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*Executive office:*
Türk Kalp ve Damar Cerrahisi Derneği
Ataşehir Mah., Ataşehir Bulvarı, 48 Ada,
Mimoza 2/2, K: 2, D: 6,
34758 Ataşehir, İstanbul, Turkey
Tel: +90 216 - 456 14 54
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e-mail: info@tkdcd.org
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*Responsible Manager*
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*Editorial Contact Person*
Anıl Z. Apaydın, MD.
e-mail: anil.apaydin@ege.edu.tr

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Örnek Mah., Dr. Suphi Ezgi Sok., Saray Apt.,
No: 11, D: 6, 34704 Ataşehir, İstanbul, Turkey
Tel: +90 216 - 317 41 14
Fax: +90 216 - 317 63 68
e-mail: info@baycinartibbiyayincilik.com

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Cardiovascular Surgery and Interventions

Case Report Open Access

# Ruptured spontaneous coronary artery dissection in a postmenopausal woman

Atike Tekeli Kunt,[1] Serdar Akgün,[2] Koray Ak,[3] Selim İsbir,[3] Sinan Arsan[3]

Received: March 27, 2014 Accepted: July 03, 2014

Spontaneous coronary artery dissection and rupture are extremely rare conditions. We report a 54-year-old postmenopausal female case of ruptured spontaneous coronary artery dissection who presented with severe chest pain. No electrocardiographic abnormalities were seen. Serum troponin level was normal. Although acute aortic dissection was suspected, contrast computed tomography revealed pericardial effusion. The patient was taken to catheterization laboratory for coronary angiography. Catheterization showed a ruptured spontaneous dissection of the left anterior descending artery without any other atherosclerotic lesions. She underwent coronary artery bypass grafting. Spontaneous coronary artery dissection and rupture should be kept in mind in differential diagnosis of chest pain.

Keywords: Coronary artery; dissection; spontaneous; rupture.

Spontaneous coronary artery dissection (SCAD) and rupture are extremely rare conditions with a poor prognosis. Ruptured SCAD is often fatal, unless the patient arrives late and early diagnosis can be made. It is associated with an increased rate of cardiac tamponade and requires an urgent bypass surgery. Mostly, SCAD is seen in young women (mean age of onset: 35 years) with the left anterior descending artery (LAD) being the most affected artery.[1,2] Herein, we present a clinical case of ruptured SCAD of LAD.

## CASE REPORT

A 54-year-old postmenopausal female patient was admitted with a severe chest pain. Physical examination and vital signs were normal on admission. No electrocardiographic abnormalities were seen. Serum troponin level was normal. Contrast computed tomography (CT) was performed based on the suspicion of acute aortic dissection. However, CT revealed an isolated pericardial effusion. The patient was then transferred to cardiac catheterization laboratory. Catheterization showed a ruptured spontaneous dissection of LAD with a double lumen structure without any other atherosclerotic lesions (Figures 1 and 2). She was successfully managed with coronary artery bypass grafting (CABG) where the left internal thoracic artery was grafted to the LAD with cardiopulmonary bypass and the proximal LAD was ligated for bleeding management. Subepicardial hematoma was evacuated. Following a normal postoperative course, the patient was discharged in the sixth postoperative day.

## DISCUSSION

Although SCAD is known to be mainly the disease of young women in the peripartum period, it can be associated with immunological disorders including systemic lupus erythematosus, Ehler Danlos syndrome (type IV) and Kawasaki disease.[1,3] It can also be seen in middle and older aged men and women having the risks of atherosclerosis. In our case, the patient was a 54-year-old postmenopausal woman having no underlying atherosclerotic coronary artery disease. Percutaneous coronary interventions, aneurysm, trauma, Kawasaki disease and SCAD are the known factors resulting in coronary artery rupture.[1,3] The clinical presentation of SCAD usually includes the entire signs and symptoms of acute coronary syndromes, mostly mimicking myocardial infarction. However, patients usually present with

[1]Department of Cardiovascular Surgery, Kocaeli Derince Training and Research Hospital, Kocaeli, Turkey
[2]Department of Cardiovascular Surgery, Bahçelievler Medicana Hospital, İstanbul, Turkey
[3]Department of Cardiovascular Surgery, Medical Faculty of Marmara University, İstanbul, Turkey

**Corresponding author:** Atike Tekeli Kunt, M.D. Kocaeli Derince Eğitim ve Araştırma Hastanesi Kalp ve Damar Cerrahisi Kliniği, 41900 Derince, Kocaeli, Turkey.
Tel: +90 532 - 746 78 69 e-mail: atikemd@gmail.com

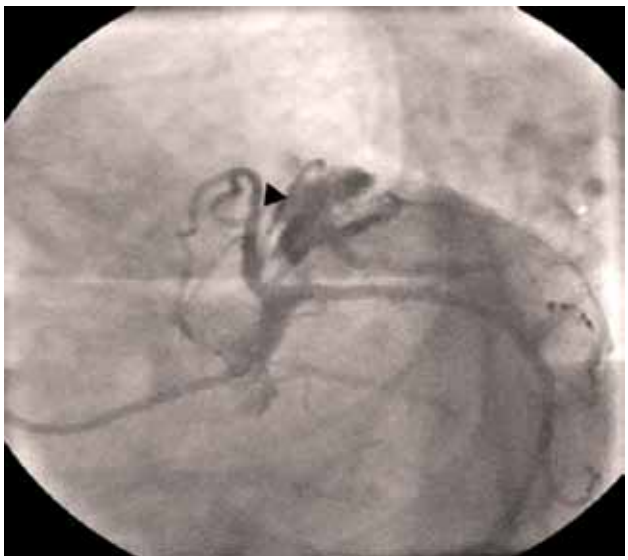

**Figure 1.** Coronary angiography showing ruptured coronary artery and pericardial contrast agent depot. (▶ denotes the extravasation area).

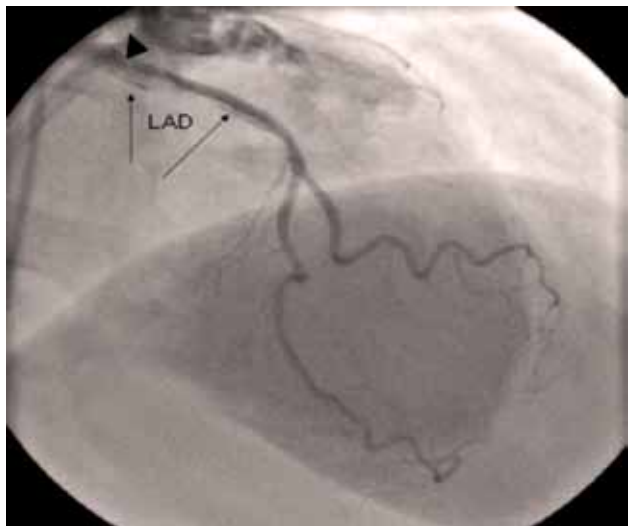


**Figure 2.** Coronary angiography showing double lumen structure of left anterior descending artery (arrows denote dissection and double lumen structure of LAD) (▶ denotes extravasation of the contrast).

cardiac tamponade and sudden death, even in case of a suspected coronary artery rupture.

The treatment strategy of SCAD solely depends on the extension of the dissection, involving coronary artery, and most importantly on the clinical presentation and overall health status of the patient. Patients with pericardial effusion or tamponade should be managed with surgery immediately. The selection of the graft and use of cardiopulmonary bypass also depends on the patient. In addition, pregnant women can be successfully managed by off-pump CABG, if surgery is indicated. Other treatment modalities include percutaneous transluminal coronary angioplasty, coronary artery stenting with cover-stents, thrombolytic and medical therapies in patients with SCAD. Spontaneous healing of SCAD has been also reported in the literature.[4-6]

In conclusion, although rare, SCAD and rupture should be kept in mind in the differential diagnosis of acute chest pain in adult patients of all ages and sex and should be individually managed.

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Case Report Open Access

# A rare location for cardiac hydatid cyst in the interventricular septum

Funda Yıldırım,[1] Barış Tuncer,[1] Adnan Taner Kurdal,[1] Tülün Öztürk,[2] İhsan İşkesen[1]

Received: February 28, 2014 Accepted: May 12, 2014

Hydatid cyst is an endemic disease seen in different regions of the world due to the Echinococcus granulosus tapeworm. The lung and liver are the most affected organs. Cardiac involvement is 0.5% to 2% of patients with hydatid disease and the interventricülar location is seen more rarely. A 17-year-old female patient with systemic hydatid cyst disease was consulted to our clinic. The patient presented with dyspnea, exertional dysrhythmia, and chest pain. Transthoracic echocardiography and cardiac magnetic resonance imaging showed the location of the interventricular septal hydatid cyst. The patient underwent elective surgery and discharged without any complications.

Keywords: Cardiac hydatid cyst; interventricular septum; operative view.

Hydatid cyst is an endemic disease seen in different regions of the world due to the *Echinococcus granulosus* tapeworm. The lung and liver are the most affected organs. Among patients who have hydatidosis, hepatic cysts occur in 70% and pulmonary cysts in 20%, but cardiac cysts are reported 0.5% to 2% of patients with hydatid disease and the interventricular location is seen rarely.[1] Cardiac hydatid cysts may rupture and cause cardiac tamponade, fatal arrhythmias or systemic infection.[2] The diagnosis of cardiac hydatid cyst is made through transthoracic echocardiography. Computed tomography (CT) and magnetic resonance imaging (MRI) are the other diagnostic tools. The treatment of cardiac hydatid cysts is surgery.

## CASE REPORT

A 17-year-old female patient with systemic hydatid cyst disease was consulted to us with cardiac interventricular septal cyst. The patient had an intracranial cyst operation approximately one month ago. Imaging studies showed two small renal cysts. There were no cysts in the liver and lung. The patient had complaints of dyspnea, effort-related dysrhythmia and chest pain for approximately three months. We planned the cardiac cyst surgery electively 54 days after the intracranial cyst operation.

On physical examination, systolic murmur was heard over the left second intercostal interval. The electrocardiogram showed right bundle branch block and the routine laboratory tests were normal. Transthoracic echocardiography showed 5x4.3 cm sized cystic lesion located in the interventricular septum (Figure 1). Before the operation, cardiac MRI imaging was obtained to identify the cyst dimensions in the septal anatomy. The cardiac MRI was reported as the compression signs to the both of the ventricles (Figure 2).

The operation was performed through median sternotomy. After opening the pericardium, the structure of cyst was palpable over the septal area. The cardiopulmonary bypass was instituted with ascending-aortic, bicaval cannulation and antegrade cardioplegia. The polyvinylpyrrolidone iodine-soaked sponges were placed into the pericardial cavity to prevent contamination. The incision was parallel to the interventricular septum. The cyst was reached by performing some dissection on the right ventricular side of the interventricular septum without opening any cardiac chamber and avoiding to damage to the left anterior descending coronary artery. The cyst content was aspirated and hypertonic solution was injected into the cyst. The germinal membranes were removed. The cavity was washed with hypertonic solution and iodinated solution, closed by capitonnage sutures, and then filled with Tisseel (Tisseel lyo 4 mlt, two component fibrin sealant, Baxter Healthcare Ltd. Caxton Way, Thetford, Norfolk, UK IP24 3SE

Departments of [1]Cardiovascular Surgery and [2]Anesthesiology and Reanimation, Medical Faculty of Celal Bayar University, Manisa, Turkey

**Corresponding author:** Funda Yıldırım, M.D. Celal Bayar Üniversitesi Tıp Fakültesi Kalp ve Damar Cerrahisi Anabilim Dalı, 45030 Manisa, Turkey.
Tel: +90 236 - 232 31 33 e-mail: fundanizamoglu@yahoo.com

**Figure 1.** Transthoracic echocardiographic image of the septal cyst.

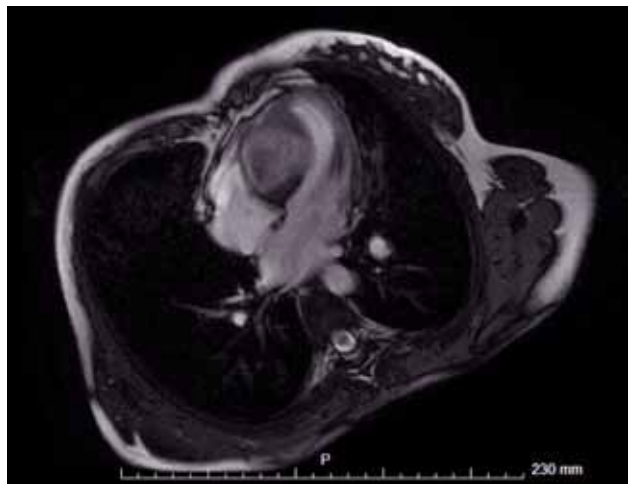


**Figure 2.** Cardiac magnetic resonance image of the septal cyst.

PL 00116/0321). The sides of myotomy were closed primarily by using Teflon felts (Figure 3, 4).

After the operation, there was no problem in the intensive care unit. The patient was discharged at the first postoperative week. The right bundle branch block was disappeared at the postoperative electrocardiographic records. The patient is still on albendazole treatment.

## DISCUSSION

Hydatid cyst disease is endemic in Turkey.[3] It is often asymptomatic at early stages. Clinical suspicion is important for the definite diagnosis. As it may affect multiple organs and systems, full body screening should be done. Cardiac involvement is 0.5-2% of the hydatid cyst patients. The interventricular septum is affected from the 4% of the cardiac cyst cases. Cardiac hydatid cysts can rupture and cause cardiac tamponade, fatal arrhythmias or systemic infection.[3] Angina secondary to coronary artery compression, anaphylactic reaction and profound circulatory collapse may follow intracavitary rupture,[4] therefore treatment is surgery. Removal of the cyst improves myocardial compliance and myocardial perfusion.[4] It also corrects cardiac deformation and strengthens myocardial contraction.[4] It has been reported that electrical activity and contractile function can be restored by removal of interventricular septal hydatid cyst.[5] Improved electrical activity was also observed in our case report, as the right bundle branch block disappeared after the removal of the

**Figure 3.** The operation view, extraction of germinal membrane of the hydatid cyst, arrows indicate cyst membrane and interventricular septal incision.


