

Endoscopic resection of co-existing severe dysplasia and a small esophageal leiomyoma

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INTRODUCTION

Leiomyoma is the most common benign mesenchymal tumor in the esophagus^[1,2]. A small and asymptomatic leiomyoma covered with endoscopically normal mucosa does not require surgical treatment such as resection because it has a characteristic endoscopic ultrasonographic appearance, slow growth rate, and negligible risk of malignant transformation^[3]. A few cases involving co-existing tumors such as epithelial tumor and subepithelial tumor in the esophagus have been reported. However, in all of these reports the tumors were larger than 1 cm, were squamous cell carcinoma^[4-8], and most were treated with surgery. A subepithelial lesion smaller than 1 cm combined with epithelial dysplasia is extremely unusual. We present a patient with an epithelial lesion of severe dysplasia overlying a small leiomyoma in the muscularis mucosa of the esophagus. Both lesions were completely removed simultaneously by endoscopic mucosal resection (EMR).

CASE REPORT

A 67-year-old male patient visited our hospital following the incidental finding of a subepithelial lesion at the mid-esophagus. He was a non-smoker, with no previous medical history and had good performance. Esophago-gastroscopic examination showed a protruding mass, measuring 1 cm × 1 cm in dimension, without any ulcer or erosion, covered with normal mucosa (Figure 1A). We examined the lesion using narrow-band imaging (NBI) which showed scattered brown dots, and dilated and tortuous vessels on the top of the lesion (Figure 1B). We judged that the lesion might have dysplasia. Endoscopic ultrasonography was performed in order to determine the depth and nature of the tumor. Findings indicated

Abstract

Leiomyoma is the most common benign mesenchymal tumor of the esophagus. A small leiomyoma covered with endoscopically normal mucosa has a characteristic endoscopic ultrasonographic appearance, slow growth rate, and negligible risk of malignant transformation; therefore the histology does not need to be proven. Synchronous tumors such as an epithelial tumor and small subepithelial tumor in the upper gastrointestinal tract are uncommon. We describe a case with a co-existing small leiomyoma and severe dysplasia in the esophagus which were completely resected by endoscopic mucosal resection.

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Key words: Synchronous tumor; Leiomyoma; Dysplasia

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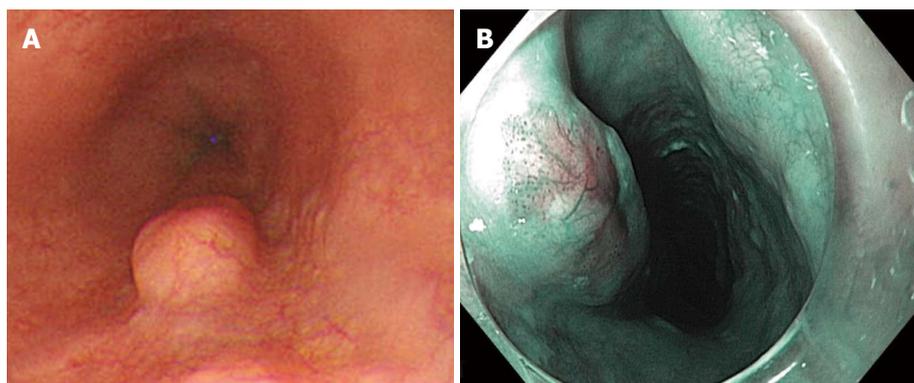


Figure 1 Endoscopic findings. A: Endoscopic findings demonstrate a subepithelial tumor with intact overlying mucosa; B: Chromoendoscopy with narrow band imaging showed scattered brown dots, and dilated and tortuous vessels on the top of the lesion.

