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Iatrogenic segmental artery aneurysmal hemorrhage: diagnosis, management, and prognosis: Case report and review of the literature

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SUMMARY

Objective: Segmental arteries are responsible for a remarkable part of the spinal circulation and aneurysmal rupture of these arteries and their branches might happen in rare cases.

Summary of background data: Here we report a unique iatrogenic segmental Artery Aneurysmal injury with the possibility of a thrombosed aneurysm, successfully treated with endovascular catheter-directed glue administration.

Methods: In this study, we would review the literature on the diagnosis, management, and prognosis of segmental Artery Aneurysmal injuries. Lower back pain and neurologic impairments are the most common symptoms of segmental Artery aneurysms.

Results: In most situations, MRI is the primary diagnostic option; however, depending on the patient's health, a brain CT scan and lumbar puncture may be required. Lumbar puncture and CT scan may indicate SAH in the case of ruptured ones. Spinal angiography that include both subclavian and vertebral and sacral artery provide for the most reliable identification of segmental artery aneurysm. Treatment is tailored to the individual, with conservative, surgical, and endovascular options available.

Conclusion: Endovascular therapy with catheter-directed adhesive application appears to be safe.

Keywords: Aneurysm; Segmental artery; Subarachnoid hemorrhage

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INTRODUCTION

Since the advent of modern neurosurgery, anatomists have provided the basis for understanding the musculoskeletal systems and the vessels supplying the spine. However, the inaccessibility of the spine was accustomed to the scarce anatomical knowledge. In 1882, Adamkiewicz, a Polish scientist researching a specific complication of neurosyphilis, called tabes dorsalis, investigated the spine arterial anatomy driving meticulously drawings of the spine arteries based on his necropsy studies [1]. Later, one of these vessels, the great anterior radiculomedullary artery, was named after him [2]. Adamkiewicz artery is located mostly on the left side of any anterior root from Dorsal 6 (D6) to Lumbar 4 (L4) [1]. These findings became more important when it became clear that spinal cord lesions might be consequences of major thoracic and abdominal operations as well as postoperative paraplegia and paraparesis, and the mechanism causing the neurologic involvement was controversial [3]. Hypo perfusion in the anterior spinal artery, where the Adamkiewicz artery (AKA) pertains in the thoracolumbar area, was indeed the cause of these serious consequences [4].

The momentousness of illustrating the detailed spine blood circulation is closely linked to the management of spinal lesions, spinal cord ischemia (SCI), and Spinal hemorrhages; where all could have burdensome consequences [5]. In non-traumatic injuries, the iatrogenic or medical complication could lead to spinal cord ischemia (SCI). Upon open and endovascular management of aortic aneurysms, SCI might happen in 11% of individuals, based on a meta-analysis of thoracoabdominal aortic surgeries [6]. Endovascular, aortic, and non-aortic surgeries are the most common reasons for SCI [7]. Spinal hemorrhages have a similar category to cerebral hemorrhages. Arteriovenous malformation (AVM) and arteriovenous fistula (AVF) of spinal arteries could cause subarachnoid hemorrhage (SAH) [8]; intradural hemorrhage could happen with ruptured aneurysms [9]. While being rare, various case reports have revealed the eventuality of any type of hemorrhage in the spine as well as segmental aneurysmal hemorrhages [10]. Here, first, we will review the anatomies of the spinal arteries prior to the narrating segmental hemorrhages and ischemia.

To briefly overview the current knowledge about the spine arterial anatomy, longitudinal arteries of the anterior spinal artery (ASA) and posterior spinal arteries are supplying the spinal circulation, being amplified by segmental arteries (SAs), mostly originating from the subclavian artery [11].

The segmental arteries in the thoracic and upper lumbar spine originate in pairs from the posterior aspect of the descending aorta adjacent to the spinal column. The segmental arteries in the thoracic spine include the posterior intercostal arteries (nine pairs) and subcostal arteries (one pair); there are typically four pairs of lumbar segmental arteries arising from the descending aorta, corresponding to the top four lumbar vertebrae.

