CASE REPORT

A massive left-to-right shunt due to delayed spontaneous perforation of polyvinyl alcohol membrane of atrial septal occluder

Atriyal septal defekt kapama cihazının polivinil alkol membranının spontan gecikmiş perforasyonuna bağlı yoğun soldan sağa şant

Serdar Bozyel, M.D.,[1] Tayfun Şahin, M.D.,[2] Emir Dervis, M.D.,[2] Müjdat Aktaş, M.D.,[2] Hüseyin Şaşkın, M.D.[3]

[1]Department of Cardiology, Derince Training and Research Hospital, Kocaeli, Turkey
[2]Department of Cardiology, Kocaeli University Faculty of Medicine, Kocaeli, Turkey
[3]Department of Cardiovascular Surgery, Derince Training and Research Hospital, Kocaeli, Turkey

Summary– Percutaneous closure of an atrial septal defect (ASD) has emerged as an alternative to surgery. A 54-year-old woman with a history of percutaneous ASD closure with a 30-mm Cardia Ultrasept septal occluder (Cardia Inc., Eagan, MN, USA) comprising 2 discs made of Nitinol wire mesh covered with polyvinyl alcohol (PVA) membrane, was admitted to the hospital with unstable angina pectoris. In a routine examination, transthoracic echocardiography revealed a left-to-right shunt through the device. Transesophageal echocardiography (TEE) also demonstrated significant left-to-right shunt through the central portion of the prosthesis. Coronary angiography was performed, which disclosed severe stenosis in the right and left anterior descending coronary arteries. Three-dimensional TEE showed multiple perforations of the PVA membrane with intact nitinol frame. Surgical removal of failing device and closure of the ASD with a pericardial patch was performed together with coronary artery bypass graft surgery. On perioperative view, the device appeared to have been correctly implanted, and the device frame was completely intact; however, the PVA membrane of both the right and left discs had almost completely disappeared and there was incomplete endothelialization around the frame. Surgeons must be aware of this rarely seen complication and they should re-examine all patients implanted with Cardia devices in regular follow-up examinations for a long period of time.

Özet– Perkütan girişimle atriyal septal defektin (ASD) kapatılması cerrahiye seçenek olarak ortaya çıkmıştır. Polivinil alkol (PVA) ile kaplı iki nitinol diskten yapılmış 30-mm Cardia Ultrasept Septal oklüder (Cardia Inc, Eagan, MN, USA) cihazı ile perkütan yolla ASD kapatılma öyküsü olan 54 yaşında bir kadın hasta kararsız anjina pektoris tanısı ile hastanemize kabul edildi. Transtorasik ekokardiyografide cihazın içinden soldan sağa şant izlendi. Transözofajiyal ekokardiyografide (TÖE) ise cihazın orta bölümünden geçen soldan sağa geçişli önemli miktarda şant gösterildi. Hastaya koroner anjiyografi yapıldı ve sağ koroner ve sol ön inen koroner arterlerde ciddi darlık saptandı. Üç boyutlu TÖE'de nitinol çatı sağlam ve PVA membran üzerinde çok sayıda perforasyon görüldü. Hastanın koroner arter baypas greft ameliyatı ile birlikte ASD kapama cihazı çıkarıldı ve ASD perikardiyal yama ile tekrar kapatıldı. Ameliyat sırasındaki incelemede, cihazın doğru bir şekilde yerleştirildiği cihazın çatısının tamamen sağlam olduğu ve çatı etrafındaki eksik endotelizasyon ile sağ ve sol disklerin tamamen kaybolduğu görüldü. İşlemcilerin bu nadir görülen komplikasyonun farkında olmaları ve Cardia cihaz yerleştirilen tüm hastaları tekrar muayene etmeleri ve uzun süre düzenli izlemeleri gerekmektedir.

Percutaneous closure of atrial septal defect (ASD) has emerged as an alternative to surgery and several devices have been developed. Cardia ASD prostheses have been available since 1998 and have been redesigned several times. The Ultrasept (Cardia Inc., Eagan, MN, USA) septal occluder consists of 2 disc frames made of Nitinol covered with polyvinyl alcohol (PVA) membrane. PVA is a water-soluble poly-

Received: December 25, 2016 *Accepted:* January 25, 2017
Correspondence: Dr. Serdar Bozyel. İbni Sina Mahallesi, Sopalı Mevki, Lojman Sokak, 41900 Derince, Kocaeli, Turkey.
Tel: +90 262 - 317 80 00 e-mail: seribra85@gmail.com

mer, which becomes insoluble for medical purposes with formaldehyde or glutaraldehyde cross-links.[1] A case of dissolution of the PVA membrane detected 2 years after implantation of Cardia Ultrasept ASD occluder in a 54-year-old woman is described.

Abbreviations:

ASD	*Atrial septal defect*
CABG	*Coronary artery bypass graft*
PVA	*Polyvinyl alcohol*
TEE	*Transesophageal echocardiography*

CASE REPORT

A 54-year-old woman with a history of percutaneous ASD closure with a 30-mm Cardia Ultrasept septal occluder in September 2014 was admitted to the emergency department with unstable angina pectoris in September 2016.

