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Understanding esophageal neurofibroma: A case series and systematic review

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ABSTRACT

INTRODUCTION: Esophageal neurofibroma is a rare benign esophageal neoplasm. With very few cases documented in the literature, not much is known about the demographics and clinicopathologic features of this tumor. This study was aimed at presenting a case report of an esophageal neurofibroma, and to conduct a systematic review of published cases.
METHOD: This review was performed according to the PRISMA guidelines. Literature search was conducted through PubMed, SCOPUS, and Cochrane Databases from inception until May 2020 for all histologically confirmed cases of esophageal neurofibroma.
RESULTS: 28 cases, including the newly reported case, were included in the review. The mean age at diagnosis was 53.3 years ±12.1. 53.6% were male. Dysphagia was the most common presenting symptom (53.6%). Most of the reported cases involved the upper esophagus (39.3%). The most utilized diagnostic test was esophagogastroduodenoscopy (57.1%). The mean tumor size was 6.1 cm ± 5.1. Preoperative biopsy was done for 9 cases, out of which seven were negative or inconclusive. In 17 cases (60.7%), immunohistochemical (IHC) staining of the resected tumor was not performed. S100 was the most utilized IHC stain. Enucleation (39.3%) was the most common treatment, followed by esophagectomy (28.6%).
CONCLUSION: Esophageal neurofibroma should be considered in the setting of dysphagia caused by a subepithelial tumor. Accurate preoperative histologic diagnosis by using a well-defined biopsy algorithm, in conjunction with IHC analysis, will favor less aggressive surgical treatment and surveillance of asymptomatic lesions. Minimally invasive surgical treatment is feasible and should be considered when the expertise is available.

1. Introduction

Benign esophageal tumors are not common and the majority of these tumors are leiomyomas which make up about 80% [1,2]. Neurofibromas are very rare benign neoplasms of the esophagus with less than 30 reported cases in the literature [1]. Due to the rarity of these tumors, there is limited data to understand the disease and there is currently no consensus on management algorithm. In this study, a case report of distal esophageal neurofibroma managed by minimally invasive esophagectomy is presented. A systematic review of the published literature on esophageal neurofibroma was also conducted. Cases were extracted from the literature review to generate data for analysis. This work has been reported in line with the PROCESS guidelines 2018 [3].

2. Methods

2.1. Ethical consideration

The case report component of this study was reviewed by the institutional review board (IRB) and approval was granted. An informed consent was obtained from the patient for treatment, data collection, and reporting. The systematic review component of the study did not meet criteria for IRB review and an IRB exempt was obtained. This study was registered with the Research Registry (UIN: researchregistry6084).

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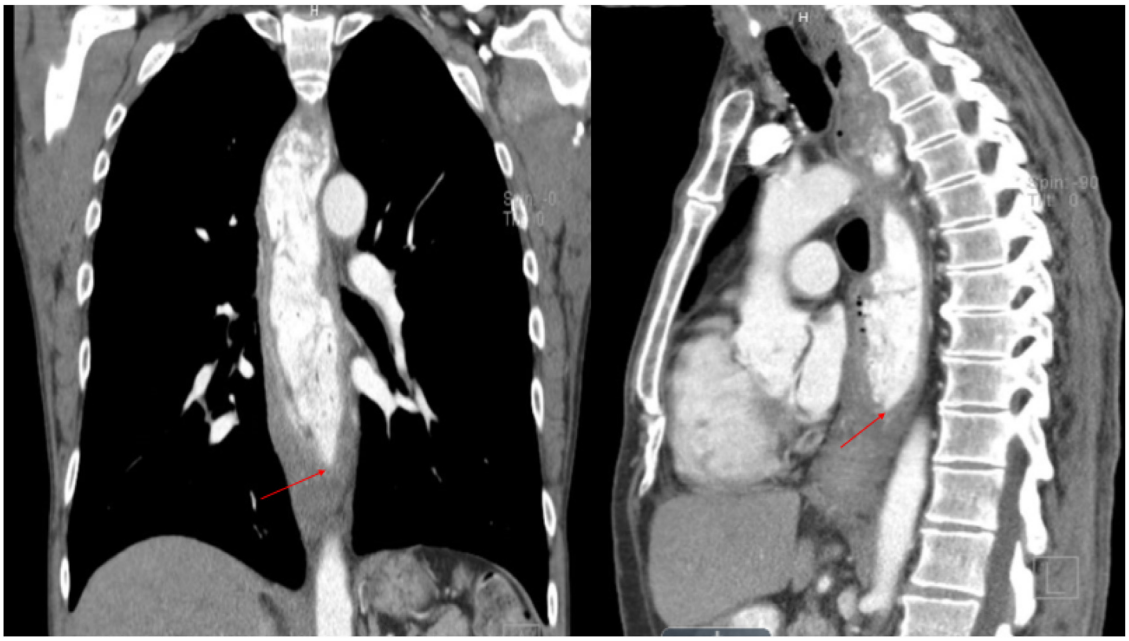

