

CASE REPORT

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A Cause of Mortal Massive Upper Gastrointestinal Bleeding: Aorto-esophageal Fistula

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ABSTRACT

Introduction: Aorto-esophageal fistula is an uncommon but mortal cause of massive upper gastrointestinal bleeding. The most common causes are thoracic aortic aneurysm, foreign body reaction, malignancy and postoperative complication. It can be seen in different pattern on upper gastrointestinal endoscopy. There are surgical, endoscopic and interventional radiological treatment options, however, definitive treatment is surgical intervention. Diagnosis and treatment decision should be made quickly because of rapid and mortal course. **Case report:** In this article, a case of aorto-esophageal fistula was presented that resulted in mortality as a result of massive bleeding.

Key words: Aorto-esophageal fistula, upper gastrointestinal bleeding, endoscopy, computed tomography.

1. INTRODUCTION

Aorto-esophageal fistula (AEF) is a rare cause of upper gastrointestinal (GI) bleeding, which can be life-threatening. It can occur as a secondary to thoracic aortic aneurysm, foreign body reaction, esophageal malignancy and postoperative complication (1). In cases that are diagnosed or under suspect, treatment decisions should be made quickly because of the high mortality risk. Treatment options can be surgical, endoscopic or interventional radiological procedures. In this article we presented an aorto-esophageal fistula secondary to thoracic aortic aneurysm that resulted in mortality as a result of massive bleeding in a patient who applied due to hematochezia.

2. CASE REPORT

A 69-year-old male patient was taken to emergency service by their relatives due to bloody stools, dizziness and fainting. He had no history of known diseases or drug use. He

looked pale in the physical examination, his blood pressure was 90/60 mmHg, pulse rate was 102/min, and his bowel sounds were increased. Stool in the form of hematochezia was seen in the rectal examination. No pathological finding was found in laboratory tests, except that haemoglobin level was 7,9 g/dL. The patient was hospitalized, his oral intake was stopped, and 3 units of erythrocyte suspension transfusion were made. Colonoscopy was performed after stabilization and some residual stool content in the form of hematochezia and melena was found in the colon, however, no clear lesion that can be the focus of bleeding was seen. In the upper GI endoscopy performed on the second day of hospitalization, a mildly bulging lesion with overlying adherent clot and fibrin was detected at approximately 25 cm from the incisors in esophagus (figure 1). Adrenaline injection was applied around this area. Urgent chest computed tomography (CT) was requested with the suspicion of fistula or a vascular lesion. Partial

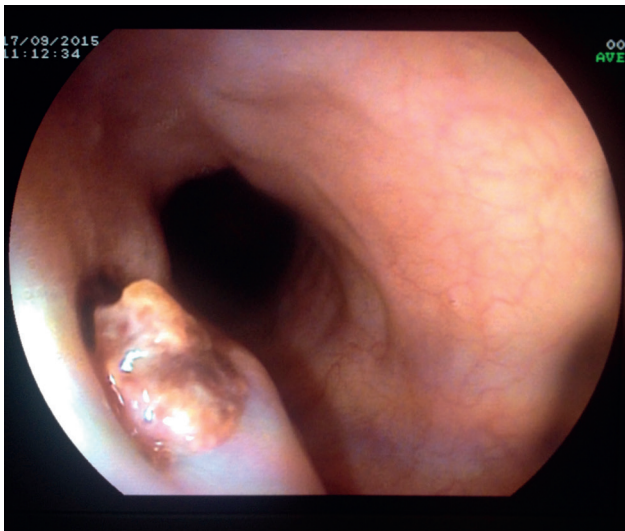


Figure 1. Upper GI endoscopy shows a mildly bulging lesion with overlying adherent clot and fibrin, at the esophagus.

thrombosed saccular aneurysm in distal aortic arch that showed protrusion towards the mediastinum and deviated the esophagus and increased wall thickness in the adjacent esophagus (fistula ?) were reported (Figure 2-3). Thoracic Surgery department was also consulted, but the patient passed away due to rapidly developing massive bleeding.

