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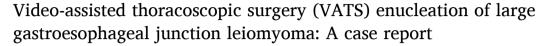
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Case report



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ABSTRACT

Introduction and importance: Esophageal leiomyomas are the most common benign esophageal tumors. They are typically smaller than 3 cm, but larger tumors can impede local structures to cause symptoms, including dysphagia and epigastric pain. Surgical treatment of esophageal leiomyomas has historically involved open thoracotomy, but this approach is being replaced by minimally invasive approaches, including video-assisted thoracoscopic surgery (VATS).

Case presentation: A 46-year-old female patient presented with upper abdominal pain. Computerized tomography (CT) scanning of the abdomen and chest revealed a large $(6.0 \times 4.0 \times 3.0 \text{ cm})$ gastroesophageal junction (GEJ) mass. An endoscopic ultrasound (EUS) with fine needle aspiration confirmed diagnosis of esophageal leiomyoma. A right VATS esophageal mass resection was performed to enucleate the mass. An intraoperative EGD was performed to check mucosal integrity, ensure adequate lumen patency, and visualization and insufflation was negative for a mucosal leak. The post-operative course was unremarkable.

Clinical discussion: This case report adds to the emerging evidence that VATS can be utilized for enucleation of larger leiomyomas (>5 cm in largest dimension). Additionally, the use of direct intraoperative endoscopic evaluation via esophagoscopy suggests that larger esophageal masses could potentially be enucleated with a combined VATS and endoscopic approach.

Conclusion: The purpose of this report is to add to the limited literature on minimally invasive surgical treatment of a relatively large GEJ leiomyoma. This case highlights that VATS, in addition to simultaneous endoscopic visualization, is an efficacious and safe option for treatment of larger leiomyomas (>5 cm) and can be associated with minimal risk.

1. Introduction and importance

Leiomyomas are benign tumors of smooth muscle origin, most commonly of the uterus, small bowel, or esophagus [1,2]. While esophageal leiomyomas are rare, only accounting for 1 % of total esophageal neoplasms, they are the most common mesenchymal esophageal tumors [3,4]. Esophageal leiomyomas can vary in size, but are typically smaller than 3 cm [5]. Patients with tumors of this size are usually asymptomatic, and the tumor is discovered incidentally via imaging. However, larger tumors can impede on local structures to cause dysphagia and epigastric or chest pain [5,6].

Surgical treatment is typically only recommended for leiomyomas that are symptomatic or larger than 5 cm [7]. The historical surgical approach for esophageal leiomyoma is an open thoracotomy followed by extra mucosal blunt enucleation [2]. This approach has been replaced over time by minimally invasive thoracoscopic approaches, including video-assisted thoracoscopic surgery (VATS) [8,9]. However, the size and location of the tumor can modify the surgical difficulty and the risk of the procedure. For surgeries that are considered to be higher risk, an open thoracotomy may still be the method of choice [2]. Here we present a case of a 46-year-old female patient with a large gastroesophageal junction (GEJ) leiomyoma that was treated with VATS. This work has

Abbreviations: VATS, video-assisted thoracoscopic surgery; GEJ, gastroesophageal junction; EGD, esophagogastroduodenoscopy; EUS, endoscopic ultrasound; GIST, gastrointestinal stromal tumor.

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been reported in line with the SCARE 2020 criteria [10].

2. Case presentation

A 46-year-old Hispanic female patient self-presented with two weeks of progressive worsening upper abdominal pain, as well as acute nausea and anorexia. She had no relevant personal or family medical history and has no history of smoking. No other abnormalities were detected on physical exam or laboratory tests. Computerized tomography (CT) scanning of the abdomen and chest revealed a large $(6.0 \times 4.0 \times 3.0 \text{ cm})$ GEJ mass displacing the esophagus to the left (Fig. 1), as well as a hiatal hernia.

An esophagogastroduodenoscopy (EGD) found submucosal bulging without an endoluminal component. There were no histopathological abnormalities found in the endoluminal biopsy samples. An endoscopic ultrasound (EUS) with fine needle aspiration was then performed on the submucosal lesion at the GEJ and revealed a hypoechoic mass of the muscular propria layer at the GEJ. Pathology confirmed the diagnosis of leiomyoma of the esophagus, distinguishing the leiomyoma from a gastrointestinal stromal tumor (GIST) [5].

The patient underwent general anesthesia and a right VATS esophageal mass resection was performed by an attending thoracic surgeon at a regional general hospital to enucleate the entire mass two months after initial presentation. The excised mass was 6.0 cm at largest diameter $(6.0\times4.0\times3.0$ cm) and weighed 31.3 g (Fig. 2). An intraoperative EGD was performed to ensure adequate lumen patency given the location at the GEJ and to check mucosal integrity by direct visualization. Insufflation was negative for a mucosal leak, and there were no surgical complications. The patient was discharged home the following day on a liquid diet and advanced as tolerated. Her post-operative course was unremarkable, as assessed by telephone consultation. She is currently doing well without symptoms and is tolerating a regular diet, and long-term surveillance is not currently indicated.

