

Retrospective study of patients with asymptomatic, incidentally discovered, thoracic aortic pseudoaneurysm treated in a single institution

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Abstract:

Introduction: Spontaneous thoracic aortic rupture can result in disastrous complications. Rarely, such an event can be asymptomatic and in these cases it may escape diagnosis, posing an immediate threat to patient's life.

Methods: We conducted a single-center retrospective analysis of all patients treated with thoracic aortic endovascular repair [TEVAR] for various pathologies, between 01/2019 - 01/2024. The subgroup of patients with incidentally found pseudoaneurysms of the thoracic aorta were identified and further analyzed. Demographic information, procedural details and follow-up data of these cases were collected.

Results: Initially, 40 patients were identified, among which only three fulfilled the criteria for an incidental finding of a spontaneous, concealed, asymptomatic rupture of the descending thoracic aorta, presenting as a pseudoaneurysm which was revealed in computed tomography [CT], performed for other medical reasons. All patients were treated, in an emergency setting, with TEVAR. Branched endografts and chimneys were used in 2 out of 3 cases. In one case, a proximal extension with a branched endograft was necessary to treat a type IA endoleak after an initial implantation of standard TEVAR. There was one in-hospital mortality due to unrelated medical reasons.

Conclusion: Asymptomatic presentation of thoracic aortic pseudoaneurysm is possible and in these patients prompt endovascular repair can reduce the risk for serious complications or death.

INTRODUCTION

Spontaneous rupture of the thoracic aorta is a devastating event¹. Most commonly it presents with hemodynamic collapse of the patient, requiring immediate treatment. However, concealed, asymptomatic rupture can be challenging to diagnose, as it may present with atypical symptoms or be completely asymptomatic and be identified as an incidental radiological finding days, weeks or even months after the event.^{1,2} This clinical entity poses a unique threat for the patient, as he is unaware of the pathology and the potential complications can be life-threatening. The first 24h mortality of patients with contained rupture of a thoracic aortic aneurysm is around 80%².

As stated by the ACC/AHA 2022 Guidelines, a pseudoaneurysm of the thoracic aorta following a blunt traumatic thoracic aortic injury [BTTAI] is classified as Grade 3 BTTAI and should be managed urgently, as they are at a high risk of progres-

sion and rupture³. However, in the present study incidental finding of non-traumatic thoracic aortic pseudoaneurysm was our focus. To our knowledge, data are very limited regarding pathophysiology, natural history, and prognosis of this clinical entity and consequently robust diagnostic and therapeutic algorithms are not currently available.

MATERIALS AND METHODS

Study design:

We conducted a retrospective observational study of all patients treated with TEVAR, in our department, between 01/2019 - 01/2024, according to the STROBE statement for observational studies.⁴ Our study population consisted of patients treated for an asymptomatic, incidentally found thoracic aorta pseudoaneurysm.

Inclusion criteria:

All patients with thoracic aortic pathology undergoing interventional treatment throughout the study period were identified. This included patients with thoracic aortic aneurysm or pseudoaneurysm, thoracoabdominal aneurysms, patients with acute aortic syndromes that involved the thoracic aorta [aortic dissection - AD, intramural hematoma - IMH, penetrating aortic ulcer - PAU or thoracic aorta rupture] and also patients with acute thoracic aortic injuries after blunt or penetrating trauma.

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After the initial screening, only patients with an incidental finding of a thoracic aortic pseudoaneurysm, were analyzed.

Exclusion criteria:

Patients with aortic pathology that did not involve the descending thoracic aorta were excluded [for example, pathology including the ascending aorta, abdominal aorta and iliac arteries]. Additionally, patients with thoracic aortic pathologies that were managed conservatively were also excluded.

Endpoints

Information that were collected included patients' demographic characteristics [age, gender, ethnicity], comorbidities and past medical history, clinical presentation, the modalities that were used for the diagnosis, the lesions' characteristics [meaning its' diameter, neck length, aortic branches involved] and type of treatment, including treatment technical characteristics. Endpoints included technical and clinical success, need for re-intervention, length of hospital stay and/or need for stay in the ICU, patients' mortality and morbidity and follow-up outcomes.

