



Treatment of a Refractory Angiolipoma with Thoracic Extension

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Abstract

The spinal angiolipoma is a rare disease. It is a benign lesion that composed of angiomatous and lipomatous structure. It can be diagnosed *via* the magnetic resonance imaging. Spinal cord compression is common and the neurological symptoms may also appear. Surgical resection is the most important treatment that relieved the symptoms. Spinal angiolipoma rarely recurred. However, some residual lesions may cause newly developed neurological symptoms. Adjuvant radiotherapy is a choice after the surgical resection.

Most of the patients underwent surgical resection. After the surgical treatment, the neurological deficit in this patients vanished. However, some studies shows treatment of infiltrating angiolipomas is aimed at wide excision with radiotherapy indicated for cases of recurrence.

Surgical removal should be followed by closely monitoring with physical examination and image even when the pathologically reported benign tumor. Further radiotherapy may be taken into consideration if this benign tumor has any malignant potential.

Keywords: Angiolipoma; Tumors; Treatment

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Introduction

Spinal angiolipomas are benign tumors histologically, and most of them are found in the spinal axis, especially the thoracic epidural space. However, it is a kind of rare neoplasm. In the spinal region, spinal cord compression syndrome caused by the angiolipoma can be usually resolved with surgical intervention [1]. Aspinal angiolipoma with malignant behavior extending outside of the spinal region has not been reported previously. Here, we describe a refractory case of a benign tumor that was diagnosed as an angiolipoma.

Case Presentation

Consent was obtained from the patient to publish her case. A 60-year-old woman presented to our hospital with numbness and weakness of the lower limbs and unstable gait for 6 months. The physical examination revealed Lhermitte's sign and positive Spurling's sign. The blood hemogram revealed normal results. The chest roentgenogram showed spondylosis, degenerative disc disease as marginal spur formation, and a mass lesion at the aortic level. Chest Computed Tomography (CT) revealed an oval mass measuring 3.7 cm × 2.6 cm in the left posterior mediastinum with surrounding bone destruction. The thoracic spine CT showed a 4.8 cm enhancing tumor in the long axis of the spinal canal with left lateral extension into the pleural space (Figure 1). Magnetic Resonance Imaging (MRI) of the thoracic spine showed a 4.85 cm × 4 cm lobulated soft-tissue mass involving the extradural space at the T5 level on the right side (Figure 2A, 2B).

Result

To relieve the patient's symptoms, total laminectomies of the T4-6 vertebrae with resection of the spinal tumor and Video-Assisted Thoracic Surgery (VATS) for resection of the paraspinal tumor were completed. After surgical treatment, her symptoms improved; however, the presence of malignancy was unclear, and her muscle strength was still poor.

Postoperatively, the microscopic examination (Figure 3) showed an angiolipoma composed of medium-sized to small-sized vessels and many capillaries with mature adipocytes as well as

Table 1: Characteristics of similar patients treated by surgical resection reported in the literature and our case.

Study	Age (years)/sex	Symptoms	Duration of symptoms	Spinal location	Extraspinal tumor	Treatment	Follow-up duration	Outcome	Time of symptom relief
Reyes et al. [9]	68/M	Lumbago	3 years	T12 to L1-L2	No	Surgery	N/A	Resolved	1 year
Rkhami et al. [10]	65/F	Paraplegia and sphincter disorders	7 months	T7-T10	No	Surgery	5 years	Recovered	1 year
Mohammed and Ahmed [11]	35/F	Lower limb paralysis and urinary incontinence	Acute onset	T5-T8	No	Surgery	1 year	Recovered	1 year
Ramdasi et al. [12]	58/M	Neck and interscapular pain, paraplegia, and sphincter incontinence	48 hours	C7-T1	No	Surgery	2 years	Rapid recovery	2 years
Chotai et al. [13]	68/M	LBP and progressive paresthesia	5 years	T9-T11	No	Surgery	N/A	Improved	N/A
Tsutsumiet al. [14]	26/F	Paraplegia and incontinence	Acute onset	T3-T4	No	Surgery	1 year	Improved	1 year
Akhaddar et al. [15]	47/m	Back pain, paresthesia, complete neurological palsy	Acute onset	T2-T3	No	Surgery	>2 years	Improved	Immediately
Lacour et al. [16]	17/M	Acute paraplegia	Acute onset	T7-T12	No	Surgery	N/A	Improved	2 months
Present case	60/F	Numbness and weakness of the lower limbs	6 months	T4-6	Yes	Surgery	6 months	Recurrence	

