

Review article

Spinal angiolioma: Presentation of two cases and review of the literature for the years 2012–2017



Sotirios Apostolakis^{a,*}, Aikaterini Karagianni^a, Athanasios Mitropoulos^a, Constantinos Mantas^a, Ioannis Mavridis^a, Panagiotis Filias^a, Christos Eftychiadis^b, Konstantinos Vlachos^a

^a Department of Neurosurgery, 'KAT' General Hospital of Attica, Athens, Greece

^b Department of Pathology, 'KAT' General Hospital of Attica, Athens, Greece

ARTICLE INFO

Article history:

Received 20 November 2018

Accepted 17 April 2019

Available online 8 June 2019

Keywords:

Embolisation

Fine needle biopsy

Infiltrative spinal angiolioma

Spinal fusion

ABSTRACT

Introduction and objectives: Angiolipomas of the spinal canal are a rare condition of unknown origin. They are considered histologically benign; however, some have the potential to infiltrate adjacent structures. The aim of this systematic review was to suggest a potential mechanism for the pathogenesis of spinal angiolipomas, along with a useful approach for their preoperative management.

Materials and methods: A literature review of cases of spinal angiolipoma was performed. In addition, two of the cases encountered in our practice are presented. The first case refers to a 35-year-old male patient with a history of spinal fusion because of a T9 fracture, while the second concerns a 46-year-old male patient with an epidural mass extending outside the spinal canal, who underwent fine needle biopsy and embolisation of its feeding vessel. **Results:** From the review of the literature performed, we were unable to identify any correlation between the infiltrative potential and the patients' demographic and tumour characteristics.

Conclusions: Angiolipomas are considered to be sporadic, yet theories concerning their pathogenesis include reaction to harmful stimuli and congenital malformation of the adipose tissue. Fine needle biopsy may be mistakenly considered non-diagnostic, due to the presence of well-differentiated adipocytes.

© 2019 Sociedad Española de Neurocirugía. Published by Elsevier España, S.L.U. All rights reserved.

* Corresponding author.

E-mail address: sotapostolakis@gmail.com (S. Apostolakis).

<https://doi.org/10.1016/j.neucir.2019.04.005>

1130-1473/© 2019 Sociedad Española de Neurocirugía. Published by Elsevier España, S.L.U. All rights reserved.

Angiolipoma espinal: descripción de 2 casos y revisión de la bibliografía durante los años 2012-2017

RESUMEN

Palabras clave:

Embolización
Biopsia con aguja fina
Angiolipoma espinal infiltrante
Artrodesis vertebral

Introducción y objetivos: Los angiolipomas del canal vertebral son una enfermedad rara de origen desconocido. Se consideran histológicamente benignos, aunque en algunos casos existe la posibilidad de que se infiltren en estructuras adyacentes. El objetivo de esta revisión sistemática es sugerir un posible mecanismo para la patogenia de los angiolipomas espinales, junto con un enfoque útil para su tratamiento preoperatorio.

Materiales y métodos: Se realizó una búsqueda bibliográfica de los casos de angiolipomas espinales. Además, se presentan 2 de los casos encontrados en nuestra práctica clínica. El primer caso corresponde a un paciente varón de 35 años con antecedentes de artrodesis vertebral debido a una fractura en T9, mientras que el segundo corresponde a un paciente varón de 46 años con una masa epidural que se extendía fuera del canal vertebral, al que se realizó una biopsia con aguja fina y una embolización del vaso nutricio.

Resultados: A partir de la revisión bibliográfica realizada, no pudimos identificar ninguna correlación entre el potencial de infiltración, los datos demográficos y las características de los tumores de los pacientes.

Conclusiones: Los angiolipomas se consideran esporádicos, existiendo, no obstante, teorías referentes a su patogenia que incluyen la reacción a estímulos nocivos y la malformación congénita del tejido adiposo. La biopsia con aguja fina puede considerarse erróneamente como no diagnóstica, debido a la presencia de adipocitos bien diferenciados.

© 2019 Sociedad Española de Neurocirugía. Publicado por Elsevier España, S.L.U. Todos los derechos reservados.

Introduction

Angiolipomas of the vertebral canal are a fairly rare condition accounting for 0.14–1.2% of all spinal axis tumours and less than 3% of all extradural tumours.^{1,2} These tumours are not exclusively found in the vertebral canal but they have also been described in other systems as well, like the breast,³ gastrointestinal tract⁴ and the respiratory tract,⁵ just to name a few. Even though it has already been over a century since the first description of this clinical entity⁶ and more than 50 years since the documentation of its microscopic features,⁷ little is known about its pathogenesis. Most cases are considered to be sporadic, while theories about their pathogenesis include reaction to harmful stimuli and congenital malformation of the adipose tissue.^{8–10} In addition, cases of familial angiolipomatosis have also been described.^{11,12} The mean age of diagnosis is within the fifth decade of life, regardless of gender,¹ however a female to male ratio of about 1.4 has been documented.¹³

Interestingly, spinal angiolipomas are predominately found in the midthoracic region, with over half of the cases located there,^{1,13} whereas purely cervical or lumbar localisation are extremely rare, with less than 10% of all epidural angiolipomas reported below the T12 level.¹⁴ The reason for this predilection has not been clarified yet. It is also noteworthy, that thoracolumbar localisation has been reported in 6 cases.¹⁵ In addition, due to the benign and slow-growing nature of the mass, most patients present with tumours extending up to four vertebral bodies² and with signs and symptoms of spinal cord compression. Nevertheless rare cases of paraplegia or paraparesis because of spontaneous

epidural^{16–18} or subarachnoid haematomas¹⁹ or even intratumoural abscesses²⁰ have also been reported as initial clinical presentations of the mass. Occasionally, the tumour may infiltrate the vertebral bodies or the adjacent soft tissues, a condition which is more common in masses of the anterior compartment.²¹ To date, no definitive conclusion exists regarding the origins of this infiltrative subtype, in other words, if it is primarily epidural extending outside the vertebral canal, or vice versa.²¹

In the present paper, we report two cases of spinal angiolipomas, one in a patient with an atypical presentation of an infiltrating epidural angiolipoma and the other in a patient with a history of spinal fusion. Through the presentation of these cases and the systematic review that was conducted, our aim is to suggest a useful approach for the preoperative management along with a potential mechanism for the pathogenesis of this rare tumour.

Material and methods

In order to record the number of infiltrating angiolipomas, not having been already documented in previous review articles, a Scopus search from 2012 through the 31st of August 2017 was performed, using the term “spinal angiolipoma”. Prior to 2012 two works have reviewed the available literature, one for the period 1890 and 2006¹³ and one for the period 2007–2011.²² Only original manuscripts, written in English were considered. In order to be included, the manuscript needed to contain information about patient demographics, tumour location (axial and coronal) and evidence of infiltrating potential.

