

LUMBOSACRAL ANGIOLIPOMA

Case report

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ABSTRACT - We present a case of a 46-year old woman with a ventral epidural angioliopoma at the lumbosacral level with erosion of the sacrum. About ninety cases of spinal angioliopomas have been previously described in the literature, most of them situated on the thoracic region, dorsal to the dural sac. Angioliopomas can be radically excised with a good prognosis even in the presence of bone erosion. We did not find any other angioliopoma at the sacral level surgically explored in the review of the literature.

KEY WORDS: angioliopoma, spinal tumors, sacral tumors.

Angioliopoma lombosacral: relato de caso

RESUMO - Descrevemos o caso de uma mulher de 46 anos com um angioliopoma lombosacral ventral ao saco dural que erodia o sacro. Cerca de noventa casos de angioliopomas foram publicados na literatura, sendo na maioria localizados na região torácica e dorsal ao saco dural. Angioliopomas podem ser ressecados de maneira radical, com bom prognóstico mesmo na presença de infiltração óssea. Não encontramos, em revisão da literatura, nenhum outro caso semelhante de angioliopoma lombosacral com erosão do sacro abordado cirurgicamente.

PALAVRAS-CHAVE: angioliopoma, tumores medulares, tumores sacrais.

Spinal angioliopoma is a benign tumor of the epidural space. It is a rare cause of spinal cord compression, accounting for 0.14% to 1.2% of the spinal tumors¹. It is considered a distinct clinical and pathological entity traditionally grouped as a variant of lipoma¹. Characteristically the tumor lies over the dorsal aspect of the dura at the thoracic level²⁻⁷. Its port-wine color or dark brown appearance contrasts very well with the normal epidural fat^{1,8,9}. Sometimes the tumor can be more aggressive and invade the contiguous bone and adjacent soft tissues^{10,11}. We report a patient with a lumbosacral angioliopoma with bone erosion associated with a L4-L5 left sided disc herniation.

CASE

A 46-year old female with a history of ten years of low back pain had a worsening of the symptoms in the three months before diagnosis. The pain radiated down the posterior aspect of the left thigh, calf and ankle, and increased with walking and physical strength. The patient also referred a progressive numbness of the perineum. A neuro-

logical examination demonstrated a mild paresis of the plantar flexion of the left toe and hypoactive left jerk reflex. A positive straight leg-raising test at ten degree at the left side could be elicited. Superficial hypoesthesia at the lateral aspect of the left foot, buttocks and perineum was noted. An X-ray of the lumbar spine and sacrum showed erosion of the posterior aspect of the sacrum and widening of the sacral canal. A MRI scan revealed an epidural mass displacing the dural sac posteriorly, eroding the bone and projecting to the anterior sacral foramina with the sacral root. The mass was isointense in T1-weighted and hyperintense in T2-weighted and showed a homogeneous and intense enhancement with gadolinium infusion. There was also a left sided disc herniation associated with the upper limit of the tumor at the level of L4-L5 space (Fig 1).

She was submitted to a L4-L5 laminectomy and a posterior opening of the sacral canal with a wide exposure of the dura and sacral roots bilaterally. The dural sac was pushed back and ventrally compressed by a firm and large port-wine highly vascularized mass, which partially encased the sacral roots and infiltrated the sacrum (Fig 2). The mass was totally resected with preservation of the roots.

