

Intermuscular cavernous lymphangioma presenting with overgrowth of fat: MRI findings

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Abstract

We present a case of an intermuscular cavernous lymphangioma with predominantly composed of fatty tissue. A 58-year-old man complained a palpable painless mass over his right arm for 1 month. Magnetic resonance imaging (MRI) showed a 14-cm mass with predominantly composed of fatty tissue between long and short heads of triceps muscle. There were some serpiginous structures with hyperintensity within the tumors on gradient-echo and gadolinium enhanced T1-weighted images. Histologic examination revealed a picture of intermuscular angioma predominantly composed of cavernous lymphangioma.

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1. Introduction

Soft tissue tumors of lymphatic origin are rare [1]. Lymphangiomas in the upper extremity and in adults are extremely rare. Magnetic resonance imaging (MRI) findings of soft tissue lymphangiomatous tumors have rarely been reported [2–5]. Most of these tumors appear as well-defined cystic masses with high signal intensity on T2-weighted images. We herein present what appears to be a rare feature of an intermuscular cavernous lymphangioma predominantly composed of fatty tissue in the arm of a 58-year-old man.

2. Case report

A 58-year-old man presented with a painless growing mass over his right upper arm for 1 month. No preceding

injury or trauma was noted. Physical examination revealed a non-movable, solid mass in the posterolateral aspect of the right upper arm. No tenderness was noted. There was no focal discoloration of the skin or atrophic change of muscles. Laboratory data were normal.

Conventional radiography showed soft-tissue swelling in the right upper arm. No bony changes of the humerus were noted. MRI (1.5-T, Horizon LX, General Electric, Wis.) showed a lobulated, well-defined soft-tissue mass, measuring 14×7×6 cm, between long and short heads of triceps brachii muscle. The mass was composed of fatty tissues interspersed serpiginous structures on T1-weighted images (Fig. 1a), some area of the mass turned to hyperintense signal and the serpiginous structures showed hyperintensity on proton-density weighted fat saturation images and gradient-echo T2* weighted images (Fig. 1b and c), and enhancement on gadolinium-enhanced T1-weighted images (Fig. 1d). The adjacent humerus was not involved. A lipoma variant with vascular channels was proposed before surgery.

The patient received wide excision of the tumor eight days later. At surgery, the tumor mass was identified deep into the space of the long and short heads of the triceps muscle. The tumor adhered to and compressed the radial nerve. Grossly, a partially encapsulated yellow mass was seen. Histologic examination revealed a picture of

