ISSN 2307-8960 (online)

World Journal of *Clinical Cases*

World J Clin Cases 2023 February 16; 11(5): 979-1223

Published by Baishideng Publishing Group Inc

World Journal of Clinical Cases

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WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

INDEXING/ABSTRACTING

The *WJCC* is now abstracted and indexed in Science Citation Index Expanded (SCIE, also known as SciSearch®), Journal Citation Reports/Science Edition, Current Contents®/Clinical Medicine, PubMed, PubMed Central, Scopus, Reference Citation Analysis, China National Knowledge Infrastructure, China Science and Technology Journal Database, and Superstar Journals Database. The 2022 Edition of Journal Citation Reports® cites the 2021 impact factor (IF) for *WJCC* as 1.534; IF without journal self cites: 1.491; 5-year IF: 1.599; Journal Citation Indicator: 0.28; Ranking: 135 among 172 journals in medicine, general and internal; and Quartile category: Q4. The *WJCC*'s CiteScore for 2021 is 1.2 and Scopus CiteScore rank 2021: General Medicine is 443/826.

RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: *Hua-Ge Yu*; **Production Department Director:** *Xiang Li*; **Editorial Office Director:** *Jin-Lei Wang*.

NAME OF JOURNAL
World Journal of Clinical Cases

ISSN
ISSN 2307-8960 (online)

LAUNCH DATE
April 16, 2013

FREQUENCY
Thrice Monthly

PUBLICATION DATE
February 16, 2023

World Journal of Clinical Cases

Submit a Manuscript: https://www.f6publishing.com

World J Clin Cases 2023 February 16; 11(5): 1094-1098

DOI: 10.12998/wjcc.v11.i5.1094

ISSN 2307-8960 (online)

CASE REPORT

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

Xiao-Song Wang, Cheng-Guang Zhao, Hui-Ming Wang, Xiao-Yan Wang

Specialty type: Medicine, research and experimental

Provenance and peer review: Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's scientific quality classification

Grade A (Excellent): 0
Grade B (Very good): B
Grade C (Good): C
Grade D (Fair): D
Grade E (Poor): 0

P-Reviewer: Atanasova EG, Bulgaria; Beji H, Tunisia; Vyshka G, Albania

Received: November 1, 2022
Peer-review started: November 1, 2022
First decision: November 11, 2022
Revised: November 17, 2022
Accepted: January 16, 2023
Article in press: January 16, 2023
Published online: February 16, 2023

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Abstract

BACKGROUND

Myxofibrosarcoma (MFS) is a fibroblast-derived sarcoma that mainly occurs in 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 for a week. Computed tomography and electronic gastroscopy showed that a giant mass was located 30 cm from the incisor and extended to the cardia. There was incomplete esophageal stenosis. Endoscopic pathology showed spindle cell lesions, which were considered inflammatory myofibroblast like hyperplasia. Considering the strong demands of the patient and his family, and the fact that most inflammatory myofibroblast tumors are benign, we decided to perform endoscopic submucosal dissection (ESD) even if the tumor size was giant (9.0 cm × 3.0 cm). Postoperative pathological examination resulted in a final diagnosis of 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 for the first time describe the successful treatment of a giant esophageal MFS by ESD, suggesting that ESD may be an alternative treatment for primary esophageal MFS, especially in elderly high-risk patients with obvious dysphagia symptoms.

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

Core Tip: For the first time, we report a giant esophageal myxofibrosarcoma (MFS) measuring about 9.0 cm × 3.0 cm. 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 in aged high-risk patients with obvious dysphagia symptoms.

Citation: Wang XS, Zhao CG, Wang HM, Wang XY. Giant myxofibrosarcoma of the esophagus treated by endoscopic submucosal dissection: A case report. *World J Clin Cases* 2023; 11(5): 1094-1098
URL: https://www.wjgnet.com/2307-8960/full/v11/i5/1094.htm
DOI: https://dx.doi.org/10.12998/wjcc.v11.i5.1094

INTRODUCTION

Myxofibrosarcoma (MFS) is a fibroblast-derived sarcoma that 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*[3-5]. MFS rarely occurs in the gastrointestinal tract, especially in the esophagus. Surgical resection and local adjuvant radiotherapy are the first choice to improve the prognosis. In this paper, we report a patient with a giant MFS whose family chose endoscopic submucosal dissection (ESD) as a treatment strategy since they considered it was relatively safe and could maintain the anatomical integrity of the esophagus. The challenges for endoscopists were that the MFS with a size of about 9.0 cm × 3.0 cm was too large to be completely removed, and that there would be 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. This case suggests that ESD may be an alternative treatment for primary MFS in the esophagus.

