



Spinal Angiolipomas : Clinical Characteristics, Surgical Strategies and Prognosis

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Objective : Angiolipomas are usually found in the subcutaneous tissue of the trunk and limbs. Spinal angiolipomas (SALs) are uncommon and have rarely been reported. We report a series of nine SALs patients who received surgical treatment in our department. To summarize the clinical characteristics of SALs, propose our surgical strategies, and evaluate the effects of the operation.

Methods : This was a retrospective review of nine SALs patients who received surgical treatment from August 2015 to March 2020. Total or subtotal resection was determined by the axial localization (dorsal or ventral) and the extent of intervertebral foramen involvement. The outcomes were assessed based on the modified Japanese Orthopaedic Association (mJOA) scoring system utilized before surgery and at various follow-up points.

Results : Among the nine patients, the mean mJOA score before surgery was 6.6 ± 2.3 , compared with 10.1 ± 1.1 at the last follow-up time point (33.4 ± 11.8 months). All patients achieved good outcomes, even in cases of subtotal resection.

Conclusion : Early surgical resection of SALs is recommended, and the specific procedures should be determined by the axial localization (dorsal or ventral) and the extent of intervertebral foramen involvement. Most of the patients had a good prognosis, even in cases of subtotal resection.

Key Words : Angiolipoma, spine · Diagnosis · Surgery · Prognosis.

INTRODUCTION

Angiolipomas are benign tumors composed of mature fatty tissue and abnormal vascular elements, and they are usually found in the subcutaneous tissue of the trunk and limbs⁵⁾. Spinal angiolipomas (SALs) are uncommon in the clinic and have rarely been reported in the literature. Because of this low occurrence, reports are always case reports. In this article, we report a series of nine SALs patients to summarize their clinical

characteristics and evaluate the surgical strategies and the effects of the operation.

MATERIALS AND METHODS

This study was approved by the Ethics Committee of Beijing Tsinghua Changgung Hospital (18013-0-01). This type of study does not require informed consent.

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General information

Nine patients with SALs (confirmed by final pathological results) were retrospectively reviewed at our center from August 2015 to March 2020. Those patients included four males and five females aged 12–73 years (46.6±17.9 years). The mean disease duration (history before diagnosis) was 15.1±19.1 months (range, 2–60), and the SALs locations included thoracic segments (n=5), lumbar segments (n=3), and cervical segments (n=1). Regarding the clinical symptoms, the patients' complaints were mostly sensory disorders (numbness, paresthesia, and pain), motor deficits (limb weakness) and sphincter disturbances. None of the patients underwent invasive treatment before the operation. The patients' general information is listed in Table 1.

Radiological classification

According to the classification methods proposed by Lin and Lin¹⁵⁾, we had seven noninfiltrating SALs cases and two infiltrating SALs cases. The different subtypes of SALs are shown in Fig. 1.

Surgical management

Electrophysiological monitoring, including somatosensory-evoked potentials and motor-evoked potentials, was used during every operation. The operations were performed with the patient in a semiprone position and with a posterior midline incision. The vertebral laminae of the corresponding lesion segments were removed to provide adequate exposure of the tumors.

In cases with a dorsal location and those in which the intervertebral foramens were not seriously affected, total resection of the tumors and the capsule was achievable. On the other hand, if the tumors had a ventral location or if the intervertebral foramens were seriously affected, wider resection was performed.

After resection of the tumors, the laminae were replaced and fixed with plates and screws to reconstruct the stability of the spine. Radiotherapy and chemotherapy were not administered after routine surgery, even in cases of subtotal resection.

The tumors were examined by two independent experienced pathologists after the surgery.

