

Case Report

Lumbar spinal angioliipomas: report of two cases and review of the literature

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Study design: Case report and review of the literature.

Objectives: To describe two patients with angioliipoma in the ventral aspect of the lumbar epidural space, to discuss the clinical, radiologic, and surgical features of these lesions, and to review previously reported cases.

Setting: Rome, Italy.

Methods: Two cases, a 60-year-old man and a 54-year-old woman presented with lumbar–sciatic pain but with no abnormal neurological signs. Investigation (CT and MRI) demonstrated lumbar tumours.

Results: Laminectomy and excision of the tumors were performed, and symptoms improved immediately.

Conclusions: Magnetic resonance imaging with suppression fat sequences allows the recognition of these lesions. The prognosis after surgical removal of spinal angioliipoma is favorable.

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Keywords: angioliipoma; epidural; epidural spinal tumor; magnetic resonance imaging

Introduction

Spinal angioliipomas are rare benign tumors. They contain mature lipomatous elements and proliferating vessels. These tumors have been suggested as an intermediate entity of a spectrum ranging from angiomas to lipomas. Angioliipomas account for only 0.14–1.2% of all tumors of the spinal axis and 3% of extradural spinal tumors^{1–4}. A review of the international literature since 1892 to 2002 revealed 83 cases of spinal extradural angioliipomas including our two cases.^{4–9}

Most spinal angioliipomas are found at thoracic level and arise in the posterior extradural space.^{4–9} Pure lumbar localization is extremely rare. Only six cases^{6–12} of lumbar angioliipomas are reported in the literature accessible to us (Table 1). Several excellent review articles^{4–9} are already available. The purpose of this report is to report two exceptionally rare cases of lumbar angioliipomas localized in the ventral aspect of the epidural space and to review the pertinent literature.

Case report 1

A 60-year-old man was admitted to our institution with a 2-year history of lumbosciatalgia. Neurological examination was negative. CT showed a hyperdense anterior epidural mass located at level L3–L4. The mass exhibited homogeneous contrast enhancement. MRI showed an area with signal intensity similar to that of the subcutaneous adipose tissue (Figure 1).

After performing a laminectomy, a brown, soft, well-vascularized tumor was found in the epidural space. The vertebral body presented an erosion of 1 × 0.5 cm. The lesion was dissected from the dura without difficulties and totally removed. Histology was that of angioliipoma (Figure 2). Postoperative period was uneventful and 2 years after operation the patient is asymptomatic.

Case report 2

A 54-year-old woman presented with a 12-month history of lumbosciatalgia. On admission, neurological examination was negative. MRI showed an L3 anterior epidural lesion with both lipomatous and vascular components and homogeneous contrast enhancement (Figure 3). The lesion eroded into the posterior wall of

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Table 1 Pure lumbar angioliipomas reported in the literature

References	Age/sex	Duration of symptoms (years)	Level	Localization	Variety	Treatment	Results
Golzales-Crussi 10	20/F	3	L1–L4	E.A.	Infiltrating	S + RT	Recovered
Lo Re and Michelacci 11	16/M	Congenital	Cauda	E.P.	Noninfiltrating	S	Recovered
	35/F	Unknown	'Lumbar'	E.P.	Noninfiltrating	S	Recovered
Pagni and Canavero 6	56/F	12	L3	E.A.	Infiltrating	S	Recovered
	59/F	27	L4–L5	E.A.	Noninfiltrating	S	Recovered
Provencale and McLendon 12	38/F	3	'Lumbar'	E.P.	Noninfiltrating	Unknown	Unknown
Present case 1	60/M	2	L3–L4	E.A.	Noninfiltrating	S	Recovered
Present case 2	54/F	1	L3	E.A.	Noninfiltrating	S	Recovered

E.A. = epidural anterior, E.P. = epidural posterior, S = surgery, RT = radiotherapy

