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Foreign body–induced aorto-oesophageal fistula: a review of five cases and their management

五宗異物引致食道主動脈瘻的病例及其治療方法的回顧

Foreign body–induced aorto-oesophageal fistula is a rare cause of massive upper gastro-intestinal bleeding and, in the absence of a timely diagnosis and surgical intervention, can be fatal. During a period of 25 years, five patients with foreign body–induced aorto-oesophageal fistula underwent surgery in our department. Three patients survived. All survivors required more than one surgical intervention. The clinical course of these five patients and the management of this potentially fatal condition are reviewed.

異物引致食道主動脈瘻極少導致上部胃腸嚴重出血，然而若此情況發生而沒有及時診斷和進行手術，卻有可能導致死亡。在二十五年內，有五名異物引致食道主動脈瘻的病人在本部門進行手術，其中三人獲救。獲救的病人均須接受多於一次的手術。本文回顧這五名病人的臨床發病過程和治療方法。

Introduction

When a foreign body (FB) is retained in the oesophagus, it erodes the wall and the adjacent aorta, and an aorto-oesophageal fistula (AEF) may be formed. Episodic minor haematemesis usually occurs and without timely diagnosis and intervention, the patient invariably succumbs to exsanguinating haemorrhage. It is generally accepted that FB-induced AEF is rare, but its exact incidence is unknown. In 1978, a review[1] of 2394 cases of FB in the oesophagus identified two cases of AEF. In the last 25 years, five patients with AEF have undergone surgery at the Grantham Hospital, Hong Kong. Three of them survived. This report documents these five cases of AEF and illustrates the importance of early diagnosis and surgical intervention.

Case series

Case 1 (1978)

A 38-year-old man presented to the Accident and Emergency Department with low retrosternal pain after swallowing a piece of chicken bone. Plain X-ray films of the neck and chest showed no abnormalities. After discharge, he noticed intermittent mild retrosternal pain. Ten days later, he presented again with a history of vomiting several small blood clots and passing one tarry stool. While in the department he developed massive haematemesis and went into cardiac arrest. Resuscitation was successful and he was transferred to the operating room with a diagnosis of upper gastro-intestinal haemorrhage, suspicious of AEF. Oesophagoscopy revealed a piece of chicken bone embedded in the wall of the oesophagus, 28 cm from the incisors, that moved with aortic pulsation. The chicken bone was not touched and the oesophagoscope was carefully retrieved; simultaneously the patient developed massive arterial bleeding and an emergency left thoracotomy was performed. A 10-cm long haematoma around the descending aorta distal to the origin of the left subclavian artery was identified. The patient died on the operating table of uncontrollable haemorrhage.

Case 2 (1978)

A 59-year-old man was admitted to a general hospital with mid-retrosternal pain that worsened on swallowing. He had swallowed a piece of chicken bone 2 days

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previously. At oesophagoscopy, a piece of chicken bone situated 27 cm from the incisors was removed uneventfully. He was discharged 1 week later after completion of a course of intravenous antibiotics. Five days later, he developed mid-retrosternal pain and vomited a large quantity of blood. He was given one unit of whole blood and commenced on intravenous antibiotics. Chest X-ray and gastrografin swallow disclosed no abnormality. A second episode of haematemesis necessitated emergency left thoracotomy. At thoracotomy the descending aorta, 5 to 7 cm below the origin of the left subclavian artery, was found to be strongly adherent to the adjacent oesophagus. With the aorta cross-clamped above and below this site, further dissection of the area of adherence revealed a 4 to 5 mm communication between the aorta and oesophagus. There was no pus but a small amount of granulation and fibrous tissue was found. Both openings were closed with interrupted 3-0 prolene. He was transferred to our unit for definitive management of potentially infected suture lines. After deliberation, the patient was re-explored the next day. The operative field was clean and the tissues appeared healthy: a 1-cm wedge of aortic wall was excised and the defect closed transversely. The left hemithorax was thoroughly rinsed with a large volume of warm saline. Bacterial culture of a swab taken from the mediastinum was negative. The patient recovered uneventfully and was discharged after completion of a course of intravenous antibiotics. He was alive and well at 18-month follow-up. This case has been reported as the first recorded survival of FB-induced AEF.[2]

Case 3 (1989)

