

Thoracic epidural angiolipoma: A case report and review of the literature

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Author contributions: Meng J and Du Y contributed equally to this work; Meng J, Du Y, Yang HF and Hu FB collected information about the patient; Meng J, Du Y, Yang HF and Zee CS designed the research; Meng J, Du Y, Yang HF, Hu FB, Huang YY and Li B collected the data; Meng J, Du Y, Yang HF and Hu FB analyzed the data; Meng J and Du Y wrote the paper.

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Received: November 6, 2012 Revised: December 18, 2012

Accepted: January 14, 2013

Published online: April 28, 2013

surgical specimen showed a typical angiolipoma. We review the previously documented cases of spinal extradural angiolipomas performed with MRI.

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Key words: Angiolipoma; Spinal epidural tumor; Spinal cord compression; Histopathology

Meng J, Du Y, Yang HF, Hu FB, Huang YY, Li B, Zee CS. Thoracic epidural angiolipoma: A case report and review of the literature. *World J Radiol* 2013; 5(4): 187-192 Available from: URL: <http://www.wjgnet.com/1949-8470/full/v5/i4/187.htm> DOI: <http://dx.doi.org/10.4329/wjr.v5.i4.187>

Abstract

Angiolipoma of the spine is a benign neoplasm consisting of both mature fatty tissue and abnormal vascular elements, and usually presents with a slow progressive clinical course. Our patient presented with bilateral lower extremity weakness and chest-back numbness. Physical examination revealed adipose elements superficial hypesthesia below the T5 level and analgesia below the T6 level. Magnetic resonance imaging (MRI) scan showed an avidly and heterogeneously enhancing mass which was located in the posterior epidural space. Compression of the thoracic cord by the fusiform mass was seen between T3-T4. During the operation, a flesh pink vascular mass (4.7 cm × 1.0 cm × 1.0 cm) with obscure margin and strong but pliable texture was found in the posterior epidural space extending from T3 to T4. There was no infiltration of the dura or the adjacent bony spine. Histopathological study of the

INTRODUCTION

Angiolipoma of the spine is a benign neoplasm consisting of mature fatty tissue and abnormal vascular elements, predominantly in middle-aged, female patients and situated mainly in the mid-thoracic region. There are only 142 cases with spinal extradural angiolipoma reported to date^[1]. They account for about 0.14%-1.2% of all spinal axis tumors and 2%-3% of spinal epidural tumors^[2]. We report another case of spinal angiolipoma in an elderly patient which showed a typical appearance on magnetic resonance imaging (MRI). The pathology, clinical features, diagnostic evaluation, and treatment of spinal angiolipoma were reviewed.

CASE REPORT

A 63-year-old man presented with a 1.0-year history of bilateral lower extremities numbness and a 6-mo history of difficulty in urination. Concurrently, he noticed bilateral lower extremity weakness and chest-back numbness one month prior to admission. Physical examination revealed a superficial hypesthesia below the T5 level and



Figure 1 Magnetic resonance imaging scan showed a fusiform posterior epidural mass compressing the thoracic cord over two vertebral body segments between T3-T4. A: Sagittal T1 weighted magnetic resonance imaging (MRI) shows a posterior epidural mass with inhomogeneous isointensity constricting the spinal cord (arrow); B: Sagittal T2 weighted MRI shows a fusiform mass with slightly inhomogeneous hyperintensity (arrow); C: Sagittal fat-saturated T2-weighted image shows a hyperintense tumor (arrow); D: Post-contrast sagittal T1-weighted MRI with fat-saturation technique shows an inhomogeneously enhancing mass (arrow); E: Post-contrast axial T1 weighted MRI show crack like low signal between spinal cord and the lesion, with compression and displacement of the spinal cord (arrow).

