CASE REPORT

Spinal angioliipoma manifesting with apoplexy

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SUMMARY

We report a 58-year-old man presenting with acute paraplegia. MRI showed a haematoma within a well-defined epidural lesion at C7-D1. Intraoperatively, organised epidural haematoma surrounded by tumour tissue was found. The final histopathology report was angioliipoma. The patient had dramatic recovery. Angioliipomas should be considered in the differential diagnosis of acute paraplegia when imaging shows well-circumscribed haematoma.

BACKGROUND

Spinal angioliipomas account for 0.04–1.2% of spinal tumours and 2–3% of extradural spinal tumours. These generally present as gradually progressive compressive spinal cord lesions.¹ Four cases of sudden onset of paraplegia have been reported in the literature (table 1). We report acute paraplegia due to cervicodorsal angioliipoma with bleeding in a patient on aspirin. We review apoplexy and unusual presentations of spinal angioliipoma.

CASE PRESENTATION

A 58-year-old hypertensive man had a sudden onset of neck and interscapular pain, paraplegia and sphincter incontinence for 48 h. There was no history of trauma. On examination he had paraplegia, weakness of both hand grips and anaesthesia below D3 level. He was on long-term treatment with aspirin due to ischaemic heart disease.

INVESTIGATIONS

MRI showed a well-defined, 3.3×1.0×1.4 cm, extradural lesion compressing the posterior aspect of the spinal cord at C7 and D1 levels (figure 1). The lesion was isointense to hyperintense on T1-weighted image and hyperintense centre with isointense to hypointense periphery on T2-weighted imaging (figure 1A,B). The lesion showed heterogeneous enhancement on gadolinium administration (figure 1C). His bleeding time was prolonged which was corrected with fresh platelets.

DIFFERENTIAL DIAGNOSIS

We had preoperative differential diagnosis of apoplexy in an extradural tumour such as plasmocytoma, metastasis or cavernoma or aspirin-induced spontaneous haematoma.

TREATMENT

The patient was taken up for emergency C7-D1 laminectomy with complete excision of the lesion. Intraoperatively there was a non-suckable, dark clot with soft consistency in the epidural space

encapsulated by vascular grey tissue. The lesion was completely removed in a piecemeal manner under the operating microscope.

OUTCOME AND FOLLOW-UP

The patient had rapid recovery in postoperative period. At follow-up of 2-years the patient had no deficit. Histology report revealed an angioliipoma with intratumoural bleeding and showed a haematoma surrounded by a pseudocapsule consisting of proliferating blood vessels and loose areas of mature adipose tissue with few foci of haemorrhage and congested blood vessels (figure 2A, B).

DISCUSSION

Spinal angioliipoma was first described by Berenbruch in 1890. These may present anywhere in the spinal canal but most frequently found in the thoracic region.¹ More than 95% tumours are epidural in location. These have female predominance, most being diagnosed during the fifth decade of life. The usual presentation is one of the progressive spinal cord or nerve root compression over many months; however, some cases may be present with remitting and relapsing course.^{2–3} Rarely do they present acutely with sudden onset of back pain and paraplegia (table 1).

Review of the literature show some rare presentations of spinal epidural angioliipoma. Raghavendra *et al* have presented a case of acute neck pain with bifrontal headache on the background of chronic limping, which was diagnosed as a spinal subarachnoid haemorrhage due to spinal angioliipoma. The authors could not give an explanation for spontaneous subarachnoid haemorrhage.⁴ Boockvar *et al* reported subacute onset of myelopathy due to spontaneous venous thrombosis in a case of angioliipoma. The authors quoted the cause of subacute paraparesis as venous stasis and tumour engorgement.⁵ Gelabert-Gonzalez *et al*, in their review of literature of 118 cases, reported pregnancy as an aggravating factor in 11 (15.7%) of the 70 female patients. They quoted interference with spinal venous drainage due to an increased epidural venous pressure from compression of pelvic and abdominal veins, the redistribution of blood, the increase in extracellular fluid, the forceful Valsalva manoeuvres during vaginal delivery, hormonal influences leading to increased adipose tissue production, which in turn lead to an increase in the size of spinal angioliipoma as the probable causes for worsening of symptoms during pregnancy.⁶ These symptoms regress following completion or termination of pregnancy.^{3–6–7} Petrella *et al* reported acute paraplegia with fever in a 16-year-old boy. The lesion was found to be