3. DISCUSSION

AEF is a rare condition, which has high morbidity and mortality. More than half of the cases develop secondary to thoracic aortic aneurysm just as in our case (1). Its classic triad was defined by Chiari as chest pain and/or difficulty in swallowing, a sentinel arterial bleeding and a massive bleeding after an asymptomatic period (2). However, this triad may not be observed in approximately half of the patients (3). It was seen that massive bleeding developed in our patient after the quiet period following a leading bleeding, but patient was not defined any cilinical symptom before the first application. Furthermore, hematemesis was also not observed in the first application of the patient, and it was present with hema-tochezia. Although this may also be the finding of a massive upper GI bleeding, first we performed a colonos-



Figure 2. Postcontrast MDCT axial image shows aortic fistula originating from distal aortic arcus and thickened esophageal wall at this level (Arrow).

copy because of the our patient was initially considered as lower GI bleeding and then upper GI endoscopy was performed as the bleeding focus could not be detected in colonoscopy. Endoscopy, contrast-enhanced CT and angiography have an important place in the diagnosis of AEF. AEF may not be determined initially in endoscopy due to massive bleeding or it can be seen as a slightly raised lesion covered by clot or fibrin in esophagus, pulsatile lesion covered by clot on the swollen ground towards the lumen, or actively pulsatile bleeding lesion (3-7). In our case, AEF was endoscopically observed as a mildly bulging lesion covered by clot and fibrin at the proximal esophagus. CT can be guiding in showing the localization of the lesion and its relationship with the esophagus. Angiography shows the presence of aneurysm, but it may not show the fistula in the period when there is no active bleeding (3).

While the prominent treatment in the existence of AEF is surgery, the options of endoscopic and endovascular treatment are also present. In the cases reported in the literature, treatments such as the endoscopically heater prob coagulation and application of hemoclip alone remained insufficient in the treatment (6, 7). Furthermore, the risk of the development of massive bleeding should



Figure 3. 3D surface shaded image shows aortic fistula originating from distal aortic arcus (Arrows).

also be taken into consideration during endoscopic treatment. However, there are also studies reporting that the use of covered esophageal stents in combination with other treatments is effective in the treatment of bleeding (8-11). Thoracic endovascular aortic repair is a treatment that is proposed as an alternative to surgery (12-14). This treatment was found related to various complications, especially with graft contamination in the long term, while it seems effective, the minimally invasive and beneficial in the early period. However, it was indicated that this method can be applied in combination with the surgical treatment or act as a bridge until open surgery

is applied (15,16). In a recent study reported by Akashi et al., it was indicated that aortic endovascular interventions alone is not a definitive method, and open surgical interventions such as aortic replacement through prosthesis or homograft, esophagectomy and repair with omentum are more effective in survival in the long term (17). Consequently, based on literature information, it can be said that the definitive treatment is the surgical method, but treatments such as the application of endovascular treatment and endoscopic esophageal covered stent can also be used as a bridge to surgical treatment and in combination with surgical treatment. Although initially no active bleeding was observed in endoscopy in our case, adrenalin injection was applied around the lesion due to the look that is considered as visible vessel like lesion covering with clot. Diagnosis of fistulised thoracic aortic aneurysm in esophagus was verified through CT. However, the hemodynamic situation of the patient was impaired due to massive bloody vomiting that developed within hours after the diagnosis, and the patient passed away without being able to make any endovascular intervention or another surgical intervention.

4. CONCLUSION

Consequently, it must be noted that aorto-esophageal fistula is a cause of upper GI bleeding, which can be result high rate of mortality. In the suspected patients with clinical and endoscopic findings, the diagnosis should be quickly verified, and a quick treatment decision should be made in collaboration with gastroenterology, interventional radiology and surgery depending on patient properties.

- **Author's contribution:** Author and all co-authors of this paper have contributed in all phases if it's preparing. Final proof reading was made by first author.
- **Conflict of interest:** The authors declare that they have no conflict of interest.

