

CASE REPORT

PEER REVIEWED | OPEN ACCESS

Esophageal leiomyoma: Laparotomic enucleation in a specialty center in the Amazon

Maria Letícia da Silva Martins, Higino Felipe Figueiredo,
Daniel Lourenço Lira, Rubem Alves da Silva Neto,
Thaís Caroline Sales Raposo, Larissa Pessoa de Oliveira

ABSTRACT

Introduction: Leiomyoma is considered a rare esophageal tumor, it is more common in the middle and distal esophagus and affects women from 20 to 69 years old. The objective of this article is to report the case of a young patient with digestive symptoms secondary to esophageal Leiomyoma that progressed to enucleation via laparotomy.

Case Report: Young woman, 24 years old, experiencing symptoms of retrosternal pain and dry cough for two years, associated with progressive dysphagia from solids to liquids and weight loss. Upper digestive endoscopy showed a heterogeneous lesion in the distal esophagus and gastrointestinal seriography suggested extramucosal esophageal tumor. Computed tomography (CT) of the chest showed a lobulated formation in the distal esophagus, measuring $5.5 \times 3.7 \times 3.6$ cm (L $\times$ T $\times$ AP), with a reduction in the luminal diameter in the distal esophagus. Conventional laparotomy treatment was chosen due to the size and location of the lesion at the esophagogastric confluence with tumor enucleation via the transesophageal abdominal route. The patient was

discharged on the 4th postoperative day, stable, without symptoms, eating a soft diet orally.

Conclusion: The diagnosis of esophageal leiomyoma, a rare disease with insidious evolution, was observed in a young patient, using an alternative access route to the established thoracotomy access. The appearance of symptoms is related to the size of the lesion, therefore, it should be investigated in patients with dysphagic symptoms with the aim of providing early diagnosis and less complex surgical treatment.

Keywords: Enucleation, Esophagus, Leiomyoma

How to cite this article

da Silva Martins ML, Figueiredo HF, Lira DL, da Silva Neto RA, Raposo TCS, de Oliveira LP. Esophageal leiomyoma: Laparotomic enucleation in a specialty center in the Amazon. Int J Case Rep Images 2024;15(2):5–9.

Article ID: 101460Z01MM2024

doi: 10.5348/101460Z01MM2024CR

Maria Letícia da Silva Martins¹, Higino Felipe Figueiredo², Daniel Lourenço Lira², Rubem Alves da Silva Neto², Thaís Caroline Sales Raposo¹, Larissa Pessoa de Oliveira³

Affiliations: ¹General Surgery Service at Samel Hospital, 1755, Joaquim Nabuco Av., Centro, Manaus, Amazonas, Brazil; ²Surgical Oncology/Abdominal Department at Samel Hospital, 1755, Joaquim Nabuco Av., Centro, Manaus, Amazonas, Brazil; ³General Practitioner of Faculty of Medicine of the Federal University (UFAM), 1053, Afonso Pena St, Praça 14 de Janeiro, Manaus, Amazonas, Brazil.

Corresponding Author: Maria Letícia da Silva Martins, 14, Stênio Neves St, Manaus, Amazonas, Brazil; Email: marialeticiaadm@gmail.com

Received: 03 May 2024
Accepted: 24 June 2024
Published: 31 July 2024

INTRODUCTION

Leiomyoma is considered a rare esophageal tumor, with an incidence of 0.4–1% of all tumors around the world; however, it is the most common benign tumor of this site, corresponding to 67–80% of cases. It is more common in the middle and distal esophagus, and uncommon in the proximal region. In most cases, it is a single lesion, ranging in size from 1 to 29 cm in diameter, generally smaller than 6 cm [1–3].

It has a higher prevalence among the age groups from 20 to 69 years old, with a higher incidence from the fifth

decade onward, and a predominance of males in the order of 2:1, in relation to females. About half the tumors are smaller than 5 cm, and asymptomatic most of the time, being incidental findings in periodic examination. Generally, when symptoms appear, the tumors are already larger than 5 cm in size. The most frequent symptoms are dysphagia, chest pain, vague retrosternal discomfort, heartburn, and occasionally regurgitation [4, 5].

Clinical investigation is carried out through esophagram or endoscopic ultrasound, and an anatomopathological study is required for the diagnosis. It reveals an appearance of bundles of smooth muscle cells with eosinophilic and fibrillar cytoplasm, which may present calcifications, requiring a complement by immunohistochemical study [6, 7].