The citations of identified articles were reviewed, and additional manuscripts were included when inclusion criteria were fulfilled. Two of the authors independently assessed the quality of the studies and any disagreement was resolved by discussion and consensus. The data collected were patient age and gender, axial location of the tumour, which served as a tumour size index by measuring the spinal levels between which the mass extended, anteroposterior tumour location and evidence of infiltration of the surrounding tissues.

Student t-test was used to test for differences between groups and Spearman's correlation to examine potential correlations between the parameters. The level of statistical significance was $p < .05$. Figures were created using Adobe Illustrator CS3 (Adobe Systems, 2007) and MATLAB 2016 (The MathWorks Inc., 2016).

Case 1

A 35 years old male patient presented in the outpatients department with reduced lower limb muscle strength bilaterally, lower limb numbness and gait instability. He also reported

erectile dysfunction and faecal incontinence. He reported his symptoms to have started following a motor vehicle accident, which resulted in a tibial plateau fracture that was managed surgically, about one year prior to his evaluation in our hospital and his condition had considerably deteriorated in the previous few days. In addition, at the age of 19 the patient underwent spinal fusion at the levels of T7-T11 vertebrae because of a T9 fracture, again resulting from another motor vehicle accident. Upon presentation he had decreased muscle strength of the lower limbs bilaterally, hypoesthesia below the level of T6, loss of proprioception, increased muscle tendon reflexes of the lower limbs and positive Babinski's sign bilaterally.

Computed tomography (CT) and magnetic resonance imaging (MRI) scans of the thoracic region with intravenously administered contrast revealed the presence of an inhomogeneously enhanced, epidural mass with soft tissue characteristics at the levels of T7-T9 measuring about 9 cm in its maximal diameter, causing stenosis of the spinal cord (Fig. 1). The mass was hypointense on T1 weighted (W) and iso- to hyperintense on T2W images.

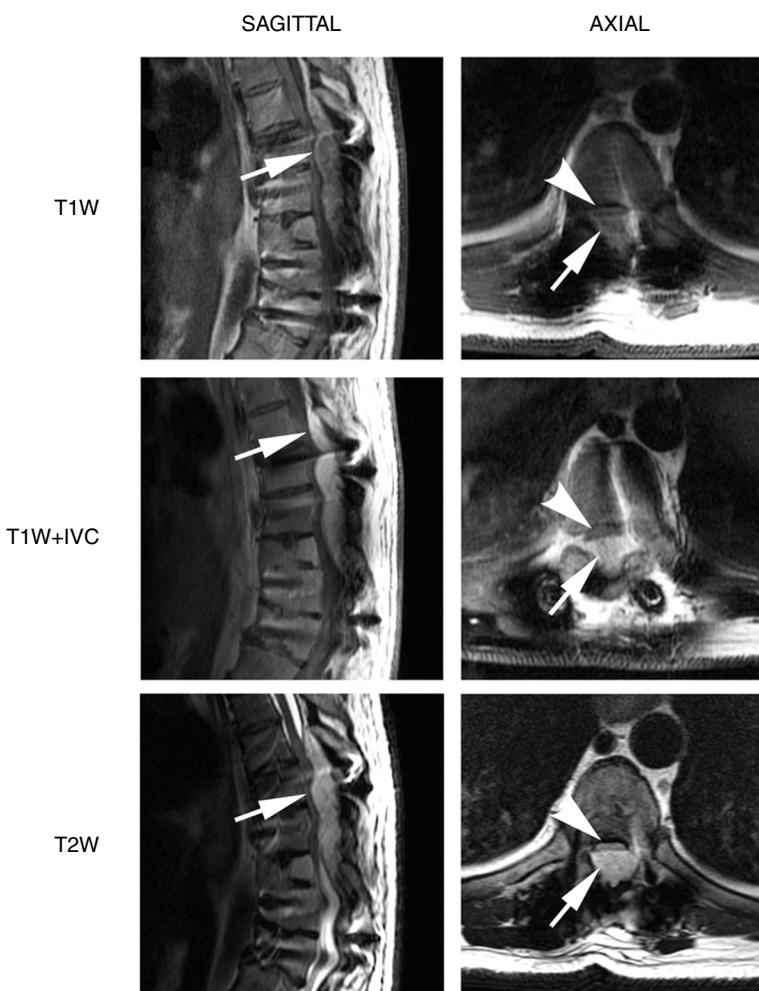


Fig. 1 – MRI of the thoracic area showing an epidural mass at the levels of T7-T9 that was hypointense on T1W and iso- to hyperintense on T2W (arrow) causing significant compression of the spinal cord (arrowhead). The mass was inhomogeneously enhanced after administration of intravenous contrast. All axial images were taken at the level of T8.

