

Case Report

Spinal thoracic extradural angiolipoma manifesting as acute onset of paraparesis: Case report and review of literature

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Abstract

Background: Angiolipomas are benign tumors most commonly found in the thoracic spine. They are composed of mature adipocytes and abnormal vascular elements that usually present with a slowly progressive course of neurological deterioration.

Case Description: A 35-year-old female, with a prior history of back pain, acutely developed paraparesis. When the thoracic magnetic resonance imaging (MRI) revealed a dorsal epidural mass at the T3-T5 level, she underwent a laminectomy for gross total excision of the lesion that proved to be an angiolipoma. On the second postoperative day, the patient was again able to ambulate.

Conclusion: The angiolipomas of spine are rare causes of spinal cord compression, and those presenting with acute neurological deficits should be immediately treated.

Key Words: Angiolipoma, extradural spinal tumor, spinal cord compression, spinal tumor

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INTRODUCTION

Angiolipomas are benign tumors consisting of mature fat cells and proliferating abnormal blood vessels. They generally occur in the subcutaneous tissue of the trunks and limbs. They can be further categorized into two subtypes – noninfiltrating (more common) and infiltrating. They account for 0.004–1.2% of all spinal axis tumors and 2–3% of extradural spinal lesions.^[2,4] Here, the diagnosis, pathology, and treatment of a spinal angiolipoma are reviewed.

CASE REPORT

A 35-year-old female, with 4 weeks of bilateral lower limb numbness, acutely presented with 24-hour evolution of an acute paraparesis, accompanied urinary retention. Physical examination revealed 4/5 motor strength in both the legs, diffuse lower extremity hyperreflexia, and superficial hypoesthesia below the T4 level.

The magnetic resonance imaging (MRI) scan showed a large fusiform dorsal lesion measuring 8.0 cm × 2.8 cm × 1.1 cm compressing the thoracic spinal cord extending over the T3-T5 levels [Figure 1]. It was inhomogeneous, isointense on T1-weighted images, and hyperintense on T2-weighted images, showing intense enhancement after gadolinium administration and occupying the posterior aspect of the spinal canal [Figure 2].

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Surgery

A laminotomy of T3-T5 was performed under general anesthesia with intraoperative neurophysiological monitoring. The posterior epidural space was filled with a fatty, highly vascular brown-pink mass that was extremely hemorrhagic. It was easily mobilized away from the compressed dura, and a gross total resection was accomplished (e.g., in one piece). There were no changes in the intraoperative motor evoked potentials [Figure 3].

Postoperative course

On postoperative day 2, the patient was able to ambulate, and she had fully recovered within 2 weeks. After 2 years, the patient remains asymptomatic and shows no signs of spinal deformity. There is also no evidence of radiographic recurrence.

Pathology

The analysis of the tumor revealed a lesion with adipocytes and capillary sized vessels [Figure 4].

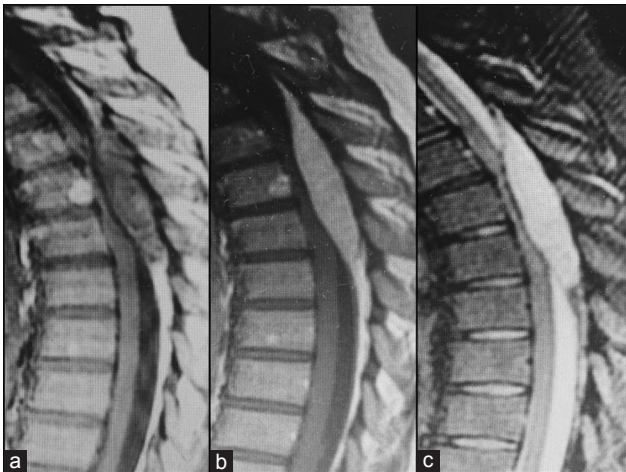


Figure 1: Sagittal MRI images showing heterogeneous epidural posterior mass compressing thoracic cord over three body segments between T3-T5. (a) T1-weighted MRI shows inhomogeneous iso to hypointensity mass; (b) Post-contrast T1-weighted MRI shows enhancing mass; (c) T2-weighted MRI shows a hyperintense tumor

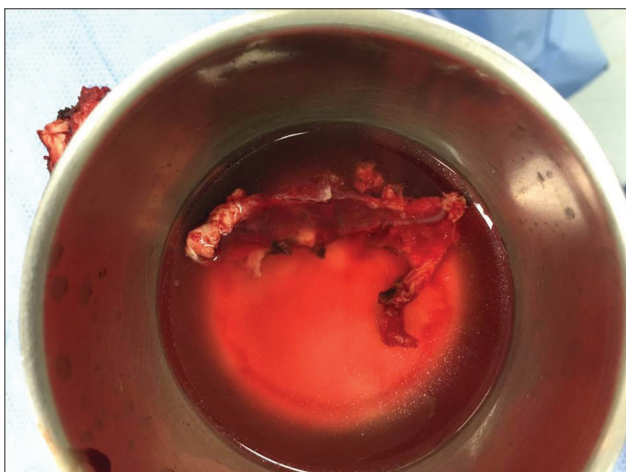


Figure 3: Total resection of a fleshy pink-brown vascular mass

DISCUSSION

Histology

Berenbruch in 1890 was the first to report extradural spinal angioliopoma; the first pathological report was made by Howard in 1960.^[1,4,6]