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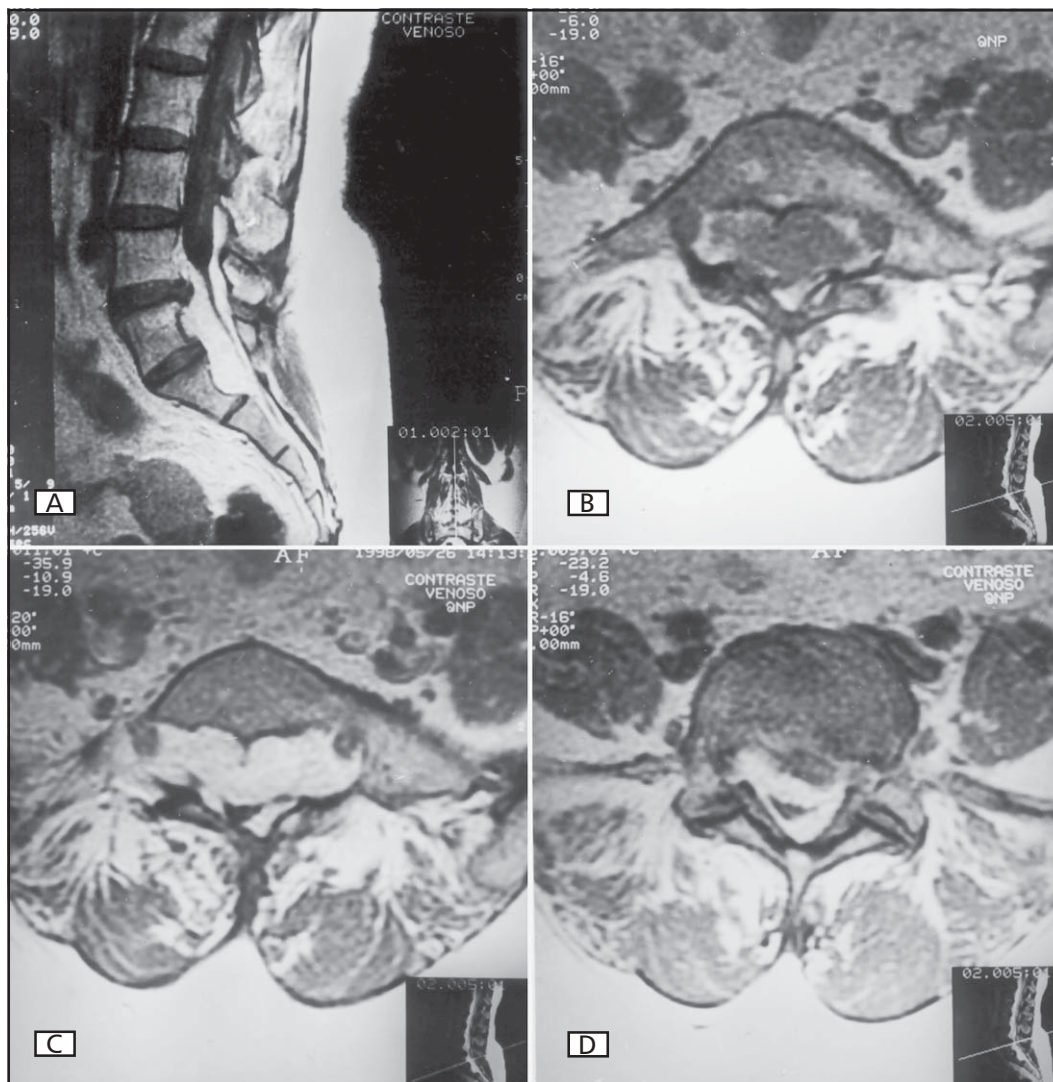


Fig 1. Preoperative MRI scans. A, sagittal T1-weighted scan with gadolinium infusion of the lumbosacral spine showing a ventral mass from L4 to S2 with homogeneous contrast enhancement. B, axial T1-weighted scan, without contrast at the S1 level. C, axial T1-weighted scan with contrast showing erosion of the sacrum and extension of the tumor to the sacral foramina. D, axial T1-weighted scan displaying the upper limit of the tumor associated with a lumbar disc herniation at L4-L5 level.

The herniated disc was dissected from the inferior aspect of the left L5 root and resected. Subcutaneous fat was harvest and then used in hemostasis and for filling the large epidural space ventral to the dural sac.

The patient recovered completely of the previous symptoms and could return to her previous activities. Two repeated MRI studies six and eighteen months after the surgical procedure confirmed the total resection and did not show any evidence of tumor recurrence (Fig 3). Histopathological analysis was characteristic of an angiolipoma and showed that the lesion was composed by adult fatty tissue intermingled with many thin walled blood vessels (Fig 4).