Segmental arteries divide into two main branches:

1. Radiculomeningeal (Dural artery), which supplies the nerve root sleeve and the dura
2. Radiculomedullary artery that divided into the anterior and posterior radicular artery that supply anterior and posterior nerve roots respectively; and also Medullary artery that joins and reinforces the anterior spinal artery and posterior spinal artery

Two main and important radiculomedullary arteries are:

1. Dominant cervical radiculomedullary artery (Artery of Lazorthes) Occurs between the C5-C7 levels
2. Dominant thoracic radiculomedullary artery (Artery of Adamkiewicz)

Tends to have a left-sided origin (75%) and arises from T9-T12 (60%)

Less common origins in the lumbar region (25%) and from T6-T8 (10%).

Understanding this complex anatomy is crucial in any surgical and interventional procedures for spinal vascular lesions [12]. The AKA is a segmental artery as well [13]. Also, the spine has an axial network of tiny arteries that anastomose with each other and with the spinal cord's nutrition arteries. The segmental vessels, as well as the subclavian and hypogastric arteries and their branches, are all inputs into this network [14].

A study summarizing identical anatomical characteristics of AKA in 5437 individuals revealed the presence of the AKA in 84.6% of the population and two AKAs in 11.3%. The presence of three AKA was extensively rare [15]. Most times Left-sided and getting originated from T8 and L1 and getting continued from Aorta to ASA [15].

As literature shows that most clinicians and researchers are cautionary about segmental artery injuries as its well determined that segmental artery is responsible for a remarkable part of spine circulation, in this study, we present a case report of spontaneous segmental artery aneurysmal hemorrhage and review the literature for its etiologies, diagnosis, management, and prognosis [16-19].

CASE PRESENTATION

A woman in her 30s with no previous medical history presented to a tertiary neurosurgery center with chronic radicular back pains and neurological symptoms. Lumbosacral MRI was requested that revealed a lesion that suspected the surgeon of a potential malignancy and tumor. Surgery was planned and during the surgery, with access to the objected area, the surgeon encountered massive ungovernable bleeding. So, the area was packed and the patient was transferred to a primary neurosurgery center with advanced facilities in CT Angiography and an MRI. MRI results were suggestive of a thrombosed lesion in the left epidural space; while the T2 view was showing a hyper signal non-flow-void area that was suggestive of an arterial injury (**Fig. 1. and 2.**). Spinal angiography was performed; while due to the packing's in the area, good visualization of the bleeding was not established. A super-selective angiography was performed that showed thin extravasation. Putting the evidence together, the injured artery might be the consequence of a thrombosed aneurysm or more (**Fig. 3.**). Re-exploration was performed and the surgical site decompressed. In the second surgery, massive bleeding was encountered, therefore surgery was stopped and the patient was transferred to the angiography room and a second look of angiography was done.

A pigtail catheter was guided to the abdominal aorta and selective angiography of the lumbar and segmental artery was performed. After injection of contrast material into the lumbar artery, extravasation of injected material through the L1 foramen on the left side was visible.

Extravasation was repaired by the endovascular method. Successful embolization of ruptured segmental artery aneurysm was performed by the super selectively catheterized with a Marathon microcatheter (Medtronic, Minneapolis, MN, USA). After proper positioning under General anesthesia from the right femoral artery, an aneurysm was confirmed by DSA. Next, 25% n-butyl cyanoacrylate (NBCA) glue (glue: lipiodol ratio, 1:3) was injected into the distal part of the feeder under fluoroscopic visualization. After embolization with cyanoacrylate glue, no definite residual was visualized.

