



# First report of two synchronous but separately placed intramedullary angioliipomas located in the dorsal spine causing progressive compressive myelopathy: management strategies and outcome review

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## ABSTRACT

Spinal angioliipoma is a benign lesion and presents with compressive myelopathy. Typically, it is located in epidural compartment. However, intramedullary angioliipoma is extremely uncommon, and till date only eight cases are reported in the literature and all reported cases had isolated solitary lesion. Authors report an interesting case of intramedullary spinal angioliipoma (ISAL) in a - 48- years male, presented with compressive myelopathy, magnetic resonance imaging study revealed presence of two separate angioliipomas, which were located at D8-D9 and D10-D12 vertebral levels respectively, underwent successful near total surgical resection with good neurological outcome. Current case represents first of its kind in the western literature. Management of such rare pathology along with pertinent literature is briefly discussed.

## INTRODUCTION

Angioliipoma is a rare benign tumour most commonly found in the subcutaneous tissue of the trunk and extremities but other sites are also reported. [1] Histopathologically, it consists of mature fatty tissue interspersed with abnormal vascular elements. [2]. Spinal angioliipoma represent a distinct clinical and pathological entity, which account for approximately 0.04-1.2 % of all spinal axis tumours. [3]. Spinal angioliipoma is mostly located in the epidural space and accounting for 2-3 % of spinal tumours. Only eight cases of intramedullary lipoma are reported in literature. [4-10]. Due to rarity of lesion, these cases are usually missed as differential causes of compressive myelopathy, which have good prognosis following early surgical resection. Authors report a case of intradural spinal angioliipoma (ISAL), which was located in intramedullary- subpial located, who underwent successful surgical excision with good neurological recovery in the postoperative period.

## CASE ILLUSTRATION

A 48 -year- male presented to our neurosurgical outpatient services with complaint of progressive spastic paraparesis for last two years. He also noticed tingling and numbness in involving trunk below

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umbilicus and both lower limbs. He had no associated history of diabetes mellitus, hypertension, coronary artery disease or pulmonary tuberculosis. Examination on admission, revealed healthy male with vital stable, had scissoring gait, increased tone with motor power of 4/5 in bilateral lower limbs along with graded sensory loss below the level D10 dermatomes, which was comparatively more marked on left side. The deep tendon jerks were brisk with ill sustained patellar and ankle clonus with plantars was extensor response bilaterally.

MR Imaging of the spine revealed, presence of two separate intradural intramedullary mass lesions located at D8 to D9 level and D10 to D12 levels (Fig-1), showing heterogeneous hyper-intense signals on T1 and T2 weighted image, showing with characteristics suggestive of angioliipomas. Patient was diagnosed to two separate D8-D10 and D10-D12 level spinal angioliipoma (Fig-2, 3). He was advised for surgical management with intraoperative electrophysiological monitoring.

He was taken up for surgery in prone position

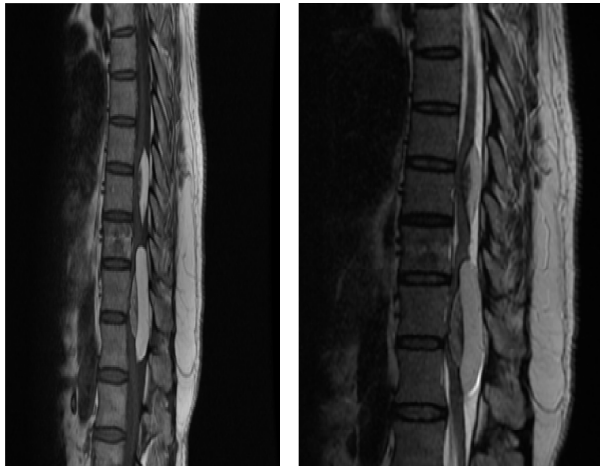


FIGURE 2. Dorsal spine, Magnetic resonance imaging, sagittal section, T2WI showing heterogeneous mass, vascular component showing hyperintense signal on T2-weighted image.

FIGURE 3. Magnetic resonance imaging of dorsal spine, T1Weighted axial section image showing ventrally located angioliipoma.

FIGURE 4. Magnetic resonance imaging of dorsal spine, T2 Weighted image, axial section showing ventrally located angioliipoma with heterogeneous hyperintense signal.

## DISCUSSION

Spinal angioliipoma represents a distinct, benign lesion and characterized by presence of mature fat cells interspersed with excessive abnormal vascular proliferations, commonly observed in the subcutaneous locations. However, spinal angioliipoma represents a separate entity with extremely rare

under general anaesthesia, intraoperative level was localized with image intensifiers, and underwent D7-L1 laminectomy, after laminectomy bulging of dura sac was noted, however, no epidural mass was observed. But dura was bulging. Midline durotomy was carried out; two separate lesions were noted at D8-D9 and D10- D12 levels respectively. Eccentrically placed mass was observed, after pial incision, pale coloured fat with larger branching vessels were noted, near total decompression with Cusa was carried out. The lesions were intra medullary in location containing lipomatous mass interspersed with dilated veins. Subtotal excision was carried out as further resection attempt led to electrophysiological abnormality. After securing hemostasis, primary dural closure was carried out and the wound was closed in layers. He tolerated surgical procedure well with no appearance of fresh neurological deficits and discharged on fifth day following surgery. At last follow-up six months after surgery, has mild improvement in motor power, however sensory deficit was persisting.