Outcome evaluation

The outcomes were assessed based on the modified Japa-

Table 1. Patients' general information

No.	Sex/age	Symptom	Disease duration (months)	Level	Axial localization	Infiltration	Preop/postop mJOA score	Follow-up duration (months)	Evolution
1	M/67	Weakness in both legs (severe on the right side)	24	L5	Ventral and RIF	Yes	8/11	54	Improved
2	F/47	Thoracic back pain, diminished position sensation of left leg, slight sphincter disturbance	4	T6–T8	Dorsal and BIF	No	6/11	50	Recovered
3	F/45	Paresthesia in left leg; weakness in both legs; slight sphincter disturbance	2	T6–T7	Dorsal and BIF	No	6/10	36	Improved
4	F/37	Numbness and pain in both legs	60	T11–T12	Dorsal	No	9/11	34	Recovered
5	M/36	Numbness, paresthesia and pain in both legs; slight sphincter disturbance	12	T8–T10	Dorsal and RIF	Yes	8/10	33	Improved
6	F/54	Thoracic back pain	2	T4–T6	Dorsal and BIF	No	9/11	26	Recovered
7	M/12	Weakness and numbness in both legs; slight sphincter disturbance	2	C4–C6	Dorsal	No	3/10	25	Improved
8	M/48	Paresthesia and pain in right leg	6	L1–S3	Dorsal and BIF	No	7/9	22	Improved
9	F/73	Numbness and pain in right leg; weakness in both legs; sphincter disturbance	24	L3–L5	Dorsal and RIF	No	3/8	21	Improved

mJOA: modified Japanese Orthopaedic Association, M: male, RIF: right intervertebral foramen, F: female, BIF: bilateral intervertebral foramen



Fig. 1. A-D : It presents the magnetic resonance imaging (MRI) scans of patient No. 4. The dorsally located epidural mass (T11–T12) was isointense on T1-weighted images (WI), hyperintense on T2WI, and homogeneously enhanced on contrast-enhanced images. E-H : It presents the MRI scans of patient No. 1. The mass was located in the ventral part, and the vertebral body (L5) and right intervertebral foramen were also affected. The mass and the affected vertebral body were hypointense on T1WI, hyperintense on T2WI, and inhomogeneously enhanced on contrast-enhanced images. A and E : Sagittal T1WI. B and F : Sagittal T2WI. C and G : Sagittal contrast-enhanced image. D and H : Axial contrast-enhanced image.

nese Orthopaedic Association (mJOA) scoring system before and 3, 6, and 12 months after the surgery and then once every 12 months. The mean follow-up period was 33.4 ± 11.8 months (range, 21–54).

Statistical analysis

Data are expressed as the mean \pm standard deviation. The mJOA scores before and after surgery were compared using t-test. All statistical analyses were performed using SPSS (version 17.0; SPSS Inc., Chicago, IL, USA), and $p < 0.05$ was used to determine statistical significance.

RESULTS

Among the nine patients analyzed, the mJOA score was 6.6

± 2.3 before surgery and 10.1 ± 1.1 at the last follow-up time point ($p < 0.001$, t-test). Total resection (confirmed by intraoperative discovery and follow-up magnetic resonance imaging [MRI] scans) of the tumors was achieved in seven patients, and subtotal resection was achieved in two patient (No. 1 and 5). At the last follow-up time point, three patients exhibited recovery, six patients exhibited improvement, and no recurrence was observed in any patients. Some of the postoperative MRI scans are shown in Fig. 2.

According to the histological examination, the tissue was composed of mature lipocytes and abnormal blood vessels, consistent with a diagnosis of angiolipoma (Fig. 3).

Immunohistochemistry was performed in seven patients. The main results were CD31 (+), 6/7; CD34 (+), 6/7; S-100 (+), 5/7; Ki-67 (+), 4/7, $< 3\%$.

