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Noninfiltrating extradural angiolipoma of Spine: A case report

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Abstract

Angiolipomas are found most commonly in the subcutaneous tissues of trunk and extremities. They are also found in other locations. Angiolipoma of spine is a rare benign tumour consisting of variable amount of mature adipose and abnormal vascular tissue. They are found mostly as extradural tumor in spine but intramedullary and intracranial origins have also been described. They constitute 0.14-1.2 percent of all spine tumors and 2.3 percent of all epidural tumors. Authors report a case of 52 year old man who presented with progressive weakness in bilateral lower limbs with difficulty in walking for the last 6 months. Hypoaesthesia was present below L1. He had difficulty in controlling urine for the last 15 days.MRI showed lesion extending from D8 to D10, was heterogeneous hyperintense on both T1,T2 and enhanced on contrast uptake. D8 to D10 laminectomy was done and tumor was removed and sent for histopathology which showed the tumor to be angiolipoma. There was no evidence of recurrence 36 months later after surgery.

Keywords

extradural; noninfiltrating; angiolipoma; spine tumors

Introduction

Angiolipomas are common subcutaneous benign lesions; in contrast, spinal angiolipoma is a specific and an uncommon clinicopathological entity. These benign tumors are composed of varying proportions of mature fat cells and abnormal capillary, sinusoidal, venous, or arterial vascular elements. Complete surgical removal is the treatment of choice for these tumors. It is important to identify it early and surgically treat this entity as prognosis for both infiltrating and noninfiltrating types is excellent even after incomplete removal. This report details the clinical, radiological, and histological characteristics of a middle aged man who presented with paraparesis and underwent surgical excision of an epidural angiolipoma.

Case Report

This 52 year old gentleman presented to us with progressively increasing weakness in bilateral lower limbs with bladder involvement. MRI was done which showed a heterogeneous hyperintense lesion on both T1 and T2 (Fig1 & Fig2) which enhanced on contrast uptake (Fig3) extending from D8 to D10. Patient was taken up for surgery and laminectomy was done from D8 to D10 with surgical removal of the mass. Tumor was sent for histopathology which revealed the tumor to be angiolipoma (Fig4). It was

reported on microscopic examination to have a mixture of mature fatty cells and abundant vascular components consisting of a mixed pattern of capillary, cavernous, and venous type. Patient was started on aggressive rehabilitation and physiotherapy after the surgery. He showed improvement in his neurological status and regained his sphincter control. Patient had returned to work 1 year after the surgery, and remains symptom free at 3 years follow up.

Discussion

Spinal Angiolipoma is a rare clinico - pathological condition; on reviewing the available literature, the first described case of spinal angiolipoma was published in 1890 in a doctoral thesis about a 16-year-old boy with numerous cutaneous lipomas who developed a progressive paraparesis with upper motor neuron signs and hyperreflexia. The tumors were removed, but the patient did not survive beyond 4 hours post surgery. The postmortem autopsy examination revealed a thoracic cutaneous lipoma infiltrating the spinal canal with a predominantly vascular component and compressing the spinal cord from C6 to T5 [1].

About a decade later in 1901, a primary spinal angiolipoma was reported by Liebscher for the first time [2]. Howard and Helwig established spinal angiolipoma as a distinct anatopathological entity [3] in 1960. Most of these tumors are located in middorsal region of spine. More than 120 cases have been described in literature till now [4]. Noninfiltrating constitute the majority of these tumors, though infiltrating tumors have also been described [5].

Spinal angiolipma contributes to 3% of extradural spinal tumors and 24% of spinal lipomas. It is a rare benign tumor with incidence of .14 to 1.2 percent of all spine axis tumors. Females outnumber males by ratio of 1.5 [4]. Most of the patients are in fourth and fifth decades of life. Almost all of the noninfiltrating tumors occupy the posterior epidural space while infiltrating tumors invade body along with posterior elements and can occupy anterior epidural space.In rare circumstances, angiolipomas occur intracranially. Noninfiltrating and infiltrating angiolipomas are considered histologically benign, although infiltrating angiolipomas have a locally aggressive nature and can erode the local bone, muscle, and neural and fibrocollagenous tissues [5].