M: Male; F: Female; LBP: Low Back Pain; N/A: Not Applicable



Figure 1: Preoperative thoracic spine contrast-enhanced computed tomography scans showing a lobulated mass in the left paraspinal region with thoracic extension into the extraspinal canal and cord compression at the dorsal horn.

occasional myxoid change of the mediastinal tissue. Regarding immune histochemistry findings, the angiolipoma stained diffuse and strongly positive for CD34. These cells failed to express S100, the glial fibrillary acidic protein, CK, epithelial membrane antigen, and D2-40. Therefore, the patient was diagnosed as having an angiolipoma.

Three months later, the postoperative spinal thoracic MRI with contrast showed a mass lesion measuring about 3.0 cm × 1.9 cm × 5.8 cm in the extradural space at the T4-T6 level with extension into the left T5/6 neural foramen. T5 vertebral body involvement (Figure 4A) and spinal cord compression were also indicated. The tumor with mass extension into the left paraspinal region that was observed previously was not seen (Figure 4B). Thus, she underwent regular Stereotactic Radiation Therapy (SRT) for the suspicious recurrent lesion.

Currently, she is undergoing regular outpatient follow-up and weekly SRT. She has remained in a relatively stable condition. Muscle strength of her extremities has not worsened, and she has not



Figure 2A: Preoperative T2-weighted magnetic resonance imaging scan at the T5 level in the sagittal view. The enhanced intraspinal dumbbell-shaped lesion involves the extradural space at the T5 level and causes spinal cord compression, most likely in the extra medullary intradural direction.

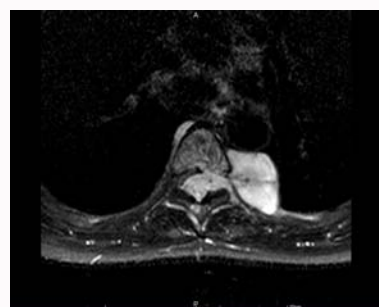


Figure 2B: Axial T2-weighted magnetic resonance imaging scan.

developed new neurological symptoms under regular SRT.

Discussion

In 1960, the term “angiolipoma” was first introduced [2]. This tumor commonly originates from subcutaneous tissue in the

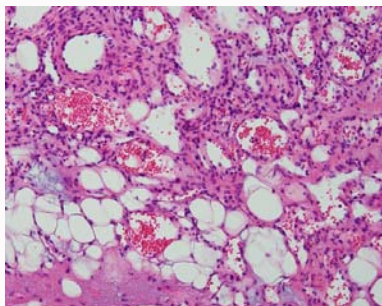


Figure 3: Mature adipose tissue is interspersed with blood channels of various sizes (hematoxylin and eosin stain).



Figure 4A: A mass lesion measuring about 3.0 cm × 1.9 cm × 5.8 cm involving the extradural space at the T4-T6 level, extending into the left T5/6 neural foramen. T5 vertebral body involvement is also seen.

