Anatomically Based Approach for Endovascular Treatment of Vertebro-Vertebral Arteriovenous Fistula

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Summary

Vertebro-vertebral arteriovenous fistula (VV-AVF) is a rare vascular disorder with an abnormal high-flow shunt between the extracranial vertebral artery (VA), its muscular or radicular branches and an adjacent vein. To date, there are no guidelines on the best treatment for VV-AVF. We present our experience of VV-AVF treatment with covered stents in three patients and detachable coils in two patients. One patient with fistula at the V3 segment had rapid fistula recurrence one week after covered stent treatment. The possible causes of failed treatment in this patient are discussed. The currently available treatment modalities for VV-AVF are also summarized after a literature review. At the end of this article, we propose a new concept of anatomically based approach for endovascular treatment of VV-AVF. Fistula in the V1-2 segments of vertebral artery could be treated safely and effectively by covered stent with the benefit of preserving VA patency. Embolization with variable embolizers should be considered first for fistula in the V3 segment because of the tortuous course and flexibility of the VA in this segment.

Introduction

Vertebro-vertebral arteriovenous fistula (VV-AVF) is a rare vascular disorder with abnormal high-flow shunt between the extracranial vertebral artery (VA), its muscular or radicular branches and an adjacent vein [1]. Most VV-AVF patients have lesions that are spontaneous or caused by neck trauma [2]. Some patients with VV-AVF are asymptomatic. Others may have tinnitus or neurologic deficit because of high flow arteriovenous shunting, steal phenomenon or compression mass effect from enlarged venous pouches [2-5].

The location of VVAVF is also variable with most cases above the C2 vertebra or below the C5 vertebra [6].

Surgical ligation or endovascular closure of the high-flow arteriovenous fistula is the main goal of treatment for VV-AVF [2,3,7,8]. However, there are no guidelines on the best treatment for VV-AVF to date. This article retrospectively reviews our experience in the management of VV-AVF, and proposes our suggestion for endovascular management.

Materials and Methods

Our institution is a referral center for endovascular treatment of neurovascular diseases. From 2006 to 2013, five patients (three men and two women) with vertebro-vertebral arteriovenous fistula (VV-AVF) treated with endovascular surgery in our institution were retrospectively reviewed after we obtained institutional review board approval (Table 1). All these patients were middle-aged, between 42 to 52 years. Three patients had a history of neck injury: one patient with a history of anky-

Table 1

Case	Gender	Age	Fistula location*	Etiology	Presentation	Treatment	Outcome	Follow-up
1	Man	45	V3 (C2)	Trauma (C2 fracture)	Neck pain	GDC	Complete	6 month
2	Man	52	V3 (C1)	Trauma (neck massage)	Pulsatile tinnitus, Headache	1st session: Covered stent (Jostent) 2nd session: GDC, platinum coil & NBCA	Complete, but recurrent Complete	1 week 5 years
3	Woman	52	V1 (C7)	Spontaneous	Headache, SAH+	Covered stent (Jostent)	Complete	14 months
4	Man	42	V1 (C6-7)	Spontaneous	Headache, SAH	Platinum coil & NBCA	Complete	2.5 years
5	Woman	43	V2 (C3)	Trauma (blunt injury)	Pulsatile tinnitus, Neck pain	Covered stent (Viabahn stent)	Complete	8 months

** V1: from origin to transverse foramen of C6, V2: from the transverse foramen of C6 to the transverse foramen of C2, V3: from C2 to the dura.*
+ SAH: subarachnoid hemorrhage.

losing spondylitis had a cervical spine (C2) fracture due to a traffic accident, another had her posterior head and neck hit by a car door two months before symptoms onset, the other had a neck massage hours before symptoms onset.

All these five patients presented with associated symptoms: one patient had neck pain, two patients presented with pulsatile tinnitus, and the other two patients had an initial presentation of severe headache and subarachnoid hemorrhage. VV-AVF was initially suspected on CT angiography (CTA) in one patient and by MR angiography (MRA) in two patients. All five patients had the diagnosis confirmed by digital subtraction angiography (DSA) before endovascular treatment.