It is presented a case of distal esophageal leiomyoma in a 24-year-old woman who complained of progressive dysphagia from solids to liquids, and retrosternal pain after meals associated with gastroesophageal reflux disease (GERD). During clinical investigation, upper digestive endoscopy showed a lesion in the distal esophagus and the pathological anatomical study, complemented by Immunohistochemistry, confirmed the diagnosis of leiomyoma. Due to the clinical conditions of the lesion and the patient, surgical treatment via laparotomy was chosen for enucleation of esophageal leiomyoma.

CASE REPORT

Female young adult, without comorbidities, with a two years history of progressive dysphagia from solids to liquids, tight retrosternal pain after ingestion of food, persistent dry cough and gastroesophageal reflux, begins outpatient follow-up to investigate gastrointestinal and respiratory symptoms within various specialties.

Attend the first medical appointment with a gastroenterologist and digestive system surgeon, with laboratory and radiological investigation. Return with laboratory tests showing no changes; colonoscopy without changes; upper digestive endoscopy showing a heterogeneous hypoechoic lesion in the distal esophagus, with probable origin in the muscular layer propria and erosive gastritis of the antrum; upper gastrointestinal seriography: oval filling defect, regular surface in the distal thoracic esophagus, left lateral, reducing its lumen and causing slight stasis upstream, suggesting extramucosal esophageal injury.

Computed tomography chest shows a lobulated formation with soft tissue density inside the distal esophagus, extending to the esophagogastric transition and measuring approximately $5.5 \times 3.7 \times 3.6$ cm (L $\times$ T $\times$ AP), reducing the luminal diameter at this level.

After a clinical-surgical meeting, surgical treatment was chosen, since endoscopic treatment was ruled out given the size of the tumor. Preliminary decision was made to access via laparotomy due to the location at the esophagogastric confluence; in the case of distal

esophagectomy, a gastric tube would be created in the second operative stage. So, there was no need for access via thoracotomy since tumor enucleation would be performed via the transesophageal abdominal route.

In the surgical approach via laparotomy, a large tumor lesion was revealed, constricting the esophageal lumen and cardia, lobulated, whitish, and avascular, with penetration and adherence to the mucous plane, firm consistency, determining stenosis >50% of the esophagogastric lumen, with no anatomical and vascular relationship with other surrounding structures (Figure 1).

The dissection plane was performed, with removal of esophageal pillars, allowing complete enucleation of the tumor. However, the dissection extended to the mucosal plane, requiring suturing in planes, at separate points in the mucosa and serosa, with nonabsorbable monofilament synthetic polypropylene suture.

An intraoperative test was performed with methylene blue for possible complications, such as fistulas in the esophageal tract, with a negative result for leaks. An intracavitary silicone drain was placed.

The anatomopathological result showed: Fusocellular neoplasia and immunohistochemistry concluded the diagnosis of leiomyoma with positivity for desmin and negativity for CD34 and KIT (CD117). Pathological anatomical exams collected from the esophagus, antrum, and gastric body showed moderate inflammatory changes, without other findings.

The patient was discharged on the 4th postoperative day without complications with a drain, clinically stable, without pain, dysphagia, regurgitant or emetic symptoms, eating orally with a soft diet and complaining of a dry cough without elimination of pathological secretions. She was instructed to return to the outpatient clinic on the 7th postoperative day. On the 10th postoperative day, she was admitted to the emergency department presenting a clinical postoperative complication, complaining of 1 fever spike (measured at 37.9°), mild/moderate dyspnea, chest pain, dry cough, and nasal congestion with hyaline discharge and history of recent contact with a person with the flu, with the pathological agent of SARS-COV-2 being considered as one of the hypotheses. A new hospital admission was carried out for clinical treatment and laboratory tests, ruling out COVID-19 infections and other surgical complications related to fistulous processes. The new hospitalization was concluded due to the viral process with clinical and laboratory improvement in three days and a new outpatient return.

Postoperative follow-up was given at 24 months, without evidence in the control, radiological and endoscopic examinations of stenosis, fistulas, or dehiscences.

Patient undergoing outpatient follow-up, with scattered complaints of gastrointestinal reflux under pharmacological control with good clinical control.