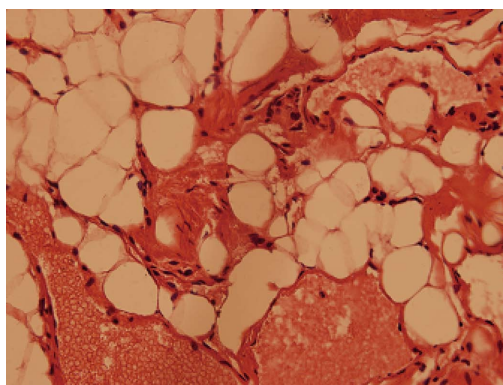


Figure 2 Histomicrograph of the surgical specimen shows typical angiolipoma. Composed of mature fat cells and abnormal vascular elements.

analgesia below the T6 level, varicose vein of the left lower limb, decreased muscle strength and increased muscle tension, as well as hyperreflexia of the low extremities.

A MRI scan showed a fusiform posterior epidural mass compressing the thoracic cord over two vertebral body segments between T3-T4. The mass was inhomogeneous, isointense on T1-weighted images (Figure 1A), slightly hyperintense on T2-weighted image (Figure 1B), hyperintense on fat-saturated T2-weighted images (Figure 1C) and inhomogeneously enhanced on fat-saturated T1-weighted image (Figure 1D). The lesion's long axis paralleled the long axis of the spinal cord, tapering at both ends. On the axial T1 weighted images, a crack like low signal between spinal cord and the lesion (Figure 1E) which is the typical appearance of the case can be seen. A laminectomy with gross total resection of the lesion was performed. During the operation, a flesh pink vascular mass with obscure margin and strong but pliable texture was found in the posterior epidural space from T3 to T4. There was no infiltration of the dura or the vertebrae. Histopathological study of the surgical specimen showed a typical angiolipoma (Figure 2) composed of mature fat cells and abnormal vascular elements.

DISCUSSION

Pathology

Berenbruch *et al.*^[3] reported the first case of spinal angiolipoma (AGL) in 1890 in about a 16-year-old with numerous cutaneous lipomas, while the first pathological report was made by Howard *et al.*^[4] in 1960. It is composed of varying proportions of mature fat cells and abnormal capillary, sinusoidal, venous or arterial vascular elements. Subsequently, AGL has been further subdivided by Lin *et al.*^[5] into two categories: noninfiltrating and infiltrating. The former is encapsulated and well demarcated, not infiltrating the dura or the vertebrae, often in the dorsal aspect of the spinal canal. Whereas, the latter is very rare, entirely or partially unencapsulated, situated in the anterior or anterolateral aspect of the spinal canal with ill-defined margins and infiltrates the surrounding tissues. Our case is type 1, unencapsulated, but not infiltrating the dura or the vertebrae.

The origin and pathogenesis of AGLs is unknown. Histologically, the lesion is mainly composed of mature fat cells and blood vessels. The fat composition is similar to the general adipose tissue and the vascular components consist of capillaries, sinusoids, thin-walled blood vessels or thick-walled blood vessels with smooth muscle and occasionally well-developed small arteries can be seen. A diagnostic feature is the presence of fibrin thrombi in the lumen of capillaries. Degenerative changes (*i.e.*, myxoid change, hyalinization and fibrosis) may be present in some longstanding cases^[6]. Traditionally, AGL is considered a subtype of spinal lipomas, but more recent clinicalpathological studies^[7] considered them as a specific entity different from pure lipomas because they were not associated with spinal dysraphism. AGLs usually contain a much greater number of mature, thick-walled vessels than liposarcomas^[5]. Angiomyolipomas are a variant of angiolipoma characterized by vascular smooth muscle proliferation extending into the surrounding fat^[8].

