Surgical treatment for secondary aortoesophageal fistula

Kayo Sugiyama[1], Toru Iwahashi[2], Nobusato Koizumi[3], Toshiya Nishibe[2], Toshiki Fujiyoshi[4], and Hitoshi Ogino[5]

[1]Aichi Medical University
[2]Tokyo Medical University Hospital
[3]Affiliation not available
[4]Pediatric Cardiovascular Surgery
[5]Tokyo Medical University

May 6, 2020

Abstract

Background Aortoesophageal fistula (AEF) is a rare but life-threatening condition. Secondary AEF is a complication of previous surgery, and can be more critical and challenging than primary AEF. The number of cases of secondary AEF is increasing due to increasing number of thoracic endovascular aortic repair (TEVAR) performed. Although TEVAR has become a successful alternative surgical strategy for thoracic aortic aneurysms, secondary AEF after TEVAR may occur because of severe adhesion between the esophagus and residual thoracic aortic wall. Methods This study analyzed six patients with secondary AEF who were treated at Tokyo Medical University Hospital between 2011 and 2016. These participants included four patients who had undergone TEVAR and two who had undergone total arch replacement. Results Open surgical repair was completed in two patients who had undergone total arch replacement. TEVAR alone was performed in two patients who had undergone TEVAR. Combined repair of TEVAR as a bridge to open surgery was planned for two patients who had undergone TEVAR. However, reconstruction of the aorta and esophagus could not be completed in these patients due to severe adhesions, and they died during hospitalization. Conclusions Definitive open repair was successfully performed in patients with secondary AEF after total arch replacement. However, in the patients with secondary AEF after TEVAR, severe adhesion between the aorta and esophagus led to difficulty in performing a successful definitive open repair. The strategy for secondary AEF should, therefore, be decided considering the etiology of secondary AEF.

Surgical treatment for secondary aortoesophageal fistula

Kayo Sugiyama[1] , MD, Toru Iwahashi[2] , MD, PhD, Nobusato Koizumi[2] , MD, PhD, Toshiya Nishibe[2] , MD, PhD, Toshiki Fujiyoshi[2] , MD, Hitoshi Ogino[2] , MD, PhD

[1] Department of Cardiac Surgery, Aichi Medical University, 1-1 Yazakokarimata, Nagakute, Aichi 480-1195, Japan

[2] Department of Cardiovascular Surgery, Tokyo Medical University, 6-7-1 Nishishinjuku, Shinjuku-ku, Tokyo 160-0023, Japan

Short title : Surgery for secondary AEF

Address for correspondence :

Kayo Sugiyama, MD, PhD

Assistant Professor

Department of Cardiac Surgery

Aichi Medical University Hospital

Address: 1-1 Yazakokarimata, Nagakute, Aichi, 480-1195, Japan

Tel: +81-561-62-3311,

Fax: +81-561-63-6841

E-mail: kayotaro3@gmail.com

Conflict of Interest: The authors declare they have no conflicts of interest.

Funding : None.

Author contributions

Study conception: KS; Data collection: KS, SS, TF; Analysis: KS, NK, MS, HO; Investigation: KS, NK, HO;

Writing: KS; Funding acquisition: KS; Critical review and revision: all authors; Final approval of the article: all authors; Accountability for all aspects of the work: all authors.

Abstract

Background

Aortoesophageal fistula (AEF) is a relatively rare condition that is often life-threatening. Secondary AEF is a complication of previous surgery, and can be more critical and challenging than primary AEF. The number of secondary AEF is increasing due to increase in the number of thoracic endovascular aortic repair (TEVAR). Although TEVAR has become a successful alternative surgical strategy for thoracic aortic aneurysms, secondary AEF after TEVAR may occur because of severe adhesion between the esophagus and residual thoracic aortic wall.

Methods

This study analyzed six patients with secondary AEF who were treated at Tokyo Medical University Hospital between 2011 and 2016. These participants included four patients who had undergone TEVAR and two who had undergone total arch replacement.

Results

Open surgical repair was completed in two patients who had undergone total arch replacement. TEVAR alone was performed in two patients who had undergone TEVAR. Combined repair of TEVAR as a bridge to open surgery was planned for two patients who had undergone TEVAR. However, reconstruction of the aorta and esophagus could not be completed in these patients due to severe adhesions, and they died during hospitalization.

