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Intracranial subarachnoid hemorrhage resulting from non-cervical spinal arteriovenous lesions: Analysis of possible cause of bleeding and literature review

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ABSTRACT

Subarachnoid hemorrhage (SAH) or intraventricle hemorrhage (IVH) with negative cerebral digital subtraction angiography (DSA) results, which are due to non-cervical spinal arteriovenous lesions, are uncommon. In this article we presented three cases from our hospital and nineteen cases from prior published literature and discussed clinical features, possible mechanisms underlying the hemorrhage and therapeutic strategies for managing this unusual entity. Our analysis revealed that headache was the most common initial symptom. Almost 60% of patients had symptoms related to the spinal cord at admission. Intramedullary arteriovenous malformations (AVM) were the most common type of malformation, and the thoracic segment was the most common location of the non-cervical spinal arteriovenous lesions. More than half of the patients had additional aneurysms. Surgery was chosen as the primary treatment modality in this series. Therefore, we speculate that thoracolumbar spinal arteriovenous lesions are an unusual cause of intracranial SAH with negative cerebral DSA results. If non-cervical spinal AVMs were associated with DSA-negative SAH, the pattern of hemorrhage could be manifested as the blood in supratentorial cisterns, the fourth ventricle or no copious blood around the foramen magnum as well (somewhat paradoxically), it depends on the timing of detection and image evaluation. The formation and the rupture of associated aneurysms were the most likely immediate cause of the intracranial SAH. If non-cervical spinal AVMs were not associated with DSA-negative SAH and all cases were genuine cases of 'SAH-of-unknown origin', the spinal AVM could be considered as incidental finding. Magnetic resonance imaging (MRI) of the complete spinal neuraxis is recommended to either exclude or identify a spinal lesion in these patients. Catheter-based spinal angiography remains the gold standard for the diagnosis of spinal vascular diseases. The decision regarding a therapeutic strategy is based on the angioarchitecture and on the type of spinal arteriovenous lesions.

1. Introduction

Although spontaneous subarachnoid hemorrhage (SAH) is commonly caused by ruptured cerebral aneurysms [1,2], it can also result from a variety of rare conditions in about 5%–10% of patients. Approximately 1% of cases of intracranial SAH are related to spinal pathologies, such as arteriovenous lesions [3], spinal artery aneurysms [4,5] and hemorrhagic and metastatic tumors [6]. Most spinal arteriovenous lesions resulting in intracranial SAH are located at the craniocervical junction or in the cervical region [7–11] and are rarely observed in other segments of the spinal cord, even in the conus medullary [11,12]. Such a location would appear to be too far away to result in an intracranial SAH, and the mechanism of SAH induced by non-cervical spinal vascular disease remains unclear. Spinal arteriovenous lesions usually present with diffuse back pain and progressive myelopathy due to venous hypertension. However, acute hemorrhage is also a mode of presentation in approximately 10% of patients. Here, we report on 3 patients treated in our hospital who presenting with intracranial SAH and subsequent negative cerebral digital subtraction angiography (DSA) results, but were diagnosed with thoracolumbar spinal arteriovenous fistula (AVF) with associated aneurysms according to the spinal DSA. In addition, the clinical features, possible mechanism of the hemorrhage and therapeutic strategy are discussed based on the data from our cases and on a review of the literature.

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2. Methods

Screening of our in-hospital database was performed to identify patients with non-cervical spinal arteriovenous lesions presenting as intracranial SAH. Our inclusion criteria in this study are the following: (1) patients presented with visible intracranial SAH confirmed by initial computed tomography (CT) scan and negative cerebral DSA results; (2) patients are confirmed as having spinal arteriovenous lesions by DSA and surgical findings; (3) lesions located at a non-craniocervical junction and the non- cervical region.

For a literature review, we developed search strategies and extracted data according to the PRISMA guidelines. Two independent investigators searched the PubMed database by using the search terms “subarachnoid hemorrhage”, “negative”, “spinal arteriovenous malformation”, and “spinal arteriovenous fistula”. We defined following inclusion and exclusion criteria.: inclusion criteria: (1) the research object was a human being; (2) the language of the article was English; (3) other inclusion criteria as stated in the case series. Exclusion criteria: (1) the publication lacked full-text documents, such as abstracts, letters to the editor, and comments; (2) a large amount of valuable information was missing from the article. (3) patients with SAH was confirmed by CSF test from lumber puncture but initial CT scan was normal. All studies were also reviewed by two investigators to decide whether they should be included. A chart of the retrieval process is presented in Supplemental Fig. 1.

Two independent researchers collected the following data: first author, year of publication, age, sex, pattern of intracranial SAH, symptoms related to the spinal cord, type and location of the spinal AVM, presence of an associated spinal aneurysm, treatment modality and outcome. Any discrepancy between the two investigators was resolved by discussion to reach a consensus.

