

Endovascular Treatment of Aorto-esophageal Fistula Caused by Ingestion of a Fragment of Beef Bone: Case Report and Review of the Literature

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Abstract Context: Aorto-esophageal fistula is a rare but catastrophic complication that can occur either primarily or after aortic reconstruction. Because of its rarity, no standardized protocols for its diagnosis and treatment have been established. **Objective:** The objective of this study was to report on a case of aorto-esophageal fistula caused by a fragment of beef bone, which was treated by means of an endoprosthesis in the descending thoracic aorta to control the bleeding. **Case report:** A 72-year-old female patient was initially admitted to the emergency service of a public hospital with dysphagia and intense pain in the dorsal region after having ingested a fragment of beef bone. Upper digestive endoscopy was performed to remove the foreign body. Nine days after the patient had been released, she was again admitted to the emergency service with back pain, voluminous hematemesis and signs of hypovolemia. Emergency angiotomography was performed, which revealed the presence of a pseudoaneurysm in the thoracic aorta just below the aortic arch that was suggestive of an aorto-esophageal fistula. The patient was taken to the hemodynamics center and underwent an endovascular procedure to insert an endoprosthesis into the descending thoracic aorta, to control the bleeding. She was released after a hospital stay of 94 days, without further signs of infection or aortic bleeding. She evolved without symptoms during a follow-up of around 30 days, but then returned to the emergency service with persistent fever and signs of sepsis. She underwent left thoracotomy for drainage and debridement, but her condition worsened and she died during the immediate postoperative period. **Conclusion:** Aorto-esophageal fistula is a serious clinical condition that needs to be dealt with early on, because of the risk of severe blood loss. Endovascular treatment is very important in this regard, since it promotes control over bleeding through an efficient and less invasive approach.

Keywords: aorto-esophageal fistula, esophageal foreign body, endovascular stenting, upper gastrointestinal hemorrhage, abdominal aortic aneurysm, endoscopy

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1. Introduction

Aorto-esophageal fistula is a rare cause of gastrointestinal bleeding that may be fatal. It can occur secondarily to aneurysm of the thoracic artery, foreign body reaction, esophageal malignancy or postoperative complications. [1] In cases that are diagnosed or suspected, treatment decisions need to be made rapidly because of the high risk of mortality. The treatment options may be surgical or endoscopic, or may involve interventional radiology. [2,3] Despite the large number of surgical treatment strategies, aorto-esophageal fistula remains a catastrophic complication with high morbidity and mortality.

[4] However, worldwide, few cases of aorto-esophageal fistula caused by foreign bodies have been successfully treated. [5,6] The objective of this study was to report on a case of aorto-esophageal fistula caused by a fragment of beef bone that was treated by means of an endoprosthesis in the descending thoracic aorta, to control the bleeding.

2. Case Report

A 72-year-old female patient was initially admitted to the emergency service of a public hospital with dysphagia and intense pain in the dorsal region one hour after having ingested a fragment of beef bone. Upper digestive endoscopy was performed to remove the foreign body

with approximately 2,3 cm of length (i.e. the bone fragment), which was located 20 cm beyond the upper dental arch, and she was released soon afterwards. Nine days later, she was again admitted to the emergency service with back pain, voluminous hematemesis and signs of hypovolemia. A vascular surgery assessment was requested. Upon physical examination, she was found to be pallid, tachycardic and hypotensive, and blood volume replacement was started immediately. The patient underwent upper digestive endoscopy again, which showed an area of esophageal perforation with

the presence of coagulum, 20 cm beyond the upper dental arch, in the same area from which the foreign body had been removed previously. During the 1st and the 2nd upper digestive endoscopy no bacterial culture was performed considering the patient did not have any clinical signs of infection. Emergency angiotomography was performed, which revealed the presence of a pseudoaneurysm in the thoracic aorta just below the aortic arch, indicating a grade IV esophageal injury, [7] that was suggestive of an aorto-esophageal fistula (Figure 1 and Figure 2).

