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**Case Reports**

# Co-existence of Early Esophageal Carcinoma and Leiomyoma: a Case Report

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There have been several reports of co-existing esophageal squamous cell carcinoma and esophageal submucosal tumor. However, there is no previous report describing a submucosal tumor located within an area of early esophageal cancer. This report presents the case of a 64-year-old man who developed early esophageal cancer with leiomyoma situated within the lesion in the upper third of the esophagus. Since leiomyoma existed within the area of the esophageal cancer, it was misdiagnosed as a component of esophageal cancer and the depth of esophageal cancer invasion was overdiagnosed by endoscopic ultrasonography. Therefore, surgery was chosen as treatment for esophageal cancer. If the leiomyoma had been diagnosed correctly as a submucosal tumor by endoscopy and endoscopic ultrasonography, an endoscopic mucosal resection would have been the therapeutic procedure of choice for an esophageal tumor.

*Key words: esophageal carcinoma – leiomyoma*

## INTRODUCTION

Endoscopic treatment of esophageal cancer has improved quality of life for patients because it is less invasive than open surgery. Thus, endoscopic mucosal resection (EMR) is becoming an important treatment option for patients with early esophageal cancer. In the present case, because the esophageal submucosal tumor (SMT) was located within an area of esophageal cancer, the depth of the invasion of esophageal cancer was diagnosed as more than it actually was. Therefore, open surgery was chosen as the treatment option for the esophageal tumor. There have been several reports of co-existing esophageal squamous cell carcinoma (SCC) and esophageal SMT, for example, leiomyoma (1–8) and lipoma (5,6,9). In these cases, part of the surface of the submucosa was overlaid by superficial esophageal cancer. To our knowledge, there has been no previous report describing an SMT located within the area of early esophageal cancer as a small nodule.

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## CASE REPORT

A 64-year-old man was referred to our department for treatment of an esophageal tumor. Endoscopy demonstrated a sessile polypoid lesion with a smooth surface within a shallow depressed area in the proximal third of the esophagus (Fig. 1). The surface of the depressed area was uneven and irregular. This area did not take up dye at all when sprayed with Lugol's solution (Fig. 2). Endoscopic biopsy demonstrated SCC. Based on these findings, the polypoid lesion was diagnosed as having a component of esophageal cancer. Endoscopic ultrasonography demonstrated a hypoechoic tumor, 13 mm in diameter, located in the submucosa, with clear margins and a smooth contour, with the muscularis propria layer intact (Fig. 3). Esophageal cancer, superficial type, 0–I + IIc was tentatively diagnosed based on the Guidelines for Clinical and Pathologic Studies of Carcinoma of the Esophagus from the Japanese Society for Esophageal Disease (10). It was diagnosed by endoscopic ultrasonography that the depth of tumor invasion was the submucosal layer at the nodule.

Endoscopy also demonstrated other lesions. One was a protruding lesion resembling an SMT, located just oral to the esophagogastric junction. The other was a small protruding tumor, 8 mm in size, in the lower gastric body, with greater

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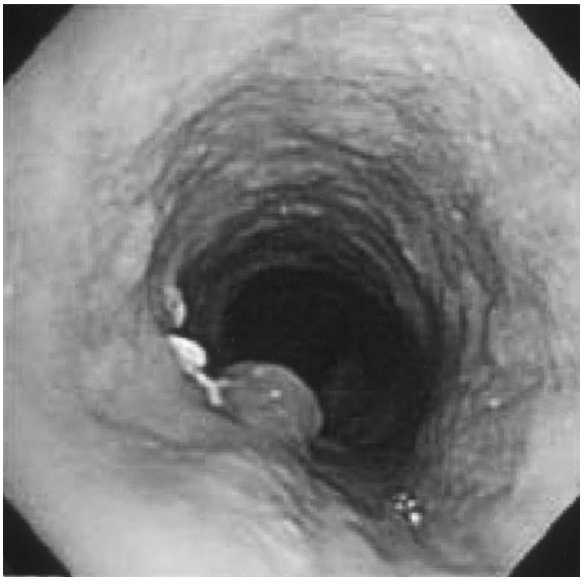

**Figure 1.** Endoscopic examination demonstrated a sessile polypoid lesion with a smooth surface within a shallow depressed area in the proximal third of the esophagus.

