










Spinal Intraosseous Hibernoma: A Case Report and Review of Literature

척추에서 발생한 골 내 동면종: 증례 보고와 문헌 고찰

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Hibernoma is a rare benign tumor that arises from vestiges of brown fat. Spinal intraosseous hibernoma has only recently been described in the literature, and only 12 cases have been reported to date due to its extreme rarity. Here, we report the case of a patient who was incidentally diagnosed with an intraosseous hibernoma in the thoracic spine, following a diverse imaging work-up and pathologic confirmation. We correlate the clinical presentation and imaging features of our case with those of previously reported cases during our review of the literature.

Index terms Lipoma; Spine; Computed Tomography, X-Ray; Magnetic Resonance Imaging; Positron-Emission Tomography

INTRODUCTION

Hibernoma is a rare benign tumor consisting of brown fat, the adipose tissue found in hibernating animals, that is commonly observed in the soft tissue of the thigh, followed by the shoulder, back, and neck (1). Intraosseous hibernomas are extremely rare (2), and its typical imaging features are poorly recognized. The few reports that include imaging findings include only a few case reports (2-5).

We present the clinical, radiological, and pathological features of a case of spinal intraosseous hibernoma. In addition, we comparatively review the clinical and radiologic features of our case with those of the 12 cases that were previously reported in the literature.

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
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CASE REPORT

A 54-year-old female presented with positive Interferon Gamma Release Assay test results following a tuberculosis screening. At the time of presentation, a low-dose chest computed tomography (CT) exam was performed to evaluate for pulmonary tuberculosis. The patient had experienced no local or systemic symptoms. The chest CT scan revealed a part-solid nodule in the right upper lobe, approximately 1 cm in diameter (Fig. 1A). A well-defined sclerotic lesion with intervening lucency was detected on the left side of the T7 vertebral body (Fig. 1B). A MRI scan of the thoracic spine revealed a well-defined, intraosseous lesion within the T7 vertebral body, approximately 2.6 cm in diameter. This lesion was characterized by heterogeneous hyperintensities on T2-weighted images (T2WI), and intermediate to hypointensities on T1-weighted images (T1WI), as compared with the paraspinous muscle or intervertebral

Fig. 1. A 54-year-old female with a spinal intraosseous mass detected on chest CT.

A. CT images of the right upper lobe demonstrate a partially solid nodule, approximately 1 cm in diameter.

B. Coronal (left image) and axial (right image) CT scans demonstrate a well-defined eccentric area of osteosclerosis with intervening lucencies in the T7 body that extends from the superior to the inferior endplate.

C. An axial T2WI image shows a well-defined and slightly inhomogeneous lesion primarily characterized by hyperintensity (left image). Axial T2WI image shows a hyperintense lesion with interspersed serpentine hypointense lines (left image, arrow). Axial T1WI image shows an intermediate signal intensity with interspersed hyperintense areas (middle image). Image of an axial T1 with fat saturation after contrast media injection; note the heterogeneous and intense enhancement (right image). The T2WI image finding of serpentine hypointense lines corresponds to persistent hypointensities in areas of intense enhancement on the axial T1 image following fat saturation (right image, arrow).