Ninety percent of these tumors are located in dorsal spine. Even though its a very slow growing tumor, the anatomical location of these tumors predisposes to early compression. Progressive motor weakness and other symptoms related to spinal cord compression are the initial symptoms noted by most of the patients. The symptomatology progresses over months; and there might be a clinical course consisting of multiple relapses mimicking multiple sclerosis. Xray is mostly negative, but might in some cases show the mass effect by pedicle erosions and canal widening, whereas infiltrating tumors may cause vertebral body trabeculations. Myelogram may reveal cord compression. On contrast CT an angiolipoma appears as a hypodense mass relative to the spinal cord. Some tumors are isodense, which might signify high degree of vascularization. FDG positron emission tomography may also be used for diagnosis.

MRI is the preferred modality and the gold standard in the diagnosis and treatment of spinal angiolipoma. Angiolipomas are usually hyperintense on T1 and T2 and enhance on contrast although the picture may vary depending on the composition of adipose tissue and vascularity of tumor. They are

usually heterogeneous on MRI due to two different tissue compositions. Vascularity of the tumor can be judged by areas of hypointensity on T1 section [6]. Gala and Aswani [12] in their description of MR imaging stated that spinal angiolipoma were usually noninfiltrating, and occured in the posterior epidural space.[13] Infiltrating spinal angiolipoma, though had a tendency to invade adjacent structures including vertebral bodies and might extend beyond the neural foramen.[13,14] Contrast-enhanced MRI was the best modality to diagnose these lesions as well as to differentiate spinal angiolipoma from epidural lipomatosis. On MRI, Hu et al.[11] divided spinal angiolipoma into two types based on the ratio of fat to vessels. Type 1lesions were predominantly fatty, and hence appeared hyperintense on T1W and T2W images. Blood vessels within the lesion appeared as T1 hypointense strips or spots, which showed post-contrast enhancement. In Type 2 lesions, vascular component formed more than 50% of the lesion and appeared heterogenous on T1W and T2W images and most of the lesion showed intense enhancement. It was also described that the absence of flow voids in spinal angiolipoma distinguished them from high flow arteriovenous malformations and capillary hemangiomas. In addition, the margins of capillary hemangioma with adjacent fat (which was not part of tumor) were clear as compared to spinal angiolipoma whose fat and vascular interface was irregular and vague [15].

The histogenesis of the angiolipoma is still controversial. It is widely accepted that angiolipomas arise from primitive mesenchymal tissues and they are intermediate between hemangioma and lipoma at either spectrum [7].

Pregnancy has been associated with sudden aggravation of symptoms in female patients. The mechanism hypothesized to cause the enlargement of tumor is venous engorgement because of compression of pelvic and abdominal veins, vascular steal phenomenon which can cause ischemia of neural tissues and obesity in pregnancy which can increase the adipose tissue and can cause tumor enlargement [8]. Tumor thrombosis and haemorrhage has been reported to cause sudden neural deterioration in patients with spinal angiolipoma [9].

The biological behavior of the infiltrating and noninfiltrating spinal angiolipoma implicates a different treatment approach. The noninfiltrating type is often encapsulated, and complete removal of the tumor along with the capsule is possible, and might assure of low recurrence rates and better recovery from the neurological symptoms. In cases of infiltrating type, resection (which might be piecemeal) followed by adjuvant radiotherapy could be considered, though some authors have described no role of any adjuvant treatment [4]. A larger lesion or a tumor with a very high vascularity might require a preoperative angiography and embolisation prior to surgical removal [5].

