

Research Article

Aneurysm of the Artery of Adamkiewicz: A Systematic Review and Descriptive Analysis

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Abstract

Aneurysms originating from the Artery of Adamkiewicz are incredibly uncommon, with very few reported cases in the current literature. Here, we perform a systematic review of the few cases available in the literature, yielding a total of 15 eligible patients. We extracted certain procedural information and outcomes from these

patients, including how this Artery of Adamkiewicz aneurysm was treated and outcomes from this surgery. We find that a slight majority of patients are treated surgically and that there is some improvement in regard to complete recovery for patients who have been surgically managed. Although there is insufficient

evidence to make any definitive conclusions about treatments for this rare aneurysm, this information remains exciting and relevant for patients who may present with this and for whom surgical versus conservative treatment options may not be a clear decision. We hope that this will provide more information on how this unique finding has been treated based on the current literature and that this will help to guide patient care in rare cases such as these aneurysms.

Keywords: Spinal Sub-Arachnoid Hemorrhage; Spinal Artery Aneurism; Artery of Adamkiewicz; Spinal Cord Infraction; Anterior Spinal Artery

1. Introduction

The Artery of Adamkiewicz [AKA], also known as the great anterior radiculomedullary artery, is one of the most important arterial supplies of the anterior spinal cord [1]. Injury to this artery can result in anterior spinal cord syndrome, affecting the anterior two-thirds of the spinal cord and leading to motor paralysis below the level of the lesion [2]. Aneurysms of the Artery of Adamkiewicz are an extremely rare occurrence, with very few reported cases in the literature. On a broader scale, spinal artery aneurysms as a whole are uncommon, but in these few cases, they are often due to vascular malformations [3]. Those unrelated to vascular malformation or solitary aneurysms are even less likely to be seen in patients [4]. The rupture of spinal aneurysms does not frequently occur, but it can lead to a spinal subarachnoid hemorrhage in a very small percentage of cases (<1%) [4, 5]. Spinal aneurysms are often not found until the patient is symptomatic, at which point there may already be some amount of damage [6]. Taken all together, this demonstrates the rarity of lesions such as these ones, although these are clinically difficult to manage due to the lack of a best-practice consensus on how to approach unique cases involving spinal artery aneurysms of any location.

More specifically, aneurysms of the AKA make up an incredibly small percentage of the causes of subarachnoid hemorrhages of the spinal cord, making it difficult to diagnose and treat patients who may present with this [7]. Patients with a rupture of an aneurysm of the AKA can have symptoms of a sudden severe headache with lower extremity paraparesis, explained by the previously mentioned role of the AKA as an arterial supply to the anterior spinal cord [8]. The extent of the symptoms will depend on the location of lesion following a rupture of the aneurysm of the AKA. In this paper, we present a systematic review of cases involving aneurysms of the Artery of Adamkiewicz based on the current literature available. We find a total of 14 papers fitting our inclusion criteria, yielding 15 patients total. From this, we present relevant information, including the patient's symptoms, comorbidities, diagnosis, and treatment (surgical versus conservative). Interestingly, we find that patients treated surgically had slightly better outcomes in regard to complete recovery. Finally, we acknowledge the limited amount of cases available due to the rarity of these aneurysms and their rupture, but we hope that it will shed light on how treatment plans may be approached for a patient presenting with an uncommon case such as these.

2. Methods

2.1 Study design

Our study was conducted under the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guideline and in accordance with the study protocol developed and agreed by all authors (Figure – 1 PRISMA 2019 flow diagram). We did not require the permission of an institutional review board or patient consent because we only used previously reported data. We used formulated PICO (Population, Intervention, Comparison and Outcomes) question to identify eligible studies-

- Population: Patients with radiological evidence of aneurysm of artery of adamkiewicz.
 - Intervention: Surgical (embolization, clipping etc.) or conservative management.
 - Comparison: Not applicable.
 - Outcomes: Occurrence of spinal or intracranial subarachnoid hemorrhage, outcome of aneurysm, clinical outcome of patient.
- Studies containing original data (case reports, case series, etc.) published in the english language were considered eligible for inclusion.



