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Peratrial Device Closure of a Congenital Coronary Artery Fistula through a Right Parasternal Approach: Innovative Use of Available Technology

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ABSTRACT

Current treatments for congenital coronary artery fistulas (CAFs) include surgical obliteration and transcatheter occlusion. However, surgical techniques involve significant trauma. Transcatheter occlusion is performed under fluoroscopy and angiography, in which radiation injury is inevitable. We present a patient, with a CAF from the left coronary artery to the right atrium, who underwent peratrial device closure of the CAF with a right parasternal approach under transesophageal echocardiography guidance. Complete occlusion was achieved by a symmetric ventricular septal occluder. We suggest that peratrial device closure of a congenital coronary artery fistula through a right parasternal approach may be a safe and effective option.

INTRODUCTION

Congenital coronary artery fistulas (CAFs) are a type of rare congenital cardiovascular anomaly that may result in severe complications, including congestive heart failure, pulmonary hypertension, myocardial ischemia or infarction [Kayalar 2009]. Previously, the most effective treatment for CAFs was surgical closure. Recently, with advances in interventional therapy, transcatheter closure has been predominantly used instead of traditional open surgery because of advantages such as decreased invasiveness and decreased recovery time [Wang 2014]. In this study, unlike the transcatheter closure, a peratrial device closure through a right parasternal approach, which was previously reported to occlude atrial septal defects [Hongxin 2003; Hongxin 2007; Liang 2006], was successfully performed on a CAF.

CASE REPORT

A 3-year-old girl who presented with a heart murmur was admitted to our hospital for further treatment. The patient was asymptomatic, and her blood pressure was 102/56 mmHg. Upon physical examination, a notable grade 3/6 continuous murmur was audible at the left upper sternal border.

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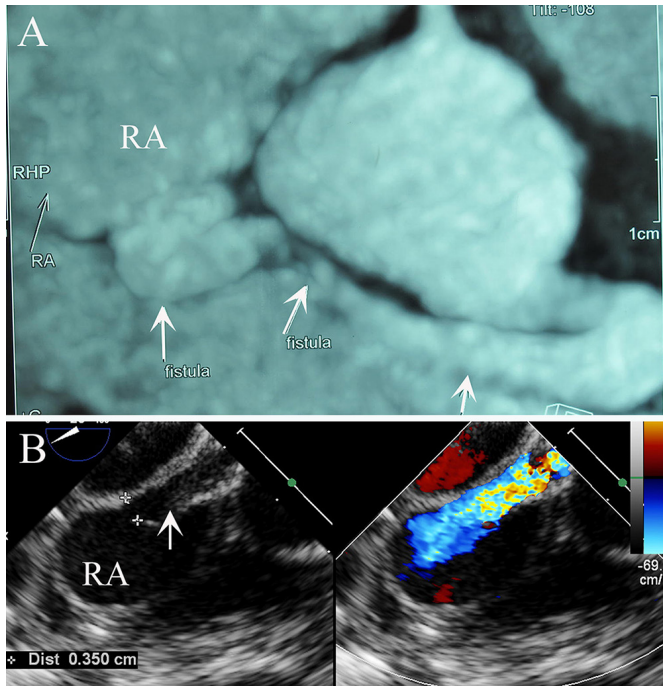

Figure 1. The computed tomography (A) and transesophageal echocardiography (B) revealed a dilated, tortuous left main coronary artery and a coronary fistula drained into the right atrium.

Left ventricular hypertrophy was observed on an electrocardiogram. Computed tomography demonstrated a dilated left main coronary artery and a tortuous fistula with a diameter of 5 to 9 mm (Figure 1, A). Transesophageal echocardiography (TEE) showed a CAF originating from the left coronary artery and draining into the right atrium. The CAF orifice, which was 3.5 mm in diameter (Figure 1, B), was located close and inferior to the superior vena cava.