CASE PRESENTATION

Chief complaints

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

History of present illness

The patient had dysphagia in the last week and had, which was more obvious when eating solid food. He did not have nausea, vomiting, or hoarseness.

History of past illness

The patient had no remarkable medical history.

Personal and family history

The patient had no remarkable personal and family history.

Physical examination

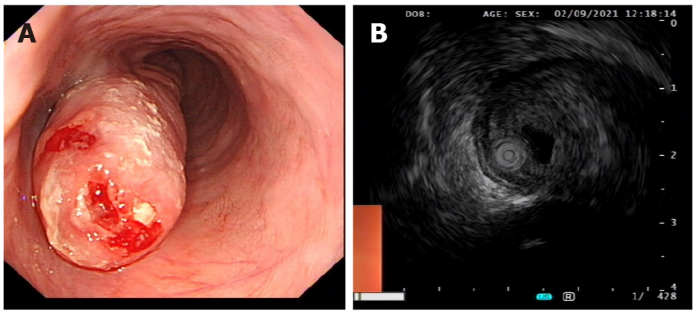
No abnormality was found in the physical examination.

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, the tumor looked 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, had mixed echo changes, and exhibited some inner cystoid structures (Figure 1B). Endoscopic pathology showed spindle cell lesions, which were considered inflammatory myofibroblast like hyperplasia.



DOI: 10.12998/wjcc.v11.i5.1094

Figure 1 Imaging examinations. A: Gastroscopy showed that a giant mass was located 30 cm from the incisor and extended to the cardia; B: Endoscopic ultrasonography showed that the mass was located in the submucosa, had mixed echo change, and exhibited some inner cystoid structures.

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 was MFS.

TREATMENT

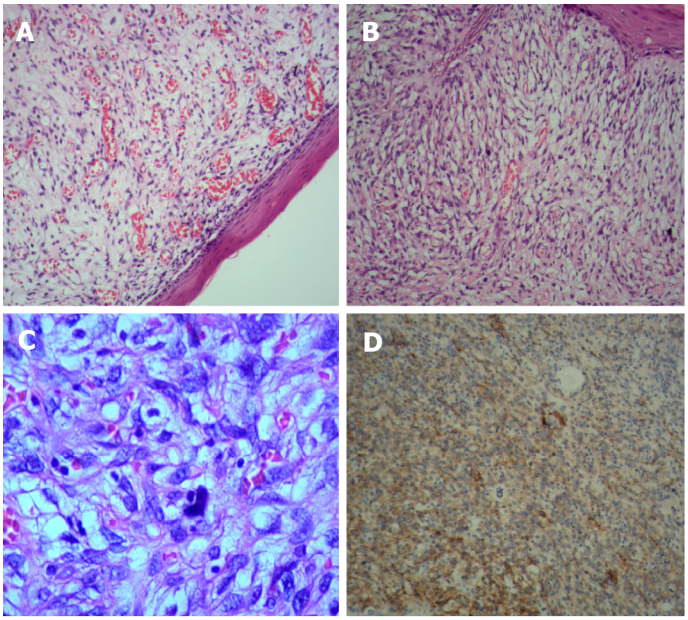
The operation was performed under general anesthesia with transparent cap assisted endoscopic therapy. After injecting the cerium mixture (composed of indigo, adrenaline, sodium hyaluronate, and normal saline) under the mucosa on the oral side, we made an incision using a fixed insulated tip (IT) knife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