PRISMA Flow Diagram

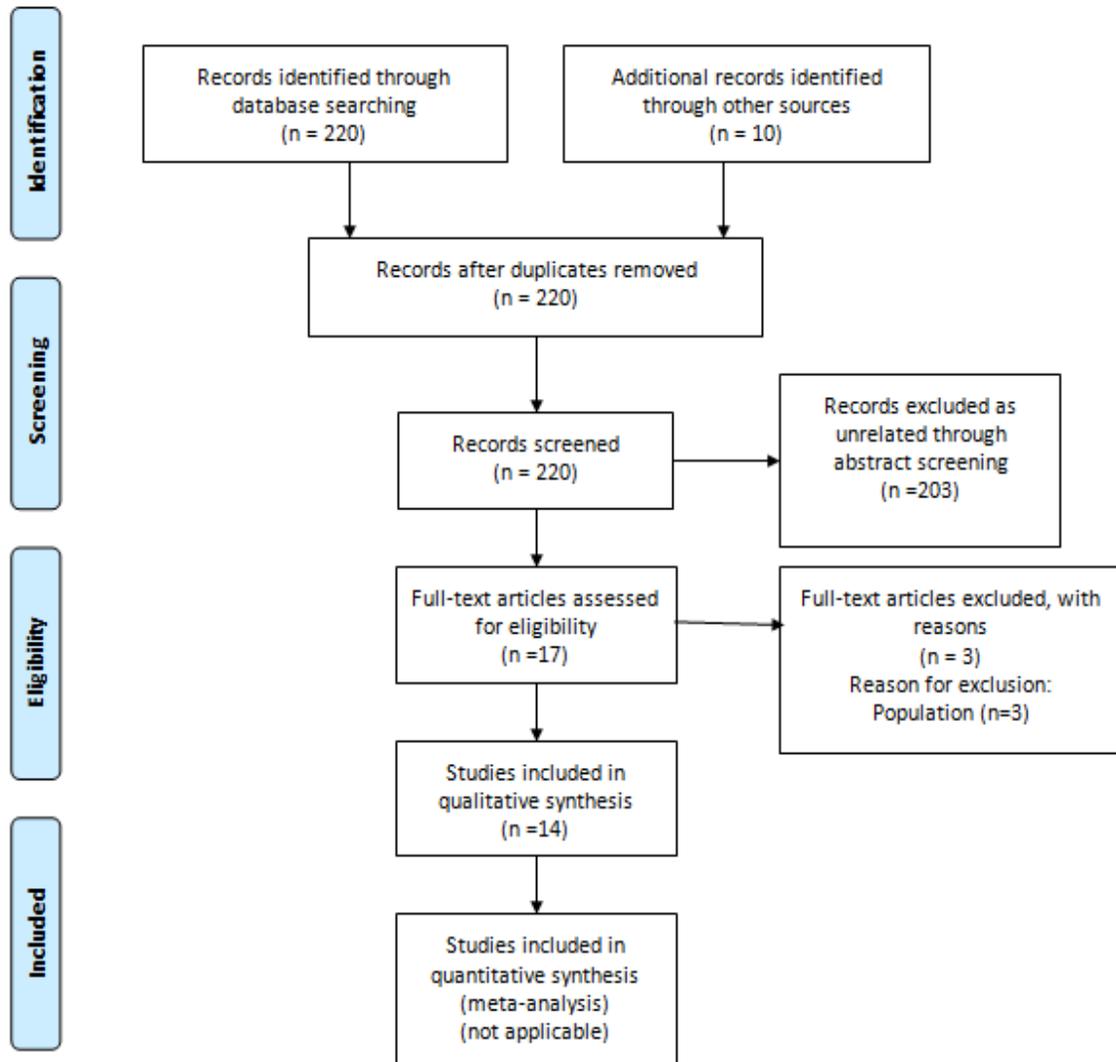


Figure 1: PRISMA 2019 flow diagram.

2.2 Literature search strategy

A comprehensive search of multiple databases from 1960 to February 24th, 2021 in English language, was conducted. The databases included Ovid MEDLINE(R) and Epub Ahead of Print, In-Process & Other Non-Indexed Citations, and Daily, Ovid EMBASE, Ovid Cochrane Central Register of Controlled Trials, Ovid Cochrane Database of Systematic Reviews, and Scopus. A professional librarian performed the search, with direct feedback from the study's principal investigator. We used following search term "Adamkiewicz"; "Aneurysm"; "Subarachnoid Hemorrhage"; "Hemorrhage"; "Pseudoaneurysm"; "dissecting"; "dissection" along with boolean operators "AND" and "OR". Animal and non-human studies were excluded. Two independent reviewers made individual decisions regarding inclusion of studies. All disagreements were resolved after reaching consensus.