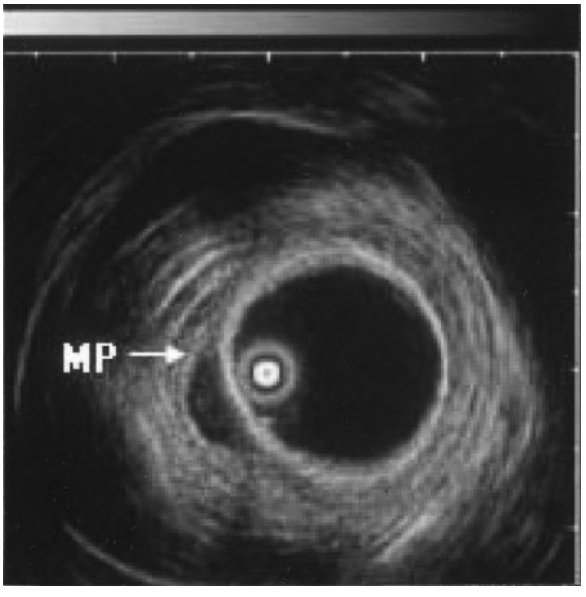


**Figure 3.** Endoscopic ultrasonography demonstrated a hypoechoic tumor, 13 mm in diameter, located in the submucosa, with clear margins and a smooth contour, with the muscularis propria layer (MP: low echoic layer) intact.

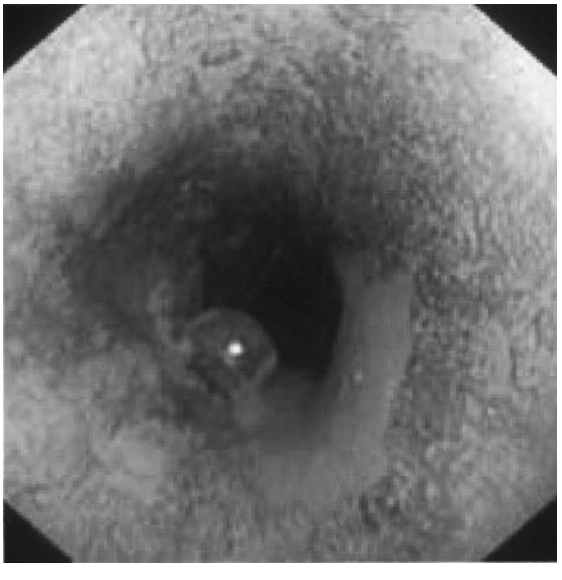

**Figure 2.** Chromoendoscopy with Lugol's iodine solution demonstrated that the sessile polypoid lesion located within the non-staining area and part of the surface was covered by normal mucosa.