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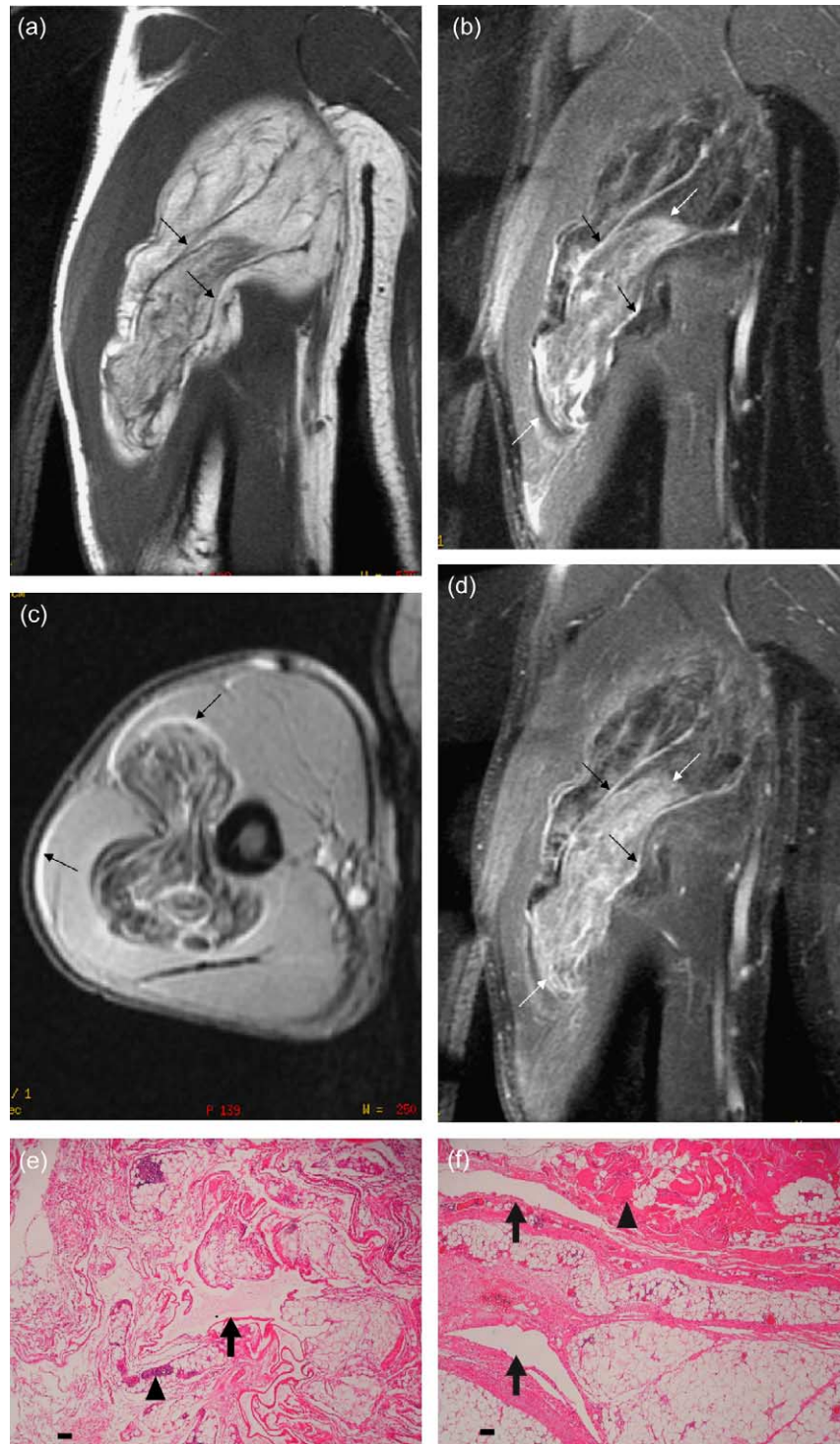


Fig. 1. Fifty-eight-year-old man with intermuscular cavernous lymphangioma over his right arm. (a) Coronal T1-weighted MR image (TR/TE, 450/11.9) shows a well-defined, lobulated high-signal-intensity mass in the right upper arm, suggesting predominantly fatty components. Some serpiginous structures (arrows) within the tumor are seen. (b) Coronal proton-density weighted MR image with fat saturation (TR/TE, 2000/31) shows the fatty tissue is of low signal intensity, and an area of high signal intensity (white arrows) is seen. The serpiginous structures (black arrows) turn to high signal intensity as compared to low signal intensity on T1-weighted images, suggestive of slow-flow vessels. (c) Axial gradient-echo T2* weighted MR image (TR/TE, 417/15; flip angle, 20°) shows hyperintense serpiginous structures within the lobulated soft-tissue mass. Note presence of enhanced chemical-shift artifact (arrows). (d) Coronal gadolinium-enhanced T1-weighted MR image with fat saturation (TR/TE, 566.7/10.2) shows enhancement of the serpiginous structures (black arrows). The area with heterogeneous enhancement (white arrows) may be attributed to the tumor with numerous lymphocytes, as documented in our histologic findings. (e) Histologic specimen reveals mature adipocytes mixed with dilated, thin-walled lymphatic channels containing eosinophilic lymph (arrow). Note that many lymphocytes (arrowhead) are seen. (H and E, $\times 40$) (Scalebar=50 μm). (f) The tumor composed of adipose tissue and many dilated lymphatic channels (arrows). Note that the skeletal muscle tissue (arrowhead) within the tumor. (H and E, $\times 40$) (Scalebar=50 μm).

intermuscular cavernous lymphangioma with clusters of ectatic lymphatic channels. Marked proliferation of adipose tissue around the lymphatic channels was noted (Fig. 1e and f). There was no lipoblast in the adipose tissue. Skeletal muscle tissue replaced by fatty tissue was identified.

