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ORIGINAL ARTICLE

Clinical features and treatment strategy of paraspinal arteriovenous shunt (PAVS): a systematic review with individual participants data meta-analysis

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Abstract

Background Because of the rarity of the disease, paraspinal arteriovenous shunt (PAVS) is not well recognized, and therapeutic options remain controversial. To introduce a rare disease of PAVS and demonstrate its etiology, clinical features, treatment options, and outcomes, we presented a case report and conducted a systematic review and individual participants data (IPD) meta-analysis.

Methods Studies regarding on PAVS were integrated and IPD were obtained including patients' demographics, disease etiology, clinical and radiologic features, clinical courses and outcomes. Clinical manifestation and treatment outcomes were reviewed, and comparison analysis (cervical versus thoracolumbar) were performed. Further, logistic regression analyses were conducted to identify the poor prognostic factors (incomplete obliteration).

Results Fifty-two articles were selected, and 88 patients enrolled. General and location-specific characteristics of PAVSs were identified: '3/4 of the isolated and 1/4 of the associated etiology', 'bruit, thrill, or murmur (cervical) and weakness (thoracolumbar) as common symptoms', '40% multiple feeders', and '22% intradural venous involvement'. Endovascular treatment was usually preferred (75%). Of 88 enrolled patients, 18 patients showed incomplete obliteration (20.5%). In multivariate analysis, 'etiologies of systematic genetic dysplasia ($P = 0.031$) and trauma (negatively, 0.038)' were significantly associated with incomplete obliteration. The parameters of 'multiple feeders (0.066)' and 'combined approach (negatively, 0.065)' are verified only in univariate analysis.

Conclusion General as well as location-specific characteristics of PAVS is successfully demonstrated. Approximately 20% of the incomplete obliteration is noted, and three potential poor prognostic factors are identified, namely, 'etiology of systematic genetic dysplasia (positive) and trauma (negative)', 'combined approach (negative), and 'multiple feeders'.

Keywords Spinal · Paraspinal · Arteriovenous · Fistula · Shunt

Introduction

Over several decades, clinicians have made efforts to classify spinal arteriovenous shunts (AVSs) with respect to their location, venous drainage pattern, and etiology [1–5].

To date, **"**Paraspinal (or paravertebral) AVS (PAVS)**"** has not received attention in the literature owing to its rare incidence and unfamiliarity. They vaguely belonged to spinal AVS as epidural or extradural types, until Rodesch first described it with the term "PAVS" [5]. Usually, they are located outside the spinal canal and are responsible for neurological symptoms related to possible mechanisms underlying cord compression by ectatic veins, venous congestion, or arterial steal [5]. Recently, a new classification of PAVSs has been reported with respect to their origins and drainage pathways (Stuttgart classification, Table 1) [6]. Due to the pathophysiological and locational differences between 'spinal' and 'paraspinal' AVS, a tailored approach for assessing PAVS is mandatory. However, studies on PAVSs are limited to case reports, and their etiology,

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Table 1 Stuttgart classification of PAVSs

Origins	Venous drainage patterns
Isolated	Intraspinal or extraspinal
(1) Acquired	
(2) Traumatic	
(3) Congenital without identifiable genetic hereditary disorder	
Associated with,	
(4) Metameric link	
(5) Systematic genetic dysplasia	

clinical manifestations, and treatment strategies remain uncertain.

Herein, the authors introduce a patient with a large PAVS on the lumbar spine for better understanding of the disease. Further, we integrated the currently available literature on PAVS and tried to demonstrate its etiology, clinical manifestation, and treatment strategy by conducting a systematic review and individual participant data (IPD) meta-analysis.

Illustrative case of ours

A 21-year-old male who suffered from backache visited our institute. Preoperative lumbar magnetic resonance imaging (MRI) and computed tomographic angiography (CTA) revealed engorged vascular structures located along the L4 vertebral body into the left L4 neural foramen. PAVS was fed by the left L4 lumbar segmental artery, which directly originated from the abdominal aorta and drained into the epidural venous structures of the intraspinal and extraspinal canals. The diameter of the feeding artery was 13 mm. Spinal catheterized angiography followed: the arterialized veins formed stepladder patterns through the foramens. We additionally examined the common iliac artery and every adjacent radicular artery; however, only a single feeder of the left L4 artery was found (Fig. 1a).

The clinical decision for surgery was made considering that a shunt could be eliminated by direct surgical ligation of 'a single feeder', whereas endovascular intervention could confer a risk due to the shunts' large diameter, uncertain margins, and potential mass effect with root compression. The conventional retroperitoneal approach was performed, and ligation of the feeder was achieved successfully (Fig. 2a). However, postoperative CTA showed that the proximal feeder was occluded, but retrograde contrast filling into the distal pouches was noted. An additional angiography revealed multiple feeders that were not previously visible in the preoperative images (Fig. 1b).

Therefore, secondary stage of embolization was scheduled. We embolized the venous pouches located on the extraforaminal area because most feeders ran from the outside of the foramen into the epidural veins and embolization of the foraminal and intraspinal parts might cause root and thecal sac compression. Seventeen detachable coils were embolized at the extraforaminal pouches through the collateral vessel of the lateral sacral artery (Fig. 2b), and final angiograms showed complete obliterations (Fig. 1c). The patient recovered soon after. Postoperative MRI (6-month) showed collapsed venous structures and decompressed thecal sacs at every vertebra.

Methods

For understanding of the disease, the systematic review and IPD meta-analysis were conducted following the Preferred Reporting Items for a Systematic Review and Meta-analysis of Individual Participant Data (The PRISMA-IPD Statement) [7].

Eligible criteria and search strategy

We enrolled every publication that reported patients with PAVS. Articles had to be written in English and limited to the human subjects including all case reports, technical notes, and original articles and were eligible if they presented the details of the neurologic status of a patient, radiologic feature of PAVSs, treatment modality, and clinical courses of patients. Articles were excluded if they reported patients who were misdiagnosed as other types of spinal AVS, were written in other languages, or published in the pre-embolization era.

Articles are extracted through PubMed and Google Scholar searches of the terms ["paravertebral" OR "paraspinal"] AND ["(arteriovenous) fistula" OR "(arteriovenous) malformation" OR "(arteriovenous) shunt"] with every combination. In addition, we included articles by searching the terms ["vertebro-vertebral" OR "metameric"] AND ["shunt" OR "malformation" OR "fistula"], because these terms have the potential of being classified as PAVS. The search was supplemented by manually surfing the reference lists within all relevant articles.

