

## CASE REPORT

# Esophageal spindle cell lipoma

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### Abstract

Symptomatic ordinary esophageal lipomas are rare tumors. Spindle cell lipomas (SCLs) of this location are even more infrequent. To our knowledge, only a previous esophageal SCL case has been reported. We describe herein the case of a 62-year-old woman with a long history of heartburn and feeling of abdominal distension. Preoperative investigations, including a Barium meal, gastroscopy, and echoendoscopy revealed a lipomatous polypoid mass attached to the middle esophageal segment. The lesion (3.5×2×1 cm) was excised endoscopically under deep sedation. The final histopathology diagnosis was pedunculated SCL. An accurate diagnosis of esophageal SCL is crucial to rule out malignant lesions, relieve symptoms, and undertake suitable treatment. The main differential diagnosis includes well-differentiated sclerosing liposarcoma, atypical spindle cell/pleomorphic lipomatous tumor, giant fibrovascular polyp, and fat-forming solitary fibrous tumor. Although rare, SCL should be added to the list of lipomatous tumors that can affect the esophagus. Complete excision is the appropriate treatment.

**Keywords:** esophagus, lipoma, spindle cell lipoma.

### Introduction

Lipomas of the esophagus are uncommon, accounting for only 0.4% of all digestive tract benign neoplasms [1]. Most lesions are clinically unapparent because of their small size. Thus, they are usually found incidentally during imaging studies. However, large lesions have been reported to cause symptoms and complications, including dysphagia, regurgitation, epigastralgia, central ulceration with bleeding, globus sensation, and asphyxia [2].

Spindle cell lipoma (SCL) occurs mainly in male patients between 26 and 82 years old and involves the regions of the shoulder, posterior neck, and upper back almost exclusively [3]. The tumor is uncommon and can exhibit atypical locations [4] and histopathological variants [3] that may cause diagnostic difficulties.

Esophageal SCL is an extremely rare neoplasm. As far as we are aware, only one previous case has been described [5].

### Aim

We herein report the clinicopathological features, including an immunohistochemical study, of a new case of esophageal SCL. Additionally, we have reviewed the literature on ordinary esophageal lipoma and exceptional esophageal SCL.

### Case presentation

A 62-year-old woman presented in 2015 with a long history of heartburn, attributable to gastroesophageal

reflux, and feeling of abdominal bloating. Past medical history was significant for arterial hypertension, dyslipidemia, and lactose intolerance. She had been a heavy smoker and drinker for 15 years. The patient underwent an upper gastrointestinal Barium transit study that was normal. Symptoms improved with the treatment of a proton-pump inhibitor.

In September 2020, a gastroscopy was performed that revealed an intraluminal, soft, mobile, submucosa bulging pedunculated polypoid mass yellowish in color with a smooth surface located 30 cm away from the incisors in the middle esophageal portion (Figure 1A). The mass showed a sausage-like shaped, about 4 cm long, occasionally penetrating the gastric cavity. Upper echoendoscopy performed one month later showed the polypoid mass extending caudally to the supracardial level. The lesion was homogeneous and hyperechoic with the appearance of a pedunculated lipomatous polyp (Figure 2B). In May 2021, a hot loop polypectomy was performed endoscopically under deep sedation. Two hemostatic endoclips were placed at the base of the stalk. The postoperative period was uneventful. The patient was doing well one month after the intervention.

### Histopathological features

The macroscopic evaluation of the surgical specimen showed a well-capsulated, soft, yellowish elongated, mucosa-covered mass measuring 3.5×2×1 cm (Figure 2).

The entire surgical specimen was fixed in 10% neutral buffered formalin. Representative tissue samples were embedded in paraffin. For routine microscopy, 4-µm-thick

sections were stained with Hematoxylin–Eosin (HE). Immunohistochemical (IHC) staining was performed using the EnVision FLEX+ Visualization System (Dako, Agilent Technologies, SL, Las Rozas, Madrid, Spain). The IHC reaction was performed using appropriate tissue controls for the antibodies utilized. Automatic staining was accomplished on a Dako Omnis stainer (Agilent Technologies, SL). Antibodies used are detailed in Table 1.

