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CASE REPORT

상피하종양에 대한 내시경 점막절제술 후 발생한 식도 방선균증 1예

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Case of Esophageal Actinomycosis Occurred after Endoscopic Mucosal Resection for Subepithelial Tumor

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Esophageal actinomycosis is a rare, chronic granulomatous disease caused by Actinomyces species. Endoscopy and biopsy are essential for making a diagnosis. This paper reports a case of esophageal actinomycosis that developed after an endoscopic mucosal resection (EMR) for a subepithelial tumor (SET). A 74-year-old male patient had a 3 cm flat, smooth elevation in the esophagus without symptoms. The SET was partially resected, and histology revealed “nonspecific degenerated mesenchymal tissue”. Three months later, the patient exhibited a persistently large ulceration at the EMR site, and a biopsy revealed actinomycosis. CT of the chest and abdomen revealed no abnormal findings. Ampicillin treatment was administered for six months, and the ulceration on the esophageal SET improved. **(Korean J Gastroenterol 2023;82:137-139)**

Key Words: Esophageal actinomycosis; Endoscopic mucosal resection

INTRODUCTION

Esophageal actinomycosis is caused by Actinomyces species, which are gram-positive anaerobic or microaerophilic bacteria belonging to the family Actinomycetaceae.[1] Actinomycosis typically occurs in patients who are chronically ill or immunocompromised but is extremely rare in healthy adults, particularly in the esophagus.[2] Most patients with esophageal actinomycosis present with dysphagia, weight loss, or anemia.[3] Treatment requires prolonged antibiotic therapy for six–12 months. This paper presents a case of esophageal actinomycosis without clinical symptoms after endoscopic mucosal resection (EMR) for a subepithelial tumor (SET).

CASE REPORT

A 74-year-old male with no significant medical history underwent a routine health checkup. Esophagogastroduodenoscopy (EGD) revealed a flat, smooth, and elevated SET measuring 3 cm in the distal esophagus (Fig. 1A). The patient had no symptoms associated with the lesions. We obtained individual written informed consents for the procedure. An esophageal EMR was performed to remove a portion of the tumor for

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a histopathological examination (Fig. 1B). The histological examination revealed nonspecific degenerated mesenchymal tissue with calcification. Three months later, a follow-up EGD showed large ulceration with easy bleeding on the esophageal SET (Fig. 1C). A histological examination revealed actinomycotic sulfur granules with filamentous structures, which are characteristic features of actinomycosis. (Fig. 2). The patient showed no symptoms, such as dysphagia, chest pain, or weight loss. The laboratory tests showed a normal white blood cell count (WBC: 4,900 /mm^3), 49.7% neutrophils, and a 0.03 mg/L CRP level. The hemoglobin level (12 g/dL) and platelet count (146,000 /mm^3) were decreased slightly. CT of the chest and abdomen revealed no abnormal findings.

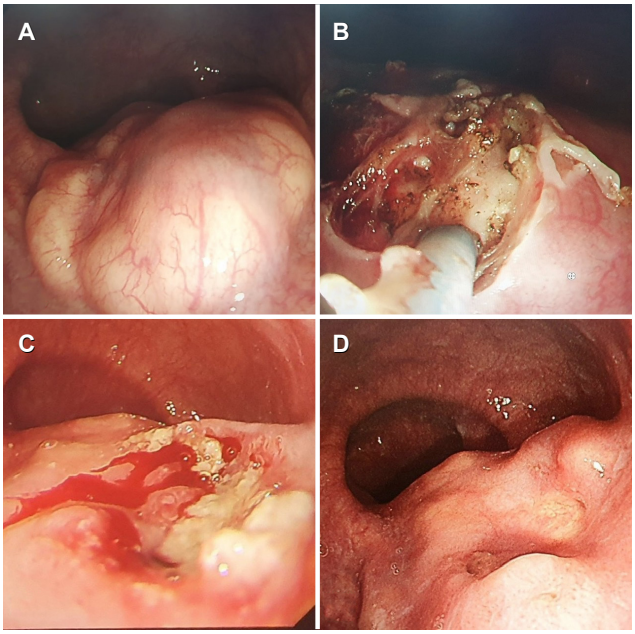

Fig. 1. Endoscopic findings. (A) Flat lobulated smooth elevation was noted, (B) EMR was done. (C) Three months later, a large ulcer on the Esophageal SET was seen. (D) The ulcer was improved markedly four months after antibiotic treatment. EMR, endoscopic mucosal resection; SET, subepithelial tumor.

