Spinal epidural angiolipomas: Clinical characteristics, management and outcomes

Sofiene Bouali, Nidhal Maatar, Asma Bouhoula, Khansa Abderrahmen, Imed Ben Said, Adnen Boubaker, Jalel Kallel, Hafedh Jemel

Department of Neurosurgery, National Institute of Neurology “Mongi Ben Hmida”, Faculty of Medicine, University of Tunis El Manar, Tunis, Tunisia

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 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. 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. There have been more than 128 cases reported to date. 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 features, and outcomes. This is an open access article distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 3.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

Address for correspondence:
Dr. Sofiene Bouali, Department of Neurosurgery, Faculty of Medicine, National Institute of Neurology, University of Tunis El Manar, Tunis, Tunisia.
E-mail: sofienebouali@hotmail.fr

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 homogeneously 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.

In 1960, Howard and Helwig established angiolipoma as a clinicopathological entity containing vascular and...

---

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

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

---

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.
Bouali, et al.: Spinal epidural angiolipomas

Asian Journal of Neurosurgery
Vol. 11, Issue 4, October‑December 2016

350

mature adipose elements.[8] Various terms including vascular lipoma, hemangiolipoma, and fibromyolipoma have been used to describe these lesions.[8] 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.[11] 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

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

<table>
<thead>
<tr>
<th>Age (years)</th>
<th>Sex ratio</th>
<th>Duration of symptoms (months)</th>
<th>Localization</th>
<th>Site</th>
<th>Surgery treatment</th>
<th>Results</th>
</tr>
</thead>
<tbody>
<tr>
<td>Our study</td>
<td>51 (29‑65)</td>
<td>5 male/4 female</td>
<td>12 (1‑24)</td>
<td>Posterior: All cases</td>
<td>Thoracic: 7/9 cases</td>
<td>100%</td>
</tr>
<tr>
<td>Literature</td>
<td>44.03±5.9</td>
<td>1.2 and 1.6</td>
<td>20.29±6.6</td>
<td>Posterior 85%</td>
<td>Thoracic 78%</td>
<td>100%</td>
</tr>
<tr>
<td>Authors</td>
<td>Gelabert et al., 2009</td>
<td>Kussel et al., 1989</td>
<td>Fernandez et al., 1994</td>
<td>Turgut et al., 1999</td>
<td>Anson et al., 1990</td>
<td>100%</td>
</tr>
</tbody>
</table>

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)
hypodense relative to the spinal cord and has variable degrees of enhancement after contrast administration.[19] 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.[6] 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.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

**References**