Clinical presentation

The clinical presentation of spinal AGLs is not different from any other benign epidural tumor. We found that the AGLs have been reported to occur predominantly in women (female:male = 22:17; Table 1) and are more common in the fifth decade^[9]. In our review, age of presentation ranged from 4 to 81 years old, with mean age at presentation of 46 years. The mode of onset may be acute, subacute or chronic, may show radicular, paraplegic, progressive or remitting-relapsing clinical types. The most common initial symptoms are back pain, lower extremity numbness or paresthesias and leg weakness (Table 1), and progressive neurological symptoms secondary to spinal cord compression may develop later on. The symptoms usually evolve over a period of months to years, but the progression can be accelerated by vascular steal phenomena, vascular engorgement, venous stasis with thrombosis, bleeding into the tumor and rarely intratumoral abscess^[10]. Bleeding is extremely rare in angioliipomas. Akhaddar *et al*^[10] reported a case presenting with spontaneous bleeding causing acute paraplegia. Like other vascular lesions, onset or deterioration may occur during pregnancy^[11] or with weight gain. This was not the case in our patient.

Diagnostic evaluation

The appearance of angioliipomas correlated well with their histological composition, so the AGL is often misdiagnosed. In the majority of cases, plain vertebral radiography demonstrates normalities. If causing adjacent bone destruction, AGL must be differentiated from epidural metastases. The metastasis is the most common malignancy of epidural space, which is typically an irregular soft tissue mass with adjacent bone destruction. Trabeculation of the affected vertebral body and erosion of the pedicle may be identified in tumors infiltrating bone^[12,13]. Computed tomography (CT) usually demonstrates a hypodense lesion with fat density, provides information on the degree of bony involvement^[7] and also can demonstrate variable degree of enhancement after contrast injection. However, CT may not be specific for spinal epidural angioliipomas and could be misleading in some cases^[14].

MRI is the imaging modality of choice for detecting angioliipomas. MRI was performed in approximately 70 cases (since 1988), but there were only 39 cases with adequate data (Table 1). The angioliipomas appear as an isointense or hyperintense extradural mass on T1-weighted images, but occasionally a hypointense mass^[11,12,15,16] may be seen. The degree of central hypointensity on T1-weighted images is predictive of the degree. On T2-weighted images, the tumors have variable signal intensity, with a predominance of hyperintensity^[10,15,17-29]. Significant heterogeneity in the imaging studies is attributed to the variable vascular and adipose elements of the tumor^[12]. In our case, the tumor is isointense with fewer areas of hypointensity on T1-weighted images and hyperintense on fat-saturated T2-weighted

images. There is slightly heterogeneity which is consistent with few vascular elements, less adipose elements of the tumor. Gadolinium enhancement reflects the vascularity of these tumors. In our case, the lesion was strongly enhanced after contrast injection.

Spinal hemangiomas also present as mixed signal intensity lesions on MRI, although the hyperintensity on T2-weighted images is more striking. Suppression of high signal intensity on fat-saturated T1-weighted images may be very useful for distinguishing between angioliipomas and melanomas or sub-acute hemorrhage. Most spinal angioliipomas show enhancement with contrast medium which better defines the borders of the tumor^[30]. In our case the tumor has an obscure margin, while after contrast injection its border becomes very clear. Contrast enhancement allows for differential diagnosis between spinal angioliipoma and extradural lipomatosis as the latter does not show contrast enhancement^[31]. Unlike some other vascular tumors (*e.g.*, glomus jugulare tumors), angioliipomas do not typically contain vascular flow voids on magnetic resonance images^[32,33], there is only one case containing vascular flow voids in the literature. This is probably because of the preponderance of capillaries and venous channels in angioliipomas, which distinguish them from malformations with arteriovenous shunting and from those lesions with predominantly arteriolar circulation both of which produce fast flow, seen on MRI as flow-void phenomena. In our case, there is no flow-void phenomena which is identical with those reported in literature.

