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Journal of the Chinese Medical Association 75 (2012) 474–478

www.jcma-online.com

Case Report

Endovascular treatment of a nontraumatic left subclavian artery pseudoaneurysm

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Received November 9, 2010; accepted August 9, 2011

Abstract

Mycotic subclavian artery pseudoaneurysms are rare. There are controversies over the surgical or endovascular approach as the treatment of choice for these lesions. The standard surgical debridement might not be a choice for poorly surgically reachable lesions or for patients with multiple comorbidities. Endovascular aneurysm repair may be an effective alternative in selected cases. This treatment was rarely reported previously. Herein, we present a high-surgical-risk case with a highly suspected left subclavian arterial mycotic pseudoaneurysm, which, although difficult to approach surgically, was successfully managed with stent grafting and a complete antibiotic treatment course. An 89-year-old male was admitted due to intermittent fever and hemoptysis for 2 months. *Salmonella* group B was cultured from his sputum, and a 3.5 cm pseudoaneurysm was identified by chest multidetector-row computed tomography (MDCT) angiogram. Endovascular treatment with a graft stent was chosen due to high surgical risk and difficult surgical access to the lesion. The intervention was well planned *ad hoc*, based on MDCT images and meticulously performed by dual endovascular approaches. Antibiotics were continued after the procedure, and the patient was discharged from the hospital. As MDCT disclosed near-complete regression of the pseudoaneurysms 2 months later and the patient was in healthy status, antibiotics were continued for 6 months. He was readmitted 11 months later due to lacunar infarction with minor pneumonia over the left lower lung in which *Salmonella* enteritis was also diagnosed. After this acute event, he was again hospitalized 14 days later due to sepsis with adult respiratory distress syndrome and shortly expired despite all emergent treatment measures. No evidence of local subclavian infection recurrence was noted throughout or related to subsequent events. In conclusion, endovascular treatment of an infected subclavian artery pseudoaneurysm could be a choice in selected patients, but treatment of underlying infection determines the clinical outcome. Copyright © 2012 Elsevier Taiwan LLC and the Chinese Medical Association. All rights reserved.

Keywords: endovascular treatment; mycotic aneurysm; pseudoaneurysm; stent graft

1. Introduction

Aneurysms of the subclavian arteries are rare and usually atherosclerotic or post-stenotic.¹ However, pseudoaneurysms of the subclavian artery are mostly traumatically induced, with most cases following central-line catheter placement, which may also injure unilateral vertebral arteries.² These

pseudoaneurysms of the subclavian artery could be treated by ultrasound-guided local thrombin injection,^{2,3} coil embolization,⁴ or endovascular treatment.⁵ On the other hand, mycotic subclavian artery pseudoaneurysms are rare,^{6–9} occurring mostly as a complication of drug addiction. They can present with pulsatile mass, chest pain, distal thromboembolism, or signs of local tissue compression,^{10–12} but they can also be lethal due to local perforation. Standard surgical treatment consists of surgical debridement with extra-anatomic bypass or *in situ* graft placement, followed by long-term antibiotic treatment.¹³ However, it might not be a choice for poorly surgically reachable lesions or for patients who are

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immunocompromised⁸ or have multiple comorbidities. On the other hand, endovascular aneurysm repair may be a less invasive, but effective, alternative to open repair of mycotic aneurysms, especially in high-risk patients and for surgically inaccessible lesions. In certain cases, it could be the only choice. This treatment was rarely reported earlier.⁸

