



Aortoesophageal and aortobronchial fistula caused by *Candida albicans* after thoracic endovascular aortic repair

Aortoezofagusna i aortobronhijalna fistula posle endovaskularnog lečenja torakalne aorte od infekcije koju je izazvala *Candida albicans*

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Abstract

Introduction. Endovascular stent-graft placement has emerged as a minimally invasive alternative to open surgery for the treatment of aortic aneurysms and dissections. There are few reports on stent graft infections and aortoenteric fistula after endovascular thoracic aortic aneurysm repair, and the first multicentric study (Italian survey) showed the incidence of about 2%. **Case report.** We presented a 69-year-old male patient admitted to our hospital 9 months after thoracic endovascular aortic repair, due to severe chest pain in the left hemithorax and arm refractory to analgesic therapy. Multislice computed tomography (MSCT) showed a collection between the stent graft and the esophagus with thin layers of gas while gastroendoscopy showed visible blood jet 28 cm from incisive teeth. Surgical treatment was performed in collaboration of two teams (esophageal and vascular surgical team). After explantation of the stent graft and *in situ* reconstruction by using Dacron graft subsequent esophagectomy and graft omentoplasty were made. After almost four weeks patient developed hemoptisia as a sign of aortobronchial fistula. Treatment with implantation of another aortic cuff of 26 mm was performed. The patient was discharged to the regional center with negative blood culture, normal inflammatory parameters and respiratory function. Three months later the patient suffered deterioration with the severe weight loss and pneumonia caused by *Candida albicans* and unfortunately died. The survival time from the surgical treatment of aortoesophageal fistula was 4 months. **Conclusion.** Even if endovascular repair of thoracic aortic diseases improves early results, risk of infection should not be forgotten. Postoperative respiratory deterioration and finally hemoptisia could be the symptoms of another fistula.

Key words:

aortic aneurysm, thoracic; surgical procedures, minimally invasive; stents; postoperative complications; reoperation; candida albicans.

Apstrakt

Uvod. Endovaskularno lečenje aortnih oboljenja je minimalno invazivna alternativa otvorenim hirurškim procedurama. Infekcije implantiranog stent-grafta do sada su opisane sa incidencijom od 2%, ali nije bilo slučajeva aortoezofagusne i aortobronhijalne fistule kod istog bolesnika. **Prikaz bolesnika.** Bolesnik star 69 godina, primljen je u našu ustanovu devet meseci nakon endovaskularnog lečenja posttraumatske hronične aneurizme torakalne aorte zbog naglo nastalih bolova u grudnom košu i ruci refraktarnih na terapiju. Multislijsna kompjuterska tomografija pokazala je nakupinu između stent-grafta i ezofagusa sa tankim slojem gasa, dok je gastroskopija pokazala krvarenje na 28 cm od sekutića. Hirurško lečenje primenjeno je u saradnji vaskularnog tima i tima za hirurgiju jednjaka. Nakon eksplantacije stent-grafta i *in situ* rekonstrukcije Dakronskim graftom učinjena je ezofagektomija i omentoplastika grafta. Nakon četiri nedelje bolesnik je dobio hemoptizije kao znak aortobronhijalne fistule koja je uspešno lečena implantacijom aortne ekstenzije. Bolesnik je otpušten u regionalnu ustanovu sa negativnom hemokulturom, normalnim inflamatornim parametrima i respiratornom funkcijom. Nakon tri meseca, usled novog respiratornog pogoršanja u vidu pneumonije uzrokovane gljivicom *Candida albicans* bolesnik je preminuo. Vreme preživljavanja od operacije aortoezofagusne fistule iznosilo je četiri meseca. **Zaključak.** Iako je endovaskularno lečenje oboljenja torakalne aorte značajno unapredilo rane rezultate, ne treba zaboraviti na rizik od infekcije stent-grafta. Pogoršanje respiratorne funkcije i hemoptizije nakon operacije mogu biti znak nove fistule kod ovakvih bolesnika.

Ključne reči:

aneurizma, torakalna; hirurgija, minimalno invazivne procedure; stentovi; postoperativne komplikacije; reoperacija; candida albicans.

Introduction

Endovascular stent graft placement has emerged as a minimally invasive alternative to open surgery for the treatment of aortic aneurysms and dissections^{1,2}. The incidence of early and major complications is low, however long term results are about to come. There are few reports of stent graft infections and aortoenteric fistula (AEF) after endovascular thoracic aortic aneurysm repair; small series have reported rates of up to 5% after stent graft placement, and the first multicentric study (Italian survey) showed the incidence of about 2%^{3,4}.

