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Esophageal Left Subclavian Artery Fistula Caused by The Esophageal Foreign Body: A Case Report

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ABSTRACT

Left subclavian artery esophageal fistula usually occurs after esophageal cancer surgery, which is a rare complication, and it is even rarer after stent implantation of left subclavian artery pseudo-aneurysm. This paper reports the case of a 21-year-old male patient with left subclavian artery pseudo-aneurysm. Two-plus months after stent implantation, he stopped anticoagulant and antiplatelet drugs and developed pain in his left upper limb. The patient was diagnosed with arterial fistula. He was discharged from the hospital successfully after several operations, such as thoracic aortic stent implantation, left common carotid artery left axillary artery artificial vascular bypass.

Conclusion: Early diagnosis and positive treatment lead to a good prognosis for patients with esophageal left subclavian artery fistula.

INTRODUCTION

Esophageal left subclavian artery fistula is a rare and critical disease in clinical practice. The main reason for its occurrence is that the rupture of the esophagus leads to inflammatory infiltration of surrounding tissues, forming an esophageal fistula and connecting the left subclavian artery [Zhao 2019]. Once the disease is formed, there is a risk of fatal massive hemorrhage at any time, and the mortality is very high.

CASE REPORT

A 21-year-old young male was admitted to the respiratory department of our hospital on April 19, 2020, due to "pain in the left upper limb for 1 month and hemoptysis for 1 day." Physical examination on admission showed shallow and fast breathing, a little moist rale in the left upper lung, and limited movement of the left upper limb, due to pain. His HRCT examination showed a large irregular mass in the left upper mediastinum, an unclear boundary with the left subclavian artery, and a high possibility of an aneurysm with hematoma (Figure 1A and 1B). (Figure 1)

The patient was transferred to ICU and the cardiothoracic surgery department for treatment. The patient had a history of fishbone obstruction 20 days before admission. According to the patient's history of fishbone entrapment and the signs of pain and hemoptysis in the left upper limb on admission, the imaging data suggested there might have been an aneurysm with mediastinal hematoma. Our department considered that there could be a causal relationship between these four factors. Next, we preliminarily summarized and considered that the patient may have had an esophagus rupture after swallowing the fishbone. The patient developed inflammation, invasion of the left subclavian artery, local pseudo-aneurysm, affecting the blood supply of the left upper arm, pain in the left upper limb,

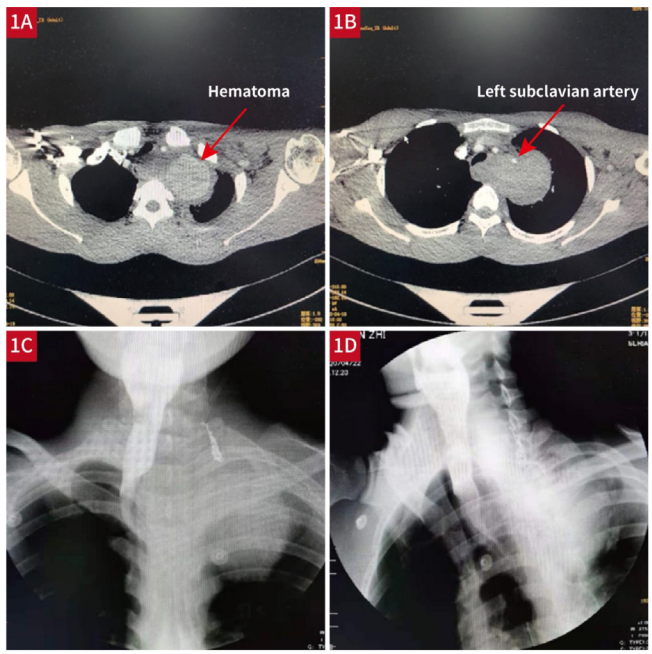

Figure 1. (A)(B) Large irregular mass in the left upper mediastinum with unclear boundary with the left subclavian artery. Considering the possibility of aneurysm with hematoma. (C)(D) No definite signs of esophageal fistula were found after improved upper gastrointestinal radiography.

