CASE REPORT

Aortobronchial fistula after esophagectomy for esophageal cancer—A very rare complication

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Abstract Most aorto-respiratory fistulas are related to aortic pathology or procedures, but fistula formation after esophageal resection has never been reported in the literature. We are now reporting a case of hemoptysis that occurred after esophagectomy for locally advanced esophageal cancer. Aortobronchial fistula was detected by computed tomography scan. The patient was finally saved by emergency surgery—Dacron graft interposition of the descending thoracic aorta. There was no malignant cell in the postoperative specimen of the fistula. The erosion of the ligaclips (Johnson & Johnson) might be responsible for the aortobronchial fistula formation. For esophageal surgery, avoidance of trauma to aortic wall and careful using of ligaclips are important to circumvent this complication.

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Case presentation

A 60-year-old man suffering from middle-third thoracic esophageal squamous cell carcinoma received transthoracic esophagectomy and gastric tube reconstruction with cervical esophagostomy after a thorough workup under the clinical stage of T3N1M0. Dense adhesions between the esophagus and the descending aorta were found during the procedure. The entire operative and postoperative course was uneventful except for very minimal cervical anastomotic leakage. The pathology of the resected specimen revealed that the esophageal cancer had extended to the adventitia layer but not to the adjacent organ; 4 out of 17 dissected lymph nodes had cancer cell metastasis. The esophageal cancer stage American Joint Committee on Cancer 7th edition was T3N2M0, Stage IIIB. He was in good condition, and his oral intake was unremarkable at home after discharge. Only mild chest tightness with occasional radiation to the back was sometimes noted. The adjuvant radiotherapy and chemotherapy were scheduled 2 months later after surgery.

About 2 months after discharge, seemingly aggravating hemoptysis was diagnosed before his scheduled chemoradiotherapy. The chest computed tomography (CT) scan revealed a pseudoaneurysm arising over the descending aorta with a fistula connecting the left main bronchus (Figs. 1 and 2). For further confirmation, emergency bronchoscopy was performed, which revealed a mucosal defect coating with a blood clot in the middle of the left main bronchus (Fig. 3).

Under the assumption of aortobronchial fistula, emergency left thoracotomy was performed. After establishing cardiopulmonary bypass through femoral access, dense adhesions between the aorta and the left main bronchus were carefully dissected. After proximal and distal aortic clamping, aortotomy was performed. A 1.5-cm perforation over the intima, which fistulized into the main bronchus, was noted. A 7-cm Dacron graft interposition for descending thoracic aorta was the procedure for the pseudoaneurysm. Direct repair or resection of the perforated bronchus was not done because of the dense adhesion around left mainstem bronchus and lung parenchyma. A pedicle intercostal muscle flap was placed in the fistula tract from aortic end instead to obliterate the fistula and separate the Dacron graft and the left main bronchus. Securing the muscle circumferentially on the adjacent aortic wall and covering the Dacron graft by remainder aortic wall was the finishing touch. The patient recovered uneventfully with the absence of hemoptysis. Bronchoscopy 2 months later showed a complete resolution.
healing of mucosa. No fistula with the Dacron graft in excellent position was detected through CT scan 5 months later.

Discussion

The development of aortobronchial fistula after esophagectomy for esophageal cancer is extremely rare. To the authors' knowledge, it has never been reported in the literature. Aortoesophageal fistula is not uncommon and occurs when the tumor invades the aortic wall, especially after chemoradiotherapy [1]. In our case, the cancer invaded the longitudinal muscle layer and extended to the adventitia; that is, the aortic wall was free of cancer and certainly would not have been damaged during the esophagectomy. However, as we reviewed the enhanced chest CT scan in detail, a ligaclip was found just at the base of the pseudoaneurysm (Fig. 2). It is reasonable to suspect that the aortic wall was gradually eroded by the vascular clip during the 2-month postoperative period, resulting in pseudoaneurysm. As the pseudoaneurysm grew, it perforated into the bronchus, causing hemoptysis. This process would require a long latent period after the esophagectomy, and it fits the clinical progression of this patient. From the surgical oncologic point of view, dissection for en bloc esophagectomy should be performed from the descending aortic adventitia and left main bronchus directly and sharply. However, it is possible that the ligaclip was applied in an angle against aorta, which induced aortic wall injury. If the injury progressed, an iatrogenic pseudoaneurysm might arise from this weak point. As the growing pseudoaneurysm invaded the bronchus, the aortobronchial fistula was formed. To prevent aortobronchial fistula, meticulous dissection, avoidance of any injury to all adjacent structure, and parallel application of ligaclip to aorta to control all esophageal branches are the essential points.

