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Spontaneous Cardioesophageal Junction Perforation in an Immunocompetent Patient with Esophageal Candidiasis

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Abstract

Background: Esophageal perforation related to esophageal candidiasis is extremely rare in immunocompetent individuals with poor prognosis.

Case report: We report a case of spontaneous cardioesophageal junction perforation due to esophageal candidiasis in a 17-year old immunocompetent male patient. He was treated with systemic antifungal therapy and although surgical repair did not achieve impermeability, he survived from septic shock and multiorgan failure.

Conclusion: Spontaneous esophageal perforation due to esophageal candidiasis is an extremely rare complication. Aggressive medical and surgical treatment may lead to complete recovery.

Keywords: Spontaneous esophageal perforation, Esophageal candidiasis, Antifungal therapy, Candida.

Introduction

Spontaneous esophageal perforation is an uncommon clinical entity, associated with increased morbidity and mortality. Among different reasons that may lead to esophageal perforation, Candida infection is extremely rare, although Candida may be detected in as many as 25% of normal oesophagus. This infection is due to the ability of C. albicans to penetrate the epithelium. This latter plays a dominant role in the infectious disease caused by microorganisms as bacteria and fungi [1].

Candida esophagitis is described most commonly in immunocompromised patients. We report a case of spontaneous cardioesophageal perforation due to candidiasis, where although surgery did not achieve impermeability, the patient, treated with systemic antifungal therapy, survived from septic shock and multi-organ failure.

Case Report

A 17 year-old male patient was admitted to the Emergency Department for acute retrosternal and upper abdominal pain. From history, no existing co-morbidities were mentioned except long time starvation due to low socio-economic background.

As the initial plain chest and abdominal x-ray finding was the presence of free subdiaphragmatic air, an urgent explorative laparotomy was performed. More than 3500 mls of yellowish fluid were aspirated, while meticulous investigation of the peritoneal cavity revealed a one cm long perforation of the lower oesophagus, close to the esophagogastric junction. The perforation was closed with one layer, intermittent, absorbable sutures. Penrose type drains were placed close to the perforation, as also to the right and left side of the peritoneal cavity. No biopsies were taken, since no signs of malignancy or other condition were evident.

His admission to the Intensive Care Unit (ICU) for further treatment and support was judged as absolutely necessary. An excessive right hydrothorax was immediately treated with thoracic drainage (Figure 1). The situation was further complicated with septic shock and multi-organ failure. White, plaque-like mucosal lesions were noted on the oropharynx. Cultures from both the pleural and peritoneal fluid were positive for Candida albicans and intravenous voriconazole (200 mg twice daily) was started. Because of relapsing hydrothorax, a right thoracotomy was performed. Fever persisted and an abdominal CT scan revealed a left subdiaphragmatic abscess formation which was treated surgically. Ten days later, a water-soluble contrast esophagogram showed a leakage through the sutured perforation (Figure 2). Surgical intervention was rejected because of the good position of the drains and further treatment comprised continuous nasogastric suction, total

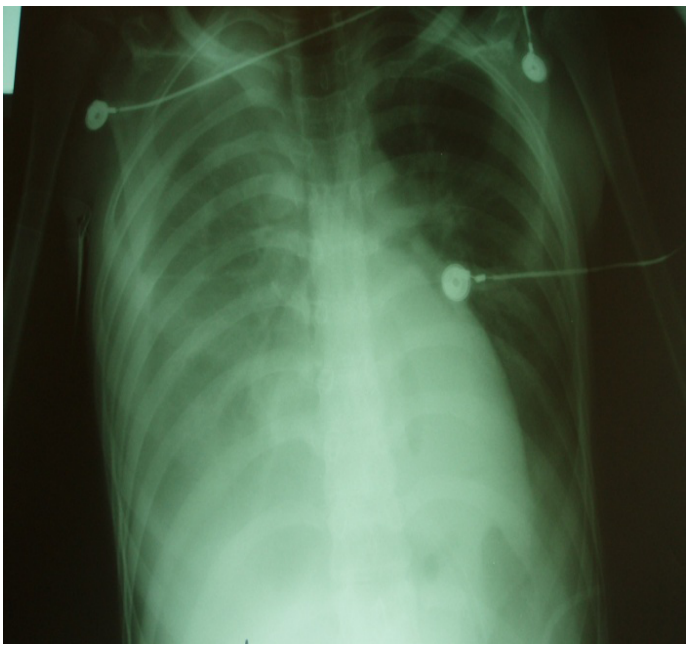

Figure 1: Excessive right hydrothorax on chest film

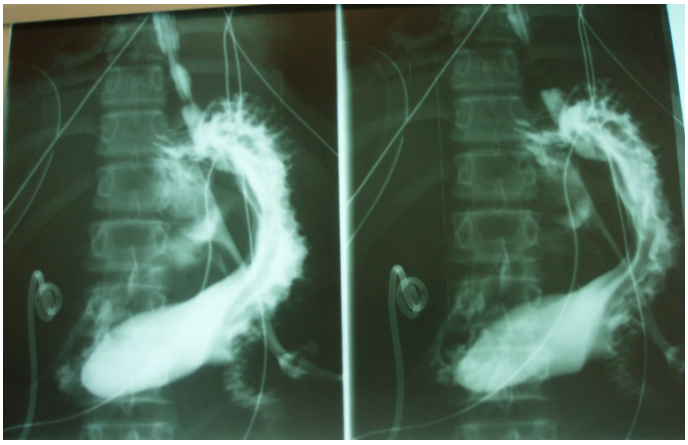

Figure 2: Contrast media leakage revealing perforation on esophagogram (arrows)

