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Case Report

Severe esophageal candidiasis in a megaesophagus patient: a rare case

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ABSTRACT

Megaesophagus is a largely dilated esophagus, an uncommon condition that could be the end-stage of preceding esophageal achalasia. Chronic food stasis such as in achalasia and megaesophagus could cause fungi infection in the esophagus. This report aimed to raise awareness of these rare cases. A 68-year-old male with two decades of swallowing difficulty was referred to Sardjito Hospital. Computed tomography esophagography showed megaesophagus and bird beak sign suggesting achalasia. Both esophagoscopy and esophageal tissue biopsy showed consistent results of severe esophageal candidiasis (EC). The patient was also suspected to have a mass at the distal part of the esophagus and gastric cardia, which could also contribute to the esophageal obstruction. We consulted the gastroenterologist for gastrostomy feeding and exploration of the suspected tumor. EC is known mainly in patients with immunodeficiency, while food stasis is a lesser-known cause. End-stage achalasia not only could lead to megaesophagus, but chronic food stasis is the perfect environment for fungi growth. Other causes of food stasis such as distal esophageal and gastric cardia tumors could also be the predisposition of EC. We reported a rare case of a patient with EC as a complication of megaesophagus and esophageal achalasia.

Keywords: Esophageal candidiasis, Megaesophagus, Esophageal achalasia, Gastroesophageal junction tumor

INTRODUCTION

Megaesophagus, a largely dilated esophagus, is an uncommon condition in esophageal diseases. It is mostly known to be related to the higher prevalence of Chagas disease, especially in Western countries. However, other etiology might also cause this entity, such as esophageal achalasia. This is even rarer, as only 5% of patients with achalasia progress to this end-stage state of megaesophagus. To complicate things, chronic food stasis such as in achalasia and megaesophagus could also cause fungi infection in the esophagus.[1-3]

Candida is a symbiont of the esophagus, which could become pathogenic in predisposed patients such as immunocompromised ones. Esophageal candidiasis (EC) is the second most common candidiasis in the gastrointestinal tract after oropharynx, while *Candida albicans* is the most common pathogen (88%) in infectious esophagitis, followed by herpes simplex virus (10%) and cytomegalovirus (2%). Several distinct species of Candida are the culprit, such as *C. albicans* as the most common, *C. glabrata*, *C. tropicalis*, and *C. krusei*. The invasion ability of Candida is enhanced after the host's immune system is impaired. Its morphology, adherence to tissues, and production of extracellular proteases are the hallmark of Candida's pathogenicity.[3-5]

EC manifests as unspecific symptoms such as odynophagia, dysphagia, and retrosternal pain. Thus, clinicians should always consider this entity as one of the possible diagnoses in patients presenting with complaints as such. The diagnosis itself is relatively straightforward with esophagoscopy and esophageal tissue biopsy.[3,4]

The treatment of EC is not complex at all; often, the patient will show improvement with systemic antifungal.[3,4] However, severe degree of infection and

other conditions such as megaesophagus, achalasia, mass at the distal part of the esophagus, or gastric cardia might complicate the management of the patients. Thus, this report aimed to raise awareness of such cases.

CASE REPORT

A 68-year-old male was referred to Sardjito Hospital, a tertiary referral hospital, with gradually progressing swallowing difficulty for the past two decades. He could drink liquid effortlessly, but great effort was needed to swallow solid food. The symptoms worsened rapidly in the past six months, up to the point he could only drink liquid, which would be followed by regurgitation of foul-smelling bolus after a certain amount of intake. He lost approximately 10 kg of his weight during this period. He was a heavy smoker, while his other medical history was not remarkable.