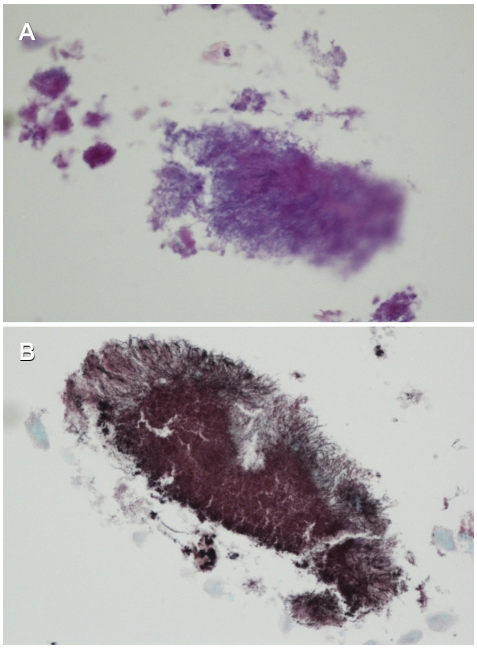

Fig. 2. Histopathological findings. (A) Bacterial colony with hematoxyphillic filamentous bacteria was seen (hematoxylin & eosin stain, ×400). (B) The edge of a granule showed filaments (Grocott's methenamine silver stain, ×400).

The patient was treated with intravenous ampicillin for three weeks, which was then replaced with oral maintenance. After four months, the ulceration improved markedly, and the histological examination revealed no evidence of actinomycosis (Fig. 1D). The antibiotics were discontinued after six months of treatment.

DISCUSSION

Actinomyces are found in humans in the oral cavity, nasopharynx, gastrointestinal tract, or female genitalia and can cause disease in humans when they enter the body through damaged tissue.[4] Actinomycosis can be observed in the case of dental interventions, after surgery, and in immunocompromised patients, such as those with diabetes mellitus, acquired immunodeficiency syndrome (AIDS), or those who have undergone organ transplantation.[5,6] This disease is more common in the abdomen, pelvis, and oral cervicofacial areas than in the esophagus.[7]

Esophageal actinomycosis is a rare, chronic infectious disease with nonspecific symptoms. Therefore, its diagnosis remains challenging.[8] Most cases of esophageal actinomycosis have been reported with symptoms of dysphagia or painful swallowing. Endoscopy reveals ulcers, fistulas, strictures, or abscesses in the esophagus.[9] In the present case, the ulcer resulting from the diagnostic EMR did not improve with treatment. Therefore, a biopsy was performed to confirm the presence of actinomycosis. The patient did not complain of symptoms, such as dysphagia, pain, or chest discomfort, and he was not immunocompromised. A histology examination is vital for definitively diagnosing the disease and can exclude tuberculosis or cancer while sometimes detecting co-infections, such as Candida. The characteristic features of acti-

nomycosis include sulfur granules and filamentous structures. Bacterial cultures are of great value in the diagnosis of this disease. On the other hand, not all testing centers can perform the necessary culture because this bacterium requires a special culture medium.

The clinicians' diagnostic orientation greatly helps pathologists diagnose the disease. There have been case reports in which esophageal actinomycosis was observed in patients with normal immunity and no underlying diseases or secondary to esophageal stent insertion or chest trauma.[5,10,11] Localized damage to the esophageal mucosa produces an opportunity for opportunistic infection by actinomycetes, which are part of the normal oral flora.[5] Therefore, the possibility of actinomycetes should be considered even in patients with normal immunity, even if the patient does not complain of symptoms, if there is a risk of local damage to the esophageal mucosa, such as EMR, biopsy, stent insertion, or trauma, or if esophagitis or ulcers do not respond well to treatment.

The mainstay of treatment for actinomycosis is antibiotics lasting six–12 months or longer. The surgical method is only indicated in cases with extensive abscesses and fistulous tracts.[12] High-dose Penicillin G, Ampicillin, or Amoxicillin is administered intravenously for the first two–six weeks, after which alternative oral therapy is indicated. In some cases of penicillin allergy, an appropriate change in antibiotics, such as ceftriaxone, doxycycline, macrolides, or carbapenems, can be selected. Patients should be monitored and evaluated periodically during the treatment period. EGD, blood tests, and CT of the abdomen and chest are also often performed to evaluate the condition and complications of esophageal actinomycosis, helping to improve the prognosis and treatment. The patient was treated with ampicillin for six months, and the esophageal lesion improved.

This is the first case report of esophageal actinomycosis, which developed after EMR. Intractable ulceration after an esophageal injury raises suspicion of actinomycosis as a potential underlying cause.

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