Study selection and data extraction

Pairs of reviewers independently assessed each abstract for eligibility. Every discrepancy was modulated by a third reviewer. Included articles were retrieved as full texts and screened in duplicate. Articles which were not accessible for the individual data were excluded. The PRISMA-IPD flow diagram is shown in Fig. 3.

After the study selection and patient enrollment, the following information was obtained: (1) author names and the year of publication, (2) number of patients and their age and gender, (3) etiology and history including trauma, prior catheterization, hereditary, or genetic disease, (4) initial physical and neurologic status, (5) radiologic features of PAVSs, (6) clinical decisions and courses according to the treatment, (7) outcomes including mortality, morbidity, and complications.

Assessment of risk of bias

As shown in Table 2, the searched articles were limited to the form of case reports or technical notes, and no comparative study was observed. Given that we enrolled articles from which the individual data (in form of each case) could be accessed, no risk of bias from selected articles exists.

Patient classification and data management

Of 92 patients from 52 selected articles, 5 patients were excluded due to the misdiagnosis, and 1 patient of the present case report (ours) was added. Totally, 88 patients were enrolled (Table 2). After data gathering, outcomes were evaluated and classified into four categories: (1) complete obliteration without neurologic deficit, (2) complete obliteration with neurologic deficit, (3) incomplete obliteration, and (4) not determined. 'Incomplete obliteration' was defined under the following conditions: (1) the initial surgery failed and required revision; (2) embolization was performed partially; or (3) recurrence of shunts was identified during follow-up.

Finally, the enrolled populations were classified according to two different standards of location (cervical vs. thoracolumbar spine) and prognosis (complete obliteration vs. incomplete obliteration, after extracting 7 not-determined patients). The flowchart of article selection, patient enrollment, and classification are shown in Fig. 3.

Statistical analyses

Statistical analyses were performed using SPSS software version 23 (IBM, Chicago, IL, USA). Continuous variables were expressed as mean ± standard deviation. Categorical variables were expressed as frequencies and percentages. To clarify the differences between PAVSs located on cervical and thoracolumbar spines, *independent t tests, Chi-square tests,* and *Fisher's exact tests* were conducted. Statistical significance was defined as $p < 0.05$. *Univariate* and *multivariate logistic regression analyses* were conducted to identify the factors associated with poor outcomes (= incomplete obliteration).

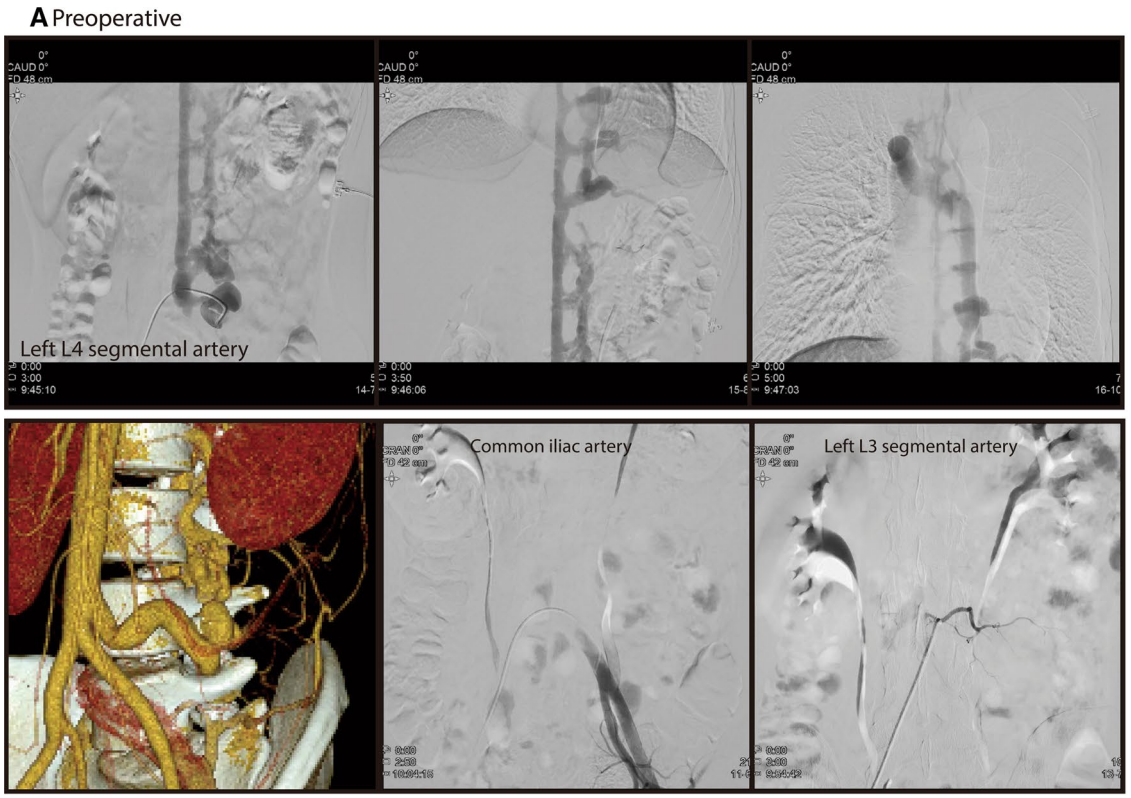
Results

Table 2 shows the list and individual data of 88 enrolled patients (from 53 articles including ours).[8–59]

In Table 3, the general demographics of enrolled patients and baseline characteristics of PAVS are presented, and we can identify the clinical manifestation as well as treatment outcomes of PAVS. Specifically, PAVSs were diagnosed on the cervical spines of 50 patients, and on the thoracolumbar spines of 38 patients. Several characteristic features of PAVS were demonstrated including mean age of 29 years, no gender predominance, 27.3% of 'associated etiology of Stuttgart classification (metameric link or systematic genetic dysplasia)', 'bruit, thrill, or murmur' and 'weakness' as common symptoms, 40% of 'multiple feeders', and 20% of 'intradural venous involvement'. In general, 66 patients received endovascular interventions (75%) and treatment failure of incomplete obliterations are observed in 18 patients (20.5%).

The location-specifical clinical features of PAVS are also identified by conducting the comparative analysis (cervical vs. thoracolumbar PAVSs), and parameters of 'female gender ($p = 0.027$)', 'traumatic etiology ($p = 0.001$)', 'bruit, thrill, or murmur ($p = 0.004$)', and 'cranial symptom or sign ($p = 0.001$)' are significantly highly observed in cervical PAVSs. On the contrary, parameters of 'male gender ($p = 0.027$)', 'acquired etiology ($p = 0.001$)', 'weakness ($p = 0.046$)', 'deformity ($p = 0.003$)', 'bowel or bladder signs ($p = 0.011$)', and 'multiple feeders ($p = 0.004$)' were significantly frequently noted in thoracolumbar PAVSs. In terms of treatment and outcomes, endovascular treatment was preferred in cervical PAVSs ($p = 0.032$); however, the proportion of "incomplete obliteration" was similar between the groups (18.0% vs. 23.7%, $p = 0.078$).