2.3 Study selection and data extraction

Duplicate studies were omitted from all citations found during the comprehensive literature search. Pairs of unbiased reviewers screened titles and abstracts for inclusion. The same pairs of independent reviewers registered related abstracts, retrieved and screened full text by using standardized, pre-piloted spreadsheets.

The extraction was completed with a focus on the following data - 1) Study characteristics (first author, year of publication, number of patients); 2) Patient characteristics (age, gender, clinical presentations, comorbidities, proposed etiology); 3) Aneurysm related characteristics (location, morphology, aneurysm size, bleeding, imaging findings); 4) Treatment-related characteristics (treatment provided; surgery and/or conservatives, nature of surgical approach); and finally

5) outcome (clinical outcome, fate of aneurysm). After reaching a consensus with further input and cross-checking from a senior reviewer, we resolve all disagreements.

3. Results

Our search strategy and rigorous inclusion protocol yielded 14 articles eligible for review [4-16]. A total of 15 patients was our study population. Among them the mean age was 46.93 years (Range - 15-78 years). Male female ratio was 8:7 (53%:47%). Most common presenting complaint was back pain (n=12) followed by headache (n=5); being present in 80% and 33% of the study population respectively. Hypertension was the most common comorbidity among the group (n=5). 86% of the aneurysm in the study population was fusiform in shape (n=13) and apart from 3 cases all were confirmed by a spinal angiography (Table: 1).

Keeping consistent with the most common complication of aneurysm being hemorrhaged most of our patients had evidence of either spinal or intracranial subarachnoid hemorrhage (SAH) or both. 80% of the study population (n=12) had spinal SAH and 40% had intracranial SAH (n=6). 6 of the patients had evidence of both (40%). 8 out of 15 patients were managed conservatively (53%); while 6 underwent various surgical procedures (40%). One patient died before any treatment was attempted. Complete recovery was attained in 66% of patients (n=10) while recovery was partial for the rest. Among the group who were treated conservatively the rate of complete recovery was decreased slightly upto 62.5% while in the group who was treated with surgical methods rate of complete recovery was up significantly upto 83% compared to the whole study cohort as a single group (Table: 2).

Author	Patients	Presenting symptoms	History of Hypertension	Proposed etiology	Type of Aneurysm	Morphology
Vishteh AG	30, M	Headache, back pain, and lower extremity paresis	No	Exertion	Pseudoaneurysm	Fusiform
Aguilar-Salinas P	54, F	Headache, back pain, emesis, lower extremity paresis	Yes	Idiopathic	Aneurysm	Fusiform
Son S	45, F	Headache, back pain, nausea, lower extremity paresis	No	Idiopathic	Pseudoaneurysm	Fusiform
Todeschi J	57, M	Epigastric pain with back pain, diffuse lower limb paresthesia	No	Idiopathic	Aneurysm	Fusiform
Berlis A	48, M	Abdominal pain and paraplegia with bilateral anesthesia below T12 & nuchal rigidity, neck pain.	No	Systemic mycosis (Candida)	Aneurysm	Fusiform
	69, F	Back pain and walking impairment; paraplegia; urinary retention	No	Idiopathic	Pseudoaneurysm	Fusiform
Doberstein CA	59, M	Severe back pain and walking impairment	No	Idiopathic	Aneurysm	Fusiform
Garcia CA	34, F	Headache, emesis, paraplegia	Yes	Pregnancy or Mycotic aneurysm	Aneurysm	Fusiform
Massand MG	30, M	Back pain, lower extremity weakness, paresthesia	N/A	N/A	Aneurysm	Fusiform
Iihoshi S	60, F	Back pain, headache, lower limb pain, nausea, moderate neck stiffness.	Yes	Idiopathic	Pseudoaneurysm	Fusiform
Aljuboori Z	78, M	Back pain and weakness in both lower limbs	Yes	Idiopathic	Aneurysm	Fusiform
Nakamura T	66, M	Posterior cervical pain, lower back pain, fever, neck stiffness	Yes	Exertion	Aneurysm	Fusiform
Heran MKS	42, M	Paraplegia, seizures, emesis, decreased level of consciousness	No	MDMA abuse	Pseudoaneurysm	Fusiform