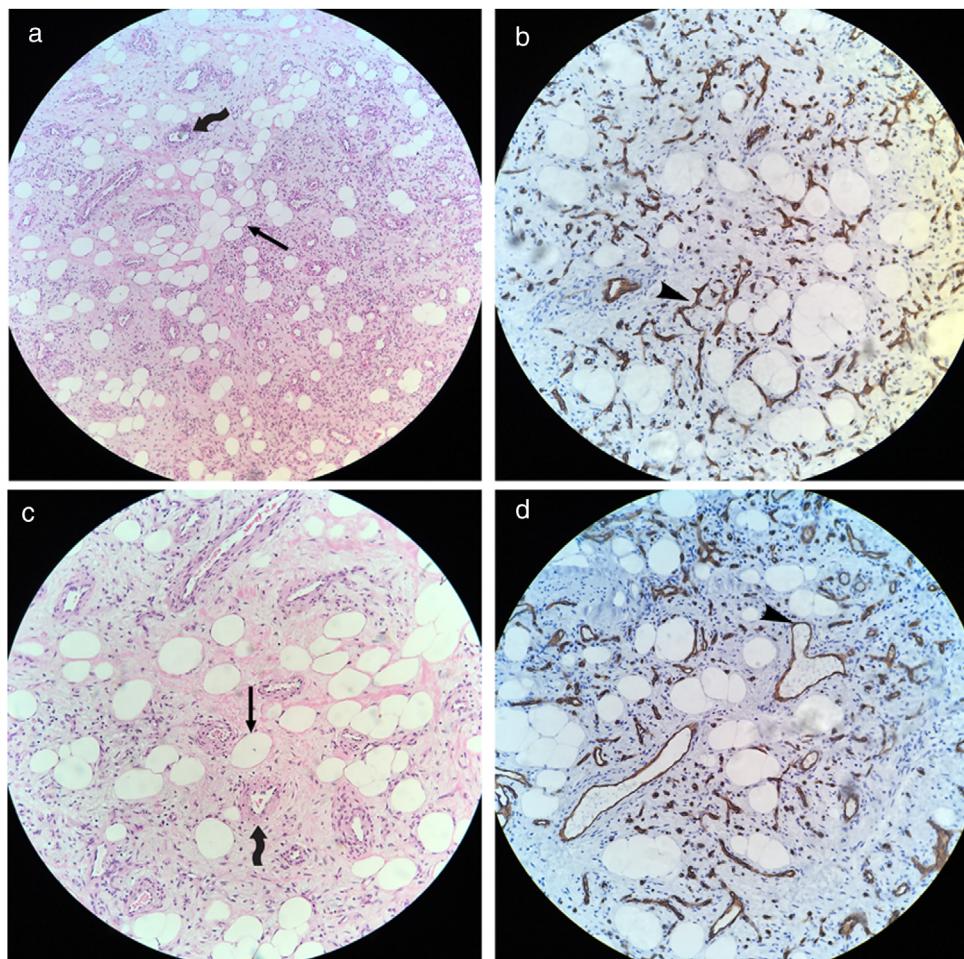


Fig. 2 – Photomicrograph of the pathological specimens of the presented cases. Top row (images a, b): Case 1, bottom row (images c, d): Case 2. Histologic examination of the excised tissue (image a: H&E stain, magnification 4x, image c: H&E stain, magnification 20x) revealed the presence of mature adipocytes without indications of atypia (arrow) and thin-walled capillary sized vessels containing fibrin thrombi (curved arrow), with CD34 immunostain highlighting the thin-walled capillary-sized vessels (images b and d: arrowhead, magnification 20x).

Due to the recent deterioration of his neurologic status, no further work-up was conducted and the patient was scheduled for operation and laminectomy with total excision of the mass was performed. Macroscopically, the tumour was multilobular and yellowish in colour. Histological examination revealed the presence of mature adipocytes without indications of atypia and an abundant network of capillaries containing fibrin thrombi (Fig. 2a, b), findings consistent with the diagnosis of epidural angiomyoma. The patient underwent postoperative MRI that demonstrated decompression of the spinal cord (Fig. 3). The recovery of the patient was uneventful, with partial remission of the symptoms and discharge on postoperative day seven. The patient was reevaluated 6 months postoperatively with MRI that demonstrated full decompression of the spinal canal (Fig. 3) and complete remission of his symptoms was identified clinically.

Case 2

A 46 years old male patient presented in the outpatients department with low back pain, lower limb numbness and gait instability of 6 months duration. Upon presentation he had decreased strength of the right quadriceps muscle, increased muscle tendon reflexes of the lower limbs and positive Babinski's sign bilaterally. His history was insignificant of any pathology.

CT and MRI scans with IV contrast of the thoracic and lumbar regions revealed the presence of an inhomogeneously enhanced, epidural mass with soft tissue characteristics, extending outside the vertebral canal through the intervertebral foramina (Fig. 4). The epidural part was hypointense on T1W and iso- to hyperintense on T2W, while the

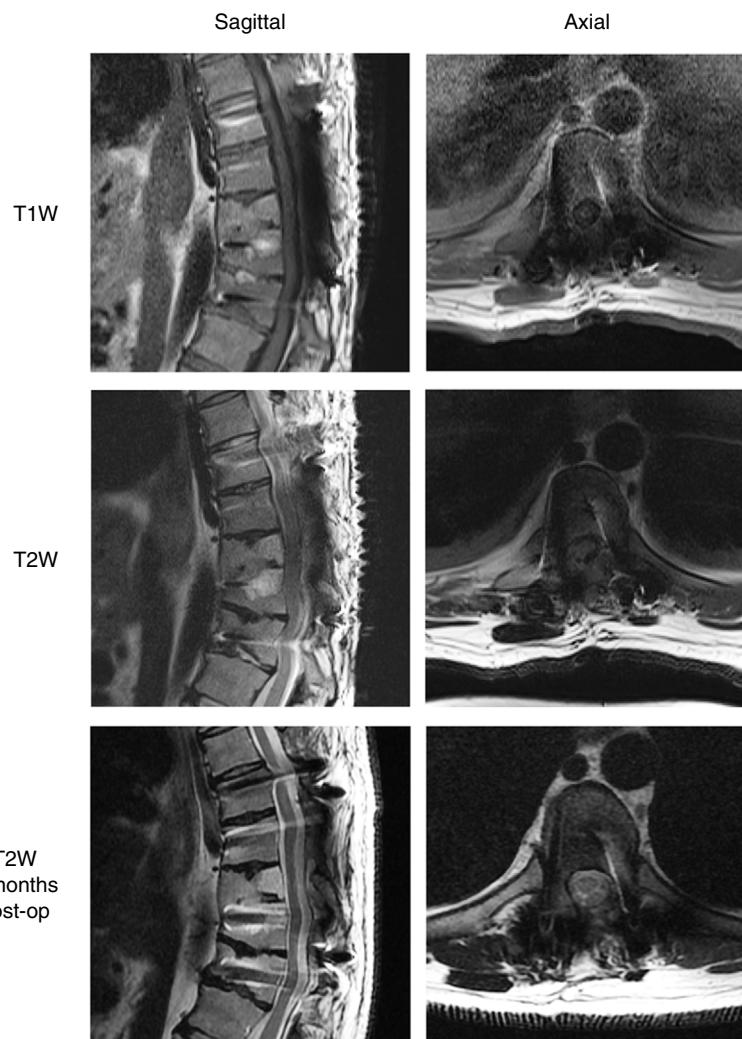


Fig. 3 – The patient underwent MRI on postoperative day 6, as well as a follow-up 6 months later, that demonstrated decompression of the spinal cord. All axial images were taken at the level of T8.

extracanalicular part gave an inhomogeneous signal with hypointense and hyperintense areas on T1W and T2W MRI. The intracanalicular part measured 8 cm in maximal diameter, causing stenosis of the spinal canal at the levels of T11-L1, while the extracanalicular one was 19 × 4 × 4 cm, extending subcutaneously between the levels of T9-L3.

Fine needle biopsy (FNB) under CT guidance revealed the presence of mature adipocytes and thus was deemed to be non-diagnostic. Following that, the patient underwent digital subtraction angiography (DSA) and elective embolisation of the feeding vessel of the extracanalicular part to assist its surgical removal, as magnetic resonance angiography had demonstrated high vascularity of the mass (image not shown). Elective operation was scheduled and laminectomy with subtotal excision of the extradural and total excision of the extracanalicular parts was performed. Importantly, no major bleeding occurred during the operation and no need for intraoperative or postoperative transfusion of packed red blood cells or other blood products was required, as the patient's haemoglobin concentration (Hb) did not drop

significantly after the operation (10.4 g/dL postoperative vs 12.5 g/dL preoperative).

