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Original Article

Diagnosis and comprehensive treatment of esophageal leiomyoma: clinical analysis of 77 patients

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Abstract: To investigate the diagnosis and comprehensive treatment of esophageal leiomyoma. The clinical data of 77 cases of esophageal leiomyoma patients were analyzed between 2005 and 2013. Its diagnosis, treatment and prognosis were analyzed. 39 cases of patients were with eating choking feeling, 18 cases presented with chest pain and weight loss and 20 cases without any symptoms. Preoperative endoscopic ultrasonography of each patient was diagnosed as possibility of esophageal submucosal tumor. 3 All patients underwent tumor enucleation, in which tumor electrotomy under gastroscope were done for 2 cases, complete video-assisted thoracoscopic (CVATS) resection of tumor for 24 cases, thoracoscope assisted small incision tumor resection for 29 cases, conventional thoracic tumor resection for 22 cases. The comparison and the difference of complete video-assisted thoracoscopic surgery group and the thoracoscope assisted small incision group for the operation time, bleeding volume, drainage volume, extubation time, hospitalization time and fasting time were not statistically significant ($P < 0.05$). All the patients recovered well and postoperative pathology of each patient was esophageal leiomyoma. They were followed up for 6 months to 8 years, average for 4 years, not recurrence of esophageal leiomyoma. Endoscopic ultrasonography is the most accurate method in diagnosis of esophageal leiomyoma. Esophageal leiomyoma which less than 1.0 cm in diameter, regular shape, originated in the muscularis mucosa, endoscopic electrotomy can be used as the preferred; Surgical operation is the main treatment of esophageal leiomyoma, three kinds of operation way has its own corresponding clinical indications, according to the clinical characteristics of patients and operator' habits to choose the corresponding operation way, all can achieve good treatment effect.

Keywords: Esophageal leiomyoma, diagnosis, comprehensive treatment, prognosis

Introduction

The majority of esophageal submucosal tumor is benign, of which leiomyoma is the most common type, accounting for about 90% [1, 2]. The development of endoscopic ultrasonography and thoracoscopic techniques has opened the door for the diagnosis and treatment of esophageal leiomyoma widely. 77 cases with esophageal leiomyoma were treated in our division from 2005 to 2013, accounting for 91.7% of the same period of benign tumors of the esophagus operation (77/84). By analyzing the 77 cases of patients with clinical data and combing with the literature, we investigate the diagnosis, comprehensive treatment and prognosis of esophageal leiomyoma.

Materials and methods

Patients

Between 2005 to 2013, a total of 77 cases of patients with esophageal leiomyoma were treated in our division, 56 cases were male, 21 were female, ratio of male to female was 2.7:1, the age ranged from 17 to 76 years old, average for 43 years old. Drinkers were 32 people (male for 28, female for 4). Smokers were 44 people, all male.

39 cases were with eating choking feeling, among which 11 cases could only take liquid diet. 18 cases with chest pain and thin, those without any symptoms were 20 cases. Except

Figure 1. Barium meal examination revealed esophageal lumen barium filling defect.

Figure 2. CT scan showed tumor was irregular shadow, with clear boundary.

for those no clinical symptoms, medical history was 3 months to 10 years.

Diagnostic methods

77 patients underwent preoperative chest CT, esophageal barium meal, electronic gastroscope and ultrasonic gastroscopy. Of which 8 patients received preoperative gastroscopic biopsy. Esophageal barium meal examination revealed esophageal lumen barium filling defect with different sizes, the local mucosal looked like "daubing syndrome" (**Figure 1**). CT scan of the chest showed the tumors were round homogeneous or irregular shadow, with clear boundary, tumor not enhanced after enhancing (**Figure 2**). Electronic endoscopy showed esophageal mucosa with eminence

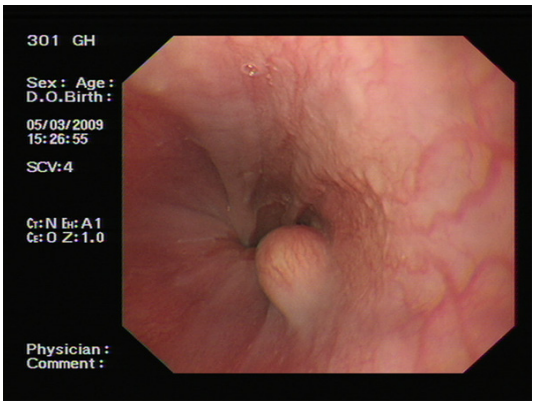

Figure 3. Endoscopy showed esophageal mucosa with regular eminence lesion, mucosal surface was smooth.