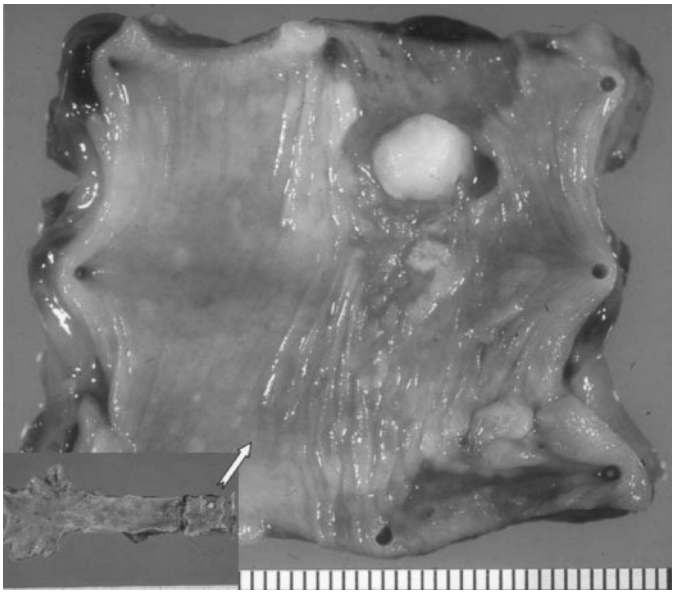

**Figure 4.** The tumor in the resected specimen had a shallow depression, 30 × 15 mm in size, with a whitish nodule.

curvature. Endoscopic biopsy of the gastric tumor demonstrated a well differentiated tubular adenocarcinoma limited to the mucosa. Because we planned resection and reconstruction using the stomach for esophageal substitution for esophageal cancer, EMR was performed using the strip-biopsy technique with removal of the gastric tumor en bloc without complication. Microscopic findings showed adenocarcinoma limited to the mucosa.

In August 2002, subtotal esophagectomy with a three-field (cervical, mediastinal and abdominal) lymph node dissection and esophagogastrostomy was performed through a right thoracotomy and abdominal incision. The tumor in the resected specimen had a shallow depression, 30 × 15 mm in size, with a whitish nodule, 7 × 6.5 mm in size (Fig. 4). Microscopic findings showed poorly differentiated SCC penetrating as far as the lamina propria mucosae, without lymph node metastasis. The nodule within the lesion was composed of interlacing bundles of spindle-shaped tumor cells, and originated in the muscularis mucosae (Fig. 5). No mitotic figures were found. Immunohistochemical staining showed that the tumor was strongly α-smooth muscle actin positive, but S-100 protein, c-kit and CD34 negative. In the light of these findings, the

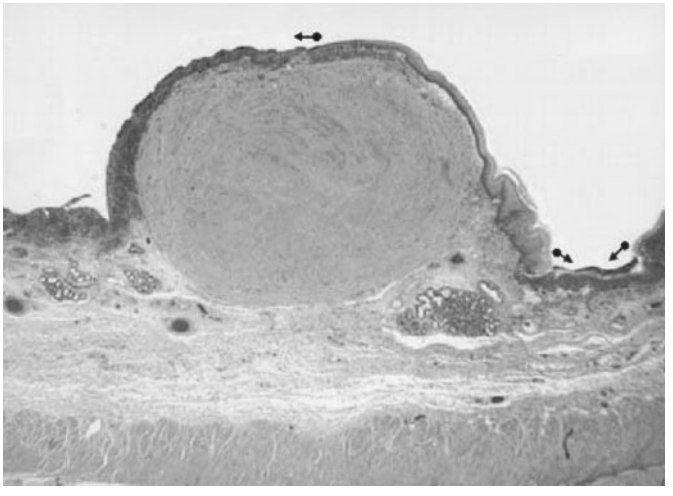

**Figure 5.** Photomicrograph of a cross-section of the resected lesion, showing a poorly differentiated SCC (arrows) and the leiomyoma of the esophagus.

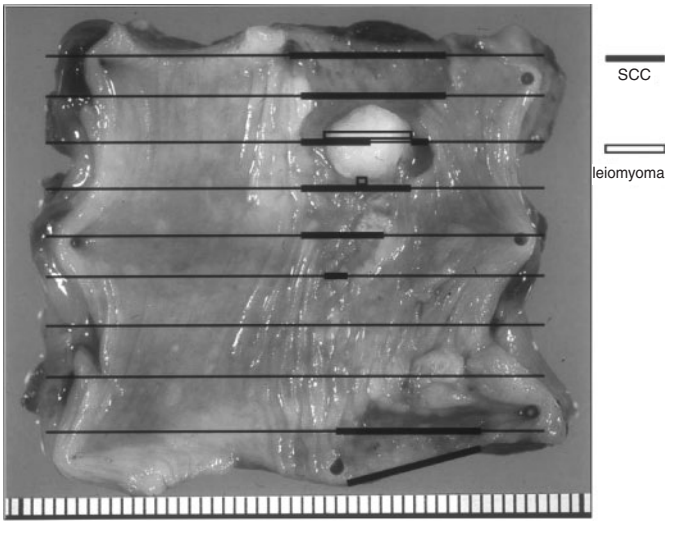


**Figure 6.** Pathological mapping of the SCC and leiomyoma, demonstrating that the leiomyoma locates within an area of early esophageal cancer.

