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A Rare Case of Aortico-Cameral Fistula Treated Surgically at BSMMU-Case Report

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Abstract: *Aortico -Cameral fistula is a relatively rare cardiac anomaly. It can be either congenital (Present at birth) or acquired due to factors like trauma, infection, or complications from procedures such as (transcatheter aortic valve implantation, TAVI). Congenital cases are more common. Their presentation ranges from being asymptomatic in early life to becoming symptomatic and leading to complications as the patient ages. Treatment may range from medication to manage symptoms to surgical intervention to close the fistula. While percutaneous closure with embolization is an option, surgical closure remains the preferred standard treatment. In this case, a 19-year-old male with effort-induced angina and an atypical continuous murmur was found to have an aortico-cameral fistula via echocardiography and cardiac CT. Surgical management included fistula closure with a Poly Tetra Fluoro Ethylene (PTFE) patch, ligation of the distal end of the fistula near the Posterior descending artery (PDA), and a reverse saphenous vein graft between aorta & distal PDA. The patient had a smooth recovery and was discharged in stable condition. A high degree of suspicion, timely diagnosis, and appropriate surgery were crucial for the successful treatment. Timely diagnosis and surgery were essential for successful treatment.*

Keywords: *Cardiac anomaly, Congenital or acquired, Fistula closure, Echocardiography and CT, Surgical management.*

1. Introduction

Aortico-cameral fistula is an uncommon congenital condition involving an abnormal fistulous communication between aorta & one of the heart chambers. In contrast, coronary arteriovenous fistulas involve connections between a coronary artery and systemic or pulmonary circulation [1]. Aorto ventricular tunnels are more frequently connected to the left ventricle than the right one. Among the aorto ventricular tunnels reported in the literature, more than 90% are in communication with left ventricle, with an incidence estimate from 0.5% of fetal cardiac malformations to less than 0.1% of congenitally malformed heart [2]. The majority of coronary arteries (90%) drain into right-sided heart chambers or major vessels, while drainage into left-sided chambers is extremely uncommon [3]. Here, we are reporting the case of a 19-years-old male who underwent successful surgical closure of the Aortico-cameral fistula. Aneurysmal dilatation of right coronary artery was seen from its origin at right coronary cusp.

2. Case Report

A 19-year-old male presented to us with a history of effort angina. On general examination, pulse-88bpm, regular, low in volume; blood pressure -120/80 mm of Hg. Cardiovascular examination revealed atypical continuous murmur over precordium. The rest of the physical examination were normal.The

Color Doppler echocardiogram showed a dilated aortic sinus (44mm). The right coronary sinus was dilated & there was tunnel like fistula (Aortico-Cameral fistula) originating from right sinus which ultimately drained into the basal lateral part of RV cavity through the right coronary artery. His ventricular function was quite good and the LVEF was 61%.Cardiac CT confirmed the aortico-cameral fistula. The length of the fistula was about 58mm & the diameter was about 8.8mm. McGoon ratio 1.9. Aneurysmal dilatation of right coronary artery was seen from its origin at right coronary cusp up to giving of the posterior descending artery branch. Posterior descending artery &postero lateral branches of RCA appears normal.

After proper evaluation, patient was diagnosed as a case of Aortico-cameral fistula and surgery was planned.After opening the chest by median sternotomy inspection revealed dilated right coronary artery (RCA) (Fig. 1) having a tortuous course over the right atrio-ventricular groove and the fistula terminating into right ventricle (RV). The cardiopulmonary bypass was established by aortic and two stage venous cannulation. The patient was cooled to 32° Celsius. After applying cross clamp, the aorta was opened and ostial antegrade cold cardioplegia was administered. The proximal opening of fistula was identified (Fig. 2) and closed by PTFE patch (Fig. 3). The fistulas tract was ligated near the PDA (Fig.4) and a reverse saphenous vein graft was placed between the aorta and posterior descending. Two chest drains kept in each pleural space and a third drain was placed in the mediastinal cavity respectively. After proper hemostasis, wound was closed in layers. Aortic cross-clamp time was 120 minutes & cardiopulmonary bypass time was 198 minutes.The postoperative course of the patient was uneventful. We discharged the patient on the 12th postoperative day in a hemodynamically stable state.