Tables 4 and 5 demonstrate the results of univariate and multivariate logistic regression analyses for identifying the poor prognostic factors of incomplete obliteration. Factors of 'etiology of trauma (negatively, $p = 0.006$), congenital disease (negatively, $p = 0.012$), or systematic genetic dysplasia ($p = 0.012$)', 'multiple feeders ($p = 0.002$)', and 'treatment modality of combined approach (negatively, $p = 0.030$)' were significant in univariate analyses. In multivariate analyses, 'etiology of systematic genetic dysplasia ($p = 0.031$)' was identified as a significant poor prognostic factor and 'etiology of traumatic ($p = 0.038$)' was inversely related to incomplete obliteration.


A Preoperative
Left L4 segmental artery
Common iliac artery
Left L3 segmental artery
B Post-surgery
Internal iliac artery
Left L3 segmental artery
C Post-embolization
Common iliac artery
Left L3 segmental artery

◂**Fig. 1** CTA images and catheterized angiograms in chronological order. Preoperatively (**a**), angiograms of the left L4 artery revealed a high-flow PAVS that drained into the hemiazygos and azygos venous systems through paravertebral and epidural venous pouches with ectatic changes in a stepladder pattern (first low). No visible other feeders in super-selective angiograms (second low). After surgery (**b**), distal venous structures filled by retrograde invisible feeders reappeared (white arrowhead). After coil embolization (**c**), abnormal vasculatures shrunk and disappeared

Discussion

Herein, we report a case of a patient with a large lumbar PAVS who underwent surgery, but it was not successful. To the best of our knowledge, our case is the largest PAVS in the literature with a feeding artery diameter of 13 mm. Regarding the systematic review and IPD meta-analysis, several characteristic features of PAVS were identified: '27.3% of associated etiology', 'bruit, thrill, or murmur on cervical and weakness on thoracolumbar PAVS as common symptoms', '40% of multiple feeders', and '22% of intradural venous involvement'. Particularly, 18 patients showed incomplete obliterations (18/88, 20.5%), and the parameters of 'systematic genetic dysplasia', 'presence of multiple feeders', and 'treatment modality other than combined approach (negative association)' were potentially associated with poor outcomes.

Clinical features of PAVSs

Traditionally, the subject of most previous investigations focused on 'Dural AVS' [60], which are usually small and both surgical and endovascular interventions showed good outcomes. On the contrary, PAVS is rare and usually forms relatively large vascular malformations (as in our case) [6]. Its complexity of hemodynamics can be challenging to the physicians and it have a higher possibility of association with genetic or hereditary disease. However, publications regarding PAVSs are scattered and its clinical features are not fully established.

In terms of the etiology of the disease (Stuttgart classification), 64 of the 88 patients were classified as 'isolated,' whereas 24 were 'associated' PAVSs (72.7% vs. 27.3%). In other words, a quarter of the patients is associated with etiology of metameric link or systematic genetic hyperplasia, and they presented high rate of treatment failure (11/21, 52.38%). It is generally accepted that the PAVS associated with genetic or hereditary disease usually shows complex pathological hemodynamics and affect larger lesions than that of isolated PAVS; therefore, incomplete obliteration frequently occur [20]. On contrary, nearly all patients with trauma-associated PAVS are successfully treated (21/22, 95.5%) and they usually located in the cervical spines ($p = 0.001$). It might be related to the often-reported cases of jugular catheterization injuries [15, 27, 33, 56], which usually showed relatively simple hemodynamic pathology of a single fistulous point.

In terms of clinical presentation, symptomatic PAVSs mostly present reflux into the epidural venous system with venous hypertension which leads to venous stenosis, large pouches, or thrombosis due to longstanding high-flow shunts [35]. By conducting this meta-analysis, we could determine commonly reported symptoms and signs with respect to the location of the PAVS. Specifically, cervical PAVSs were significantly associated with 'bruit, thrill, or murmur' which might be related to the large vascular malformation located beneath the skin of the posterior neck, and 'cranial signs or symptoms (such as cranial nerve palsy, and subarachnoid hemorrhages in the cranium)' potentially owing to venous hypertension. On the other hand, various presentations of 'weakness', 'spinal deformity', and 'bladder and bowel dysfunction' were frequently observed in thoracolumbar PAVSs. A large formation of arterialized venous pouches or chronic venous hypertension might lead to such myelopathic symptoms or signs.

Interestingly, the incidence of intradural venous drainage was 21.6%, which was lower than that reported in usual spinal AVSs [61]. With pathophysiological evidence, PAVSs usually arise from relatively large arteries of the vertebral, intercostal, or lumbar segmental arteries and are located either at the main trunk and/or its branches. Every feeder supplies all tissues on a given metamere, with the exception of the spinal cord [14]. In other words, PAVSs are located on the outer spinal canal and rarely lead to intradural venous changes because the communication between intra- and extra-dura is relatively narrow and far to be affected.

Treatment strategy of PAVSs

In terms of general outcome, 18 of 81 patients (excluding 7 not-determined cases) showed unsuccessful clinical courses including failed surgery, partial embolization, or recurrence during follow-up (22.2%). Regarding the regression analyses, three factors were associated with poor outcomes: (1) *etiology (Stuttgart classification),* (2) *presence of multiple feeders, and* (3) *treatment modality*.

