CASE REPORT

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Esophageal leiomyoma and simultaneous overlying squamous cell carcinoma: a case report and review of the literature

Saadat Mehrabi¹, Mohammad Javad Yavari Barhaghtalab^{1*}, Safoora Hejazinia² and Hossein Saedi³

Abstract

Background: Squamous cell carcinoma is the most common epithelial tumor of the esophagus. Upper endoscopy with multiple minimally invasive biopsies should be performed to confirm the diagnosis. Leiomyoma of esophagus is rare, but it's the most common benign submucosal mesenchymal tumor of the esophagus. The simultaneous occurrence of an overlying epithelial lesion and a mesenchymal lesion is very rare. This study aims to show a case operated due to squamous cell carcinoma of esophagus that was postoperatively diagnosed with coexistent esophageal leiomyoma and give a clear overview of the existing literature on it.

Case presentation: The patient was a 41-year-old woman who underwent three field esophagectomy (McKeown). Pathological evaluation was done, and the patient had poorly differentiated squamous cell carcinoma and multiple leiomyomas. A leiomyoma was found with an invading overlying squamous cell carcinoma.

Conclusion: It is concluded that esophageal carcinomas may coexist with leiomyomas; preexisting benign tumors may have played an important role in the development of the carcinoma by inducing constant stimulation of the overlying mucosa; endoscopic ultrasonography is recommended to avoid overestimating the extent of tumor invasion and the resultant aggressive radical surgery. As the developing countries had limited equipment, esophageal resection could be the modality of choice in the treatment.

Keywords: Esophageal squamous cell carcinoma, Leiomyoma of esophagus, Simultaneous occurrence, Overlying, Esophageal resection, Case report

Background

Although esophageal leiomyoma is rare, it's the most common benign submucosal mesenchymal tumor (SMT) of the esophagus, originates from the cells of the smooth muscle, and form near the two-thirds (60-70%) of all benign tumors of the esophageal [1-3]. It almost appears as a single tumor, and multiple leiomyomas of the esophagus are extremely rare [1, 4]. Since esophageal

leiomyoma is generally a slow-growing tumor and the size of the tumor may not change for many years, most affected patients are asymptomatic [2]. Often, a diagnosis of esophageal leiomyoma is made as an incidental finding during routine investigation or screening for upper gastrointestinal (GI) pathology [3]. Endoscopic ultrasonography (EUS) and computerized tomography (CT) are used for the diagnosis of leiomyoma. The diagnosis is difficult when multiple leiomyomas coexist with carcinoma lesions. In cases where a carcinoma overlies a submucosal leiomyoma, there is a possibility of overestimating the extent of tumor invasion, and multiple minute leiomyomas are sometimes misdiagnosed as intramural metastasis [2]. Although esophageal leiomyoma is



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conventionally treated by surgical removal via open thoracotomy for the tumors in the upper two-thirds of the esophagus, minimally invasive approaches like videoassisted thoracoscopic surgery (VATS) and endoscopic resection are other alternative methods used for enucleation of the tumor [3, 5].

The most common epithelial tumor of the esophagus is the squamous cell carcinoma (SCC) [2]. For the identification of an SCC, an upper GI endoscopy with multiple minimally invasive biopsies should be implemented. To assess the size and extent of the primary tumor and to check for the existence of liver metastases and celiac lymphadenopathy, a CT scan of the thorax and abdomen should be done. A precise preoperative staging leads to a proper selection of the treatment. There are some treatment recommendations proposed: (1) Superficial and limited mucosa disease (less than T1a), could undergo endoscopic resection, (2) Lesions penetrating the submucosa with negative lymph nodes (LNs) (more than T1b) could undergo direct surgical resection with lymphadenectomy, (3) Resectable lesions invading muscularis propria with positive LNs (less than T2N1) could receive neoadjuvant chemoradiation, (4) Locally advanced, unresectable or metastatic disease could receive palliative systemic therapy [6].

The simultaneous occurrence of an overlying epithelial lesion (SCC) and a mesenchymal lesion (leiomyoma) is very rare. This study aims to show a case operated due to SCC of esophagus that was postoperatively diagnosed with coexistent esophageal leiomyoma and give a clear overview of the existing literature on it.