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Table 1 A list of the previously reported cases of acute onset paraplegia due to apoplexy in angioliipoma

Author	Age/sex	Precipitating factor	Symptoms	Duration (h)	Location	Treatment	Recovery
Anson <i>et al</i> ¹¹	65/M	HTN, IHD	Backache, acute paraplegia	24	D1-D6	Surgery	Partial
Akhaddar <i>et al</i> ⁹	47/M	Strenuous exercise	Backache, acute paraplegia	Not described	D2-D3	Surgery	Complete
Sankaran <i>et al</i> ¹²	77/M	Trauma	Progressive paraparesis	48	D8-D11	Surgery	Complete
Tsutsumi <i>et al</i> ¹⁰	26/F	Pregnancy	Backache, acute paraplegia	Not described	D3-D4	Surgery	Complete
Present case	58/M	HTN, IHD, aspirin	Backache, acute paraplegia	48	C7-D1	Surgery	Complete

F, female; HTN, hypertension; IHD, ischaemic heart disease; M, male.

intratumoral abscess in an angioliipoma. They could not find any precipitating factor for abscess formation and suggested the haematogenous spread of systemic infection to the spinal epidural space.⁸

There are five reports, including the present case, of acute paraplegia in an angioliipoma due to tumour apoplexy. Patients are aged between 26 and 77 years. Although incidence of angioliipoma is more in women, apoplexy in angioliipoma is more common in men (four of five cases). The location of the tumour was in the dorsal region except our case. All the patients presented with acute back pain and paraplegia. Three patients were operated within 48 h of presentation of which two had complete recovery and the third had partial recovery. The authors have not mentioned the reason for partial recovery in spite of operating in less than 24 h of presentation. In the remaining two cases the duration of onset of symptoms has not been mentioned. They too had complete recovery (table 1).

Various reasons have been mentioned in the literature for acute presentations such as rapid tumour expansion due to enlarging blood vessels,⁵ local vascular steal by tumour vessels leading to reduced blood to the cord,⁷ tumour necrosis and sudden haemorrhage into the tumour (table 1). Akhaddar *et al* stated that haemorrhage into an angioliipoma can occur due to vigorous exercise which would augment blood flow size into the tumour to produce epidural bleeding and compression of the

cord.⁹ In a case report by Tsutsumi *et al*, the patient was pregnant and the above said factors might have led to the bleeding in the tumour.¹⁰ In a case presented by Anson *et al*, the patient was a known case of hypertension and ischaemic heart disease. The authors have not mentioned about antiplatelet or anticoagulation therapy or precipitating factors for the bleeding.¹¹ Our patient had hypertension and ischaemic heart disease and was on antiplatelet therapy. He did not have any history of trauma or vigorous exercise which could have precipitated bleeding. We believe that in abnormal vasculature of the tumour which is fragile, vascular changes due to prolonged hypertension, antiplatelet therapy (a risk factor for spontaneous spinal epidural haematomas) might have acted as precipitating factors. The exact source of blood is not clear; an arterial source of clot seems to be more probable than venous because of the rapid onset and progression of symptoms.

Irrespective of the cause for apoplexy, emergency decompression is the treatment of choice and is curative. Rapid correction of the coagulopathy should be done in patients who are on antiplatelets and anticoagulants. Most of the patients have excellent clinical recovery.

Figure 1 MRI sagittal scan of the cervicodorsal region. (A) T1-weighted image showing a biconvex extradural lesion of heterogeneous intensity compressing the spinal cord at C7 and D1 level (B) T2-weighted image showing an oval lesion with hyperintense core and heterogeneous periphery compressing the cord. (C) Postgadolinium T1-weighted image showing heterogeneous contrast enhancement in the peripheral part of the lesion.

Figure 2 Histopathological photomicrograph showing mature adipocytes (shown by arrow) and small-medium calibre blood vessels (shown by triangle) suggestive of angioliipoma (H&E stain, A x10, B x40).

Learning points

- ▶ Spinal angioliipoma may present with acute paraplegia.
- ▶ Angioliipoma with apoplexy should be considered in differential diagnosis of well-circumscribed epidural haematoma with cord compression.
- ▶ Emergency excision of the lesion is curative with excellent outcome.

Competing interests None.

Patient consent Obtained.

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