All procedures were performed in accordance with the guidelines of the institutional review board, which approved our retrospective medical chart review and analysis.

Results

Locations of the VV-AVF, modality for endovascular treatment, and outcome of our five patients are listed in Table 1. Two patients were treated with coils (GDC in one patient, platinum coil and n-butyl cyanoacrylate (NBCA) glue in the other). Three patients had endovascular reconstruction with stent graft (Jomed covered stent grafts in two patients and Viabahn stent graft in one patient). After stent graft reconstruction, dual antiplatelet therapy with 75 mg of clopidogrel orally every day for two months and lifelong low dose aspirin (100 mg) orally every day was advised. The VV-AVF in all these patients was complete obliterated on post-treatment control angiography. Unfortunately, one patient who underwent endovascular stent graft reconstruction had recurrent VV-AVF due to type 1b endoleak one week after initial treatment. Therefore, a second ses-

sion of endovascular trapping of the left VA was performed using GDC, platinum coil, and NBCA glue smoothly. All patients had long-term follow-up (from eight months to six years) without symptom or fistula recurrence.

Selected Case

Case 1

A 43-year-old woman visited our Neurology Clinic for progressive left pulsatile tinnitus, and left posterior neck soreness and tightness for one month. She had her posterior head and neck hit by car door about one month before symptoms onset. CT angiography (CTA) was arranged for evaluation of possible vasculopathy and showed prominent epidural and paraspinal engorged venous structures in upper neck (Figure 1A). Suspected direct connection between the left VA and adjacent venous structure at the C3 level was also seen (Figure 1B). Therefore, DSA was arranged and confirmed the left high-flow VV-AVF at the C3 level (Figure 1C,D). Endovascular reconstruction was arranged five days later. A 6-Fr shuttle sheath with continuous heparinized-saline flush was placed in the left subclavian artery. After identifying the fistula site, a self-expanding covered stent (Viabahn, Gore, 5×50 mm) was navigated to the left VA and deployed at the C2-5 level of left VA. After post-stenting angioplasty with balloon catheter (PTA balloon, Covidien, 5x15 mm) the fistula was completely obliterated. Post-stenting angiography of bilateral VA showed no further VV-AVF (Figure 1E). CTA follow-up eight months later showed patency of the covered stent and no recurrent fistula (Figure 1F).

Case 2

A 52-year-old man heard pulsatile bruit a few hours after having a neck massage. The next day, he felt occipital headache. The bruit and headache both progressed gradually over time. Brain MRA was arranged in the Neurology Clinic two weeks later and revealed enlarged veins with arterialized flow surrounding the left VA at the C1-2 level (Figure 2A). Cerebral DSA confirmed the high flow arteriovenous shunt arising from the left VA and draining into adjacent veins (Figure 2B). After explanation and discussion, the patient chose to have endovascular stent graft reconstruction first. After establishing bilateral femoral arterial access, a 6-Fr straight-tip guiding catheter with continuous heparinized-saline flush was navigated to left VA. In addition, a 4-Fr H-1 diagnostic catheter with continuous heparinized-saline flush was placed in the right VA for mapping of the left VA distal to the VV.AVF. We obtained angiography of bilateral VAs to confirm the exact fistula location, size of the parent artery, and venous drainage status. Two balloon-expandable covered stents (Jostent, Abbott, 4×19mm and 4×16mm) were overlapped and deployed to cover the fistula completely. Post-stenting angiography of bilateral VAs showed complete obliteration of the VV-AVF (Figure 2D). The patient had no further bruit or headache after endovascular treatment. However, he heard recurrent pulsatile bruit again one week after initially treatment, and recurrent VV-AVF because of type 1b endoleak was shown on the DSA (Figure 2E). This time, the left VA was trapped using GDC, platinum coils, and NBCA glue (Figure 2F). The patient had no recurrent AVF during the subsequent follow-up for five years.

