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Giant myxofibrosarcoma of esophagus resected by endoscopic submucosal dissection: A case report

Wang XS *et al*. ESD for giant MFS of esophagus

Abstract

BACKGROUND

Myxofibrosarcoma (MFS) is a fibroblast-derived sarcoma, which mainly occurs in the subcutaneous tissue. MFS rarely occurs in the gastrointestinal tract, especially in the esophagus.

CASE SUMMARY

A 79-year-old male patient was admitted to our hospital for dysphagia evolving for a wk. Computed tomography and electronic gastroscope showed that a giant mass was located 30 cm from the incisor and extended to the cardia. There was incomplete esophageal stenosis. Pathologic view showed spindle cell lesions, considered inflammatory myofibroblast like hyperplasia. Considering the strong demands of the patient and his families, and the fact that most inflammatory myofibroblast tumors are benign, we decided to perform endoscopic submucosal dissection (ESD) even if the tumor size was 9.0 cm × 3.0 cm. Postoperative pathological examination revealed MFS. MFS rarely occurs in the gastrointestinal tract, especially in the esophagus. Surgical resection and local adjuvant radiotherapy are the first choices to improve the prognosis. This case report firstly described the ESD for esophageal giant MFS. It suggests that ESD may be an alternative treatment for primary esophageal MFS.

CONCLUSION

This case report suggests that it may be an alternative treatment for esophageal MFS, especially in elderly high-risk patients with obvious dysphagia symptoms.

Key Words: Endoscopic submucosal dissection; Giant myxofibrosarcoma; Esophagus; Case report

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Core Tip: In this study, we first reported a giant esophageal myxofibrosarcoma (MFS) about 9.0 cm × 3.0 cm in size. We managed to remove the MFS completely through endoscopic submucosal dissection (ESD) without severe complications. This case suggests that ESD may be an alternative treatment for esophageal MFS, especially with obvious dysphagia symptoms and aged high-risk patients.

INTRODUCTION

Myxofibrosarcoma (MFS) is a fibroblast-derived sarcoma, which mainly occurs in the subcutaneous tissue[1,2]. Most cases of MFS occur in the extremities, and only about 20% of cases occur in the trunk, retroperitoneum, heart, *etc*[1-4]. MFS rarely occurs in the gastrointestinal tract, especially in the esophagus. Surgical resection and local adjuvant radiotherapy are the first choices to improve the prognosis. In this report, the families of the patient considered that endoscopic submucosal dissection (ESD) was relatively safe and maintained the anatomical integrity of the esophagus, so they chose ESD as a treatment strategy. The challenge for endoscopists is that the MFS with a size of about 9.0 cm × 3.0 cm is too large to be completely removed. And there is a high risk of gastrointestinal perforation and bleeding. Fortunately we managed to remove the MFS completely through ESD without severe complications. And the patient recovered well. It suggests that ESD may be an alternative treatment for primary MFS in esophagus.

CASE PRESENTATION

Chief complaints

A 79-year-old male patient was admitted to our hospital for dysphagia evolving for a week.

History of present illness

The patient had eating obstruction in the last week and had obvious symptoms when eating solid food. He did not have nausea, vomiting or hoarseness.

History of past illness

The patient has a healthy medical history.

Personal and family history

No abnormality in personal and family history.

Physical examination

No abnormality found in physical examination.

Laboratory examinations

Laboratory examinations showed no abnormalities.

Imaging examinations

Computed tomography (CT) showed thickening of the lower esophageal wall and stenosis of the lumen. Gastroscopy revealed a giant mass located 30 cm from the incisor and extended to the cardia. There was incomplete esophageal stenosis. Macroscopically, it looks like a serpentine, with a size of about 9.0 cm × 3.0 cm (Figure 1A). Endoscopic ultrasound revealed that the tumor originated from the submucosa, mixed echo changes, and exhibited some cystoid in the inner-structures (Figure 1B). Pathologic view showed spindle cell lesions, which was considered inflammatory myofibroblast like hyperplasia.

Further diagnostic work-up

Immunohistochemical staining showed KI67(55%+), P53(3+, Mutant), Vimentin(3+), P40(-), CK5/6(-), SMA(-), Desmin(-), HMB45(-), CD34(-), CD117(-), DOG-1(-), CKpan(-), and EBER(-)(Figure 2).

FINAL DIAGNOSIS

The final diagnosis of the presented case is MFS.

TREATMENT

The operation was performed under general anesthesia with transparent cap assisted endoscopic therapy. After injecting the cerium mixture (composed of indigo, adrenaline, sodium hyaluronate and normal saline) under the mucosa on the oral side, we made an incision using a fixed insulated tip (IT) knife (Olympus, Japan). Then we used IT knife and double knife (Olympus, Japan) to gradually resect the tumor as a whole without serious complications (Figure 3).

OUTCOME AND FOLLOW-UP

The patient recovered well and was discharged from the hospital a week later without eating obstruction.

DISCUSSION

MFS is a fibroblast-derived sarcoma, which mainly occurs in the subcutaneous tissue[1,2]. MFS often occurs in elderly patients, with the extremities and girdles being the most frequent sites[6]. Although some have shown a higher incidence in men, the current evidences have no obvious gender preference[4,7]. MFS is a malignant fibroblastic neoplasm characterized by a high risk of local recurrence[8]. Surgical resection and local adjuvant radiotherapy are the first choice to improve the prognosis. As a soft tissue tumor mainly occurring in subcutaneous tissue, MFS rarely occurs in gastrointestinal tract, especially in esophagus. This entity is challenging in diagnosis due to rare and nonspecific clinical manifestations. MFS presents as a heterogeneous

soft tissue mass on CT. Magnetic resonance imaging (MRI) is another choice to help diagnosis. T1-weighted MRI shows a low to intermediate signal. T2-weighted MRI presents that the solid and myxoid components are high signal intensity, and the myxoid component are higher than the fluids[9]. Surgical excision and histologic tests are considered the gold standard for MFS diagnosis, particularly if the tumor is present in a rare location. Thoracoscopic surgery may be the first choice to remove this unusual tumor. However, compared with endoscopic surgery, the surgical method is invasive, expensive, and often leads to additional complications especially in elderly patients. The advantages of ESD are relatively safe and keep the integrity of esophageal anatomy. And some cases have used ESD to treat giant esophageal masses[10,11]. However, ESD may not be able to completely remove the huge tumor. In addition, the risk of esophageal perforation and bleeding is high. If the above situations occur, surgery is still required to remove the tumor. In this case, considering the obvious symptoms of dysphagia, the elderly, and the strong demand of patient and his families, we decided to perform ESD even if the tumor size was 9.0cm × 3.0cm. Fortunately, the tumor was *en bloc* removed by ESD without severe complications. Combined with immunohistochemistry, the tumor was diagnosed as MFS in the Department of Pathology of Jiangsu Provincial People's hospital. This is the first case of MFS occurring in the esophagus and being resected by ESD. With the increasing incidence of gastrointestinal tumors, we believe this case report will help promote the ESD as an alternative treatment of esophageal MFS.

CONCLUSION

Resection of esophageal MFS with ESD has not been reported before. This case report suggests that ESD may be an alternative treatment for esophageal MFS, especially in elderly high-risk patients with obvious dysphagia symptoms.

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