Case report

Successful treatment of persistent aortoesophageal fistula after thoracic endovascular aortic repair (TEVAR) with esophageal bypass surgery: a case report

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Abstract This study reported a case of persistent aortoesophageal fistula after successful thoracic intravascular aneurysmorrhaphy. An esophageal bypass operation by gastric pull up procedure was performed in this case with a successful outcome. Chiang Mai Med J 2015;54(3):139-45.

keywords: Thoracic aortic aneurysm, esophageal perforation, thoracic aortic stent, aortoesophageal fistula, surgical technique

Background

Aortoesophageal fistula is a rare but devastating complication of thoracic aortic aneurysm[1,2]. Nowadays, there is still no consensus regarding a treatment strategy for this condition, especially in this era of endovascular surgery, which is currently spreading over the Southeast Asian region. Previously, thoracic aortic aneurysm could be managed by thoracotomy that repaired the aneurysm and closed the fistula; however, this method resulted in a very high morbidity and mortality rate. After the endovascular technique developed and became standard treatment, it showed superior results, with lower early post operative morbidity and mortality[3,4]. The 30 day-mortality from thoracic endovascular aortic repair (TEVAR) vs open repair was 3.5% vs 9.4% and morbidity 16% vs 68%, accordingly[5]. After TEVAR had become a standard technique, the troublesome complication of aortoesophageal fistula was reported recently. Persistent aortoesophageal fistula was found, despite the thoracic aortic aneurysm being managed by thoracic intravascular aneurysmorrhaphy (TIVA), and the fistula between the vascular sac and esophagus had not healed completely. This problem occurred in about 2% of patients after successful TIVA procedure[6]. Despite the low incidence of persistent fistula, it inflicted an almost 100% morbidity rate, as reported by Eggebrecht et al. They reported that patients who developed persistent fistula often died; even if they were treated by a different strategy[7]. This study reported the successful management of persistent aortoesophageal fistula with esophageal bypass surgery.
Case presentation

A 60-year-old man presented with hematemesis. His vital signs were Temperature = 36 degrees Celsius, Pulse rate = 98 beats per minute, Blood pressure = 149/87 mmHg, Respiratory rate = 16 breaths per minute and Oxygen saturation = 100%. He was examined by endoscopy and a bleeding area was found at the mid thoracic esophagus. The point of bleeding resembled a tunica intima, in which intravascular plaque was visible. A computed tomography (CT)-scan was carried out, which showed a large thoracic aortic aneurysm from the T-5 to T-7 level. A hematoma occupied almost the entire area of the posterior mediastinum, where the esophagus was indistinguishable from the hematoma. As the general condition and vital signs of the patient were stable, it was decided to use the endovascular technique, due to accessible vascular anatomy. The first CT-scan is shown in Figure 1.

The procedure was initiated under general anesthesia. An arterial line and central venous pressure were placed, before the guide wire and endovascular stent (Medtronic, Valiant type, proximal =36 mm, distal =32 mm, length = 160 mm) were passed through the right femoral artery, which was accessed by an open technique. The procedure was carried out under real time fluoroscopic guidance. After the stent was placed successfully and the appropriate stent position confirmed, no immediate post operative complication occurred.

However, after the first session of TIVA, the patient still experienced recurrent hematemesis on the second postoperative day. He vomited 200 mL of fresh blood and complained of back pain. His vital signs were Temperature = 37.1 degrees Celsius, Pulse rate = 70 beats per minute, Blood pressure = 160/60 mmHg, Respiratory rate = 16 breaths per minute and Oxygen saturation = 100%. A repeated CT-scan identified a possible endovascular leakage. A follow-up CT-scan showed a type II endoleak and the patient was taken back to the endovascular suite immediately. The CT-scan that demonstrated the endoleak is shown in Figure 2. After the second endovascular stent was deployed, a further CT-scan revealed no leak-

Figure 1. CT scans of a thoracic aortic aneurysm showing an aortoesophageal fistula with contrast leakage to the esophagus. The arrow identifies an aortoesophageal fistula. E: esophagus, T: thoracic aorta.
age. All aortoesophageal patients were nil per os (NPO) routinely for two weeks, and then evaluated by CT-scan and esophagoscopy to confirm that the esophagus had healed. During this period, patient nutrition was managed with enteric access via jejunostomy feedings, which were inserted via the open technique three days after the second endovascular session, and supplemented with partial parenteral nutrition, as needed. The patient also was encouraged to expectorate all saliva.

After the two weeks of no oral feeding, an endoscopic examination showed that the aortoesophageal fistula persisted, although the endovascular stent was visible through esophagoscopy. The finding from esophagoscopy is shown in Figure 3. With the persistent fistula being found, esophageal bypass procedure was scheduled and performed with a gastric conduit, which resulted in remaining esophageal diverticulum.

The operation began with an exploratory laparotomy in order to prepare the gastric conduit, and the right gastric and right gastroepiploic vessels were preserved. The lesser curve of the stomach was divided with a linear stapler...
before a retrosternal passage was created. An incision in the anterior was then made to the left of the sternocleidomastoid muscle, followed by dissection until the esophagus was encircled. The gastric conduit was anastomosed to the proximal esophagus after it has passed through the retrosternal plane. The complete operation was uneventful, with no postoperative complication. The operative time was three hours and the endotracheal tube was removed on the second post operative day. The patient was admitted to the intensive care unit (ICU) for three days and resumed an oral diet on the ninth post operative day. He was discharged on the 14th day post operation. The length of hospital stay was 78 days. Details of the operative procedure are shown in Figure 4.

