

Management of intractable hemolysis after transcatheter ventricular septal defect closure with Nit Occlud® Lê ventricular septal defect coil

Nit Occlud® Lê ventriküler septal defekt koil ile ventriküler septal defektin transkateter yolla kapatılması sonrası dirençli hemolizin yönetimi

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ABSTRACT

The Nit-Occlud® Lê ventricular septal defect coil is a novel device designed for transcatheter closure of ventricular septal defects. Despite continued developments in the area of transcatheter closure of ventricular septal defects, complications remain a common occurrence. In this article, we present two perimembranous ventricular septal defect cases who required re-intervention after developing intractable hemolysis secondary to ventricular septal defect closure with a Nit-Occlud® Lê ventricular septal defect coil.

Keywords: Child; hemolysis; Nit-Occlud® Lê ventricular septal defect coil; transcatheter treatment; ventricular septal defect.

The transcatheter closure of ventricular septal defects (VSDs) was first described in 1988 by Lock et al.,^[1] in which a Rashkind double-umbrella was used for the closure. Although many different devices have been developed since along with increasing procedure success rates and significant improvements in alternative treatment methods, biomedical engineers are still searching for an optimum device for transcatheter VSD closure. The Nit-Occlud® Lê VSD coil (PFM: Produkte für die Medizin AG, Cologne, Germany) is a novel device designed for transcatheter closure of VSDs. There are very few reports on its success rates in the literature.^[2,3] Despite continued developments in the area of transcatheter VSD closure, complications remain a common occurrence.^[3,4] In this report, we present two perimembranous VSD cases who

ÖZ

Nit-Occlud® Lê ventriküler septal defekt koil, ventriküler septal defektlerin transkateter yolla kapatılması için tasarlanmış olan yeni bir cihazdır. Ventriküler septal defektlerin transkateter yolla kapatılması hususunda devam eden gelişmelere rağmen, komplikasyonlar yaygın olarak devam etmektedir. Bu yazıda, Nit-Occlud® Lê ventriküler septal defekt koil ile ventriküler septal defektin kapatılmasına ikincil gelişen dirençli hemoliz sonrası yeniden girişim gerektiren iki perimembranöz ventriküler septal defektli olgu sunuldu.

Anahtar sözcükler: Çocuk; hemoliz; Nit-Occlud® Lê ventriküler septal defekt koil; transkateter tedavi; ventriküler septal defekt.

required transcatheter re-intervention after developing intractable hemolysis secondary to VSD closure with a Nit-Occlud® Lê VSD coil.

CASE REPORT

Case 1– A 10-year-old girl weighing 28 kg was admitted for transcatheter closure of a perimembranous VSD. Doppler echocardiography showed a restrictive filling pattern and a pressure gradient of 90 mmHg. The left ventricular end-diastolic diameter Z-score was +2.1. Under general anesthesia, left ventricular angiography and real-time transesophageal echocardiography (TEE) were performed to determine the size and location of the defect (Figure 1a). The VSD was measured 9 mm from the left ventricular side and 4 mm from the right ventricular side. The mean pulmonary artery pressure



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was 19 mmHg and the pulmonary-to-systemic flow (Qp/Qs) ratio was 1.7. Transcatheter closure of VSD was performed by deploying a 12/6 mm Nit-Occlud® Lê VSD coil using the routine technique. There was small residual shunting at the center of the device at the end of the procedure (Figure 1b).

Within five hours after the procedure, the patient presented with dark brown urine. Transthoracic echocardiography (TTE) showed good positioning of the device, mild residual shunting across the ventricular septum at the center and upper edge of the device (Figure 1c), and mild tricuspid regurgitation. The next day, the patient needed an infusion of one unit of erythrocyte suspension due to low hematocrit levels. Aspartate transaminase and alanine transaminase levels increased and an increased level of urobilinogen was detected in the urine test without increased serum creatinine level. Fluid intake and intravenous hydration increased and urine was alkalinized by sodium bicarbonate. Due to persistent shunting through the device, a decision was made in favor of percutaneous re-intervention at the post-procedural day four. Under general anesthesia, left ventricular angiography and real-time TEE were performed and showed residual flow across the ventricular septum at the center and upper edge of the device. A 6.5x5 mm detachable coil was deployed close to the central axis of the original coil (Figure 1d). After deployment, left ventricular angiography showed residual flow remaining at the superior margin of the device; however, shunting was reduced at the center of the device. Due to the continued residual flow and the continuing hemoglobinuria, the patient underwent a surgical repair one day after the second procedure (Figure 1e). After surgery, urine color returned to normal within the same day and biochemical abnormalities normalized over the next several days.

