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CASE REPORT

Spinal Angiolipoma

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ABSTRACT

Spinal angiolipomas are rare benign tumours composed of mature lipocytes admixed with abnormal blood vessels. Spinal angiolipomas account for 0.14% to 1.20% of all spinal axis tumours and 2% to 3% of extradural spinal tumours. Extradural tumours usually predominate. Spinal angiolipomas commonly present with pain at the local site, spastic or peripheral paraparesis of the lower extremities, paraesthesia, and impaired sensitivity. This report describes a 58-year-old otherwise healthy man with spinal angiolipoma, who presented with chronic epigastric pain for 3 years and sudden onset of acute myelopathy. Magnetic resonance imaging confirmed the presence of a space-occupying lesion at T7, T8, and T9. The lesion was posterior in location, and spindle in shape, measuring 6.5 cm in length. Laminectomy was performed, with gross total resection of the mass. Pathology was consistent with angiolipoma. Follow-up scan at 1 year did not show any evidence of recurrent tumour.

Key Words: *Angiolipoma; Pain; Spine*

INTRODUCTION

Spinal angiolipomas are rare benign tumours composed of mature lipocytes admixed with abnormal blood vessels. Spinal angiolipomas account for 0.14% to 1.20% of all spinal axis tumours and 2% to 3% of extradural spinal tumours.[1] Extradural tumours usually predominate. Spinal angiolipomas commonly present with pain at the local site, spastic or peripheral paraparesis of the lower extremities, paraesthesia, and impaired sensitivity. This report describes a 58-year-old otherwise healthy man with spinal angiolipoma, who presented with chronic epigastric pain for 3 years and sudden onset of acute myelopathy.

CASE REPORT

A 58-year-old otherwise healthy man presented in 2008 with chronic pain in the upper abdomen radiating to the back for the previous 3 years. The pain had been investigated, and he was diagnosed with irritable bowel syndrome. The condition was treated with anxiolytics and analgesics, but these did not help him, and the pain was affecting his quality of life.

The patient developed acute bilateral leg weakness, which worsened over the next 24 hours. He reported to the local hospital emergency room, where his symptoms became worse and he developed urinary retention and rectal incontinence. He had had no back injury or neurological symptoms in the past. However, the patient lifted a heavy object before developing the symptoms, which may have increased the resistance to venous return. Physical examination revealed paraparesis. The sensory impairment was below the T10 level, with no sacral sparing. Deep tendon reflex of the lower legs was increased. There was no relevant family history. No dysraphism was seen.

Magnetic resonance imaging (MRI) of the spine showed a space-occupying lesion at the T7 to T9 level (Figure 1). The lesion was posterior in location, and spindle in shape, measuring 6.5 cm in length and 10 mm at its maximum width. T1- and T2-weighted sequences and post-contrast with fat suppression images suggested a meningioma. The rest of the spine was normal. The patient underwent T7 to T9 laminectomy, with total removal of the epidural mass. The postoperative period was uneventful.

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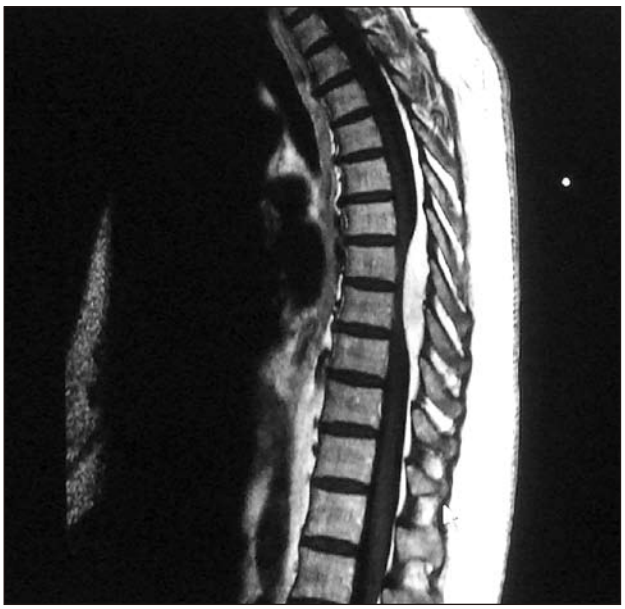

Figure 1. Magnetic resonance imaging of the spine showing a space-occupying lesion at the T7 to T9 vertebral level.