**Figure 4.** The operation view, arrows indicate: 1- Inferior vena cava cannula, 2- Sutures of the myotomy with Teflon felts, 3- Aortic cannula. RV: Right ventricle; LV: Left ventricle.

hydatid cyst. We performed cardiac operation 54 days after the cranial cyst operation, therefore there was no risk of intracranial bleeding due to heparinization. In addition, serological markers against *Echinococcus granulosus* was negative. It decreases the risk of systemic dissemination due to the two major surgical procedures for this patient.

### Conclusion

The cardiac cysts are rare and may present with various clinical findings. Clinical suspicion is the critical issue for the definite diagnosis. Due to the possible risks of systemic distribution, cardiac cysts should be treated surgically. During surgery, perioperative measures should be taken to prevent systemic dissemination.

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# Surgical treatment of two different interventional cardiological complications at the same patient

Mustafa Seren,[1] Ömer Faruk Çiçek,[2] Ali Cemal Düzgün,[1] Salih Fehmi Katırcıoğlu[1]

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With the introduction of technological improvements, invasive cardiologic interventions have become increasingly used alternatives to surgery. Despite its numerous benefits, serious and potentially life-threatening complications of invasive cardiology interventions may occur. Herein, we report a case who developed two different complications due to an invasive cardiology intervention at different times. One of them was device embolization after transcatheter closure of atrial septal defect and the other was right ventricle perforation due to late pericardial tamponade-related percutaneous pericardiocentesis. Both complications associated with percutaneous intervention were surgically treated.

Keywords: Atrial septal defect; complication; occluder.

Atrial septal defect (ASD) is one of the most common congenital heart defects requiring procedural intervention. Transcatheter closure of ASD has gained wide popularity thanks to its high success rates, lack of scar, low morbidity, and shorter hospital stay. However, several complications have been reported, although rare, such as arrhythmia, embolization, erosion, fracture, malfunction, malposition, stroke, thrombus on device.[1] The reported incidence of early device embolization after transcatheter ASD closure with Amplatzer septal occluder (AGA Medical Corp, Golden Valley, Minn) is approximately 0.5%.[1] In this article, we report a case who developed two different complications due to an invasive cardiology intervention at different times: device embolization after transcatheter closure of ASD and right ventricle perforation due to late pericardial tamponade-related percutaneous pericardiocentesis. Both complications associated with percutaneous intervention were surgically treated.

## CASE REPORT

A 41-year-old female patient was admitted to cardiology outpatient clinic with complaints of shortness of breath and tachycardia. Transthoracic echocardiography (TTE) and transesophageal echocardiography (TEE) demonstrated a 20 mm secundum ASD with adequate rims. The patient was scheduled for the deployment of percutaneous closure device, as ASD's morphology was suitable for percutaneous closure. A 26 mm Amplatzer device was deployed successfully without any complications through catheterization. On the first day after implantation, repeated echocardiography and fluoroscopy detected device migration into the left ventricle (Figure 1). The patient was taken to operation room immediately. After institution of cardiopulmonary bypass, a longitudinal right atriotomy was performed. The edges of the ASD were retracted and the device was detected under anterior leaflet of the mitral valve. The device was gently detached from chordae and retrieved (Figure 2). Mitral valve leaflets and chordae were examined carefully for a possible traumatic injury, however, no pathology was noted. Atrial septal defect was repaired with the running suture technique. The postoperative course was uneventful. Warfarin was initiated due to atrial fibrillation. The patient was discharged on the fifth postoperative day. At two months postoperatively, the patient was admitted with shortness of breath and hypotension. International normalized ratio (INR) was high (>7) and TTE detected massive pericardial effusion. With fresh frozen plasma, INR was normalized. Echocardiography-guided percutaneous

[1]Department of Cardiovascular Surgery, Ankara Training and Research Hospital, Ankara, Turkey
[2]Department of Cardiovascular Surgery, Türkiye Yüksek İhtisas Training and Research Hospital, Ankara, Turkey

**Corresponding author:** Mustafa Seren, M.D. Ankara Eğitim ve Araştırma Hastanesi Kalp ve Damar Cerrahisi Kliniği, 06340 Altındağ, Ankara, Turkey.
Tel: +90 312 - 595 30 00 e-mail: drmseren@hotmail.com

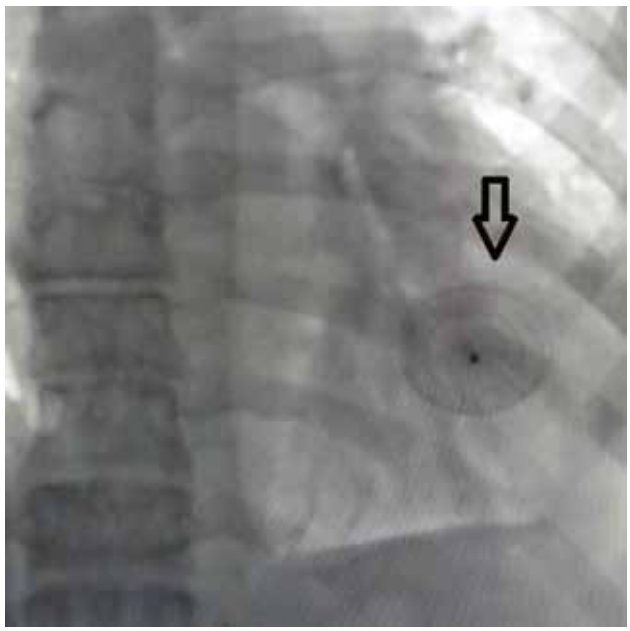

**Figure 1.** Fluoroscopy showing the Amplatzer occluder (arrow) in the left ventricle.

pericardiocentesis was performed with the subxiphoid approach. Catheter was seen in right ventricle during procedure. The patient was then taken to operation room immediately. Following median sternotomy, 1500 mL of bloody effusion was drained. Catheter from the right ventricle was removed and the puncture hole was sutured (Figure 3). The patient was discharged on the fourth postoperative day.

## DISCUSSION

Percutaneous closure of ASD in adults has emerged as an alternative to surgery. Acute failure of these devices may occur due to several reasons, the most critical condition being poor patient and/or device selection.[2] The other suggested mechanisms of acute failure are as follows: operator-related failure resulting from inadequate experience (learning curve), inaccurate placement, inadequate defect rim to hold the device, tearing of the interatrial septum, at the lower rim of the ASD during catheter, particularly, and device manipulation.[3] According to Boysan et al.[4] coughing may be an interesting reason for device embolization.

A part of the device or the whole device may embolize to the right or left atrium, to the main pulmonary artery, or even to the other parts of the

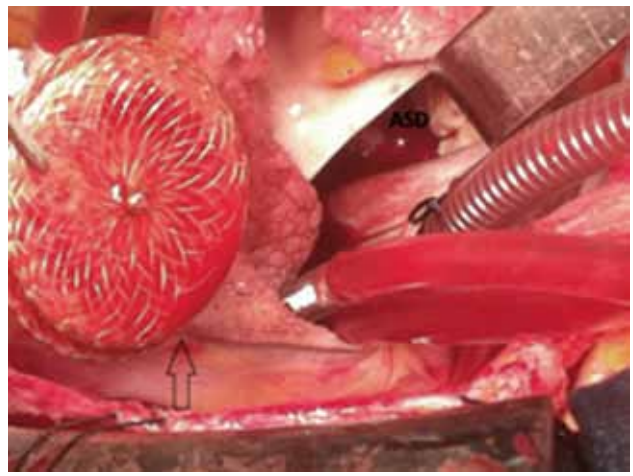


**Figure 2.** Intraoperative view of Amplatzer occluder (black arrow) and atrial transseptal approach to access the left ventricle. ASD: Atrial septal defect.

vascular tree. Embolization into left ventricle is rarer compared the right ventricle and pulmonary artery. Percutaneous retrieval of the embolized device is possible in about 70% of cases, and several techniques have been described, including the use of large sheaths, snares, or bioptomes.[5] However, some authors suggest that embolization of device is always an indication for emergency surgical retrieval, which also permits a direct inspection of intra-cardiac structures that may have become injured.[6] In our

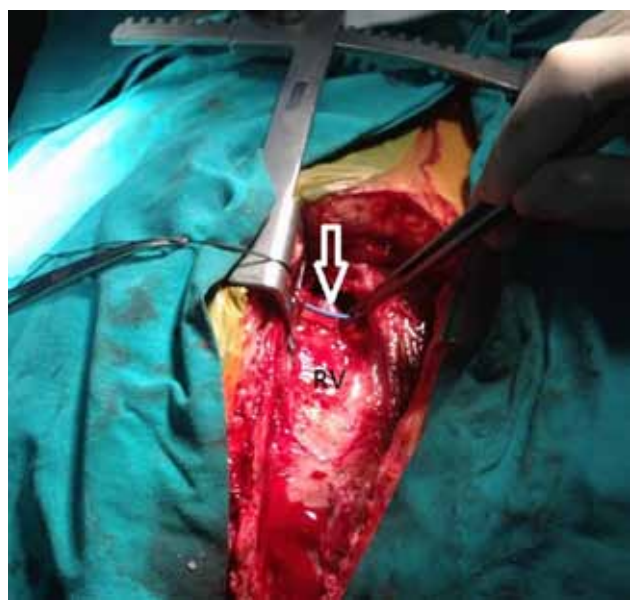


**Figure 3.** White arrow shows pericardiocentesis catheter in the right ventricle (RV).

patient, we also preferred surgical approach due to close relation of device with mitral valve in the left ventricle.

Late postoperative cardiac tamponade is an uncommon, but potentially lethal condition. Several authors have asserted that excessive anticoagulation in the postoperative period is responsible for late postoperative tamponade. Pericardiocentesis with catheter placement is highly effective and patients can be re-anticoagulated safely. In a Mayo Clinic series, echocardiography-guided pericardiocentesis was successful in withdrawing pericardial fluid or relieving tamponade in 97% of the procedures.[7] Major complications including chamber laceration, intercostal vessel injury, pneumothorax requiring a chest tube, sustained ventricular tachycardia (VT), bacteremia, and death occurred in 1.2% of patients.[8] Tsang et al.[9] from Mayo Clinic reviewed 245 patients necessitating pericardiocentesis after cardiac surgery and showed 0.8% incidence of ventricular perforation. In our case, perforation of right ventricle was diagnosed during pericardiocentesis and the patient was then taken into operation urgently.

In conclusion, a careful echocardiographic assessment and procedure planning should be done for a percutaneous intervention. In addition, surgical back-up must be available in the hospital to cope with potentially lethal acute complications.

### Declaration of conflicting interests

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Cardiovascular Surgery and Interventions

Case Report Open Access

# Ross procedure after aortic balloon valvuloplasty: the youngest case in Turkey

Tayyar Sarıoğlu,[1] Ahmet Arnaz,[2] Ersin Erek,[3] Yusuf Yalçınbaş[2]

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In this study, we report a Ross procedure which was performed on an emergent basis for severe aortic regurgitation following balloon aortic valvuloplasty in an infant with congenital critical aortic stenosis. To the best of our knowledge, this is the youngest case who was applied the Ross procedure in Turkey.

Keywords: Balloon valvuloplasty; congenital heart disease; left ventricular outflow tract obstruction; Ross procedure.

Left ventricular outflow tract obstruction (LVOTO) remains a significant challenge in neonates and infants in pediatric cardiology and cardiac surgery.[1] Residual stenosis or severe aortic regurgitation may develop in neonates and infants with critical aortic stenosis undergoing surgical valvotomy or balloon aortic valvuloplasty (BAV).[2-4] The Ross procedure and Ross-Konno procedure are one of the treatment of choices for such small babies with severe left ventricular dysfunction and heart failure.[1,5]

In this study, we report a Ross procedure which was performed on an emergent basis for severe aortic regurgitation following BAV in small baby with congenital critical aortic stenosis. To the best of our knowledge, this is the youngest case who was applied the Ross procedure in Turkey.

## CASE REPORT

A two and half-month-old premature male infant with a weight of 2.5 kg was referred to our clinic due to severe heart failure. The patient had previous BAV, when he was 30 days old. Echocardiographic examination revealed severe aortic regurgitation and valvular aortic stenosis with a gradient of 44 mmHg. Anatomy of aortic annulus, pulmonary annulus and pulmonary valve were normal. Severe left ventricular dysfunction was detected (fractional shortening: 17%). The patient with a poor health status underwent mechanical ventilation via endotracheal intubation in intensive care unit. The patient was stabilized using inotropic agents, diuretics, and blood gas analysis, and then urgently operated.

### Operative technique

Standard cardiopulmonary bypass (CPB) was initiated through median sternotomy, aortic, and bicaval cannulation. Hypothermia at 28 °C and intermittent hypothermic blood cardioplegia were applied. Aortic valve was unicuspid and complicated by rupture of the aortic annulus, clinging to one side of the valve. Pulmonary artery was transected proximal to its bifurcation and pulmonary autograft was prepared. Right and left coronary buttons were excised and the pulmonary autograft was implanted in the aortic position using aortic root replacement technique.[2] Sutures were ligated over the three thin autologous pericardial strips to strengthen neoaortic annulus and ensure blood management. Right ventricular outflow tract was reconstructed using 14 mm Bovine Jugular Vein (Contegra, Medtronic Inc., Minneapolis, Minnesota, USA). Aortic cross-clamp time was 146 minutes and CPB time was 235 minutes. On Day 7, postoperative echocardiography revealed normal left ventricular function (fractional shortening: 36%) and neoaortic valve (very mild regurgitation) (Figure 1). No pulmonary conduit stenosis was observed.