3. Results

3.1. Case series

Between 1 July 2010 and 1 July 2018, three cases were identified from our hospital database. We retrospectively analysed the clinical data of each patient, including sex, age, presentation, CT scan, SAH location, spinal DSA results, treatment modality and final outcomes. Outcomes were assessed using the modified Rankin Scale (mRS) score system at the most recent follow-up (cases 1, 2 and 3).All the image data are shown on the supplemental materials.

3.1.1. Case 1

A 31-year-old man who suffered a thunderclap headache, nausea and vomiting was admitted to a local hospital. A brain CT scan (14 Mar 2011) showed diffuse SAH in the supratentorial cistern and the fourth ventricle, including the carotid cistern and bilateral sylvian cistern. The pan-cerebral DSA was negative for aneurysms and other vascular lesions (17 Mar 2011). After a month of conservative treatment, the patient was transferred to our hospital. He received the second cerebral

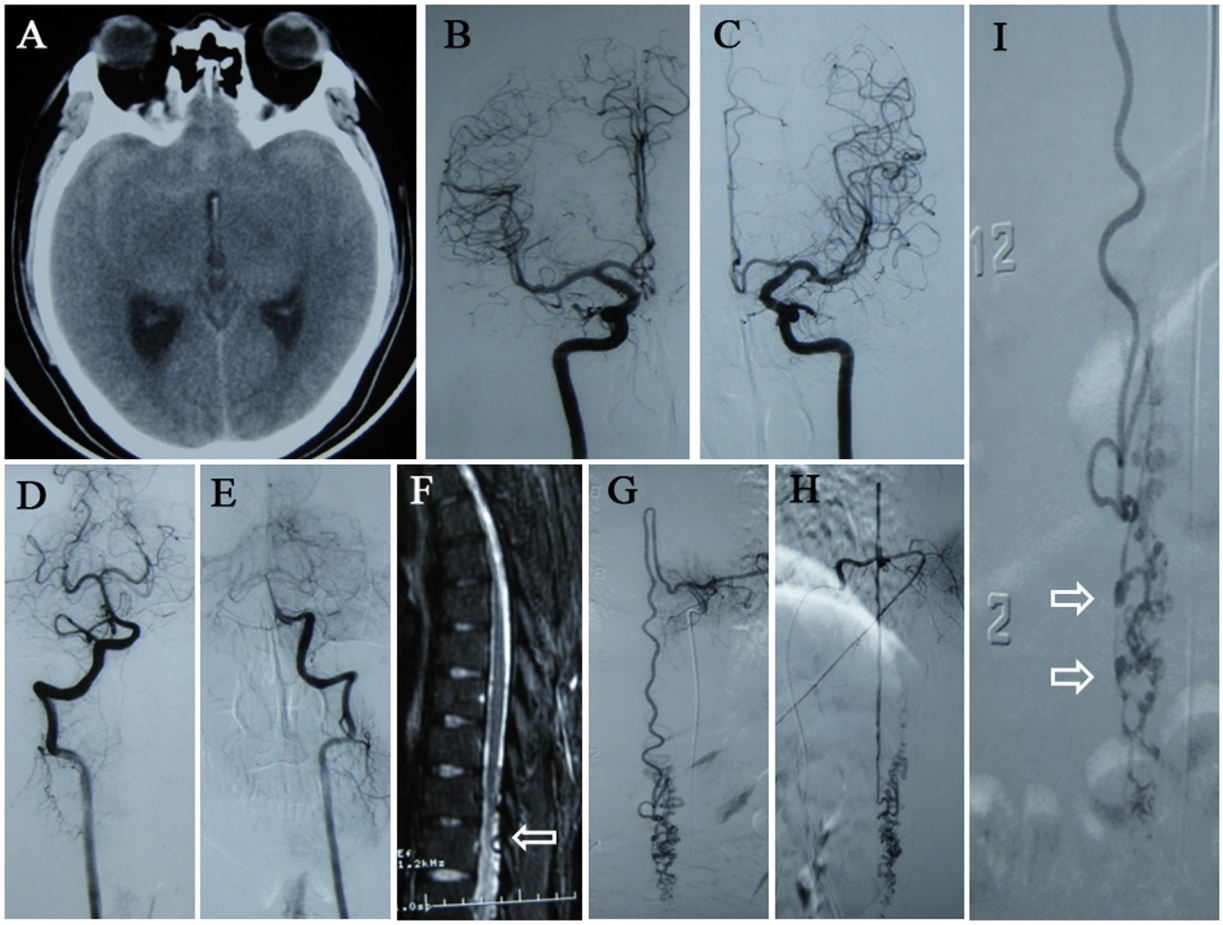

Fig. 1. Case 1. A: On admission to the local hospital, axial CT images of the brain showed diffuse SAH in the supratentorial cistern. B-E: Cerebral DSA from the right internal carotid artery (B), left internal carotid artery (C), right vertebral artery (D) and left vertebral artery (E) were negative for aneurysms and other vascular lesions. F: T2-weighted MRI image showing perimedullary vascular flow void signal *(Arrowhead)* on the dorsal aspect of the T10-L1 spinal cord. G-H: Spinal DSA of the anteroposterior (G) and lateral projection (H) showed intradural ventral AVF at the L1 level fed by the anterior spinal artery from selective injection of the left T9 intercostal artery. I: Spinal DSA of the anteroposterior projection of the enlarged image shows two aneurysms on the draining vessels *(Arrowhead)*.

