Surgical treatment of esophageal leiomyoma larger than 5 cm in diameter: A case report and review of the literature

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ABSTRACT Although leiomyoma is the most common benign esophageal neoplasm, it is a rare condition. Resection of the tumor is recommended in symptomatic patients, and observation is recommended in asymptomatic patients with small lesions. We discussed here a patient admitted to our hospital for dyspepsia in whom a calcified mediastinal neoplasm was diagnosised preoperatively and esophageal leiomyoma was diagnosised postoperatively. Enucleation of a leiomyoma of the esophagus is recommended and the optimal approaches should be tailored based on the location and size of the tumor.

KEY WORDS Benign esophageal tumor; enucleation; esophageal leiomyoma

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Introduction

Benign tumors of the esophagus are rare and leiomyoma is the most common benign tumor of the esophagus (1). It is reported that most leiomyomas originate the inner circular muscle layer of the distal and midthoracic esophagus, particularly at the esophagogastric junction (2,3). Middle-aged men are most frequently affected (4,5). The main symptoms usually are dysphagia and epigastric pain, but they are not specific for the disease. The size of the esophageal leiomyoma may change, a size of 1 to 29 cm has been defined in the literature (1,6,7). But most of them was samller than 5 cm in diameter. Tumor that size lager than 5cm are rare. It was easily misdiagnosised as mediastinal mass, esophageal cancer (8) and esophageal stromal tumor. Its clinical feature and management are differente with other smaller esophageal leiomyoma. Here, a patient with large calcified esophageal leiomyoma who was treated in our institute is presented, initial discuss its diagnosis and management against the background of previously published cases and series.

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Case report

A 42-year-old woman was admitted to our hospital with dyspepsia and esophageal reflux. There was no nausea, vomiting, or weight loss. Results of a physical examination and standard laboratory tests were normal. A chest radiograph showed a mass in the right low mediastum, and a filling defect was apparent on esophagography. Computerized tomography (CT) scanning of the chest revealed a completely calcified 70 mm × 60 mm mass at the right lower posterior mediastinum narrowing the esophagus lumen and the cacified benign mediastinal neoplasm was diagnosis (Figure 1A, 1B). Growth over the organ borders or infiltration in neighboring structures was not detected. There were no enlarged lymph nodes, and there was no evidence for distant metastases. A right thoracotomy was performed. During surgical exploration, a unregular cacified mass in the distal esophagus (Figure 1C). The mass was enucleated completely. Histopathologic examination revealed a tumor of 70 mm × 60 mm × 50 mm and cacified spindle cell fascicles without mitosis or atypia was observed (Figure 1D).

Discussion

Leiomyomas are benign tumors descending from smooth muscle cells of the esophagus. They are the most common benign tumors of the esophagus and they may occur in all parts of the esophagus, but 60% occur in the distal third, 30% in the middle, and 10% in the proximal esophagus (4). Although gastrointestinal bleeding is a common finding in gastric leiomyomas, esophageal leiomyomas rarely bleed, which may be because they do not ulcerate (9). Leiomyomas grow slowly, and half of the patients are asymptomatic and the symptoms