Conclusions

Definitive open repair was successfully performed in patients with secondary AEF after total arch replacement. However, in the patients with secondary AEF after TEVAR, severe adhesion between the aorta and esophagus led to difficulty in performing a successful definitive open repair. The strategy for secondary AEF should, therefore, be decided considering the etiology of secondary AEF.

Keywords : secondary aortoesophageal fistula; thoracic endovascular aortic repair; total arch replacement

Main Text

INTRODUCTION

Aortoesophageal fistula (AEF) is a relatively rare condition that is often life-threatening.[1-3] Secondary AEF, a complication of surgery in the posterior mediastinum such as open repair, thoracic endovascular aortic

repair (TEVAR), and esophageal surgery, can be more critical than primary AEF, and the management can be more challenging because of the presence of adhesion in the pleural space and the poor general status of patients. The number of secondary AEF is increasing due to increase in the number of TEVAR. Although there have been few studies comparing post TEVAR and post graft replacement secondary AEF cases, [4,5] secondary AEF after TEVAR can be more critical than secondary AEF after graft replacement.

MATERIALS AND METHODS

We retrospectively reviewed the clinical charts of six patients who required surgical intervention for the treatment of secondary AEF at Tokyo Medical University Hospital between 2011 and 2016. Primary AEF resulting from esophageal malignancy, thoracic aortic aneurysm, foreign body ingestion, prolonged gastric tube intubation and etc. was excluded. Secondary AEF resulting from esophageal malignancy was not found. Diagnoses were made based on enhanced computed tomography and endoscopic examination. According to the results of blood cultures, antibiotics were initiated based on the recommendations of the infection control team in our institute. This study was conducted in compliance with the Declaration of Helsinki. All patients provided written informed consent for using their clinical data for scientific presentations or publications.

The surgical treatment strategy for each case was designed based on the patient's frailty and infection severity.[1-3] Treatment strategies were considered by a multidisciplinary team that included cardiac and vascular surgeons, general surgeons, radiologists, and anesthesiologists. Optional treatment strategies for AEF included open surgical repair, TEVAR alone, and combined repair with TEVAR and open surgery. In patients with severe infections indicated by air bubbles between the aorta and esophagus on computed tomography (Figure 1a, 1b) or an obvious fistula on endoscopy (Figure 1c, 1d), open surgical repair was considered. In severely deteriorated patients, TEVAR alone was considered. In severely deteriorated patients with severe infection, combined repair with TEVAR as a bridge to open surgery was considered.

Open surgical repair was performed in stages. Following esophagectomy, the proximal stump of the esophagus was pulled out to the left side of the neck and an esophagostomy was performed. Then, reconstruction of the descending aorta using a rifampicin-soaked Dacron graft was performed. After aortic reconstruction, the pleural cavity was left open and copiously irrigated with 12 to 24 liters of 0.2% Gentian violet solution[3] per day for about 3 days. After then, omental wrapping around the artificial graft in the left pleural space was performed and the chest was closed. After several months, when the patient's condition had stabilized, reconstruction of the esophagus using colon interposition was performed through the anterosternal route (Figure 2a). Combined repair was also planned in stages. Following TEVAR to control bleeding due to hematemesis and gain hemodynamic stability, open debridement and reconstruction of esophagus and aorta were planned. In combined repair cases, and cases of TEVAR alone, GORE® TAG® Thoracic Stent Graft (Gore & Associates, Flagstaff, AZ) was chosen as stent-graft because of the durability to infection of expanded polytetrafluoroethylene. Open surgical repair was done by one experienced cardiac surgeon and TEVAR was done by one experienced vascular surgeon.

We assessed clinical outcomes including complications of early and late stages. The early and late mortalities, cause of death, major adverse aortic events and recurrence of infection were also evaluated. Feasibility of the treatment strategy was considered based on these outcomes.

RESULTS

Patient characteristics, management strategies, and treatment outcomes are summarized in Table 1. The mean age was 67 years (range, 41–78 years); five patients were male (83%). Four patients had undergone TEVAR, two for thoracic aortic aneurysm, and two for chronic aortic dissection. Three (75%) of the four post-TEVAR cases underwent emergency TEVAR for impending rupture of the thoracic aorta. Two patients had undergone total arch replacement, one for thoracic aortic aneurysm, and one for chronic aortic dissection. There were no patients who had previously undergone esophageal surgery.

