Case report - Congenital

Surgical repair of aortico-left ventricular tunnel arising from the left aortic sinus

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Abstract

Aortico-left ventricular tunnel is a rare congenital cardiac defect, which bypasses the aortic valve via the paravalvar connection from the left ventricle to the aorta. In most of the cases, the tunnel arises from the right aortic sinus. We herein report a case of aortico-left ventricular tunnel, of which the aortic orifice was arising from the left aortic sinus, requiring special attention for avoiding left coronary artery injury at the time of surgical repair.

Keywords: Aortico-left ventricular tunnel; Aortic insufficiency; Coronary artery

1. Introduction

Aortico-left ventricular tunnel (ALVT) is a rare congenital heart anomaly presenting paravalvar communication between the ascending aorta and the left ventricle, for which surgical closure has been recommended at the time of diagnosis due to inadequacy of medical therapy. Since the first surgical treatment reported by Levy and colleagues in 1963 [1], approximately 100 cases of ALVT were reported. According to the literature review [2–4], the aortic orifice is mostly opened from the right coronary sinus, and the right coronary artery is occasionally involved in the tunnel requiring special consideration for surgical repair. We report herein an extremely rare case of ALVT in which the aortic orifice arose from the left sinus requiring special care for avoiding left coronary artery injury at the time of surgical repair.

2. Clinical summary

A 10-month-old baby was admitted for the diagnosis of severe aortic regurgitation. Echocardiography demonstrated trivial regurgitant flow through aortic valve and a tubular communication between the ascending aorta and the left ventricle bypassing the aortic valve showing massive regurgitate flow (Fig. 1a,b). The left ventricle was moderately dilated, but ventricular function was preserved with a fraction shortening of 39%. Cardiac catheterization confirmed a runoff of contrast material from the left ventricular outflow tract through a tunnel into the ascending aorta (Fig. 1c).

An elective operation was performed with cardiopulmonary bypass and moderate hypothermia (27°). After the aorta was cross-clamped and incised horizontally, inspection from inside and outside of the aortic wall revealed that the tunnel was located on the posterior aspect and leftwards of the aorta close to the left coronary artery (Fig. 2a). The myocardial protection was achieved by selective administration of cold crystalloid cardioplegia. The aortic opening of the tunnel arose from the left coronary sinus at the level of sinotubular junction (Fig. 2b), and the tunnel lay between the left coronary artery and the ascending aorta. As the aortic orifice was slit-like and thick, primary closure of the aortic orifice was performed with extra care so as not to injure the left coronary artery (Fig. 2c). Then, plication of the tunnel was added (Fig. 2d). After closure of the aortic incision and declamping the aorta, the patient was weaned from cardiopulmonary bypass without any difficulty. Intra-operative trans-esophageal echocardiography showed complete closure of the tunnel with no aortic valve regurgitation. The patient was discharged home on the 6th postoperative day, and trans-thoracic echocardiography at three months of follow-up demonstrated trivial aortic insufficiency.

3. Discussion

The incidence of ALVT is low. Martins et al. reported it was 0.001% in patients with congenital heart disease at their institution experience [2]. There were approximately 100 patients with ALVT reported who received surgical
treatment. In most of the cases, the aortic orifice of the tunnel arose from or above the right coronary sinus, and the tunnel was located anterolaterally to the ascending aorta. However, our case demonstrated that the aortic orifice arose from the left coronary sinus and the tunnel lay posterolaterally to the ascending aorta. We could find only three reports in which the aortic orifice is opened from the left aortic sinus [5–7]. In this situation, incision of the tunnel is difficult and the tunnel should be closed through aortic incision.

As for the operative technique, Serino and colleagues [8] report that closing the aortic defect by direct suture distorts the cusps by pulling them toward the weak aortic wall, which remains unsupported within the dilated aortic sinus. From this point of view, the patch technique is believed to reduce that risk. Our case demonstrated a slit-like opening at the aortic end with no valve distortion. Then, primary closure of the aortic orifice was performed in addition to plication (obliteration) of the tunnel. The short-term result is satisfactory, but late development of severe aortic regurgitation remains a matter of concern [9]. Careful observation is needed in this anomaly after the surgical repair.

References


eComment: Congenital aortico-left ventricular tunnel: anatomic variations and surgical experience

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Congenital aortico-left ventricular tunnel (ALVT) is a rare cardiovascular anomaly, and the presented case with aortic orifice being located in the left sinus of Valsalva is only the fifth description of such variation of ALVT in world literature; three cases are mentioned in the paper [1], one case is described by the authors themselves, and one case was found in German-language literature [2].

We congratulate the authors for precise diagnostics and successful correction of this extremely rare congenital heart defect.

In connection with this case we would like to make some comments concerning the classification of ALVT. In 1988, H. Hovaguimian et al. [3] suggested a new surgical classification of ALVT, which was subsequently adopted by most cardiac surgeons and pediatric cardiologists [4]. In our view it would be reasonable, along with the identification of ALVT type in accordance with this classification, to precise the aortic sinus where the tunnel’s orifice is located. Surgical significance of this precision is obvious, as in cases with the orifice location in the right aortic sinus the ALVT passes anterior to the ascending aorta and can be corrected from the approach through the tunnels’ wall. On the contrary, if the orifice is located in the left aortic sinus, the ALVT passes posteriorly and laterally to the ascending aorta. In such rare cases the section of the tunnel’s wall is difficult and aortotomy is the most optimal approach for the correction of ALVT.

Fourteen patients with ALVT have been operated on at the Bakoulev Center for Cardiovascular Surgery Russian Academy of Medical Sciences. The
patients’ age varied from 1 month to 43 years, there were 11 males. In 71% (10/14) of the cases the following concomitant defects were revealed: aortic valvular insufficiency (n=5, in 4 of them – associated with the ascending aortic aneurysm), the atresia of the right coronary artery ostium (n=2), PDA (n=2) and VSD (n=1). In two out of 14 (14.3%) cases the aortic orifice of ALVT was located in the left sinus of Valsalva. In one patient (a boy aged 11 years 7 months) the ostium of the right coronary artery was absent, in another (a girl aged 8 years 2 months) there was a severe aortic valvular insufficiency. Plastic closure of the aortic orifice with a patch was performed in 10 patients (in one case in association with aortic valve replacement). In three out of 10 cases the left ventricular orifice of ALVT was closed by a second patch. Aortic orifice of ALVT was sutured in four patients: in association with aortic valve and ascending aorta replacement (n=2), in association with aortic valve replacement (n=1) and in association with VSD closure (n=1). Hospital mortality in our series was 0%. One infant with recanalization of ALVT underwent re-intervention for the closure of ALVT fistula on day 6 after the first operation.

References