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Spinal angiolipomas in pregnancy: Natural history and surgical treatment

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ABSTRACT

Spinal angiolipomas (SALs) are rare extradural tumors frequently located at the level of the thoracic spine and they are associated with spinal cord compromise that might result in severe myelopathy. While the first macroscopic description dates 1890, histologically these tumors where not described as angiolipomas until 1986 by Haddad et al.

Occurrence in pregnancy is even more infrequent. Since their first macroscopic description, spinal angiolipomas were reported anecdotally in pregnant women.

We present a case of spinal angiolipoma in pregnancy with confirmed histologic diagnosis.

In the present paper, we reviewed the literature regarding spinal angiolipomas in order to characterize their clinical manifestation, natural history, radiologic and histologic appearance. We add also a further case of spinal angiolipoma in a pregnant woman. Finally, we provide suggestions for the management of such rare tumors in pregnancy.

1. Introduction

SALs are rare extradural tumors described anecdotally in 1890 by Berenbruch [1] and in 1901 by Liebscher [2] based on macroscopic observations.

A primary histologic description was not published until 1986 by Haddad and coworkers [3]. In general, SALs have a peak of occurrence in women during the fifth decade of age (female/male ratio of 1.4) [4–7], mostly located at the level of the thoracic spine.

Their manifestation during pregnancy is even rarer and only fifteen cases were reported in the current literature [8–10]. According to the review from Turgut et coworkers [11], 22% of female patients with spinal angiolipomas become symptomatic during pregnancy. Three of the fifteen reported cases could only be retrospectively diagnosed as angiolipomas, since the occurrence was before the histologic description in 1986 [3].

This work reports on the fifteenth case of spinal angiolipoma in after the histologic description of such tumors by Haddad in 1986 [3]. We provide details of clinical presentation, the operative treatment as well as a thorough histopathologic description of such rare tumors. Based on this case, we also review the existing literature about SALs in pregnancy to reflect our experience in the body of literature, in order to provide an overview of clinical manifestation, natural history, radiologic and histologic appearance. Finally, we derived treatment suggestions due to the scarcity of treatment strategies for these rare tumors in pregnancy [8–10,12]

2. Case report

2.1. History and clinical presentation

A 29-year-old pregnant woman presented with severe dorsal pain, gait imbalance and bilateral leg weakness from the 29th week of gestation to her uncomplicated on-term delivery.

After delivery, the patient's neurologic condition worsened with development of a dorsal myelopathy with severe paraparesis (M3/5 in both legs), diffuse hyperreflexia, bilateral Babinski sign and posterior cordonal deficit with a T6 neurological level (Nurick score IV).

MRI of the entire spine (Fig. 1, Panel A and B) revealed an epidural mass extending from T5 to T9 with hyperintense signal at the periphery and hypointense core in T1 and T2 sequences. No gadolinium enhancement and no fat suppression were present (Fig. 1 Panel C and D).

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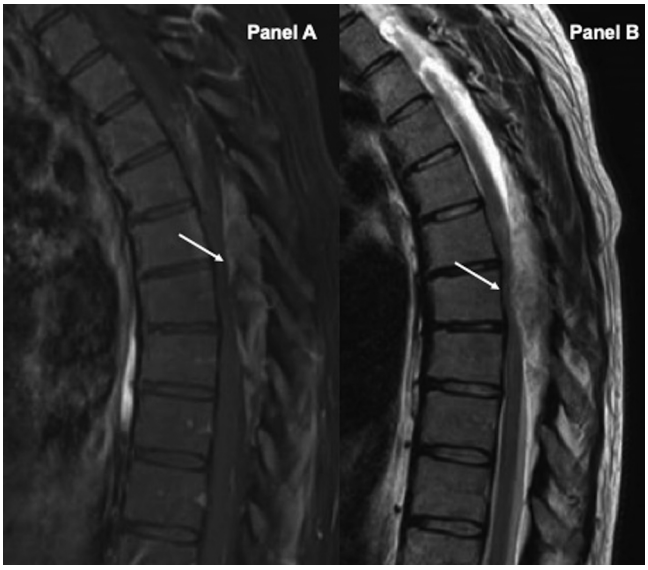

Fig. 1. Preoperative MRI.
Panel A: T1 inversion recovery sagittal images with gadolinium showing a T5-T9 hyperintense heterogeneous epidural mass with gadolinium enhancement and no fat suppression compatible with spinal angiolipoma.
Panel B: T2 sagittal images showing a T5-T9 hyperintense heterogeneous epidural mass with no spinal cord hyperintensity.