RESULTS

Study population

After searching our database, 43 patients were at first identified. Fifteen patients were treated for thoracoabdominal aortic aneurysm, 17 patients presented with thoracic aortic dissection treated either in the acute or subacute phase and 8 patients presented contained aortic rupture after trauma and were subjected to endovascular repair. Finally, 3 patients fulfilled the criteria for an asymptomatic, incidentally found, thoracic aortic pseudoaneurysm, who were treated with TEVAR. The above-mentioned results are also depicted in the flow-diagram presented in Figure 1.

Overall results

All patients were treated <24h after initial diagnosis. One patient was treated with Chimney TEVAR, while the remaining two were initially treated with standard TEVAR, with LSA coverage in order to achieve an adequate proximal seal. In one of those cases this was not successful and the deployment of an additional branched device was required to treat a Type IA endoleak. Post-procedurally all patients were admitted to the ICU. Two patients were discharged in good general condition, while one patient died after several weeks of hospitalization due to unrelated medical reasons. The results regarding patient information and procedural data are summarized in Table 1. Endpoints of the analysis are summarized in Table 2. In the following section a detailed report of these cases is provided.

Case 1

The first patient is a 78 years old male who was hospitalized two months ago for fever of unknown origin, which was finally attributed to lung infiltrations. During his hospitalization he



Figure 1: Flow chart summarizing selection of the study population.

underwent chest x-ray, where a radiopaque saccular mass in the left mediastinum was depicted, and which wasn't present in a previous x-ray from the patient's history six months ago (Figure 2). However, at that time, this finding wasn't further assessed. After his discharge, he was advised to undergo a CT scan, in order to review the aforementioned lung infiltrations. In the CT scan a pseudoaneurysm of the thoracic aorta was identified, measuring 60mm of maximum diameter. Then, he was referred to our department where he underwent CT angiography [CTA] and a concealed rupture of the thoracic aorta, originating proximal to the origin of the left subclavian artery [LSCA] was diagnosed. Specifically, a ruptured penetrating aortic ulcer [PAU] was identified as the probable cause of rupture (Figure 3). He was at first admitted to our department and then was treated, urgently, with chimney - TEVAR [Ch-TEVAR] under general anaesthesia. A TEVAR endograft Terumo RelayPro 34/30 154mm was deployed with the proximal extent in the Z2 zone, just proximal to the origin of the LSCA and distal extent to T5 zone, while two balloon expandable covered stents Atrium Advanta V12 [BE] 12x61mm were deployed in the LSCA, with the chimney technique, in order to preserve LSCA patency (Figure 4). The patient had a high-grade left internal carotid artery stenosis, measured around 90%, so assuring that the left vertebral artery remained patent was important in order to preserve brain perfusion. In the completion angiography adequate coverage of the rupture point was verified, while the LSCA & left vertebral artery remained patent, without endoleak or active extravasation. The patient was transferred post-operatively to the ICU, awake, for further monitoring where he remained for 24h and then was transferred to the Vascular Surgery Unit in good general condition, fully mobilized. During his hospitalization he developed contrast-induced acute kidney injury [CI-AKI], with a peak cre-