Macroscopically, the tumour was multilobular, with brownish and in parts yellowish colour. Histological examination of both the epidural and extracanalicular parts revealed the presence of mature adipocytes without indications of atypia and thin-walled capillary sized vessels containing fibrin thrombi, with CD34 immunostain highlighting the thin-walled capillary-sized vessels (Fig. 2c, d). Based on the histological findings the diagnosis of angiolioma was made. The patient underwent postoperative MRI that demonstrated the decompression of the spinal cord (Fig. 4). The recovery of the patient was uneventful, with partial remission of the symptoms and discharge on postoperative day four. The patient was reevaluated 6 months postoperatively with MRI that demonstrated full decompression of the spinal canal (Fig. 4). Neurologically, full remission of his symptoms was identified.

Both patients provided a written informed consent for the release of their case history and of the visual material published in the present paper.

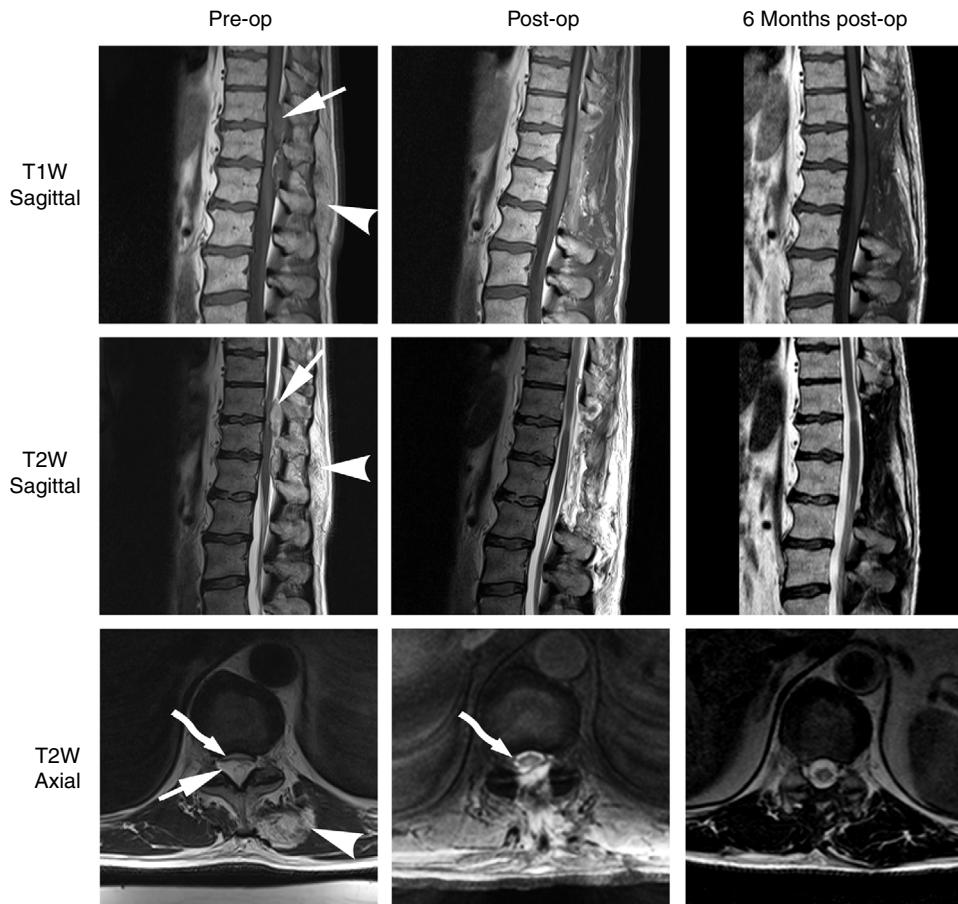


Fig. 4 – MRI of the thoracolumbar area (left column) showing an epidural mass on T11-L1 levels that was hypointense on T1W and iso- to hyperintense on T2W (arrow) and caused significant compression of the spinal cord (curved arrow), as well as an extra-canicular part that gave inhomogeneous signal with hypointense and hyperintense areas on T1W and T2W MRI (arrowhead). MRI on the 4th postoperative day (middle column) and 6 months later (right column), demonstrated the complete decompression of the spinal cord. All axial images were taken at the level of T11-T12 intervertebral disk. All images were taken in turbo spin echo sequence, except for the postoperative axial that was taken in gradient echo 2-D.

Results

From the 40 publications that resulted from our search, 10 were excluded for being irrelevant to the topic of our study and 5 were excluded for not being published in English. Finally, the search yielded 25 publications that were included in the present study.

Between 1890 and 2006, 15 out of the 118 cases of angioliomas reported were of the infiltrating type,¹³ while from 2007 to 2011 15 cases of spinal angioliomas were described, of which 3 were infiltrating.²² From the literature review performed, of the 81 cases reported from 2012 to 2017, 10 were infiltrating (Table 1). The vast majority of the tumours were located posteriorly, whereas 4 cases were located laterally and 8 posterolaterally. Purely cervical, thoracic or lumbar localisation was reported in 2.4% (N=2), 71.6% (N=58) and 12.3% (N=10) respectively. For either cervicothoracic or lumbosacral localisations, 4 cases (5%) have been reported and finally 3.7% (N=3) of the tumours extended from the thoracic to the lumbar region of the spinal cord.

For the infiltrating cases, no gender difference was identified (male to female ratio: 1), while overall a slight female predominance was found (58%). Mean age of diagnosis regardless of infiltrating potential was 48.4 years (Fig. 5a), while the same index was found to be 55.3 years for the infiltrating cases (Fig. 5b). Taken all the available data to date, about 12.3% of all angioliomas reported were infiltrating. As far as our database is concerned, no statistically significant difference was found on the age of diagnosis between the infiltrative and non-infiltrative groups ($t_{75} = 1.63, p = .1$). No statistically significant correlation was found between the infiltration potential and gender ($r_s = -0.022, p = .84$), anteroposterior localisation ($r_s = -0.158, p = .16$), spinal cord region ($r_s = -0.1, p = .36$) or tumour size ($r_s = 0.08, p = .48$).