The differential diagnosis was between a late subacute thoracic epidural hematoma and a fat epidural mass. We performed a spinal arteriography with selective catheterism of the intercostal and lumbar arteries bilaterally to rule out spinal vascular malformation.

Due to the severe myelopathy, the patient underwent surgical decompression. The operative strategy was to attempt the evacuation of the hematoma through a T8 and T9 alternate levels hemilaminectomy. Resection of the ligamentum flavum permitted to expose a soft burnish epidural mass with intermingled small vessels (Fig. 2) without infiltration of the underlying dura. Biopsies were taken for microbiologic and histopathologic workup. Complete macroscopic resection of the mass was realized. To achieve complete tumor removal, we extended the bone removal with a bilateral laminectomy from T5 to T7 with sparing of the articular processes. We did decide not to perform posterior fixation due to the intrinsic stability of the thoracic chest.

Operative time was 180 min, blood loss was estimated at 400 mL.

Postoperatively the patient was monitored in the neurosurgical intermediate care unit for 24 h.

The patient was mobilized at day 1 and prophylactic anticoagulation with 40 mg sodium enoxaparin subcutaneous was started. Under intensive neurologic rehabilitation, the patient completely recovered the muscular strength in both legs (M5/5) with persistence of a mild ataxia (Nurick I).

MRI (Fig. 4) showed near-total resection of the tumor with two small tumor remnants at both cranial and caudal extremities of the resection cavity.

The histopathologic analysis (Fig. 3) was carried out using the Van Gieson-elastine staining. In the absence of neither mitosis, nor focal necrosis [13] the major finding was the presence of mature adipocytes intermingled with prominent capillary and veins throughout the tumor as seen with the Van Gieson-elastine staining. The diagnosis of angiolipoma was thus established due to the prevalent proportion of mature adipose cells over the vasculature as originally described by Haddad et al [3].

After multidisciplinary tumor board discussion, we decided for a clinical and radiologic follow-up schedule at 6 weeks, 3 and 6 months postoperatively, then once per year. At 6 months of follow up, the patient is asymptomatic and the last MRI shows no progression in size of the two non-compressive tumor remnants. Standing Xrays of the entire spine did not show postoperative kyphosis.

3. Discussion

SALs are rare tumors accounting for only 2–3% of all spinal epidural tumors [3,12,14,15].

The coincidence of a SAL and pregnancy is even rarer. In summary, since the first macroscopic description of this tumor, 15 cases (including our case) were reported in pregnant patients (Table 1).

3.1. History and clinical presentation

SALs in pregnancy manifest generally at the third trimester with acute dorsal pain, which may be referred to the inter-scapular region and chest, associated with signs of myelopathy (Table 1), like in our case. Only 2 patients described by Preul et al [12] and Mohammed et al

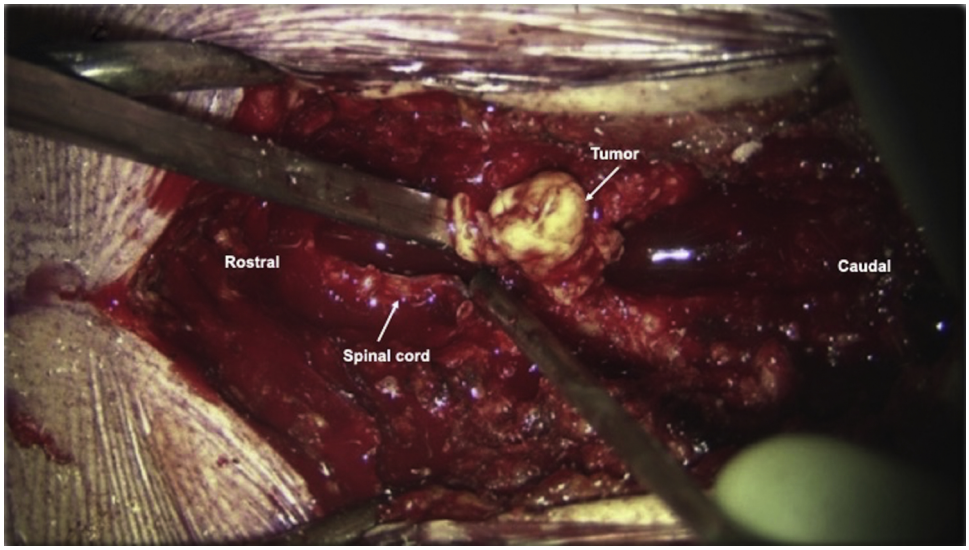

Fig. 2. Intraoperative picture.
After posterior thoracic laminectomy and ligamentum flavum resection, a soft burnish epidural mass was visible with intermingled small vessels, easily dissected from the underlying dura.

