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Case Report

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Clinical, Radiological and Histological Characteristics of Thoracic Epidural Angiolipoma: A Case Report

Torasik Epidural Anjiyolipomun Klinik, Radyolojik ve Histolojik Özellikleri: Olgu Sunumu

Tayfun Hakan,[1] Erhan Çelikoğlu,[1] Jülide Hazneci,[2] Adnan Somay[3]

[1]Department of Neurosurgery, University of Health Sciences, Hamidiye Faculty of Medicine, Istanbul, Türkiye
[2]Department of Neurosurgery, Fatih Sultan Mehmet Treaning and Research Hospital, Istanbul, Türkiye
[3]Department of Pathology, Fatih Sultan Mehmet Treaning and Research Hospital, Istanbul, Türkiye

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Correspondence:
Dr. Tayfun Hakan.
Department of Neurosurgery, University of Health Sciences, Hamidiye Faculty of Medicine, Istanbul, Türkiye

Phone:
+90 532 324 32 84

e-mail:
tayfunhakan@yahoo.com

ABSTRACT

Spinal epidural angiolipomas are benign tumors composed of mature fat cells and abnormal vessel structures. They are often misdiagnosed, and there is limited information on their clinical features and surgical strategies based on outcomes. A 59-year-old female presented with chronic neck pain. Magnetic resonance imaging revealed an epidural mass at the T1-T2 levels. It was isointense on the T1 and hyperintense on the T2-weighted images. It showed intense and homogenous enhancement. The same tumor was identified in a previous MRI examination conducted three years ago. However, the patient had declined the recommended surgical treatment at that time. There were no notable differences between the previous and current MRI scans. A laminectomy was performed on the T1 and T2 laminae, and the tumor was found to be soft, fragile, and reddish-purple. Pathological analysis revealed mature adipose tissue with varying levels of vascular proliferation. The blood vessels within the tumor contained fibrin thrombi, which was characteristic of angiolipoma. It had a low proliferation index (low Ki-67) and showed a positive stain for CD31 in immunohistochemical staining. The patient had an uneventful postoperative recovery. In conclusion, angiolipomas can remain asymptomatic for up to three years; patients may still have normal neurological function even if the tumors occupy half of the spinal canal. It is critical to consider angiolipomas as a possible cause when diagnosing epidural spinal lesions. MRI features can aid in the diagnosis, and surgical removal is the recommended treatment with a high success rate.

Keywords: Angiolipoma; Magnetic resonance imaging; Pain; Spine; Spinal cord compression; Spinal epidural tumor.

ÖZET

Spinal epidural anjiyolipomlar, olgun yağ hücreleri ve anormal damar yapılarından oluşan iyi huylu tümörlerdir. Sıklıkla yanlış tanı alırlar ve klinik özellikleri ile sonuçlara dayalı cerrahi stratejiler hakkında bilgiler sınırlıdır. 59 yaşında kadın hasta kronik boyun ağrısı şikayetiyle başvurdu. Manyetik rezonans görüntülemede T1-T2 düzeyinde epidural kitle saptandı. T1 ağırlıklı görüntülerde izointens, T2 ağırlıklı görüntülerde ise hiperintens idi. Yoğun ve homojen kontrast tutulumu mevcuttu. Aynı tümör, üç yıl önce yapılan MR incelemesinde de tespit edilmiş, ancak hasta önerilen cerrahi tedaviyi reddetmişti. Önceki ve mevcut MR taramaları arasında kayda değer bir fark saptanmadı. Hastaya T1 ve T2 laminektomi ile tümör eksizyonu ameliyatı yapıldı. Tümör yumuşak kıvamlı, kırılgan ve kırmızımsı-mor renkteydi. Patolojik incelemede değişen düzeylerde vasküler proliferasyona sahip olgun yağ dokusu görüldü. Tümör içindeki kan damarları, anjiyolipomun karakteristiği olan fibrin trombüsünü içeriyordu. Düşük bir proliferasyon indeksine sahip (düşük Ki-67) ve immünohistokimyasal boyamada CD31 pozitiftir. Ameliyat sonrası iyileşme süreci sorunsuz geçti. Sonuç olarak, anjiyolipomlar üç yıla kadar asemptomatik kalabilir; tümörler omurilik kanalının yarısını işgal etse bile hastaların nörolojik fonksiyonları hala normal olabilir. Epidural spinal lezyonların ayırıcı tanısında anjiyolipomların dikkate alınması kritik öneme sahiptir. MR özellikleri tanıya yardımcı olabilir. Önerilen tedavi yöntemi cerrahi olarak çıkartılmalarıdır.

