Case Report

Evolution of spontaneous dissecting mycotic Superior Mesenteric aneurysm in a 12-year-old child: A case report

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ABSTRACT

Visceral artery aneurysms are uncommon especially in children. One of the main complications before surgery is rupture.

This 12-year-old child presented with a large, fast growing, mycotic superior mesenteric aneurysm that had all the favourable conditions for rupture. There was spontaneous dissection of the weakened media with partial erosion of the aneurysm into the wall of the third part of the duodenum. However, this aneurysm formed a thrombus that gradually occluded the lumen. This led to formation of collateral vessels for the continued vascular supply of the midgut.

The uniqueness of this case report has been highlighted from several points (the rarity of the condition in children, the favorability of the conditions to rupture, the gradual but complete luminal occlusion with the eventual formation of collaterals to supply the midgut, and the spontaneous medial dissection with partial duodenal wall erosion without causing rupture).

Although there is no standard surgical approach, early elective surgery is recommended. Non-operative approach is an option that should aim at reducing the risk for rupture. Control of blood pressure is key.

This child underwent surgery. Under total vascular exclusion, the aneurysm was opened. After total luminal occlusion and collateral blood supply to midgut was noted, infected thrombus was evacuated and the aneurysm walls debrided.

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INTRODUCTION

Aneurysms and pseudoaneurysms of the visceral arteries are uncommon. They have a prevalence of 0.1 – 2% in the general population.[1,2] This is especially true for mycotic aneurysms; and more so true for the paediatric population.[3] The aetiology of dissecting superior mesenteric artery branch aneurysm are unspecific. Generally, the abdominal aneurysm may be associated with atherosclerosis (32%), medial degeneration/ dysplasia (24%), abdominal trauma (22%), infection and inflammatory disease (10%) mainly mycotic aneurysm. Other associated conditions include connective tissue disorders and hyperflow conditions.[4]

Mycotic aneurysm are said to account for 60% of Superior Mesenteric Artery aneurysms and pathogenesis involves destruction of arterial wall by infection.[5] The inflammation of the vascular wall and formation of mycotic aneurysm starts with the disturbance of flow in the vasa vasorum that occurs with narrowing or obstruction of such blood vessels.[6] The natural course of Superior Mesenteric Aneurysms is usually fatal with high risk of rupture.[7]

Keywords: Superior mesenteric artery, visceral artery aneurysm, aneurysm, midgut.

Superior Mesenteric Artery supplies the midgut and therefore has risks of rupture, shock and loss of midgut in case of sudden occlusion.[8]

Clinical diagnosis is difficult and, in many cases,, the aneurysm is discovered and diagnosed only after rupture.[9] This report is aimed at raising awareness on this natural course of the disease despite the presence of all the risk factors for rupture of the aneurysm.

CASE PRESENTATION

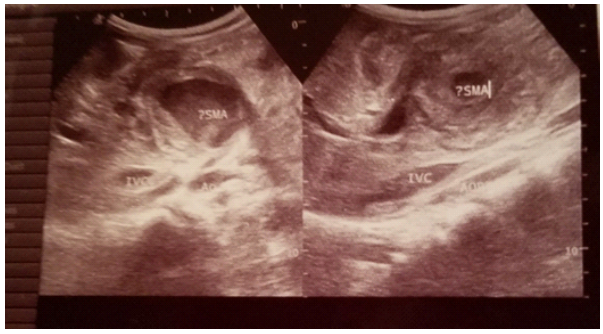
A 10-year old male referred from local hospital with a month history of abdominal pains, occasional vomiting, constipation and fever on and off. There was no history of abdominal trauma prior to onset of the symptoms, history of loss of appetite or weight loss. Previously well; non significant past medical and surgical history. Non significant drug history. No similar illness in the family. He had no history of connective tissue disorder or metabolic diseases or autoimmune diseases in the family.

The patient was conscious, oriented, emaciated, but neither pale nor jaundiced. Patient was afebrile. He had tinea capitis.He had normal heart sounds and no joint hyperextensibility.

His admission temperature was 36.5°C and Bp 119/45mmHg. The subsequent Bp profile ranged from 97 – 112mmHg for SBP and 63 – 71mmHg DBP. There were scarification marks seen in suprapubic region. The abdomen was soft, and non-tender. There was a pulsating abdominal mass; with bruits noted.

His full blood count showed hemoglobin of 7.99g/dl. The other parameters were normal. Peripheral smear, ESR, sickling test, liver function tests, urea and electrolytes were normal. The HIV test was non reactive. The first abdominal ultrasound scan and doppler ultrasound showed an epigastric complex cystic mass with swirling motion of blood in it, and in communication with the abdominal aorta. It measured 5.75cm x 5.47cm x 4.74cm. After 3 months a second scan showed a dilated superior mesenteric artery with lumen of 3.53cm diameter with thick walls (see figure 1.