A 35-year-old man was admitted to a general hospital with retrosternal chest pain. He had swallowed a piece of goose bone 3 days earlier. Radiological examination of the neck and chest showed no abnormality. Upper endoscopy revealed no FB; a small abrasion was noted on the left lateral oesophageal wall 20 cm from the incisors and he was discharged home. Four days later he collapsed in the street following an episode of retrosternal pain and haematemesis. Soon after re-admission to the general hospital, he vomited about 1 L of fresh blood. Emergency upper endoscopy revealed an ulcer overlying a haematoma, 20 cm from the incisors on the left lateral wall of the oesophagus. A Sengstaken-Blakemore tube was inserted and the oesophageal balloon inflated. Two units of blood were transfused and he was transferred to our hospital for further management. Repeated chest X-ray films showed a widened left superior mediastinum with streaks of free gas in addition to the in-situ Sengstaken-Blakemore tube. An aorto-oesophageal fistula was diagnosed and an emergency left thoracotomy was performed. At operation, 30 mL of turbid fluid was found in the left pleural cavity with the left lung apex adherent to the mediastinum. The left subclavian artery was encased by a 4 x 3 x 2 cm inflammatory mass composed of blood clots and foul-smelling necrotic tissue. A 1-mm perforation was noted on the posteromedial wall of the left subclavian artery at its origin. No oesophageal fistula opening or FB was found. The inflammatory mass, together with a segment of the left subclavian artery, was excised. The arterial stump was closed. A decision was made to manage the oesophageal lesion conservatively because it was inadvisable to dissect the mediastinum further and open more tissue planes. Bacterial culture of the abscess wall yielded a mixed growth of aerobes and anaerobes, and a course of appropriate antibiotics was prescribed. A gastrografin swallow on the second postoperative day showed no leakage of contrast material and feeding via a nasogastric tube was started. The patient was transferred back to the general hospital for further management. On postoperative day 9, the patient developed clinical features of sepsis, and the findings of a repeated chest X-ray were compatible with a mediastinal abscess. Re-exploration revealed a 2 x 1 cm perforation of the thoracic oesophagus above the aortic arch. The oesophagus was transected and its distal stump closed. A terminal cervical oesophagostomy and a gastrostomy were performed to divert oropharyngeal secretions and allow feeding. Continuity of the gastrointestinal tract was later restored with a retrosternal jejunal oesophagoplasty in order to avoid potentially difficult mediastinal dissection. A completion oesophagectomy was performed 1 year later because of a recurrent chest wall abscess caused by a persistent cutaneous-oesophageal fistula. The patient remains well. This case has also been reported previously.[3]

Case 4 (1999)

A 64-year-old man on long-term steroid therapy for psoriasis, was admitted to a general hospital with dysphagia that developed after swallowing a fish bone. Upper endoscopy was normal and he was discharged. Ten days later he was re-admitted with fever and dysphagia. A chest X-ray showed a widened mediastinum and a repeated upper endoscopy disclosed a 1-cm oesophageal mucosal ulcer on an 8-cm haematoma, 35 cm from the incisors. Computed tomography (CT) of the thorax showed a pseudoaneurysm at the distal aortic arch, 4 cm in size and encased by a haematoma. He was transferred to our hospital for further management. While waiting for surgery, the patient developed massive haematemesis and went into cardiac arrest. An emergency left thoracotomy was performed after successful resuscitation. Bleeding was controlled by cross-clamping the aorta above and below the pseudoaneurysm. The wall of the pseudoaneurysm was composed of necrotic tissue and dense granulation tissue. An interpositional 22-mm-knitted Dacron arterial graft was sutured in place. The patient developed disseminated coagulopathy and it was decided not to proceed with the oesophageal repair. The patient died of multi-organ failure the following day.