(1) *Etiology*: In the literature review, patients who had an accompanying genetic or hereditary disease showed a more severe presentation of clinical symptoms and signs, such as a severe deformity of the thoracic spine or huge nevi and mass formation on the back. These lesions sometimes accompanied complex nidus formation, which might

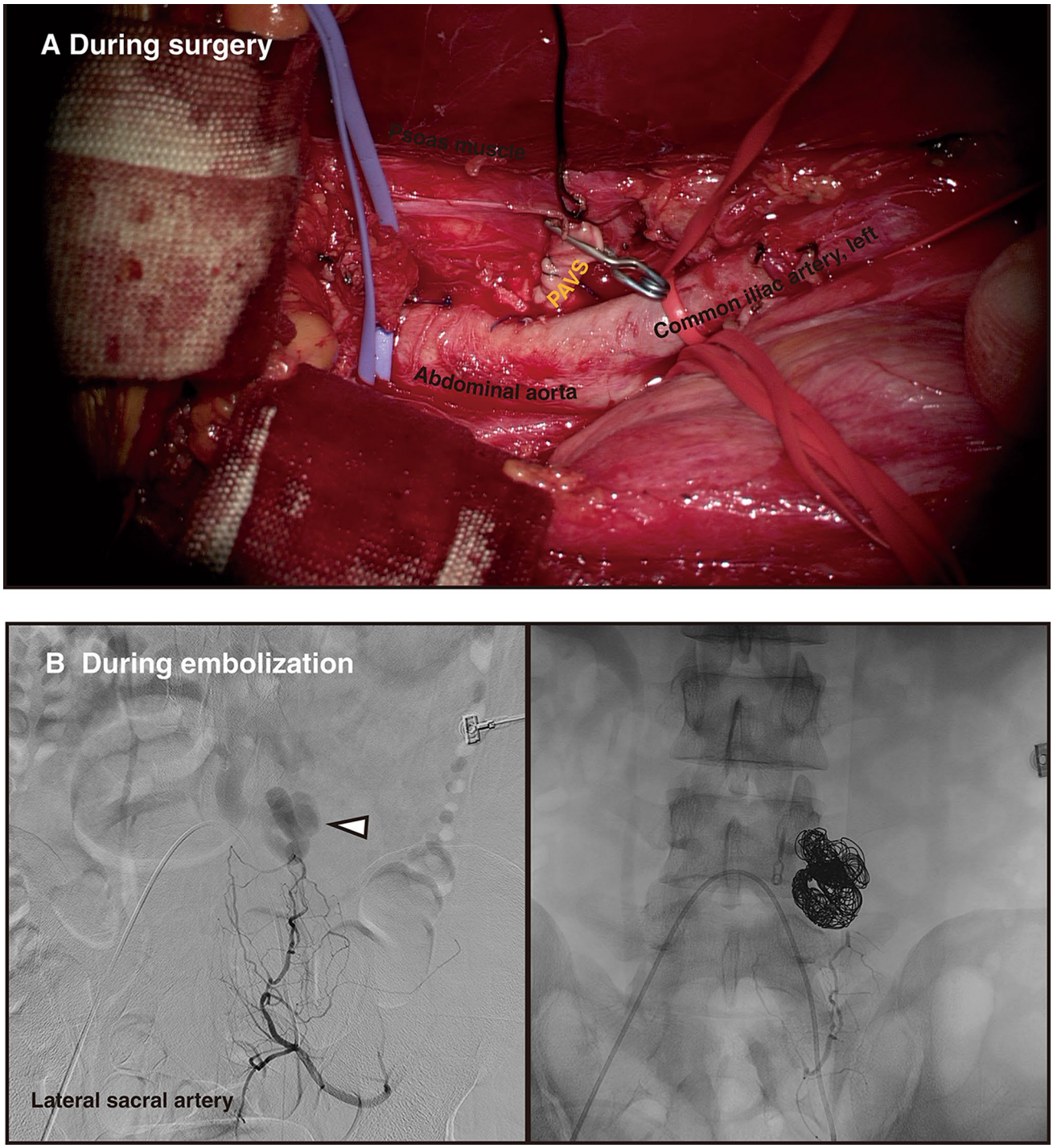


Fig. 2 Images during surgery and coil embolization. In surgical view (A), a large feeder originating from the abdominal aorta adjacent to the iliac artery bifurcation was observed and successful ligation was performed. Coil embolization through the transarterial route of 'the lateral sacral artery' was performed with a 5-Fr guide catheter (JL4, Cordis, Miami, FL), a microcatheter (Excelsior SL-10, Stryker, Kalamazoo, MI), and a microguide wire (Synchro 10, Stryker). Seventeen detachable coils were deployed at the extraforaminal area (B)

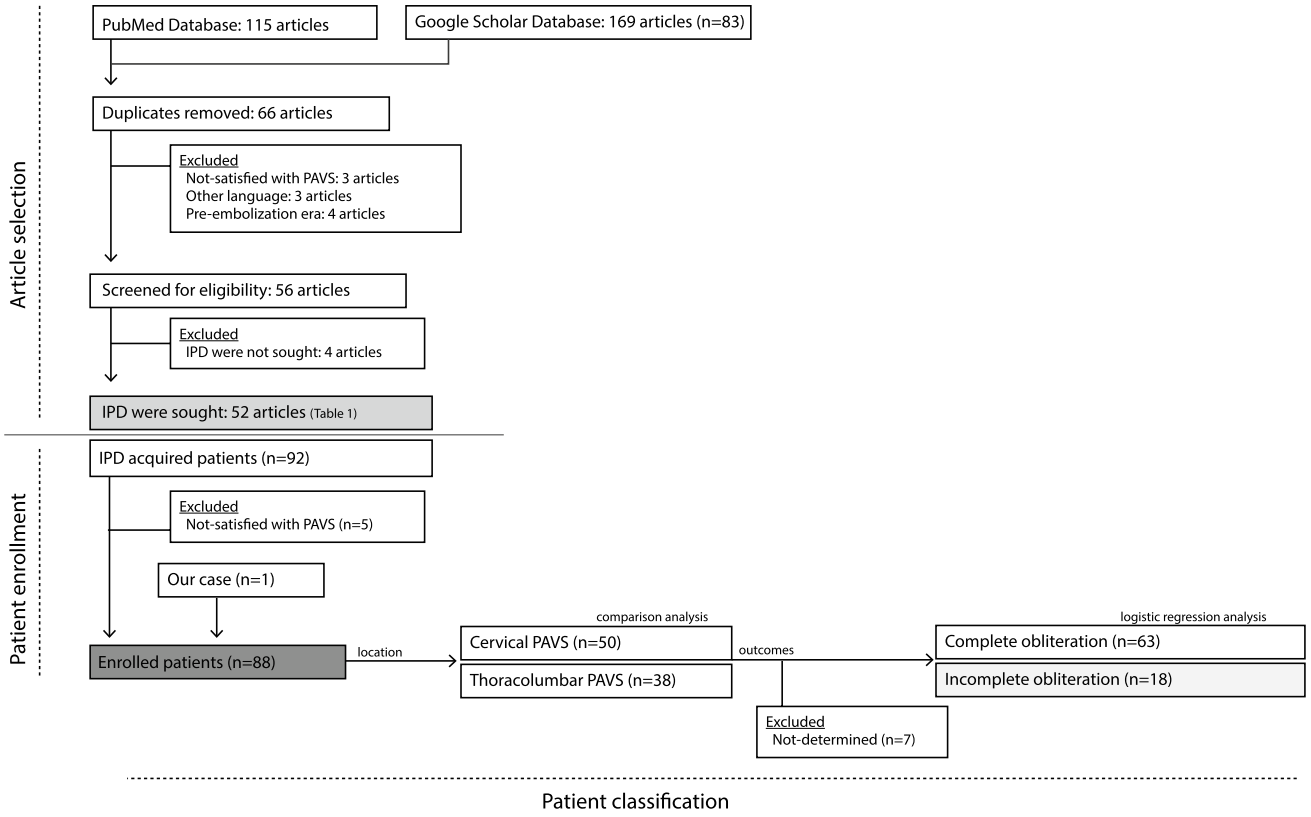


Fig. 3 Flowchart of the article selection, patient enrollment, and classification