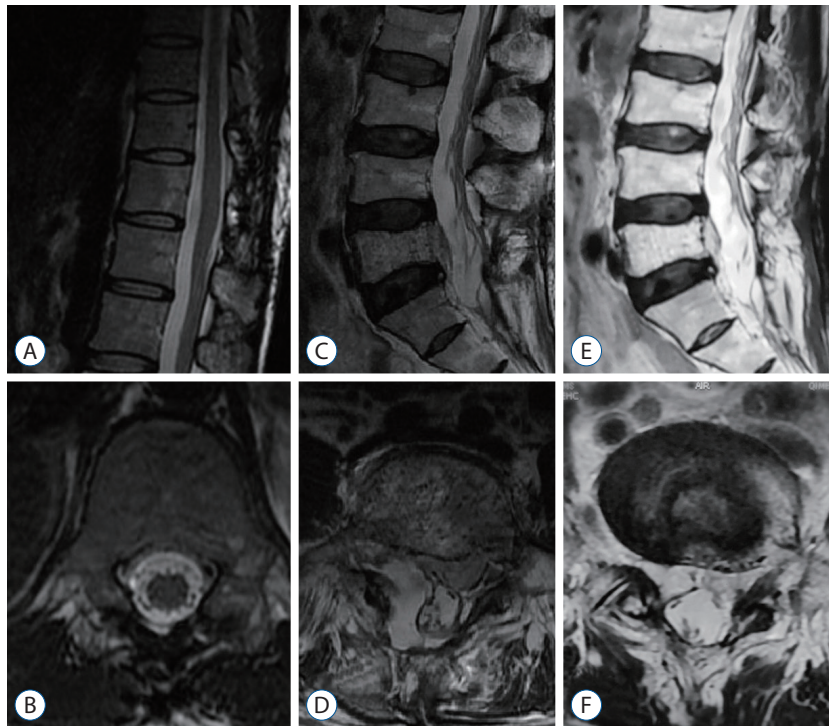


Fig. 2. A and B : It presents the postoperative the magnetic resonance imaging (MRI) scans (2 years after surgery) of patient No. 4. MRI shows that the tumors were completely removed without recurrence. C-F : It presents the postoperative MRI (1 month and 4 years after surgery) scans of patient No. 1. The results show that the tumors were partially removed, and there was no sign of further growth. A : Sagittal T2WI. B : Axial T2WI. C : Sagittal T2WI 1 month after surgery. D : Axial T2WI 1 month after surgery. E : Sagittal T2WI 4 years after surgery. F : Axial T2WI 4 years after surgery.

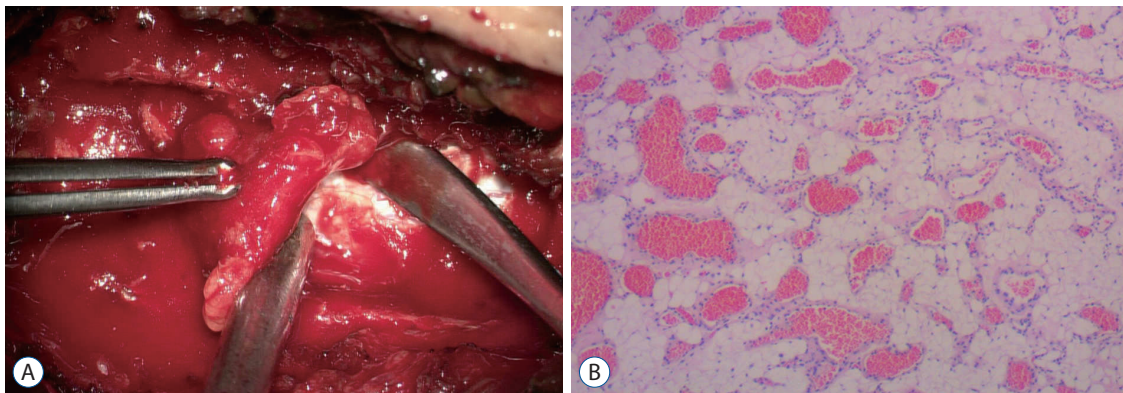


Fig. 3. A : Intraoperative photo. The tumors were mainly composed of adipose tissue mixed with vascular structures that easily bled. B : Photomicrograph. The tumor was composed of mature adipose tissue and proliferating vascular elements (H&E, original magnification $\times 40$).

DISCUSSION

SALs were first reported by Berenbruch in 1890¹⁾. In 1960, Howard and Helwig¹⁰⁾ described SALs as anatomopathologic entities containing vascular and mature adipose elements.

Demographic characteristics

As reported in previous studies, SALs are considered benign and rare lesions of the spinal axis, and they account for 0.14–1.2% of all spinal tumors and 2–3% of epidural spinal tumors^{4,12,18)}. SALs occur mostly in adults aged between 40 and 60 years and have a female predominance, and the duration of disease is usually long. The most common site of SALs is the

posterior epidural space at the thoracic level, and multiple segments are often involved^{6,14,23,28}.

