Endovascular treatment of perforating aortic ulcerations coexisting with thoracoabdominal aneurysm: case report and literature review

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Abstract
The aim of the study was to introduce endovascular repair of excessive PAUs and thoracoabdominal aneurysm by staged Zenith alpha and T-branch application in a patient with PAU, thoracoabdominal aneurysm and subclavian steal syndrome, additionally managed by carotid–subclavian bypass.

We present a case of a 71-year-old woman with four excessive life-threatening thoracic penetrating aortic ulcerations (PAUs, maximum diameter 57 mm), accompanied with thoraco-abdominal aortic aneurysm (TAAA) — Crawford IV (max diameter 65 mm) and severe asymptomatic left subclavian artery (LSA) stenosis due to its compression by proximal PAU. In response to this serious life-threatening condition, we performed staged endovascular repair, starting with thoracic stent-graft implantation with coverage of LSA ostium, using thoracic stent-graft (The Cook Zenith Alpha). The patient unexpectedly presented postoperative symptomatic subclavian steal syndrome and left-hand ischaemia. On the fourth postoperative day, a carotid-subclavian bypass was performed, and the patient was discharged in a good general condition. Three months later the patient underwent endovascular thoraco-abdominal aneurysm repair using a multibranched off-the-shelf stent-graft (Zenith T-branch), a week later angio-CT was performed to confirm treatment efficacy, and the patient was discharged in a good general condition.

Endovascular treatment of PAUs coexisting with TAAA is possible and feasible even with total aorta exclusion.

Key words: endovascular treatment, perforating aortic ulcerations, thoracoabdominal aneurysm, carotid–subclavian bypass, thoracic endovascular aortic repair, multibranched off-the-shelf stent-graft

Introduction
Penetrating aortic ulcers (PAUs) of the aorta were first described in 1934 by Shennan [1]. PAU is a rare pathological condition of the aortic wall, often asymptomatic, but requiring urgent surgical intervention in most patients; otherwise, the ulcers may rupture, leading to haemorrhage and death. PAUs occur mostly in elderly patients with massive atherosclerosis [2]. The evolution in vascular imaging and endovascular repair has provided a major improvement in aortic disease management and has reduced the morbidity and mortality rates compared to open surgery, especially in elderly multimorbid patients. Endovascular treatment of PAU has become the method of choice since the introduction of this method in 1998 by Murgo et al. [3]. Since the introduction of endovascular treatment in 1996 for juxtarenal aneurysm repair by Park et al. [4], widespread use of thoracoabdominal endovascular aneurysm repair via fenestrated and branched stent grafts has been observed [5, 6].

The aim of this study is to introduce endovascular repair of excessive PAUs and thoracoabdominal aneurysm by staged Zenith alpha and T-branch application in a patient with PAU, thoracoabdominal aneurysm and
subclavian steal syndrome, additionally managed by carotid–subclavian bypass.

**Case report**

A 71-year-old woman was admitted to our department in November 2016 through the emergency ward. She was haemodynamically stable with four excessive life-threatening thoracic penetrating aortic ulcers (PAUs, maximum diameter 57 mm) (Fig. 1), accompanied with thoraco-abdominal aortic aneurysm (TAAA) — Crawford IV (max diameter 65 mm), as well as severe asymptomatic left subclavian artery (LSA) stenosis due to its compression by proximal PAU. In anamnesis, the patient had poorly controlled hypertension, stable ischaemic heart disease, tricuspid valve regurgitation grade II, permanent atrial fibrillation (CHAD-VASc5, HASBLED 3), chronic immunologic thrombocytopaenia, chronic obstructive pulmonary disease (COPD), complicated type 2 diabetes mellitus, atonic neurogenic bladder and chronic renal disease. She had a history of panhysterectomy and cholecystectomy.

