

CASE REPORT

ANOMALOUS VENA CAVAL RETURN TO LEFT ATRIUM
PRESENTING AS RECURRENT ABORTIONS: A CASE REPORTMohsin Masud[✉], Yasir Shafi, Sajid Abaidullah

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Opening of inferior vena cava into the left atrium with atrial septal defect is an extremely rare congenital abnormality. We report a case of 28-years-old female presented with repeated respiratory tract infections and repeated abortions 19 years after the repair of atrial septal defect and partial anomalous pulmonary venous return. Investigations showed that she was also having abnormal drainage of inferior vena cava into the left atrium. This drainage was causing hypoxemia leading to repeated abortions. Second surgery was performed which confirmed the drainage of inferior vena cava into the left atrium. The repair was carried out with subsequent correction of hypoxemia and successful pregnancy.

Keywords: Anomalous vena caval return; Atrial septal defect; Repeated abortions.

Citation: Masud M, Shafi Y, Abaidullah S. Anomalous vena caval return to left atrium presenting as recurrent abortions: A case report. J Ayub Med Coll Abbottabad 2025;37(2):265–8.

DOI: 10.55519/JAMC-02-13046

INTRODUCTION

Anomalous drainage of the inferior vena cava (IVC) into the left atrium (LA) is a rare congenital disorder that can occur alone or in association with other cardiovascular anomalies like anomalous pulmonary venous drainage or atrial septal defects (ASD).^{1–4} ASD is a congenital cardiac anomaly where an abnormal communication persists between right and left atrium. Among its various types, sinus venosus type is a rare one representing only 5 to 10% of all ASD cases.^{5,6}

We discuss a case of a 28-years-old female with sinus venous atrial septal defect (SVASD) with partial anomalous pulmonary venous return (PAPVR) and abnormal communication of IVC with LA who had repeated abortions, a unique presentation of a rare congenital anomaly.

CASE PRESENTATION

A 28-years-old female presented to outdoor medical clinic with the complaint to cough for last four days. The cough was not associated with fever and episodes were more at night. Patient gave the history of frequent episodes of cough for past few years and shortness of breath on intense exertion. There was no history of hemoptysis, palpitations, or syncope. The patient was married for past 2 years, had 4 spontaneous miscarriages and no live issue. None of her pregnancies progressed beyond 7 weeks of gestation. The patient was being thoroughly evaluated by the gynecologist for the possible causes of repeated miscarriages for past one year, but none was found yet. The patient also gave the history of cardiac surgery done at the age of 9 years for the repair of Atrial septal defect (ASD) and Partial anomalous pulmonary

venous return (PAPVR). The review of her surgical record revealed subsequent normal echocardiograms. She was not taking any medications. The patient was not diabetic or hypertensive and her family history was not significant regarding any medical or congenital ailments. On examination, the patient was sitting comfortably, with no apparent pallor or cyanosis. Clubbing was present (since childhood). She had normal hemodynamic parameters and was afebrile. Her heart rate was 62 bpm and her pulse was regular. Her saturation at room air was 92%. Auscultation of the chest revealed normal vesicular breathing with no added sounds. First and second heart sounds were audible with no murmur. Her oxygen saturation was rechecked after activity, i.e., walking for a block which turned out to be 63%. The patient was mildly dyspneic, and her oxygen saturation gradually improved back to 90% after resting. Laboratory investigations of the patient were then carried out. Her complete blood count (CBC) showed Hb of 16.8 g/dl. Her WBC and platelet counts were within normal range ($6.4 \times 10^9/L$ and $174 \times 10^9/L$ respectively). Her chest X ray and electrocardiogram (ECG) did not reveal any abnormality. HRCT and pulmonary function tests (PFTs) were performed which also turned out to be normal. Her transthoracic echocardiography was done which concluded surgically corrected ASD, trace mitral regurgitation (MR) and tricuspid regurgitation (TR) with no pulmonary hypertension. Considering the level of hypoxemia the patient developed on exertion, her transesophageal echocardiography (TEE) was done. It revealed abnormal superior vena cava (SVC) drainage in right atrium (RA). There was delayed appearance of contrast from left upper pulmonary vein in left atrium

