CASE REPORT

Mycotic iliac artery aneurysm with appendicitis in a pediatric patient

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SUMMARY

Iliac aneurysms are rare in children, especially mycotic aneurysms. Re-vascularization is challenging given the infected field and concern on patency due to their growth potential and a longer life-span. We report a complex case of a mycotic iliac aneurysm in a child. A 12-years-old boy with a previous history of infective endocarditis was referred to us for a right common iliac mycotic aneurysm after presenting with pain. A balloon-expandable stent-graft was deployed across the aneurysm during the acute presentation. He improved post-operatively, but developed abdominal pain four weeks later. A repeat computed tomography (CT) imaging showed a new inflammation of the appendix which was adhered to the calcified wall of the aneurysm and an endoleak from the internal iliac artery. A laparotomy was performed and the right internal iliac artery ligated along with an appendicectomy and omental pedicle. Postoperatively the patient was well and discharged home. Six-month surveillance revealed a healthy child and imaging showed a patent stent-graft and no residual collection.

INTRODUCTION

Arterial aneurysms are rare in children, especially mycotic iliac aneurysms (MIA).[1] Conventional open repair of MIA raises the challenge of a suitable conduit and morbidity in an ill child. We report our management of a 12-year-old boy who developed a MIA due to infective endocarditis which was subsequently complicated with acute appendicitis. Herein we describe our approach of the management.

CASE REPORT

A 12-year-old boy presented with an acute lower abdominal pain of a week duration associated with fever and diarrhoea. He had previously been treated for infective endocarditis when he was 9 years old with a long course of penicillin after blood cultures grew Streptococcus pneumoniae. At that point he had no abdominal symptoms and there was no suspicion of an iliac aneurysm. Surveillance echocardiograms showed resolved vegetation and no valvular heart disease. On presentation at the Department of Surgery, Hospital Kuala Lumpur a tender mass was palpable on his right iliac fossa. He was febrile and tachycardic, though no significant leucocytosis and C-reactive protein (CRP) of 12mg/L. Computed tomography (CT) scan revealed a saccular aneurysm of the distal right common iliac artery (CIA), and a normal appendix (Figure 1).

An endovascular approach was chosen for the treatment as open surgery would be associated with a higher morbidity. Furthermore, there was no suitable native conduit and we felt a synthetic graft was better suited once the child had completed adolescence. While under general anaesthesia the right femoral artery was exposed via a groin incision. Percutaneous access was not attempted as the profile of the stent-graft was 9Fr and we did not have access to vascular closure devises. After catheter-directed retrograde cannulation of the CIA, angiography revealed a stenotic proximal external iliac artery (EIA) due to the mass effect of the aneurysm. After dilatation with an 8mm PTA dilatation catheter, a 7mm x 58mm Lifestream BE stent-graft (Becton Dickinson, UK) was deployed across the mycotic aneurysm, proximally landing at the CIA and distally at the EIA. Post-deployment angiography revealed no filling of the sac, and the right internal iliac artery opacified from retrograde flow (Figure 2). Postoperatively the condition of the patient improved and had reduced CRP from 74.6mg/L to 9.4mg/L. Surveillance ultrasonography revealed an endoleak from the right IIA. As it was small, we planned for conservative treatment, and empirical ertapenem was given during the subsequent recovery as blood cultures did not yield any organisms.

Unfortunately his sepsis and abdominal pain recurred four weeks later while he was an inpatient. A repeat CT scan revealed gas pockets in the previous aneurysm sac, and the appendix was grossly inflamed and was adhered to the sac, which was not present on the previous imaging. Open drainage was performed as there was no accessible percutaneous window. Intraoperatively the sac was filled with pus and a perforated appendix densely adhered to the calcified wall. An appendicectomy was performed, the right internal iliac artery was ligated and an omental pedicle anchored into the sac after drainage and deroofing, avoiding exposure of the endograft. Intraoperative cultures showed mixed growth.

The patient made a full recovery after completing a course of intravenous metronidazole and ampicillin & salbactam over two months. On discharge he was prescribed oral metronidazole and ampicillin & salbactam for another two months. Erythrocyte sedimentation rate (ESR) and CRP continued to reduce and also the resolution of leucocytosis. All subsequent cultures were negative. He is on long term aspirin 100mg daily and oral ampicillin/sulbactam and remains under follow up. Surveillance imaging at 6 months showed no endoleak and good antegrade flow.

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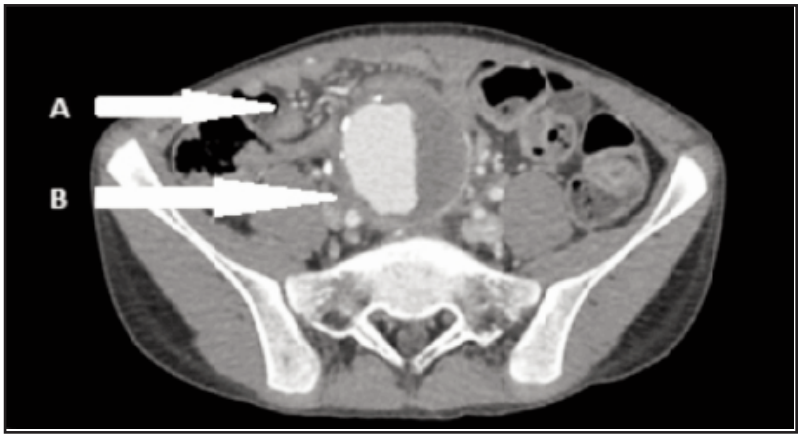

Fig. 1: CT abdomen, A- Normal appendix, B – Right CIA aneurysm.