In response to this serious life-threatening condition, we performed staged endovascular repair, starting with thoracic stent-graft implantation to cease PAUs’ blood flow to avoid rupture with coverage of LSA ostium. Moreover, due to severe LSA stenosis, we decided not to perform revascularisation. TAAA repair was left for the next surgical stage due to high risk of spinal cord ischaemia (SCI), when a long segment of the aorta was covered with a stent-graft in one operation. Endovascular thoracic PAU exclusion was performed under general anaesthesia, under digital subtraction angiography (DSA) control in a hybrid operating room. The surgical approach was via the right femoral artery, by placement and deployment of a thoracic stent-graft (The Cook Zenith Alpha) starting just below the origin of the left common carotid artery, and aortic coverage extended from the LSA to the distal descending aorta 2 cm above the coeliac trunk, including two stent-graft components (209 and 142 mm), both with a diameter of 34 mm. The total length of thoracic aorta coverage was 27 cm. Postoperative angio CT demonstrated patent stent-grafts; all the PAUs were successfully sealed, with no evidence of endoleak (Fig. 2). The proximal part of LSA was occluded at a length of 50 mm, and the distal was contrasted through the left vertebral artery. However, the patient unexpectedly showed postoperative symptomatic subclavian steal syndrome and left-hand ischaemia, despite the above-mentioned preoperative LSA near total occlusion. On the fourth postoperative day, a carotid-subclavian bypass was performed under general anaesthesia using a 6-mm polytetrafluoroethylene (PTFE) graft, and the patient was discharged in a good general condition.

The patient had not decided for the second-stage operation until three months later, when she agreed and was admitted. A control CT angiography revealed patent stent-grafts without endoleaks and excluded all PAUs and occluded LSA in the proximal part. Carotid-subclavian bypass was patent. The maximum transverse dimensions of the thoraco-abdominal aneurysm were 58 x 68 mm at the infrarenal level (Fig. 3). The patient underwent endovascular thoraco-abdominal aneurysm repair under general anaesthesia. Intraoperatively, heparin was administered via intravenous infusion to maintain activated clotting time (ACT) of 250 to 300 seconds and was con-
trolled every 30 minutes. The operation was performed under DSA control in a hybrid operating room. The surgical approach was via the right brachial artery and the left femoral artery. The device was a multibranched off-the-shelf stent-graft (Zenith T-branch), 34 mm in diameter at the top and 18 mm at the bottom, with a length of 202 mm, with four branches dedicated for each visceral artery in a specific distance and clock rotation (Fig. 4) [7]. After insertion and deployment of the t-branch, bifurcated stent-graft and ipsilateral extension

in proper position, all sheaths were removed with closure of the left femoral artery for early pelvic and lower limb reperfusion. The pull-through wire was left to secure the position of the right axillary approach, through which the branches were connected with the visceral vessels with self-expandable covered bridging stents (Fluency Bard Peripheral Vascular, Tempe, Arizona, USA), reinforced with self-expandable bare stents (Zilver Cook). Furthermore, the right common iliac artery extension was inserted and deployed via an axillary approach using Fluency reinforced with Zilver stents due to right groin scar formation after a previous intervention. Final angiography revealed proper contrast perfusion of stent-grafts and branches. The operative time was 140 minutes, the radiative time was 30.8 min and the dose of radiation was 11.58 mGy/m². The patient underwent our postoperative management algorithm, with 48-hours monitoring in the intensive care unit. Mean arterial blood pressure was maintained at >90 mm Hg, and administration of antihypertensive drugs was suspended to enhance spinal cord perfusion. At this period, a heparin infusion pump was administrated, with target APTT 2–2.5 × normal ratio. We did not drain cerebrospinal fluid in this patient. Then, low-molecular-weight heparin (LMWH, until discharge) was prescribed in prophylactic doses in association with acetylsalicylic acid, 75 mg and clopidogrel 75 mg per day. Before the patient discharge, CT was performed to confirm the treatment efficacy (Fig. 5). The postoperative period was uneventful, and the patient was discharged a week later in a good general condition. The control angio CT scan was performed three months after the second-stage operation and showed patent stent-grafts with no endoleaks. Further follow-ups of the patient,
including CT investigation, are scheduled twelve months after the operation and annually thereafter.

**Discussion**

PAUs have been categorised as an acute aortic syndrome (AASs) and represent only 2.3%–7.6% of patients diagnosed with AASs, which also involve intramural haematoma, classic aortic dissection and aortic rupture [8–12].