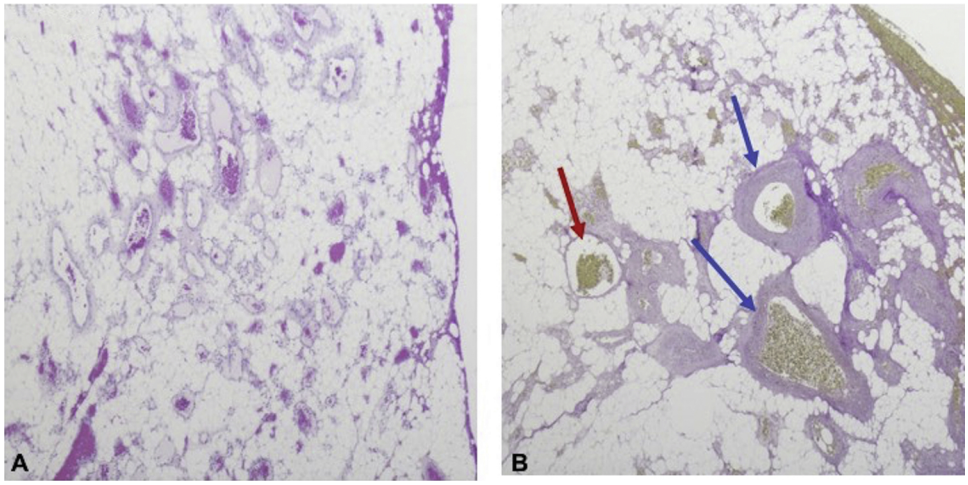

Fig. 3. Histopathologic photomicrographs. Panel A: Hematoxylin and eosin stain, magnification 2x showing mature adipocytes intermingled with vessels of variable size; Panel B: Van Gieson Elastine staining (VGEL) magnification 4x showing the absence of internal elastic lamina and the presence of capillary (red arrow) and veins (blue arrows). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article).

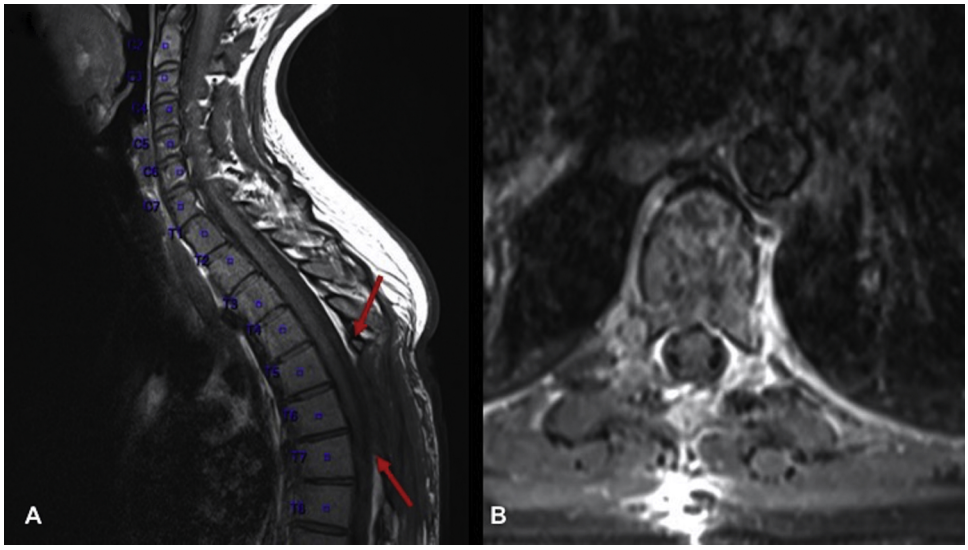

Fig. 4. Postoperative MRI.
Panel A and B: T1 sagittal and axial images showing near total resection of the spinal angiolipoma with resolution of spinal cord compression; small tumor remnants are visible at both cranial and caudal extremities of the resection cavity (red arrows). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article).

[16] became symptomatic after delivery. There is no defined time free interval between spinal pain and onset of neurological deficit.