**Table 1 – IHC antibodies used in this study**

Antibody	Source	Clone	Dilution	Retrieval solution pH (Dako)
$\alpha$ -SMA	Dako	1A4	FLEX RTU	High
Anti-human melanosome	Dako	HMB45	FLEX RTU	High
Melan A	Dako	A103	FLEX RTU	High
Calponin	Dako	CALP	1:200	High
CDK4	Gennova	DCS-35	1:25	High
MDM2	Gennova	IF2	1:50	High
P53 protein	Dako	DO-7	FLEX RTU	High
CD34	Dako	QBEnd 10	FLEX RTU	High
S100 protein	Abcam	Ab55787	1:100	High
CD117	Dako	Polyclonal	1:200	High

Abcam, Cambridge, UK; Dako (Agilent Technologies), SL, Las Rozas, Madrid, Spain; Gennova Scientific, SL, Sevilla, Spain;  $\alpha$ -SMA: Alpha-smooth muscle actin; CD: Cluster of differentiation; CDK4: Cyclin-dependent kinase 4; HMB45: Human melanoma black 45; IHC: Immunohistochemical; MDM2: Mouse double minute 2; RTU: Ready-to-use.



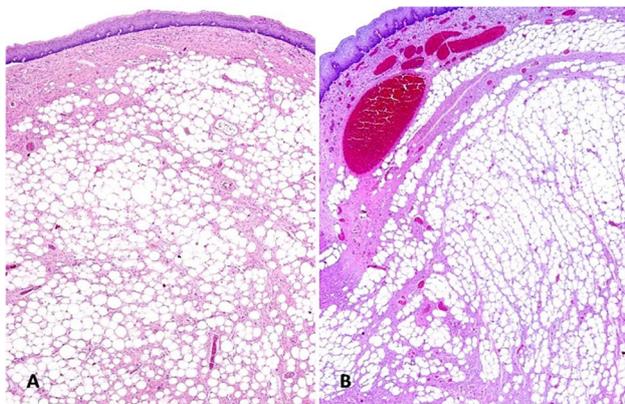
**Figure 1 – Gastroscopy and esophageal sonography:** (A) Gastroscopy shows an intraluminal, yellowish, polypoid mass originating from the esophagus; (B) Sonographically, the lesion is hyperechoic, and homogeneous suggesting an esophageal lipoma.

Microscopically, a well-delimited, mostly encapsulated, neoplastic lesion was observed. A benign squamous mucosa completely covered the tumor (Figure 3A). In the lamina propria of the mucosa, there were occasional dilated vessels filled with blood (Figure 3B). The tumor consisted of a mixture of mature adipocytes and bland mitotically inactive, short, spindle mesenchymal cells. The adipocytes dominated the tumor and showed scant variation in size. Lipoblasts or multinucleated giant cells with a floret-like nuclear pattern were not present. The spindle cells were randomly oriented and had ovoid nuclei with inconspicuous nucleoli and scant cytoplasm (Figure 4A). Scant spindle-cell dominant areas were present (Figure 4B). Between the spindle cells thick, eosinophilic, refringent, rope-like collagen bands were observed (Figure 5). Scattered, abundant mast cells were present along the neoplasm. The surgical border was free of tumor.

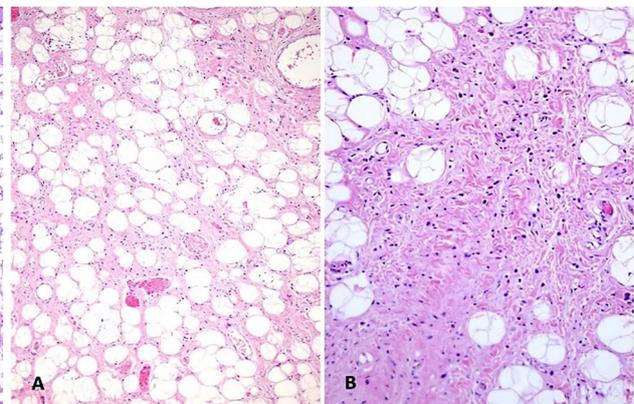
IHC study revealed strong reactivity of the spindle cells for cluster of differentiation (CD)34 (Figure 6A). These cells were negative for S100, alpha-smooth muscle actin ( $\alpha$ -SMA), human melanoma black 45 (HMB45), Melan A, and calponin. In addition, nonreactivity of the neoplastic cells was observed for mouse double minute 2 (MDM2), cyclin-dependent kinase 4 (CDK4), and p53. Abundant mast cells were revealed by CD117 (Figure 6B).



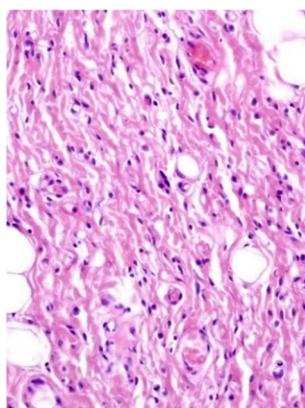
**Figure 2 – Macroscopic aspect of the intraluminal esophageal polypoid mass.** The lesion is covered entirely by mucosa.



