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Finding The Chameleon: A Massive Upper Gastrointestinal Bleeding Associated to A Double-Lumen Esophagus

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ABSTRACT

Double-Lumen Esophagus/Esophagogastric Fistula has rarely been reported in the literature. We report an unusual case of a 70-year-old woman undergoing emergency endoscopy for massive gastrointestinal bleeding with unexpected evidence of double lumen esophagus.

KEYWORDS: Esophagogastric Fistula; Hb; Orotracheal

INTRODUCTION

Double-Lumen Esophagus/Esophagogastric Fistula has rarely been reported in the literature. We report an unusual case of a 70-year-old woman undergoing emergency endoscopy for massive gastrointestinal bleeding with unexpected evidence of double lumen esophagus.

CASE REPORT

A 70-year-old woman with past medical history of hypertension, thyroidectomy for thyroidcancer, and recently dysphagia for solids and dyspnea for short efforts, was referred to our emergency roomfor melena. Barium X-ray performed a year earlier showed the presence of an esophageal ectasia; furthermore, mass tracheal compression was observed. At the admission in the emercency room Hb was 8.9 mg/dL, there was red blood from the nasogastric tube and rectal exploration confirmed the melena. An esophagogastroduodenoscopy was done: a large amount of blood was observed in the proximal third of the esophagus. Furthermore, in the middle third there was a diverticulum in whichbleeding seemed to originate but without obtaining a clear image of the source. The procedure was suspended to perform general anesthesia and orotracheal intubation which required the use of the video laryngoscope. Hb on blood gas analysis was 7 mg/dL therefore the patient was transfused by2 units of concentrated red blood cells. However, the exact source of the bleeding was not identifiedat the new endoscopy: the presence of red blood and clots in the lower third of the esophagus and inthe gastroduodenal lumen limited the examination. Since the endoscopic procedure had not found the hemorrhagic source, an angiography and an abdominal thoracic CT

scan with contrast were performed. In both procedures no active contrast spreading was reported; an anomalous course of the right mammary artery was observed: it ran near the paraesophageal formation which was 82.5 x 70 mm on the axial plane and with a longitudinal extension of 109 mm. This formation was posterior to the trachea and the right brachiocephalic trunk and gave compression of the cervical and upper thoracic tract of the esophagus. Its walls were irregular and hyperemic. Reviewing the images of previous radiography, the paraesophageal formation was identified as a double lumen esophagus that originated immediately under the pyriform sinuses. Then, another esophagogastroduodenoscopy was performed: red blood was aspirated and 2 esophageal pockets were observed with an entrance orifice about 15 cm from the dental arch and the junction between the 2 pockets found at about 28 cm; the mucosa had varicose cords in the absence of active bleeding. Respiratory and hemodynamic exchanges remained good throughout all procedures. Therefore, the patient was hospitalized at ICU where she was transfused with 2 other units of concentrated red blood cells, was extubated on the first day after a rapid weaning; no episode of melena occurred; the Hb values were always stable. On the third day, the patient was discharged to the emergency surgery department and sent to a referral center for surgical treatment.

DISCUSSION

"Double-lumen esophagus" is a fascinating endoscopic finding. Altough the definition of this termis not clear, it has been applied to the presence of "false" and a "true" esophageal lumen separated by a septum [1].

Double-lumen esophagus and esophagogastric fistula are terms used interchangeably in the literature. Double lumen of the gastrointestinal tract may be congenital (duplication) or acquired (fistula) [2]. It was postulated by Choudhry and Shenoy that the term 'double-lumen esophagus' should be used when both lumens are of equal diameter [3].

To our know ledge esophagogastric fistula is a rare entity usually as a result of an inflammatory pattern secondary to reflux esophagitis, a complicated Nissenfundoplication, Crohn's disease or malignancy [4]. Esophagealdilationprocedures done for strictures were also found to be a predisposingfactor [3].

The exact mechanism of fistula formation is still understudied due to the rarity of this disease process [5].

