

## Aortopulmonary fistula occurring 19 years after repair of aortic coarctation with Dacron patch aortoplasty

Dacron yama aortoplasti tekniğiyle aort koarktasyonu onarımından 19 yıl sonra gelişen aortopulmoner fistül

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A 39-year-old male patient presented with complaints of cough and hemoptysis. He had a history of aortic coarctation repair with the use of Dacron patch aortoplasty 19 years before. Thoracic aortography showed significant narrowing of the aortic lumen and some extravasation of contrast material in the patch area. Computed tomography angiography of the chest revealed obvious narrowing at the repair site and a double lumen appearance. There was no evidence for a true aneurysm or pseudoaneurysm. Following a left thoracotomy and on partial cardiopulmonary bypass, the Dacron patch and coarctation segment were completely removed. A new Dacron graft of 20 mm was used to restore aortic continuity. A pleural flap was placed on the new Dacron graft to separate it from the lung tissue. The postoperative course was uneventful and the patient was discharged on the fifth postoperative day. Hemoptysis disappeared following the procedure. Magnetic resonance angiography performed three months after the repair showed a patent graft and no coarctation.

**Key words:** Aortic coarctation/surgery; fistula/surgery; hemoptysis/etiology; postoperative complications.

Aorto-airway fistula is a rare but potentially life threatening complication of surgery of the descending thoracic aorta. Most cases occur following surgical repair of aortic coarctation or after repair of descending thoracic aneurysms.<sup>[1]</sup>

We hereby present a patient who developed an aorto-pulmonary fistula following repair of aortic coarctation with Dacron patch aortoplasty.

### CASE REPORT

A 39-year-old male patient presented with cough and hemoptysis of three-week history. The amount

Otuz dokuz yaşında erkek hasta öksürük ve hemoptizi yakınmalarıyla başvurdu. Hastaya 19 yıl önce Dacron yama aortoplasti tekniğiyle aort koarktasyonu onarımı yapılmıştı. Torasik aortografide aort lümeninde belirgin daralma ve yama bölgesinde bir miktar kontrast madde kaçağı görüldü. Bilgisayarlı tomografi anjiyografide eski tamir bölgesinde belirgin daralma ve çift lümen görünümü izlendi. Anevrizma veya psödoanevrizma varlığına ilişkin bir bulgu yoktu. Sol torakotomiden sonra ve kısmi baypas altında, önceki Dacron yama ve koarktasyon segmenti tümüyle eksize edildi. Aort devamlılığını sağlamak için 20 mm'lik yeni bir Dacron yama yerleştirildi ve yamayı akciğer dokusundan ayırmak için üzerine pleural flep kondu. Ameliyat sonrası dönemi sorunsuz geçiren hasta ameliyatın beşinci gününde taburcu edildi. Onarımdan sonra hemoptizinin tümüyle kaybolduğu görüldü. Üçüncü ayda yapılan manyetik rezonans anjiyografide greftin açık olduğu izlendi ve koarktasyon görülmedi.

**Anahtar sözcükler:** Aort koarktasyonu/cerrahi; fistül/cerrahi; hemoptizi/cerrahi; ameliyat sonrası komplikasyon.

of hemoptysis had progressively increased over the previous week, with a change in its character from blood-tinged sputum to large quantities of bright red blood. There was also a history of spontaneous efflux of bright red blood given off with Valsalva posturing. The only significant finding in the patient's history was aortic coarctation repair with the use of Dacron patch aortoplasty performed 19 years before. A thoracic aortogram was obtained which showed significant narrowing of the aortic lumen in the same location as well as a pressure gradient of 39 mmHg across the repair (Fig. 1). A small

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amount of contrast material extravasation was also noted at the site of Dacron patch plasty. A diagnosis of recoarctation was made with a possible aortopulmonary fistula. Computed tomography (CT) angiography of the chest revealed obvious narrowing at the repair site as well as a double lumen appearance. However, a direct communication could not be demonstrated between the aorta and the pulmonary parenchyma. There was no evidence for a true aneurysm or pseudoaneurysm. Considering the presence of a significant pressure gradient and persistent hemoptysis with a possible etiology of aortopulmonary fistula, a decision of a redo coarctation repair was made.

