Aneurysms in pediatric age: a challenging and rare disease entity

Ahmed Sayed[a], Ahmed Elsadat[c] and Marwa Mashaal[b]

Introduction **Pediatric arterial aneurysms are extremely rare. Their etiology can be congenital, mycotic, following infective endocarditis, post-traumatic pseudoaneurysms, or vasculitis. The treatment strategy in children is not very clear because of the small number of cases.**

Methods **This study included eight children with arterial aneurysms, which included one thoracic, two abdominal aortic, one iliac, three upper-extremity, and two carotid aneurysms. Revascularization was performed using an autogenous vein whenever feasible. Anastomoses were performed with interrupted sutures with nonabsorbable material to allow for future growth of the vessels. Ligation was allowed only after ensuring that the distal collateral circulation was adequate.**

Results **Eight aneurysms were reconstructed and one carotid aneurysm was ligated. No neurologic events occurred after the carotid ligation. Follow-up ranged between 4 months and 4 years and showed no recurrences or occlusion of reconstructed aneurysms, as detected clinically and radiologically. The grafts used for reconstruction – including the synthetic ones – were found to be growing with the age of the children. In one aortic case, there was mild stenosis in one of the iliac limbs, but that was asymptomatic. The extremity aneurysms repair were uneventful, with good flow in the affected extremity.**

Conclusion **Repair of aneurysms in children is feasible and yields good midterm results. Management is usually individual and tailored to each case. Finding the suitable conduit is a challenge, and autogenous veins are preferred whenever available.** ***Ann Pediatr Surg*** **11:192–196 © 2015 Annals of Pediatric Surgery.**

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Introduction

Arterial aneurysm is one of the most serious vascular diseases in adults, and is most commonly caused by atherosclerosis. Its management was established long ago by exclusion and/or repair by open surgery or by endovascular intervention. In children aneurysms are extremely rare. Its etiology can be infection, trauma, connective tissue disorder, or vasculitis [1]. In many patients the cause remains unknown, and is described as idiopathic. The treatment strategy in children is not very clear because of the small number of cases.

Patients and methods

This study prospectively collected data between January 2012 and January 2014 of children presenting with arterial aneurysms at an age of less than 12 years. The data collected included age, sex, presentation, investigations conducted for diagnosis, and imaging studies used to evaluate the extent of the aneurysms and their morphology. The etiology of the aneurysms was reported and was diagnosed from the child's history and examination results. Aneurysms following an attack of infective endocarditis were considered infective (mycotic) aneurysms. Other aneurysms preceded by a history of trauma at the site of the aneurysm were considered post-traumatic aneurysms, provided they were pseudoaneurysms. Aneurysms with no evidence of infection, trauma, connective tissue disorders, or autoimmune diseases were considered idiopathic (or congenital) aneurysms.

The treatment option and operative details were recorded. Management was by resection with revascularization or ligation according to individual variables. Revascularization was performed using an autogenous vein whenever feasible. Synthetic grafts were used when an autogenous vein of an adequate caliber was not available. Anastomosis was performed by means of interrupted sutures with nonabsorbable material to allow for future growth of the vessels. Ligation was allowed only after ensuring that distal collateral circulation was adequate by measuring the pressure in the distal artery stump after ligation.

The aneurysms were followed up clinically and radiologically to confirm good circulation and absence of recurrence after the procedure. Recurrences and occurrence of aneurysms elsewhere during follow-up were also managed according to the individual circumstances and the data were reported.

Results

Eight patients manifesting nine arterial aneurysms were reported. Their ages ranged between 6 months and 11 years at the time of presentation. Table 1 shows patients' data. There were one thoracic, two abdominal aortic, one iliac, three upper-extremity, and two carotid aneurysms. The aortic and iliac aneurysms were infective aneurysms following infective endocarditis. One upper-extremity aneurysm was post-traumatic following a trial of venous cannulation in an 8-month old child lacking proper sized veins, and ended in the injury of the axillary artery,

