

Paradoxical Cerebral Embolism in a Patient with Paget-Schroetter Syndrome

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A 28-year-old man presented with transient speech disturbance and right hemiplegia. Computed tomography of the brain revealed a low-density area in the right cerebellum. A ventilation/perfusion lung scintiscan detected multiple perfusion defects in the both lungs and catheterization revealed pulmonary hypertension. Venography of the upper extremities revealed obstruction of the left subclavian vein. Furthermore, Doppler echocardiography revealed a right-to-left shunt via a patent foramen ovale. Those examinations demonstrated paradoxical cerebral embolism caused by Paget-Schroetter syndrome, which is a rare complication of the disorder. We hypothesize that the source of thrombi was the left subclavian vein and surgery was needed to prevent further thromboembolic events. At surgery, the upper half of the sternum was incised in the midline, and the left brachiocephalic vein was ligated. No thromboembolic episodes have occurred postoperatively. (Ann Thorac Cardiovasc Surg 2005; 11: 429–31)

Key words: paradoxical cerebral embolism, Paget-Schroetter syndrome, patent foramen ovale, pulmonary embolism

Introduction

Primary subclavian vein thrombosis, or Paget-Schroetter syndrome, rarely causes thromboembolism. However, we describe a patient with Paget-Schroetter syndrome, who suffered paradoxical cerebral embolism and pulmonary embolism. We performed surgery to treat this complication of Paget-Schroetter syndrome.

Case

A 28-year-old man, who was a field hockey player, complained of dyspnea on exertion. Three months later, he experienced transient speech disturbance and right hemiplegia. Computed tomography of the brain revealed a low-density area in the right cerebellum. He was admitted to our hospital to determine the origin of the embolus.

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No abnormal neurologic findings were detected at admission. The pulse was regular and electrocardiography showed normal sinus rhythm. A systolic murmur (Levine I/VI) was detected at the left sternal border in the fourth intercostal space. There was no difference in the bilateral circumferences of the upper or lower extremities. The superficial veins of the upper extremities were not dilated. There was no decrease in pulsation in either radial artery on Adson's test. There was no evidence of varicose veins or pretibial edema in either lower extremity.

A ventilation/perfusion lung scintiscan revealed multiple perfusion defects in both lungs and suggested the presence of pulmonary embolisms. Catheterization revealed pulmonary hypertension (systolic pressure: 49 mmHg, mean pressure: 26 mmHg). Although no deep venous thrombosis was detected on venography of the lower extremities, venography of the upper extremities revealed obstruction from the left subclavian vein to the left brachial vein and the presence of well-developed collaterals. Furthermore, retrograde venography revealed obstruction of the left subclavian vein between the clavicle and the first rib (Fig. 1). Echocardiography showed dilatation of the right ventricle and severe tricuspid regurgi-



Fig. 1. Retrograde venography of the left brachiocephalic vein showing complete obstruction.



Fig. 2. Doppler echocardiography revealing a right-to-left shunt (arrow).
RA, right atrium; LA, left atrium.

tation. The maximum pressure gradient between the right ventricle and atrium was 79 mmHg. Moreover, Doppler echocardiography revealed a right-to-left shunt via a patent foramen ovale (Fig. 2). Laboratory tests of coagulation and fibrinolysis were normal. The partial pressure of oxygen in the arterial blood was 95 mmHg on room air. It was postulated that pulmonary embolism resulted from thrombi from the left subclavian vein. This caused pulmonary hypertension, which resulted in a right-to-left shunt via a patent foramen ovale. Subsequently, paradoxical embolism occurred.

We decided to perform surgery to prevent recurrent thromboembolism. The upper half of the sternum was incised in the midline. We exposed the left brachiocephalic vein and identified the position of the left brachiocephalic vein flowing into the superior vena cava. We confirmed that there was no thrombus or venous dilatation in the brachiocephalic vein. Then we ligated the bra-

chiocephalic vein.

Although the circumference of the left arm was increased by 2 cm just after surgery, the left arm returned to its preoperative size 1 week after surgery. The patient was started on anticoagulation therapy with warfarin potassium. No thromboembolic episodes have occurred since the operation. Sixteen months after surgery, echocardiography demonstrated trivial tricuspid regurgitation and the maximum pressure gradient between right ventricle and atrium had decreased to 35 mmHg. Furthermore, shunt flow via the patent foramen ovale disappeared.

Discussion

Paradoxical embolism is arterial embolization caused by venous thrombus through a right-to-left shunt. Deep venous thrombosis in the lower extremities is the major

source of these emboli. However, the origin of the paradoxical cerebral embolus in the present case was a primary subclavian vein thrombosis. Such a source of thromboembolism is very rare.

Deep venous thrombosis occurring in the upper extremities is also rare, accounting for only 2% of all deep venous thromboses.^{1,2)} Although subclavian vein thromboses are often reported to be secondary to trauma or the presence of a central venous catheter, primary subclavian vein thromboses occurs in about 30% of cases.³⁾ Primary subclavian vein thrombosis is known as Paget-Schroetter syndrome, which is a subtype of thoracic outlet syndrome.⁴⁾ Because the subclavian vein runs through the costoclavicular space, the subclavian vein is compressed frequently and repeatedly. Therefore, thrombosis of the subclavian vein is induced by repetitive muscular activity at work or during sports. Our patient was a field hockey player who used his upper extremities repeatedly.

Subclavian vein thrombosis causes pulmonary embolism less frequently than does deep venous thrombosis of the lower extremities.^{5,6)} Paradoxical cerebral infarction caused by Paget-Schroetter syndrome has not previously been reported. Subclavian vein thrombosis is usually treated initially by anticoagulation and thrombolytic therapy. Machleder et al.⁷⁾ reported that local catheter-delivered thrombolytic therapy is effective. These types of conservative treatments can improve symptoms by inducing growth of collateral routes. Because paradoxical cerebral infarction occurred in this case, surgical treatment was considered to be better than conservative treatment. The methods and timing of surgery for the treatment of subclavian vein thrombosis are controversial. Although thrombectomy and veno-venous bypass⁸⁾ are available, those procedures cannot prevent recurrence of thrombosis. With respect to methods to prevent recurrence of thrombosis, rib resection to decompress the costoclavicular space⁹⁾ and venous stenting¹⁰⁾ have been reported. In the present case, it was important to prevent recurrent

paradoxical embolism from the left subclavian thrombus securely. Venography confirmed that the collateral vein was well developed. Therefore, ligation of the left brachiocephalic vein was selected as the treatment. The postoperative course was satisfactory, and the patient is doing well without swelling of the left arm over 3 years after surgery.

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