2. Case report

An 89-year-old man was admitted due to intermittent fever and hemoptysis for a total of 2 months. He was first diagnosed to have pneumonia and treated at a local hospital. However, fever and hemoptysis relapsed 1 week prior to current admission. He had hypertension and was under medical control for about 10 years. No chest trauma or surgical history could be elicited. During the current hospital stay, *Salmonella* group B was cultured from his sputum and antibiotic treatment

with cefotaxime was initiated. Chest roentgenogram showed a mass lesion in the left retroclavicular region, extending to the aortic knob (Fig. 1A). Chest multidetector-row computed tomography (MDCT) angiogram showed a contained ruptured pseudoaneurysm, 35 mm in diameter with a neck of 13 mm, at the proximal left subclavian artery and proximal to the vertebral artery origin (Fig. 1B–D). Therefore, left subclavian arterial mycotic aneurysm was highly suspected 3 days after antibiotic treatment. Complete aneurysm extrusion was achieved by endovascular approach with a graft stent, but no open repair was intended due to high surgical risk and poor lesion access to the retroclavicular area. The occlusion procedure was planned ad hoc in detail, based on MDCT images, and meticulously performed by bilateral arterial access with an 11F introducer sheath placed via left brachial cutdown and an 8F sheath in the right femoral artery. A 10 × 50 mm self-expandable stent graft (Wallstent, Boston Scientific, Galway,

