

Presacral hibernoma: Radiologic-pathologic correlation

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Abstract

Hibernoma is a rare benign tumor of brown fat origin. It is found in locations where brown adipose tissue is present in the fetus. These locations include the neck, axilla, thorax, and extremities. Retroperitoneal hibernoma is an extremely rare site of presentation. We present a rare case of presacral hibernoma. Preoperative embolization of this highly vascular tumor was performed. To the best of our knowledge, this is the first reported case of a presacral hibernoma in the English literature.

Key words: Brown fat tumor; computed tomography; hibernoma; presacral mass

Introduction

Hibernoma is a rare, benign, slow-growing tumor of brown fat origin the majority occurring in the extremities and the neck.^[1] The presacral location is rare, although a few cases in the retroperitoneum have been described in the literature.^[2-5] We describe a case of presacral hibernoma with radiologic-pathologic correlation. The imaging findings are nonspecific but, given the rarity of this tumor, when there is a large well-defined heterogeneous retroperitoneal mass with dilated intratumoral vessels, the possibility of a hibernoma should be considered in the appropriate clinical setting.

Case Report

A 36-year-old female presented with abdominal distention and dull, vague, left lower quadrant pain since 2 years. There was no history of weight loss or menstrual irregularities. On clinical examination, a large solid pelvic mass was found and pelvic ultrasound was requested. On

ultrasound, there was an ill-defined large heterogeneous mass with increased vascularity seen posterior and superior to the uterus, with poor demarcation from the uterus and the left ovary. The possibility of a uterine leiomyosarcoma or an ovarian neoplasm was considered. The patient being unwilling for MRI, CT scan of the pelvis was performed for further evaluation. CT scan revealed a large presacral, heterogeneous, hypervascular mass, measuring approximately 10.6 × 10.0 × 5.8 cm and causing compression and displacement of the rectum anteriorly [Figure 1A-C]. This mass had multiple tortuous arterial and venous channels, with the dominant arterial supply being from branches of the inferior mesenteric artery (IMA). The possibility of retroperitoneal leiomyosarcoma was raised. A CT scan guided True-Cut® biopsy was performed [Figure 2]. On microscopy, there were numerous multivacuolated adipose cells with eosinophilic cytoplasm and eccentric nuclei, consistent with hibernoma [Figure 3].

As the mass was hypervascular, preoperative embolization of this mass was clinically requested. On selective IMA catheter angiogram, there were dilated vascular branches feeding this mass, with an early tumor blush, but no arteriovenous communication was seen [Figure 4A]. Selective particle embolization of the feeding branch was performed [Figure 4B]. The tumor was then surgically resected.

On gross examination, the mass was lobulated and well encapsulated, measuring 13.1 × 12.6 × 4.2 cm. The cut surface was brownish yellow in color, with multiple coursing

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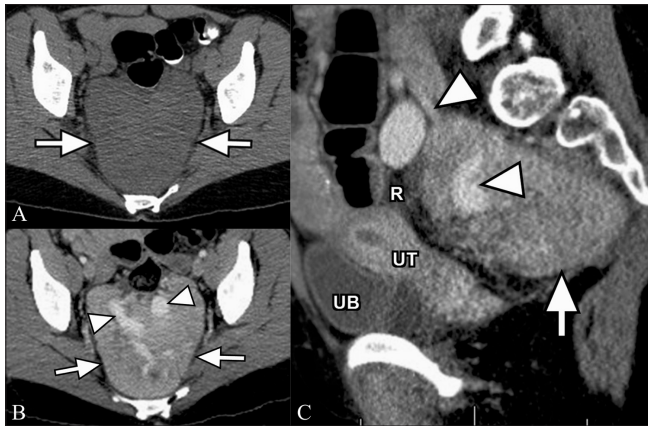


Figure 1 (A-C): Noncontrast (A) and contrast-enhanced (B) axial CT scans through the pelvis demonstrate a large well-defined, heterogeneous, enhancing presacral mass (arrows), with dilated tortuous vessels (arrowheads). Sagittal reformatted contrast-enhanced image (C) demonstrates the mass pushing the rectum (R) and uterus (Ut) anteriorly. UB = urinary bladder