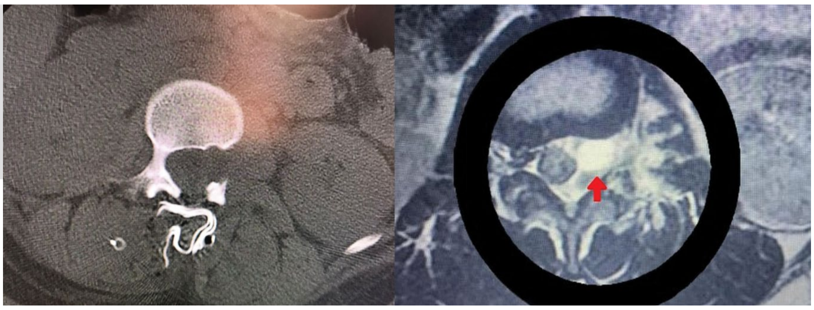


Fig. 1. Axial spiral CT scan of L1 vertebrae shows posterior body erosion & oval shape iso dense extra-axial lesion anterior to the medial facet in the spinal canal, showing a 3.5 cm cyst-like hyper density that was not flow-void.

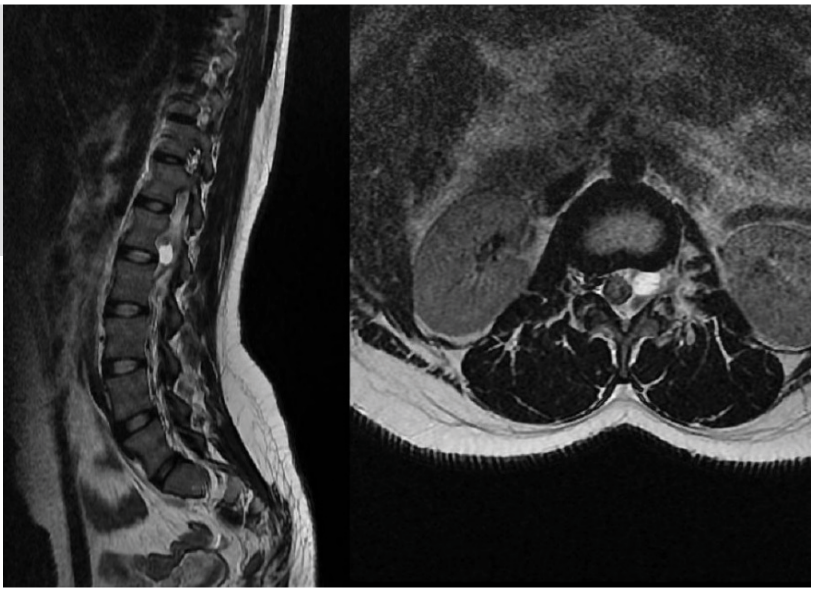


Fig. 2. Axial & sagittal MRI of thoraco-lumbar spine show cystic structure extended from the posterior aspect of the left side of the body of L1 vertebrae adjacent to the cord to the posterior midline of cord with significant pressure of cordon left side neural foramen in level L1-L2 and erosion L1 vertebral body and pressure effect on exiting nerve is noted.

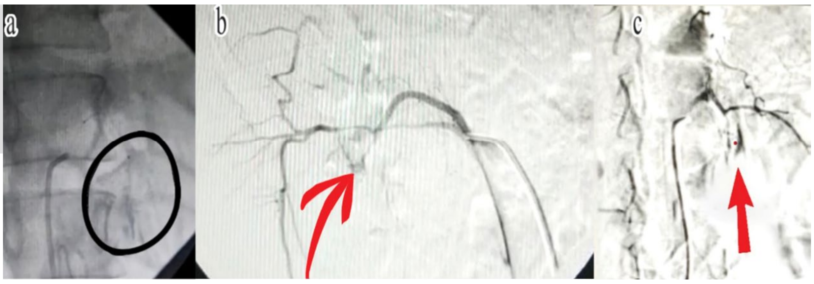


Fig. 3. (a) Angiography and (b) super-selective spinal angiography with micro catheter show extravasation of dye from the left L1 foramen, and (c) after glue administration.