The patient was discharged at 25 days following surgery and followed every six months. At 42 months, his motor-mental development was very good with a NYHA Class I functional capacity. He remained under follow-up without any medication excluding aspirin. Repeated echocardiography every six months also revealed normal growth of autograft

[1] Department of Cardiovascular Surgery, Medical Faculty of Acıbadem University, İstanbul, Turkey
[2] Department of Cardiovascular Surgery, Acıbadem Bakırköy Hospital, İstanbul, Turkey
[3] Department of Cardiovascular Surgery, Acıbadem Atakent Hospital, İstanbul, Turkey

**Corresponding author:** Ahmet Arnaz, M.D. Acıbadem Bakırköy Hastanesi Kalp ve Damar Cerrahisi Kliniği, 34140 Bakırköy, İstanbul, Turkey.
Tel: +90 532 - 275 39 44 e-mail: ahmetarnaz@yahoo.com

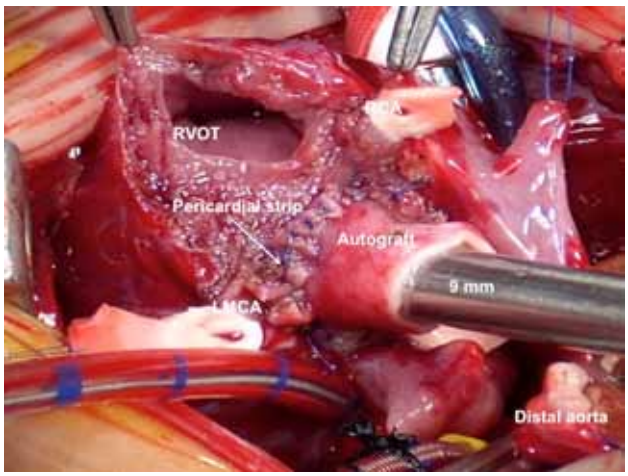


**Figure 1.** Intraoperative view. RVOT: Right ventricular outflow tract, RCA: Right coronary artery; LMCA: Left main coronary artery.

(annulus: 2.2 cm) without annular and sinotubular dilatation. There was no progress in the aortic regurgitation. The left ventricular function was very good (fractional shortening: 42%). However, valvular stenosis in the right ventricle to pulmonary artery (RV-PA) conduit was observed with a gradient of 62 mmHg.

## DISCUSSION

Open or closed surgical valvotomy or BAV may offer interim palliation in infants with critical aortic stenosis with normal biventricular heart. However, severe aortic regurgitation or residual aortic stenosis may subsequently develop.[3,4] Therefore, a rapid and radical management approach may be required, as in our case. The main disadvantages of prosthetic valve replacement in neonates and infants with critical aortic stenosis include implanting an appropriate prosthetic valve by expanding aortic annulus, need for re-do surgery in the following years, and challenges in using anticoagulants.[6] On the other hand, aortic valve replacement (AVR) using homografts is not a reasonable alternative due to the lack of growth potential and rapid degeneration of allografts in pediatric population, as well as difficulty in homograft supplying.[7]

The Ross procedure is the most reasonable treatment of choice in neonates and infants requiring AVR. However, an optimal compliance between pulmonary autograft and aortic annulus, growth potential, acceptable durability profile of pulmonary autograft in the mid- and long-term and no need for anticoagulants are the main advantages of the procedure. In addition, it can be combined with the Konno procedure in the presence of complex LVOTO.[1,5,8-10] In our case, aortic annulus was within normal size (9 mm). We would also perform the Ross-Konno procedure in case of annulus hypoplasia.[9]

Although the Ross procedure is a technically challenging and time-consuming intervention, it can be safely applied with a mortality rate below 5%.[1,4,8-10] However, there are still concerns regarding putting at a risk of both valves, autograft dilatation in the long-term and need for re-do surgery of the right ventricular outflow tract conduit.[11-13] There are several studies showing less annular and sinotubular dilatation of the autograft and aortic regurgitation in neonates and infants.[1,4] Maeda et al.[1] reported that 74% (7±12.9) of infants had very mild aortic regurgitation at five years following the Ross-Konno procedure. Shinkava et al.[5] also reported that 95.2% of infants had excellent autograft functions which tended to grow with increasing age in a 10-year follow-up period. This may be explained by an ongoing histological structuring of semilunar valves (mucopolysaccharide and collagen balance) in neonates and infants. Pulmonary arteries and valves may release histological adaptation against systemic pressure, thereby leading to less autograft dilatation and dysfunction in this population.[5] In addition, improved neoaortic valve functions in the long-term following arterial switch operation support this assumption.

Nonetheless, the most important disadvantage of the Ross procedure is the risk of re-do surgery due to the possible RV-PA conduit-related complications. The Contegra valved bovine jugular vein graft (Contegra; Medtronic, Inc., Minneapolis, Minn) have a very low early re-do surgery rate for RVOT reconstruction and could be therefore used in neonates and children under the age of three years, unless a blood group-compatible homograft can be found.[13,14]

There are several reports demonstrating functional RV-PA conduit in 50-70% of the patients at 10 years.[5,7,10-12] A large-size conduit as much as possible should be placed during the initial operation to ensure a more durable RV-PA conduit. It is well-established that conduits less than 14 mm in diameter may result in early re-do surgery. In our study, we implanted a 14 mm Contegra conduit (Medtronic Inc., Minneapolis, Minn, USA), as his weight was 2.5 kg. At 42 months of surgery, conduit stenosis was

observed preserving its function. We believe that we can increase the lifespan of the conduit by pulmonary balloon angioplasty.

In conclusion, the Ross procedure and Ross-Konno procedure are among the first treatment of choices in the management of aortic valve pathologies and LVOTO in neonates and infants.

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Cardiovascular Surgery and Interventions

Case Report Open Access

# Partial endocardial cushion defect with Raghib's syndrome: a rare case report

Muhammet Akyüz, Onur Işık, Mehmet Fatih Ayık, Yüksel Atay

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Raghib's syndrome is characterized by the combination of abnormal drainage from the left superior vena cava into the left atrium, the presence of an atrial septal defect, and the absence of a coronary sinus. Although surgical repair of Raghib's syndrome is often indicated, its correction in combination with partial endocardial cushion defect is extremely rare. In this article, we present a 24-year-old female patient with partial endocardial cushion defect accompanied by Raghib's syndrome who was corrected successfully with surgery.

Keywords: Coronary sinus; endocardial cushion defect; superior; vena cava.

Absence of coronary sinus is a rare eventuality in persistent left superior vena cava (PLSVC), which is usually associated with interatrial defect.[1] In general, PLSVC drains into the right atrium through the coronary sinus (92%) or directly into the left atrium (8%, unroofed coronary sinus).[2] The latter may lead to right-to-left shunting, cyanosis, paradoxical embolism, and a misdiagnosis of Eisenmenger's syndrome.

In this article, we report a 24-year-old female case with partial endocardial cushion defect and coronary sinus atresia, unroofed coronary sinus, and PLSVC draining directly into the left atrium, which was diagnosed intraoperatively and corrected successfully.

## CASE REPORT

A 24-year-old female patient was admitted to our clinic with symptoms of dyspnea and fatigue. She had no history of rheumatologic disease or rheumatic fever or Down syndrome. The results of the physical examination were within normal limits without clubbing or cyanosis. On cardiac examination, the patient had a pansystolic murmur in the fifth intercostal space on the left side at the midclavicular line. Her vital signs were normal with an oxygen saturation of 90%. An electrocardiogram showed sinus rhythm and left axial deviation. Echocardiography demonstrated a large ostium primum defect and third-degree mitral regurgitation (Figure 1). No other associated cardiac malformations were demonstrated.

After written informed consent, median sternotomy and aortic-bicaval cannulation were performed under cardiopulmonary bypass. In the inspection of the venous anatomy, the innominate vein was absent. After aortic cross-clamping and antegrade blood cardioplegia administration, right atriotomy was performed. The coronary sinus was not visualized. Multiple Thebesian vein ostiums in the right atrium were observed. Persistent left superior vena cava was draining to the left atrial roof. A vent sucker was placed in the PLSVC and snared for good operative exposure and prevention of brain-related complications. There were normal pulmonary venous connections. There was also a cleft in the mitral valve anterior leaflet.

Surgical correction was performed initially. The cleft in the mitral valve was repaired using multiple interrupted 6-0 polypropylene sutures and the flow of the PLSVC was diverted to the right atrium including Thebesian veins with an 8 mm polytetrafluoroethylene (PTFE) prosthetic tube graft, which was cut vertically (Figure 2). The atrial septum was reconstructed with an autologous pericardial patch which was used for closing primum ASD (Figure 2).

Postoperatively, oxygen saturation improved to 99%. Following an uneventful recovery, the patient was discharged in the fifth postoperative day. Postoperative anticoagulant therapy was administered for eight weeks. At sixth months, she was free of symptoms and in sinus rhythm. Repeated echocardiography also

Department of Cardiovascular Surgery, Medical Faculty of Ege University, İzmir, Turkey

**Corresponding author:** Muhammet Akyüz, M.D. Ege Üniversitesi Tıp Fakültesi, Kalp ve Damar Cerrahisi Anabilim Dalı, 35100 Bornova, İzmir, Turkey.
Tel: +90 505 - 943 79 89 e-mail: drmak100@gmail.com

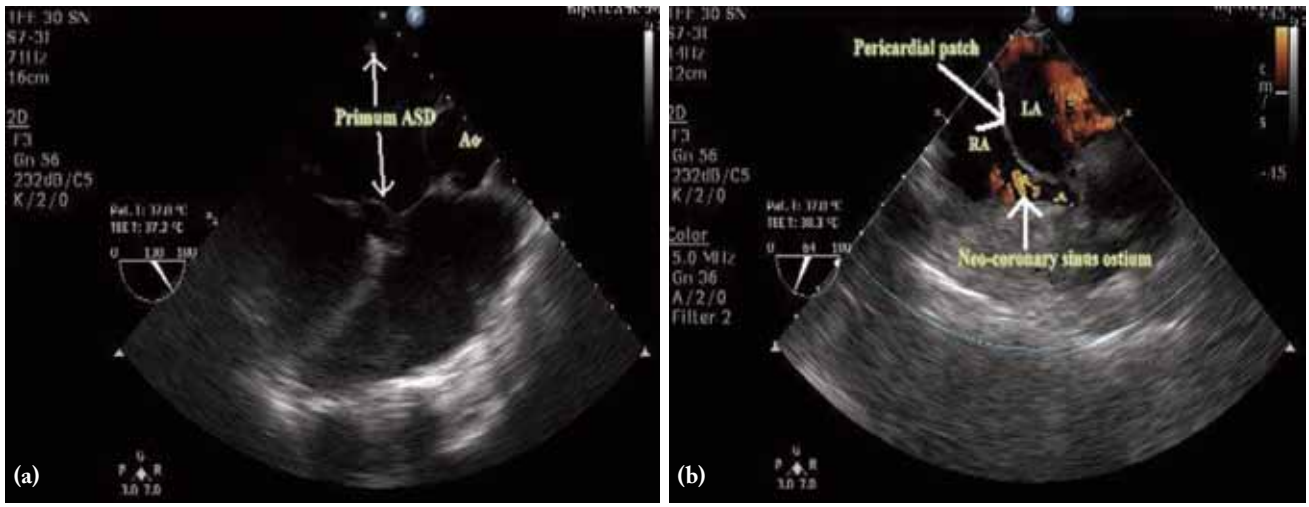


**Figure 1.** **(a)** Preoperative and **(b)** postoperative transesophageal echocardiography.

showed a mild mitral insufficiency and patent flow of the PLSVC (Figure 1).

## DISCUSSION

Persistent left superior vena cava is a congenital anomaly which affects 0.3% of healthy population and 1.3-10% of patients with cardiovascular disease.[3] An association between PLSVC and absence of coronary sinus is an extremely unusual condition, usually associated with an inter-atrial defect.[3,4] More unusual is the PLSVC, absence of coronary sinus, and partial atrioventricular septal defect.

Some authors hypothesized that this unusual anatomy is due to the failed process of embryonic venous system lateralization at the level of the left horn and left anterior cardinal vein.[5] The left anterior cardinal vein follows the same development of the right, while the last part of the left horn remains high on the left and behind the left atrium instead of becoming the coronary sinus. Because the left horn does not mitigate, the ostium of coronary sinus will not form.[5]

Furthermore, clinical complications of this anomaly are cyanosis and reduced strain tolerance.[2] Therefore, PLSVC carries more importance in the event of cardiovascular surgery (absolute contraindication of retrograde cardioplegia), central venous, and permanent transvenous pacing lead placement.[6,7] This anomaly may also cause a misdiagnosis such as right-to-left shunting or Eisenmenger's syndrome.

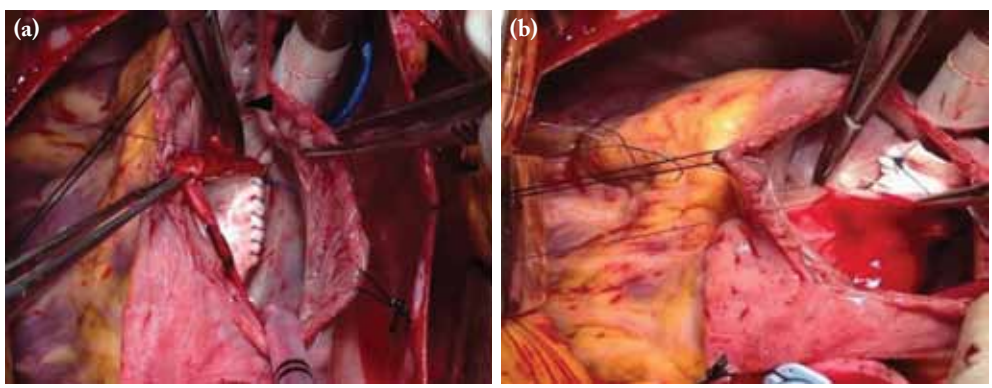


**Figure 2.** **(a)** The orifice of polytetrafluoroethylene baffle, which was used to divert the blood from the orifices of the persistent left superior vena cava into the right atrium seen under the autologous pericardial patch. ▶ The clamp was inside the venous baffle. **(b)** The primum atrial septal defect was closed with an autologous pericardial patch over the polytetrafluoroethylene baffle and the completed venous baffle directed persistent left superior vena cava flow to the right atrium.

