Case Report

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Endoscopic submucosal dissection of coexisting early esophageal carcinoma and leiomyoma: a case report and review of the literature

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Abstract

The occurrence of early esophageal cancer located within an area of leiomyoma is extremely rare, and its clinical features and treatment methods have not been well described. We herein report the clinical characteristics, diagnosis, and treatment methods of early esophageal cancer that developed on top of a leiomyoma in the upper third of the esophagus in a 78-year-old woman. All tumor marker concentrations were normal. The leiomyoma was correctly diagnosed as a submucosal tumor by endoscopy and endoscopic ultrasonography. Endoscopic biopsy revealed esophageal squamous cell carcinoma. Both lesions were successfully treated by endoscopic submucosal dissection. The patient was followed up for 6 months without recurrence. Endoscopic submucosal dissection was a successful initial treatment method for esophageal carcinoma coexisting with esophageal leiomyoma in this case.

Keywords

Endoscopic submucosal dissection, squamous cell carcinoma, leiomyoma, esophageal cancer, coexisting tumors, endoscopic biopsy, case report

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Introduction

Esophageal squamous cell carcinoma (SCC) and esophageal submucosal tumors (SMTs) are distinct neoplasms originating from different cell layers.[1] Although simultaneous development of such carcinomas within the esophagus is common,[2,3] early esophageal cancer located within an area of an SMT is relatively rare. We encountered a patient with acid reflux, heartburn, and dysphagia who was eventually diagnosed with early esophageal carcinoma combined with a leiomyoma. After endoscopic submucosal dissection (ESD), the patient's clinical symptoms were relieved.

Case report

A 78-year-old man presented to the inpatient service of the Department of Gastroenterology, the First People's Hospital of Changzhou with a 6-month history of acid reflux and heartburn and a 2-week history of dysphagia after meals. He had no history of melena, weight loss, fever, or joint pain. However, he had a 10-year history of hypertensive disease. Physical examination findings upon admission were unremarkable. As shown in Table 1, his laboratory evaluation results, including his coagulation function, liver function, and serum tumor markers, were normal. A contrast-enhanced thoracic computed tomography scan showed thickening of the esophageal wall on the right side of the upper third of the thoracic esophagus (Figure 1). Endoscopy revealed a protruding lesion with a light reddish surface of 0.3×0.3 cm within a shallow depressed area in the proximal third of the esophagus. The margin of the light reddish area (arrows in Figure 2(a), (b)) showed disappearance of the vascular network in the mucosa. Endoscopic biopsy revealed highly differentiated SCC. Endoscopic ultrasonography (EUS) demonstrated a hypoechoic tumor, 11 mm in diameter, confined to the submucosa with a smooth, well-demarcated outline and intact muscularis propria layer. Moreover, EUS demonstrated that the SCC overlay a leiomyoma originating in the muscular layer, suggesting that the cancer may have invaded only as far as the mucosal layer (Figure 2(c)). The tentative diagnosis was superficial type IIc esophageal cancer based on the Guidelines for Clinical and Pathologic Studies of Carcinoma of the Esophagus from the Japanese Society for Esophageal Disease.[4] After obtaining informed consent, ESD was carried out by aspiration lumpectomy. The

Table 1. Laboratory observations upon admission.

Characteristics	Index	Reference range
Blood		
WBC count ($\times 10^9$/L)	8.36	4.0–10.0
RBC count ($\times 10^{12}$/L)	4.0	3.5–5.5
Hb (g/L)	131	120–155
PLT count ($\times 10^9$/L)	180	100–300
Coagulation function		
PT (s)	11.5	9.0–13.0
APTT (s)	23.9	19.0–34.5
Liver function		
ALT (μ/L)	24	9–50
AST (μ/L)	36	10–45
γ-GT (μ/L)	32	10–60
ALP (μ/L)	84	40–125
TP (g/L)	72.3	60–82
ALB (g/L)	45.0	35–55
CHE (μ/L)	7975	3000–8000
Serum tumor markers		
AFP (ng/mL)	4.25	0–8
CEA (ng/mL)	1.05	0–5
CA19-9 (U/mL)	9.16	0–37
T-SPOT	Negative	Negative

WBC, white blood cell; RBC, red blood cell; Hb, hemoglobin; PLT, platelet; PT, prothrombin time; APTT, activated partial thromboplastin time; ALT, alanine transaminase; AST, aspartate aminotransferase; γ-GT, glutamyl transpeptidase; ALP, alkaline phosphatase; TP, total protein; ALB, albumin; CHE, cholinesterase; AFP, alpha fetoprotein; CEA, carcinoembryonic antigen; CA19-9, carbohydrate antigen 19-9.