DSA (2 May 2011) 45 days after the first hemorrhage, the result was still negative. Magnetic resonance imaging (MRI) of the thoracic and lumbar segment of the spine revealed a flow void signal in the T2-weighted image (4 May 2011). Spinal DSA (9 May 2011) showed an intradural ventral AVF at the L1 level fed by the anterior spinal artery with two aneurysms on the draining vessels (***Arrowhead,*** Fig. 1I) from selective injection of the left T9 intercostal artery (Fig. 1A-I). The patient had a successful T12-L2 laminectomy with incision of the fistula, although the first embolization attempt failed. He suffered decreased muscle strength in the left extremity after the operation, however the symptoms resolved completely during the stay in the hospital.

3.1.2. Case 2

A 47-year-old man was admitted to our hospital in Oct 2014 with lumbar back pain and numbness of the lower extremity for 2 months. He suffered from an SAH and intraventricular hemorrhage (IVH) in 28 Sep 2013, with negative computed tomography angiography (CTA) and DSA results. Cervical MRI did not identify any source of bleeding at that time (8 Oct 2013). Neurologic examination was unremarkable except for level 4 of right lower extremity strength. MRI of the thoracic and lumbar region of the spine (9 Oct 2014) revealed a flow void signal in T2-weighted images and hyperintensities at the centre of the spinal cord in the lower thoracic segment. Spinal DSA (20 Oct 2014)showed an intradural ventral AVF at the T10 level fed by the posterior spinal artery and an aneurysm near the fistula ***(Arrowhead,*** Fig. 2F***)*** from selective injection of the right T10 intercostal artery **(**Fig. 2**A-F)**. The patient had a successful T9-T12 laminectomy with incision of the fistula. He suffered transiently decreased muscle strength in the right lower extremity, but showed complete recovery within one month of rehabilitation.

3.1.3. Case 3

A 31-year-old man who suffered sudden onset of a headache and neck pain was transferred to our hospital. A previous CT scan performed by a local hospital showed a diffuse SAH in the supratentorial cistern, interpeduncular cistern and the fourth ventricle (12 Mar 2017). The pan-cerebral DSA and CTA were negative for aneurysms and other vascular lesions (16 Mar 2017, See supplemental material). MRI of the cervical and thoracic segment of the spine (20 Mar 2017) revealed multiple tortuous and vascular flow void signals in T2-weighted images and extensive oedema in the cervical and upper thoracic parenchyma of the spinal cord. Magnetic resonance angiography (MRA) showed an abnormal and tortuous vascular structure in the spinal canal. Spinal DSA (27 Mar 2017) showed an intradural ventral AVF at the T6 level fed by the anterior spinal artery with multiply aneurysms inside the lesion from selective injection of the right T5 intercostal artery, (***Arrowhead,*** Fig. 3E). He had a successful T4-T6 laminectomy, and the fistula was cut off with three clips (AESCULP 740,710,720) with the aid of spinal intraoperative DSA (IO-DSA) in a hybrid operating room. Postoperative MRI (17 Apr 2017) did not show any vascular flow void signal in T2-weighted images or on MRA. He suffered transiently decreased muscle strength in the right extremity, but was recovered at discharge. Postoperative spinal DSA (4 Jun 2017) revealed no aneurysms or abnormal vessels at the 3-month follow-up **(**Fig. 3**A-I)**.

3.2. Review of the literature

Our analysis revealed nineteen cases of non-cervical spinal vascular disease presenting with intracranial SAH or IVH, published in sixteen papers. Data from our three case series and the nineteen cases identified from the literature research were pooled together prior to data analysis. The details of the collected data are listed in Supplemental Table 1. All 22 patients presenting with SAH or IVH were confirmed by CT scan. Out of all 22 cases, 15 patients were male, and 7 patients were female. Headache was the most common initial symptom in the series (13/22). On admission, 13 patients had symptoms related to the spinal cord, such as back pain, radicular pain or motor and sensory deficits of the lower extremity. 19 cases were negative for cerebral DSA but positive for spinal MRI or DSA. Intramedullary AVMs were identified in 11 patients, intradural ventral AVFs in 8 patients, and intradural dorsal AVFs in 3 patients.The presence of aneurysm was found in 12 patients. In 10 patients, lesions of the spinal cord occurred in the thoracic segment, in 5 patients in the thoracolumbar segment, in 3 patients in the lumbar segment and in 4 patients in the conus medullaris. Regarding the mode of primary treatment, surgery was performed in 10 patients, embolization in 8 patients, and both procedures in 2 patients; 2 patients did not undergo any surgical or interventional treatment. A good recovery was achieved in 15 patients, only 1 patient died. A summary of the characteristics is listed in Table 1.

