Surgical Treatment of a Ruptured Isolated Spinal Artery Aneurysm with Negative Angiography Findings: A Case Report

Jun-Woo Ha1, Yangkyu Lee2,3, Kyung Hyun Kim1, Bong Ju Moon1, Jeong Yoon Park1, Dong Kyu Chin1, Keun Su Kim1, Hyun Jun Jang1

1Department of Neurosurgery, Spine and Spinal Cord Institute, Gangnam Severance Hospital, Yonsei University College of Medicine, Seoul, Republic of Korea
2Department of Pathology, Gangnam Severance Hospital, Yonsei University College of Medicine, Seoul, Republic of Korea
3Institute for Breast Cancer Precision Medicine, Yonsei University College of Medicine, Seoul, Republic of Korea

INTRODUCTION

Isolated spinal aneurysms are rare vascular lesions of the spinal cord. Due to its rarity, the natural disease course and treatment guidelines have not been clearly defined to date3. Isolated spinal aneurysms refer to aneurysmal lesions of the spinal cord vasculature that are unrelated to other well-defined spinal vascular lesions, such as arteriovenous malfor-

Received: January 24, 2024
Revised: March 16, 2024
Accepted: March 26, 2024

Corresponding author:
Hyun Jun Jang
Department of Neurosurgery, Spine and Spinal Cord Institute, Gangnam Severance Hospital, Yonsei University College of Medicine, 211, Eonju-ro, Gangnam-gu, Seoul 06273, Republic of Korea
Tel: +82-2-2019-3406
Fax: +82-2-3461-9229
E-mail: janghj0@yuhs.ac

Isolated spinal artery aneurysms are rare vascular lesions of the spinal cord. Due to their rarity, the natural disease course and treatment guidelines have not been clearly defined. Here, we report a case of an angiography-negative isolated spinal aneurysm in the thoracic spine surgically that was treated without neurological compromise using indocyanine green (ICG) and intraoperative neurophysiological monitoring (IONM). A 52-year-old man without any prior medical history presented to the ER with acute lower back and bilateral leg pain accompanied by worsening voiding and difficulty defecating. Magnetic resonance imaging (MRI) of the lumbar spine showed a diffuse subarachnoid hemorrhage in the lumbar spine. The patient was initially treated conservatively with painkillers, but experienced a rapid recurrence of symptoms. A follow-up MRI scan showed subacute transformation and expansion of the subarachnoid hematoma, as well as a non-enhancing, intradural, extramedullary lesion at the T12/L1 level. Angiography did not show any remarkable findings, and surgical exploration revealed a thrombosed aneurysmal lesion. The lesion did not show ICG uptake, and temporary clipping of the caudal end of the lesion did not lead to changes in motor-evoked potential signals. A pathological examination revealed a capillary vascular structure in granulation tissue with organizing thrombi, favoring a thrombosed, granulated lesion over a vascular neoplasm. Ruptured, isolated spinal aneurysms can be especially difficult to diagnose and treat when angiography findings are negative. We report that a spinal artery aneurysm can be safely excised using intraoperative ICG and IONM.

Keywords: Aneurysm; Angiography; Indocyanine green; Monitoring, intraoperative; Spine
Rupture of spinal aneurysms results in subarachnoid hemorrhage of the spine and causes a variety of symptoms ranging from back pain to motor, and sensory deficits. Various treatment methods have been suggested in the literature, including endovascular intervention, surgery, and conservative care. Here, we report a case of angiography-negative, ruptured, ventrolaterally located isolated spinal aneurysm in the thoracic spine surgically treated without neurological compromise with the employment of indocyanine green (ICG) and intraoperative neurophysiological monitoring (IONM).

CASE REPORT

A 52-year-old male without any prior medical history presented to the emergency room (ER) with acute, lower back and bilateral leg pain accompanied by worsening voiding and defecation difficulty. The pain did not radiate along specific dermatomes, but was rather migratory along the buttocks, anterior and posterior thighs. The patient’s symptoms first occurred 10 days prior to the ER visit. Physical exam did not show motor, sensory deficits, and anal tone, saddle was intact. The only remarkable finding was a positive straight leg raising test at 30 degrees in both legs. Prior to visiting our ER, the patient had visited a local clinic and had undergone an magnetic resonance imaging (MRI) scan of the lumbar spine, which showed diffuse subarachnoid hemorrhage at the lumbar spine (Fig. 1). He was initially treated conservatively with painkillers, which were effective for a few days until the symptoms recurred. Physical therapy and nerve block procedures were not effective, and the patient underwent a follow-up MRI scan, which showed subacute transformation and expansion of the subarachnoid hematoma (Fig. 2A). Additionally, the MRI scan revealed a non-enhancing, ventrolateral, intradural, extramedullary mass-like lesion at the T12/L1 level (Fig. 2B). The patient was admitted for further evaluation and angiography was performed to rule out vascular anomalies such as arteriovenous fistula or AVM. Angiography did not show any remarkable findings, suggesting two possibilities: a vascular lesion that has bled and been thrombosed or the lesion is not vascular in nature (Fig. 3). The patient’s symptoms persisted, and surgical exploration was performed to prevent further neurologic deterioration. Subtotal laminectomy of T11 lower, L1 upper lamina and laminotomy of total T12 lamina was performed. Upon opening of the dura by vertical incision, dark-brownish cerebrospinal fluid gushed out, and further opening of the arachnoid membrane revealed hematoma compressing the spinal cord (Fig. 4A, B). The hematoma was removed by gentle dissection and irrigation, which led to the exposure of a thrombosed, fusiform-shaped aneurysmal lesion in the subarachnoid space within a thin, whitish vessel running parallel to the spinal cord (Fig. 4C, D). The lesion did not show ICG uptake, and temporary clipping of the caudal end of the lesion did not show changes in intraoperative motor evoked potential signals (Fig. 5). The cranial and caudal ends were subsequently coagulated and the aneurysm was excised and sent for pathological diagnosis. Dura repair was done by watertight dura suture and T12 lamina was re-attached to the laminotomy site using titanium plates. The patient experienced immediate relief in pain after the surgery and gradual improvements of bladder/bowel function followed. Follow-up MRI showed complete excision of the lesion (Fig. 6). Pathological examination revealed capillary vascular structure in granulation tissue with organizing thrombi, favoring a thrombosed, granulated lesion over a vascular neoplasm (Fig. 7). At one-month follow-up, the patient is free of neurological symptoms and only minor residual tingling sensations of the lower extremities remain.

