

Hibernoma of the axillary region: a rare benign adipocytic tumor

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Abstract

Hibernoma is a rare benign tumor considered to arise from remnants of fetal brown adipose tissue. It tends to occur in sites where brown fat persists beyond fetal life, such as the interscapular region, but can occur in sites where brown fat is usually absent in adults. Clinically, hibernomas are slow-growing, asymptomatic tumors. However, unlike lipomas, MRI findings sometimes mislead clinicians to diagnose a malignant neoplasm. We describe a 63-year-old male with an axillary hibernoma involving the brachial neurovascular bundles and mimicking a well-differentiated liposarcoma, from which it should be distinguished.

Introduction

Hibernoma is a rare benign adipose tumor composed in part of brown fat cells with granular, multivacuolated cytoplasm.¹ In contrast to a lipoma, which originates from white adipose tissue and is among the most common soft-tissue tumors, hibernoma is listed among the rarest. It has been recognized since the very early years of the last century, and was first described by Merkel in 1906 as being composed of brown adipose tissue;² then the term of hibernoma was proposed in 1914 by Gery. Hibernoma occurs predominantly in young adults, in a wide variety of locations but usually in sites where brown fat persists beyond fetal life. The most common site is the thigh, followed by the trunk, upper extremity, and head and neck.³ We describe here a case of a 63-year-old male with a large axillary hibernoma involving the brachial neurovascular bundles and mimicking a well-differentiated liposarcoma.

Case Report

A 63-year old man was referred to our institute because of a large soft-tissue mass in his left axilla. The patient was very healthy and completely asymptomatic as regards the tumor, having full, normal left arm functions and no lymphedema in his left arm and hand. The overlying skin and underlying tissues of the mass were moveable and not very adhesive. The palpation of the tumor revealed a soft mass compatible with a lipoma, without localized tenderness or Tinel's sign. A magnetic resonance image (MRI) of the shoulder showed a 150x90x60 mm long, well-defined tumor in the left axillary region (Figure 1A). Both T1 and T2 weighted imaging showed relatively homogeneous high intensity with scatter hypo- or iso-signal intensity areas. There was an area in the lower apex region of the tumor that was not suppressed in fat suppression imaging, suggesting the presence of a nonlipoma-like component (Figure 1B) The radiologist suggested the possible differential diagnosis of a well-differentiated liposarcoma. The neurovascular bundles were involved, running through the tumor (Figure 1C). Because brachial neurovascular bundles should be preserved, the tumor mass was completely excised with dissection of the neurovascular bundles from within the tumor, which underwent histopathological examination. Macroscopically, the fresh specimen was a well circum-

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scribed, lobulated, tan-brown, homogeneous tumor with a hypervascular surface (Figure 2).