4. Discussion

We report on three patients treated in our hospital who presenting with intracranial SAH and negative cerebral DSA results, but were diagnosed with thoracolumbar spinal AVF associated with aneurysms according to the spinal DSA and the surgical findings. Moreover, our study also presents several patients described in the literature with non-cervical spinal vascular disease presenting with intracranial SAH or IVH. Most spinal arteriovenous lesions resulting in intracranial SAH are located at the craniocervical junction or in the cervical region but are rarely observed in the other segments of the spinal cord, which appear to be located too far away to result in an intracranial SAH, even in the conus medullary. The clinical features, the possible mechanism underlying the hemorrhage and the therapeutic strategy for managing this non-cervical spinal vascular disease remain unclear.

4.1. Significance of a spinal neuraxis MRI screening in patients with intracranial SAH with negative cerebral angiographic results

Intracranial SAH with negative cerebral angiographic imaging at the first DSA screening requires a pragmatic approach to detect the possible source of bleeding. The possibility of spinal SAH should at least be considered. In a prospective observational study of 75 cases of spontaneous non-aneurysmal, non-perimesencephalic SAH and absence of clinical / neurological findings MRI of the complete spinal axis showed a very low diagnostic yield in detection of a spinal atiology and is therefore not recommended [13]. More recent studies have recommended the use of only a cervical-MRI in selected cases if signs and symptoms raise the suspicion of spinal pathology, or patients present with a second clinical episode [14]. However, in clinical situations, initial symptoms, such as neck or back pain and transient lower-extremity weakness, are usually overshadowed by the ensuing severe headache or loss of consciousness. The clinical findings and information that suggest a spinal aetiology are often overlooked by a careless physical examination. Frankly spoken, a minor trace of spinal SAH is difficult to demonstrate on spinal MRI several days after the onset, and when you come to realize the spinal origin of the hemorrhage. Kocak et al. reported a patient with an intracranial SAH due to a spinal aneurysm who died of a rebleed within 24 h of onset, even though surgery had been planned, and emphasized that an accurate diagnosis and timely treatment to prevent rebleeding is extremely critical [15]. As the likelihood of finding a spinal etiology is very low in this population, the cost of a spinal MRI of the neuroaxis needs to be considered. Nonetheless, it is worth considering that a few patients would either become severely disabled or incur death due to the missed or delayed diagnosis. Therefore, while on the one hand, the clinical history and the information derived from the physical examination should be studied with care to identify a possible spinal cause of SAH; on the other hand, it is necessary to identify or exclude spinal aetiologies of intracranial SAH to prevent recurrent hemorrhage that may have catastrophic outcomes [16].

