

Tricuspid Stenosis: A Rare and Potential Complication of Ventricular Septal Occluder Device

Abstract

Asymmetrical septal occluder device (ASOD) has made percutaneous closure of ventricular septal defect an easy and effective management option. Although there are reports of aortic and tricuspid valvular regurgitation after deployment of ASOD, only few cases of tricuspid stenosis (TS) has been reported so far in the literature. We report a case of malaligned ASOD that occurred after successful device closure resulting in TS along with mild tricuspid and aortic regurgitation requiring surgical retrieval. Transesophageal echocardiography played crucial role in detecting the cause of tricuspid valve dysfunction besides providing continuous monitoring during the procedure. We intend to emphasize the need of echocardiographic evaluation of the tricuspid valvular apparatus and aortic valve during and after the device deployment even after the successful device closure to prevent this rare complication.

Keywords: *Asymmetrical septal occluder device, tricuspid stenosis, ventricular septal defect*

Introduction

Ventricular septal defects (VSDs), the 2nd most common congenital heart disease (CHD) only after the bicuspid aortic valve, is often treated using percutaneous techniques.^[1] With the development of newer asymmetrical septal occlude device (ASOD), many VSDs which were thought unsuitable for percutaneous closure are being treated using these devices with acceptable morbidity and very low mortality.^[2-6] Sometimes, after deployment of ASOD, tricuspid valve regurgitation (TR) and/or aortic valve regurgitation (AR) develop due to the proximity of the aortic valve and tricuspid valve to the perimembranous VSD. The occurrence of tricuspid stenosis (TS) is very rare.^[6] We report an unusual case of TS along with mild TR and AR in a patient after successful device closure of a perimembranous VSD that required surgical retrieval of ASOD. Transesophageal echocardiography (TEE) after device retrieval showed ruptured chordae tendinae of septal leaflets of TV causing TR.

Case Report

A 12-year-old male child presented to our institution with a complaint of respiratory

distress while playing. His general physical examination was unremarkable. Auscultation of chest revealed a grade 4/6 pan-systolic murmur over precordium. Transthoracic echocardiogram (TTE) showed an 8 mm perimembranous VSD with left to right shunt, normal biventricular function, and no TR. The gradient across VSD was 40 mmHg with no evidence of pulmonary arterial hypertension and Qp/Qs ratio >1.5:1.

He underwent percutaneous device closure for VSD under general anesthesia with endotracheal intubation after confirmation of VSD suitability for same on TEE and cath study. A 10 mm ASOD (Cera™ membrane VSD occluder, Life tech scientific, China) was placed across the VSD through femoral approach, and the device was deployed in place after confirming correct position on TEE and angiography. Postdeployment angiography confirmed the position of ASOD across the VSD with minimal flow across it and normally functioning tricuspid and aortic valve [Figure 1]. After the procedure, anesthesia was reversed, and tracheal tube was extubated while avoiding coughing and bucking.

On post-ASOD placement day 1, TTE revealed the device located in the tricuspid valve inlet causing turbulent flow across tricuspid valve and mild aortic

**Ganesh Kumar
Munirathinam,
Bhupesh Kumar,
Anand Kumar
Mishra¹**

*Departments of Anesthesia
and Intensive Care and
¹Cardiothoracic and Vascular
Surgery, Postgraduate Institute
of Medical Education and
Research, Chandigarh, India*

Address for correspondence:
*Dr. Bhupesh Kumar,
Department of Anesthesia and
Intensive Care, Postgraduate
Institute of Medical
Education and Research,
Chandigarh - 160 012, India.
E-mail: bhupeshkr@yahoo.com*

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regurgitation [Figure 2a and Video 1]. The continuous wave Doppler across the tricuspid valve showed peak velocity of 1.2 m/s [Figure 2b]. Clinically, the patient remained asymptomatic and hemodynamically stable. A decision for surgical removal of the ASOD and closure of the VSD was taken.