Pathology examination showed sections with benign adipose tissue with a prominent vascular network. The vessels were congested and were predominantly capillaries with some fibrin thrombi. The cellular variant included angiomatous tissue and spindle cells, which were abundant in the cellular areas and associated with mild pleomorphism. The histopathology result was consistent with a diagnosis of angiolipoma (Figures 2 and 3). There was no evidence of malignancy. The patient made a full neurological recovery, and the epigastric pain completely subsided. Follow-up MRI at 1 year showed postoperative changes, with no residual tumour (Figure 4). The spinal canal was well maintained. The patient was clinically and radiologically disease free at the 15-month follow-up visit.

DISCUSSION

Approximately 80 patients with spinal angiolipoma have been described in the literature since 1996. Liebscher, in 1901, was the first clinician to clearly describe angiolipoma of the spine.[2] In 1960, Howard and Helwig established angiolipoma as a distinct entity.[3] Spinal angiolipomas are rare benign lesions composed of mature lipocytes admixed with a vascular element. Spinal epidural angiolipomas predominantly affect women and middle-aged men.[4] A slow progressive clinical course is usually seen and, rarely, a fluctuating course may be experienced by pregnant women. The symptoms in pregnant women are probably exacerbated by weight gain, suggesting that vascular engorgement and obesity influence the evolution. The preponderance of spinal

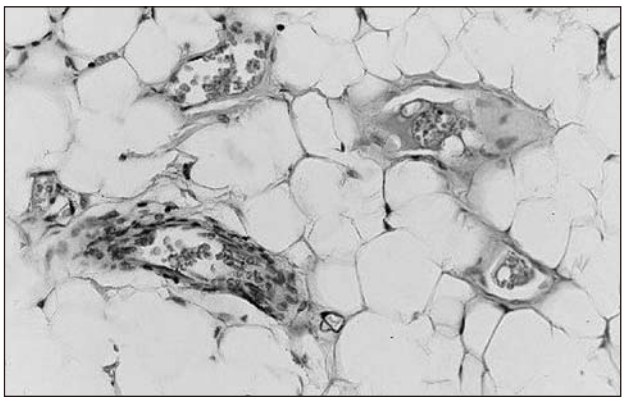

Figure 2. Histopathology showing mature adipose tissue and prominent, mostly thin-walled, vascular elements with varying numbers of fibrin thrombi in the capillaries (haematoxylin and eosin stain).

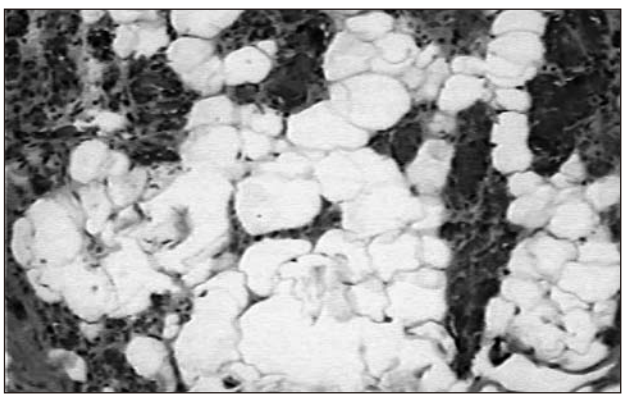

Figure 3. Mature adult fat cells without pleomorphic or myxoid degeneration, embedded in a network of vascular elements (haematoxylin and eosin stain).

angiolipomas in older peri- or postmenopausal women and the clinical exacerbation in pregnant women support a role for hormonal influence.[5]

Based on studies by Lin and Lin, angiolipomas are subdivided into 2 histological types: infiltrating and non-infiltrating.[6] Infiltrating angiolipoma is characteristically not encapsulated. The clinical behaviour is similar to that of haemangioma. The prognosis is good for both types of angiolipoma if they are removed completely.[6] Infiltrating angiolipomas are usually diagnosed in older patients. The vast majority occur in the lower extremities or in the paraspinal region, which can lead to muscular pain and neural deficits.[7] Most angiolipomas are epidural based but there are reports of spinal intramedullary angiolipoma. Non-infiltrating or circumscribed angiolipomas are encapsulated lesions limited to the subcutaneous compartment. Their size almost never exceeds 4 cm. These lesions are more common in young people, and they are equally distributed between the sexes.[8] Trabulo et al reported that 8 of 11 patients with