Discussion

Vertebro-vertebral arteriovenous fistula (VV-AVF) is a rare vascular disorder with an abnormal shunt between the extracranial vertebral artery (VA), its muscular or radicular branches and adjacent veins [1]. Trauma is the most common cause, including knife wounds, gunshot injury and blunt trauma. Other patients have spontaneous fistulas, partly associated with fibromuscular dysplasia [1,2]. Incidental iatrogenic injury during jugular vein puncture and traumatic fracture and dislocations of the cervical spine are also possible etiologies [4,8,9]. Our case series comprised three traumatic fistulas after blunt neck injury and two spontaneous fistulas without associated fibromuscular dysplasia or other vasculopathy. Therefore, the possibility of VV-AVF in patients without neck trauma or underlying vasculopathy cannot be excluded in our experience and in the literature.

The locations of fistula are variable. According to the literature, the fistula is located at C1 to C2 segments (46%) or below the C5 segment (44%) in most cases of VV-AVF, and fistulas at C2 to C5 segment are uncommon (11%) [6]. However, the VV-AVF at C2-C5 vertebral segment could be more common in pa-

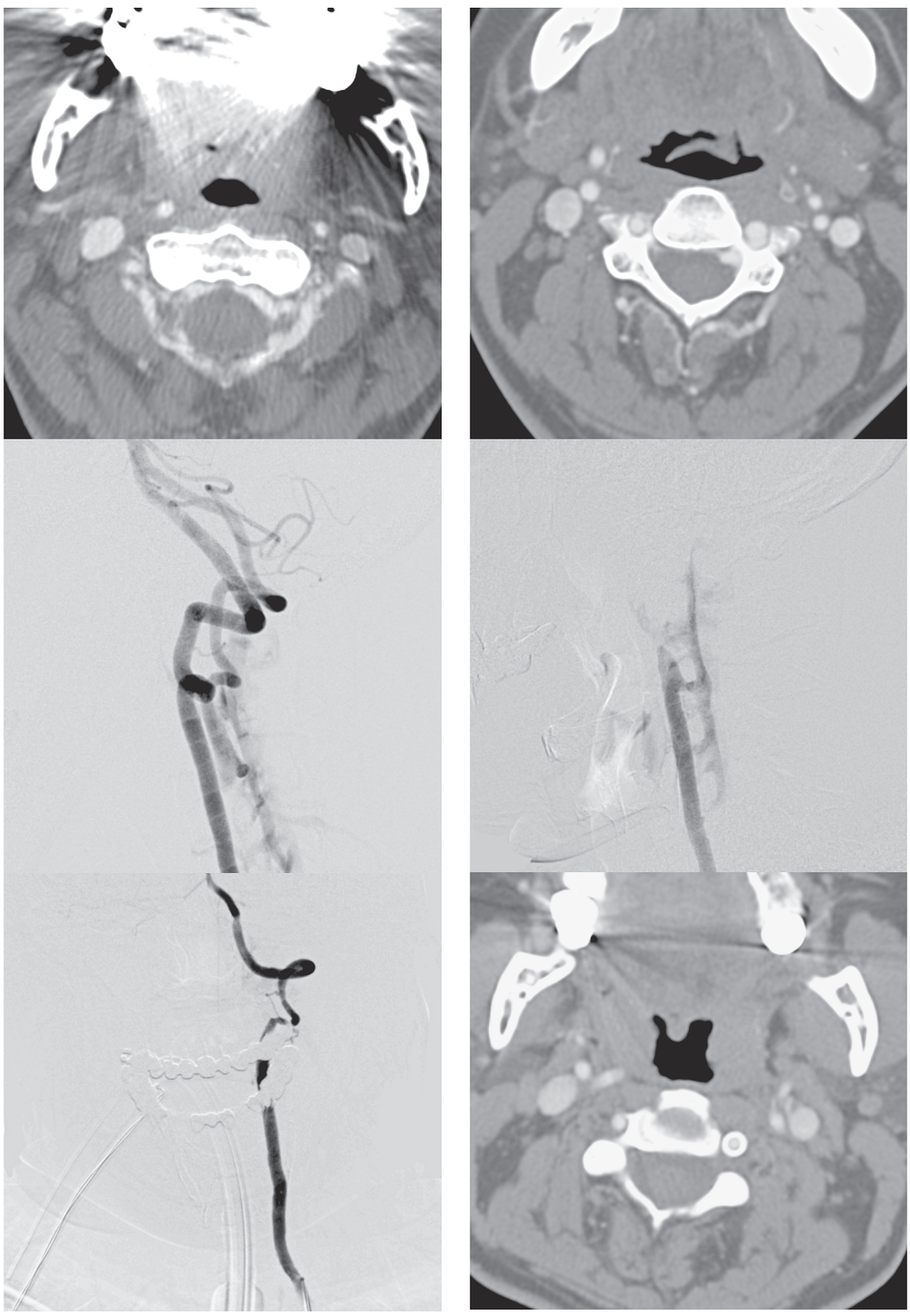

Figure 1 A 43-year-old woman with a history of blunt neck injury had pulsatile tinnitus and neck soreness. A) CT angiography (CTA) showed prominent epidural and paraspinal engorged venous structures in upper neck. B) Suspected direct connection between the left VA and adjacent vein at the C3 level. C,D) Digital subtraction angiography (DSA) of the right and left VA in lateral projection confirmed the left high-flow VV-AVF at the left C3 level. E) After deployment of a covered stent at the C2-5 level and post-stenting angioplasty, the fistula was completely obliterated. F) CTA follow-up 8 months after treatment showed lumen patency of the stent without recurrent fistula.

Figure 2 A 52-year-old man felt pulsatile bruit and progressive neck pain after having neck massage. A) Brain MR angiography revealed enlarged veins with arterialized flow surrounding the left VA at the C1-2 level. B) High flow arteriovenous shunt arising from the left VA and draining into adjacent veins was confirmed by DSA. C,D) The VV-AVF was completely obliterated after deploying two overlapping balloon-expandable covered stents. E) One week after initial treatment, recurrent VV-AVF because of type 1b endoleak was shown on the DSA. F) The left VA was trapped using GDC, platinum coils, and NBCA glue with no further recurrence during the subsequent 5-year follow-up..