Several surgical procedures in the correction surgery have been reported including ligation of the left SVC, intra-atrial redirection of flow from the left SVC to the right atrium, and re-implantation of the left SVC into the right atrium, pulmonary artery or SVC.[8] Ligation of the vein obliterates the intracardiac shunt, however, this procedure is risky, unless there are large collateral links in the head which allow non-obstructed head and neck venous return into the heart. Re-implantation of the persistent left SVC is preferable, when there is a possibility that an intra-atrial baffle may obstruct systemic or pulmonary venous return due to the location of the veins orifices, in particular.[8-10]

In conclusion, our case had partial endocardial cushion defect with an absent coronary sinus and PLSVC without any communication between SVCs. In our case, thebesian veins were opening on way of the baffle and there was the absence of intercaval communication and adequate left atrium volume. Therefore, we used a tube graft PTFE material, as we thought that the selected material could be more resistant to compression from the left atrium pressure.

### Declaration of conflicting interests

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Cardiovascular Surgery and Interventions

Case Report Open Access

# Aortic coarctation and descending aortic aneurysm involving the subclavian artery: Hybrid approach to treatment

İbrahim Özsöyler,[1] Hasan Uncu,[1] Hacı Ali Uçak,[1] Mehmet Acıpayam,[1] Dilek Uçak[2]

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Aortic aneurysms are complex and difficult to treat conditions and are associated with high mortality rates. Alternative methods of treatment continues to show development day by day. One of them is the main subject of this article: the hybrid operation. Herein, we present the case of hybrid repair in a 37-year-old female with 56 mm descending aortic aneurysm and aortic coarctation Aortic debranching, carotid-to-subclavian bypass, and thoracic endovascular aortic repair (TEVAR) operations were performed sequentially in this case. The follow-up computed tomography angiography demonstrated thrombus formation in the aneurysm lumen, no endoleak, and the aortic arch and bypass graft were all patent.

Keywords: Aortic coarctation; descending aorta aneurysm; hybrid procedure; thoracic endovascular aortic repair.

Treatment of aortic aneurysms is performed by less invasive methods with the growing experience and knowledge of hybrid procedures and the developments in recent years.[1-3] Conventional surgical methods still have total circulatory arrest caused by neurological complications and high mortality and morbidity rates are reported.[4,5] Hybrid approach continues to evolve with each passing day. Arch hybrid procedures include three basic approaches: *(i)* debranching of aortic arch and anastomosis; *(ii)* preparation of the proximal and distal landing zones; *(iii)* thoracic endovascular aortic repair (TEVAR). Stenting procedure can also be performed together or sequentially in different sessions. In this case report, we present a hybrid two-stage surgical procedure in a patient with descending aorta aneurysm involving subclavian artery and postductal coarctation of the aorta.

## CASE REPORT

A 37-year-old female patient was admitted with complaints of back pain. On physical examination, her blood pressure (BP) measurements were 123/97 mmHg and 114/92 mmHg at her right and left arm, while 84/67 mmHg and 82/65 mmHg at her right and left ankle. Chest X-ray showed a calcified mass and then transthoracic echocardiography was performed. On echocardiography, aortic coarctation with 38 mmHg peak gradient and aneurysm of the descending aorta was detected. Thoraco-abdominal computed tomography angiography, coronary angiography, and aortography was performed (Figure 3, 4). According to the Ishimaru classification, aortic coarctation and descending aortic aneurysm localized zone 2 and zone 3 (Figure 1) was identified (Figure 2). Bilateral carotid and vertebral artery Doppler ultrasonography showed no pathology.

Surgery was performed under general anesthesia with cerebral oximetry monitoring by near-infrared reflectance spectroscopy (NIRS). Heparin administrated to maintain activated clotting time (ACT) over 300 s. Median sternotomy was performed and innominate vein agenesis was observed. An additional anatomical variations or venous anomaly was not detected. Truncus brachiocephalicus and left common carotid artery were explored. The left subclavian artery was located in the aneurysm site. Aorta-to-left common carotid and aorta-to-truncus brachiocephalicus bypass operations

[1] Department of Cardiovascular Surgery, Adana Numune Training and Research Hospital, Adana, Turkey
[2] Department of Anesthesiology & Critical Care Medicine, Adana Numune Training and Research Hospital, Adana, Turkey

**Corresponding author:** İbrahim Özsöyler, M.D. Adana Numune Eğitim ve Araştırma Hastanesi Kalp ve Damar Cerrahisi Kliniği, 01030 Yüreğir, Adana, Turkey.
Tel: +90 322 - 355 01 01 e-mail: ibrahimozsoyler@yahoo.com

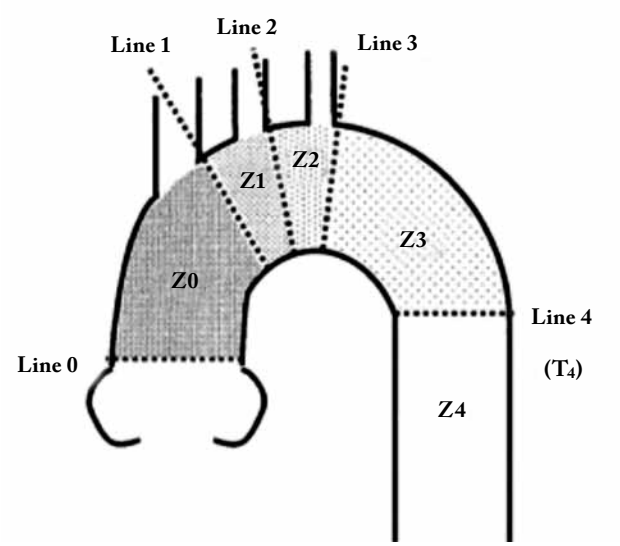


**Figure 1.** Ishimaru classification.

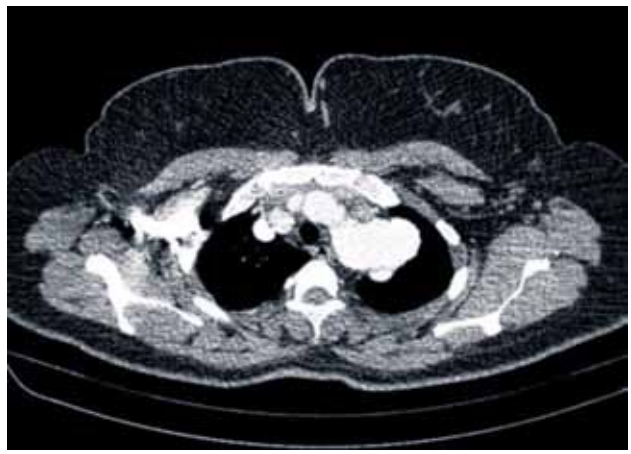

**Figure 2.** Preoperative thoraco-abdominal computed tomography angiography.

were performed with side clamping by using 7/14 mm Y-shaped Dacron graft without cardiopulmonary bypass (Figure 5). Zone of the stent will be placed on the ascending aorta were marked with wire of pacemaker.

At the same session, left common carotid-to-left subclavian artery bypass surgery was also performed. The left femoral artery was explored before the end of the operation for TEVAR. Artery calibration viewed approximately 10 mm. The patient after surgical procedures was taken to angiography unit. A 20 mm self-expandable nitinol stent (E-XL Endoluminal Aortic Stent Prosthesis 20x30 mm, JOTEC GmbH, Hechingen, Germany) was deployed across the coarctation and aneurysm site without previous balloon dilatation procedure. The proximal end of the stent to the ascending aorta and distal end of the stent was placed to distal zone of coarctation site (Figures 6, 7).

**Figure 3.** Preoperative three dimensional computed tomography angiography.

**Figure 4.** Preoperative aortography.

**Figure 5.** Aorta-to-left common carotid and aorta-to-truncus brakiosefalikus bypass.

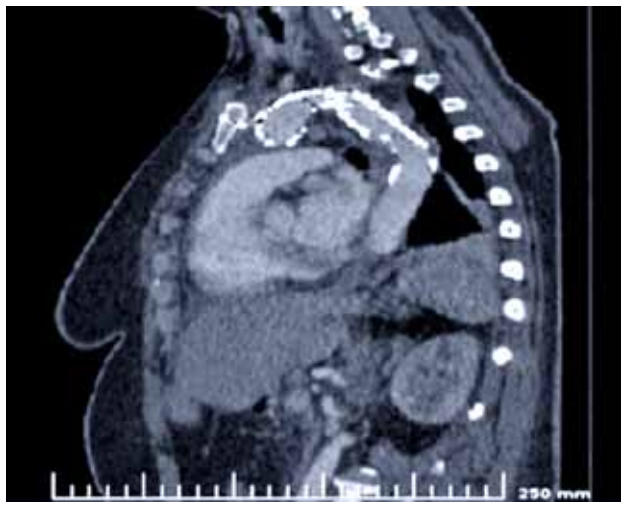


**Figure 7.** Postoperive computed tomography.

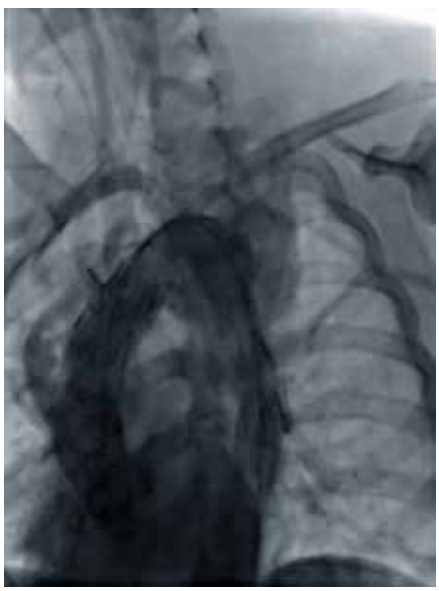

**Figure 6.** Postoperative aortography.

No further balloon dilatation was done immediately after the stent deployment to avoid unnecessary tissue injury. There was no endoleak at the end of the procedure and the patient was taken to the intensive care unit. The patient was discharged at the sixth postoperative day. At two months and every six months, follow-up was scheduled and no endoleak was observed. The BP measurement of four limbs evaluated (Table 1).

## DISCUSSION

With the introduction of developments in the treatment of aortic aneurysms in the last 10 years, less invasive methods can be used. These processes performing without cardiopulmonary bypass and hypothermic cardiac arrest attracts the complication rates, morbidity and mortality rates linked to aneurysm surgery down.[6,7] The hybrid method combining cardiovascular surgery and catheterization procedures has been rapidly spreading all over the world. There are publications showing that hybrid method is effective in cases of dissections, ruptures requiring an emergency intervention, and elective cases.[8] Proximal landing zone is the distal site of left subclavian artery, therefore it is zone 2 (Z 2) or zone 3 (Z 3). It may be possible, if the aneurysm is localized in the distal site of the left subclavian artery or thoracic aortic aneurysm or in some cases of type B dissections. If the pathology located proximally, left common carotid-to-left subclavian artery

**Table 1**
Blood pressures (systolic/mean/diastolic, mmHg) of four limbs

| | Right arm | Left arm | Right ankle | Left ankle |
|---|---|---|---|---|
| Before the stent insertion | 123/106/97 | 114/100/92 | 84/73/67 | 82/71/65 |
| Five days after the stent insertion | 130/113/103 | 128/107/96 | 119/98/88 | 123/104/95 |
| Six months after the stent insertion | 116/97/87 | 119/102/93 | 109/89/79 | 109/90/81 |

bypass surgery should be performed. In the literature, TEVAR procedures to fix the complications developing secondary to the coarctation surgery performed.[9,10] Hybrid treatment of a newly diagnosed coarctation together with an aneurysm of descending aortic is less invasive and safer than convantional surgery.

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Cardiovascular Surgery and Interventions

Case Report Open Access

# Aortic coarctation treated with a self-expandable stent graft

Funda Yıldırım,[1] Adnan Taner Kurdal,[1] Alper Özbakkaloğlu,[1] Tülün Öztürk,[2] İhsan İşkesen[1]

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In this article, we presented a 19-year-old patient with juxtaductal aortic coarctation. The coarcted segment was observed with angiographic study. We planned to implant a self-expandable stent graft. At the angiography laboratory, endovascular aortic stent graft was implanted through the right femoral artery with direct small femoral incision. Then, we dilated the narrowed stent segment by balloon catheter. After the procedure, the patient had a significantly dilated aorta distal to the previously coarcted segment. A stent graft was used to treat coarctation. No access site complications were observed.

Keywords: Neosinus; pericardial patching; right ventricular outflow tract stenosis.