Case presentation

The patient was a 41-year-old woman who was first seen at Shahid Mofateh Clinic affiliated to Yasuj University of medical sciences in 2019 with complaints of having progressive dysphagia to solid foods, and a ten-kilograms weight loss for the preceding 6 months (Weight = 50.5, BMI = 19.77); the patient also had anorexia, decreased intake, and general fatigue and lethargy as accompanying symptoms, but the patient had no nausea and vomiting. Physical examination findings were typically normal. There was no neck (laterocervical or supraclavicular) lymphadenopathy and no hepatomegaly. In past medical and drug history, the patient had asthma and used fexofenadine and salbutamol spray. There was no family history of malignancy in her parents and siblings. The patient did not use tobacco, smoke cigarette and drink alcohol. Laboratory values were all within normal limits.

The patient underwent upper GI endoscopy. In esophagus, the upper esophageal sphincter, cricopharyngeus and upper third of esophagus were normal. A large fungating and ulcerative mass was found in the middle and the lower third of esophagus, 25–33 cm from the upper incisors. The Z line was normal (Fig. 1). In the stomach, the cardia (retro-vision maneuver), fundus, body and antrum were all normal. In the duodenum, the bulb and 2nd part were normal. The patient underwent an inci-

squamous cell carcinoma in pathology evaluation. In spiral chest CT scan with and without intravenous (IV) contrast, obtained axial images, represented a 29×18 mm soft tissue fullness at middle to distal esophagus (at the level of the carina to the level of the main pulmonary artery), which was suggestive for a tumoral lesion. No lung metastasis was found. In spiral abdomen and pelvic CT scan with oral and IV contrast, there was a 12×9 mm LN within gastrohepatic ligament (Fig. 2). In the abdomen and pelvic sonography, there was a 10×9 mm hypoechoic lesion between the left liver lobe and greater curvature which was suggestive of lymphadenopathy (LAP) (Fig. 3.).

sional biopsy and was found to have poorly differentiated

Afterward a multidisciplinary team decision-making meeting (weekly general surgery grand round at the department of surgery, Shahid Beheshti Hospital, Yasuj University of Medical Sciences) was held, and it was decided that the patient would receive 3 sessions of chemotherapy without radiotherapy before undergoing the operation. Then, the patient underwent three field esophagectomy (McKeown) including laparotomy, right thoracotomy, and gastric pull-up with cervical anastomosis including extended thoracotomy, esophagectomy, gastric pull-up, pyloroplasty, cervical esophagostomy, jejunostomy, and chest tube insertion. An abdominal LN at the gastrohepatic ligament was also resected. The postoperative course was uncomplicated. There was no evidence of the leak at the anastomotic site by administration of methylene blue dye on the fifth postoperative day and then nasogastric tube was removed and the patient was told to start drinking liquids. The patient advanced to a regular diet by the tenth postoperative day. She was discharged from the hospital on the 14th day of admission. At a follow-up visit of 1 week and then 2 weeks after discharge from the hospital she was feeling well. The patient's condition after surgery came back to her normal habitual life within 3 months. The patient had no complaint at the 9th month follow-up visit after surgery. A spiral chest and abdomen CT scan was done and showed a pulled-up stomach, filled with fluid and air at the right paraspinal region (Fig. 4). In the last follow-up, the patient had no complaints and said that she ate a satisfying meal recently, and that she is optimist about the future.

The specimen was sent in formalin for pathology and immunohistochemistry evaluation. The received specimen was consisted of the esophagus and a segment