In a routine examination, transthoracic echocardiography revealed an important, recurrent, left-to-right shunt through the device. Transesophageal echocardiography (TEE) demonstrated significant left-to-right shunt through the central portion of the prosthesis and a mobile thrombus on the right atrial disc. (Figure 1a, b). Coronary angiography disclosed severe stenosis in the right and left anterior descending coronary arteries (Figure 1c, d). The Ultrasept ASD device is made of 2 discs with Nitinol frame covered with PVA membrane (Figure 2a, b). Three-dimensional TEE revealed multiple perforations of the PVA membrane of the device with intact metallic structure (Figure 2c).

Surgical removal of the failing device and closure of the ASD with a pericardial patch was performed successfully, together with a coronary artery bypass graft (CABG) surgery. On perioperative view, the device appeared to have been correctly implanted, the device frame was completely intact. On both the right and left discs of the device, the PVA membrane had almost totally disintegrated, and there was incomplete endothelialization around the frame (Figure 2d, e). Analysis of the explanted device did not indicate any evidence of infection and culture remained sterile. The device manufacturer was informed of the adverse event.

DISCUSSION

The Ultrasept ASD occluder device is the seventh generation Cardia prosthesis. It has a double, round disc design; the frame is made of Nitinol and covered

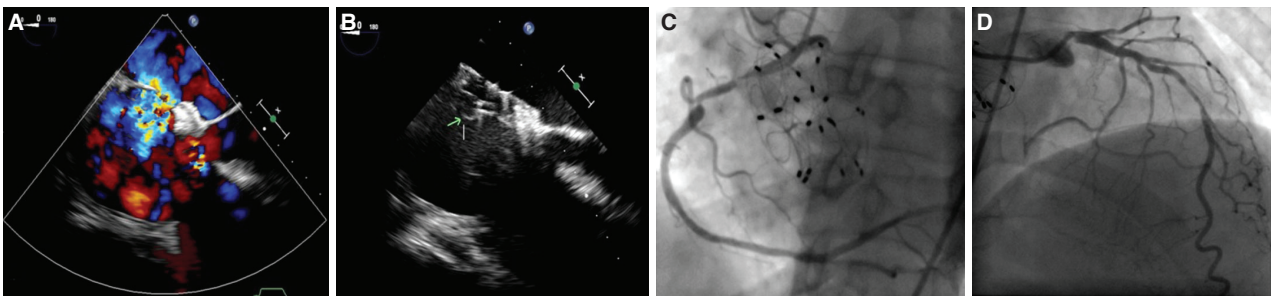

Figure 1. (A and B) Transesophageal echocardiography demonstrated important left-to-right shunt through the central portion of the prosthesis and a mobile thrombus on the right atrial disc (green arrow). **(C and D)** Coronary angiography disclosed severe stenosis in the right and left anterior descending coronary arteries.

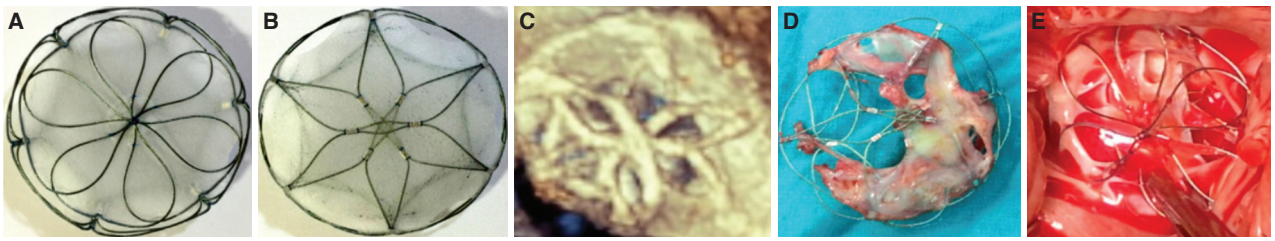

Figure 2. (A) Right atrial disc of the Cardia Ultrasept (Cardia Inc., Eagan, MN, USA) closure device, comprised of a Nitinol frame covered with polyvinyl alcohol (PVA) membrane. **(B)** Left atrial disc made of a Nitinol frame covered with PVA membrane. **(C)** Three-dimensional transesophageal echocardiography showed multiple perforations in the PVA membrane of the device with intact Nitinol frame. **(D)** Left disc of the removed device with missing PVA membrane and incomplete endothelialization around the intact frame. **(E)** Intraoperative view of the right disc of the device with virtually no PVA membrane remaining.

with PVA membrane. PVA is a synthetic polymer commonly used in medical devices due to its biocompatibility, high water solubility, and chemical resistance. This device offers a minimal metallic frame and both discs have rounded edges to minimize the risks of erosion and/or perforation. It was also developed to allow percutaneous access to the left atrium through the inserted device.[1,2]

We report a case of dissolution of the PVA membrane observed after 2 years in a patient treated with 30-mm Ultrasept ASD occluder. Similar cases have previously been reported with many versions of Cardia ASD prostheses (Table 1). Bartel et al. reported 2 cases of device failure in patients treated with the Atriasept II (Cardia Inc., Eagan, MN, USA), an older version of the device we used.[3] Bhattacharyya et al. demonstrated disintegration of PVA with the same version of the device as in our case.[4] Several other cases of device failure have also been reported with latest generation of Cardia devices (Ultrasept II ASD occluders).[5–7]