Case report

We presented a 69-year-old male patient admitted to our hospital due to dysphagia, hoarseness and chest discomfort. After examination by means of chest radiography and multislice computed tomography (MSCT), posttraumatic thoracic aneurysm of 65 mm in diameter was revealed, due to frontal chest trauma 30 years ago in a car accident. The patient was successfully treated by endovascular stent graft implantation (Medtronic, Santa Rosa, CA-Valiant) using 12% of oversize. Perioperative antibiotics were used in usual prophylactic dosage (cephazolin 1.5 g i.v. at the beginning of the procedure, and every 8 hours for 72 hours). Postoperative recovery was uneventful and the patient was discharged after control MSCT had shown complete exclusion of aneurysm with the correct position of the stent graft, and preservation of left subclavian artery flow. First month control MSCT was also correct, while 9 months later the patient was admitted to the regional hospital with severe chest pain in the left hemithorax and the left hand refractory to analgesic therapy. The patient was treated with symptomatic therapy and 3 weeks later referred to our hospital with the same symptoms of inflammatory syndrome (leucocytes 18×10^9 , sedimentation rate 105 of the first hour, C-reactive protein 150 mg/L, incipient anemia, fever and malaise), and blood culture test positive on *Candida albicans*. MSCT was performed showing the normal stent graft position, no fracture, no endoleak, and a collection between stent graft and the esophagus with thin layers of gas (Figure 1).

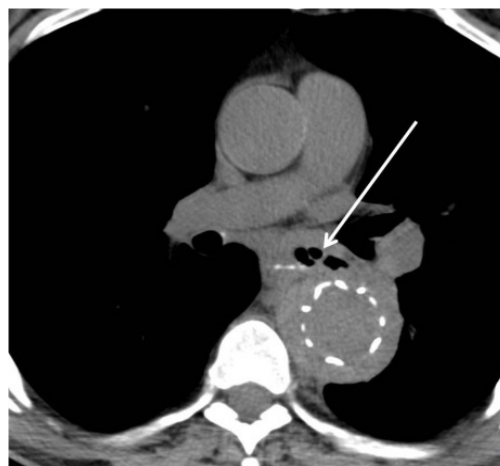


Fig. 1 – Axial image of the stent-graft implanted in the thoracic aorta with a gas collection in the surrounding tissue (arrow).

Gastroendoscopy was performed, and visible blood jet 28 cm from incisive teeth was visualized. Short after this examination the patient had first time hematemesis of 500 mL of fresh blood. Blakemore tube was used to control hemorrhage temporarily. Surgical treatment was performed in collaboration of two teams (esophageal and vascular surgical team). In condition of partial femoro-femoral extracorporeal bypass (ECBP), through left thoracotomy in the 4th intercostal space, there was no visible hematoma, and no visible hemorrhage, but diffuse inflammation was noticed, with adhesions between the thoracic aorta, the esophagus and the left lung. Proximal control was gained between the origins of the left common carotid and the left subclavian artery, while distal control was secured in the distal thoracic aorta, 3 or 4 cm distal from the lower margin of the stent graft. After resection of the thoracic aorta and explantation of the stent graft, *in situ* reconstruction by using Dacron graft was made. After that it was much easier to divide esophagus from severely inflammation of periaortic tissue, and to perform esophagectomy. Midline laparotomy, mobilization of peritoneum and its transposition through diaphragm incision, and omentoplasty of Dacron graft were followed by nutritive gastrostoma and cervical jejunostoma (Figures 2a, b and c).

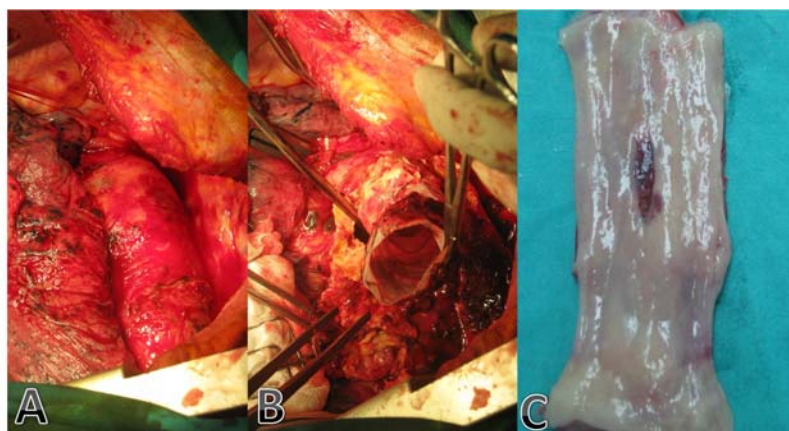


Fig. 2 – A) Inflammation of the thoracic aorta and surrounding tissue without extravasation of blood; B) Explantation of the thoracic stent-graft; C) Short lesion on the inner wall of the explanted esophagus.