REFERENCES

1. Hollander JE, Quick G. Aorto-esophageal fistula: a comprehensive review of the literature. *Am J Med* 1991; 91: 279–87.
2. Chiari H. Über Fremdkörperverletzung des Ösophagus mit Aortenperforation. *Berl Klin Wochenschr* 1914;51:7–9 (in German).
3. Heckstall RL, Hollander JE. Aorto-esophageal fistula: recognition and diagnosis in the emergency department. *Ann Emerg Med* 1998;32(4):502-5.
4. Fang YH, Zhou XL, Liu JC. Aorto-esophageal fistula (AEF): Fatal upper gastrointestinal haemorrhage. *Turk J Gastroenterol* 2014;25(Suppl 1):321-3.
5. Jiao Y, Zong Y, Yu ZL, Yu YZ, Zhang ST. Aorto-esophageal fistula: a case misdiagnosed as esophageal polyp. *World J Gastroenterol* 2009;15(47):6007-9.
6. Yozgat A, Akmangit I, Altinbas A. A Rare Cause of Massive Upper Gastrointestinal Tract Bleeding in an Older Adult: Aorto-esophageal Fistula. *J Am Geriatr Soc* 2015;63(9):1966.
7. Hsu WF, Lin CC, Chang KM, Lee TH. Primary aorto-esophageal fistula: a rare but fatal cause of upper gastrointestinal bleeding. *J Dig Dis* 2013;14(12):676-8.
8. Onodera M, Inoue Y, Fujino Y, Kikuchi S, Endo S. A case of secondary aorto-esophageal fistula inserted a covered self-expanding esophageal stent to control gastrointestinal bleeding. *Case Rep Gastrointest Med* 2013;2013:857135.
9. Ishikawa M, Toyota N, Kakizawa H, Matsuura N, Hieda M, Awai K. Aorto-esophageal fistula after stent-graft treatment of an aortic arch mycotic aneurysm: treatment with embolization and covered esophageal stent. *J Vasc Interv Radiol* 2011;22:578–81.
10. Tsai SM, Chen YY, Chin-Yuan Y, Lai WL. Closure of an aorto-esophageal fistula with an esophageal stent and hemoclip. *Endoscopy* 2011;43 Suppl 2 UCTN:E302-3.
11. Rodrigues-Pinto E, Pereira P, Macedo G. Esophageal fully covered self-expanding metal stent for combined treatment of an aorto-esophageal fistula. *Endoscopy* 2015;47 Suppl 1 UCTN:E73-4
12. Dake MD, Miller DC, Semba CP, Mitchell RS, Walker PJ, Liddell RP. Transluminal placement of endovascular stent-grafts for the treatment of descending thoracic aortic aneurysms. *N Engl J Med*. 1994;331(26):1729–34.
13. Mitchell RS, Miller DC, Dake MD. Stent-graft repair of thoracic aortic aneurysms. *Semin Vasc Surg*. 1997;10(4):257–71.
14. Nienaber CA, Fattori R, Lund G, Dieckmann C, Wolf W, von Kodolitsch Y, et al. Nonsurgical reconstruction of thoracic aortic dissection by stent-graft placement. *N Engl J Med*. 1999;340(20):1539–45.
15. Mok VW, Ting AC, Law S, Wong KH, Cheng SW, Wong J. Combined endovascular stent grafting and endoscopic injection of fibrin sealant for aortoenteric fistula complicating esophagectomy. *J Vasc Surg*. 2004;40(6):1234–7.
16. Topel I, Stehr A, Steinbauer MG, Piso P, Schlitt HJ, Kasprzak PM. Surgical strategy in aorto-esophageal fistulae: endovascular stentgrafts and in situ repair of the aorta with cryopreserved homografts. *Ann Surg*. 2007;246(5):853–9.
17. Akashi H, Kawamoto S, Saiki Y, Sakamoto T, Sawa Y, Tsukube T, et al. Therapeutic strategy for treating aorto-esophageal fistulas. *Gen Thorac Cardiovasc Surg* 2014;62(10):573-80.