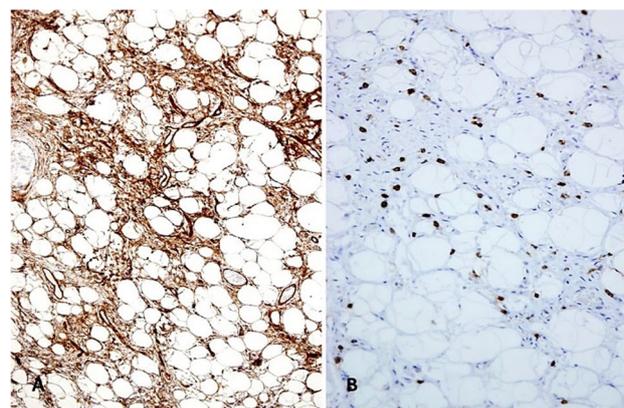
**Figure 3 – Panoramic view of the neoplasia:** (A) A squamous mucosa covers the well-demarcated lipomatous tumor; (B) In the lamina propria of the mucosa, occasional venous vessels are filled with blood. Hematoxylin–Eosin (HE) staining: (A and B)  $\times 40$ .



**Figure 4 – Tumor components in varying proportions:** (A) The neoplasia contains mature adipose cells and randomly oriented bland spindle cells; collagen bands are seen between the cellular elements; (B) Area showing prominent spindle cell component and abundant rope-like collagen bands. HE staining: (A)  $\times 100$ ; (B)  $\times 200$ .



**Figure 5** – Spindle cells have ovoid, slender, uniform nuclei with inconspicuous nucleoli and scant cytoplasm; between them, abundant thick, eosinophilic, refringent, ropy collagen bands can be observed. HE staining,  $\times 400$ .



**Figure 6** – Immunohistochemical study: (A) Intense positivity of the spindle cells for CD34 ( $\times 100$ ); (B) Scattered abundant mast cells are revealed by CD117 ( $\times 200$ ). CD: Cluster of differentiation.

## Discussions

Lipomas can be present in all segments of the gastrointestinal tract (GIT). These tumors throughout the GIT have an incidence of approximately one in 600 necropsies [6]. Lipomas are most commonly found in the colon, the region of the ileocecal valve, and rectum, followed by the small intestine [6]. Uncommon locations include the hypopharynx, esophagus, and stomach [7, 8].

Esophageal lipomas are more common in men than in women with a ratio of 2.1:1. The tumors have been observed in patients between the ages of four and 80 years old (mean age 52.4 years, median age 55 years) [2, 9]. Lipomas more frequently occur in the cervical and upper thoracic esophagus [10]. Thus, Ferrari *et al.* [2] in a review of the literature found that 85.7% occurred in the cervical esophagus, 8.0% in the distal esophagus, and 6.3% in the mid-thoracic esophagus. However, they can potentially appear anywhere from the pharynx to the distal esophagus. Most tumors are small, solitary, asymptomatic, and incidentally detected. Symptoms are related to the size of the lipoma. Tumors over 2 cm in maximum diameter can produce symptoms [9]. Symptomatic patients can undergo dysphagia, regurgitation, odynophagia, recurrent melena, anemia, epigastralgia, substernal fullness, weight loss, cough, aspiration pneumonia, or episodes of asphyxia [2]. Sudden death from asphyxiation can occur due to the regurgitation of a pedunculated esophageal lipoma [2, 11]. The most common complaint is dysphagia [2]. There is also a danger of starvation because obstructive growth interferes with deglutition. In addition, an esophageal diverticulum can complicate a voluminous lipoma [12].

Lesions can be intramural or intraluminal [2, 9]. Most symptomatic lipomas are intraluminal polypoid lesions (88.6%) with a stalk arising from the cervical esophagus. Intramural lipomas (11.4%) are mostly located in the thoracic and distal esophagus [2].

SCL is an uncommon lipoma variant. It occurs predominantly in the subcutaneous tissue of the upper back, posterior aspect of the neck, or shoulder. It is found principally in older men (4.4:1 male/female ratio) with a mean age of 57 years (median 59 years, range 26–82 years) [3]. These tumors are infrequent compared with conventional lipomas (1:60 ratio) [13]. SCL has been

exceptionally described in locations of the digestive tract, such as the hypopharynx [14], esophagus [5], and small intestine [15].

