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Brief Report

Catheter closure of coronary sinus atrial septal defect using Amplatzer Septal Occluder

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Abstract Coronary sinus defect is a rare type of atrial septal defect. We report two patients who had a coronary sinus atrial septal defect without persistent left superior caval vein, where the orifice of the coronary sinus was closed using the Amplatzer Septal Occluder. The procedure was successful, without any complications including conduction disturbance.

Keywords: Adult congenital cardiac disease; catheter intervention; atrial septal defect

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Coronary sinus defect is a rare type of atrial septal defect in which communication occurs between the coronary sinus and left atrium as a consequence of a partial or complete absence of the coronary sinus septum, and it comprises less than 1% of atrial septal defects.[1] Coronary sinus defect can be considered as a type of unroofed coronary sinus syndrome. Unroofed coronary sinus syndrome is frequently associated with persistent left superior caval vein and other cardiac anomalies. Therefore, surgical repair is the standard treatment for patients with unroofed coronary sinus.[2–4] Closure of the coronary sinus orifice, resulting in coronary sinus flow draining into left atrium, is one of the operative options in patients who have unroofed coronary sinus without persistent left superior caval vein.

We report two adult patients who had coronary sinus defect without persistent left superior caval vein, and the orifice of the coronary sinus was closed using the Amplatzer Septal Occluder (AGA Medical Corporation, Plymouth, Minnesota, United States of America).

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Case 1

A 67-year-old man with exertional dyspnoea was referred to our institution for catheter closure of the atrial septal defect. The transthoracic echocardiography showed left-to-right shunt across the atrial septal defect, which was difficult to distinguish from the coronary sinus orifice. The transoesophageal echocardiogram showed unroofed coronary sinus, and the maximum diameter of the atrial septal defect was 8 millimetres, with an adequate rim around the defect, except for the deficient posterior rim. Cardiac contrast-enhanced computed tomography showed normal pulmonary venous return and the absence of the persistent left superior caval vein.

Catheter closure was performed under general anaesthesia. Pulmonary-to-systemic flow ratio was 1.57. Balloon-sizing with stop-flow technique resulted in a stretched diameter of 9 millimetres. A 9-millimetre device was deployed in a stable and proper position without any complications. The transoesophageal echocardiogram images clearly showed the unroofed portion of the septum between the coronary sinus and left atrium (Fig 1a and b). Systemic oxygen saturation before the procedure was 98% and did not change after the atrial septal

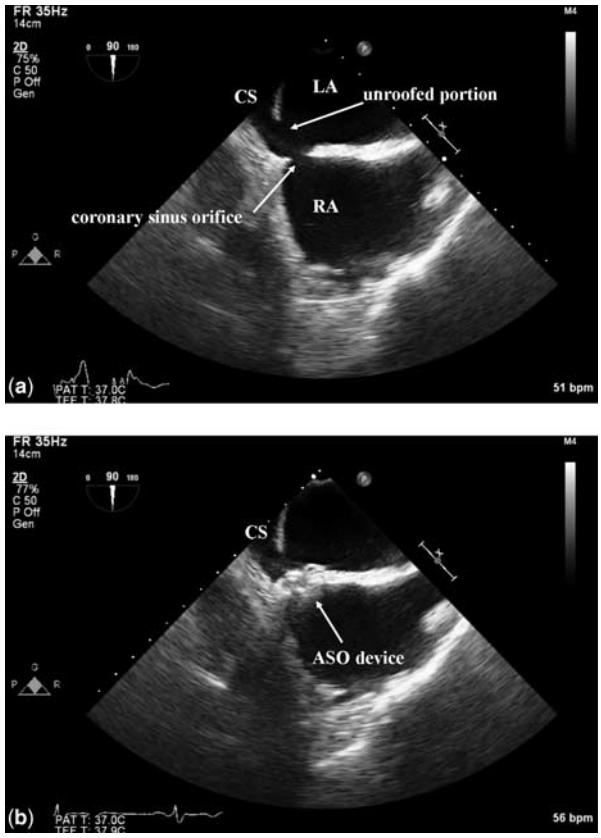

Figure 1.
*(**a**) The transoesophageal echocardiogram shows the CS opening into both the LA and the RA. (**b**) The device is deployed in the interatrial septum with the unroofed portion of the CS remaining open (CS = coronary sinus; LA = left atrium; RA = right atrium; ASO = Amplatzer Septal Occluder).*