In the operating room, after establishing intravenous access, ASA standard monitoring and invasive arterial

monitoring, anesthesia was induced with titrated dose of propofol and fentanyl. Injection vecuronium was used to facilitate endotracheal intubation. The maintenance of anesthesia was achieved with inhalation of isoflurane, fentanyl 1 µg/kg/h and intermittent boluses of vecuronium. A TEE probe (iE 33, Philips Ultrasound, Bothell WA, USA) was inserted into the esophagus after securing the airway. Central venous cannulation was done through the left internal jugular vein with continuous monitoring under TEE to reduce the possibility of ASOD displacement by the guide wire or catheter. Intraoperative TEE confirmed the TTE finding of tricuspid inflow obstruction and showed the presence of mild TR [Figure 3a and b and Video 2]. Transgastric inflow view confirmed the gradient across tricuspid valve (peak/mean 7/3 mmHg) [Figure 3c]. Mid-esophageal 4 chamber view and modified 4 chamber view with retroflexion of probe showed that the device is malaligned such that the right ventricular rim of the device override the septal tricuspid leaflet and encroached on the anterior and posterior tricuspid valve leaflets resulting in improper opening and closing of the valve [Figure 4a, b and Videos 3 and 4]. Cardiopulmonary bypass (CPB) was instituted using bicaval cannulation under continuous monitoring of TEE to prevent potential ASOD embolization. Intraoperatively, the surgeons found the ASOD stuck in the tricuspid valve. The device was partly entangled in the margin of anterior tricuspid leaflet



Figure 1: Postdeployment angiography showing the normally positioned Asymmetrical septal occluder device across the Ventricular septal defect with minimal flow across the device

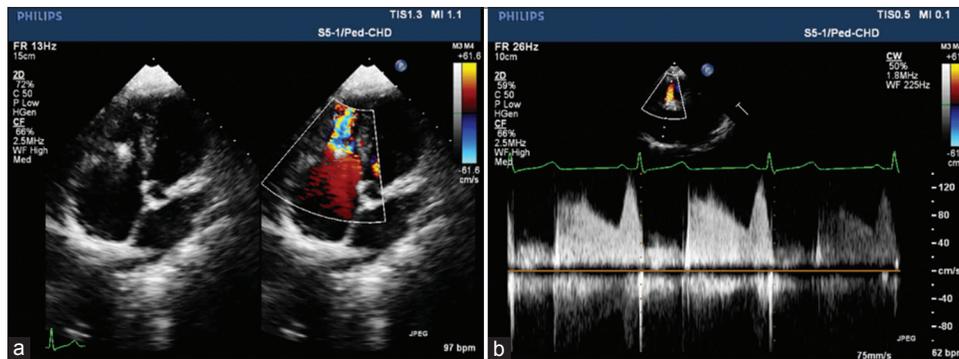


Figure 2: Transthoracic apical 4 chamber view showing the Asymmetrical septal occluder device in the tricuspid valve inlet causing turbulence across the tricuspid valve and mild aortic regurgitation (a) and continuous wave Doppler across the tricuspid inlet measuring peak velocity of 1.2 m/s (b)

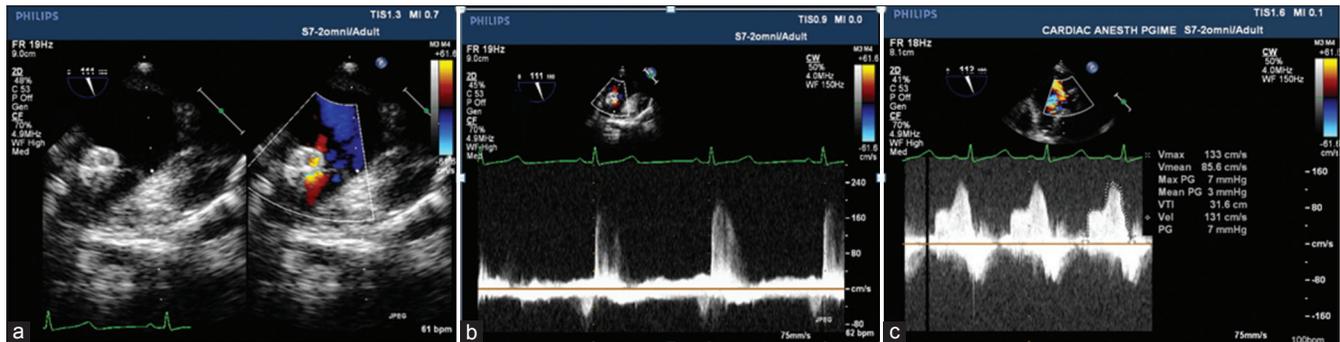


Figure 3: Midesophageal modified bicaval view showing Asymmetrical septal occluder device in the tricuspid valve inlet causing mild tricuspid regurgitation (a), Continuous Wave Doppler across tricuspid valve measuring tricuspid regurgitation (b), Transgastric tricuspid inflow view with Continuous Wave Doppler across tricuspid valve measuring a gradient of 7/3 mmHg (c)

