ORIGINAL ARTICLE

Spinal epidural angiolipomas: Clinical characteristics, management and outcomes

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ABSTRACT

Purpose: The spinal epidural angiolipomas are rare expansive processes made of mature lipomatous and angiomatous elements. They often have a benign character. Their etiology, pathogenesis remains uncertain, and it is a cause of spinal cord compression. The magnetic resonance imaging is the most important neuroradiological examination. Histological examination is the only examination to confirm the diagnosis. Surgery is the treatment of choice.

Methods: A retrospective study of all patients operated on for a spinal epidural angiolipoma at the Department of Neurosurgery at the National Institute of Neurology of Tunis between January 2000 and December 2014 (15 years) was performed. The aim of this study is to describe the clinical, radiological, histological characteristics and the treatment of this tumor.

Results: A total of nine patients were operated from January 01, 2000 to November 30, 2014. The average age of our patients was 51 years with ages that ranged from 29 to 65 with a male predominance. The period between onset of symptoms and diagnosis ranged from 24 months with an average 12 months. Posterior localization of the tumor was seen in all patients. Surgical resection was performed for all cases. The postoperative course has been satisfactory, with a complete recovery of neurological functions in all patients.

Conclusions: The spinal epidural angiolipomas is rare expansive process causing spinal cord compression. Treatment is exclusively surgical resection. The functional outcome of spinal epidural angiolipomas is particularly favorable with a complete neurological recovery is if the patient was quickly operated.

Key words: Angiolipoma, epidural tumor, magnetic resonance imaging, pathology, spinal

Introduction

Spinal angiolipomas are rare benign tumors containing mature fat cells with an excessive degree of vascular proliferation.[1-3] They are estimated to account for between 0.04% and 1.2% of all spinal axis tumors and are predominantly found in the epidural space, where they represent 2–3% of spinal tumors.[1,4,5] There have been more than 128 cases reported to date.[6] In this paper, we present our cases of spinal epidural angiolipomas and discuss the relevant literature.

Methods

A retrospective study of all patients operated on for a spinal epidural angiolipoma at the Department of Neurosurgery at the National Institute of Neurology of Tunis between January 2000 and December 2014 (15 years) was performed.

The purpose of this paper is to report our cases and review the clinical presentation, radiological appearance, pathological

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aspects, and treatment of this distinct clinico-pathological entity.

Lipomas and pure angioma were excluded from our study.

Data are collected and analyzed from patient medical files in the hospital archives.

Results

A total of nine patients were admitted and operated during the period mentioned above.

The average age of our patients was 51 years with ages that ranged from 29 to 65 years. Four women aged 40, 47, 59, and 61, and five men aged 29, 48, 49, 62, and 65 [Table 1].

The diagnosis was established in all cases based on clinical findings, computed tomography (CT) scan, magnetic resonance imaging (MRI), and pathological findings.

The period between onset of symptoms and diagnosis ranged from 24 months with an average of 12 months. Six patients were presented with a progressive neurological deficit in the lower limbs, ranging from a mild deficit to complete motor loss as a consequence of spinal cord compression. A CT scan of the spine was done for one patient.

MRI was performed to the rest of 8 cases and showed homogeneous isointensity on T1-weighted images, which was slightly higher than the spinal cord, and homogeneously slightly high signal on T2-weighted images and homogenously strongly enhanced. All lesions were noninfiltrating [Figures 1 and 2].

Of the nine patients, the lesions were located at the thoracic segment of the spinal canal in seven cases and a lumbar segment in two cases.

All lesions were resected by open surgery, and pathologic examination was performed for all cases. During surgical procedures, the lesions showed dark red or purple red color and as an intact capsule.

On histological examination, the tissue consisted mainly of fat cells and some regular vessels. Mature adipocytes and multifocal vascular proliferation, consistent with an angiolipoma [Figure 3].

The postoperative course has been satisfactory, with a complete recovery of neurological functions in all patients [Tables 1 and 2].

Discussion

Berenbruch first described a case of spinal angiolipoma in 1890.[7] In 1960, Howard and Helwig established angiolipoma as a clinicopathological entity containing vascular and

