

Giant esophageal lipoma: complicated surgical treatment (case report)

Gigantinės stemplės lipomos komplikuotas chirurginis gydymas: klinikinis atvejis

Justina Katinaitė¹, Žymantas Jagelavičius², Algis Kybartas³, Vygantas Gruslys²,
Ričardas Janilionis²

¹ *Vilniaus universiteto Medicinos fakultetas, M. K. Čiurlionio g. 21, LT-03101 Vilnius, Lietuva*

² *Vilniaus universiteto Medicinos fakulteto Infekcinių, krūtinės ligų, dermatovenerologijos ir alergologijos klinika, Krūtinės chirurgijos centras, Santariškių g. 2, Vilnius, Lietuva*

³ *Vilniaus universiteto ligoninės Santariškių klinikos, Krūtinės chirurgijos skyrius, Santariškių g. 2, Vilnius, Lietuva*
El. paštas: zymant@yahoo.com

¹ *Vilnius University Faculty of Medicine, M. K. Čiurlionio Str. 21, LT-03101 Vilnius, Lithuania*

² *Centre of Thoracic Surgery, Clinic of Infectious and Chest Diseases, Dermatovenerology and Allergology, Faculty of Medicine, Vilnius University, Santariškių Str. 2, Vilnius, Lithuania*

³ *Department of General Thoracic Surgery, Vilnius University Hospital Santariskiu Klinikos, Santariškių Str. 2, Vilnius, Lithuania*
E-mail: zymant@yahoo.com

Background

Lipomas are distinctly uncommon benign tumours of the esophagus. Lipoma and other benign tumours should be considered for patients admitted to the hospital with non-specific complaints of upper gastrointestinal tract disease. Radiological and endoscopic suspicion of a tumour with benign histology and correct preoperative histological diagnosis are important factors in order to choose the best surgical treatment option.

Case report

A 59-year-old woman was admitted with dysphagia lasting for about 1.5 months. Endoscopy and computed tomography scan revealed intraluminal pedunculated submucosal mass, 13 cm in length and 2 cm in diameter, –67 Hounsfield units in density, suggesting esophageal lipoma to be the most likely diagnosis. Tumour was excised through right lateral thoracotomy with esophagotomy. Barium esophagogram in the postoperative period showed esophageal leakage, hence rethoracotomy was performed and the defect was attempted to close by sutures. Repeated barium esophagogram revealed persisting evidences of esophageal leakage so biodegradable coated 10 cm in length and 31/25/31 mm in diameter SX-ELLA stent was used and successfully sealed the leakage.

Conclusion

Giant esophageal lipoma is a rare benign esophageal tumour causing dysphagia. Open surgery is the best treatment option for giant esophageal lipomas. However, postoperative complications such as esophageal leakage are common in patients after open esophageal surgery. Biodegradable stent could be successfully used dealing with persistent esophageal leakage.

Key words: dysphagia, esophageal lipoma, esophagotomy, mediastinitis, esophagopleural fistula.

Ižanga

Lipoma – retas nepiktybinis stemplės navikas. Tiriant pacientus dėl nespecifinių viršutinio virškinamojo trakto ligos simptomų, reikėtų nepamiršti nepiktybinių stemplės navikų. Vaizdiniais tyrimais įtartas nepiktybinis stemplės navikas ir tiksliai nustatyta histologinė diagnozė priešoperaciniu laikotarpiu yra svarbūs veiksniai pasirenkant tinkamiausią gydymo metodą bei siekiant išvengti sudėtingesnių chirurginių procedūrų.

Klinikinis atvejis

Moteris, 59 metų amžiaus, buvo paguldyta į chirurgijos skyrių dėl disfagijos, truncančios apie 1,5 mėnesio. Endoskopiškai įtartas stemplės pogleivio navikas. Atlikus krūtinės ląstos kompiuterinės tomografijos tyrimą rastas 13 cm ilgio ir 2 cm pločio, –67 Hounsfieldo vienetų tankio intraluminalinis pogleivio darinys ant kojytės. Radiologiškai labiausiai tikėtina diagnozė buvo stemplės lipoma. Navikas pašalintas atlikus ezofagotomiją per dešinės pusės torakotominį pjūvį. Pooperaciniu laikotarpiu atlikta kontrastinė ezofagograma parodė stemplės nesandarumo požymius. Buvo atlikta retorakotomija ir stemplės defektas užsiūtas. Kartotinis radiologinis stemplės tyrimas parodė, kad stemplės nesandarumas išliko. Tuomet į stemplę buvo implantuotas biodegraduojantis SX-ELLA dengtas 10 cm ilgio, 31/25/31 mm išorinio skersmens stentas, kuriuo defektas stemplėje sėkmingai užgydytas.