The thoracic predominance of SALs was also found in our series of patients (5/9). This could be explained by the regional variation of the blood supply in the mid-thoracic spine, where the spine is least perfused. This may allow spinal lipomas to transform into SALs because of neovascularization caused by possible ischemic events¹³.

We also noted some differences in our series. 1) We had a 12-year-old cervical SALs (C4–C6) patient (No. 7) with a relatively short disease duration (2 months). This situation has also been reported in other studies^{7,24}, but we did not find spontaneous epidural hematomas during surgery. And 2) we had a patient with infiltrating SALs (No. 1) whose tumor was located in a lumbar segment (L5) and was completely in the ventral part. This is not common and has rarely been reported before^{9,16,22}. We performed subtotal resection, and the long-term effect was good.

Clinical syndromes

The clinical syndromes of epidural angioliipomas are basically the same as those of other benign space-occupying spinal tumors²⁰. Because of the benign nature of angioliipomas and with the prolongation of the disease course, patients may gradually develop sensory disorders, weakness in the lower limbs and sphincter dysfunction.

Sudden deterioration of neurological symptoms can occur due to intratumoral hemorrhage or thrombosis^{11,19,27,30}. Patient No. 6 in our group experienced sudden aggravated radicular pain due to hemorrhagic SALs after receiving a back massage. The sudden intratumoral hemorrhage was confirmed by changes in imaging data and actual conditions during surgery. Additionally, these sudden deteriorating phenomena can occur during pregnancy and with a history of weight gain in obese patients, as reported elsewhere^{8,17,26,27}, probably because of hormonal changes and an increase in the fatty component of the tumor.

Radiological manifestations and classification

Lin and Lin¹⁵ categorized SALs into noninfiltrating and infiltrating in 1974. MRI is the standard diagnostic tool for SALs. The fat content appears hyperintense on T1- and T2-weighted images, has a weakened signal due to fat suppression, and has no enhancement performance, while the vascular

Table 2. A comparison with the published single center report on treatment and outcomes of SALs

Study	N	Age (years)	Duration (months)	Resection	Radiotherapy	Evolution	Complications	Recurrence	Follow-up time
Wang et al. ²⁹ (2014)									
Infiltrating	2	50.0±5.7	17.5±5.0	2 sub-total resection	No	1 recovery, 1 improve.	No	No	40 months
Noninfiltrating	10	47.6±5.5	16.4±8.3	10 total resection	No	5 recovery, 5 improve.	No	No	From 3 to 192 months
Bouali et al. ³ (2016)									
Infiltrating	0								
Noninfiltrating	9	51.1±11.8	10.9±7.9	9 total removal	No	9 recovery	No	No	NA
Si et al. ²⁴ (2014)									
Infiltrating	3	56.7±8.4	126.7±202.2	3 total removal	No	Recovery rate : from 75% to 100%	No	No	Approximately 10 years
Noninfiltrating	18	51.6±14.2	15.6±14.2	18 total removal	No	Recovery rate : from 50% to 100%	No	No	
This study									
Infiltrating	2	51.5±21.9	18.0±8.5	2 sub-total resection	No	2 improve	No	No	33 and 54 months
Noninfiltrating	7	48.6±12.5	14.3±21.6	7 total resection	No	3 recovery, 4 improve	No	No	From 21 to 50 months

SALs : spinal angioliipomas, NA : not available

component appears isointense on T1-weighted images and hyperintense on T2-weighted images, is not weakened due to fat suppression and is strongly enhanced after the injection of contrast medium. Because SALs are composite tumors composed of mature fat tissue and proliferated vasculature, their appearance on MRI is often protean¹¹⁾. Computed tomography scanning is helpful for evaluating the degree of bone destruction when the tumor infiltrates the vertebral body. Differential diagnoses include extradural lipomatosis, nerve sheath tumor, meningioma, metastasis, and malignant lymphoma.