Fig. 1. CT chest showing the distal esophageal tumor with obstruction.

2.2. Search strategy

This review was performed according to the Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) guidelines. Three of the authors (CO, SS and TO) independently performed a literature search in the SCOPUS (Elsevier), PubMed (NLM NIH) and Cochrane (Wiley) databases using the keywords "Esophageal Neurofibroma". Databases were searched from inception until May 31, 2020. The search was limited to human case reports and case series with no limitations to the date of publication, language, and text availability. The references from the articles obtained were reviewed and additional relevant papers were hand searched and reviewed.

2.3. Selection criteria

All case reports and case series involving patients with histologic confirmation of esophageal neurofibroma were included in the review.

2.4. Data extraction

All selected articles were reviewed and the following data were retrieved: age, gender, presenting symptoms, presence of predisposing genetic condition such as neurofibromatosis, diagnostic tests, number of tumors, location of the tumor, size of the tumor, immunohistochemical analysis and type of surgery. We also extracted the authors' names and year of publication of the papers.

2.5. Statistical analysis

Descriptive statistics were used to present the demographic, clinical and pathologic features of the pooled data from all the selected studies. Continuous variables were presented as mean with standard deviation while categorical variables were presented as proportions. Statistical analysis was performed using SPSS version 26.

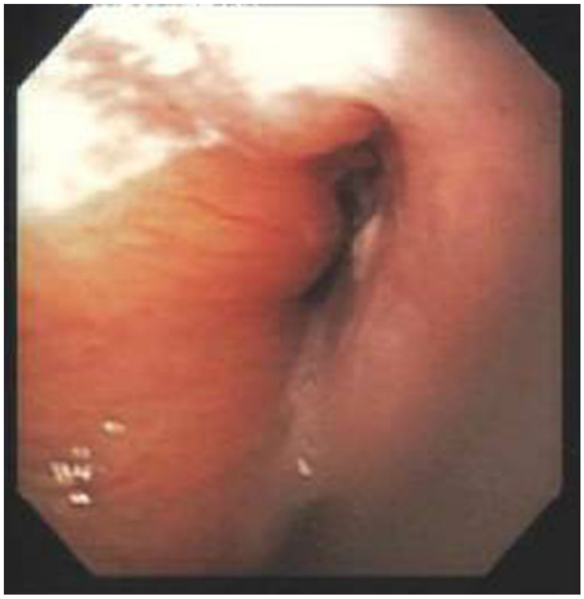

Fig. 2. EGD showing narrowing of the distal esophagus.

3. Case presentation

The patient is a 60-year-old African American male with a ten-year history of dysphagia and a prior diagnosis of esophageal stricture that failed serial esophageal dilation. The last esophageal dilation was 5 years prior to his present evaluation. At presentation, he complained of worsening dysphagia to solids and liquids. He was barely able to tolerate a clear liquid diet. There was associated 36 kg weight loss over the preceding 12 months. He has a 40 pack-year history of cigarette smoking and daily alcohol use for several years. The past medical history is not significant for any other comorbidities. He underwent splenectomy for traumatic ruptured spleen following motor vehicle crash thirty years prior to presentation. The