Case 2– A transcatheter VSD closure procedure was performed to a 33 kg, 10-year-old girl whose echocardiographic findings were similar to the first case. The size of the VSD was 8 mm from the left ventricular side and 4 mm from the right ventricular side (Figure 2a). The Qp/Qs ratio was 1.8 with a normal pulmonary artery pressure. A 10/6 mm Nit-Occlud® Lê VSD coil was used for VSD closure. At the end of the procedure, there was minor residual shunting at the center of the device (Figure 2b).

During her follow-up in the ward, the patient presented with dark brown urine at the seventh hour. Transthoracic echocardiography showed mild residual shunting at the center of the device (Figure 2c) and mild tricuspid regurgitation. On the next day, the

patient required erythrocyte suspension transfusion due to hemolysis and aforementioned medical treatment was administered to reduce possible side effects of hemolysis. A second percutaneous intervention was performed due to persistent shunting through the device on the third day. Left ventricular angiogram and real-time TEE showed residual flow across the ventricular septum at the center and upper edge of the device. A 6x5 mm Amplatzer® Duct Occluder II was deployed close to the central axis of the original coil (Figure 2d). Repeated angiogram showed no residual flow across the VSD (Figure 2e). After the procedure, urine color returned to normal within the same day.

DISCUSSION

Many studies have demonstrated the development of intravascular hemolysis following transcatheter closure of atrial septal defects (ASDs), VSDs, and patent ductus arteriosus (PDA). In the study conducted by Zuo et al.,^[5] out of 301 patients in whom the VSDs were closed using the Amplatzer® perimembranous VSD device, intravascular hemolysis developed in two cases (0.7%) and the patients reportedly recovered with medical treatment within one week. In another study by Gu et al.,^[6] it was reported that out of 26 patients with postoperative residual VSDs which were closed using symmetrical and asymmetrical perimembranous VSD devices, three of them developed hemolysis within 12 hours after the procedure and that these patients recovered after four days, eight days, and four weeks, respectively. In the study carried out by Li et al.,^[7] it was shown that out of 223 patients with perimembranous VSDs closed transcatheterly, three of them (1.3%) developed hemolysis and recovered with corticosteroids. Similarly, in our previous report, three out of 20 VSD patients implanted with the Nit Occlud® Lê VSD coil, two had moderate residue, one had mild residue, and all three developed hemolysis.^[3] In two patients, hemolysis regressed during the follow-up without any need for medical treatment. One of our patients with intractable hemolysis (Case 1) required surgery after an additional detachable coil deployed inside the original device proved ineffective. Of 24 patients who underwent VSD closure with a Nit Occlud® Lê VSD coil at our center to date, Case 2 was the fourth case of hemolysis requiring re-intervention using an Amplatzer® Duct Occluder II. In the first case, we used a detachable coil; however, the patient required surgery eventually. Therefore, we believe that it is more advisable to use a device with an acute occluding property instead of an additional coil in patients who develop intractable hemolysis after VSD closure with a Nit Occlud® Lê VSD coil.

The design of the Nit Occlud® Lê VSD coil is specifically appropriate to close the defects without an aortic rim particularly with ventricular septal aneurysms.^[3] It is also considerably useful to close a defect without any major complication in the cardiac conduction system.^[2,3] Thanks to its mobility and stretchable structure, it offers an opportunity to shape the device for the defect. Nonetheless, if a residual shunt remains after procedure, the development of

hemolysis is much more possible than the other devices, as the angle between the Dacron fibers of the coil and the direction of the shunt flow creates a more suitable situation for mechanical destruction of erythrocytes.

In conclusion, it should be kept in mind that hemolysis may develop after transcatheter closure of VSDs with a Nit-Occlud® Lê VSD coil. Although hemolysis usually tends to be self-limiting or responsive to treatment, it may also be intractable in certain cases.

