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Lumbar Epidural Angiolipoma - Three Case Reports and Review of the Literature

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ABSTRACT

Purpose: A spinal angiolipoma is a rare benign tumor of the spinal axis. It is most common in the mid-thoracic spine and found predominantly in females. Pure lumbar epidural angiolipomas are very rare. We report three cases of epidural angiolipomas involving solely the lumbar region and review the associated literature.

Methods: We reviewed three cases of lumbar angiolipomas from two academic centers from 2012 to 2016. The tumor character was confirmed by the preoperative imaging study and pathological report. All had similar clinical manifestations, lower back discomfort or bilateral lower limb radicular symptoms and characteristic presentation in magnetic resonance imaging (MRI).

Results: From 2012 to 2016, there were three cases included for analysis, including two males and one female. All cases presented with symptomatic spinal pain, and MRI study showed the tumors were located at lumbar spine level. We performed laminectomy according to the locations of the tumors to achieve grossly total tumor excision or en bloc resection. All patients recovered uneventfully with fair post-operative neurological outcomes.

Conclusion: Spinal angiolipomas are rare benign tumors. For the patients with symptomatic presentation, MRI provides pre-operative clues for such tumor. En bloc resection should be considered in the symptomatic patients to relieve the compression of spinal cord or nerves.

Keywords: Spinal tumor, Spinal angiolipoma, En bloc resection

INTRODUCTION

Spinal angiolipomas are rare benign tumors, and they account for 0.14-1.2% of all spinal axis tumors [1]. The lesions are most common in the mid-thoracic spine and found predominantly in females [1,2]. The pathological presentation is mature adipose tissue and abnormal vessels [1,3-5]. The two subtypes of spinal angiolipomas are non-infiltrating and infiltrating [5]. We report three cases of epidural non-infiltrating angiolipomas, located solely at the lumbar spine, which is a rare occurrence, and discuss the clinical presentations, pathogenesis, radiography and management of the lesions.

CASE REPORT 1

A 79 year old man presented with a 2 year history of progressive pain, numbness and weakness in the bilateral lower extremities. The pain was improved by medication in the beginning but not resolved in recent months. Physical examination revealed intermittent claudication such that he was able to walk for 5 min or 200 m only. Neurological examination revealed bilateral positive straight leg rising of 45° and paresthesia below the L4 dermatome. The muscle strength of bilateral ankle dorsiflexion was 4/5. No urinary incontinence was noted during this period. The lesion was heterogeneous and slightly hyperintense in T1-weighted images (T1WI) and T2-weighted images (T2WI) of

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magnetic resonance imaging (MRI). The lesion was enhanced by post-contrast T1-weighted fast spin echo (FSE) **(Figures 1A-1C)**.

