Review Article

Spinal angiolipomas: A puzzling case and review of a rare entity

ABSTRACT

Patients with spinal epidural abscesses (SEAs) may have a variable presentation. Such an infection has a typical appearance on magnetic resonance imaging (MRI) and enhances with gadolinium. We present a case that was a diagnostic challenge where pre- and intra-operative findings resulted in conflicting impressions. The mimicker was a spinal angiolipoma (SAL). The authors then provide a thorough review of this rare spinal neoplasm. A 55-year-old man presented with back pain, paresis, paresthesia, and urinary retention. MRI was indicative of a longitudinal epidural thoracic mass with a signal homogeneous to nearby fat, curvilinear vessels, and lack of enhancement. Although at emergent surgery, the lesion was found to contain abundant purulent material. Microbiology was positive for methicillin-resistant *Staphylococcus aureus* and consistent with SEA without evidence of neoplasia. While the imaging features were suggestive of an angiolipoma, the findings at surgery made SEA more likely, which were validated histopathologically. The diagnosis of SEA is often clear-cut, and the literature has reported only a few instances in which it masqueraded as another process such as lymphoma or myelitis. The case highlights SEA masquerading as an angiolipoma, and further demonstrates to clinicians that obtaining tissue diagnosis plays a crucial role diagnostically and therapeutically. SALs, on the other hand, are slow-growing tumors that can be infiltrating or noninfiltrating. They typically present with chronic symptoms and T1-MRI shows an inhomogeneous picture. Complete surgical excision is standard of care and patients tend to do well afterward.

Keywords: Angiolipoma, back pain, epidural abscess, magnetic resonance imaging, spinal cord compression, spine neoplasms

INTRODUCTION

Angiolipomas, by definition, tend to be benign tumors that may be found in areas such as the neck, trunk, and forearm usually presenting subtly and have a benign course.[1,2] Although rare, spinal angiolipomas (SALs) also form an important subset from a clinical perspective. In the literature, around 200 cases of SAL have been recorded; accounting for <2% of all spinal neoplasms.[3-6] They have been documented as early as 1890, but the first description was in the 1900s by Liebscher. Sixty years later, Howard and Helwig would coin the term angiolipoma after describing the defining characteristics of the tumor.[9]

The neoplasm occurs predominantly in the thoracic region of the spinal cord, particularly T2–T5 and both intra- and extra-dural forms have been reported.[6-11] The predilection to this location might be due to the relatively poor blood

Access this article online	
Website: www.jcvjs.com	Quick Response Code
DOI: 10.4103/jcvjs.JCVJS_23_17	

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How to cite this article: Shweikeh F, Sangtani A, Steinmetz MP, Zahos P, Chopko B. Spinal angiolipomas: A puzzling case and review of a rare entity. J Craniovert Jun Spine 2017;8:91-6.

supply within this area.[9] Angiolipomas are similar to lipomas histologically, but include an additional vascular component and are therefore considered to be similar to hemangiomas as well.[12] The clinical presentation of SAL may mimic that of other spinal space occupying lesions such as spinal epidural abscess (SEA) and therefore adds a certain degree of diagnostic complexity.[14]

In terms of SEA, estimates have indicated that it occurs in approximately 2–3 admissions per 10,000 hospital admissions.[16] The diagnosis is usually suspected with characteristic clinical signs and symptoms, distinctive radiographic findings, and confirmed microbiologically.[16-18] The thoracolumbar regions are a common location.[12,16] Moreover, a number of risk factors have been elucidated along with SEA, including diabetes mellitus, intravenous drug abuse, alcohol abuse, immunodeficiency, and invasive spine procedures.[16-18] We present a case of a patient with a SEA that mimicked an angiolipoma on both clinical and imaging features and review the literature.

CASE REPORT

A 55-year-old man with an unremarkable medical history presented with a multiple week history of lower back pain radiating to the upper scapula and a 2-day history of urinary retention. He had a history of chronic back pain that had worsened in the more recent weeks before the presentation. The patient was seen 4 days prior for an upper respiratory infection and back pain in the emergency department and given nonsteroidal anti-inflammatory medications. His vital signs were within normal limits. Initial physical examination showed irregular gait, stiffness, and weakness in his left lower extremity. His weakness was in the L4–L5 and S1 area, and he exhibited decreased sensation in the L5 distribution and positive straight leg raise. He had no clonus, negative Babinski, and 2+ reflexes. The rectal tone was normal.