In our hospital, we strongly recommend that cerebral angiograms

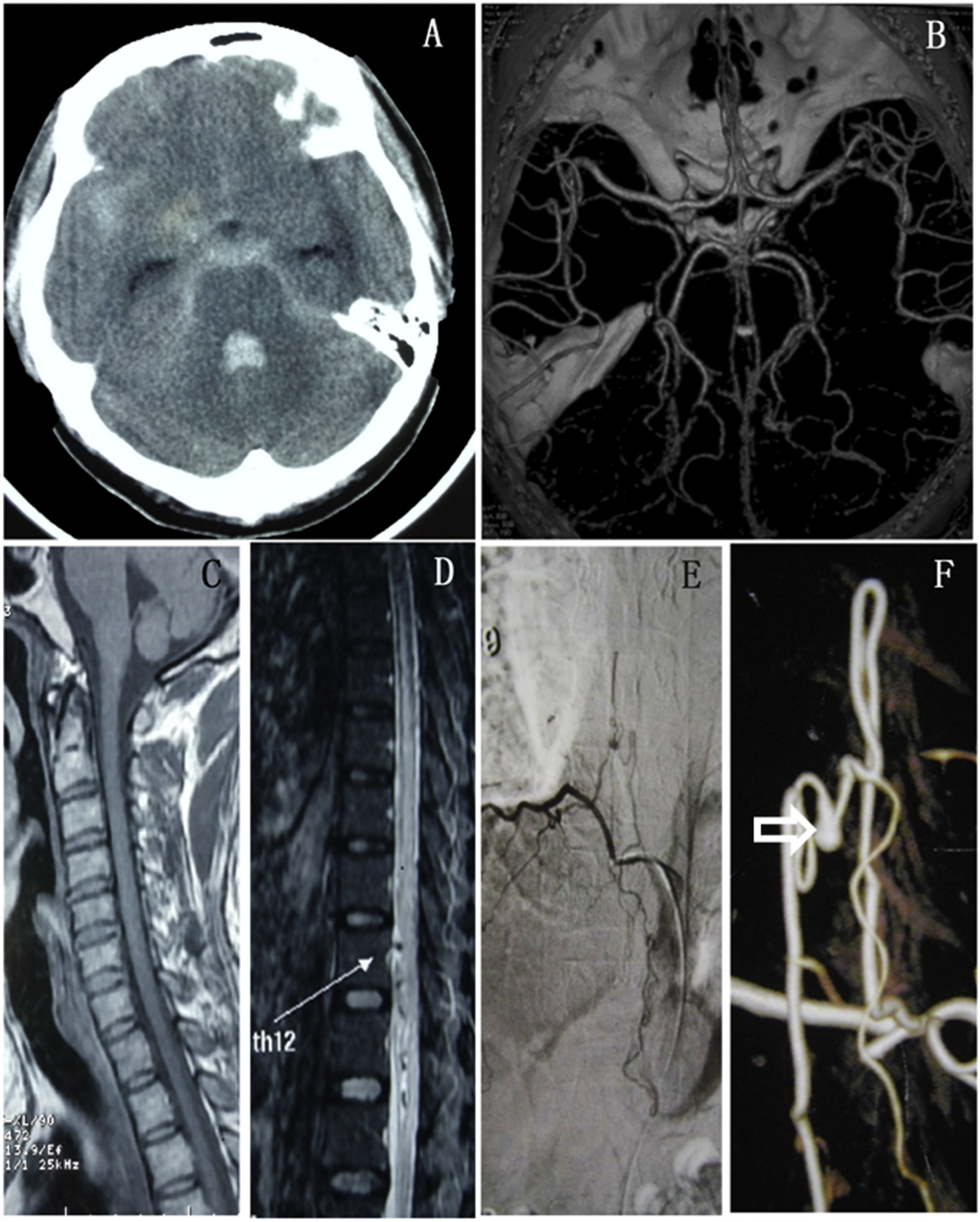

Fig. 2. Case 2. A: Axial brain CT image showed bleeding in the prepontine cistern, ambient cistern and in the fourth ventricle at the first onset. B: CTA was negative for aneurysm and other sources of vascular lesions. C: T1-weighted MRI of the cervical segment did not demonstrate any source of bleeding. D: MRI of the thoracic and lumbar region of the spine revealed a flow void signal in the T2-weighted image, and hyperintensities in the centre of the spinal cord in the lower thoracic segment. E: Spinal DSA of anteroposterior projection showed intradural ventral AVF at the T10 level fed by the posterior spinal artery from selective injection of the right T10 intercostal artery. F: Three-dimensional rotation of the spinal DSA showed an aneurysm near the fistula *(Arrowhead)*.

need to be repeated in patients with a non-perimesencephalic SAH pattern within 4 weeks if the initial DSA of the cerebral vessels did not reveal any intracranial source of spontaneous SAH [17,18]. During the interval until the second DSA, MRI of the whole spinal neuraxis, not a cervical-MRI alone, should be performed to exclude or identify a spinal lesion [19].