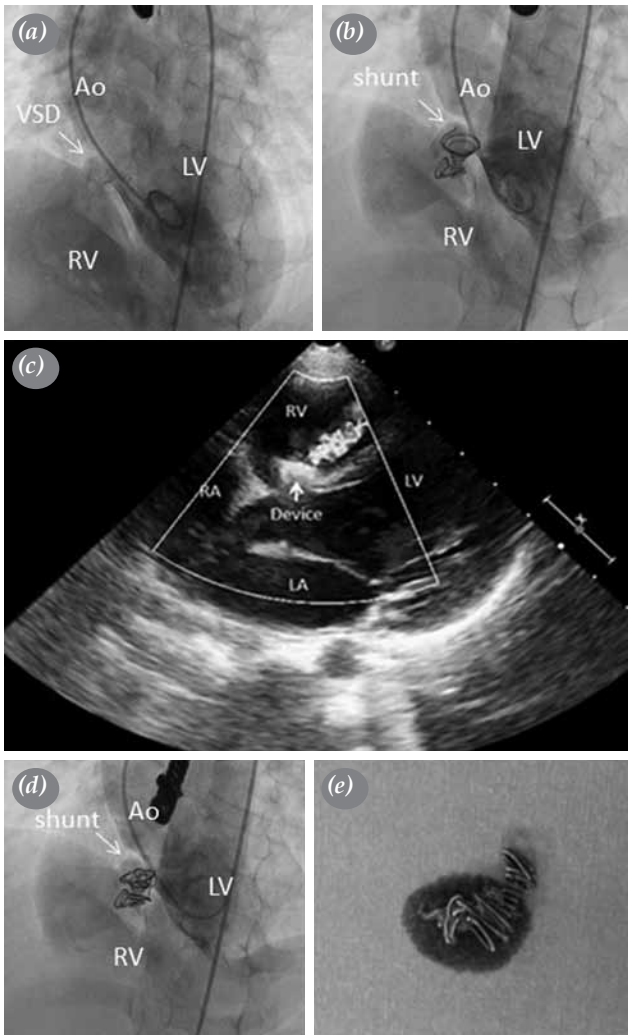


Figure 1. (a) Angiographic view of left ventricular injection showing a ventricular septal defect with significant left-to-right shunting. (b) Post-procedural control injection shows mild residual shunting through the Nit-Occlud® Lê ventricular septal defect coil. (c) Long-axis view of the left ventricle showing residual shunting through the coil. (d) Angiographic view after the second procedure showing a detachable coil implanted inside the Nit-Occlud® Lê ventricular septal defect coil and continued residual shunt. (e) Surgically removed material of the detachable coil implanted in Nit Occlud® Lê VSD coil.

Ao: Aorta; VSD: Ventricular septal defect; LV: Left ventricle; RV: Right ventricle; RA: Right atrium; LA: Left atrium.

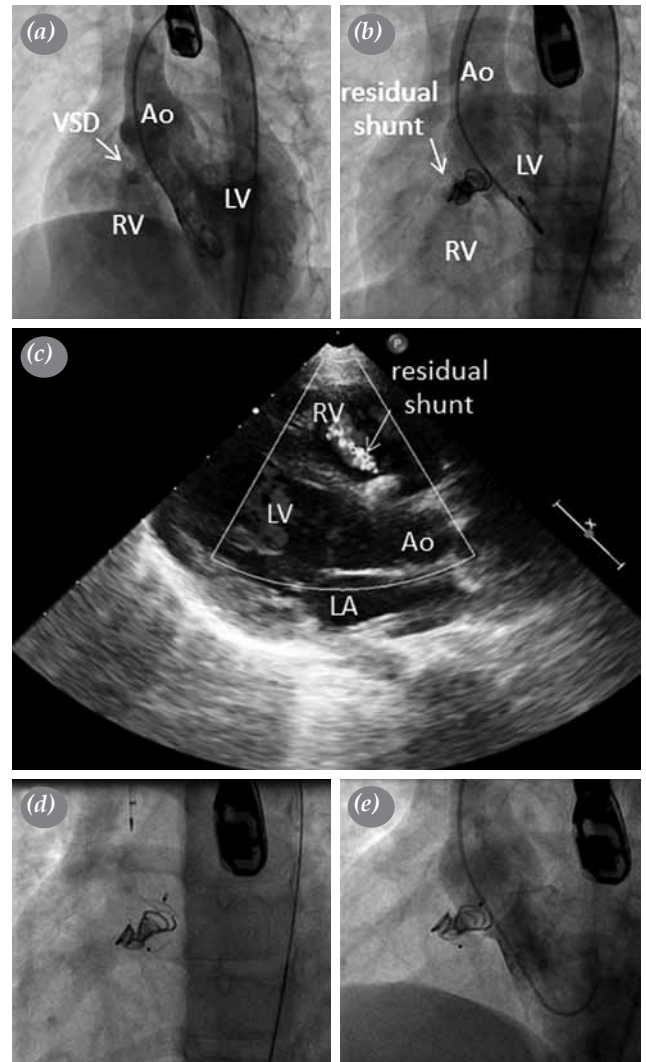


Figure 2. (a) Angiographic view after left ventricular injection showing a ventricular septal defect with significant left-to-right shunting. (b) Post-procedural control injection showing mild residual shunting through the Nit-Occlud® Lê ventricular septal defect coil. (c) Long-axis view of the left ventricle showing residual shunting through the coil. (d) Angiographic view of the Amplatzer® Duct Occluder II device implanted inside the Nit-Occlud® Lê ventricular septal defect coil. (e) Repeated left ventricular angiogram showing no residual shunt.

Ao: Aorta; VSD: Ventricular septal defect; LV: Left ventricle; RV: Right ventricle; LA: Left atrium.

In such cases, re-intervention is required to deploy an additional device with an acute occlusive property.

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