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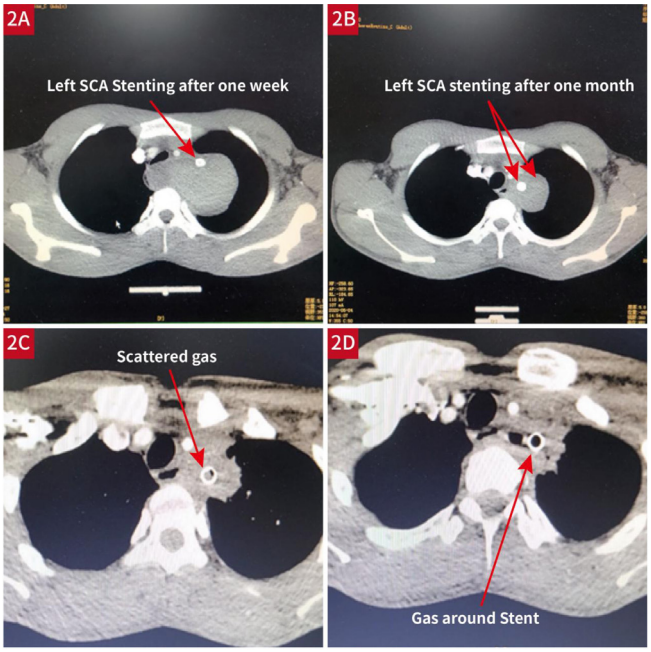

Figure 2. (A) One week after the left subclavian artery covered stent, the enhanced CT showed that the left subclavian artery was unobstructed and the mass volume was slightly absorbed. (B) Re-examination of the left subclavian artery with stent graft for more than one month after the operation with enhanced CT showed that the left subclavian artery was usually, and the mass was absorbed more obviously than before. (C)(D) After the left subclavian artery stent for more than two months, the patient came to the hospital for reexamination due to discomfort. Enhanced CT showed there was no contrast agent in the lumen, embolization is possible, scattered air in and around the stent, left upper mediastinum. Irregular mass shadows are smaller than on June 4, 2020.

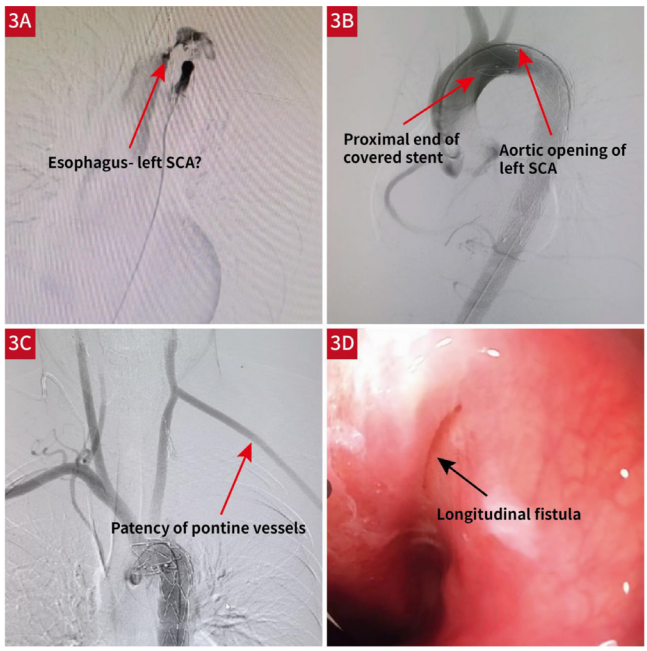

Figure 3. (A) Angiography through the left subclavian artery showed no contrast medium passed through the left subclavian artery, and the contrast medium overflowed in the overlapping area of the stent and flowed into the esophagus. (B) Thoracic aortic covered stent placement through the femoral artery. The stent covered the proximal end of the left subclavian artery to prevent blood flow from passing through the fistula and save the patient's life. (C) After left common carotid artery left axillary artery artificial vascular bypass grafting, angiography showed that the bridging vessel was unobstructed. (D) Gastroscope shows a longitudinal fistula with a width of 2mm and a length of about 1cm at 24cm from the incisor, confirming the esophageal fistula.

and hemoptysis. In order to clarify the esophageal rupture, the gastroenterology department arranged gastroscopy, but the family refused gastroscopy and agreed to conduct upper gastrointestinal radiography. There was no definite sign of esophageal fistula after upper gastrointestinal angiography (Figure 1C and 1D).

