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CASE REPORT

What are MRI findings of Spine Benign Metastasizing Leiomyoma? Case report with literature review

Junseok W. Hur · Sunhye Lee · Jang-Bo Lee · Tai-Hyoung Cho · Jung-Yul Park

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Abstract
Introduction Benign Metastasizing Leiomyoma (BML) is a rare disease that results from metastasis of uterine leiomyoma to distant sites with benign pathologic features. Spine BML is very rare so the information of its features and pathophysiology is seldom known.
Materials and Methods We experienced a case of 42-year-old woman who presented with right buttock and leg pain with paresthesia. She had a surgical history of uterine myomectomy. Magnetic resonance imaging (MRI) of the lumbar spine revealed a well-circumscribed mass lesion in the posterior compartment of the L4 vertebral body, with extension into the ventral epidural space and both foramina. The mass showed hypointensity on T1-, T2-weighted images and strong homogeneous enhancement on gadolinium enhanced T1-weighted images. Tumor removal was conducted, and permanent biopsy revealed the mass as leiomyoma. Nine previous spine BML reports, which are known for all, were reviewed along with our case. We collated the clinical information and MRI findings of spine BML to figure out its common denominators.
Results Premenopausal women, previous history of uterine myoma, myomectomy/hysterectomy, and lung BML seemed to be predisposing clinical factors. For the imaging findings, posterior vertebral body invasion with bony destruction, neural foramen invasion, and canal encroachment were shown as common denominators. Especially in MRI findings, low T1 and T2 signal intensities with strong homogeneous enhancement were their common features.
Conclusion We gathered the fragmentary information of the spine BML for the first time, especially the MRI findings. Although spine BML is rare, it surely exists. Accordingly, spine surgeons should be suspicious of spine BML given its typical clinical history and MRI findings.

Keywords Benign metastasizing leiomyoma · Metastatic spinal tumor · Spine · Batson plexus · MRI

Introduction

Benign metastasizing leiomyoma (BML) is a rare disease that results from metastasis of uterine leiomyoma to distant sites with benign pathologic features [1]. BML has been reported in the lung (the most common site), skin, trachea, bladder, esophagus, liver, adrenal gland, skull base, and spine and is usually associated with a history of hysterectomy or myomectomy [1–8]. Among various BMLs based on locations, there is no organized literature of spine BML as an individual disease entity, which makes it difficult to suspect diagnosis of spine BML. We present a case of spine BML and have analogized its clinical and radiologic characteristics with other previous reports.

Case report

A 42-year-old woman presented with right buttock and leg pain with paresthesia. Her symptoms had started 2 years previously and showed aggravation 1 week ago. No

J. W. Hur · J.-B. Lee (✉) · T.-H. Cho · J.-Y. Park
Department of Neurosurgery, Korea University Anam Hospital, College of Medicine Korea University, #126, 5-GA, Anam-Dong, Sungbuk-Ku, Seoul 136-705, Korea
e-mail: jblee42@gmail.com

S. Lee
Department of Radiology, Korea University Anam Hospital, College of Medicine Korea University, Seoul, Korea

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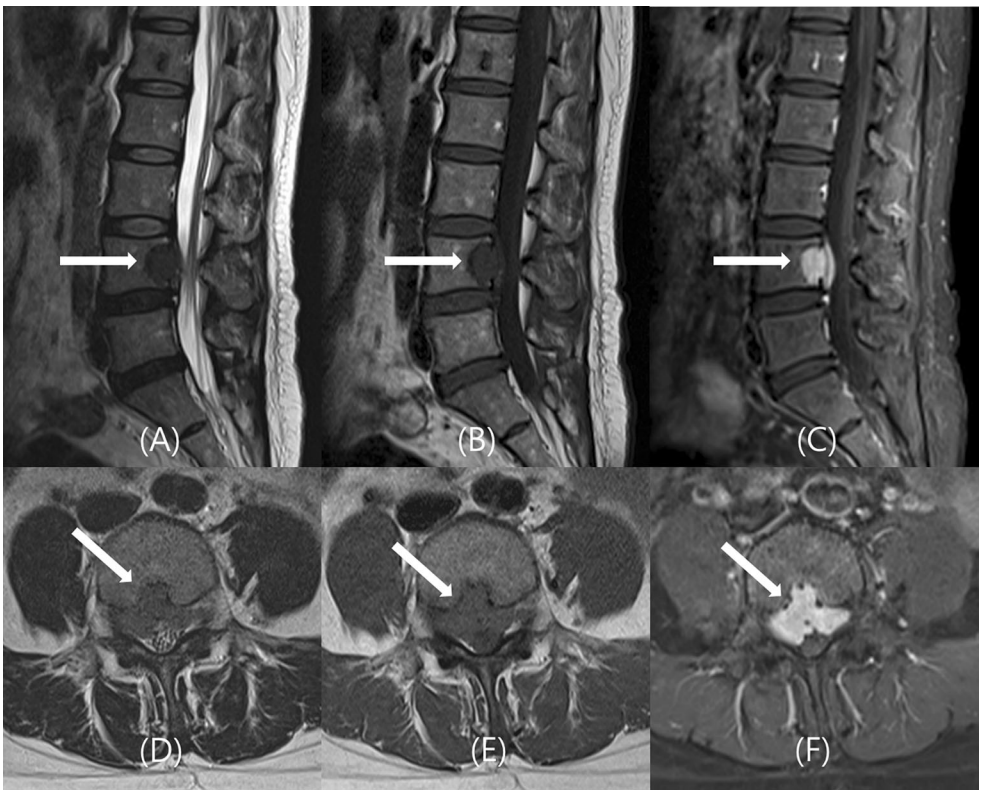