Išvados

Stemplės lipoma yra retas nepiktybinis stemplės navikas, sukeliantis disfagiją. Atvira stemplės operacija yra geriausias gydymo būdas esant dideliems nepiktybiniams stemplės navikams. Tačiau tokios pooperacinės komplikacijos kaip stemplės nesandarumas nėra retos pacientams po atvirų stemplės operacijų, o jų gydymas sudėtingas. Biodegraduojantis stentas galėtų būti tinkamas įsisenėjusiam stemplės nesandarumui gydyti.

Reikšminiai žodžiai: disfagija, stemplės lipoma, ezofagotomija, mediastinitas, ezofagopleurinė jungtis

Introduction

Lipomas of the alimentary tract are uncommon benign tumours rarely reported in the literature. Hence, diagnosis and treatment often poses a challenge for surgeons. Overall incidence of alimentary tract lipomas is 4.1%, but that of the esophagus is extremely rare with an incidence of only 0.4% [1]. Most lipomas are small, do not cause symptoms and may be found incidentally during imaging studies [2]. Rarely, lipomas become large and cause symptoms such as dysphagia, odynophagia when surgical excision is required [3].

We present a case of a large pedunculated esophageal lipoma treated by surgery in a patient with non-specific complaints of upper gastrointestinal (GI) tract disease lasting for about a year prior to initial diagnosis. Successful management of postoperative complications which occurred after excision of the tumour is described, emphasizing the need to treat such patients in a medical centers capable of providing highly qualified medical care and best treatment facilities.

Case presentation

A 59-year-old woman was admitted to our hospital complaining of progressive dysphagia that existed for about 1.5 months. Other complaints included heartburn, regurgitation and nausea. Patient also recalled

having intermittent pain attacks in epigastric region first appeared a year ago and nausea leading to repeated vomiting with small amount of gastric contents and foreign body sensation in the esophagus. She denied weight loss or dispnea. There was no other significant medical, family or social history. On physical examination no abnormalities were found and laboratory tests were unremarkable. Upper GI endoscopy revealed a submucosal mass with normal mucosa arising from esophageal wall on the right located 18 cm away from the incisors and with its body extending downward to the lower esophagus, occupying half of the lumen. There was also a sliding hiatal hernia. The biopsy at this time showed chronic active esophageal ulcer.

Taking into consideration patient's symptoms and endoscopic suspicion of submucosal tumour, computed tomography (CT) of the chest was performed and revealed an intraluminal pedunculated submucosal mass, 13 cm in length and 2 cm in diameter, elongated from the upper to the middle third esophagus with its pedicle in upper esophagus. The mass had a density of –67 Hounsfield units (HU) and showed no apparent invasion into surrounding tissues, suggesting esophageal lipoma to be the most likely diagnosis (Figure 1). Due to large size of the lipoma with its body located in the thoracic esophagus and the uncertainty about the length of adherence with which the tumour pedicle attached to