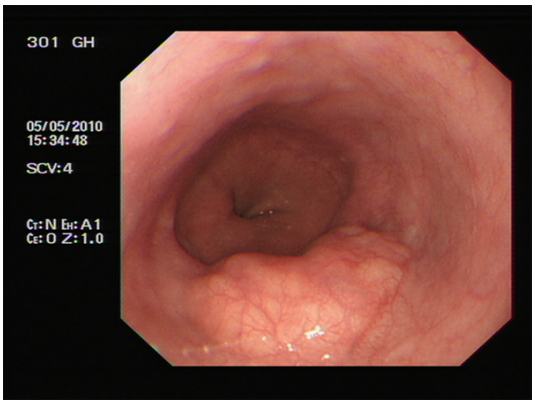

Figure 4. Endoscopy showed esophageal mucosa with irregular eminence lesion, mucosal surface was smooth.

lesions, mucosal surfaces were smooth, touch with the sense of movement (**Figures 3**, **4**). Under gastroscope, for 39 cases esophageal lesions were stenosis, including 11 cases with obvious stenosis. Ultrasound gastroscope examination confirmed that 2 cases of the tumors were originated from the muscularis mucosa, muscularis propria for 75 cases (**Figures 5**, **6**). 75 cases with single tumor, 2 cases were multiple. Tumor size and location were seen in **Table 1**. Tumor size was measured in length diameter by transrectal ultrasonography. All the patients were diagnosed as esophageal submucosal tumor before the operation.

Treatment methods

Under gastroscope electric excision: under local anesthesia of throat place of gastroscope,

Figure 5. Ultrasound gastroscope examination confirmed that tumor was originated from the muscularis mucosa.

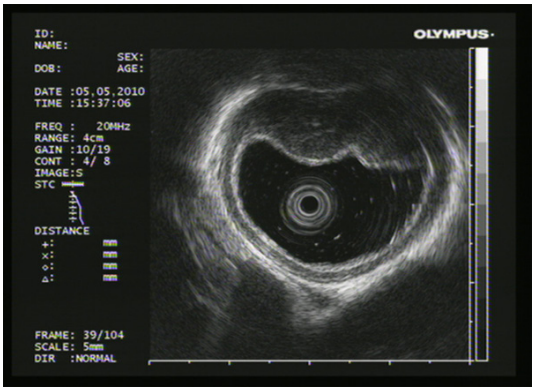

Figure 6. Ultrasound gastroscope examination confirmed that tumor was originated from the muscularis propria.

after determining the location of lesion and range, injected of saline in the lesion of the distal, proximal and both sides of submucosal, made the lesions completely uplift, then conducted the cavity ultrasonography to confirm tumor originated in the muscular layer of mucosa, and separated from the sticky submucosa and muscularis propria. Direct ligation by snare after electrifying tumor resection, hemostasis was done by wound electric coagulation.

The surgical treatment

Double lumen endotracheal intubation anesthesia, one lung for ventilation. Lateral position, way rate leaning forward, the operator stands at the back side of the patient. Operation approach depends on the lesion site, in middle and upper esophageal leiomyoma, uses the right thoracic approach; the lower esophageal leiomyoma often uses the left thoracic approach. Three kinds of surgical incision selection are as follows: (1) complete thoracoscopic operation: Thoracoscopic observation hole selects eighth intercostal space of posterior axillary line, choose another 2 operated holes for 1.0~2.0 cm or single operated hole for 3.0~5.0 cm according to the location of the lesion, not pulling ribs. (2) The thoracoscope assisted small incision operation: Thoracoscopic observation hole selects eighth intercostal space of posterior axillary line, according to the lesion location select subaxillary small incision for 3.0~5.0 cm, slightly enlarged incision for intercostal muscles, with special small incision retractor for the rib, only operation apparatus can enter the intrathoracic operation. (3) The conventional open chest operation: take lateral outside incision, length is between 15~25 cm.