The procedure was performed under general anesthesia. After intubation, a 1.5-cm parasternal incision was made in the right fourth intercostal space (Figure 2, A). The pericardium was incised and cradled with 4 pericardial stay sutures. Following heparinization (1.0 mg/kg), two purse-string sutures of 5-0 polypropylene (Ethicon, Somerville,NJ) were placed on the lateral wall of the right atrium. A specially designed probe-assisted delivery system (ZL 201010169667.6) was applied during the procedure (Figure 2, B). A right atrial puncture was made within the purse-string sutures, and the hollow probe was inserted. With the outside hole covered,

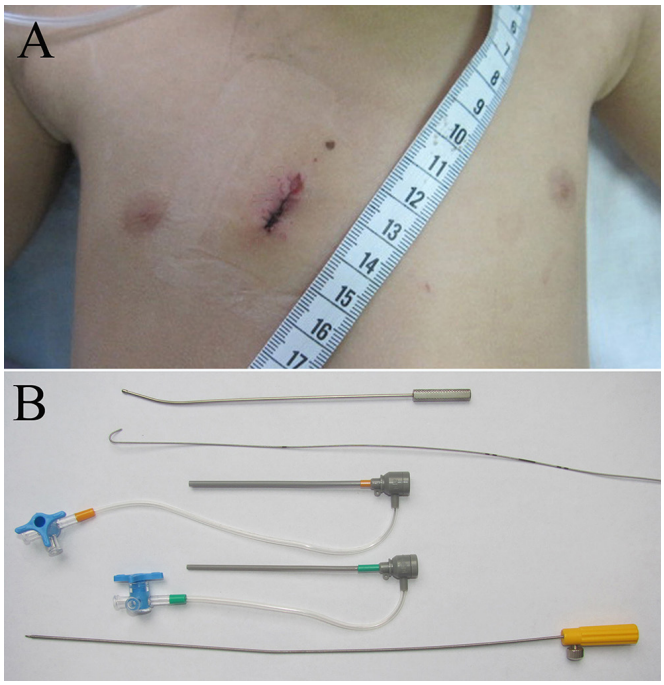

Figure 2. The right parasternal incision (A) and the probe-assisted delivery system (B), which consists of a J-tipped hollow probe, a flexible guidewire, a delivery sheath, a loader sheath and a delivery cable.

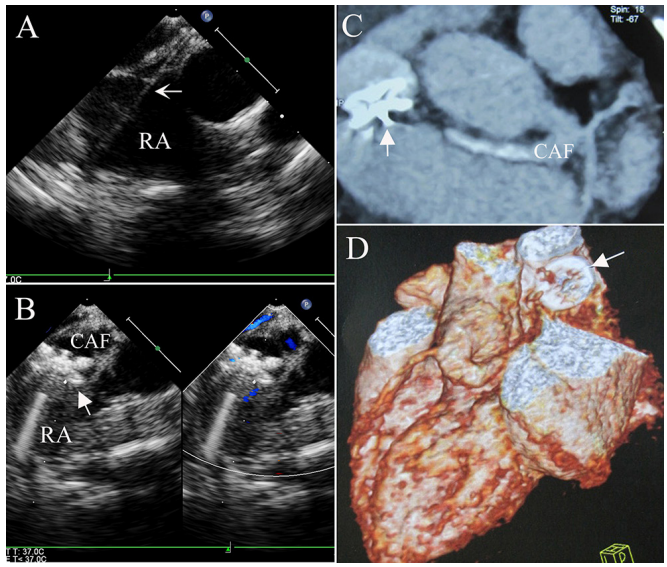

Figure 3. The transesophageal echocardiography (A) showed a flexible guidewire was introduced into the fistulae (arrow), and a symmetric ventricular septal occlude (B) was deployed in place (arrowhead). The follow-up computed tomography scan (C, D) demonstrated the appropriate device position (arrowhead) and the obviously decreased size of the CAF aneurysm (arrowhead). RA indicates right atrium; CAFs, coronary artery fistulas.