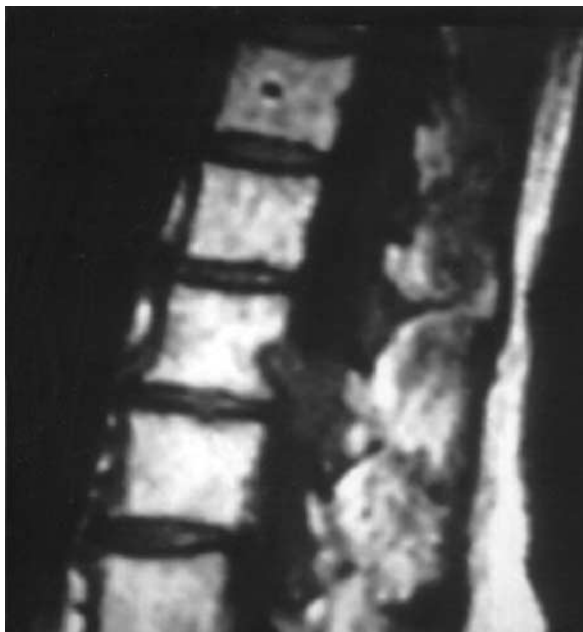


Figure 1 MRI showed an area with signal intensity similar to that of the subcutaneous adipose tissue

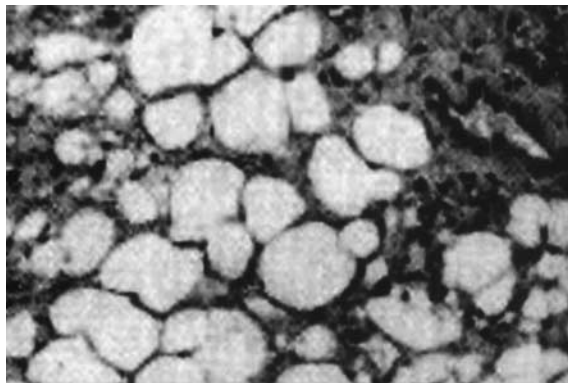


Figure 2 Tumor section showing admixture of mature fat cells and bands of small vessels

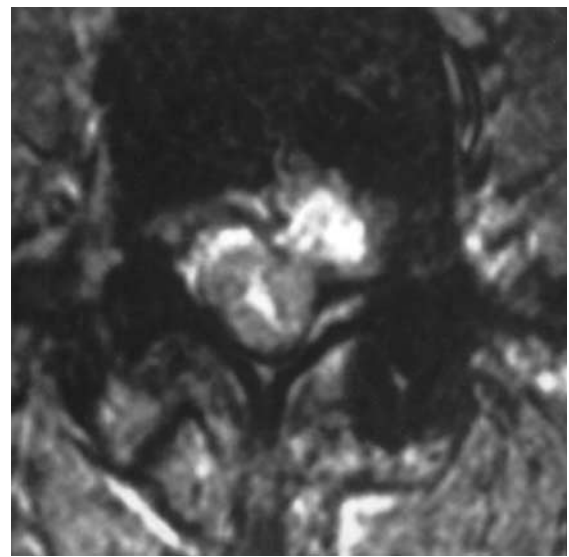


Figure 3 MRI showed a L3 anterior epidural lesion with both lipomatous and vascular components and homogeneous contrast enhancement

the vertebral body and left pedicle. A bilateral laminectomy was performed and a mass of yellow–brown tissue was found. The lesion was located both into bone, with an erosion of 1×0.8 cm, and in the epidural space. The histopathological study revealed the tumor to be an angioliipoma (Figure 4). The postoperative course was uneventful. The patient is currently free of her previous symptoms.

Discussion

The first case of spinal angioliipoma was described by Berenbruch.¹³ Spinal angioliipomas were initially considered a hypervascular variant of spinal lipomas. In 1961, Howard and Helwig¹⁴ established angioliipoma as a clinico-pathological entity. These authors reported that the majority of these tumors are located in subcutaneous vessel, muscle, bone, and kidney.