tients with penetrating traumatic patients (38.8%) [3], probably because of the increased neck trauma in zone II of the neck (zone II lies between the cricoid and the angle of the mandible) [10]. None of our patients had a penetrating neck injury, and accordingly fistula at C2 to C5 is uncommon in our case series. In addition, comprehensive angiography study of the carotid or subclavian arteries is necessary for detection of possible collateral circulation in accordance with the fistula location. For instance, the occipital artery is a possible accessory feeding artery to fistula in the C1-C2 segment. Fistula at the C5-C7 segment may also receive arterial supply from the ascending cervical artery.

The clinical symptoms of these patients are also variable. Some asymptomatic patients were incidentally discovered at routine clinical examination. Tinnitus is a common symptom due to rapid arteriovenous shunting across the fistula. Neurologic symptoms of vertigo and diplopia could be secondary to the vertebral arterial steal phenomena by the high-flow arteriovenous shunting. Venous congestion or direct compressive mass effect from engorged epidural venous pouches can cause headache, cervical neuralgia, compressive radiculopathy or even myelopathy [2-5]. All of our patients had symptomatic fistulas regardless of their etiologies. But surprisingly, two patients with spontaneous fistulas both had abrupt explosive headache and diffuse subarachnoid hemorrhage (SAH), which is a typical scenario encountered in cerebral aneurysmal bleeding. Spontaneous VV-AVF with initial symptoms of SAH is not common, and only reported by a rare case report in the literature [11]. Therefore, VV-AVF should be considered in patients with spontaneous SAH without cerebral aneurysm or AVF. Especially when the patient had negative brain DSA study repeatedly, we should consider imaging the extracranial VA before come to the diagnosis of perimesencephalic venous hemorrhage.

