

CASE REPORT

Intraosseous Hibernoma

A Case Report and Review of the Literature

Imran Hafeez, MD,* Steven Shankman, MD,* Jon Michnovicz, MD,† and Vincent J. Vigorita, MD*

Study Design. A case report and a literature review are presented.

Objective. To describe and review the clinical presentation and characteristic imaging and pathology findings of intraosseous hibernoma.

Summary of Background Data. Hibernomas are lesions of brown fat. Brown fat is typically found in newborn mammals and is rich in mitochondria, thus enabling its role in thermoregulation. It represents a small proportion of adult fat and is distinct from the more common "white fat." Rarely does a hibernoma occur within bone. To the authors' knowledge, 5 cases in all have been reported in the literature.

Methods. We report the first case to our knowledge of an intraosseous hibernoma occurring within the lumbar spine as well as a review of the literature.

Results. Characteristic findings from magnetic resonance studies include variable T1W signal relative to skeletal muscle and hyperintense signal on fluid-sensitive imaging. Computed tomography has consistently demonstrated a sclerotic lesion with variable definition. Pathologic findings include sheets of multivacuolated cells with centrally placed nuclei and numerous tiny surrounding cytoplasmic vacuoles overlying bony trabeculae.

Conclusion. Our review of the literature demonstrates that intraosseous hibernoma is most likely an incidental finding with a predilection for the lower extremities in middle-aged females.

Key words: chordoma, hibernoma, lumbar spine, middle-aged females, benign intraosseous lesion, brown fat, lipoma, hemangioma, lower extremity.

Level of Evidence: 4

Spine 2015;40:E558–E561

Hibernomas are rare benign tumors consisting of brown fat, the type of adipose tissue seen in hibernating animals. Although present in greatest abundance in human newborns, its presence steadily decreases thereafter and accounts for less than 1% of total body mass in adult humans.¹ Hibernomas in humans are usually observed in the soft tissue of the neck, axillae, mediastinum, and periaortic perirenal zones.² Intraosseous hibernomas are extremely rare.^{3,4} Herein, we report such a case and discuss the clinical, radiographical, and pathological features of this unusual entity.

REPORT OF A CASE

A 67-year-old female at a long-term nursing facility with a long-standing history of diabetes and immobilization secondary to a complex psychiatric history presented with a pressure sore. Physical examination revealed a soft tissue sacral decubitus ulcer with overlying warmth and redness and a clinical concern for underlying osteomyelitis. The patient is mentally disabled but did not report any pain, fever, chills, night sweats, or weight loss. Her medical history was otherwise noncontributory and she has no history of malignancy.

RADIOLOGICAL FINDINGS

Magnetic resonance image of the lumbar spine, initially obtained to rule out osteomyelitis, demonstrates diffusely hypointense signal of the entire L3 vertebral body with T1WI and diffusely hyperintense signal with T2WI (Figures 1–2). Computed tomographic scans obtained just prior to computed tomography-guided biopsy reveal speckled sclerosis with intervening lucency at the L3 vertebral body most suggestive of hemangioma (Figure 3). Bone scintigraphy and positron emission tomography were not performed.

PATHOLOGIC FINDINGS

The computed tomography-guided core biopsy of the L3 vertebral body yielded tan-red/tan-brown soft and bony tissue. The biopsy revealed diffusely thickened sclerotic bone with irregular contours. The bone had features of both lamellar and woven bone (Figure 4). Although foci of normal hematopoietic marrow were present within the lesional tissue, the bulk of the marrow was replaced by cells of various appearances. Some foci showed polygonal cells with a finely granular brown cytoplasm. Other foci showed cells with abundant cytoplasm

From the *Maimonides Medical Center, Brooklyn, NY; and †National Institute for People With Disabilities, New York, NY.

Acknowledgment date: April 20, 2014. Revision date: August 28, 2014. Acceptance date: August 30, 2014.

The manuscript submitted does not contain information about medical device(s)/drug(s).

No funds were received in support of this work.

No relevant financial activities outside the submitted work.