3. Clinical discussion

Esophageal leiomyomas are the most common benign esophageal tumors [1]. About half of known leiomyomas are symptomatic, and the leiomyoma in the patient presented was likely causing her upper abdominal pain [11]. The patient's presentation is relatively typical in that most esophageal leiomyomas present in the distal third of the esophagus [1] and detection peaks between ages 30 and 50 [5]. Esophageal leiomyomas are more common in men by about a factor of two [1]. This patient's leiomyoma was larger than most, as esophageal leiomyomas larger than 5 cm are relatively rare [7]. As a result, this patient was recommended for surgery under the current guidelines that recommend surgical management for esophageal leiomyomas that are larger than 5 cm or that are symptomatic [2,7]. This patient's esophageal leiomyoma was located at the GEJ, which can be a difficult area to enucleate large leiomyomas due to potential stricture at the GEJ [2]. The patient was consented for possible minimally invasive esophagectomy (MIE) if the GEJ were to become compromised.

This case report adds to the emerging evidence that VATS can be utilized for enucleation of larger leiomyomas (>5 cm in largest dimension), and particularly that simultaneous endoscopic visualization can be useful in challenging locations such as the GEJ. The use of VATS for enucleation of esophageal leiomyoma was first described by Everitt in 1992 [9]. However, VATS did not become the preferred approach over open thoracotomy for treatment of esophageal leiomyomas until 2011 [8]. In the years since, minimally invasive surgical management of leiomyomas has continued to rise in popularity, and has even been shown to be efficacious in the treatment of giant esophageal leiomyomas (defined as leiomyomas >10 cm in largest diameter) [12,13]. In fact, recent studies have suggested that the size of the leiomyoma is less important as a determinant of which surgical approach should be used, and that surgeon experience may be a more important factor [12]. Location at the GEJ adds additional difficulty and complexity to the operation, especially for a larger leiomyoma. However, the use of direct endoscopic evaluation via esophagoscopy during this patient's procedure while the mass was enucleated provides further visual and anatomic

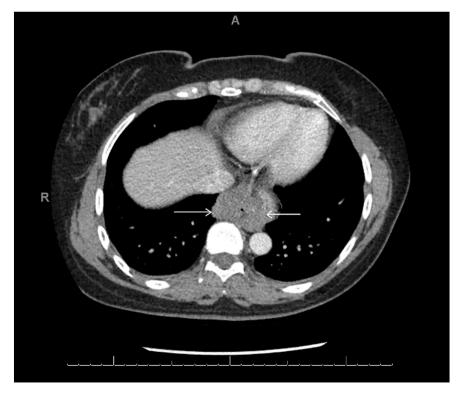


Fig. 1. CT Scan. 6.0 × 4.0 × 3.0 cm gastroesophageal junction mass seen in the submucosa and muscularis propria, displacing the esophagus to the left.

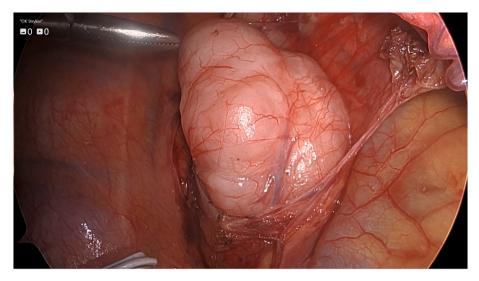


Fig. 2. Intraoperative image. Image from thoracoscopy shows $6.0 \times 4.0 \times 3.0$ cm esophageal mass.

reassurance that the underlying esophageal mucosa is not disrupted or narrowed. As such, this case suggests that larger esophageal submucosal masses (>5 cm) at the GEJ can potentially be enucleated with a combined VATS and endoscopic approach.

When comparing VATS with open surgery, some previous studies have shown that patients undergoing VATS had significantly shorter operative time, less blood loss, faster time to oral intake, and shorter length of postoperative stay [14,15]. Additional studies have shown that VATS is associated with decreased intraoperative and post-operative complication rates in comparison to open thoracotomy [8]. Notably, the vast majority of the patients in these prior studies comparing the two surgical approaches had tumors that were smaller than 5 cm [8,14]. In line with these findings, the patient presented in this report had an unremarkable surgical and post-operative course. She was able to be discharged on post-operative day 1 tolerating a clear liquid diet without issue.

4. Conclusion

Here we present a patient with a 6.0 cm GEJ leiomyoma that was enucleated operatively via a combined endoscopic and right VATS approach. The purpose of this report is to add to the limited literature on minimally invasive surgical treatment of a relatively large GEJ leiomyoma. This case highlights that VATS combined with endoscopic visualization is an efficacious and safe option for treatment of larger leiomyomas (>5 cm) of the GEJ and can be associated with minimal risk.

Provenance and peer review

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Ethical approval

This study is exempt from ethical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the

written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Rachel K. Wile: data curation, writing – original draft and editing; Katherine E. Barnes: data curation, writing – original draft and editing; Kian C. Banks: conceptualization, writing – review and editing, supervision; Jeffrey B. Velotta: conceptualization, writing – review and editing, supervision.

Research registration

None.

Guarantor

Dr. Jeffrey B. Velotta.

Declaration of competing interest

None.

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