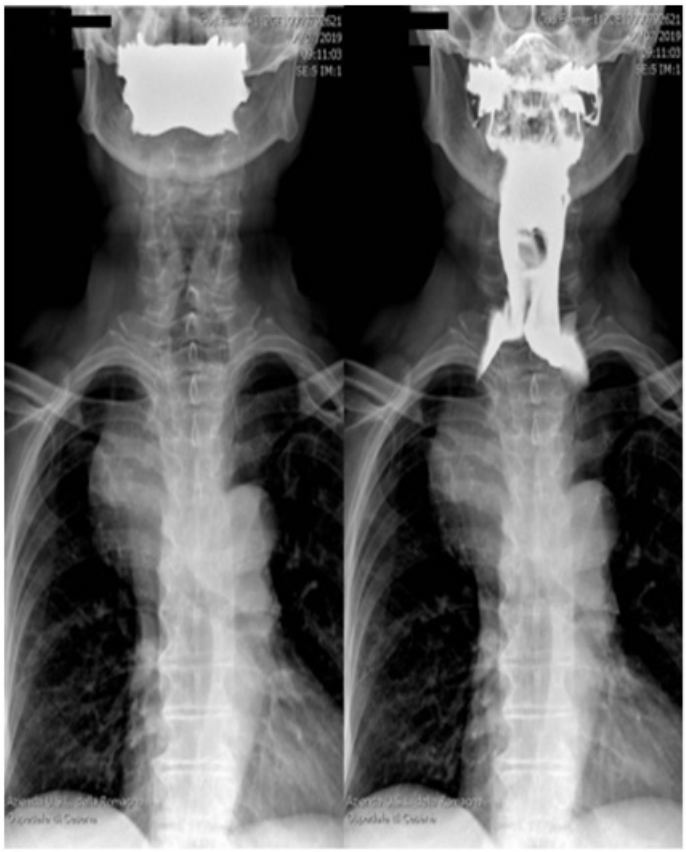

Figure 1

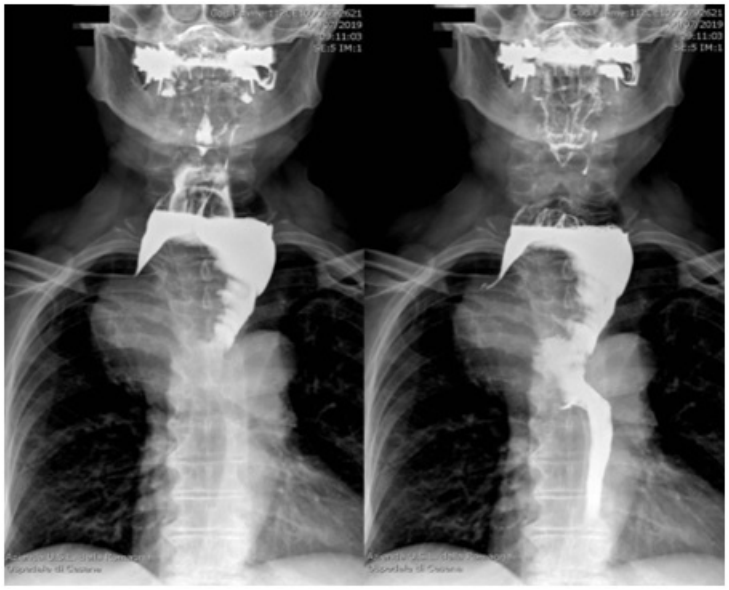

Figure 2

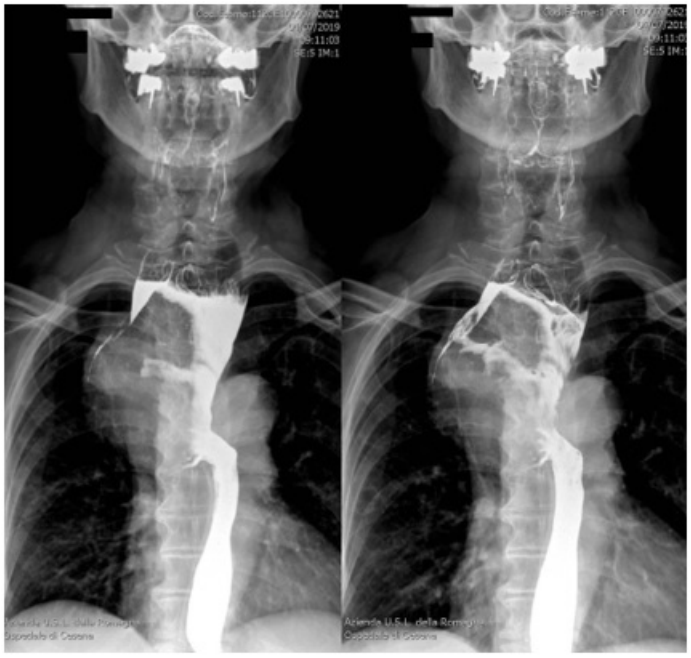

Figure 3

Symptoms are non-specific. Dysphagia is almost always reported by patients sometimes with associated odynophagia and respiratory impairment resulting from an accumulation of contents within the false lumen. Although barium swallow study or CT can be useful, endoscopy remains theprocedure of choice for diagnosing esophagogastric fistula [6].