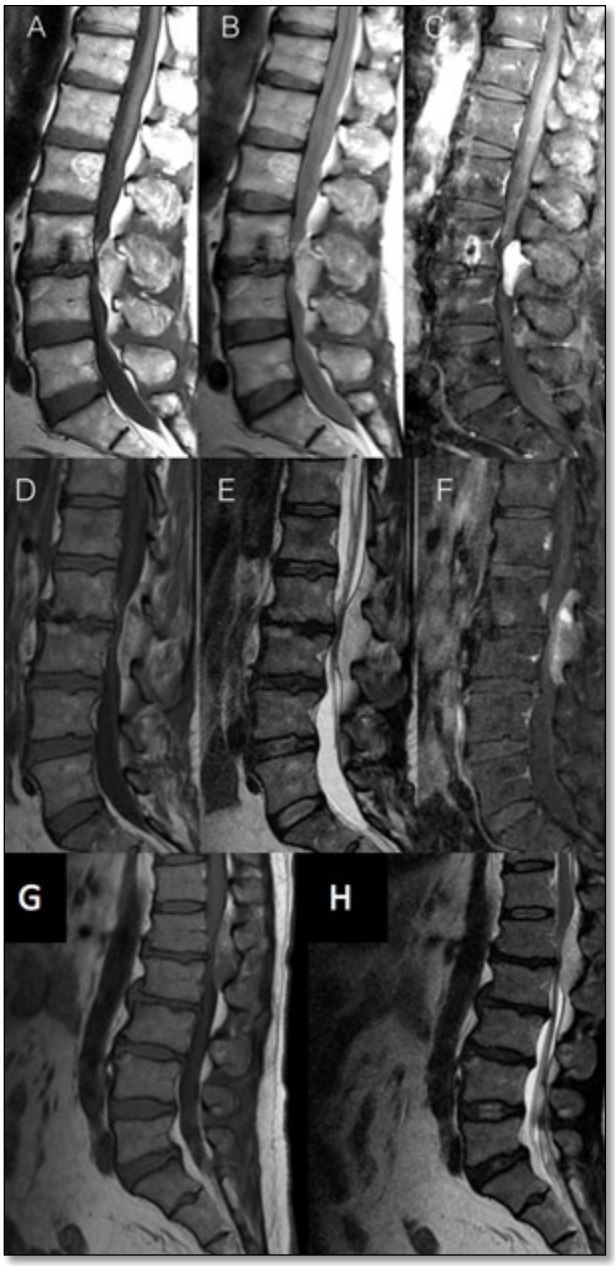

Figure 1. A non-contrast T1-weighted MRI showing a heterogeneously slightly hyper intense lesion in the L3-4 epidural space, causing spinal canal stenosis. B T2-weighted MRI showing slight hyper intensity. C Post-contrast T1-weighted MRI showing homogeneous enhancement. D Non-contrast T1-weighted MRI showing a heterogeneous iso- to hyper-intense lesion at L2-3 posterior epidural space. E T2-weighted MRI showing homogeneous hyper intensity, causing severe spinal canal stenosis. F Post-contrast T1-weighted MRI showed heterogeneous enhancement. G Non-contrast T1-weighted MRI showing a heterogeneous iso- to hyper-intense lesion in the L1-2 epidural space. H T2-weighted MRI showing hyper intensity.

We performed L2-4 laminectomy and tumor excision. The operative findings revealed severe L2-4 thecal sac compression by a well-demarcated neoplasm of about 2.5 × 1.5 cm in size. The pathological presentation was an angiolipoma composed of a collection of mature adipose tissue with varying numbers of vascular structures **(Figure 2)**. The patient recovered well and had no further neurological complaints. There was no recurrence on follow-up MRI 2 years after the operation.

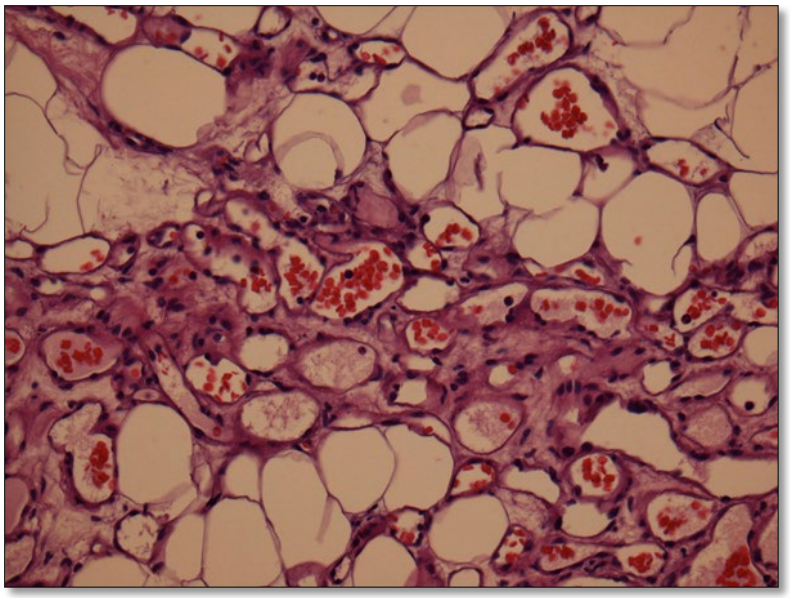

Figure 2. Microscopically, the section showed mature adipose tissue mixed with multiple branching and blood-filled vessels.

CASE REPORT 2

A 45 year old female had complained of progressive bilateral lateral thigh pain and numbness for two years prior to her visit to the hospital. Progressive claudication had developed rapidly in recent months and she could not walk more than 100 m when she visited the clinic. There was no urinary or stool incontinence. MRI showed a fusiform mass of about 53 mm in cranio-caudal diameter, with extension to the bilateral neural foramen, in the posterior epidural space of the L2-3 level, with heterogeneous but predominant high T1W and T2W signal intensities. There was enhancement in the post-contrast series **(Figures 1D-1F)**.

We performed L2 laminectomy for en bloc tumor excision. One adipose-like, well-demarcated neoplasm with profound vascularity was noted in the epidural space of L2-3 **(Figure 3)**. The neoplasm was located on the dorsal side of the thecal sac with extension to the bilateral neural foramen of L2-3. The thecal sac and bilateral L2 roots were deviated anteriorly. The pathology indicated angiolipoma as well **(Figure 4)**.