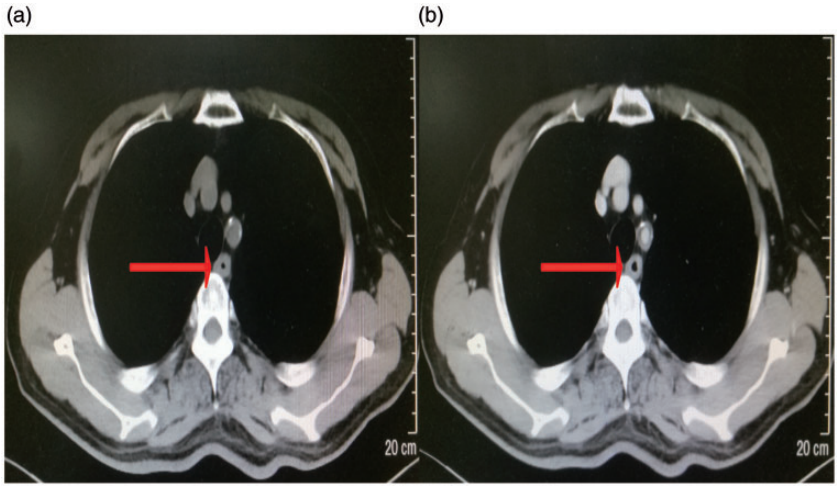

Figure 1. Conventional and enhanced computed tomography (CT) images. Thoracic contrast-enhanced CT showed thickening of the esophageal wall on the right side of the upper third of the thoracic esophagus. (a) Conventional CT images. (b) Enhanced CT images.

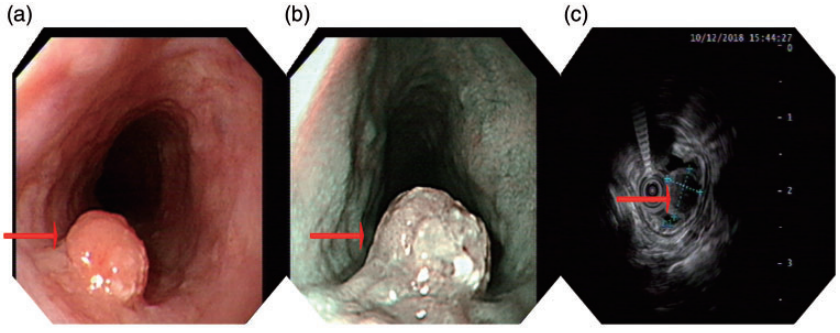

Figure 2. Endoscopic appearance. (a) A protruding lesion with a smooth surface was observed within a shallow depressed area in the proximal third of the esophagus. (b) Chromoendoscopy with narrow band imaging demonstrated a polypoid lesion and normal mucosa covering part of the surface. (c) Endoscopic ultrasonography demonstrated a hypoechoic tumor, 11 mm in diameter, confined to the submucosa with a smooth, well-demarcated and smooth outline and intact muscularis propria layer.

resected specimen measured 35×33 mm and consisted of two independent histological types of neoplasms. One was a leiomyoma that originated from the muscularis mucosa. Histopathological examination revealed low overall cellularity and interlaced smooth muscle cells with hypovascularity and no mitosis. Immunohistochemically, the tumor cells were positive for smooth muscle actin but negative for CD117 and CD34. The other neoplasm was a highly differentiated esophageal SCC confined to the mucosa overlying the leiomyoma (Figure 3). The resected specimen showed an M2 (lamina propria) esophageal cancer with negative lateral margins. There was no evidence of lymphovascular invasion. The patient's clinical symptoms improved significantly within