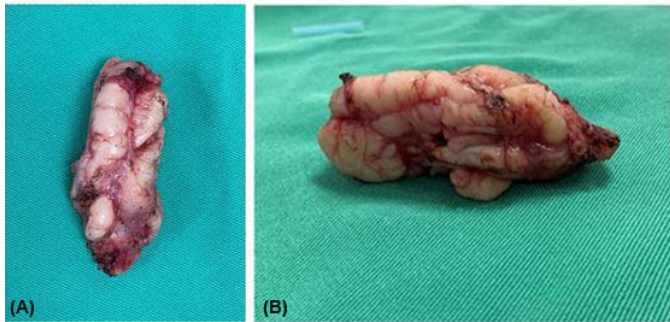


Figure 1: (A) and (B) Esophagus surgical piece as a result of laparotomic enucleation.

DISCUSSION

We present a case of esophageal leiomyoma with laparotomy enucleation with supraumbilical median access and transesophageal excision.

Esophageal leiomyomas are extremely rare tumors with insidious growth that arise from the mesenchymal tissue of the muscles and grow intramurally. Generally, this type of lesion does not exceed 5 cm, being more common in the middle and distal region of the esophagus and can affect patients of any age—being more common from the fifth decade of life onward in males in a 2:1 ratio. However, in the case presented, we note a young, female patient with significant symptoms without a history of comorbidities or family history [8–10].

Most patients are asymptomatic. When present, the condition is generally associated with a tumor larger than 5 cm and the most common symptoms are dysphagia, epigastric pain, and nausea due to extrinsic compression of neighboring organs [11, 12].

The diagnosis is confirmed by anatomopathological and immunohistochemical studies. The first diagnostic investigation can be done through esophagram or endoscopic ultrasound, which is used to define the limits of the tumor and shows five layers of the esophagus: superficial mucosa, muscularis mucosa, submucosa, muscularis propria, adventitia [13].

Complementary exams such as upper digestive endoscopy and computed tomography (CT) are important to describe the location and appearance of the lesion, in addition to evaluating adjacent structures and the presence of adhesions in neighboring organs. Endoscopic incisional biopsy is contraindicated in cases of leiomyoma since it is a submucosal tumor, therefore, it presents a high incidence of false-negative results associated with the risk of perforation and adherence of the tumor, making later enucleation difficult. Furthermore, incisional biopsy does not tend to change the surgical indication when present. Consequently, this procedure is reserved for ulcerated lesions or those with exposed muscle tissue [14]. In the case presented, there was no such clinical presentation when digestive endoscopy was performed, so the patient underwent surgical treatment.

The treatment of choice for this type of esophageal injury is surgery. In cases of questionable appearance with large and imprecise tumors associated with an exuberant clinical picture, surgical treatment is indicated. In asymptomatic patients with a benign-looking lesion after evaluation of complementary exams and without involvement of adjacent structures, clinical follow-up is chosen [15, 16].

In the case reported, the surgical indication was clear since the patient was experiencing progressive manifestation of symptoms with severe clinical evolution such as weight loss.

With the advancement of technology in the area of videolaparoscopy, lesion excision through thoracotomy was no longer performed due to high complication rates such as respiratory difficulty and anastomosis failures. Currently, the procedure can be performed using a minimally invasive method through thoracoscopy or laparoscopy access depending on the location of the tumor [6].

The choice of the operative method depends on some factors such as: size of the leiomyoma; location along the esophageal tract; the presence and extent of adhesion in the esophageal mucosa; mucosal injury after resection; suspicion of malignancy in the macroscopic aspect; the approach familiar to the surgeon and accordingly to the intraoperative infrastructure favorable to the execution of the procedure (arrangement of advanced energy material, appropriate drains, multidisciplinary team for continuity of health care) [17]. In the case reported, due to the size and location of the tumor, it was decided to complete enucleation of the lesion without evidence of fistulas or perforations in the postoperative period. The patient progressed well in the ward with outpatient follow-up thereafter.

CONCLUSION

Esophageal leiomyoma is a benign, rare tumor, with insidious growth and generally asymptomatic. Symptoms appear as the lesion increases.

In the reported case, a young patient is noted, with gradual evolution of symptoms and impaired quality of life, highlighting the rarity of the case. This type of lesion is treated by surgery, which depends on the size and location of the leiomyoma.

With the advancement of technology in the area of videolaparoscopy surgery, the resection of these tumors using this method provides satisfactory results. In the case in question, regarding the most comfortable access route to resolve the problem, abdominal access was chosen, highlighting the ease of access to the lesion due to its topographical characteristics and the absence of previous surgeries via this same route.

The importance of a detailed assessment of each patient and the correct indication of complementary exams are highlighted with the aim of establishing the

diagnosis early and increasing the possibilities of surgical resection.