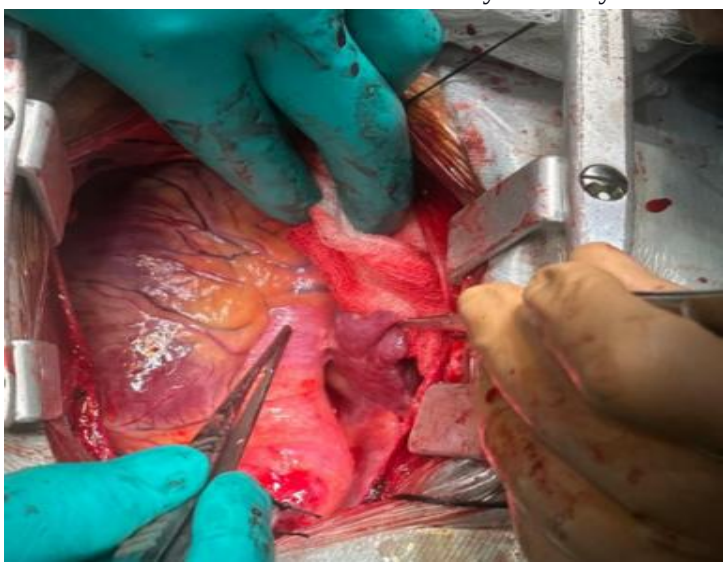

Figure1. *Dilated right coronary artery on the heart surface.*

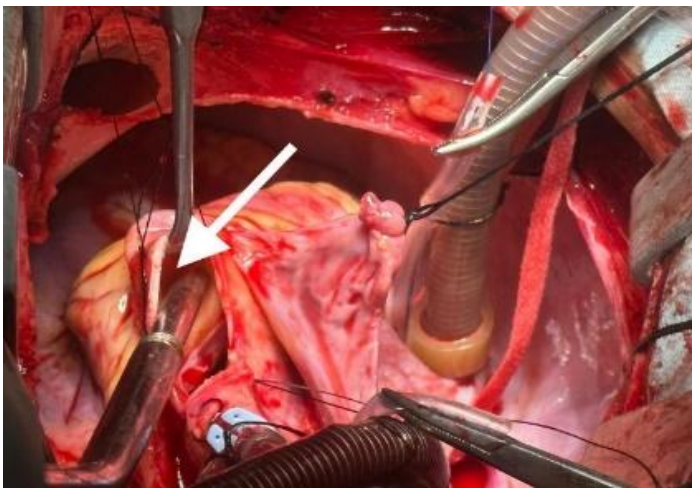

Figure2. *Showing the dilated fistulas Right Coronary Artery*

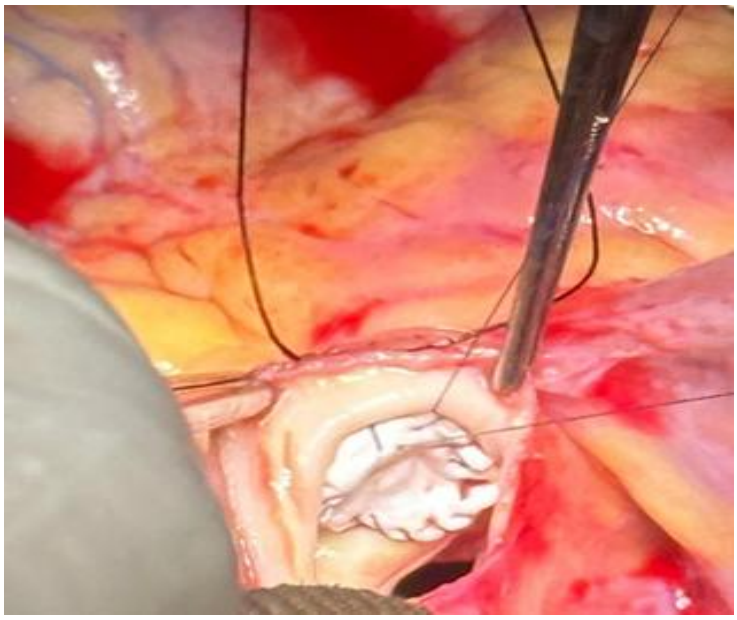

Figure3. *PTFE patch closure of right coronary artery*

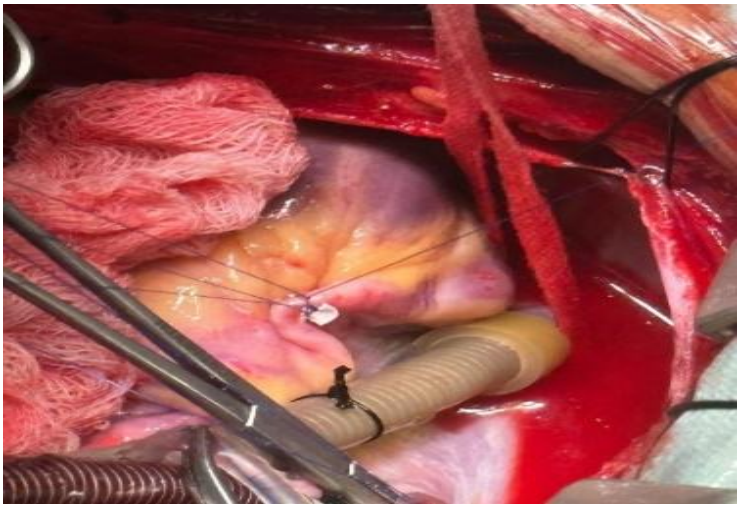

Figure4. *Ligation of right coronary artery with pledgeted stitch.*

3. DISCUSSION

Aorto-ventricular tunnel is an extracardiac channel that connects the AA to the RV, passing outside the heart into the tissue plane between the muscular sub pulmonary infundibulum and the AV sinuses [4]. It must be differentiated from a ruptured sinus of Valsalva aneurysm/fistula, which is an intracardiac channel that connects a sinus of the AV to the LV [5].The aortico-cameral fistula can drain into any of the heart's four chambers, with the majority originating from the right coronary artery (RCA) and most frequently terminating in the right ventricle (RV) [6]. In this case, a 19-year-old male presented with an aortico-cameral fistula, experiencing effort-related angina and an atypical continuous murmur over the precordium. A Doppler echocardiogram showed a dilated aortic sinus (44 mm) and an aortico-cameral fistula from the right sinus draining into the RV. Cardiac CT confirmed the fistula's presence (58 mm long, 8.8 mm in diameter) and revealed aneurysmal dilation of the RCA. Banday et al [7]. described a case involving a 10-year-old boy with a coronary cameral fistula connecting the proximal RCA to the right atrium (RA).In one study, a 66-year-old patient with aortic dissection developed a fistula from the aorta to the right ventricle [8]. Another case involved a coronary cameral fistula, where a connection formed between the coronary artery and the heart’s atrial or ventricular chambers [9].The aorto-ventricular tunnel is an uncommon congenital anomaly resulting from abnormal development of the aortic valve (AV) sinuses during the embryonic formation of the

cushions within the aortopulmonary outflow tract. The abnormal thinning of the distal margins of the cushions produces the openings of the tunnel, while the abnormal growth of the central part of the fused cushion mass in the plane between the infundibulum and the aortic root explains the formation of those tunnels. The tunnel is histologically different in its parts: the arterial end of the tunnel resembles the aorta with fibrous tissue, elastic fibers, and smooth muscle cells, while within the