be related to poor outcomes ($p=0.031$) [10, 11, 13, 14, 24]. Instead, traumatic vertebro-vertebral AVS usually occurred after implantation of internal jugular catheters and were observed near after the events. They were usually treated with simple embolization of fistulous points; therefore, the traumatic etiology was negatively related with poor outcomes ($p=0.038$).

(2) *Multiple feeders*: Even though verified only in the univariate analysis ($p=0.002$), the presence of multiple feeders increased the complexity of pathologic hemodynamics and potentially led to a higher chance of treatment failure. Generally, identifying the number of feeding arteries and planning the treatment based on hemodynamics is important to achieve higher treatment success [6]. However, fully understanding the hemodynamics is sometimes difficult, because PAVSs usually have a larger feeding artery diameter and affect larger lesions. Small feeding arteries or collateral vessels can be masked due to the major large flows with high pressure. Missing these small multiple feeders may lead to treatment failure, thus, preoperative catheterized angiography should be precisely performed and carefully interpreted.

Figure 4 illustrates the major feeding artery of enrolled PAVSs. Several cervical PAVSs originate from the branches of the subclavian artery and lumbosacral PAVS can be fed by branches of the internal iliac artery. These arteries should be super-selected in preoperative angiography for reducing the missing feeders.

(3) *Treatment modality*: Fig. 5 presents the flow of the clinical courses according to the treatment modality. Surgery previously had shown poor outcomes before the development of endovascular interventions, so endovascular embolization has become the accepted first-line treatment method [9, 47]. In corroboration with previous reports, surgery alone showed poor outcomes (5/6, 83.3%). However, endovascular interventions too showed relatively high rates of incomplete obliteration (13/66, 19.7%). Although "combined treatment" was only verified in univariate analysis ($p=0.03$), it would be recommended in complicated cases of PAVS to achieve higher success.

Limitations

Several limitations exist in the current study. First, the interpretation of the results cannot be generalized due to the wide time period between 1994 and 2018 of the published articles in which vast improvements occurred in endovascular

Table 2 The list and general demographics of the selected articles

Author	Year	No. (excl.)	Age	Sex	Associated factors	Major feeding artery (No. of patients)	Treatment	Outcomes
Hui F[47]	1994	3	0~10	M (1), F(2)	trauma (1), congenital (2)	Vertebral artery (1), T7 intercostal artery (1), Ascending cervical artery, posterior cervical artery (1)	TAE c coil, glue (3)	complete obliteration (3)
Cognard C[21]	1995	1	17	F	Trauma	Iliolumbar artery (main), lateral sacral artery	TAE c balloon	complete obliteration
Guglielmi G[37]	1995	1	66	M	Catheterization	Vertebral artery	TAE c coil	complete obliteration
Ricolfi E[56]	1995	5 (2)	20~77	M (1), F (4)	Catheterization	Vertebral artery (4), Deep cervical artery (1)	Surgery with intraoperative embolization (1), TAE c glue, balloon (4)	complete obliteration (5)
Kominami S[41]	1996	1	12	M	Trauma	Vertebral artery	TAE c coil	complete obliteration
Chen C[18]	1997	1	39	M	Acquired	T12 intercostal artery	TAE c glue	complete obliteration
Nagashima T[13]	1998	1	51	M	SAMS	T4-6 intercostal arteries	Surgery after embolization	complete obliteration: ambulatory without assist
Miralbes S[52]	1998	1	67	F	Catheterization	Vertebral artery	TAE c coil	complete obliteration
Szajner M[19]	1999	1	48	F	Klippel-Trenaunay syndrome	Thyrocervical trunks	TAE c glue (failed), TVE c glue	recurred AVS, finally complete obliteration
Ushikoshi S[31]	1999	1	51	F	NF1	Vertebral artery and multiple collaterals	TAE c glue, TVE c Coil	complete obliteration
Goyal M[39]	1999	10	1~73	M (7), F (3)	Acquired (4), Trauma (1), SAMS (1), NF(1), Congenital(3)	Lateral sacral artery (1), Vertebral artery (1), Thoracic intercostal and lumbar segmental arteries (7), Thoracic paravertebral artery (1)	Surgery (2), TAE (7), Not performed (1)	incomplete (3), complete obliteration (6), ND (1)
Pascual-Castroviejo I[42]	2002	1	7	M	Cobb syndrome	T5-6 intercostal arteries	Not performed d/t high-risk	ND
Kähärä V[40]	2003	1	43	F	Acquired	Internal iliac arteries	Embolization after failed surgery	incomplete obliteration: bladder dysfunction
Corr P[48]	2003	1	37	M	Acquired	Superior gluteal artery	TAE c balloon, glue	complete obliteration; mild weakness
Siddhartha W[57]	2003	1	36	F	NF1	Vertebral arteries, multiple collaterals	TAE c coil (partial)	complete obliteration; ambulatory with assist, residual weakness
Niimi Y[9]	2005	5	2~3	M	Congenital	Single thoracic intercostal artery (4), lumbar segmental artery (1)	TAE c coil, glue, balloon (5)	complete obliteration (5)
Tenjin H[29]	2005	1	72	F	Catheterization	Vertebral artery	TAE c coil	complete obliteration
Fotso A[12]	2006	1	3	M	Congenital	T11 intercostal artery	TAE c coil	complete obliteration
Akiyama Y[15]	2006	1	38	M	Lhemitte-Duclos syndrome	Vertebral artery	TAE	complete obliteration

Table 2 (continued)