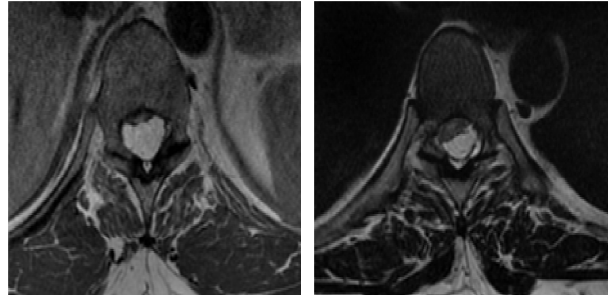


FIGURE 1. Magnetic resonance imaging, T1WImage, dorsal spine sagittal section showing two separate intradural intramedullary lesion at D8-D9 and D10-D12 causing severe distortion of spinal cord with secondary canal stenosis. Vascular component showing hypointense and fatty component with hyperintense signals.

occurrence, predominantly located in the epidural space but extremely rarely can also occur in intradural spinal compartment. Liebscher was credited to first describe the angioliipoma. [10]

Spinal angioliipoma may contain variable proportions of mature adipose tissue interspersed with abnormal vascular elements. The proportion of

adipose to vascular tissue is variable, being predominantly in lipomatous lesion to the very scanty dense mixed with vascular and stromal elements. [11] Benvenuti-Regato et al. carried out a detailed literature search in 2015 and could find out only a total of 177 patients suffering with spinal epidural angioliipoma. [12] the average age for epidural spinal angioliipoma was 46 years with predilection for female gender [59%]. The most common presenting symptom was paraparesis (30 %), others were thoracic or low back pain in (24. %). 12 The majority of epidural spinal angioliipoma occurred, in the order but and least involved site was cervical spine 2.3%. [12]

Histopathologically, ISAL lesions consist of mature adipocytes and network of branching capillary-sized vessels, which usually contain fibrin thrombi and usually positivity for CD31, Factor VIII and Factor XIII a [13].

The clinical feature of ISAL is compressive myelopathy related to spinal cord compression and usually presents early. Case with long standing history may show rapid or acute deterioration as result of enlarging vessels, engorged vein or vascular steal phenomenon, venous thrombosis or occurrence of fresh haematoma. [14].

X-ray spine may show enlargement of spinal canal and moulding of adjoining lamina. Angioliipomas typically displays heterogeneous slight hypointense signal compared to typical epidural fat on T1-weighted images and inhomogeneous enhancement on contrast is observed in T1-weighted images using fat-saturation techniques. [2, 15]. A high vascular content correlates with the presence of large hypointense regions on T1-weighted images. ISALs are typically hyperintense on non-contrast T1-weighted images relative to other spinal tumors or epidural lipomatosis. [16].

additionally, spinal lipomas occur most commonly in the midline of the lower. [15]

Surgery is the main treatment modalities; however, the extent of surgical resection is variable in reported cases, varying from simple biopsy, and subtotal resection, and near total resection and complete surgical resection have been carried out. Surgical resection may be augmented if surgery is carried out with electro-physiological monitoring. In our case we could only manage with subtotal resection, as this is limitation, as further attempt of resection led to neurophysiological alteration as, we had to stop resection.

Garg et al. reported two cases of ISAL who underwent near total surgical excision.2 Maggi et al. in 1996 reported an eight-year-old female child suffering with D11-L2 intramedullary angioliipoma, who underwent total surgical resection. [7]. Klisch et al. analysed in a 34- year -old female, with intramedullary angioliipoma at C6-D4 level, underwent partial resection and observed the clinical presentation is nonspecific, however, MRI findings was helpful in the preoperative diagnosis and helped in planning of surgical management. [5]. similarly, a 36-year female suffering with C6-D2 intramedullary angioliipoma, also underwent partial surgical resection. [9].

Prasad et al reported a -26-year- female harbouring D5-D9 ISAL, who underwent successful near total surgical excision. [4]. Preul et al. reported a 36-year female with ISAL at D7-D11 underwent only subtotal resection with improved postoperative outcome. [6]. However, Weil et al advised biopsy as treatment modality in a case with large non -resectable ISAL, and illustrated a case- 27-year-old female with extensive ISAL at C 5-D8 was managed with surgical biopsy for confirmation of diagnosis. [8].

TABLE 1. Review of Previously reported intradural angioliipoma.

S.no.	Author/ references	Year	Age/sex	location	Surgical management	Neurological outcome
1	Prasad and Sinha <sup>4</sup> .	2014	26 year /male	D5-D9	Neat total excision	Incomplete recovery
2a	Garg et. al. <sup>2</sup>	2002	26 year/male	D3-D7	Subtotal resection	Incomplete recovery
b		2002	28year/male	C6-D2	subtotal	Incomplete recovery
3	Klisch et al. <sup>5</sup>	1999	34 year/ female	C6-D4	Partial resection	Good outcome
4	Maggi et al. <sup>7</sup>	1996	8 year/ female	D11-L2	total resection	Good outcome

5	Preul et al. <sup>6</sup>	1993	36 year /female	D7-D11	subtotal	Good outcome
6	Weill et al <sup>8</sup>	1991	27 year/female	C5-D8	Only biopsy	No improvement
7	Palkovic et al. <sup>9</sup>	1988	27 year/male	C6-D2	partial	Not available
	Current case	2017	48year/male	D8-D9 and D10-D12	Subtotal resection	Incomplete recovery

## CONCLUSION

Spinal angioliopoma is a benign lesion and surgery can provide good outcome. Spinal angioliopoma is mostly located in epidural compartment and only eight cases of intramedullary angioliopoma is reported. Preoperative MRI and detailed history and clinical evacuation are helpful in preoperative diagnosis. Current case is extremely rare as two isolated intramedullary angioliopoma were present separately, and underwent successful surgical resection, representing first case- report in the western literature. A high degree of suspicion for spinal angioliopoma should also be kept as differential of intramedullary pathology with typical neuroimaging findings.

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