Treatment

There is no clear consensus as to what combination of therapy is optimal for spinal AGL. The biological behavior of the infiltrating and noninfiltrating AGL implicates a different treatment approach^[18]. To date, the main treatment is total surgical resection. Most extradural non-infiltrative tumors are amenable to complete excision *via* laminectomy. Total removal of infiltrating angioliipomas that often involve the vertebral body has been recommended using the anterolateral approach and stabilization of the affected vertebrae is desirable^[9]. Extent of tumor resection in infiltrative angioliipoma has been debated, but most authors agree that risking neurological function is not necessary with aggressive attempts to attain gross total removal^[9,17,34]. In spite of vascularization of the tumor, profuse hemorrhage has rarely been reported^[10]. Although complete removal of the lesion is not always easily achievable, recurrence is exceptional^[35]. In case of recurring or infiltrative SALs, wider resection followed by radiotherapy should be considered^[17]. Most patients have a good prognosis because the tumors are usually slow growing and do not undergo malignant transformation. In our case, the patient's clinical symptoms improved postoperatively. There are no signs of tumor recurrence and no neurological deficit during the two year follow-up period.

In conclusion, AGL is a rare but benign clinico-

Table 1 Reported cases of spinal extradural angiolipomas performed with magnetic resonance imaging

Ref.	Age (yr)	Sex	Clinical presentation (signs/duration)	MRI finding (T1/T2, post-contrast)	Pathological finding
Turgut <i>et al</i> ^[1]	26	F	Acute onset of paraplegia, in week 31 of pregnancy	T1 and T2: Hyperintense	Angiolipoma
Diyora <i>et al</i> ^[6]	20	M	Upper abdominal pain/6 mo, and lower limb weakness/1 wk	Isointense/hyperintense; enhancement	Angiolipoma
Turgut <i>et al</i> ^[9]	54	F	Weakness, urinary incontinence, and constipation/5 mo	Very hyperintense/ nearly isointense	Angiolipoma
Akhaddar <i>et al</i> ^[10]	47	M	Sudden back pain, paresthesia and complete neurological palsy/a few-minutes period	Isointense/ slightly hyperintense; no gadolinium enhancement	Angiolipoma
Park <i>et al</i> ^[11]	74	F	Lower back pain/5 mo	Low signal intensity/iso-or high signal intensity; High signal intensity after gadolinium injection	Angiolipoma
Provenzale <i>et al</i> ^[12]	38	F	Lower back pain/3 yr	Moderately hypointense/nearly isointense; Slightly inhomogeneous enhancement	Angiolipoma
	61	F	Paraparesis/2 yr	T1: Slightly hypointense; Inhomogeneous enhancement	Angiolipoma
	42	F	Midthoracic back pain/2 yr	Iso-hypo-intense/nearly isointense	Angiolipoma
Leu <i>et al</i> ^[15]	81	M	Unstable gait, and lower limbs weakness/2 wk	Inhomogeneous hypointensity/high signal intensity; Strongly enhanced	Angiolipoma
Yen <i>et al</i> ^[16]	71	M	Acute paraparesis	T1: Homogeneously hypointense	Angiolipoma
Fourney <i>et al</i> ^[17]	46	F	Feet and legs numbness/4 yr	Homogeneously hyperintense/hyperintense; enhanced	Angiolipoma
Shibata <i>et al</i> ^[18]	38	F	Paraparesis/6 mo	T1 and T2: High-intensity signal	Angiolipoma
Bouramas <i>et al</i> ^[19]	27	F	Diminution sensation/2 mo	Heterogeneous signal intensity/high signal intensity	Angiolipoma
Boockvar <i>et al</i> ^[20]	34	F	Interscapular back pain/5 mo	Hyperintense/hyperintense; Enhanced homogeneously	Angiolipoma
Amlashi <i>et al</i> ^[21]	36	M	Back pain, and both legs weakness	T1 and T2: Homogenous, hyperintense	Angiolipoma