Noninfiltrating angiolipomas are easily accessible by posterior laminectomy approach because of their location in posterior epidural space. Infiltrating tumors can be removed best with anterolateral approach. [16] Guzey et al. stated that a total resection could be more difficult for infiltrating types, however, most patients still had a good prognosis even with subtotal removal as these lesions were slowly growing and did not undergo malignant transformation.[16] Our case was a non infiltrating variety, and a piecemeal resection of the tumor was done. A moderately high amount of bleeding was encountered while dissection and removal of the tumor mass, which spontaneously reduced once all the lesion was removed completely. The patient has completely recovered neurologically, and has been free from any recurrence at 3 yrs followup with no residual symptomatology. Only one case of recurrence after 12 yrs

of surgery has been reported till date in literature [10] which again shows that this tumor is associated with a good prognosis following surgical removal. We also support Zevgaridis et al who have stated thatthe rare nature of this condition does not allow the formulation of treatment guidelines for this condition [17].

Figures

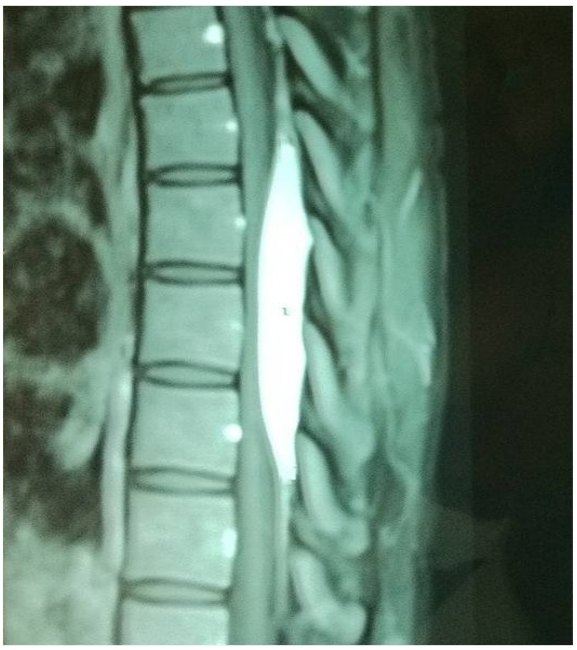

Figure 1: showing T2 saggital section showing hyperintense tumor extending from D8 to D10.

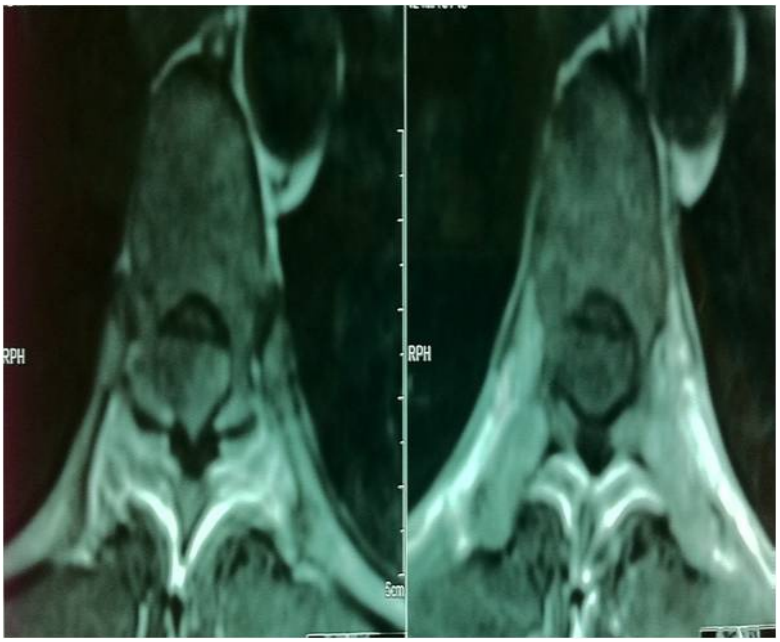

Figure 2: showing T1 axial section showing tumor occupying posterior epidural space compressing the cord.

