BRIEF COMMUNICATION

CEREBRAL INFARCTION IN CHILDREN DUE TO THROMBOEMBOLISM FROM LIGATED BLALOCK-TAUSSIG SHUNT

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In recent decades, there has been a marked reduction in surgical mortality for many forms of congenital heart defects. However, neurologic sequelae remain a widely recognized and potentially devastating complication of pediatric cardiac surgery.^{1,2} The best-known cerebral lesions in children with congenital heart defects are caused by thromboembolism from the heart.^{2,3} We describe 3 unique cases of cerebral thromboembolism from a ligated Blalock-Taussig (BT) shunt after definitive repair of congenital heart defects. To our knowledge, no previous reports describe a ligated BT shunt as a potential source of embolism.

Clinical summaries

PATIENT 1. The patient was a girl in whom a diagnosis of tetralogy of Fallot {I,L,L} was made soon after birth. A right classic BT shunt operation was performed at 6 months of age. The diameter of the subclavian artery measured at the BT shunt operation was 4.5 mm. At the age of 6 years, she underwent patch closure of a ventricular septal defect and enlargement of the right ventricular outflow tract. The subclavian artery used for the BT shunt had grown to 9 mm in diameter and was doubly ligated. The patient made an uneventful recovery and was discharged to the ward 2 days after the operation. On postoperative day 12, she had the abrupt onset of headache and severe left hemiplegia. A computed tomographic (CT) scan of the brain revealed an infarct involving the right basal ganglia with brain edema suggesting thromboembolism in the middle cerebral artery. Transthoracic echocardiography did not show any evidence of thrombus formation in the cardiac cavity. She was treated with D-mannitol and dexamethasone. The patient gradually recovered from the hemiplegia. She was transferred to the rehabilitation center 22 days after the development of throm-

bosis. She is currently leading a normal life with slight left hemiplegia 12 years after the operation.

PATIENT 2. The patient was a boy in whom the diagnosis of tetralogy of Fallot {S,L,L} with pulmonary atresia was made soon after birth. A right classic BT shunt operation was performed at 3 months of age. At the age of 9 years, he underwent a Rastelli procedure without correction of the discordant connection. The subclavian artery used for the BT shunt had grown to 15 mm in diameter and was doubly ligated. The patient made an uneventful recovery until experiencing the abrupt onset of severe left hemiplegia and loss of consciousness 19 days after the operation. A CT scan of the brain revealed an infarct involving the right basal ganglia with brain edema suggesting a thromboembolism in the middle cerebral artery. Transthoracic echocardiography showed no evidence of thrombus formation in the cardiac cavity. He was treated with glycerol and heparin. The patient awoke the next day and gradually recovered from the hemiplegia. He was transferred to the rehabilitation center 45 days later. He is currently leading a normal life with slight left hemiplegia 4 years after the operation.

PATIENT 3. The patient was a boy in whom tetralogy of Fallot {S,D,D} with pulmonary atresia and major aortopulmonary collateral arteries had been diagnosed soon after birth. At the age of 1 month, he underwent a right classic BT shunt operation and ligation of the major aortopulmonary collateral artery, followed at the age of 3 months by left unifocalization. At the age of 8 years, he underwent patch closure of a ventricular septal defect and reconstruction of the right ventricular outflow tract with the use of autologous pericardium and a monocuspal valved outflow patch (MVOP; Polystan, Copenhagen, Denmark). The subclavian artery used for the BT shunt had grown to 10 mm in diameter and was doubly ligated. The patient made an uneventful recovery until the abrupt onset of headache and severe left hemiplegia 13 days after the operation. A CT scan of the brain revealed an infarct involving the right basal ganglia with brain edema suggesting a thromboembolism in the middle cerebral artery. Transthoracic echocardiography showed no evidence of thrombus formation in the cardiac cavity. He was treated with glycerol and ticlopidine hydrochloride. The patient gradually recovered from the hemiplegia. Postoperative cardiac catheterization was performed before discharge from the hospital. Angiography demonstrated that contrast material entered the ligated BT shunt, outlining the thrombus (Fig 1). He was transferred to the rehabilitation

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Fig 1. Angiogram in patient 3 demonstrated that contrast material entered the ligated BT shunt outlining the thrombus.

center and is currently leading a normal life with slight left hemiplegia 2 years after the operation.

Discussion. These 3 patients had certain anatomic similarities and similar clinical courses. They underwent definitive repair of tetralogy of Fallot, with ligation of the classic BT shunt. This ligation creates a blind stump that appears to be a potential nidus for thrombus formation. Although recent advances in pediatric cardiac surgery allow early definitive repair in many patients, some patients with complex cardiac anomalies still require a BT shunt, especially in the neonatal and early infancy periods.⁴ The classic BT shunt shows good long-term patency and offers the possibility for growth.⁴ However, ligation of the grown subclavian artery used for the BT shunt creates a large blind sac that may promote thrombus formation, presumably because of the presence of stagnant flow, and thromboembolism in the common carotid artery.

Pediatric cardiac surgery includes a particular risk of cerebral complications.^{1,2} Among these, thromboembolism causes serious problems that can result in a life-threatening complication. Several cases complicated by cerebral infarction due to thromboembolism originating from the heart have been reported.^{3,5} Hana and associates⁵ showed that the patent foramen ovale was closely related to cerebral infarction. Rosenthal and associates³ reported some cases of thrombosis in the pulmonary artery stump after distal ligation of the main

pulmonary artery as part of a Fontan or Glenn procedure. We observed thrombus formation in the stump of the ligated BT shunt by postoperative angiography. To the best of our knowledge, thrombosis in this location has not previously been reported. We believe that this finding has clinical importance for aggressive evaluation of patients with a ligated classic BT shunt, because thrombolytic recanalization of occluded arteries may reduce the degree of injury to the brain if it is done before the process of infarction has been completed.⁶ Recognizing the ligated subclavian artery used for the classic BT shunt as a potential source of thromboembolism also has implications regarding the optimal medical and surgical management during and after definitive repair in such patients. Intensive and earlier anticoagulant therapy, including warfarin sodium and antiplatelet agents, may be needed to prevent this particular complication in these patients. Also, as surgical prophylaxis, the volume of the stump should be reduced as much as possible to decrease the risk of thrombus formation.

In summary, the blind stump of the ligated subclavian artery used for the classic BT shunt can become a source of thromboembolism after definitive repair. To prevent this complication, one should consider adequate postoperative anticoagulant therapy and attempt to reduce the size of the blind stump in such cases.

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