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Approach to esophageal leiomyoma: a report of eight cases

Özofageal leiyomyoma yaklaşım: Sekiz olgu sunumu

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Background: This study aims to evaluate outcomes of surgical treatment of esophageal leiomyomas.

Methods: Between January 1999 and December 2011, eight cases (3 females, 5 males; mean age 41.5 years; range 30 to 66 years) who were operated due to leiomyoma were retrospectively analyzed. Age and sex of the patients, symptoms, localization of lesions, diagnosis, surgical modality, length of hospital stay and morbidity and mortality rates were reviewed.

Results: Six patients had dysphagia. Leiomyoma was localized at 1/3 mid-esophagus in five patients and at 1/3 distal esophagus in three patients. Three patients underwent thoracoscopic enucleation. Two patients underwent right thoracotomy, two with left thoracotomy and one with laparotomy and enucleation. The mean diameter of lesions was 5 cm (range, 2 to 7 cm). At three months following surgery, pseudodiverticulum was seen in one patient who underwent thoracoscopic enucleation. No intraoperative mortality was seen. The mean length of hospital stay was eight days (range, 5 to 12 days). During a mean follow-up of 16.4 months (range, 4 to 56 months), no recurrence or malignancy were observed.

Conclusion: Enucleation is an effective and safe diagnostic and therapeutic modality in patients with leiomyoma. Muscular layer should be also closed to prevent possible mucosal defects.

Key words: Esophagus; leiomyoma; surgery.

Amaç: Bu çalışmada özofageal leiyomyomlarda cerrahi tedavi sonuçları değerlendirildi.

Çalışma planı: Kliniğimizde Ocak 1999 - Aralık 2011 tarihleri arasında leiyomyom nedeni ile ameliyat edilen sekiz olgu (3 kadın, 5 erkek; ort. yaş 41.5 yıl; dağılım 30-66 yıl) retrospektif olarak incelendi. Hastaların yaşı ve cinsiyeti, semptomlar, lezyonun yerleşim yeri, tanısı, cerrahi yöntem, hastane yatış süreleri ile morbidite ve mortalite oranları gözden geçirildi.

Bulgular: Altı hastada disfaji vardı. Leiyomyom beş hastada özofagus 1/3 orta, üç hastada 1/3 distal kesimde yerleşmiş idi. Üç hastaya torakoskopik enükleasyon uygulandı. İki olguya sağ torakotomi, iki olguya sol torakotomi, bir olguya ise laparotomi ve enükleasyon uygulandı. Lezyon çapı ortalama 5 cm (dağılım, 2-7 cm) idi. Torakoskopik olarak enükleasyon uygulanan bir hastada ameliyattan üç ay sonra psödodivertikül gelişti. Ameliyat sırasında mortalite gözlenmedi. Ortalama hastane yatış süresi sekiz gün (dağılım 5-12 gün) idi. Ortalama 16.4 aylık (dağılım, 4-56 ay) takip süresince nüks veya malignensi gözlenmedi.

Sonuç: Leiyomyom olgularında enükleasyon etkili ve güvenli bir tanı ve tedavi yöntemidir. Olası mukozal defektleri önlemek için de musküler tabaka kapatılmalıdır.

Anahtar sözcükler: Özofagus; leiyomiyom; cerrahi.

Leiomyoma is a smooth muscle cell tumor of the esophagus that originates from the muscularis propria. Although it is the most common benign tumor in this organ, leiomyoma has only been found in 0.1-0.006% of autopsies.[1] In addition, it constitutes less than 1% of all esophageal neoplasms[2] and makes up 10% of gastrointestinal tumors along with 70-80% of esophageal benign tumors.[1,2]

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Leiomyomas of the esophageal wall grow slowly. They are usually asymptomatic and are discovered incidentally from observations made for other reasons. Resection in leiomyoma patients remains controversial; therefore, this study aims to evaluate the results of this surgical treatment.