Anahtar sözcükler: Ağrı; Anjiyolipom; Bası; Manyetik rezonans görüntüleme; Omurga; Spinal epidural tümör; Spinal kord.

Angiolipomas are rare, slow-growing, benign tumors composed of mature fat cells and abnormal vessel structures. They primarily occur in the subcutaneous tissues of the extremities and trunk. Spinal angiolipomas account for only 0.04 to 1.2% of all angiolipomas found in the epidural and posterior thoracic regions.[1] These tumors typically cause pain and progressive neurological deficits due to the compression of neural structures, usually around one year after onset.[2] Misdiagnosis of angiolipomas is common due to incomplete reports on their clinical features, and the surgical strategies based on outcomes are limited.[3]

In this case report, we presented a description of a patient who underwent surgical resection of a spinal extradural angiolipoma in the superior thoracic region. The report included detailed information about the patient's clinical, radiological, and histological characteristics.

Case Report

In June 2022, a 59-year-old female patient visited the outpatient clinic due to chronic neck pain. Her neurological examination showed no abnormalities, as she did not experience any radiating pain or numbness, and her deep tendon reflexes were normal. A spinal magnetic resonance imaging (MRI) was conducted, which revealed a mass lesion measuring 36x16x12 mm on the right side of the spinal canal at the T1-T2 level. The lesion extended two vertebral body lengths in the craniocaudal direction (Fig. 1). On the T1-weighted images, the lesion appeared iso-intense, while on the T2-weighted images, it was slightly hyper-intense. Additionally, the lesion exhibited homogenous enhancement on the post-contrast T1-weighted images.

The same tumor was also present in the previous MRI examination performed in November 2018 due to neck pain, but she had refused the recommended surgical treatment at that time. There was no difference between the previous

