CASE REPORT

Acute Testicular Ischaemia Following Endovascular Aneurysm Repair on the Opposite Side to Intentional Internal Iliac Artery Occlusion

Nikolaos Kontopodis ^a, Alexandros Kafetzakis ^a, Androniki Kozana ^b, Konstantinos Tzirakis ^c, Ioannis Peteinarakis ^b, Christos V. Ioannou ^{a,*}

Introduction: Testicular ischaemia is a potential complication after endovascular aneurysm repair (EVAR), which has only rarely been reported in the literature. This is the report of a patient who presented with acute testicular ischaemia in the immediate post-EVAR period.

Report: A 65 year old patient underwent EVAR for an aortic and bilateral iliac aneurysms. During the procedure, the right internal iliac artery was intentionally occluded to facilitate treatment of the common iliac aneurysm; however, the left internal iliac artery was preserved. The procedure was uneventful. On the second post-operative day the patient gradually developed symptoms of acute left testicular ischaemia. Clinical and ultrasonographic findings constituted the bases of diagnosis and the patient received conservative treatment with gradual improvement. To the authors' knowledge, this is the ninth case of testicular ischaemia after endovascular aneurysm repair reported in the literature.

Conclusion: Testicular ischaemia, although rare, is a possible complication post-EVAR. Acute and chronic testicular damage found in association with an abdominal aortic aneurysm or its treatment has not been well studied in the literature and therefore may be under reported.

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INTRODUCTION

Pelvic ischaemia after endovascular aneurysm repair (EVAR) has been mostly associated with uni- or bilateral intentional occlusion of the internal iliac artery and usually presents as buttock claudication, sexual dysfunction, or intestinal ischaemia. Pelvic embolic complications after EVAR have been reported at a rate of 1%. Testicular ischaemia post-EVAR is a very rare complication, which has scarcely been reported in the literature. Specifically, to date, only eight cases have been reported with variable presentation, management, and outcome. This report describes a patient who developed acute testicular ischaemia after EVAR.

CASE REPORT

A 67 year old male patient presented with an infrarenal aorto-iliac aneurysm with a maximum aortic diameter of 42

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mm, a left common iliac diameter of 36 mm, and right common iliac diameter of 35mm (Fig. 1). His medical history was remarkable for coronary artery disease (conservative management), smoking, arterial hypertension, hyperlipidaemia, previous abdominal surgery (small bowel resection caused by bleeding of a benign vascular malformation), and resection of vocal cord cancer. The patient was scheduled for EVAR, taking into account that occlusion of the right internal iliac artery would be required. The left internal iliac artery could be preserved because sealing was feasible in the distal left common iliac artery (20 mm diameter for a sealing length of 15 mm). An iliac branched device was not used in this case because the left internal iliac artery could be preserved. Preservation of one internal iliac artery is in accordance with current guidelines and provided confidence that pelvic blood supply would not be compromised after occluding the right internal iliac artery. 10

Initially, through a contralateral approach, the right internal iliac artery was occluded with an 11 mm EOS vascular plug (Art Ventive Medical Group, Carlsbad, CA, USA). Next, a percutaneous EVAR procedure was performed, under local anaesthesia and conscious sedation, with the Ovation iX endograft (Endologix, Irvine, CA, USA) (main body $29\!\times\!80$ mm, right iliac limb $28\!\times\!100$ mm with a $14\!\times\!160$ mm extension to the right external iliac and left iliac limb

^a Vascular Surgery Unit, Department of Cardiac, Thoracic and Vascular Surgery, University of Crete Medical School, Heraklion, Crete, Greece

^b Department of Medical Imaging, University of Crete Medical School, Heraklion, Crete, Greece

^c Biomechanics Laboratory, Department of Mechanical Engineering, Technological Educational Institute of Crete, Estavromenos, Heraklion, Crete, Greece

^{*} Corresponding author. Department of Cardiac, Thoracic and Vascular Surgery, University Hospital of Heraklion, University of Crete Medical School, PO Box 1352, 711 10, Heraklion, Greece.

E-mail addresses: ioannou@med.uoc.gr; xioannou@uoc.gr (Christos V. loannou).

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Figure 1. Axial CT slices of the abdominal aortic (A), left (B), and right common iliac (C) aneurysms pre-operatively, where maximum diameters are displayed.

