77231_Auto_Edited.docx

WORD COUNT 1541
TIME SUBMITTED 13-JUL-2022 05:35PM
PAPER ID 88356019

Name of Journal: *World Journal of Clinical Cases*

Manuscript NO: 77231

Manuscript Type: CASE REPORT

Misdiagnosis of an elevated lesion in the esophagus: A case report

The value of endoscopic diagnosis and therapeutic strategy for esophageal carcinosarcoma

Xing-Bin Ma, Huai-Yuan Ma, Xing-Fang Jia, Fei-Fei Wen, Cheng-Xia Liu

Abstract

BACKGROUND

Esophageal carcinosarcoma (ECS) is a rare biphasic tumor and a type of esophageal malignancy, which presents as protruding or elevated lesions. ECS patients are not hospitalized until they have severe dysphagia. ECS is easily misdiagnosed as a benign tumor due to its atypical characteristics under endoscopy. With the popularization of endoscopic treatment, these patients are often referred to endoscopic treatment, such as endoscopic submucosal dissection (ESD). However, ECS lacks consensus on its endoscopic features and therapies. Here, we report a case of ECS and discuss the value of endoscopic diagnosis and therapeutic strategies.

CASE SUMMARY

A 63-year-old man was admitted to the hospital with dysphagia. During the endoscopic examination, an elevated lesion was found with an erosive and hyperemic surface covered with white pseudomembranous inflammation. Endoscopic ultrasonography (EUS), biopsies, and enhanced thoracic computed tomography (CT) were performed, suggesting that it was a benign lesion and located within the submucosal layer. This lesion was diagnosed as a fibrovascular polyp with a Paris classification of 0-Ip. The patient was then referred to ESD treatment. However, the post-ESD pathological and immunohistochemical (IHC) study showed that this lesion was ECS with a vertical positive margin (T1b), indicating that we made a misdiagnosis and achieved a noncurative resection. Due to the potential tumor residue, additional open surgery was performed at the patient's request. In the postoperative pathological study, no tumor remnants or metastases were discovered. The patient was followed for 1 year and had no recurrence.

CONCLUSION

ECS can be misdiagnosed at the initial endoscopy. EUS can help to identify the tumor stage. Patients with T1b ECS cannot be routinely referred to ESD treatment due to the high risk of metastasis and recurrence rate.

Key Words: Esophageal carcinosarcoma; Misdiagnosis; Endoscopic ultrasonography; Endoscopic submucosal dissection; T1 stage; Case report

Ma XB, Ma HY, Jia XF, Wen FF, Liu CX. Misdiagnosis of an elevated lesion in the esophagus: A case report of esophageal carcinosarcoma and literature review. *World J Clin Cases* 2022; In press

Core Tip: ECS is a rare type of esophageal malignancy. ECS commonly presents as a pedunculated characteristic (0-Ip), which is often misdiagnosed due to the lack of specific features. EUS can help to evaluate whether ECS invasion is within the submucosal layer (T1 or T2) stage but cannot further distinguish whether it is T1a or T1b stage. Due to the high risk of metastasis and recurrence based on the literature review, ESD treatment cannot be routinely recommended for ECS patients with T1b stage disease.

INTRODUCTION

Patients routinely undergo endoscopic evaluation for dysphagia, during which protruding or elevated lesions are frequently found. Some of the lesions are presented as pedunculated lesions, including esophageal adenoma, inflammatory polyp, fibrovascular polyp, carcinosarcoma [1-3], *etc.* However, they lack specific features and have similar EUS characteristics. Therefore, it is difficult to conclusively diagnose the lesion without the support of the postsurgical pathology.

Herein, we report a rare case of ECS, which was assessed as a benign tumor and treated with ESD. Nevertheless, post-ESD pathology indicated that it was preoperatively misdiagnosed. Therefore, we systematically evaluated the endoscopic

and clinicopathological characteristics of ECS and analyzed the feasibility of endoscopic treatment.

CASE PRESENTATION

Chief complaints

Dysphagia for 3 mo.

History of present illness

A 63-year-old man was admitted to the hospital with dysphagia for 3 mo. The patient can only swallow semi-solid food for 2 wk, with intermittent swallowing pain.

History of past illness

The patient was in good health in the past.

Personal and family history

The patient had no personal and family history.

Physical examination

The patient was in good condition. The physical examination was completely normal.

Laboratory examinations

Routine laboratory tests were all within the normal range.

Imaging examinations

During the endoscopic examination, an elevated lesion with an erosive and hyperemic surface covered with white pseudomembranous inflammation was found. It had a short peduncle connected to the mid-esophagus wall and was 30 x 40 mm in size. EUS revealed a lesion derived from the submucosal layer with an intact inherent muscle layer, and this lesion was hypoechoic and consisted of internal multicystic components. Ultrasonic elastography of the lesion was blue–green, indicating a tough texture.

Multiple biopsies showed necrosis and active fibroblast proliferation (Figure 1). An enhanced thoracic CT showed a protuberant lesion in the middle of the esophagus, suspecting a benign tumor. A multidisciplinary consultation was performed, and we preliminarily diagnosed this lesion as a fibrovascular polyp.