the probe was advanced toward the fistula orifice under TEE guidance. The tip of the probe was adjusted to point toward the fistula jet. A flexible guidewire was inserted into the probe channel and advanced into the CAF to establish a delivery pathway. The probe was withdrawn from the heart while maintaining the guidewire in its original position (Figure 3, A). The delivery sheath was introduced into the CAF over the wire and positioned with its tip near the CAF orifice. After an unsuccessful attempt with a cone-shaped ductal occluder, a 10-mm symmetric ventricular septal occluder (Starway Medical Technology, Beijing, China) connected with a device stay suture was chosen and retracted into the loader sheath. Following loader sheath connection, the device was introduced into the delivery sheath and subsequently deployed to occlude the orifice. The device stability was tested by repeatedly performing a "push-and-pull" maneuver. A 15-minute occlusion test was well tolerated; no evidence of myocardial ischemia was observed on a continuous electrocardiogram. Once optimal device position and normal left ventricular function were confirmed, the device was released by disconnecting the delivery cable. Then, the sheath and the device stay suture were withdrawn. The pericardium and the incision were closed in layers without placement of a drainage tube. The intracardiac manipulation time was 50 minutes and the procedural time was 85 minutes.

The patient returned to the intensive care ward in a satisfactory condition and was extubated 2 hours after the procedure. Serial CKMB and troponin levels in the first 48 hours were within normal limits. She recovered uneventfully and was discharged on the fourth postoperative day. A residual shunt was not observed (Figure 3, B). The left ventricular size was notably decreased. Anticoagulation therapy was continued for 6 months with 3 mg/kg of aspirin per day. The patient had echocardiographic, electrocardiographic, radiographic (optional), and CT (optional) follow-up at discharge and 1, 3, 6, 12, and 24 months after the procedure. The transthoracic echocardiography and CT scan demonstrated the appropriate position of the device and the obvious decreased aneurysm size of the CAF (Figure 3, C and D).

DISCUSSION

CAFs are uncommon and mostly asymptomatic. However, their progressive nature and potential complications may be disastrous. Currently, the main available treatment options for CAFs are surgical and transcatheter closure. Surgical closure is generally reserved for single, large, and symptomatic fistula. But it has the disadvantage of significant trauma and complications associated with the sternotomy approach, such as bleeding and an unsightly scar. Transcatheter closure is challenging in small children with low weight or poor vascular access. It also has some limitations such as exposure to medical radiation (for patients and medical workers). Ionizing radiation is associated with a spectrum of malignancy, especially in children who are sensitive to radiation exposure [Kleinerman 2006]. Moreover, this procedure is associated with some potential risks related to manipulation of stabilizing catheters and wires in the coronary vasculature, including coronary artery spasm, vessel injury, ventricular dysrhythmias, and heart perforation [Mangukia 2012].

In this case, a new minimally invasive surgery of peratrial device closure through a right parasternal approach was performed, which has been used for secundum atrial septal defects [Hongxin 2003; Hongxin 2007; Liang 2006] and perimembranous ventricular septal defects [Hongxin 2014], but never used for CAFs. During this procedure, a J-tipped hollow probe, which has been described in previous reports [Hongxin 2014], was used to cross the fistula orifice. The probe allowed the guidewire to be introduced into the CAF to establish a delivery pathway with high efficiency.

Device selection was based on the anatomic features of the fistula. At first, we implanted a duct occluder. But the device was unstable and replaced with a ventricular septal occluder as a result of the small orifice and large ampulla of the CAF. The device stay suture can make it possible to retrieve a suboptimally placed device through a larger delivery sheath and avoids device embolization [Hongxin 2014]. This improves the security of this technique.

The peratrial approach is performed through an intercostal access port without sternotomy, which results in less pain, less invasiveness, and better cosmetic results compared with surgical closure. Moreover, the parasternal approach has no vascular access limit and was performed under TEE guidance without radiation exposure. The short delivery system need not pass through the coronary arteries or any cardiac valve, which further increases the safety of this procedure. The follow-up results demonstrate that this procedure represents a safe and effective alternative therapy for selected patients.

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