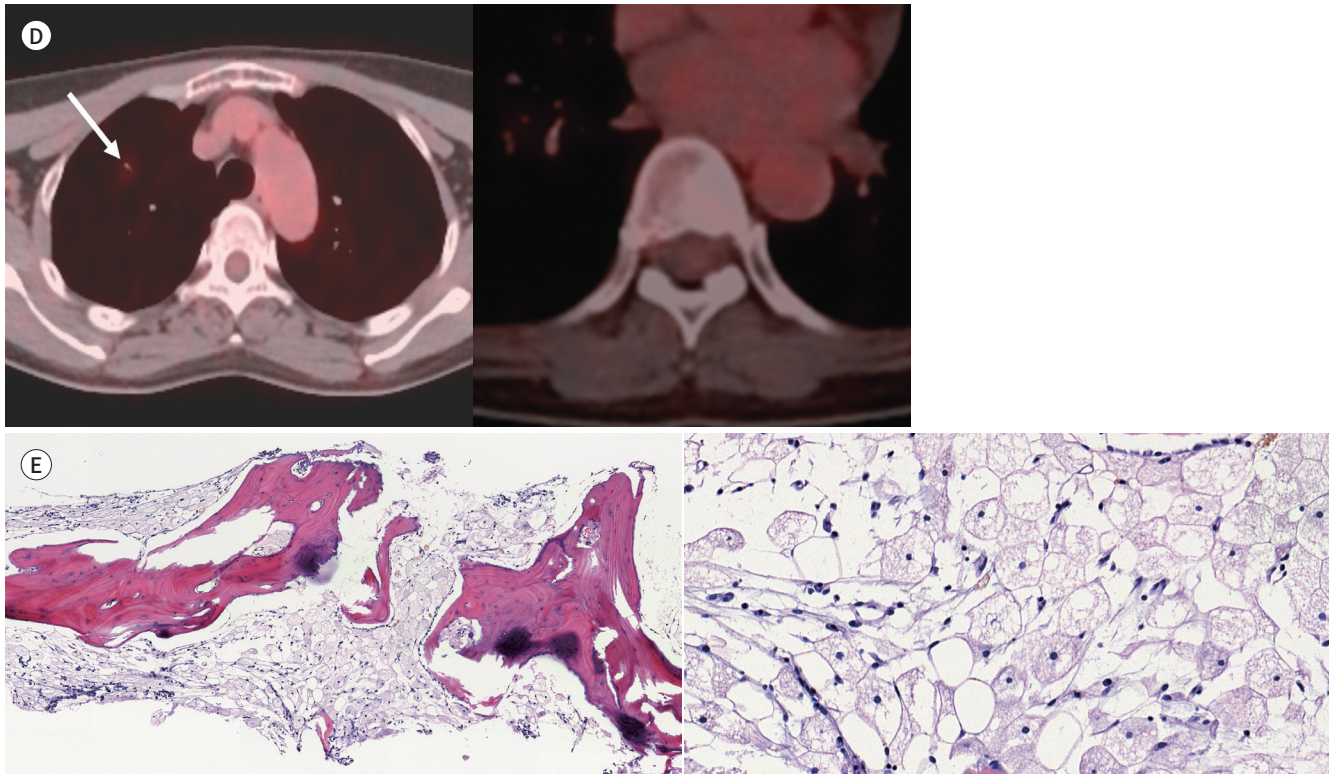
T1WI = T1-weighted image, T2WI = T2-weighted image



Fig. 1. A 54-year-old female with a spinal intraosseous mass detected on chest CT.

D. PET scans show low-grade fluorodeoxyglucose uptake of the part-solid nodule in the right upper lobe with a standardized uptake value of 1.1 (left image, arrow). Note the absent uptake of the sclerotic lesion in the T7 vertebral body (right image).

E. Pathologic features of intraosseous hibernoma. Transparent pale-brown fat cell infiltration is observed in the bone marrow cavity (left image, H&E stain, $\times 100$). The tumor contains a large number of brown fat cells with multi-vacuolation and central nuclei (right image, H&E stain, $\times 400$). H&E = hematoxylin and eosin



tebral disc. A post-contrast fat-saturated T1WI revealed heterogeneous contrast enhancement of the lesion (Fig. 1C). PET-CT was used to evaluate the lung nodule and the intraosseous mass at the level of T7. During the PET-CT scan, we observed mild fluorodeoxyglucose (FDG) uptake (max standardized uptake value: 1.1) in the right upper lung nodule. In contrast, no hypermetabolism was detected in the sclerotic T7 vertebral lesion (Fig. 1D). Based on these findings, we considered a differential diagnosis of atypical presentation of the hemangioma, or a possible blastic metastasis due to an intrapulmonary lesion.