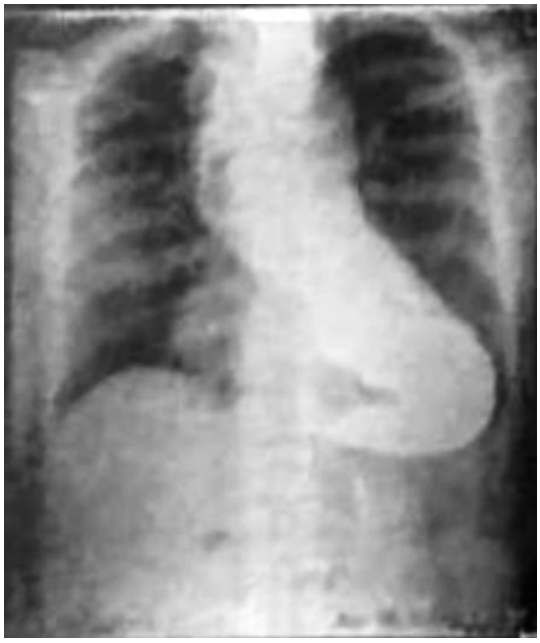

Figure 1: The esophagogram of the patient showed barium contrast filling a dilated esophagus, tapering at the distal end (bird beak sign).

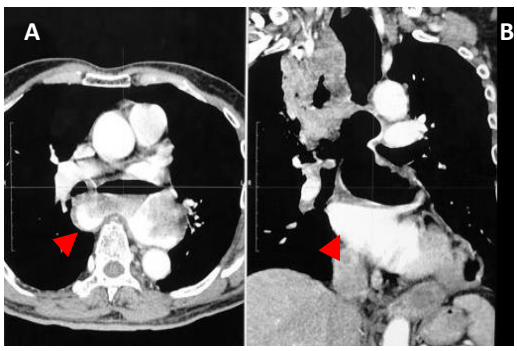

Figure 2: CT esophagography with double-contrast (esophageal and vascular). Arrowhead, showing megaesophagus on axial (A) and coronal plane (B).

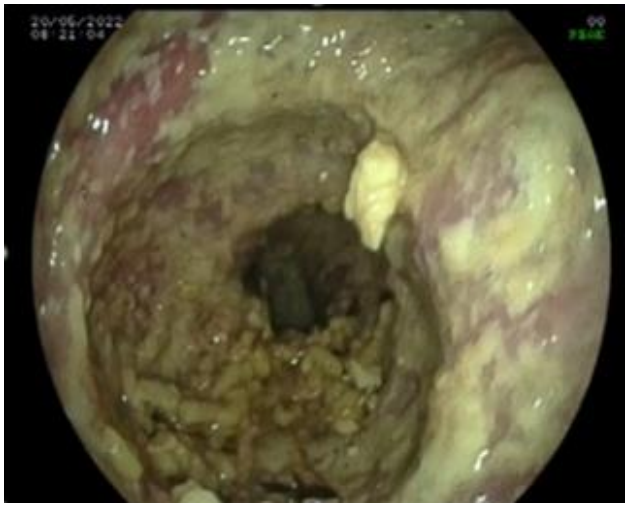

Figure 3: Flexible esophagoscopy revealed white plaque covering the entire length of the esophageal wall, food debris, and the desquamated plaque.

The esophagogram performed at the previous hospital showed a bird beak sign of a dilated esophagus tapering at the distal end, suggesting esophageal achalasia (Figure 1). The patient was then referred to Sardjito Hospital for further workup and management. Initial physical examination showed no remarkable finding. The patient could only swallow liquid, some parts were regurgitated a few seconds after swallowing. Double-contrast computed tomography scan confirmed the esophagogram's result of thoracal megaoesophagus with esophagogastric junction narrowing (Figure 2).

The flexible esophagoscopy revealed dilated esophagus with white patches covering the entire length of the esophagus wall. The esophagus was filled with food debris and desquamated plaque. The distal part of the esophagus was narrowed, hindering further scope advancement. We suspected a possible mass distal to this site which could not be visualized either by the scope or imaging. We then decided to carry out biopsies at four sites of the esophagus wall. The histopathology examination discovered pseudohyphae and spores, compatible with candidiasis, implicating suppurative fungal esophagitis.

We then consulted the gastroenterologist for gastrostomy tube placement and exploration of the possible gastroesophageal junction mass. Systemic antifungal was given in the meantime. However, the patient refused further treatment and lost to follow up.