There was no complication after embolization of catheter-directed glue (N-butyl cyanoacrylate). The next day in open surgery, L1-L2 was explored revealing normal neural roots. A unilateral fixation and fusion at the same level of the lesion were conducted through the unilateral resection of pars interarticularis. During the surgery, patient was monitored for neurological deficits that showed it did not occur. Two days later, the patient did not have any symptoms with no further neurological deficits. Patient follow-up was performed routinely for 2 years which was satisfactory (**Fig. 4.**).

DISCUSSION

Aneurysmal hemorrhage of segmental artery is scarcely reported in the literature. Sixteen case reports (15 studies) were found to report ruptured aneurysms of segmental artery and AKA, as shown in **Tab. 1.** [20-34]. Aneurysms and pseudo aneurysms of Intercostal arteries that AKA originates from are reported in the literature with stenting treatment approaches to preserve AKA to avoid spinal cord ischemia [35,36].

Diagnosis

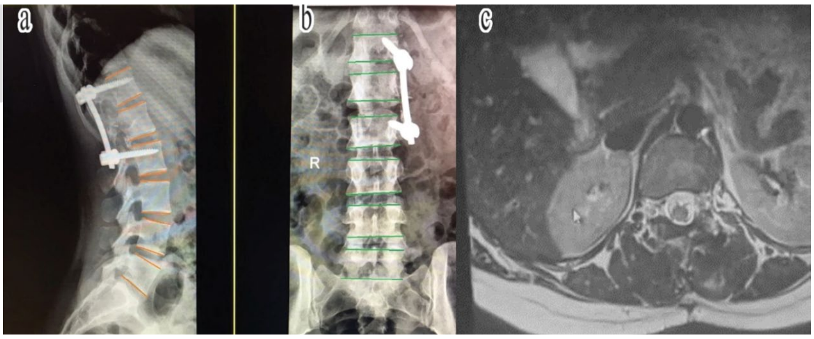
We reviewed all collected case reports for the diagnostic approach. In absence of an MRI device, lumbar metrizamide myelography was first used for the diagnosis of spine lesions along with CT scan methods but most newer studies have used MRI as the preliminary choice of diagnosis [37]. In case of symptoms, el Mahdi, et al. reported patient presented with involuntarily urination, low back pain, and sciatica. As well as our case, they were suspected of an aneurysm of an artery during the open surgery. While most cases are reporting gradual onset manifestations [25]; AKA aneurysms can also manifest more offensively. Acute severe thoracic pain presented in a 13 years old female who was operated on for laminectomy at 1.5 years old [21]. Also, the symptoms as well as neck stiffness and fever could be mimicking other differential diagnoses that should be addressed [32,33]. Guillain-Barre syndrome also deserves to be reviewed in the differential diagnosis list. A lumbar puncture seems to be diagnostic in case of ruptured aneurysmal lesions, being indicative of SAH and for rolling out some other differentials [38].

As we mentioned that our first angiogram was not discriminant and a second selective angiography showed up extravasation in segmental artery, Decker, et al. used selective angiography and segmental artery aneurysm was found [21]. Limaye, et al. used a spinal angiogram following the MRI for the patient having low back pain and paresthesia [17]. MRI seems the most useful primary diagnostic tool as most patients were presenting with low back pain [30].

Armel Junior Tokpo, et al. reported a pseudo aneurysm of the spinal segmental artery after spinal trauma that was treated with endovascular embolization [39].

Seong Son, et al. reported spontaneous resolution of AKA [10].

Fig. 4. Two years follow up radiographic in sagittal (a) & coronal (b) and transverse (c) planes.



Tab. 1. Studies included in the critical review.