Fig. 1. Magnetic resonance imaging scan at the patient’s initial presentation with symptoms showed a diffuse subarachnoid hemorrhage in the lumbar spine. The left is a T2-weighted image and the right is a T1-weighted image.
To our knowledge, only a few cases of angiography-negative, isolated spinal aneurysms have been reported to date. Surgical exploration confirmed the suspected diagnosis of a ruptured, thrombosed isolated spinal aneurysm in two cases. In another case, initially occult spinal artery aneurysm appeared on a follow-up computed tomography angiogram, in which the patient did not experience a re-bleeding event until 16 months follow-up and refused to pursue further treatment or follow-up imaging. Yet, in another case, an occult spinal aneurysm treated conservatively was identified on a follow-up angiogram after a re-bleeding event. Other reports of ruptured isolated spinal aneurysms, however, report natural regression of the aneurysm with conservative treatment on serial follow-up angiograms. As such, selecting the optimal treatment modality for angiography-negative spinal aneurysms can be tricky due to their unpredictable clinical course.

In our case, surgical intervention was decided due to persistent neurological deterioration. Since the parent artery was not identified in the preoperative angiogram, ICG and IONM was employed to minimize complications related...
Fig. 4. Upon incision of the dura, dark-brownish cerebrospinal fluid gushed out (A), and opening the arachnoid membrane revealed a hematoma compressing the spinal cord (B). Removal of the hematoma showed a dark, thrombosed aneurysmal lesion located laterally to the rootlets (C). The lesion (white arrowhead) was within a thin whitish vessel (blue arrowheads) running parallel to the spinal cord (D).

Fig. 5. The lesion did not show indocyanine green uptake (A), and temporary clipping did not lead to changes in intraoperative motor evoked potential signals (B).
to ligation of the parent artery. Intraoperative ICG staining revealed that the parent artery did not have remarkable arterial blood flow, and IONM coupled with temporary clipping ensured that ligation and excision of the aneurysm would not result in infarction of the spinal cord and cause-related neurological deficits. Thus, ICG and IONM can be useful tools in surgical excision of angiography-negative spinal aneurysms, where the parent artery of the aneurysm cannot be identified preoperatively.

Most isolated spinal aneurysms have been reported to be pseudoaneurysms rather than true aneurysms. Its pathophysiology is yet to be elucidated, but it has been

---

**Fig. 6.** Postoperative follow-up magnetic resonance imaging showed complete excision of the aneurysmal lesion and evacuation of the hematoma. T2-weighted image, sagittal (A), T1-weighted contrast-enhanced image, sagittal (B) and T1-weighted contrast-enhanced image, axial (C).

**Fig. 7.** Histological diagnosis. (A) Capillary vascular structures in granulation tissue with organizing thrombi (hematoxylin and eosin stain, ×200 magnification). (B) CD34 positivity in the small vasculature (CD34 immunohistochemistry, ×200 magnification).
reported to be associated with dissection of the arterial wall due to underlying inflammatory conditions. Pathological diagnosis in our case reported that the excised lesion mainly consisted of granulation tissue with organizing thrombi, favoring the diagnosis of a pseudoaneurysm over a true aneurysm. Such pathological finding suggests that the aneurysm may have been in a natural regression process, as several previous reports documented cases of spontaneous regression of ruptured isolated spinal aneurysms. Thus, excision of angiography-negative, ruptured spinal aneurysms may not be absolutely necessary, especially if there is a risk of neurological complication due to ligation of the parent artery.

CONCLUSION

Ruptured, isolated spinal aneurysms can be especially difficult to diagnose and treat when the angiography finding is negative. However, surgical exploration and intervention may be necessary in cases when neurological deficits are present. We report that isolated spinal artery aneurysms can be safely excised with the use of intraoperative ICG and IONM and believe that they should be actively utilized in surgical treatment of angiography-negative spinal artery aneurysms.

CONFLICTS OF INTEREST

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

REFERENCES