Robert E. Decker	14, F	Midline pain in the lower thoracic region, transient weakness of legs, episodes of sudden back pain and neck stiffness	No	N/A	1 true aneurysmal, 1 pseudoaneurysmal sac	saccular
M A el Mahdi	17, F	Low back pain, right sciatica, urgency of micturition, relative weakness of the plantar flexors of the right ankle.	No	Idiopathic	N/A	N/A

MDMA- methylenedioxyamphetamine, N/A- not available

Table 1: Demographic characteristics of patients.

First Author	Age, Sex	Location of aneurysm	Aneurysm size (mm)	Evidence of Spinal SAH	Evidence of Intracranial SAH	Treatment Approach	Aneurysm outcome	Evidence of ASA preserved	Recovery	Duration of recovery (Weeks)
Vishteh AG	30, M	T11	N/A	Yes	No	Muslin wrapping	N/A	Yes	Complete	52
Aguilar-Salinas P	54, F	Left T10	4x8	Yes	No	Conservative	complete occlusion	N/A	Partial	43
Son S	45, F	Left L1	3x11	Yes	Yes	Conservative	Residual intradural collection	N/A	Complete	9
Todeschi J	57, M	T9	N/A	Yes	Yes	Conservative	N/A	N/A	Partial	N/A
Berlis A	48, M	T12	5x7	Yes	No	Conservative	complete resolution	No	Partial	N/A
	69, F	Left L1	2.5x8	Yes	No	Conservative	complete resolution	Yes	Complete	3
Doberstein CA	59, M	Right T10, T11	N/A	Yes	No	Conservative	N/A	N/A	Complete	28
Garcia CA	34, F	T6	5x3	Yes	No	N/A	N/A	N/A	Death	N/A

Massand MG	30, M	T11	N/A	No	No	Partial T11 vertebrectomy with muslin wrapping	complete resolution	N/A	Complete	N/A
Iihoshi S	60, F	Left T11, T12	2x8	Yes	Yes	Conservative	complete resolution	Yes	Complete	N/A
Aljuboori Z	78, M	T9	5.8x2.8	Yes	Yes	Surgery and clipping	complete resolution	N/A	Complete	197
Nakamura T	66, M	T12-L1	10 in diameter	Yes	Yes	Conservative	complete resolution	Yes	Complete	N/A
Heran MKS	42, M	Left T9, T10	N/A	Yes	Yes	Endovascular embolization	complete resolution	Occluded at T9 level	Partial	26
Robert E. Decker	14, F	T12	NA	No	No	Embolization	complete resolution except proximal 3/4mm	N/A	Complete	N/A
M A el Mahdi	17, F	T12	30 x 30 x 22	No	No	clipping then emptying by aspiration	N/A	N/A	Complete	4

Table 2: Aneurysm’s characteristics and outcome.

4. Discussion

The artery of Adamkiewicz or AKA arises from a variable region like one of the 8th-12th intercostal arteries and joins the anterior spinal artery to supply the anterior two-thirds of the thoracolumbar spinal cord. It is commonly found on the left side in approximately 75% of cases. As the artery of Adamkiewicz supplies T8 to the spinal cord's conus medullaris, so obstruction or rupture of this artery results in anterior spinal artery syndrome.

4.1 Spinal SAH due to aneurysm of artery of adamkiewicz rupture

Subarachnoid hemorrhage (SAH) accounts for 10-15% of strokes. Among SAH most common causes are intracranial, so the spinal cause of SAH is rare. The most frequent causes of spinal SAH are trauma, spinal tumor, vascular malformation. Rarely, it can be due to an aneurysm in the artery of Adamkiewicz.

4.2 Clinical feature

Like other spinal SAH causes, patients can present with sudden-onset back pain, often accompanying lower extremity symptoms including weakness and/or paresthesia, sciatic pain, paraparesis, sensory disturbance, myelopathy, urinary incontinence.

