Lumbar Spinal Extradural Angiolipomas —Two Case Reports—

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Abstract

Spinal extradural angiolipomas are benign tumors mostly localized in the thoracic region. A 50-year-old woman and a 36-year-old man presented with rare lumbar spinal angiolipoma manifesting as low back pain but without neurological signs. Magnetic resonance imaging showed lumbar extradural tumors at the L4-5 and L1-2 levels, respectively. Each patient underwent complete surgical resection of the epidural tumors. Histological examination revealed characteristics of angiolipomas in both tumors. The symptoms of both patients improved postoperatively and no recurrence of the tumors was found 1 year after surgery.

Key words: extradural spinal tumor, spinal angiolipoma, spinal cord

Introduction

Spinal angiolipomas are benign lesions consisting of mature adipocytes and abnormal blood vessels that vary in size from capillary to sinusoid or venular to arterial.²⁶⁾ Spinal angiolipomas are rare, accounting for only 0.14-1.2% of all spinal axis tumors and 2-3% of extradural spinal tumors. 22,26) Spinal angiolipomas are subdivided into two types: noninfiltrating and infiltrating.¹⁷⁾ Most cases show no tendency for involvement of the surrounding tissue, but some cases demonstrate infiltration into the bony compartment of the vertebral column. 13-15,20, ^{22,23,28)} The tumors can occur anywhere from C-6 to L-4, but mainly in the posterior midthoracic region, whereas pure lumbar localization is extremely rare. 15,16,29) Several reviews 15,22,29) and approximately 100 case reports³⁰⁾ have identified only nine cases in the lumbar region. Here we report two cases of lumbar angiolipomas, one of which was infiltrating.

Case Reports

Case 1: A 50-year-old woman with a 2-year history of lumbosciatalgia was admitted for a worsening of the symptoms in the previous month. On admission, neurological examination and radiography of the lumbar spine demonstrated no abnormalities.

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Magnetic resonance (MR) imaging showed an extradural mass in the anterolateral L4–5 region, displacing the dural sac and extending into the right foramen, and partially infiltrating the posterior wall of the L-4 vertebral body. The mass was isointense on T₁-weighted images and hyperintense on T₂-weighted images, with hyperintense homogeneous enhancement by contrast medium (Fig. 1A, B).

Right L4-5 hemilaminectomy was performed. The epidural space dural sac was compressed by a purple, highly vascularized lesion that extended into the foramen and eroded the anterior aspect of the L-4 body. The lesion was dissected and completely removed. Histological examination revealed angiolipoma consisting of well-differentiated adipose tissue with admixed vascular structure (Fig. 1D).

The patient recovered completely and lumbosacral MR imaging confirmed total resection and no evidence of recurrence 1 year after surgery (Fig. 1C).

Case 2: A 36-year-old man presented with an 8-month history of low back pain radiating from the anterior aspect of the right thigh and increasing with walking and standing. Neurological examination and radiography of the lumbar spine found no abnormalities. MR imaging revealed an epidural mass in the right L-2 neural foramina. The lesion was isointense on T₁-weighted images and hyperintense on T₂-weighted images, with homogeneous enhancement after gadolinium administration (Fig. 2A, B).

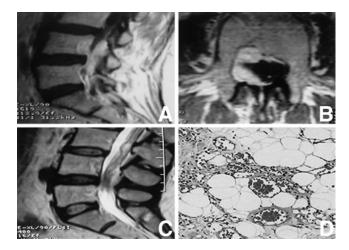


Fig. 1 Case 1. A: Sagittal T₁-weighted magnetic resonance (MR) image with contrast medium demonstrating an L4-5 anterolateral extradural lesion with homogeneous signal intensity. B: Axial T₁-weighted MR image showing partial infiltration of the lesion into the posterior wall of L-4. C: Postoperative sagittal T₁-weighted MR image with contrast medium showing no evidence of tumor at 12 months after surgery. D: Photomicrograph of the tumor showing mature adipose tissue and irregular vascular structures. HE stain, original magnification × 200.

Right L1-2 laminectomy was performed. The foraminal space was filled with a red, soft, fatty, and hypervascular tumor, which had compressed the L-2 root. The tumor was totally removed. Histological examination revealed angiolipoma (Fig. 2D).

The postoperative course was uneventful. MR imaging confirmed total resection and no evidence of recurrence 12 months after the surgical procedure (Fig. 2C).