tumor was diagnosed as a leiomyoma. The SMT just oral to the esophagogastric junction was also diagnosed as a leiomyoma. Pathological mapping of SCC and leiomyoma demonstrated that the leiomyoma located within the area of early esophageal cancer, though part of its surface was overlaid by non-cancerous epithelia (Fig. 6).

The post-operative course was uneventful and the patient was discharged 29 days after surgery. He is doing well 2 years after surgery without any recurrence.

## DISCUSSION

EMR is a safe treatment for superficial esophageal cancer (11). Endoscopic treatment for esophageal SCC is indicated in the tumor with invasion limited to the lamina propria mucosae (12). In the present case, the tumor was diagnosed by endoscopic ultrasonography as invading the submucosal layer at the nodule, which was actually an SMT, leiomyoma, but was misdiagnosed as a component of esophageal cancer. Surgery was therefore chosen as the treatment option for this esophageal tumor.

In the present case, the sessile polypoid tumor was located within the esophageal cancer. However, endoscopic ultrasonography demonstrated that the tumor had clear margins and a smooth contour, which suggested a benign tumor, and chromoendoscopy with Lugol's iodine solution showed part of the tumor was covered by normal mucosa. Moreover, there was another tumor, resembling an SMT, in the esophagus. It was reported previously that 2.4–7.8% of esophageal leiomyomas were multiple (13,14). Based on the available findings, the sessile polypoid tumor in the present case should be diagnosed as an SMT before surgery.

An SMT of the esophagus presents good indications for minimally invasive surgery, such as endoscopic (15–17) or thoracoscopic resection (18–20). When choosing treatment for SMT, it is important to determine by endoscopic ultrasonography from which layer the tumor originates (15). If a small SMT is diagnosed as originating from the muscularis mucosa, endoscopic resection is the treatment of choice (15,16). When the tumors are large and originate in the muscularis propria, this technique has the risk of severe complications, such as esophageal perforation and massive bleeding. Hence, in those cases, open surgery or thoracoscopic resection is chosen. In the present case, microscopic findings showed that the leiomyoma originated from the muscularis mucosae and its size was 7 mm in diameter. Retrospective analysis of pre-operative endoscopic ultrasonography also revealed that the submucosal tumor originated in the muscularis mucosae. Therefore, this case is a good indication for EMR regarding SMT. Leiomyomas typically arise from the muscularis propria, Takubo et al. (21) reported that the incidence of leiomyomas arising from the muscularis mucosae was 18.4% in 38 esophageal leiomyomas.

There have been two reports of co-existing esophageal leiomyoma and early esophageal SCC treated by curative EMR (7,8). These cases were diagnosed by endoscopy and endoscopic ultrasonography, which showed that superficial SCC overlaid the SMT. Therefore, in the present case, an aggressive endoscopic approach appeared to have been the therapeutic procedure of choice for this esophageal tumor, if a sessile polypoid lesion was diagnosed as SMT originating from the muscularis mucosa by endoscopy and endoscopic ultrasonography. This is because the depth of invasion of the esophageal cancer was histologically limited to the lamina propria mucosae.

## References

1. Callanan JG. Simultaneous occurrence of simple and malignant tumors in the esophagus. *J Thorac Surg* 1954;28:4–11.
2. Lu YK, Li YM, Keng CC. The coexistence of benign and malignant tumors of the esophagus. *Chin Med J* 1963;82:805–8.