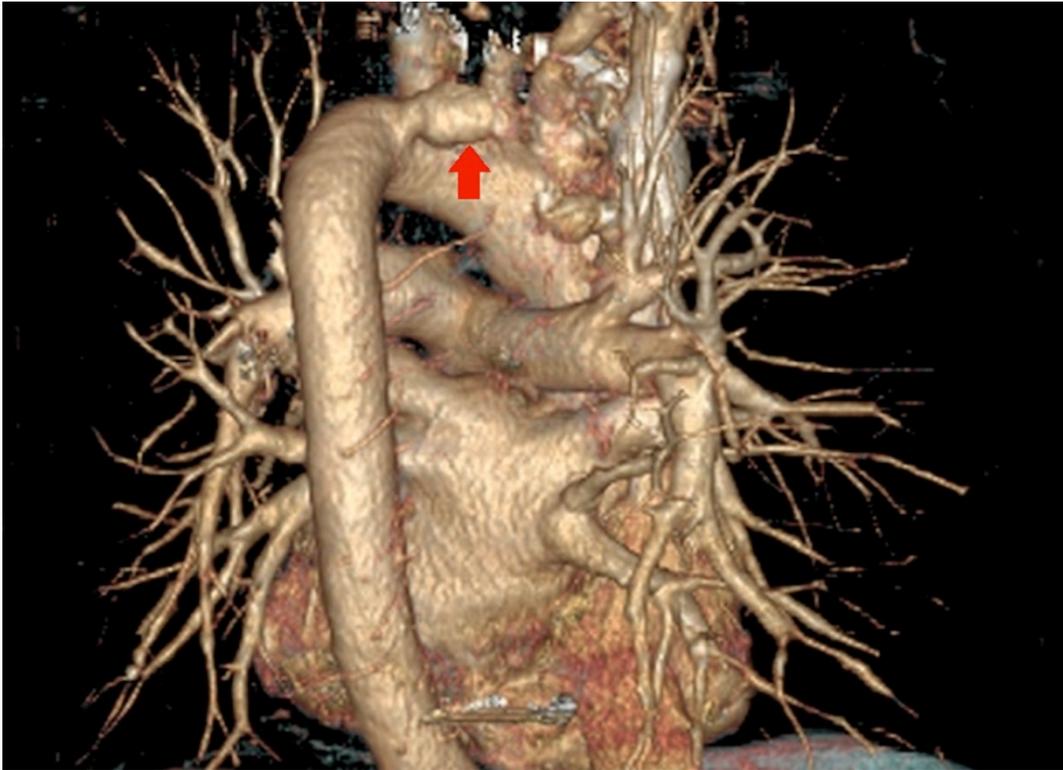


Figure 1. Pseudoaneurysm of the thoracic aorta shown in a 3D reconstruction from chest computed tomography scans