All six patients presented with fever and complained of general fatigue. The causative bacteria were detected in five patients (83%);*Streptococcus* in three and *Staphylococcus* in two. Antibiotics were administered

preoperatively and continued for at least 4 weeks following surgery. Gallium scintigraphy successfully detected the primary origin of sepsis in four patients (Figure 1e, 1f, 1g). Computed tomography was performed in all patients and additional endoscopy was performed in five (83%) patients to confirm the diagnosis. In three patients, computed tomography demonstrated severe infection with air bubbles between the aorta and esophagus (Figure 1a, 1b). In two patients, endoscopy clearly identified the fistulas (Figure 1c, 1d).

Surgical indication and strategy were decided considering each patients' frailty and infection severity. Two patients who had undergone total arch replacement underwent definitive open surgical repair (Figure 2a); of the patients who had undergone TEVAR previously, two underwent TEVAR alone and two underwent combined repair with TEVAR and open surgery (Figure 2b). In the two patients who had open surgical repair (Figure 2a), a longer period until treatment completion was necessary (229 days and 209 days), but they were discharged without complications. The patients who underwent TEVAR alone had required urgent surgery to control bleeding, and TEVAR was, therefore, performed without esophagectomy or reconstruction of the aorta. These patients were discharged without major complications shortly following surgery, in 28 and 86 days. In one of the two patients who underwent combined repair, the aorta could not be resected due to severe adhesions, and esophageal reconstruction could not be completed following esophagectomy due to the severely deteriorated state of the patient. This patient died of an aortobronchial fistula during hospitalization. In the other patient, esophageal repair was performed without esophagectomy. An additional extra-anatomical bypass from the ascending aorta to the abdominal aorta bypass with closure of the aortic stump was performed due to severe adhesions (Figure 2b). This patient died of sepsis during hospitalization.

Overall, in-hospital mortality occurred in two (33%) of the total 6 patients, who underwent combined repair. The condition of both these patients was too deteriorated to undergo definitive repair of the aorta and esophagus, and they were subsequently hospitalized for a long period (118 days and 265 days). Furthermore, they had severe adhesions between the aorta and esophagus. Late mortality occurred in two (50%) of the remained four patients. One patient who underwent TEVAR alone died of rupture of the remaining thoracic aortic aneurysm 6 months following surgery, and one patient who underwent surgical open repair died of pneumonia 1 year following surgery.

DISCUSSION

AEF is a relatively rare condition that is often life-threatening. Despite significant refinement in surgical techniques, the operative mortality of open repair ranges from 25% to 100%.[1-3]There are several etiologies of primary AEF including thoracic aortic aneurysm, foreign body ingestion, esophageal malignancy, and prolonged gastric intubation.[6,7] There are several etiologies of secondary AEF resulting from surgery in the posterior mediastinum such as aortic arch replacement, TEVAR to descending aorta, and esophageal surgery. [4,8] The number of secondary AEF is increasing due to increase in the number of TEVAR being performed. Treatment of secondary AEF can be more challenging than that of primary AEF due to the presence of adhesion in the pleural space and the poor general status of patients. In the patients in this study who underwent combined repair, reconstruction of the aorta and esophagus could not be completed due to severe adhesions caused by previous TEVAR. There have been few studies comparing the degree of adhesion between post-TEVAR cases and post-graft replacement cases. Moreover, there have been few studies regarding outcomes between post-TEVAR and post-graft replacement in secondary AEF cases. Further studies are warranted, however, according to the present study, secondary AEF after TEVAR can be more critical than secondary AEF after graft replacement.

The exact mechanism of secondary AEF after TEVAR remains unknown. Eggebrecht et al. reported that the incidence of AEF was 1.9% and occurred 1–16 months following intervention in a series of 268 patients who underwent TEVAR.[5] Some reports suggested that its pathogenesis was related to inflammation of the aneurysmal wall.[9,10] They also suggested that the pathogenesis was related to esophageal ischemia secondary to elevated pressure in the posterior mediastinum, inflammation due to the resorbed hematoma, and mechanical compression by a large aneurysm following TEVAR.[9,10] In post-TEVAR cases, fistulas were reportedly caused by endoleaks into the residual aneurysmal sac, erosion of the stent-graft through the aorta, and ischemic necrosis of the esophageal wall due to compression of its feeding arteries by the stent-

graft.[4,5] In accordance with these reports, severe adhesion between the descending aorta and esophagus might occur more frequently in secondary AEF after TEVAR than after graft replacement. In aortic rupture cases, endovascular stenting does not remove the hematoma or the thrombosis, and thoracic compartment syndrome is likely to occur. Therefore, emergency TEVAR is associated with an increased risk of AEF occurrence.[4,5,11] In the present study, in three (75%) patients who had undergone TEVAR previously, AEF developed after emergency TEVAR.