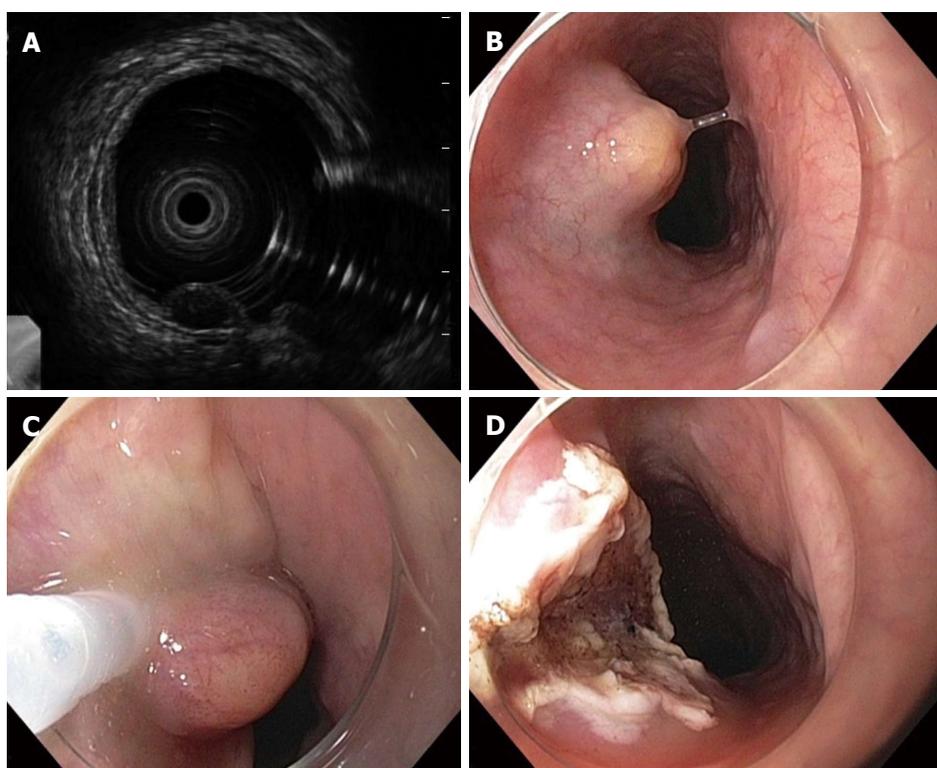


Figure 2 Endoscopic ultrasound findings and procedures for endoscopic mucosal resection. A: High frequency ultrasonographic findings showed a clear homogenous hypoechoic mass derived from the second layer of the esophagus; B: After making a submucosal bleb; C: The subepithelial lesion was captured using a snare; D: No immediate complications occurred after endoscopic mucosal resection.

that the lesion, which measured 5.0 mm × 4.0 mm in size, was a homogenous hypoechoic mass, confined to the second layer, the muscularis mucosa (Figure 2A). We performed a mucosal biopsy. Histological examination of the specimen indicated severe dysplasia. We diagnosed severe dysplasia overlying a small leiomyoma from the muscularis mucosa. Therefore, we planned *en bloc* resection by endoscopy for removal of the synchronous lesions. Endoscopic resection was performed using the EMR method. Following injection of hyaluronic acid into the submucosa, resection was performed using a snare (Figure 2B and C). No complications related to the procedure were observed (Figure 2D). Histopathological

examination of the tumor revealed high-grade dysplasia with a clear resection margin and the results of immunohistochemical analyses revealed that the leiomyoma was negative for C-Kit and CD 34, and positive for smooth muscle actin (Figure 3).

DISCUSSION

The present case of severe epithelial dysplasia overlying a small subepithelial tumor was treated with EMR.

Due to improvements in diagnostic tools and prolonged lifespan rates, the diagnosis of esophageal subepithelial lesions is increasing. Leiomyoma is the most