After angiography, we found the initial segment of the left subclavian artery obviously was dilated, and there was no obvious contrast agent spillover. After the catheter was inserted into the right vertebral artery again, the contrast agent retrogradely entered the aneurysm cavity through the left vertebral artery, and the distal part of the left subclavian artery was visualized. We conducted a multidisciplinary discussion and finally discussed two options: 1. Left vertebral artery embolization + left subclavian artery covered stent implantation; 2. Left subclavian artery, vertebral artery embolization + left common carotid artery left subclavian artery bypass + mediastinal tumor resection. Compared with the two schemes, the second scheme had a higher risk, and the whole operation needed to be carried out under general anesthesia. But it was difficult to intubate the patient's airway under pressure, and the risk of bleeding was higher. However, the first scheme could be carried out under local anesthesia without reconstructing the blood supply of the left upper limb, and the operation relatively was simple. After communicating with the patient's family members, the first scheme finally was chosen. On April 21, 2020, the left vertebral artery embolization and left subclavian artery-covered stent implantation successfully were completed under local anesthesia. The next day, the patient was transferred back to the general ward for further treatment. Because the upper gastrointestinal radiography did not show the signs of esophageal fistula before, the patient gradually began to resume diet after being transferred to our department. On April 28, 2020, a re-examination of HRCT showed the left subclavian artery was unobstructed, and the mass volume was slightly absorbed (Figure 2A). (Figure 2)

On May 9, 2020, the patient was discharged and ordered to take oral antiplatelet drugs. On June 4, 2020, another follow-up visit was conducted in our outpatient department. HRCT showed that the left subclavian artery had obvious mass absorption (Figure 2B).

At the time, we expected the patient would continue to recover, but things didn't develop that way. On June 30, 2020, the patient was admitted to the hospital again because of "fever for 7 days, with left upper limb pain for 3 days."

Physical examination at admission: t 38.00C, P 105/min, R 20/min, BP 115/68mmhg; the cardiopulmonary examination was negative, left upper limb movement was limited due to pain, no swelling, and local skin temperature was slightly higher. Auxiliary examination on admission: blood routine: WBC 14.27 * 10 ^ 8/L, n% 80.7%, Hb 123g/L, PCT 3.97mg/ml; other biochemical, coagulation and heart damage indexes were normal. The patient stopped taking antiplatelet drugs three weeks before admission. After admission, HRCT showed that there was no contrast agent in the lumen of the left subclavian artery after stent implantation, and embolization was possible. There was scattered gas accumulation in and around the stent, and the irregular mass shadow in the left upper mediastinum was smaller than on June 4, 2020. Because there still was scattered gas in and around the stent, the possibility of bacterial embolus was high (Figure 2C and 2D).

The patient's condition was aggravated, and the left radial artery pulsation was weakened on July 1, 2020, so we considered the need for surgical treatment. On July 2, 2020, we planned to perform a left common carotid artery left subclavian artery bypass under general anesthesia. However, repeated exploration during the operation failed to detect the left subclavian artery. We considered that the left subclavian artery embolism caused distal occlusion, which could not be detected. For further diagnosis and treatment, interventional angiography immediately was arranged, and the feasibility of thrombectomy was evaluated. However, after angiography, no contrast medium was found in the left subclavian artery, and the contrast medium overflowed into the esophagus in the stent overlap area. Esophageal left subclavian artery fistula was considered. The risk was very high, and the patient's life was in danger (Figure 3A). (Figure 3)

We immediately came up with two options: 1. Surgical thoracotomy exploration. But at the time, we considered the possibility of pus embolus in the left subclavian artery, peripheral inflammatory invasion, tissue edema and decay, so the risk of bleeding was very high and it would not be easy to stop bleeding during thoracotomy exploration; 2. Thoracic aortic-covered stent implantation via the femoral artery, which would block the proximal end of the left subclavian artery and prevent blood flow through the fistula, to rescue the blood supply of the left upper limb. Color Doppler ultrasound showed there was no blood flow signal in the left brachial artery, and collateral circulation could be seen around the lumen. At the same time, the cubital fossa was about 2 * 3cm in size, and the possibility of abscess in the liquid dark area was high. Although there was collateral circulation, the risk of left upper limb necrosis could exist. After communicating with family members, we considered saving his life first, so we chose the second plan. At the same time, incision and drainage of cubital fossa and extraction of pus were performed. The whole process was smooth, and the patient returned to ICU after the operation (Figure 3B).