These lesions consist of varying portions of mature fat cells and abnormal capillary, sinusoidal, venous, or arterial vascular elements. The ratio of fat to vessels is variable and ranges from 1:3 to 2:3. Tumors with an abundance of smooth muscle proliferation are further subclassified as angiomyoliopomas. Angioliopoma generally arise from the dorsal aspect of spinal canal at thoracic levels compressing the spinal cord and causing symptoms.^[2,4-6]

Origin

The origin and pathogenesis of angioliopomas are thought to arise from pluripotential mesenchymal stem cells with secretory activity. Early inclusion of pluripotent stem cells during the developmental ossification of neural arch is believed to be a prerequisite for spinal angioliopoma formation. Degenerative changes may be present in some longstanding cases but malignant transformation and neural tissue infiltration have never been reported.^[2-4,6,7]

Clinical presentation

Angioliopomas predominantly occur in females (female:male ratio = 22:17) in their fifties who

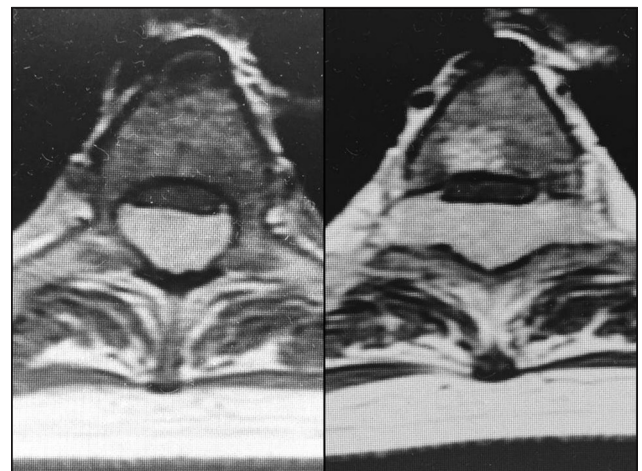


Figure 2: Axial T2 MRI show compression and anterior displacement of the spinal cord

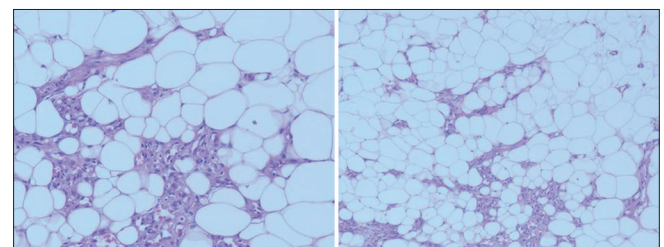


Figure 4: Angioliopoma consist of mature adipocytes and branching capillary sized vessels, which usually contain fibrin thrombi. H and E ×200

typically suddenly develop paraparesis. The rapid onset of symptoms is usually due to vascular factors; e.g., anomalous vessels, intralesional thrombosis, and hemorrhage or steal phenomena.^[4,6,7]

Demyelinating disease must be considered among the differential diagnoses as occasionally these lesions present with relapsing/remitting course.^[2]

Imaging

MRI, T1-weighted images typically demonstrate a high signal accompanied by an inhomogeneous mass. On T2-weighted images, the signal intensity seems to be similar to adipose tissue. The lesion markedly enhances with gadolinium administration [Table 1]. Other differential diagnoses include benign and malignant neoplasms; e.g., angiomyolipomas, lymphoma, and epidural metastases.^[4,6,7]

Treatment and prognosis

Spinal angioliipomas are treated exclusively by surgical removal, and most may be completely excised with good clinical results (e.g., normal postoperative

examinations).^[4-7] If magnetic resonance suggests a highly vascular tumor, angiography and embolization can be performed before surgery to facilitate surgical excision. These lesions rarely recur; even infiltrating subtypes treated with subtotal resection did not recur after an average 37.6 months follow-up in one study.^[4]

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Conflicts of interest

There are no conflicts of interest.

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Table 1: Previous reported cases of extradural angioliipomas performed with magnetic resonance imaging

Age (mean)	Gender (n)	Duration of symptoms (mean)	MRI finding (T1, T2/post-contrast)
46.5	F=26 M=20	13 months	T1/T2 very variable - usually hyperintensity inhomogeneous marked enhancement