Exposed esophageal bed, explored the tumor, if the location was not clear, could be implanted into the gastroscope to assist positioning. When necessary, we could free part of the esophagus in order to expose the tumor on the opposite wall. After accurate positioning of tumor, incision esophagus muscular layer of tumor surface, blunt dissection of tumor; wire transfixing suture to pull the tumor body to facilitate the complete removal of tumor. The stomach tube was back to the tumor segment of esophagus, clipped lower esophagus of tumor section, thoracic cavity was injected of water, rapid injected of air by stomach tube, observed if esophageal mucosa was damaged, no gas escape meant the esophageal mucosa was intact. When gas escaping, seek for mucosal crack, if the crack was small, repaired by 5-0 absorbable suture. If the esophageal mucosa crevasse was bigger, mucosal defect range accounted for more than 1/2 of the local esophageal wall, that forcibly sutured mucosal would lead to severe stenosis of local esophageal lumen, at this time mostly adopt subtotal esophagectomy, esophageal gastro anastomosis. Examine of tumor specimens, if the tumor regular shaped, surface was smooth, capsule was integrity and hard, did not need rapid intraoperative pathological examination; otherwise needed to do intraoperative frozen pathological examination to exclude malignancy stromal tumors, if proved as malignant tumor, operation way would be changed to subtotal esophagectomy, esophageal gastro anastomosis. At

Table 1. Comparison of patients' general information of operation way for three groups

Group	Case	Age	Tumor size (cm)	Tumor site			Operation way	
				Upper	Middle	Lower	Tumor dissection	Esophageal resection
CVATS	24	37.12±2.23	2.40±0.50	6	8	10	24	0
Small incision	29	38.31±1.90	3.00±0.24	6	10	13	29	0
Thoracotomy	22	49.80±4.51	8.60±3.45	4	8	10	20	2

Table 2. Operation effect comparison of total endoscopic group and small incision group

Group	Operation time (min)	Bleeding volume (ml)	Drainage volume (ml)	Extubation time (day)	Fasting time (day)	Complications
CVATS	180.91±12.50	97.13±15.20	213.10±70.24	4.11±0.99	4.12±1.12	none
Small incision	175.60±14.32	103.11±2.23	209.19±88.29	4.19±1.18	4.13±1.21	none
t value	0.984	-1.348	1.046	-1.543	-1.435	
p value	0.221	0.190	0.284	0.145	0.179	

the end put the stomach tube into stomach again, closed thoracic cavity layer by layer.

The operator and pathologists are PLA General Hospital attending physician, and the research method has been agreed by the medical ethics committee of the hospital.

Statistical analysis

Statistical analysis was performed using SPSS19.0 software. Measurement data was presented with the mean and standard deviation. Difference comparison in counted data between groups were analyzed by chi square test, the difference comparison of measured data between groups were used by independent samples t test, *P* value < 0.05 means the difference with statistical significance.

Results

42 patients were with right pleural operation, 33 cases with left chest operation. Patients general information of operation way for three groups were seen in **Table 1**, between group data for complete thoracoscopic group and thoracoscope assisted small incision group, for the comparison with the age, size and location of the tumor, the difference was not statistically significant (*P* > 0.05). Between group data for thoracoscopic minimally invasive group and conventional thoracotomy group, for the comparison with age, tumor size, the difference was statistically significant (*P* < 0.05). 11 cases of patients with ruptured mucosa during operation, 9 cases were repaired with 5-0 absorbable suture, 2 cases underwent subtotal esophagectomy and esophageal-gastro anastomosis for too large mucosa defect. 4 cases were found tumor by gastroscopy for their tumor too small or multiple. Endoscopic transurethral resection were 2 cases, tumor diameter were 0.2 cm and 1.0 cm, respectively.

All patients operated smoothly, no esophageal fistula or anastomotic leakage occurred, no cases of death. Operation effect comparison for complete thoracoscopic surgery group and the thoracoscope assisted small incision group is in **Table 2**, between the two groups, for the comparison of the operation time, bleeding volume, drainage volume, extubation time, hospitalization time and fasting time, the difference was not statistically significant (P > 0.05).

Pathological analysis

Tumor of 64 cases was round (**Figure 7**), 13 cases of irregular shape (**Figure 8**). Intraoperative frozen examination were 16 cases, all were benign leiomyoma. The postoperative pathology was confirmed as esophageal leiomyoma, tumor tissue was composed of smooth muscle cells for a large number spindle shaped arrangement (**Figure 9**).