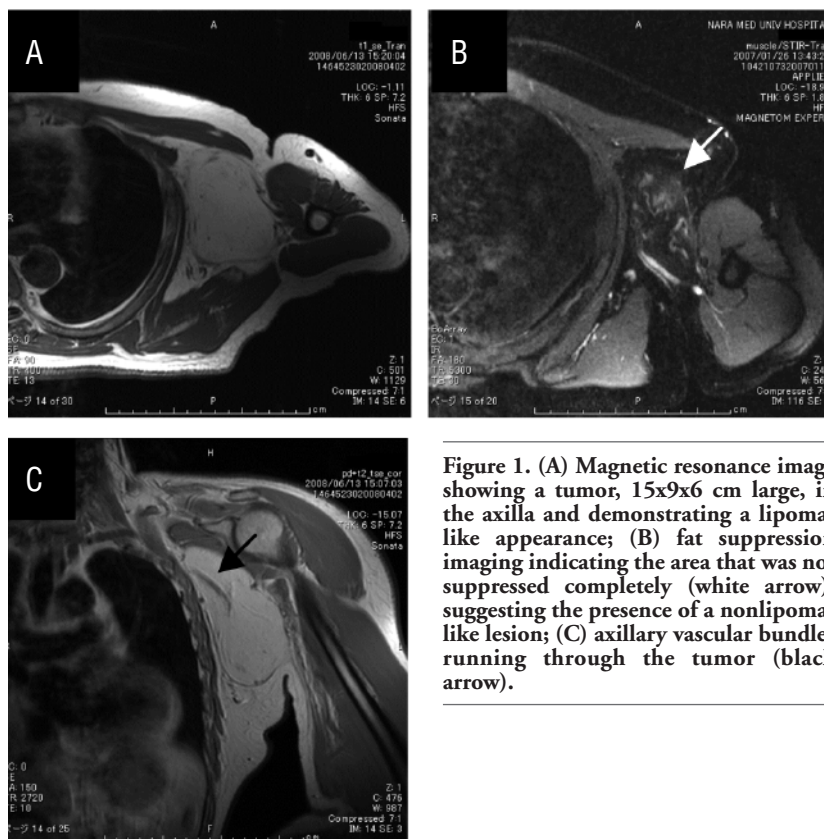


Figure 1. (A) Magnetic resonance image showing a tumor, 15x9x6 cm large, in the axilla and demonstrating a lipoma-like appearance; (B) fat suppression imaging indicating the area that was not suppressed completely (white arrow), suggesting the presence of a nonlipoma-like lesion; (C) axillary vascular bundles running through the tumor (black arrow).

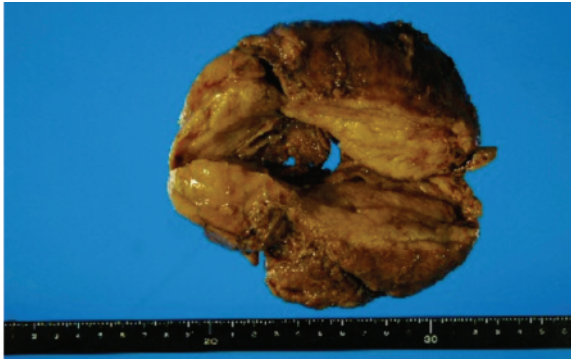


Figure 2. The macroscopic appearance of the hibernoma, showing a lipoma-like encapsulated, lobular, greasy, tan-brown cut surface.

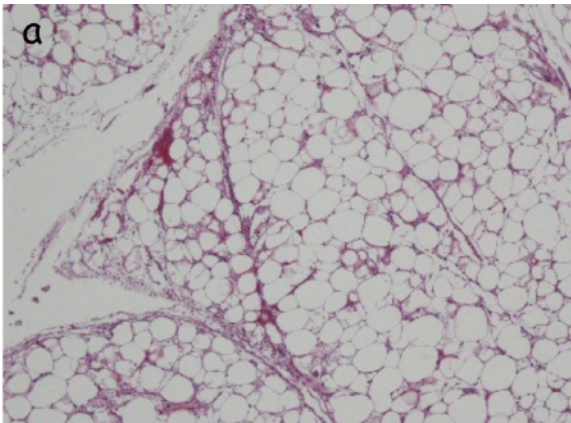
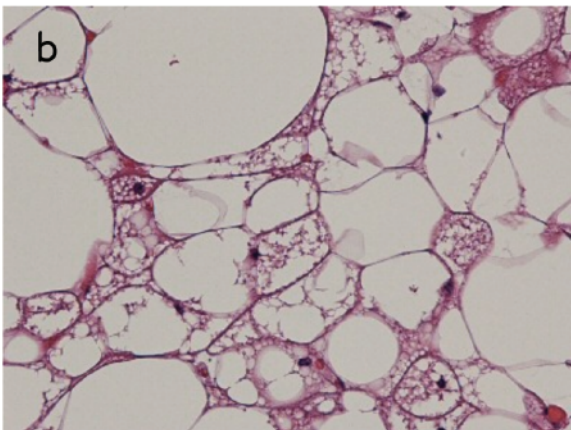


Figure 3. (a) The tumor was composed of cells arranged in lobules separated by fine reticulin fibers (magnification: 40x); (a) both univacuolated lipoma-like cells and round or polygonal cells with granular or multivacuolated lipid-containing cytoplasm and small, centrally placed nuclei were observed (magnification: 100x); (hematoxylin and eosin stain).



Light microscopic examination showed tumor cells arranged in lobules separated by fine reticulin fibers (Figure 3A). The cells were both univacuolated lipoma-like cells and round or polygonal cells, with granular or multivacuolated lipid-containing cytoplasm and small, centrally placed nuclei (Figure 3B). This led to the diagnosis of hibernoma. In addition, small capillary proliferation was seen, and neither mitosis nor atypia was observed by close examination of multiple, representative sections. Immunohistochemistry showed that only S-100 protein was focally positive, and other indicators including TOPOII and CD68 were negative. There was no local recurrence or aggressive disease observed after the removal of the tumor, and no functional loss was observed in the patient's left upper limb.