Figure 1: First ultrasound scan images showing cross section of suspected SMA aneurysm 5.75cm x 5.75cm x 4.74cm

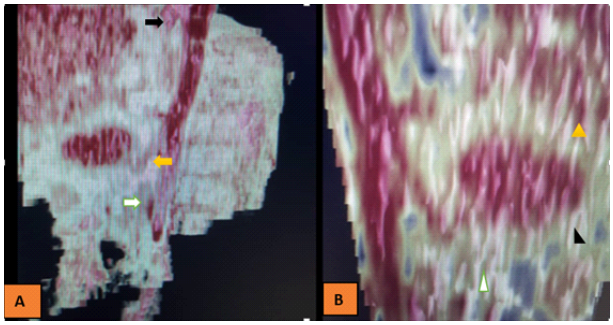


Septic screen including several blood and urine cultures yielded no bacterial or fungal growths.

Colour-flow doppler imaging showed a pattern of swirling blood that was suggestive of an aneurysm.

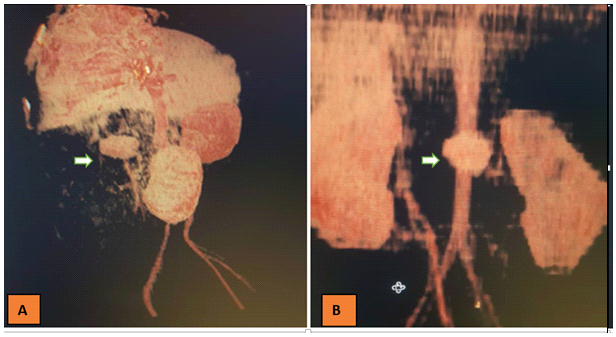
The initial CT Scan (plain and with contrast) showed a well circumscribed saccular aneurysm along the superior mesenteric artery measuring 43mm x 39mm (see figure 2 below). It had thick irregular walls of approximately 15mm at point of maximal thickness. Free fluid was noted in the lesser sac and pelvis. Patient was planned for aneurysmorrhaphy or grafting with a native blood vessel. Surgery was delayed as the surgeon with vascular surgery expertise was unavailable.

Figure 2: CT images of SMA aneurysm in 2D. A, in relation to Celiac Trunk and Inferior Mesenteric Artery (arrows). B, in relation to collaterals arteries supplying the midgut (arrow heads)



Six months later, CT Scan was repeated. This time, it showed a superior mesenteric artery aneurysm measuring 6.0 x 6.6cm (see Figure 3 below).

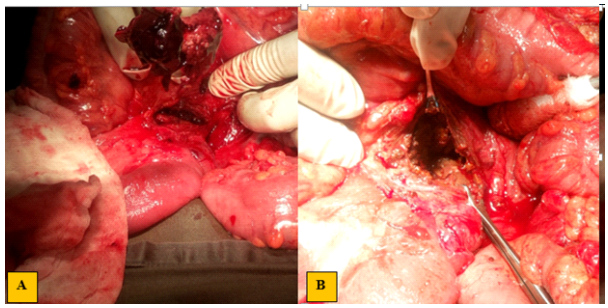
Figure 3: CT 3D reconstruction 6 months later. A, Lateral view SMA aneurysm 6.0 x 6.6cm (arrows). B, AP view



He was evaluated for cardiac disease (endocarditis) using echocardiography and ECG. The two investigations were normal.

Patient was taken to theatre by a combined team of surgeons (including pediatric surgeons, vascular surgeon, paediatric anaesthetist and a cardiac theatre nurse). Patient was placed under GA and abdomen opened using upper transverse abdominal incision. Intraoperative findings: Left iliac abscess (inflammatory mass) involving proximal sigmoid, omentum and left colic gutter. This mass measured approximately 5cm x 5cm. Excision of mass was done. Pus was suctioned out and abscess cavity washed. Sigmoid colon was repaired. Walled off aneurysm measuring 80mm X 80mm found approximately 2cm from the aorta. It was adherent to small bowel and pancreas. Collateral blood vessels found between the SMA proximal to the aneurysmal mass and the SMA branches distal to the mass. All bowels and organs were well perfused. The SMA was contolled and aneurysmal mass opened.

Figure 4: intra-operative images. A, shows evacuated clot from SMA aneurysm. B, shows thick irregular aneurysm wall partially eating into 3rd portion of duodenum



Old blood clots were found in the core of the aneurysm, attached to what appeared was a mycotic mass. No bleeding was noted even after evacuation of clots (see Figure 4 above). Aneurysmal wall incisional biopsy was done.