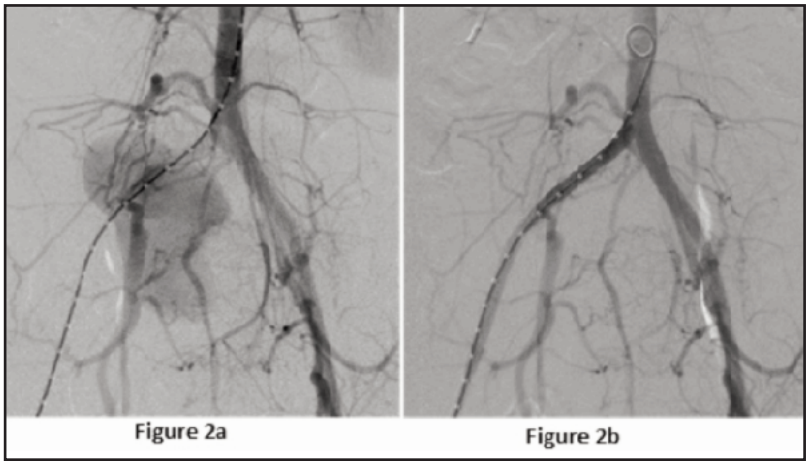

Fig. 2: DSA of right iliac artery before (2a) and after (2b) endograft deployment.

DISCUSSION

The etiology of the aneurysm in our patient can be attributed to his previous bout of infective endocarditis in his childhood, as the calcified wall suggests a chronic process. In the classification proposed by Sarkar et al, this type of aneurysm is due to arterial infection from a systemic process and accounts for a third of reported cases.[1] The appendicitis in our patient developed while during recovery from the first surgery was not in fact the cause of the aneurysm, as initial imaging showed a normal appendix. We postulate that the appendicitis developed due to lymphoid hypertrophy secondary to inflammation from the mycotic aneurysm and foreign-body response after the stent-graft deployment.

In the literature, the surgical approach to treat mycotic aneurysms in children has generally been via an open technique. Iliac mycotic aneurysms due to infective endocarditis and tuberculosis have been described in an 8-year-old and a 12-year-old respectively, both having successful bypasses with synthetic grafts.[2,3] Nevertheless, patency is an issue in paediatric vascular bypasses, wherein late primary graft failures occur in up to a third of cases. Due to somatic growth, the dimensions of the conduit and technique of anastomosis play an important role. Both the diameter and length need to be considered when choosing and fashioning a graft. Autologous graft in this patient was not used due to the small calibre of the native veins of the child.

Stent-grafts on the other hand have a relatively fixed geometry, and are usually not performed in children. However, its role as a bridging procedure in an ill child is very attractive and can be extrapolated from adult series.[4] Stent-grafts have proven to be a feasible mode for treatment in MIA in adults, at least as a bridge to later definitive therapy when the physiologic condition of the patient is more favourable. Stenting in an adolescent in an acute setting has been reported using balloon-expandable stent-grafts.[4] Our choice of balloon-expandable stent-graft allows maximum inflation dilatation to 10mm. As the average adult Asian iliac artery is 11mm, this allows for future redilatation to nearly accommodate the increased blood flow required during his pubertal growth spurt. This hypothesis extrapolated from pediatric data on stent-graft use in coarctation of aorta has several drawbacks though. Neointimal hyperplasia and the risk of arterial wall injury have to be considered prior to redilatation, along with stent-graft shortening.

Keeping in mind the afore-mentioned growth spurt, the postoperative management entails various aspects, mainly

infection control and stent-graft patency. As this is a mycotic aneurysm which was complicated with an acute appendicitis, there remains a risk of endograft infection and subsequent pseudoaneurysm and blow-out. Choice of antimicrobial therapy is based on culture results though there is no consensus on duration of therapy.[5] Surveillance for infected grafts may be enhanced with white blood cell scintigraphy and single-photon emission computed tomography (SPECT/CT), though these modalities are hampered by its practicality.[5] Hence biochemical and clinical parameters remain the cornerstone of surveillance. Until the end of the adolescent growth spurt, surveillance should be done frequently as children may be asymptomatic of endograft thrombosis due to collateral vessels.

CONCLUSION

To our knowledge, this is the first reported case of a mycotic aneurysm in a child associated with a perforated appendix, successfully treated with a staged hybrid approach as a bridging therapy. The use of a stent-graft reduces the morbidity of the index surgery in an ill patient and allows for future bypass, at which point both target arteries and autologous conduits are bigger. Postprocedural surveillance is vital in the management to detect endograft thrombosis, especially during the growth spurt of puberty and detection of endograft infection.

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REFERENCES

1. Sarkar R, Coran AG, Cilley RE, Lindenauer SM, Stanley JC. Arterial aneurysms in children: clinicopathologic classification. J Vasc Surg 1991; 13(1): 47-57.
2. Bardi I, Ben Chehida F, Barsaoui S, Hammou A, Ouerghi M, Bousnina S, et al. Mycotic aneurysm of the iliac artery disclosed by an abdominal mass. Arch Fr Pediatr 1993; 50(7): 609-11.
3. Cox SG, Naidoo NG, Wood RJ, Clark L, Kilborn T. Tuberculous iliac artery aneurysm in a pediatric patient. J Vasc Surg 2013; 57(3): 834-6.
4. Osmán I, Barrero R, León E, Medina R, Torrubia F. Mycotic pseudoaneurysm following a kidney transplant: a case report and review of the literature. Pediatr Transplant 2009; 13(5): 615-9.
5. Björck M, Earnshaw JJ, Acosta S, Bastos Gonçalves F, Cochennec F, Debus ES, et al. Editor's Choice - European Society for Vascular Surgery (ESVS) 2020 Clinical Practice Guidelines on the Management of Acute Limb Ischaemia. Eur J Vasc Endovasc Surg 2020; 59(2): 173-218.