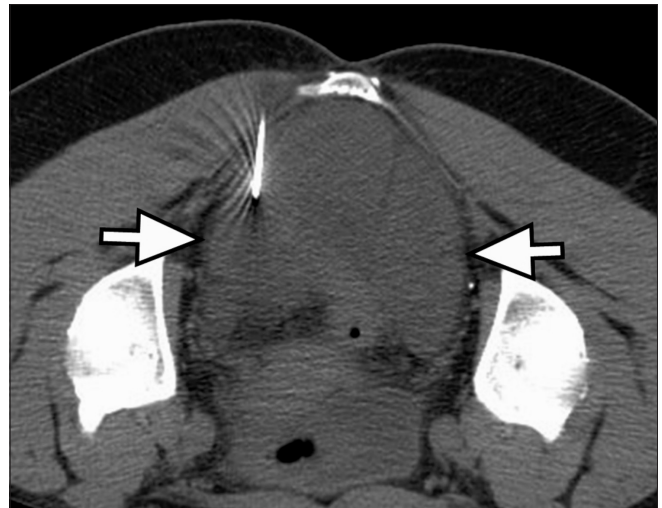


Figure 2: Prone, axial, plain CT scan through the pelvis, with the biopsy needle (arrow) in the presacral mass

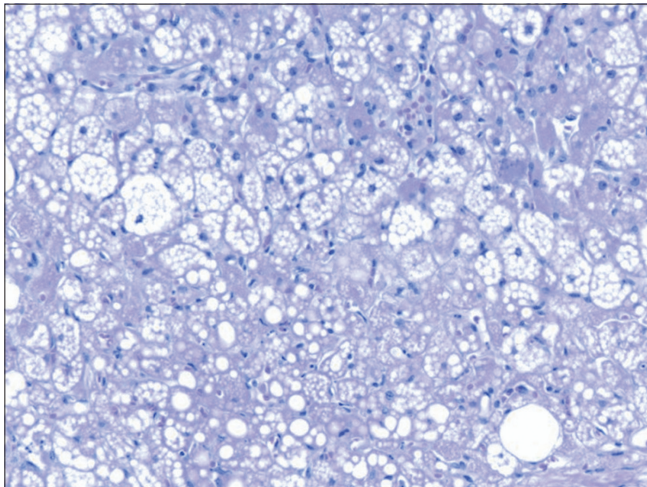


Figure 3: Photomicroscopic image of the biopsy specimen shows numerous multivacuolated adipose cells, with eosinophilic cytoplasm and eccentric nuclei

vessels and without any areas of hemorrhage or necrosis [Figure 5].

Discussion

Hibernoma is a rare, benign, slow-growing tumor of brown fat origin.^[1] The tumor is named hibernoma due to its similarity to brown fat, which has a thermogenic role in hibernating animals.^[6] The tumor was first termed as pseudolipoma by Merkel in 1906 and subsequently renamed hibernoma by Gery in 1914.^[7,8] The imaging features of this rare tumor have been mostly described for lesions occurring the extremities and neck region, with only six cases of retroperitoneal hibernoma described in the

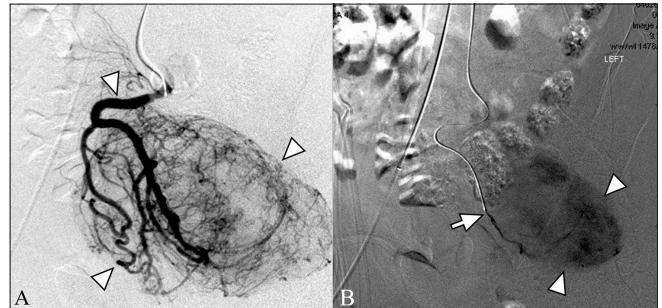


Figure 4 (A, B): Selective catheter angiogram of the inferior mesenteric artery (IMA), (A) shows a feeding vessel (arrow) and early tumor blush (arrowhead). Selective IMA angiogram, (B) after particle embolization of the feeding branch of the IMA shows the embolized feeding branch (arrow) with near complete loss of tumor blush (arrowhead)