Early locoregional recurrence of cancer is another explanation; however, its possibility is very scarce as (1) no macroscopic residual tumor was noted during esophagectomy; (2) there was no microscopic cancer cell invasion beyond the adventitia; and (3) no cancerous cells were found in the specimen of aortic operation.

To diagnose such a fistula requires a substantial clinical history and vigilance [2]. However, a history of descending aortic disease and aortic surgery was absent in this patient. Also, there were no obvious findings in the chest radiograph—no mediastinal widening or infiltration—hence, chest CT scan became imperative in this scenario. In patients with esophageal cancer, the locoregional recurrence in the first year is around 40% [3]. Hemoptysis is usually the first sign [4]. Combined chest CT scan with CT–angiography would provide a wide range of intrathoracic evaluation and reveal aortic abnormality [5]. Tumor recurrence, consolidation, or hemorrhage of pulmonary parenchyma, aneurysm, or aortobronchial fistula would be clearly identified.

Moreover, bronchofiberscopy is necessary to rule out the possibility of tumor recurrence. On the other hand, it would be fatal if the orifice of the fistula is occluded by clot and dislodged during the procedure [6]. It would be better to perform the bronchofiberscopy inside the operating room just before the thoracotomy to ensure that the anesthetist is well prepared to establish an emergency airway any time. Double-lumen endotracheal tube should be used for anesthesia, and we chose a left side one. To prevent possible injury to the fistula by the endotracheal tube, we introduced the tube through bronchofiberscopic guidance. The cuff of left side double-lumen tube can also compress the fistula orifice and stop further blood leaking into the airways.

The surgical procedure consists of two parts, the aorta and the bronchus. Cardiopulmonary bypass through femoral cannulation is mandatory. In the review of Piciche et al. [7], various procedures can be considered: 32.3% chose graft interposition, 15.2% chose primary repair, and 11.2% used the extra-anatomic bypass graft. Because of the relatively large defect of the aortic wall after adequate debridement, repairing it with a patch was not practical. We decided to use in situ graft interposition during the operation for several reasons. First, compared with extra-anatomic bypass graft, the in situ graft has a better hemodynamic effect. Another reason was that we could anastomose both ends of the Dacron graft in a normal aorta structure after adequate debridement. The length of the Dacron graft was about 7 cm, located between the levels of T6 and T9. About 75% of the artery of Adamkiewicz arises between T9 and T11; hence, the effect of our graft on this artery was relatively acceptable. On the other hand, if infection occurs unfortunately, we can arrange another operation for extra-anatomical bypass graft. For the bronchus, if the fistula is related to the locoregion of the cancer, repair of the bronchus would be difficult if its resection is to be avoided. If it is caused by nonmalignancy or trauma, pedicle flap repair or wrapping of the bronchus is preferred [8,9], and vascularized flap has better resistance to infection. Recurrence of fistula can be prevented by the interposition of the vascularized tissue.

As the development of aortic endovascular stent, surgical intervention for aortobronchial fistula could be challenged. Compared with surgery, endovascular stenting procedure is easier, less invasive, and prevents from cardiopulmonary bypass [10]. The success rate is higher and mortality rate is lower. Certainly, complications, such as migration [11], infection [12], and blockade of spinal arteries [13], are still likely. The experiences of endovascular stem graft are scarce, however, and only 15 cases have been reported. Moreover, the long-term follow-up data are limited. Piciche et al. [2] suggest stent graft for aortobronchial fistula involving descending aorta because of higher mortality of surgical repair. Some authors thought that it could be used in the initial insult of bleeding followed by staged surgical repair [14]. The endovascular repair is thought to be an alternative option for aortobronchial fistula, and there is no clear indication right now.

In conclusion, this complication after esophagectomy should always be kept in mind. Chest CT scan and bronchofiberscopy are the necessary means for diagnosis. Emergency aortic procedure under cardiopulmonary bypass or endovascular stenting is the recommended course of action to ensure preservation of life.
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References