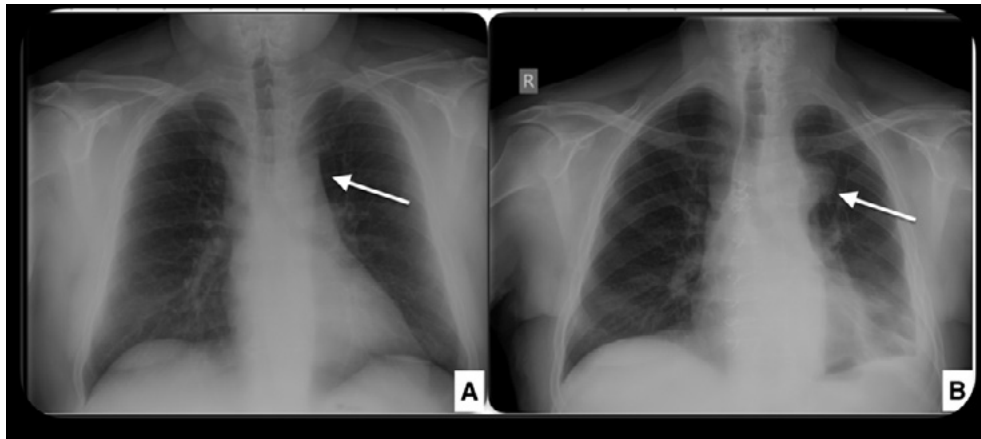


Figure 2: X-ray of the 1st patient at the time of presentation (B) and 6-months ago (A). White arrow indicates a mediastinal mass which was not apparent in the 1st examination.



Figure 3: CT angiography of the 1st patient with the white arrow indicating the pseudoaneurysm of the thoracic aorta.

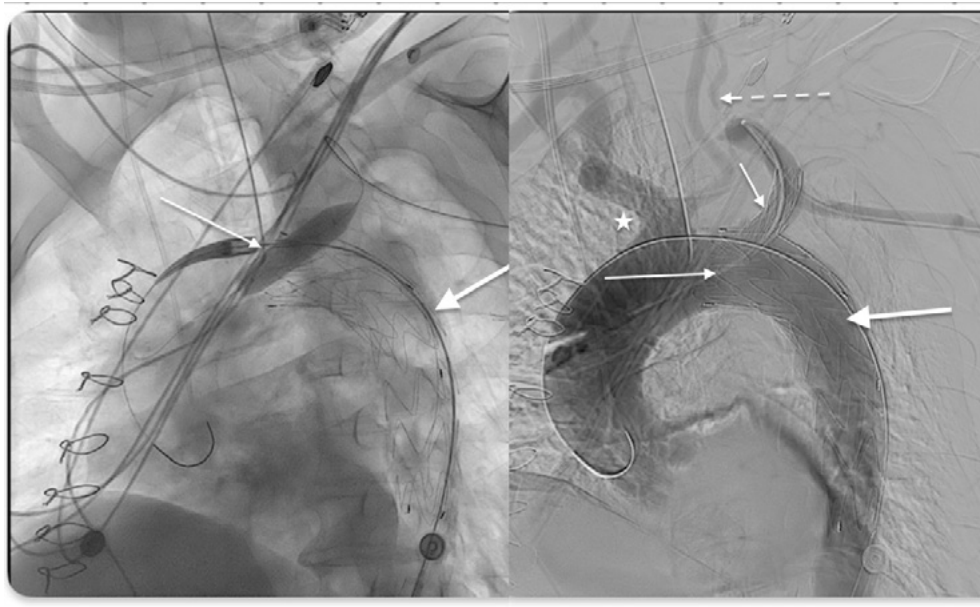


Figure 4: Intra-operative images of the 1st patient. In the left panel the main endograft (thick white arrow) and the balloon expandable chimney stent in the LSCA (thin white arrow) can be seen. In the right panel an intraoperative angiography indicates successful sealing with patent main graft and chimney graft (thin white arrows). Additionally, the left vertebral artery (dashed arrow) and the common origin of the brachiocephalic truck and the left common carotid artery (star) are depicted.

atinine level of 1.67mg/dl, which prolonged his stay for five days. The fourth postoperative day the patient underwent a CTA, for post-operative monitoring, where a gutter endoleak between the main endograft and the Chimney graft, to the saccular pseudoaneurysm, was identified, without clinically significant growth of the aneurysm diameter. A third scan was performed during the eighth postoperative day, for investigation of an episode of shortness of breath, where spontaneous occlusion of the endoleak was observed, without growth of the pseudoaneurysm sac. He was discharged the ninth post-op day, with baseline renal function restored. The patient has successfully completed short-term follow-up, at one month after the discharge, where in the CTA both no endoleak and sac regression were identified. He has yet to complete however one year follow-up.