Aortic coarctation describes wide spectrum of diseases with the congenital narrowing of any part of the descending thoracic or abdominal aorta.[1] It typically refers to the narrowing of the proximal thoracic aorta at the level of the ductus or ligamentum arteriosum.[1,2] Surgical treatment has been used for more than 70 years and coarctation is defined as primary (native) phenomenon or secondary to previous corrective surgery.[2] Currently, endovascular techniques have become more widespread for adult patients, in particular, and these techniques are less invasive than conventional surgery. Endovascular techniques are either balloon angioplasty or primary stent implantation. Today, catheter intervention is an established treatment option for adult coarctation with a good success rate and safety profile.[3]

## CASE REPORT

A 19-year-old female patient with juxtaductal aortic coarctation was admitted to our clinic. Physical examination revealed no femoral pulses. The upper extremity blood pressure was measured 160/80 mmHg and 3/6 systolic murmur was heard over precordial and interscapular areas. The laboratory test results were normal. Echocardiography did not reveal any associated abnormality. The coarcted segment was demonstrated with the angiographic study (Figure 1). The sufficient diameter of the coarcted segment and sufficient length of proximal precoarcted segment was shown for direct stent graft implantation.

We planned to implant a self-expandable stent graft (Medtronic Valiant Thoracic Stent Graft with the Captiva Delivery System, USA). We prepared the patient under epidural anesthesia and bilateral femoral exposure. After the femoral exposure, we observed both of the femoral arteries which were 4-5 mm in diameter due to less blood supply of lower half of the body. At the angiography laboratory, endovascular aortic stent graft was implanted through the right common femoral artery with direct small femoral artery incision (Figure 2). Left femoral artery exposure was used for angiographic control views. We implanted the graft with 22 mm in radius and 112 mm in short and 124 mm with anchoring system in length. Then, we dilated narrowed stent segment by balloon catheter application (Medtronic Reliant Stent Graft Balloon catheter; 12 F) (Figure 3).

After the procedure the patient had a significantly dilated aorta in comparison with the previously coarcted segment (Figure 4). One stent graft was enough to dilate the coarcted segment. There were no access site complications. We observed no complications. Blood pressure in the upper extremity was decreased to normal and the patient was discharged without any complication. At six months, a computed tomography angiographic image was obtained. The preoperative and postoperative images were then compared (Figures 5-7). There was no coarcted segment at the

Departments of [1]Cardiovascular Surgery and [2]Anesthesiology and Reanimation, Medical Faculty of Celal Bayar University, Manisa, Turkey

**Corresponding author:** Funda Yıldırım, M.D. Celal Bayar Üniversitesi Tıp Fakültesi Kalp ve Damar Cerrahisi Anabilim Dalı, 45030 Manisa, Turkey.
Tel: +90 236 - 232 31 33 e-mail: fundanizamoglu@yahoo.com

**Figure 1.** Aortic coarctation preoperative angiographic view.

**Figure 2.** Aortic coarctation postoperative angiographic view just after direct stent implantation.

postoperative views. The blood pressure measurements were within normal range.

## DISCUSSION

Patients with primary adult aortic coarctation have a decreased life expectancy, unless treated. Surgical repair was the only effective treatment option available in the past and was demonstrated to improve the postoperative treatment of hypertension.[1] These patients with primary adult aortic coarctation may suffer from upper extremity hypertension, exercise intolerance and shortness of breath. These patients may have many associated cardiovascular comorbidities such as pulmonary hypertension, left ventricular hypertrophy and arrhythmias. After surgery, serious complications requiring revision can be seen. These are paradox hypertension, postcoarctation syndromes

**Figure 3.** Aortic coarctation angiographic view during balloon dilatation.

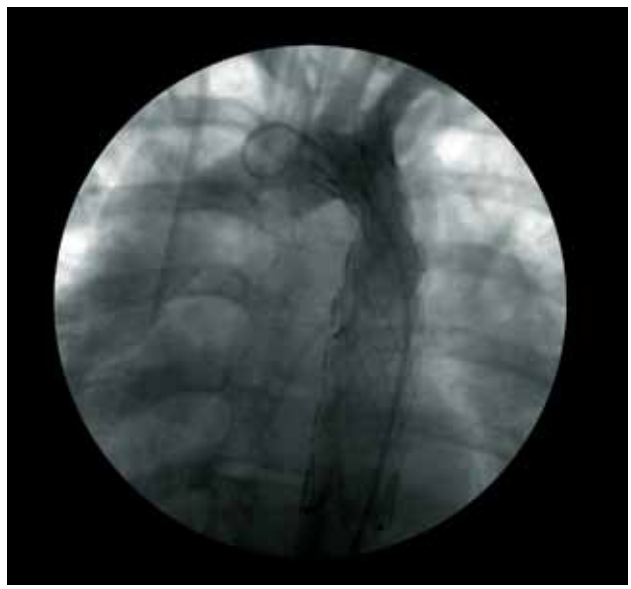

**Figure 4.** Aortic coarctation postoperative angiographic view.

**Figure 5.** Aortic coarctation preoperative computed tomographic image, white arrow shows coarted segment.

**Figure 6.** Aortic coarctation postoperative six months computed tomographic image, white arrow shows corrected segment.

characterized by abdominal complaints, paraplegia, stroke. Hemothorax, chylothorax, left recurrent nerve paralysis, left phrenic nerve paralysis, Horner's syndrome also occur. As late complications restenosis, ischemia of the left arm and aneurysm formation at the sugicaly corrected site could be seen. For this reason, less invasive methods like balloon angioplasty or stent implantation were preferred for primary adult aortic coarctation instead of surgery. Since recurrent coarctation might have seen or complications such as pseudoaneurysm at the anastomosis or aneurysm formation around the aortic tissue after the primary surgical correction, less invasive endovascular interventions have been described with good results despite high operative risk.[5] However, aortic coarctations in older adults have thicker, more resilient and extensive calcifications which increase the theoretical risk of rupture with vigorous balloon dilatation.[4] Furthermore, appropriate patient selection may decrease complication ratio.

Wheatley et al.[1] reported a single center case series with 16 patients in an eight-year period in 2010. The authors observed no perioperative complications and no open surgical conversions, however, further long-term follow-up results were needed. Fink-Josephi et al.[5] reported an endovascular repair of thoracic adult aortic coarctation experiences with 12 patients without complications.

In this case report, we performed direct stent implantation, as there was enough diameter to permit the stent placement into the coarcted segment. Then, we opened the self-expandable stent. We also performed balloon dilatation to the previously coarcted segment to prevent mismatch between the post-stenotic dilated segment and coarcted segment diameter and we observed no complication. Balloon dilatation was a simple procedure following the endovascular stent implantation. The patient had no problem related to the coarctation. Hypertension was not observed during the six-month follow-up period.

In conclusion, as long-term results of stenting have not been fully reported in the literature, this method should be restricted to selected cases. Our primary criterion for primary stent implantation is the enough diameter of the coarcted segment to pass through endovascular stent graft device, being at least 5-10 mm.

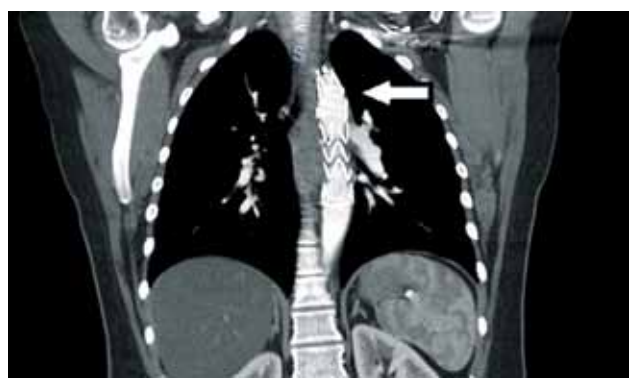

**Figure 7.** Aortic coarctation postoperative six months computed tomographic image, white arrow shows corrected segment.

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Cardiovascular Surgery and Interventions

Case Report Open Access

# Open abdominal surgery for migration of patent ductus arteriosus occluder device

Fatih Gümüş, Mehmet Taşar, Fatih Ada, Zeynep Eyileten, Levent Yazıcıoğlu, Bülent Kaya, Adnan Uysalel

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Although percutaneous interventions have been increasing for closure of patent ductus arteriosus, there may be situations where procedure-related complications are encountered and surgical help is required. In this article, we present a five-year-old girl in whom an Amplatzer duct occluder was first dislodged into the descending aorta and then into abdominal aorta. The device was removed through open abdominal surgery by temporarily occluding the aorta.

Keywords: Abdominal aorta; ductus arteriosus; transcatheter closure.

Although satisfactory results have been obtained in the closure of small ducts (<2 mm using detachable coils), there is a higher incidence of residual shunt, hemolysis and embolization in larger ducts.[1] In 1998, Masura et al.[2] published the first series of cases of percutaneous closure of the arterial duct by using the Amplatzer device, which was specifically designed for medium to large ducts. Complications related to the percutaneous closure of patent ductus arteriosus (PDA) by using an Amplatzer duct occluder (ADO) are rare. However, procedure-related difficulties may lead to severe complications. In this article, we report a case of PDA device embolization into abdominal aorta which was removed successfully through open abdominal incision, a rare surgical approach for this complication.

## CASE REPORT

A-five-year-old girl was referred to the Department of Cardiac Surgery with the diagnosis of a large PDA (3.5-4 mm diameter, 2.2 mm length) and descending aorta embolization as a consequence of implant migration after failed percutaneous closure with the ADO I (8/6 mm). After deployment of the device, the fluoroscopy confirmed that it was dislodged into the juxtaductal descending aorta improperly (Figure 1). Several attempts by catheter retrieval failed. The girl was eligible for a surgical procedure of PDA closure with simultaneous minimally invasive removal of the implant via median sternotomy. Median sternotomy was performed in the operating room and careful dissection was carried out around the aorta, pulmonary artery and PDA. Intraoperative heparin was administered to reduce the risk of thromboembolization. The device was not felt in the aortic edge of PDA. Preoperatively monitorized femoral pulses became feeble. Then, PDA was immediately closed with double ligation. Repeated fluoroscopy showed that the occluder device was located at the bifurcation of abdominal aorta. After an abdominal incision, the device was held with artery forceps and removed from abdominal aorta (Figure 2). Postoperative period was uneventful.

## DISCUSSION

Technological advances in interventional transcatheter closure of PDA provide simple and routine techniques with shorter hospital stay, less mortality and morbidity rates.[3] Moreover, it also reduce surgical risk factors and inevitable operation scar of surgery. Since it was first described by Porstmann et al.[4] in 1967, variable devices have been introduced into the clinical practice. Gianturco and Cook detachable coils have been proven both safe and effective in closure of small to moderate size of PDAs,[5] while ADO device and Nit-occlud® device have been developed to meet relatively favorable outcomes in moderate to large PDAs.[6] However, the procedure is not free of complications, which may include residual shunt with or without hemolysis, protrusion or migration of the device into aorta or pulmonary artery, endocarditis,

Department of Cardiovascular Surgery, Medical Faculty of Ankara University, Ankara, Turkey

**Corresponding author:** Mehmet Taşar, M.D. Ankara Üniversitesi Tıp Fakültesi Kalp ve Damar Cerrahisi Anabilim Dalı, 06100 Sıhhiye, Ankara, Turkey.
Tel: +90 505 - 442 47 80 e-mail: mehmet.tasar@hotmail.com

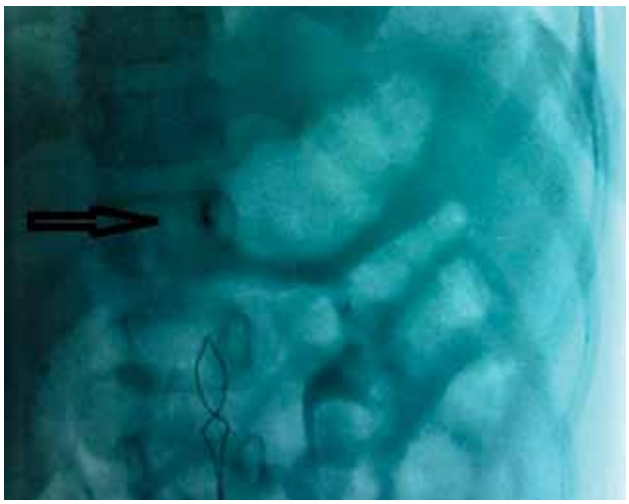

**Figure 1.** Angiography showing migrated device in juxtaductal descending aorta.

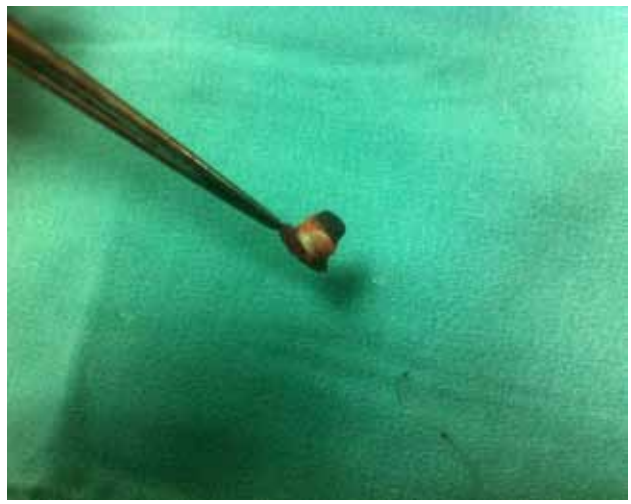

**Figure 2.** Occluder device removed by open abdominal surgery.

thromboembolization and wire fracture or device disruption.[7] Major complication risk is almost 10% in some studies.[8] Embolization of the device has been identified as one of the most significant complications of intervention.[9] It may occur in unexpected sites of circulatory system and cause serious damage. In the present case report, the device was first embolized into the descending aorta and then into the abdominal aorta.

Immediate surgical intervention to remove migrated device is indicated in patients who are hemodynamically unstable. Even in patients who are hemodynamically stable, immediate surgical intervention is preferred, as it facilitates the removal of the device before embolization. Most of the surgical attempts to remove those devices which migrated into descending aorta are done through median sternotomy with or without the aid of cardiopulmonary bypass. In our case, we initially performed median sternotomy to remove the partly dislodged device from juxtaductal descending aorta; however, we were unable to reach, due to the remigration of the occluder into the abdominal aorta. Therefore, we performed an abdominal incision.

