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OLGU SUNUMU / *CASE REPORT*

THORACIC SPINAL ANGIOLIPOMA: A CASE REPORT

Serdar ÖZGEN[1], Deniz KONYA[1], Özlem KURTKAYA[2], M. Necmettin PAMİR[1]

SUMMARY

The objective of this report is to describe a case of spinal angiolipoma in the thoracic region. A 61-year-old woman presented to the outpatient clinic with a 6-month history of back pain. Magnetic resonance imaging showed an extradural mass at T8 level that was isointense on T1-weighted images and hyperintense on T2-weighted images. Surgery was performed, and T8 laminectomy revealed an extradural tumor. The mass was totally excised. Histopathological examination showed that the neoplasm was composed of mature adipose tissue and blood vessels. The diagnosis was spinal angiolipoma. At follow-up 36 months later, there was no evidence of recurrence. Spinal angiolipomas are very rare, but should always be considered in the differential diagnosis for any spinal cord mass. Surgical excision is almost always curative.

Keywords: *Angiolipoma, spinal tumor, excision.*

ÖZET

Bu yazıda torakal bölge yerleşimli bir spinal anjiolipoma olgusu sunulmaktadır. Polikliniğe altı aydır devam eden sırt ağrısıyla başvuran altmışbir yaşında bir kadının magnetik rezonans incelemesi yapıldı ve T8 seviyesinde T1 ağırlıklı kesitlerde izointens, T2 ağırlıklı kesitlerde hiperintens ekstradural kitle saptandı. Laminektomi yapıldığında dura dışı yerleşim gösteren tümör gözlendi. Kitle tümüyle eksize edildi. Histopatolojik incelemede tümör dokusunun matür adipoz doku ve kan damarlarından oluştuğu gözlendi ve spinal anjiolipoma tanısı konuldu. Otuzaltı ay sonraki izleminde nüks gözlenmedi. Spinal anjiolipomalar nadir olmakla beraber herhangi bir spinal kord kitlesi ayırıcı tanısında akılda tutulmalıdır. Cerrahi eksizyon hemen her zaman tedavi sağlar.

Anahtar Kelimeler: *Anjiolipoma, spinal tümör, eksizyon.*

[1] *Department of Neurosurgery, Marmara University Faculty of Medicine and*
[2] *Marmara University Neurological Sciences Institute*. Istanbul. Turkey*

INTRODUCTION

Spinal angiolipomas are very unusual extradural neoplasms that account for approximately 0.14-1.2 % of all spinal axis tumors[3,5,6,8,11,15,20]. Since the first spinal angiolipoma tumor case was described in 1890, 84 spinal angiolipomas cases have been reported to date[4,25]. Angiolipomas are benign mesenchymal neoplasms that are composed of mature adipose tissue and contain abnormal blood vessels ranging from capillary size to arterial size[14]. Spinal angiolipoma may occur anywhere in the spine[4,5,6,8,16,28]. In this report, we present a case of spinal angiolipoma of the thoracic region and discuss the relevant literature.

CASE REPORT

61-year-old woman presented to the outpatient clinic with the complaint of back pain for about 8 months. Her neurological examination was unremarkable, and she was referred for physical therapy to address the chronic pain. Three months later, she was readmitted to hospital with the same complaint. Neurological examination revealed intact motor function but sensory deficits below T8 level on the patient's left side. Her deep tendon reflexes were all normal and no pathologic refiexes were detected.

Routine plain x-rays of the thoracic region showed enlargement of T8 foramen. Computed Tomography (CT) imaging show the lesion was marked to invade and to dilate the left neural foramen at the T8 level and the lesion showed contrast enhancement (Fig 1).

Magnetic resonance imaging (MRI) revealed an extradural mass at T8 level that was isointense on T1-weighted and hyperintense on T2-weighted images (Fig 2). The lesion had partially eroded the left interior part of T8 vertebral body and extended into the lefi T8 neural foramen.