Case 2

The second patient was a 73 years old male with a history of lung lymphoma with multiple chemo- and radiation-therapy sessions, who was at that time hospitalized in the Hematology department for dyspnea, cough and hoarseness of voice investigation. He underwent non contrast thorax-CT, for possible respiratory tract infection. However, in the CT scan, a pseudoaneurysm of the descending thoracic aorta was identified. Following the above-mentioned finding, the patient underwent CTA, where a concealed rupture of the thoracic aorta, originating distal to the origin of the LSCA was diagnosed, with a pseudoaneurysm measuring 85mm of maximum diameter (Figure 5). He was then admitted to our department

and treated, urgently, based on the proposed guidelines, with the deployment of a TEVAR endograft TTerumo Relay-Pro 32/32x160mm with proximal extent in the Z2 zone just proximal to the LSCA and with distal extent to T5 zone. In the completion angiography a Type IA endoleak was depicted (Figure 6), and therefore the patient underwent a 2nd procedure 24h later, with the insertion of a proximal extension of the endograft with the deployment of a Castor Endograft Endovastec Lombard 36/30x200mm, branch 10x25mm@10mm (C363010-2002510), with a branch to the left common carotid artery [LCCA]. During the deployment, the endograft slightly migrated proximally, causing severe stenosis of the orifice of the brachiocephalic artery [BCA]. Immediately, the right common carotid artery [RCCA] was dissected and cannulated and a balloon expandable covered stent Atrium V12 12x61mm was deployed in the BCA, with the chimney technique (Figure 7). In the completion angiography no endoleak was identified, with good patency of the BCA and LCCA. Postoperatively, he had a prolonged stay at the ICU, which lasted for six days, due to severe face and larynx swelling which didn't allow extubation. Moreover, the patient acutely developed severe heart failure, with a left ventricle ejection fraction [LVEF] 15% and had multiple episodes of acute respiratory distress syndrome [ARDS] which were finally attributed to bronchiolitis. The patient was transferred to the Pulmonology department for further treatment, on the forty-ninth post-op day. However, he passed away ten days later, due to aplastic anemia and acute respiratory failure.



Figure 5: CT angiography of the 2nd patient with the white arrow indicating the pseudoaneurysm of the thoracic aorta.

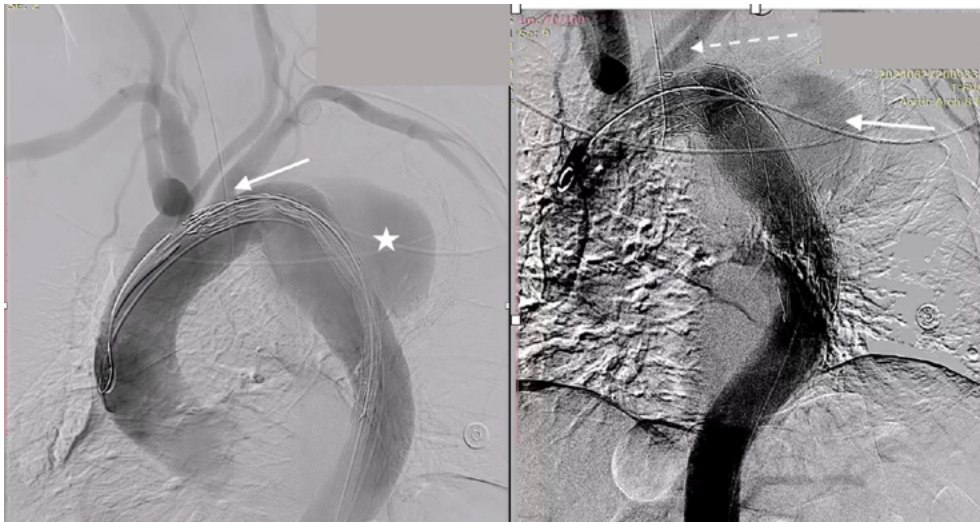


Figure 6: In the left side an intraoperative angiography shows the endograft before its deployment proximal to the origin of the LSCA (white arrow) and the sac of the pseudoaneurysm (star). In the right side, postoperative CT angiography indicates continuous perfusion of the pseudoaneurysm sac due to Type IA endoleak (white arrow) despite successful deployment of the endograft at the level of the LSCA (dashed arrow).