In conclusion, percutaneous closure of ductus arteriosus is a safe and effective alternative to surgery, however, complications may be seen in those with unfavorable duct anatomy. Possible mismatch between implanted occluder size and anatomical PDA diameters could be the reason for the phenomenon of PDA underestimation and subsequent complications. Surgical back-up is also important for such interventional procedures. Although rarely seen, re-migration of migrated devices may occur and open abdominal surgery may be required as a life-saving emergency procedure for device retrieval.

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Cardiovascular Surgery and Interventions

Case Report Open Access

# Removal of a missed guide wire in central vein with endovascular intervention: a case report

Eyüpserhat Çalık,[1] Mehmet Tort,[1] Pelin Aydın,[2] Ziya Yıldız,[1] Mehmet Ali Kaygın[1]

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During invasive vascular procedures, missing the guide wire in the vascular lumen is a rare complication. It was identified with direct X-ray images in a 29-year-old male patient with tetraplegia developed after a car accident and followed in the intensive care unit that the guide wire was missed from the right femoral vein after central catheter intervention performed and extended to superior vena cava. The guide wire in the vein was removed with endovascular snare method with the femoral vein attempt. In this article, missing the guide wire into the vascular lumen after invasive vascular interventions as a rare complication was discussed.

Keywords: Catheterization; complication; intravascular foreign body/guide wire; percutaneous intervention.

Invasive vascular interventions (IVI) are performed with increasing frequency in intensive care units (ICU), operating rooms, emergency, and units of cardiologic and radiologic procedures. Critical complications may occur during IVI in the early and late periods. Anatomical structures, individual experiences and skills and the quality of the materials used are crucial factors in the development of complications. Often pneumothorax, hemothorax, vascular injury and bleeding, infection, thromboembolic events, air embolism may be accompanied. The embolization of catheter fragments is a serious complication and the incidence was reported to be 0.1-1%.[1] The missing of a guide wire into the vascular lumen is seen rarely, however, it may cause mortality with a rate of 24-60% leading to complications such as thrombosis, infections, cardiac arrhythmia and perforation.[2-4] In this article, a case of missing guide wire into the vein during central venous catheterization was presented in the light of the literature.

## CASE REPORT

A 29-year-old male patient who was admitted with tetraplegia developed after a car accident was put on a mechanical ventilator in the ICU. The patient who was stabilized was transferred to the re-animation ICU of our hospital after being performed first interventions in the emergency department. He had a dislocation of the C6-7, compression fracture in C7 and cord compression. Necessary correction operations were made by a neurosurgeon. Since he was admitted to the ICU, he had persistent fever, increased white blood cell counts and CRP values. On telecardiography, abdominal and pelvic radiographs, the image of a guide wire extending from the superior vena cava to the right femoral vein was identified (Figures 1-3). It was understood that the guide wire was missed during the placement of central venous catheter into the right femoral vein in the external emergency service. The guide wire was easily removed with snare catheter by the endovascular snare method by performing intervention from the right femoral vein under fluoroscopy (Figure 4). No additional complications were seen before and after the intervention.

## DISCUSSION

Central venous catheterization (CVC) has been increasingly performed with several purposes in the operating rooms, ICUs, emergency units, invasive intervention units, sometimes in services. The intervention is usually done by the Seldinger technique, and jugular, subclavian and femoral veins are mostly used. Some complications such as infection, air or thrombus embolism, arrhythmia, hematoma, pneumothorax, hemothorax, hydrothorax, chylothorax, cardiac perforation, cardiac tamponade, trauma to

Departments of [1]Cardiovascular Surgery, [2]Anesthesiology and Reanimation, Erzurum Regional Training and Research Hospital, Erzurum, Turkey

**Corresponding author:** Eyüpserhat Çalık, M.D. Erzurum Bölge Eğitim ve Araştırma Hastanesi Kalp ve Damar Cerrahisi Kliniği, 25240 Erzurum, Turkey. Tel: +90 505 - 251 54 45 e-mail: eyupserhatcalik@hotmail.com

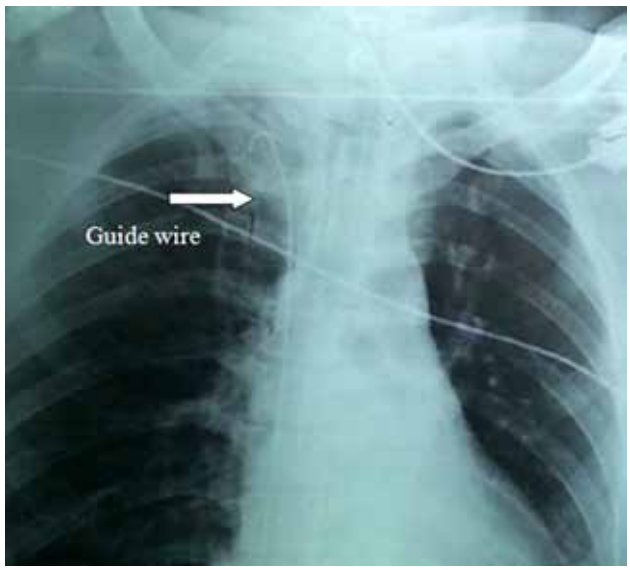


**Figure 1.** Image of guide wire in superior vena cava on telecardiography.

adjacent nerves and blood vessels can be seen during and in the early stages of CVC. The missing of guide wire into the vein; perforation of the superior vena cava, which can be fatal, aortic injury, acute cardiac tamponade and rupture of catheter have been reported as a very rare complication.[3] The complications such as venous thrombosis, superior vena cava syndrome, endocarditis, and sepsis may develop in the late period.[2,3] Doğan et al.[3] reported that a forgotten guide wire in the lumen six months ago caused sepsis in the patient. In our case, although persistent fever and infection were observed due to the presence of intravascular catheter, the culture was negative.


**Figure 2.** Image of guide wire in inferior vena cava on abdominal radiography.


**Figure 3.** Image of central venous catheter in right femoral vein and guide wire on pelvic radiography.

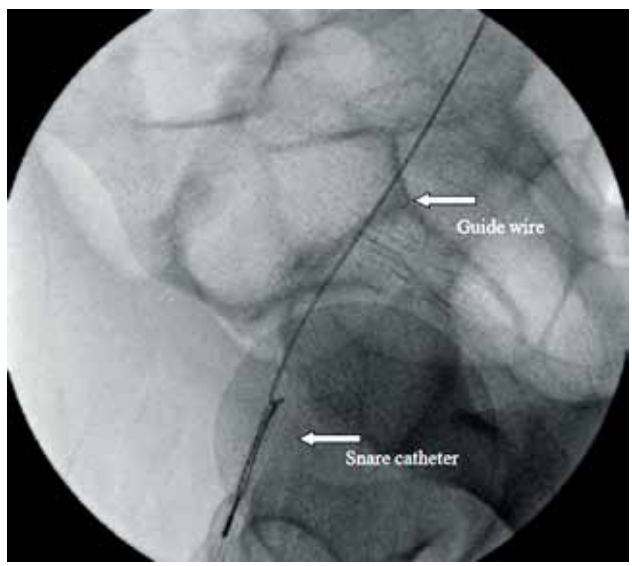


**Figure 4.** Image of fluoroscopy. Removal of the guide wire.

The most important factors which may cause complications in the early period are as follows: poor use of the technique, the clinician's experience while placing the catheter, anatomical region and the quality of the materials used.[3,5] It was considered in our case that the missing of the guide wire into the vein could be due to the catheter insertion performed in emergency conditions and the poor use of the technique.

Complications which may occur after central venous catheterization, can be detected with inexpensive direct radiographies which can be performed easily at an early stage. The direct radiography performed after the procedure also provides information about the placement and function of the catheter. After CVC, plain radiographs will be performed initially and will allow the early diagnosis of complications and early correction before more serious complications occur.[3,5] In our case, the presence of intravascular guide wire was detected by direct radiography at an early stage.

Removal of foreign bodies missed into vascular lumen can be made by endovascular or surgical interventions. As surgical technique, thoracotomy, retroperitoneal approach or vascular exploration can be needed. Schechter and Parisi[6] described the snare technique in 1972 and they had removed the pieces of catheter from two patient's pulmonary arteries. Removal of foreign bodies missed into the vessel has continued to increase over time with the snare technique.[4] Taşoğlu et al.[5] and Özcan et al.[7] removed an intravascular guide wire with endovascular method similar to our case which they were unable to remove it by a simple surgical intervention. We also removed the guide wire easily in our case by endovascular snare technique without performing open surgery because of its additional morbidity potential.

Furthermore, the cases of missing of foreign bodies into the vessels have been increasing with the increased IVI. The removal foreign bodies missed into the vessels with endovascular procedures protects the patient from major surgery and morbidity that can arise in these interventions. Although surgical intervention remains important in patients in whom the removal of foreign bodies with endovascular interventions is not possible or in cases of failure, we conclude that performance of endovascular snare technique primarily is preferred. As a result, removal of intravascular foreign body can be performed rapidly, easily and safely with the endovascular snare technique.

### Declaration of conflicting interests

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Cardiovascular Surgery and Interventions

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# Histopathological findings of an excised varicose vein complicated with chemical phlebitis

Seyhan Yılmaz,[1] Eray Aksoy,[1] Güven Güney[2]

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Sclerotherapy aims to obliterate the varicose veins after injection of the sclerosing substance into vein lumen by causing stenosis, fibrosis and thrombosis through endothelial cell damage, spasm and inflammatory reaction mechanisms. However, there is a limited number of studies investigating the histopathological effects of sclerotherapy to the human veins. In this case report, we evaluated the histopathological results of the in vivo liquid sclerotherapy [Lauromacrogol 400 (40 mg-2% Aethoxysklerol)] on a vein wall specimen surgically excised after development of a painful and non-resorbing thrombus.

Keywords: Local anesthesia; sclerotherapy; varicose vein.

Minimally invasive techniques in varicose vein treatment have gained an increased popularity thanks to their satisfactory outcomes, low complication rates and faster return to normal activities.[1] Physical and chemical methods are available to obliterate the lumen of varicose veins and sclerotherapy is one of the chemical methods being used over the last 60 years.[2] Sclerosing substance may cause stenosis, fibrosis and permanent thrombosis through endothelial cell damage, spasm, and inflammatory reaction mechanisms to obliterate varicose veins. Data regarding *in vivo* histopathological findings after sclerotherapy is also limited.

In this case report, we evaluated the histopathological results of the *in vivo* liquid sclerotherapy [Lauromacrogol 400 (40 mg-2% Aethoxysklerol)] on a vein wall specimen surgically excised after development of a painful and non-resorbing thrombus.

## CASE REPORT

A 32-year-old woman who received liquid sclerotherapy for calf varicose veins three months ago presented with a painful phlebitis at the area of injection. Her complaints began about two months after receiving sclerotherapy. She had no previous history of chronic comorbidities, steroid or oral contraceptive use, having hypercoagulation disease or allergies. She had a history of 10 pack-year smoking. The patient's CEAP classification (the Clinical-Etiology-Anatomy-Pathophysiology) was varicose veins ($C_2$), primary (Ep), superficial (As), reflux (Pr). According to the information from hospital registry and patient's statement, she received our standard protocol of practice: 2 ml of lauromacrogol 2% was injected into the vessel without diluting, a 32 G needle was used, the area of injection was compressed for several minutes, the leg was bandaged, and the patient was discharged with resting instructions. Although she used topical anti-inflammatory agents for three months, the pain and stiffness over the phlebitis varicosity were not relieved. The patient requested removal of the diseased varicose vein. Physical examination revealed 4 cm in length and 6 mm in width hyperpigmented lesion on the diseased vessel segmented, which was firm and tender with palpation. Duplex ultrasound scanning showed thrombus formation in formerly treated varicose veins. Under local anesthesia, a 3 cm long diseased segment of the vein was ligated at both ends and excised between the tied ends. The specimen was fixed in normal saline solution and stained with hematoxylin and eosin for histopathological examination. Histopathological findings were as follows: vascular lumen was almost totally obliterated

Departments of [1]Cardiovascular Surgery, [2]Pathology, Hitit University Çorum Training and Research Hospital, Çorum, Turkey

**Corresponding author:** Seyhan Yılmaz, M.D. Hitit Üniversitesi Çorum Eğitim ve Araştırma Hastanesi Kalp ve Damar Cerrahisi Kliniği, 19200 Yeniyol, Çorum, Turkey.
Tel: +90 364 - 223 03 23 e-mail: drlabarna@gmail.com

by a well-organized and partly recanalized thrombus; tunica media had decreased in thickness, and the endothelium was eroded in focal sectional areas; and tunica intima was lacking in sites where thrombus was adherent. There were also extensive involvement with neutrophil leukocytes, lymphocytes and degenerated smooth muscle cells (Figure 1). The patient was discharged and postoperative course was uneventful (The patient presented herein did not allow us to use a photograph of the lesion before the operation, however, she gave consent to have her information used for scientific publishing).

## DISCUSSION

Sclerosing agents not only results embolization of the target vein, but also disrupt its endothelium limiting proliferation and collateralization.[3] Although earlier studies suggested that hypertonic solutions might cause fewer side effects (i.e. hyperpigmentation) than chemical agents,[4] recent data suggests that sclerosing agents are superior to placebo in terms of efficacy and patient satisfaction.[5] A recent prospective randomized study showed no superiority of hypertonic saline solution over polidocanol and also a higher rate of pain during its injection.[6] Currently, sclerotherapy with sclerosing agents is regarded as the choice of therapy in treating spider and reticular veins.[7] According to a report from England, the number of patients requesting for sclerotherapy increased by 300% over seven years, although number of patients presenting with varicose veins declined by 34%.[1]

Complications related to sclerotherapy are extremely rare[2,8] and may not only be dependent on chemical irritation or iatrogenic factors; however, they also be related with patients related factors such as hypercoagulable state.[9] There have been a number of case reports in the past about development of sclerotherapy-related severe life threatening complications including acute ischemic stroke in a 73-year-old woman,[10] myocardial infarction in a 61-year-old patient with foramen ovale,[11] arterial occlusion due to intra-arterial injection in a 59-year-old woman,[12] middle cerebral arterial embolism,[13] and visual loss in a 66-year-old woman.[14] It is likely that foam sclerotherapy in elderly may rarely cause serious complications, while related complications are usually mild, transient and underreported.