Patients with device failure were asymptomatic in most cases, but it may be revealed in dyspnea, fatigue, or multiple stroke.[4,5,7] Although minimal shunt through the device was noticed at ninth month, our patient remained asymptomatic for 2 years. Surgical removal of the failing device and repair with a patch was preferred in most reported cases. Covering the damaged membrane with a second device has been reported and may be considered an alternative to surgery.[4,7] The space between the Nitinol struts makes passage of long sheath possible in order to implant a second device without damaging the structure of the first device. Since our patient also had severe coronary artery disease at the same time, we performed a concomitant CABG and patch closure.

Table 1. Review of reported cases of patients with malfunction of polyvinyl alcohol membrane of septal occluder

First author (year)	Patient age/sex	Device Type	Device size	Recurrence of shunt after procedure	Symptom	Management
Bartel (2010)	62/NA	Atriasept II	24 mm	6 weeks	No	Endoscopic device removal
	42/NA	Atriasept II	20 mm	3 months	No	Endoscopic device removal
Aubry (2014)	77/M	Ultrasept II	24 mm	3 months	No	Surgical device removal and Gore-Tex patch repair
Bhattacharyya (2015)	69/M	Ultrasept	28 mm	10 months	Several transient neurological events	Covering the damaged membrane with a second device
Ramoglu (2016)	4/M	Ultrasept II	20 mm	1 week	No	Surgical device removal and Gore-Tex patch repair
Labombarda (2016)	19/F	Ultrasept II	26 mm	1 months	Dyspnea	Surgical device removal and Gore-Tex patch repair
	30/M	Ultrasept II	26 mm	6 months	No	
	44/F	Ultrasept II	16 mm	6 months	No	
	75/F	Ultrasept II	18 mm	11 months	No	
Chamie (2016)	28/F	Ultrasept II	20 mm	6 months	Exercise-induced fatigue	Device in device technique
	33/F	Ultrasept II	16 mm	4 months	No	Device in device technique
	49/F	Ultrasept II	16 mm	6 months	No	Device in device technique
	17/F	Ultrasept II	14 mm	14 months	No	Device in device technique

NA: Not available; M: Male; F: Female.

Neither previous authors nor the manufacturer has been able to explain the mechanism of PVA membrane disappearance. ASD device sizes used in reported cases varies from 14 mm to 28 mm, so this complication is independent of device size. We could not find any evidence of infection, a possible cause of degradation. It may be caused by early reabsorption of the PVA sail, associated with late endothelialization of the device. After a declaration of adverse events made by Labombarda et al., the manufacturer announced the development of a new consolidated device with the addition of a Gore-tex (W. L. Gore and Associates, Newark, DE, USA) patch between the 2 Nitinol discs.[5]

Conclusion

We have reported the malfunction of the PVA membrane in ASD device detected in the second year after the implantation procedure. Disintegration of the PVA membrane is rare and usually requires surgical intervention. Surgeons must be aware of this potential complication and they should re-examine all patients implanted with Cardia devices at the earliest opportunity and conduct regular follow-up examinations for a long period of time.

Conflict-of-interest: None declared.

REFERENCES

1. Alexandre N, Ribeiro J, Gärtner A, Pereira T, Amorim I, Fragoso J, et al. Biocompatibility and hemocompatibility of polyvinyl alcohol hydrogel used for vascular grafting--In vitro and in vivo studies. J Biomed Mater Res A 2014;102:4262–75.
2. Huang Y, Kong JF, Venkatraman SS. Biomaterials and design in occlusion devices for cardiac defects: A review. Acta Biomater 2014;10:1088–1101. [CrossRef]
3. Bartel T, Bonaros N, Muller S. Device failure weeks to months after transcatheter closure of secundum type atrial septal defects. Heart 2010;96:1603. [CrossRef]
4. Bhattacharyya S, Ilsley CD, Baltabaeva A. Disintegration of polyvinyl alcohol membrane covering atrial septal defect closure device. Eur Heart J Cardiovasc Imaging 2015;16:1153.
5. Labombarda F, Roule V, Beygui F. Delayed spontaneous perforation of polyvinyl alcohol membrane-Covered atrial septal defect closure devices. Catheter Cardiovasc Interv 2016.
6. Ramoglu MG, Ucar T, Tutar E. Early malfunction of polyvinyl alcohol membrane of septal occluder. Catheter Cardiovasc Interv 2016;87:151–3. [CrossRef]
7. Chamié F, Maia J, Giuliano LC. Device-in-device: A transcatheter alternative to surgical explantation of a failing atrial septal defect intracardiac prosthesis. Catheter Cardiovasc Interv 2016;88:239–43. [CrossRef]

Keywords: Atrial septal occlude; polyvinyl alcohol membrane; perforation.

Anahtar sözcükler: Atriyal septal oklüder; perforasyon; polivinil alkol membran.