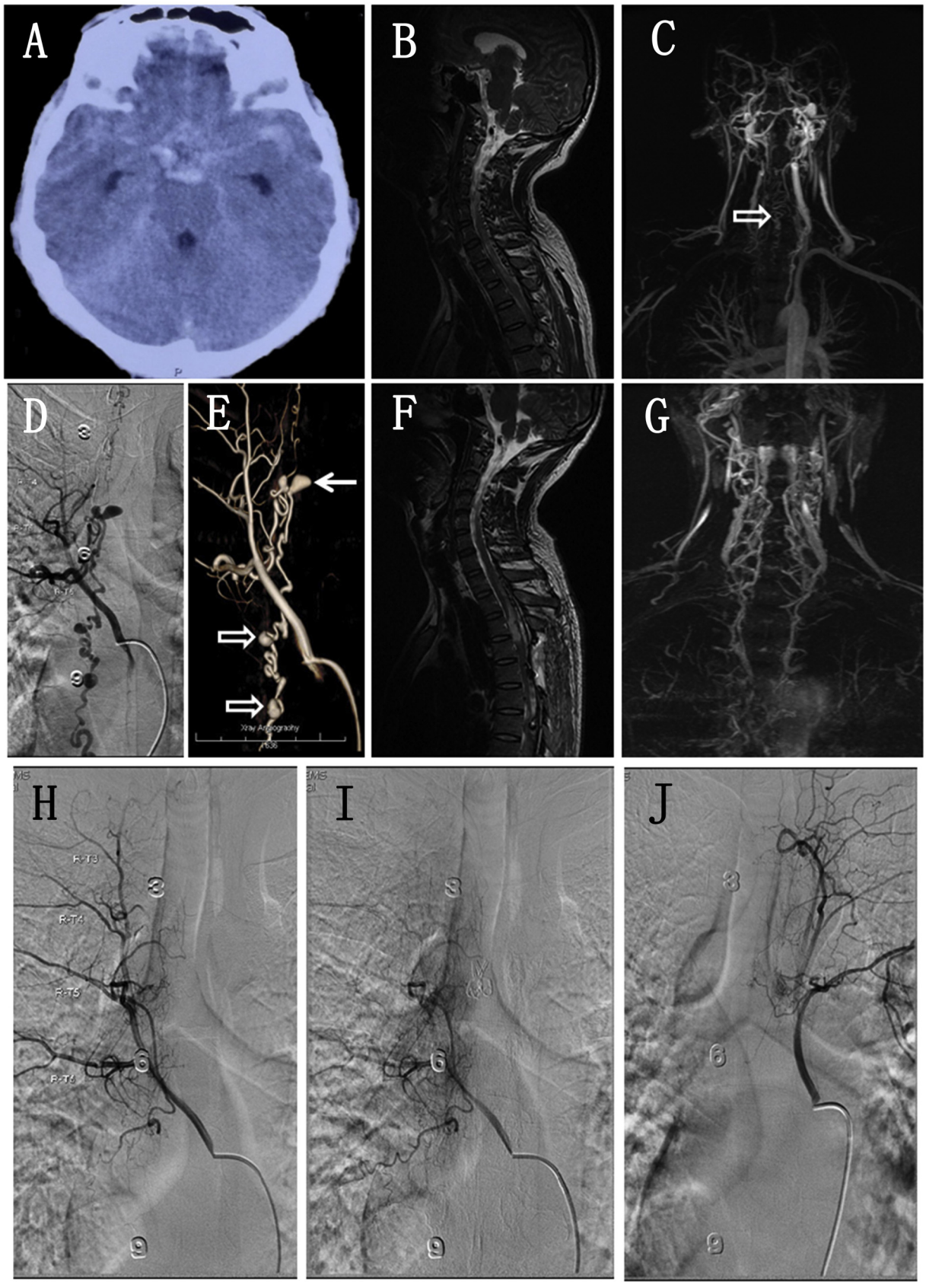

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Fig. 3. Case 3. A: Axial brain CT image showing diffuse SAH in the carotid cistern, interpeduncular cistern and ambient cistern. B: T2-weighted MRI of the cervical and thoracic segments revealed a flow void signal on the dorsal aspect of the spinal cord and hyperintensities in the centre of the spinal cord in the upper thoracic segment of the spinal cord. C: Spinal MRA shows an abnormal and tortuous vascular structure in the spinal canal *(Arrowhead)*. D: Spinal DSA of anteroposterior projection showed intradural ventral AVF at the T6 level fed by the anterior spinal artery from selective injection of the right T5 intercostal artery. E: Three-dimensional rotation of spinal DSA shows two aneurysms near the fistula *(Arrow)* and multiple aneurysms *(Arrowhead)* on the draining vessels. F: Postoperative T2-weighted MRI of the cervical and thoracic segments did not show any flow void signal on the dorsal aspect of the spinal cord. G: Postoperative spinal MRA did not show the presence of an original abnormal vascular structure. H-I: Postoperative spinal DSA of the anteroposterior projection from selective injection of the right and left T5 intercostal artery revealed no aneurysm and abnormal vessel.

Table 1
Summary of 22 cases of non-cervical spinal arteriovenous lesion presenting intracranial subarachnoid hemorrhage or intra-ventricular hemorrhage.

	Data statistics
Age (years)	25.27 ± 18.5(y)
Sex	
Male	68.2%(15/22)
Female	31.8%(7/22)
Clinical manifestation	
Headache	59.1%(13/22)
Loss of consciousness	18.2%(4/22)
Symptoms related to the spinal cord	59.1%(13/22)
Initial CT scan	
SAH	50%(11/22)
IVH	4.5%(1/22)
SAH + IVH	45.5%(10/22)
Type of spinal arteriovenous lesion	
AVM	50%(11/22)
Ventral AVF	36.4%(8/22)
Dorsal AVF	13.6%(3/22)
Lesion location	
Thoracic segment	45.5%(10/22)
Thoracolumbar segment	22.7%(5/22)
Lumbar segment	13.6%(3/22)
Conus medullary	18.2%(4/22)
Presence of aneurysm	
Yes	54.5%(12/22)
No	22.7%(5/22)
Not described	22.7%(5/22)
Treatment modality	
Surgery only	45.5%(10/22)
Embolization only	36.4%(8/22)
Surgery + Embolization	9.1%(2/22)
Not treated	9.1%(2/22)