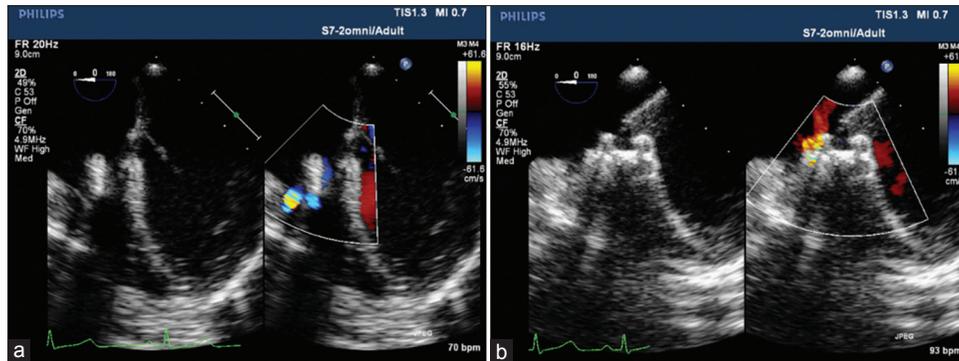


Figure 4: Mid esophageal 4 chamber view showing malaligned Asymmetrical septal occluder device with right ventricular rim overriding the septal tricuspid leaflet and encroaching upon anterior tricuspid leaflet (a) and modified mid esophageal 4 chamber view showing malaligned Asymmetrical septal occluder device with right ventricular rim overriding the septal tricuspid leaflet and encroaching on the posterior tricuspid valve leaflet (b)

and the chordae of septal, anterior and posterior leaflets. The device was retrieved with utmost care to prevent further damage to the leaflets, chordae tendineae, and the conduction system. Visual inspection of the tricuspid valve after removal of device showed tear in the anterior leaflet and ruptured chordae tendineae of septal leaflet. The anterior tricuspid leaflet was repaired with primary suturing. The VSD was closed with poly tetra fluoro-ethylene patch. CPB was terminated using nor-adrenaline 0.05 µg/kg/min. TEE after termination of CPB revealed no residual VSD; the pulse wave Doppler across tricuspid valve showed no gradient (3/1 mmHg) and ruptured chordae tendineae of septal leaflet causing eccentric TR jet [Figure 5 and Video 5]. After surgery, the patient was shifted to intensive care unit for elective mechanical ventilation which was weaned off after 4 h. He was discharged home after an uneventful stay of 5 days in the hospital.

Discussion

Isolated VSD comprises 20% of all CHD.^[7] The development of congestive heart failure, pulmonary over-circulation causing recurrent respiratory tract infection, and pulmonary hypertension necessitates the closure of these VSD once detected with few exceptions like small muscular VSD which may close spontaneously as the child grows.^[8] Surgical closure of VSD is associated with complications like residual defects, damage to the adjacent tricuspid valve, aortic valve, and conduction systems besides inflammatory and stress response of CPB on various organ system. Despite advancement in the surgical and CPB techniques reducing the above-mentioned complications, the ease of closure and the avoidance of CPB-related complication make the percutaneous technique a preferred approach over surgical closure. First attempt at closing a VSD using device was done using Rashkind umbrella device designed for closing patent ductus arteriosus.^[9] With the development of ASOD, most of the isolated VSD are now successfully closed in the catheterization laboratory. The need of the adequate rim for the placement of these ASODs makes perimembranous and

muscular VSD suitable for this approach. Since in our case the defect was perimembranous with adequate rims we planned for ASOD closure.

Even though the success rate of complete closure using the ASOD is high (>90%),^[10] it is not without risks. In the immediate postdeployment period, device malposition and embolization often occur while in the long-term conduction block, injury to the valvular structure causing TR and/or AR, left ventricular outflow tract obstruction and hemolysis may occur.^[1,11-16] The incidence of postdevice deployment conduction blocks vary from 1% to 5%.^[5,6,17] TR and AR are not infrequent due to the associated damage to these valvular apparatus during device deployment.^[3,6,18] Mertens *et al.*^[18] reported severe TR 1 month after the successful device closure of the VSD. On retrospective analysis of the cine imaging, they noted the entrapment of the chordae into the device during deployment which did not cause much problem in the immediate postprocedure period whereas the repeated stress on the device screw assembly due to each cardiac contraction may have led to metal fatigue leading to its rupture and entrapment of the ruptured segment in the chordal structures.