There were no any abnormalities on his blood tests, including no elevation in white blood cells or a left shift. A magnetic resonance imaging (MRI) of the thoracic spine showed an epidural longitudinal mass spanning T3–T8. The majority of the mass demonstrated a signal homogeneous to nearby fat. Curvilinear vessels were also present within the mass. There was significant spinal cord compression most evident at T6 and T7 without enhancement [Figure 1]. There was no apparent spinal cord or disk enhancement with contrast. There was no evidence of hemorrhage or concerning findings for an abscess. The most likely findings on imaging combined with the signs, symptoms, and chemistries were those related to an angiolipoma.

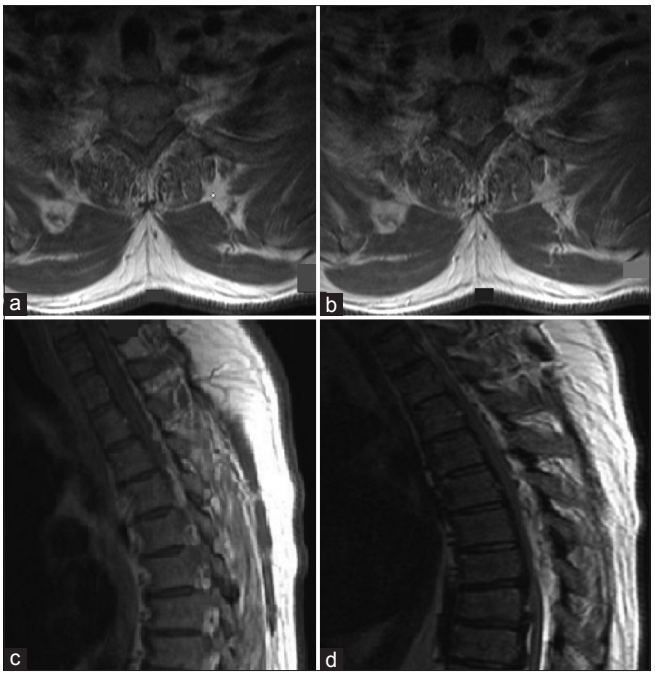

Figure 1: Axial T1-weighted magnetic resonance imaging without contrast (a) and postcontrast (b) of the thoracic spine lesion. Saggital postcontrast T1-weighted (c) and T2-weighted magnetic resonance imaging without contrast (d) of the same lesion

The patient was emergently taken to the operating room for decompression of the spinal cord. He underwent posterior T3–T8 laminectomy, with resection of epidural fat and saline irrigation. The lesion was found to contain purulent material with necrotic epidural fat from T3–T8. No tumor remnant or capsule was found. Initial gram stain was positive for *Staphylococcus aureus*, with final cultures positive for methicillin-sensitive *S. aureus*. Overall, the histopathology was consistent with epidural abscess. A thorough examination of necrotic fat found no evidence of angiolipoma or any other neoplasia.

Postoperatively, the patient's weakness significantly improved with mild residual weakness persistent on his left extremities. The patient was sent to rehab for strengthening, and eventual resolution of all symptoms. He completed a 6-week course of intravenous antibiotics. At most recent outpatient follow-up, MRI revealed no lesions (though the Institutional Review Board/Ethics Committee approval and patient consent were not sought for this case presentation, neither were required).

DISCUSSION

In usual cases, patients with SEA have a significant history of medical problems and arrive with acute back pain and constitutional systemic signs and symptoms. Imaging of the lesion is distinctive, demonstrating iso- or hypo-intensity

on T1-MRI and hyperintensity on T2.[15,19] Gadolinium administration usually results in a typical ring-enhancing lesion with a nonenhancing center.[16] Diagnosis should be confirmed using tissue histopathological analysis, with *S. aureus* being the most frequently isolated microbe.[16] SAL, on the other hand, is an even less common entity, comprising 0.04%–1.2% of all spinal neoplasms.[10,14,20,21] These tumors occur predominantly in the thoracic spine, particularly T2–T5. While similar to lipomas, they include an additional vascular component liking them to hemangiomas, representing an intermediate subsistence between the two neoplasms.[10,14,20,21]

Pathology

The tumor is composed of mature adipose fat cells and vascular components, and to be classified as a SAL, more than 50% of the cells must be fat cells.[8,22,23] The benign nature of the tumor has also been described as a mixture of developed adipocytes and dividing miniature vessels containing fibrin, as well as a neoplasm or a congenital malformation of pluripotent mesenchymal stem cell origin.[2,7] The tumor has been described, in certain cases, to deteriorate more rapidly resulting in hemorrhage, thrombosis, vascular steal phenomena, or expand into a larger tumor volume.[2,12,13,17] In certain cases of SAL, significant amounts of smooth muscle can be appreciated, further classifying the tumor as an angiomyolipoma. Without an adventitia, the smooth muscle may mesh into the surrounding tissue, or into cartilage or osteoid tissue in the vicinity.[29] The tumor can result in eroded pedicles, trabeculation of vertebral bodies or mediastinum, and/or spinal cord compression.[22,30] In certain cases, the trabeculations present as vertical striations of the vertebral bodies, which is also characteristic of spinal hemangiomas.[30]