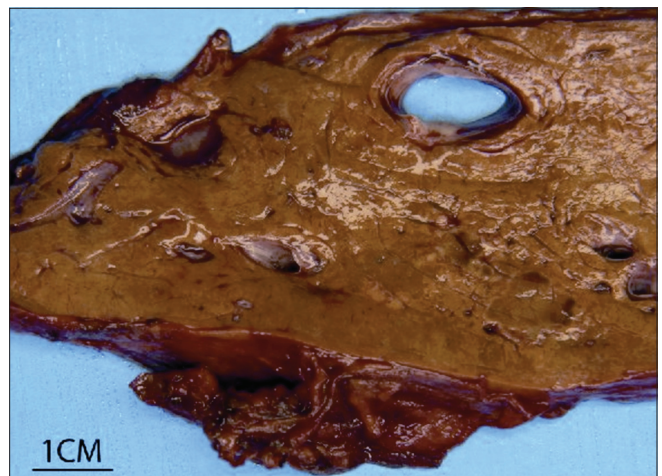


Figure 5: Cut surface of the gross specimen shows the brownish-yellow color of the mass, with multiple vessels but without any areas of hemorrhage or necrosis

English literature.^[1,2-5] To the best of our knowledge, this is the first case report of a presacral hibernoma, describing cross-sectional CT imaging and conventional catheter angiographic feature of this tumor with preoperative vascular embolization.

Hibernoma is seen in the 20- to 50-year age-group, with a female preponderance. However, a large series by the Armed Forces Institute of Pathology (AFIP) did show a slight male preponderance.^[9] The intra-abdominal hibernoma, by itself, does not produce symptoms and as a result the tumor is often large at initial presentation, the large size of the tumor causing abdominal discomfort and mass effect on adjacent structures.^[10]

The USG findings of intra-abdominal hibernoma have not been described, but subcutaneous and intramuscular hibernomas are seen as well-defined heterogenous masses with variable echogenicity; some may appear as hypoechoic masses, with increased vascularity on color Doppler flow.^[11]

On CT scan, the hibernoma is seen as a large heterogeneous, well-circumscribed mass, with attenuation ranging between fat and muscle.^[1,5] The heterogeneity of the hibernoma is attributed to the presence of varying amount of fat, muscle, and vessels. In our case, the mass demonstrated soft-tissue attenuation, without any definite fat attenuation or necrosis. Following intravenous contrast administration, the hibernoma shows heterogeneous enhancement, with hypertrophic branching intratumoral vessels as was seen in our case.^[12] Hibernomas have rarely been described to show minimal postcontrast enhancement.^[2,4] Retroperitoneal hibernoma causes displacement of the adjacent organs and structures but no infiltration.^[13]

On MRI, hibernoma demonstrates high signal intensity on T1W and T2W images; however, the intensity is less than that of subcutaneous fat and there is incomplete fat suppression on fat-saturated pulse sequences.^[14] The most important feature on MRI is the presence of large branching vessels.

The differential diagnosis of a large well-defined heterogenous retroperitoneal and/or presacral mass containing large intratumoral vessels could include liposarcoma, angioliipoma, and hemangiopericytoma. Teratoma does not demonstrate internal vascularity. Liposarcoma are heterogenous soft-tissue masses with areas of necrosis, hemorrhage, and/or calcification, but without large intratumoral vessels.^[15] Also, in contrast to the slow-growing well-defined hibernoma, the liposarcoma has a poor margin, with infiltration into adjoining structures.^[2] A mature teratoma classically presents as a complex mass containing fat, calcification, rarely a fat-fluid level, without intralesional vascularity.^[16] Angioliipoma and hemangiopericytoma may have increased vascularity;

however, none of these lesions show both increased signal intensity on T1W and T2W images and large branching intratumoral vessels.