Fig. 1 A well-defined mass lesion was noticed on MRI in the posterior compartment of the L4 vertebral body (*arrows*), with extension into the ventral epidural space. The mass showed hypointensity on both T2- (**a**, **d**) and T1- (**b**, **e**) weighted images. The lesion showed homogeneous strong enhancement on gadolinium enhanced T1-weighted images (**c**, **f**). An osteolytic lesion was observed on the posterior aspect of the L4 vertebral body and expanded into the central spinal canal and both foramina

neurologic deficit including motor impairment was noted. Magnetic resonance imaging (MRI) of the lumbar spine revealed a well-circumscribed mass lesion in the posterior compartment of the L4 vertebral body, with extension into the ventral epidural space on axial images. On both T1- and T2-weighted images, the mass lesion showed hypointensity relative to bone marrow. The lesion showed strong homogeneous enhancement and was gently displacing vessels on gadolinium enhanced T1-weighted images. In addition, an osteolytic lesion was observed on the posterior aspect of the L4 vertebral body with expansion into the central spinal canal and both neural foramina (Fig. 1). She had surgical history of uterine myomectomy 14 years previously. There was no medical history including any malignancy. Nevertheless, to rule out any possibility of a primary or metastatic tumor, a whole body Positron Emission Tomography (PET) scan was performed as a screening test. However, there were no hot uptakes in any organ including the spinal lesion (Fig. 2). We performed surgery for diagnosis and treatment. Under general anesthesia, right L4 and 5 paravertebral muscles were dissected, and facetectomy was held. A whitish and adhesive mass was found ventral to the thecal sac, expanding into both neural foramina and posterior vertebral body. Frozen biopsy was reported as benign spindle cell tumor. Subtotal tumor removal and pedicle screw fixation with lateral fusion were performed on L4/5. Post-operatively, her symptoms were improved. Permanent pathology result was

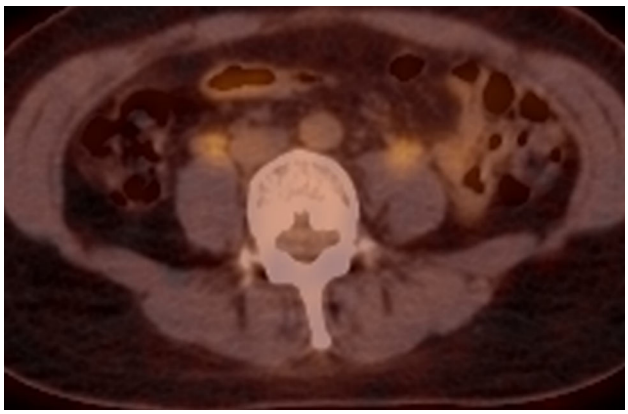

Fig. 2 PET scan of equivalent level of L4, where the mass was observed on MRI. There was no evidence of hypermetabolism, which reflects a benign feature of the mass