Follow-up

The period of follow-up was 6 months to 8 years, average 4.8 years, all patients after operation recovered to normal life or work. No recurrence of esophageal leiomyoma.

Figure 7. Tumor was elliptical and regular.

Figure 8. Tumor was irregular shape.

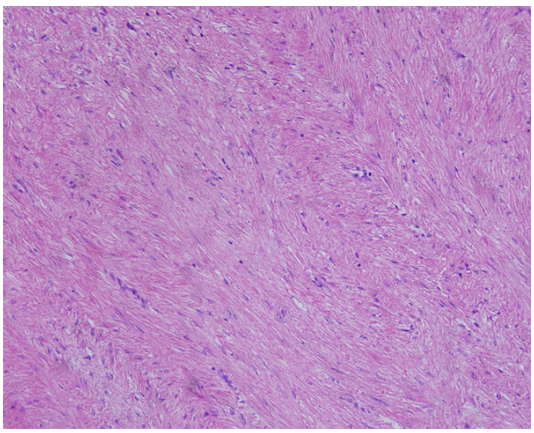

Figure 9. Tumor tissue was composed of smooth muscle cells with spindle shaped arrangement.

Discussion

Esophageal leiomyoma is the most common benign tumor of esophagus. The male to female ratio is about 3:1, the age is above 40 years old. Multiple tumors are in lower esophageal [2, 3]. It is still unclear for the etiology and pathogenesis of esophageal leiomyoma. Tumor growth is slow, long diameter is within 5 cm. Huge leiomyoma of esophagus has been reported, weight up to 5 kg [4].

The diagnosis technology of esophageal leiomyoma: the most common clinical symptoms of esophageal leiomyoma patients is dysphagia, followed by chest pain; these symptoms are due to esophageal stenosis or compression of surrounding tissue caused by large tumor size. When the tumor diameter is less than 2 cm, no obvious symptoms are found except for checking the body. Esophageal barium meal examination is the most commonly means for diagnosis of esophageal leiomyoma, manifested as partial lumen barium filling defect, the local mucosal liked “daubing syndrome”. The method is simple and easy, which has an important significance in diagnosis of esophageal leiomyoma [5]. Chest CT scan showed mass with homogeneous density in the esophageal wall, when strengthen with weak enhancement [2, 5]. It’s visible for tumor protruding into the lumen under gastroscope, a hemisphere or polypoid shaped, esophageal mucosa was smooth [6, 7].

Endoscopic ultrasonography has become the most important means for diagnosis of esophageal leiomyoma, which can clearly display the 5 layer structure of esophageal wall, from inside to outside were mucous layer (high echo), muscular layer of mucosa (low echo), submucosa (high echo), muscularis propria (low echo) and adventitial layer (high echo). The vast majority of esophageal leiomyoma originated from the muscularis propria layer, a small part may origin from muscular layer of mucosa. The tumor showed low acoustic shadow with clear boundary, homogeneous density [7, 8]. Endoscopic ultrasonography in this group showed the tumor origin from muscular layer of mucosa were 2 cases, the origin from muscularis propria were 75 cases. The judgment by preoperative endoscopic ultrasonography of esophageal submucosal tumor origin showed complete agreement with the operation.

The manifestation of leiomyoma of esophagus and esophageal stromal tumors under endoscopic ultrasonography are very similar, if the

tumor is larger or irregular shaped, endoscopic doctors tend to esophageal stromal tumor [7, 8]. In this group, under endoscopic ultrasonography, 61 cases were with preoperative diagnosis of esophageal leiomyoma possibility, 16 cases with esophageal stromal tumor possibility; endoscopic ultrasonography qualitative diagnosis rate of esophageal submucosal tumors was 93.2% (61/77). Currently endoscopic ultrasonography has become the gold standard for clinical diagnosis of esophageal leiomyoma.

For esophageal submucosal tumor, preoperative endoscopic biopsy of the tumor is not desirable; for biopsy will lead to tumor and local esophageal mucosa adhesion closer, which increasing the risk of esophageal mucosa rupture during operation [9, 10]. 8 cases in this group underwent routine preoperative gastroscopic biopsy, did not achieve positive pathological results, and 5 cases occurred esophageal mucosal tears. Therefore we suggest that if the ultrasonic gastroscopy showed lesions of esophageal submucosal tumors, no endoscopic biopsy.