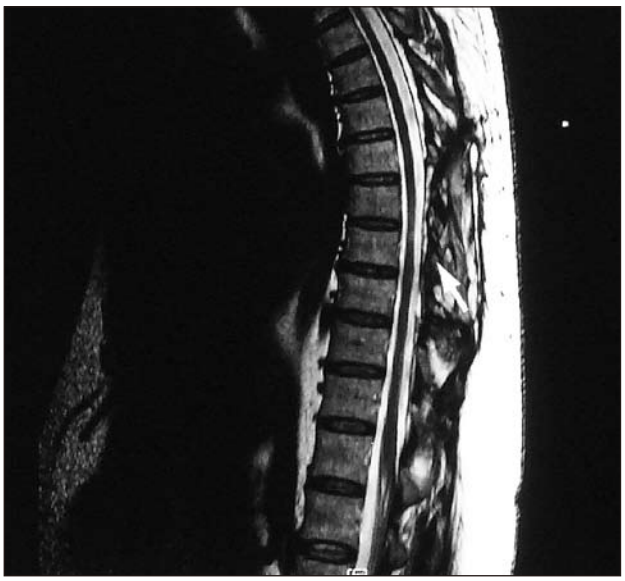

Figure 4. Postoperative magnetic resonance imaging of the spine showing intact spinal space with no residual tumour.

infiltrating angiolipomas that were partially removed experienced neurological improvement.[9]

Myelogram is abnormal in approximately 40% of patients with spinal angiolipoma, and there is a complete block in 63% of these abnormal myelograms. The computed tomography appearance of a spinal angiolipoma is of a mass that is typically hypodense relative to the spinal cord and has a variable degree of enhancement. Rarely, there may be calcification. Vertebral bodies infiltrated by disease show little or no contrast enhancement, which is a reliable indicator for distinguishing angiolipoma from haemangioma.[10] Contrast may show enhancement of the epidural angiolipoma. MRI demonstrates a fatty mass with an abundant vascular component that suggests the diagnosis of angiolipoma. The mass is usually hyperintense on T1-weighted signals, rather than on T2-weighted signals, but it can also be heterogeneous or of mixed intensity. The mass may be isointense with epidural fat on MRI.[11] Unlike other vascular tumours, angiolipomas do not typically contain vascular flow demonstrable on MRI. This may be due to the presence of capillaries and venous channels, which distinguish them from other lesions that predominantly contain arteriolar circulation. Lesions such as arteriovenous malformation usually show evidence of fast flow indicated by a flow void phenomenon.[12]

Pathologically, angiolipoma is composed of mature fat cells with numerous small blood vessels. Thin fibrous capsules with fibrous septa usually divide the mass into lobules. The presence of fibrinous microthrombi in the lumen is a diagnostic feature. The vascular component may be patchy, and is frequently accentuated in the subcapsular area. The vessels are predominantly capillaries containing some fibrin thrombi. Fibrosis may be associated with the vascular component. The cellular variant includes angiomatous tissue, which may be of the spindle-cell type, abundant in cellular areas, and may be associated with mild pleomorphism. Numerous mast cells are usually seen throughout the tumour. Degenerative changes of hyalinisation, myxoid change, and fibrosis may be present in long-term disease. Mitotic figures are usually inconspicuous, but have no clinical significance.[13] Immunochemistry can be helpful, especially CD34 and CD31, which stains the endothelium of the blood vessels. Genetic studies have generally shown no abnormalities in the karyotype of patients with this condition.[14] Angiomyxolipoma is a common differential diagnosis, which is characterised by myxoid stroma. Unlike angiolipoma, which has normal karyotype, angiomyxolipoma shares abnormal cytogenesis with lipoma, spindle-cell lipoma, and myxoma. Other differential diagnoses include metastatic well-differentiated angiosarcoma of the breast, Kaposi sarcoma, or primitive capillary haemangioma. The histogenesis is unknown but angiolipoma can be regarded as maldevelopment of the embryo. The hypothesis of a hamartomatous origin from primitive pluripotent mesenchymal cells is gaining wide acceptance. However, it may still be a neoplastic rather than a hamartomatous disease. The more invasive disease type may be a shift towards the haemangioma end of this spectrum.[15]

The mainstay of treatment is complete surgical extirpation. Non-infiltrating extradural angiolipomas are amenable to total resection, but infiltrating angiolipomas are difficult to remove entirely, especially if they are anterior or anterolateral in location.[16] Staged removal, including an anterolateral approach with spinal stabilisation, may be necessary. Since these lesions are slow growing and do not undergo malignant transformation, even partial surgical resection will provide substantial symptomatic relief. The role of external beam radiation is controversial. However, postoperative radiation has been given to 3 patients following partial excision.[17] Angiolipomas are benign entities, so the prognosis is good even with partial surgical resection, and postoperative radiation should be given with caution.

Spinal angiolipoma is a rare but well-defined clinical entity with distinguishing histology and imaging features. The treatment for both infiltrating and non-infiltrating angiolipoma is total surgical excision. Radiation therapy

is not advocated, even if there is some residual tumour after surgery.

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