Next, we started to think about restoring blood supply to the left upper limb. We also considered how to deal with a mediastinal abscess and whether there was any foreign matter. MDT was started again. The results showed the blood supply should be reconstructed as soon as possible to save the left upper limb, the risk of bleeding of mediastinal abscess should be reduced under the protection of a covered stent, and the abscess should be removed. In addition, attention should be paid to finding out whether there were foreign bodies while removing the abscess. On July 3, 2020, according to the results of the MDT discussion, left common carotid artery left axillary artery bypass grafting + mediastinal infection debridement were performed under general anesthesia. The procedure was smooth, and no foreign body was detected. The artificial blood vessel can be seen by angiography after the operation, and the blood supply successfully was established (Figure 3C).

Because the patient with esophageal left subclavian artery fistula needed long-term fasting and drinking prohibition, our department considered asking the Department of Gastroenterology to place a duodenal nutrition tube under a gastroscope for nasogastric enteral nutrition. Intraoperative gastroscopy showed there was a longitudinal fistula with a width of 2mm and a length of 1cm 24cm away from the incisors, which confirmed the esophageal fistula (Figure 3D).

The results of pus culture obtained at the left cubital fossa also showed that he had a Streptococcus pharyngitis infection. The Streptococcus pharyngitis came from the oropharynx, confirming the diagnosis of esophagus left subclavian artery fistula. At the time, the diagnosis was confirmed, the blood supply of the upper limb was recovered, the blood flow of the left subclavian artery had been blocked, and the esophageal fistula forbade the patient to eat and drink. We thought the condition gradually would improve. However, on July 23, 2020, the patient had hemoptysis again, aggravated on July 26. Hemochromatin collapsed, a large amount of bright red bloody fluid was drained from the mediastinal drainage tube, and there was continuous fresh blood exudation around the drainage tube. Considering the active bleeding, the patient immediately was transferred to ICU. Pulmonary artery CTA was reexamined, and a patchy, dense shadow was found around the left subclavian artery and aortic arch. There also was a patchy, increased-density shadow found in adjacent lung tissue. For high-density shadow, our department considered that the patient's condition was aggravated, and the intrapulmonary hemorrhage was obvious. So, we asked the interventional department to clarify the responsible pulmonary vessels, but the interventional department considered the bronchial artery embolization could not be performed after aortic stent implantation (Figure 4A). (Figure 4)

We were puzzled by the sudden appearance of hemoptysis. We tried to get to the bottom of this and inquired about medical history. The patient told us that around July 21, 2020, he ate biscuits and milk after we repeatedly told him not to eat or drink. Our department considered the reason for the aggravation of infection may have been caused by diet, leading to further tissue edema, causing endoleak, or the possibility of infection-corroding blood vessels, so we started MDT again. After the final discussion, we decided to carry out aortic arch replacement + descending aorta replacement + thoracic aorta covered stent removal + esophageal exclusion + jejunostomy under general anesthesia.