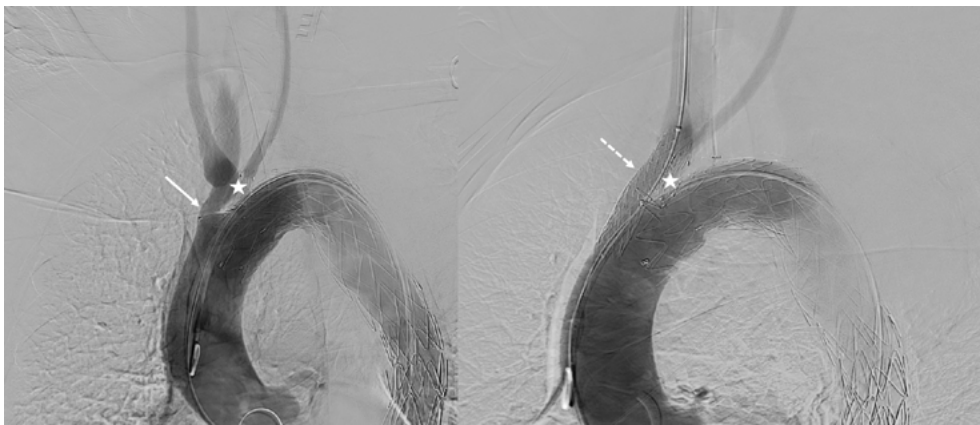


Figure 7: Intraoperative images of the 2nd patient during the deployment of the branched endograft. In the left side the proximal part of the endograft partially covers the origin of the brachiocephalic artery (white arrow). As bailout, a chimney graft was deployed in the brachiocephalic artery through the right common carotid artery, as shown in the right image (dashed arrow). Star indicates the branch inside the left common carotid.

Case 3

The third patient is a 44 years old female, with a history of Adamantiades - Behcet's disease [ABD], under therapy with colchicine and methotrexate, who was hospitalized at that time in the Pulmonology department, for thoracic pain, migrating to the back and possible upper respiratory tract infection. During her hospitalization she underwent a non-contrast thorax-CT. Just as the above-mentioned case, the CT showed a periaortic hematoma in junction with the descending thoracic aorta, which was described as a potential concealed rupture of the thoracic aorta forming a saccular pseudoaneurysm measuring 60mm of maximum diameter. Then, she underwent CTA, where a concealed rupture of the thoracic aorta, originating distal to the origin of the LSCA was identified (**Figure 8**). She was then transferred to our department for further treatment. She underwent urgent TEVAR, under general anaesthesia. A TEVAR endograft Terumo Relay PRO 28x130mm,

with proximal extent to the Z2 zone, just proximal to the LSCA and distal extent to T5 zone, was deployed. In the completion angiography, adequate coverage of the rupture point was verified, while LSCA & left vertebral artery remained patent, without endoleak or active extravasation. Her postoperative course was uncomplicated and was fit for discharge at the fourteenth post-op day. The patient has to-date completed successfully three-year follow up, where complete sac regression was identified in the most recent CTA.

DISCUSSION

After conducting a detailed search of all the patients treated in our department, for pathology regarding the thoracic aorta, during the last five years, we identified 3 patients that fulfilled the aforementioned criteria and who formed our target-group.