Discussion

Hibernomas are very rare benign adipocytic tumors originating from brown adipose tissue. Brown adipose tissue is believed to have a role in thermoregulation (in relation to "hibernation"), first recognizable in human fetuses.⁴ Therefore, hibernoma is also termed as lipoma of immature adipose tissue, lipoma of embryonic fat, or fetal lipoma.⁵ In the adult, brown fat persists in the neck, axilla, mediastinum, and perirenal and perirenal areas.^{4,6} In a report of a large series of hibernomas, the most frequent site is the limbs; 30% of tumors were located in the thigh.³ According to the persistent presence of brown fat tissue, the neck, axilla, interscapular area, and intrathoracic

area are regarded as frequent sites as well.^{7,8} The peak incidence of hibernomas is in the third decade of life, younger age-groups than for conventional lipomas, and slightly more predominant in females than in males.^{3,5,9} Clinically, the typical presentation of this tumor is as a progressive, painless enlargement, and localized tenderness is rare.

The macroscopic appearance of the tumor is a usually well-defined, soft, and mobile mass with the color of tan to brown.⁵ Microscopically, four histological subtypes have been described as a lipoma-like variant, nonlipoma-like (typical) variant, myxoid variant, and spindle cell variant.^{1,3} The most common or typical variant of hibernoma displays a distinct lobular pattern, and is composed of cells that show varying degrees of differentiation, ranging from uniform, round or ovoid, granular, eosinophilic cells with a distinct cellular membrane to multivacuolated cells with multiple lipid droplets and centrally placed nuclei.^{5,10} The differences between the typical variant of hibernoma and the lipoma-like variant or lipoma can be distinguished according to the ratio of multivacuolated adipocytes (brown fat) and univacuolated adipocytes (white fat). Tumors containing more than 70% multivacuolated adipocytes are generally accepted as typical hibernomas.¹⁰ Immunohistochemistry is not necessary, but staining for S-100 protein is usually positive.^{3,5}

The nonlipoma-like subtype always presents particular characteristics on MRI; for example, being heterogeneous, hypo- or iso-intense, with hypervascularity.¹¹ Thus, the appearance of nonlipoma-like hibernomas on MRI sometimes misleads clinicians to confuse the diagnosis with well-differentiated liposarcomas or atypical lipoma variants. In such cases, the computerized topography-guided biopsy or incisional biopsy would be required for a definitive preoperative diagnosis.

Curative treatment of hibernomas is complete excision, preserving vital structures. The vascular supply is considerably more prominent in hibernomas than in lipomas, therefore it should be treated with care to avoid postoperative bleeding or hematoma formation. Follow-up in 66 patients (mean period of 7.7 years) revealed no local recurrences or evidence of aggressive behavior, although many of these tumors were incompletely excised.³

We concluded that hibernoma is a very rare tumor, but should be included in the differential diagnosis of fatty soft-tissue tumors. It can be well managed surgically, and the outcome is usually excellent without any local and distant aggressive disease.

References

1. Miettinen MM, Fanburg-Smith JC, Mandahl N. Hibernoma. In: WHO Classification of tumours. Pathology and genetics of tumours of soft tissue and bone. Fletcher CDM, Unni KK, Mertens F, editors. Lyon (France): IARC Press 2002; pp33-4.
2. Merkel H. On a pseudolipoma of the breast (peculiar fat tumor). *Beitr Pathol Anat* 1906;39:152-7.
3. Furlong MA, Fanburg-Smith JC, Miettinen MM. The morphologic spectrum of hibernoma. A clinicopathologic study of 170 cases. *Am J Surg Pathol* 2001;25:809-14.
4. Kösem M, Karakök M. Hibernoma: A case report and discussion of a rare tumor. *Turk J Med Sci* 2001;25:175-6.
5. Enzinger FM, Weiss SW. *Soft tissue tumors*. 5th ed. Philadelphia: Mosby-Elsevier, 2009, pp466-70.
6. Stephen H, Sanjay RP, John WB. Hibernoma of the pericardium presenting as pericardial effusion in a child. *Ped Pathol* 1990;10:575-80.
7. Ahn C, Harvey JC. Mediastinal hibernoma. A rare tumor. *Ann Thorac Surg* 1990;50:828-30.
8. Udadia ZF, Kumar N, Bhadura AS. Mediastinal hibernoma. *Eur J Cardiothorac Surg* 1999;15:533-5.
9. Rigor VU, Goldston E, Jones J, et al. Hibernoma: A case report and discussion of a rare tumor. *Cancer* 1986;57:2207-11.
10. Peycru T, Tarat E, Schwartz A, et al. Hibernoma of the neck: a rare benign tumour. *J Can Chir* 2009;52:E52-3.
11. Ritchie DA, Aniq H, Davies AM, et al. Hibernoma – correlation of histopathology and magnetic-resonance-imaging features in 10 cases. *Skeletal Radiol* 2006;35:579-89.