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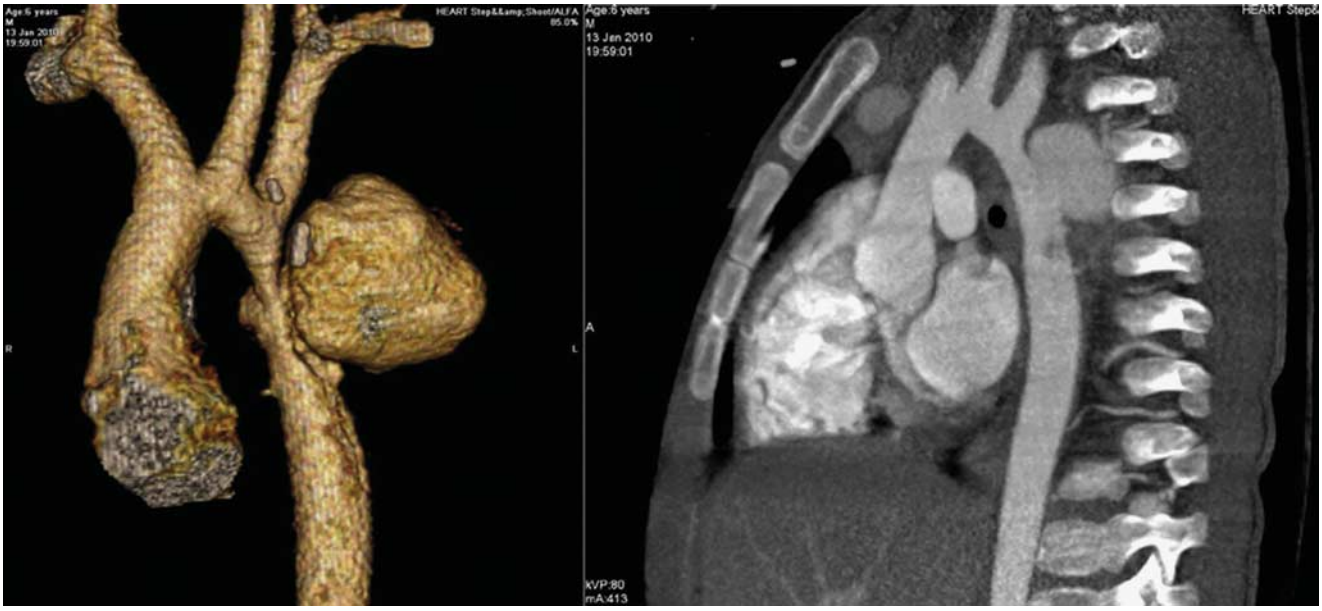
Table 1 Patients' data

	Site	Age	Sex	Etiology	Presentation
1	Thoracic aorta and AAA	6 years	M	Mycotic	Lumber and lower-extremity pain
2	AAA	11 years	F	Mycotic	Right loin and thigh pain
3	Right common iliac artery	7 years	M	Mycotic	Right lower-extremity pain
4	Left brachial artery	6 months	M	Idiopathic	Cubital fossa swelling
5	Left axillary artery	3 years	M	Idiopathic	Axillary swelling
6	Left axillary artery	8 months	M	Iatrogenic trauma	Axillary swelling
7	Left internal carotid artery	5 years	F	Idiopathic	Left neck swelling
8	Left internal carotid artery	2 years	M	Idiopathic	Left neck swelling

AAA, abdominal aortic aneurysm.

Fig. 1



Mycotic thoracic aortic aneurysm.

forming a pseudoaneurysm. The remaining extremity aneurysms were considered idiopathic due to the absence of evidence of other pathologies. Both appeared gradually, until diagnosed clinically. Both carotid aneurysms presented as neck swellings. Diagnosis was made by duplex and/or Computerized tomography angiography.

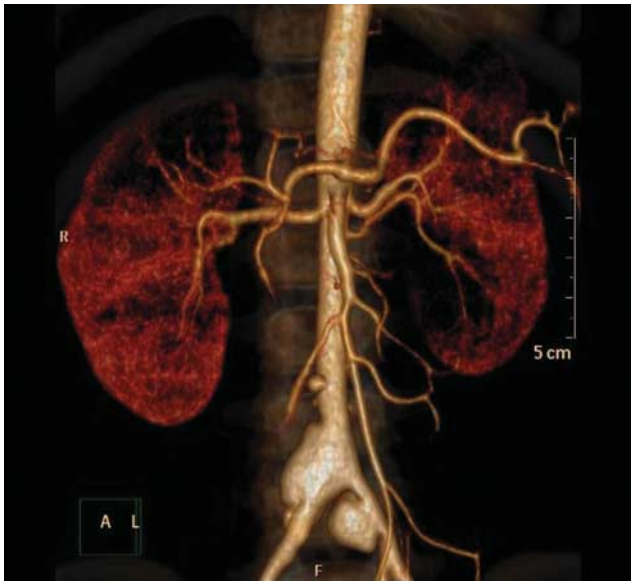
Eight aneurysms were reconstructed and one carotid aneurysm was ligated. Follow-up ranged between 4 months and 4 years, and showed no recurrences or occlusion of reconstructed aneurysms as detected clinically and radiologically.

