An unusual case of giant coronary sinus causing left ventricular inflow obstruction

Deepak Gowda, DNB, C. H. Praveen, MBBS, Aaditya Sirohi, MD, and Neelam Desai, MCh, DNB, Andhra Pradesh, India

From the Department of Cardio Thoracic and Vascular Surgery, Sri Sathya Sai Institute of Higher Medical Sciences, Prasanthigram, Andhra Pradesh, India.

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Address for reprints: Deepak Gowda, DNB, Department of Cardio Thoracic and Vascular Surgery, Sri Sathya Sai Institute of Higher Medical Sciences, Prasanthigram, Room No. 251, Puttaparthy, Anantapur District, 515134 Andhra Pradesh, India (E-mail: drdeepakgowda@gmail.com).

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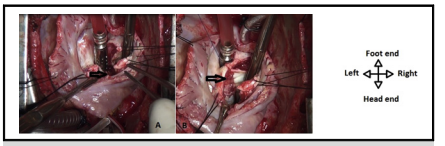


Cut back of coronary sinus (A) and partial plication (B).

Central Message

An unusual case of left ventricular inflow obstruction caused by left superior vena cava draining to the coronary sinus, supramitral ring, and absent right superior vena cava and its successful management.

The left-sided superior vena cava (LSVC) is thought to be a remnant of the embryologic left anterior cardinal vein. Its prevalence has been reported to be approximately 3% in isolation and up to 2.8% to 4.3% in the presence of congenital heart disease.[1] Patients with persistent LSVC and dilated coronary sinus seem to have a higher incidence of associated left-sided obstructive lesions.[2] We describe successful relief of left ventricular inflow obstruction (LVIO) caused by a dilated coronary sinus, which had been misdiagnosed as caused by a supramitral ring.

Persistent left superior vena cava draining to the coronary sinus causes dilation of the coronary sinus. A dilated coronary sinus that is adherent to the thickened posterior wall of the left atrium produces a tubular obstruction at the left ventricular inlet and leads to blood flow across the mitral valve.[2] An echo-free structure in the posterior atrioventricular groove suggests an enlarged coronary sinus, which can be confirmed in a parasternal long-axis view and a modified apical four-chamber view with posterior tilt (Figure 1).

CASE PRESENTATION

A male child aged 3 years and 9 months was admitted with the complaint of recurrent respiratory tract infection. Transthoracic echocardiography revealed a supramitral

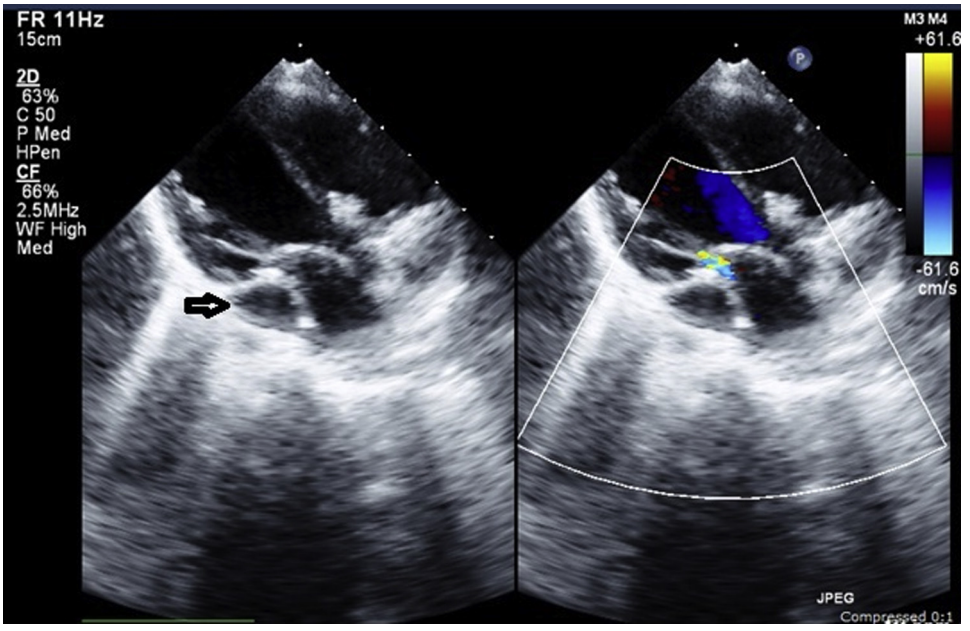


FIGURE 1. Parasternal long-axis view showing a hugely dilated coronary sinus.