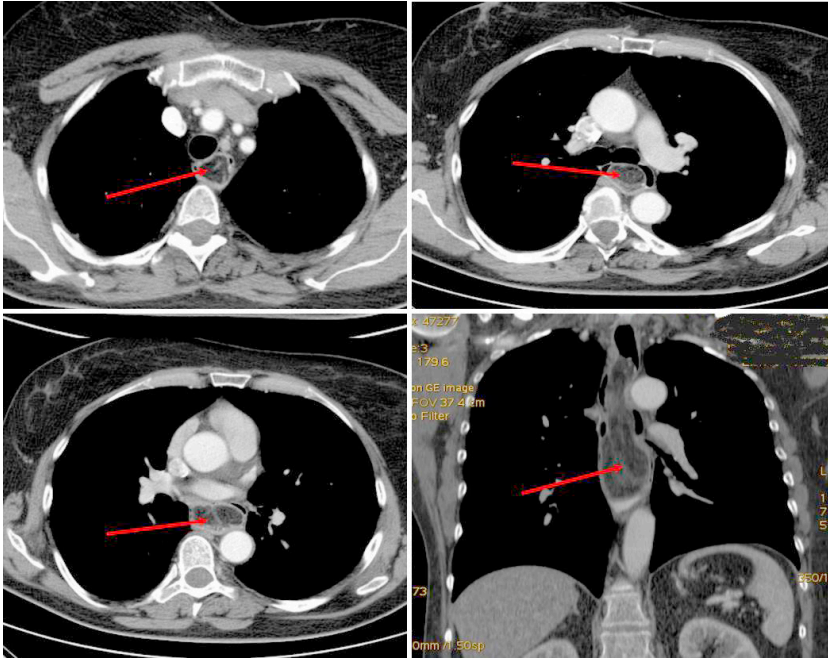


Figure 1. Computed tomography scan showing giant mass in the posterior mediastinum (arrows)

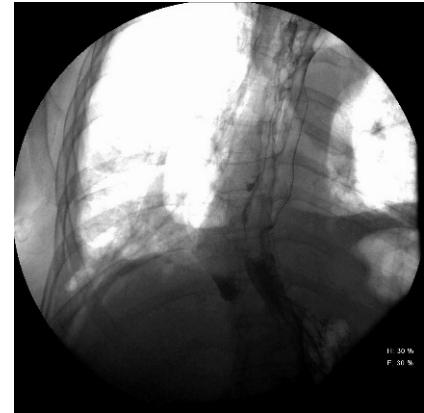


Figure 2. Esophageal leak on the 6th postoperative day

the esophageal wall, decision was made to excise the tumour through right – sided transthoracic approach with esophagotomy. Intraoperative gross examination of the specimen showed elastic intraluminal mass, measured 12 × 7 cm, resembling lipoma with its pedicle extending upwards to thoracic outlet so it can not be reached and radically excised. The tumour was excised leaving a part of the pedicle. Esophagotomy was closed in two layers and the closure assessed intraoperatively. Three chest tubes one of which mediastinal were inserted before wound closure. The final biopsy results of the excised mass confirmed the diagnosis of lipoma as the mass was composed of mature adipose tissue with ulcerations in mucosa and neither increased mitotic activity nor lipoblasts was found.

During the postoperative period patient received metronidazole with gentamicin, enteral nutrition was administered through the nasoduodenal tube. Despite antimicrobial therapy patient had fever as high as 38.3 °C, elevated C-reactive protein (CRP) – 144.6 mg/L, together with fluid in both pleural cavities seen on chest X-ray. Regarding the clinical picture, barium swallow X-ray study of the esophagus was perfor-

med and showed right sided esophageal leak (Figure 2). Considering radiological signs of esophageal leakage and clinical manifestation of mediastinitis, it was decided to perform rethoracotomy. Esophageal wall defect was found 5 cm above diaphragm and was 2 cm in diameter. The defect was closed by sutures, periesophageal mediastinum and pleural cavity were drained by chest tubes. Patient received ceftriaxone with metronidazole postoperatively. Microbiologic study of patient's pleural fluid specimen was positive for methicillin-resistant coagulase negative staphylococci (MRCNS). A ten day course of meropenem was added to the therapy with continuous administration of metronidazole. Patient also developed moderate anaemia after surgery: red blood cell count (RBC) was $2.71 \times 10^{12}/l$, hemoglobin (Hb) – 82 g/l and hematocrit (Hct) – 0.237 respectively. On the tenth day after rethoracotomy repeated barium swallow X-ray of the esophagus revealed persisting evidences of esophageal leakage (Figure 3). Chest X-ray showed remaining pleural effusions in both pleural cavities with positive dynamics. Patient was in a stable condition with no clinical signs of infection, so decision was made to wait for the defect to close itself.

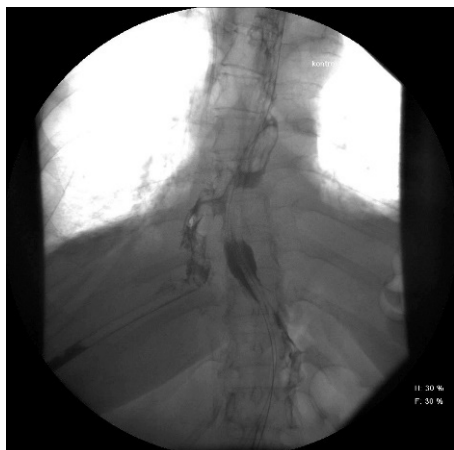


Figure 3. Esophageal leak on the 10th day after rethoracotomy



Figure 4. Persistent esophageal leak (before stent placement)

