Spontaneous Pseudoaneurysm of the Vertebral Artery in Behçet's Disease

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A pulsatile mass and severe neck pain developed suddenly in a 15-year-old female patient suffering from Behçet's disease. Magnetic resonance imaging showed a pseudoaneurysm at the C_3-C_4 level that was $51 \times 49 \times 45$ mm in size, originating from the left vertebral artery, with a thin neck and thrombus inside. Repair of the vertebral artery wall by percutaneous transluminal intervention was not successful. Because of the possibility of rupture, the patient underwent surgical repair. Ligation of the left vertebral artery was applied 1 cm above the origin of the subclavian artery. During the subsequent postoperative period, no further complications were seen. From our review of the literature, this is the first reported case of surgical treatment of spontaneous development of a pseudoaneurysm at the vertebral artery in association with Behçet's disease. Ligation of the vertebral artery can be safely used to control a pseudoaneurysm related to Behçet's disease.

Behçet's disease is a rare, chronic, autoimmune, multisystem disorder that causes inflammation of blood vessels (vasculitis) anywhere in the body. Pseudoaneurysm is dilatation of an artery with arterial disruption of one or more layers of the wall, as can occur with puncture as a complication of percutaneous arterial catheterization, rather than expansion of all layers of the wall.

Pseudoaneurysm formation in the arterial system can be seen in Behçet's disease. The most common site of pseudoaneurysm formation is the femoral artery.[1] Spontaneous pseudoaneurysm of the vertebral artery, treated surgically, has not been reported. We present such a case here. In this study, we explain our strategy for management of the pseudoaneurysm that developed at the vertebral artery through Behçet's disease, and review the related literature.

CASE REPORT

A 15-year-old female patient was admitted to our outpatient clinic with a spontaneously occuring pulsatile mass and severe pain on the left side of her neck. The patient had been followed and treated in our outpatient clinic for deep venous thrombosis of the lower extremities for 5 months. Five months previously the patient had come to our outpatient department because of swelling and pain in the lower extremities. In the patient's history, recurrent oral and genital ulceration and arthralgia, especially in the left shoulder and mandibular joint, were found. Laboratory findings were mainly nonspecific indices of inflamation, such as leukocytosis and elevated erythrocyte sedimentation rate and C-reactive protein level. After neurologic, ophtalmologic, dermatologic, and rheumatologic consultation, the diagnosis was neuro-Behçet's disease, according to the criteria of the International Study Group for Behçet's Disease.[2] The patient was hospitalized and systemic corticosteroid and heparin therapy was administrated. The patient was discharged and followed with anticoagulant therapy in the outpatient clinic.

When the patient returned to our outpatient clinic with spontaneous pulsatile mass formation, color Doppler ultrasonographic examination was done and pseudoaneurysm of the left vertebral artery was diagnosed. After ultrasonographic examination, magnetic resonance angiography of the vertebral and carotid arteries was performed. A pseudoaneurysm originating from the vertebral artery at the C_3-C_4 level, $51 \times 49 \times 45$ mm in size, was shown (Figs. 1 and 2). The first choice of

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Fig. 1. Magnetic resonance angiographic scan of pseudoaneurysm originating from the left vertebral artery (*arrows*).

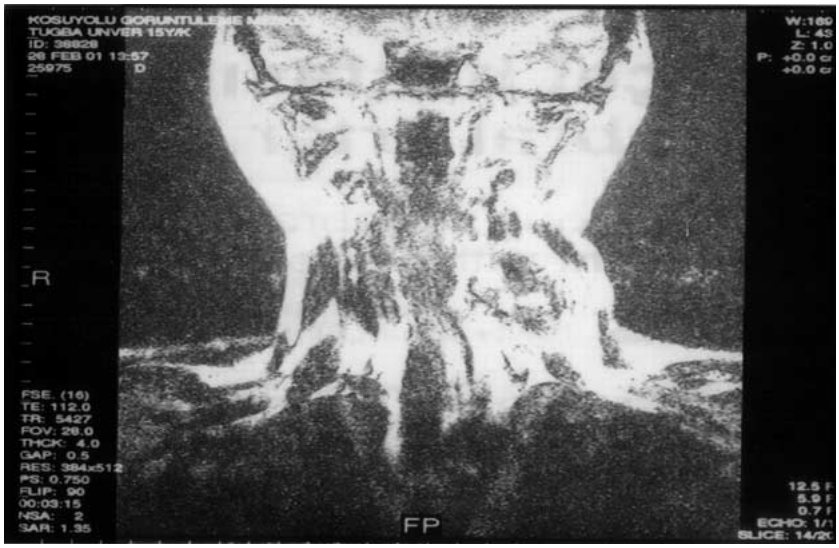

Fig. 2. Magnetic resonance image of pseudoaneurysm.

treatment was percutaneous transluminal intervention. But repair of the vertebral artery wall by percutaneous transluminal covered stenting was not successful. When the size of the pseudoaneurysm increased to the point that it was perceptible from outside and there was numbness in the left shoulder extending to the forearm, because the possibility of vessel rupture the patient underwent emergency surgical repair. The left vertebral artery was exposed by supraclavicular incision under cervical blockage and local infiltration anesthesia. Then ligation of the vertebral artery was performed 1 cm above the origin of the subclavian artery.

During the subsequent postoperative period, no complication was seen. The pulsatile mass on the neck and neurologic symptoms of the patient disappeared and the patient was discharged from the hospital after 4 days. After 1 week, the patient underwent an magnetic resonance angiographic scan of the vertebral and carotid arteries (Fig. 3). Despite retrograde flow of the ligated vertebral artery on magnetic resonance imaging, this flow did not cause re-formation of the aneurysm of the vertebral artery. We decided to follow this patient at 6-month intervals.

DISCUSSION

Pseudoaneurysm of the vertebral artery is a rare condition because of its anatomic site and proper-