

Spontaneous Regression of an Arteriovenous Fistula Between Paravertebral Venous Plexus and the Vertebral Artery: A Rare Case Report

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Case report

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Abstract

Background: Spontaneous regression of huge vertebral arteriovenous fistula (AV fistulas) due to acupuncture procedure is a rare phenomenon. The clinical presentations, management, prognosis of this type arteriovenous fistula has been rarely reported. In addition, we exclusively analyzed the possible reasons for its spontaneous regression.

Case presentation: A 57-year-old Chinese woman presented with a 2-month history of progressively worsening headache and high-pitched tinnitus after acupuncture and massage in the neck and the left mastoid region. Later Digital Subtraction Angiography (DSA) confirmed the presence of arteriovenous fistulas at cervical 2-3 (C2-3) level, lying between the paravertebral venous plexus and the left vertebral artery. The patient had a background of hypertension, hypothyroidism and hysterectomy. The arteriovenous fistulas disappeared mysteriously, possibly because of the motion of the left vertebral artery and the compression of guide wire against the blood vessel wall. The patient recovered well following conservative treatment of taking atorvastatin calcium tablets (20mg/day), despite delayed recognition and invasive investigation.

Conclusion: Adequate evaluation and standardized procedures with Doppler may minimize the associated risks while performing acupuncture. Early application of vascular examination should be considered in patients with typical high-pitched tinnitus and medical history associated with acupuncture treatment. Statins are effective in repairing the lining of blood vessels.

Background

Acupuncture is a traditional alternative therapeutic option that has been widely used in pain management [1]. It involves the insertion of fine sliver needles through the subject's skin at specific points and varied depths, to achieve a desirable result. Individuals suffering from chronic neck and back pain can benefit from the treatment of an experienced acupuncturist[1]. However, lack of precautions at acupuncture point may lead to severe complications while performing needling. Here, we report a rare case of arteriovenous fistula of the left vertebral artery possibly induced by acupuncture procedure and discussed the potential mechanism for its spontaneous disappearance.

Case Presentation

A 57-year-old Chinese woman presented to our comprehensive stroke center with a 2-month history of neck discomfort, progressively worsening headache, and pulsatile tinnitus. The patient also felt upset and had difficulty falling asleep after the symptoms above shown up. She had cervical spondylosis for years. A week before these symptoms shown up, she had undergone a single session of acupuncture therapy for pain control related to the cervical spondylosis. During the treatment, multiple needle punctures had been administered around the neck and the left mastoid region. No other trauma to the cervical region had occurred. She had experienced sharp pain in the neck and required that the needles be removed. The

sharp pain faded after the removal of the needles and she did not notice any abnormality in the neck region. Two months before presentation to our stroke center, however, the patient had suffered a sudden high-pitched tinnitus and shooting pain in the neck region while she was sleeping in the night. The pain and tinnitus did not resolve in the following days, though she continued to seek help and acupuncture therapy from doctors at local Chinese medicine hospital. No vascular examinations were performed during this session of treatment. The direction, however, seemed to be wrong.

Almost 2 months after typical symptoms onset, fortunately, she was arranged for Computed Tomography Angiography at local hospital, showing a huge mass of abnormal vascular shadows at C2-3 level of the left vertebral artery. Considering the complexity and severity of this lesion, she was recommended to the superior hospital for further treatment.

Pertinent clinical findings on presentation to our hospital included insomnia and a high-pitched murmur over the left mastoid and neck region. Vital signs on arrival were unremarkable except for a mild elevation of blood pressure to 145/97 mmHg. The patient had a background of hypertension, hypothyroidism, and hysterectomy. Risk factors for vascular disorder like unhealthy diet and tobacco or alcohol abuse were absent.

Given the patient's presenting features, imaging findings and in combination with lack of therapeutic response previously, the presumptive diagnosis of acquired arteriovenous fistula was made. Later she was started on atorvastatin calcium tablets (20 mg/day). Additional medications include levothyroxine sodium tablets and amlodipine besylate tablets.

After the preoperative examination was ready on the 4th day of hospitalization, cerebral angiography was adopted and then confirmed an arteriovenous fistula between the paravertebral venous plexus and the left vertebral artery at the level of C2-3 (Fig. 1. b, c, d). This fistula was not visualized by right vertebral angiography (Fig. 1. e, f). Not surprisingly, symptoms of persistent headache and pulsatile tinnitus still existed after this examination. However, one day later, the patient unexpectedly fell asleep for the first time in 2 months. In addition, the tinnitus faded away spontaneously, and the high-pitched murmur was inaudible in the next morning. Sudden alterations in the features of murmurs suggest that the orifice, the flow direction or the size of the arteriovenous fistula may have changed. To figure out what had happened to the fistula, cerebral angiography was performed under general anesthesia and endovascular treatment was prepared. Multi-angle angiography revealed that the arteriovenous fistula had disappeared spontaneously (Fig. 1. i, j). However, the vascular wall was not smooth and there seemed to be a partial filling defect in the C2-3 segment of the left vertebral artery, where was presumed to be the location of the previous fistula. Two months later, she remained asymptomatic and no abnormal vascular shadows were detected by cerebral angiography (Fig. 1. k, l). In addition, the left vertebral artery displayed a relative smooth intima, which might be the result of taking statins for two months.