Address correspondence and reprint requests to Imran Hafeez, MD, Department of Radiology, Maimonides Medical Center, 4805 Fort Hamilton Pkwy, Brooklyn, NY 11219; E-mail: ihafeez@maimonidesmed.org

DOI: 10.1097/BRS.0000000000000851

E558 www.spinejournal.com

Copyright © 2015 Wolters Kluwer Health, Inc. Unauthorized reproduction of this article is prohibited.



Figure 1. Sagittal T1W image of the lumbar spine demonstrating diffusely hypointense signal of the L3 vertebral body.

filled with a multivacuolated appearance mimicking lipoblasts (Figure 5). There were no identifiable mitoses and no significant cytologic atypia. Initial diagnostic considerations included storage disease, chordoma or notochordal rest, and a lipomatous



Figure 2. Sagittal T2W image of the lumbar spine demonstrating diffusely hyperintense signal of the L3 vertebral body.

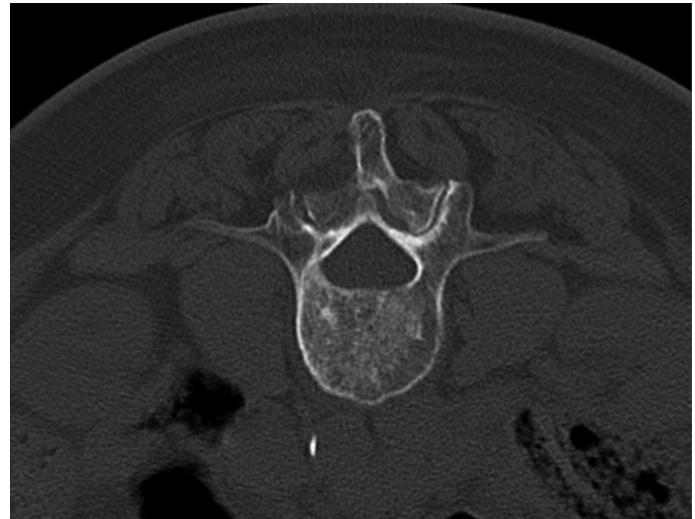


Figure 3. Axial computed tomographic scan of the L3 vertebral body demonstrating focal speckled sclerosis with intervening lucency.

type of tumor. Immunohistochemical stains were positive for S-100 and CD68 and negative for epithelial membrane antigen and AE1/AE3, ruling out a storage or chondromatous lesion. The combination of histological and immunohistochemistry findings confirms the diagnosis of hibernoma.

DISCUSSION

Intraosseous hibernoma is an exceedingly rare entity. The limited number of cases in the literature have demonstrated characteristic clinical, radiological, and pathologic findings. Intraosseous hibernomas are most commonly diagnosed in females (6:1) in the fifth and sixth decades of life. Intraosseous hibernomas on biopsy examination reveal a range of cellular features that could lead to initial difficulty in diagnosis. The presence of clusters of macrophage-like cells can mimic a storage disease such as Gaucher disease, and vacuolated cells can mimic a range of fatty tumors including lipoma and

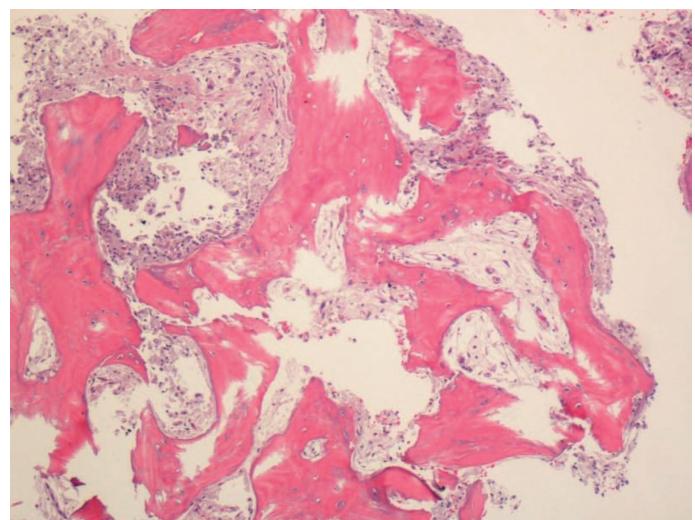


Figure 4. Biopsy reveals diffusely thickened sclerotic bone with irregular contours. The bone has features of both lamellar and woven bones.

