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Surviving a Catastrophic Upper Gastrointestinal Bleeding Caused by Esophageal-Subclavian Fistula: Case Report

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Abstract

Arterio-esophageal fistula (AEF), whether congenital or acquired, is a rare condition which can lead to fatal upper gastrointestinal bleeding. We report here a young man who developed a subclavian-esophageal fistula (SEF) secondary to chicken bone impaction in the upper esophagus. The diagnosis was reached by urgent upper endoscopy and Computed Tomography of the chest which showed pseudo-aneurysmal changes at left subclavian artery with leaked contrast through the fistula towered the esophagus. Urgent endo-vascular angiography confirmed the subclavian arterio-esophageal fistula that was managed uneventfully using covered 6mm Viban stent-graft. The patient survived this serious condition and was discharged home in good condition.

Keywords

Gastrointestinal Bleeding, Esophageal-Subclavian Fistula

1. Introduction

Bleeding from an arterio-esophageal fistula (AEF) is a rare and mostly fatal condition. The first case of aorto-esophageal Fistula was described by the French naval surgeon Dubreuil in 1818 [1]. Although this arises from an aberrant subclavian artery in most of occasions, it can result as a complications of foreign body impaction in the upper esophagus leading to an esophageal-aortic or esophageal-subclavian fistula in a normal anatomy setting [2] [3] [4]. Generally, this condition should be suspected if a patient reported difficulty in swallowing, massive vomiting of blood, or underwent recent aortic or esophageal surgery [1].

Chiari and colleagues published the first AEF due to boney food impaction in 1914 and described the specific symptoms that constituted a specific syndrome [5]. However, few cases of non-aberrant left subclavian AEF caused by foreign body impaction were reported [1] [6], and most of such patients succumbed to massive bleeding. Here we document one patient who survived a massive upper gastrointestinal bleeding caused by a left subclavian artery esophageal fistula induced by chicken bone impaction. Fortunately, this serious complication was discovered early and well managed by endovascular stent grafting [6] [7].

2. Patient Presentation

A previously well 16-year-old man who had experienced food impaction following a chicken meal presented to the emergency department complaining of discomfort at the root of the neck associated with difficulty in swallowing. The patient was discharged from the ED after normal chest and neck X-rays.

However, he returned back 6 days later with retrosternal discomfort and difficulty in swallowing, as well as having had few episodes of vomiting bright red blood, the last one of these was a large volume bleed.

Examination revealed that he was unstable with sinus tachycardia of 10^5 /min, hypotension at 100/60, pallor, and clinical sign of hypovolemic shock. The rest of the examination was unremarkable His hemoglobin was 6.6 md/dl (14 - 17 gm./dl) with normal biochemistry and INR (normal < 1.1) The patient was admitted to the intensive care unit and resuscitated with 6 units packed RBCs.

Urgent upper gastro-intestinal endoscopy showed traumatic ulceration at upper third of esophagus around 17 cm from incisor with sign of chronic changes suspicious of foreign body injury.

Urgent Computed Tomography of the chest with arterial phase contrast revealed a left subclavian-esophageal traumatic fistula with pseudo-aneurysmal changes of the left subclavian artery and demonstrated the leaked contrast through the fistula towards the esophagus (Figures 1(a)-(b)). Urgent endovascular angiography confirmed the subclavian arterio-esophageal fistula that was managed uneventfully using covered 6mm Viban stent-graft (Figure 2).

The patient improved dramatically with obvious stabilization of his clinical signs and his hemoglobin increased to 14 g/dl with complete resolution of bleeding.

He was discharged 48 hours post management on good clinical condition and remained well on regular clinic follow-up after 2 weeks, then after 3 & 6 months.

3. Discussion

As described by Chiari, Arterio-esophageal (AEF) syndrome consists of initial esophageal injury followed by a symptom-free interval preceding the "signal bleeding," and finally culminating in exsanguinating hemorrhage hours or days later [5]. The patient presented here exhibits a similar course to what was described in the literature. He initially complained of difficulty of swallowing after

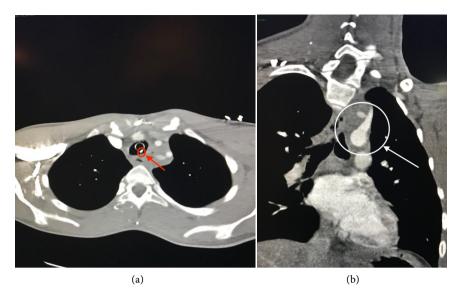


Figure 1. (a) CT scan showing the clip inserted initially to control the bleeding (arrow); (b) Sagittal view computed tomography of chest showed pseudo-aneurysmal changes at left subclavian artery with leaked contrast through the fistula towered the esophagus (arrow).

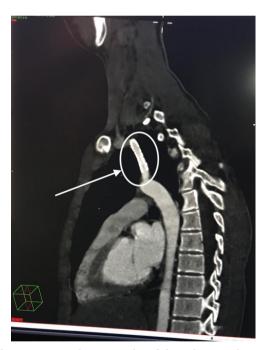


Figure 2. Sagittal view computed tomography of chest showing the stent-graft (arrow).

he ingested a bony chicken meal, and then he remained stable for few days. After that, he started to vomit blood (the index bleed) which ended up with a massive upper gastrointestinal bleeding.

Over the past years, more than hundred cases of AEF secondary to foreign body ingestion have been documented but only few, including our patient, have survived for months [4] [6]. Floréa reported a similar patient to ours, but involving an infected fistula [7]. Most of the cases of AEF occurred about 1 - 5 cm from the origin of the left subclavian artery which is explained by the narrowing

of the esophagus secondary to food impaction [5]. In contrast to most reported cases, our patient has normal anatomy of the subclavian artery and thanks to the early diagnosis & intervention, he survived this serious event [7].

The currently accepted management comprises vascular stent-graft interposition and if no success, this can be followed by esophageal surgery and mediastinal drainage. Fortunately, our patient did not need open thoracic surgery which usually carries a high mortality [8] [9] [10].

4. Conclusion and Recommendations

Arterio-esophageal fistula secondary to food impaction is rare but is life-threatening and should be discovered early by Computed Tomography imaging if suspected. The key message to be learned from this report is to exercise a high index of suspicion for AEF fistula in patients presenting with difficulty in swallowing or vomiting of blood after taking a bone-containing meal, or who ingest a sharp foreign body. This is because early recognition contributes to a better prognosis following relevant workup and subsequent management with endovascular stent grafting, or open thoracic surgery.

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