DISCUSSION

Berenbruch in 1890 described the first case of angiolipoma in a sixteen-year-old male confirmed in

an autopsy¹². Since then about ninety cases of angiolipoma have been reported at the literature. In the past, the finding of fatty tumors in the epidural space would be considered a matter of little interest and many cases would be unreported¹³. With the advance of MRI and the widespread use of operating microscope, even those small tumors have been distinguished from the epidural fat, and the number of cases being reported is increasing¹³. Otherwise those slow growing tumors would be unrecognized by many surgeons^{1,13}.

In the review of Preul¹ spinal angiolipomas account for 0.14% to 1.2% of all spinal tumors and 2% to 3% of extradural spinal tumors. Males and females are almost equally affected. The mean age of patients is 41.6 years (the youngest was 6 years and the oldest

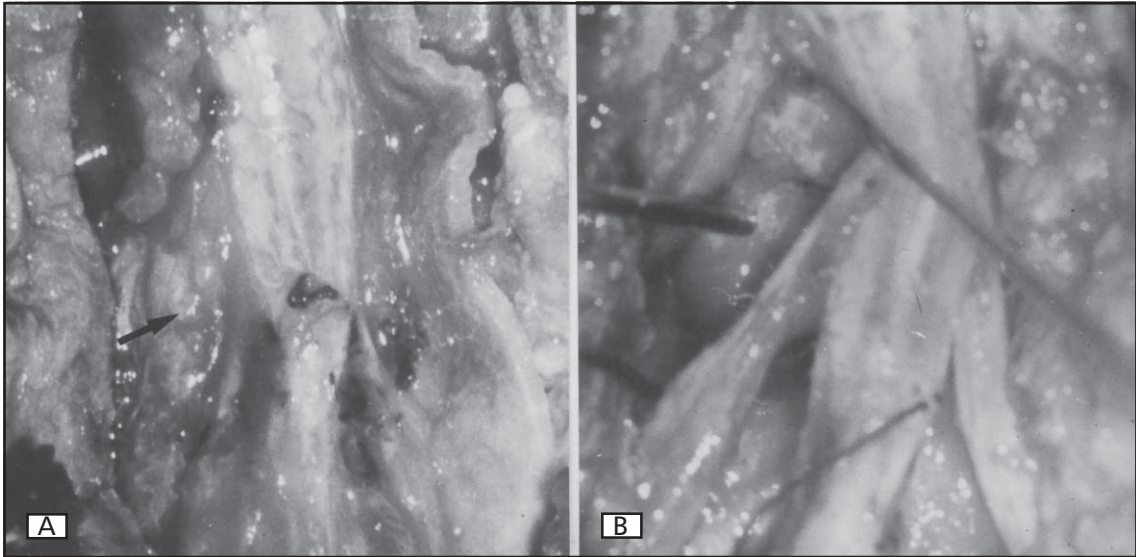


Fig 2. A, intraoperative view of the surgical field showing a large epidural mass ventral to the dural sac (arrow) encasing the sacral roots. B, same view as in A after the tumor resection. The dural sac was displaced to show the L4-L5 left side disc herniation.

72 years old)¹. If the lipomas secondary to prolonged high corticosteroid intake or following epidural injections are excluded, most of the symptomatic fatty tumors in the epidural space are angioliopomas^{4,14}. Spinal angioliopoma typically presents as a slow growing mass causing compression of the spinal cord. Lower-extremity numbness, back pain and leg weakness are frequent initial complaints¹. The mean duration of the symptoms is usually longer than one year^{4,15,16}. There were rare described cases of sudden installation of the symptoms or abrupt neurological deterioration¹⁷. Increasing of the body weight and pregnancy appear to exacerbate the symptoms due to changes in the tumor mass and volume^{1,4,16}. The concomitant presence of spinal tumors and discal hernia is probably rare. However, before the introduc-

tion of CT and MRI studies, spinal tumors might be misdiagnosed as herniated intervertebral disk and vice-versa¹³. In the present case the simultaneous occurrence of a lumbar disc herniation could be crucial in the development of the symptoms of radicular compression. The sacral location of the tumor in this case explained the atypical clinical presentation. The association of spinal angioliopoma and lumbar disc herniation was found in two previous reported cases^{9,13}.

