Hemorrhagic onset of spinal angiolipoma

Case report

Marcos Devanir Silva da Costa, M.D.,1 Daniel de Araújo Paz, M.D.,1 Thaigo Pereira Rodrigues, M.D.,1 Ana Camila de Castro Gandolfi, M.D.,1 Fabricio Correa Lamis, M.D.,1 João Norberto Stavale, M.D., Ph.D.,2 Italo Capraro Surião, M.D.,1 Luiz Daniel Marques Neves Cetl, M.D.,1 and Sérgio Cavalheiro, M.D., Ph.D.1

Departments of 1Neurosurgery and 2Pathology, Federal University of São Paulo, Brazil

Spinal angiolipomas are rare benign tumors that generally induce slow progressive cord compression. Here, the authors describe a case of sudden-onset palsy of the lower extremities caused by hemorrhagic spinal angiolipoma. An emergent laminectomy was performed to achieve total lesion removal. Follow-up examinations indicated neurological improvement and the absence of recurrence.

(http://thejns.org/doi/abs/10.3171/2014.9.SPINE131162)

Key Words • spinal angiolipoma • spine tumor • cord compression • oncology

Angiolipomas are benign tumors that comprise mature fat cells and proliferating blood vessels and commonly affect the subcutaneous tissues of the trunk and extremities.4,6 Spinal angiolipoma accounts for approximately 0.14%–1.2% of spinal axis tumors,3,6,15 which are predominantly found in the epidural space.4 Approximately 39% of angiolipomas present with bone infiltration.12 They predominantly distribute in the thoracic region, followed by the lumbar and cervical spine in the dorsolateral region.4,11 These tumors induce slow progressive spinal cord or radicular compression, and acute myelopathy is exceedingly rare in such cases.1 Surgery is currently the best line of treatment for these tumors, based on the lower rate of recurrence due to the nonadherent characteristics of the lesion and the feasibility of the posterior approach.4,5

Since 1892, 128 cases of spinal angiolipoma have been reported.5 This case represents the second reported instance of the hemorrhagic onset of spinal angiolipoma.1

Case Report

History and Examination. A 43-year-old man presented with sudden-onset thoracic pain followed by paraplegia; he visited the hospital at 32 hours after onset. We found no evidence of medication, alcohol, or tobacco use or any history of disease. Neurological examination revealed the presence of urinary retention and paraplegia, absence of any sensation below the nipple line, and absence of lower extremity reflexes. Magnetic resonance imaging showed a mixed lesion with T2-weighted hypointense nuclei surrounded by isointense tissue (Fig. 1). T1-weighted imaging with Gd contrast showed slightly peripheric tissue capitulation (Fig. 2).

Operation. An emergent laminectomy of the affected spinal level was performed. Underneath the ligamentum flavum, a distinct layer of excessively vascularized fatty tissue was found surrounding a clot. This lesion was totally removed via a piecemeal approach and dissected from the dura mater. Pathological examination indicated that the removed lesion comprised mature adult fat cells embedded in a network of vascular elements and clots, and it was diagnosed as an angiolipoma by the pathologist (Fig. 3).

Postoperative Course. The patient was discharged 5 days postsurgery, with no signs of neurological recovery, and was referred to a rehabilitation center that provided all the required physiotherapy modalities as well as physiatric support. Six months after the operation, the patient presented with bladder function recovery and Grade 3 muscle strength in the lower extremities. Magnetic resonance imaging examination showed signs of laminectomy of the second thoracic vertebra, gross-total resection of the tumor, and an isointense T1-weighted (Fig. 4 left) and hyperintense T2-weighted (Fig. 4 right) area of 8 mm in the anterior portion of the spinal cord at the same level, probably related to myelomalacia. At the 10th postoperative month, the patient...
had muscle strength of Grade 4 in the legs, with spasticity of the left leg. He can stand and walk short distances without support and long distances with support.

Discussion

Berenbruch described the first case of spinal angiolipoma in 1890; however, this entity was first defined in 1945 by Ehni and Love as a benign neoplasm with abnormal blood vasculature.