It has been previously reported that aggressive treatment for patients with AEF was associated with good outcomes[5,10,12] and reduced short-term mortality. In the present study, although patients who received open surgical repair were hospitalized for a longer period, they were discharged without complications. However, the patients who underwent TEVAR alone were also discharged without complications. The treatment strategy should be carefully decided especially in patients with secondary AEF. In severely deteriorated cases, palliative treatment can be considered as an alternative.

Although TEVAR was proposed as an alternative surgical management strategy for open surgery,[13] secondary AEF after TEVAR can have severe consequences when left untreated.[4,5]With the increasing numbers of TEVAR procedures being performed and the longer follow-up periods, late complications of TEVAR are becoming increasingly evident.[4,5] The treatment strategy for secondary AEF after TEVAR should be carefully designed considering the presence of severe adhesions between the aorta and esophagus. Moreover, the patient's frailty should be considered in the decision of treatment. Further studies on surgical treatment for secondary AEF cases, especially regarding patient inclusion and exclusion criteria are warranted.

This study had several limitations. First, a small number of patients was included owing to the rarity of secondary AEF. Therefore, accurate evaluation might be difficult because combined surgery was performed on severely ill patients. Second, it was a retrospective study and the data were obtained from a single institution. Therefore, the study results may not reflect the general features of patients with AEF. Third, endovascular treatment strategy has evolved during the time period of this study.

CONCLUSIONS

Previous TEVAR may cause severe adhesions between the aorta and esophagus which may result in secondary AEF and make definitive repair of this condition very difficult. In secondary AEF cases, the surgical treatment strategy should be decided depending on each patients' status and the etiology of the fistula.

Acknowledgements

We would like to thank the Honyaku center for reviewing and editing the manuscript and our colleagues for their insightful comments. Additionally, we would like to thank Dr. Tachibana for his support with esophageal surgeries.

Conflict of Interest

The authors have no conflicts of interest to declare.

REFERENCES

1) Akashi H, Kawamoto S, Saiki Y, et al. Therapeutic strategy for treating aortoesophageal fistulas. Gen Thorac Cardiovasc Surg 2014;**62** : 573-80.

2) Kubota S, Shiiya N, Shingu Y, et al. Surgical strategy for aortoesophageal fistula in the endovascular era. Gen Thorac Cardiovasc Surg 2013; **61** : 560-4.

3) OkitaY, Yamanaka K, Okada Y, et al. Strategies for the treatment of aorto-oesophageal fistula. European Journal of Cardio-Thoracic Surgery 2014; **46** : 894-900.

4) Chiesa R, Melissano G, Marone EM, et al. Aorto-oesophageal and Aortobronchial Fistuale Following Thoracic Endovascular Aortic Repair: A National Survey. Eur J Vasc Endovasc Surg 2010; **39** : 273-9.

5) Eggebrecht H, Mehta RH, Dechene A, et al. Aortoesophageal fistula after thoracic aortic stentgraft placement: a rare but catastrophic complication of a novel emerging technique. JACC Cardiovasc Interv 2009;**2** : 570-6.

6) Amin S, Luketich J, Wald A. Aortoesophageal fistula: case report and review of the literature. Diag Dis Sci 1998; **43** : 1665-71.

7) Pipinos II, Reddy DJ. Secondary aortoesophageal fistula. J Vasc Surg 1997; **26** : 144-9.

8) Svensson LG, Crawford ES, Hess KR, et al. Experience with 1509 patients undergoing thoracoabdominal aortic operations. J Vasc Surg 1993; **17** : 357-68.

9) von Segesser LK, Tkebuchava T, Niederhäuser U, et al. Aortobronchial and aortoesophageal fistulae at risk factors in surgery of descending thoracic aortic aneurysms. Eur J Cardiothorac Surg 1997; **12** : 195-201.