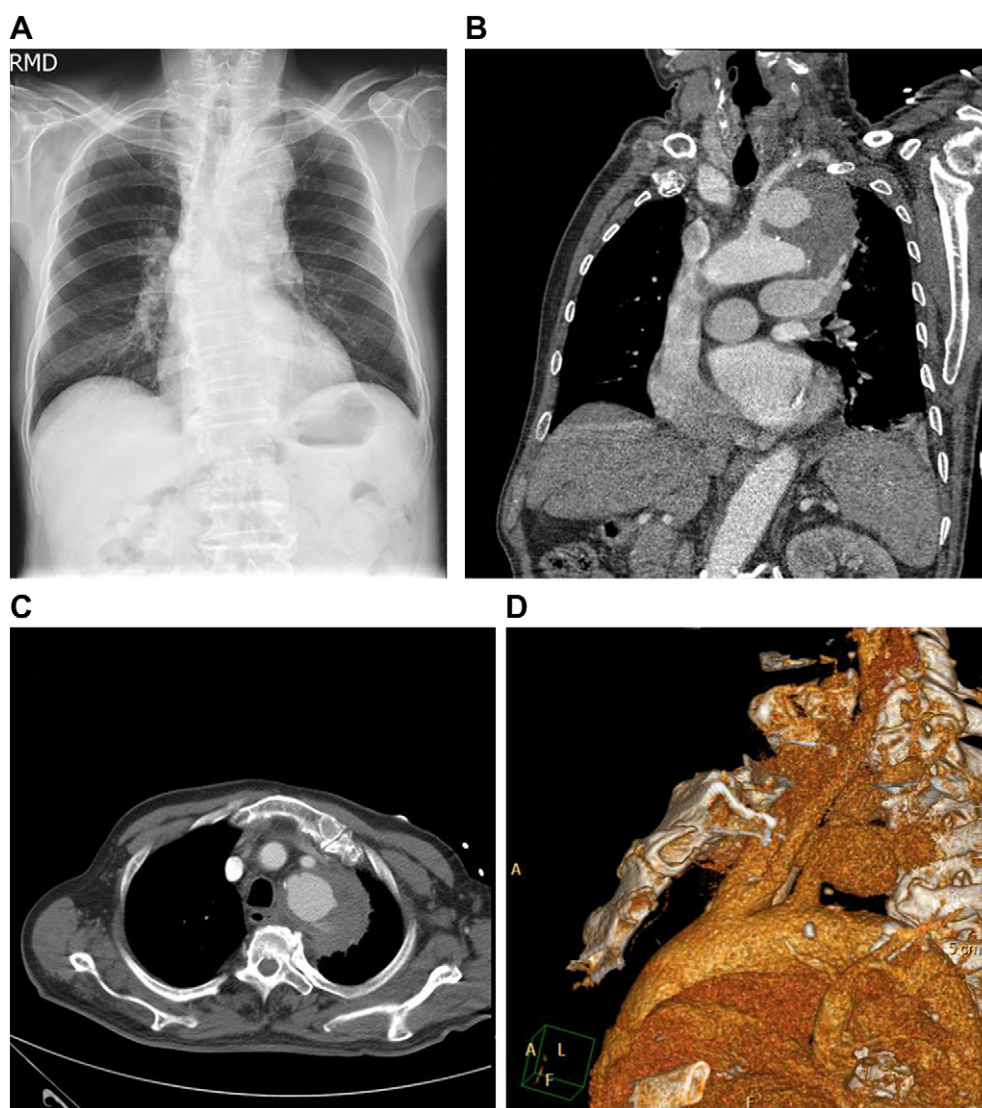


Fig. 1. (A) The CXR showed a mass lesion in the left upper lung field involving the retroclavicular region and extending to the aortic knob. (B and C) The contrast medium MDCT in saggital section showed a $3.2 \times 2.7 \text{ cm}^2$, previously ruptured mycotic aneurysm (arrows) at the proximal left subclavian artery contained by surrounding blood clot and reacting tissues ($7.2 \times 6.8 \text{ cm}^2$ in size). (D) The volume-rendered reconstructive image is shown here. CXR = chest X-ray; MDCT = multidetector-row computed tomography.

Ireland) was introduced retrogradely via the left brachial sheath and positioned precisely under contrast guidance with an 8F JR4 guide through the femoral sheath to avoid jailing in the left vertebral artery and, at the same time, to prevent stent dislodgement into the aorta. Complete sealing-off of the aneurysm was achieved by postdilatation with a 10×20 mm balloon up to 10 bar along the entire device for multiple times. Minimal contrast leakage into the pseudoaneurysm was still present on final control angiogram, but the flow was very sluggish. There was no procedural complication or neurological sequela. After the procedure, hemoptysis and fever subsided until 2 weeks later, when the patient again suffered from hemoptysis that was more massive than before, severe dyspnea, and desaturation of blood oxygen. Emergency intubation was done and MDCT was repeated, but it revealed no endoleak (Fig. 2A). Bronchoscopy was also performed, and no active bleeding point could be identified. The bleeding was presumed to be due to bronchial artery erosion by smoldering infection in the extruded mycotic aneurysm. After conservative treatment for 1 week, the patient was weaned from the ventilator successfully. Owing to suspected antibiotic-related neutropenia 4 weeks later, cefotaxime was substituted by ciprofloxacin. Finally, the patient was discharged from hospital 55 days after his admission. Ciprofloxacin was continued for another 5 months and then stopped as the patient's general condition was quite stable, and MDCT follow-up 2 months post intervention revealed dramatic reduction in the aneurysm size. The patient was readmitted 11 months post intervention and was hospitalized for 11 days due to lacunar infarction with minor pneumonia over the left lower lung, but the left upper lung adjacent to the previous mycotic aneurysm was completely clear. During the hospitalization, *Salmonella* enteritis was diagnosed based on abdominal distension, dull pain, gastrointestinal tract bleed, and positive stool culture growth. However, an abdominal CT scan showed negative finding. After antibiotic treatment, he was discharged. However, he was admitted again 14 days later due to sepsis

with adult respiratory distress syndrome, hypotension, bradycardia, and hypothermia. The patient soon died despite all emergent treatment measures. No evidence of local subclavian infection recurrence was noted throughout or related to subsequent events.

3. Discussion

Pseudoaneurysms of the subclavian artery are mostly traumatic, with most cases following central-line cannulations.² These catheterization-injury-related pseudoaneurysms are usually located at or distal to the subclavian–vertebral junctions,² and endovascular strategy is the treatment of choice in modern practice.⁵ They can also occur as a consequence of shoulder dislocation,¹⁴ local balloon dilatation,¹⁵ Blalock–Taussing shunting,¹⁶ or type IV Ehlers–Danlos syndrome.¹⁷ It may also follow infection of previously placed stents.¹⁸ Our reported case did not have such histories. Instead, the pseudoaneurysm of our patient was proximal to the vertebral orifice. In view of the clinical presentations, the milieu of active infection, the image of a large expanding pseudoaneurysm with surrounding hematoma and compression on the neighboring lung that had relatively clear parenchyma, and the uneventful recovery course after antibiotics treatment, it was strongly proposed that the aneurysm in our patient was primary and mycotic despite our having no direct microscopic or blood culture evidence. The source of infection in our case also remained elusive.

