

Tricuspid Pouch can Cause Systemic Embolization in Adulthood

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A 63-year-old man with significant left hemiplegia was admitted to hospital. He had experienced a transient cerebral ischemic attack 10 years ago. Computed tomography revealed hypodensity along the right lateral ventricle, which corresponded to the left paralysis. Echocardiography and left ventricular angiography revealed an aneurysm of the membranous septum (AMS) without a ventricular septal defect (VSD). Therefore, the embolism was thought to be of cardiac origin, but surgery revealed that it was not caused by AMS. The aneurysm was created when the septal leaflet of tricuspid valve formed a giant capsule during the process of natural closure of the VSD. It was a large pouch, 2.0 cm in diameter, adjacent to the septal leaflet. Anomalies of the tricuspid valve, including pouches, can resemble AMS. (Circ J 2006; 70: 631–633)

Key Words: Aneurysm; Cerebral infarction; Embolism; Membranous septum; Tricuspid pouch

The tricuspid pouch is a pouch-like structure that bulges into the right ventricle near the interventricular membranous septum. It is commonly reported at postmortem examination of infants, but is rarely seen in adults. Hamby et al reported aneurysm of the pars membranacea for the first time in 1970.¹ The septal leaflet of the tricuspid valve formed an irregular large capsule during the process of natural closure of a ventricular septal defect (VSD). Aneurysm of the interventricular membranous septum (AMS) is similar to anomalies of the tricuspid valve, including pouches, and misinterpretations appear in the literature. Complications such as rupture, endocarditis or thrombosis are unusual for either a tricuspid pouch or AMS.² We report a patient with a large tricuspid pouch that we thought was AMS until cardiac surgery.

Case Report

A 63-year-old man was admitted to hospital because of left hemiplegia. Motor function of the left side was diminished, possibly as a sequel of a prior neurological accident (transient cerebral ischemic attack) 10 years ago and a more recent stroke. Brain computed tomography revealed a low-density area within the right lateral ventricle, which corresponded to the left paralysis.

Clinical examination revealed blood pressure of 120/70 mmHg, heart rate of 80 beats/min and respiratory rate of 15 breaths/min. Chest X-ray was normal and electrocardiogram (ECG) showed sinus rhythm and left ventricu-

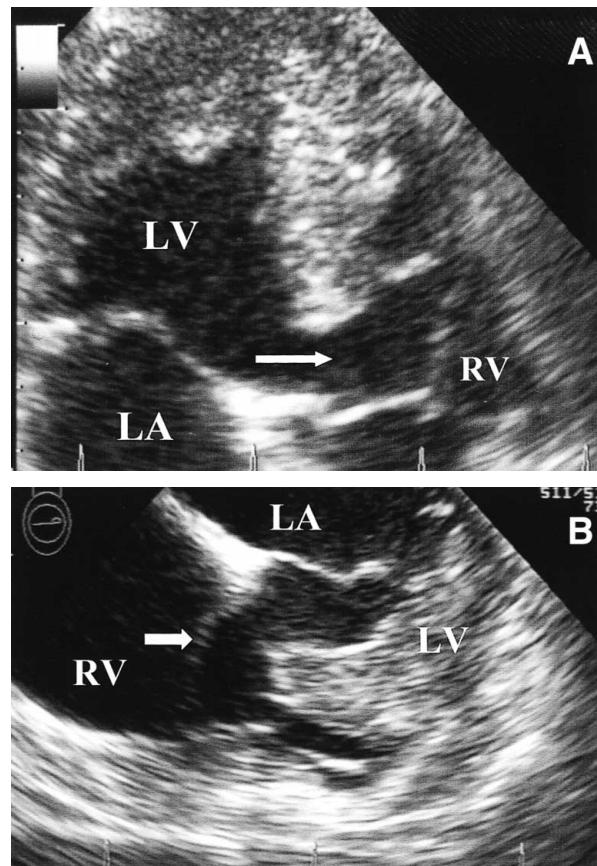


Fig 1. (A,B) Preoperative transthoracic and transesophageal echocardiograms in the 4-chamber view showing the tricuspid pouch at inflow of the right ventricle. LA, left atrium; LV, left ventricle; RV, right ventricle.

