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Aortico-Left Ventricular Tunnel: A Case Report

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Abstract **A 12 year-old girl who underwent PDA ligation since she was 1 year old came to the hospital with a problem of easy tiredness. After surgery, there was no residual PDA but mild aortic insufficiency. One year previously, she complained of easily getting tired and experienced occasional abnormal heartbeat. On physical examination, diastolic murmur was detected at the left parasternal border. Echocardiogram demonstrated prolapsed aortic valve cusps and moderate regurgitation with diastolic reversal flow in the descending aorta. The presence of perimembranous VSD was suspected but completely occluded by the prolapsed right coronary cusp. The regurgitant flow of aortic insufficiency was eccentric and a tunnel was demonstrated. Therefore, Aortic-Left Ventricular Tunnel (ALVT) was diagnosed. Because of the progressive course of aortic insufficiency and larger size of LV than in normal population, correction of aortic insufficiency was proposed and performed.**

She underwent surgery with standard aortic cannulation, single venous cannula and LV vent. All orifices, coronary ostia and the aortic end of the tunnel were probed. The aortic end of the tunnel was closed with a piece of autologous treated pericardium. The right coronary cusp was minimally prolapsed and no subaortic VSD was detected. Thus, no other intervention was done. Intraoperative transesophageal echocardiogram showed no patency of the tunnel which was closed at the aortic end and no aortic regurgitation but prolapse of the right cusp was still detected.

Postoperatively, transthoracic echocardiogram showed no flow in the tunnel. At 6 months postoperatively, the patient was doing well and could attend full-scale school activities. No abnormal heartbeat was detected.

INTRODUCTION

Aortico-Left Ventricular Tunnel (ALVT) is a tunnel connecting between the aorta and the left ventricle. It causes blood flows through it in the diastolic period, causing aortic regurgitation. ALVT is subdivided into 4 types.[1] Large percentage of patients presented with congestive heart failure and almost half of them were asymptomatic.[2] ALVT is a rare cause of aortic regurgitation.[3] We report a case of such condition at Ramathibodi Hospital.

CASE REPORT

The patient was a 12 year-old girl who came to the hospital with a problem of easy tiredness. She underwent PDA ligation since she was 1 year old. After the procedure, early diastolic murmur was detected over the left parasternal border. Conclusively, there was no residual PDA but mild aortic insufficiency. Because of the mild degree of regurgitation and being asymptomatic, she had been followed at our cardiology clinic. She could attend school classes and social

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activities very well. However, one year previously, she complained of getting tired easily and experienced occasional abnormal heartbeat. Her current medication was Enalapril.

By physical examination, her pulse was regular and not bounding. There was no heaving of the ventricles. S1 and S2 were normal. However, there was a diastolic murmur at the left parasternal border. Other parts were normal.

Her chest film and ECG showed some abnormalities (Figure 1, 2). Echocardiogram demonstrated prolapsed aortic valve cusps and moderate regurgitation with diastolic reversal flow in the descending aorta. The presence of perimembranous VSD was suspected but completely occluded by the prolapsed right coronary cusp. The regurgitant flow of aortic insufficiency was eccentric and a tunnel was demonstrated. Therefore, ALVT was diagnosed. Cardiac catheterization was not performed since the detailed information was considered adequate.

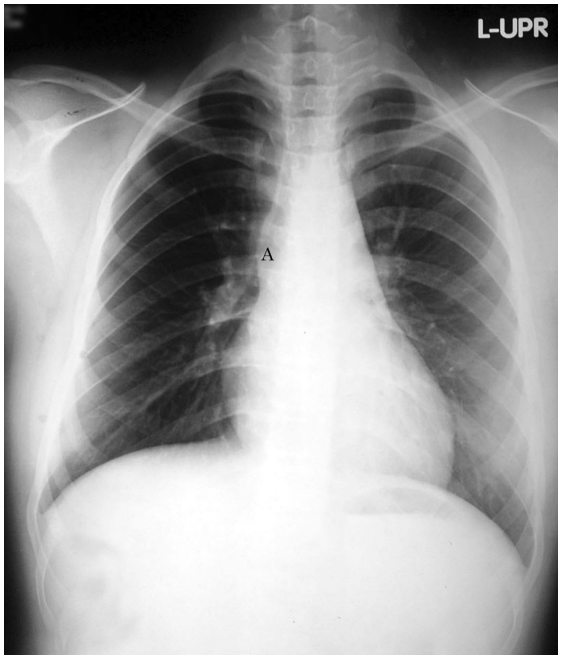

Figure 1 Note the bulging shadow of ascending aorta (A)

Her LV diastolic diameter was 5.6 cm which is higher than expected in normal population, based on Feigenbaum in 1993[4] (normal value should be 4.0 cm for her body size). Because of the progressive course of aortic insufficiency and larger size of LV than in normal population, correction of aortic insufficiency was proposed to her parents. An informed consent was obtained.