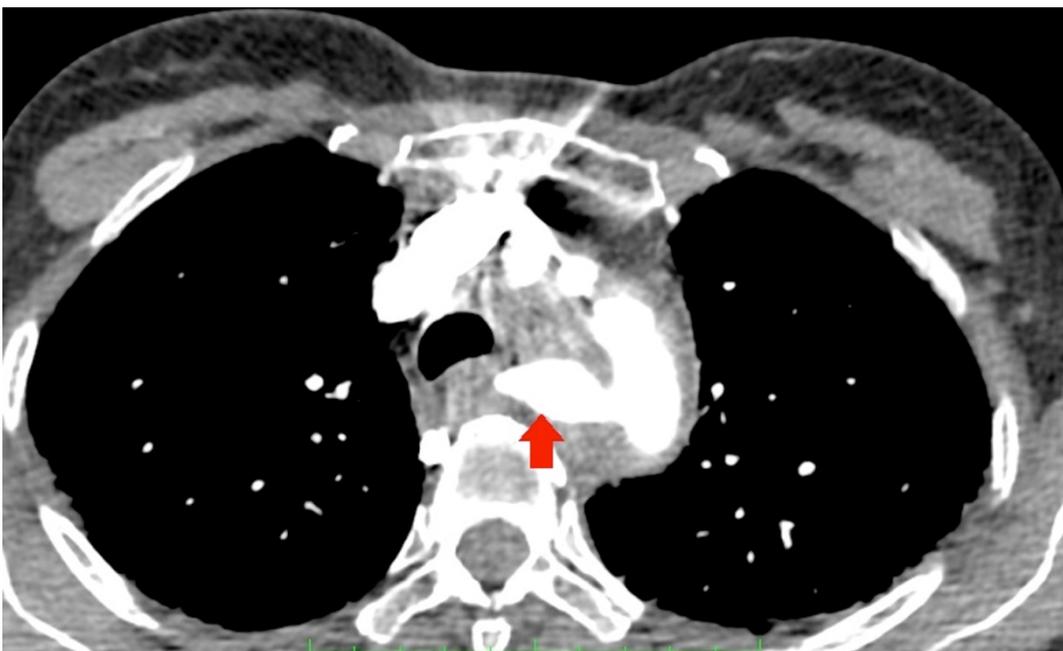


Figure 2. Pseudoaneurysm of the thoracic aorta adjacent to the esophagus, shown on chest computed tomography scans



Figure 3. After endovascular treatment of pseudoaneurysm of the thoracic aorta by means of implantation of an endoprosthesis, shown in a 3D reconstruction from chest computed tomography scans

On the same day, the patient presented a new episode of hematemesis and evolved with hemorrhagic shock. She was taken to the hemodynamics center and underwent an endovascular procedure to insert an endoprosthesis into the descending thoracic aorta (Figure 3), to control the bleeding. The final angiography showed that the aortic lesion had been eliminated and that there was no extravasation of contrast. On the next day, the patient underwent exploratory cervicotomy to deal with the esophageal lesion, which consisted of esophagostomy, esophageal cerclage and gastrostomy. Over the course of the postoperative evolution, she presented persistent fever and evidence of an abscess in the mediastinum, close to the aortic arch, which was shown on chest computed tomography (CT) scans performed three days after admission, from this moment the Early Goal Directed Therapy

(EGDT) protocol was initiated. [8] Video pleuroscopy was then performed in order to drain the abscess.

Over the course of the hospital stay, the patient's evolution continued to include daily episodes of fever, without leukocytosis, despite prolonged antibiotic therapy. Upper digestive endoscopy was performed again and showed that a labiated ulcerative lesion persisted in the region where the esophagus had been perforated, this time a bacterial culture of the ulcer, a blood culture and a pleural fluid culture were performed all showing negative results. It was therefore decided to endoscopically implant an esophageal prosthesis to avoid persistence of mediastinal contamination. After this procedure, the patient evolved with improvement of the fever and she was released after a hospital stay of 94 days, without any signs of infection or aortic bleeding.

The antibiotic therapy was continued for a further 15 days after the patient had been released from hospital and she evolved without any symptoms over a follow-up period of around 30 days. However, she then returned to the emergency service with persistent fever and signs of sepsis. Chest CT scans were performed, which showed the mediastinal abscess and left-side pleural empyema. The patient then underwent left-side thoracotomy for drainage and debridement. However, her condition worsened and she died during the immediate postoperative period.

3. Discussion

Aorto-esophageal fistulas can evolve clinically in an insidious manner, with initial manifestations of fever and pain that precede hemorrhage of the digestive tract. It may also evolve rapidly, with sudden massive hematemesis, depending on the different mechanisms for the pathogenesis of the aorto-esophageal fistula [9].