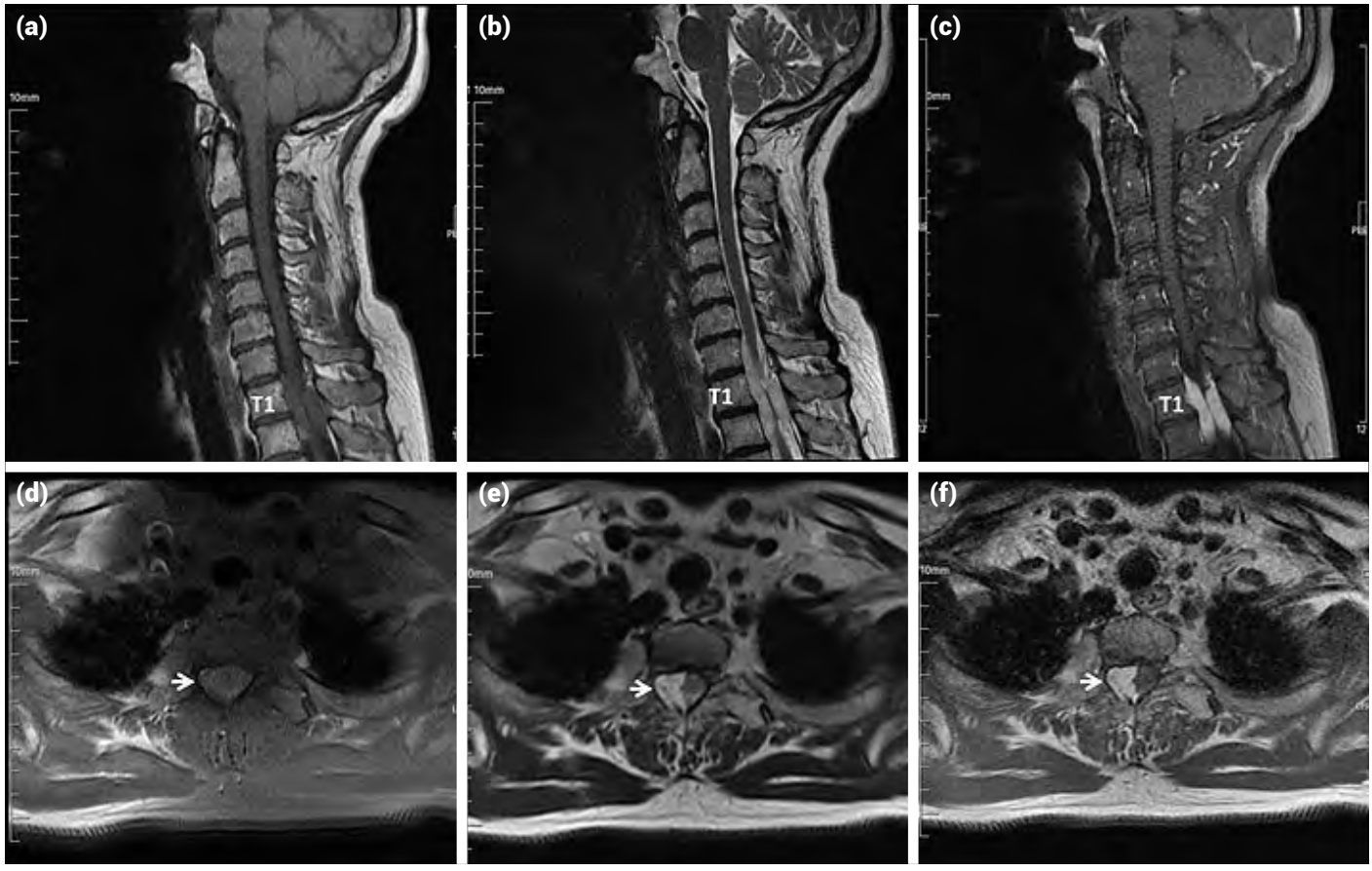

Figure 1. Magnetic resonance images of the thoracic epidural lesion. An isointense lesion is seen throughout the T1-T2 levels on sagittal **(a)** and axial **(d)** T1-weighted images (white arrow). The same lesion appears slightly hyperintense on sagittal **(b)** and axial **(e)** T2-weighted images (white arrow). The lesion occupies the right half of the spinal canal and compresses the spinal cord. The lesion exhibits an intense and homogeneous enhancement on the post-contrast sagittal **(c)** and axial **(d)** T1-weighted images (white arrow).

and current MR images; only the lesion size had increased by 2-3 mm in length. Additionally, there was no lesion in the MRI conducted in September 2014 (Fig. 2) for her neck pain. A laminectomy was performed on the T1 and T2 laminae to decompress the spinal cord and remove the epidural mass. The tumor was soft, fragile, and reddish-purple. It lined the right posterolateral borders of the T1 and T2 vertebrae, measuring 10 mm in thickness, and surrounding the spinal cord from the right side like a sleeve. However, it was easily movable and showed no signs of invading adjacent structures. The tumor was successfully removed as gross total, and the patient was discharged without any complications.

The histopathological examination revealed a partially capsulated nodular stromal tumor (Fig. 3). Among the uni-vacuolar adipocyte groups, there were structures of proliferating vessels of varying sizes. Some blood vessels were small and rounded, while others were dilated. Furthermore, some blood vessels contained fresh blood clots consisting of concentrated red blood cells and plasma in their lumens. In certain areas, the vascular structures had sinusoidal patterns. The endothelial cells and adipocytes lining the vessels showed no signs of atypia. In immunohistochemical staining, the blood vessels tested positive for CD34 and CD31. The Ki-67 proliferation index was low (≤3). The tumor was diagnosed as an angiolipoma and did not require any further adjuvant therapy. At the 7-month follow-up, there was no clinical and radiological evidence of recurrence in the patient.