Our histopathological assessment supports the concept that sclerosing agent not only resulted in thrombus formation within the vein lumen, but also induced a substantial structural damage to the vessel wall. We observed that vein endothelium was irreversibly eroded and tunica media was almost completely disappeared in focal sectional areas. Such changes should be regarded as the natural consequence of the treatment. Presence of both inflammatory and necrotic cells in histopathological specimen is likely to explain why the patient has been suffering from pain and sensitivity despite using topical medications. Our patient had no further complications such as ascending thrombophlebitis which occurred previously in a 48-year-old patient undergoing foam sclerotherapy. The authors reported that the patient presented with some symptoms similar to ours after receiving foam sclerotherapy for varicose veins at lower extremity. That patient was eventually diagnosed with breast cancer.[15]

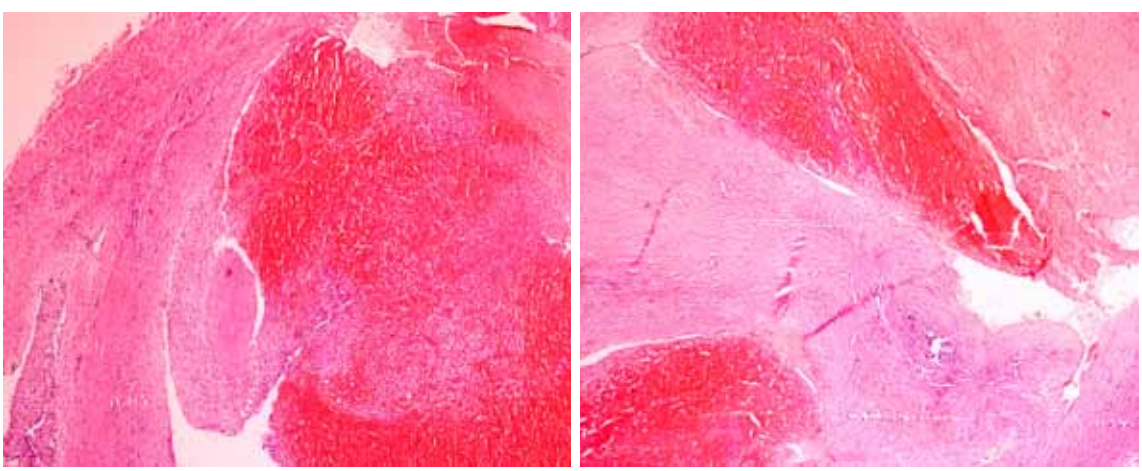

**Figure 1.** Photomicrograph of the sectional areas from excised vein sample material (H-E x 40).

### Declaration of conflicting interests

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Cardiovascular Surgery and Interventions

Case Report Open Access

# Iatrogenic arteriovenous fistula of tibialis posterior artery following surgery of inserting plates and screws on a broken ankle: a case report

Bahar Temur, Mustafa Aldağ, Tolga Can, Adlan Olsun, Mehmet Kaplan

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Iatrogenic arteriovenous fistulas (AVFs) are seen after vascular trauma or injury. Peripheral arteriovenous fistulas may cause many problems like limb pain, edema, aneurysmal degeneration of the artery, ischemic symptoms, ulceration related to arterial insufficiency, and congestive cardiac failure. We report a 50-year-old man admitted to our clinic with a lower extremity bleeding ulcer and a history of orthopedic surgery a year ago. Following Doppler ultrasound and computed tomography findings, the fistula was treated surgically. Although endovascular stenting is the procedure of choice for such fistulas, surgical treatment is still an option.

Keywords: Iatrogenic arteriovenous fistula; surgery; traumatic arteriovenous fistula.

Iatrogenic arteriovenous fistulas (AVFs) are harmful complications caused by different surgical operations and vascular injuries.[1] Most peripheral iatrogenic AVFs need treatment. The shunt increases venous volume and pressure and decreases peripheral vascular resistance. Reduced blood flow to the lower extremity can lead to the onset or worsening of lower extremity ischemic symptoms. The increase in stroke volume and heart rate may lead to a dramatic rise in cardiac output. This case report discusses a unique case of an AVF between the right posterior tibial artery and vein in a patient presenting with leg pain, edema and bruising (Figure 1). The examination and treatment strategies are also discussed.

## CASE REPORT

A 50-year-old man was admitted to our emergency service with pain and bleeding from a varicose vein on his right leg. He had complaints of limb pain and edema. The previous history of surgery of inserting plates and screws on his broken ankle suggested an AVF, and the patient was hospitalized.

During examination, there were many varicose veins in the great saphenous vein (GSV) distribution in the distal leg. Peripheral pulses were palpable. There was a continuous bruit on the left side, if the extremity with a palpable thrill.

A Doppler ultrasound was made to the lower extremity and it only showed venous insufficiency in the GSV. Contrast enhanced computed tomography of lower extremity was performed which revealed a connection between the right posterior tibial artery to its adjacent vein. In the arterial phase, the varicose veins were filled with blood due to the fistula.

Laboratory analysis revealed no abnormality. Echocardiography demonstrated normal left ventricular systolic and diastolic functions. All valves were reported to be functioning normally without stenosis or insufficiency.

The patient was operated under general anesthesia. A longitudinal incision was made 1 cm below the knee. Trifurcation of the popliteal artery was exposed. Large varicose veins derived from the GSV were ligated. Posterior tibial artery was exposed till the fistulized part. Systemic heparinization was instituted and vascular clamps were applied to the vessels.

The narrow-based AVF between the posterior tibial artery and vein was ligated and divided at arterial and venous ends. The arterial side was primarily repaired. After this procedure, the GSV was ligated distally and proximally and excised.

Surgical repair was successfully performed and clinical signs and symptoms relieved dramatically

Department of Cardiovascular Surgery, Dr. Siyami Ersek Thoracic and Cardiovascular Surgery Training and Research Hospital, İstanbul, Turkey

**Corresponding author:** Bahar Temur, M.D. Dr. Siyami Ersek Göğüs, Kalp ve Damar Cerrahisi Eğitim ve Araştırma Hastanesi, Kalp ve Damar Cerrahisi Kliniği, 34668 Haydarpaşa, İstanbul, Turkey.
Tel: +90 216 - 542 44 44 e-mail: bahartemur@hotmail.com

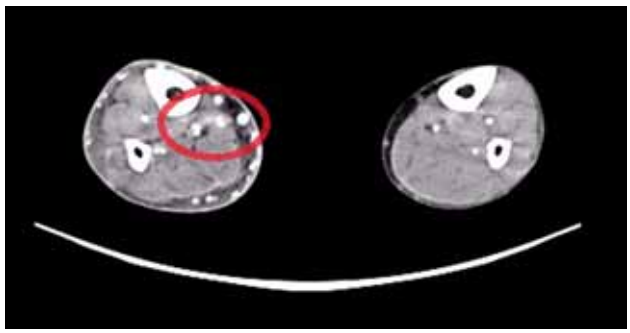

**Figure 1.** Computed tomography-scan showing the fistula between posterior tibial artery and its adjacent vein.

subsequently (Figure 2). The patient was discharged from the hospital at the fifth postoperative day.

Repeated Doppler ultrasound was done and there was no sign of the fistula. At the third postoperative week, the leg was decent with no edema and the venous ulcerations were nearly all treated.

## DISCUSSION

Iatrogenic arteriovenous fistulas usually occur following percutaneous interventions, surgical operations, and trauma. They may remain asymptomatic for many years. Swelling, thrill, and pulse deficit in the limb are the clinical features of the fistula.[2] Peripheral AVFs may also cause intermittent claudication or venous hypertension. Progressive symptoms of heart failure may be also seen eventually.

The diagnosis of AVFs can be made by history and physical examination.[3] Duplex ultrasonography, computed tomography, magnetic resonance imagining, and conventional angiography may be used to find the localization of the fistula.[4] Color-coded Doppler ultrasound is also a non-invasive and simple method, but in our case this method failed to show the AVF.

One-third of all AVFs close spontaneously in one year and for asymptomatic AVFs, watch and wait policy may be used. The type of treatment depends on the cause, acute or chronic nature, size and location of the AVF. Pressure with an ultrasound probe can be applied or bandaging may be done for spontaneous closing of the fistula.[2]

Endovascular techniques and surgical repair are two different options in AV fistula treatment. Although endovascular techniques reduce patient morbidity and

**Figure 2.** Edema, hyperemia and pain on the right leg, decreased significantly in the postoperative period.

hospital stay, surgery is still the gold standard.[2] Surgical repair offers a 96% chance of closure of the fistula.[5] Endovascular modalities of treatment include covered stent implantation, coil embolization and utilization of glue. Using covered stents is technically easy, and said to have a high success rate and a low complication rate.[6,7]

Endoluminal coils can be easily traced through the high flow fistula and enter the venous circulation. Therefore, it must be avoided for the treatment of the AVF.[8] The use of covered stents and stent grafts in small arteries has a higher risk of thrombosis and vascular stenosis.[9]

In conclusion venous hypertension and ischemic complications can be seen in peripheral arteriovenous fistulas. To avoid vascular stenosis and thrombosis, distal AVFs must be treated surgically in trauma patients.

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# Giant external iliac artery pseudoaneurysm following percutaneous coronary intervention: a rare case

Süleyman Sürer, Yüksel Beşir, Orhan Rodoplu, Ömer Tetik

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Invasive cardiac interventions have an important role in development of lower extremity arterial pseudoaneurysms. A 27-year-old male patient presented with the complaint of difficulty in walking following the discharge from the hospital after a successful emergent percutaneous transluminal coronary angioplasty and stent implantation for acute myocardial infarction in a cardiology clinic. Arterial Doppler ultrasonography showed a 90 mm of pseudoaneurysm in the right external iliac artery. Closure of aneurysm by compression and thrombin injection under ultrasonographic guidance was planned, but failed. Aorto-femoral bypass was performed eventually.

Keywords: Iliac artery; percutaneous intervention; pseudoaneurysm.

Invasive cardiac interventions play an important role in development of lower extremity arterial pseudoaneurysms. In this paper, we report a patient operated for a giant external iliac artery pseudoaneurysm developed after coronary artery intervention.

## CASE REPORT

A 27-year-old male presented with the complaint of difficulty in walking, after he was discharged from a cardiology clinic following a successful emergent percutaneous transluminal coronary angioplasty and stent implantation for acute myocardial infarction. He was on dual antiplatelet therapy after the stent implantation.

At admission, he was complaining of spasm in the right foot, difficulty in walking erect, and distension in the right lower abdominal quadrant. Physical examination revealed a pulsatile mass in the right lower quadrant. An arterial Doppler ultrasonography showed a 53x28 mm pseudoaneurysm in right external iliac artery (Figure 1). Closure of aneurysm by compression and thrombin injection under ultrasonographic guidance failed. Thus, a surgical intervention was scheduled. Meanwhile, the aneurysm continued to grow in size. The patient was taken into operation under general anesthesia in appropriate position. A right flank and femoral incisions were made and joined. Femoral artery was exposed first, followed by aneurysm exploration via retroperitoneal approach (Figure 2). Near-full thickness injuries and dissections were detected in many parts of right external iliac artery up to the level of internal iliac artery. Following control of the iliac arteries with a vascular tape, aneurysmectomy was performed after 5000 IU intravenous heparin injection. A 90x50 mm pseudoaneurysm sac was removed (Figure 3). As severely dissected, the external iliac artery was excised. Since the common iliac artery was extremely fragile, we planned to perform an aortofemoral bypass by means of the abdominal incision. Abdominal aorta was controlled by abdominal incision. Aorto-femoral bypass was performed via retroperitoneal approach using a 6 mm polytetrafluoroethylene graft. Surgical layers were closed anatomically. Postoperative course was uneventful.

## DISCUSSION

Cases of pseudoaneurysm in the artery of vascular access have been increasingly reported with the growing number of invasive cardiac (coronary angiography, angioplasty, and stenting) and vascular interventions each year.[1] Furthermore, rates of pseudoaneurysm are increased with antithrombotic therapy (heparin, warfarin, antiplatelet agents)

Department of Cardiovascular Surgery, Bursa Yüksek İhtisas Training and Research Hospital, Bursa, Turkey

**Corresponding author:** Süleyman Sürer, M.D. Bursa Yüksek İhtisas Eğitim ve Araştırma Hastanesi Kalp ve Damar Cerrahisi Kliniği, 16330 Yıldırım, Bursa, Turkey.
Tel: +90 505 - 494 08 28 e-mail: drsuleyman1@hotmail.com

**Figure 1.** Doppler ultrasonographic view of pseudoaneurysm.

given as an adjunct to interventional therapies due to inhibition of coagulation cascade.[2] Norwood et al.[3] reported in a retrospective study that pseudoaneurysm was located in femoral artery in 79 (83%) of 95 patients, whereas iliac arteries (common, external) were affected in only three (3.1%) cases. In our patient, the pseudoaneurysm was situated in the external iliac artery. The aforementioned study also found that the risk of pseudoaneurysm significantly increased in patients receiving Glycoprotein IIb/IIIa inhibitors.[3] Coronary angiography (with or without angioplasty/stenting) is the leading cause of pseudoaneurysms involving femoral artery (40.5% of cases), followed by pseudoaneurysms due to vascular graft anastomoses (29.15%), and peripheral vascular angiography (10.1%).[3] Currently, ultrasound-guided thrombin injection (UGTI) remains the mainstay of radiological pseudoaneurysm treatment. Studies have shown that UGTI is more effective than ultrasound-guided compression used in the past.[3-6] Surgery remains the treatment of choice in 41.8% of patients, followed by graft revision in 27.8%, UGTI in 24.1%, UG compression in 2.5% and stenting in 1.3%.[3] Management approaches to pseudoaneurysms have been changed, since 1995.[3] Today, surgical approach has been increasingly less common, while UGTI has become the treatment of choice.[3,7] We first employed UGTI method and performed surgical intervention, when the former failed.