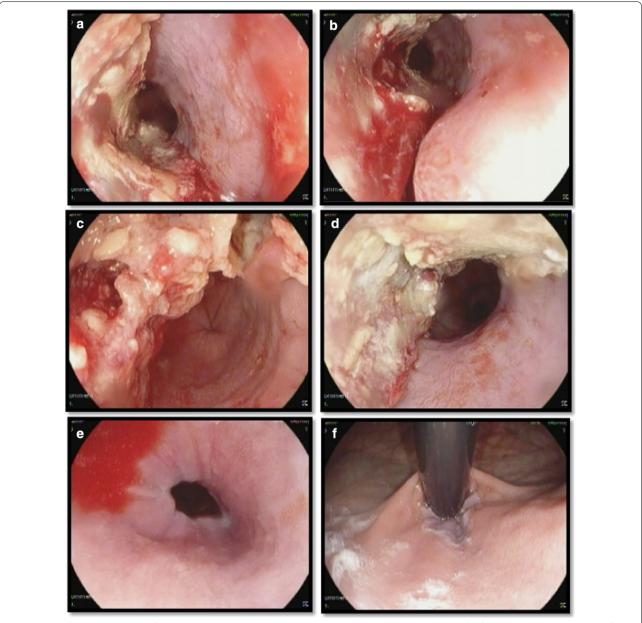
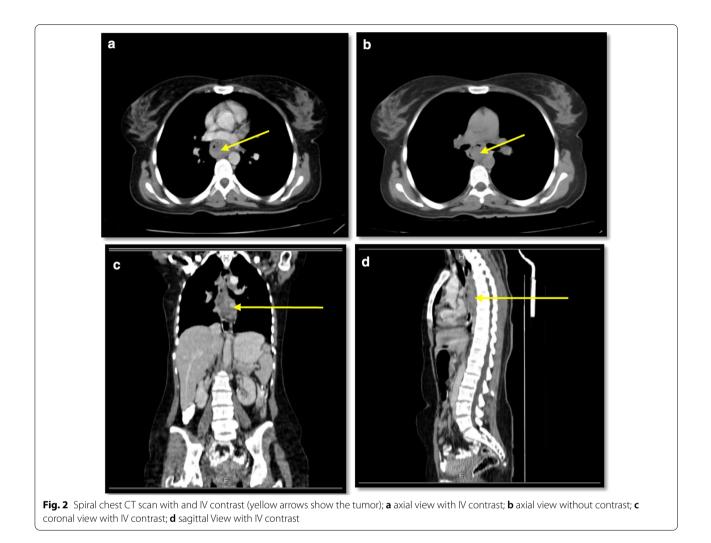
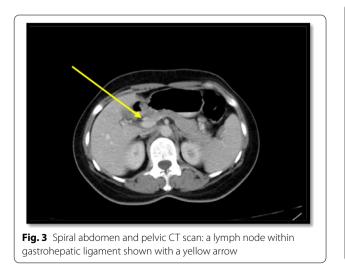


Fig. 1 Upper GI endoscopy: large fungating and ulcerative mass in middle (a and b) and lower third (c and d) of esophagus, Z-line (e), Cardia (f)

of the stomach. The resected esophagus was 12 cm in length and 3 cm in the greatest diameter. The adventitial surface was congested. On opening, there were four intramural creamy round firm masses measuring from 1 to 2 cm. No perforation site was found. Proximal and distal margins were not grossly involved. The resected segment of stomach was 8 cm in greater curvature and 6 cm in lesser curvature and showed no mass (Fig. 5). Multiple LNs were identified measuring from 0.5 to 1 cm in diameter. Pathological evaluation was done, and the patient had small groups of residual poorly differentiated squamous cell carcinoma and also multiple leiomyomas (four). A leiomyoma was coexisted with an invading overlying SCC (Fig. 6). There were 5 isolated LNs within the pathology specimen, of which two were involved with the tumor, but pathology did not show any sign of metastasis in the LNs resected at the gastrohepatic ligament. Tumor pathology characteristics are summarized and shown in Table 1.







region shown with a yellow arrow

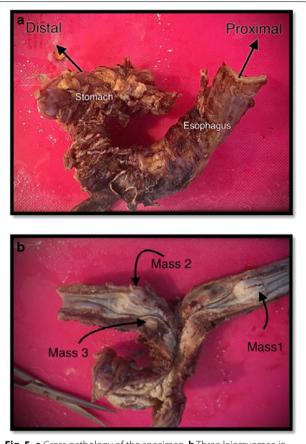


Fig. 5 a Gross pathology of the specimen, **b** Three leiomyomae in gross pathology of the specimen

