Thoracic aortic aneurysm complicated by secondary aortoesophageal fistula after thoracic endovascular aortic repair : a case report

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Short abstract

This is a case report of a patient with a thoracic aortic aneurysm (TAA) presenting with dysphagia and weight loss as primary symptoms. She was treated via thoracic endovascular aortic repair (TEVAR). The procedure was complicated with a secondary aortoesophageal fistula (AEF) for which open surgical repair of the esophageal defect was done. Long term (i.e. more than 30 days) antibiotics were given. The recovery was uneventful. (Acta gastroenterol. belg., **2017**, 80, **527-529**).

Key words: Thoracic aortic aneurysm, aortoesophageal fistula, thoracic endovascular aortic repair, dysphagia, case report.

Case report

We report a case of a 71 year old woman with a history of gastroesophageal reflux disease for which she took no medication. Her chief complaint was progressive dysphagia since a few weeks with significant weight loss of 4 kilograms for which she sought medical attention in another hospital abroad. She was given the diagnosis of esophageal cancer. Her family brought her to our hospital for re-evaluation and treatment. The lab works showed mild normocytic anemia, iron deficiency and elevated CRP and sedimentation. Liver function tests were all within normal range. Tumor marker CEA was normal, CA 19-9 was only mildly elevated. An upper gastrointestinal endoscopy showed a bulging necrotic mass located in the mid-esophagus (Fig. 1). The lesion was stenosing hence unpassable with the endoscope. Nutritional support was provided using total parenteral nutrition. Subsequent CT scan of the thorax confirmed a multinodular heterogeneous stenosing mass in the mid-esophagus (Fig. 2) with some peripheral calcifications. The tumor was biopsied but the pathological report returned negative showing only necrotic material. Two more attempts to get a tissue sample were carried out via an endoscopic ultrasound guided fine needle aspiration cytology and via endobronchial ultrasound guided fine needle cytology. Both of these biopsies showed only hemorrhagic fluid with few nonpathological cells, not otherwise specified.

In an attempt to obtain a larger piece of tissue; potential distant metastatic lesions or adenopathies were looked for using FDG PET-CT. No other pathological lesions could be found besides the primary thoracic lesion which was hypermetabolic on PET. It was however impossible to differentiate between a primary esophageal tumor and a paraesophageal mass resulting in extrinsic compression on the esophagus. Interestingly on the PET/CT, which was done ten days after the first CT, the lesion appeared smaller in size correlating with the patient having less difficulty in swallowing liquids and even soft foods. In absence of a clear histopathological or clinical diagnosis, further imaging, by means of contrast enhanced MRI, was needed. This showed a large thrombosed aortic aneurysm (TAA) of the descending aorta with secondary dislocation of the esophagus and trachea (Fig 3).

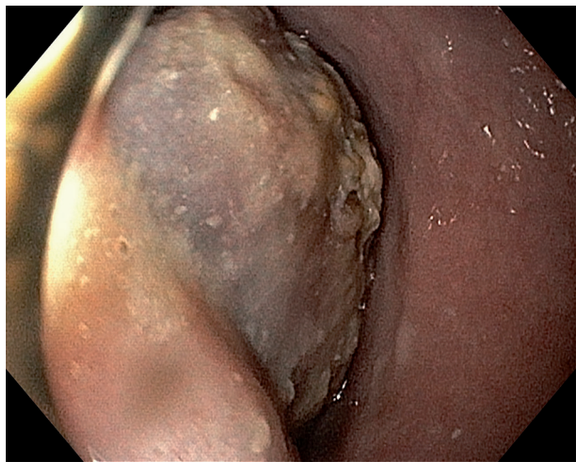

Fig. 1. — Endoscopic image showing a bulging mass with a rather smooth surface covered with necrotic debris and food at 20cm from the incisors. The mass caused a stenosis of the mid-esophagus.