Repeated barium swallow X-ray test of the esophagus performed a month after rethoracotomy clearly defined persistent esophageal wall defect in the lower esophagus with leakage of the contrast (Figure 4). Decision was made to seal the leakage with a biodegradable coated stent. Esophageal endoscopy showed roughly deformed lumen of the esophagus and esophagopleural fistula, 6 mm in diameter located 30 cm away from the incisors in the right wall of the esophagus. A 10 cm in length and 31/25/31 mm in diameter biodegradable self-expanding SX-ELLA coated stent was placed to seal the leakage, leaving its upper edge 28 cm away from the incisors

and lower edge above cardioesophageal junction. On the barium swallow follow up after stent placement there was no evidence of leakage (Figure 5).

The patient was discharged from the hospital with complete resolution of symptoms. Further patient's follow up 1.5, 6 and 8 months after discharge showed no stent migration, full stent biodegradation after its implantation. At the last follow up 2 years after discharge no evidence of esophageal leakage nor disease relapse was found (Figure 6). A patient has no complaints, can consume liquid as well as solid food without any signs of dysphagia.

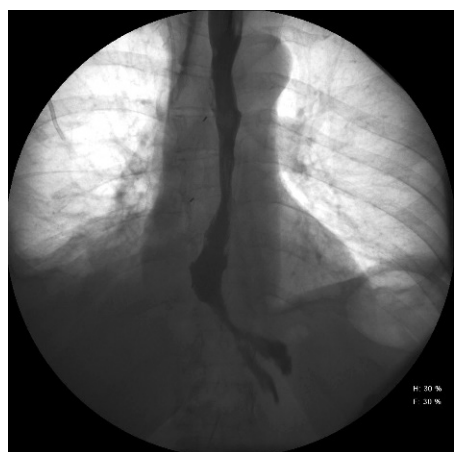


Figure 5. Esophagogram after biodegradable stent placement



Figure 6. Esophagogram 2 years after discharge

Discussion

Lipomas have been found in all segments of the digestive tract: lesions in the colon occur most frequently, followed, in decreasing order, by lesions in the small intestine, stomach, and esophagus [1, 4, 5]. Esophageal lipomas have been reported in patients between the ages of 4 and 80 years old, with a mean age of 50 [2, 6]. Usually, esophageal lipomas originate in the cervical and upper thoracic esophagus [2]. The exact etiology of lipomas is uncertain, however, one theory suggests an association between previous trauma and subsequent lipoma formation [7]. Although lipomas are rare in the esophagus, early diagnosis and resection should be recommended for all symptomatic cases [2]. Establishing a correct diagnosis of an esophageal lipoma demands a careful history, thorough radiographic examination and/or inspection with upper GI endoscopy [3]. There may be symptoms of odynophagia, recurrent melaena [8] and mechanical compression of the upper respiratory tract [9] most often due to displacement of the trachea. Esophageal lipomas generally become symptomatic only when they are large enough to cause dysphagia, when they merit surgical excision [2]. Our patient had dysphagia, intermittent pain in epigastric region, nausea and other symptoms suggesting the possibility of a space occupying process located in esophagus. On CT, a homogeneous mass with HU between -80 and -120 is nearly pathognomonic for a lipoma [10]. On magnetic resonance imaging (MRI), lipomas can be identified by following fat signal as T1-weighted hyperintensity that becomes hypointense on fat-suppressed images [3]. In our case CT scan showed mass with -67 HU.

As lipoma is a rare benign tumour of the esophagus, differential diagnosis from more common benign and malignant lesions is mandatory. Leiomyomas, the most common benign esophageal tumours, has subtle differences from that of a lipoma. Radiologically, they appear as a well-defined mass with muscle density and occasionally contain nodular calcifications. This translates to a hypoattenuating lesion on CT, a hypoechoic lesion with fine echoes in the setting of calcifications on endoscopic ultrasound and a tan mass on upper GI endoscopy

[11]. Presentation of a lipoma on upper GI endoscopy is a lesion of yellow color, pliability and a smooth surface while malignant lesions tend to be friable and irregular, with an ulcerated surface [2]. Other useful signs to differentiate lipomas from other benign or malignant lesions include a “squeeze” sign – mass changes in contour and configuration as a result of peristalsis [12].