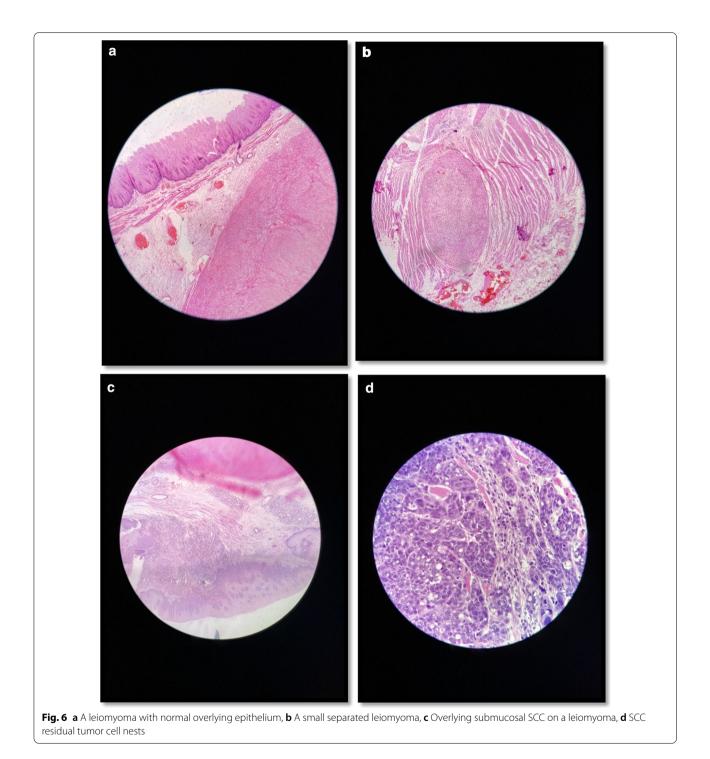
Discussion and conclusion

There are several reports of co-existing overlying esophageal SCC and leiomyoma in the literature. In this situation, the esophageal submucosal layer is overlaid by the superficial cancer. This coexistence is very rare, and this is usually detected following surgery. Callanan et al., reported the first case of its kind in 1954 [7]. In this regard, all previously published reports on cases with leiomyoma overlying with esophageal SCC were searched and found on the web and only available full-text original articles were encountered. Accordingly, fifteen cases in fourteen reports were found. These reports were evaluated and summarized in Table 2 according to the originated country and published year of case report, patient's gender, age, chief complaints, risk factors, comorbid diseases, upper GI endoscopy, EUS, other diagnostic modalities, pathological results, location of tumor, and treatments applied [1, 2, 4, 8–18].

Of the previous published reports, 10 reports were from Japan, 2 reports were from South Korea, and one report from Iran and Turkey each. It shows that this pathology is much more common in the East Asia, especially in Japan. There was only one female patient previously reported, so our case was the second one. This displays the male majority in this concern. Ishida et al. reported a male to female ratio of 4:1 in patients with SCC overlying leiomyoma [16]. Mean age was 59.62 ± 9.46 years (minimum 41 and maximum 75 years). Our case was the youngest patient in all, as the youngest patient was previously from Turkey with an age of 44 years [4].

In chief complaints, there were 5 cases with dysphagia or stenotic sensation in the pharyngeal region, 3 cases with significant weight loss, 3 cases with esophageal tumor which were found at the routine health checkups, and one patient with epigastric pain. From the reports in which risk factors were mentioned, heavy smoking was seen in three cases. Comorbid diseases were found negative in 4 cases, and malignant lymphoma of the thyroid and hyperglycemia and osteoporosis were found in one case each. Upper GI endoscopy showed an elevated or protruding lesion in 5 cases, a sessile polypoid lesion in 3 cases, and a submucosal tumor in 3 cases. Chromoendoscopy with iodine staining was done in 7 cases and showed a non-staining area in all. EUS was done in 8 cases and showed a hypoechoic homogeneous submucosal tumor with a well demarcated and clear margin in the muscularis mucosae (MM) covered with the intraepithelial layer indicative of leiomyoma in most of the cases. Barium esophagogram study was done in 4 cases. In pathological evaluation, squamous cell carcinoma in situ (high-grade intraepithelial neoplasia) was seen in 4 cases, poorly differentiated SCC in 2, moderately differentiated SCC in 2, and malignant SCC in one case. More than one leiomyoma was seen in 3 cases. According to the location of the lesions, proximal, middle and distal third of the esophagus accounted for 6, 6 and 4 cases each respectively. Surgery (esophageal resection) was done in 10 cases, and this is comparable with endoscopic removal which was done in 5 cases.