Case 5 (2003)

A 30-year-old woman was admitted to a general hospital following an episode of haematemesis. She gave a history of fish bone ingestion 2 weeks earlier and difficulty with swallowing since. Upper endoscopy revealed an actively oozing haematoma 28 to 30 cm from the incisors. Computed tomography of the thorax showed a descending

aortic false aneurysm. She was transferred to our hospital for further management. An emergency left thoracotomy revealed a false aneurysm at the descending aorta, just above the hilum of the left lung. A Gott shunt bypassing the false aneurysm was inserted. The descending aorta was cross-clamped above and below the false aneurysm. On opening the false aneurysm, a 1.5-cm defect with ragged edges was found on the aortic wall. It was excised together with 3 cm of the descending aorta. Aortic continuity was re-established with a woven Dacron arterial graft. The oesophageal perforation was not found: it was presumed to be small and well-contained within the mediastinum and likely to respond to drainage and antibiotics. Bacterial culture grew *Streptococcus milleri*. Postoperative upper endoscopy confirmed no frank perforation and only a mucosal haematoma. Oral feeding was resumed 3 weeks later when gastrografin study revealed no leakage and repeated CT of the thorax showed a resolving mediastinal haematoma. She was discharged on completion of a 6-week course of antibiotics. Two months later she was re-admitted to the general hospital with osteomyelitis of her left femur. Routine repeated upper endoscopy disclosed that the aortic Dacron graft had eroded through the oesophageal wall. She underwent thoracoscopic oesophagectomy and drainage of the left femur with multiple drill holes. Six weeks later, she was re-admitted to our hospital and underwent extra-anatomical bypass grafting from the ascending to descending aorta via a median sternotomy followed immediately by a repeated left thoracotomy and excision of the infected thoracic aortic Dacron graft. Continuity of the alimentary tract was restored by end-to-end oesophago-gastrostomy via a right thoracotomy 3 months later. The patient recovered uneventfully.

Discussion

In addition to an FB retained in the oesophagus, predisposing causes of AEF include aneurysms of the thoracic aorta, congenital thoracic aortic anomalies, thoracic malignancy, mediastinal infection, oesophagogastric reflux, corrosive exposure, and prior thoracic surgical intervention. In a review of 2394 cases of FB in the oesophagus,[1] only two cases of AEF were identified. Until 2004, only nine survivors of FB-induced AEF have been reported.[4] There is a general consensus on the pathogenesis, clinical features, and prognosis of AEF. Nevertheless, the optimal management of FB-induced AEF remains controversial.

On reviewing the literature, there are different management strategies available to formulate a theoretically sound approach to this potentially fatal clinical problem. All individuals suspected of having an FB in the oesophagus should undergo upper endoscopy/oesophagoscopy with or without retrieval of the FB. If there is evidence of oesophageal injury, the individual should be kept fasted, commence parenteral/nasogastric tube feeding, and be prescribed intravenous broad-spectrum antibiotics until repeated endoscopy shows satisfactory healing. In retrospect, if case 3 had been subjected to a very strict regimen of parenteral nutrition and a full course of intravenous antibiotics, the mediastinal infection and AEF probably would not have developed. On discharge, the individual should be informed of the possibility of AEF and its clinical features. Patients should be instructed to seek immediate medical advice if they experience persistent or recurrent retrosternal pain/dysphagia or become febrile.

Attending surgeons should remain alert to the possibility of AEF developing in an individual with a history of ingested FB, irrespective of whether it has been retrieved. When an evolving AEF (oesophageal ulcer with or without submucosal haematoma) is diagnosed, some precautions should be taken to prevent exsanguinating haemorrhage prior to definitive surgical intervention: endoscopic application of haemoclips,[5] injection of fibrin sealant,[6] or insertion of a Sengstaken-Blakemore tube.[3] If one of these measures had been taken in cases 1 and 4, both patients might have survived.

The descending aorta is usually best approached through a left thoracotomy but in rare cases involving the right aortic arch and right descending aorta, a right thoracotomy may be a better alternative. Endovascular stent grafting[6] is a feasible alternative to open repair in a high-risk patient. If aortic repair is expected to be difficult—for example, a pseudoaneurysm is present—prior insertion of a temporary Gott shunt as in case 5 is a preferred option. In extreme cases, hypothermic circulatory arrest can be life-saving.[7] There are two options for aortic repair: wedge excision of the involved aortic wall and primary repair or segmental aortic resection plus auto-topic interpositional Dacron grafting/extra-anatomical bypass grafting. The decision rests on the extent of aortic involvement, the severity of infective mediastinitis if present, and the experience of the attending surgeon. Similarly, oesophageal repair can vary from primary repair to oesophagectomy, or cervical oesophagostomy plus staged restoration of continuity of the proximal alimentary tract as in cases 3 and 5, respectively. Potential or established mediastinal infection should be controlled with broad-spectrum antibiotics and drainage, with or without daily topical irrigation.[6]

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