Si et al.²⁴⁾ proposed a new and impressive classification of SALs. According to their classification strategy, subtype IB SALs usually have lipomatosis in their upper and/or lower segments. In our patients, the MRI manifestations were consistent with those reported in most studies, but we did not observe any cases of lipomatosis in the upper and/or lower segments of the tumors, even in overweight patients. This could be because we had a limited number of cases.

Pathology and diagnosis

Histologically, SALs are tumors composed of mature lipocytes and proliferating abnormal vessels, including capillary, sinusoidal, venous or arterial vascular elements. The ratio of fat to vessels is variable, ranging from 1 : 3 to 2 : 3^{15,21,29)}. Immunohistochemical assays were performed in seven of our nine patients, and we observed positive staining for CD31, CD34, and S-100 and a low proliferation rate of Ki-67. The results were in agreement with those reported in the literature^{6,24,25)}. A diagnosis of SALs should be based on a combination of clinical, radiological and pathological examination findings.

Surgical plans and outcomes

The extent of resection for infiltrative SALs has always been a topic of debate. Because patients' symptoms are caused by tumors in the spinal canal, surgeons generally do not deal with the affected vertebral bodies. Therefore, infiltration does not determine the choice of surgical procedure. In our opinion, the axial localization (dorsal or ventral) and the extent of intervertebral foramen involvement are the factors that should determine the surgical plan.

Most cases in which tumors are located dorsally and those in which the intervertebral foramens are not seriously affected

can be totally resected. In such cases, we usually found that the tumors were compressive epidural fatty tissue intermingled with vascular structures that bled easily. Total resection was relatively easy to achieve without dura injury because adhesions between the tumors and the dura were limited.

However, if tumors are located ventrally, if the intervertebral foramens are seriously affected, or if the thoracic cavity is involved, wider resection should be planned. The extent of laminectomy should be appropriately expanded, especially on the symptomatic side. Additionally, according to Si et al.²⁴⁾, the sternocostal joint might need to be removed in some cases. In these cases, surgeons can cut the tumors into pieces and remove them in a piecemeal fashion rather than in an intraleisional fashion. For the cases whose facet joint were partially removed, and the cases whose sternocostal joint need to be removed, appropriate internal fixation is recommended to ensure spinal stability. Radiotherapy and chemotherapy are not recommended for these cases of subtotal resection.

According to the literature, most SALs patients have a good prognosis, even in cases of subtotal resection^{3,24,29)}. According to the largest single center report, 21 cases of SALs had a good prognosis and no recurrence with a follow up of approximately 10 years²⁴⁾. As to Wang et al.²⁹⁾, case series (12 cases of SALs), two cases of infiltrating SALs with subtotal resection had no recurrence with a mean follow-up of 40 months. The recurrence of SALs after surgery is rare, and only two cases were reported : one after gross-total removal²⁾ and one infiltrating tumor following subtotal resection¹²⁾. According to literature, the first case recurred 12 years after surgery²⁾. A comparison with the published single center report on treatment and outcomes of SALs is listed in Table 2.

In our group, six cases (including two infiltrating SALs) achieved improved outcome rather than recovered. It may be related to the nerve root invasion by the tumor, the duration of symptoms and the timing of surgery. As to recurrence, all seven patients who underwent total tumor resection had good prognoses with a mean follow-up of 30.6 months, and there was no recurrence. The patient who underwent subtotal resection (patient No. 1) exhibited an improved outcome 54 months after the surgery, which was a relatively long follow-up period, with no obvious signs of recurrence. From our point of view, early surgical resection is recommended because of the improvement or recovery of symptoms after surgery and the extremely low recurrence rate.

CONCLUSION

SALs are rare but well-defined benign tumors. In addition to clinical characteristics and radiological manifestations, the diagnosis of SALs depends strongly on pathological findings. Early surgical resection is recommended, and the specific procedures used should be determined by the axial localization (dorsal or ventral) and the extent of intervertebral foramen involvement. Most patients have a good prognosis, even in cases of subtotal resection.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

INFORMED CONSENT

This type of study does not require informed consent.

AUTHOR CONTRIBUTIONS

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