4.3 Diagnosis/Imaging

All patients with suspected spinal SAH require urgent contrast-enhanced MRI of the spine with MR angiography to identify the source of bleeding and the extent of spinal cord involvement. CT scan angiography may also help to identify the extent of SAH and rule out any bony injury. However, digital subtraction angiography or DSA provides a definitive assessment of the lesion's anatomy, identifies the entering and exiting blood vessels, and rules out any associated vascular lesions that may accompany the aneurysm. If an aggressive intervention is not immediately called for,

the aneurysms may be evaluated for progression or regression with follow-up angiography. When and if surgical or endovascular interventions are considered, the most crucial consideration to be made is whether or not the parent vessel can be preserved [17].

4.4 Morphology

In contrast to intracranial aneurysms, mostly saccular and found on bifurcation sites, spinal aneurysms are fusiform and found on non-bifurcation sites. Since they arise from minute parent arteries, it makes any intervention modalities difficult without a distinct neck to clip and or an aneurysm sac to coil. Therefore, alternative ways must be considered, including parent vessel sacrifice, lesion resection, or wrapping. From a technical point of view, either microsurgical or endovascular intervention could result in the sacrifice of the parent vessel (0.5mm) [6]. The latter technique is quite challenging to perform as it bears significant risks. The risks arise from the complexity of spinal vasculature, evidently, the small or tortuosity of the AKA. A spinal angiogram is the first step to evaluating the features of an aneurysm in this artery and whether it is associated with any vascular malformations. An exciting and rare case finding was reported in 1943 regarding a phenomenon called Arachnoiditis secondary to spinal SAH, which occurs months to years after the causative agent [5, 18-20]. Although the pathophysiology is entirely not understood, it is suspected that it occurs due to prolonged chronic inflammatory reactions in the pial-arachnoid membrane [5, 21, 22].

4.5 Management and outcome

This systematic review examined the published case of patients with radiological evidence of aneurysm at the artery of Adamkiewicz, focusing on mortality, clinical outcome in surgical versus nonsurgical management. Among included fifteen patients, eight and six patients managed conservatively and surgically, respectively. In

between 3 weeks to 46 months, follow-up clinical outcomes like the complete recovery rate are slightly higher in the surgically treated group (83%) than the nonsurgical management group. Due to such a case's

extreme rarity and the lack of data, detailed analysis has not been done and remains equivocal. To improve patient care, we need adequate data to conclude.

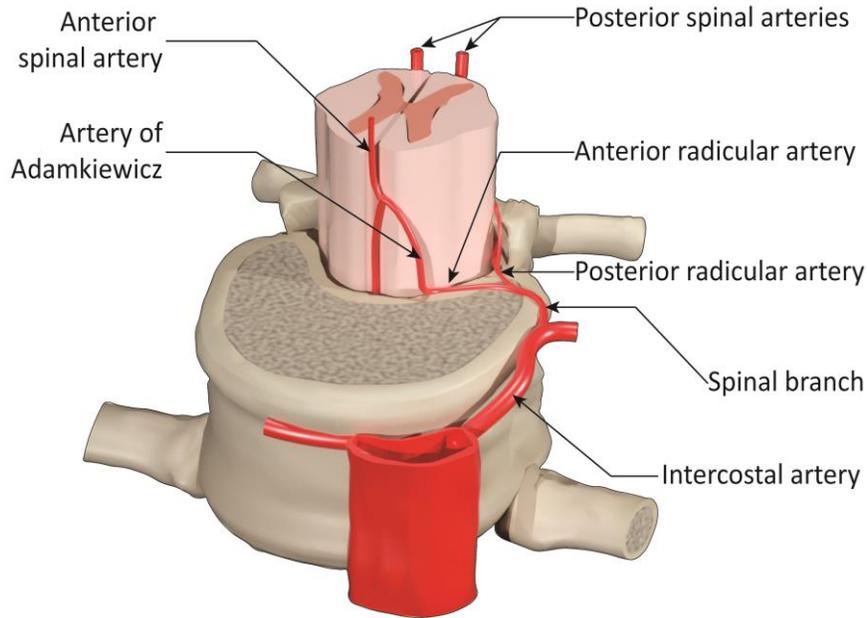


Figure 2: Illustration of artery of Adamkiewicz: origin and reinforcement of anterior spinal artery.