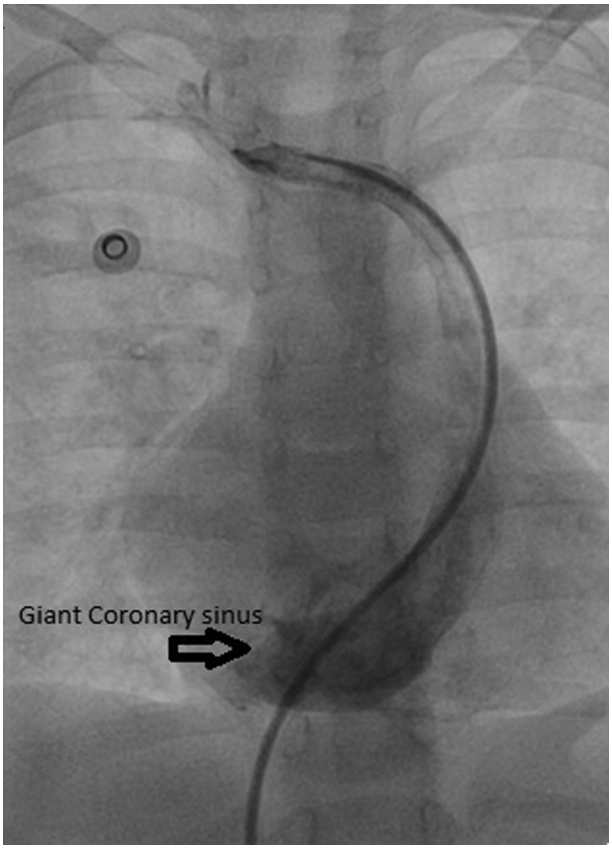
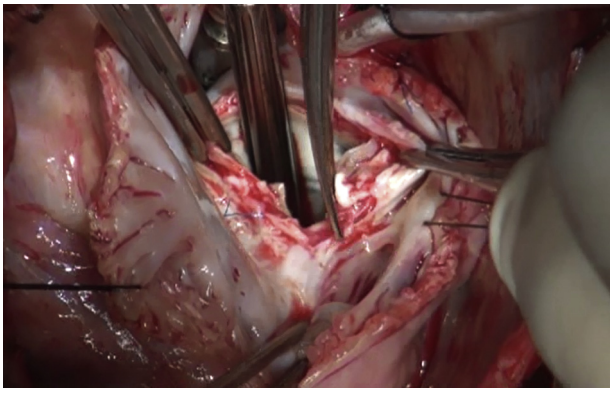


FIGURE 2. Catheterization study showing absent right superior vena cava. Catheter is seen in the left superior vena cava, which is draining into dilated coronary sinus.

ring with a mitral valve gradient of 19/7 mm Hg, a tiny perimembranous ventricular septal defect (PM VSD) restricted by a septal leaflet of the tricuspid valve, and moderate tricuspid regurgitation with severe pulmonary arterial hypertension. The patient underwent cardiac catheterization, which revealed a mean gradient of 16 mm Hg across the left atrium and the left ventricle, an absent right superior vena cava, with persistent large draining of the left superior vena cava to the coronary sinus (Figure 2), and an operable small PM VSD.



VIDEO 1. Video demonstrating the giant coronary sinus causing the LVIO and the reduction plasty of the coronary sinus in process to relieve the LVIO. Video available at http://www.jtcvs.org.

The supramitral ring was excised intraoperatively and the VSD was closed with a Dacron patch. After cardiopulmonary bypass, transesophageal echocardiography showed no residual VSD and no significant gradient across the mitral valve and trivial mitral regurgitation and no tricuspid regurgitation.

The patient gradually developed noncoapting tricuspid valve leaflets with severe tricuspid regurgitation, ascites, hepatomegaly, and moderate mitral regurgitation, and a catheterization study revealed a mean gradient of 10 mm Hg across the left ventricular inflow tract. Because of the child's deteriorating clinical condition, we decided to repair the tricuspid valve. During the surgery, an abnormally giant coronary sinus was present and thought to be the cause of the LVIO. Hegar dilators of appropriate size were passed through the mitral valve, and we then made the decision on the amount of plication of the coronary sinus needed. Partial plication of the dilated coronary sinus was done through the right atrium (Figure 3). The coronary sinus was cut back, as shown in Figure 3, and the cut ends were then plicated, thus reducing the orifice of the coronary sinus, which was the cause of the LVIO (Video 1). The interatrial septum was closed with a Dacron patch,

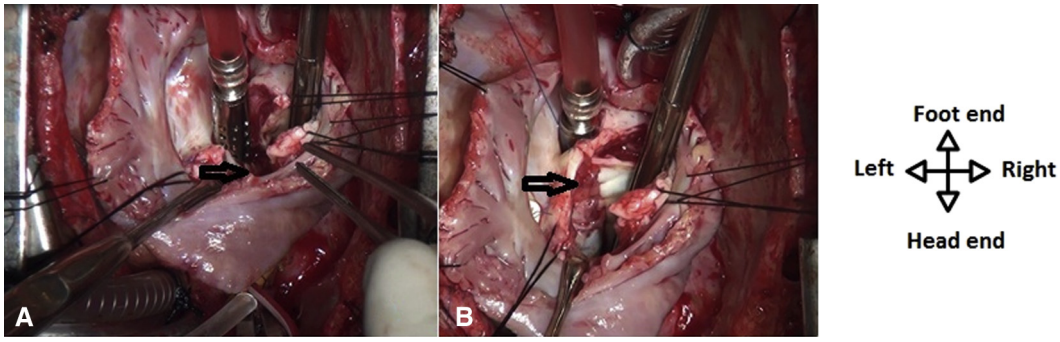


FIGURE 3. Cut back of the coronary sinus (sucker inside the coronary sinus) (A) and plication with the sucker inside the coronary sinus (B).