Histology sections from the hematoma showed hemorrhage with bands of collagen. The sections from the abscess cavity demonstrated cysts containing amorphous oesinophilic materials and had linning of low cuboidal cells. Surrounding the cysts wass a mixed inflammation cell infiltrate of lymphocytes and neutrophils. Some areas characterised by replacement of cysts by inflammatory cell infiltrate and abscess. There was no organism isolated. The patient made an uneventful recovery. After termination of antibiotics and antifungals he did not go through Superior mesenteric arteriography or angiogram. Three weeks later he was discharged from the hospital and was symptom free at 1 month and 12 months reviews.

DISCUSSION

Mycotic aneurysms occur from septic emboli in patients with infective endocarditis and may involve any artery, although not frequently detected before autopsy. The most common sites include brain, abdominal aorta, Superior mesenteric artery, to mention but a few.[3] Though a rare entity and often

asymptomatic, visceral artery aneurysm are known to rupture if size greater than 55mm and expansion is fast. Those aneurysms expanding more than 5mm per year are more prone to early damage and rupture especially when infected or traumatised.[5] This case had all the favourable conditions for rupture (location, rapid rate of expansion, size greater than 55mm, infection, young age). However, It did not. Instead, it ran a more subclinical course, developed collateral blood supply and clotted. It is not clear whether there was a spontaneous dissection of the main trunk leading to thrombosis and eventual obliteration of the aneurysmal lumen. It is possible this is what happened in this case.

Scholars have reported on the natural history of spontaneous dissecting aneurysm of the proximal superior mesenteric artery.[10] Senocak writes that rupture of visceral artery aneurysms is rare and that it is uncertain whether the aneurysms should be actively treated.[3]This case adds to a body of evidence of visceral arterial aneurysms running that clinical course.

Treatment of aneurysms has evolved overtime. A number of authors have reported on both successful surgical treatment and conservative management.[11] Surgical treatment include aneurysmorrhaphy, bypass or interposition grafts such as repair with native vessels, repair with cyopreserved allografts, and repair with various synthetic grafts.[12] However, surgical treatment with a bypass or interposition graft have potential complications such as massive bleeding and anastomotic dysruption with average mortalities that are way over from 15%.[13] Interventional radiology treatment methods including endovascular aneurysm repair (such angiographic transluminal embolisation and repair with stent grafts) has become popular and, treatment of choice in many settings. This is especially true for treatment of aneurysms that occur in life threatening anatomic locations.[8] Not only is it less invasive treatment modality than open repair, it also offers obvious advantages in terms of reduced perioperative morbidity and mortality in high risk patients with mycotic aneurysms.[14] Many researchers recommend obliteration of the aneurysms larger than 2cm in diameter and or the rapidly expanding ones.[15]

Like in aortic aneurysm, non-operative management must be aimed at minimising the risk of aneurysmal expansion and rupture; corner stone being the control of blood pressure.[12] However, generally, early elective repair is preferred to prevent rupture. There is no optimal or standard operative approach that overrides the others. Literature reports multiple methods.

Our patient was planned to have aneurysmorrhaphy or graft with native vessel done. Multidisciplinary team was constituted. This included a vascular and transplant surgeon, a team of paediatric surgeons, a team of anaesthetists and theatre nurses led by cardiac surgery nurse specialist. A laparotomy was done and a lesion with features that were consistent with spontaneously dissecting aneurysm of the proximal superior mesenteric artery with obstructed blood flow; but had formed collaterals vessels to the midgut. Using the principles of total vascular exclusion the aneurysmal lesion was opened, and when found with infected thrombus, was carefully evacuated and debrided. It was tested for bleeding. There was no bleeding. The abdomen was closed. Patient completely recovered and remains symptom free.

CONCLUSION

A superior mesenteric aneurysm can either progress, rupture or occlude. In case of the latter, as in our case, gradual occlusion may lead to formation of collateral vessels that either bypass the aneurysm or otherwise take over the blood supply of the midgut. These courses in the natural history of the disease should inform the principles of surgical care.There is no standard surgical option for treatment of visceral aneurysms. Surgery should be aimed at reducing the risks of aneurysmal expansion, rupture and bleeding while ensuring continued blood supply to the affected viscera. Early repair is recommended. Conservative approach is still much of an option. However, it should be abandoned once the aneurysm

fulfills the criteria of rapid expansion and/ or grows larger than 20mm in diameter. Limitations: Interventional radiology treatment methods that could provide more access to anerysmal treatment are still under explored in our settings; the challenges being lack of skilled manpower and lack of facilities, to mention but a few.

This condition is uncommon. However, it provides insights into the principles of surgery that can be applied across the whole spectrum of aneurysms.

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