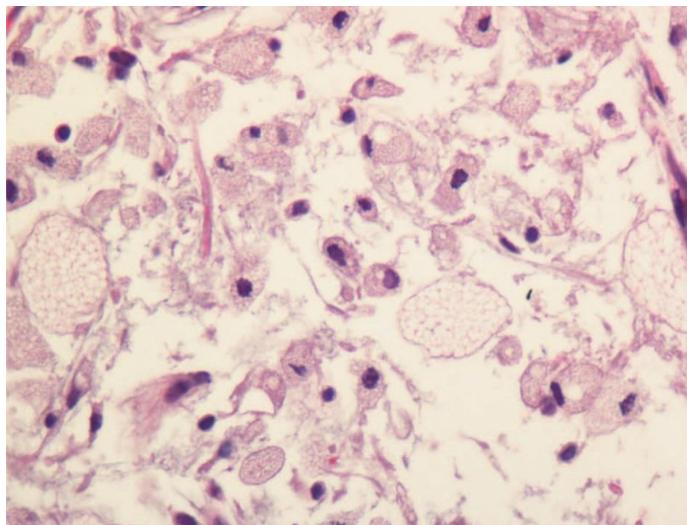


Figure 5. Some foci show polygonal cells with a finely granular brown cytoplasm. Other foci show cells with abundant cytoplasm filled with a multivacuolated appearance mimicking lipoblasts. There are no identifiable mitoses and no significant cytologic atypia.

even liposarcoma. The multivacuolated “physaliferous”-like cells can mimic a chordoma or notochordal rest. Immunohistochemistry profile of intraosseous hibernoma (+)S100, (−) cytokeratin, and (−) brachyury is helpful in excluding notochord lesions. Magnetic resonance imaging has demonstrated variable T1W signal relative to skeletal muscle and hyperintense signal on fluid-sensitive imaging, with variable enhancement and, as reported by Botchu *et al*,⁵ an additional rim of low signal on T1W and high signal on Short T1 Inversion Recovery. Computed tomography has consistently demonstrated a sclerotic lesion with variable definition. Pathologic findings are consistent with the expected findings of a soft-tissue hibernomatous lesion,^{6–8} with sheets of multivacuolated cells with centrally placed nuclei and numerous tiny surrounding cytoplasmic vacuoles overlying bony trabeculae. We report the first case to our knowledge of such a lesion involving the lumbar vertebrae and which in addition displayed radiological findings reminiscent of a hemangioma.

Hibernomatous lesions are derived from brown fat and appear grossly brown secondary to high vascularity and mitochondrial number.⁵ In mammals, brown fat gradually disappears with age with the loss of mitochondria and becomes white fat.¹

Our review of prior cases of intraosseous hibernoma (Table 1) suggests an overall predilection for the lower extremities and pelvis as well, and, as in our case, the lumbar spine. In only 1 case, an intraosseous hibernoma was successfully treated with radiofrequency ablation and which was followed by resolution of both clinical symptoms and radiological findings.⁹

The strong predilection for middle-aged females is of interest and not understood. The lesion most likely represents an incidental finding in the clinical workup of unrelated musculoskeletal disorders.

The lesion is benign and does not require treatment. Malignant transformation has not been reported.