Author	Year	No. (excl.)	Age	Sex	Associated factors	Major feeding artery (No. of patients)	Treatment	Outcomes
Vergouwen MDI[27]	2006	1	17	F	Catheterization	Vertebral artery	TAE c coil	complete obliteration
Hauck EF[30]	2006	1	51	F	NF1	Thyrocervical and costocervical trunks	Surgery with intraoperative embolization (hybrid)	complete obliteration; ambulatory without assist
Nakano S[54]	2006	1 (1)	0	ND	Congenital	Vertebral artery	TAE c glue	complete obliteration
Shirakawa M[35]	2008	1	58	M	Stab injury	Vertebral artery	TAE c coil	complete obliteration
Kitagawa RS[8]	2009	1	12	F	Acquired	T2 intercostal artery	TAE c Onyx	complete obliteration; ambulatory with assist
Patro SN[26]	2009	1	29	F	NF1	Vertebral artery and multiple collaterals	Embolization after failed surgery	complete obliteration; ambulatory with assist
Mortimer A[51]	2009	1	40	F	Trauma	Vertebral artery	TAE c coil	complete obliteration
Kalhorn SP[10]	2010	1	17	F	SAMS	T4-7 intercostal arteries	Surgery after embolization	complete obliteration
Fairhall JM[11]	2010	1	22	M	Acquired	T6-9 intercostal arteries	Surgery after embolization	complete obliteration
Núñez F[53]	2010	1	1	F	Congenital	Vertebral artery	TAE c balloon, glue	complete obliteration; remnant motor weakness, ataxic gait
Toi H[16]	2011	1	60	M	Acquired	Lateral sacral artery (main) and inferior gluteal artery	Surgery after failed embolization	complete obliteration; ambulatory with assist, bladder/sphincter dysfunction
Alomari AI[20]	2011	6	4 ~ 17	M (3), F (3)	CLOVES syndrome	Vertebral artery (1), Thoracolumbar paravertebral arteries (2), Thoracic intercostal and lumbar segmental arteries (3)	Surgery after embolization (3), multiple failed surgeries (1), not performed (2)	complete obliteration (3), recurred AVS (1), ND (2)
Santillan A[25]	2011	1	77	F	SAMS	T11-L1 intercostal and segmental arteries	TAE c coil, Onyx	recurred AVS
Spiotta AM[36]	2011	1	ND	ND	Cobb syndrome	Internal iliac artery (multiple branches)	Surgery after failed embolization	recurred AVS, finally complete obliteration
Wang Q[59]	2011	1	20	F	Acquired	Vertebral artery	TAE c coil, Onyx	complete obliteration
Teramoto S[17]	2012	1	41	M	Acquired	Right thyrocervical trunk	TAE c coil	complete obliteration
Iwakura T[23]	2012	1	93	F	Trauma	Descending aorta	Stent insertion	complete obliteration
Hughes DG[43]	2012	1	29	F	NF1	Vertebral artery, ECA, thyrocervical trunk	TAE c coil, glue (partial), TVE	recurred AVS: ambulation difficulty
Briganti F[34]	2013	3	34 ~ 61	M (1), F (1)	Trauma (2), NF1(1)	Vertebral artery (3)	Surgery after failed embolization (1), TAE c balloon, coil (2)	complete obliteration (2), recurred AVS (1)
Honarmand AR[49]	2013	1	8	M	Congenital	Vertebral artery, deep cervical artery	TAE c coil	complete obliteration
Walcott BP[55]	2013	1	69	M	Acquired	Vertebral artery	TAE c coil, Onyx	complete obliteration; mild cognitive deficit
Martínez-Galdámez M[22]	2014	1	60	F	Acquired	L3 segmental artery	TAE c Onyx	complete obliteration

Table 2 (continued)

Author	Year	No. (excl.)	Age	Sex	Associated factors	Major feeding artery (No. of patients)	Treatment	Outcomes
Komiyama M[24]	2014	1	0	F	SAMS	Vertebral artery (main), deep cervical artery, ascending pharyngeal artery	TAE c coil	recurred AVS
Li F[28]	2014	1	40	F	Stab injury	Vertebral artery	Stent insertion	complete obliteration
Yeh C[50]	2014	5	42 ~ 52	M (3), F (2)	Acquired (2), Trauma (1)	Vertebral artery (5)	TAE c coil, glue (2), Stent insertion (3)	complete obliteration (4), recurred AVS (1)
Elkordy A[14]	2015	1	15	M	SAMS	T5-8 intercostal arteries	Surgery after embolization	complete obliteration
Farhat N[45]	2015	2	1	M	Congenital	Descending aorta (1), vertebral artery (1)	Not performed d/t young age	ND (2)
Narayana RV[46]	2015	1	42	M	NF1	Vertebral artery, multiple cervical arteries	TAE (failed), TAE	unsuccessfully embolized, remnant weakness
Guneyli S[33]	2016	4 (2)	4 ~ 17	M (2), F (2)	Acquired (2), Congenital (2)	Vertebral artery (4)	TAE c balloon, coil	unsuccessfully embolized (1), successfully treated (3)
Ashour R[38]	2016	2	4, 8	M (1), F (1)	Congenital	Vertebral artery (2)	TAE c coil	complete obliteration (2)
Tse GH[44]	2017	1	18	F	Acquired	Vertebral artery	TAE c coil	complete obliteration
Mitchell P[32]	2018	1	10	M	SAMS	Vertebral artery, subclavian and carotid collaterals	Not performed	ND
Breda MS[58]	2018	1	36	M	Acquired	Vertebral artery, ascending cervical artery	TAE c coil	complete obliteration
Our case	2020	1	21	M	Acquired	L4 segmental artery, L3, internal iliac arteries	Surgery first	incomplete obliteration

No, number; excl., excluded; FMD, fibromuscular dysplasia; AVS, arteriovenous fistula; NA, not assessed; TVE, trans-venous embolization; TAE, trans-arterial embolization; ND, not determined; NF1, neurofibromatosis type 1; SAMS, spinal arteriovenous metameric syndrome; CLOVES, congenital-lipomatous-overgrowth-vascular malformation-epidermal nevi-and spinal/skeletal anomalies or scoliosis; ECA, external carotid artery

Table 3 The general demographics of pooled data and the results of statistical analyses comparing cervical to thoracolumbar PAVS

