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Case Report

Iatrogenic Diversion of the Inferior Vena Cava to the Left Atrium After Atrial Septal Defect Closure: Misdiagnosis of Chronic Dyspnea

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Abstract

Background: Iatrogenic diversion of the inferior vena cava (IVC) to the left atrium (LA) can occur in patients who have undergone atrial septal defect (ASD) closure. This can result in hypoxemia and life-threatening complications, such as stroke, due to paradoxical embolism. Currently, this complication is rare owing to transesophageal echocardiography (TEE) performed during the surgery in the operating room. **Case:** We report a case of a patient with a longstanding, unrecognized chronic dyspnea after ASD closure who subsequently experienced a stroke. Following the diagnosis of iatrogenic diversion of the IVC into the LA, surgical correction was undertaken, and the dyspnea was resolved completely. **Conclusions:** Some patients who undergo ASD closure during childhood can reach adulthood with undiagnosed or misdiagnosed dyspnea.

Keywords: atrial septal defect; complication; congenital heart disease; inferior vena cava; left atrium

1. Introduction

Iatrogenic diversion of the inferior vena cava (IVC) to the left atrium (LA) is a rare complication that can occur after closure of a large posteroinferior atrial septal defect (ASD). Notably, this complication is uncommon owing to the routine use of intraoperative transesophageal echocardiography (TEE) during cardiac surgery. Moreover, after induction of general anesthesia, an anesthesiologist performs TEE before the operation to obtain a clear image of the defect and, after closure of the defect, to assess the surgical success of repair [1,2].

However, postoperative hypoxemia or exertional dyspnea may occur in some cases immediately after ASD closure, or chronic hypoxemia may present years after surgery [3,4,5,6,7]. This delayed diagnosis can result in potentially life-threatening complications such as stroke or recurrent foetal loss [8,9,10]. Meanwhile, particular care must be taken to visualize the lower corner of the defect in low-IVC-type ASD. Indeed, failure to visualize this region, or placing stitches too close to the IVC cannula and not tying them securely, can result in either a residual shunt or, more importantly, partial or total drainage of the IVC into the LA. The suture line should begin with an open technique in the lower corner of the ASD, with careful visualization of the IVC orifice and the Eustachian valve.

This case report describes a patient with a longstanding unrecognized chronic dyspnea after surgical closure of ASD, who subsequently experienced a stroke, a life-threatening complication.

2. Case Presentation

We report the case of a 16-year-old girl who was diagnosed with ASD at birth. She underwent surgical ASD closure at 18 months of age. During the growth years, she walked and ran more slowly, got out of breath earlier, and had difficulty moving around compared to others. However, since this did not markedly impact her daily life, she was simply presumed to be weak. During childhood, she faced limitations in exercise-based activities owing to mild dyspnea.

At 15 years, the exercise-induced dyspnea progressed to the modified Medical Research Council (mMRC) dyspnea scale grade 2 to 3, and the patient subsequently visited the pediatric department. Pulse oximetry revealed an average oxygen saturation of 90 % with peripheral cyanosis. Transthoracic echocardiography (TTE) of the heart and contrast-enhanced computed tomography (CT) of the chest failed to reveal the cause of dyspnea. As pulmonary hypertension and restrictive lung function due to congenital heart disease were suspected, the patient was administered sildenafil (phosphodiesterase-5 inhibitor). However, there was no improvement in symptoms, and the dyspnea persisted.

During the pediatric department follow-up, dysarthria developed due to cerebral infarction. At 16 years, the patient developed sudden gaze deviation, right upper and lower limb weakness, and dysarthria, necessitating an emergency department visit. Brain magnetic resonance imaging revealed cerebral infarction in the left middle cerebral artery territory. Since the most common risk factor for pediatric stroke is cardiac origin [10,11], the patient was re-evaluated by the neurology department. More invasive

