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Late Presentation of Cor Triatriatum Sinister in Adulthood: Successful Surgical Treatment of Two Cases

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Abstract | **Case Report**

Cor triatriatum sinister is a rare congenital malformation, and its diagnosis in adulthood remains exceptional. Delayed recognition is often due to the silent evolution of non-obstructive forms, compounded in low-resource countries by limited access to advanced imaging. We report two adult female patients presenting with exertional dyspnea in whom cor triatriatum sinister associated with an atrial septal defect was diagnosed. Both underwent surgical correction. The postoperative course was uneventful, with no recurrence or complications over a 36-month follow-up. These cases illustrate the feasibility and effectiveness of surgical management for this rare anomaly in sub-Saharan Africa.

Keywords: Cor triatriatum, Atrial septal defect, Adult congenital heart disease, Cardiac surgery, Sub-Saharan Africa.

INTRODUCTION

Cor triatriatum sinister, also known as divided left atrium, is a rare congenital anomaly accounting for 0.1% to 0.4% of congenital heart diseases. It is characterized by the abnormal persistence of a fibromuscular membrane within the left atrium, which creates an obstruction to left ventricular filling. This anomaly is frequently associated with other congenital cardiac defects [1]. Although typically diagnosed in childhood, non-obstructive forms may remain asymptomatic and be revealed only in adulthood [2]. In low-resource countries, the diagnostic delay is exacerbated by limited access to advanced cardiac imaging. We report two cases of cor triatriatum sinister diagnosed in adulthood and successfully managed surgically in Benin.

CASE PRESENTATION

Case 1

A 50-year-old woman with no prior cardiovascular history presented with presyncopal episodes and exertional dyspnea (NYHA class III) evolving over three months. Electrocardiogram (ECG) revealed a regular sinus rhythm, right ventricular overload, and incomplete right bundle branch block. Transthoracic echocardiography (TTE) showed a high-positioned ostium secundum atrial septal defect (ASD) measuring 14 mm with a left-to-right shunt, marked right heart chamber dilatation (right atrial area: 42 cm²), and a membrane dividing the left atrium (Figure 1). The left ventricle was non-dilated, non-hypertrophied, with preserved ejection fraction. Severe tricuspid regurgitation and a dilated main pulmonary artery (47 mm) were observed, with an estimated systolic pulmonary artery pressure (sPAP) of 62 mmHg. Transesophageal echocardiography (TEE) confirmed these findings. The patient underwent surgery via median sternotomy with cardiopulmonary bypass (CPB) established between the aorta and both venae cavae. The procedure included ASD closure using a pericardial patch, complete resection of the left atrial fibromuscular membrane, and tricuspid annuloplasty using a modified De Vega technique reinforced with three pledgeted sutures (figure 2). CPB and aortic cross-clamp times were 97 and 60 minutes, respectively. Postoperative TEE confirmed a watertight ASD closure and an effective tricuspid repair. The postoperative course was uneventful, with no complications or recurrence during 36 months of follow-up.

Case 2

A 42-year-old woman with no identifiable cardiovascular risk factors presented with NYHA class II exertional dyspnea of six months' duration, accompanied by palpitations and precordial chest pain. ECG showed regular sinus rhythm, biventricular overload, and incomplete right bundle branch block. TTE revealed a 15 mm ostium secundum ASD with left-to-right shunt, moderate right heart dilation (right atrial area: 25 cm^2), and left atrial fibromuscular membrane. The left ventricle was dilated and hypertrophied with preserved left ventricular ejection fraction. There was marked dilatation of the main and branch pulmonary arteries, pulmonary regurgitation, and an estimated sPAP of 132 mmHg. Significant tricuspid regurgitation was also noted. TEE confirmed all findings. Surgery was performed via median sternotomy under aorto-bicaval CPB. The intervention included ASD closure using a Dacron patch, complete resection of the left atrial fibromuscular membrane, and tricuspid annuloplasty with a Sovering Band No. 32 ring. CPB and aortic cross-clamp times were 51 and 26 minutes, respectively. Postoperative TEE showed complete ASD closure with no residual tricuspid regurgitation. The postoperative course was favorable, with no complications and no recurrence at 36 months' follow-up.