Culture of the aneurysmatic sac and explanted stent graft was positive on *Candida albicans*, as well as was sputum culture. In the long lasting postoperative recovery time a lot of problems occurred. During almost four weeks of slow recovery, several evacuations of 500–1000 mL of pleural effusion (positive on *Candida albicans*) were performed. Eventually, the patient developed hemoptysis. Control MSCT revealed a n aortobronchial fistula at the level of proximal anastomosis and the left principal bronchus. According to the patient general condition we decided to perform endovascular treatment with implantation of another stent graft. The only stent graft available on shelf was aortic cuff of 26 mm that was successfully implanted and patient respiratory function recovered slowly, as was further general recovery (Figures 3a and b).

delayed diagnosis partly because of the fact that clinical presentation was unusual. The patient complained of left arm and hemithoracic chest pain with no inflammatory syndrome. Very thin periaortic inflammation was visible but inexperience made us not be act. Probably it could be much more easier to treat this initial infection – periaortitis. Later on, the patient was admitted with severe infection and active bleeding. Surgical repair of infection in thoracic cavity is much more demanding than the same complication in the abdominal position when extra-anatomic reconstruction (axillobifemoral) is feasible. Extra-anatomic reconstruction due to thoracic graft infection considers a two-stage procedure: bypass from the ascending to distal descending thoracic aorta, or supraceliac aorta, as the first and extirpation of the stent graft and ligation of the thoracic aorta as the second



Fig. 3 – A) Periaortic hematoma and extravasation of blood at the level of the proximal anastomosis causing hemoptisia; B) Short aortic cuff implanted in the thoracic aorta due to suspected aortoesophageal fistula.

All these procedures and difficult recovery caused severe muscle atrophy and needed weeks to recover. The patient was discharged and referred to the regional center with negative blood culture, normal inflammatory parameters and respiratory function. Three months later the patient suffered deterioration with a severe weight loss and pneumonia caused by *Candida albicans* and unfortunately died. The survival time from the surgical treatment of aortoesophageal fistula was 4 months.

Discussion

Introduction of new technologies and minimally invasive surgery did not change our relation to one of the biggest challenges in vascular surgery – graft infection: it still happens! Minimally invasive surgery increased the number of treated patients, as well as the total number of patients with this complication. Thoracic endografting not only increased the number of treated patients, and the frequency of comorbid conditions, but it also consequently devastated the immune system making the patient more susceptible to infection. In addition, endovascular stent grafts are more susceptible to infection than grafts for open surgery⁵.

Treatment of this severe and devastating condition in the presented patient was even more difficult because of the

stage⁶. This kind of procedure is possible when the diagnosis is established timely, because the first stage does not resolve bleeding that can be dreadful. Alternative could be another endovascular procedure as a bridge to control bleeding, and then the two stage procedure with removal of the infected graft and adjusted structures in the second stage. However, since we did not have any stent grafts on the shelf, we performed the one-stage procedure due to unstable condition with esophageal bleeding of the patient.

Causes of stent graft infections have several explanations: it could be an already present fistula, or contamination from intraluminal aneurysmatic thrombus, or surgical primary contamination during surgical manipulation (more frequent in radiology suits). There are some other theories like ischemia of esophageal or bronchial wall caused stent implantation, or aggressive oversizing^{7,8}. A lot of possible causes of this complication suggest that the multifactorial pathogenesis is most probable. An isolated infected organism could reveal a possible cause, if skin bacteria are isolated than skin contamination is more probable. In the presented patient *Candida albicans* was isolated from blood, aneurysmatic sac and stent graft specimens. Even more in postoperative recovery *Candida albicans* was isolated from pleural effusions as well as from sputum during pneumonia just before death - it has never been eradicated. Only three cases with *Candida albicans*

contamination were reported and all with lethal outcome. The early diagnosis could give us time for initial antifungal therapy with modern developed drugs, however due to unstable condition in the presented case there was no time for this.

Conclusion

Although endovascular repair of thoracic aortic diseases improves early results, risk of infection should not be forgotten. Close follow-up and the timely diagnosis could give us

time for preparing a patient with potential antimicrobial medications, and for extra-anatomic, two-stage reconstruction. Postoperative respiratory deterioration and finally hemoptisia could be the symptoms of another fistula.

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