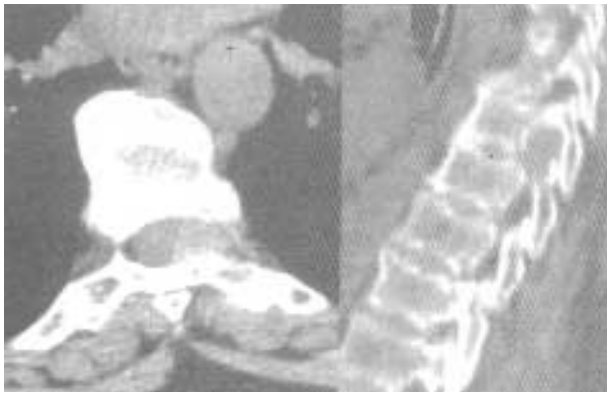

Figure 1. Left: The lesion had been enhanced after contrast injection at thoracic CT. Right The lesion dilated the left neural foramen at the T8 level.

Figure 2. The extradural mass at T8 level was isointense on T1-weighted (left) and hyperintense on T2-weighted images (right).

The mass showed marked enhancement after contrast injection. The diagnosis was spinal tumor, and surgery was performed.

Surgery: The patient was operated in prone position under general anesthesia. The procedure was done using a posterior approach through a T7-9 midline incision. Laminectomy was performed at T8, and this revealed a 25x 18-mm extradural mass that had no capsule and did not involve the dura. Total excision was achieved.

Pathological Examination: The tissue was fixed in 10% buffered formalin and embedded in paraffin. Serial sections were cut from the lesion. All sections were stained with routine haematoxylin-eosin (H&E) stain (Fig 3).

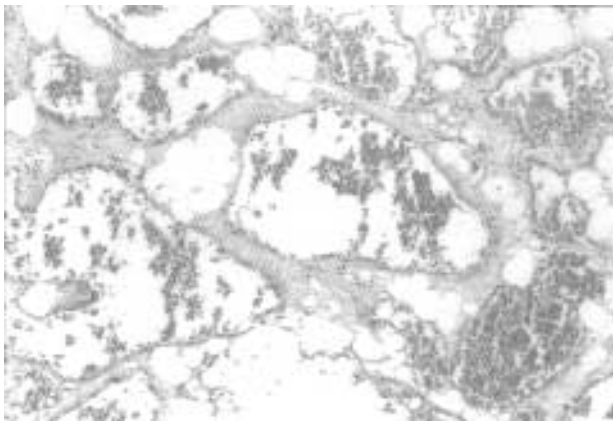

Figure 3. The lesion consisting of a mixture of mature adipocytes and large branching, blood pilled cavemous vascular channels (H&E x 200).

Immunohistochemical staining was also done using the peroxidase-antiperoxidase technique with monadanal antibody against desmin (Clone 33, Biogenex, CA). Examination of sections showed that the mass was composed of mature adipose tissue and blood vessels. Most of the vessels were thick-walled medium-sized vascular channels with elastic lamina. There were also a few mature cells that varied in size. No pleomorphism, mitotic activity, or necrosis was noted. Immunostaining revealed cytoplasmic immunopositivity for desmin in occasional mature smooth muscle cells of the thick vessels. The histopathologic diagnosis was angiolipoma.

The postoperative period was uneventful and the patient was discharged 5 days after surgery. Postoperative MRI confirmed complete removal of the tumor. At follow-up 36 months later, there was no evidence of recurrence.

DISCUSSION

Spinal epidural angiolipomas are benign, wellcircumscribed tumors that usually arise in the thoracic region[3,7,8,15,28]. The majority of these tumors are non-infiltrating and do not involve the surrounding tissue[18,21]. However, some infiltrating forms have also been described, and invasion of vertebral bodies and the thoracic cavity has been documented[9,10,23]. Several studies showed that most non-infiltrating angiolipomas are situated in the dorsal portion of the epidural space[6,15,21] Our patient's tumor was located in the thoracic region and had partially eroded the left interior part of T8 vertebral body and extended into the left T8 neural foramen.