Corrective Procedure

Before opening her chest, intraoperative transesophageal echocardiogram was performed to look for the VSD which was not identified preoperatively and no VSD was found. She underwent surgery with standard aortic cannulation, single venous cannula and LV vent. Cardiopulmonary bypass was performed at 28 °C. Myocardial protection strategies were direct-antegrade cold blood cardioplegia and topical hypothermia. Aortic clamp time was 46 minutes and cardiopulmonary bypass time was 62 minutes. All orifices, coronary ostia and the aortic end of the tunnel were probed (Figure 3, 4). The aortic end of the tunnel was closed with a piece of autologous treated pericardium. The right coronary cusp was minimally

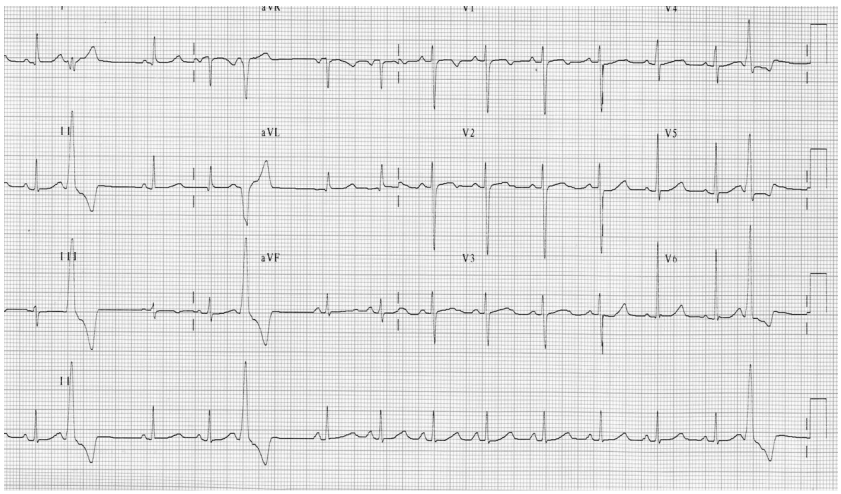

Figure 2 Preoperative ECG showing frequent PVCs

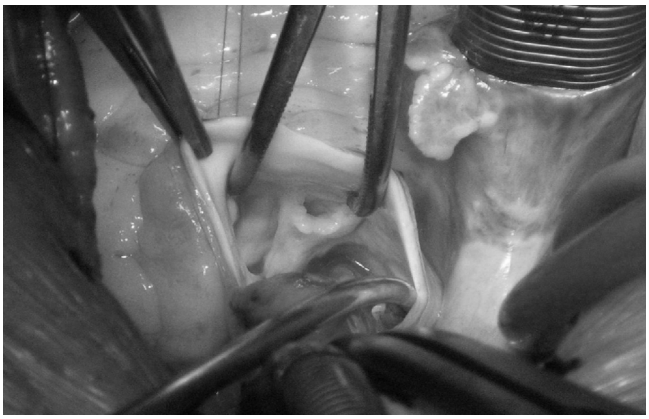

Figure 3 After opening the aorta, a hole (aortic end) just above the sinotubular junction is seen

Figure 4 The tunnel being probed from the aortic end down to the ventricular end

prolapsed and no subaortic VSD was detected. Thus, no other intervention was done. Intraoperative transesophageal echocardiogram showed no patency of the tunnel which was closed at the aortic end and no aortic regurgitation, but prolapsed of the right cusp was still detected.

After surgery, the patient was transferred to ICU and transferred back to ward on the next day. Her total hospital stay was 7 days. Postoperatively, transthoracic echocardiogram showed no flow in the tunnel. At 6 months postoperatively, she was doing well and could attend full-scale school activities. No abnormal heartbeat was detected anymore.

Discussion

Causes of aortic regurgitation are several. However, focusing on pediatric population, most of the time, it is because of congenital malformation in origin. ALVT is one of those that can cause aortic regurgitation. Due to its rarity, each series reported only a small number of patients. No matter what causes aortic insufficiency, significant degree of insufficiency leads to dilatation of the left ventricle and heart failure eventually. Therefore, fixing the culprit lesion that causes aortic regurgitation should be done before left ventricle is damaged.

Fixing aortic regurgitation in children is unique. In adult population, most of the times aortic regurgitation can be fixed with aortic valve replacement and less commonly, aortic root surgery. In children their aortic annulus is smaller than in adult and this could result in a more complex surgery, ie. aortic root enlargement or Ross Procedure or combination of many operations.

ALVT is a tunnel connecting between the aorta and the left ventricle. It causes blood flows through it in the diastolic period, causing aortic regurgitation. In a collective review by Hovaguimian, et al. in 1988, ALVT is subdivided into 4 types.[1] Our patient was classified as type II. A single largest series of ALVT from Boston's Children Hospital published in 2004[2] reported 11 cases of ALVT from their 35-year period. Large percentage (6/11) of patients presented with congestive heart failure and almost half of them (5/11) were asymptomatic. Interestingly, they reported a case of spontaneous closure.

Repairing the ALVT is quite straight forward, ie. tunnel closure at the aortic end. Good long-term results of the operation could be expected.[2,5,6] However, this lesion is associated with other anomalies, most of them are problems of the left ventricular outflow tract, as reported by Hovaguimian, et al.[1] and Martins, et al.[2] The patient in this report has no other aortic root abnormality but persistent ductus arteriosus and prolapsed right coronary cusp of the aortic valve which need further follow up.

Conclusions

ALVT is a rare cause of aortic regurgitation. Corrective procedure is quite simple, however, there are some anomalies which are associated with it. Careful follow-up is suggested.

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