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Huge Liposarcoma of Esophagus Resected by Endoscopic Submucosal Dissection: Case Report with Video

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Liposarcoma is one of the most common soft tissue sarcomas occurring in adults, but it rarely occurs in the gastrointestinal tract and more uncommonly in the esophagus. To the best of our knowledge, there are only 19 reported cases of esophageal liposarcoma in the literature published in English language up to the year 2008, and they were all treated by surgical methods. Here, we report a case of primary liposarcoma of the esophagus which was treated with endoscopic submucosal dissection (ESD). ESD was well tolerated in this patient, suggesting that it may be a therapeutic option for primary esophageal sarcomas.

Key Words: Esophageal neoplasms; Liposarcoma; Endoscopy

INTRODUCTION

Liposarcomas typically occur in the retroperitoneum or at the lower extremities. The occurrence of liposarcomas in the gastrointestinal tract is very low and the reported incidence of gastrointestinal liposarcoma is 0.1% to 5.8% at autopsy. The incidence of liposarcomas occurring at the esophagus is particularly low, where they form only 1.2% to 1.5% of all gastrointestinal lipomas. The optimal treatment method for primary esophageal liposarcoma has not been established yet, but all reported cases so far have been treated by surgical means. Although the use of therapeutic esophageal intervention has become increasingly common, nonsurgical therapy of primary esophageal liposarcomas has not been described yet in the literature. Here, we present a case of a large esophageal liposarcoma which was resected by endoscopic submucosal dissection (ESD) under general anesthesia.

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CASE REPORT

A 44-year-old male patient was admitted to our hospital for 4 months of dysphagia which was exacerbated during the recent weeks. An abdominal computed tomography (CT) scan showed a 15 cm long mass protruding into the esophageal lumen on coronal image. The majority of the tumor showed fat attenuation, internal septa-like structures, and soft tissue density (Fig. 1).

On esophagogastroduodenoscopy (EGD), there was a yellowish, soft, and movable subepithelial tumor (SET) located 20 to 35 cm away from the upper incisor. There was 4 cm sized stalk just below the upper esophageal sphincter (Fig. 2A). Endoscopic ultrasonography showed a nonhomogeneous submucosal mass located in the posterior wall of upper to mid esophagus (Fig. 2B). Although the tumor had a stalk, the stalk was too huge to use a snare. So we performed ESD on the patient under general anesthesia with nasal intubation. The procedure was done using a cap-fitted gastroscope (GIF-Q260; Olympus, Tokyo, Japan). After submucosal injection of cerol mixture (glycerin 10%, fructose 5%; Cheiljedang Pharma Corp., Seoul, Korea), the first incision was made using a fixed flexible knife (Kachu Technology, Seoul, Korea) at the center of the stalk. After exposing the submucosal tissue, the mass was dissected along the stalk using the IT-2 knife (KD-611L; Olympus Medical Systems Co., Tokyo, Japan) (Fig. 3A, Supple-

mentary Video 1, available online at <http://www.e-ce.org/>).

Macroscopically, the tumor had a three fingers-like shape and was measured 8.7×6.0×3.0 cm. Its surface was covered by normal esophageal mucosa with small erosion in some area. The cut end of the mass revealed multilobulating contoured mass, which was located in the submucosal layer and had yellow color (Fig. 3B). Histologically, the neoplasm consisted predominantly of mature looking fat tissues which were in variable sizes and the shape of adipocytic element. The lipocytes were divided by fibrous septa. Some lipocytes showed big and bizarre nucleuses, suggesting liposarcoma rather than lipoma (Fig. 3C).

The tumor had the characteristic features of well-differentiated liposarcoma in general. In some other areas, there were lobules of mature adipose tissue with a delineated myxoid tu-

mor in the subepithelial stroma. The cellularity was low and cellular pleomorphism was mild to moderate. Tumor necrosis was not seen. Following ESD, the patient was followed up for 4 months by chest CT and EGD, and signs of recurrence and metastasis have not been found so far (Fig. 4).

DISCUSSION

Four main subtypes of liposarcoma include well-differentiated liposarcoma, myxoid liposarcoma, pleomorphic liposarcoma, and dedifferentiated liposarcoma.