The first patient developed infective endocarditis following repair of an aortic coarctation. He developed a descending thoracic aortic aneurysm (Fig. 1), which was managed, as an emergency under cover of antimicrobials, by resection and synthetic patch repair. One month later an aneurysm at the aortic bifurcation appeared because of embolization by infected material (Fig. 2). This was resected and reconstructed by a 6 mm Dacron graft, which was tailored to match the size of the aorta and iliac arteries. No venous conduit was available, and the aneurysm was operated after complete resolution of infection, as evidenced by repeated blood cultures. Infection recurred at the thoracic aorta after 1 year and the patient was reoperated upon with excision and replacement of 15 cm of the descending aorta using a 20 mm Dacron graft, and the graft remained patent after 48 months of follow-up. In this patient there was also a mild stenosis in one of the reconstructed iliac limbs, but that was asymptomatic.

The second infective abdominal aortic aneurysm (AAA) was leaking and was reconstructed using the superficial femoral vein (SFV) to avoid the use of synthetic material in an infected field. The iliac aneurysm was reconstructed with an iliofemoral bypass using a PTFE graft after complete resolution of infection.

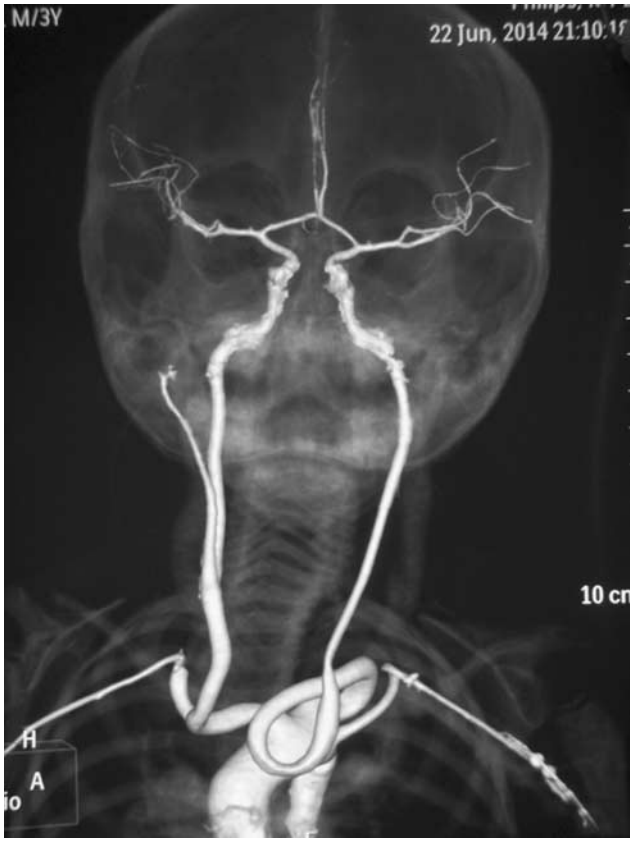
The three upper-extremity aneurysms were managed with resection and revascularization by reversed saphenous graft in one patient with axillary aneurysm, with end-to-end anastomosis in a patient with brachial aneurysm, and by sac opening with primary suture of a small tear in the third patient with an axillary pseudoaneurysm. All extremity aneurysm repairs were uneventful with good flow in the affected extremity.

Fig. 2



Mycotic aortoiliac aneurysms.