Sudden hematemesis is the first clinical sign in situations in which communication between the esophagus and the aorta is created through immediate penetration of both walls after ingestion of a foreign body. This mechanism is extremely rare and does not always provide enough time to institute treatment. On the other hand, aorto-esophageal communication may occur gradually, with an intense inflammatory reaction in the esophageal wall where the foreign body is found, which may lead to necrosis of the adjacent tissue and formation of an aortic pseudoaneurysm and an aorto-esophageal fistula. [10] In our case, this last mechanism would explain the nine-day period that elapsed between the patient's ingestion of the foreign body and the occurrence of aortic perforation, shown through hematemesis.

Hematemesis consequent to aorto-esophageal fistula may be catastrophic, since it puts the patient at imminent risk of death due to the large volume of blood loss. [7] Hence, early diagnosis and haste in instituting surgical treatment are critical factors for increasing the survival rate among patients with aorto-esophageal fistula [10].

A history of dysphagia and removal of a foreign body preceding the episode of hematemesis are important within the clinical history, given that these make it possible to rapidly direct the diagnosis towards aorto-esophageal fistula. According to Zhang et al., [10] an early differential diagnosis of upper digestive hemorrhage is of fundamental importance, given the possibility of confounding the diagnosis with other pathological conditions such as peptic ulcer or acute lesion of the esophageal mucosa. Making these other diagnoses would delay making the diagnosis of aorto-esophageal fistula, which is a rare condition but requires rapid clinical management. For this reason, obtaining a detailed clinical history during the investigation is fundamental.

Upper digestive endoscopy has the capacity to indicate the location of the bleeding in the digestive tract, but it does not confirm the presence of an aorto-esophageal fistula. This confirmation can be achieved through angiotomography, which is a simple, safe and noninvasive examination that has high sensitivity and specificity for diagnosing aorto-esophageal fistula. In comparison with arteriography, angiotomography with image reconstruction

is capable of more clearly showing the extent of the aortic lesion and the relationship between the aorto-esophageal fistula and the foreign body.¹⁰ For this reason, it is the examination of choice in investigating such cases, given that it can precisely indicate the area of the aorta that presented the pseudoaneurysm and thus suggested the aorto-esophageal fistula.

Despite the surgical alternatives for treating aorto-esophageal fistula that have been reported in the literature, there is no consensus regarding which of these would be the best method. [2,10] The classical treatment for aorto-esophageal fistula is conventional surgery. [11] However, this has been correlated with a high mortality rate, especially among patients who present hemorrhagic shock or who are in an unfavorable clinical condition. [2] In the case reported here, the patient was admitted to the emergency service already showing signs of hypovolemia and rapidly evolved to a state of hemorrhagic shock. This would have made conventional surgery a risky choice, given the clinical conditions at that moment. For this reason, we decided to use an endovascular technique that had been used more recently and which would provide treatment alternatives in situations like this.

Successes achieved through endovascular treatment have been reported over the last few years. [11] However, although this technique is now widely used, there is relatively little information available regarding results from endovascular treatment of aorto-esophageal fistula. [9] This is because of the rarity of this condition, the fact that this treatment has only recently been applied, the scarcity of Nonetheless, an increasing number of studies have been confirming that the endovascular procedure consisting of insertion of a coated stent has a central role relating to control over hematemesis and stabilization of the hemodynamic status. In cases of instability, this procedure should be applied without hesitation. It is a safe and feasible method for treating patients with aorto-esophageal fistula, provided that an early diagnosis is made and that it is applied aggressively and without delay. [12] This procedure has shown several advantages over conventional surgical treatment with an arterial bypass, given that it can be performed with the patient under sedation and local anesthesia, which minimizes the blood loss. It is well tolerated by high-risk patients and also reduces the mortality rate and the chances of cardiovascular diseases. [13]

Because endovascular treatment is faster, less complex and safer than conventional surgical treatment in unstable patients, some studies have considered that endovascular treatment is the first-line method for achieving immediate control over aortic bleeding. [2,12] This justified our choice of endovascular treatment for this case, given that the patient presented massive hematemesis and hemorrhagic shock.