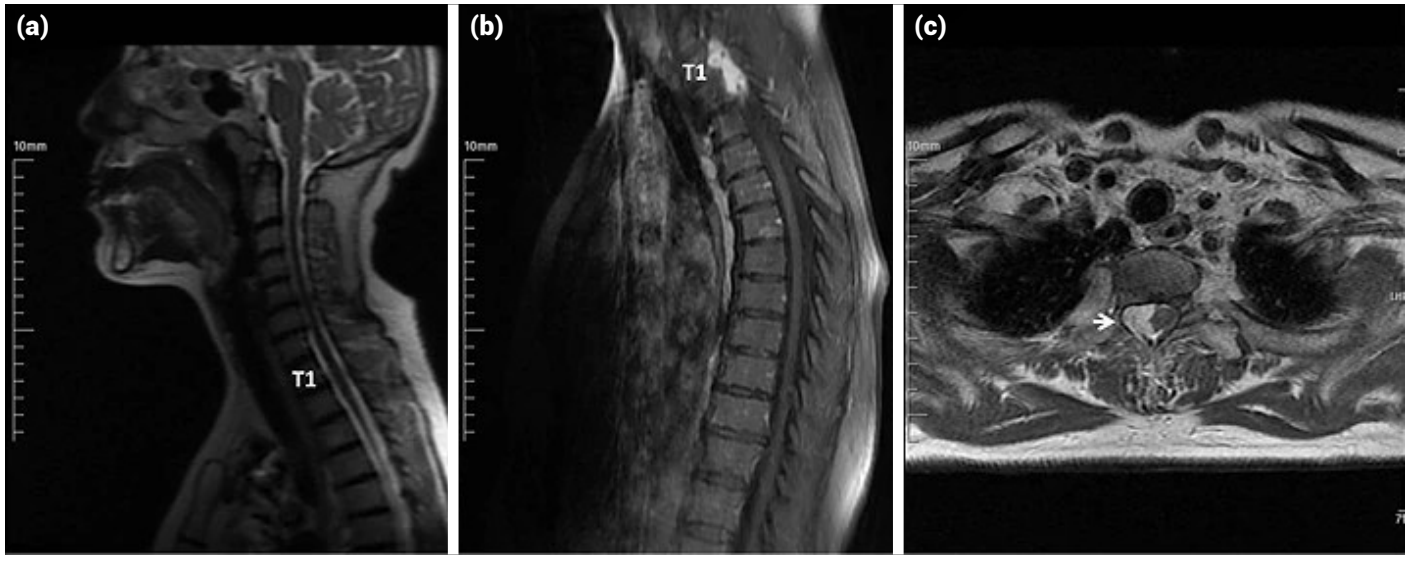

Figure 2. Images of two different magnetic resonance studies performed four years apart. **(a)** A sagittal T2-weighted image of the patient in 2014 showed no mass lesion. Postcontrast sagittal **(b)** and axial **(c)** T1-weighted images performed in 2018 revealed a mass lesion with contrast enhancement at the T1-2 levels occupying the right half of the spinal canal (white arrow).

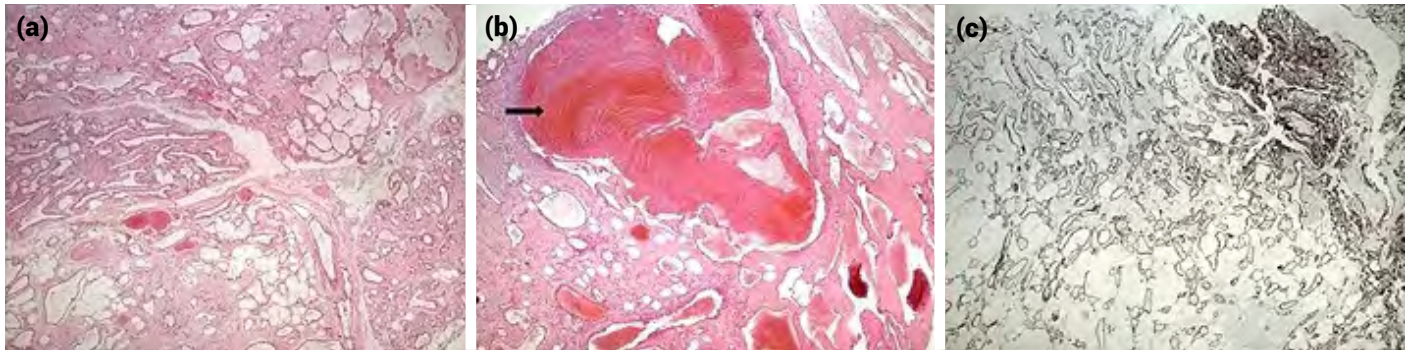

Figure 3. Histopathological images of the angiolipoma. **(a)** Tumor consisting of vascular structures of various calibrations among the uni-vacuolar adipocyte groups. Hematoxylin-eosin X 100. **(b)** Large-caliber vascular structures with fresh thrombus (arrow) in their lumen. Hematoxylin-eosin X 100. **(c)** Proliferated vessels staining positive for CD31. X40.