Histopathologically angioliopomas are characterized by mature adipose tissue containing copious vascular elements that vary from sinusoids, thin-walled vessels or thick-walled vessels with proliferation of the smooth muscle layer⁴. Mitotic figures are infrequent and malignant changes have not been identi-

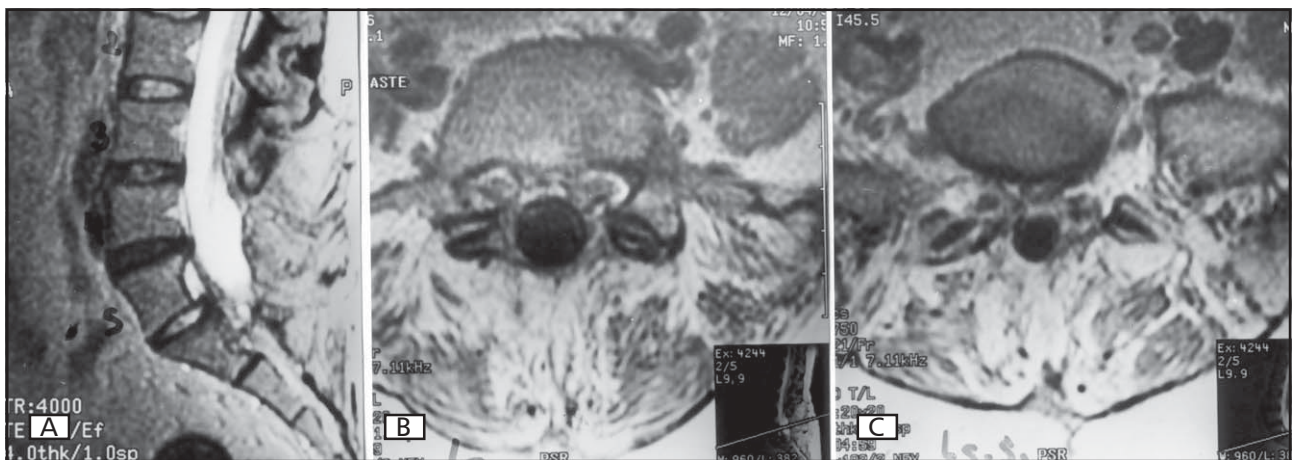


Fig 3. Postoperative MRI scans. A, sagittal T2-weighted scan. B,C axial T1-weighted scans with gadolinium infusion showing complete resection of the tumor.