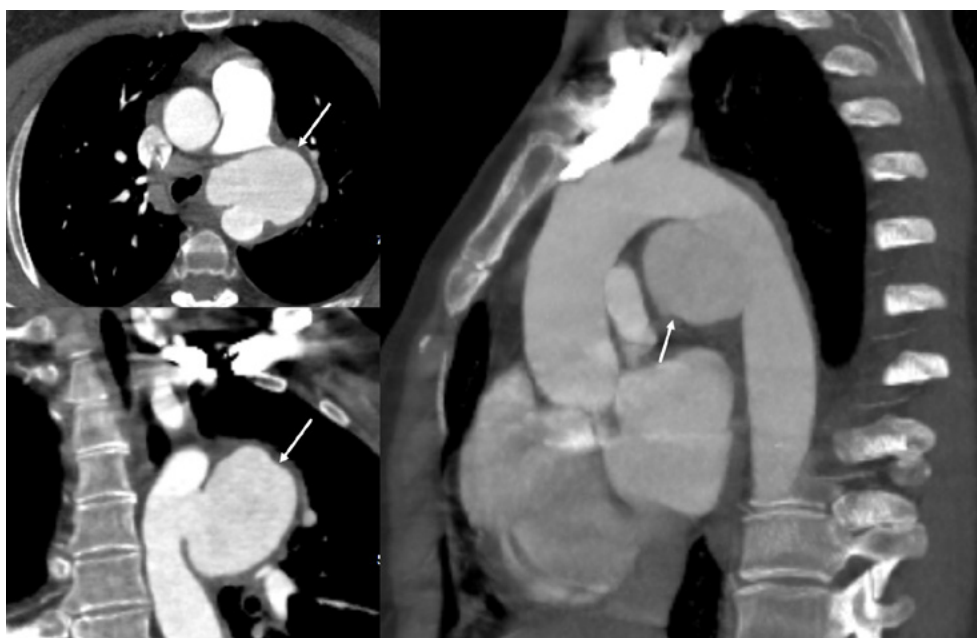


Figure 8: CT angiography of the 3rd patient with the white arrow indicating the pseudoaneurysm of the thoracic aorta.

The first patient [patient #1] suffered a concealed spontaneous rupture of the descending thoracic aorta, due to a ruptured penetrating aortic ulcer [rPAU], at the level of origin of the LSCA, causing the formation of a saccular pseudoaneurysm. PAUs most commonly present with symptoms imitating aortic dissection, a tearing-sensation chest pain, migrating to the back or the shoulder, with concomitant pleural effusions, identified in up to 30% of ruptures, whether more atypical symptoms include cardiac and pulse rhythm abnormality, signs of stroke, vascular insufficiency and end-organ infarction^{5,6}. There is scarce information in the literature regarding spontaneous, asymptomatic, ruptured PAU.

He had a medical history of arterial hypertension, dyslipidemia, coronary artery disease, COPD, AMI, inactive gastrointestinal cancer and he was an active, heavy smoker (110packs/year), making him high risk for developing PAU, as it's described in the literature^{7,8}. The first imaging modality that he underwent was a chest x-Ray, where a saccular radiopaque mass was depicted, which was later identified as a hematoma. Most commonly the chest radiograph findings of a PAU are unremarkable⁵, or pleural effusions or widened mediastinum may be present⁹. However, in this case, there was a chest x-Ray to compare, six months ago, where this exact finding was absent and that could have raised clinical suspicion, but was misidentified and not assessed clinically by the treating physicians at that time.

The second patient [patient #2] had a medical history of lymphoma that was in regression when he was referred to our department. Based on the literature, there is an association between aortic aneurysm and developing malignancy, and vice versa, especially pulmonary and hematology malignancies¹⁰. Also, there is suspicion that radiation therapy and chemotherapeutic drugs, such as antimetabolites, have been associated with altered aneurysm growth and thus need in-

creased aneurysm surveillance. However, increased risk of aortic rupture, in patients with active malignancy, with or without synchronous chemotherapy or radiation therapy, has not been documented¹⁰.