There are two types of coexistence: (1) the carcinoma covers the benign tumor in the overlying type, and (2) completely separated lesions elucidated through or after the esophagectomy for esophageal SCC [2, 12, 19]. In our case study, the two kinds of coexistence were present and this result was compatible with the results of two other studies done by Iwaya et al. and Geramizadeh et al. [2, 15]. Two concepts might be important in the pathogenesis of the disease and carcinoma development over a leiomyoma: (1) A bulging of the leiomyoma into the esophageal lumen may cause the esophageal mucosa to be irritated chronically and predispose it to dysplasia; (2) The underlying leiomyoma itself may prevent the overlying SCC from spreading and also a deep invasion [2, 13, 15].



For the differentiating esophageal carcinoma and leiomyoma, an upper GI endoscopy is usually used, but as the esophageal leiomyomas are submucosal lesions, it may not always lead to a precise diagnosis. Detection and management of these tumors could be done by the use of EUS by revealing the five-layered structure of the gastrointestinal wall as well as by enabling exact localization and origin, tumor margin, echogenic pattern, and exact size measurement [4, 5]; but unfortunately we didn't have EUS in our hospital, so we were unaware about coexistence of any leiomyoma until it had been removed through esophagectomy.

Tumor type	SCC, poorly differentiated (G3)
Residual tumor size	0.6 cm
Tumor extension	Tumor invaded submucosal (at least)
Proximal margin	Free of tumor
Distal margin	Free of tumor
Radial margin	Free of tumor
Treatment effect	Near complete response, score 1 (single cell and small groups of cancer cells
Lymphovascular invasion	Present
Perineural invasion	Present
Lymph-node invasion	Total: 5, involved: 2; metastatic poorly differentiated squamous cell carcinoma (LN2)
Additional pathologic findings	Multiple leiomyomas, the largest one measuring: 2 cm in diameter
Pathologic staging	ypT1bN1Mx
Immunohistochemistry	Negative for C-KIT and CDX2 proto-oncogenes
	Positive for Desmin and P63 proto-oncogenes
Note	This staging categorizes the extent of tumor actually present at the time of examination (after the chemotherapy). The "y" categorization is not an estimate of tumor before chemotherapy

 Table 1
 Tumor pathology and immunohistochemistry characteristics

In narrow-band imaging (NBI) endoscopy, optical filters are used which increase absorbance and scattering of light, and this would boost vessels and other structures appearance, and could afford a high tissue contrast throughout the endoscopy. So with the means of magnifying endoscopes and chromoendoscopy, description of histological tissue and assessment of esophageal lesions are now possible [5]; but unfortunately we did not have any of these to help us evaluate the tumor or esophageal mucosa.

In SMTs which are not accompanied by carcinomas, it is important to find out which layer the tumor originates from by the use of EUS. Endoscopic mucosal resection (EMR) is the treatment of choice in a small SMT originating from the muscularis mucosa. Open surgery or thoracoscopic resection is chosen in a large SMT originating from the muscularis propria. In these cases EMR can't be used because of high risk of complications, such as esophageal perforation and massive bleeding [13]. In our case study, we didn't have EUS in our hospital, so we were unaware about coexistence of any leiomyoma until it had been removed through esophagectomy.

Locally advanced oesophageal cancer is usually treated with neoadjuvant therapy and then surgery, but it's still controversial in choosing between neoadjuvant chemoradiotherapy and chemotherapy, and moreover, there are no benefits according to the patients' survival in using neoadjuvant chemotherapy or neoadjuvant chemoradiotherapy in these patients. This result was understood from a study done by von Döbeln et al., in which a greater tumor tissue reaction in neoadjuvant chemoradiotherapy recipients was found, contrary to no increasing change in patients' survival. He concluded that the radiotherapy should not be added to the neoadjuvant chemotherapy on a routine basis in the patients with resectable esophageal cancer, and should be used in selected patients [20]. In another study done by Visser et al., no differences between neoadjuvant chemotherapy and neoadjuvant chemoradiotherapy were seen in postoperative complications and in-hospital mortality in patients treated for esophageal adenocarcinoma [21]. In the current study, preoperative chemotherapy without radiotherapy was done, which was actually a decision made in a multidisciplinary team due to the facts mentioned above.