The natural history of patients with SALs is depicted by Balado in a patient presenting with dorsal pain and myelopathy due to a thoracic SALs treated conservatively with partial regression of myelopathy after parturition [8]. The patient underwent other seven gestations with recurrence of myelopathy at each pregnancy during the third trimester. The patient died during her 8th pregnancy due to severe myelopathy and cardiovascular complications.

The relapsing clinical presentation of SALs is also described in non-pregnant patients. Taylor described two patients with SALs and relapsing myelopathy, the first presenting with a two years' history of dorsal myelopathy and spontaneous regression after several months, the second presented myelopathy with two deteriorations over a nine-year period [17].

We suppose two mechanisms to explain the acute clinical manifestation of SALs as well as their relapsing behavior in pregnancy: 1) hormone trigged rapid tumor growth and 2) vascular "steal" phenomenon.

Tumor growth in pregnancy is stimulated by estrogenic hormones that prompt fat tissue proliferation within the SALs as well by blood accumulation, intra-lesion thrombi formation and epidural venous congestion due to venous stasis in the inferior cava vein [12,18].

The "steal phenomenon" is due to the dense vascular structure of SALs causing blood accumulation within the tumor and reduced perfusion of the adjacent spinal cord, thus leading to ischemia of the watershed mid-thoracic spinal cord levels (T5-T9), where SALs are frequently located (Table 1) [4–6,10,19–27]

3.2. Imaging

The necessary radiologic workup in pregnant women is limited by the fetal sensitivity to radiation. Thus, whole spine MRI is the choice in pregnant women presenting with unusual dorsal pain and myelopathy to rule out epidural tumors and other conditions [28].

The following MRI sequences are recommended: T1W images with and without gadolinium, T2W images and inversion recovery sequences (STIR) to detect the adipose component [29]. We suggest to perform whole spine MRI to exclude the presence of associated spinal lesions. In the present case, we did not perform a staging whole body CT scan due to the benign histology of the resected tumor.

MRI appearance of SALs variates depending on the fat/vascular component [30]. SAL are high vascular tumors and they show avid contrast enhancement in T1W images [31–33]. The spontaneous fat hyperintensity on T1WI may mask gadolinium enhancement [30,31,33]. Thus, inversion recovery sequences (STIR) are needed to suppress the fat hyperintensity and to show the contrast enhancement of SALs [31,32]. In cases of low fat to vessel ratio, SALs may not present fat suppression, like in our case (Fig1), thus mimicking epidural hematomas, cavernomas or neurogenic tumors [30,33].

Table 1
Clinical presentation and management of spinal angiolipomas in pregnancy.

Author (Year)	Balado (1928)[8]	Ehni (1945)[9]	Cull (1978)[10]	Cull (1978)[10]	Preul (1993)[12]	Bouramas (1995)[21]	Trabulo (1996)[26]
Age (years)	NA	33	37	40	36	27	26
Clinical manifestation	Lower limb Weakness	Weakness numbness and loss of sensation	Lower limbs weakness and numbness	Lowe limb weakness	Dorsal pain, Lower limbs numbness, right leg weakness	Lower limb weakness and sensory loss	Dorsal pain, loss of sensation, numbness, gait disturbance
Time of symptoms onset during pregnancy	Third trimester (not specified)	Third trimester (not specified)	5^{th} and 7^{th} month	7^{th} month at the 2 pregnancies	1 week after delivery	Third trimester (not specified)	7^{th} month
Level	T (not specified)	T8	T8-T9	T8	T5-T8	T4-T7	T6-T8
Anterior/Posterior	NA	Posterior	Posterior	Posterior	Posterior	Posterior	Posterior
Dura infiltration (Yes/No)	NA	NA	No	No	Yes	No	No
Time to surgery (from symptoms onset, months)	No surgery	36	216	60	12	2	8
GTR	–	–	Yes	Yes	No	Yes	Yes
Clinical outcome	Death	Improved	Improved	Improved	Improved	Improved	Improved
Number of symptoms recurrences	8	2	2	2	3	No	No
Histological diagnosis	Heamangioma	Lipoma	Heamangiolipoma	Heamangiolipom-a	Angiolipoma	Angiolipoma	Angiolipoma