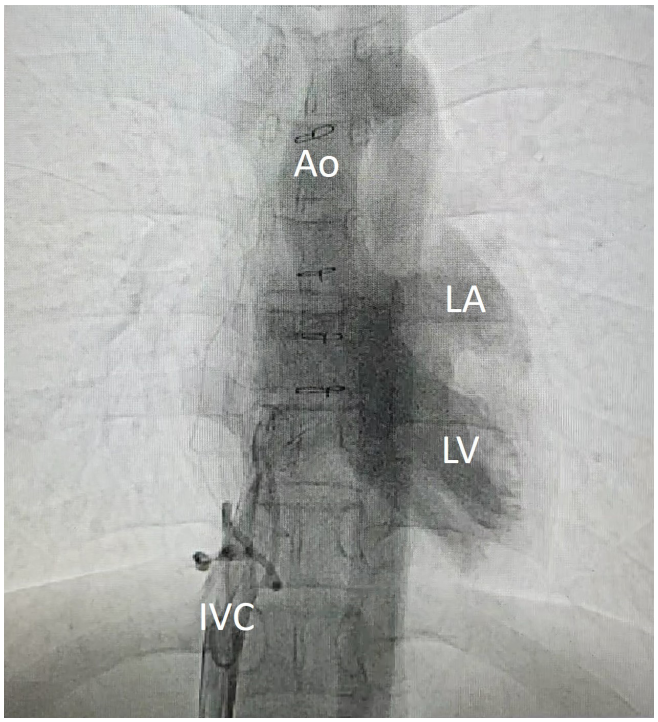


Fig. 1. IVC angiogram. IVC angiogram of the contrast flow filling from the IVC to the LA, left ventricle, and aorta. Confirming diversion of the IVC into the LA. Ao, aorta; IVC, inferior vena cava; LA, left atrium; LV, left ventricle.

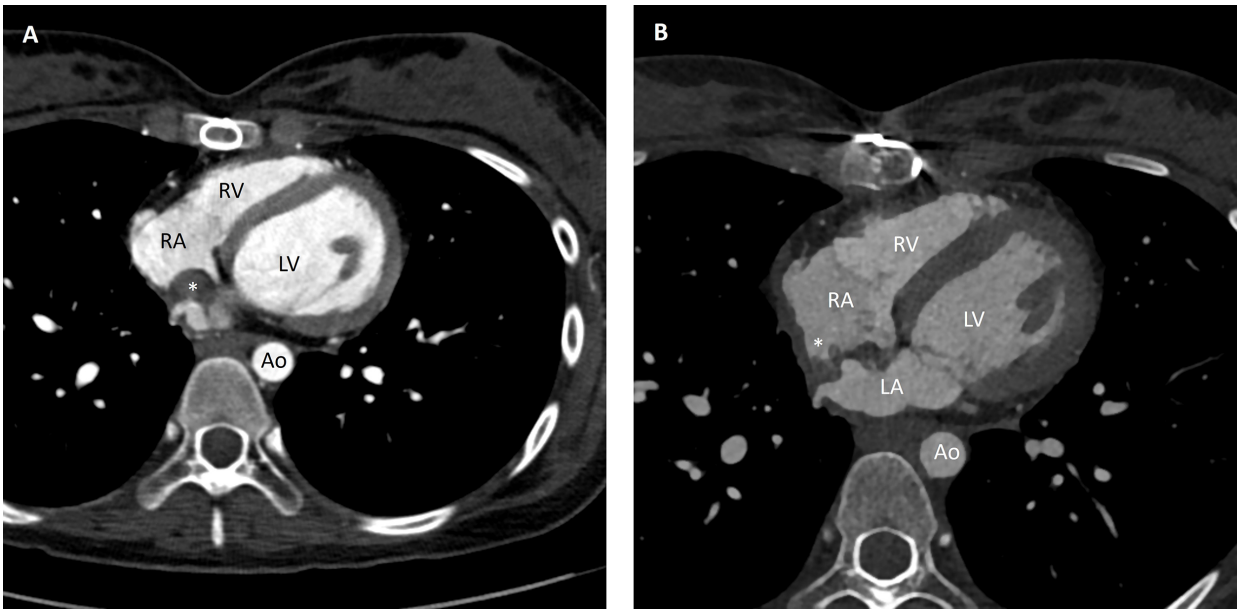
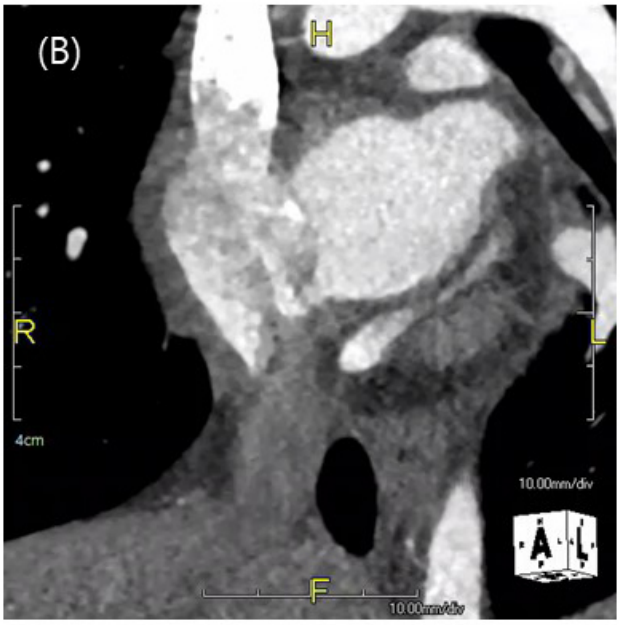


Fig. 2. Contrast-enhanced chest computed tomography (CT) of the patient. Preoperative (A) and postoperative (B) CT. (A) The contrast-filling defect in the RA demonstrates the abnormal connection of the inferior vena cava (IVC, asterisk). (B) After the surgical correction, there is no contrast fillingdefect in the RA; meaning well connection of the inferior vena cava (asterisk) to the right atrium. Ao, aorta; LA, left atrium; LV, left ventricle; RA, right atrium; RV, right ventricle.