To assess an effective esophagectomy with proper LN dissection, the number of LNs retrieved are counted as a quality indicator [22, 23]. In a new study done by van der Werf et al., the number of LNs harvested was associated with more precise staging, but did not have an influence on patient's survival or morbidity and mortality [24]. The number of LNs retrieved is related to the several patient and disease features, including preoperative weight loss, low Charlson comorbidity score, higher clinical N stage, using neoadjuvant therapy, surgical approach (transthoracic or transhiatal), year of resection, and hospital volume [22, 23]. Neoadjuvant chemoradiotherapy would cause the tumor and lymph node down-staging and result in more resections with negative margins and lymph nodes [23, 25]. In our study only five LNs were harvested, and as the patient received three sessions of chemotherapy before the operation, we could propose that the few numbers of the LNs harvested in this study, could be due this fact.

	Country and year	Gender and age	Country and Gender and Chief Risk factors Comorbid Upper Gyear age complaints diseases Endosco	Risk factors	Comorbid diseases	Upper Gl Endoscopy	EUS	Other diagnostic modalities [1]	Other diagnostic modalities [2]	Pathology	Location	Treatment
-	Japan, 1984 [8]	Male/ 75	Stenotic sensation in the pharyngeal region	ing ing	Not avail- able (NA)	An elevated erosive lesion in the posterior wall of the esophagus	۲ Z	Barium esoph- agogram: a tumorous shadow,5 cm, middle thoracic esophagus	¢ Z	with overly- ing SCC	Middle third of the esophagus	Surgery: blunt dissection of the esopha- gus without thora- cotomy, esophago- gastroanas- tomosis
7	Japan, 1995 [9]	Male/45	Dysphagia	ΥN	NA	AN	NA	ΥN	NA	Leiomyoma with overly- ing SCC	Middle third of the esophagus	Esophageal resection and recon- struction
m	Japan, 1995 [9]	Male/71	Not men- tioned, referred for treat- ment of an esophageal tumor	Ч И	Ϋ́Υ	A mucosal irregularity on the sur- face of the submucosal tumor	Ч Х	ΥX	Ч Ч	Leiomyoma with overly- ing SCC	Middle third of esopha- gus	Esophageal resection and recon- struction
4	Japan, 1995 [10]	Male/52	Asympto- matic	Heavy smok- ing	Negative	Sessile polypoid lesion at abdominal esophagus	A 9 mm submucosal tumor with smooth contour, leiomyoma	Barium swallow: as health checkup	Double con- trast barium esoph- agogram: semispheri- cal tumor, 1.5 cm	Leiomyoma with overly- ing poorly differenti- ated SCC	Distal third of the esopha- gus	Surgical Resec- tion
Ŋ	1997 [11]	Male/61	Ϋ́́Υ	٧	Ą	Sessile polypoid lesion with a smooth surface at the esopha- gus	A hypoechoic tumor just below the epithelial layer	Υ	Ч Ч	Moderately differ- entiated SCC on leiomyoma originat- ing from muscularis mucosa	Proximal third of the esophagus	Endoscopic resection by aspiration lumpectomy

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	year year	dender and age	complaints	KISK FACTORS	diseases	upper di Endoscopy	EUS	otner diagnostic modalities [1]	Otner diagnostic modalities [2]	rathology	Location	Ireatment
o ا	Japan, 2002 [12]	Male/62	Not men- tioned, referred for treat- ment of an esophageal tumor	₹ Z	₹ _Z	A protrud- ing lesion, resembling a submu- cosal tumor, in the proximal third of the esophagus	A hypoechoic tumor con- fined to the submucosa with a well demar- cated, smooth outline	Chromoendos- copy: non- staining	₹ _Z	Moderately differenti- ated SCC overlying the leio- myoma	Proximal third of the esophagus	Endoscopic mucosal resection (EMR)
	Japan, 2004 [13]	Male/65	₹ Z	۲ Z	¢ Z	A sessile polypoid lesion with a smooth surface in the proximal third of the esophagus	A hypoechoic tumo, 13 mm in diameter, located in the submu- cosa, with cosa, with cosa, with cosa, with contour, with the muscularis propria layer intact	Chromoendos- copy: non- staining	¥ Z	Leiomyoma with overly- ing poorly differenti- ated SCC	Proximal third of the esophagus	subtotal esophagec- tomy with a three-field (cervical, mediastinal and abdomi- nal) lymph nad dis- section and esophago- gastrostomy
ω	Japan, 2006 [2]	Male/71	Firstly, presented with a cervical mass	Ч Z	Malignant Jymphoma of the thyroid	A superficial ulcerative tumor [24 mm in diameter] in the lower third of the esophagus, overlying the smaller leiomyoma	Cancer overlaid the smaller leiomyoma and invaded the submucosal layer	Barium swallow Esophagogram: two smoothly rounded defects defects	Chromoen- doscopy: non-stain- ing	Squamous cell carcinoma in situ overlying a leiomyoma, separated multiple leiomyo- mas	Distal third of the esopha- gus	Surgery: esophagec- tomy, gastric pull-up tion, cervical anasto- mosis via abdominal, left cervical and right