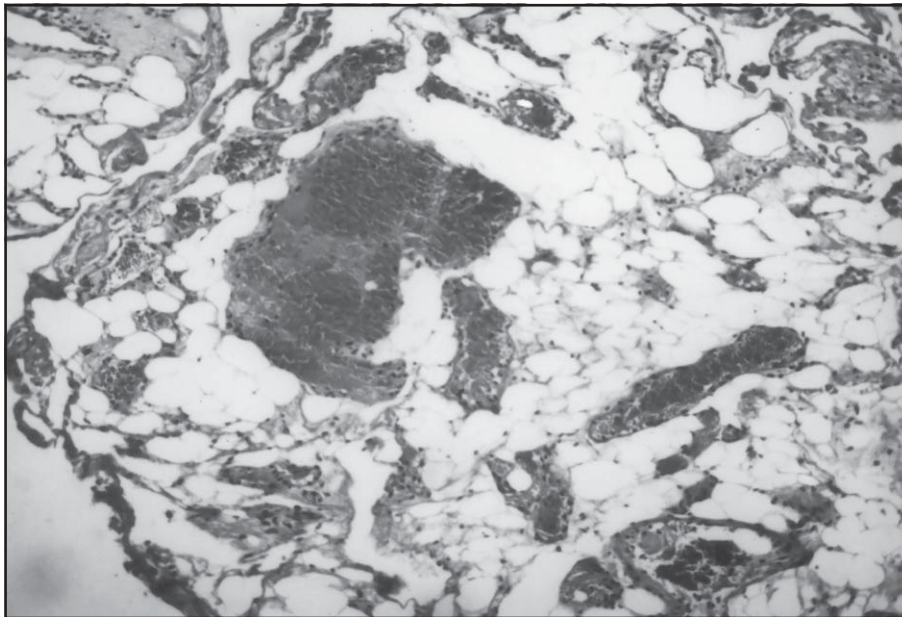


Fig 4. Photomicrograph of a tumor section showing the characteristic angioliopoma pattern with mature adipose tissue intersected by blood vessels (H&E, X 100).

fied¹⁸. They have a gross aspect that vary in color from whitish-yellow to a grayish-purple and can be usually well distinguished from the normal epidural fat^{1,9,13,19}. The pathogenesis of angioliopomas is unknown. They may result from abnormal development of the primitive, pluripotential mesenchymal cells from which adipose tissue and vascular endothelium arise or may be hamartomatous in nature^{1,16,17}.

Almost all-spinal angioliopomas are located at the epidural space. The great majority of the epidural angioliopomas are located in the thoracic region at the posterior surface of the dura^{1,4,13,16,20-23}. There have been described eight cases of angioliopomas involving the lumbar spine^{16,24}. They were frequently located anteriorly in the spinal canal in contrast with the regular dorsally located angioliopomas at the thoracic levels^{1,15,17}. Kasper et al. in 1929 reported an autopsy study with angioliopomas in the lumbosacral area (from L3-S3) and in the cervical spine canal in a 6-years old boy²⁵. Nishiura et al. described a case of left L5 and S1 radicular compression pre-operatively diagnosed as disc herniations that was explored through foraminotomies⁹. They found a hemorrhagic mass similar to our case compressing the L5 and S1 roots which was subtotally removed and the final diagnosis was hemangioliopoma. Palkovic et al.²⁸ reported the first case of intramedullary angioliopoma and Preul et al.¹ described another similar case.

In 1966 Gonzalez-Crussi et al described the first case of infiltrating spinal angioliopoma²⁷. They considered that bone infiltration was associated with a

more aggressive behavior and a worse prognosis. Subsequent studies failed to demonstrate differences in prognosis in the infiltrating angioliopomas in comparison with the non-infiltrating group¹⁶. These invasive angioliopomas are more common at the ventral aspect of the dura in the lumbar spinal canal^{13,27}. Total resection is the goal of the surgical treatment and could be achieved less frequently in the infiltrating angioliopomas specially if they are anterior and anterolateral in location^{1,28}. However, angioliopomas sub-totally resected have also a good prognosis with rare recurrences described even in the infiltrating variant¹². Almost all the patients have improvement of the neurological deficits after the surgery with total or near-total recovery¹⁷.

Plain X-rays are normal in the majority of the cases. Computed tomography studies are not conclusive in angioliopomas, with the epidural mass more often isodense or slightly hyperdense with light or absent homogeneous contrast enhancement^{1,29}. In MRI studies the extradural components of angioliopomas are isointense or hyperintense in T1-weighted images, probably due to the fat component and usually hyperintense in T2-weighted images. Those areas show an early enhancement after gadolinium administration and are considered the vascular component^{1,16}. Slow-growing masses dorsally located in the thoracic spinal canal with these MRI findings should be suspected of angioliopoma. With the widespread availability of MRI, spinal angioliopomas have been more frequently recognized and diagnosed.

In conclusion, spinal angioliipomas are rare tumors with a benign behavior. When localized in the thoracic spinal canal they have usually a well-defined clinical-radiological presentation with MRI studies. The authors describe a case of a lumbosacral angioliipoma diagnosed in vivo and totally resected with a good outcome. To the best of our knowledge we could not find any other angioliipoma with erosion of the sacrum reported in the literature.

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