 22×100 mm landing just prior to the left iliac bifurcation) (Fig. 2). The procedure was uneventful.

On the second post-operative day, the patient complained of left flank pain radiating to the left testicle. His abdomen was soft, with normal bowel sounds and no signs of tenderness or guarding. No signs of haematoma were apparent at either access site. He was haemodynamically stable, while blood and urine laboratory examinations were unremarkable. Urgent CT angiography showed a patent endograft with good positioning and successful exclusion of the aneurysm with no signs of an endoleak (Fig. 3). The patient reported gradual localisation of the pain to the left scrotum, which became painful and tender on the third post-operative day. Initial ultrasonography using a conventional grey scale and colour Doppler, revealed an enlarged, inhomogeneous left testis with the absence of flow on colour Doppler examination and architecture distortion, representing infarct/necrosis (Fig. 4).

Urological consultation suggested that the clinical presentation was compatible with the abovementioned



Figure 2. Completion intra-operative angiography showing good positioning of the endograft, occluded right internal iliac artery, and patent left internal iliac artery, with no evidence of an endoleak.

diagnosis but no urgent surgical exploration was needed at that time and a strategy of watchful waiting was applied. Opioid analgesics were administered and the pain subsequently improved. The patient was discharged from the hospital on the eighth post-operative day. At the two month follow up visit, the patient reported symptom resolution. Repeat ultrasound confirmed the absence of flow in the affected testicle, as shown with the power Doppler image (Fig. 5).

Retrospective examination of the pre-operative CT scan revealed the presence of a patent left testicular artery originating from the aorta just distal to the level of the renal arteries whereas a right testicular artery was not observed (Fig. 6).

DISCUSSION

Testicular ischaemia or infarction after EVAR is a rare complication, which has scarcely been reported in the literature. Specifically, only eight previously published cases were identified.²⁻⁹ The characteristics of previous reports are summarised in Table 1. It is noteworthy that the reported cases had remarkably variable presentations, treatments, and outcomes. For example, most patients (n = 7)developed symptoms immediately post-operatively, while one had delayed presentation, six weeks post-EVAR.² Additionally, among the reported cases, four patients underwent orchidectomy^{2,4,7,8} while another four were treated conservatively, 3,5,6,9 which resulted in gradual symptom improvement. Ultrasonography has an established role in investigating acute or chronic scrotal pathology and remains the imaging modality of choice for assessing testicular vascularity. In the present case, infarction was hypothesised because of a hypoechoic left testis which presented architectural distortion and absence of blood flow.

Another issue that varies among reported cases is the contribution of iliac artery embolisation in development of testicular ischaemia. In three cases the internal iliac artery was occluded during the procedure,^{2–4} whereas in the remaining cases it was not. Taking into account that the main blood supply of the testis is the testicular artery, which originates from the infrarenal aorta, while important collaterals, mainly the cremasteric artery and the vas deferens artery, arise from the external and the internal iliac artery,

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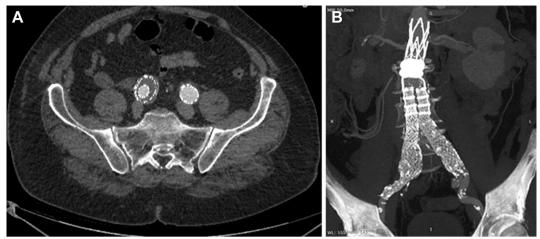


Figure 3. Axial CT image (A) and multiplanar reconstruction (B) post-operatively. The distal sealing zone at the level of the left iliac bifurcation can be seen.

respectively, two possible mechanisms have been proposed for the post-EVAR testicular ischaemia. These are inflow occlusion and distal embolisation. In the absence of collaterals symptoms of testicular ischaemia may arise after the

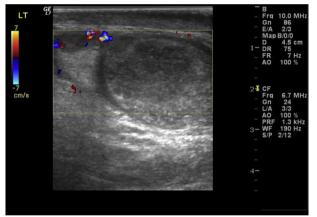


Figure 4. Ultrasonography using colour Doppler. The left testis appeared inhomogeneous and oedematous. The absence of blood flow is noted representing infarct/necrosis.