Subsequently a thoracic endovascular aortic repair (TEVAR) was undertaken. Under general anesthesia a percutaneously aortic endoprosthesis was placed (Valiant 34-34-100mm). CT scan three days after the procedure showed a correct placement of the endoprosthesis. However a rupture of the esophagus in connection with the aneurysm was noted, confirming an AEF (Fig. 4). She was put on amoxicillin clavulanic acid and subsequently referred to a tertiary center for further treatment, total

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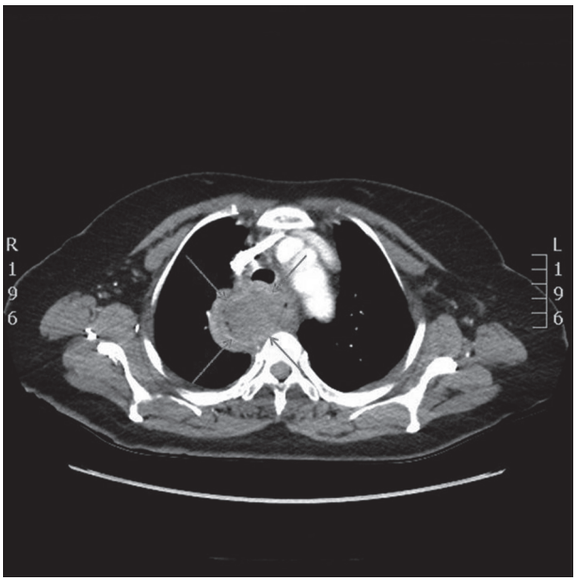

Fig. 2. — CT scan of the thorax showing a multinodular stenosing mass (10cm x 5cm) in the mid-esophagus with some calcifications in the border of the mass (arrows). The mass caused deviation of the trachea and narrowing of the main bronchi.

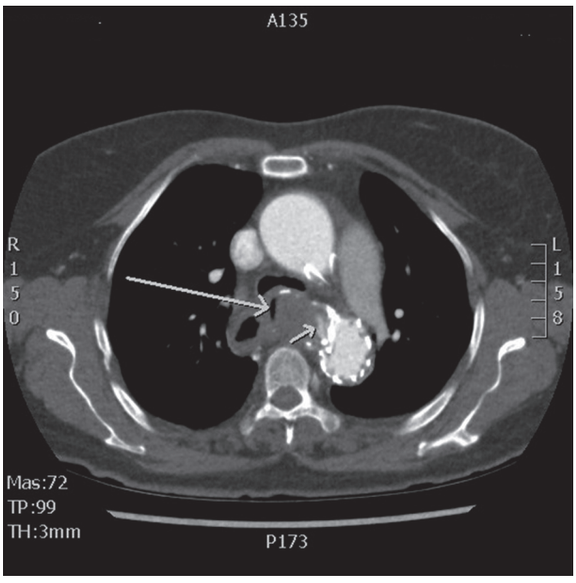

Fig. 4. — CT angiogram showing the endoprosthesis in the descending aortic arch with contrast medium leaking to the largely thrombosed aneurysm (48 x 37 mm) which implicates an endoleak (short arrow). The longer arrow points to the secondary aortoesophageal fistula

parenteral nutrition was continued.

A subsequent upper endoscopy showed an esophageal defect at twenty centimeters from the incisors with a diameter of 12 millimeters. The margins appeared smooth. An open surgical approach was considered most appropriate. The esophageal defect was covered using an intercostal bundle via right thoracotomy. The details