4.2. Mechanism underlying the intracranial SAH resulting from non-cervical spinal arteriovenous lesions

In this study, a modified classification system of spinal AVM, which is based on the specific anatomical and pathophysiological factors proposed by Spetzler et al., was used. In this system, spinal AVMs are classified into 4 groups: extradural AVF, intradural dorsal AVFintradural ventral AVF, extradural-intradural AVM and intramedullary AVM (which can be characterized as compact or diffuse depending on the angioarchitecture of the nidus) and conus medullaris AVM [20]. Intradural ventral AVFs, also known as perimedullary AVFs, were first described in 1977 by Djindjian et al. [21] and are estimated to comprise 20% of spinal AVMs. Intradural ventral AVFs are located on the pial surface of the spinal cord and are characterized as arteriovenous shunts without a nidus of abnormal vessels between the feeding artery and the draining vein. They are fed by the anterior spinal artery (ASA) and/or the posterior spinal artery (PSA) and drain directly into abnormal, dilated, and tortuous perimedullary veins [22]. They are occasionally presented concurrently with other rare diseases, such as hereditary haemorrhagic telangiectasia (HHT), Cobb syndrome or Klippel-Trenaunay-Weber syndrome and even tumours [23].

The majority of intradural ventral AVFs frequently present with progressive myelopathic deterioration, such as gradually worsening sensory disturbances and weakness, diffuse back and muscle pain due to venous hypertension or less commonly spinal hemorrhage [24]. However, SAH are more commonly associated with vascular lesions located at the craniocervical junction or in the cervical region, but rarely in the other segments of the spinal cord [11]. It is reported that intracranial SAH can result from a thoracolumbar spinal AVM and AVF, even in the conus medullary, the location of which is far from the cranial and cervical region [12,24–26]. The most common types of lesions are intramedullary spinal AVMs (50%, 11/22), followed by intradural ventral AVFs and intradural dorsal AVFs, according to our summary data. The mechanism underlying the development of intracranial SAH from spinal arteriovenous lesions remains uncertain.

In a few reported cases, spinal AVM or AVF may have venous drainage that extends as far as the intracranial veins or sinuses. It may present with intracranial hemorrhage resulting from venous hypertension and rupture of the intracranial draining vein [29]. Among the 22 cases collected from our hospital and from those previously reported, the rate of the presence of associated aneurysms was 54.5% (12/22). We presume that the rupture of the associated spinal aneurysm of non-cervical AVM is most likely an immediate cause of the intracranial SAH, which is similar to the risk factors for bleeding in cases of intracranial AVMs or AVFs. Spinal AVMs may have one or more arterial feeders, and the relatively fast flow combined with venous hypertension may lead to dilation of the draining vein in a tortuous and ampullary fashion [25] and to formation of the aneurysms on the feeding artery or the draining vessel [27]. Spinal aneurysms, which are very rare, can be divided into two groups: those associated with AVMs and isolated aneurysms [28]. In the study of Madhugiri et al., 42.9% of the spinal aneurysms were associated with spinal AVMs, and 57.1% were isolated spinal aneurysms not associated with spinal AVMs [29]. Jung SC et al. defined the aneurysms into three types according to the location in relation to the nidus of the AVM: prenidal (arterial), intranidal, and postnidal (venous). A postnidal aneurysm presents as a dilated pouch or aneurysmal varix identified at the venous side [28]. In our case series, patient 1 had multiple aneurysms on the draining vessels, patient 2 had an aneurysm near the fistula, and patient 3 had multiple aneurysms on both of the above sites. A fragile arterial wall and haemodynamic factors may contribute to the rupture of aneurysms. Studies have shown that spinal AVMs with aneurysms have a greater tendency to present with hemorrhage than those without aneurysms. Patients with an intranidal aneurysm had a higher incidence of hemorrhage than those with an aneurysm on the feeding vessel of the AVM [29].

The most likely mechanism underlying this phenomenon is the extension of subarachnoid bleeding from the spinal subarachnoid space into the intracranial subarachnoid space [28]. The blood was predominant in the basal cisterns, the perimesencephalic space and in the fourth ventricle in our case series and a few cases reported in the literature, but not a predominance around the foramen magnum. The presence of the blood both in the intracranial and spinal subarachnoid space simultaneously, with more blood around the spinal lesion seems to be the most straightforward evidence to support the blood of spinal origin. Unfortunately, we could not acquire the direct evidence because it is difficult to perform brain and spinal CT scan or the whole spinal neuroaxis MRI and spinal angiography at the same time, except for the presentation with symptoms and signs definitely related to the spinal cord at admission. So we could speculate that if non-cervical spinal AVMs were associated with DSA-negative SAH, the pattern of hemorrhage could be manifested as the blood in supratentorial cisterns, the

fourth ventricle or no copious blood around the foramen magnum as well (somewhat paradoxically), it depends on the timing of detection and image evaluation. But if non-cervical spinal AVMs were not associated with DSA-negative SAH and all cases were genuine cases of 'SAH-of-unknown origin'. The spinal AVM could be considered as incidental finding by chance.