(Received November 18, 2005; revised manuscript received February 6, 2006; accepted February 9, 2006)

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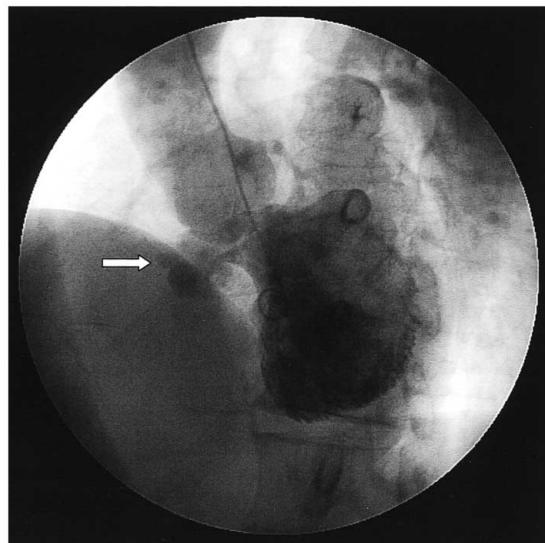


Fig 2. Left ventricular angiography in left anterior oblique view shows a pouch-like structure bulging into the right ventricle during systole.

lar hypertrophic pattern. Transthoracic and transesophageal echocardiography (TEE) revealed an aneurysm protruding into the right ventricle (Fig 1A,B) with eddying of blood on TEE, but we did not recognize the thrombus in the pouch or its mobility. The findings of left ventricular angiography (LVA) in the left anterior oblique projection were similar to those for AMS (Fig 2). It protruded during inflow of the right ventricle and its surface was regular and cauliflower-like. There was no left-to-right shunt flow, no dilatation of the right ventricle and trivial tricuspid regurgitation. We could not confirm if the cerebral thrombosis had a cardiac origin or not, but brain magnetic resonance imaging suggested that the cerebral infarction had been caused by embolism because of the site and the sudden onset of hemiparesis. Therefore, we considered that the protruding aneurysm was the most likely culprit for blood stasis and thrombus formation and that cardiac operation was necessary to prevent stroke attack in the future.

At operation, besides a perimembranous outlet type of VSD, a pouch of 2.0 cm in diameter was found adjacent to the septal leaflet of the tricuspid valve (Figs 3A,B). There was not a residual membranous septum and the surface of right ventricle that was adhered to the septal leaflet was irregular and discolored. The tendon of the tricuspid valve was slightly extended. The VSD was closed with a patch. Based on these findings, we speculated that the tricuspid pouch was formed by the effect of a jet stream through the VSD.

After surgery, anticoagulant therapy was instituted for 3 months and to date the patient is healthy and has not had other neurological events.

Discussion

A pouch-like structure that bulges into the right ventricle on LVA is almost always considered to be AMS, which is a rare congenital lesion that is almost always associated with congenital anomalies such as VSD and endocardial cushion defect.^{3,4} Many investigators have reported the occurrence of AMS^{2,5-7} but its mechanism of development is a matter of argument. Baron et al² reported that AMS is formed by