The patient complained about dyspnea, hoarseness of voice and cough. At first, the aforementioned symptoms were attributed to possible respiratory tract infection [RTI] and that's the reason he underwent a CT scan. However, after diagnosing the large pseudoaneurysm sac a possible explanation would be that the sac was applying pressure on the left bronchi / lung, causing dyspnea and cough, as well on the left recurrent laryngeal nerve [RLN] causing hoarseness. RTI was ruled out since there were no inflammatory markers and no imaging findings supporting this diagnosis.

There are no known contraindications for deploying a thoracic endograft in a patient with synchronous active malignancy, and as described by Danial P. et al, TEVAR for patients with T4 lung cancer with aortic evasion, before open resection of the tumor, had excellent results, with no late thoracic endograft-related complications observed, during the follow-up¹¹. As no active infection was officially diagnosed and the patient's anatomy was suitable for endovascular treatment, he was treated, as proposed in current the guidelines, with TEVAR³.

The third patient [patient #3] had a history of Adamantiades-Behcet's disease. ABD is an idiopathic, systemic inflammatory vasculitis, which presents clinically with recurrent oral and genital ulcers and ocular involvement. Vascular involvement has been described in up to 7-38% of the cases. Pathology of the aorta has been described in about 1.5-2.7% of the cases and most commonly involves pseudoaneurysm or saccular aneurysm of the abdominal aorta¹². Due to significantly increased risk of rupture in these patients, attributed to inflammatory processes and fibrotic reactions in the sur-

rounding tissues, treatment is indicated regardless of the aneurysm size. However, in this case, no aneurysmal dilation of the thoracic aorta was identified. The treatment modalities of an aortic aneurysm in a patient with ABD involves conservative management, consisting of corticosteroid and immunosuppressants, open surgical and endovascular repair¹². Conservative treatment was not a feasible solution for a ruptured thoracic aorta. Open surgical treatment has a high risk for complications and is technically challenging¹². 30-day mortality for open and endovascular repair is around 33% and 19%, respectively¹³. The patient was a good candidate for endovascular repair, as it was anatomic feasible and there was no active infection, therefore she was treated, as suggested by the guidelines, with TEVAR^{13,14}.

The initial clinical suspicion for all the patients was set when they underwent a non-contrast thorax CT, where a pseudoaneurysm of the thoracic aorta was identified. Especially for patient #1, non-contrast CT could depict hyperdensity in PAU, indicating intimal hematoma with an increased risk of rupture¹⁵. CTA is the preferred imaging modality for identifying a PAU, as it is seen as a contrast-filled outpouching or crater-like morphology, with irregular margins and extending beyond the expected boundaries of the aorta^{5,6,8}.

Then, they all were referred to our department, where they urgently underwent CTA, which is the gold standard for conclusive diagnosis and planning the treatment, in cases that thoracic aorta rupture is suspected¹⁶. Pseudoaneurysm of the descending thoracic aorta was diagnosed. In patient #1 a ruptured type B PAU, originating just proximal to the origin of the LSCA was depicted, whether in patients #2 and #3 the rupture point was identified distal to LSCA orifice. Aortic pseudoaneurysm is defined as a leakage of blood from the aorta, forming a contained hematoma, with persistent communication between the originating artery and the hematoma. It is actually a contained rupture of the aorta, and thus, urgent treatment is indicated. Aortic pseudoaneurysms are proposed to be treated with TEVAR, with excellent technical success rates around 100% and low rate of device-related complications, around 2.4%¹⁷.