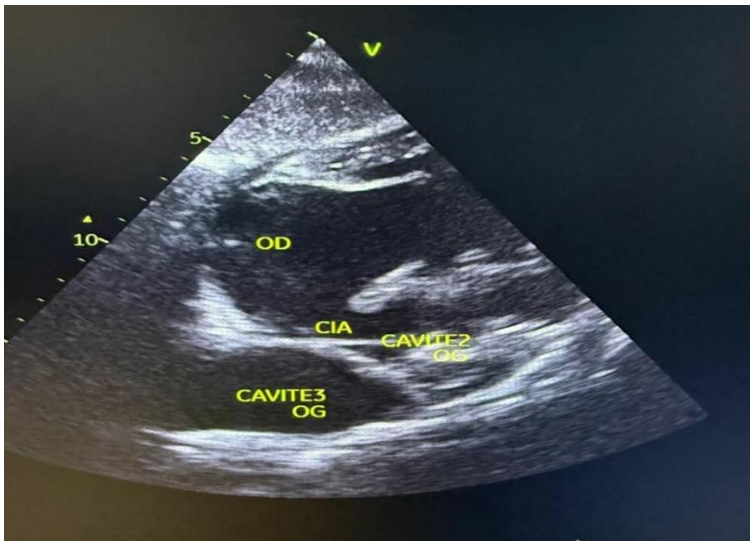

Figure 1: Subcostal transthoracic echocardiographic view demonstrating a cor triatriatum sinister and a secundum-type ASD (atrial septal defect)

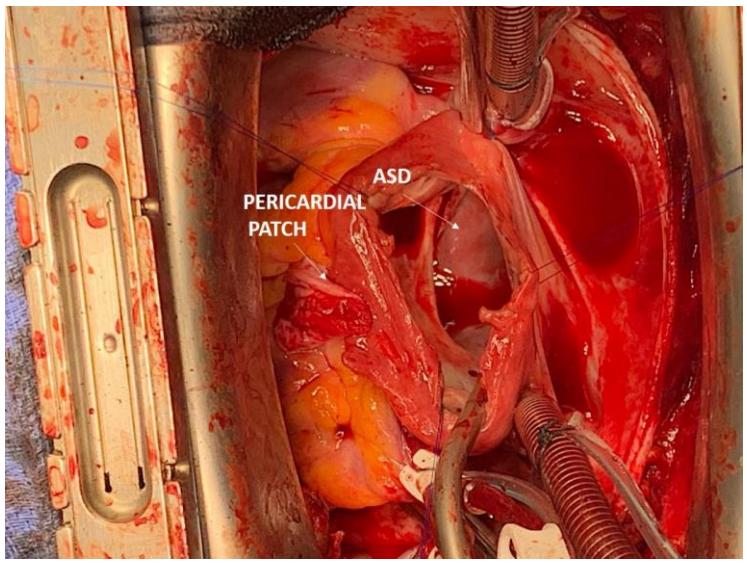

Figure 2: Intraoperative view of ASD (atrial septal defect) closure using a pericardial patch

DISCUSSION

Cor triatriatum sinister is a rare congenital cardiac anomaly. Its pathophysiology remains unclear. Unlike cor triatriatum dexter, which results from persistence of the right sinus venosus valve, no definitive embryological mechanism has been established to explain the formation of the left atrial membrane [2].

Clinical presentation depends on the degree of interatrial obstruction. Severe obstructive forms typically present in the neonatal period with cardiogenic shock, pulmonary edema, and respiratory distress. In contrast, non-obstructive variants may remain asymptomatic into adulthood [2]. Our cases illustrate this late presentation, consistent with the literature, which reports a median age of diagnosis around 43 years in adults. No sex predominance has been established [2,3].

Diagnosis is primarily based on transthoracic echocardiography and may be supplemented by TEE or 3D imaging for better visualization of the left atrial fibromuscular membrane [2]. In approximately one-third to three-quarters of cases, this anomaly is associated with other congenital heart defects. In adults, the most common associated anomaly is ASD, followed by persistent left superior vena cava [2]. Both of our patients had an ostium secundum ASD, consistent with this association.

Medical treatment is reserved for asymptomatic non-obstructive forms under close echocardiographic monitoring [2]. Surgery remains the gold standard for symptomatic forms [2,4]. Surgery involves complete resection of the left atrial fibromuscular membrane to restore free pulmonary venous return, along with correction of associated anomalies. Minimally invasive alternatives, such as transcatheter dilatation, hybrid procedures, and robotic techniques, have been reported [2,3,5,6]. A systematic review of 171 adult cases of cor triatriatum sinister found that 41.5% underwent curative treatment (mostly surgical), 45.6% required no intervention, and 7% refused treatment [2,4,7].

Postoperative prognosis is generally favorable, with reported operative mortality ranging from 0% to 4%, depending on the complexity of associated anomalies, degree of pulmonary hypertension, and timing of intervention [2,8]. Reintervention is rare, typically indicated for recurrent pulmonary vein stenosis, residual ASD, or persistent left atrial obstruction [2,8,9].

Conclusion

Cor triatriatum sinister remains a rare congenital cardiac anomaly, exceptionally diagnosed in adulthood. In low-resource settings, delayed diagnosis due to limited access to advanced imaging is a major obstacle to optimal care. Our two cases show that, despite these limitations, carefully conducted surgical intervention can achieve safe and durable outcomes in adults. This experience highlights the need to strengthen local diagnostic capacities and promote multidisciplinary collaboration to improve the prognosis of adult congenital heart diseases in sub-Saharan Africa.

Conflicts of Interest: The authors declare no conflicts of interest related to this publication.

Ethical Approval: Not Required

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