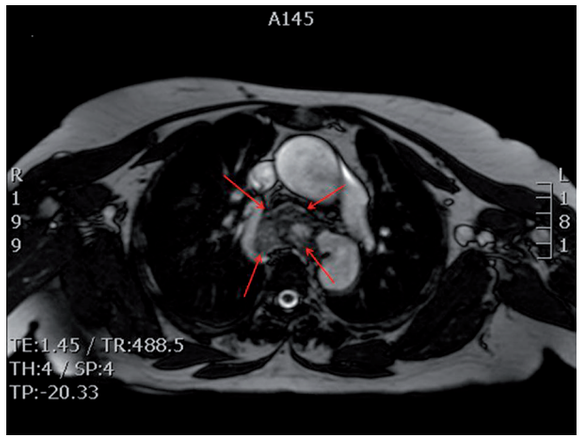

Fig. 3. — T2 weighted MRI image of the mediastinum on which a biloculair mass is seen (3,8 cm x 4,6 cm x 6,4 cm) (arrows). The lesion is heterogenic hypo and hyper echogenic on T2 weighed imaging. The esophagus is displaced to the right. The trachea is ventrally displaced. After injecting IV gadolinium (image not shown) a small part of the mass showed equal intensity than the adjacent descending thoracic aorta allowing the diagnosis of a largely thrombosed aortic aneurysm to be made.

of the operative technique will not be discussed in this report. Her recovery was uneventful. Solid foods were progressively introduced in the weeks after surgery.

Antibiotics were continued after follow-up consultation one month later. There was no evidence of mediastinitis or stent-graft infection.

Discussion

TAA is a rare disease (10.4 per 100,000 inhabitants) (1). Most often it is an accidental finding on CT scan or cardiac ultrasound carried out for other reasons. There are no signs or symptoms indicative of the presence of the disease. Dysphagia is only rarely a presenting symptom.

TEVAR is considered the preferred treatment when compared with open surgical repair because of less morbidity and equal outcome in terms of mortality (2). However randomized controlled trials directly comparing these two treatment modalities are lacking. (1) Since TEVAR is done more and more, the long term complications are becoming apparent and can be serious. These include leaks, graft migration, need for re-intervention and formation of fistulas.

An AEF can present with massive upper gastro-intestinal bleeding. Since the patient was already treated with an endoprosthesis and the aneurysm was filled with thrombus, this was not the case. The fistula was thought to be secondary to the placement of the endoprosthesis. Because the aneurysm had already caused the esophageal wall to become necrotic, the formation of a fistula was likely. Direct erosion into the esophagus is considered the mechanism of the formation

of a secondary AEF (3). It is a rare complication after TEVAR (4). In the case series from Canaud et al. there was only one case of secondary AEF in 236 patients (4). The patient mentioned in this case-series was treated with total esophagectomy (the esophageal defect was three centimeter in diameter) combined with removal of the endovascular stent-graft and an interposition aortic tube graft repair of the descending thoracic aorta was performed. A conservative approach is associated with a mortality of up to 100% (5).

Secondary AEF can arise as late as 12 months after TEVAR (4). The treatment of a secondary AEF proposed by Canaud et al. consists of intensive medical treatment followed by drainage and primary repair or resection of the esophagus alone or combined removal of the stent-graft and aortic reconstruction (4).

The placement of an esophageal stent in the combined treatment of AEF has been described in a case report (6) and has been advocated as an alternative in patients with severe comorbidities or who are too ill to undergo open surgery (7).

In our case the treatment of the secondary fistula was done at a tertiary hospital. The decision to treat the patient surgically, with the placement of an intercostal bundle, was partially based on the endoscopic finding of 'non healing' margins of the esophageal defect; the placement of an esophageal stent was therefore not considered suitable. She was also fit to undergo open surgery. The stent graft was not removed since broad spectrum antibiotics were started almost immediately after diagnosing the AEF and there was no evidence of stent-graft infection. Long-term antibiotics are associated with better survival. (2,4)

Conclusion

Dysphagia is a rare symptom of TAA. Therefore this case provided a diagnostic and therapeutic challenge. In our patient the placement of an endoprosthesis for the treatment of TAA in the thoracic arch was complicated with a secondary AEF. TEVAR is considered the treatment of choice when compared with open surgery, however randomized trials comparing both treatment strategies are lacking. An open surgical repair of the esophageal defect was considered the most appropriate treatment modality with the endoprosthesis left in place in our patient. Long-term antibiotics were continued as it is associated with better outcome.

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