The TS is a rare and serious complication that requires attention in the postprocedure period. Till now only two cases of device-induced TS has been reported in the literature, Arora *et al.*^[6] reported a case of TS in the study of a large group of VSD device closure patients that was relieved by balloon dilatation. Christiani *et al.*^[10] found TS immediately following postdevice deployment due to entrapment of the right ventricular disc in the chordae of the anterior tricuspid leaflet which was relieved by chance when they tried to retrieve the device. They proposed that right coronary Judkin catheter that was passed through the defect from the arterial side would have passed through the chordae of the anterior leaflet of tricuspid valve given its proximity to the VSD, thus causing the entrapment of the right ventricular disc into the chordal structure during the device deployment. They suggested that device entrapment in the adjacent structures may be suspected whenever the ventricular disc does not get its prescribed shape after

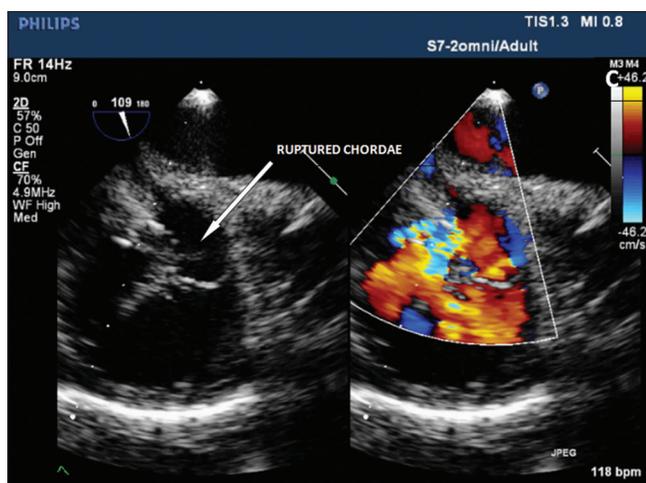


Figure 5: Mid esophageal modified 4 chamber view showing ruptured chordae tendinae of septal leaflet of tricuspid valve causing eccentric regurgitant jet

deployment of the device and to try manipulation of the adjacent structures such as tricuspid valvular apparatus to relieve the device entrapment before approaching surgical retrieval; however, danger of device displacement and embolisation should be kept in mind before manipulation. The device malalignment may be fatal at times as in the Christiani *et al.*^[10] report, TS caused pressure elevation in the right atrium leading to shunting of blood through patent foramen ovale (PFO) into the left atrium causing desaturation and relief of right atrial pressure, however, in the absence of PFO same incident may have been fatal before detection of the cause.

In our case, immediately after deployment cine study showed the normal shape and placement of the device with the closure of the VSD, however, postdevice placement day 1 TTE showed device in the tricuspid valve inlet causing TS. TEE confirmed the TTE finding and showed that TS was due to malalignment of the device. Probably in our case, part of tricuspid valve chordae got entrapped in the device during deployment that was not significant enough to cause tricuspid valve functional abnormality at that time. With the repeated cardiac contraction, the device may have got entangled into the tricuspid valve apparatus causing malalignment resulting in TS and damage to the valve leaflets due to friction force caused by repeated movement of valve on the ASOD.

Few strategies suggested to prevent entrapment of chordal structure in the device includes straightening of the wire after snaring to look for any kinking in the wire, monitoring for TR or TS during the passage of the device delivery sheath across the VSD and using curved catheter to cross the VSD from left ventricular side in place of Judkin catheter thus reducing the chance of wire passing through the chordae.^[18] In our case, although Judkin catheter was used there was no signs of abnormal flow pattern across tricuspid valve after device deployment, suggesting that

entrapment of the tricuspid valve apparatus was not severe enough to produce a change in the inflow pattern in the immediate postprocedure period. Once the complications are diagnosed, the best treatment strategy should be surgical removal of the device and closure of the defect, as an attempt to remove through percutaneous technique may lead to adverse effects such as damage to tricuspid valve apparatus, adjacent conduction system, and device embolization. This case report adds to the importance of echocardiographic evaluation of the tricuspid valve apparatus both during the device deployment, in the immediate postdeployment period and during long-term follow-up.

Conclusion

Tricuspid valve stenosis although rare may present few days after device deployment warranting monitoring before discharge from the hospital. A special attention should be given to avoid entanglement of device in the tricuspid valve chordal structures to avoid this complication. TEE may play crucial role to find the cause of valve dysfunction after device deployment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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