4.3. Imaging evaluation

Intradural ventral AVFs are typically shown on T2 MRI images as serpentine flow voids in the subarachnoid extramedullary space and as changes in the spinal cord due to venous congestion. MRI is the most sensitive diagnostic approach to evaluate hemorrhage of a suspected spinal origin and to localize the lesions [13]. Other non-invasive methods, including spinal MRA, multi-detector-row CTA or 4D CTA [30,31], are available to diagnose and localize the feeding artery, fistulas and draining vein. However, catheter-based angiography is still the gold standard for evaluating spinal angioarchitecture when there is suspicion of a spinal arteriovenous lesion. Observations from the three clinical cases suggest that MRI of the complete spinal neuraxis should be performed to exclude or identify a spinal lesion in patients with negative results on cranial imaging studies [19], and, when the MRI has positive findings, spinal DSA should also be performed.

4.4. Treatment options

The therapeutic goal for intradural ventral AVFs is to interrupt arteriovenous communication while preserving the normal arterial supply and venous drainage of the spinal cord [32]. The most popular classification includes the following categories: i) Type A fistulas are characterized by a single feeding artery with low flow through the arteriovenous shunt; ii) type B fistulas are intermediate in size, usually have multiple feeding arteries and show more marked venous enlargement than the type A; iii) type C fistulas are multipediculated, with high blood flow and enlarged, tortuous draining veins [33]. Current therapeutic modalities include surgery, endovascular therapy and a combination of these two approaches [34]. The treatment decision is based on angioarchitecture as shown by spinal DSA and the type of AVF [35]. For the patients with an SAH, which has been confirmed as a spinal AVM associated with aneurysms, the main goal is to eliminate the aneurysm or reduce the risk of rupture by decreasing the hemodynamic impact. A recent study of 8 spinal AVM patients who had associated aneurysms treated by endovascular management showed that obliteration of the responsible aneurysm can result in immediate improvement in a patient's symptoms and can clinically stabilize the AVM during follow-up [28]. In our cases, surgical occlusion was applied as the main treatment for all the patients for the following reasons: patient 1, the first attempt at superselective microcatheterization was unsuccessful as the feeding artery was too thin and too far from the fistula point; patient 2, the site of the fistula was fed by the posterior spinal artery, which is on the dorsal surface of the spinal cord and had less risk of injury to the spinal cord; patient 3 presented with type B AVF, and we could accurately localize the fistula and surgically excise it with the aid of spinal IO-DSA in a hybrid operating room, which is the most sensitive tool for intraoperative assessment of the spinal vasculature. Regarding the surgical approach, ICG video angiography associated with intraoperative doppler ultrasonography is an important adjunct in the treatment of spinal and cranial AVFs [36,37]. Successful resection of the fistula was achieved in all the cases, and no rebleeding occurred during the follow-up. The therapeutic strategy of spinal AVF associated with an aneurysm should be based on angioarchitecture and the classification of intradural ventral AVF.

5. Conclusions

Intracranial SAH and IVH caused by non-cervical spinal vascular lesions need to be considered when the results of a routine cerebral DSA are negative. The clinical history and the information obtained from the physical examination should be studied with care to identify the possible spinal cause of SAH. MRI of the complete spinal neuraxis is necessary to either exclude or identify a spinal lesion in patients with negative results on cranial imaging studies. Catheter-based spinal angiography remains the gold standard for the evaluation of spinal vascular diseases. If non-cervical spinal AVMs were associated with DSA-negative SAH, the pattern of hemorrhage could be manifested as the blood in supratentorial cisterns, the fourth ventricle or no copious blood around the foramen magnum as well (somewhat paradoxically), it depends on the timing of detection and image evaluation. The formation and the rupture of an associated aneurysm were the most likely immediate cause of the intracranial SAH. If non-cervical spinal AVMs were not associated with DSA-negative SAH and all cases were genuine cases of 'SAH-of-unknown origin', the spinal AVM could be considered as incidental finding. The decision regarding a therapeutic strategy is based on the angioarchitecture and on the type of spinal arteriovenous lesions.

Conflict of interest

All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest or non-financial interest in the subject matter or materials discussed in this manuscript.

Ethical approval

All procedures performed in the studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. For this type of study, formal consent is not required.

Informed consent

Informed consent was obtained from all individual participants included in the study.

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Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.clineuro.2019.105371.

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