The only established methods for the diagnosis of liposarcoma are surgical excision and histological examination. Although indications for resection have not been established, unconditional resection is not generally recommended be-

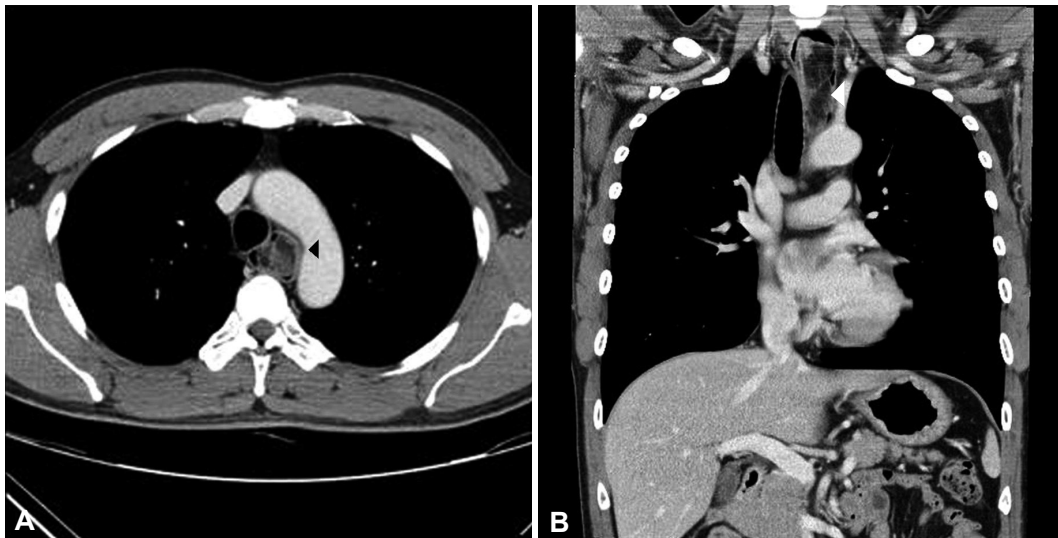


Fig. 1. Contrast-enhanced computed tomography scan (A, transverse view; B, coronal view) images demonstrate a tumor (arrowheads) protruding into the esophageal lumen. The large tumor is almost completely obstructing the esophagus.

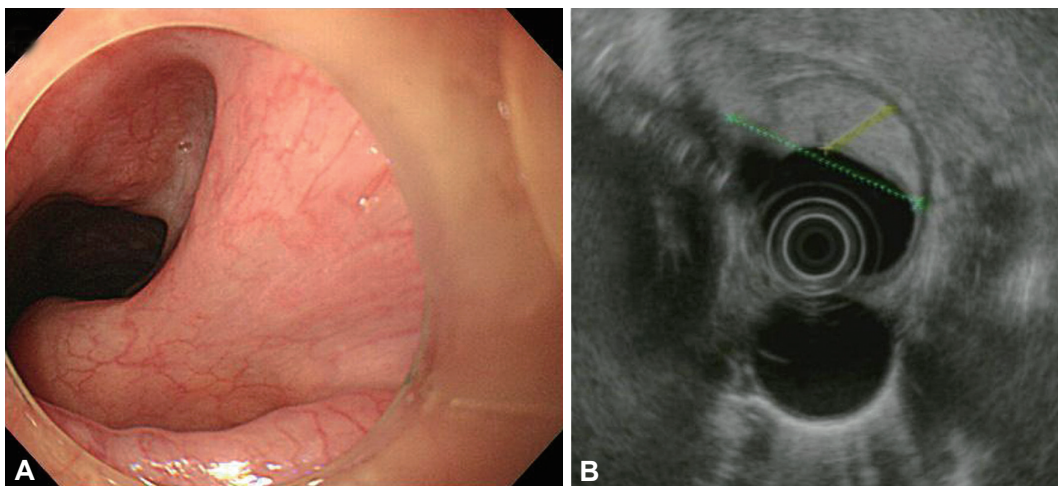


Fig. 2. (A) Esophagogastroduodenoscopy image shows a huge mass with stalk. (B) Endoscopic ultrasonography image shows a submucosal mass located in the posterior wall of upper to mid esophagus. The diameters of the mass were 12.7 and 37.1 mm, respectively.

