ABSTRACT

Coronary insufficiency caused by unruptured left sinus of Valsalva aneurysm (SVA) is exceedingly rare in the literature. Herein, we present a successful surgically treated case of giant left SVA with severe aortic regurgitation and coronary insufficiency, thus introducing a tailored valve-sparing aortic root repair technique.

INTRODUCTION

Sinus of Valsalva aneurysm rarely arises from the left coronary sinus as the pulmonary trunk and the right ventricle externally supports the left coronary sinus [Ott 2006]. Isolated left sinus of Valsalva aneurysm (SVA) is sporadically reported and is predominantly congenital, while acquired SVAs are caused by infections, trauma, or degenerative diseases [Martinez-Comendador 2013]. Notably, when the aneurysm enlarges, it causes mitral valve obstruction or insufficiency, coronary insufficiency, and aortic valve regurgitation [Cho 2013; Abe 2012]. For this reason, operative repair of the SVA is usually required. Herein we present a case of giant left SVA causing severe aortic regurgitation (AR) and coronary insufficiency, which was successfully managed using the limited aortic root remodeling operation informally known as “uni-Yacoub” [Chiu 2016].

CASE REPORT

A 56-year-old woman with progressive shortness of breath for 2 years was admitted to our institution. Physical examination revealed a 3/6 diastolic murmur at the 3rd intercostal space of the left sternal border. Electrocardiogram exhibited normal sinus rhythm, while the chest roentgenogram showed a protrusion of the left superior cardiac border (Figure 1A, arrow). Transthoracic echocardiography showed severe AR and an isolated left SVA with a size of 33 mm × 79 mm × 52 mm (Figure 1B, arrow). The isolated left SVA was further confirmed by cardiac computed tomography angiography (CTA), which also showed an abnormal origination of the left main coronary artery (Figure 1C and 1D).

The aorta was transected 1 cm above the sinotubular junction (STJ) following cardiopulmonary bypass initiation and cardioplegia administration through a median sternotomy. The aortic leaflets were normal, although the aortic annulus measured 24 mm in diameter with evident stenosis of the left coronary artery (Figure 2A). A tongue-shaped Dacron patch with its fabric corrugation oriented longitudinally was tailored to the SVA size, although slightly larger than its orifice.

After aneurysm excision (Figure 2B), the aneurysm remnant and the patch’s proximal end were anastomosed to the aortic annulus using 4-0 polypropylene sutures starting...
from the nadir of the left sinus towards an area above the commissure (Figure 2C) and reinforced using a pericardial patch. Each suture’s specific width was about 1-2 mm smaller on the native side than the prosthesis side to make a bulge. The left cusp was noted to have an elongated free edge and prolapsed under static pressure testing. The cusp was plicated using a 6-0 polypropylene suture near the left coronary-noncoronary commissure to achieve aortic valve competency. A proximal anastomosis of the 26 mm tubular Gelweave Valsalva graft to the reconstructed aortic root was then performed and reinforced. The aortic valve’s competency was reassessed, and the Dacron graft was tailored to the appropriate length and anastomosed distally. We then constructed a coronary artery bypass graft (CABG) with the great saphenous vein connecting the innominate artery to the proximal left anterior descending coronary artery (Figure 2D). A coronary artery bypass graft (CABG) with the great saphenous vein connecting the innominate artery to the proximal left anterior descending coronary artery was also performed.

**DISCUSSION**

SVA, a rare cardiac condition commonly arising in the right coronary sinus and non-coronary sinus, is thought to occur due to the incomplete fusion of the distal bulbar septum and truncal ridges. The left SVA is exceedingly rare and seen in <1% of all sinus of SVAs [Feldman 2006]. The incidence rate discrepancies are primarily attributed to the various anatomic asymmetries concerning stress and strain variations. A three-dimensional finite-element model [Grande 1998] showed that stress in the right coronary sinus and non-coronary were 21% and 10% greater than the left sinus. Significantly, the left coronary ostium provides diastolic pressure relief. This inherent nature of aortic leaflet and sinus could help us better understand the pathogenesis of left SVA and the low incidence of its rupture from a biomechanical perspective.