Bone biopsy was performed with a 14-gauge core needle using the left transpedicular approach under fluoroscopic guidance. Histopathological examination of the core biopsy specimen confirmed the presence of transparent pale-brown fat cell infiltration in the bone marrow cavity. A large number of brown fat cells with multi-vacuolation and central nuclei were observed in the tumor mass (Fig. 1E). The morphological features of the tissue were consistent with brown fat, which indicated that the lesion was an intraosseous hibernoma. CT-guided core biopsy of the part-solid right lung nodule confirmed the diagnosis of adenocarcinoma and the patient was referred for lobectomy.

Table 1. Clinical Summary of Patients with Spinal Intraosseous Hibernoma that Have Been Previously Reported in the Literature

Reference	Age/Sex	Site	Reason for Imaging	Clinical Presentation	CT Finding (X-Ray)	MR Finding	Other Imaging
This case	54/F	T7 VB	Tuberculosis screening	Incidentally detected	Sclerotic lesion with intervening lucency	T1: heterogeneous intermediate to hypointensity T2: heterogeneous hyperintensity CE: heterogeneous enhancement	PET-CT: no hypermetabolism
Bonar et al. (5)	48/F	T5 VB	Swallowed glass	Incidentally detected	Sclerosis	T1: heterogeneous hypointensity T2: hyperintensity CE T1: peripheral enhancement T1: isointensity to muscle T2: hyperintensity	PET: mild hypermetabolism (max SUV 3.3) Bone scan: mild increased uptake Bone scan: subtle uptake
Hafeez et al. (4)	67/F	L3 VB	Pressure sore	Incidentally detected	Speckled sclerosis with intervening lucency	T1: diffuse hypointensity T2: diffuse hyperintensity	
Jermain et al. (9)	67/F	Left sacrum	Low back pain	Incidentally detected	PET-CT: well-defined sclerotic lesion	T1: hypointensity STIR: heterogeneous, hyperintense peripheral rim CE T1WI: moderate enhancement throughout the lesion and in the peripheral rim	Bone scan: increased metabolic activity PET: mild hypermetabolism
Kumar et al. (2)	57/F	Left sacral alar	Low back pain	Incidentally detected	Lucent center, sclerotic rim	T1: heterogeneously hyperintense; STIR/T2 fat-sat: heterogeneously hypointense, CE-FS T1: mild heterogeneous enhancement	
Ringe et al. (10)	70/F	Left mass lateralis of sacral bone	Low back pain	Low back pain	Sclerotic, well-demarcated	T1: Hypointensity T2: Hyperintensity	
Song et al. (3)	65/M	T12 VB	Hepatocellular carcinoma	Incidentally detected	Sclerosis	T1: heterogeneous hypointensity T2: hyperintensity	Bone scan: increased uptake
	71/F	L3 VB	Low back pain	Three patients had other cause of pain on imaging, one patient only showed intraosseous lesion, but relieved with analgesic	Sclerosis	T1: heterogeneous hypointensity T2: hyperintensity	PET: mild hypermetabolism
	49/M	T12 VB	Low back pain		X-ray: sclerosis	T1: heterogeneous hypointensity T2: hyperintensity	
	68/M	Sacral ala	Low back pain		Osteolysis with peripheral sclerosis	T1: heterogeneous hypointensity T2: hyperintensity	
	56/F	L3-4 VB	Low back pain		Sclerosis	T1: heterogeneous hypointensity T2: hyperintensity	
Westacott et al. (6)	84/F	Sacrum	Right hip and low back pain	Cause of pain Relieved with analgesics	Lytic bone lesion	N/A	

CE = contrast-enhanced, FS = fat-saturated, N/A = not available, RFA = radiofrequency ablation, STIR = short T1 inversion recovery, SUV = standard uptake value, T1WI = T1-weighted image, VB = vertebral body

DISCUSSION

Spinal intraosseous hibernoma is an exceedingly rare entity. Its characteristic clinical, radiological, and pathologic features are reported within only 12 case reports. The clinical and radiologic features of these cases are summarized in Table 1. Patients with spinal intraosseous hibernomas tend to be female (male-to-female ratio of 3:9), with an age range of 49–84 years. Reported lesion locations include the sacrum ($n = 5$), thoracic spine ($n = 4$), and lumbar spine ($n = 3$). All lesions were detected incidentally during clinical workups for unrelated musculoskeletal disorders, and were mostly unrelated to the patients' symptoms.