PATIENTS AND METHODS

In this study, we retrospectively reviewed the cases of eight consecutive patients (3 females, 5 males; mean age 41.5 years; range 30 to 66 years) who were surgically treated for esophageal leiomyoma in the Department of Thoracic Surgery, Medical Faculty of Atatürk University, between January 1999 and December 2011. All cases were preoperatively examined using direct chest radiography, computed tomography (CT), and esophagoscopies, with magnetic resonance imaging (MRI) also being employed in two cases.

Our data was obtained through patient files, recordings of surgical interventions, esophagoscopy and pathology reports, and outpatient clinical notes, and the results were compared with the data from the available literature.

RESULTS

Six of the patients had dysphagia while two were diagnosed with dyspepsia. In addition, one suffered from a cough, one had sputum, and one had chest pain. All of the patients underwent a standard examination, and intact mucosal and submucosal lesions were detected via esophagoscopy in all cases. After endoscopic and tomographic evaluations, the preliminary diagnosis was leiomyoma for all of the patients, with five having tumors in the middle third of the esophagus and three having them in the distal third. Three of the patients with leiomyomas in the middle third of the esophagus underwent thoracoscopies, and the other two had right thoracotomies. Additionally, two of patients with leiomyomas in the distal section of the esophagus underwent a left thoracotomy while a laparotomy was performed on the other.

Furthermore, all of the patients underwent esophageal myotomies. Enucleation was performed on the lesions, and solitary lesions were detected in all of the cases (Figures 1-3). After examination of the frozen sections, leiomyoma was confirmed, with the tumors having a mean lesion diameter of 5 cm (range, 2-7 cm). After the enucleation, saline was inserted into the enucleated area, and air was injected into the nasogastric tube to investigate the possibility of mucosal injuries, but none were reported. The esophageal muscle layers were then primarily closed with absorbable sutures, except for the first two cases which underwent a thoracoscopy. At the third postoperative month, a pseudodiverticulum was seen in one of these patients. The muscle layer of the third thoracoscopic case was also primarily closed. No perioperative mortality was seen in any of the cases, and the average length of hospital stay was eight days (range, 5-12 days). In addition, there was no recurrence or malignancy during the follow-up period, which averaged 16.4 months (range, 4-56 months). The clinical characteristics of the patients and treatment methods are summarized in Table 1.

DISCUSSION

Leiomyomas are usually seen in patients ranging from their second decade of life to their fifth decade, with twice the number of cases occurring in men. In addition, 80% of the cases occur in the middle or lower third of the esophagus, and they are usually seen as single, smooth-edged submucosal masses. In rare instances, multiple tumors can appear.[1-8] Intramural lesions are round or oval with smooth edges that come in various sizes. At times they may be lobulated and can wind

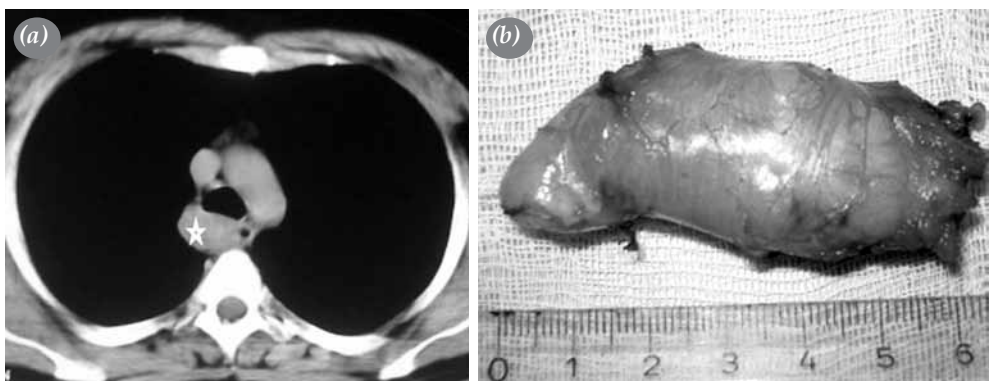

Figure 1. (a) Tomographic view of a 30-year-old male patient with leiomyoma located in the middle third of the esophagus. **(b)** The view after resection.