Compared with non-coronary or right SVAs presenting between 20 to 40 years of age [Feldman 2006], left SVAs are commonly reported in older adults [Martinez-Comendador 2013; Jiang 2017; Pedroza 2018], consistent with the present reported case (sixth decade of life). The significant age difference was primarily attributed to the distinct disease manifestation and progression patterns. Right and non-coronary SVA were mostly diagnosed upon the onset of a heart murmur, acute chest pain, dyspnea, and heart failure caused by sudden SVAs rupture [Feldman 2006]. On the other hand, the unruptured left SVA related pathophysiologic processes, such as AR2 and compression of the coronary artery [Martinez-Comendador 2013; Jiang 2017], were prone to chronic phenotype and delayed diagnosis due to slowly progressive mass effect.

Thus, our experience shows that giant left SVA should be included in the differential diagnosis once a paracardiac space-occupying lesion is accidentally found on a chest X-ray. Suppose SVA is suspected in a patient at low risk for coronary artery disease; in that case, TA is recommended as a precaution against possible iatrogenic injury caused by invasive coronary angiography, especially in large SVA patients whose coronaries may be hard to access. Notably, AR does not directly occur secondary to an unruptured left SVA. Thus, as the aneurysm expands, a distal displacement of the commissure or even STJ dilation occurs, leading to coaptation failure, causing AR and subsequent secondary change in the cuspal tissue such as stress fenestrations [David 2016]. In the present study, the coronary arterial flow was always hindered by the large left SVA. Compression and elongated distortion of the left main coronary artery were primarily caused by a prominent mass effect or intramural course of the vessel [Martinez-Comendador 2013; Jiang 2017; Pedroza 2018].
Valve sparing patch repair [Jiang 2017] modified reimplantation technique [Pedroza 2018] and composite graft aortic root replacement [Martinez-Comendador 2013] had been reportedly used to treat left SVA. Nonetheless, the mentioned techniques depend on the aortic valve’s reparable possibilities; in that case, there was a concomitant CABG procedure [Martinez-Comendador 2013; Jiang 2017; Pedroza 2018]. Notwithstanding, we carried out CABG instead of direct coronary reimplantation due to the challenging anastomosing tension and unpredictable coronary patency restoration.

The remodeling technique is advantageous for older patients with a degenerative aneurysm and a non-dilated annulus [David 2016]. Significantly, the remodeling operation appears to retain aortic root dynamism, and restore near-normal geometry and flow characteristics [Yacoub 2018]. Considering there is a mismatch between the asymmetrical size of the patient’s sinuses and the commercial symmetrical sinuses of the vascular graft, tailored remodeling of the aortic root is ideal for isolated left SVA versus reimplantation technique, which is more suitable in a condition where all three aortic sinuses must be replaced [David 2016].

Our method of inverting the Dacron pleats’ direction to generate a tailored sinus that would expand horizontally in response to the dynamic stress recreated an individualized left neo-sinus configuration. A physiology was assembled mimicking root environment to resuspend the aortic leaflets. Moreover, we reconstructed a neo-STJ within the suture line between the aortic root and the Dacron graft tube [Urbanski 2012]. The reinforced felt strip was for ensuring hemostasis and supporting the root to prevent further dilatation. Our modified remodeling technique included aortic root repair, aortic valve repair, STJ reconstruction, ascending aorta replacement, and CABG.

Herein, we described the successful experience of individualized treatment of an unruptured left SVA, causing severe AR. Valve sparing aortic root repair procedure was safe and effective in the short-term follow-up. To our knowledge, using a modified remodeling technique for the repair of a giant left SVA is the first to be reported in the literature.

REFERENCES