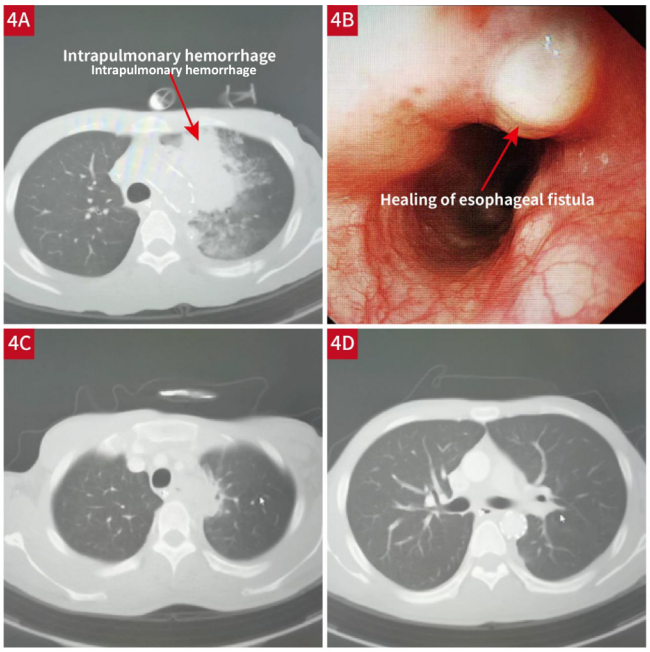

Figure 4. (A) CTA of pulmonary artery showed there was mass density increasing shadows around the left subclavian artery and above the aortic arch, patches of slightly high-density shadows in adjacent lung tissues, and obvious intrapulmonary hemorrhage. (B) On August 28, 2020, the gastroscope showed local mucosal changes in the esophagus, considering the healing performance of the esophageal fistula. (C)(D) On August 31, 2020, chest-enhanced CT showed the scope of pseudoaneurysm around the proximal end of the left subclavian artery was significantly reduced compared with before.

However, the risk of this operation was huge, and we also were faced with a mediastinal infection. The operation was several times more dangerous than the previous ones. After repeated communication with the patient's family, his family members gave up on the operation and chose conservative treatment. Fortunately, after symptomatic hemostasis and blood transfusion treatment, the patient's condition gradually stabilized. On August 3, 2020, the patient was transferred back to the general ward of our department. Because of the previous bleeding, the anticoagulant and antiplatelet drugs were stopped, and they're still was the risk of embolism. After returning to the general ward of our department, the patient's condition was stable, and he continued to recover. On August 28, 2020, gastroscopy showed that the local esophageal mucosa changed, and the healing performance of the esophageal fistula was considered (Figure 4B).

On August 31, 2020, chest enhanced CT showed that the range of pseudoaneurysm around the proximal left subclavian artery was significantly reduced (Figure 4C, 4D). On September 28, 2020, gastroscopy again showed the local granulation tissue hyperplasia of the esophagus. We gradually had the patient resume diet, and reexamination a week later showed no obvious abnormalities. The patient was smoothly discharged.

DISCUSSION

Pathogenesis: Through the treatment of this case and reference to related literature, we believe the pathogenesis of this type of disease is foreign bodies cause esophageal injury, perforation, or puncture of blood vessels to form internal fistulas; at this time, fasting and drinking are not strictly performed and digestive juice overflows, erosion, and foreign bodies remain [Stringari 2013]. It can cause inflammation, edema and even infection of surrounding tissues, such as the esophagus, mediastinum, and large blood vessels. Some may form localized abscesses, while others may cause systemic infections to worsen the condition. Due to the high pressure of the thoracic aorta, the high-pressure blood enters through the fistula, at low pressure, repeated hemoptysis, hematemesis, thrombosis, hematoma, pseudoaneurysm, etc. occur [Morisaki 2014]. Once the foreign body or thrombus falls off and the hematoma or pseudotumor ruptures, it can cause fatal bleeding. After treatment of this case, we concluded this type of disease may have "signal bleeding" in the early post-injury period, and esophageal injuries, internal fistulas, foreign bodies, infections, and arterial hypertension bleeding can affect each other and make the condition worse. It can also affect the outcome of the disease, due to the degree of injury, location of the fistula, size, and order of treatment [Uno 2017].

Principles of treatment: By consulting the relevant literature, we know that once the esophagus-thoracic large vascular fistula occurs, it is extremely dangerous and may cause death due to hemorrhage at any time, with a mortality rate of 97.2%. In recent years, many scholars believe that emergency surgery in thoracic surgery should be the main treatment method. According to the treatment of this case, we believe that the principle of treatment of this type of disease should be based on saving lives, and the key to surgery is to control blood flow and prevent digestive juice leaks, handle the aorta and esophageal fistula, thoroughly debride, remove foreign bodies, and control infection [Al-Thani 2021].