Discussion

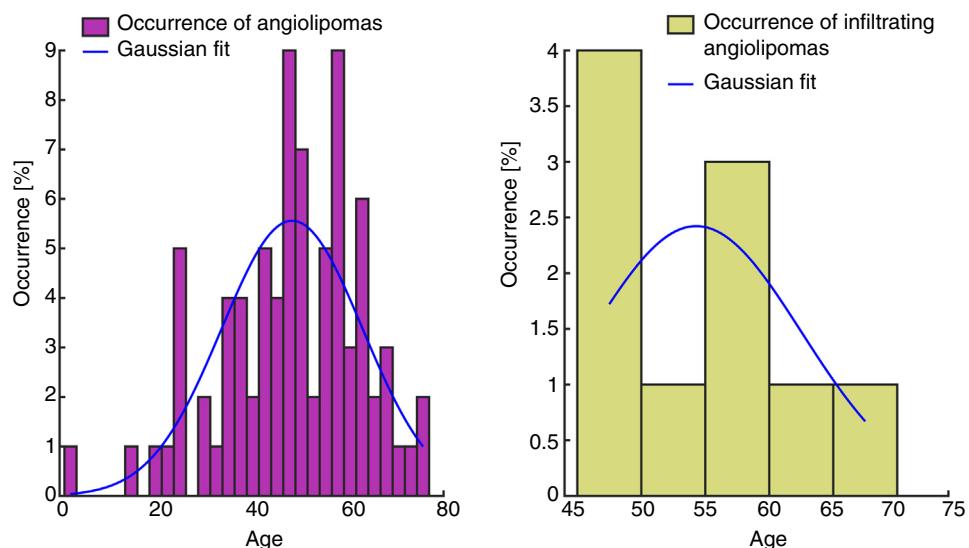
Here, we report two cases of spinal angiolioma with a particular focus on its pathogenesis, as well as its challenging differential diagnosis and preoperative management.

Table 1 – Summary of published cases of spinal angiomyomas from 2012 to 31 August 2017. M: male, F: female.

References	Age/sex	Spine level	Localisation	Infiltrating
Ghanta et al. ²³	56/M	T4-T5	Posterior	No
Han et al. ²⁴	58/M	T4-T5	Posterior	Yes
Rodrigues et al. ²⁵	71/F	T5-T6	Lateral	Yes
Fujiwara et al. ²⁶	64/F	T5-T8	Posterior	No
	65/M	T5-T7	Posterior	No
Hu et al. ²⁷	44/M	T3-T5	Posterior	No
	24/F	T4-T5	Posterior	No
	77/M	T4-T5	Posterior	No
	49/F	T5-T9	Posterior	No
	29/F	T4-T5	Posterior	No
	60/M	T5-T7	Posterior	No
	58/M	T8-T10	Posterior	No
	67/M	T4-T6	Posterior	No
	50/F	T11-T12	Posterior	No
	50/F	T9-T11	Posterior	No
Ramdas et al. ¹⁷	58/M	C7-T1	Posterior	No
Wang et al. ²⁸	47/F	T3-T10	Posterior	No
	36/F	L3-S2	Posterior	No
	46/F	T2-T4	Lateral	Yes
	50/F	T1-T3	Posterior	No
	47/F	L2-L4	Posterior	No
	46/F	T11-T12	Posterior	No
	54/F	L3-L4	Posterolateral	Yes
	44/F	T8-T11	Posterior	No
	55/F	L5-S1	Posterior	No
	47/F	L2-L3	Posterior	No
	49/F	L2-L3	Posterior	No
	55/F	T6-T8	Posterior	No
Prasad et al. ²⁹	26/M	T5-T9	Posterior	No
Awang et al. ³⁰	15/F	T6-T9	Posterolateral	No
Bovier ³¹	39/M	T4-T5	Posterior	No
Si et al. ³²	50/M	L3-L4	Posterior	No
	53/M	T4-T7	Posterior	No
	58/M	T9-T10	Posterior	No
	41/F	T5-T6	Posterior	No
	19/F	C3-C6	Posterior	No
	26/F	C4-C6	Posterolateral	No
	63/F	T7-T10	Posterior	No
	62/F	T8-T10	Posterior	No
	74/F	L4-L5	Lateral	No
	55/M	T3-T5	Lateral	No
	62/M	L4-L5	Posterior	Yes
	61/M	T11-L3	Posterior	Yes
	43/F	T7-T9	Posterolateral	No
	57/M	T5-T8	Posterior	No
	69/F	T4-T6	Posterolateral	No
	62/M	T4-T6	Posterior	No
	47/F	T2-T3	Posterior	Yes
	50/F	T2-T4	Posterolateral	No
	37/M	T4-T7	Posterior	No
	51/F	T4-T5	Posterior	-
	59/F	T8-T11	-	-
Ramdas et al. ¹⁷	58/M	C7-T1	Posterior	No
Nakao et al. ³³	32/F	T1-T6	Posterior	No
Costa et al. ³⁴	43/M	T2-T3	Posterior	No
Sandvik et al. ¹⁸	1/M	C6-T7	Posterior	No
Nadi et al. ³⁵	50/F	T6-T9	Posterior	Yes
Eap et al. ³⁶	22/M	C7-T3	Posterior	No
Mohammed and Ahmed ³⁷	35/F	T5-T8	Posterior	No
Bouali et al. ³⁸	59/F	T5-T8	Posterior	No
	40/F	T4	Posterior	No
	62/M	T4-T6	Posterior	No
	49/M	T4-T6	Posterior	No
	29/M	T1-T8	Posterior	No

Table 1 (Continued)

References	Age/sex	Spine level	Localisation	Infiltrating
Glynn et al. ³⁹	48/M	T4–T10	Posterolateral	No
	61/F	L1–L2	Posterior	No
	65/M	T3–T5	Posterior	No
	47/F	L1–L4	Posterior	No
Glynn et al. ³⁹	37/F	T6–T9	Posterior	No
Sim et al. ⁴⁰	58/F	T2–T6	Posterior	Yes
	42/F	T11–L2	Posterior	No
	39/M	T3–T6	Posterior	No
	26/F	L5–S1	Posterolateral	No
Wang et al. ⁴¹	25/F	L3–L4	Posterior	No
	77/F	T2–T4	Posterior	No
	45/F	T4–T6	Posterior	No
Shweikeh et al. ⁴²	55/M	T3–T8	Posterior	No
Mohammed et al. ⁴³	68/M	T4–T8	Posterior	No
Onishi et al. ⁴⁴	35/F	T3–T5	Posterior	No
Present Case 1	35/M	T7–T9	Posterior	No
Present Case 2	46/M	T11–L1	Posterior	Yes

**Fig. 5 – Occurrence of angioliomas by age group regardless of infiltrating potential (a) and of the infiltrating type (b).**

Epidural angioliomas are rarely encountered before the 4th decade of life and demonstrate a slight female predominance.¹³ Our results are in agreement with the former statement as only 11 of the 81 cases involved patients under 30 years old with a female to male ratio of 1.38. No causative factor has yet been identified, however it is speculated that this tumour grows as a reaction to harmful stimuli,^{7,8} that eventually give rise to an inflammatory cascade. Taking into consideration the demographics of the Case 1 along with his history of spinal cord injury, we could speculate that this tumour might have resulted from persistent irritation of the adipose tissue, induced from the operation in the spinal cord, rather than being sporadic. This assumption is further supported by the fact that the bulk of the mass is located between the T7–T8 vertebrae, rather than in the T9, which was the location of the fracture.