REFERENCES

1. Milito P, Asti E, Aiolfi A, Zanghi S, Siboni S, Bonavina L. Clinical outcomes of minimally invasive enucleation of leiomyoma of the esophagus and esophagogastric junction. *J Gastrointest Surg* 2020;24(3):499–504.
2. Bardini R, Segalin A, Ruol A, Pavanello M, Peracchia A. Videothoroscopic enucleation of esophageal leiomyoma. *Ann Thorac Surg* 1992;54(3):576–7.
3. Hu X, Lee H. Complete thoracoscopic enucleation of giant leiomyoma of the esophagus: A case report and review of the literature. *J Cardiothorac Surg* 2014;9:34.
4. Maggi G, Dei Poli M, Gaetini A, et al. Our experience in 6 cases of leiomyoma of the esophagus. [Article in Italian]. *Minerva Chir* 1981;36(15–16):1035–42.
5. Beji H, Bouassida M, Kallel Y, Tormane MA, Mighri MM, Touinsi H. Leiomyoma of the esophagus: A case report and review of the literature. *Int J Surg Case Rep* 2022;94:107078.
6. Furtado WS, Mello DACPG, Santos VM, Oliveira Júnior WP, Schroff WLA. Leiomioma de esôfago tratado por via endoscópica em mulher de 41 anos etários. *Brasília Med* 2012;49(4):284–8.
7. Priego P, Lobo E, Alonso N, Gil Olarte MA, Pérez de Oteyza J, Fresneda V. Surgical treatment of esophageal leiomyoma: An analysis of our experience. *Rev Esp Enferm Dig* 2006;98(5):350–8.
8. Pereira F de B, Leão B da C, Leli LF, Lima M de P. Leiomioma de esôfago com características epidemiológicas anormais: Relato de caso. *Rev Med* 2018;97(3):357–60.
9. Inderhees S, Tank J, Stein HJ, Dubecz A. Leiomyoma of the esophagus: A further indication for robotic surgery? [Article in German]. *Chirurg* 2019;90(2):125–30.
10. Lorencetti RR da G, Goes JFV, Mattion ÉA, Módena SF, Paula RA de. Excision of esophageal leiomyoma through thoracotomy. *Rev Col Bras Cir* 2006;33(1):55–6.
11. Samphire J, Nafteux P, Luketich J. Minimally invasive techniques for resection of benign esophageal tumors. *Semin Thorac Cardiovasc Surg* 2003;15(1):35–43.
12. Singla V, Gupta E, Bhattacharjee H, Joshi M, Sharma R, Parshad R. Thoracoscopic enucleation of a large esophageal leiomyoma in the lower esophagus: Challenges and solutions. *Indian J Thorac Cardiovasc Surg* 2021;37(6):694–7.
13. Ray S, Saluja SS, Gupta R, Chattopadhyay TK. Esophageal leiomyomatosis – An unusual cause of pseudoachalasia. *Can J Gastroenterol* 2008;22(2):187–9.
14. Luh SP, Hou SM, Fang CC, Chen CY. Video-thoracoscopic enucleation of esophageal leiomyoma. *World J Surg Oncol* 2012;10:52.
15. Cheng BC, Chang S, Mao ZF, et al. Surgical treatment of giant esophageal leiomyoma. *World J Gastroenterol* 2005;11(27):4258–60.

16. Pinheiro FAS, Campos AB, Matos JRF, Araripe DPA. Tratamento videoendoscópico do leiomioma de esôfago. *Arq Bras Cir Dig* 2013;26(3):234–7.
17. Kent M, d'Amato T, Nordman C, et al. Minimally invasive resection of benign esophageal tumors. *J Thorac Cardiovasc Surg* 2007;134(1):176–81.

Author Contributions

Maria Letícia da Silva Martins – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Higino Felipe Figueiredo – Conception of the work, Design of the work, Acquisition of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Daniel Lourenço Lira – Conception of the work, Design of the work, Acquisition of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Rubem Alves da Silva Neto – Conception of the work, Design of the work, Acquisition of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Thaís Caroline Sales Raposo – Conception of the work, Design of the work, Acquisition of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Larissa Pessoa de Oliveira – Conception of the work, Design of the work, Acquisition of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

Source of Support

None.

Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

Copyright

© 2024 Maria Letícia da Silva Martins et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

Access full text article on
other devices



Access PDF of article on
other devices