Indications for surgery: We believe that such diseases usually have a clear history of foreign bodies and corresponding clinical symptoms. If there is a history of foreign bodies, patients start to have “bleeding.” If there is a shunt or a cotton ball phenomenon, it will indicate an esophageal fistula. This case was admitted to the hospital for the first time and suspected of esophageal injury, but considering the high risk, a gastroscopy was not performed. The upper gastrointestinal angiography showed no clear signs of esophageal fistula [Georvasili 2016]. After the second admission, the condition was critical, and the risk of gastroscopy confirmed the esophageal fistula. But at the time, the upper gastrointestinal angiography still showed no exact esophageal fistula, so perhaps the patient’s esophagus was damaged the first time but it was not diagnosed. At the same time, many domestic and foreign documents also suggest that upper gastrointestinal angiography has false negatives in the diagnosis of esophageal fistula, so we believe that we should first choose esophageal gastroscopy in the diagnosis of esophageal fistula [Sladojevic 2017].

Treatment of arterial fistula: We mentioned in the

treatment principle that the most important thing for this type of disease is to control bleeding first, with the main purpose of saving lives. You can first pass the strapping method at both ends of the fistula, the blocking forceps method, and the trapezoidal non-damage forceps side of the fistula. The wall clamp method and other methods control bleeding and fully expose the visual field. Do the following methods to completely stop bleeding:

(1) Ligation method. We can choose to ligate the corresponding artery of the disease, according to the patient's condition. After the second admission of this case, we chose a safer way to block the blood flow of the left subclavian artery by implanting the aortic stent to save lives. However, we must be cautious when choosing such methods because the rate of disabling amputation or brain disease (including mortality) after vascular ligation is high [Li 2020]. Therefore, we believe that a preligation experiment first can be performed to block more than 60 minutes of blood flow. If there is no noticeable change in the color and temperature of the corresponding blood supply area, ligation or permanent blockade can be considered. At the same time, autologous or artificial blood vessel bypass should be performed as soon as possible. Transplantation recanalize the corresponding blood supply area to reduce the risk of disabling amputation and brain disease [Zhao 2019].

(2) Repair method. Most scholars believe that if the fistula is small and the inflammation is mild, the fistula can be repaired. However, the inflammation, in this case, is severe, which is not suitable for this method. If the repair is forced, it will cause irreversible fatal bleeding due to tissue edema or aggravated infection [Morisaki 2014].

(3) Resection, closure, and bypass surgery. This method is suitable for cases with large fistulas, severe inflammation, fragile tube walls, difficult repairs, or severe stenosis. In this case, the method adopted the descending aorta stent implantation to seal the left subclavian artery to block the blood supply to the proximal heart, and then through the left common carotid artery-left subclavian artery artificial blood vessel bypass surgery to reopen the blood supply area to save the affected limb reaches the corresponding therapeutic effect [Al-Thani 2021]. However, we believe that the method of implanting the left subclavian artery stent on the first admission, in this case, is not advisable, because when the esophageal fistula invades the blood vessel, inflammation, edema, and necrosis of the surrounding tissues are serious, so implantation is performed at this time. The stent is very easy to cause the formation of bacterial clots in the stent to aggravate bleeding and infection.

Treatment of esophageal fistula: For those with small fistulas, mild inflammation, or unobvious shunting, fasting and drinking can be strictly performed, and enteral nutrition can be achieved by inserting duodenal-jejunal nutrition tubes to preserve intestinal function [Sladojevic 2017]. For patients with large fistulas and severe inflammation, esophagectomy, diversion, or external placement decisively should be adopted, and second-stage surgery should be pursued. After the operation, anti-infection should still be emphasized, and measures such as fasting and drinking, mediastinal drainage, and chest drainage should be adopted.

Treatment of mediastinal inflammation: According to the treatment of this case, we believe that we should actively explore whether foreign bodies are remaining, thoroughly debride the wound, repeatedly wash and drain, use effective antibiotics for anti-infection treatment, and strengthen lung function training to promote lung recruitment [Georvasili 2016].

CONCLUSION

When the upper gastrointestinal radiography is negative, if the esophageal fistula is highly suspected, gastroscopy should be performed for further confirmation. As a surgeon, not every operation is beneficial to the patients. Sometimes appropriate conservative treatment may also get gratifying results. MDT multi-disciplinary diagnosis and treatment will occupy an increasingly important position in the future. Human diseases are complex, and sometimes we cannot solve problems in one department. When we encounter complex problems, we must remember multi-disciplinary cooperation.

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