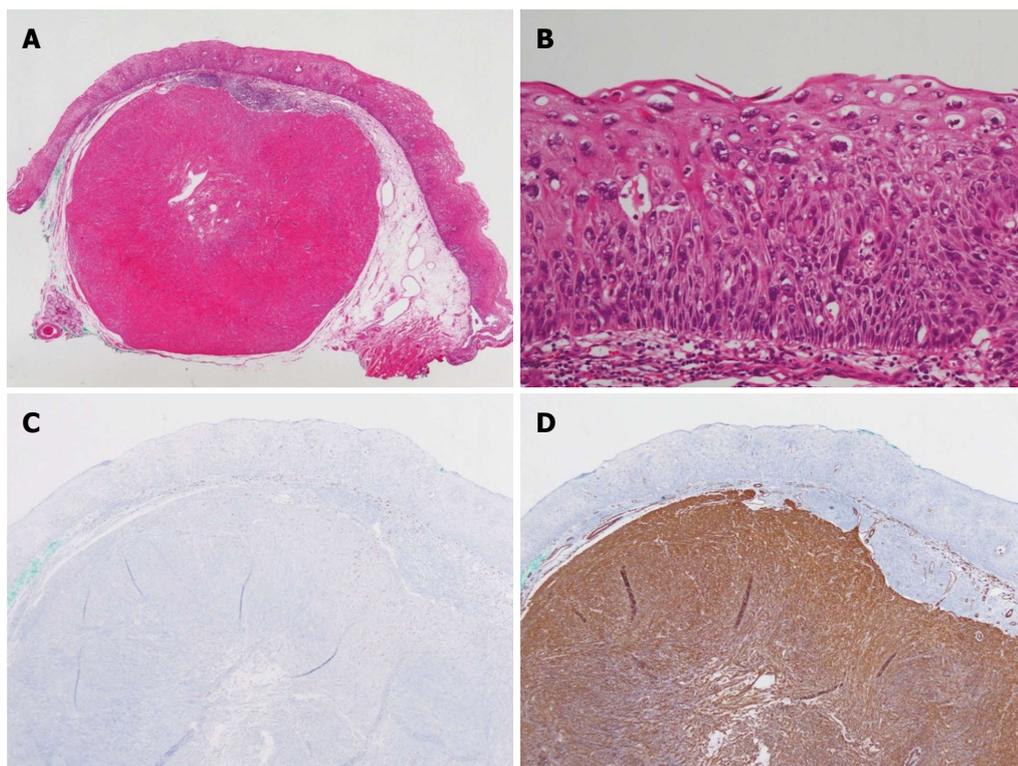


Figure 3 Histopathologic findings. A: Esophageal leiomyoma arising from the muscularis mucosa (HE, 20 ×); B: Atypical cells in all three areas of the epithelium, but not full thickness, showed severe dysplasia (HE, 200 ×); C: The esophageal leiomyoma was negative for CD117 (C-Kit); D: The esophageal leiomyoma showed strong and diffuse positive staining for smooth muscle actin (200 ×).

common subepithelial tumor of the esophagus. Esophageal leiomyomas account for approximately 12% of all gastrointestinal leiomyomas^[9]. Esophageal leiomyomas may occur in the muscularis propria layer or the muscularis mucosa of the esophagus. With the development of the high frequency ultrasonic endoprobe, detection of the origin of leiomyomas, even in small lesions, appears to be easier^[10]. On occasion, those arising from the muscularis mucosa can present as polypoid intraluminal tumors. In a review of 838 cases, only 1% were the polypoid intraluminal type^[11]. The question of whether a leiomyoma can show malignant degeneration is controversial. If it were possible, the risk would be negligible. Thus, when a small leiomyoma covered with normal mucosa is encountered on endoscopy, a biopsy is not recommended^[1].

The co-existence of an epithelial lesion and a subepithelial lesion is rare. Twelve patients in ten case reports with carcinoma located in the mucosa overlying a benign tumor have been reported^[4-6,12]. All cases were squamous cell carcinoma and the subepithelial lesions were larger than 1 cm. Eight patients were treated surgically. However, this is the first case of epithelial dysplasia overlying a leiomyoma measuring less than 1 cm in the esophagus.

It is not clear whether both lesions occurred at the same time and whether there were any causative relationships. However, we suggest that there was synchronous development of the subepithelial and epithelial lesions. After development of a leiomyoma, external stimuli may

change the mucosa. Epithelial dysplasia is the principal precursor lesion of esophageal squamous cell carcinoma. Studies have shown that esophageal squamous cell carcinoma develops through a progressive sequence from mild to severe dysplasia^[13]. Predisposition to esophageal dysplasia may be related to certain carcinogenic stimuli, dietary factors, and individual genetic susceptibility^[13]. The patient was a non-smoker and did not have any relevant medical history. He did not have any risk factors for esophageal cancer. We assume that focal mechanical stimuli might induce mucosa dysplasia. However, determination of a dysplastic lesion on conventional endoscopy is difficult, even when using an iodine stain. The prevalence of severe dysplasia derived from iodine-stained tissue is quite low (< 1%)^[14]. The specificity of NBI for severe dysplasia performed by experienced endoscopists has been reported to be up to 100%^[15]. Therefore, a subepithelial lesion presenting with an intraluminal polypoid mass should be examined closely using electronic chromoendoscopy with NBI and tissue biopsy. However, there is a risk of bleeding and scattering malignant cells with tissue biopsy. Furthermore, scarring after tissue biopsy can create problems in the complete removal of lesions. The examination of simultaneous lesions is important. If NBI findings suggest dysplastic change, it is better to resect the lesion without tissue biopsy. Severe dysplasia overlying a leiomyoma originating from the muscularis mucosa can be removed safely using an endoscopic technique^[10].

In conclusion, a polypoid subepithelial tumor of the

esophagus is usually very small; however, it can occur with simultaneous epithelial dysplastic change. If the lesions are detected early using novel endoscopic imaging, they can be removed endoscopically.

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