10) Czerny M, Eggebrecht H, Sodeck G, et al. New insights regarding the incidence, presentation and treatment options of aorto-esophageal fistulation after thoracic endovascular aortic repair: the European Registry of Endovascular Aortic Repair Complications. Eur J Cardiothorac Surg 2014; **45** : 452-7.

11) Eggebrecht H, Baumgart D, Radecke K, et al. Aortoesophageal fistula secondary to stent-graft repair of the thoracic aorta. J Endovasc Ther 2004; **11** : 161-7.

12) Kieffer E, Chiche L, Gomes D. Aortoesophageal fistula: value of in situ aortic allograft replacement. Ann Surg 2003; **238** : 283-90.

13) Canaud L, Ozdemir BA, Bee WW, et al. Thoracic endovascular aortic repair in management of aortoesophaegeal fistulas. J Vasc Surg 2014;**59** : 248-54.

Figure Legends

Figure 1a: Preoperative computed tomography scan of an open repair case demonstrating air bubbles (white arrow heads) around the artificial graft

Figure 1b: Preoperative computed tomography scan of a combined repair case demonstrating massive air bubbles around the previous stent-graft

Figure 1c: Endoscopic view showing protrusion (white arrow heads) of the stent at the previous stent-graft site

Figure 1d: Endoscopic view showing exposure of the sutures and pledget (white arrow head) of the previous graft

Figure 1e, 1f, 1g: Preoperative gallium scintigraphy demonstrating enhancement between the aorta and esophagus

Figure 2a: Intra-operative photograph showing reconstruction of the esophagus using the terminal ileum and right colon

Figure 2b: The resected stent-graft in a combined repair case that was treated with an extra-anatomical bypass

Hosted file

`AEF manuscript Table.docx` available at https://authorea.com/users/317850/articles/447925-surgical-treatment-for-secondary-aortoesophageal-fistula