All patients were treated successfully, based on the proposed guidelines, with TEVAR³. Regarding the revascularization of the LSCA, when the patient is being operated electively it is suggested to maintain the patency either with endovascular techniques or by open surgical revascularization, whether in emergency cases it is common practice to cover the orifice of the LSCA if needed, in order to achieve the minimum length of proximal landing zone required for the endograft deployment³. Exception to the above is when there is an internal mammary artery to coronary artery bypass or a clearly dominant left vertebral artery, where, even in an emergency setting, patency of LSCA has to be preserved¹³. In patient #1, due to severe >90% stenosis of the left common or internal carotid artery, and a clearly dominant left vertebral artery, we considered it important to preserve patency of the vertebral artery in order to avoid cerebral hypoperfusion. The severe comorbidities deemed the patient as extremely high risk for open surgery [carotid-subclavian artery bypass], chimney

grafting of the left subclavian artery was chosen instead, with the pitfall of not complete sealing of the rupture point and persistent endoleak. Despite the aforementioned risk, the two endografts were successfully deployed, without endoleak or extravasation identified in the completion angiography. Regarding patients #2 and #3, there weren't any special considerations to preserve patency of the LSCA, so this was covered.

Post-TEVAR type IA EL was identified in two of the three patients [patient #1 and #2]. Endoleak can be identified in up to 9-38% of patients undergoing TEVAR, with the most common type being Type I EL, around 8.4% of the patients¹⁸. As it is well established, all type IA EL, once identified, should be aggressively treated, since it represents high-pressure endoleak that can potentially cause sac expansion, rupture and death¹⁹. In-between the treatment modalities of type IA EL are proximal extension with aortic cuff placement, placement of a large-caliber balloon expandable stent in the proximal extent of the endograft in order to further promote sealing, deployment of a custom-made or off-the-shelf branched device, use of endo-anchor fixation system and transcatheter embolization of the leaking site²⁰. However, some ELs can resolve spontaneously. Moreover, EL can be classified as slow or fast, regarding the time needed to visualize the aneurysmal sac, during aortography, as proposed by De León Ayala et al¹⁹. These authors concluded that the majority of slow IA EL cases spontaneously occluded in the 6 months follow up, with no recurrence reported in the one-year follow up. In patient #1, despite the fact that no endoleak was present in the completion angiography, in post-operative CTA, a gutter type IA EL with filamentous perfusion of the pseudoaneurysm sac, without clinically significant growth of its diameter was depicted. Four days after the first post-op CTA, a second scan was performed, in order to screen for acute onset of dyspnea and to exclude pulmonary embolism [PE], where spontaneous occlusion of the endoleak was revealed with no further growth of the aneurysm sac. Therefore, no further treatment was indicated. Patient #2 developed a fast EL IA and therefore further treatment was indicated. In the literature, spontaneous occlusion of type I endoleaks is not usually anticipated²¹. Accordingly, the endoleak in patient #2 was treated successfully, endovascularly, without further impact on overall survival of the patient²¹.

Patient #1 developed acute-on-chronic kidney disease, as defined by the 2011 ESUR Contrast Media Safety Committee guidelines²², although all the proposed guidelines regarding hydration therapy as a preventive measure were followed^{23,24}. However, with proper management he restored renal function to his baseline creatinine [1.60mg/dl].

Patient #2, who underwent staged endovascular aortic arch debranching, with LSA coverage and TEVAR, developed post-op, acute severe heart failure [HF], with a LVEF of 15% and recurrent episodes of ARDS without any indication of an acute coronary syndrome. Among the common complications after complete endovascular debranching of the aortic arch are retrograde dissection, stroke, graft stenosis or occlusion, spinal cord ischemia and type IA EL²⁵. It is worth mentioning that in a previous heart U/S the LVEF was 40%. As stated in

the literature, after the deployment of TEVAR, an increase of arterial stiffness is noted, with multiple effects in the heart, central hemodynamics and heart systole^{26,27}. However, such acute and severe HF was an unexpected complication in the present case, and burdened heavily his post-op course.

CONCLUSION

Spontaneous, asymptomatic rupture of the thoracic aorta, is a rare vascular condition. Patients with different medical background may present with such lesions. On the absence of robust evidence regarding the clinical course of these cases, prompt endovascular treatment even if this means operating under suboptimal conditions without waiting until complete preoperative work-up is completed, may be appropriate in order to treat these patients and avoid serious adverse events.

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