VOLUME - 13, ISSUE - 04, APRIL - 2024 • PRINT ISSN No. 2277 - 8160 • DOI : 10.36106/gjra

Original Research Paper

General Surgery

EXTREMELY RARE CASE OF HAEMOPTYSIS IN PAEDIATRIC AGE GROUP: IDIOPATHIC AORTIC ANEURYSM WITH AORTO-BRONCHIAL FISTULA

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ABSTRACT Aneurysms of the post-coarctation portion of the aorta are exceptionally rare. When they emerge de novo, they are even rarer, and they are very fatal if not surgically treated on time. Post coarctation aneurysms most commonly occur after surgical repair of aortic coarctation and present as pseudoaneurysms which may erode into surrounding tissue and may lead to aorto-bronchial fistulas. The prognosis of such aorto-bronchial fistulas is poor with a high morbidity and mortality. Here we present an exceedingly rare case of haemoptysis due to an auto-bronchial fistula in an 11-year-old boy which was diagnosed on time and could therefore be successfully treated. Presenting this case, we would like to emphasize that haemoptysis can occur due to an aorto-bronchial fistula which requires immediate diagnosis to allow life-saving treatment either by open surgery or by endovascular techniques.

KEYWORDS : Haemoptysis, Idiopathic Aortic Aneurysm, Aorto-Bronchial Fistula

CASE REPORT

An 11-year-old boy came to the hospital after vomiting/coughing a huge amount of blood. He was not sure if he coughed or vomited blood but described "a trickling feeling" in his throat. He was immediately and thoroughly evaluated by a physician in the emergency department. His blood work showed severe anemia, and he was hypotensive (BP 90/70mmHg). Therefore, necessary resuscitation measures were taken to stabilize him in the ER. His bedside chest X-ray film was unremarkable and did not uncover an unequivocal cause for haemoptysis/hematemesis. Upper GI endoscopy by a gastroenterologist was also unremarkable letting us suspect haemoptysis rather than haematemesis. A chest CT (computed tomography) scan was therefore performed and revealed a massive aortic aneurysm.

An urgent Cardiothoracic and Vascular surgery team consult was arranged, and an emergency aortogram was done. Aortogram showed a large saccular aneurysm arising from the posterior part of the upper thoracic segment of the Aorta (figure 1) justifying urgent surgical treatment

Through a left posterior thoracotomy approach, blood was shunted via an aorto-iliac bypass circuit. We could reveal that the massive aneurysm eroded into the anterior segment of the left upper lobe. Hence, the aneurysm was separated from the lung followed by a wedge resection of the same segment. The aorta was clamped above and below the aneurysm, an autotomy was done and extended above and below, and it was repaired by an 18 mm PTFE graft (figure 2). Surgery was performed without intraoperative complications and the patient was extubated on the table after the procedure. (Figure 3 & 4)

The patient's recovery was uneventful for 5 days. On the 6th day, the attending doctor noticed that the patient was developing weakness in his left lower limb and peripheral pulses were feeble on palpation. As his weakness did not improve with conservative management, angiography was performed and revealed sclerosis of the left external iliac artery, probably due to the insertion of a 10F cannula in the iliac artery for aorto-iliac bypass circuit, which might have been too big for the young patient. On the same day, a PTFE patch angioplasty was performed on the left external iliac artery.

The patient's recovery was uneventful and no further complications occurred. 10 days after the initial presentation at the ER the patient was discharged.

Follow-up:

The patient presented at the hospital for regular follow-ups. 18 months after surgery he was free of any symptoms.

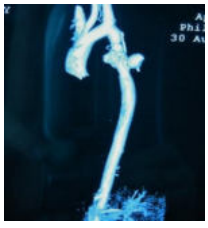

Figure 1: Aneurysm of descending thoracic aorta

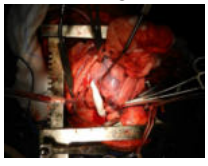

Figure 2: Repair of Aneurysm with PTFE graft

Figure 3: Erosion of the left upper lobe by the Aneurysm

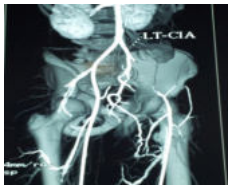

Figure 4: Sclerosis of left common iliac artery because of the use of a larger cannula

DISCUSSION

Haemoptysis can be massive and life-threatening. The most common aetiologies of haemoptysis are pulmonary diseases such as tuberculosis, bronchial malignancy, or bronchiectasis.1,2

Rarely do the causes involve cardiac origin. De-novo aneurysms of the post-coarctation segment of the aorta are rare.

An aorto-bronchial fistula was first described in 1914 by Giardet. Six cases of aorto-bronchial fistulas were described in a series of autopsies by Keeper and Mallory. Jones successfully repaired an aorto-bronchial fistula in an 11-year-old girl who had undergone repair of a patent ductus arteriosus before. Today, most cases of aorta-bronchial fistulas arise as a sequela of aneurysm or pseudoaneurysm of the thoracic aorta. The most common causes in the past were atherosclerosis or infections like syphilis.3,4,5

When these lesions are corrected by surgery, the approach is usually by left thoracotomy, and repair is done using patch aortoplasty, graft replacement, and subclavian artery flap repair.6

Most cases are noticed after using the 'Dacron patch'7 which is done either for congenital anomaly correction or correction of aneurysms due to atherosclerosis in older age groups. Moreover, they develop in a span of 15-20 years after surgery by open technique. There are reports of earlier presentations after endovascular stent graft repair.8

The case reported here is unique because there was no prior history of aortic surgery, the aneurysm was arising from a very unusual site at the post-coarctation segment, the aneurysm had eroded into the pulmonary parenchyma, and the patient presented with massive haemoptysis rather than with intermittent haemoptysis which is the more typical presentation reported so far.

The reported case also demonstrates the challenge of reliably determining the source of bleeding in such situations and deciding whether an investigation of the upper GI tract and/or the respiratory system should be prioritized. CT scans are increasingly done as one of the primary investigations, but may not necessarily reveal a fistula as in our case. Thus, an angiogram should be the preferred investigation if the CT scan does not reveal the cause of bleeding.

Vascular interventions such as endovascular stent graft repair have become increasingly popular to treat vasculopathy as they are comparably successful, are less time-consuming, and are associated with reduced morbidity. However, long-term results of endovascular techniques are yet to be seen. The long-term results of repair of ABFs are good and recurrence is extremely rare.

CONCLUSION

When a young patient presents with haemoptysis with no prior history of cardiovascular surgery an aorto-bronchial fistula or a ruptured aortic aneurysm should be considered as a potential cause of bleeding. E.R. physicians should be alert, especially in patients with a history of prior aortic surgery including repair of congenital pathologies.

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