PAUs are pathologically defined as an atherosclerotic plaque ulceration invading the internal elastic lamina with lesion penetrating into the lamina media of the aorta [2, 11, 13, 14]. A rupture of the degenerated internal lamina occurs that it exposes the lamina media directly to the arterial blood pressure, which leads to haematoma formation in the lamina media, resulting in external bulging of the arterial wall. In more than 90% of cases, the pathology involves the descending part of the thoracic aorta. However, it may involve the ascending and aortic arch or present in multiple parts of the aorta [15, 16]. According to the location of PAU, the disease can manifest as chest, abdominal or back pain [17].

The endovascular treatment of PAUs was introduced and expanded in the last two decades by the progression of techniques in vascular imaging and endovascular surgery. This gives a greater opportunity for early diagnosis and treatment.

Despite this progression of vascular imaging techniques, an early diagnosis of PAU is still a challenge in some cases due to its rarity, which may lead to poor prognosis as a result of management delay.

The threat of rupture increases in patients with lesions beyond 20 × 10 mm in width and depth, respectively. PAUs rupture in approximately 40% of cases [10]. In addition, other life-threatening complications, such as aorto-oesophageal or aortobronchial fistulas, may develop and induce death [18, 19].

Eggebrecht et al. reported a multicentre analysis of 19 published studies from 1994 to 2008 concerning 209 PAU cases, which proved the efficiency and safety of endovascular repair with 98% technical success and 96% complete PAU sealing. Postoperative mortality was 7%, and mortality in follow-up period (mean of 14 months) was 2%. Endovascular reinterventions were required in 5% of cases [10].

In our case, the endovascular treatment of PAUs had 100% technical success even though short neck of the first PAU, the stent-graft covered the LSA, which already was nearly totally occluded before. Postoperatively, the patient unexpectedly presented symptomatic subclavian steal syndrome, which was managed by carotid-subclavian bypass.

The first brachiocephalic trunk and subclavian bypass by transthoracic approach was documented in 1958 by De Bakey et al. [20]; in 1967, the first carotid-subclavian bypass graft through extrathoracic approach was performed by Diethrich et al. [21].

Currently, the methods of choice for subclavian artery stenosis or occlusion treatment are extrathoracic procedures such as carotid-subclavian bypass, rarely, subclavian-carotid transposition and axillary-to-axillary artery bypass [22].

AbuRahma et al. [22] reported a study with 20-year experience, which proved the safety and efficiency of PTFE grafts using for carotid-subclavian bypass as a treatment for subclavian artery stenosis or occlusion with superb long-term outcomes. Moreover, many studies [23–25] have been documented showing that the PTFE gives better long-term patency outcomes, up to 95%, compared with Dacron and vein grafts.

Our patient had type IV TAAA, which is not associated with subclavian artery occlusion. However, we had to treat TAAA as functional type I due to the place of the proximal PAUs, which compressed the LSA.

Moreover, according to the literature, the treatment of types I and II TAAAs, proximal aortic sealing may affect the left subclavian artery orifice. In such elective cases, staged operation is recommended to maintain arm and vertebral blood perfusion by debranching of the LSA [5, 26]. We considered our patient to have an aortic emergency and therefore did not perform a bypass preoperatively.

The elaboration of endovascular strategies in patients with TAAA is evident, owing to the fact that open TAAA repair is associated with high rates of morbidity (SCI) and mortality, even in highly qualified centres [27–29].

Counter to the open approach, the endovascular technique is associated with lower morbidity and mor-
tality rates, as it prevents prolonged aortic exposure and visceral ischaemia as a consequence to aortic clamping [5, 30–33]. However, the requirement for secondary reinterventions is greater in the endovascular approach than in the open surgery [34].

The first endovascular repair of a TAAA was reported by Chuter et al. [1] in 2001 [35], and they were the first who reported the safety and efficiency of multibranched endograft used for endovascular aneurysm repair (mbEVAR) with satisfactory outcomes and acceptable perioperative mortality rate [30].