(LA). The interatrial septum (IAS) was repaired and intact with no evidence of shunt, interventricular septum (IVS) was also intact, there was mild TR and MR and a normal pulmonary artery pressure (PAP) of 19 mmHg. Further proceeding with the work-up, her pulmonary CT angiogram was done. There was right to left shunt due to direct communication between inferior vena cava (IVC) and LA. SVC gram showed communication of SVC to the RA and right ventricle (RV). Small residual ASD was also identified. However, there was no extra-cardiac shunt identified in the lung fields. The patient was referred to the

cardiac surgeon who performed the corrective surgery. The operative findings confirmed baffling of IVC to LA. There was a small residual ASD and the Dacron patch used for ASD repair previously was heavily calcified. The patch was excised partially and ASD closed using autologous pericardial patch, ensuring IVC connection to RA. The patient did well postoperatively and was discharged. The patient was kept in regular follow-up. Her symptoms gradually improved. The patient conceived subsequently. The pregnancy remained uneventful, and she gave birth to a healthy baby.

Table-1: Timeline of disease course

1998	Diagnosed with ASD + PAPVR	Corrective surgery performed
2014	Follow up echocardiography	Normal study, Repaired ASD
2015	Follow up echocardiography ECG	Normal study, Good repair of ASD Normal sinus rhythm
April 2017	First pregnancy, confirmed by beta HCG levels	CBC, Blood glucose levels normal, TTE showed normal study with good ASD repair
May 2017	Spontaneous abortion at 8 weeks	Conservatively managed and discharged No RPOC on pelvic ultrasound
July 2017	Second pregnancy, confirmed by beta HCG levels and pelvic ultrasound	CBC, Blood glucose levels normal
August 2017	Spontaneous abortion at 7 weeks	No RPOC on follow up pelvic ultrasound
Oct 2017	Work up for abortions started Antiphospholipid antibodies, ANA, AMA, ASMA, Rubella antibodies Glucose tolerance test	Negative Normal study
Nov 2017	Third pregnancy, confirmed by beta HCG levels and pelvic ultrasound	Started antiplatelets and dydrogesterone
Dec 2017	Spontaneous abortion at 7 weeks	
March 2018	Continued workup for repeated abortions TORCH Profile report	Non-reactive
Sept 2018	Fourth pregnancy, confirmed by beta HCG levels and pelvic ultrasound Indirect Coomb's test Thyroid profile	Negative Normal
Oct 2018	Spontaneous abortion at 7 weeks	
Feb 2019	Presented with c/o cough for four days Gave history of similar episodes for past few years	SpO ₂ at rest = 92% SpO ₂ after walk = 63%, gradually improved with rest, Normal chest Xray, Hb = 16.8g/dl, advised echocardiography and PFT
March 2019	Transthoracic echocardiography done HRCT chest	Normal study with good LV systolic function Unremarkable study
April 2019	Transesophageal echocardiography done	Showed repaired ASD with no evidence of shunt, grade-I MR, Delayed appearance of contrast into LA Pulmonary CT angiogram advised
May 2019	Pulmonary function test performed	Showed mild restriction
June 2019	Pulmonary arteriography done Case referred to cardiac surgeon Corrective cardiac surgery performed	Reveled right to left shunt due to IVC emptying into LA, small residual ASD Operative findings: IVC baffled to LA, Dacron patch over previously repaired ASD heavily calcified Previous patch partially excised, defect closed with autologous pericardial patch, ensuring IVC connection to RA.
Follow up in subsequent months	Patient was doing well, no symptoms regarding cardiovascular system Conceived subsequently, normal term pregnancy Patient was much satisfied with all the management	Delivered a healthy baby

DISCUSSION

Abnormal drainage of IVC into LA is a rare congenital vascular anomaly first described by Gardner in 1957. 70% of such cases are also associated with ASD.