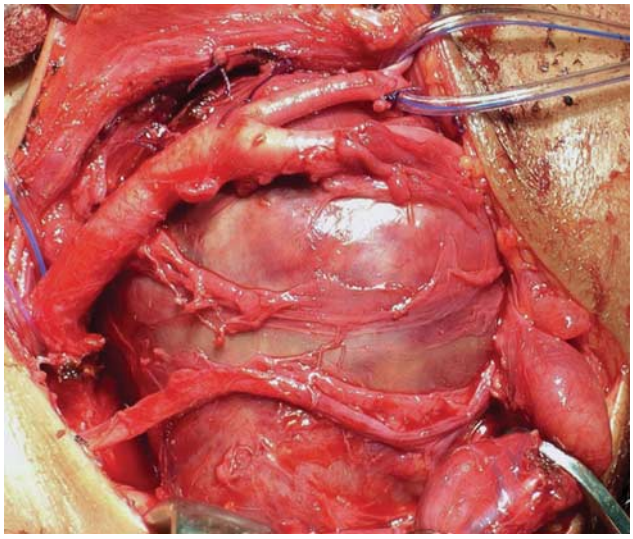
Fig. 3



Patent left internal carotid artery after reconstruction using a reversed saphenous graft. The common carotid artery is elongated and shows a coil.

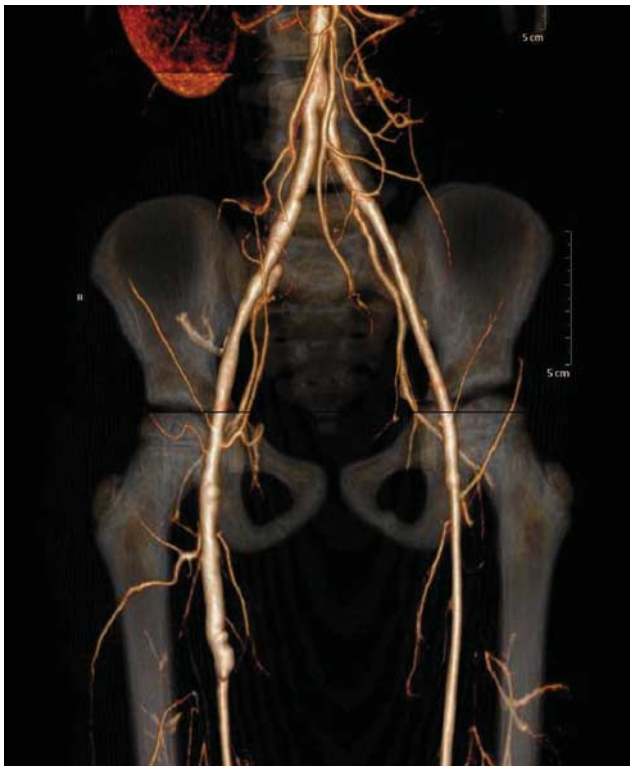
One carotid aneurysm was reconstructed using a saphenous graft with excision of a cutaneous hemangioma. One year later it remained patent (Fig. 3), but a swelling

Fig. 4



Carotid arteries stretched over a huge aneurysm arising from the distal ICA just below the skull base. ICA, internal carotid artery.

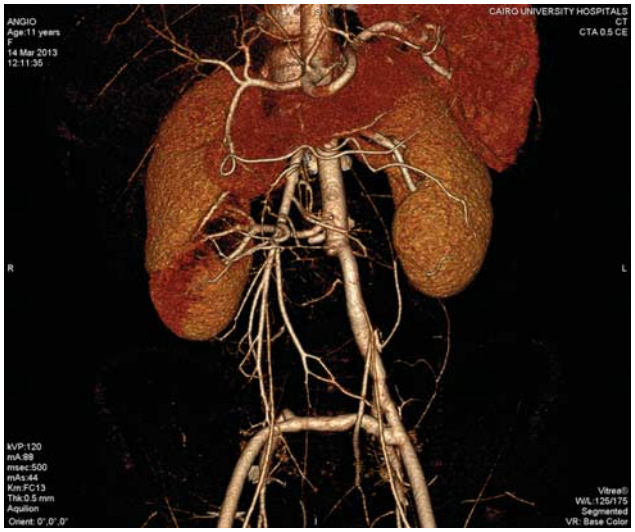
Fig. 5



Aortoiliac reconstruction using a 6 mm Dacron graft at 24-month follow-up. The graft diameter has increased to 7.5 mm.

was evident on follow-up. This was resected safely without affecting the saphenous graft, and pathologic examination revealed a hamartoma. The other carotid aneurysm was ligated because of very small and friable distal internal carotid artery (ICA) (Fig. 4). Adequate collateral circulation was confirmed by the presence of a stump pressure in the distal ICA after clamping of

Fig. 6



Reconstructed aortoiliac segment using SVF at 3 months postoperatively. SFV, superficial femoral vein.