Mycotic aneurysm was first described by William Osler in 1885. It often involves the bifurcation or the narrowing portion of the arterial wall. Clinically, mycotic aortic aneurysm is relatively rare, with an incidence of about 0.65–2% of all aortic aneurysms. *Staphylococcus aureus*, *Salmonella*, and *Streptococcus* species are the most common microorganisms. *Salmonella* species frequently involve gastrointestinal tract, but often there is no history of gastroenteritis. The most common presentation is fever or abdominal pain. *Salmonella*

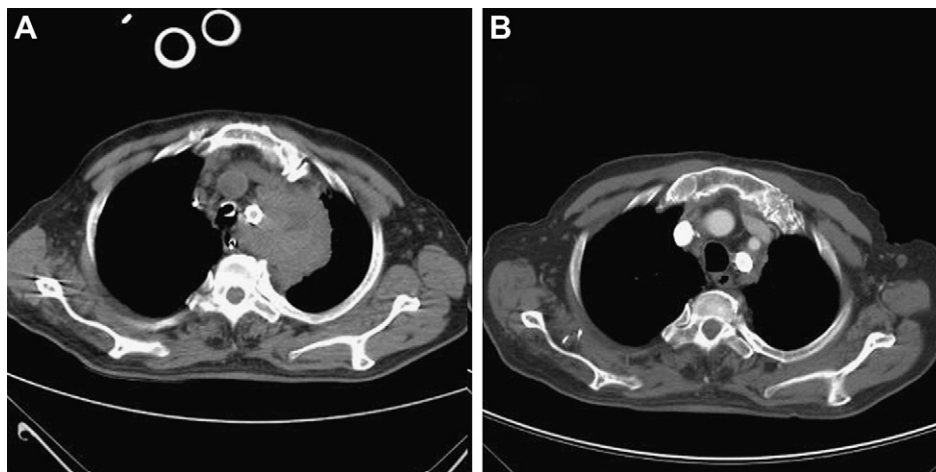


Fig. 2. The transverse-view contrast medium MDCT image taken 14 days after endovascular stent grafting showed adequate stent positioning, full stent expansion, and no endoleak. (A) The blood clot was still noted around the treated mycotic aneurysm. (B) The contrast medium MDCT image done 3 months after endovascular treatment in the right clearly showed almost complete resolution of both the mycotic aneurysm and the surrounding hematoma. The vessel lumen was obscured due to partial volume effect. MDCT = multidetector-row computed tomography.

mycotic aneurysm is difficult to diagnose unless there is a positive culture growth. Because it carries a high rupture and mortality rate, we should always keep this possibility in mind, especially in immunocompromised patients. Although the sputum culture of our patient grew *Salmonella* group B, a frequent microorganism for mycotic aneurysm, his blood culture was negative. This might be due to partial antibiotic treatment for alleged pneumonia at the local hospital prior to current admission. Although pneumonia may be the source of infection, it was more probable that the patient's pneumonia was secondary to the mass effect of the large pseudoaneurysm and the surrounding hematoma. In the patient, the bacterial source should be in the gastrointestinal tract as *Salmonella* enteritis was firmly diagnosed 11 months later in his second admission, presenting with typical symptoms and signs. Mycotic aneurysms involving peripheral arteries are much less frequent than those involving the aorta. The most frequent ones are those involving femoral arteries, occurring mostly in intravenous drug abusers. Blood culture can be negative in up to 25–50% of cases. Mycotic subclavian artery pseudoaneurysms are rare,^{6–9} usually seen in drug addicts. Risk factors for mycotic aneurysms include chronic alcoholism, diabetes mellitus, and chronic renal failure. Our patient did not have any of these risk factors, and the cause was vague.

