Erosion of a retroesophageal subclavian artery by an esophageal prosthesis

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Supplemental material is available online.

Arterioesophageal fistula is a rare cause of upper digestive hemorrhage. It usually presents with a fatal outcome. It may happen in the course of esophageal or thoracic disease, or in the setting of foreign body ingestion, and involves the thoracic aorta or the subclavian artery.

Arterioesophageal fistula can also occur in the presence of a retroesophageal subclavian artery (RESCA), also known as *arteria lusoria*. It is usually asymptomatic but may cause dysphagia (*dysphagia lusoria*) and may demonstrate symptomatic atherosclerotic and aneurysmal degeneration. However, spontaneous fistulization in the esophagus is not described. It has mostly been associated with prolonged nasogastric tube presence. The present report describes the first case of arterioesophageal fistula with a RESCA caused by an esophageal prosthesis and discusses treatment based on a review of the literature.

Clinical Summary

Fourteen months before the actual admission, a 9-year-old girl with Down syndrome was admitted for caustic ingestion. She presented with severe ulceration from the middle esophagus down to the stomach. The acute episode was treated conservatively. Nonetheless, dysphagia rapidly developed in the patient. Two months after the accident, an esophagoscopy disclosed active stenotic esophagitis of the last third of the esophagus. Iterative pneumatic dilations were performed, which were discontinued because of esophageal fissuration and mediastinitis. A 14-mm Polyflex stent was inserted that rapidly migrated into the stomach; it was extracted and replaced by a 16-mm stent.

In June 2005, the patient was admitted for hematemesis and melena. Emergency esophagoscopy showed blood oozing around the prosthesis. The endoscopic procedure was interrupted, and the

child was brought to the catheterization laboratory where the prosthesis could be mobilized under angiographic control. When the prosthesis was pushed toward the stomach, a major hemorrhage occurred and a big aortic branch was identified as the source of bleeding. First it was regarded as a major bronchial artery. The prosthesis was pulled back to enable partial control of the bleeding, and coil embolization was performed in the so-called bronchial artery. This maneuver did not control the hemorrhage, and a RESCA was identified as the source of bleeding (Figure 1). RESCA embolization was attempted unsuccessfully, despite satisfactory angiographic images (Figure 2), and the patient presented sustained bleeding with hemorrhagic shock and cardiac arrest. After successful reanimation the patient underwent emergency surgery consisting of ostial RESCA ligation while the endoesophageal prosthesis was left in place. An 8-mm polytetrafluoroethylene (Gore-Tex, Flagstaff, Ariz) prosthetic bypass was constructed between the lateral aspect of the ascending aorta and the axillary artery. The patient had a prolonged intensive care unit stay (23 days) but fully recovered and was discharged without after-effects. Although the first esophagoscopic control showed embolized coils visible through the ulcerated esophageal mucosa (Figure E1), the lesions finally healed.

Comment

The interruption of the fourth right aortic arch between the notches for the common carotid artery and subclavian artery causes the advent of a RESCA. It is the most common aortic arch anomaly. It is present in 1 in 200 persons in the general population and is even more prevalent in patients with Down syndrome. Fistula between the esophagus and a RESCA is an exceptional event. We performed a PUBMED search of the literature and found 20 such cases reported between 1977 and 2005.

In 17 of those cases a prolonged period of nasogastric tube insertion was mentioned; in the remaining 3 cases this information is not reported. Four patients were reported to have survived the acute event.

In the 4 surviving patients, the upper gastrointestinal bleeding was controlled by a multidisciplinary approach consisting of endoscopic balloon compression followed by angiographic intervention, and the definitive cure was provided by urgent surgery. It is worth mentioning that arterial embolization was only a bridge to surgery whenever it was tried. In the successful cases, the therapeutic sequence might be summarized as angiographic localization of the arterial bleeding source^{2,3} followed by endoscopic compression (in the present case provided by the prosthesis itself).¹ After the angiographic finding of RESCA (with or without embolization, or occlusion catheter) the patient requires emergency operation consisting of RESCA ligation⁴ associated with a subclavian bypass to prevent upper limb ischemia.

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Received for publication Nov 14, 2005; accepted for publication Dec 1, 2005.

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J Thorac Cardiovasc Surg 2006;131:1183-4

0022-5223/\$32.00

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doi:10.1016/j.jtcvs.2005.12.026

Figure 1. Abnormal subclavian artery (*white arrow*); contrast dye oozing around the oesophageal prosthesis (*plain arrows*).

Despite the liberal use of the nasogastric tube and the relatively high prevalence of RESCA, fistulization is uncommon and one cannot recommend screening of this abnormality before any nasogastric intubation, even if it is prolonged. However, in the increasingly frequent case in which an esophageal prosthesis is implanted in a vulnerable tissue, such as in our patient, ultrasonographic evaluation of the esophagus is a simple and sensitive method that can be considered.⁵

Figure 2. Aorto-axillary bypass (*black arrow*); embolized RESCA (*white arrow*); backflow to RESCA before distal ligation at the site of the grey arrow.

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Figure E1. Esophageal erosion with visible coils (black arrow).