90 mmHg and pulsatile back bleeding. No neurologic events occurred during the follow-up.

Pathologic examination of the idiopathic aneurysms revealed no specific pathology suggestive of vasculitis or connective tissue disorders. Serial postoperative imaging revealed that the grafts used for reconstruction – including the synthetic ones – were slowly growing with the age of the children, as documented by duplex and computed tomography imaging (Figs 3, 5 and 6).

Discussion

The current study showed successful management of nine aneurysms in eight children aged between 6 months and 11 years. The causes of the aneurysms were infection in three patients and trauma in one patient, and remained unidentified in four patients. A large series from Michigan proposed in 1991 a clinicopathologic classification for pediatric aneurysms [2]. However, this was not widely used because of the inability to determine the cause. The small number of reported cases made it difficult to determine the exact incidence, causes, natural history, and prognosis of these aneurysms. The firm identification of cause is often complicated by nonspecific late pathologic findings in aneurysm specimens regardless of the prior disease state responsible for their development [2]. The most common etiology found in the literature was infection [3].

The small size of the arteries and the need to allow growth of the child made the choice of reconstruction challenging. We used synthetic conduits for repair in two patients with aortic and iliac aneurysms. In both patients, autogenous veins were thought to be inadequate. The use of synthetic grafts has been reported and has shown good results [3,4]. The SFV was used in one infected aneurysm where it was of suitable diameter and length.

The choice of bypass conduit is controversial. It is preferable to use autologous grafts [5]. However, the use of prosthetic conduits has been reported with satisfactory results [4–6]. Barral and colleagues reported that the greatest potential problem lies in the size discrepancy between the aorta and the graft. Therefore, they recommended deferring surgery until the patient was older to mitigate this discrepancy [4]. Robicesk *et al.* [6] have reported elongation of synthetic graft material with progression of body morphology. This feature was observed in our patient with AAA repaired with a Dacron graft on 30-month follow-up images.

The surgical management of mycotic aneurysms should include the following: (i) preoperative control of infection; (ii) aneurysm resection and debridement of infected tissues and arterial reconstruction through uninfected tissue planes with selected use of interposition grafting through the bed of the resected aneurysm; (iii) soft tissue coverage; and (iv) the use of autologous tissue for reconstruction whenever possible [7].

We used the SFV for revascularization of the AAA where the operative field showed obvious signs of tissue infection, making a fatal secondary hemorrhage an anticipated risk. Moreover, SFV exploration showed good caliber and wall thickness. Thus, it was decided to use a part of the vein as an aorto-left common iliac bypass. The second part the vein was anastomosed between the left CIA and the right external iliac artery avoiding any anastomoses at the site of infection around that artery. SFV grafts proved to be an excellent conduit for vascular reconstruction after drainage and debridement of infected tissue and graft material [8]. The use of a single SFV for reconstruction of the aortoiliac segment was reported with good patency and less operative time [9].

Ligation of arteries without concomitant revascularization can be accepted only in hostile anatomic and pathologic circumstances, provided the collateral circulation is adequate to maintain sufficient distal circulation. In adults carotid ligation is only allowed when distal stump pressure exceeds 70 mmHg [10]. In a series of carotid Behcet aneurysms, seven aneurysms were ligated without any major neurologic events or stroke [11]. In general, younger patients have a better tolerance to carotid occlusion, but this is not reported in the pediatric age group. We ligated the ICA in one patient and this did not result in any neurologic morbidity.

The other carotid aneurysm showed a rather complex pathology, with reappearance of a soft tissue neoplasm. However, the carotid revascularization remained patent and quite separable from the resected mass.

Conclusion

Repair of aneurysms in children is feasible and yields good midterm results. Management is usually individual and tailored to each case. Finding the suitable conduit is a challenge, and autogenous veins are preferred whenever available. Proper technique of revascularizations is associated with good durability.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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