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Hybrid Repair of Spontaneous Aortic Arch Rupture with Left Subclavian Artery Disinsertion

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Abstract

Spontaneous thoracic aortic rupture (STAR) is an infrequent yet critical manifestation of acute aortic syndromes, demanding immediate and precise intervention. This case study details the treatment of a 50-year-old male presenting with STAR and left subclavian artery disinsertion via a hybrid surgical approach. Despite lacking conventional risk factors such as aneurysm or connective tissue disorder, the patient's lifestyle, and health profile—marked by smoking, obesity, hypertension, diabetes, and dyslipidemia—underscored the necessity for acute clinical awareness. Diagnostic imaging revealed an aneurysmal dilatation and subsequent rupture of the aortic arch, leading to a tailored treatment comprising bilateral aorto-carotid bypass, endovascular stent-graft implantation, and coil embolization of the left subclavian artery. The intervention was successful, with the patient demonstrating no postoperative limb ischemia and favorable recovery, aside from a transient ischemic attack due to non-adherence to anticoagulation therapy. This case exemplifies the complexity and efficacy of hybrid repair strategies in treating STAR, advocating for increased vigilance and comprehensive assessment in at-risk populations. It also calls for further exploration into predictive diagnostics for better management of acute aortic conditions.

Keywords: Aortic arch spontaneous rupture; Left subclavian artery disinsertion; Hybrid repair; Bilateral aorto-carotid bypass; Coil-embolization

Abbreviations: AVK: Vitamin K Antagonist; BD: Bis Die; COPD: Chronic Obstructive Pulmonary Disease; CMN: Cystic Medial Necrosis; CTA: Computed Tomographic Angiography; ESC: European Society of Cardiology; INR: International Normalised Ratio; IV: Intravenous; OD: Omni Die; STAR: Spontaneous Thoracic Aortic Rupture

Introduction

Spontaneous thoracic aortic rupture is a pathology, part of the heterogenous group of acute aortic syndromes, that occurs in the absence of known aneurysm, dissection, infection, inflammation, trauma, or connective tissue disorder prior to the event. Despite being very rare, it is a life-threatening condition, that requires high clinical suspicion, immediate diagnosis, and prompt surgical and/or percutaneous therapeutic intervention for the salvage of the patients [1-3]. The disinsertion of the left subclavian artery is an even rarer complication that aggravates the prognosis and rises the complexity of the repair [4]. We present a case of a 50-year-old man with a spontaneous rupture of the aortic arch and left subclavian disinsertion managed by a hybrid approach consisting in bilateral aorto-carotid bypass, percutaneous stent-graft implantation of the aortic arch and closure of the left subclavian artery by coil embolization. This article also discusses the status quo of this condition in the literature, the different treatment modalities and raises awareness of the existence of possible preventive and diagnostic approaches for better assessing the patients at high risk.

Case Report

Written informed consent was obtained from the patient.

We present the case of a 50-year-old man, known heavy smoker and obese, with no other relevant medical history, besides type 2 diabetes mellitus and arterial hypertension, complaining of precordial pain with interscapular radiation, of variable intensity, doubled by an irritating cough with haemoptysis, 3 days prior

to presentation to the emergency department of the nearby county hospital. His clinical examination was unremarkable. The electrocardiogram objectified sinus tachycardia with no pathological changes and the cardiac echocardiogram was within normal parameters. On the other hand, the blood tests displayed negative levels of high sensitive I troponin, normal renal and hepatic function, and an inflammatory syndrome with neutrophilic leucocytosis (17,180/mm^3) and an elevated C-reactive protein (110mg%).

In regard of the suspicion of an acute aortic syndrome, a computed tomographic angiography (CTA) was performed that objectified an aneurysmal dilatation in the initial portion of the left subclavian artery, partially thrombosed, with a maximum transverse diameter of 69mm and cranio-caudal diameter of 58mm, with oedematous infiltration and a small amount of reactive fluid adjacent to the aneurysmal sac, likely superinfected.

In the context of the superinfection of the collection, the patient was started on intravenous antibiotic therapy consisting of Ceftriaxone IV 1g BD and Vancomycin IV 1g BD and was referred to the Surgical Clinic of the Timisoara Institute of Cardiovascular Diseases for the salvage procedure. Given his hemodynamically stable state, no emergency procedure was performed, and the patient was admitted to the Intensive Care Unit for continuous surveillance for the Heart Team to assemble and decide the best repair strategy. Meanwhile, the CTA was repeated with the additional evaluation of the vasculature of the left superior limb for the assessment of the feasibility of an interventional approach via the brachial artery. The second examination revealed the rupture of the aortic arch and the complete disinsertion of the left subclavian artery at 9mm distally to the insertion of the left common carotid artery, despite the patient maintaining hemodynamically stable parameters and a level of Hb in plateau at 12G/dL. After thorough analysis by the Heart Team, the patient was proposed a 3-step hybrid approach. The first operation was a bilateral aorto-carotid bypass with a 16/8/8mm bifurcated Dacron prosthesis, with insertion from the ascending aorta to both the common carotid arteries. The second step was the endovascular implantation of a stent-graft at the level of the aortic arch, extending all the way down to the descending aorta. The third and final step consisted in the closure of the left subclavian artery by coil embolization, via the left brachial artery. The only unwanted, event to have taken place was the closure of the left vertebral artery along with the left subclavian artery, due to the proximity of its insertion.

Post-operatively, the patient was transferred to the Intensive Care Unit and was started on triple antithrombotic therapy (Aspirin, Clopidogrel and Enoxaparin) for a total of 7 days. He developed post-procedural atrial fibrillation with rapid ventricular response and prompt chemical cardioversion by IV Amiodarone. The persistance of the inflammatory syndrome in the context of acute pneumonia, led to the continuation of the antibiotic treatement with Vancomycin and Ceftriaxone IV for a total of 14 days with favourable response. Postoperative onset of productive cough and persistance of ronchi at auscultation led to the introduction of beta 2 agonist and corticosteroids inhaling therapy in the context of a newly undiagnosed COPD.