Author (Year)	Al-Anazi (1998)[22]	Turgut (1998)[23]	Cubillos (2005)[25]	Gelabert-Gonzalez (2008)[34]	Tsutsumi (2010)[24]	Glynn (2016)[27]	Mohammed (2016)[16]	Maduri (2017)
Age (years)	38	32	40	45	26	37	36	29
Clinical manifestation	Lower limbs numbness, loss of sensation and weakness	Lower limb numbness and weakness Impairment of bladder emptying	Lower limb numbness and weakness, Impairment of bladder emptying	numbness and weakness in the right leg, Low-back pain	Back pain Lower limb weakness and sensory loss	Lower Limb weakness	Lower limb weakness and sensory loss	Dorsal pain, gait disturbance and bilateral leg weakness
Time of symptoms onset during pregnancy	8^{th} month	7^{th} and 8^{th} month	7^{th} month	5^{th} month	7^{th} month	Third trimester (not specified)	1 month after delivery	7^{th} month
Level	T5-T9	T4-T9	T4-T7	L5-S1	T3-T4	T7-T6	T5-T8	T5-T9
Anterior/Posterior	Posterior	Posterior	Posterior	Anterior	Lateral	Posterior	Posterior	Posterior
Dura infiltration (Yes/No)	No	No	No	No	No	No	No	No
Time to surgery (from symptoms onset, months)	–	264 (22 years)	1	6	2 weeks (during pregnancy)	12	0	2
GTR	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Clinical outcome	Improved	Improved	Improved	Improved	Improved	Improved	Improved	Improved
Number of symptoms recurrences	No	3	No	No	No	1	No	0
Histological diagnosis	Angiolipoma	Angiolipoma	Angiolipoma	Angiolipoma	Angiolipoma	Angiolipoma	Angiolipoma	Angiolipoma

Legend: GTR: gross total resection; T: Thoracic; L: Lumbar; S: Sacral; NA: Not available.

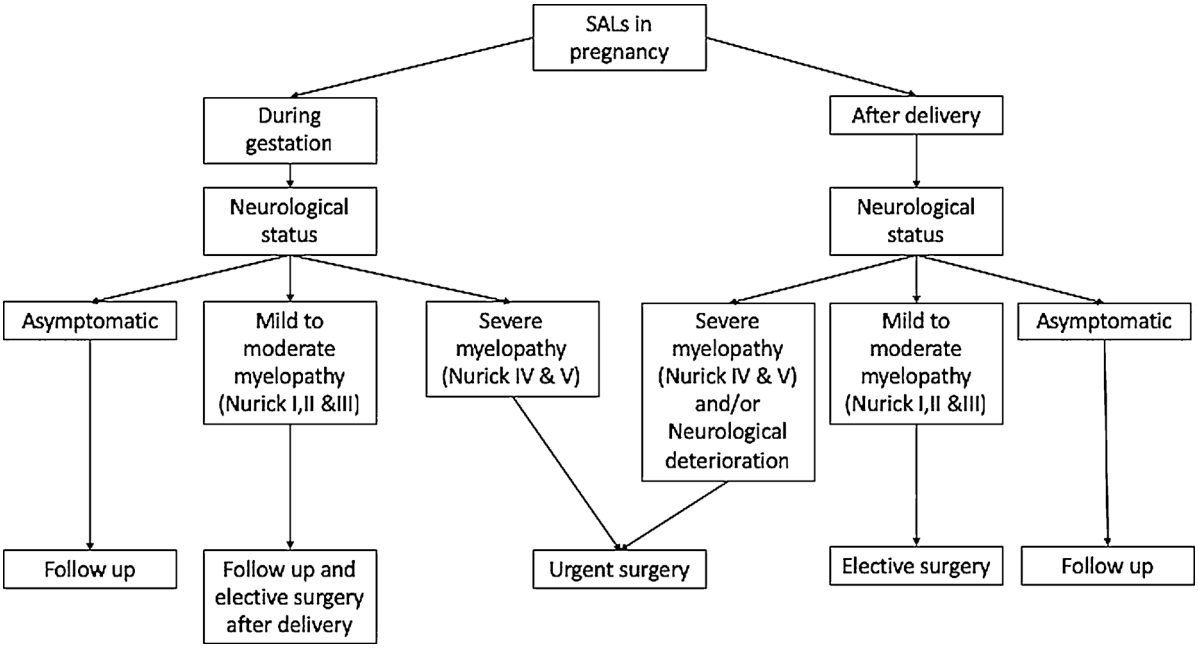

Fig. 5. Recommended treatment algorithm for Spinal angiolipomas in pregnancy.