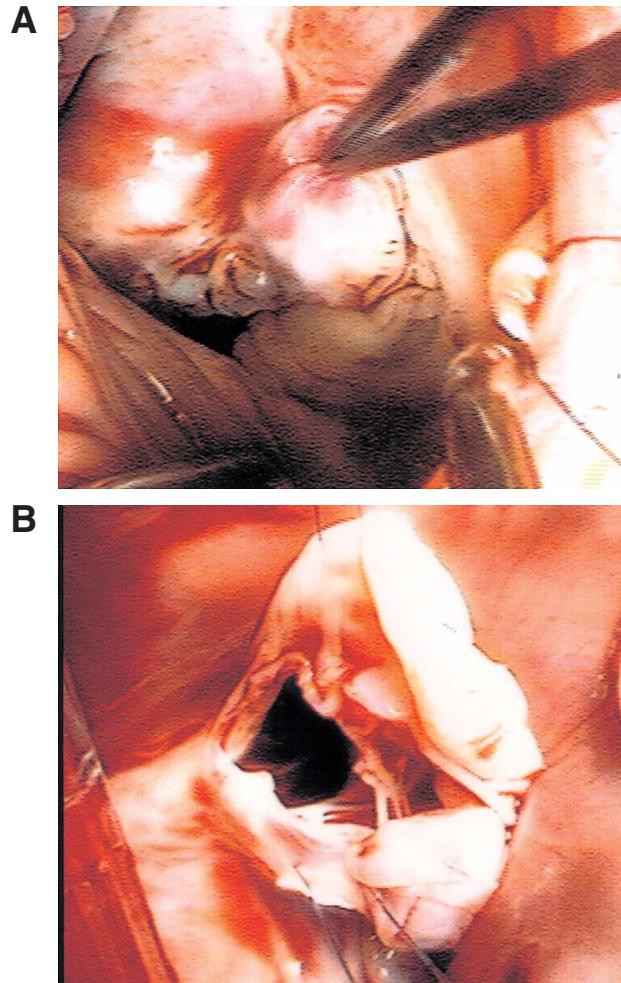


Fig 3. (A) Right atrium is opened to reveal a spherical aneurysmal pouch 2cm in diameter and formed by the adherence of the septal leaflets. (B) Incision of the tricuspid leaflet exposes the true ventricular septal defect.

projection of left ventricular pressure during spontaneous closure of a VSD, which occurs in a significant percentage of patients, usually during infancy. Except for muscular defects, the mechanism of closure is not clearly understood. It may occur as a result of deposition of fibrin over the margins of the defect or by attachment to the septum of the septal leaflet of the tricuspid valve.^{1,6} In the presence of a VSD, left-to-right shunt flow jet toward the tricuspid valve may occur after adhesion of the septal leaflet of the tricuspid valve to the margin of the VSD. But this is not AMS, rather it is a tricuspid pouch. Idriss et al³ reported that tricuspid pouch was occurred in 12.2% of cases of VSD. Tricuspid pouch sometimes causes subpulmonary obstruction in association with transposition of the great arteries, but rarely does so without transposition.⁸ Generally, the development of a tricuspid pouch is secondary to ineffective treatment of endocarditis. However, in the present patient, the surface of the tricuspid valve was smooth and there was no inflammatory change. The pouch was created by a jet lesion of the VSD. Tricuspid pouch can be confused with AMS and both are diagnosed by 2-dimensional echocardiography and LVA, although it is difficult to distinguish them before operation, as in the present case.

The clinical course of most cases of tricuspid pouch is

silent and complications, such as rupture, endocarditis or thrombosis, are unusual. In the present case, the pouch was not associated with interventricular communication and repeated cerebrovascular embolism occurred. The risk of thrombo-embolic complications for AMS and tricuspid pouch is almost same. In cases with a small or negligible VSD, conditions are favorable for blood stasis and thrombus formation. Thrombo-embolic complications with tricuspid pouch are very rare, but may be underestimated, as in the past with aneurysm of the interatrial septum, which was considered to be a potential cause of thrombo-embolism. Previous investigators have reported complications of AMS, such as cerebral infarction⁹⁻¹² and all of these previous cases underwent surgery because there was possibility of thrombus in the future. A report of tricuspid pouch is rare because the final diagnosis of tricuspid pouch is made during cardiac surgery.

Rhythm disturbances have been reported in associated with AMS,¹³ but none of the reported cases of AMS with thrombo-embolism had cardiac arrhythmias,¹² nor was there evidence of atrial fibrillation or ventricular premature contraction on Holter ECG in the present case.

We could not confirm thrombus in the aneurysm, but after the patient had had a transient ischemic attack approximately 10 years ago, he was administered aspirin and nevertheless he had another severe stroke. Surgical investigation for a tricuspid pouch is essential when cerebral embolism occurs despite anticoagulant therapy.

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