From the literature search that was performed, we were unable to identify similar cases reported to date. While most cases are considered to be sporadic, a recent study argues that

mutations in the protein kinase D2 gene may play a role in the tumourigenesis of angioliomas.⁴⁵ Even though it is not possible to draw a definitive conclusion on the pathogenesis of the tumour in our case, the possibility of its development in the spinal fusion region out of mere coincidence seems quite unlikely. Supposing that indeed the mass developed as a defence mechanism against prolonged irritation of the adipose tissue, one question still remaining to be addressed is whether the stimulus was provided by the spinal fusion implants or from the operative technique. If this is indeed the case, an idiopathic component must also have contributed to the pathogenesis of the mass, provided on the one hand the rarity of this tumour and on the other hand the wide application of prosthetic materials in the spinal cord. On the other hand, it is also possible that we are facing an underdiagnosed condition due to the impaired visualisation of the spinal canal following the positioning of metallic implants. In view of the lack of available data on the subject, and the advances in the treatment of spinal cord lesions and visualisation techniques,

these questions are expected to be answered in the years to come.

With regard to Case 2, infiltrative angioliomas of the spinal canal are rarely encountered, with about one in ten (12.34%) of the cases identified from our search demonstrating infiltrative potential, a number which is consistent with the findings of previous studies.^{13,22} Peak mean age of occurrence was found to be comparable between the infiltrating and non-infiltrating types, while no case of the former type has yet been reported in patients younger than 45 years old. Furthermore, we were unable to correlate the occurrence of the infiltrative type neither with a specific demographic parameter, nor with the characteristics of the tumour. This finding suggests that the infiltrating potential of the tumour could be most likely attributed to genetic factors.

Despite their aggressive behaviour, infiltrative spinal angioliomas are considered to be histologically benign.⁴⁶ Microscopic findings include mature adipocytes and a rich network of vessels, which are of larger diameter than those found in subcutaneous angioliomas.⁴⁷ In addition, some authors have found the infiltrating type to be lacking partially or entirely the capsule and having ill-defined borders compared to its non-infiltrating counterpart.⁴⁶ It is, therefore, not surprising the fact that in the present case, FNB was considered non-diagnostic. At this instance, it should be noted that FNB was conducted only in the second patient, due to the fact that the extracanalicular part of the tumour was more easily accessible and hence the procedure was considered safer than accessing the intracanalicular part. In addition, the rapid neurologic decline of the first patient imposed rapid decompression of the spinal canal and as a result, a potential preoperative biopsy was considered superfluous.

As far as the imaging features of the mass are concerned, it has been previously reported that the signal intensity emitted by the mass depends on its vascularity. Hence, a mass rich in blood vessels is hypointense on T1W and hyperintense on T2W MRI and demonstrates intense enhancement after intravenous contrast administration. In contrast, a tumour with low vascularity, presents hyperintense on both T1W and T2W MRI and hypointense on fat-suppressed Images.²⁷ Based on the aforementioned statements, we can deduce that in our case, the subcutaneous part of the mass of Case 2 had a higher vascular component than its epidural counterpart (Fig. 4), which was further supported by the DSA image.

Taken the above into account, it is strongly recommended to consider angiolioma of the spinal canal in the differential diagnosis, when encountering a mass with the aforementioned characteristics on MRI and a – seemingly – non-diagnostic biopsy. The differential diagnosis of a spinal epidural mass apart from angiolioma, includes a variety of pathologies such as lipomatosis, abscess, metastasis, haematoma, arteriovenous malformation and herniated nucleus pulposus.⁴⁸ Of these, haemangiomas are probably the most difficult to differentiate between, based on their imaging characteristics, as they are most commonly located posteriorly, have a high signal intensity on T2W and low on T1W and are greatly enhanced by intravenous contrast.⁴⁹ It should be stressed however, that given the rarity of the condition and its overall benign nature, angioliomas should be kept low in the list when differentiating a mass with

similar radiologic characteristics in order not to miss other more frequent and potentially more harmful entities.

Definitive management of this entity is provided through surgical resection. Both total and gross total resections provide good neurological results with low chances of recurrence⁴¹ and virtually no difference in the outcomes between the infiltrating and non-infiltrating types.⁵⁰ The preoperative embolisation of an infiltrative spinal angiolioma can be of great benefit for both the patient and the surgeon, as it considerably reduces intraoperative blood loss. To the best of our knowledge there are only two other reports of preoperative embolisation of such tumours published to date.^{2,51}

Preoperative embolisation of spinal tumours is a relatively safe and highly efficacious option for their preoperative management.^{52,53} The benefits of preoperative embolisation of spinal tumours and particularly of metastases, hemangiomas, hemangioblastomas have been well established and include improved perioperative visualisation and postoperative outcome, resulting from the reduced morbidity and more aggressive surgical resection, thanks to the reduction of perioperative blood loss.⁵⁴ Taking the above into account, and considering the particularities of the tumour encountered in Case 2, and especially of its increased dimensions and high vascularity, the final decision of preoperative embolisation was made, with excellent postoperative outcomes. In conclusion, we would strongly recommend the application of preoperative embolisation to assist resection of selected cases of spinal angioliomas that are the most likely to benefit from what this invasive technique has to offer.

Conclusions

Spinal angioliomas are rare tumours typically found in the midthoracic region. Here we suggest that some cases of spinal angioliomas might result from persistent irritation of the adipose tissue, as for example in reaction to harmful stimuli, giving rise to an inflammatory cascade. From the literature search that was performed, we were unable to identify similar cases involving patients with spinal fusion implants. Furthermore, in some cases, the mass may extend beyond the spinal canal, infiltrating the adjacent structures. From the review of the available literature, this infiltrating potential of the tumour was not found to be significantly correlated with the patients' demographics and tumour characteristics. This suggests that the infiltrating potential of the tumour is most likely attributed to the intrinsic characteristics of the tumour, rather than to its host. Regarding the preoperative management of the patient, biopsy may be mistakenly considered non-diagnostic, while preoperative embolisation can greatly assist surgical removal by reducing considerably intraoperative blood loss.

Funding

No funding was received for this research.

Conflicts of interest

The authors report no conflicts of interest.

Informed consent

Informed consent was obtained from all individual participants included in the study. Additional informed consent was obtained from all individual participants for whom identifying information is included in this article.