Parameters	Total ($n=88$)	Cervical ($n=50$)	Thoracolumbar ($n=38$)	P value
General				
Age (year-old) [a]	29.14 ± 24.413	32.90 ± 22.946	24.07 ± 25.710	0.095
Female [b]	42 (47.7%)	29 (59.1%)	13 (35.1%)	**0.027***
Stuttgart classification [c]				
Acquired	20 (22.7%)	8 (16.0%)	12 (31.6%)	**0.001***
Traumatic	22 (25.0%)	20 (40.0%)	2 (5.3%)	
Congenital	22 (25.0%)	11 (22.0%)	11 (28.9%)	
Metameric link	6 (6.8%)	1 (2.0%)	5 (13.2%)	
Systematic genetic dysplasia	18 (20.5%)	10 (20.0%)	8 (21.1%)	
Symptom or sign				
Bruit, thrill, or murmur [b]	36 (40.9%)	27 (54.0%)	9 (23.7%)	**0.004***
Cranial symptom or sign[1, c]	9 (10.2%)	9 (18.0%)	0 (0%)	**0.001***
Weakness [b]	38 (40.2%)	17 (34.0%)	21 (55.3%)	**0.046***
Deformity [c]	5 (5.7%)	0 (0%)	5 (13.2%)	**0.003***
Bowel or bladder dysfunction [c]	10 (11.4%)	2 (4.0%)	8 (21.1%)	**0.011***
Mass, nevi, skin lesions [b]	14 (15.9%)	6 (12.0%)	8 (21.1%)	0.250
Non-specific symptoms [2, c]	6 (6.8%)	1 (2.0%)	5 (13.2%)	**0.036***
Imaging feature				
Hematoma, hemorrhage [c]	10 (11.4%)	6 (12.0%)	4 (10.5%)	0.829
Multiple feeders [b]	38 (40.2%)	15 (30.0%)	23 (60.5%)	**0.004***
Intradural venous involvement [b]	19 (21.6%)	8 (16.0%)	11 (28.9%)	0.144
Treatment [c]				
Surgery	6 (6.8%)	1 (2.0%)	5 (13.2%)	**0.011***
Endovascular	66 (75.0%)	44 (89.0%)	22 (57.9%)	
Combined	9 (10.2%)	3 (6.0%)	6 (15.8%)	
Observed	7 (8.0%)	2 (4.0%)	5 (13.2%)	
Follow-up (month) [a]	15.36 ± 26.06	13.37 ± 20.85	18.28 ± 32.43	0.447
Outcome [c]				
Complete obliteration w/o N/D	54 (61.4%)	36 (72.0%)	18 (47.4%)	0.078
Complete obliteration w/ N/D	9 (10.2%)	3 (6.0%)	6 (15.8%)	
Incomplete obliteration	18 (20.5%)	9 (18.0%)	9 (23.7%)	
Not determined	7 (8.0%)	2 (4.0%)	5 (13.2%)	

*P value < 0.05

[1]Cranial symptom/sign includes tinnitus, cranial nerve palsy, and subarachnoid hemorrhage in cranium

[2]Non-specific symptoms include dysesthesia or pain along spinal axis

[a]Unpaired t test, [b]Chi-square test, and [c]Fisher's exact test

treatment. The clinical decisions, interventional techniques, and outcomes were thus not the same between the different time points. Second, as a limitation of meta-analysis itself, there might be several missing cases, which may lead to selection bias. In the current study, we tried to enroll all cases of PAVSs in the literature; however, the term "paravertebral or paraspinal" was only recently defined and a certain portion of PAVSs may have been missed due to being previously defined as "epidural or extradural" AVSs.

Table 4 The results of *univariate logistic regression analysis* for identifying factors associated with incomplete obliteration

Parameters	Complete obliteration ($n=63$)	Incomplete obliteration ($n=18$)	Univariate logistic analysis		
			Hazard ratio	95% Confidential index	*P* value
General					
Age (years)	30.0 ± 25.06	33.4 ± 23.39	1.007	0.986–1.029	0.513
Female	31 (50.0%)	10 (58.8%)	1.429	0.482–4.235	0.520
Stuttgart classification					
Acquired	15 (23.8%)	4 (22.2%)	0.233	0.052–1.044	0.057†
Traumatic	21 (33.3%)	1 (5.6%)	0.042	0.004–0.395	**0.006***
Congenital	17 (27.0%)	2 (11.1%)	0.103	0.017–0.612	**0.012***
Metameric link	3 (4.8%)	3 (16.7%)	0.875	0.132–5.819	0.890
Systematic genetic dysplasia	7 (11.1%)	8 (44.4%)	1.000		**0.012***
Symptom or signs					
Bruit, thrill, and murmur	29 (46.0%)	4 (22.2%)	0.335	0.99–1.131	0.078†
Cranial	7 (11.1%)	1 (5.6%)	0.471	0.054–4.099	0.495
Weakness	25 (39.7%)	11 (61.1%)	2.389	0.816–6.989	0.112
Deformity	2 (3.2%)	1 (5.6%)	1.794	0.153–20.988	0.641
Bowel/bladder	6 (9.5%)	3 (16.7%)	1.900	0.425–8.499	0.401
Mass/nevi	7 (11.1%)	4 (22.2%)	2.286	0.586–8.914	0.234
Non-specific	3 (4.8%)	2 (11.1%)	2.500	0.384–16.257	0.337
Location					
Cervical	39 (61.9%)	9 (50.0%)	1.000		0.460
Thoracic	17 (27.0%)	5 (27.8%)	0.404	0.097–1.681	0.213
Lumbosacral	7 (11.1%)	4 (22.2%)	0.515	0.106–2.504	0.411
Radiologic features					
Multiple feeders	18 (28.6%)	13 (72.2%)	6.500	2.023–20.886	**0.002***
Intradural venous involvement	11 (17.5%)	5 (27.8%)	1.818	0.537–6.155	0.337
Treatment					
Surgery	1 (1.6%)	5 (27.8%)	8.077E + 9		0.999
Endovascular	53 (84.1%)	13 (72.2%)	396,248,552		0.999
Combined	9 (14.3%)	0 (0%)	1.000		**0.030***

***P** value < 0.05, †*P* value < 0.1

Table 5 The results of *multivariate logistic regression analyses* for identifying factors associated with incomplete obliteration

Parameters	B	S.E	Wald	Multivariate logistic analysis			
				Hazard ratio	(95% Confidential index)		*P*value
Stuttgart classification							
Acquired			7.191				0.126
Traumatic	− 2.782	1.340	4.309	0.062	0.004	0.856	**0.038***
Congenital	− 2.167	1.104	3.855	0.115	0.013	0.996	0.050
Metameric link	19.429	10,879.178	0.000	274,204,256.1	0.000		0.999
Systematic genetic dysplasia	2.790	1.297	4.628	0.061	0.005	0.780	**0.031***
Symptom and signs							
Bruit, thrill, murmur	− 0.793	0.989	0.643	0.453	0.065	3.143	0.423
Radiologic finding							
Multiple feeders	1.612	0.876	3.384	5.015	0.900	27.947	**0.066**†
Treatment							
Surgery	44.014	15,444.120	0.000	1.30267E + 19	0.000		0.998
Endovascular	40.240	15,444.120	0.000	2.99303E + 17	0.000		0.998
Combined			5.462				**0.065**†

