Clinical case

SPINAL EPIDURAL ANGIOLIPOMA

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SUMMARY
Spinal epidural angiolipoma is a rare benign tumor predominantly located in the mid-thoracic region. The authors report a case of spinal epidural angiolipoma in a 36-year old woman who presented with subacute paraplegia. Clinico-pathological and MRI findings of this uncommon tumor are discussed.

Key words : epidural tumor, spine, angiolipoma, MR imaging.

RÉSUMÉ
Angiolipome épidural rachidien
L’angiolipome épidural rachidien est une tumeur rare, bénigne et prédominant au niveau du rachis thoracique moyen. Les auteurs présentent le cas d’une femme âgée de 36 ans ayant un angiolipome épidural rachidien révélé par une paraplégie d’évolution subaiguë. Ils discutent les aspects cliniques, radiologiques et étiologiques de cette pathologie.

Mots-clés : tumeur épidurale, rachis, angiolipome, IRM.

INTRODUCTION
Spinal angiolipomas are rare benign tumors, composed of angiomatous and lipomatous components, situated predominantly in the mid-thoracic region. They are more common in the epidural space, followed by vertebral body with epidural extension. Intradural location is very rare. They account for 0.14 % to 1.2 % of all spinal axis tumors and 2 % to 3 % of spinal epidural tumors [21].

In this report, we describe a typical form of pure epidural spinal angiolipoma and discuss its clinico-pathological and radiological aspects.

CASE REPORT
A 36-year-old woman was referred to our department on an emergency basis, complaining of back pain, progressive weakness of both legs with gait disturbance and urinary incontinence. 10 days previously she had a minor trauma to the back. She had no other significant medical history.

Physical examination revealed weakness of lower extremities (3/5) accompanied with a sensory loss below the T8 level, including pinprick, touch and proprioception. There were hyperactive deep tendon reflexes of the lower extremities with Babinski’s sign. She could not walk without help and the gait was unsteady.

Radiographs of the thoracolumbar spine were normal. Magnetic resonance imaging (MRI) revealed a fusiform posterior epidural mass lesion compressing the thoracic cord over four vertebral body segments between T2-T6. The mass was homogeneous, hyperintense on both T1- and T2-weighted images (figures 1-4). The suggested radiological diagnosis was possibly a spontaneous subacute epidural hematoma or a tumor with fatty and vascular components.

An emergency surgical intervention was planned. Bilateral T2-T6 laminectomy was performed. A dark brown hypertensive mass was harvested. The mass occupied the posterior epidural space was seen. The mass compressed the cord from T2 to T6. Dissection of the lesion from dural sac was easy. There was no invasion of the dura or the vertebrae.

Histopathological study of the surgical specimen showed mature adipocytes associated with numerous

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small and medium sized vascular channels with relatively thin walls containing variable amounts of smooth muscle cells compatible with an angiolipoma.

The postoperative course was uncomplicated. She regained her sensation in 3 days and the full control of her sphincter 10 days later. Over a 6-month follow-up period, the patient was ambulatory without assistance.
DISCUSSION

The first case of spinal angiolipoma (AGL) was reported by Berenbruch in 1890 [1], while the first pathological description was made by Howard and Helwig in 1960 [4]. Spinal localization of AGL is very rare. Turgut [21] in the last review of the literature found only 75 cases. In the last 10 years with the routine use of MRI, their frequency increased significantly.

Lin & Lin [8] defined two different pathological types of AGL: noninfiltrating and infiltrating. The latter is very rare, being partially or entirely unencapsulated, ill-defined and infiltrates the surrounding tissues, whereas the former is encapsulated and well demarcated. The majority of the spinal AGLs are noninfiltrating type, generally occupying posterior thoracic epidural space. They are not adherent to the dura and have no tendency to involve surrounding tissues.

Although it is not clear in the literature, whether infiltrating lesions originate from the epidural space and infiltrate the bony compartment or conversely, it seems more likely that these lesions primarily involve the spine, because in all infiltrating type lesions, the tumor involves only one vertebral segment, while the epidural component of the same lesion extends more than one level [20]. They are generally situated in the anterior epidural compartment [14]; this phenomenon is well-defined in the other benign lesions of the spine such as hemangiomas [7]. Comparing the outcome and recurrence of these lesions, the two main types fail to show any difference in the spinal region [20].

In our opinion, a classification according to anatomical localization of the tumor into “pure epidural” and “vertebral” types is more justifiable. Considering the tumor behavior, we found only three cases possessing the infiltrating characteristics, all of them have been associated with the Proteus syndrome [12, 16, 19].

The clinical presentation of spinal AGLs is not different from any other benign epidural tumor. They are more common in fifth decade [21]. The mode of onset may be acute, subacute or chronic and may show radicular, paraplegic, progressive or remitting-relapsing clinical types.

The radiological findings of spinal AGL are very similar to the spinal angiomomas [6]. Computed tomography reveals a hypodense mass lesion with a variable degree of enhancement after contrast injection. Rarely, foci of calcification within the tumor are reported [9]. In the vertebral AGL, the lesion produces trabeculations of the vertebrae, simulating vertebral hemangioma.

Noncontrast T1-weighted MR images show a hyperintense epidural mass. Rarely, there are a large number of small hypointense regions interspersed through the high signal forming a mosaic pattern [15]. AGLs that contain large hypointense foci on noncontrast T1-weighted images can be expected to have a high degree of vascularity [15].

The tumor may exhibit two regions of varied signal: a dorsal homogeneously hyperintense portion or hyperintensity in the extremities of the lesion, corresponding to the lipomatous component, with an isointense area relative to the spinal cord situated in the middle or the ventral portion of the lesion, corresponding to the vascular component [5, 14]. Postcontrast T1-weighted MRI shows a slight inhomogeneous enhancement, that is more prominent in the vascular hypointense regions of the noncontrast images. It might be difficult to visualize contrast enhancement on the hyperintense T1-weighted portion of the tumor. Thus, contrast administration in conjunction with fat saturation seems to better define the postcontrast enhancement and tumoral limits [15]. On T2-weighted images, the lesion is generally isointense with the epidural fat.

The morphology of the lesion in the pure epidural type is generally fusiform, lying on the posterior surface of the dura and molding the posterior epidural space. There is a predilection for the thoracic region. The two tail-like extremities are in continuous with the normal epidural fat. It has nipple-like projections into the intervertebral foramina, and rarely extensions form a dumbbell lesion [17]. In one case, the lesion showed a diffused thickening of the epidural adipose tissue in the thoracic region simulating an epidural lipomatosis [13].

The origin and pathogenesis of AGLs has been a topic of discussion. Traditionally, AGL is grouped as a variant of spinal lipomas, but more recent clinicopathological studies [10] considered them as a specific entity different from pure lipomas, because they are not associated with spinal dysraphism and because they are more common in the epidural space. Also, it has been proposed that AGL may represent an intermediate step of a spectrum ranging from pure lipomas to hemangiomas resulting from the differentiation of primitive pluripotential mesenchymal stem cells [2, 3].

In a recent cytogenetic analysis of subcutaneous angiolipoma [18], all lesions had a normal karyotype (which is also seen in hemangiomas). In contrast, the different forms of lipoma have characteristic chromosomal aberrations. This finding raises the possibility that the angiomatos component is the primary event, and the adipose tissue, although prominent, a secondary component.

The treatment of spinal AGL is total surgical resection. In spite of vascularization of the tumor, profuse hemorrhage has rarely been reported. Although complete removal of the lesion is not always easily achievable, recurrence is exceptional [12, 16, 19].
REFERENCES