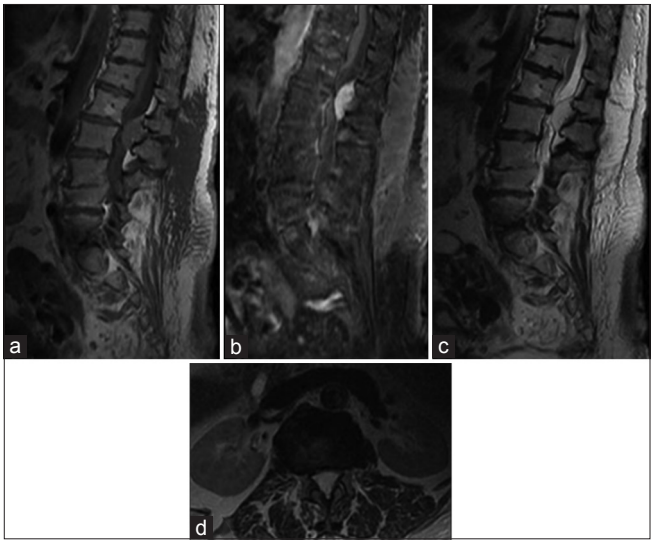

Figure 1: Lumbar magnetic resonance imaging of 61-year-old woman, sagittal T1-weighted fast-spin echo (a), sagittal T1-weighted fast-spin echo, fat-saturated images after contrast (b), T2-weighted fast spin-echo (c), and axial T2-weighted (d), showed a L1–L2 posterior epidural lesion with both lipomatous and vascular components and homogeneous contrast enhancement

Table 1: Summary of clinical characteristics, management, and results of our patients

Case	1	2	3	4	5	6	7	8	9
Age (years)	59	40	62	49	29	48	61	65	47
Sex	Female	Female	Male	Male	Male	Male	Female	Male	Female
Symptoms	Lower limb monoparesis	Paraplegia	Paraparesis	Paraparesis	Paraparesis	Paraparesis	Lumbosciatalgia	Back pain	Paraplegia
Duration of symptoms (months)	18	1	4	12	10	24	16	12	>1
Site	D5-D8	D4	D4-D6	D4-D6	D1-D8	D4-D10	L1-L2	D3-D5	L1-L4
Axial localization	Posterior	Posterior	Posterior	Posterior	Posterior	Left posterolateral	Posterior	Posterior	Posterior
Treatment	Total surgical removal	Total surgical removal	Total surgical removal	Total surgical removal	Total surgical removal	Total surgical removal	Total surgical removal	Total surgical removal	Total surgical removal
Result	Recovery	Recovery	Recovery	Recovery	Recovery	Recovery	Asymptomatic	Asymptomatic	Recovery

Table 2: A comparison with published literature reports of spinal epidural angiolipomas

	Age (years)	Sex ratio	Duration of symptoms (months)	Localization	Site	Surgery treatment	Results
Our study	51 (29-65)	5 male/4 female 1, 2	12 (1-24)	Posterior: All cases	Thoracic: 7/9 cases	100%	Total recovery 100%
Literature	44.03±5.9	1.2 and 1.6	20.2±9.6	Posterior 85%	Thoracic 78%	100%	Total recovery 100%
Authors	Gelabert *et al.*, 2009	Akhaddar *et al.*, 2000 Turgut *et al.*, 1999	Kussel *et al.*, 1989 Fernandez *et al.*, 1994 Anson *et al.*, 1990	Wilts *et al.*, 1993	Akhaddar *et al.*, 2000 Turgut *et al.*, 1999	Howard *et al.*, 1974 Enzinger *et al.*, 1995 Do souto *et al.*, 2002	Preul *et al.*, 1993 Gelabert *et al.*, 2009

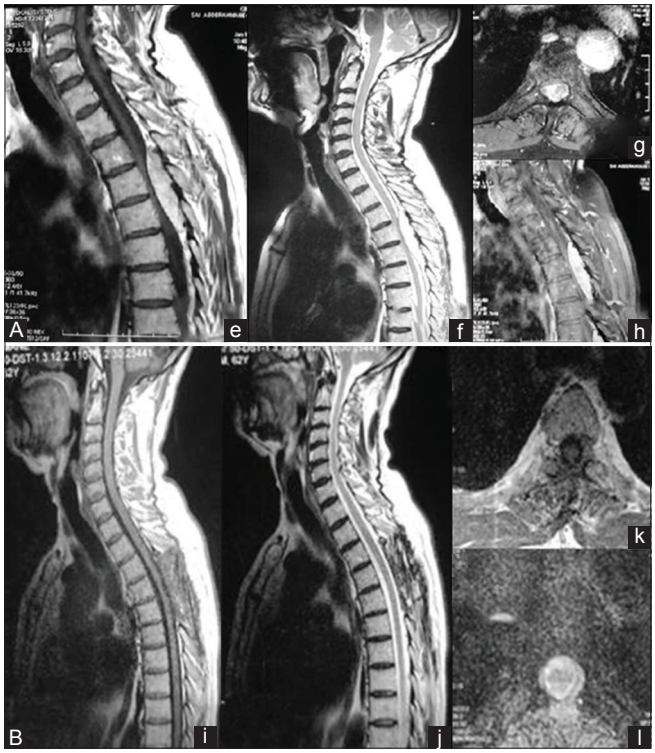

Figure 2: Spinal magnetic resonance imaging in a 65-year-old woman. (A) sagittal T1-weighted (e), sagittal T2-weighted (f), axial (g), and sagittal (h) T1-weighted, showing posterior spinal epidural mass with inhomogeneous high signal on all sequences, extending from T3 to T4 and causing thecal sac compression. (B) The postoperative spinal magnetic resonance imaging in the same patient, sagittal (i) T1-weighted, sagittal (j) and axial (l) T2-weighted, axial (k) T1-weighted after contrast, showing the disappearance of the lesion