Tab	Table 2 (continued)	(pi										
	Country and year	Gender and age	Chief complaints	Risk factors	Comorbid diseases	Upper Gl Endoscopy	EUS	Other diagnostic modalities [1]	Other diagnostic modalities [2]	Pathology	Location	Treatment
6	Japan, 2008 [14]	Male/61	Mild epigas- tralgia	₹ Z	¥	A submucosal tumor with a smooth and mildly red surface mucosa in the proximal third of the esophagus	A hypoechoic lesion originating from the muscularis mucosae with a clear margin	Chromoen- doscopy with iodine staining: non- staining area	۲ Z	Leiomyoma with overly- ing SCC	Proximal third of the esophagus	Endoscopic submucosal dissection (ESD)
01	10 Iran, 2009 [15] Male/47	Male/47	Progressive dyspha- gia and epigastric pain and about 20 kg weight loss	∀ Z	ч Х	A depressed area with uneven and irregular border	۲ Z	¢ Z	۲ Z	Several leiomyo- mas, the largest of which was covered by malignant SCC	۲ Z	Laparotomy, transhiatal esophagec- tomy, cervical esophagas- trostomy (Orringer), and bilateral chest tube insertion
2	Japan, 2009 [16]	Male/65	Asympto- matic	Heavy smok- ing and alcohol consump- tion	Negative	An esopha- geal tumor in the mid- dle thoracic esophagus	Hypoechoic tumor in the muscularis mucosae covered with intraepi- thelial layer indicative of leiomyoma	Chromoen- doscopy with iodine staining non- staining area	Magnifying endoscopy with narrow band imag- ing (NBI): dilated and elongated intrapapil- lary loop (IPCL), indicated intraepi- thelial carcinoma	Leiomyoma with overly- ing SCC	Middle third of the esophagus	ESD

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Turkey 201 Maler 4 Weight Los Negative statistics in markets in the statistic in the addres in the statistic in the addres in the statistic in the addres in the addre		Country and year	Gender and age	Chief complaints	Risk factors	Com orbid diseases	Upper Gl Endoscopy	EUS	Other diagnostic modalities [1]	Other diagnostic modalities [2]	Pathology	Location	Treatment
Japan, 2013 Female/75 Exophageal NA Hyperglyce- tumorat Aubmucosal NA NA Coexist- erce of the inci- sory pageal 20m from the inci- sory pageal 1171 the inci- tumorat the inci- tumorat the inci- tumorat the inci- sory pageal	2	Turkey 2012 [4]	Male/44	Weight loss and dys- phagia for two month	Negative	Negative	A tumoral mass with a partially irregular surface in the distal third of the esophagus	¥ Z	CT scan of the thorax and abdomen: minimally diffuse esophageal wall enlarge- ment in a 5 cm seg- ment of the distal esopha- gus with no lymph node involvement or distant metastasis	Ž	SCC infil- trated to the submucosa and two separated leiomyoma along with five reactive and three metastatic lymph nodules	Distal third of the esopha- gus	Distal esophagec- tomy and an esophago- gastrostomy together with a lapa- rotomy and thoracotomy (lvor-Lewis approach)
South Korea, Male/53 Esophageal NA NA An elevated NA Chromoen- NA Squamous Proximal Si tumor at tumor at mass, tumor at is cell third of the deactory the deactory with indine carcinoma esophagus vith irregue staining: non- overlying submucosal small erosion in the upper thoracic esophagus submucosal sector in the upper thoracic esophagus submucosal solution to esophagus submucosal sector in the upper thoracic esophagus submucosal solution the upper thoracic esophagus solution the upper thoracic esophagus solution the s	ő	Japan, 2013 [17]	Female/75	Esophageal tumor at health checkup	۲ Z	Hyperglyce- mia and osteopo- rosis	A submucosal tumor, measuring 5 x 4 mm in diameter, 20 cm from the incisors	ч Х	¥ Z	Ч И И	Coexist- ence of esophageal SCC in situ (high-grade intraepi- thelial neoplasia) overlying elomyoma	20cm from the inci- sors (proxi- mal third of the esopha- gus)	EMR
	4	South Korea, 2014 [19]	Male/53	Esophageal tumor at health checkup	۲ Z	₹ Z	An elevated mass, 1.8 cm in diameter, with irregu- lar central small erosion in the upper thoracic esophagus	A Z	Chromoen- doscopy with iodine staining: non- staining area	¢ Z	Squamous cell carcinoma in situ overlying submucosal leiomyoma	Proximal third of the esophagus	Subtotal esophagec- tomy