Obliterating the high-flow arteriovenous shunting is the primary goal for treatment of VV-AVF. However, there are still no guidelines on the best treatment for VV-AVF to date. Surgical exposure and ligation of the fistula could be difficult because of extensive hemorrhage from the surrounding arterialized Batson plexus and only applicable in selected case [1,8]. Endovascular embolotherapy using detachable balloons has been used safely and effectively in the treatment of VV-AVF for more than 25 years and is still the treatment of choice in countries that still have access to the balloons [1-3]. Detachable coils are an effective alternative when detachable balloons are unavailable [3]. Recently, successful treatment of VV-AVF by transarterial Onyx or vascular plug embolization was also reported in some cases [12,13]. However, embolizer misplacement or migration to downstream arterial or venous structures may occur and cause ischemic complications or fistula recurrence. Moreover, the patient may suffer from vertebrobasilar ischemia caused by atherosclerotic stenosis in the contralateral VA later in life. Therefore, to occlude the fistula site and preserve VA patency simultaneously may benefit the patients. Combined therapy using detachable coils and uncovered stent could be used to reconstruct the VA [3]. A self-expanding or balloon-expandable covered stent is another emerging option to reconstruct the injured head and neck arteries. Preliminary results suggest that placement of stent-grafts is a safe and effective method of treating carotid and vertebral arterial pseudoaneurysms [14,15]. Covered stent reconstruction for VV-AVF was also performed successfully in some case reports [16,17]. However, stent graft reconstruction necessitates long-term antiplatelet therapy at the expense of increased risk of major bleeding, especially when subsequent orthopedic or cerebrospinal fluid diversion surgery is necessary for patients with traumatic fistula or subarachnoid hemorrhage. Guidelines for perioperative management of antiplatelet therapy are available in the literature [18].

Despite the promising results of covered stent in our case series and in the literature, one of our patients who underwent endovascular reconstruction using covered stent had rapid fistula recurrence, which required a second treatment by parent artery occlusion. This may be explained by two reasons. First, the V3 segment of the VA is very flexible with a frequent change in its conformation in accord with the wide range of motion at the craniocervical junction. A covered stent without enough compliance may kink, fracture or migrate during posture change of head, causing insufficient seal of the fistula. Second, the tortuous course in the V3 segment of VA precludes the operator from introducing a long, large-profile, and rigid covered stent into this segment. Like the situation we encountered in the reported case 2, instead of a longer and larger stent graft, we had to deploy two overlapping covered stents to occlude the fistula completely. The risk of

stent migration and fistula recurrence is surely higher when inserting two overlapping stents into a tortuous and very flexible arterial structure (V3 segment) than deploying a single stent in a straight and inflexible artery (V1-2 segment). Therefore, for VV-AVF in the V3 segment, treatment using a covered stent could be a very difficult and dangerous with a potential risk of stent migration, kinking, fracture and fistula recurrence.

According to our experience in treating VV-AVF in these patients and the experience of other authors in the literature, we propose an original anatomically based approach for endovascular treatment of this rare vascular disorder. In patients with a fistula at the V1-2 segment of the VA, covered stent reconstruction is a safe and effective treatment choice with the benefit of preserving ipsilateral VA flow. If the fistula is located in the V3 segment, transarterial embolization with detachable coils, vascular plug, Onyx, or NBCA glue is preferable to a covered stent for the increased vertebral artery mobility and greater risk of fistula recurrence.

There are some limitations of this study. First, the study is retrospective in nature and therefore not controlled for selection bias, detection of events, and data collection. Second, because VV-AVF is a relatively rare vascular disorder, the number of cases in our study is fairly small to devise definite guidelines on how this disease should be treated. Third, the follow-up period in our study is short compared with the known long-term results of detachable balloons. Further, prospective randomized trials with long-term follow-up study are required to monitor the long-term durability of stent grafts in extracranial vertebral arteries.

Conclusions

In conclusion, we retrospectively reviewed our experience in the treatment of VV-AVF and propose a new anatomically based approach for VV-AVF. Fistula in the V1-2 segments could be treated safely and effectively by covered stent with the benefit of preserving VA patency. Embolization with variable embolizers should be considered first for fistula in the V3 segment.