Under general anesthesia and using a double-lumen endotracheal tube, a spinal drainage catheter was placed at the L<sub>4-5</sub> interspace. Cerebrospinal fluid drainage was performed throughout the procedure and during the first 24 hours after the procedure to keep the intraspinal pressure between 10 to 12 mmHg. Flexible fiberoptic bronchoscopy was performed after the patient was completely prepared and draped. Blood seemed to emanate from the bronchial orifice of the left upper lobe. There was no evidence for any other source of bleeding. A redo left thoracotomy was performed. The left upper lobe of the lung was densely adherent to the previous repair site. These adhesions were left untouched. The aortic arch was circumferentially dissected between the left common carotid artery and the left subclavian artery as well as in the mid-descending thoracic aorta approximately 5 cm below the Dacron patch repair. The patient was heparinized and placed on partial cardiopulmonary bypass via a left atrial-left common femoral cannulation. We did not use an oxygenator. The aortic arch was clamped between the left common carotid artery and the left subclavian artery as well as distally in the mid-descending aorta. Lung adhesions were then removed sharply to prevent traumatic lung tears. The lung parenchyma was fragile, hemorrhagic, and partly necrotic. There was a defect-like appearance in the visceral pleura adjacent to the anastomotic line of the previous operation. The Dacron patch and coarctation segment were completely removed. An interposition Dacron graft of 20 mm was used to restore aortic continuity. The necrotic area in the left lung was removed using a GIA stapler. A pleural flap was placed on the new Dacron graft to separate it from the lung tissue. Aortic cross-clamp time was 34 minutes. The postoperative course was uneventful and the patient was discharged on the fifth postoperative

day. Hemoptysis disappeared following the procedure. Magnetic resonance angiography performed three months after the repair showed no coarctation and a patent graft (Fig. 2).

## DISCUSSION

A high index of suspicion is necessary for aortopulmonary or aortobronchial fistulas in patients presenting with recurrent hemoptysis and a history of surgery in the proximal descending aorta. This is a potential-



**Fig. 1.** Preoperative thoracic aortogram demonstrating recurrent coarctation at the site of Dacron patch aortoplasty and contrast extravasation through the patch.



**Fig. 2.** Magnetic resonance angiography three months after the repair showing the new interposition graft.

ly fatal complication of operations involving the descending aorta if not diagnosed and treated in an expeditious manner.

Clinical suspicion of a communication between the aorta and the bronchopulmonary tree is the key to correct diagnosis and management of these patients. Although diagnostic imaging techniques may be helpful, the fistula can only be demonstrated in a minority of patients prior to surgery. Standard chest X-rays may be completely normal, may show infiltrates in the adjacent lung parenchyma or may suggest an enlarged descending thoracic aorta. Computed tomography may help identify abnormalities in the descending thoracic aorta or in the adjacent lung. Standard contrast aortography can show changes in the aortic lumen due to previous repair or recoarctation. Rarely, contrast material can be seen to extravasate from the aorta into the lung tissue<sup>[2]</sup> or into the airways.<sup>[3]</sup> Pooling of contrast material in the lung or in the airways after the injection of dye provides indirect evidence for the presence of a fistula. Aortography is now less commonly performed due to the widespread use of CT angiography, which is currently the imaging modality of choice when there is a suspicion of an aorto-airway fistula. Magnetic resonance imaging does not provide any additional information to that obtained by CT angiography.

Fiberoptic bronchoscopy may be helpful to rule out other causes of hemoptysis. However, it may displace the thrombus overlying the fistula, which may result in massive hemoptysis.<sup>[4]</sup> It should be performed in the operating room under general anesthesia after the patient is draped as was done in this case.

Dacron patch plasty repair of aortic coarctation may be associated with several long-term complications, i.e. true or false aneurysms, recoarctation or fistulization into the adjacent lung parenchyma or into the airways. Aorto-airway fistulas developing after Dacron patch plasty repair of aortic coarctation have been reported in association with a pseudoaneurysm or a true aneurysm.<sup>[5]</sup> There was no coexisting aneurysm in our case and the fistula originated from the anastomotic suture line. Surgical repair should be planned without delay when there is a strong suspicion and when there is no other cause for hemoptysis. There have been reports of massive hemoptysis and death while awaiting surgery.<sup>[6]</sup> A double-lumen endotracheal tube may be lifesaving in cases of massive hemoptysis to protect

the contralateral lung. Cardiopulmonary bypass via the femoro-femoral route should be established prior to thoracotomy in hemodynamically unstable patients. The surgical approach to the fistula is through a left posterolateral thoracotomy in the fourth interspace. The patient is placed on left atrial-femoral bypass and proximal and distal aortic control should be obtained. The distal descending aorta can be used for outflow in cases where femoral arteries are of small caliber and not suitable for cannulation.<sup>[7]</sup> The fistula should be exposed after the aorta is clamped. The Dacron patch should be excised completely together with all coarctation tissue. Aortic continuity is restored using a Dacron tube graft of appropriate size. The damaged lung is either resected or repaired with sutures. Rarely, lobectomy or even pneumonectomy may be required due to extensive destruction of lung parenchyma. A pedicled pleural or pericardial fat flap should be placed between the new graft and the lung to prevent recurrent fistulas or graft infections.<sup>[4]</sup> Operative mortality for repair of aortopulmonary fistulas is reported to be around 16%.<sup>[1]</sup> Endovascular stent grafting has also been used in this group of patients.<sup>[8,9]</sup> This approach is associated with less mortality and morbidity compared with open surgical repair. However, there is still a need for long-term follow-up for early diagnosis and timely treatment of possible stent-related complications. We did not consider stent grafting in our case due to the presence of recoarctation accompanying the fistula.

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