REFERENCES

1. Preul MC, Leblanc R, Tampieri D, Robitaille Y, Pokrupa R. Spinal angioliomas. Report of three cases. *J Neurosurg*. 1993;78:280–6, <http://dx.doi.org/10.3171/jns.1993.78.2.0280>.
2. Andaluz N, Balko G, Bui H, Zuccarello M. Angiolipomas of the central nervous system. *J Neurooncol*. 2000;49:219–30.
3. Kahng HC, Chin NW, Opitz LM, Pahuja M, Goldberg SL. Cellular angiolioma of the breast: immunohistochemical study and review of the literature. *Breast J*. 2002;8:47–9.
4. Liu YJ, Karamchandani DM. Gastric angiolioma: a rare entity. *Arch Pathol Lab Med*. 2017;141:862–6, <http://dx.doi.org/10.5858/arpa.2016-0239-RS>.
5. Wu Z, Wan H, Shi M, Li M, Wang Z, Yang C, et al. Bronchoscopic resection of bronchial angiolioma: a rare case report. *Mol Clin Oncol*. 2016;5:850–2, <http://dx.doi.org/10.3892/mco.2016.1069>.
6. Berenbruch K. Ein fall von multiplen angioliomen kombiniert mit einem angiom des ruckenmarks. Universität Tübingen; 1890.
7. Howard WR, Helwig EB. Angiolipoma. *Arch Dermatol*. 1960;82:924–31.
8. Putney FJ, Adkins WY, Pratt-Thomas HR. Perivenous angiolioma of the internal jugular vein. *Trans Sect Otolaryngol Am Acad Ophthalmol Otolaryngol*. 1977;84:105–7.
9. Ehni G, Love JG. Intraspinal lipomas: report of cases; review of the literature, and clinical and pathologic study. *Arch Neurol Psychiatry*. 1945;53:1–28, <http://dx.doi.org/10.1001/archneurpsych.1945.02300010011001>.
10. Pagni CA, Canavero S. Spinal epidural angiolioma: rare or unreported? *Neurosurgery*. 1992;31:758–64, discussion 64.
11. Garib G, Siegal GP, Andea AA. Autosomal-dominant familial angioliomatosis. *Cutis*. 2015;95:E26–9.
12. Abbasi NR, Brownell I, Fangman W. Familial multiple angioliomatosis. *Dermatol Online J*. 2007;13:3.
13. Gelabert-Gonzalez M, Garcia-Allut A. Spinal extradural angiolioma: report of two cases and review of the literature. *Eur Spine J*. 2009;18:324–35, <http://dx.doi.org/10.1007/s00586-008-0858-8>.
14. Rocchi G, Caroli E, Frati A, Cimatti M, Savlati M. Lumbar spinal angioliomas: report of two cases and review of the literature. *Spinal Cord*. 2004;42:313–6, <http://dx.doi.org/10.1038/sj.sc.3101535>.
15. Benvenutti-Regato M, De la Garza-Ramos R, Caro-Osorio E. Thoracic epidural spinal angiolioma with coexisting lumbar spinal stenosis: case report and review of the literature. *Int J Spine Surg*. 2015;9:67, <http://dx.doi.org/10.14444/2067>.
16. Akhaddar A, Alouzidi A, Elmostachid B, Gazzaz M, Boucetta M. Sudden onset of paraplegia caused by hemorrhagic spinal epidural angiolioma. A case report. *Eur Spine J*. 2008;17 Suppl. 2:S296–8, <http://dx.doi.org/10.1007/s00586-008-0591-3>.
17. Ramdasi RV, Avinasha KM, Mahore A, Kawale J. Spinal angiolioma manifesting with apoplexy. *BMJ Case Rep*. 2014;2014, <http://dx.doi.org/10.1136/bcr-2014-204379>.
18. Sandvik U, Svensdotter E, Gustavsson B. Spinal cavernous extradural angiolioma manifesting as a spontaneous spinal epidural hematoma in a child. *Childs Nerv Syst*. 2015;31:1223–6, <http://dx.doi.org/10.1007/s00381-015-2760-0>.
19. Raghavendra S, Krishnamoorthy T, Ashalatha R, Kesavadas C. Spinal angiolioma with acute subarachnoid hemorrhage. *J Clin Neurosci*. 2007;14:992–4, <http://dx.doi.org/10.1016/j.jocn.2006.04.024>.
20. Petrella G, Tamburrini G, Lauriola L, Di Rocco C. Spinal epidural angiolioma complicated by an intratumoral abscess. Case report. *J Neurosurg*. 2005;103:166–9, <http://dx.doi.org/10.3171/ped.2005.103.2.0166>.
21. Trabulo A, Cerqueira L, Monteiro J, Roque P, Reis FC, Coelho MR. Spinal angioliomas revisited: two case reports. *Acta Neurochir (Wien)*. 1996;138:1311–9, <http://dx.doi.org/10.1007/bf01411061>.
22. Reyes D, Candocia FJ. Thoracolumbar spinal angiolioma demonstrating high signal on STIR imaging: a case report and review of the literature. *Spine J*. 2013;13:e1–5, <http://dx.doi.org/10.1016/j.spinee.2013.06.057>.
23. Ghanta RK, Koti K, Dandamudi S. Spinal epidural angiolioma: a rare cause of spinal cord compression. *J Neurosci Rural Pract*. 2012;3:341–3, <http://dx.doi.org/10.4103/0976-3147.102617>.
24. Han SR, Yee GT, Choi CY, Lee CH. Infiltrating spinal angiolioma. *J Korean Neurosurg Soc*. 2012;52:161–3, <http://dx.doi.org/10.3340/jkns.2012.52.2.161>.
25. Rodrigues JCL, Mortimer AM, Love S, Renowden SA. A rare cause of neural foraminal widening. *J Radiol Case Rep*. 2012;6:1–8, <http://dx.doi.org/10.3941/jrcr.v6i12.1236>.
26. Fujiwara H, Kaito T, Takenaka S, Makino T, Yonenobu K. Thoracic spinal epidural angiolioma: report of two cases and review of the literature. *Turk Neurosurg*. 2013;23:271–7, <http://dx.doi.org/10.5137/1019-5149.jtn.4860-11.0>.
27. Hu S, Hu C-h, Hu X-y, Wang X-m, Dai H, Fang X-m, et al. MRI features of spinal epidural angioliomas. *Korean J Radiol*. 2013;14:810–7, <http://dx.doi.org/10.3348/kjr.2013.14.5.810>.
28. Wang B, Yang Z, Yang J, Wang G, Xu Y, Liu P. Spinal angiolioma: experience of twelve patients and literature. *Neurol India*. 2014;62:367–70, <http://dx.doi.org/10.4103/0028-3886.141232>.
29. Prasad GL, Sinha S. Spinal intradural subpial angiolioma: case report and review of literature. *Surg Neurol Int*. 2014;5:164, <http://dx.doi.org/10.4103/2152-7806.145770>.
30. Awang MSB. Hemorrhagic thoracic spinal epidural angiolioma: a case report. *Int Med J Malays*. 2014;13:73–6.
31. Bovier EG. Acute paraplegia by spinal angiolioma. Case report and literature review. *Coluna*. 2014;13:147–9, <http://dx.doi.org/10.1590/S1808-18512014130200403>.
32. Si Y, Wang Z, Pan Y, Lin G, Yu T. Spinal angiolioma: etiology, imaging findings, classification, treatment, and prognosis. *Eur Spine J*. 2014;23:417–25, <http://dx.doi.org/10.1007/s00586-013-3073-1>.
33. Nakao Y, Shimokawa N, Tsukazaki Y, Terada A, Nakajo K, Fu Y. Radical excision combined with instrumented fixation in the management of thoracic epidural angiolioma: a case report. *J Med Case Rep*. 2014;8:377, <http://dx.doi.org/10.1186/1752-1947-8-377>.
34. Costa MDSd. Hemorrhagic onset of spinal angiolioma. Case report. *J Neurosurg Spine*. 2014;21:913–5, <http://dx.doi.org/10.3171/2014.9.SPINE131162>.
35. Nadi MM, Nadi AM, Zabara MY, Ahmad TM. Management of infiltrating spinal epidural angiolioma. *Neurosciences*. 2015;20:159–63, <http://dx.doi.org/10.17712/nsj.2015.2.2.20140463>.
36. Eap C, Bannwarth M, Jazeran JF, Kleber JC, Theret É, Duntze J, et al. Spontaneous epidural hematoma due to cervico-thoracic angiolioma. *Neurochirurgie*. 2015;61:398–400, <http://dx.doi.org/10.1016/j.neuchi.2015.09.005>.
37. Mohammed ZI, Ahmed MMZ. Spinal extradural angiolioma manifested after normal vaginal delivery. *BMC Res Notes*. 2016;9:132, <http://dx.doi.org/10.1186/s13104-016-1944-3>.