Garg <i>et al</i> ^[22]	12	F	Paraparesis/the previous year	Hyperintensity/hyperintensity; Heterogeneous contrast enhancement	Angiolipoma
	26	M	Paraparesis/3 mo	T1 and T2: Homogeneous high signal intensity; contrast enhancement	Angiolipoma
	28	M	Bowel and bladder impairment, paraparesis/the previous year	T1 and T2: Homogeneously hyperintense	Angiolipoma
do Souto <i>et al</i> ^[23]	46	F	Low back pain/10 yr	Isointense/hyperintense; Homogeneous and intense enhancement	Angiolipoma
Rabin <i>et al</i> ^[24]	47	M	Legs paresthesias/6 mo	T1 and T2: High signal	Angiolipoma
Samdani <i>et al</i> ^[25]	49	F	Back pain, lower extremity weakness/3 yr	Intermediate-signal intensity/hyperintensity; homogenous contrast enhancement	Angiolipoma
Dogan <i>et al</i> ^[26]	50	F	Lumbosciatalgia/2 yr	Isointense/hyperintense; Homogeneous enhancement	Angiolipoma
	36	M	Low back pain/8 mo	Isointense/hyperintense; Homogeneous enhancement	Angiolipoma
Guzey <i>et al</i> ^[27]	41	F	Low back and leg pain/18 mo	Iso-hyperintense/hyperintense; homogeneously enhanced	Angiolipoma
Hungs <i>et al</i> ^[28]	52		Thoracolumbar pain/1.5 yr	T1: Hyperintense; Diffusely intense enhancement	Angiolipoma
Sankaran <i>et al</i> ^[29]	77	M	Paraparesis/48 h	Isointense/inhomogeneous hyperintensity; Inhomogeneous enhancement	Angiolipoma
Weill <i>et al</i> ^[33]	46	F	Paraparesis/1 yr	Iso-hypointensities/mixed signal	Angiolipoma
	27	F	Right leg weak/several week	Very-hyperintense/mixed-signal; Inhomogeneous enhancement	Angiolipoma
Gelabert-González <i>et al</i> ^[34]	16	M	Low back pain/6 mo	Slightly inhomogeneous/heterogeneously hypointense	Angiolipoma
	45		Both feet numbness, and leg weakness/6 mo	T1 and T2: Moderately hyperintense relative to spinal cord	Angiolipoma
Sakaida <i>et al</i> ^[36]	72	M	Legs abnormal sensation/4 mo	Inhomogeneous enhancement	Angiolipoma
Oge <i>et al</i> ^[37]	72	M	Paraparesis/4 d	T1: Hyperintense	Angiolipoma
al-Anazi <i>et al</i> ^[38]	38	F	Both feet numbness, 8-mo pregnant housewife	T1: Hyperintense	Angiolipoma
Gelabert-González <i>et al</i> ^[39]	4	M	Back pain, and both legs weakness/2 d	T1: Mixed-intensity	Angiolipoma
Rocchi <i>et al</i> ^[40]	60	M	Lumbosciatalgia/2 yr	T1: Signal intensity similar to that of the subcutaneous adipose tissue	Angiolipoma
	54	F	Lumbosciatalgia/12 mo	Homogeneous contrast enhancement	Angiolipoma
Chotai <i>et al</i> ^[41]	69	M	Back pain, paresthesias, and hypesthesia/5 yr	T1: Slightly high intensity with areas of hypointensity; Inhomogeneous enhancement	Angiolipoma
Konya <i>et al</i> ^[42]	60	F	Low back pain/6 mo	T1: Hyperintense; marked enhancement	Angiolipoma
Current study	63	M	Lower extremities numbness/1 yr	Isointense/ slightly hyperintense; Obviously inhomogeneous enhancement	Angiolipoma

Age, sex, clinical presentation, magnetic resonance imaging findings, and pathological findings are shown for each case. Authors and year of publication are shown for each case reported. F: Female; M: Male.

pathological entity which is composed of fatty tissue and vascular elements. It grows in a spindle shape along the spinal canal, without associated malformations. The postoperative outcome after surgical management of this lesion is favorable. Accurate pre-operative diagnosis is very important. MRI typically shows an iso- to hyperintense mass on T1 weighted images and hyperintense mass without flow voids on T2 weighted images in the posterior epidural space. Following intravenous injection of contrast material, avid inhomogeneous enhancement is seen.

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