Immediate action aimed at controlling and preventing fatal bleeding is essential. [9] This will make the next surgical step safer. [12] This next step needs to be a procedure that is capable of halting the continual contamination through the esophageal fistula and should precede definitive vascular treatment comprising interposition of an aortic prosthesis, so as to avoid infection of the prosthesis [11,12,13].

However, because of the high risks of contamination of the endoprosthesis [2,12] and recurrent sepsis, [15] use of endovascular treatment alone does not provide a definitive

cure for aorto-esophageal fistula. [2] Nonetheless, both endovascular and conventional treatment prevent major blood loss in these patients, despite the high risk of infectious complications. This is because the esophageal cavity is not sterile and therefore the risk of contamination of this area is significantly high in cases of unrepaired lesions. [11]

It has been suggested that removal of this source of infection can be achieved through resection of the esophagus [7,12] or, in some cases, installation of an esophageal stent by means of endoscopy. [16] in addition to esophagectomy, antibiotic therapy [2,11,14,15] and complete debridement of the infection of the mediastinal tissue are fundamental. [9] A prior endovascular approach is useful because it enables debridement of greater aggressiveness, [9] which should be done early on, i.e. as soon as the patient's clinical condition allows. [2,12]

Ultimately, endovascular repair by means of a coated stent reduces the risk of aortic rupture. Thoracoscopy, performed subsequently, serves to treat mediastinal infections. [14] Combining these procedures has resulted in less trauma and faster recovery than has immediate conventional thoracotomy. [13,16,17]

In the case reported here, antibiotic therapy and the surgical approach to the esophagus to halt the bleeding were performed early on (less than 24 hours after the endovascular treatment). However, deviation of the gastrointestinal transit was performed only by means of esophagostomy, esophageal cerclage and gastrostomy. No initial debridement of the mediastinal tissue (as has been recommended in the literature) was performed. This might explain the episodes of persistent fever and the formation of a mediastinal abscess, which was only dealt with later on, by means of video pleuroscopy. The persistence of the ulcerated esophageal lesion and fever were the reason for implantation of an esophageal prosthesis. Despite a short period of clinical improvement through antibiotic therapy, the infectious condition of the mediastinum progressed and led to a severe septic condition that could not be reversed, even after the thoracotomy for drainage and debridement that was subsequently performed.

In the present case, the diagnosis was made rapidly, as was the intervention of the endovascular procedure that enabled control over the aortic bleeding. However, as foreseen in reports in the literature, an infectious condition due to mediastinal contamination was present and this was a determinant of evolution of the condition. This led to an unfavorable outcome before the patient could undergo definitive surgical treatment of the aortic lesion. This demonstrates the importance of early clinical and surgical approaches, which need to be implemented because of the mediastinal infection that accompanies aorto-esophageal fistula.

4. Conclusion

Aorto-esophageal fistula is a serious clinical condition that needs to be dealt with early on because of the risk of severe blood loss from the patient. In this regard, endovascular treatment has great importance, since it promotes control over the aortic bleeding by means of an effective and less invasive approach that serves as a bridge

to conventional surgical treatment. The latter should include repair of the esophageal lesion and aggressive debridement of the area adjacent to the lesion, followed by resection of the aorta and replacement of the endoprosthesis, which remains at risk of infection due to local contamination.

Recurrent sepsis is common under these conditions and should be combated by means of broad-spectrum antibiotic therapy, in association with surgical procedures to remove the focus of infection.

Although the reports published so far have shown that endovascular treatment can be used successfully in cases of aorto-esophageal fistula, further studies are needed to assess the results, given that use of this procedure has been limited because of the rarity of the condition and because this is a technique that has only been applied relatively recently.

Statement of Competing Interests

We do not have any conflicts of interest.

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