References

1 Halbach VV, Higashida RT, Hieshima GB. Treatment of vertebral arteriovenous fistulas. Am J Roentgenol. 1988;150(2):405-412. doi: 10.2214/ajr.150.2.405.
2 Beaujeux RL, Reizine DC, Casasco A, et al. Endovascular treatment of vertebral arteriovenous fistula. Radiology. 1992;183(2):361-367.
3 Herrera DA, Vargas SA, Dublin AB. Endovascular treatment of traumatic injuries of the vertebral artery. Am J Neuroradiol. 2008; 29 (8): 1585-1589. doi: 10.3174/ajnr.A1123.
4 Vinchon M, Laurian C, George B, et al. Vertebral arteriovenous fistulas: a study of 49 cases and review of the literature. Cardiovasc Surg. 1994; 2 (3): 359-369.
5 Ito O, Nishimura A, Ishido K, et al. Spontaneous vertebral arteriovenous fistula manifesting as radiculopathy. No Shinkei Geka. 2011; 39 (8): 775-781.
6 Desouza RM, Crocker MJ, Haliasos N, et al. Blunt traumatic vertebral artery injury: a clinical review. Eur Spine J. 2011; 20 (9): 1405-1416. doi: 10.1007/s00586-011-1862-y.
7 Briganti F, Tedeschi E, Leone G, et al. Endovascular treatment of vertebro-vertebral arteriovenous fistula. A report of three cases and literature review. Neuroradiol J. 2013; 26 (3): 339-346.
8 Ishiguro T, Kawashima A, Yoneyama T, et al. Two cases of iatrogenic vertebral arteriovenous fistulas successfully treated by surgery. No Shinkei Geka. 2011; 39 (3): 269-274.
9 Boot BP, Macdonald JH, Parker GD, et al. Teaching NeuroImages: traumatic vertebral arteriovenous fistula. Neurology. 2011; 76 (7): e29-30. doi: 10.1212/WNL.0b013e31820c2eaa.
10 Munera F, Soto JA, Palacio DM, et al. Penetrating neck injuries: helical CT angiography for initial evaluation. Radiology. 2002; 224 (2): 366-372. doi: 10.1148/radiol.2242010973.
11 Hiu T, Yoshioka T, Kitagawa N, et al. Vertebral arteriovenous fistula presenting with subarachnoid hemorrhage: a case report. No Shinkei Geka. 2009; 3 7(7): 667-671.
12 Gordhan A. Onyx embolization of high-flow spontaneous cervical vertebral arteriovenous fistula. Vasc Endovascular Surg. 2012; 46 (6): 484-486. doi: 10.1177/1538574412452156.
13 Mihlon F, Agrawal A, Nimjee SM, et al. Enhanced, rapid occlusion of carotid and vertebral arteries using the AMPLATZER Vascular Plug II device: The Duke Cerebrovascular Center Experience in 8 patients with 22 AMPLATZER Vascular Plug II devices. World Neurosurg. 2013. doi: 10.1016/j.wneu.2013.07.084.
14 Maras D, Lioupis C, Magoufis G, et al. Covered stent-graft treatment of traumatic internal carotid artery pseudoaneurysms: a review. Cardiovasc Intervent Radiol. 2006; 29 (6): 958-968. doi: 10.1007/s00270-005-0367-7.
15 Yi AC, Palmer E, Luh GY, et al. Endovascular treatment of carotid and vertebral pseudoaneurysms with covered stents. Am J Neuroradiol. 2008; 29 (5): 983-987. doi: 10.3174/ajnr.A0946.
16 Sancak T, Bilgic S, Ustuner E. Endovascular stent-graft treatment of a traumatic vertebral artery pseudoaneurysm and vertebrojugular fistula. Korean J Radiol. 2008; 9 Suppl: S68-72. doi: 10.3348/kjr.2008.9.s.s68.
17 Li F, Song X, Liu C, et al. Endovascular stent-graft treatment for a traumatic vertebrovertebral arteriovenous fistula with pseudoaneurysm. Ann Vasc Surg. 2014; 28 (2): 489 e411-484. doi: 10.1016/j.avsg.2012.12.013.
18 Darvish-Kazem S, Gandhi M, Marcucci M, et al. Perioperative management of antiplatelet therapy in patients with a coronary stent who need noncardiac surgery: a systematic review of clinical practice guidelines. Chest. 2013; 144 (6): 1848-1856. doi: 10.1378/chest.13-0459.

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