Tab	Table 2 (continued)	(pa										
	Country and Gender and year age	Gender and age	Chief complaints	Risk factors	Comorbid diseases	Upper Gl Endoscopy	EUS	Other diagnostic modalities [1]	Other diagnostic modalities [2]	Pathology	Location	Treatment
15	15 South Korea, 2015 [1]	Male/66	Esophageal tumor at health checkup	₹ Z	₹ _Z	A protruding mass that measured 1 × 1 cm in diameter and was located 25 cm from the upper incisors	A hypoechoic and homo- geneous lesion that originated from the muscularis mucosa (MM) below the epithe- lial layer	Chromoen- doscopy with iodine staining: non- staining area	₹ Z	Squamous cell carcinoma in situ overlying submucosal leiomyoma	Middle third of the esophagus	ESD
10	Iran, 2020 (current study)	Female/41	Progressive dysphagia and signiff- cant weight loss for the preceding 6 months	Negative	Negative	A large fungating and ulcera- tive mass, middle and lower third of esophagus, 25–33 cm from the upper inci- sors	Υ N	Chest CT scan: a 29*18 mm soft tissue full- ness at middle to distal esophagus, suggestive for tumoral lesion	₹ Z	Multiple leiomyo- mas, SCC overlying a leiomyoma	Middle and distal third of the esophagus	Extended thor- acotomy, esophagec- tomy, gastric pull-up, pyloroplasty, cervical esophago- stomy (Orrin- ger)

To sum up it can be concluded that (1) It must be kept in mind that esophageal carcinomas may coexist with leiomyomas, (2) Preexisting benign tumors may have played an important role in the development of carcinoma by inducing constant stimulation of the overlying mucosa, (3) EUS is recommended to avoid overestimating the extent of tumor invasion and the resultant aggressive radical surgery (4) As the developing countries had limited equipment, esophageal resection could be the modality of choice in the treatment.

Abbreviations

NA: Not available; SMT: Submucosal mesenchymal tumor; LN: Lymph node; EUS: Endoscopic ultrasonography; CT: Computerized tomography; GI: Gastrointestinal; VATS: Video-assisted thoracoscopic surgery; SCC: Squamous cell carcinoma; IV: Intravenous; MM: Muscularis mucosae; ESD: Endoscopic submucosal dissection; EMR: Endoscopic mucosal resection; NBI: Narrow band imaging.

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Authors' contributions

SM evaluated the patient clinically, operated the patient (main surgeon), read and revised the paper. MJYB evaluated the patient clinically, helped to operate the patient (co-surgeon), prepared the first draft and revised the paper. SH evaluated the pathology slides (our pathologist and revised the manuscript. HS helped in collecting the patient data, assisted with the administration and monitoring of anesthesia during patients operation and upper GI endoscopy (our anesthetic technician). All authors read and approved the final manuscript.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

Written informed consent was obtained from the patient for participation. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests

The authors of this manuscript declare no competing of interests.

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