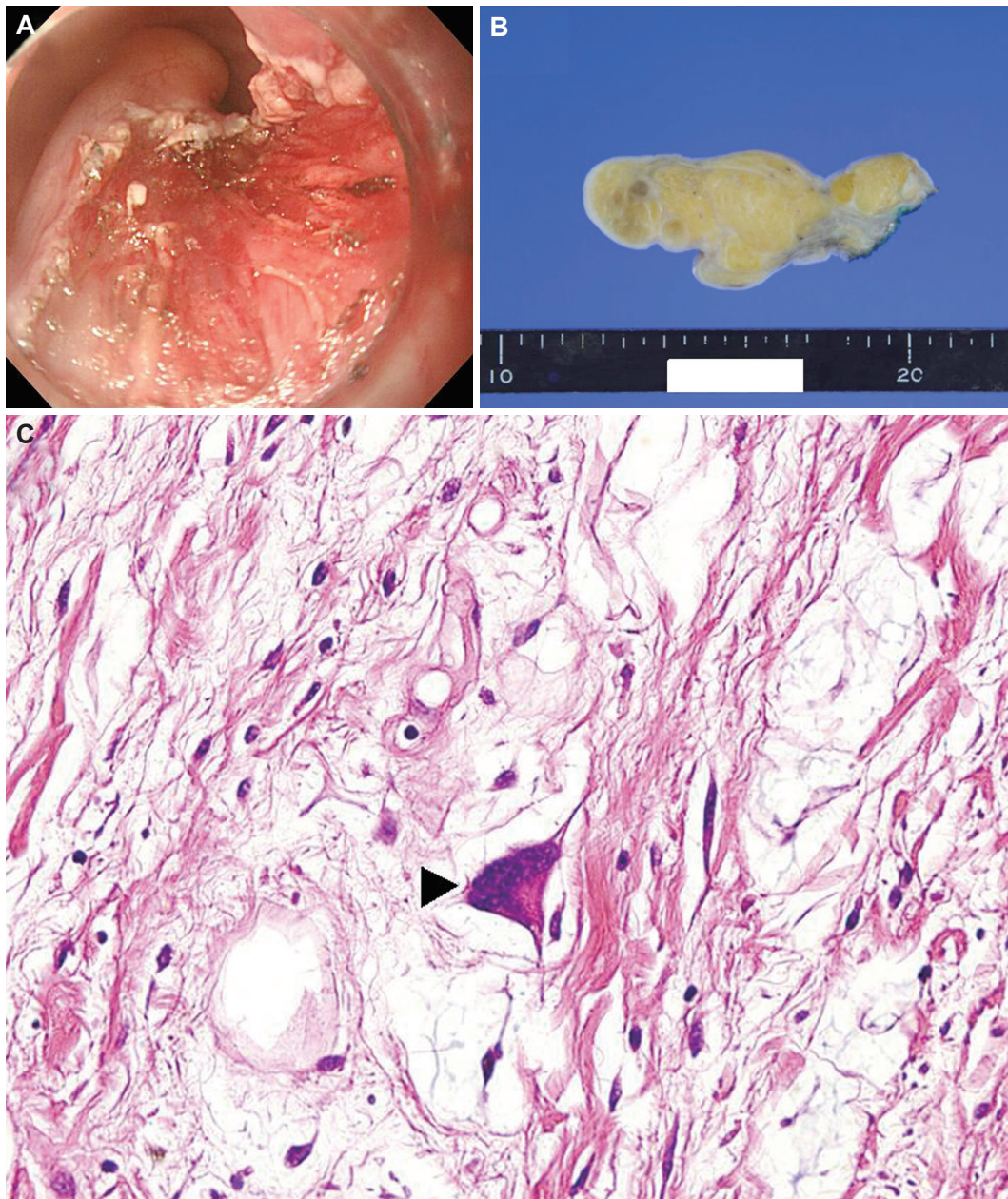


Fig. 3. (A) Esophagogastroduodenoscopy image during endoscopic submucosal dissection. (B) Gross image shows a yellowish, multilobulating mass divided by fibrous septa. (C) Microscopic finding shows a bizarre, big nucleus (arrowhead) suggesting liposarcoma rather than lipoma (H&E stain, $\times 200$).