Treatment and prognosis of esophageal leiomyoma: if patients with esophageal leiomyoma are with smaller tumor size (< 5.0 mm) and without any discomfort symptoms, can choose the regular examination of gastroscopy; otherwise it shall actively seek operation treatment, to prevent tumors grow up or malignant. There are three kinds of esophageal leiomyoma treatment, endoscopic tumor ring ligation, thoracoscopic esophageal leiomyoma stripping and thoracic esophageal leiomyoma stripping operation.

The way of operation depends on the size, shape, position of the tumor. If the tumor is less than 1.0 cm, with regular shape, and is located in the muscularis mucosa, can consider endoscopic resection of the ferrule [6, 7]. Otherwise, should try to choose the surgical operation treatment, so as to avoid the risk of esophageal perforation.

2 cases in our group underwent the gastroscopic resection, resection tumor diameter were 0.6 cm and 0.9 cm, both are derived in the muscular layer of mucosa, fasting for 3 days after the operation, with a smooth recovery.

Outside the esophageal mucosa enucleation of leiomyoma is the most widely used and best effective treatment method. Most of the patients only undergo tumor dissection, rarely patients need to choose esophagectomy and esophageal gastro anastomosis. Operation methods include conventional open chest operation and thoracoscopy assisted minimally invasive operation. Conventional open chest operation mainly for the following situations [1, 2]: 1. pleural cavity extensive adhesion or intolerant of unilateral pulmonary ventilation; 2 huge leiomyoma of the esophagus or tumor stripping is difficult; 3 intraoperative frozen pathology is malignancy. Indications of thoracoscopic minimally invasive operation are: 1 pleural cavity without adhesion and can tolerance unilateral pulmonary ventilation; diameter of 2 tumor is between 2~5 cm and with regular shape; 3 tumors are easy to peel off [10-12].

Operation approach is generally determined by the location of the lesions and whether exposure easily, on the high and middle thoracic esophageal leiomyoma, mostly take the right side of the 4 or 5 intercostal to space, the low and abdominal segment of esophagus should take the left side of the 6 or 7 intercostal to space. Conventional open chest operation is suitable for patients who were aged, tumor was difficult to peel or need for esophageal resection. Minimally invasive operation for thoracic wall small incision, has advantages include pain relieving, avoid liquefied incisional infection and others, also provides the operation opportunity for patients refusal to open operation. An operator with rich experience of conventional operations, if master of thoracoscopic instruments operation, with the help of image system of high definition, he can safely complete resection of esophageal leiomyoma, reach with the same effect of routine open chest operation treatment [12, 13].

Regardless of the way of operation, should protect esophageal tissue around it, avoid sharp separation, and ensure the blood supply of esophagus during the operation. When cut the esophagus muscular layer, should confirm the incision in esophageal tumor by side, otherwise easily break the esophageal mucosa, increasing the risk of esophageal pleural fistula after operation. The most effective method is operation located by gastroscope or touch local gas-

tric tube by finger to determine esophageal wall of tumor side, thin wall is normal esophageal wall, the thick wall is the esophageal wall of tumor side. The focus of operation is complete stripping of tumor and avoid of esophageal mucosa injury. If Difficulties in tumor dissection should be timely expansion of incision, don't neglect the operation feasibility for the pursuit of small incision.

Excision in this group of complete thoracoscopic tumor were 24 cases, thoracoscope assisted small incision tumor resection were 29 cases, conventional open chest routine open chest operation resection of tumor were 22 cases. All groups of patients recovered well after the operation, diet was good, follow-up without recurrence of esophageal leiomyoma. So we think that the choice of way of operation of esophageal leiomyoma depends on the general condition, tumor size, operation exploration results of patients and the ability and habit of the operator etc. No matter what operation way, points which decide whether the success of operation are as follows: 1. Accurately determine the orientation of tumor body in the esophageal wall. 2. Complete resection of the tumor and ensure the integrity of local esophageal mucosa. 3. Reserve vagus nerve and blood supply of the esophagus.

Conclusion

In short, esophageal leiomyoma is a benign tumor with slow progress [14], for patients who suspected esophageal leiomyoma should undergo endoscopic ultrasonography to diagnose clearly, avoiding endoscopic biopsy. Surgical operation is the most effective method, and the prognosis is good.

Disclosure of conflict of interest

None.

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