defect closure (98%). At 24 months after the procedure, there were no complications, including atrioventricular blocks, and the patient had normal systemic oxygen saturation (98%).

Case 2

A 68-year-old man with exertional dyspnoea and a history of syncope was referred to our institution for catheter closure of the atrial septal defect. The transoesophageal echocardiogram showed unroofed coronary sinus, and the maximum diameter of the defect was 7 millimetres with adequate septal rim. The contrast echocardiography from the left arm did not reveal a flow of persistent left superior caval vein.

Catheter closure was performed under general anaesthesia. Pulmonary-to-systemic flow ratio was 1.69. A 9-millimetre device was selected by balloon-sizing and the device was deployed in a stable and proper position without any complications (Fig 2a and b). The transthoracic echocardiography after the procedure showed a considerably large unroofed portion of the coronary sinus with the device deployed in a stable position closing the interatrial left-to-right shunt (Fig 2c). Systemic oxygen saturation was 98% before the catheter closure and remained at the same level at discharge (98%). Desaturation was not observed around the catheter closure. At 19 months after the procedure, there were no complications, including atrioventricular blocks, and the patient had normal systemic oxygen saturation (98%).

Discussion

To our knowledge, this is the first case report of closing the orifice of the coronary sinus using an Amplatzer Septal Occluder device in patients who had unroofed coronary sinus without persistent left superior caval vein. There are four previous case reports of catheter treatment in cases with unroofed coronary sinus.[5–8] Of the four case reports, one showed catheter closure of the coronary sinus defect with persistent left superior caval vein using the Amplatzer Septal Occluder device. In this case, the unroofed portion of coronary sinus was closed and persistent left superior caval vein was draining into the right atrium after the closure.[5] Two reports showed catheter treatment for a persistent left superior caval vein using a caval vein filter and coil,[6] as well as a vascular occlusion device.[7] Another report showed catheter treatment for the unroofed portion of the coronary sinus using a covered stent.[8] All these reported cases were complicated with persistent left superior caval vein; however, our cases were not.

Previous reports have classified the morphology of unroofed coronary sinus into four types.[1] Diagnosis of the coexistence of persistent left superior caval vein is important because it may cause systemic embolisation and/or generalised cyanosis due to right-to-left shunt and it may also alter the strategy for treatment, including the method of anatomical reparation in the surgical procedure.[4,9] Both of our cases had developed partially unroofed portion of the coronary sinus without persistent left superior caval vein, and therefore our cases belonged to partially unroofed terminal portion of the coronary sinus type of Kirklin and Barratt-Boyes classification.

In cases with partially unroofed terminal portion of coronary sinus, the coronary sinus orifice opens into the left atrium rather than the right atrium and the localised aperture of the unroofed portion is just before the orifice of the coronary sinus. In this type of unroofed coronary sinus, standard surgery involves closure of the unroofed portion or allowing the coronary sinus to open into the left atrium by closure of the orifice of coronary sinus. Our cases were not complicated with persistent left superior