The published cases share several imaging features. The majority of cases showed a sclerotic lesion with a variable definition on CT or radiograph. Eleven cases showed sclerotic masses on CT or radiograph, and only one case presented with an osteolytic mass (6). MRI revealed intermediate to hypointensities on T1WI–relative to skeletal muscle–and hyperintensities on T2WI or fluid-sensitive images. Contrast-enhanced MRI examinations were conducted with three cases. These studies revealed peripheral rim enhancement ($n = 1$), heterogeneous enhancement ($n = 1$), and moderate enhancement throughout the lesion and peripheral rim ($n = 1$). Nuclear medicine tests, such as a bone scans or PET scans, showed variable uptake or hypermetabolism.

Our case exhibited CT and MRI findings similar to those of previously reported cases. CT revealed the presence of a sclerotic mass and characteristics of intermediate to hypointensities on T1WI, and heterogeneous hyperintensities on T2WI with a heterogeneous enhancement pattern. However, unlike previously reported cases, our case did not show hypermetabolic features on PET-CT. Such variable findings may be explained by the fact that spinal intraosseous hibernoma may not show FDG uptake during PET-CT examination.

Histopathological examination of the core biopsy revealed infiltration of the tumor cells within the marrow space between the bony trabeculae with preservation of overall intact anatomy, despite slight hypertrophy of the lamellar bony trabeculae within the lesion and mild sclerosis. This pathologic feature correlated with observed imaging features, including a sclerotic lesion with an intervening lucency. In contrast, our findings stand in contrast with reported features of intraosseous lipomas, which are typically well-defined osteolytic intramedullary lesions with thin sclerotic rims.

Kumar et al. (2) reported that the MR findings of intraosseous hibernomas were similar to those of soft-tissue hibernomas. Soft-tissue hibernomas are vascular tumors that contain fibrovascular septa. These features may contribute to heterogenous nature of MR signals within the lesion (7). Song et al. (3) reportedly observed small- to medium-sized vessels within an intraosseous hibernoma. Our case showed serpentine and persistent hypointense lines on MRI, which indicated vascular elements (Fig. 1C).

For the imaging finding of a solitary sclerotic bone lesion, the differential diagnosis should include benign notochordal cell tumor (BNCT), atypical hemangioma with intramedullary sclerosis, or blastic metastasis. BNCTs present as sclerotic lesions on CT and show hypointensities on T1WI. These tumors are usually located at midline and do not show contrast enhancement on MRI. Atypical hemangioma with intramedullary sclerosis and an intraosseous hibernoma have similar conventional MR features: hyperintensities on T2WI and hypointen-

sities on T1WI. Unlike usual hemangiomas, atypical hemangiomas have lower signal intensities on T1WI due to the increased vascular components and intramedullary sclerotic portion. These features contribute to possible signal differences, in comparison with usual hemangiomas. Furthermore, both atypical hemangioma and intraosseous hibernomas have various enhancement patterns. Therefore, as in this case, if both tumors appear as sclerotic lesions, it may be challenging to distinguish between these two lesions using MR features. This is also true in cases of metastases. Usmani et al. (8) reported that atypical hemangiomas might vary in appearance and may present as sclerotic lesions on CT and hypointensities on T1WI, potentially resembling metastatic lesions. Metastatic lesions usually show lower signal intensities than intervertebral discs; however, in many cases, hibernomas show diverse imaging characteristics. This makes it challenging to distinguish hibernomas from metastases. Although PET is useful for differentiating these lesion types, in our case, there was no uptake in the lesion under PET. As with our case, it can be difficult to differentiate between various conditions based only on uptake values. In such situations, biopsy and pathologic diagnosis play a critical role.