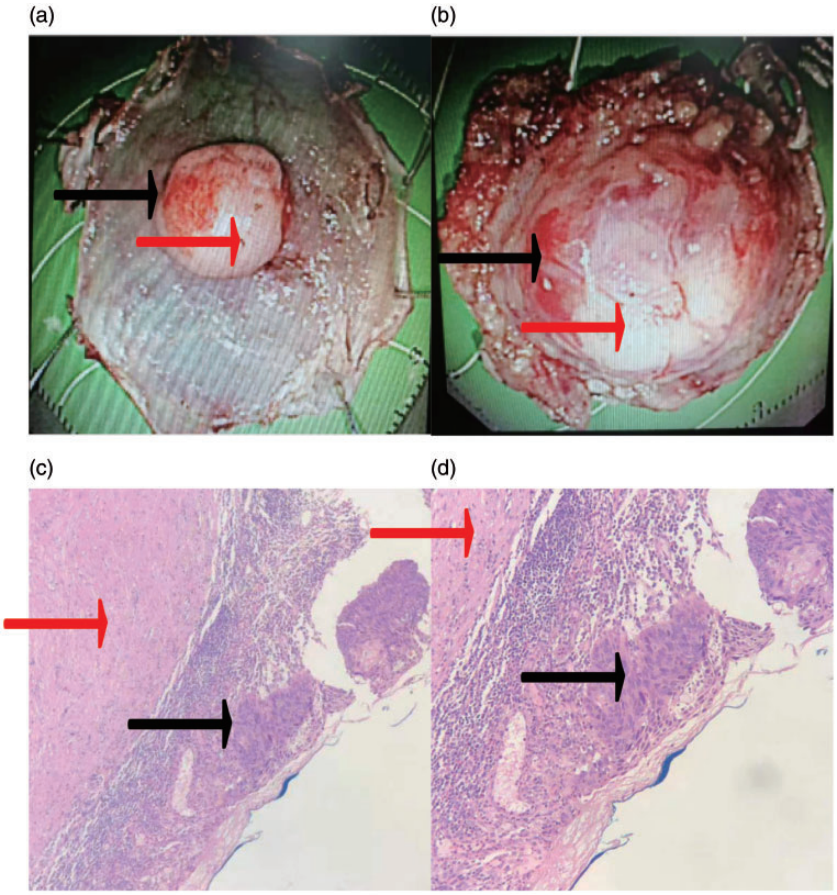

Figure 3. Histological examination. (a, b) The tumor in the resected specimen had a shallow depression and measured 35 × 33 mm. Photomicrograph of a cross section of the resected lesion, showed a highly differentiated squamous cell carcinoma (black arrows) and leiomyoma (red arrows) of the esophagus. (c) Magnification, 10×. (d) Magnification, 40×.

2 months. Repeated endoscopy at 6 months showed no evidence of recurrence.

Discussion

SCC is the most common tumor of the esophagus.[1–3] The incidence of esophageal adenocarcinoma associated with Barrett's esophagus and other types of esophageal tumors, benign or malignant, has gradually increased in recent years.[5] Moreover, the frequency of coexisting SCC and nonepithelioid tumors has reportedly increased.[1–3,6] Such coexisting tumors occur with two presentations: one is the separate type, in which the SCC and the benign tumor are two separate lesions, and the other is the overlying type, an extremely rare type in which the SCC covers the benign tumor. In the present case, the patient had the overlying type. The leiomyoma was first diagnosed by EUS, which demonstrated clear margins and a smooth contour, suggesting a benign tumor. Endoscopic biopsy then revealed SCC limited to the epithelium. To our knowledge, only two cases of SCC in the

epithelium overlying a leiomyoma have been reported to date.[1,2] This pathogenesis of the overlying type may be due to intraluminal protrusion of the SMT, leading to carcinoma development.

ESD is a safe treatment for SCC limited to the lamina propria mucosae[7] and an SMT originating from the muscularis mucosa.[8] In contrast, if a large SMT originates from the muscularis propria, ESD might cause serious complications such as massive bleeding or esophageal perforation.[7,8] Hence, thoracoscopic resection or open surgery is the treatment of choice for these patients.[9,10] In recent years, two reports[11,12] have described the performance of ESD in patients with SCC overlying SMTs. However, accurate diagnosis of the depth of SMTs using EUS is very important. For example, Mizobuchi et al.[13] reported a case in which an early esophageal SCC was overdiagnosed because the leiomyoma appeared to be a component of the carcinoma. Hence, several diagnostic modalities, such as EUS and computed tomography, allow esophageal leiomyomas to be accurately diagnosed. In the present case, we achieved a precise diagnosis and then successfully stripped the combination tumor using ESD. Eight weeks after endoscopic resection, the patient's clinical symptoms had disappeared and no evidence of tumor recurrence was observed.

The present report describes an unusual presentation of esophageal SCC located within an area of leiomyoma in the upper third of the esophagus, emphasizing the importance of accurate diagnosis of the depth of invasion using EUS. In addition, a noninvasive endoscopic therapeutic procedure should be chosen when a coexisting superficial carcinoma overlies a leiomyoma.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Ethics statement

This study was approved by the Ethics Committee of the First People's Hospital of Changzhou. Informed written consent was obtained from the patient for publication of this case report and accompanying images.

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