Discussion

Spinal angiolipomas are rare lesions first reported in 1890.⁵⁾ This tumor was defined as a benign neoplasm of the adipose tissue with abnormal blood vessels in 1945.⁹⁾ The majority of these tumors are located in the subcutaneous vessels, muscle, bone, and kidney.¹¹⁾ The etiology and pathogenesis of angiolipomas are unknown but several theories have been advanced. For example, following some undefined stimulus, this tumor may arise from pluripotential mesenchymal stem cells by divergent differentiation along both adipose and angioid lineages,⁹⁾ or be a congenital malformation⁶⁾ or a benign hamartoma.³¹⁾ Spinal angiolipomas may

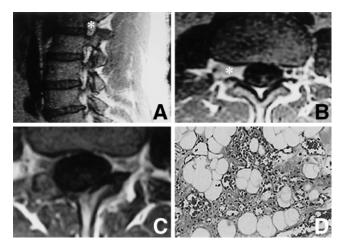


Fig. 2 Case 2. A, B: Sagittal (A) and axial (B) T₁-weighted magnetic resonance (MR) images with contrast medium showing an epidural lesion in the right L-2 neural foramina with homogeneous signal intensity (asterisk). C: Postoperative axial T₁-weighted MR image with gadolinium showing complete removal of the lesion. D: Photomicrograph showing angiolipoma with mature adipose tissue and vascular structures. HE stain, original magnification × 200.

arise from pluripotential stem cells with secretory activity.³⁾

Only nine cases of angiolipomas involving the pure lumbar spine have been described (Table 1). 10,18,20,21,23,25) The thoracic predominance of spinal angiolipomas may depend on the regional variation in blood supply in the spine, and specifically that the mid-thoracic spine is the least well-perfused segment. 15)

Most patients presented with a slow-growing mass causing compression of the spinal cord. Lower extremity numbness, back pain, and leg weakness are frequent initial complaints.^{22,29)} However, as in our cases, a few cases of angiolipoma situated in the lumbar extradural space have manifested as sciatica.8,20,25) The more rapid onset of symptoms in some cases may depend on vascular factors, such as anomalous vessels, intralesional thrombosis, hemorrhage, or steal phenomena.^{2,4,15,27)} Symptom onset and exacerbation have also occurred during pregnancy, possibly in conjunction with an increase in tumor volume resulting in impaired spinal venous drainage, or hormonal changes resulting in an increase in extravascular fluid volume. 1,7,22) Obesity may also lead to symptoms induced by greater fatty component.²²⁾ Spinal angiolipomas may be misinterpreted as a demyelinating disease because of the

Author (Year)	Age/ Sex	Spinal level	Localization of lesion	Vertebral involvement	Treatment	Results
Gonzalez-Crussi et al. (1966) ¹⁰⁾	20/F	L1-4	epidural anterolateral	body and lamina of L-3	radiotherapy	recovery
Lo Re and Michelacci (1969) ¹⁸⁾	16/M	cauda	epidural posterior	no	surgery	recovery
	35/F	lumbar	epidural posterior	no	surgery	recovery
Pagni and Canavero (1992) ²⁰⁾	56/F	L-3	epidural anterolateral	posterior wall and lt pedicle of L-3	surgery	recovery
	59/F	L4-5	epidural anterior	no	surgery	recovery
Provenzale and McLendon (1995) ²³⁾	38/F	lumbar	epidural posterior	no	unknown	unknown
Pinto-Rafael et al. (2002) ²¹⁾	85/M	L1-2	epidural posterior	no	surgery	recovery
Rocchi et al. (2004) ²⁵⁾	60/M	L3-4	epidural anterior	no	surgery	recovery
	54/F	L-3	epidural anterior	no	surgery	recovery
Present Case 1	50/F	L4-5	epidural anterolateral	posterior wall of L-4	surgery	recovery
Present Case 2	36/M	L-2	epidural intraforamen	no	surgery	recovery

Table 1 Reported cases of pure lumbar spinal extradural angiolipomas

fluctuating clinical course.²⁹⁾

Plain vertebral radiography demonstrates no abnormalities in the majority of the cases. Trabeculation of the affected vertebral body and erosion of the pedicle may be identified in tumors infiltrating bone.^{23,24} Computed tomography usually demonstrates a hypodense lesion consistent with fat density and provides information on the degree of bony involvement.¹⁹⁾ These tumors show little or no contrast enhancement, allowing a reliable way to distinguish angiolipoma from hemangioma.²²⁾ MR imaging is the imaging modality of choice for detecting angiolipomas. The extradural components of angiolipomas appear as isointense or hyperintense on T₁-weighted images and usually as hyperintense on T2-weighted images. Spinal hemangiomas also present as mixed intensity lesions on MR imaging, although the spinal intensity is greater on T₂-weighted images. Suppression of high signal intensity on fat-saturated T₁-weighted images may be useful for distinguishing between angiolipomas and subacute hemorrhage or melanomas. Most spinal angiolipomas are enhanced by contrast medium which better defines the borders of the tumor.12)

Spinal epidural angiolipomas are rare benign tumors that are best managed with surgical resection, even if the infiltrating type, and complete removal is typically possible. Nevertheless, good outcomes occur even with subtotal resection which provides substantial symptomatic relief.²⁹⁾ Only one recurrence of angiolipoma has ever been reported, and successful reoperation was performed 12 years

after the first surgery.⁴⁾ Malignant transformation of these tumors has not been noted. Radiotherapy is not suggested for these tumors.

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