Spinal angiolipoma has a female predominance, and the lesions occur most frequently during the 5th decade of life. Gelabert-González and García-Allut, in a review of 118 cases, found the following frequency rates of symptoms: progressive or sudden weakness, 40.6%; thoracic or lumbar pain, 22.8%; altered sensation (numbness), 20.3%; unilateral or bilateral sciatica, 5%; and gait disturbance, 1.7%. In a literature review, we found only 1 case in which sudden paraplegia was caused by a hemorrhagic epidural angiolipoma. Many factors contribute to the accelerated onset of symptoms, including rapid expansion of tumor volume caused by enlarging or degenerating blood vessels or hemorrhage within the lesion, as well as venous stasis with thrombosis, vascular steal, and pregnancy, which likely aggravate symptoms because of impaired spinal venous drainage or hormonal changes.

The etiology of spinal angiolipoma is unknown.
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These tumors might result from the abnormal development of primitive pluripotent mesenchymal cells, as postulated by Ehni and Love.10,12 Recently, an electron microscopy study suggested that angiolipomas originate from active secretory adipocytes that contain lipid-like materials in perivascular granules.2,10 The appearance of spinal angiolipoma can vary in imaging studies. On plain radiography, the lesions often yield negative results; however, in certain cases, they might show erosions of the pedicle and widening of the spinal canal.1,6,10,12 The lesions are hypo- to hyperdense on CT scans, depending on the extent of the vascular component or the presence of calcification. Magnetic resonance imaging is the modality of choice when diagnosing spinal angiolipoma.4,6 On MRI, angiolipomas are iso- or hyperintense on T1-weighted images and variable on T2-weighted images but usually hyperintense. The degree of central hypointensity on T1-weighted images is predictive of the degree of vascularity;2,13 therefore, heterogeneous hyperintensity lesions with focal hypointensity on T2-weighted images should suggest a diagnosis of acute/hyperacute spinal epidural hematoma.1 In a meta-analysis of 613 patients with spontaneous spinal hematoma, 29.7% of the cases were found to have no etiological diagnosis.7 We believe that a hemorrhagic event in angiolipomas could be a possible cause of acute spinal hematomas, as demonstrated in our case report.

Spinal angiolipomas are treated exclusively by surgical removal of the lesion. Noninfiltrating angiolipomas are well defined and separate from the surrounding tissue and can usually be easily removed via laminectomy and dissecting techniques. Infiltrating spinal angiolipomas ideally require a wider resection;4,5 however, even cases of subtotal resection of the infiltrating variant reportedly have good outcomes8 in terms of spinal cord decompression. One case of preoperative embolization of an infiltrating angiolipoma has been previously described, and the authors reported that this maneuver allowed complete removal with minimal intraoperative blood loss.14 Adjuvant radiation has not been recommended for patients with these benign pathological entities because the prognosis is very good, even for the infiltrating variety.3,5

Tumor recurrence following surgery is rare, and only 2 such cases (1.8%) were reported in a review of 109 cases: one recurred after gross-total removal, and the other recurred as an infiltrating tumor following subtotal resection.14 In only 3 reported cases, postoperative radiotherapy was used as complementary treatment of infiltrating angiolipomas following subtotal resection.14 The outcomes after surgical removal or decompression are satisfactory in most cases, despite the presence of severe progressive neurological deficits at the initial presentation.1,4,9

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper. Author contributions to the study and manuscript preparation include the following. Conception and design: Costa, Rodrigues, Cetl. Acquisition of data: Costa, Paz, Stavale, Cetl. Drafting the article: Costa. Critically revising the article: Paz, Rodrigues, Gandolfi, Lamis, Suriano, Cetl, Cavalheiro. Accepted submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Costa.

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Manuscript submitted January 6, 2014. Accepted September 2, 2014. Please include this information when citing this paper: published online October 10, 2014; DOI: 10.3171/2014.9.SPINE131162. Address correspondence to: Marcos Devanir Silva da Costa, M.D., Rua Napoleão de Barros, 420, CEP 04024-001, São Paulo, Brazil. email: marcosdevanir73@yahoo.com.br.