tunnel itself, there is a junction between ventricular and arterial components [10]. The involvement of the adjacent AV sinuses and leaflets in their formation explains the high incidence of associated anomalies of the coronary arteries and of the semilunar valves. The coronary anomalies can vary from an anomalous orifice origin to atresia. The ostium of the coronary artery may also lie within the aorto-ventricular tunnel itself [3]. In addition, older patients may acquire leaflet perforation or aortic insufficiency as a result of hydrodynamic trauma to the unsupported right or left coronary cusp, and progressive aortic dilatation. Stenosis of the pulmonary valve occurs less frequently, while compression of the RVOT by the tunnel may produce sub-pulmonary obstruction [5].This patient presented with a dilated aortic sinus measuring 44 mm and a tunnel-like aortico-cameral fistula originating from the right coronary sinus and draining into the right ventricle (RV) through the right coronary artery. Both left and right ventricular functions were good, with a left ventricular ejection fraction (LVEF) of 61%. Cardiac CT confirmed the fistula's dimensions at 58 mm long and 8.8 mmwide, along with a dilated right coronary artery extending to the posterior descending artery (PDA). Frías-Ordoñez JS reported a 52-year-old male with a coronary cameral fistula also presented with angina of effort, which led to further investigation and treatment [11]. Similar cases of acquired coronary cameral fistulas has been reported, where patients experienced significant symptoms due to the abnormal blood flow, including angina and ischemic complications [12]. It may be presented with quiet earlier. A 27-year-old pregnant female was also diagnosed with a right coronary artery fistula leading to the right ventricle, emphasizing the importance of early detection even during routine checkups [13].A high degree of suspicion, meticulous clinical examination, and CT angiogram were prerequisites to diagnose this case.

4. Conclusion

In conclusion, this case highlights the successful diagnosis and surgical management of a rare aortico-cameral fistula in a 19-year-old male with a history of effort angina and atypical continuous murmur. Preoperative imaging, including color Doppler echocardiogram and cardiac CT, confirmed the Aortico-cameral fistula. A conventional angiogram was excluded because the patient was young and the delineation was very obvious on cardiac CT.

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