Hence, in cases without loss of signal on fat-suppressed STIR sequences the SAL may mimic spinal epidural hematomas. On T2-weighted images, SALs have variable signal intensity with but always hyperintensity due to their vascularity [30,31,33].

In terms of localization, all of the 14 reported cases had a posterior located tumor and 13 of the 14 at the thoracic level (between T4 and T9) only the one reported by Gelabert-Gonzalez et al were in the lumbar level (L5-S1) [34].

Differential diagnosis includes spontaneous epidural hematomas and other epidural tumors like metastasis, lymphoma, multiple myelomas [33].

3.3. Treatment

The optimal treatment strategy for SALs in pregnancy is a matter of debate and several factors should be considered into the decision framework for surgery, such as the neurological status of the mother, the symptoms progression and the gestational age.By reviewing the existing literature about SALs in pregnancy, we were able to delineate a treatment algorithm for such rare condition (Fig. 5)

In pregnant asymptomatic patients or in case of mild/moderate myelopathy (Nurick I to III), a conservative expectant management may be considered with a close neurological observation in order to ensure a prompt surgical decompression in case of clinical deterioration.

The rationale for this expectant management is essentially the slow growing nature of SALs and the possibility of spontaneous tumor regression after pregnancy as reported in literature [9,10,12].

In case of severe myelopathy (Nurick IV and V), urgent surgical removal of SALs maybe performed under general anesthesia and continuous fetal monitoring, with no added risks for the fetus. The patient should be placed in a proper prone position by using specific frames high enough to hang the protuberant abdomen and to free the uterus from compression [24,28].

Since vaginal delivery could worsen the myelopathy due to the increase of abdominal pressure and unpredictable labor duration, in case of life-threatening neurological deficit and risk of preterm labor, cesarean section could be indicated prior to spinal cord decompression if the gestational age is adequate [28,35,36].

After parturition, tumor removal is strongly suggested in case of neurological deterioration or in case of persistent myelopathy in order to prevent tumor progression associated with further pregnancies.

From a surgical standpoint, midline posterior approach with bilateral laminectomies ensure adequate exposure for SALs removal. These tumors present generally a cleavage plane and are easily dissected from the underlying dura (Fig. 2). Only in some cases they may infiltrate the dura yielding to incomplete resection [12,37].

Microsurgical resection of the tumor from the underlying dura will minimize spinal cord manipulation and the risk of CSF leaks.

Regarding sagittal alignment changes after multilevel laminectomy, the exact incidence of kyphosis in adults is still unknown [38]. Multiple level lamincetomies (≥ 3 levels), resection of $> 50\%$ of the faces joints increase the risk of iatrogenous kyphosis [39]. As showed by Aizawa et co-workers, the increase in kyphosis after laminectomy for thoracic myelopathy caused by degenerative conditions of the spine is not significant when facets joints are preserved, thus spinal fusion should not be routinely performed in case of multilevel laminectomy [40]. Furthermore, in case of thoracic laminectomies for SALs removal during pregnancy, one should consider the risk of fetal irradiation during spinal stabilization.

Nevertheless, close radiologic follow-up is necessary to detect ongoing sagittal deformity.

The principal predictors of outcome after removal of SALs are the preoperative neurological status and the extent of tumor removal. In case the case of Preul [12], after incomplete resection of a thoracic spinal angiolipoma due to dura infiltration, myelopathy recurred during a second pregnancy but the patient improved spontaneously after delivery. Thus, surgery should always attempt complete removal to prevent tumor recurrence at further pregnancies. After surgery, close follow-up with MRI should be performed, ideally at 6 weeks, 3, 6 and 12 months after surgery then once per year at least for 5 years postoperatively to exclude tumor recurrence.

4. Conclusion

During pregnancy, SALs may manifest with different degrees of myelopathy and risk of severe neurological deficit. Moreover, further pregnancy are a risk factor for recurrence of these rare tumors.

By reviewing the literature, we propose a treatment strategy for SALs according to the neurological status of the patient.

Close clinical follow-up can be suggested in women with mild to

moderate myelopathy, reserving surgery in case of neurological deterioration or in patients with severe myalopathy.

Surgical decompression in pregnancy could be performed safely in centers providing multidisciplinary cooperation between spine surgeons, neurologists, gynecologists and anesthesiologists.

Complete tumor resection is mandatory to ensure a favorable neurological outcome and to prevent tumor progression with further pregnancies.

Conflict of interest

The authors have no conflict to declare.

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