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Case Report

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

G Rocchi[1], E Caroli*[,1], A Frati[2], M Cimatti[1] and M Savlati[2]

[1]*Department of Neurological Sciences, Neurosurgery, University of Rome 'La Sapienza', Italy;*
[2]*Department of Neurosurgery-INM Neuromed IRCCS, Pozzilli (Is), Italy*

Study design: Case report and review of the literature.
Objectives: To describe two patients with angiolipoma 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 angiolipoma is favorable.
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Keywords: angiolipoma; epidural; epidural spinal tumor; magnetic resonance imaging

Introduction

Spinal angiolipomas 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. Angiolipomas 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 angiolipomas including our two cases.[4–9]

Most spinal angiolipomas 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 angiolipomas 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 angiolipomas localized in the ventral aspect of the epidural space and to review the pertinent literature.

*Correspondence: E Caroli, Via Meropia, 85, 00147 Roma, Italy

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 angiolipoma (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

Table 1 Pure lumbar angiolipomas 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

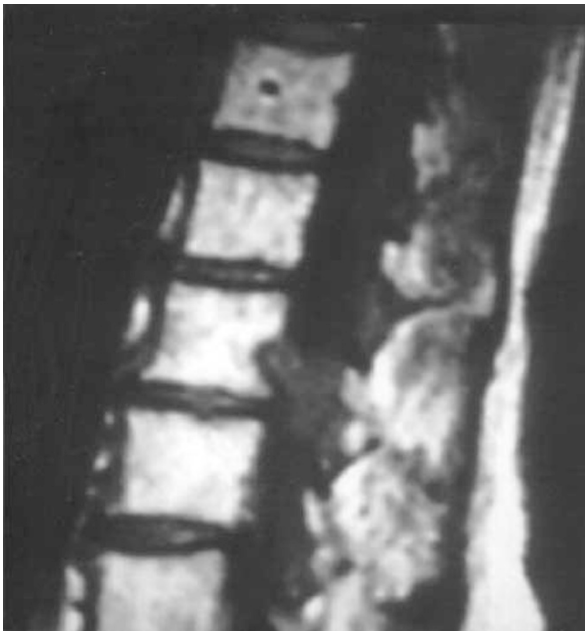

Figure 1 MRI showed an area with signal intensity similar to that of the subcutaneous adipose tissue

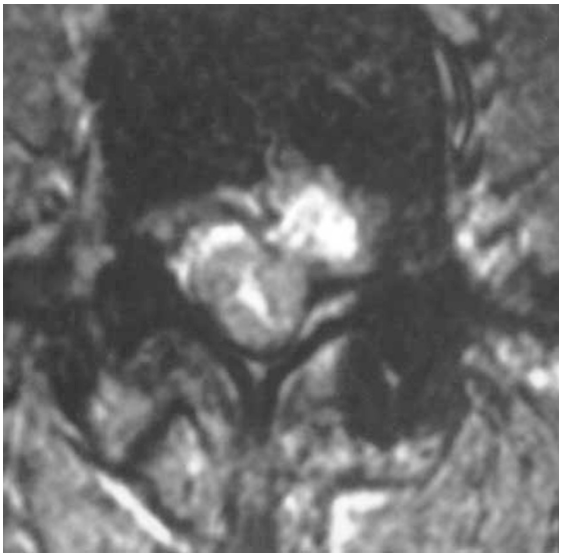

Figure 3 MRI showed a L3 anterior epidural lesion with both lipomatous and vascular components and homogeneous contrast enhancement

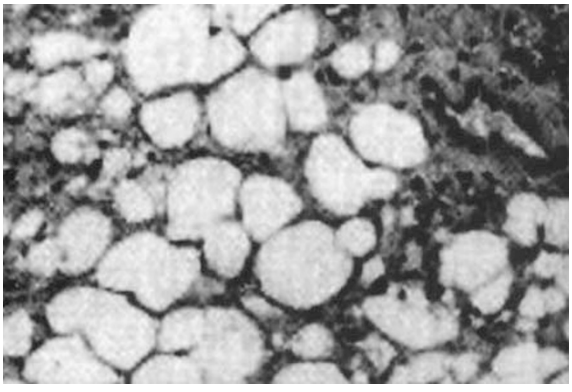

Figure 2 Tumor section showing admixture of mature fat cells and bands of small vessels

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 angiolipoma (Figure 4). The postoperative course was uneventful. The patient is currently free of her previous symptoms.

Discussion

The first case of spinal angiolipoma was described by Berenbruch.[13] Spinal angiolipomas were initially considered a hypervascular variant of spinal lipomas. In 1961, Howard and Helwig[14] established angiolipoma as a clinico-pathological entity. These authors reported that the majority of these tumors are located in subcutaneous vessel, muscle, bone, and kidney.

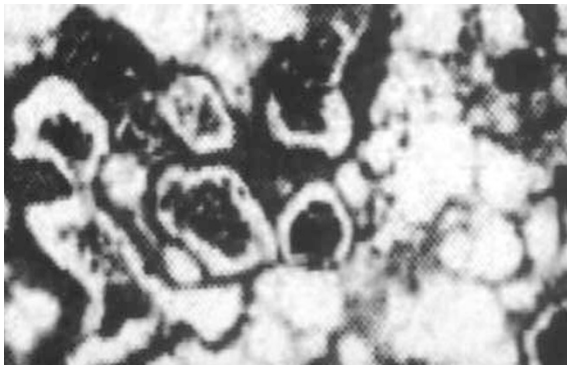

Figure 4 Features of a typical angiolipoma are again shown

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

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

The majority of reported spinal angiolipomas originate in the dorsal aspect of the thoracic segment and show no tendency to infiltrate the surrounding bone. According to Lin and Lin,[18] angiolipomas 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 angiolipomas show infiltrative behavior.[7,8,10,16,19–24] Trabulo *et al*[5] highlight that it is not clear if the infiltrative angiolipomas 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 angiolipomas, 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 angiolipomas. In most cases, the time between onset of clinical symptoms and diagnosis is 1 year or less.[25] Patients most commonly had longstanding 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, angiolipomas 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 lumbosciatalgic pain.

MRI is the imaging modality of choice in detecting angiolipomas. It must be emphasized that the true incidence of spinal angiolipomas 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 angiolipomas are easily missed. In our cases, preoperative diagnosis was suspected on the basis of simple MRI sequences and was confirmed by histologic examination. Angiolipomas 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 angiolipoma. 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 angiolipomas already hyperintense on T1 sequences. Moreover, fat suppression sequences may enhance areas of abnormal signal intensity within the fatty tumors.[28]

Spinal epidural angiolipomas 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 angiolipomas, 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 angiolipomas 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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