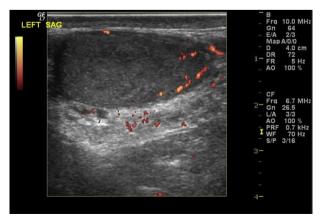


Figure 5. Ultrasonography using power Doppler performed at the two month follow up visit. Repeat ultrasound confirmed the absence of blood flow consistent with the original infarction.

testicular artery has been occluded during EVAR. In the present case, the ischaemia occurred on the side of the patent internal iliac artery, and the left testicular artery remained patent on post-operative CT angiography. Therefore, the presence of collateralisation may suggest an embolic mechanism but the contribution of gonadal occlusion from the endograft cannot be excluded. Despite the fact that a hypothesis that embolic material travels from the aorta through the narrow testicular artery along its entire length to cause focal parenchymal ischaemia may seem unlikely, at least two prior reports which have performed a histopathological examination of surgical specimens indicated the presence of cholesterol emboli inside the testicular interstitial vessels. 4,8 Furthermore, McKenna et al. have reported thrombosis of the gonadal artery in their case of

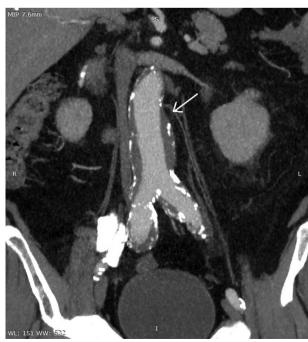


Figure 6. Pre-operative CT angiography displaying a patent testicular artery on the left side (white arrow). The size of the testicular artery is 3 mm.

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Table 1. Summary of previous reports of testicular ischaemia after EVAR.

Author	Journal and year of publication	Time of presentation (Post-op)	Clinical presentation		Intentional IIA occlusion	Possible cause of testicular ischaemia
McKenna ²	J Vasc Surg 2009	6 weeks	Testicular pain	Orchidectomy	Yes	Thrombosis of testicular artery
Hall ³	J Vasc Interv Radiol 2010	6 days	Testicular pain	Conservative	Yes	Occlusion of the testicular artery and compromise of collaterals after IIA coverage
Milburn ⁴	Eur J Vasc Endovasc Extra 2010	10 days	Asymptomatic mass-malignancy	Orchidectomy	Yes	Embolisation following dislodgement of atherosclerotic debris during graft deployment
Finnerty ⁵	Case Rep Emerg Med 2014	2 days	Testicular pain	Conservative	Not reported	Not discussed
Vervoort ⁶	Cardiovasc Intervent Radiol 2015	1 day	Testicular pain	Conservative	No	Embolism in the testicular artery
Thomas ⁷	Urol Cas Rep 2017	2 days	Testicular pain	Orchidectomy	No	Microembolisation into the hypogastric artery causing transient colonic ischaemia and left testicular ischaemia
Pathmarajah ⁷	EJVES Short Rep 2017	1 day	Testicular pain	Orchidectomy	No	Parenchymal infarction secondary to cholesterol emboli
Zebari ⁸	Urology 2018	1 day	Testicular pain	Orchidectomy	No	Covering the gonadal arteries, coupled with an absence or delayed development of adequate collateral iliac blood flow. A thrombo-embolic event, is also a possible cause
Kontopodis	Current study	2 days	Testicular pain	Conservative	No	Microembolisation/arterial collateral disturbance

testicular ischaemia.² Therefore the relative contribution of those two possible mechanisms cannot be evaluated at the moment. Notably, the right testis was universally hypoechoic in this case and presented with a reduced Doppler signal, which takes into account the absence of any symptoms, and was probably caused by long-standing, chronic ischaemia. As aneurysmal disease may be accompanied by the accumulation of intraluminal thrombus, this may hamper patency of inflow arteries and compromise collateralisation of testicular arterial perfusion, regardless of treatment. Remarkably, no cases of acute testicular ischaemia after open surgical AAA treatment have been reported. This observation may render open surgical repair preferable in selected patients, especially in young fit subjects. In the present case, the patient's comorbidities, as well as his previous abdominal surgery (hostile abdomen) accounted for the decision to manage this case endovascularly.

CONCLUSION

Changes in testicular arterial perfusion caused by aneurysmal disease and EVAR are not well understood. Distal embolisation or inflow compromise are two possible mechanisms that may result in testicular infarction after EVAR and increased awareness is needed to understand the

magnitude of the problem and identify patients at risk of such a complication. This should be added to the list of possible complications after EVAR during patient consenting.

CONFLICTS OF INTEREST

None.

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