Clinical presentation

Patients with angiolipoma present with progressive neurological deficits and pain in the affected regions related to progressive spinal cord compression, gradually worsening over the course of approximately 1 year [Table 1].[26,32-34] More rapid onset of symptoms may be attributed to vigorous exercise, which increases blood flow to the tumor and exacerbates epidural bleeding, leading to neurologic symptoms. Patients may also present with gait disturbances and urinary hesitation.[9,24] Symptoms may mimic a demyelinating disorder, such as multiple sclerosis and are worsened in pregnant females due to increased epidural pressure that further compresses the spinal cord.[12,22]

In contrast, SEA has been associated with verified comorbidities[16-18] and the infection usually manifests itself in middle-aged persons with fever, chills, and malaise.[17,18] Clinical presentation can vary with the time-frame and starts as fevers with chills that progress to spinal pain with neurological symptoms and culminates with bowl and/or bladder dysfunction.[17,18,27] The presence of a fever can be variable, but back pain is almost always present,[12,18] tending to be over the affected region and exacerbated by movement.[27]

Diagnosis

Plain film radiography is a poor indicator of a SAL, only occasionally indicating pedicle erosion.[23] Although evidence of bone deterioration may be present, computed tomography scans of angiolipomas show little to no contrast enhancement and can thereby be distinguished from vertebral. Therefore, T1-MRI is the standard because it alludes to the presence of a lipid tumor with vascular components resulting in an inhomogeneous picture.[7,9,12,21,25] The level of vascularization correlates with hypointense regions, but iso- or hyper-intense regions may also be appreciated, usually within homogenous intensity in any one patient. The T1-imaging can be used, therefore, to rule out lipomas, which would not have the hypointense regions correlated with vascularization.[4,8,12,22,24] It can be distinguished from spinal vascular malformations, such as fistulas because of the latter present with enlarged subarachnoid vessels on T1-imaging.[33]

Suppression of the high signal intensity in fat-suppressed T1-imaging can exclude methemoglobin and melanin, which would consequently differentiate between spinal epidural angiolipomas with flow voids and arteriovenous malformations as well.[34,37] In addition, T2-MRI generally shows hyperintense regions, but the results are variable.[10] Gadolinium enhancement can also contribute to diagnosis by eliminating extradural lipomatosis, which are not enhanced by IV injected contrast, from the differential diagnosis.[12,13,34] In addition, angiography may be used for embolization of the tumor, which helps with removal,[26] though it is difficult to distinguish from certain diagnoses, such as metastases and meningiomas.[34]

Treatment and outcome

Treatment depends on tumor characterization. Noninfiltrating undergo total excision, which is usually possible because the tumor does not adhere to the dura underneath.[29] In cases of infiltrating type, surgery is always recommended while radiation is sometimes included [Table 1].[22,24] That being said, consensus and hence clear guidelines remain hazy on treatment. This may be due to a combination of the disease entity being rare in addition to the heterogeneous biological behavior that ultimately entails individualizing treatment modalities.[14] Outcomes after surgical resection tend to be favorable.[32] In fact, a literature review revealed no differences in outcomes between infiltrating and noninfiltrating.[39]

Table 1: Characteristics of recent reported cases describing thoracic spinal angiolipomas