In conclusion, hibernomas demonstrate common imaging features, but may also demonstrate considerable heterogeneity. The growing number of radiological investigations of the spine is expected to increase the rate with which spinal intraosseous hibernomas are detected. Radiologists should consider spinal intraosseous hibernoma as a differential diagnosis in patients with sclerotic spinal intraosseous lesions.

Author Contributions

Conceptualization, U.M., L.E., L.J.W.; data curation, U.M., L.E.; investigation, U.M., L.E.; methodology, L.J.W., K.Y., A.J.M., K.H.S.; project administration, L.E.; resources, U.M., L.E.; supervision, L.E.; visualization, U.M., L.K.S.; writing—original draft, U.M., L.E., L.J.W., L.K.S.; and writing—review & editing, L.J.W., L.K.S., K.Y., A.J.M., K.H.S.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

REFERENCES

1. Furlong MA, Fanburg-Smith JC, Miettinen M. The morphologic spectrum of hibernoma: a clinicopathologic study of 170 cases. *Am J Surg Pathol* 2001;25:809-814
2. Kumar R, Deaver MT, Czerniak BA, Madewell JE. Intraosseous hibernoma. *Skeletal Radiol* 2011;40:641-645
3. Song B, Ryu HJ, Lee C, Moon KC. Intraosseous hibernoma: a rare and unique intraosseous lesion. *J Pathol Transl Med* 2017;51:499-504
4. Hafeez I, Shankman S, Michnovicz J, Vigorita VJ. Intraosseous hibernoma: a case report and review of the literature. *Spine (Phila Pa 1976)* 2015;40:E558-561
5. Bonar SF, Watson G, Gragnaniello C, Seex K, Magnussen J, Earwaker J. Intraosseous hibernoma: characterization of five cases and literature review. *Skeletal Radiol* 2014;43:939-946
6. Westacott L, Collins A, Dickenson I. Intraosseous hibernoma in the sacrum of an adult. *Int J Surg Pathol* 2016;24:749-752
7. Peer S, Kühberger R, Dessl A, Judmaier W. MR imaging findings in hibernoma. *Skeletal Radiol* 1997;26:507
8. Usmani S, Marafi F, Rasheed R, Al Kandari F, Ahmed N. Atypical hemangioma mimicking metastasis on 18F-sodium fluoride positron emission tomography-computed tomography and magnetic resonance imaging: gallium-68-prostate-specific membrane antigen positron emission tomography improves the specificity of bone lesions. *Indian J Nucl Med* 2018;33:171-173

9. Jerman A, Snoj Ž, Kuzmanov BG, Limpel Novak AK. Intraosseous hibernoma: case report and tumor characterization. *BJR Case Rep* 2015;1:20150204
10. Ringe KI, Rosenthal H, Länger F, Callies T, Wacker F, Raatschen HJ. Radiofrequency ablation of a rare case of an intraosseous hibernoma causing therapy-refractory pain. *J Vasc Interv Radiol* 2013;24:1754-1756

척추에서 발생한 골 내 동면종: 증례 보고와 문헌 고찰

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동면종은 갈색지방세포로부터 기원한 드문 양성종양이다. 척추에서 발생한 골 내 동면종은 매우 드물고, 근래에 들어서 문헌에 보고되기 시작하여 현재까지 단 12예만이 보고되어 있다. 우리는 우연히 발견된 흉추의 골 내 동면종의 다양한 영상 검사 소견과 병리학적 소견을 보고하고자 한다. 또한 문헌 고찰을 통해 이전에 보고되었던 증례들과 임상 양상 및 영상 소견을 비교하여 분석하였다.

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