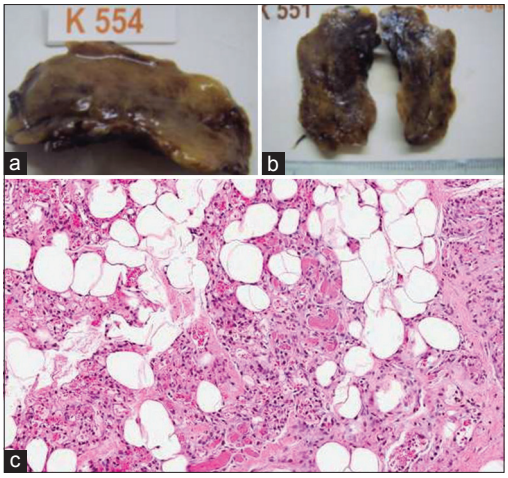

Figure 3: (a and b) The cut surface of the tumor (the same patient) is yellowish, in places hemorrhagic, firm, and spongy. (c) Histopathology examination of the tumor. Showing admixture of mature adipose tissue and vascular elements (H and E, ×40)

mature adipose elements.[8] Various terms including vascular lipoma, hemangiolipoma, and fibromyolipoma have been used to describe these lesions.[5] Spinal epidural angiolipomas commonly occur in adults in the fourth and fifth decades and have female preponderance.[1,9] Few cases of pediatric spinal epidural angiolipomas have been reported.[1] The histopathogenesis of angiolipomas is unknown. They probably arise from abnormal primitive pluripotential mesenchymal cells that can differentiate into lipomatous, angiomatous, or mixed tissue.[10] Spinal angiolipomas are categorized as one of the two types: "Noninfiltrating" and "infiltrating." In the common type, angiolipoma is encapsulated and noninfiltrating, and shows a benign prognosis. The less common type is nonencapsulated and infiltrating, showing an unfavorable prognosis.[11-13] Almost all noninfiltrating epidural angiolipomas are located in the posterior or posterolateral space of the spinal canal.[14] The infiltrating type is intramedullary or intravertebral occurrence.[13] They most frequently appear in the thoracic spine where they are called "extradural" angiolipomas since they develop between the encephalic layer of the dura mater which forms the dural sheath of the spinal cord and the osteoperiosteal layer which covers the vertebrae.[15,16] This interperiosteal dural space is improperly called the epidural space.[15,16] Pure lumbar localization is extremely rare.[17] In our cases of thoracic and lumbar angiolipomas, the tumors were in the posterior epidural space but did not show any signs of bone infiltration.

The common clinical presentations of spinal epidural angiolipomas are progressive paraparesis, back pain without radiculopathy, lower extremity sensory changes, and hyperreflexia. Sudden neurological deterioration can occur due to tumor thrombosis or hemorrhage.[6,18] Exacerbation of symptoms may occur during pregnancy and in obese patients probably because of hormonal changes and increase in a fatty component within the angiolipoma.[19] The CT appearance of spinal angiolipoma is that of a mass that is typically

hypodense relative to the spinal cord and has variable degrees of enhancement after contrast administration.[20] MRI is the investigation of choice in diagnosing spinal angiolipoma.[21]

Angiolipomas are composite tumors constituted by mature fat tissue associated with vascular proliferation. These findings help to explain the tumor's appearance when viewed by MRI. The observed hypersignal during T1-weighted magnetic resonance (MR) sequences strongly suggests the diagnosis and enhancement after gadolinium injection confirms the hypervascular nature of these tumors. T1-weighted MR sequences, with suppression of fat tissue, indicate that the tumors were derived from fat tissue.[21] Differential diagnoses include lipoma, hemangioma, malignant lymphoma, and nerve sheath tumor.[2] The biological behaviors of infiltrating and noninfiltrating spinal epidural angiolipomas imply the need for different treatment approaches.

Noninfiltrating spinal epidural angiolipomas is often encapsulated, and complete removal of the tumor ensures no recurrence and allows the improvement of neurological symptoms.[9,22] In cases of recurring or infiltrative spinal epidural angiolipomas, wider resection followed by radiotherapy should be considered.[13,23]

Our experience as well as of those in the literature show that total excision of these lesions is often possible and that total recovery after local removal was obtained in all cases.