TABLE 1. Clinicopathologic Information of Prior Intraosseous Hibernoma

Sex	Age (yr)	Site	Presentation	CT Findings	MR Finding	Other Imaging	Treatment	Source
F	61	N/A	Essential thrombo-cytopenia	N/A	N/A	N/A	None	Thoms <i>et al</i> ¹⁰
M	54	b/l femur	N/A	N/A	N/A	N/A	None	Reyes <i>et al</i> ¹¹
F	57	Left sacral ala	Low back pain	Lucent center, Sclerotic rim	T1: Heterogenously hyperintense; STIR/T2 fat-sat: heterogenously hypointense, postcontrast fat-sat T1: Mild heterogenous enhancement	None	None	Kumar <i>et al</i> ⁴
F	40	Right posterior ileum	Low back, buttock pain	Lucent center, sclerotic rim	T1: Intermediate signal with hypointense rim; STIR: high signal with high signal rim	Bone scan: Increased uptake	Analgesia	Botchu <i>et al</i> ⁵
F	50	Right ileum	Hip pain	Sclerotic, well-demarcated	T1: Hypointense, STIR: hyperintense	PET: SUV = 3.0	None	Bai <i>et al</i> ¹²
F	70	Left mass lateralis of sacral bone	Low back pain	sclerotic, well-Demarcated	T1: Hypointense, T2: hyperintense	None	RFA-ablation, successful	Ringe <i>et al</i> ⁶
F	67	L3 vertebral body	Pressure sore	Vague sclerosis	T1: Diffuse hypointensity of vertebral body, T2: Diffuse hyperintensity of vertebral body	None	None	Hafeez <i>et al</i> ^[this study]

CT indicates computed tomographic; MR, magnetic resonance; F, female; M, male; N/A, not available; STIR, Short T1 Inversion Recovery; PET, positron emission tomography; SUV, standard uptake value; RFA, radiofrequency ablation.

➤ Key Points

- Intraosseous hibernoma comprises brown fat, which plays a role in thermoregulation in mammalian newborns, and represents a small proportion of fat within adults.
- The lesion is benign and without malignant potential.
- It has characteristic clinical, radiological, and pathologic features that we describe within this report and review of the literature.
- Our review of prior cases demonstrates an overall predilection for the lower extremities in middle-aged females, the significance of which is not yet understood.

References

1. Gesta S, Tseng YH, Kahn CR. Developmental origin of fat: tracking obesity to its source. *Cell* 2007;131:242–56.
2. Furlong MA, Fanburg-Smith J, Miettinen M. The morphologic spectrum of hibernoma: a clinicopathologic study of 170 cases. *Am J Surg Pathol* 2001;25:809–14.
3. Chow LT, Lee KC: Intraosseous lipoma. A clinicopathologic study of nine cases. *Am J Surg Pathol* 1992;16:401–10.
4. Kumar R, Deaver MT, Czerniak BA, et al. Intraosseous hibernoma. *Skeletal Radiol* 2011;40:641–5.
5. Botchu R, Puls F, Hock YL, et al. *Skeletal Radiol* 2013;42:1003–5.
6. Enerbäck S. The origins of brown adipose tissue. *N Engl J Med* 2009;360:2021–3.
7. Nord KH, Magnusson L, Isaksson M, et al. Concomitant deletions of tumor suppressor genes MEN1 and AIP are essential for the pathogenesis of the brown fat tumor hibernoma. *Proc Natl Acad Sci U S A* 2010;107:21122–7.
8. Kransdorf MJ, Murphey MD. Lipomatous tumors. *Imaging of Soft Tissue Tumors*. Philadelphia, PA: Lippincott Williams & Wilkins; 1997:77–83.
9. Ringe KI, Rosenthal H, Länger F, et al. Radiofrequency ablation of a rare case of an intraosseous hibernoma causing therapy-refractory pain. *J Vasc Interv Radiol* 2013;24:1754–6.
10. Thorns C, Schardt C, Katenkamp D, et al. Hibernoma-like brown fat in the bone marrow: report of a unique case. *Virchows Arch* 2008;452:343–5.
11. Reyes AR, Irwin R, Wilson JD, et al. Intraosseous hibernoma of the femur: an unusual case with a review of the literature (poster #20). Paper presented at: Annual College of American Pathologists Meeting; 2008; San Diego, CA; 132:3.
12. Bai S, Mies C, Stephenson J, et al. Intraosseous hibernoma: a potential mimic of metastatic carcinoma. *Ann Diagn Pathol* 2013;17:204–6. doi:10.1016/j.anndiagpath.2012.07.001.

Copyright of Spine is the property of Lippincott Williams & Wilkins and its content may not be copied or emailed to multiple sites or posted to a listserv without the copyright holder's express written permission. However, users may print, download, or email articles for individual use.