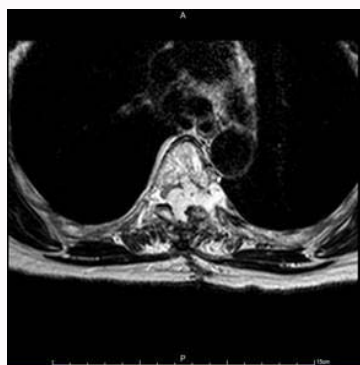


Figure 4B: The spinal cord is compressed, and the prior tumor mass that extended into the left paraspinal region has disappeared.

forearms, trunk, neck, and proximal upper extremities, but it is a rare, benign tumor when located in the thoracic posterior epidural spaces [3]. Histologically, it is a benign tumor, and it can be confused with a hamartoma or benign tumor that develops from dural mesenchymal cells [3]. An angiolipoma with a malignant tendency is very rare.

MRI is the most valuable modality for determining the characteristics of an angiolipoma [4]. A lipomatous mass has a different appearance from the spine, which appears as a high-density structure, and this type of mass has a relatively lower density than the bone material when viewed by CT; thus, an angiolipoma may appear as a lipomatous extradural mass. Bone erosion may also be visible because of tumor compression [5]. As its name suggests, an angiolipoma is composed of fat and vascular elements. The vascular

part appears as a hypodense lesion on T1-weighted imaging, whereas the fat part appears as a hypo intense lesion on T2-weighted imaging. After contrast enhancement with gadolinium, the vascular element appears as trabeculated bone tissue with a netted texture or like a reticular structure. On T1-weighted sequences, fat saturation can be seen clearly. However, the vessel part is hypointense and can manifest an obvious enhancement on T1-weighted sequences with contrast enhancement using gadolinium [6].

Because of the richness of vessels, a highly vascularized tumor can be predicted preoperatively from a T1 void signal [7]. The differential diagnosis includes lipomas and spinal vascular tumors, such as fistulas, metastases, meningiomas, and hemopathies. Angiography is helpful with the differential diagnosis, and it can be used to assess vascularization of the lesion, which is important for tumor removal using embolization [8]. On T2-weighted images, hyperintensity is a common finding of vascular content [7].

Surgical treatment was the only treatment performed in similar patients to ours with the symptom of compression, as shown in Table 1 [9-16]. All patients in previous studies showed good improvement postoperatively without recurrence. Most patients can achieve complete symptom relief. However, our patient had symptoms of paraplegia only a few days postoperatively. Because of the intra spinal origin and dumbbell shape, a neurologic tumor or malignancy was suspected. Thus, the differential diagnosis included a neurogenic tumor, ganglioma, hemangioma, hemangiopericytoma, or angiolipoma. However, the contrast-enhanced CT showed a strong enhancing lesion, which was considered hyper vascularity that was different from the presentation of a neurogenic lesion. Because of the worsening symptoms on postoperative month 3, an imaging study was performed and revealed a focal mass lesion at the spinal cord, although the prior mass disappeared. For this type of tumor, surgical treatment can be used to easily remove the tumor; nevertheless, some debris may remain without complete removal because of hyper vascularity of the tumor [17]. It has been reported that treatment of an infiltrating angiolipoma focuses on wide excision plus radiotherapy in the case of recurrence [18]. Thus, we administered regular radiotherapy in our patient because of her refractory lesion.

In our patient, the entire operation was performed without incident. After total laminectomies of T4-6 vertebrae with resection of the spinal tumor, she underwent VATS for resection of the paraspinal tumor. Some authors reported that thoracoscopic tumor removal after laminectomy is the most appropriate and reliable surgical treatment of a spinal dumbbell tumor. Because tumors are vessel-rich, some lesions may infiltrate the vertebral body. Furthermore, the residue may possibly become enlarged and cause the prior symptoms to recur. Our patient experienced partial clinical improvement with partial recovery within 1 week postoperatively. In the outpatient department follow-up, the patient experienced weakness of both lower limbs at 3 months postoperatively. Radiotherapy was administered because of the newly developed lesion in the spinal region, although surgical pathological results revealed that the prior lesion was benign.

Conclusion

Surgical resection should be followed by basic monitoring with a physical examination and imaging study even when the pathological results indicate a benign tumor. Furthermore, radiotherapy may be considered if the benign tumor has any malignant tendency.

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