Discussion

Acupuncture therapy has enjoyed overwhelming popularity in both Asian countries and western part of the world, with a low prevalence of major adverse events at approximately 5.5 cases per 100 000 patients [2, 3]. It means that severe vascular injuries caused by acupuncture are, indeed, rare. However, using this ancient technique may lead to severe complications, including regional or systemic infections, pseudoaneurysm, acquired arteriovenous fistulas, arterial dissection, as well as injuries to central nervous system[2, 4].

In this case, digital subtraction angiography was not performed until more than 2 months after the onset of symptoms, despite history of acupuncture treatment, massage, and high-pitched murmurs over the left mastoid region at presentation to local hospital. A typical history and clear evidence of vascular injury are sufficient to immediately pursue a routine vascular examination. Unfortunately, it is likely that both the patient herself and the attending physician were falsely reassured by the safety of acupuncture. However, acquired cervical vertebral arteriovenous fistulas are usually developed from penetrating injuries rather than blunt trauma[5]. In this patient, the vascular lesion is possibly caused by a deep insertion with 5–6 cm long acupuncture needles into the vertebral artery. Right after acupuncture treatment in the left mastoid and the neck region, the patient happened to receive neck massage, which might promote the arteriovenous fistula formation. In addition, cerebral angiography demonstrated that the left vertebral artery was the dominant vertebral artery (Fig. 1. a), where the vessel wall might be subjected to greater hemodynamic pressure. Reviewing the literature, we found that spontaneous or acquired vertebral arteriovenous fistula often developed at around C2 section of the vertebral artery, which might be associated with the specific anatomical structure of this site[6, 7]. It is presumed that the increased strain of this segment of artery occurring with neck rotations, coupled with intrinsic weakness of the arterial wall, might be a predisposing factor [8]. In traditional Chinese medicine, there is an acupuncture point called Tianchuang for the treatment of cervical spondylosis, neck pain and tinnitus, located exactly at the C2-3 segment of the vertebral artery. Therefore, if acupuncture is necessary, we highly recommend performing this procedure under the guidance of Doppler, which may help to eliminate some severe complications.

The pathophysiology of its sudden regression has not been well established but is often associated with minor penetrating injury. Choudhri et al[9] reported that neck rotations and vertebral arterial movements might help to reduce blood flow through the fistula, thus promoting to its spontaneous regression. In this patient, a sharp twist was observed at the very beginning of the left vertebral artery during the first digital subtraction angiography procedure (Fig. 1. a). To send the catheter into the left vertebral artery for super-selective angiography, the guide wire was placed at a very high position in the left vertebral artery, which might have a compression effect against the vessel wall during the procedure. As the catheter was sent into the targeted vessel, the vertebral artery showed a significant movement. The intima of the artery might be overturned to a proper position where it could gradually block off the fistula. Combined the indications of both cerebral angiography and cervical magnetic resonance imaging, we presumed that there was a minor dissection at the fistula. To our understanding, the guide wire might happen to serve as an intravascular stent or balloon, which eliminated the minor dissection. In addition, statins might involve in the repair of vascular intima. Recent studies have shown that endovascular occlusion of fistula is the

optimal treatment because of its low odds for long-term recurrence[8, 10]. This endovascular technique often utilizes detachable coils, stent grafts, or detachable balloons as embolic materials[7]. In general, the primary target of therapeutic management is to occlude the fistula and protect the parent artery.

Conclusions

Adequate evaluation and standardized procedures with Doppler may minimize the associated risks while performing acupuncture. Knowledge in vascular anatomy and microbiology are essential for application of this technique to reduce severe complications. Here we recommended that acupuncture be performed under ultrasound guidance. Early cerebral angiography and thoroughly physical examination are critical for diagnosis of such vascular lesions caused by acupuncture. Patients with traumatic arteriovenous fistula may yield a better result from taking atorvastatin calcium tablets. Endovascular occlusion remains an alternative medical treatment for vertebral arteriovenous fistula.

Abbreviations

AV fistulas: arteriovenous fistulas; DSA: Digital subtraction angiography; C2-3: cervical 2-3; MRI: magnetic resonance imaging.

Declarations

Ethics approval and consent to participate

The study was carried out in accordance with the Declaration of Helsinki and was approved by the Ethics Committee of The Affiliated Brain Hospital of Nanjing Medical University.

Consent for publication

Informed written consent for publication was obtained from the patient.

Availability of data and materials

The datasets are available from the corresponding author upon reasonable request.

Competing interests

The authors declare no conflict of interest relevant to this article.