cause of rare incidence of esophageal liposarcoma. The prognosis after the resection of esophageal liposarcoma is not well known because the occurrence of esophageal liposarcoma is so rare that there is no long term follow-up study. When we reviewed previous records, recurrence occurred in two patients among 15 patients whose medical records were available, so the recurrence rate of esophageal liposarcoma does not seem so high. Although there is no established guideline on the follow-up after the resection of esophageal liposarcoma yet, it seems that too frequent monitoring is not required

because esophageal liposarcoma is known as a slow-growing tumor.

Here, we showed that ESD can be a good alternative method for esophageal SET to avoid aggressive surgical procedures. Previously esophageal liposarcoma cases were treated by various surgical methods including simple enucleation and partial or total esophagectomy via transcervical, transthoracic, and transgastric routes.^{1,2} However, such surgical approaches are expensive and more invasive, resulting in longer hospital stays compared to endoscopic methods.

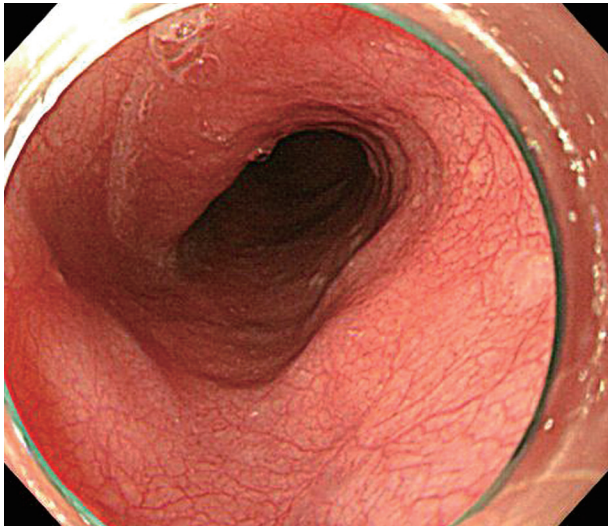


Fig. 4. On esophagogastroduodenoscopy after endoscopic submucosal dissection, there is no evidence of recurrence or metastasis.

ESD is a technique which was originally developed in Japan as a method for endoscopic resection of early gastric cancer and adenoma. The use of ESD has been expanded to include the removal of gastric SETs following reports of its usefulness in the treatment of gastric SETs.³ Today, ESD is also considered to be a useful therapeutic option for the resection of high grade dysplasia or early cancer in the esophagus. There have been several studies investigating the efficacy of ESD for the resection of esophageal SETs. Shi et al.⁴ reported the usefulness of ESD in treating esophageal SETs originating from the muscularis propria layer. In this study, the curative resection rate was 100% (28/28) and there were no recurrences during the follow-up of 3 to 27 months. Perforation, which occurred in two cases, were closed with metal endoclips and the patients recovered quickly without surgery.

Recently, there have also been some reports about cases for which submucosal tunnel dissection was used to treat upper gastrointestinal SETs.^{5,6} In these studies, SETs were success-

fully resected in patients with tumors sized 2 to 4 cm without severe complications.

To our knowledge, there have been no previous reports about the use of ESD for the treatment of esophageal liposarcoma, perhaps due to its rare occurrence. This is the first case for which ESD was performed to treat esophageal liposarcoma.

Although the postoperative follow-up has been only 4 months long, there have not been any signs of recurrence or complications. We believe that the endoscopic approach is superior to more aggressive surgical approaches (such as esophagectomies) because it allows more detailed resection as well as early recovery and less pain for the patient. Considering the fact that the use of ESD has been increasing recently, we believe this case report is meaningful and will help to validate the use of ESD as a feasible option for the treatment of esophageal liposarcomas.

Conflicts of Interest

The authors have no financial conflicts of interest.

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