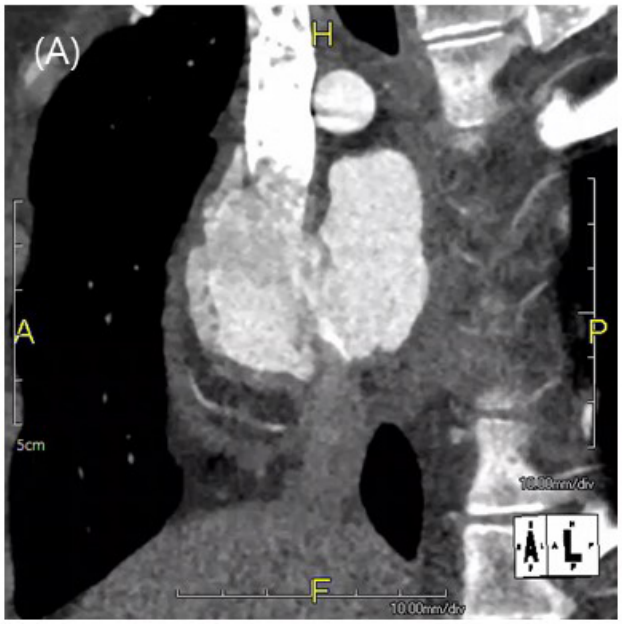
examinations, including TEE and cardiac catheterization, were undertaken. The TEE results were unclear regarding abnormal IVC drainage into the LA. An agitated saline contrast study (bubble test) via the femoral vein was performed, and a shunt lesion was suspected from the IVC to the LA [12]. Finally, an IVC angiogram confirmed that contrast initially filled the LA, confirming a diagnosis of IVC diversion into the LA (Fig. 1). Surgical correction was planned and preoperative contrast-enhanced CT was performed with injection of a contrast dye into a lower limb vein (Fig. 2A).

Redo median sternotomy and cannulation were performed. For venous cannulation, the superior vena cava and femoral veins were cannulated using a 24 Fr angled catheter and a 19 Fr straight catheter, respectively (the body weight of the patient was 43.5 kg). Under mild hypothermia, cardiopulmonary bypass was established, and pharmacological cardiac arrest was induced. The right atrium (RA) was incised. The previous patch was removed, and the anatomical structures of the IVC, left lower pulmonary vein, and mitral valve were identified. The ASD was closed correctly using a new bovine pericardial patch. Since the IVC opening was narrow, IVC angioplasty with a bovine pericardial patch was undertaken before RA closure. The cardiopulmonary bypass and aortic cross-clamp times were 84 and 45 minutes, respectively. Following an uneventful postoperative course, the patient was discharged on postoperative day 6.

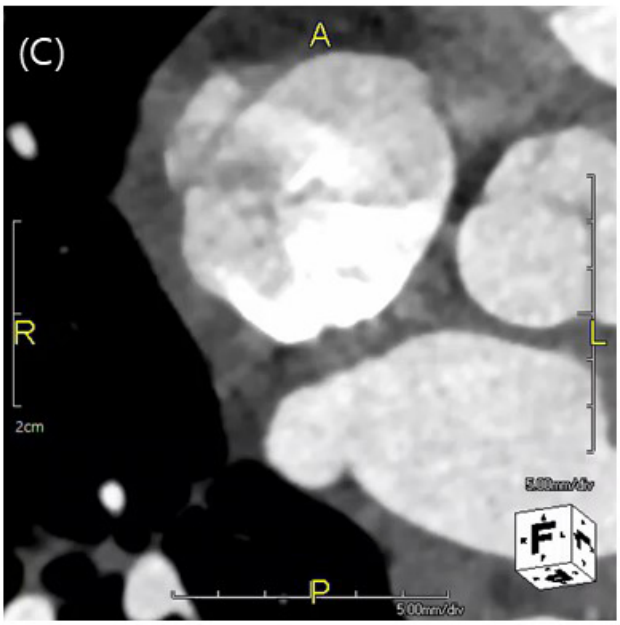
Postoperative TTE showed no residual shunt with normal cardiac function. Further, postoperative contrast-enhanced chest CT showed closure of the ASD without IVC obstruction (Fig. 2B, Videos 1A,1B,1C). Two months after discharge, the patient visited the outpatient clinic and was able to resume daily activities and exercise freely without dyspnea.