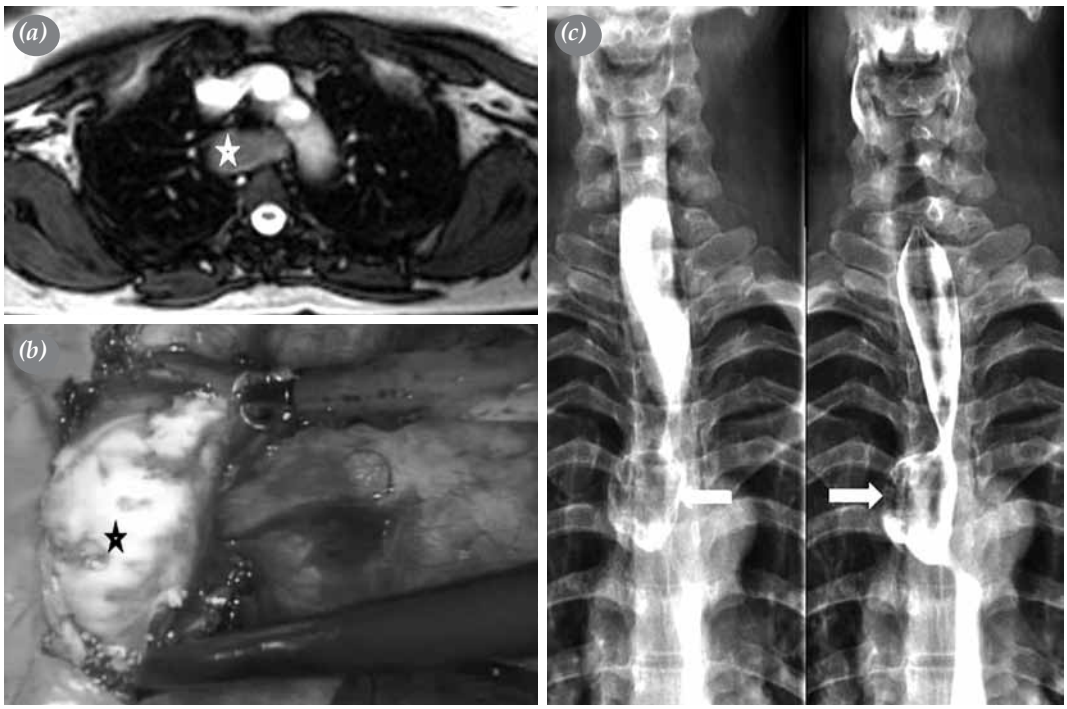

Figure 2. (a) Magnetic resonance imaging of a 49-year-old female patient with leiomyoma located in the middle third of the esophagus. **(b)** Intraoperative view of the leiomyoma. **(c)** A pseudodiverticulum in the lesion area three months postoperatively as revealed by barium esophagography.

around the wall of the esophagus. In this study, all of the patients ranged in age from their twenties to their fifties with the exception of one patient who was 66 years old. As previously mentioned, five cases had leiomyomas in the middle third of the esophagus and three in the distal third. Moreover, the subjects had single, smooth-edged leiomyomas in the form of submucosal lesions. No direct relationship between tumor size and symptoms has been verified;[7] however, Mutrie et al.[1] reported a significant association between these two variables in their case study of 53 patients. They determined that 31 cases were symptomatic and that the median tumor size of these patients was 5.3 cm. Their most common symptoms included dysphagia and nonspecific chest and retrosternal pain along with rare instances of regurgitation, epigastric tenderness, dyspnea, and weight loss. The average tumor size in our cases was 5 cm. There were six patients with dysphagia, and one patient without dysphagia had dyspepsia. Another had symptoms of cough and sputum.