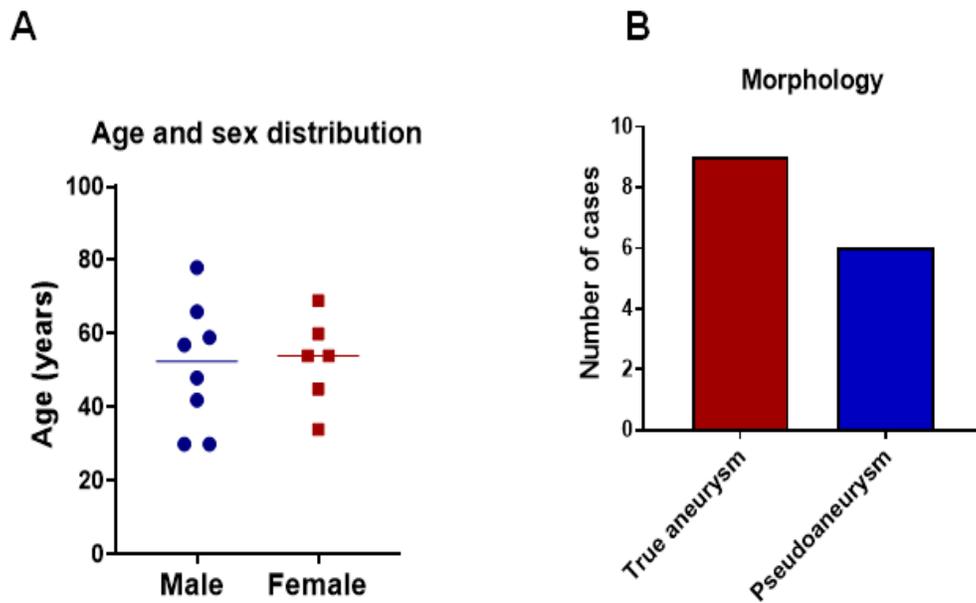


Figure 3: Age, sex and morphological distribution of cases.

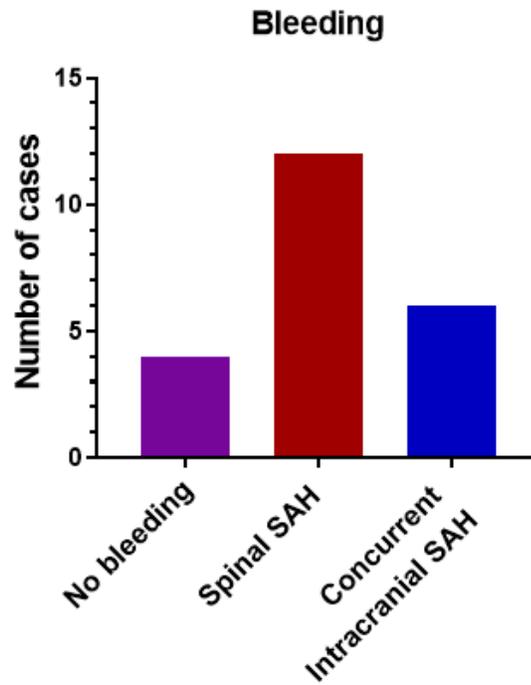


Figure 4: Pattern of bleeding from Aneurysmal rupture.