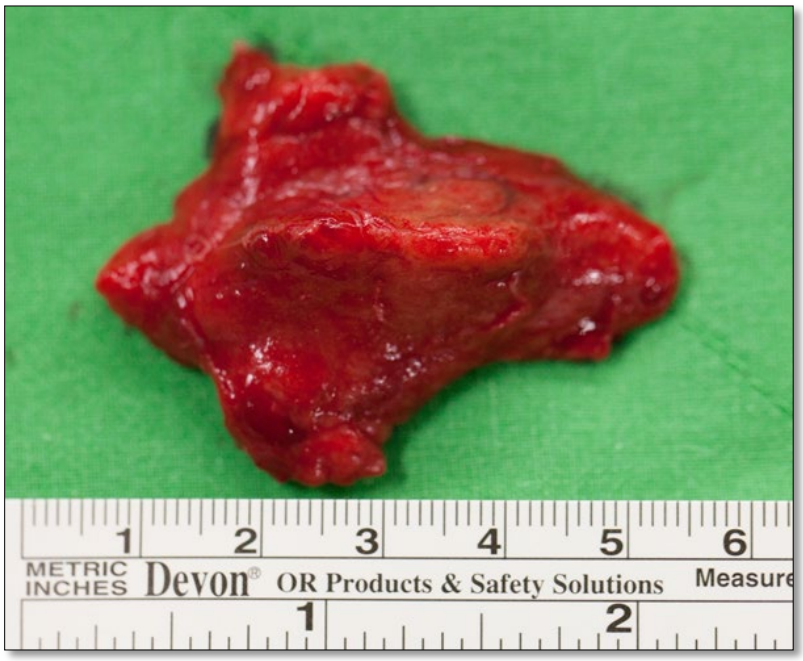

Figure 3. Grossly, the tumor was a yellowish to reddish, adipose-like, well demarcated tumor of about 5 cm diameter.

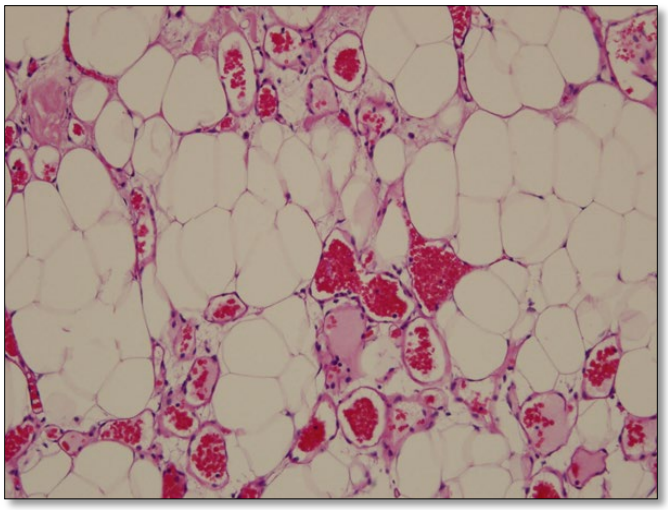

Figure 4. Microscopically, the section also showed mature adipose tissue with hypervascularity.

CASE REPORT 3

A 53 year old man presented with severe lower back soreness and pain with bilateral lower leg numbness for more than six months. Later intermittent claudication caused him to seek medical assistance in the clinic. MRI showed a 4 × 1.5 cm epidural lesion at L1-2 causing severe thecal sac compression, which was to hyper intense in TIWI and hyperintense in T2WI **(Figures 1G and 1H)**. He received conservative treatment but the effect was limited. We performed T12-L2 laminectomy for tumor excision. Operative findings showed a 4 × 1.5 × 1 cm yellowish and elastic-soft tumor in the epidural space with thecal sac compression. Post-operatively, he recovered well. The pathology represented typical characteristics of angiolipomas **(Figure 5)**.

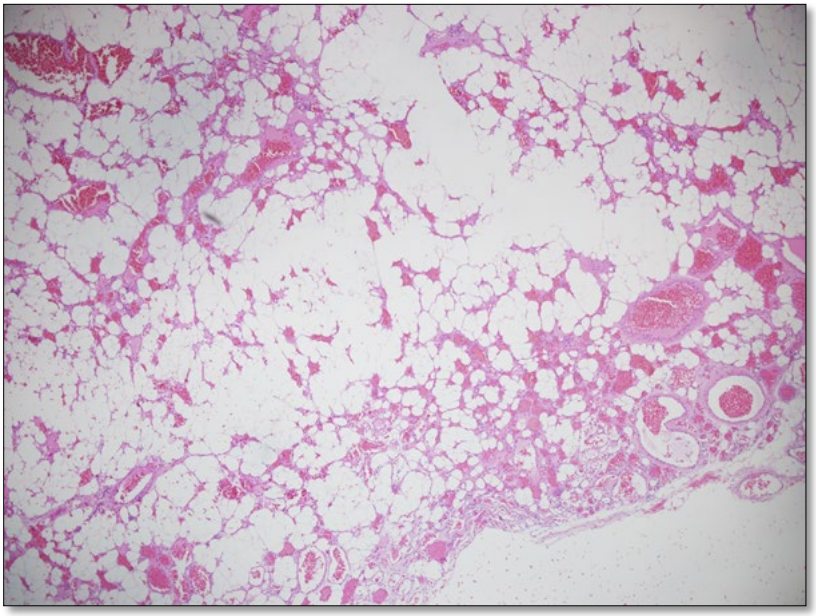

Figure 5. Microscopically, the section showed mature adipose tissue with hypervascularity and some internal hemorrhage.