38. Bouali S, Maatar N, Bouhoula A, Abderrahmen K, Said IB, Boubaker A, et al. Spinal epidural angioliomas: clinical characteristics, management and outcomes. *Asian J Neurosurg.* 2016;11:348-51, <http://dx.doi.org/10.4103/1793-5482.180901>.
39. Glynn D, Murray B, Cryan J, O'Brien D, Kavanagh E. Spinal epidural angiolioma. *Spine J.* 2016;16:e531-2, <http://dx.doi.org/10.1016/j.spinee.2016.01.205>.
40. Sim K, Tsui A, Paldor I, Kaye AH, Gaillard F. Four cases of spinal epidural angiolioma. *J Clin Neurosci.* 2016;25:134-9, <http://dx.doi.org/10.1016/j.jocn.2015.08.028>.
41. Wang FF, Wang S, Xue WH, Cheng JL. Epidural spinal angiolioma: a case series. *BMC Res Notes.* 2017;10:128, <http://dx.doi.org/10.1186/s13104-017-2432-0>.
42. Shweikeh F, Sangtani A, Steinmetz MP, Zahos P, Chopko B. Spinal angioliomas: a puzzling case and review of a rare entity. *J Craniovertebr Junct Spine.* 2017;8:91-6, http://dx.doi.org/10.4103/jcvjs.JCVJS_23_17.
43. Mohammed Y, Elhamdani S, Farooq M, Mazagri R. A case report and review of thoracic spinal angiolioma. *Surg Neurol Int.* 2017;8:113, http://dx.doi.org/10.4103/sni.sni_148_17.
44. Onishi FJ, Salem FAS, de Melo Lins DL, Dauar RFB, Stavale JN. Spinal thoracic extradural angiolioma manifesting as acute onset of paraparesis: case report and review of literature. *Surg Neurol Int.* 2017;8:150, http://dx.doi.org/10.4103/sni.sni_467_16.
45. Hofvander J, Arbajian E, Stenkula KG, Lindkvist-Petersson K, Larsson M, Nilsson J, et al. Frequent low-level mutations of protein kinase D2 in angiolioma. *J Pathol.* 2017;241:578-82, <http://dx.doi.org/10.1002/path.4865>.
46. Lin JJ, Lin F. Two entities in angiolioma. A study of 459 cases of lipoma with review of literature on infiltrating angiolioma. *Cancer.* 1974;34:720-7.
47. Hattori H. Epidural angiolioma is histologically distinct from its cutaneous counterpart in the calibre and density of its vascular component; a case report with review of the literature. *J Clin Pathol.* 2005;58:882-3, <http://dx.doi.org/10.1136/jcp.2004.023895>.
48. Gala FB, Aswani Y. Imaging in spinal posterior epidural space lesions: a pictorial essay. *Indian J Radiol Imaging.* 2016;26:299-315, <http://dx.doi.org/10.4103/0971-3026.190406>.
49. Demir MK, Ozdemir H, Unlu E, Temizöz O, Gençellac H. Differential diagnosis of spinal epidural meningioma and hemangioma at MR imaging. *Radiology.* 2007;244:933-4, <http://dx.doi.org/10.1148/radiol.2443061813>.
50. Guzey FK, Bas NS, Ozkan N, Karabulut C, Bas SC, Turgut H. Lumbar extradural infiltrating angiolioma: a case report and review of 17 previously reported cases with infiltrating spinal angioliomas. *Spine J.* 2007;7:739-44, <http://dx.doi.org/10.1016/j.spinee.2006.08.014>.
51. Rabin D, Hon BA, Pelz DM, Ang LC, Lee DH, Duggal N. Infiltrating spinal angiolioma: a case report and review of the literature. *J Spinal Disord Tech.* 2004;17:456-61.
52. Griessenauer CJ, Salem M, Hendrix P, Foreman PM, Ogilvy CS, Thomas AJ. Preoperative embolization of spinal tumors: a systematic review and meta-analysis. *World Neurosurg.* 2016;87:362-71, <http://dx.doi.org/10.1016/j.wneu.2015.11.064>.
53. Awad AW, Almefty KK, Ducruet AF, Turner JD, Theodore N, McDougall CG, et al. The efficacy and risks of preoperative embolization of spinal tumors. *J Neurointerv Surg.* 2016;8:859-64, <http://dx.doi.org/10.1136/neurintsurg-2015-011833>.
54. Jones K, Meyers P, Gobin P, Liu A-H. Embolization of spinal tumors. *Oper Tech Neurosurg.* 2003;6:156-62, [http://dx.doi.org/10.1053/S1092-440X\(03\)00041-0](http://dx.doi.org/10.1053/S1092-440X(03)00041-0).