Authors (Reference)	Age/ Gender	Location/Levels involved	Associated conditions	Duration of Symptoms	Presenting problem	Imaging (description on MRI/CT)	Treatment	Outcome
Sim, 2015	58 F 42 F 39 M 26 F	T2 – T6 T11 – L2 T3 - T6 L5 – S1	Obesity, DM2 Asthma	4 months	Burning in bilateral hands/ shoulders, back pain Bilateral lower limb numbness/ weakness	Enhancing fat w/extradural mass	T2 – T6 laminectomy	Uncomplicated post-operative course; persisting 4/5 dorsiflexion, 5/5 power in others
Nadi, 2015	50 F	T6 – T9	-	10 year	Back pain mid-dorsal area; progressive LE weakness and stiffness	Inhomogeneously enhancing hyperintensity on T2W1 and on fat suppression T1W1	T11 – L2 laminectomy	uneventful postoperative course; Mild residual spasticity and hyperreflexia
Regato, 2015	65 F	T8 - T10	-	6 weeks	LE dysesthesia and neurogenic claudication	T1: dumbbell-shaped hyperintense mass with hypo intense regions	T3-T7 laminectomy	Uneventful recovery, symptom free and no recurrence after 6 months
Da Costa, 2014	43 M	Thoracic	-	32 hours	Sudden onset thoracic pain, paraplegia	T1 w/Gd: slight tissue cavitation peripherally T2: mixed lesion, hyperintense areas, isointense surrounding	L5/S1 hemilaminectomy	At 10 months: 4/5 strength, can stand and walk short distances w/o support
Si, 2014	21 patients (1/01-2/13) 9 M; 12 F	3L; 16T; 2C	-	12 hrs – 360 months	(Various)	Fat: hyperintense on T1/T2 Vascular: isointense on T1, hyperintense on T2	Total resection in all 21	Better prognosis in IA vs IB (based on JOA score) Mean recovery rate: 93.9% in IA vs 45.5% in IB
Prasad, 2014	26 M	T5-T9	-	6 months	Progressive spastic paraparasis with autonomic involvementsensory level	hyperintensity/isointensity on T1; & hypointensties on T2	enbloc laminoplasty	Initial deterioration followed by gradual recovery
Fujiwara *et al.*, 2013	64 F 65 M	T5-T8 T5-T7	Hypertension Healthy with no prior medical history	3 months 2 years	, Left leg dysesthesia, back pain, spasticity, , gait disturbances, hyperreflexia, ankle clonus, , urinary retardation	T1WI: Isointense masses T2WI: Hyperintense masses	T5-T8 laminoplasty T5-T7 laminoplasty	Hyperreflexia was the only abnormality No abnormalities after 9 months
Meng *et al.*, 2013	63M	T3-T4	Varicose vein of left lower limb	1 year	Bilateral hypesthesia difficulty urinating, paraparesis, hyperreflexia	T1WI: inhomogeneous isointense mass T2WI: hyperintense fusiform Post-contrast T1: low signal	T3-T4 laminectomy with total resection	-
Han *et al.*, 2012	58 M	T3-T5	Not mentioned	9 months	Paraparesis and hypesthesia of the lower extremities; hyperreflexia on the R leg	CT: mass along posterior epidural space MRI: infiltrating mass	T3-T5 laminectomy	Improved muscle strength and sensation
Ghanta *et al.*, 2012	56 M	T4-T6	Multiple subcutaneous lipomas	3 months	hypesthesia in lower extremities, ambulatory difficulties	TW1: hypointense mass	T4-T6 laminectomy	no neurological abnormalities after 5 years
Haji *et al.*, 2011	65 F	T4-T7	Mild degenerative disc disease; renal mass	9 months	Bilateral hypesthesia, paraparesis, progressing to T10, hyperreflexia, difficulty urinating	hyperintense at T5-T7	T3-T7 posterior laminectomy	Patient regained muscle strength, lingering hypesthesia

Table 2: Summary of previous case reports involving spinal epidural abscess mimicking a different disease entity

Authors (Reference)	Age/sex	Location/ levels	Mimicked pathology	Presentation	Origin of infection	Imaging (description on MRI/CT)
Hanck and Muñiz[1]	75/female	C6-C7	Vertebral fracture	Low back and right leg pain	*Staphylococcus aureus* from infective endocarditis	CT: C6-C7 cortical endplate erosion and loss of disk space MRI: Spondylodiscitis and osteomyelitis with an epidural abscess
Bakar and Tekkok[15]	71/female	L5-S1	Disc herniation	Bladder infection	Not identified	CT: No abnormalities MRI: L5-S1 disc degeneration
Patel *et al*.[19]	55/male	T8-T10	Lymphoma	Severe back pain	Dental procedure	T1/T2-weighted MRI: Hypointense mass that enhanced uniformly with gadolinium
Rao *et al*.[35]	28/female	C6-T2	Transverse myelitis	Urinary retention and constipation; HIV+	Minor trauma from a pat on the back	MRI: Extradural collection

MRI - Magnetic resonance imaging; CT - Computed tomography

Overall, good clinical outcomes have been reported among the different types.[6,39]

Not many cases of SEA have been reported to mimic other forms of pathology, and none have been reported that mimicked SAL. Previously reported as masquerading entities include lymphoma, vertebral fracture, transverse myelitis, and disc herniation, as summarized in Table 2. Our case highlights SEA masquerading as an angiolipoma. Despite being a relatively rare entity, SAL is a clinically relevant subset of benign tumors.

CONCLUSION

Although patients with SEAs may present with classic signs and symptoms, this presented case highlights the potential variability in the infection's characteristics and diagnostic challenges. In this case, the indolent course in the patient's symptomology and imaging traits of the thoracic spinal mass were highly suggestive of an angiolipoma, but the findings at operation and the pathology confirmed the lesion to be a SEA. Contradictory diagnostic and therapeutic impressions in medicine remain as part of the foundation for abstract thought, intellectual curiosity, though as our case illustrates, and in most clinical situations, it presents an obstacle to the surgeon.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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