The custom-made mbEVAR was a revolution in the treatment of multimorbid patients with high risk of death due to open repair, but the important clinical problem is the necessity to wait 8–10 weeks for production and delivery, which is unacceptable in some symptomatic patients with large TAAAs [36–40]. As a solution, Cook Medical (Bloomington, IN, USA) has produced a new off-the-shelf mbEVAR (T-branch) [40], made with standard visceral cuffs positioned to be suitable for almost half of TAAA patients, that has been proven in previous studies. Moreover, in case of implementing an additional adjuvant intervention, the convenience of T-branch could be higher, in up to 60% of patients [37, 40–42].

Our patient rejected the second-stage operation (TAAA treatment) until three months later; therefore, we did not order Custom-made device (CMD) for the operation. Fortunately, she had a vascular anatomy amenable to T-branch implantation, so when she had agreed, we did not have to wait for CMD manufacture.

The first documented clinical assessment of technical success and perioperative findings by Bosiers et al. has proven the safety and efficacy of the T-branch device [43].

Bisdas et al. showed in the first technical and clinical comparability among the CMD and the new off-the-shelf mbEVAR T-branch for TAAAs endovascular repair, that using T-branch is effective and safe with superb technical success and outcomes comparable to the CMD endografts, with possibility of implantation without delay in symptomatic patients [44].

Since then, the T-branch device has been the method of choice in TAAA repair [37, 42]. In contrast, the custom-made mbEVARs are applied only in cases in which the T-branch endograft is not applicable [44].

The variations in mortality rates are determined by different factors in many centres, as using a specific algorithm to decrease the incidence of SCI, including neuromonitoring, early restoring of limb blood supply and staging procedures. Other important determinants are learning curve and clinical experience [45–49].

Regarding the high risk of long-time lower limb ischaemia during TAAA endovascular repair, we routinely reperfuse the lower limb after T-branch deployment and distal extensions by removing the delivery systems and closing the femoral approach to restore blood perfusion to the lower limb and the pelvis. This may help to avoid SCI occurrence by reducing steal syndrome from internal iliac artery [and its branches supplying the spinal cord] to the lower limbs. Then the bridging stents are inserted via axillary access.

The documented occurrence of SCI symptoms in case of thoracic endovascular aortic repair and endovascular TAAA repair are up to 10% and 30%, respectively [50].

Additional extensive covering of thoracic aorta during TAAA endovascular repair may yield a higher risk of SCI [37].

According to Bisdas et al. [51], multivariate analysis in a cohort study showed, that the incidence of SCI after endovascular TAAA repair was up to 16%; half of the patients presented with paraplegia, and the other half presented with paraparesis. In most cases, neurological symptoms were reversible; only in 2% of cases the paraplegia was irreversible. Also, cerebrospinal fluid drainage (CSFD) is recommended if patients develop SCI symptoms, since all the patients who had CSFD immediately after the appearance of symptoms demonstrated neurological improvement.

In our department, preoperative CSFD is not routinely performed unless the patients report SCI symptoms to avoid reported neurological complications reported previously [44, 51]. Furthermore, the coverage percentage of aorta, especially the thoracic part, was documented as the most important risk factor for SCI [51].

Some authors [52] observed a correlation between SCI symptoms and cardiovascular instability following endovascular TAAA repair, which is why some authors recognise a delay in symptoms of SCI development in 60% of cases, according to Bisdas [51], and 87% reported by Greenberg [31].

We apply a CSFD as a part of our postsurgical treatment algorithm only in cases complicated by spinal cord ischaemia owing to the correlation of CSFD with many neurological complications [43, 53] and the delay of spinal cord ischaemia inception following endovascular repair [31, 54].

**Conclusion**

Endovascular treatment of PAUs coexisting with TAAA is possible and feasible even with total aorta exclusion. Some adjuncts, such as early pelvis reperfusion and connection of contralateral leg from axillary access, can simplify this complex surgery and make it a success.
Conflict of interest

None.

References:

28. Coselli JS, Bozinovski J, LeMaire SA. Open surgical repair of 2286 thoracoabdominal aortic aneurysms. Ann Thorac...