On angiography, hibernoma demonstrates dilated feeding vessels, typical tumor blush, and early venous return, as was seen in our case.^[17] In the past, angiography was performed for evaluating presurgical vascular anatomy and also to differentiate hibernoma from hypovascular lesions like lipomas and fibromas.^[2] These days, CT and MRI have superseded the role of conventional angiography in the diagnosis of such lesions.

Given the nonspecific features on imaging, histological diagnosis is usually required for appropriate planning before surgery. Despite the increased vascularity of this tumor, image-guided core biopsy is reported to be without complications.^[2]

Microscopically, hibernoma is composed of multivacuolated adipocytes with granular eosinophilic cytoplasm and eccentric nuclei, univacuolated cells with peripheral nuclei, and abundant capillaries.^[5]

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References

1. Sansom HE, Blunt DM, Moskovic EC. Large retroperitoneal hibernoma--CT findings with pathological correlation. *Clin Radiol* 1999;54:625-7.
2. Cantisani V, Morteale KJ, Glickman JN, Ricci P, Passariello R, Ros PR, *et al*. Large retroperitoneal hibernoma in an adult male: CT imaging findings with pathologic correlation. *Abdom Imaging* 2003;28:721-4.
3. Delsignore A, Ranzoni S, Arancio M, Marchetti C, Landi G, Mina A, *et al*. Kidney hibernoma: Case report and literature review. *Arch Ital Urol Androl* 2010;82:189-91.
4. Gulmez I, Dogan A, Balkanli S, Yilmaz U, Karacagil M, Tatlisena A. The first case of periureteric hibernoma. Case report. *Scand J Urol Nephrol* 1997;31:203-4.
5. Rigor VU, Goldstone SE, Jones J, Bernstein R, Gold MS, Weiner S. Hibernoma. A case report and discussion of a rare tumor. *Cancer* 1986;57:2207-11.
6. Cannon B, Nedergaard J. Brown adipose tissue: Function and physiological significance. *Physiol Rev* 2004;84:277-359.
7. Merkel H. Uber ein pseudolipom der mamma (Eigenartiger Fettzellentumor). *Beitr Path Anat* 1906;39:152-7.
8. Gery L. Id. MF Bonnel's paper. *Bull Mem Soc Anat* 1914;89:111-2.
9. Furlong MA, Fanburg-Smith JC, Miettinen M. The morphologic spectrum of hibernoma: A clinicopathologic study of 170 cases. *Am J Surg Pathol* 2001;25:809-14.
10. Dardick I. Hibernoma: A possible model of brown fat histogenesis. *Hum Pathol* 1978;9:321-9.
11. Harges J, Scheil-Bertram S, Hartwig E, Gebert C, Gosheger G, Schulte M. Sonographic findings of hibernoma. A report of two cases. *J Clin Ultrasound* 2005;33:298-301.

12. Ward SC, Hornsby VP. "The Dormouse is asleep again" said the Hatter, and he poured a little hot tea upon its nose (Lewis Carroll). *Br J Radiol* 1990;63:733-4.
13. Anderson SE, Schwab C, Stauffer E, Banic A, Steinbach LS. Hibernoma: Imaging characteristics of a rare benign soft tissue tumor. *Skeletal Radiol* 2001;30:590-5.
14. Drevelegas A, Pilavaki M, Chourmouzi D. Lipomatous tumors of soft tissue: MR appearance with histological correlation. *Eur J Radiol* 2004;50:257-67.
15. Colville J, Feigin K, Antonescu CR, Panicek DM. Hibernoma: Report emphasizing large intratumoral vessels and high T1 signal. *Skeletal Radiol* 2006;35:547-50.
16. Davidson AJ, Hartman DS, Goldman SM. Mature teratoma of the retroperitoneum: Radiologic, pathologic, and clinical correlation. *Radiology* 1989;172:421-5.
17. Rossi P, Francone A, Martini S, Pavone P. Multiple imaging modalities in a case of hibernoma. *Eur J Radiol* 1988;8:125-7.

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