OUTCOME AND FOLLOW-UP

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

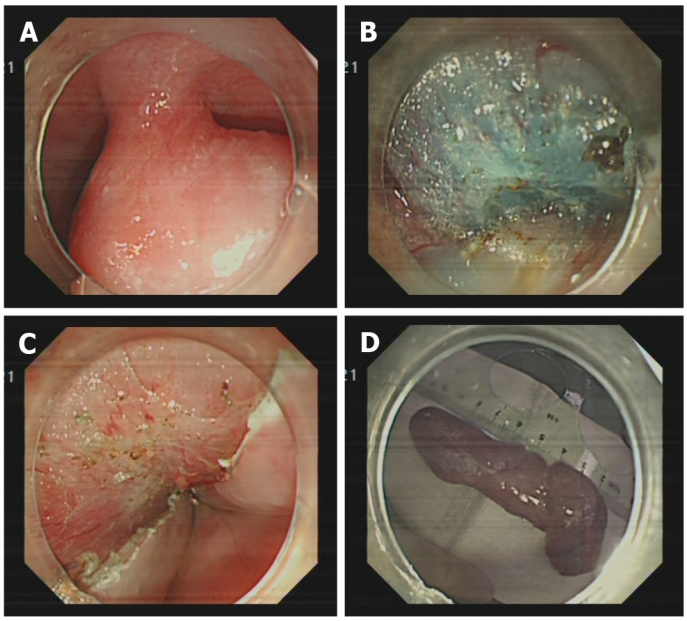
DISCUSSION

MFS is a fibroblast-derived sarcoma that mainly occurs in the subcutaneous tissue[1,2]. MFS often occurs in elderly patients, with the extremities and girdles being the most frequently affected sites[6]. Although some studies report a higher incidence in men, the current evidence suggest 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 the gastrointestinal tract, especially in the esophagus. The diagnosis of this entity is challenging due to its rare and nonspecific clinical manifestations. MFS presents as a heterogeneous soft tissue mass on CT. Magnetic resonance imaging (MRI) is another choice for diagnosis. Typically, T1-weighted MRI shows a low to intermediate signal, and T2-weighted MRI shows that the solid and myxoid components have high signal intensity, and the signal intensity of the myxoid component is higher than that of the fluid [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, open surgery is invasive and expensive, and often leads to additional complications, especially in elderly patients. ESD is relatively safe and can keep the integrity of esophageal anatomy. And some scholars 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 the present case, considering the obvious symptoms of dysphagia, the advanced age of the patient, and the strong demand of the patient and his family, we



DOI: 10.12998/wjcc.v11.i5.1094

Figure 2 Histologic images of the myxofibrosarcoma. A: Spindle tumor cells; B: Myxoid stroma; C: Karyokinesis; D: Positive vimentin.



DOI: 10.12998/wjcc.v11.i5.1094

Figure 3 The endoscopic submucosal dissection procedure. A: A submucosal tumor in the esophagus was found and marked; B: Mucosal injection and submucosal precut created using an insulated tip-2 knife; C: The wound surface after removal of the tumor; D: The specimen of the tumor measuring 9.0 cm × 3.0 cm.

decided to perform ESD even if the tumor size was big (9.0 cm × 3.0 cm). Fortunately, the tumor was *en bloc* removed by ESD without severe complications. Combined with immunohistochemistry results, the tumor was diagnosed as MFS. 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 that this case report will help promote the ESD as an alternative treatment for esophageal MFS.

CONCLUSION

Resection of esophageal MFS by 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.

FOOTNOTES

Author contributions: Wang XS and Wang HM wrote and revised the case report; Zhao CG and Wang XY were responsible for all the clinical aspects of the paper; and all authors issued final approval for the version to be submitted.

Supported by the Key Project of Jiangsu Province, China, No. ZD2022052.

Informed consent statement: Informed consent was obtained from the patient for the publication of this case report and any accompanying images.

Conflict-of-interest statement: All the authors report no relevant conflicts of interest for this article.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Country/Territory of origin: China

ORCID number: Xiao-Song Wang 0000-0002-6383-091X; Xiao-Yan Wang 0000-0002-8779-4514.

S-Editor: Wang JJ
L-Editor: Wang TQ
P-Editor: Wang JJ

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E-mail: bpgoffice@wjgnet.com
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