ID	Age	Sex	Level	Method	Outcome	ASA	Side
Garcia, et al. [21]	34	Female	T6	NA	Death	NA	NA
Vishteh, et al. [22]	30	Male	T11	Wrapping	Full recovery	Preserved	NA
Berlis, et al. [23]	48	Female	T12	Conservative	Disability remaining	Not preserved	LT
Berlis, et al. [23]	69	Female	T12	Conservative	Full recovery	Preserved	LT
Iihoshi, et al. [24]	60	Female	T12	Conservative	Full recovery	Preserved	LT
Son, et al. [25]	45	Female	T12	Conservative	Full recovery	NA	LT
Doberstein, et al. [26]	59	Male	T11	Conservative	Full recovery	NA	RT
Aguilar-Salinas, et al. [27]	54	Female	T10	Conservative	Full recovery	Not preserved	LT
Massand, et al. [28]	30	Male	T11	Wrapping	Full recovery	NA	NA
Heran, et al. [29]	42	Male	T10	Endovascular embolization	Disability remaining	Not preserved	LT
Todeschi, et al. [30]	57	Male	T9	Conservative	Disability remaining	NA	NA
Aljuboori, et al. [31]	78	Male	T9	Open surgery, clipping	Full recovery	Preserved	NA
Nakamura, et al. [32]	66	Male	T12	Conservative	Full recovery	Preserved	NA
Decker, et al. [33]	13	Female	T12	Endovascular embolization	Full recovery	NA	NA
el Mahdi, et al. [34]	17	Female	T12	Open surgery, clipping	Full recovery	NA	NA
Limaye, et al. [35]	43	Female	T11	Conservative	Full recovery	NA	LT

M. Álvarez Postigo reported an iatrogenic lumbar segmental artery aneurysm that was embolized with a coil [40].

Computed tomography angiography was not useful in diagnosis in two studies (33, 24) [15,30] whereas others reported positive findings [27,33].

Management

The Conservative method is selected by most reported individual cases of segmental and AKA aneurysm (**Tab. 1**); while most cases reported in the literature are not being presented with progressive symptoms and life-threatening conditions. MRI and spinal angiography have been used as follow-ups when conservative treatment is approached. Operative reconstruction of ruptured aneurysm/warping methods or using clipping methods or warping methods is proposed in the literature, mostly when endovascular treatment seems unsafe [25,31]. The endovascular approach might be judged risky due to the size of the aneurysm and the dominance of the spinal artery. For checking embolization safety, Amytal and Lidocaine method is widely being used for anesthetic considerations of interventional neuroradiology. Injection of those medications evaluates the tolerance to vessel occlusion. In the case of AKA aneurysms, the positive Lidocaine test in Doberstein, et al. report was dissuasive of the endovascular approach.

Only 2 studies were found in the literature that used the endovascular approach. Gelfoam was used for embolization in Decker, et al.'s reported case [21]. N-butyl-2-cyanoacrylate and ethiodized oil were used in the endovascular approach of Heran, et al. [27]. Our study reports successful embolization of ruptured segmental artery aneurysm by glue (N-butyl cyanoacrylate). The use of stents to treat aneurysms is known to prevent rupture; while is not reported to be used in segmental artery aneurysms. Riche, et al. report neuroradiology and embolization techniques *via* solids, balloons, and bucrylate to be effective in malformations related to ASA.

Recent studies are providing shreds of evidence about aneurysms in intercostal arteries related to segmental and AKA, being treated by endovascular stenting [41].

CONCLUSION

Segmental artery aneurysms are being presented mostly by lower back pain and neurologic deficits. MRI is the primary diagnostic choice; based on the patient's condition, a head CT scan and lumbar puncture are necessary in most cases. In the case of ruptured ones, lumbar puncture and brain CT scan could be indicative of SAH. The most accurate diagnosis of segmental artery aneurysm is permissible in selective angiograms. Treatment is case-based, having conservative, operative, and endovascular approaches available choices. Endovascular treatment by catheter-directed glue administration seems reputable.

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CONFITS OF INTEREST

The authors declare that there is no conflict of interest.

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