Video 1B. Postoperative contrast-enhanced chest computed tomography (CT) of the patient. Postoperative CT showing the inferior vena cava course in multiple planes. (B) Coronal plane. Video associated with this article can be found, in the online version, at https://doi.org/10.31083/HSF46821.



Video 1A. Postoperative contrast-enhanced chest computed tomography (CT) of the patient. Postoperative CT showing the inferior vena cava course in multiple planes. (A) Sagittal plane. Video associated with this article can be found, in the online version, at https://doi.org/10.31083/HSF46821.



Video 1C. Postoperative contrast-enhanced chest computed tomography (CT) of the patient. Postoperative CT showing the inferior vena cava course in multiple planes. (C) Axial plane of the CT. Video associated with this article can be found, in the online version, at https://doi.org/10.31083/HSF46821.

3. Discussion

Currently, neonatal and pediatric ASD closure is considered an extremely safe procedure with a low risk of complications and successful outcomes. Iatrogenic diversion of the IVC into the LA has been reported as a complication following closure of a low-IVC-type ASD [1,2]; however, while this complication is rare, there are some cases of undiagnosed or misdiagnosed dyspnea in adulthood.

First, surgeons should always be aware of the anatomy during surgery. Indeed, ASD closure has become a safe procedure following the development of cardiopulmonary bypass techniques. Nonetheless, the simpler the procedure, the more carefully the basic anatomy needs to be considered. During ASD closure, the lower corner of the ASD, the IVC orifice, and the Eustachian valve must be visualized. Especially in low IVC-type-ASD, care must be taken to visualize the lower corner of the defect since failure to visualize this lower corner, or placing the stitches close to the IVC cannula and not tying them securely, can result in either a residual shunt or, more importantly, partial or total drainage of the IVC into the LA. The suture line should begin with an open technique in the lower corner of the ASD, with careful visualization of the IVC orifice and the Eustachian valve [13].

Second, it is important to always listen carefully to the symptoms of each patient. In the postoperative period after ASD closure, unexplained hypoxemia or desaturation may occur. Arrhythmias can also lead to symptoms such as dyspnea and palpitations after ASD closure. The presence of residual shunts or newly formed shunts could lead to cyanosis and exertional dyspnea. Iatrogenic issues, such as our case, which includes the diversion of the IVC into the LA during surgery, can also result in significant respiratory symptoms. When performing postoperative TTE, patients with unexplained symptoms should undergo further evaluation with close consideration and high index of suspicion.

Third, when performing a test on a patient, it is essential always to consider the necessity and appropriateness of the test method. Our patient underwent a TTE and contrast-enhanced CT at the initial pediatric department visit; however, this failed to lead to a diagnosis of the cause of dyspnea. After planning the surgery, a preoperative contrast-enhanced CT was performed with injection of an iodinated contrast medium into a lower limb vein [14,15]. The contrast travels from the leg vein through the common iliac veins, which merge to the IVC, showing that the IVC drains into the RA; however, in our patient, the contrast drained into the LA (Fig. 2A). If the patient had undergone the first contrast-enhanced CT properly, the diversion of the IVC into the LA might have been discovered before the stroke occurred.

4. Conclusions

Iatrogenic diversion of the IVC into the LA can occur in patients undergoing ASD closure. If postoperative hypoxemia or desaturation occurs after ASD closure, diversion of the IVC into the LA should be suspected. Echocardiography should be performed with a high index of suspicion, and contrast-enhanced CT should be performed via injection of contrast into a lower limb vein to highlight the IVC drainage pathway. Most importantly, the chief complaint of each patient should be heard and carefully considered.

Abbreviations

ASD, atrial septal defect; CT, computed tomography; IVC, inferior vena cava; LA, left atrium; RA, right atrium; TEE, transesophageal echocardiography; TTE, transthoracic echocardiography.

Availability of Data and Materials

Data sharing does not apply to this article as no datasets were generated or analyzed.

Author Contributions

HJ conceived the report and wrote the manuscript. YL was the main surgeon and was involved in drafting the manuscript or reviewing it critically. HJ and YL were involved in patient care. HJ, SJP and YL made substantial contributions to conception and design, or acquisition of data, or analysis and interpretation of data. SJP revised the manuscript. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript. All authors have participated sufficiently in the work and agreed to be accountable for all aspects of the work.

Ethics Approval and Consent to Participate

The study was carried out in accordance with the guidelines of the Declaration of Helsinki. Informed consent was obtained from the patient's parents for this case report and the use of any accompanying images. The Institutional Review Board of Kyungpook National University Hospital approved the study, with informed written consent obtained from the patient's parent..

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Conflicts of Interest

The authors declare no conflicts of interest.

Supplementary Material

Supplementary material associated with this article can be found, in the online version, at https://doi.org/10.31083/HSF46821.

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