3. Discussion

Benign soft tissue lymphatic tumors include solitary lymphangioma, lymphangiomatosis, lymphangiomyoma, and lymphangiomyomatosis. Histological features of lymphangioma have been classified on the basis of the size of the lymphatic channels as capillary lymphangioma, cavernous lymphangioma, and cystic lymphangioma [1]. The distinction between cavernous and cystic lymphangioma is not always clear-cut and they often coexist.

Cavernous lymphangiomas are commonly encountered in the mouth, lips, cheek, tongue or other areas. About 50 to 65% of these lesions are present at birth, and 80–90% are clinically manifest within the first 2 years of life [2]. Lymphangiomas are believed to be congenital lymphatic malformations that are the result of non-communication between sequestered lymphoid tissue and the peripheral lymphatic system [2]. Histologically, cavernous lymphangioma is characterized by dilated lymph channels lined by a flattened layer of endothelial cells with separation of the dermal collagen bundles [4].

MRI findings of soft tissue lymphangiomatous tumors have rarely been reported [2–5]. Most of these tumors appear as well-defined cystic masses with high signal intensity on T2-weighted images and slightly higher signal intensity than the surrounding muscles on T1-weighted images. After gadolinium administration, a more heterogeneous enhancement of the tumor usually can be seen. Other cases with features of macrocystic spaces due to lymphatic drainage obstruction and homogeneous, septated lymphangioma have been reported [2,6].

In this report, we described MRI findings of an intermuscular cavernous lymphangioma composed of fatty tissues interspersed with slow flowing vascular channels. The low-signal-intensity serpiginous structures on T1-weighted images, hyperintense on proton density and gradient-echo T2* weighted images, and marked enhancement are compatible with slow-flow venous or lymphatic channels. On the contrary, fast-flow vessels are usually depicted by a signal-void structures on both T2-weighted and gadolinium-enhanced T1-weighted images, which was not seen in our case. The non-fatty areas of hyperintense signal on gadolinium-enhanced T1-weighted images may be attributed to the tumor with numerous lymphocytes, as documented in our histologic findings.

Conversely, intermuscular hemangiomas are usually depicted by signal intensity equal to or slightly lower than that of muscle, unless complicated by hemorrhage, infection, or prior surgery [7]. Hemangiomas may show

presence of phleboliths. Flow-voids arising from feeding arteries or draining veins can be seen in fast-flow hemangiomas. After gadolinium administration, a more homogeneous enhancement of contrast medium in the tumor is usually seen. Magnetic susceptibility effects due to deposition of blood components such as hemosiderin in the vascular lakes are often easily detected in hemangiomas. However, it can be impossible to distinguish hemangiomas from lymphangiomas on MRI.

Soft tissue masses with predominately fatty components may easily approach to the appearance of lipoma or well-differentiated liposarcoma. These tumors usually have irregular thin or thickened, linear, or nodular septa. These non-adipose structures may show a non-specific decreased signal on T1-weighted images, a variably increased signal on T2-weighted and gadolinium-enhanced T1-weighted images [8]; these features can be similar to those of our case. Higher grade liposarcomas typically contain little or no macroscopic fat. Lipoma variants, such as chondroid lipoma and osteolipoma, usually contain nodular foci of low signal intensity. Angiolipoma typically shows a fatty mass with nodular foci of high signal on T2-weighted and gadolinium enhanced images [8].

Arteriography is helpful to define a hypervascular tumor, but venous or lymphatic tumors are often not visualized and so arteriography may underestimate the extent of the lesion. Similarly, MRI angiography added little information for establishing the diagnosis of lymphangioma.

Debulking surgery is the treatment of choice for soft tissue lymphangiomatous tumors. Resection of cavernous lymphangiomas arising in and infiltrating the muscular fibers may have a higher local recurrence rate and complications of nerve palsies.

4. Summary

A case of an intermuscular cavernous lymphangioma predominantly composed of fatty tissue in the arm is presented. Although it is non-specific, some serpiginous slow-flow vascular structures within a soft tissue mass must be considered as one differential diagnosis of cavernous lymphangioma on MRI.

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