Furthermore, endovascular therapy has some challenges related to iliofemoral access, despite the ever improving logistics provided by advancing stent graft technology and delivery platforms. Murray et al.[8] in a systematic review of studies on endovascular access techniques performed between 1994 and 2005, reported that majority of access problems are brought about by overly tortuous iliac arteries, circumferential vessel wall calcification, severe vessel obstruction, and small vessel caliber. It is usually not possible to use covered endovascular stent grafts, since there is a significant size mismatch between the common and external iliac arteries.[9]

The success and complication rates of non-surgical treatments of this pathology are heavily dependent on the width and length of pseudoaneurysm neck.

**Figure 2.** Intraoperative view of pseudoaneurysm.

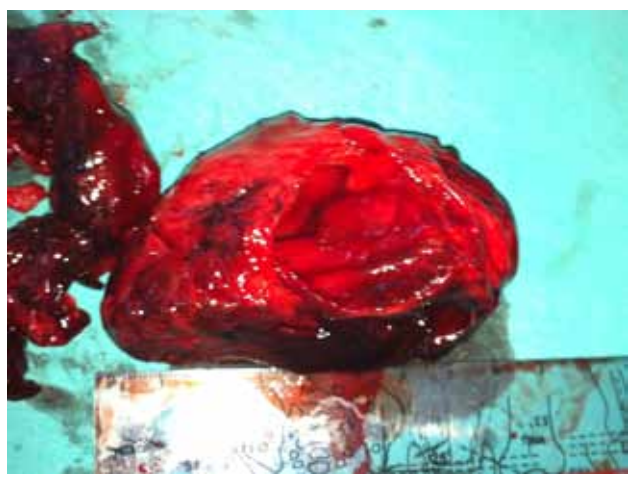

**Figure 3.** Sac of pseudoaneurysm (90x50 mm).

In conclusion, conventional surgical approach remains the gold standard for the management of pseudoaneurysms, although endovascular techniques and ultrasonographically-guided vessel compression and thrombin injection are increasingly used.[10,11] The primary surgical approach should be aneurysmectomy and arterial reconstruction. End-to-end anastomosis or primary repair with an appropriate graft should be performed, where possible.

### Declaration of conflicting interests

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Case Report Open Access

# Surgical treatment of an isolated left jugular vein aneurysm: a case report

İhsan Alur, Serkan Girgin, Bilgin Emrecan, Ali Vefa Özcan

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Jugular venous aneurysms are rare causes of neck masses which occur during straining or Valsalva maneuver and most frequently seen in children. Symptoms include painless and enlarging mass with conditions which increase intrathoracic pressure such as coughing, straining or Valsalva maneuver. Although the exact etiology has not been fully elucidated, congenital defect in the muscular layer, compression of apex of right lung or clavicle to the jugular vein, mechanical obstruction of the lower part of the neck or upper mediastinum, insufficient compliance of vein, thoracic outlet syndrome or compression of anterior scalene muscle may play a role. Jugular vein aneurysms can be treated surgically; however, timing of the surgery is controversial. Linear plication and encapsulation are the most commonly performed surgeries. Herein, we present a seven-year-old boy with left internal jugular vein aneurysm who underwent a successful surgical repair.

Keywords: Aneurysm; child; jugular vein; surgery.

Jugular vein aneurysm (JVA) is described as a fusiform or saccular dilatation of the jugular vein without torsion in the jugular vein wall.[1] The etiology of JVA is unclear, however some predisposing factors have been suggested such as congenital agenesis of the muscular layer of the vein wall, external compression of the jugular vein by the cupola of the right lung and head of the clavicle, mechanical obstruction of the inferior neck and upper mediastinum, the inflexibility of the vein wall, thoracic outlet syndrome and anterior scalen compression.[1] On histologic examination, there can be loss of elastic fiber and intimal hyalinization or the wall of the vein can be totally normal.[2] It is generally seen in children with the right jugular vein being affected most and is two times more common in males.[3] In addition, JVA can be considered as an asymptomatic and benign state, however, in some cases, there can be complaints such as shortness of breath, feeling of suffocation or weight on the chest, tongue pain, discomfort during physical exercise, and may also cause cosmetic-psychological complaints. In the differential diagnosis, laryngocele growing (swelling) with Valsalva on the neck, cystic hygroma, external laryngeal diverticula, upper mediastina cyst or tumor and other vascular pathologies should be also considered.[4] The diagnosis is based on the evaluation through Doppler ultrasound (USG) or contrast computed tomography (CT). Doppler USG codes the turbulent flow created during Valsalva, while CT demonstrates the deep anatomical structures adjacent to the jugular vein.[5]

## CASE REPORT

A seven-year-old boy applied with the complaint of a swelling on the left side of his neck which grew larger with crying or blowing into balloons for the past one year. On physical examination, a non-pulsating compressible mass of soft character was palpated on the left side of the neck (Figure 1b). The hyperelastic skin and hyperextensible joints were suggestive of Ehlers-Danlos syndrome; however, medical genetic consultation findings were normal. There was no history of trauma or infection of the neck and upper thorax. On ultrasonography, a mass on the jugular vein measuring 15x12 mm before Valsalva and 38x24 mm after Valsalva was noted. Turbulent flow through the vein lumen with a thin movable valve was prominent. This appearance was primarily interpreted as an enlargement of the jugular vein with Valsalva secondary to valve damage. There was no significant cystic-solid sonopathology. On CT angiogram, it was seen that the left jugular vein was approximately 27 mm in diameter at a level of 1 cm above where it drained into the subclavian vein (Figure 1a). An incision appropriate to the line of the left carotid artery was performed under general anesthesia. The internal jugular vein aneurysm (IJVA) was exposed (Figure 2a).

Department of Cardiovascular Surgery, Medical Faculty of Pamukkale University, Denizli, Turkey

**Corresponding author:** İhsan Alur, M.D. Pamukkale Üniversitesi Tıp Fakültesi Kalp ve Damar Cerrahisi Anabilim Dalı, 20070 Kınıklı, Denizli, Turkey.
Tel: +90 532 - 392 60 78 e-mail: alur_i@hotmail.com

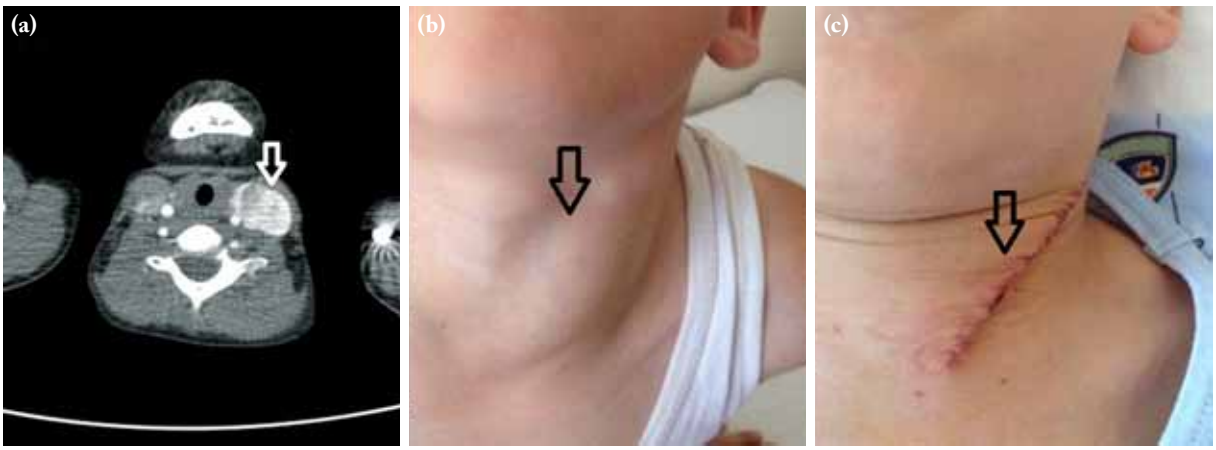


**Figure 1.** **(a)** Contrast-enhanced computed tomography image preoperatively, **(b)** preoperative view of aneurysm, **(c)** postoperative view of incision.

A segmental dilatation with a diameter of 3 cm was observed on the IJVA. The external jugular vein was normal. The aneurysmal segment was clamped using side clamps. Linear plication was performed with 6/0 prolene suture (Figure 2b). The aneurismal sac was opened longitudinally and sutured like a flap to the anterior wall of the IJV to strengthen the anterior wall (Figure 2c). The patient was discharged without any problems at the fifth postoperative day (Figure 1c).

## DISCUSSION

The surgical treatment indications for JVA are still controversial. Some authors recommend a conservative approach, while others perform surgery after diagnosis. This benign congenital venous abnormality may cause some complications.[6,7] The vein wall becomes thinner and weaker secondary to aneurysmal expansion, thereby leading to rupture and bleeding. Since the ability and awareness of self-protection against external traumas are weaker in children than in adults, there is the risk of rupture in these cases. An untreated JVA may cause bleeding during surgical procedures such as tonsillectomy.[8] Turbulent flow which develops due to the hemodynamic changes occurring in dilated veins may also cause thrombophlebitis, intramural thrombus, pulmonary embolism, and congestive heart failure.[9] A long-term follow-up has shown that dilatations of the jugular vein continue to grow.[1,4] We also need to remember the cosmetic-psychological component. This kind of abnormality is likely to affect the development of personality and inner peace psychosocially.

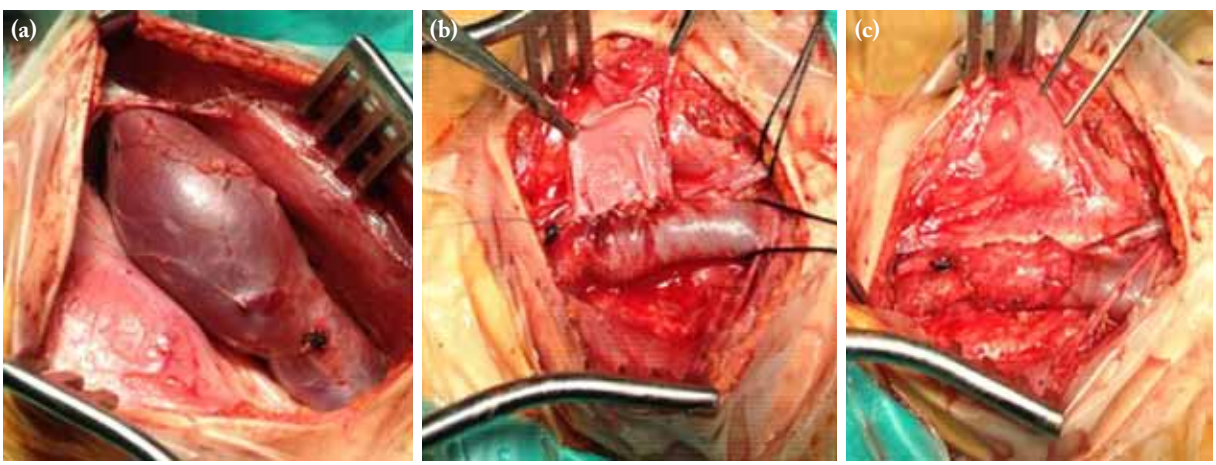


**Figure 2.** **(a)** Operative view of jugular venous aneurysm, **(b)** linear plication view, **(c)** the end view of jugular vein.

Treatment of JVA is surgical. Ligation or resection is performed, if the dilated segment is located in the external jugular vein. However, care is required with unilateral or bilateral dilatation of the internal jugular vein (IJV), because the IJV provides about 70% of the venous drainage for the brain.[6] On the other hand, ligation or resection can cause fatal complications. Jianhong et al.[6] reported vomiting due to an increased intracranial pressure, headache, unilateral edema of the neck and craniofacial region after IJV ligation and right pontin lacunar infarction on MR imaging in three of the 51 patients operated. They recommended a vein repair + encapsulation by longitudinal suturing technique instead of ligation and resection for unilateral or bilateral IJVA cases and reported that it was safer.

We applied vein repair + encapsulation by longitudinal suture technique (linear plication) in our case after placing a side clamp on the dilated segment of the IJV. After placing the side clamp, we performed a longitudinal incision along the dilated segment of the jugular vein and wrapped the encapsulation circumferentially to the jugular vein using linear suturing. We believe that this technique strengthens the vascular wall, preventing recurrences. With side clamp repair, we also protected the brain hemodynamics and did not increase the intracranial pressure. Strengthening the vascular wall with the encapsulation technique can be performed using Dacron or PTFE patches.[6] We think that our method (plication + encapsulation) is superior to using synthetic grafts in growing children.

Furthermore, an endoscopic repair case as an alternative to open surgery has been reported in the literature.[10] Although the duration of the procedure is longer and postoperative pain is more, the pathology can be improved without neck scars with the endoscopic method.

In conclusion, JVA is a rare benign venous pathology in children. It can cause severe complications, if left untreated. Also, it can adversely affect the psychosocial personality development of the child. We successfully performed the plication and encapsulation in surgical treatment of JVA. We think that our method is more advantageous than using synthetic grafts in growing children.

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