The patient got discharged 9 days after the aortic repair, being evaluated by another CTA that revealed patent aortic arch stent-graft and aorto-carotid bypass graft, with no leakage, and a residual hematoma measuring approximately 8 by 7.2cm, located in the superior left mediastinum contiguous to the aortic arch, with metallic artifacts internally (left subclavian artery catheter wires). The left subclavian artery was not visualized from its origin with slight re-injection distally at the level of the axillary artery. The left vertebral artery presented a clear flow visible approximately 6.5cm above the aortic arch.

Despite the total closure of the left subclavian artery the patient presented no signs of acute limb ischemia.

At discharge the triple antithrombotic therapy was scaled down to Aspirin and Acenocoumarin with INR targeted between 2-3.

5 days after discharge the patient had a transient ischemic cerebral attack, having reported an INR of, due to treatment noncompliance. Consequently, the AVK was replaced by Apixaban 5mg BD.

The one-month follow-up showed a non-symptomatic patient, with normal motility of the left upper limb and no signs of ischemia.

Discussion

Spontaneous thoracic aortic rupture etiology, risk factors and prevention

To the best of our knowledge approximatively 50 case reports and small case series of spontaneous thoracic aortic rupture have been published, starting with 1961 [1-3,]. Yokoyama et al. describe atherosclerotic disease as the common factor in the 18 cases analyzed in their article [7]. Malyshev et al. [8] also find atherosclerosis as the leading cause of STAR and enlist atherosclerotic penetrated atherosclerotic ulcer, neurofibromatosis type I, and cystic medial necrosis (CMN) as most common reported clinical contexts [5,8]. Due to the enhanced dynamics of pregnancy amplifying stress on the aortic wall, and the fact that pregnancy induces aortic degeneration via estrogen-mediated elastin abnormalities in the aortic media during the peri- and postpartum periods, instances of STAR have been observed with greater frequency compared to the general population [5,6]. One other well-known and important risk factor is represented by hypertension [9]. Arterial stiffness has also been associated in recent years with the presence of diabetes mellitus

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[9]. Moreover, arterial stiffness has been demonstrated to increase the risk of acute aortic dissection and rupture, several imaging modalities (aortic strain rate, tissue doppler imaging) being proposed for patients at high risk to be better assessed [10].

Our patient was hypertensive and had type 2 diabetes mellitus, with no other known pathologies or risk factors associated with STAR.

Technical particularities

Our case has several particularities. Firstly, we notice the contrast between the severity of the defect and the hemodynamically stable status of the patient, mainly due the hemorrhage being contained by the large thrombus forms in the mediastinum – an aspect previously reported in literature [11]. Secondly, the complete disinsertion of the left subclavian artery at the proximity of the left vertebral artery ostium added tremendous complexity to the case.

In accord the 2014 ESC Guidelines for Aortic Disease in case endovascular aortic repair with stent-graft, a minimum of 2cm is required to be included in the stent-graft, proximally and distally to the defect [1]. In our case this implied occluding the left common carotid artery. As a solution, the bilateral aorto-carotid bypass was performed to enable a safe and complete lesion coverage. Regarding the closure of the left subclavian artery, two possible endovascular variants could have been possible: a Amplatzer vascular plug or a coil embolization technique. We preferred the later due to local availability [12]. The obvious risk of an iatrogenic acute limb ischemia was an assumed one, with a secondary aorto-subclavian bypass prepared in stand-by as an emergency back-up solution. Fortunately, no such intervention was needed, despite the closure of the left subclavian artery at the origin of the left vertebral artery, which was arising from the very proximity of the point of the defect. This undesired effect could have probably been avoided if an arterial plug was used instead, even if the apposition of such a device would have been no less of challenge due the same reason. The surgical direct closure of the left subclavian artery might have represented another mean of sparing the left vertebral artery to facilitate a proper irrigation of the upper left limb, as previously described literature [13]. In our case, whatsoever, this gesture would have implied on-pomp surgery for a safe dissection of the hematoma, that along with the surrounding thrombus was most probably the only way through which the bleeding was contained. Considering the high risk of an on-pomp surgery in our case, a separate percutaneous closure of the subclavian seemed the better option. Despite the odds, in the absence of an evident collateral source, the patient did not present left limb ischemia, with preserved mobility. As abovementioned the control CTA revealed a slight re-injection distally at the level of the axillary artery.

Classical versus hybrid repair of STAR

Kavanagh et al. [14] wrote in 2021 a paper aiming at assessing the classical versus the hybrid approach in managing STAR, including one ongoing clinical trial and two unpublished ones, along with 2 systematic reviews. In context of these insufficient data, the classical surgical approach remains the gold standard, with the hybrid repair showing potentially favorable outcomes [14]. Our case resolved in hybrid fashion with good results stands as testimony for future development on this domain.

Conclusion

Despite being extremely rare, the spontaneous thoracic aortic rupture must be taken into consideration among the 3 pillars of the acute aortic syndrome (aortic dissection, intramural hematoma and penetrating aortic ulcer), as left undiagnosed has tragic outcomes, whilst promptly found, more than optimistic prognosis. Our complex case of spontaneous aortic arch rupture with left subclavian artery disinsertion demonstrates the benefits of a hybrid repair approach, towards which the general opinion seems to be heading to. Lastly, predictive capabilities of concepts as the arterial stiffness should be further researched to facilitate a better assessment for the patient at high risk of developing acute aortic syndromes.

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