Discussion

Spinal angiolipomas have been of interest since they were first described by Berenbruch in 1890. This study presents a rare case of epidural thoracic angiolipoma in a female. The demographic data and location of the tumor were partly similar to previously reported cases. These lesions are seen in females,[1,3,4] with symptom onset typically occurring in the fourth and sixth decades.[3,5] The tumors most commonly occur in the mid-thoracic region, particularly at T2-T5 levels.[1,2,4]

The etiology and pathogenesis of spinal extradural angiolipomas are still not well understood. Mesenchymal stem cells capable of differentiating into both angioid and adipose cells have been suggested to be involved in the formation of spinal angiolipomas.[6] Additionally, obesity and pregnancy are believed to be risk factors.[7,8] The predominance of spinal angiolipomas in females and their frequent occurrence in peri- or postmenopausal women suggest a possible hormonal influence on their development or persistence.[9]

Angiolipomas are known to present with progressive neurological deficits and pain because of progressive spinal cord compression.[2] The initial symptoms are sensory disorders and back pain in most of the patients,[3] but motor deficits and sphincter dysfunctions can be seen over time;[9,10] it is expected to gradually worsen in about 1 year.[2] Surprisingly, the presented patient had no abnormal neurological exam findings despite her intraspinal subarachnoid space being obliterated and the spinal cord displaced. The tumor in the patient was most likely incidental. The patient's neck pain was chronic and was present four years before the tumor was detected. Moreover, she only complained of chronic neck pain during the 3 years she lived with a very slow-progressing tumor. The patient's tumor should have emerged within a maximum of 4 years because there was no appearing tumor in the MRI examination performed four years ago. This case is unique as it gives an idea of the developmental time of the angiolipoma.

Most angiolipomas are located in the thoracic spine, often in the posterior region, causing the spinal cord to be pushed forward.[4,11,12] In the presented case, the angiolipoma was located at the thoracic level, and it occupied the entire right half of the spinal canal by pushing the spinal cord to the left half. Extradural angiolipomas can be identified by distinctive MRI findings. In T1-weighted images, the presence of a lipid tumor is indicated by inhomogeneous images, and areas of hypointensity correlate with vascularization.[2] The appearance of MR can vary depending on the ratio of fat to vessel. Type 1: tumors consist mostly of fatty tissue appearing hyperintense on T1-weighted and T2-weighted images.[3,10] Type 2: tumors are rich in vascular structures appearing isointense on T1-weighted and hyperintense on T2-weighted images. The case presented here was classified as type 2 with isointensity on T1 images and hyperintensity on T2-weighted images. The intraoperative appearance of the tumor was also consistent with MR classification, appearing reddish-purple. As previously mentioned, the gross appearance of the extradural tumor during surgery resembled that of a metastatic carcinoma, typically found in the usual site of malignant tumors in the spine.[13]

Angiolipomas are divided into infiltrating and non-infiltrating subgroups depending on their biological behavior.[14] Infiltrating angiolipomas are non-capsulated or rarely partially capsulated, while non-infiltrating angiolipomas are well-capsulated tumors. Most angiolipomas, including the presented case, are non-infiltrating types. Non-infiltrating spinal angiolipomas can typically be easily removed without causing any damage to the nerves or blood vessels. However, infiltrating types can be more challenging to resect due to their invasive nature.[3] The best treatment for thoracic angiolipomas is complete removal of the tumor.[1] These tumors are usually benign and slow-growing, and patients can expect to experience functional recovery after complete removal.[3] Recurrence of these tumors is exceptional.[5]

The pathological features of the presented study included mature adipose tissue with variable vascular proliferation. The blood vessels contained fibrin thrombi, characteristic of angiolipomas.[15] Additionally, the study showed a low proliferation index with low Ki-67 and a positive stain for CD31 in immunohistochemical staining.[12]

Conclusion

Angiolipomas are slow-growing, benign tumors that can remain silent for more than three years without any signs or symptoms, as was observed in the presented case. Patients may be intact neurologically even if the tumor occupies half of the spinal canal and pushes the spinal cord into the other half of the spinal canal in the superior part of the thoracic spine. Angiolipomas should be considered among the differential diagnoses of epidural spinal lesions. MRI features help make the diagnosis, and surgical resection is the recommended treatment option with a nearly excellent prognosis.

Disclosures

Informed consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

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