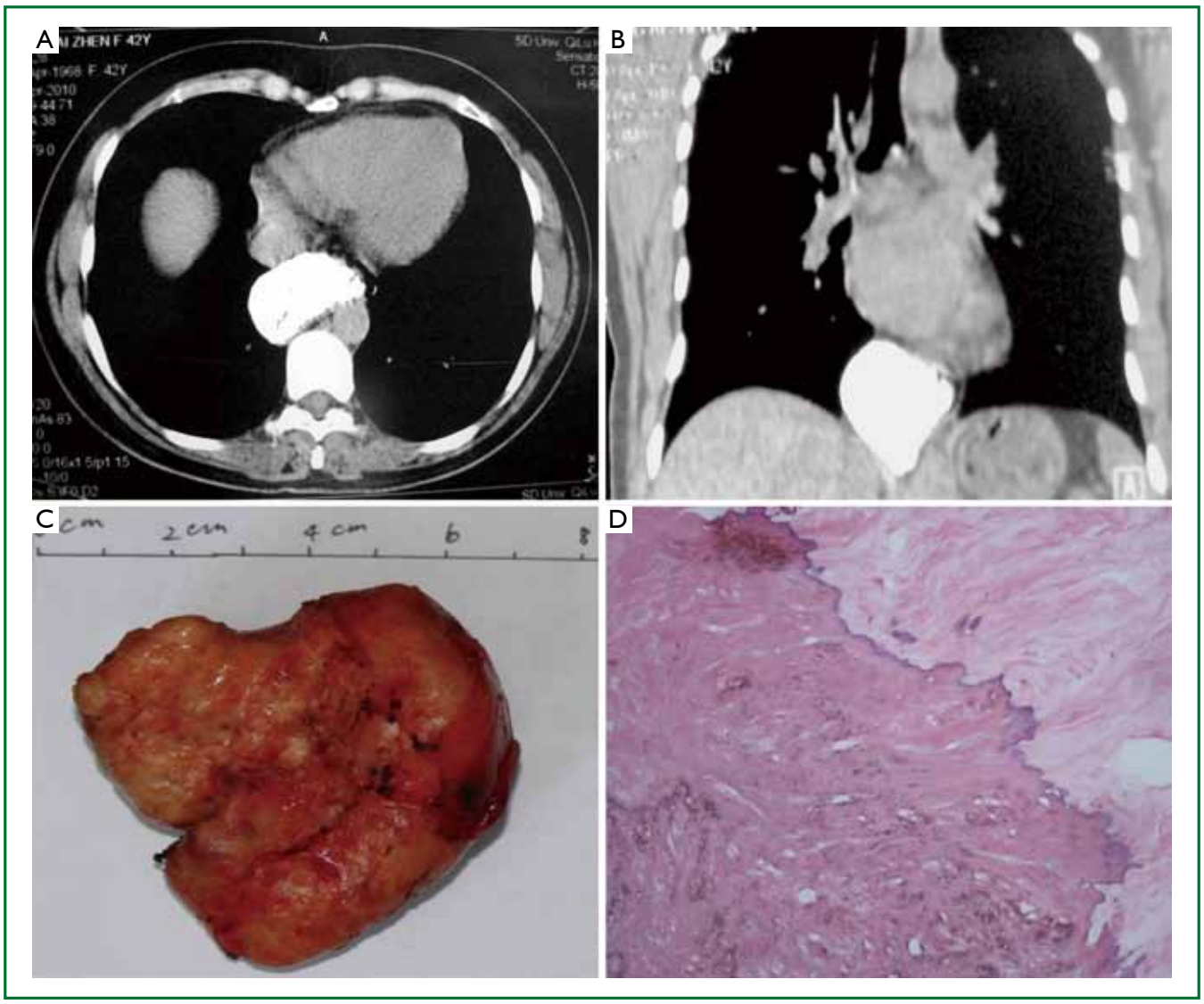

Figure 1. A/B: Computerized tomography (CT) scanning of the chest revealed a completely calcified 70 mm × 60 mm mass at the right lower post mediastinum; C: A solitary esophageal leiomyoma with a ginger-like shape after enucleation; D: Histopathologic examination revealed cacified spindle cell fascicles without mitosis or atypia (HE×100) course was uneventful, and the patient was discharged subjectively free of complaints on the 9th postoperative day.

are not specific. Dysphagia with concomitant epigastric pain or retrosternal burning usually appears when the tumor's diameter becomes larger than the critical point of 4.5-5 cm (10). It seemed that the size of tumor correlates with the severity of the symptoms, but larger leiomyoma usually grow toward outside of esophageal lumen. So dysphagia don't always proportionate to the size of tumor in larger leiomyoma. In our report, the symptoms were dyspepsia and esophageal reflux.

The diagnosis of esophageal leiomyoma is mostly not clear preoperatively. It may present as a mediastinal mass on chest radiograph and may be seen as an incidental radiologic finding. Differential diagnoses include foreign oppressed desease of esophagus, malignant esophageal tumors such as squamous or adenomatous carcinomas or leiomyosarcoma and other benign tumors. Barium swallow is the most commonly used radiologic test for esophageal lesions (4). The finding on barium swallow is a smooth filling defect in esophageal lumen without a mucosal abnormality. More esophagus was involved in large esophageal leiomyoma, the mucosa of diseased region become thinnings, and show hyperaemia. So it can be regarded as mucosa destroy when barium swallow examination, that misdiagnosis as esophageal cancer (8) or esophageal stromal tumor. Computed Tomography scans of the chest show in most cases a mass originating from esophagus without mediastinal lymphadenopathy, but in giant leiomyoma, tumor usually grow toward outside of esophageal lumen. To form soft tissue

