Esophageal Ulcer of Unknown Origin Complicated by Left Atrial Myxoma

Yuji Nishizaki¹, Shinichiro Yamagami¹, Daisuke Hayakawa¹, Shiori Takashima², Osamu Nomura¹, Eiryu Sai¹, Kazuyoshi Kon¹, Shujiro Matsuyama², Sumio Watanabe³ and Hiroyuki Daida⁴

Abstract

Myxoma induces the onset of paraneoplastic syndromes by excreting various humoral mediators and is therefore known to present with diverse symptoms. A 40-year-old woman was admitted to our hospital for the treatment of an esophageal ulcer, the cause of which could not be identified on various examinations. Notably, a left atrial tumor was incidentally found on chest enhanced computed tomography. The esophageal ulcer, which was intractable to conventional therapy, improved with the administration of 5-aminosalicylate, a drug known to inhibit IL-1β. This inhibitory action effectively suppressed the development of myxoma-induced paraneoplastic syndrome.

Key words: esophageal ulcer, myxoma, paraneoplastic syndrome

(Intern Med 54: 1365-1367, 2015) (DOI: 10.2169/internalmedicine.54.3828)

Introduction

Primary cardiac tumors are rare. In contrast, myxoma, which commonly develops in the left atrium, is a benign tumor with the highest morbidity among cardiac tumors (1). Myxoma induces the onset of paraneoplastic syndromes by excreting various humoral mediators, thereby producing diverse symptoms (2). We herein report a case of myxoma in the left atrium that was discovered incidentally during a workup and treatment for an esophageal ulcer of unknown origin. To the best of our knowledge, this is the first report of myxoma complicated by an esophageal ulcer.

Case Report

A 40-year-old woman visited a nearby clinic three months prior to the current admission with a complaint of epigastric pain. Upper endoscopy revealed an esophageal ulcer, and treatment with proton pump inhibitors and a mucosal protectant was administered. A repeat examination with upper endoscopy performed two months after the initial treatment revealed that the esophageal ulcer had worsened. The patient then visited the outpatient division of the Department of Gastrointestinal Medicine at our hospital. Two weeks later, she was hospitalized due to increased thoracic and epigastric pain. She had a past history of tonsillectomy at 30 years of age and herpes zoster at 35 years of age. In addition, she had been visiting a nearby clinic for treatment for panic disorder, and had been prescribed paroxetine hydrochloride, alprazolam and ethyl loflazepate. She had no history of drug use, food allergies, smoking or alcohol drinking.

After admission, an esophageal biopsy was performed to identify the cause of the esophageal ulcer; however, no evidence of malignancy, fungal infection or Crohn’s disease was apparent. A blood examination also showed no findings of cytomegalovirus, herpes simplex virus or human immunodeficiency virus, and there were no findings indicative of...
Behcet’s syndrome, such as aphthous ulcers in the oral mucosa or characteristic symptoms in the eyes or the skin. The patient’s medications for panic disorder were discontinued, and a proton pump inhibitor and mucosal protectant were administered under a nil per os (NPO) status to protect the esophageal mucosa. Upper endoscopy performed on day 23 after admission did not show any signs of improvement (Fig. 1A).

A left atrial tumor was found incidentally on chest contrast-enhanced computed tomography (Fig. 2A). The patient’s heart sounds were normal and she displayed no symptoms of heart failure, emboli or neurological disorders. A tumor measuring 24×14 mm in size was noted on transthoracic echocardiography (Fig. 2B), which was strongly suspicious of a left atrial myxoma. In addition, the serum interleukin-6 (IL-6) level was elevated (10.7 pg/mL).

On hospital day 24, the patient was transferred to our affiliated hospital for further treatment of the unimproved esophageal ulcer and tumor resection. The esophageal ulcer gradually improved following the oral administration of 5-aminosalicylate [PENTASA® (Kyorin Pharmaceutical, Tokyo, Japan) tablets dissolved in water (dose: 2,000 mg,
twice a day). Fig. 1B shows the findings for the esophagus on upper endoscopy performed after treatment with 5-aminosalicylate. Because the esophageal ulcer improved, we planned to perform elective surgery for the cardiac tumor; however, she developed anemia, and autologous blood donation was required for a long period. Tumor resection was subsequently performed five months after the oral administration of 5-aminosalicylate. The surgery was successful, without any complications, and the administration of 5-aminosalicylate was continued postoperatively, with no recurrence of the thoracic or epigastric pain for three months until today.

**Discussion**

Cardiac myxoma induces the onset of several paraneoplastic syndromes (2) and reportedly produces constitutional symptoms (3, 4). To the best of our knowledge, this is the first report of myxoma complicated by an esophageal ulcer. In the present case, the myxoma was found incidentally during a workup and treatment for an esophageal ulcer of unknown origin.

We hypothesized that the esophageal ulcer in this case was caused by myxoma-induced paraneoplastic syndrome for the following two reasons: 1. the cause of the esophageal ulcer could not be identified on various workups; and 2. the esophageal ulcer, which was intractable to conventional therapy, improved after the administration of 5-aminosalicylate. Originally, 5-aminosalicylate was used to treat inflammatory bowel disease, and there is no firm evidence of its efficacy against esophageal ulcers. However, this drug is known to inhibit IL-1β (5) and successfully suppressed the development of myxoma-induced paraneoplastic syndrome in the current patient. The limitation of this case study is that the increase in the IL-1β level was not examined. Furthermore, why ulcers did not develop anywhere other than the esophagus also remains unclear.

We experienced a very rare case of myxoma occurring in the left atrium that was complicated by an esophageal ulcer of unknown origin. The administration of 5-aminosalicylate effectively treated the esophageal ulcer, which was intractable to conventional therapy. We believe that the IL-1β-inhibitory effects of 5-aminosalicylate suppressed the development of myxoma-induced paraneoplastic syndrome in this case.

The authors state that they have no Conflict of Interest (COI).

**Acknowledgement**

The authors would like to thank Takeshi Asakura, LLB, for his help in translating this case report into English.

**References**

1. Silverman NA. Primary cardiac tumors. Ann Surg 191: 127-138, 1980.
2. Smith M, Chaudhry MA, Lozano P, Humphrey MB. Cardiac myxoma induced paraneoplastic syndromes: a review of the literature. Eur J Intern Med 23: 669-673, 2012.
3. Nishizaki Y, Yamagami S, Myojin M, et al. A murmur-free giant myxoma discovered incidentally on abdominal ultrasonography. Intern Med 52: 2529-2531, 2013.
4. Takizawa T, Sumino H, Kanda T, Kobayashi I, Nagai R, Ichikawa S. An interleukin-6-producing cardiac myxoma associated with mediastinal lymphadenopathy. Cardiology 92: 275-277, 1999.
5. Mahida YR, Lamming CE, Gallagher A, Hawthorne AB, Hawkey CJ. 5-Aminosalicylic acid is a potent inhibitor of interleukin 1 beta production in organ culture of colonic biopsy specimens from patients with inflammatory bowel disease. Gut 32: 50-54, 1991.