Leiomyoma diagnosis is confirmed via a barium esophagogram, esophagoscopy, or CT. Esophageal

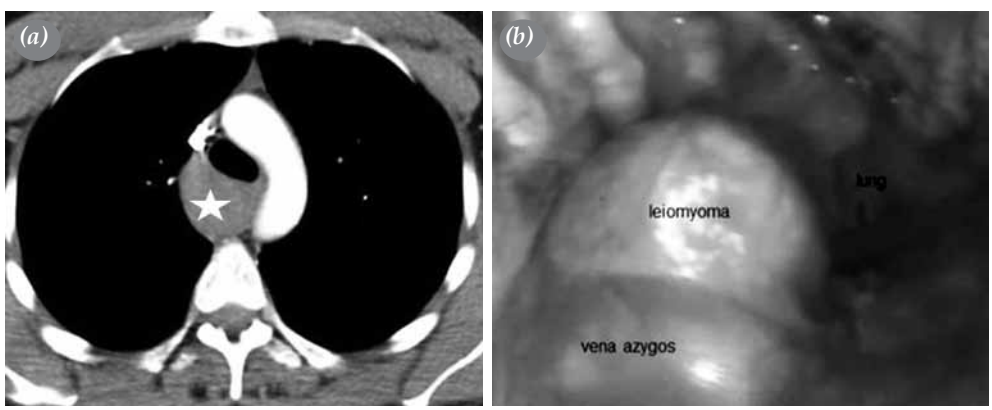

Figure 3. (a) Tomographic view of a 33-year-old male patient with leiomyoma located in the middle third of the esophagus. **(b)** Intraoperative thoracoscopic view before opening the mediastinal pleura.

Table 1. Patient characteristics

Cases	Age/gender	Presenting	Localization Symptoms	Surgical method	Tumor diameter	Complication	Hospital stay (days)
1	38/M	Dysphagia, dyspepsia	Middle third	Right thoracotomy	2 cm		12
2	30/M	Dysphagia	Middle third	Right thoracotomy	5 cm		8
3	50/M	Dysphagia	Distal third	Left thoracotomy	7 cm		9
4	34/F	Dysphagia, chest pain	Distal third	Left thoracotomy	4 cm		6
5	30/M	Cough, sputum	Middle third	VATS (right)	6 cm		5
6	49/F	Dysphagia	Middle third	VATS (right)	6 cm	Pseudodiverticulum	7
7	33/M	Dyspepsia	Middle third	VATS (right)	7 cm		11
8	66/F	Dysphagia	Distal third	Laparotomy	3 cm		6

VATS: Video-assisted thoracoscopic surgery.

leiomyomas can be seen as a posterior mediastinal mass on chest radiographs, but the radiological findings can be viewed as coincidental. A smooth, crescent-shaped filling defect can be seen at the mucosa of the anomaly-free esophagus contour on barium X-rays. The tumor is usually mobile during swallowing.[8-10] Leiomyomas have weak homogeneity in contrast CTs, and differential diagnoses resulting from neurofibroma, hemangioma, and other esophageal tumors, such as fibroma, are difficult.[7,9] They are usually seen as isointense submucosal lesions in MRI T_2 sequences and can be distinguished from esophageal carcinoma by their high signal appearance in the T_2 sequences.[9] Esophagoscopies preserve mucosal integrity, and smooth-edged lesions growing into the lumen are characteristically found via this procedure. Recently, endoscopic ultrasonography (EUS) has been extremely beneficial in the identification of benign lesions. However, it is not recommended for leiomyoma patients because needle aspiration is not suitable for them.[1,2,11] In this study, we utilized esophagoscopies and CT in all of the cases, and an MRI was also performed on two of the patients.