There are no effective pharmacological treatments for esophagogastric fistula as far as a conservative approach with PPI and follow up can be targeted to selected cases [5]. Endoscopic and surgical interventions provide the route for restoration of a single lumen. Surgery carries a significant risk of morbidity and mortality, and accordingly, endoscopic alternatives are preferable. A new endoscopic septotomy technique using a ascissor-type knife, typically used for Zenker's diverticulotomy was recently proposed by Rao and colleagues [1].

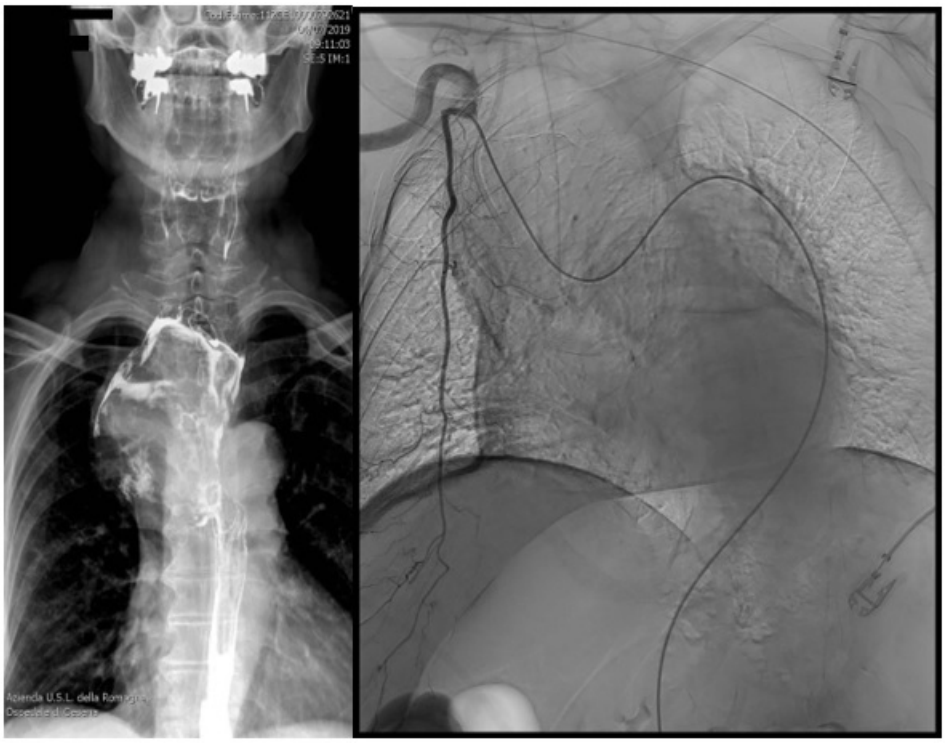

Figure 4

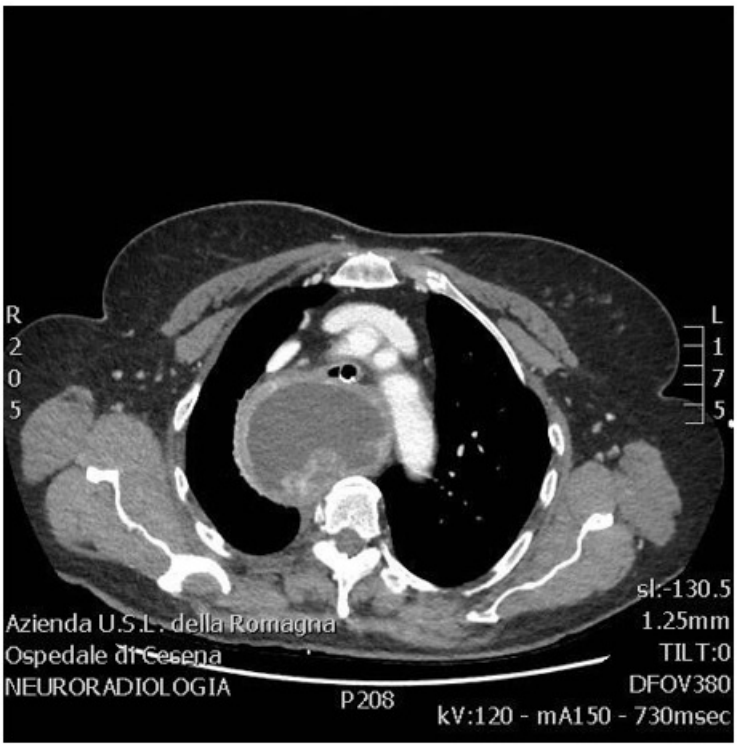

Figure 5

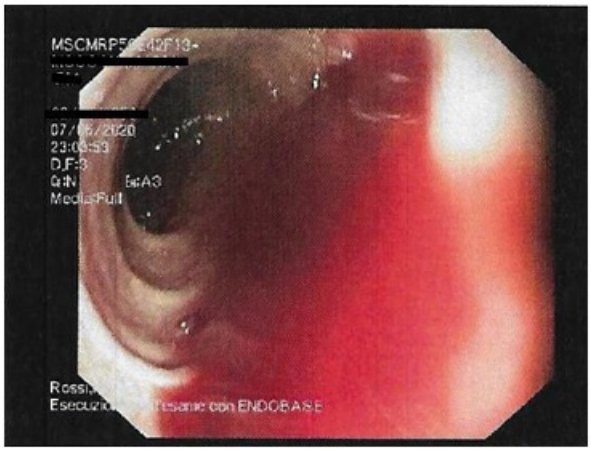

Figure 6

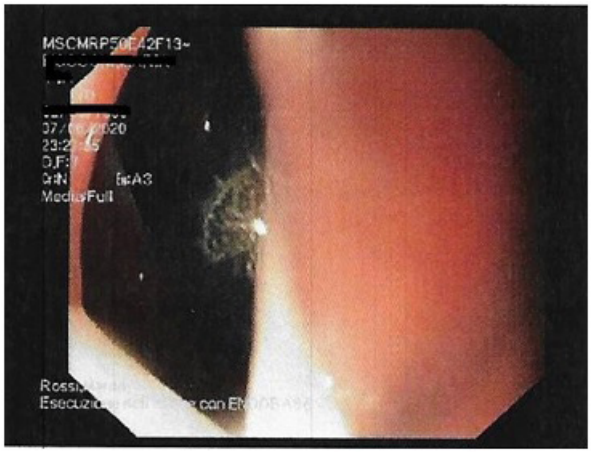

Figure 7

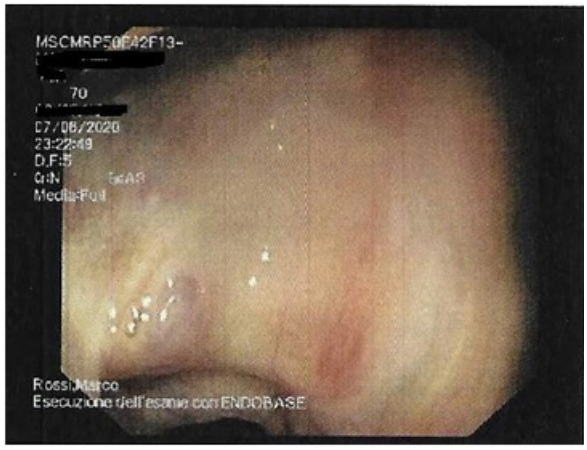

Figure 8

Our case was remarkable for several reasons: the atypical location (mid-esophagus), the absence of risk factors and the dramatic clinical presentation. The esophagogastric fistula hinderedthe management of the airways by compressing the trachea, compromised radiological images interpretation and offered a false route to the endoscopy, without allowing to identify the location and the source of bleeding. Fortunately, in our case the bleeding was self-limited.

Despite herrarity, this pathological entity may result therefore in a extremely varied clinical picture confounding the diagnosis and representing a challenge for the multidisciplinary team to "find the chameleon" and identify targeted treatment for the patient.

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