SCL is a benign lesion that has three main components: (i) adult fat cells; (ii) small, thin, uniform, spindle cells arranged in short fascicles between the fat cells; and (iii) bright, eosinophilic, dense rope-like collagen bundles. The proportion of spindle cells and the adipocytic component can vary considerably. Abundant scattered mast cells are usually present. In addition, cells with enlarged hyperchromatic nuclei arranged in semicircles (florete-like multinucleated cells) or mildly pleomorphic multinucleated cells may be observed (pleomorphic variant of SCL). Immunohistochemically, spindle and florete-like cells are reactive for vimentin (100%), CD34 (100%), B-cell lymphoma-2 (Bcl-2) (92.3%), desmin (16%), and S100 protein (15%); and non-reactive for, muscle actin, MDM2, and CDK4 [3, 16, 17]. Fluorescence *in situ* hybridization (FISH) for MDM2 amplification is negative [4]. Besides, tumor cells show loss of nuclear retinoblastoma 1 (RB1) protein expression [18].

To the best of our knowledge, only one case prior to the present one has been reported in the esophagus. Razzak *et al.* [5] described the case of a 60-year-old man with progressive dysphagia and weight loss who presented with several episodes of near asphyxiation secondary to regurgitation and aspiration of a pedunculated mass. The preoperative study included a Barium meal, esophagoscopy and computed tomography. The mass was mobile, pedunculated immediately distal to the cricopharyngeus muscle. The lesion was excised through open surgery by a cervical approach. The histopathological study revealed a 5 $\times$ 4 cm, oval-shaped encapsulated, yellowish, well-circumscribed, pedunculated mass covered with mucosa. The lesion corresponded to an SCL with focal presence of florete-like cells.

The appropriate treatment for symptomatic SCL is complete excision. An esophageal resection is not required.

SCL must be differentiated from well-differentiated sclerosing liposarcoma (WDSL) [19], atypical spindle cell/pleomorphic lipomatous tumor (ASCLT) [20], giant fibrovascular polyp (GFVP) [21] and fat-forming variant of solitary fibrous tumor (SFT) [22].

The main histopathological finding of the WDSL is the presence of scattered atypical stromal cells, showing

intense nuclear hyperchromasia placed in an extensive fibrillary collagenous stroma. Multivacuolated lipoblasts can be present. MDM2 and/or CDK4 nuclear immunopositivity is present in most cases [19]. The addition of p16 to MDM2 and CDK4 increases significantly diagnostic specificity [23]. The tumor can show recurrences. However, WDSL does not metastasize unless the tumor shows dedifferentiation [24].

ASCLT locates more commonly in the hand, foot, and thigh. It is most common in males (mean age 54 years; range, six to 87 years). Grossly, is an unencapsulated tumor with margins ill-defined. It shows variable proportions of atypical spindle cells, adipocytes, lipoblasts, pleomorphic multinucleated cells, and a myxoid to collagenous extracellular matrix. Tumor cells show variable positivity for CD34, S100, and desmin. MDM2 and CDK4 are negative. Loss of nuclear RB1 expression can be observed in about 57% of cases. The recurrence rate of this tumor is around 13% [20]. Dedifferentiation or metastases has not been reported. No case of ASCLT has been described in the esophagus; however, a case has recently been documented in gastric cardia [25].

GFVP is a large polypoid mass that occurs in adults predominantly in the 5<sup>th</sup> decade and the proximal esophagus. The lesion is poorly delimited, edematous, and hypocellular. It consists of a random mixture of spindle cells, adipocytes, and prominent vessels. Disseminated atypical cells with hyperchromatic nuclei can be detected. The atypia in some cases is subtle. Most of these cases correspond to well-differentiated liposarcomas that show IHC positivity for MDM2 and CDK4 [21, 26]. Rhabdomyomatous differentiation can rarely occur in this tumor [27].

SFT is composed of uniform spindle cells arranged around branching and hyalinized hemangiopericytoma-like (staghorn-shaped) blood vessels with a background of hyalinized (keloidal-type) collagen. Cellularity and collagenization are variable. The tumor may contain an adipocytic component (fat-forming SFT) [28] or multinucleated stromal cells (giant-cell angiofibroma pattern). Mitotic activity is usually low. Neoplastic cells show CD34, Bcl-2, vimentin, and nuclear signal transducer and activator of transcription 6 (STAT6) expression by immunohistochemistry [29]. Besides, the cells retain expression of nuclear RB1 [18].

## ☒ Conclusions

SCL is an extremely uncommon tumor of the esophagus. We are reporting the second case of the literature. The tumor may present as a pedunculated polypoid mass. When it reaches a considerable size, it can lead to symptoms and complications. SCL is potentially a life-threatening condition. The tumor should be differentiated from several subtypes of lipomatous or fat-forming tumors, including liposarcomas. The recommended treatment is complete excision.

## ☒ Conflict of interests

The authors declare that they have no conflict of interests.

## ☒ Compliance with ethical standards

No Ethics Committee approval is required in our institution for a case report involving a single patient.

## Consent

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

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