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Case Report

A rare case report of sacral epidural angiolipoma: Diagnosis and treatment

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ABSTRACT

Background: Spinal angiolipomas are rare benign tumors composed of mature adipose tissue and anomalous vascular channels. The sacral localization is extremely rare. To the best of our knowledge, there have been only two cases reported in the literature. Herein, we present an additional case of sacral angiolipoma.

Case Description: We present a case of a 54-year-old woman who presented with the right lumbosciatica and distal crural weakness. Spinal MRI showed an epidural lesion at the level of L5-S1 extending to the first right sacral foramen. The patient had total resection of the tumor and the histological study concluded to an angiolipoma. The patients' neurologic symptoms improved postoperatively and follow-up revealed no signs of tumor recurrence.

Conclusion: Despite the rarity of sacral localization in angiolipomas, it is a diagnosis to be considered in the case of an epidural tumor with foraminal extension. Magnetic resonance imaging is important for detecting and characterizing spinal angiolipomas despite diagnosis is not always obvious. After surgical removal, the functional prognosis is generally favorable.

Keywords: Benign tumor, Epidural, Sacral, Spinal angiolipoma, Surgery

INTRODUCTION

Spinal angiolipomas are benign neoplasm composed of mature lipocytes and abnormal blood vessels. They account for only 0.04–1.2% of all spinal tumors and are predominantly found in the epidural space, where they represent 2–3% of spinal tumors.[8] The majority of spinal angiolipomas are located in the mid-thoracic region or more rarely in cervical and lumbar region, sacral localization had been reported in only 0.8% of cases,[11] with, to the best of our knowledge, only two cases reported in the literature.[1,3]

CASE REPORT

This is a 54-year-old woman with no medical history who was referred to our department by family physician for progressively worsening of the right lumbosciatica associated with urinary urgency for 2 years and radicular claudication for 1 year, reducing the walking perimeter to 500 m.

On examination, she had paresis of plantar and dorsal flexion (muscle strength 2/5) associated with a poorly systematized hypoesthesia of the right lower limb. She did not have saddle anesthesia. Spinal magnetic resonance imaging (MRI) showed an epidural tissular mass at the level of L5-S1 extending to the first right sacral foramen, which was enlarged without bone lysis with extension to the presacral soft tissues, hypointense on T1-weighted image, slightly hyperintense on T2-weighted image with intense and homogeneous contrast. This lesion was suggestive of an S1 schwannoma [Figure 1].

The patient was operated in the genupectoral position. A median incision from L4 to S3 was performed followed by a right unilateral approach and hemilaminectomy of L5, S1, and S2 until exposure of the S3 foramen. A reddish hemorrhagic epidural lesion was found, pushing back the dural sac and the right S2 nerve. A careful dissection of the tumor was performed following a cleavage plane with the nerve. The resection was macroscopically complete.

The histological study concluded to an angiolipoma [Figure 2].

Postoperatively, the patient showed an improvement of her motor deficit, which was rated at 3+/5, and she underwent motor rehabilitation, which allowed her to walk normally again.

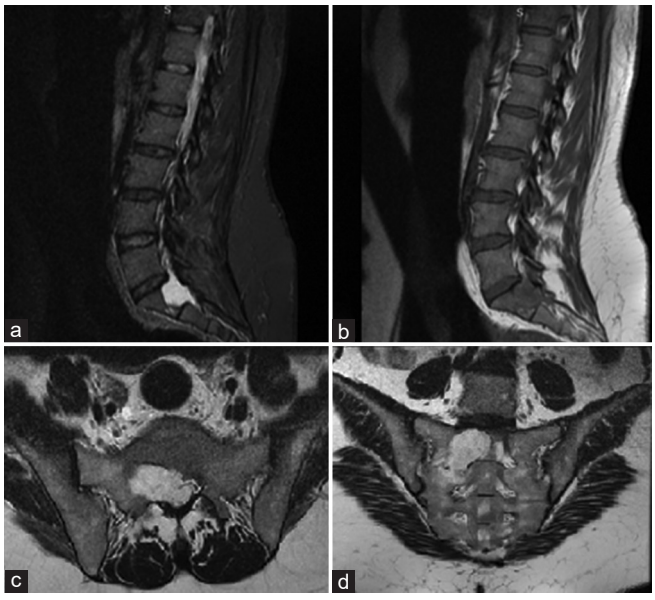

Figure 1: Magnetic resonance imaging of the lumbosacral spine: sagittal gadolinium T1-weighted with fat suppression (a), T2-weighted (b), axial T2-weighted (c), and coronal T2-weighted (d) demonstrated intracanal tissular process regarding L5-S1 extending to the level of the first right sacral foramen, which was enlarged without bone lysis with extension to the presacral soft tissues, in T1 hyposignal, moderate T2 hypersignal with intense and homogeneous contrast.

A 2-year follow-up revealed no signs of tumor recurrence.