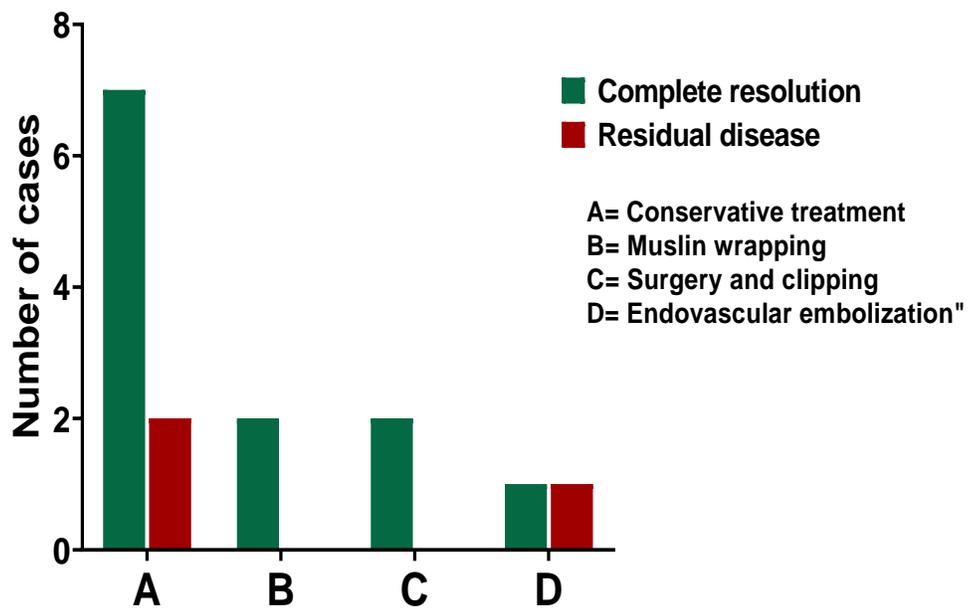


Figure 5: Outcome data.

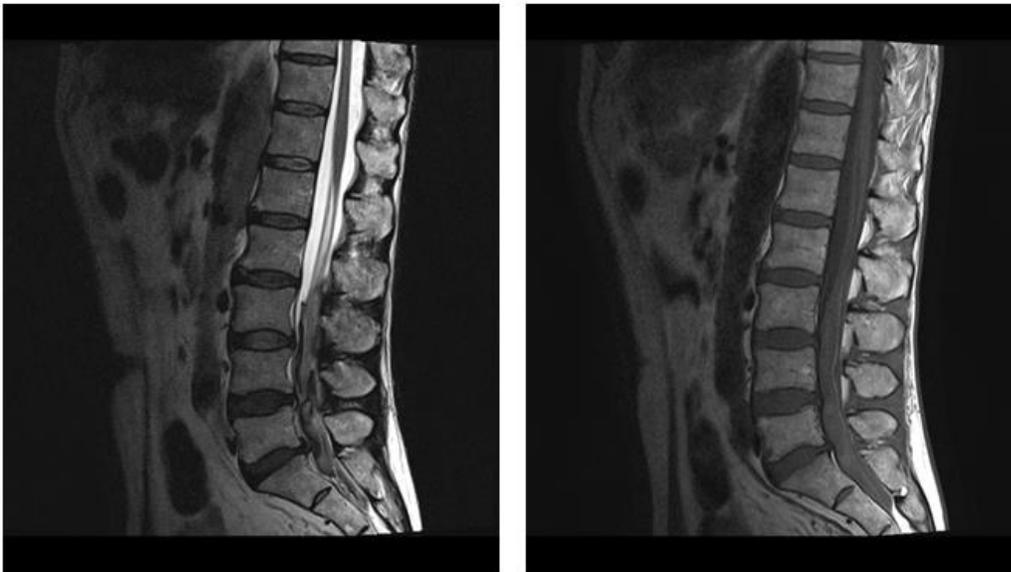


Figure 6: MRI of the lumbosacral spine: Sagittal T1 demonstrates hyperintensity, and the T2 sequence reveals hypointensity in subarachnoid space and severe compression of the cauda equina. The findings are consistent with a subarachnoid hemorrhage. Source: <https://radiopaedia.org/>

5. Conclusion

Aneurysm of the artery of adamkiewicz is extremely rare and may have multiple etiology ranging in a wide distribution of patient characteristics. While previous literature review on this very topic had heavily leaned towards treating the SAH resulting from the aneurysm with conservative approach due to risk of rebleeding from surgery [7]. our analysis found significantly better outcomes in patients treated with surgical methods. While the absolute reduction of risk of incomplete recovery was 21% with surgical modalities due to smaller size of our study population and lack of control of confounders, we suggest taking an individualized approach towards determining the treatment protocol. A randomized control trial may have been the best possible method to determine the treatment choice but due to the rare & acute nature of the condition it seems unlikely to happen.

Conflict of Interest

All authors declare no conflict of interest.

Disclosures

The authors have no financial conflicts of interest to declare.

Human/Animal Rights

This article does not contain any studies with human or animal subjects performed by any of the authors.

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