Conclusion

The clinical course in our patients was relatively brief compared to other reports, which have described a mean duration of symptoms of 20 months, but the operative results in our cases combined with a review of the published literature leads us to suggest that the functional outcome of spinal epidural angiolipomas is particularly favorable with a complete neurological recovery is if the patient was quickly operated.

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Conflicts of interest
There are no conflicts of interest.

References

1. Gelabert-González M, García-Allut A. Spinal extradural angiolipoma: Report of two cases and review of the literature. Eur Spine J 2009;18:324-35.
2. Gelabert-González M, Agulleiro-Díaz J, Reyes-Santías RM. Spinal extradural angiolipoma, with a literature review. Childs Nerv Syst 2002;18:725-8.
3. Bucy PC, Ritchey H. Klippel-Feil's syndrome associated with compression of the spinal cord by an extradural hemangiolipoma. J Neurosurg 1947;4:476-81.
4. Preul MC, Leblanc R, Tampieri D, Robitaille Y, Pokrupa R. Spinal angiolipomas. Report of three cases. J Neurosurg 1993;78:280-6.
5. Haddad FS, Abla A, Allam CK. Extradural spinal angiolipoma. Surg Neurol 1986;26:473-86.
6. Ghanta RK, Koti K, Dandamudi S. Spinal epidural angiolipoma: A rare cause of spinal cord compression. J Neurosci Rural Pract 2012;3:341-3.
7. Amlashi SF, Morandi X, Chabert E, Riffaud L, Haegelen C, Rolland Y. Spinal epidural angiolipoma. J Neuroradiol 2001;28:253-6.
8. Howard WR, Helwig EB. Angiolipoma. Arch Dermatol 1960;82:924-31.
9. Hungs M, Paré LS. Spinal angiolipoma: Case report and literature review. J Spinal Cord Med 2008;31:315-8.
10. Ehni G, Love JG. Intraspinal lipomas: Report of cases; review of the literature, and clinical and pathologic study. Arch Neurol Psychiatry 1945;53:1-28.
11. Turgut M. Spinal angiolipomas: Report of a case and review of the cases published since the discovery of the tumour in 1890. Br J Neurosurg 1999;13:30-40.
12. Pinto-Rafael JI, Vázquez-Barquero A, Martín-Laez R, García-Valtuille R, Sanz-Alonso F, Figols-Guevara FJ, *et al.* Spinal angiolipoma: Case report. Neurocirugia (Astur) 2002;13:321-5.
13. Lin JJ, Lin F. Two entities in angiolipoma. A study of 459 cases of lipoma with review of literature on infiltrating angiolipoma. Cancer 1974;34:720-7.
14. Miyamoto Y, Naka J, Asahi S, Mikami T, Sawada H, Sano K, *et al*. Spinal extradural angiolipoma. A report of two cases and review of the literature. Kanto Seisaishi 2009;40:420-4.
15. Andaluz N, Balko G, Bui H, Zuccarello M. Angiolipomas of the central nervous system. J Neurooncol 2000;49:219-30.
16. Bardosi A, Schaake T, Friede RL, Roessmann U. Extradural spinal angiolipoma with secretory activity. An ultrastructural, clinico-pathological study. Virchows Arch A Pathol Anat Histopathol 1985;406:253-9.
17. Rocchi G, Caroli E, Frati A, Cimatti M, Savlati M. Lumbar spinal angiolipomas: Report of two cases and review of the literature. Spinal Cord 2004;42:313-6.
18. Akhaddar A, Albouzidi A, Elmostarchid B, Gazzaz M, Boucetta M. Sudden onset of paraplegia caused by hemorrhagic spinal epidural angiolipoma. A case report. Eur Spine J 2008;17 Suppl 2:S296-8.
19. Diyora B, Nayak N, Kukreja S, Kamble H, Sharma A. Thoracic epidural angiolipoma with bilateral multilevel extraspinal extensions: A rare entity. Neurol India 2011;59:134-6.
20. Matsushima K, Shinohara Y, Yamamoto M, Tanigaki T, Ikeda A, Satoh O. Spinal extradural angiolipoma: MR and CT diagnosis. J Comput Assist Tomogr 1987;11:1104-6.
21. Yamashita K, Fuji T, Nakai T, Hamada H, Kotoh K. Extradural spinal angiolipoma: Report of a case studied with MRI. Surg Neurol 1993;39:49-52.
22. Okuyama K, Watanabe M, Matsumoto M, Maruiwa H, Chiba K, Fujimura S, *et al*. Spinal extradural angiolipoma. A report of five cases. Spine Spinal Cord 2000;11:196.
23. Nagano S, Ijiri Y, Yone K, Aketomi E, Nakahara S, Sako T. Extradural angiolipoma with thoracic myelopathy. A case report. Seikeigeka Saigaigeka 2000;49:315-6.