FINAL DIAGNOSIS

The post-ESD pathological study showed that this lesion was composed of a malignant fibroblast apoptosis component and a basal-like squamous cell carcinoma (BSC) component, indicating esophageal carcinosarcoma (ECS). The polypoid mass was predominantly composed of malignant fibroblast ocytosis with a vertical positive margin, horizontal negative margin, and no evidence of vascular or lymphatic invasion. Immunohistochemical (IHC) showed CK (-), vimentin (+), CD68 (+), β-catenin (+), p53 (+), S-100 (-), CD34 (-), SMA (-), desmin (-), Twist1 (+), ZEB1 (+), Snai2 (-), PDGFR alpha (-) and Ki-67 of 20%. BSC was observed in the neck of the tumor, and its vertical and horizontal margins were negative. IHC showed CK (+), E-cadherin (+), p53 (+), vimentin (-), S-100 (-), CD34 (-), CD68 (-), SMA (-), desmin (-) and Ki-67 of 30% (Figure 2).

TREATMENT

The following ESD treatment was successful with no obvious adhesion in the submucosal layer after the patient's informed consent was obtained.

OUTCOME AND FOLLOW-UP

Due to the potential tumor residue, additional open surgery was performed at the patient's request. No tumor remnants or metastases were discovered in the postoperative pathological study. The patient was followed for 1 year and had no recurrence.

DISCUSSION

ECS is a rare biphasic tumor that accounts for 0.2~2.8% of all esophageal malignancies. It is characterized by the presence of both malignant epithelial and mesenchymal components[4]. ECS usually presents as a large intraluminal polypoid mass on the upper and middle esophagus, with a median diameter of 55~75 mm. The endoscopic features of this lesion may include a hyperemia surface, erosion, ulceration, brittleness, and easy bleeding, which lack specificity for endoscopic diagnosis[5].

The diagnosis of ECS mainly relies on pathological studies[4]. However, untargeted endoscopic biopsies of this lesion usually reveal components of sarcoma, which make it easily misdiagnosed. Efforts can be made to potentially improve the biopsy accuracy by targeting the root or peduncle as the epithelial cancer component always exceeds the mass in the range[6].

EUS evaluation of the lesion plays a role in the assessment before treatment. Although lacing specificity in diagnosis, EUS can provide information on invasion depth. According to a report from Taiwan[7], five of six ECS patients were correctly assessed on the invasion depth with EUS. However, all lesions were in the deep invasion (T2) stage. Our preoperative EUS showed that the origin of the ECS was derived from the T1 stage, which was proven by postoperative pathology. However, we also noticed that EUS could not further distinguish whether it was T1a or T1b stage. The reason could be that the echo of sarcoma that invaded the submucosa was similar to the original interstitial composition and therefore could not be distinguished by EUS.

Data on lymph node metastases of ECS in the T1 stage are limited[7, 8]. In 2006, Sanada *et al* reviewed 57 cases of ECS reported in Japan between 1995 and 2004, among which 1 was a T1a case and 17 were T1b cases[9]. Seven (41%) of the T1b cases were found to have lymph node metastasis compared with none of the T1a cases. In 2021, Chen *et al* reported that none of the 10 ECS patients in the T1 stage were found to have lymph node metastasis, with no report of T1 subtypes[10]. Since lymph node metastasis is related to prognosis, a detailed assessment is required before treatment.

Data on the prognosis of ESD treatment for ECS in the T1 stage are also limited. One Korean case from Ra Ri Cha *et al* in 2014 is very similar to ours[11]. The lesion located

within the submucosal layer without evidence of metastasis was treated with ESD. The post-ESD pathological study reported ECS with a positive vertical margin (T1b stage). In contrast, the patient from this Korean study refused to receive additional surgery, and a recurrence was found during an endoscope examination 21 mo later. Two Chinese cases were also treated with ESD, and one case was followed by additional surgery. Unfortunately, neither of them had long-term follow-ups [12, 13]. Therefore, robust data on the prognosis of ESD for ECS are needed.

CONCLUSION

We report a rare case of ECS with BSC, which can be misdiagnosed as lacking specificity characteristics. Targeted biopsies on the root or peduncle after observation with narrow-band imaging or iodine staining may potentially improve the diagnostic accuracy. EUS can help to evaluate the layer of the origin (T1 or T2) but cannot further distinguish whether it is within the T1a or T1b stage. ESD treatment cannot be routinely recommended to ECS patients with T1b stage disease due to the risk of metastasis and high recurrence rate.

77231_Auto_Edited.docx

ORIGINALITY REPORT

5%
SIMILARITY INDEX

PRIMARY SOURCES

1	Shusen Chen, Yu Shi, Zhengjing Lu, Mingwei Wang, Longfei Cong, Baixia Yang, Xudong Chen, Jing Cai, Xi Yang. "Esophageal Carcinosarcoma: Analysis of Clinical Features and Prognosis of 24 Cases and a Literature Review", Cancer Control, 2021 Crossref	29 words — 2%
2	www.wjgnet.com Internet	19 words — 1%
3	mafiadoc.com Internet	17 words — 1%
4	f6publishing.blob.core.windows.net Internet	15 words — 1%

EXCLUDE QUOTES	ON	EXCLUDE SOURCES	< 12 WORDS
EXCLUDE BIBLIOGRAPHY	ON	EXCLUDE MATCHES	< 12 WORDS