DISCUSSION

Angiolipomas are benign tumors that usually appear in the forearm, trunk or neck and rarely in the spinal canal [6]. The first case of spinal angiolipoma was reported by Berenbruch in 1890 [7] in an autopsy of a 16 year old boy after an unsuccessful operation. Spinal angiolipomas are predominantly located in the mid-thoracic region and are more common in females. Lumbar spinal angiolipomas are extremely rare and the first was reported by Kasper and Cowan in 1931 [8].

The typical pathology presentation of an angiolipoma is a benign neoplasm composed of mature adipose tissue and

abnormal vessels [9]. It may be classified within the spectrum between lipoma and hemangioma [10]. The ratio of fat to vessels is variable, ranging from 1:3 to 2:3 [11]. In 1974, Lin and Lin [5] classified spinal angiolipomas into two types: non-infiltrating and infiltrating. The non-infiltrating subtype is more common. Both subtypes are benign tumors; however, the infiltrating subtype invades surrounding tissues, such as the vertebral body. Non-infiltrating lesions are almost always located in the posterior epidural space, while infiltrating lesions appear mostly in the anterior or anterolateral space with vertebral body and pedicle invasion [12,13].

Spinal angiolipomas cause a slow progressive clinical neurological presentation according to the level of the spinal canal compromised [1]. Common subjective complaints include back pain and progressive motor and sensory deficits below the affected spine level, with possible progression to sphincter dysfunction in the late stage [14]. The symptoms tend to develop slowly over months to years. However, sudden deterioration can occur as a result of tumor thrombosis or hemorrhage, as reported in two different cases by Labram et al. [15] and Anson et al. [16] Pregnancy is an aggravating factor, and pregnancy termination may cause regression of symptoms, as reported by Cull et al. [17] for two cases presenting paraparesis during pregnancy but remission after parturition. Pregnancy may interfere with the drainage of venous blood and increase epidural venous pressure due to compression of abdominal and major veins, resulting in increased extracellular fluid, and the forceful Valsalva maneuvers associated with vaginal delivery may acutely aggravate symptoms [18]. Other suggested possible factors, including vascular steal, could be a cause of spinal cord ischemia, as well as pulsatile compression on the adjacent cord because of its high vascularity [2].

In most cases, spine radiography is useful in detecting vertebral bony destruction; i.e., erosion of the pedicles or vertebral body, trabeculation, widened pedicles and foraminal widening [19]. Computed tomography will show a hypodense signal and can help in the assessment of the involved bony lesion. Magnetic resonance imaging (MRI), considered the best modality for diagnosing spinal angiolipomas, typically shows hyperintense signals in a T1-weighted image without contrast, owing to their fatty content. Provenzale and Mclendon [18] showed that large hypointense foci observed within spinal angiolipomas on non-contrast T1-weighted images are correlated with increased vascularity and most lesions are enhanced with gadolinium administration. T2-weighted imaging can be variable but is usually hyperintense.

Total surgical resection of the tumor is an ideal treatment, depending on the location [20]. The non-infiltrating type is located mainly in the posterior epidural space, so it can be removed through posterior laminectomy approach. The infiltrating type is located mainly anterior to the spinal canal and it is likely to affect the vertebral body, so it can be approached through anterior, lateral, or mixed approach methods [21] and sometimes, instrumentation of the involved vertebral body may be required.

The prognosis is excellent for both non-infiltrating and infiltrating types. Although the infiltrating type is difficult to remove completely with surgery, adjuvant or neoadjuvant radiotherapy is not recommended, even when complete removal cannot be achieved, because of the benign pathology.

CONCLUSION

Spinal angiolipomas are rare benign tumors that cause slow progressive neurological deficit according to the level of the spinal cord affected. MRI is the best modality for diagnosing spinal angiolipomas. MRI shows heterogeneous hypo intensity in T1-weighted images and hyperintensity in T2-weighted images, and most lesions are enhanced with contrast administration. Total surgical resection is an ideal treatment. Whether or not resection is complete, no adjuvant or neoadjuvant radiotherapy is needed. The prognosis of both types of spinal angiolipomas is good.

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