After the procedure, the patient was followed-up closely to detect any mediastinal infection, due to remnant of the esophageal diverticulum being left in place. After a six-month follow-up period, the patient showed no signs of serious infection. CT-scans were performed at three and six month follow-up evaluations and no remnant of air in the vascular sac or around the endovascular stent was observed. The follow-up CT-scans are shown in Figure 5.

Discussion

Aortoesophageal fistula can result from various causes, including foreign body ingestion, ruptured thoracic aortic aneurysm or esophageal disease, and it creates a difficult medical condition that currently has no consensus on management. It also requires special medical attention for the following reasons:

1. Very high rate of bleeding from the thoracic aorta, and bleeding without resuscitation could lead to high grade shock and organ failure in only a few minutes.
2. The thoracic aorta is a very dangerous and difficult area to access because it is located close to many vital organs.
3. There is a high percentage of morbidity and mortality, including stroke, renal failure and paraplegia[1,2].

After emergence of the endovascular technique, the benefits of minimally invasive operations outweigh open thoracotomy surgery. There is less operative time, and early postoperative morbidity and mortality are decreased dramatically. Nowadays, the endovascular technique is preferable for thoracic aortic aneurysm if there is no specific contraindication in each case[3,4].

However, after the endovascular technique became popular, the authors discovered that a fistula between the esophageal lumen and
Thoracic aortic aneurysmal sac does not heal spontaneously in approximately 2% of patients\[6\]. This group of persistent aortoesophageal fistulas is very problematic, and almost all patients with this problem consequently die from its complications, as sepsis and organ failure occur after repeated infections. Attempts at open thoracotomy in order to close the fistula almost always fail, due to the fragility of the patient and complexity of the procedure. Successful treatment has been reported sporadically, with no consensus on treatment guidelines\[6,8-10\].

Kubota et al reported that aortoesophageal fistula patients, who underwent TEVAR alone, had no chance of survival. That author reported four TEVAR cases in which three patients underwent additional esophagectomy and reconstruction with gastric conduit and the other patient, who did not receive additional procedure, died due to graft infection. Of the three patients who received additional procedure, only one survived to the one-year follow-up, while another one died of graft infection, sepsis and aortobronchial fistula. This study could not summarize that esophagectomy is a protective procedure against graft infection\[11\], and another choice of conduit in reconstructive surgery has been proposed. Kuroki et al reported using the colon as a conduit and they achieved a good result. However, this operation required two more anastomoses and caused longer operative time. Using a gastric conduit would be a better choice if the stomach were suitable for the operation\[12\].

After a multidisciplinary team discussed the patient of this study, a staged operation was selected to manage the fistula. The primary procedure of a gastric pull up to bypass the fistula site was considered to be the most beneficial for the patient, as it protects against aortic stent contamination, which is the most common cause of mortality in this group of patients. The esophagus was divided close to the fistula site via the trans hiatal route, without a standard thoracotomy. This procedure resulted in approximately 10 cm of esophageal diverticular remnant (proximal 10 cm and distal five cm of excised esophagus). There was still concern about infection within the remnant of the diverticulum. However, this procedure was selected first, with a thoracotomy reserved in case infection persisted. Fortunately, the patient was free from infection and recovered very well. He was still doing well at the 1 year 3 month follow-up.

Earlier, this institute had often performed a routine Erkin’s tube esphagogastrostomy as protection immediately after the endovascular procedure. However, recent cases possessed quite
a large hematoma and this procedure does not benefit the patient because a persistent fistula still occurs. Also performing the diversion causes a problem for further gastric pull up bypass procedures, because the diversion creates a scar that makes the bypass operation more difficult.

Conclusion

This study proposed the gastric pull up operation as a rescue procedure for persistent aortoesophageal fistula. A thoracotomy to resect the esophageal diverticula remnant should be utilized only when infection persists. An early esophageal diversion may not change the course of the disease and may result in much more complicated reconstructive surgery.

References

การรักษาผู้ป่วยภาวะที่มีรูว่างระหว่างหลอดเลือดแดงใหญ่ทรงกับหลอดอาหารที่เกิดภายหลังการใส่หลอดเลือดเทียมในหลอดเลือดทรงได้ด้วยเทคนิกลูกหลาม ด้วยการผ่าตัดสร้างทางเบี่ยงของหลอดอาหาร : รายงานผู้ป่วย

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ภาควิชาศัลยศาสตร์ คณะแพทยศาสตร์ มหาวิทยาลัยเชียงใหม่

ผู้นิพนธ์รายงานผู้ป่วยที่มีรูว่างระหว่างหลอดเลือดแดงใหญ่ทรงกับหลอดอาหาร ที่เกิดขึ้นภายหลังการรักษาภาวะหลอดเลือดใหญ่ทรงโป่งพองด้วยเทคนิคการใส่สายสวนหลอดเลือดผู้ป่วยได้รับการผ่าตัดทางเบี่ยงหลอดอาหารด้วยเทคนิคการใช้กระเพาะเป็นทางเดินอาหารแทน ซึ่งได้ผลลัพธ์การรักษาที่ดีเชิงメディซิน 2558;54(3):139-45.

คำสำคัญ: หลอดเลือดใหญ่ทรงโป่งพอง หลอดอาหารทะลุ ซิลิโคนหลอดเลือดใหญ่ทรงก แผลชอนทะลุระหว่างหลอดเลือดใหญ่และหลอดอาหาร วิธีการผ่าตัด