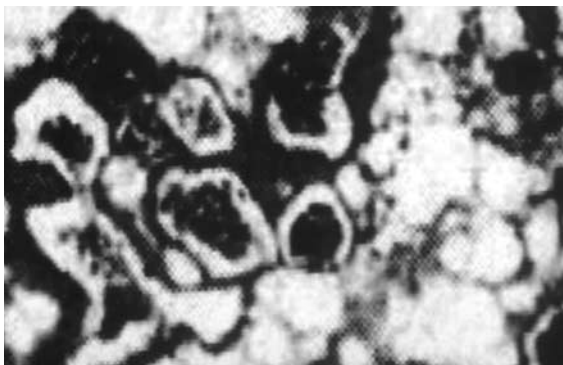


Figure 4 Features of a typical angioliopoma are again shown

Spinal epidural angioliopomas are quite rare and lumbar angioliopomas are extremely rare representing only 9.6% of all spinal extradural angioliopomas. These tumors occur in middle-aged women and preferentially affect the thoracic spine.⁹

The histopathogenesis of angioliopomas is unknown. They probably arise from abnormal primitive pluripotential mesenchymal cells that can differentiate into lipomatous, angiomatous, or mixed tissue.¹⁵ Some authors suggested angioliopomas to be true hamartomas.^{15–17}

The majority of reported spinal angioliopomas originate in the dorsal aspect of the thoracic segment and show no tendency to infiltrate the surrounding bone. According to Lin and Lin,¹⁸ angioliopomas are subdivided into two types: infiltrating and noninfiltrating. The infiltrating type is unencapsulated and contains areas in which the vascular constituent predominates. Rarely, spinal epidural angioliopomas show infiltrative behavior.^{7,8,10,16,19–24} Trabulo *et al*⁵ highlight that it is not clear if the infiltrative angioliopomas originate in the epidural space and infiltrate the bone, or if they originate in the bone and spread to the epidural space or if they arise in both compartments simultaneously. Infiltration of the vertebral body is often associated with anterior localization.⁵ In our cases of lumbar angioliopomas, the tumors were in the ventral epidural space but did not show any signs of bone infiltration. The only bone alteration observed in our cases was an erosion that contained the angioliopomas. In most cases, the time between onset of clinical symptoms and diagnosis is 1 year or less.²⁵ Patients most commonly had long-standing pain and then developed progressive neurologic symptoms secondary to spinal cord compression. Similarly to other vascular lesions, onset or deterioration during pregnancy may occur.^{1,4,26} Rarely, angioliopomas may cause sudden deterioration by thrombosis, hemorrhage, or steal phenomena.^{4,6,16,27} The extremely unusual pure lumbar anterior localization of the lesions in our patients explains the atypical clinical presentation. In fact, both patients complained of lumbosciatic pain.

MRI is the imaging modality of choice in detecting angioliopomas. It must be emphasized that the true

incidence of spinal angioliopomas could be greater than generally believed and MRI will likely increase the opportunities for detecting these lesions. However, neuroradiological diagnosis may be difficult, as the MRI findings of angioliopomas are easily missed. In our cases, preoperative diagnosis was suspected on the basis of simple MRI sequences and was confirmed by histologic examination. Angioliopomas appears as a hyperintense lesions on T1-weighted images.

Gadolinium enhancement is due to the vascularity of these tumors. This phenomenon allows differential diagnosis between extradural lipomatosis, which does not enhance, and spinal angioliopoma. We confirm that gadolinium infusion with fat saturation sequences are useful in the study of these lesions. This occurs because the gadolinium enhancement might be undetected being angioliopomas already hyperintense on T1 sequences. Moreover, fat suppression sequences may enhance areas of abnormal signal intensity within the fatty tumors.²⁸

Spinal epidural angioliopomas are benign lesions and result in a good postoperative outcome, especially if affecting the lumbar level (Table 1). Surgery is the treatment of choice and in most cases of spinal angioliopomas, even if infiltrating, complete surgical removal is possible.^{6,9–12} Turgot,⁹ in an excellent review of the literature, concluded that even if infiltrating angioliopomas can be only partially resected, subtotal resection provides substantial symptomatic relief, because these lesions are slow growing and do not undergo malignant transformation. Radiotherapy has been given only in three cases reported in the literature,^{7,9,10,29} but there is no indication to give it for these benign lesions.

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