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Authors' contributions

All authors contributed to the study conception and design. The clinical data and related images were collected by Xingcheng Bai. The manuscript was first drafted by Zhiqiang Yu and Guangxu Zhang and it was then revised by Jinbing Zhao and Jun Ma, and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

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Figures

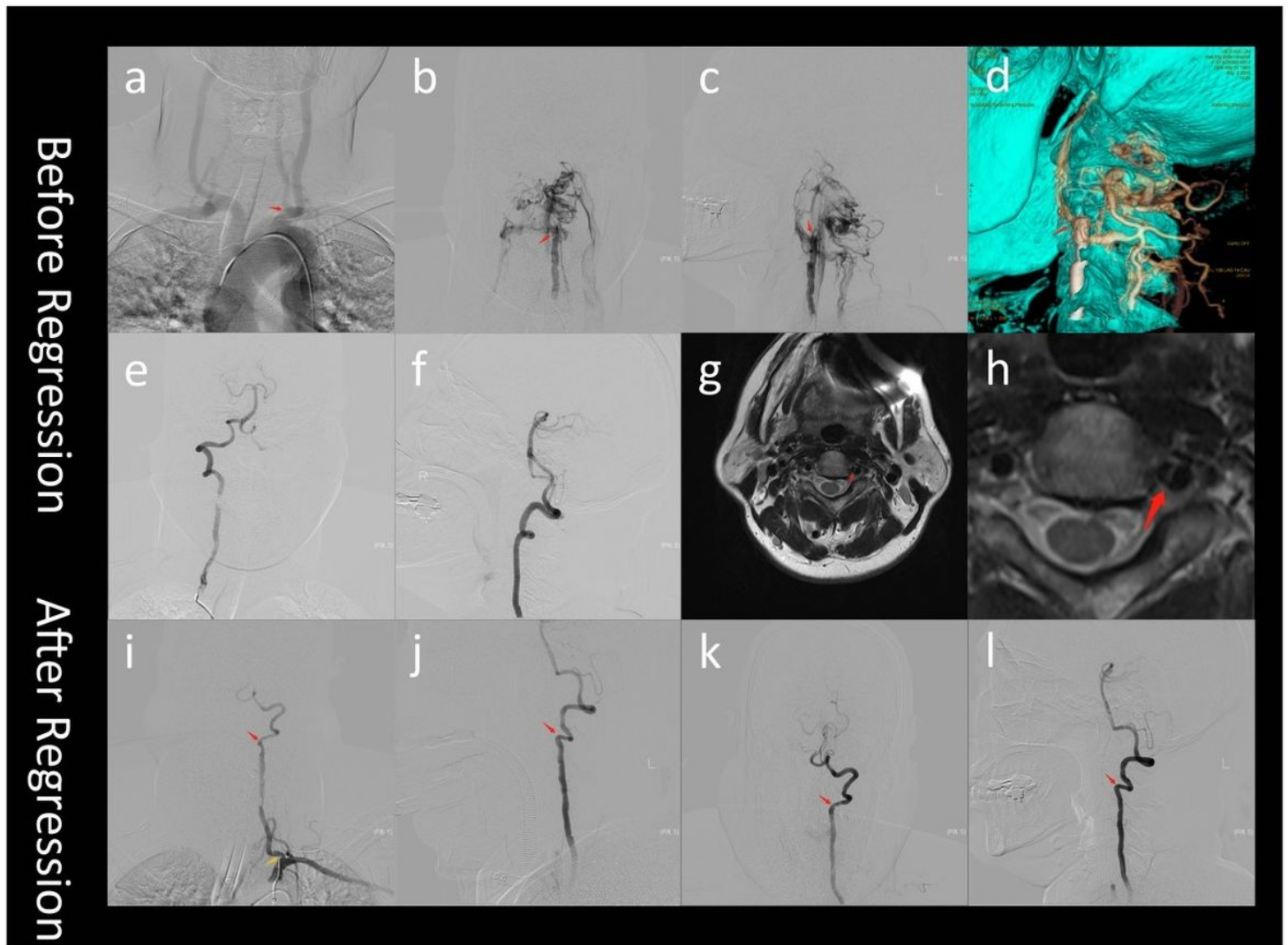


Figure 1

Representative images of digital subtraction angiography (DSA) and cervical magnetic resonance imaging (MRI) for this patient at baseline (a-h) and follow-up (i-l) examinations. (a) The aortic arch angiography showed the left vertebral artery was the predominant artery compared to that of the right one. (b-d) Baseline DSA images (anterior, lateral projections and 3D imaging) demonstrated the fistula located exactly at the C2-3 segment of the left vertebral artery (red arrow), while the right vertebral artery (e-f) appeared to be in a sound condition. (g-h) Cervical MRI revealed there might be a micro dissection of the left vertebral artery at the level of C2-3 segment (red arrow). (i-j) Spontaneously regression of the arteriovenous fistula on the 6th day of hospitalization. A sharp twist was observed at the very beginning of the left vertebral artery (yellow arrow). (k-l) Two month after discharge, the follow-up DSA images confirmed a relatively smooth intima and no recurrence of the fistula.