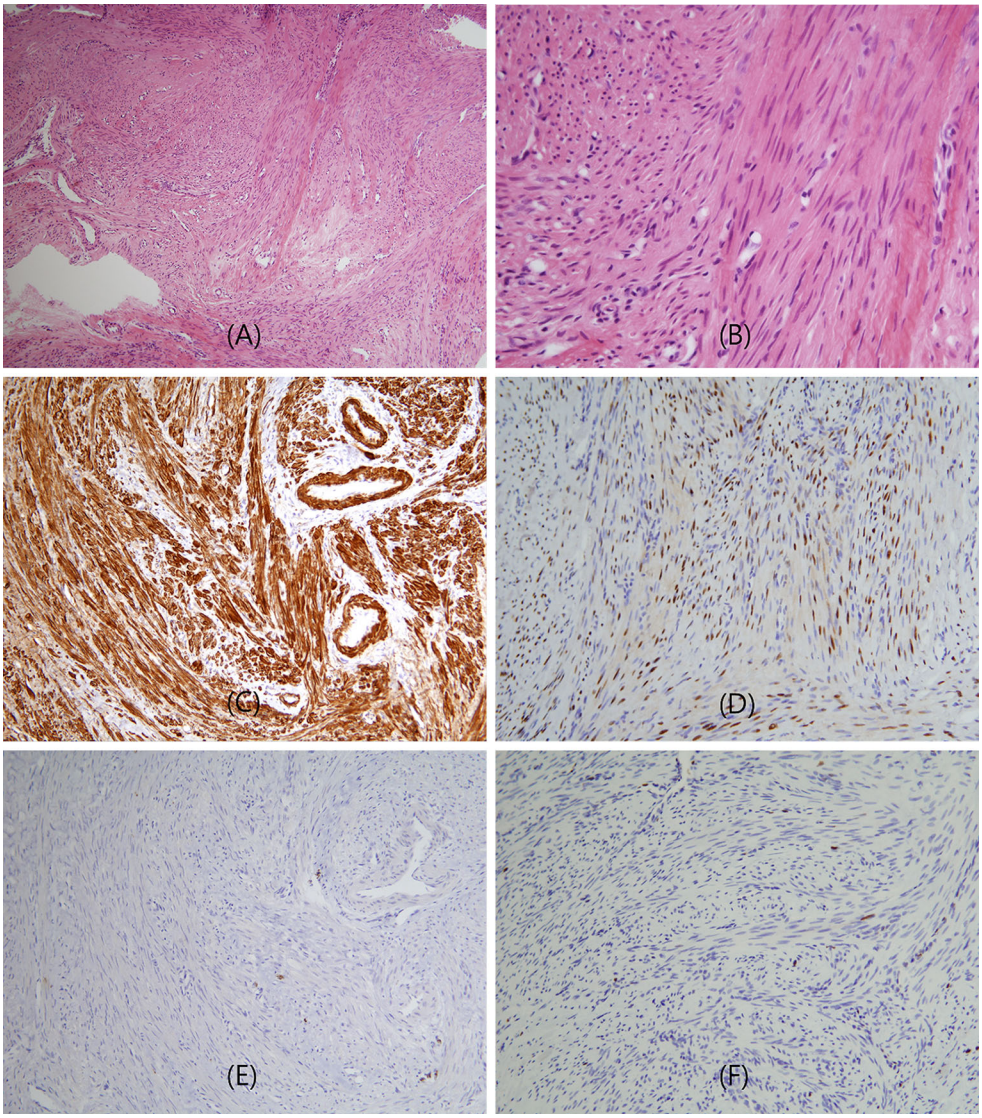

Fig. 3 Pathology findings of the specimen. Diffuse infiltration of uniform spindle cells with fascicular arrangement was observed at ×100 magnification (**a**). The tumor cells showed eosinophilic cytoplasm without nuclear pleomorphism, and no mitoses were found at ×400 magnification (**b**). In immunohistochemical stain, tumor cells were positive for smooth muscle actin (**c**), desmin (not shown), progesterone receptor (**d**), and estrogen receptor (not shown), but negative for neurofilament (**e**), S100 (not shown), and epithelial membrane antigen (not shown). The Ki-67 index was less than 1 % (**f**)

perfectly compatible for diagnosis of benign metastasizing leiomyoma (BML) (Fig. 3). After pathologic confirmation, chest computed tomography (CT) and gynecologic sonography were performed for further evaluation of BML. No definite abnormal findings were evident on chest CT, while gynecologic sonography detected three 1–2 cm sized myomas. Retrospectively, we recognized there was a history of recurred myoma, which had been diagnosed 2 years previously. However, as the sizes of the uterine myomas were small and showed no definite evidence of growing compared to previous medical records, the gynecologist planned for regular follow-up only.

Discussion

Spine BML is difficult to suspect before pathologic confirmation because of its rarity. The clinical, pathophysiologic, and imaging information of spine BML is not yet determined thoroughly. We searched the articles that are related to the spine BML, using MEDLINE/PubMed as a searching engine. Only 9 case reports were revealed; therefore, this article would be the 10th to report spine BML, to the best of our knowledge. We reviewed 9 previous reports and analyzed with our case (summarized on Table 1) [2, 4–6, 8–12]. Of these cases, three occurred in