Regarding the treatment of mycotic subclavian artery pseudoaneurysms, there remain controversies.^{7,8} Some authors are strongly opposed to the use of stent grafts in infected aneurysms,⁷ citing that the infected stents could be a source for mycotic pseudoaneurysm.¹⁸ However, the standard surgical management, especially the intrathoracic one, might not be a choice in high-risk patients with unfavorable lesion locations, like the one presented in this report. On the other hand, endovascular aneurysm repair may be a less invasive, but effective, alternative in particular clinical settings, but this was rarely reported priorly.⁸ This is because, at one extreme, endovascular treatment could be associated with smoldering infection in the prosthetic graft and aneurysmal bed. Stent thrombosis, oversizing, undersizing, migration, rupture, and endoleak could be other complications. Therefore, a prolonged and complete antibiotic treatment is an important adjunct to endovascular treatment. There is no consensus on how long the antibiotics should be used after endovascular management. However, prolonged antibiotic treatment for at least 6 weeks or even lifetime is suggested in the literature. Third-generation cephalosporins, especially ceftriaxone and newer quinolones, were used for *Salmonella* infection.

Complications after endovascular treatment of mycotic aneurysms have been reported, including sepsis, aneurysm rupture with mortality,¹⁹ type 1 endoleak with sepsis,²⁰ and aortoenteric fistula.²¹ One author even reported *Salmonella* infection flare-up years after treatment.²¹ In our case, intravenous cefotaxime was used for 1 month, and ciprofloxacin for another 5 months and then stopped based on clinical judgment. Unfortunately, the use of antibiotics was proved insufficient, and the infection relapsed 11 months post intervention and turned out to be lethal in the old patient. However, even at this time, there was no sign of local infection recurrence at the subclavian

stent graft site. One study reported that although early mortality rate of endovascular repair was relatively lower than that of open surgery, rate of late complications such as late aneurysm-related events could be higher than that of open surgery. Kan et al found a 12-month survival rate of 39% for patients with persistent infection but 94% for those whose aneurysms were healed. Although stent grafting might be a reasonable alternative to open surgical repair in certain lesions in high-risk patients, several issues deserve particular attention beforehand. It should not be considered when active infection has not been controlled by effective antibiotics. It is not suitable for lesions with vital branches that could not be sacrificed, lesions without proper landing zones, or lesions involving long segment with frail walls. Meticulous treatment planning based on thorough considerations of clinical and imaging information was key to successful endovascular stent graft treatment in our case. The lesion location and vessel characteristics were quite suitable for endovascular obliteration, and the size and length of the stent were predecided by radiographic images. The endovascular repair was approached by bilateral access technique in which the bulky stent graft was introduced retrogradely via left brachial cutdown to ensure successful device delivery, smooth device manipulation, and precise stent positioning, whereas the large guide in the aorta provided antegrade lightening and help prevent proximal stent dislodgement in case of stent migration during deployment. The entire procedure was performed successfully as planned, and there were no neurological sequelae due to distal embolism or sacrifice of the vertebral artery. Neither there was any endoleak. Complete resolution of the aneurysm in follow-up and no recurrence of local infection at the subclavian site might convey the message that active infection was eliminated and the risk of aneurysm rupture is minimal. To the best of our knowledge, only one case of subclavian mycotic aneurysm treated with stent grafting has been reported so far. The patient was an immunosuppressed young girl with *Aspergillus* infection, who died of leukemia relapse 11 months after the procedure.

In conclusion, endovascular stent graft repair of large subclavian artery mycotic aneurysm might be a safe and feasible option for certain high-surgical-risk patients and for surgically inaccessible lesions, whereas surgical debridement remains the standard treatment. Endovascular treatment should be tailored to each individual patient and his/her unique clinical settings after comprehensive treatment planning. Prolonged antimicrobial treatment should be the adjunctive therapy, and the duration may vary from case to case to reduce the risk of persistent infection and even sepsis. Treatment of underlying infection determines the clinical outcome.

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