Anomalous pulmonary venous drainage and pulmonary arteriovenous fistulae are other associated malformations¹. Our patient had a sinus venosus type ASD with PAPVR and IVC draining into the LA. SVASD is a rare type of ASD, the defect being located

near the entry point of the superior or inferior vena cava.^{5,6} The patient presented to us with a complaint of cough. She had undergone corrective surgery for the repair of ASD with PAPVR at the age of 9 years. Her subsequent echocardiograms were normal with no residual defects. The patient did not develop any symptoms related to the cardiovascular system like palpitations, dizziness, or chest pain. She had only mild dyspnea on intense exertion and occasional episodes of respiratory tract infections. However, while probing into her history, it surfaced that the patient was struggling with the problem of repeated abortions for the past two years which was a major concern to her. She was being evaluated for the reason of her repeated spontaneous miscarriages by the gynecologist. Various known etiologies of repeated abortions include cytogenetic anomalies, structural uterine anomalies, endocrine disorders like that of thyroid and diabetes mellitus, metabolic disorders, antiphospholipid syndrome (APS), non-APS thrombophilia, and environmental factors.⁸⁻¹⁰ During her workup for repeated abortions, numerous laboratory investigations were carried out. There was no structural abnormality in her reproductive tract on pelvic ultrasound. There were no adnexal masses or cysts. Her metabolic profile was within normal limits. She was not diabetic, and her thyroid profile revealed normal thyroid functioning (serum TSH = 3.1339 uIU/ml). Antiphospholipid antibodies were also negative. Anti-HCV and HBsAg were not detected. Her antibody profile for autoimmune dysfunction was carried out. ANA, AMA, and ASMA were all negative. IgG and IgM antibodies for Toxoplasma, Rubella, Cytomegalovirus, and Herpes simplex virus (TORCH infections) were non-reactive. Her direct and indirect Coomb's tests were negative. Transthoracic echocardiography revealed a normal study with no residual defect. Her hemoglobin (Hb) was within the normal range during the first three pregnancies (14.0 g/dl, 11.5 g/dl, 12.6 g/dl respectively). However, during her 4th pregnancy, her Hb was 16.1 g/dl (normal range 12–14 g/dl). During her 3rd and fourth pregnancies, she was prescribed antiplatelets (aspirin 75 mg) and anticoagulants (Low molecular weight heparin) along with progesterone, but they also ended up in spontaneous abortions in the 7th and 8th week, respectively. At her presentation in our clinic, hypoxia was detected on exertion (SpO₂ = 63%) while her resting oxygen saturation was 92%. This extent of hypoxia raised our suspicion for any residual shunt and her transthoracic echocardiogram was again performed only to reveal a normal study. To probe further into the matter TEE was done which hinted towards the underlying problem. The final diagnosis of anomalous drainage of IVC into the LA was made based on her pulmonary angiogram. This abnormal

drainage of IVC into LA could have been congenital and missed during her corrective surgery for SVASD. Choi *et al* described a case of ASD with PAPVR and drainage of IVC into LA1 where the patient presented at the age of 7 years with complaints of palpitations and chest discomfort. There is also a possibility of this being an iatrogenic diversion during the repair of ASD, a rare complication of surgery.¹¹ The eustachian valve during fetal life directs the oxygenated blood from IVC towards the foramen ovale. This eustachian valve is the embryological remnant of the IVC valve. In the case of the margin of the ASD defect lying at the superior margin of the eustachian valve, the valve can be mistakenly considered as the lower margin of the defect, thus taken into the repair, resulting in the iatrogenic diversion of IVC into the LA. This results in the bypassing of the deoxygenated blood from the lung and ending up directly into the LA, leading to hypoxemia. A few cases of iatrogenic diversion of IVC into LA while repairing low-lying ASDs have been reported.¹¹⁻¹³ The possibility of this being a missed congenital anomaly is more as the patient had adapted herself well to this shunt and the resulting hypoxemia and had developed minimal symptoms. However, the fetuses were unable to tolerate the hypoxemia and ended up in spontaneous abortions. Once the surgical correction of this diversion was performed, the hypoxemia of the patient was corrected. The subsequent pregnancy of the patient remained uneventful, and she delivered a healthy baby.

CONCLUSION

The hypoxemia resulting from the opening of IVC into the LA was the cause of repeated abortions in this patient. Once surgically corrected, the subsequent pregnancy was successful.

Conflict of interest: None

Ethical approval: The patient gave informed consent for the publication and provided the relevant data.

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*Submitted: March 6, 2024**Revised: May 8, 2025**Accepted: May 21, 2025*

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