Table 1 Summary of spine BML cases

	References	Year	Sex	Age	Level	MRI (T1)	MRI (T2)	MRI (enhance)	Vertebra body involvement	Bony destruction	Canal encroachment	Foramen or pedicle involvement	Lung BML	Uterine leiomyoma	Myomectomy or hysterectomy	Time interval†
1	Rogers and Thomas [10]	1959	F	37	T2-3, S	N/A	N/A	N/A	Posterior	O	O	N/A	N/A	O	Hysterectomy	10 years
2	Gatti et al. [9]	1983	F	56	C2, T6, T10	N/A	N/A	N/A	Antero-posterior	O	O	O	O	O	Myomectomy Hysterectomy	20 years 8 years
3	Hekster et al. [11]	1994	F	43	C6	N/A	N/A	N/A	Antero-posterior	O	O	O	N/A	O	Myomectomy	20 years
4	Pimentel et al. [12]	2001	F	30	L2	N/A	N/A	HoE	Posterior	O	O	O	N/A	O	Hysterectomy§	–
5	Alessi et al. [2]	2002	F	43	S2	Low	Low	N/A	Posterior	O	O	N/A	O	O	Myomectomy Hysterectomy	16 years 4 years
6	Joseph et al. [5]	2003	F	38	C4-6	N/A	Low	HoE	Posterior	O	O	O	O*	O*	None	–
7	Kang et al. [6]	2011	F	30	T6	N/A	N/A	HoE	Posterior	O	O	N/A	O	O	Myomectomy	3 years
8	Jayakody et al. [4]	2011	F	44	T5, 10	N/A	Low	N/A	Posterior	O	O	O	O	O	Hysterectomy	4 years
9	Wang et al. [8]	2012	F	47	L2, 4	low	Low	HoE	Posterior	O	O	O	x	O	Hysterectomy	3 years
10	Hur et al.	2014	F	42	L4	Low	Low	HoE	Posterior	O	O	O	x	O	Myomectomy	14 years

N/A data not available, *HoE* homogeneous enhancement

* Postmortem diagnosis

§ Hysterectomy was performed after spine BML diagnosis

† The time interval from myomectomy/hysterectomy to discovery the spine BML

the cervical spine, four in the thoracic spine, three in the lumbar spine, and two in sacrum, which appear to represent an equal distribution. Five patients were diagnosed with lung BML, two patients showed no evidence of lung BML, and three reports contained no data on lung involvement. As previous studies for BMLs on other locations [13, 14], we found premenopausal women (9 among 10 patients were fourth or fifth decades) and myomectomy/hysterectomy history (8 among 10 patients) as predisposing factors (including preexisting lung BML) for spine BML. We investigated the time interval from myomectomy/hysterectomy to discover the spine BML, and it varied from 3 to 20 years. This was similar to lung BML (4–12 years) [1].

Despite some debates, metastatic mechanism for lung BML is explained with pelvic venous plexus-IVC route [1, 14, 15]. The abundant blood (containing some myoma cells) moves from pelvic venous plexus to IVC, and to lung via heart, and this could explain the relative majority of lung BML compare to other sites. Nevertheless, the pelvic venous plexus also communicates with the Batson plexus. Furthermore, the IVC gives branches directly to the lumbar region via lumbar veins, and this could provide a route for lumbar BML [16–19]. For cervical and thoracic spine BML, referring to the literature of metastatic spinal tumor, we can hypothesize some possible routes. First, pulmonary vein drains into the left side of heart, and the blood spreads in a generalized pattern throughout the skeleton [16, 17]. Second, lung BML seeds directly via the segmental arteries of the cervical and thoracic spine [18].

Based on MRI information, we initially suspected the mass for differential diagnosis, to be hemangioma, hemangiopericytoma, lymphoma, benign bone origin tumor, bony invasion of neuroblastic tumor, or a metastatic tumor, but still none perfectly matched the MRI findings [20]. After receiving pathologic confirmation, we reviewed the literature to determine how BML typically appears in spine images, but none of the literature suggested the common pattern. There are only some reports characterizing lung BML features in chest CT images, presumably because of its relative abundance. Interestingly, there were several common denominators in spine BML appearance. In entire cases, all the lesions involved 'posterior' element of vertebral body. Two cases showed further mass extension toward the anterior portion of vertebral body. Bony destruction and spinal canal encroachment were noted in all cases as well. We could also verify the invasion of the neural foramen or pedicle in all legible cases (seven cases had available descriptions). Reviewing all the cases that had mentioned MRI findings, there were significant common denominators. The masses were well circumscribed and showed low signal intensity on both T1 and T2-weighted images. In case of enhance studies, strong homogeneous enhancement on T1-weighted post-gadolinium images were noted. In particular, this information regarding MRI findings represents the first attempt to organize the characteristics of spine BML.

Conclusion

In the present study, we collated the clinical information and image findings of spine BML. Spine surgeons should be suspicious of spine BML given its typical clinical history and MRI findings (1) well-circumscribed mass located on posterior vertebral body with bony destruction, neural foramen invasion, and canal encroachment, (2) low T1 and T2 signal intensity with strong homogeneous enhancement. Further clinical/imaging data collection and pathophysiologic studies should be conducted.

Conflict of interest None.

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