There is no consensus in the literature regarding the pathogenesis of spinal angiolipoma. Several theories have been postulated. Ehni and Love have suggested that this neoplasm originates from mesenchymal progenitor cells that have the potential to differentiate into angiomatous and lipomatous tissue[3]. Others have proposed that this tumor is congenital malformation or a true hematoma [12,20]. Shibata et al. suggest that the late inclusion of mature adipocyte causes spinal lipoma, which commonly occurs in the last closure site of the embryonic neural arc, wheras the early inclusion of the pluripotential stem cell into the neural arch cause spinal angiolipoma in the thoracic region, corresponding to the fim ossification site of the neural area[22]. Therefore, it is likely that the more immature pluripotential stem cell, which has more infiltrative characteristic, is included in this area. This could explain the infiltrative behavior of some angiolipomas.

Some authors have classified angiolipomas as a subgroup of lipomas[2,19,26]. Spinal angiolipomas differ from spinal lipomas in several ways. The spinal angiolipomas usually appear in adults, are almost always located in the epidural space, and lack associated congenital myelovertebral malformations. Spinal angiolipomas are frequently confused with spinal angiomyolipomas, Angiomyolipomas of the spine are very unusual and the diagnosis is based on predominance of smooth muscle in the vascular walls.

Clinically, patients with spinal angiolipomas usually present with symptoms related to spinal cord and root compression[2,7]. As evident in our case, angiolipomas in the lumbar region tend to present with radicular symptoms. Typically, the symptoms progress gradually because the tumor is benign and slow growing. Sudden onset or worsening of neurological symptoms occurs when there is a sudden increase in tumor size due to intratumoral thrombosis, hemorrhage or steal phenomenon[1,16,21]. Öge and colleagues suggested that increased blood volume due to administration of coronary vasodilator drugs in 72 years old patient might also cause sudden onset of neurological deficits[16].

Plain radiographs are usually normal in patients with spinal angiolipoma; however, these films may reveal non-specific erosion of the vertebral body, trabeculation of vertebral body and enlargement of the vertebral foramen due to erosion. Not surprisingly, plain x-ray abnormalities are more frequent with the infiltrative form. On CT without contrast, spinal angiolipomas appear hypodense and can be misdiagnosed as epidural fat tissue[27]. When contrast is administered, the sean may show parti al lesion enhancement. MRI is the most valuable radiological modality for diagnosing spinal angiolipomas. These lesions are usually hyperintense or isointense on T1- and hyperintense on T2-weighted images[5,24,27]. Compared to epidural fat tissue, spinal angiolipomas are hypointense[13]. T2-weighted imaging is less sensitive than T-1 technique and the diagnosis should always be made based on homogeneous enhancement of the lesion with contrast administration.

The treatment of choice for this neoplasm is surgery. Most non-infiltrating spinal angiolipomas are located in the dorsal portion of the epidural space, and can thus be removed via posterior laminectomy[3,4,8,14,17]. However, half of the infiltrative cases reported to date were located ventrally or ventrolaterally in the spinal canal, so they required anterior and anterolateral decompressive surgery as opposed to simple laminectomy[23,26]. Labram and co-workers described a huge angiolipoma that extended from the spinal canal into thoracic cavity. This tumor was treated with incomplete resection via a combined anterior and posterior approach[11]. Complete surgical excision is believed to be curative in most cases of spinal angiolipoma, but Preul documented one of the three cases in their article, of recurrence 12 years after complete removal (20). This patient was re-operated with good clinical results. Weight gain and pregnancy are reported to aggravate symptoms in cases of incomplete resection. Adjuvant treatment with chemotherapy or radiotherapy is not recommended for these benign lesions, even when only incomplete removal is achieved[4,20].

In conclusion, spinal angiolipoma is very rare lesion but should always be considered in the differential diagnosis of any spinal cord mass. Total cure can be achieved with surgical excision.

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Correspondence Address:
Dr. Deniz Konya, Marmara Üniversitesi Hastanesi
Nöroşirürji Anabilim Dalı
Tophanelioğlu Caddesi, Altunizade
İstanbul, Turkey
Tel: +902163264559, 05337351293
Fax: +90 216 3057961
E-mail: drdkonya@hotmail.com