3. Iizuka T, Kato H, Watanabe H, Itabashi M, Hirota T. Superficial carcinoma of the esophagus coexisting with esophageal leiomyoma: a case report and review of the Japanese literature. *Jpn J Clin Oncol* 1984;14:115–22.
4. Watanabe M, Baba T, Hotchi M. A case of leiomyoma of the lamina muscularis mucosae of the esophagus with a complication of carcinoma in situ of the overlying mucosa. *Acta Pathol Jpn* 1987;37:1845–51.
5. Yoshikane H. Tsukamoto Y, Niwa Y, et al. The coexistence of esophageal submucosal tumor and carcinoma. *Endoscopy* 1995;27:119–23.
6. Kuwano H, Sadanaga N, Watanabe M, Yasuda M, Nozoe T, Sugimachi K. Esophageal squamous cell carcinoma occurring in the surface epithelium over a benign tumor. *J Surg Oncol* 1995;59:268–72.
7. Nagashima R, Takeda H, Motoyama T, Tsukamoto O, Takahashi T. Coexistence of superficial esophageal carcinoma and leiomyoma: case report of an endoscopic resection. *Endoscopy* 1997;29:683–4.
8. Fu K, Tahara M, Muto M, et al. Carcinoma coexisting with esophageal leiomyoma. *Gastrointest Endosc* 2002;56:272–3.
9. Marcial-Rojas RA, Suau P, Juan S, Rico P. Epidermoid carcinoma in mucosa overlying a pedunculated lipoma of the esophagus. *J Thorac Surg* 1959;37:427–34.
10. Japanese Society for Esophageal Disease. Guidelines for clinical and pathologic studies on carcinoma of the esophagus, ninth edition: preface, general principles, part I. *Esophagus* 2004;1:61–88.
11. Narahara H, Iishi H, Tatsuta M, et al. Effectiveness of endoscopic mucosal resection with submucosal saline injection technique for superficial squamous carcinomas of the esophagus. *Gastrointest Endosc* 2000; 52:730–4.
12. Kodama M, Kakegawa T. Treatment of superficial cancer of the esophagus: a summary of responses to a questionnaire on superficial cancer of the esophagus in Japan. *Surgery* 1998;123:432–9.
13. Seremetis MG, Lyon WS, de Guzman VC, Peabody JW Jr. Leiomyomata of the esophagus. An analysis of 838 cases. *Cancer* 1976;38:2166–77.
14. Wang Y, Zhang R, Ouyang Z, Zhang D, Wang L, Zhang D. Diagnosis and surgical treatment of esophageal leiomyoma. *Zhonghua Zhong Liu Za Zhi* 2002;24:394–6.
15. Takada N, Higashino M, Osugi H, Tokuhara T, Kinoshita H. Utility of endoscopic ultrasonography in assessing the indications for endoscopic surgery of submucosal esophageal tumors. *Surg Endosc* 1999;13:228–30.
16. Kajiyama T, Sakai M, Torii A, Kishimoto H, Kin G, Uose S, et al. Endoscopic aspiration lumpectomy of esophageal leiomyomas derived from the muscularis mucosae. *Am J Gastroenterol* 1995;90:417–22.
17. Kinney T, Waxman I. Treatment of benign esophageal tumors by endoscopic techniques. *Semin Thorac Cardiovasc Surg* 2003;15:27–34.
18. Bardini R, Segalin A, Ruol A, Pavanello M, Peracchia A. Videothoracoscopic enucleation of esophageal leiomyoma. *Ann Thorac Surg* 1992;54:576–7.
19. Everitt NJ, Glinatsis M, McMahon MJ. Thoracoscopic enucleation of leiomyoma of the oesophagus. *Br J Surg* 1992;79:643.
20. Samphire J, Nafteux P, Luketich J. Minimally invasive techniques for resection of benign esophageal tumors. *Semin Thorac Cardiovasc Surg* 2003;15:35–43.
21. Takubo K, Nakagawa H, Tsuchiya S, Mitomo Y, Sasajima K, Shirota A. Seedling leiomyoma of the esophagus and esophagogastric junction zone. *Hum Pathol* 1981;12:1006–10.