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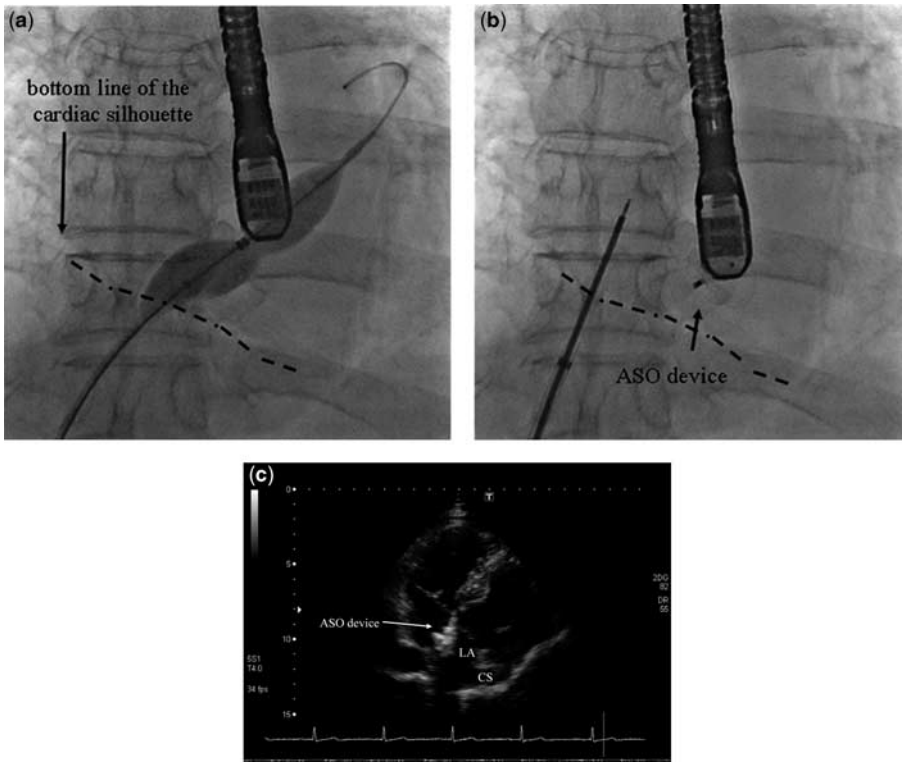

Figure 2.
*(**a**) The cineangiographic image of balloon-sizing in the anteroposterior projection shows that the waist of the balloon is positioned lower than usual for secundum-type atrial septal defect. (**b**) The device is deployed with stability in the same position as the waist of the balloon. (**c**) The transthoracic echocardiogram shows a large unroofed portion of the CS with the device deployed in the interatrial septum (ASO = Amplatzer Septal Occluder; CS = coronary sinus; LA = left atrium).*

caval vein, and therefore there was clinical validity for the closure of the atrial septal defect, which corresponded to the orifice of the coronary sinus. Catheter closure of the atrial septal defect is an accepted procedure worldwide, even in adults.[10] In patients with partially unroofed terminal portion of the coronary sinus-type unroofed coronary sinus, surgical closure of the coronary sinus orifice or aperture of the unroofed portion of the coronary sinus is generally selected. In surgical procedures, conduction disturbances, including complete atrioventricular blocks, can lead to operative complications and caution is required during the procedure to avoid these complications.

In our cases, there were no additional conduction disturbances during the procedures and during the follow-up period. In addition, even though the coronary venous flow drained into the left atrium, there was no change in systemic oxygen saturation when observed immediately after occlusion, as well as during our follow-up period. Long-term follow-up is important for the evaluation of systemic desaturation due to drainage of coronary venous flow into the left atrium. The findings from our cases suggest that catheter closure might be not disadvantageous for conduction disturbance compared with standard and conventional surgical reparation in the acute or mid-term period after the procedure. Because the compact atrioventricular node, conduction tract and the branching bundle lie beneath the transitional cells or the atrial overlay cells,[11] mild compression from the surface by device might not damage the conduction tract.

In conclusion, our experiences suggested that catheter closure of coronary sinus atrial septal defect is a simple and novel approach. A precise diagnosis of the unroofed coronary sinus and information about the presence of persistent left superior caval vein is mandatory for indication of this procedure.

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