DISCUSSION

In the review of Preul *et al.*,[8] spinal angiolipomas accounted for 0.14–1.2% of all spinal tumors and 2–3% of extradural spinal tumors. The mean age of patients was 41.6 years.[8]

The majority of spinal angiolipomas arise in women throughout their peri- or postmenopausal period. According to a literature review of 108 spinal angiolipomas in 2009, the majority of the tumors (86%) were located in the thoracic spine, followed by 12 (11%) in the lumbar spine, 2 (1.85%) in the cervical spine, and one in the sacrum.[1,11] An additional case of sacral angiolipoma was reported in 2020 and was associated with tight filum terminal, spina bifida, and spinal arteriovenous fistula.[3] Therefore, we consider this case the third case of sacral angiolipoma in the literature [Table 1].

Spinal angiolipoma usually presents as a slow-growing mass causing compression of the spinal cord. Clinically, early symptoms are typically numbness in the lower extremities, back discomfort, and leg weakness,[8] as were the case with our patient. These symptoms last on average for more than a year before diagnosis.[7] A few cases of rapid onset of symptoms or neurological impairment have been described.[4] Symptoms can be worsened in case of weight gain and pregnancy due to changes in the tumor mass and volume.[8,12]

Angiolipomas are mesenchymal neoplasms made up of mature adipose tissue and aberrant blood arteries. These lesions are considered a subgroup of lipomas.[5,6]

In some aspects, spinal angiolipomas differ from spinal lipomas. The former is more common in adults, almost always seen in the epidural space, and is unrelated to congenital myelovertebral abnormalities. The latter mainly

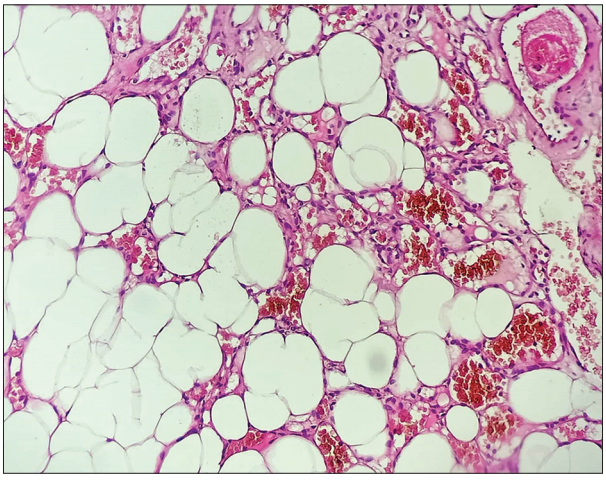

Figure 2: Epidural angiolipoma. HE ×200. The tumor is composed of both vascular structures with some thrombi (up right) and regular adipocytes.

Table 1: Summary of the previous reported cases of sacral angiolipoma with the present case

Author and year	Age	Sex	Clinical features	MRI features	Treatment	Outcome	Recurrence
Souto *et al*.,[11] 2003	46 years old	Female	Lombosciatica and paresis of the planter flexion	Ventral extradural mass from L4 to S2, with erosion of the sacrum	L4 and L5 laminectomy with total resection of the tumor	Postoperative complete recover	No recurrence after 18 months of follow-up
Iampreechakul *et al*.,[3] 2020	55 years old	Female	Paraparesis and paresthesias with a slow-growing mass at the left buttock since birth	Extradural mass at the level of S3-S4, with tight filum terminale and spina bifida coexisting with spinal arteriovenous fistula	Endovascular treatment, after 6 months surgical removal of extradural mass containing the AVF, and subsequent release of the tight filum	Gradual recover after 2 years of follow-up	No recurrence after 2 years of follow-up
Our case	54 years old	Female	Lombosciatica with urinary urgency radicular claudication paresis of planter and dorsal flexion	Extradural tissular mass at the level of L5-S1 extending to the first right sacral foramen	Unilateral hemilaminectomy of L5, S1, and S2 and complete removal of the tumor	Gradual recover after 2 years of follow-up	No recurrence after 2 years of follow-up

appears in childhood and is found in the epi- and intra-dural regions; majority are associated to congenital myelovertebral abnormalities.[2]

Noninfiltrating and infiltrating angiolipomas are the two forms of spinal angiolipomas. The majority of these tumors, including our case, are noninfiltrating.[10] The infiltrating type is encapsulated and contains areas with a predominant vascular component.[9] The infiltrating form usually arises in ventral locations within the thoracic and lumbar portion of the spinal column, and these lesions typically invade vertebral bodies and pedicles.

Extradural components of angiolipomas are isointense or hyperintense in T1-weighted images, most likely due to the fat component, and usually hyperintense in T2-weighted images in MRI studies. These areas show an early enhancement following gadolinium administration and are thought to be the vascular component.[8,9]

Surgical resection of the lesion through anterior approach or posterior laminectomy is the gold standard treatment modality.[13] We used posterior approach to access the tumor in our case and we attained total tumor resection.

The first-choice treatment is total surgical removal of the tumor. However, it may be more difficult in the infiltrating type than in the noninfiltrating type. Nevertheless, even with partial removal, most patients have a good prognosis because tumors are usually slow growing and do not progress to malignancy.[13] Furthermore, recurrence is extremely rare in spinal angiolipoma. Adjuvant radiation should not be used on patients with this benign entity even in the infiltrating type.[1]

CONCLUSION

Spinal angiolipomas are rare benign tumors of angiomatous and lipomatous tissue. Sacral location is exceptional. The diagnosis can be challenging radiologically since they may mimic the other spinal lesions. Total resection may not be achievable in some cases. We attained total resection in our case without any further neurological complication.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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