Malignant transformation is very rare in leiomyomas, with a reported rate of 0.2% in the literature.[1] In some instances, coincidental cases of esophageal carcinoma and leiomyoma have been reported.[12]

There is no consensus on the management of leiomyoma. Some authors have advocated removal, even if the patients are asymptomatic at diagnosis, whereas some have recommended frequent follow-up at regular intervals for asymptomatic patients and those with small leiomyomas. Both diagnosis and treatment are carried out at the same time during surgery. An operation is indicated in cases with a continued increase in tumor size and mucosal ulceration, which can also be used to facilitate histopathological diagnosis and other surgical procedures.[11] We think removal should be done in patients suspected of having leiomyomas, even if they are asymptomatic. If there is no treatment or if it is delayed, serious symptoms can present, and malignant transformation might even occur in the future, though this is a rare event.

Leiomyomas are located between the muscle layers and hold surrounding tissues loosely. They can be easily dissected with blunt dissection. For this reason, enucleation was our consensus choice for the surgical procedure. However, in rare cases where there is a very large tumor or leiomyomatosis, a resection may be required.[13,14]

Single or multiple tumors along with their size and localization can be used as guides for deciding whether a patient should have surgery. The surgical approach can be achieved by either a thoracotomy or thoracoscopy. Right thoracotomies are preferred since they provide excellent exposure for almost all transthoracic esophageal lesions. Left thoracotomies, thoracoabdominal incisions, or laparoscopies are preferred in lesions situated in the distal section.[11,13,15] In this study, we had five cases located in the middle third of the esophagus with right hemithorax, and we performed video-assisted thoracoscopic surgery (VATS) on three of the patients while two underwent thoracotomies. Enucleation was also performed in these cases since the surgeon had experience with this procedure. In addition, two of three cases with leiomyomas in the distal portion of the esophagus underwent left thoracotomies, and the other, which was located close to the gastroesophageal junction, had a midline laparotomy.

Bardini et al.[6] first described video thoracoscopic enucleation in esophageal leiomyomas in 1992. Since then, operative approaches have changed in favor of thoracoscopic procedures.[2] When compared with thoracotomies, VATS has cosmetic advantages

while also providing less operative trauma, less pain, and better postoperative pulmonary functions. Video-assisted thoracoscopic surgery is also preferred when there is only a single esophageal leiomyoma measuring between 1 and 5 cm. The possibility of a thoracotomy increases with larger leiomyomas.[6,11,13,15] No intraoperative mortality has been reported with regard to esophageal leiomyomas treated with VATS enucleation. However, 13.3% of the patients experience a mucosal tear, 10% experience postoperative mucosal bulging, 3.3% have pleural effusion, and 3.3% suffer from epigastric pain.[13,16] In our study, three cases underwent thoracoscopic surgery, and in two of these, the tumor diameters were 6 cm and 7 cm. However, a thoracotomy was not needed for these patients.

There is no consensus in the literature regarding closing the myotomy area after enucleation. However, the general opinion is that re-emerging muscular layers are needed in order to prevent mucosal deformation.[13,16] Bardini and Asolati[15] reported a pseudodiverticulum in a thoracoscopic resection case because the muscular layer had not been closed. In this study, all muscle layers were closed except for the two VATS cases, and a postoperative pseudodiverticulum was seen in the muscle layer in one of these patients. For this reason, we believe that the muscle layers should always be closed.

In addition, mucosal injuries should be kept in mind during leiomyoma enucleation. An unrecognized mucosal defect can lead to very serious postoperative outcomes. Hence, after enucleation, we administered saline to the enucleated area and pumped pressured air through a nasogastric probe to investigate the possibility of mucosal injuries in all of our thoracoscopic and surgical cases. In some cases, the hospital may make the patient to drink methylene blue when postoperative mucosal injury is suspected. Furthermore, draining methylene blue from a chest tube can be used for leakage control.[13,16]

Conclusion

In this study, the majority of esophageal leiomyoma patients were symptomatic, and we found thoracoscopic enucleation to be a safe and effective method for the diagnosis and treatment for leiomyomas smaller than 5 cm in diameter. We recognize that a thoracotomy may be necessary for large masses, but this should not be automatic. In addition, we conclude that the muscular layer should be closed with absorbable sutures placed separately in order to prevent mucosal defects.

Declaration of conflicting interests

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