DISCUSSION

There is no predominance of sex in EC. The median age of a person with EC is 55.5 years old. EC's incidence rate ranges from 0.32-5.2% in the general population.[5] The clinical manifestations of EC are closely related to the severity of esophageal mucosal damage, such as odynophagia, dysphagia, and retrosternal pain. Other less

common symptoms are abdominal pain, heartburn, weight loss, diarrhea, nausea, vomiting, and melena.[3]

The mucous film of the esophagus is protected by the mechanical barrier of the nonkeratinized stratified squamous epithelium. This protection allows *Candida* to make up 20% of the commensal that colonizes the esophagus. An impairment of the immune system or local lesions in the upper esophageal cortex can lead to the proliferation and colonization of fungi.[6]

Although some patients might also have oropharyngeal thrush on examination, the absence of thrush does not preclude the diagnosis of esophagitis.[4] As demonstrated in this case, even with a highly severe infection of *Candida* in the esophagus, the patient had a somewhat normal oropharyngeal appearance.

EC was commonly found in immunocompromised patients with human immunodeficiency virus (HIV). However, this trend is decreasing due to the effectiveness of HIV treatment. There is a rise of EC in non-HIV patients, with the risk factors of using proton-pump inhibitors or other medications such as antibiotics and corticosteroids. Food and secretion stasis in the esophagus, such as in cardiac achalasia, could also lead to the overgrowth of fungi and the development of EC.[3,6] However, many patients reported not having these predisposing factors. In their report, McDonald mentioned the limited data on the attributable cause of EC, thus underlining the need for further investigation.[7] Our patient presented with achalasia and megaesophagus which led to prolonged food stasis and eventually, EC.

Other causes of food stasis, such as tumors at the distal part of the esophagus and gastric cardia were also the predisposition of EC.[8] We still could not rule out a tumor at the gastroesophageal junction in this patient. Small lesions might not be visualized in imaging, thus prompting the need for direct intraluminal visualization using gastroscopy.[9]

Malignancy could also affect the EC formation, as it damages the mucosa and, in turn, impairs the antifungal host defense. On the other hand, several reports also noted the possibility of esophageal carcinoma developing after prolonged EC. There is increasing evidence that Candida has carcinogenic properties, with reports of oral and esophageal squamous cell carcinoma in patients with chronic candidiasis.[8] Although the mass in our case is still not yet visualized, the possibility of it causing EC or the other way around must not be excluded.

Endoscopic findings of white plaques, white exudates, mucosal breaks, and classic clues such as erosions and ulcers are broadly known to suggest EC diagnosis. *Candida* adheres to the esophageal mucous membrane and forms yellow-white patches that cannot be washed with water irrigation. These patches could be found at the distal or proximal part or throughout the esophageal walls.[3,6] We reported a very severe, possibly chronic, EC case whose esophageal wall has been covered entirely by yellow-white patches with no visible normal mucosa.

It was important to note, however, many low-organism burden cases were not identified from endoscopic examination only. A 2017 study found esophagoscopy had low sensitivity and high specificity rates in diagnosing EC of 46.2% and 100%, respectively. Furthermore, claims of acute inflammation as a characteristic feature of EC have also been debunked. Up to 30% of EC cases were not acutely inflamed. When it existed, inflammation was often mild or localized. Thus, endoscopy alone was not reliable in diagnosing EC.[3,6]

Isolation of *Candida* from the sputum and stool specimens is also not specific for EC since it is a normal mycotic flora in the oral and gastrointestinal tract. Only when they grow unchecked and create imbalance, does the EC develop. The patient could be suspected to have EC if they show typical clinical manifestation, *Candida* found in microbial cultures, and high-risk factors are present. However, a definitive diagnosis could only be achieved using histopathology examination from an esophageal specimen biopsy.[3,6]

Histopathology characteristic of EC is at least one fungal pseudo hyphae found which is morphologically compatible with *Candida*, infiltrating detached or intact squamous epithelium.[6] EC usually responds well to systemic antifungal therapy rather than topical, unlike oral candidiasis. Antifungal medications are advised to be given intravenously daily for patients who cannot tolerate oral medication. This regimen could then be changed to oral once the patient can tolerate oral intake.[3] However, the presenting esophageal and gastric mass poses a challenge in that the patient might not be able to tolerate oral intake for a more extended period compared to other cases of EC. Thus, we decided to proceed with gastrostomy as a means of medication and food intake.

CONCLUSION

We reported a rare case of a patient with EC as a complication of megaesophagus and esophageal achalasia.

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