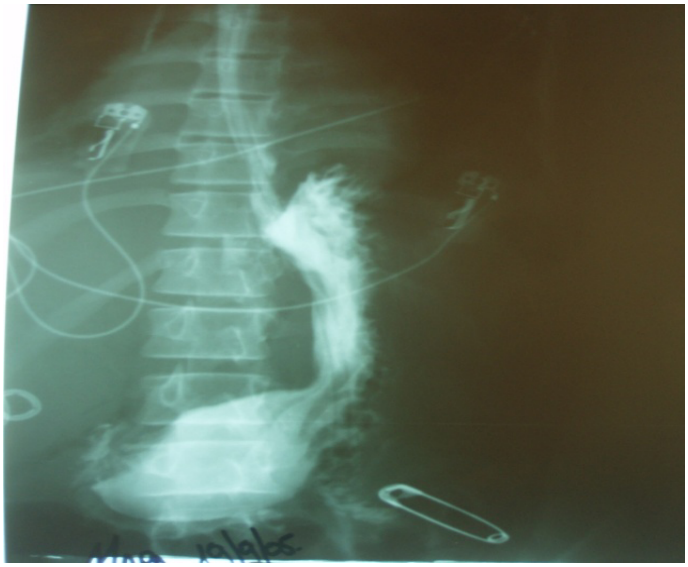

Figure 3: Esophagogram with no evident perforation

parenteral nutrition TPN and broad spectrum antibiotics. One week later, a new esophagogram excluded any leakage (Figure 3). Multiple septic episodes were conservatively treated and after a 46-days ICU stay, the patient was discharged to the ward in good general condition and established oral feeding. At follow up, he remained asymptomatic one year later.

Although the indicated surgical perforation repair did not achieve impermeability, the patient survived from chest and abdominal sepsis most probably because of an internal hollow visceral fistula formation.

Discussion

Candida infection remains an extremely rare cause of esophageal perforation. We were able to indentify only few cases in the current English language literature [2-5]. Esophageal candidiasis is most common in patients with HIV infection, hematological malignancies or after organ transplantation. Gock et al. reported esophageal perforation due to transmural Candida infection in a patient with actinic skin lesions after radiation therapy for vaginal carcinoma [2]. The patient died from septic shock and multi-organ failure, despite esophageal resection. Jones et al. reported fatal Candida esophagitis in two patients after renal transplantation who developed an esophagomediastinal fistula or an esophagobronchial fistula, despite prophylactic oral nystatin treatment [3]. Gaissert et al. reported two cases of nonfatal esophageal perforation related to invasive candidiasis, one after acute leukemia and neutropenia and one after diagnostic esophagogastroscopy for esophageal candidiasis [4]. Aghdam MR et al reported a case of invasive esophageal candidiasis with chronic mediastinal abscess and fatal pneumomediastinum. This case treated with long-term immunosuppression for psoriatric arthritis and died shortly after admission [5]. To our knowledge, our patient is the only case of spontaneous esophageal perforation related to esophageal candidiasis while immunocompetent and without previous esophagogastroscopy.

Esophageal and oropharyngeal candidiasis often occur simultaneously and many patients, as in our case, remain asymptomatic [6]. The prevalence of candida esophagitis might be changing today's rapidly among non-HIV infected patients [7]. In our patient, the presence of white mucosal plaques on the oropharynx and the isolation of Candida albicans from pleural and peritoneal fluid cultures supported the diagnosis of Candida esophagitis, as diagnostic esophagoscopy and biopsy were contraindicated after surgery.

The diagnosis of esophageal perforation may be confirmed by water-soluble contrast esophagogram (Gastrografin®). Although barium is superior in demonstrating small perforations, it causes an inflammatory response in the mediastinal or pleural cavities and is therefore not used as the primary diagnostic study. In our patient, the leakage was confirmed by a water-soluble contrast esophagogram.

Successful treatment of invasive Candida infection complicated by esophageal perforation requires both long-term systemic antifungal therapy and urgent surgical intervention. The causative organism is mainly Candida albicans. Infectious Diseases

Society of America (IDSA) guidelines for esophageal candidiasis, recommend fluconazole administration [8] but in refractory cases we can use other azoles as voriconazole or echinocandins that appear to be as effective as fluconazole [8,9]. Our patient was treated with intravenous voriconazole.

In some cases an urgent esophagectomy and cervical esophagostomy are performed followed in a later phase, by a substernal gastric conduit with cervical esophageal anastomosis [4]. On the other hand, esophageal perforation can be treated with primary closure, if recognized early [10]. Primary closure using a transabdominal approach has also been described [11]. Our patient presented within 12 hours after perforation, so an urgent laparotomy was performed and primary closure was decided. Seven days later a right thoracotomy was performed for relapsing hydrothorax with no specific findings. A second-look laparotomy was necessary for the drainage of a newly formed left subdiaphragmatic abscess. Although repair of the tear did not achieve impermeability, the patient survived from chest and abdominal sepsis probably because of an internal hollow visceral fistula formation.

Case reports have demonstrated the feasibility of treating esophageal perforation with a covered, self-expanding esophageal stent [12]. Treatment also includes continuous nasogastric suction and parenteral nutrition.

Since only a few cases have been reported so far with poor long-term outcome, no established treatment regimen has been validated.

In summary, spontaneous esophageal perforation related to esophageal candidiasis is extremely rare and even with aggressive management, prognosis remains very poor. Nevertheless, in selected cases, treatment is not futile, and every effort should be made in order to gain maximal profit from any available therapy.

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