shadow in mediastinum, it can be misdiagnosised as mediastinal mass. The diagnosis is difficault to do and may cause diagnostic confusion. So to posterior mediastinum mass that close neighbor esophagus, it maybe a esophageal leiomyoma, this is worthy think highly (11). Esophagoscopy is also used for the diagnosis of esophageal leiomyoma, but it only shows submucosal lesions and will not lead to an accurate diagnosis (4,12). The use of the EUS can clearly reveal the structure of the esophageal wall. On EUS, leiomyoma presents as a homogeneous and hypoechoic lesion with clear margins, surrounded by a hyperechoic area (12), which can easily be differentiated from a lipoma, cyst, or hemangioma in the esophageal wall. Preoperative biopsy of the tumor is a debating issue (13). Our policy is not to recommend it, because the tumor is easily adhesive to the mucosa and the mucosal damage occurs accidentally during enucleation. Moreover, in many cases biopsy could not provide enough material to establish an accurate histopathological diagnosis.

Once the clinical diagnosis of leiomyoma is established, many factors must be considered for the optimal treatment. Tumor size and location are important, but also the patient's symptoms, general condition, and comorbidities should be taken into account. The surgical indications of these tumors include unremitting symptoms, increased tumor size, mucosal ulceration, histopathologic diagnosis, and facilitation of other surgical procedures (7). Because malignant transformation in leiomyomas is rare, some authors recommend regular follow-up with barium swallow and endoscopy for asymptomatic patients with lesions smaller than 5 cm and when the preoperative workup has excluded malignancy (7,13). We suggest that a leiomyoma should be removed when diagnosed even when asymptomatic, because there is always the possibility, rarely though, of malignant transformation.

For leiomyoma, the location and size of the tumor are important factors in determining the appropriate surgical approach. Endoscopic approaches appear possible in case of small pedunculated tumors of 2-4 cm originating from the muscularis mucosae (14). Usually to execute endoscopic mucosal resection (EMR) or endoscopic submucosal dissection (ESD). Symptomatic small leiomyomas <5 cm can be enucleated either by open surgery (15,16) or by means of video-assisted thoracoscopy (VATS) (17). Transthoracic extramucosal blunt enucleation via a left- or right-sided thoracotomy is the most common procedure for small- to midsized esophageal leiomyoma, which is easier, faster, and safer compared to resection (1,7). Low tumors and tumors of the esophago-gastric junction can be approached via upper midline laparotomy (15). After nucleation, the muscular wall should be closed to avoid diverticular-like mucosal bulging and for the preservation of the muscular propulsing activity. The larger the tumor that diameter >5 cm, associated with muscle atrophy and more muscular defects. The muscular wall should be repaied with pedunculated pleural film, diaphragm valve, or omentum, lung, pericardium. Postoperation feeding time should be delayed to avoid fistula, diverticula. Strengthen the lower esophageal mucosa, esophageal mucosa even weak are protected, and can effectively prevent to form postoperative gastric acid reflux and esophageal diverticulum. For giant esophageal leiomyoma should be preferred esophageal resection and reconstruction surgery, not the same as conventional enucleation of tumor. Because: (i) It was technically difficult to only enucleate giant leiomyoma, and the defect of esophageal muscle can't ensure wound healing; (ii) esophageal muscle was pressed by giant leiomyoma that expansive growth, became thinning and membranous. So lower esophageal sphincter dysfunction or loss. Iterature has reported that patients after removal of giant leiomyoma prone to symptoms of reflux esophagitis (18); (iii) For giant esophageal leiomyoma, it may have leiomyosarcoma-like transformation or with small leiomyosarcoma-like lesions (19); (iv) Meanwhile, the huge tumor endangered the physical health of patients by dysphagia. Or may be combined esophageal cancer (20), so as soon as possible surgery. To perform partial or subtotal resection of the esophagus, and esophagogastric anastomosis, the results are satisfactory. In our patient, because of the size, the location and the confusion diagnosis of the tumor, it was extramucosal blunt enucleated using thoracotomic approach.

In conclusion, diagnosis of esophageal leiomyomas requires both endoscopic and radiologic examinations. Once the clinical diagnosis of leiomyoma is established, the operation should be performed to remove the tumor.

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