***P** value < 0.05

†*P* value < 0.1

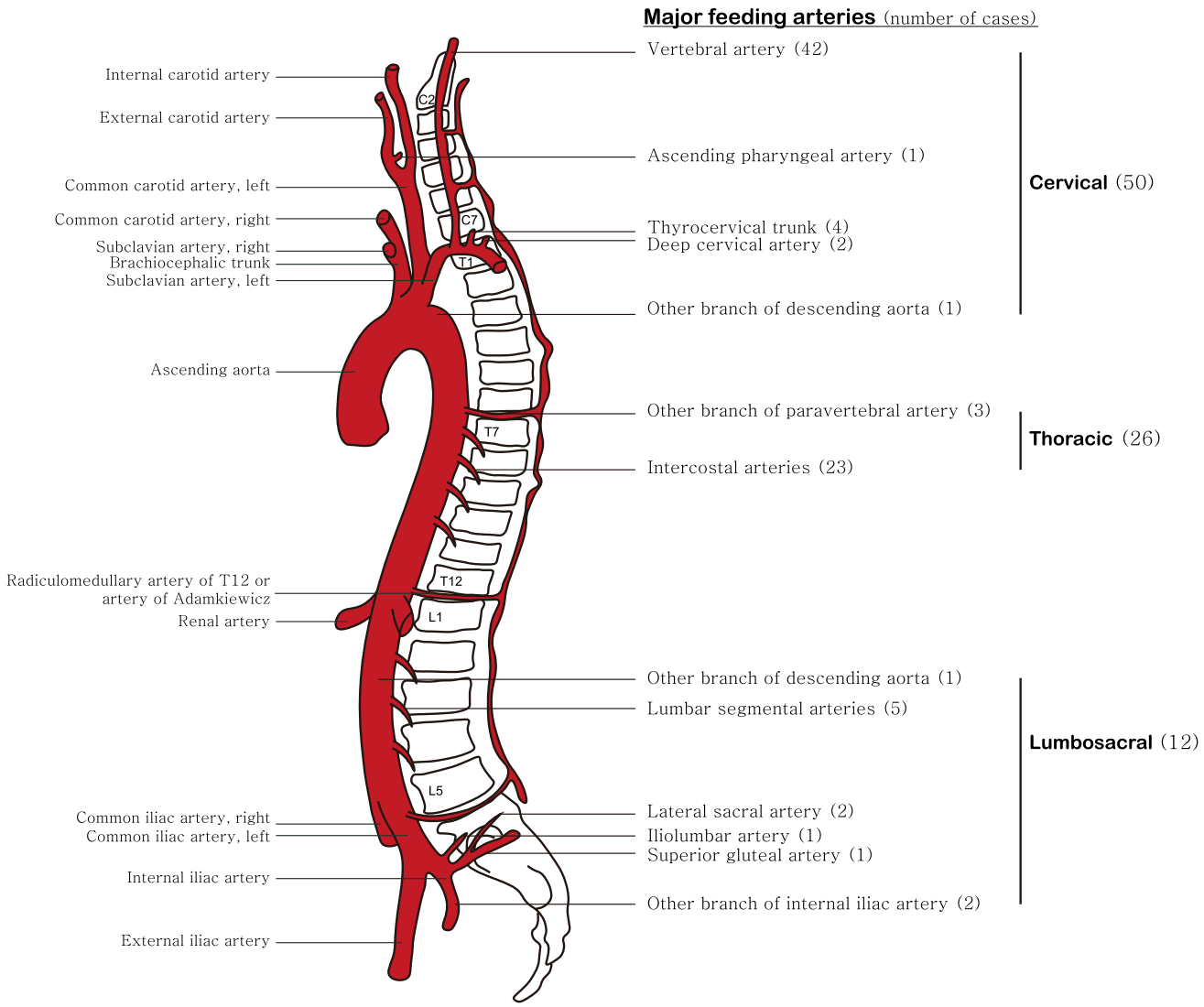


Fig. 4 A schematic illustration of the major feeding artery of the enrolled PAVSs

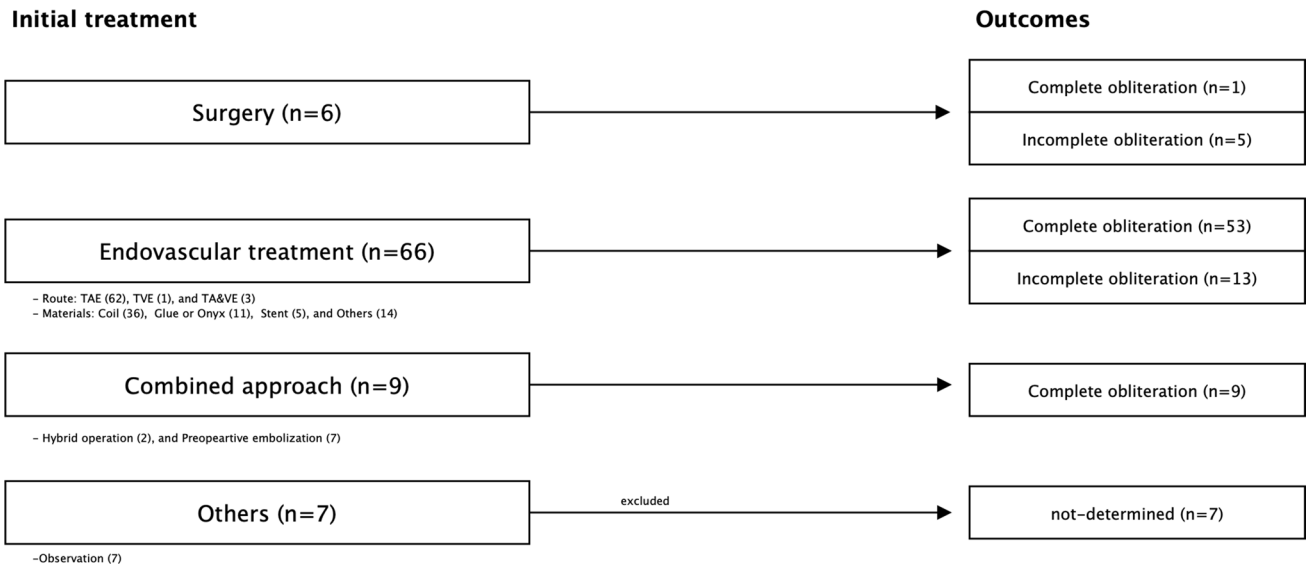


Fig. 5 Flowchart of the outcomes according to treatment modalities

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