Management of esophageal lipomas is dependent on size and location of the lesion. The options include monitoring if diagnosis is secure and symptoms are not experienced, or removal endoscopically or through either a cervical or thoracic approach [13]. Surgical excision is recommended for symptomatic benign tumours and those greater than 5 cm [3]. Although the pulmonary complications associated with open surgery can be avoided by an endoscopic approach, open surgical approach provides operators with a better visual operating field, enables better control for managing large tumours and prevent bleeding caused by endoscopic transection of the vessel-enriched pedicle [14]. In our case, the tumour was large, exceeding 13 cm, the length and diameter of the pedicle were unclear before surgery. Therefore, endoscopic resection did not seem appropriate and open surgery was chosen for tumour resection. Taking into account that during the surgery esophageal lumen was opened due to visualize the tumour, attention should be drawn to the possible postoperative complications. Primary repair of the esophageal wall carries high incidence of leakage and subsequent mediastinitis [15]. Proper management of leakage and mediastinitis becomes the priority in postoperative period.

Conclusion

Lipoma is a rare benign tumour found in esophagus. Early diagnosis should be a priority for the physicians when a patient with dysphagia is admitted. CT and histological examination are the main tests to be used in establishing accurate diagnosis and differentiating lipomas from other benign or malignant lesions. Management of esophageal lipomas depends on location and size of the lesion. For large tumours open surgery is the treatment of choice. Biodegradable stent can be useful in managing persistent esophageal leaks.

REFERENCES

1. Mayo CW, Pagtalunan RJ, Brown DJ. Lipoma of the alimentary tract. *Surgery*. 1963; 53: 598–603.
2. Wang C-Y, Hsu H-S, Wu Y-C, Huang M-H, Hsu W-H. Intramural Lipoma of the Esophagus. *J Chin Med Assoc*. 2005; 68: 240–3.
3. Qinying W, Wei L, Shuihong Z. Large pedunculated lipoma of the esophagus: Report of a case and review of literature. *J Cancer Res Ther*. 2015; 11: 1031.
4. Plachta A. Benign tumors of the esophagus. Review of literature and report of 99 cases. *Am J Gastroenterol*. 1962; 38: 639–52.
5. Feldman M. An appraisal of associated conditions occurring in autopsied cases of lipoma of the gastrointestinal tract. *Am J Gastroenterol*. 1961; 36: 413–6.
6. Kang JY, Chan-Wilde C, Wee A, Chew R, Ti TK. Role of computed tomography and endoscopy in the management of alimentary tract lipomas. *Gut*. 1990; 31: 550–3.
7. Lipomas: Background, Pathophysiology, Etiology. 2016 Sep 2 [cited 2016 Nov 1]; Available from: <http://emedicine.medscape.com/article/191233-overview>
8. Zschiedrich M, Neuhaus P. Pedunculated giant lipoma of the esophagus. *Am J Gastroenterol*. 1990; 85: 1614–6.
9. Hasan N, Mandhan P. Respiratory obstruction caused by lipoma of the esophagus. *J Pediatr Surg*. 1994; 29: 1565–6.
10. Taylor AJ, Stewart ET, Dodds WJ. Gastrointestinal lipomas: a radiologic and pathologic review. *Am J Roentgenol*. 1990; 155: 1205–10.
11. Murata Y, Yoshida M, Akimoto S, Ide H, Suzuki S, Hanyu F. Evaluation of endoscopic ultrasonography for the diagnosis of submucosal tumors of the esophagus. *Surg Endosc*. 1988; 2: 51–8.
12. Hurwitz MM, Redleaf PD, Williams HJ, Edwards JE. Lipomas of the gastrointestinal tract. An analysis of seventy-two tumors. *Am J Roentgenol Radium Ther Nucl Med*. 1967; 99: 84–9.
13. Razzak R, Bédard ELR, Hunt I, Satkunam N. Spindle cell lipoma of the esophagus. *Eur J Cardiothorac Surg*. 2009; 35: 542–3.
14. Liu C-H, Chang H-C, Goan Y-G. Large pedunculated lipoma of the esophagus. *J Formos Med Assoc*. 2008; 107: 424–7.
15. Algin C, Hacıoglu A, Aydin T, Ihtiyar E. Esophagectomy in esophageal lipoma: report of a case. *Turk J Gastroenterol*. 2006; 17: 110–2.