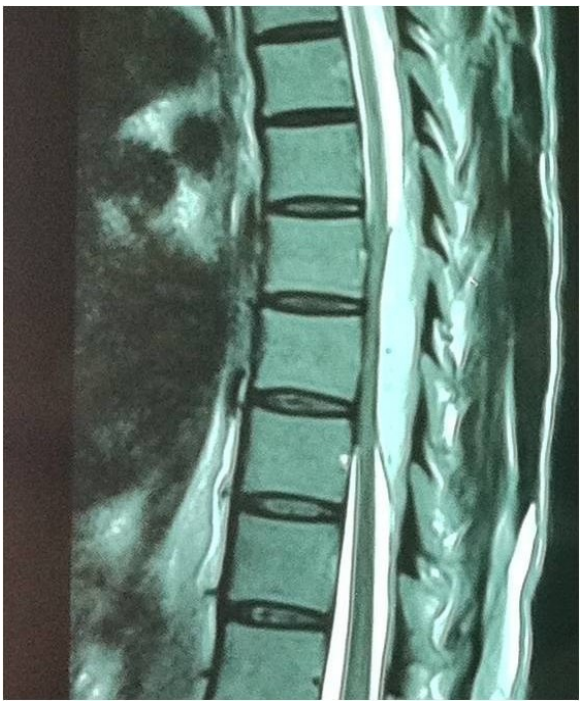

Figure 3: showing enhancement of tumor with diffuse uptake of contrast.

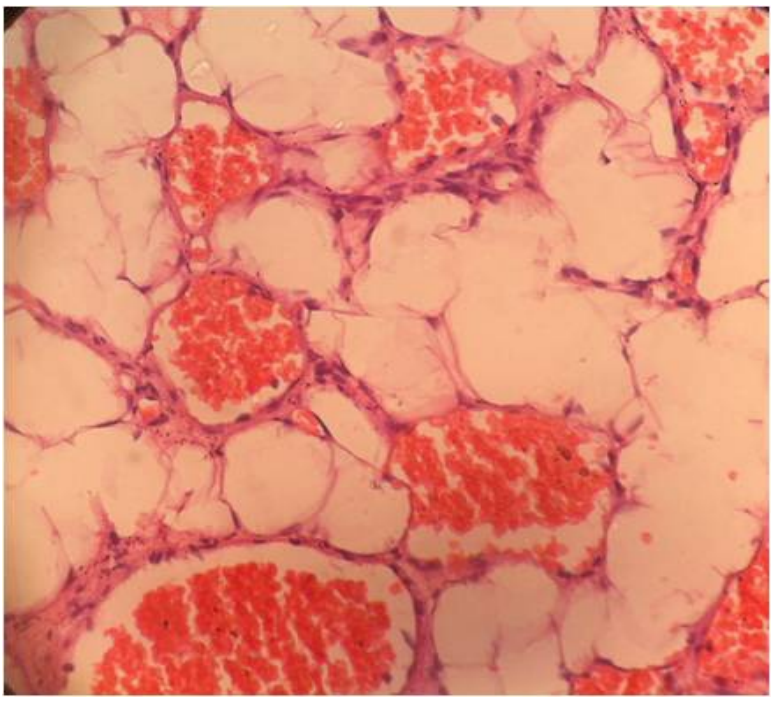

Figure 4: showing histopathological picture of angiolipoma showing blood vessels and fat cells.

Conclusion

Spinal angiolipoma is a rare pathology causing compressive myelopathy; it is important to recognise the entity and differentiate it from other serious afflictions on the basis of MR imaging, which can also be used to identify its infiltrative nature and vascularity to fat ratio, both of which might affect the surgical management in terms of approach, need for preoperative angioembolisation, and possibility of en-masse excision along with the pseudo-capsule. The tumour is slow growing; and shows an excellent prognosis even with intralesional removal, with minimal chances of recurrence.

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