Figure 1a

Figure 1b

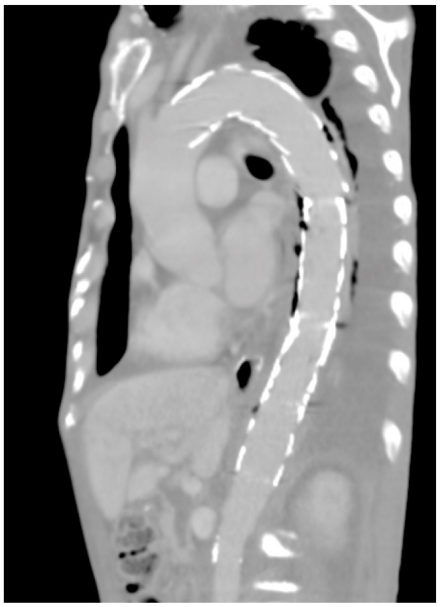

Figure 1c

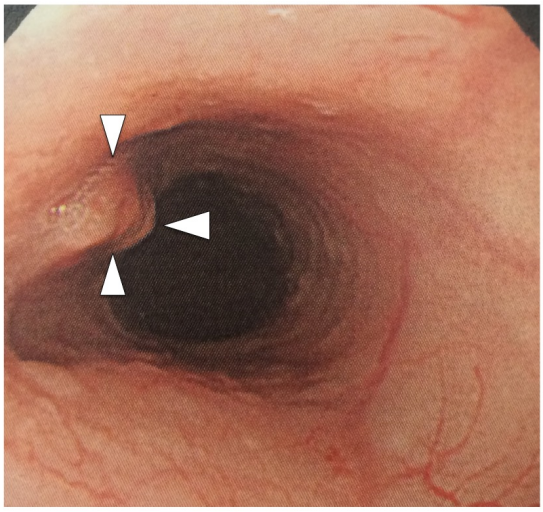

Figure 1d

Figure 1e

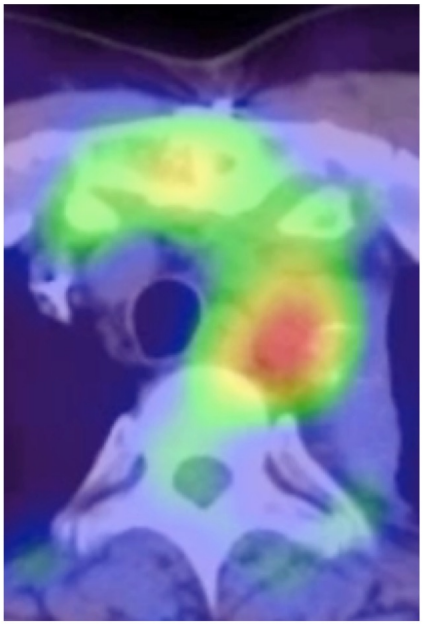

Figure 1f

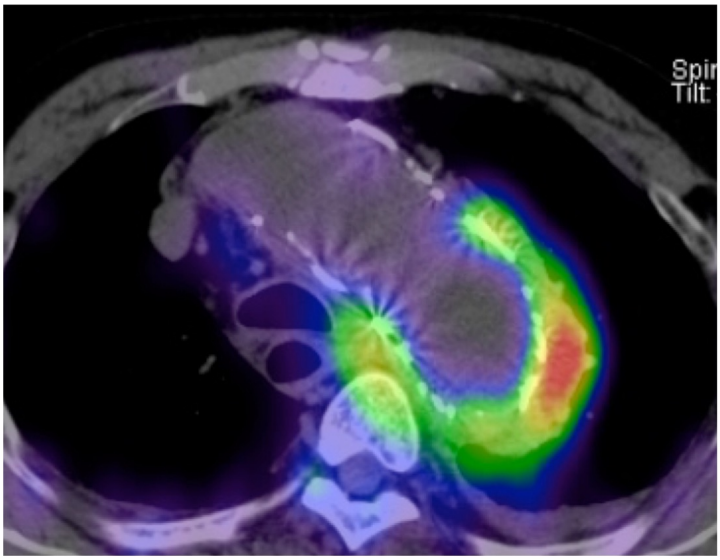

Figure 1g

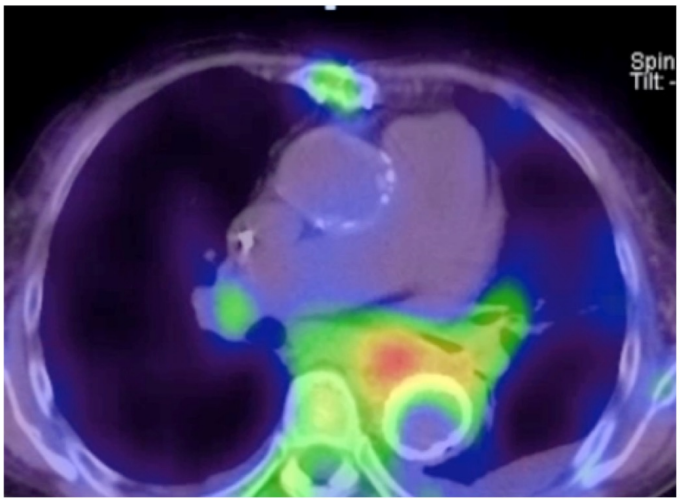

Figure 2a

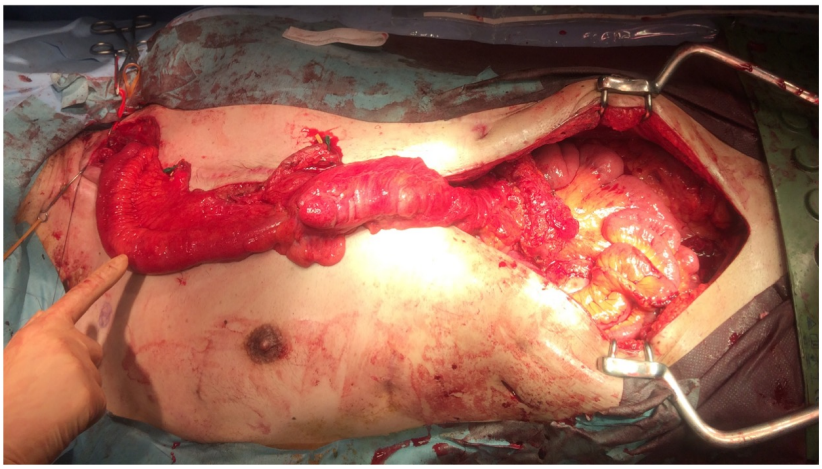

Figure 2b