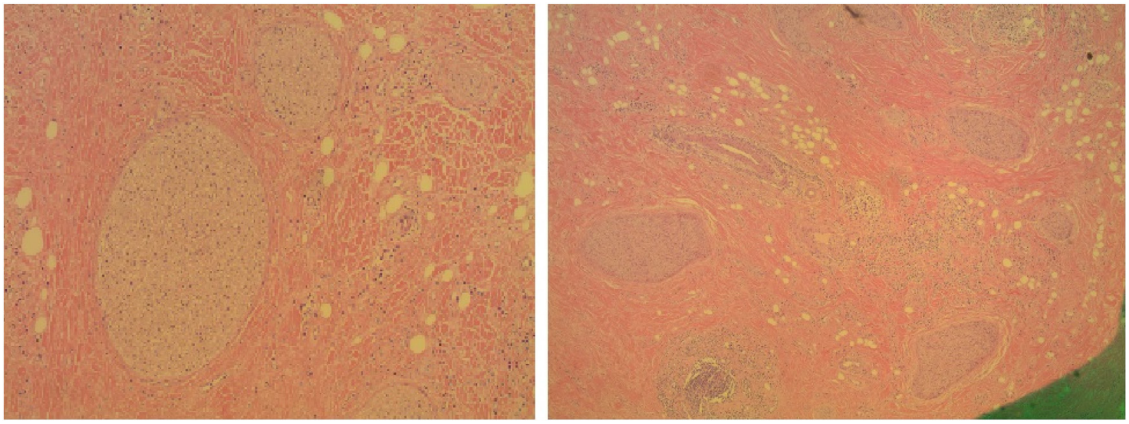

Fig. 3. Histologic stains showing submucosal stromal cells, marked fibrosis and multiple nerve bundles.

physical examination was significant for a severely malnourished middle-aged man (BMI = 15 kg/m2). The initial diagnostic work up with computerized tomographic (CT) scans of the chest, abdomen and pelvis showed a 6 cm long distal esophageal mural wall thickening which was concerning for neoplasm (Fig. 1). Mildly enlarged gastrohepatic and celiac lymph nodes were also identified. Esophagogastroduodenoscopy revealed narrowing of the distal esophagus (Fig. 2) located at 37 cm from the incisors. An endoscopic ultrasound (EUS) was also done and showed a 5.9 cm × 1.8 cm × 2.0 cm submucosal mass. A fine needle aspiration (FNA) cytology of the mass was non-diagnostic. It showed stromal tissue with evenly distributed and bland spindle cell nuclei. The specimens were CD117 and DOG 1 negative, which ruled out gastrointestinal stromal tumor. The possibility of low-grade stromal neoplasm like Leiomyoma was raised. In view of his poor nutritional status, a feeding jejunostomy tube was placed for nutritional rehabilitation. The patient was presented at the multidisciplinary tumor conference. The consensus recommendation was to continue nutritional rehabilitation and to repeat endoscopic and radiologic studies in 2 months.

A follow up CT of the chest and abdomen 2 months later showed progressive enlargement of the distal esophageal mass with eccentric narrowing and associated proximal dilation of the esophagus, as well as an increase in the size of the celiac nodes. An MRI of the chest and abdomen also demonstrated the earlier noted mass and lymphadenopathy. A repeat EUS with FNA was concerning for esophageal leiomyoma. The case was presented for discussion again at the multidisciplinary tumor conference. At this point, due to concerns for increase in size of the tumor, worsening obstruction and possible underlying malignancy, the consensus recommendation was to remove the tumor surgically. The patient underwent minimally invasive McKeown esophagectomy with gastric conduit. The immediate postoperative recovery was uneventful. He was discharged home on postoperative day #8.

The histologic examination of the specimen showed submucosal stromal fibrosis with multiple nerve bundles in a background of chronic inflammation, multinucleated giant cell reaction and lymphoid aggregates (Fig. 3). Immunohistochemical analysis was positive for S-100 (Fig. 4), SOX - 10 and focal staining for CD 34. The overall picture was consistent with pathologic diagnosis of esophageal neurofibroma.

Post operatively the patient did well. He was able to resume and tolerate a regular diet 4 weeks after discharge from the hospital. The feeding jejunostomy was discontinued after 6 weeks. There was no complaint at 12-week, 18-week, 24-week and 32-week follow up visits.

The patient was managed at a tertiary academic center and the surgeon was a board-certified surgical oncologist.

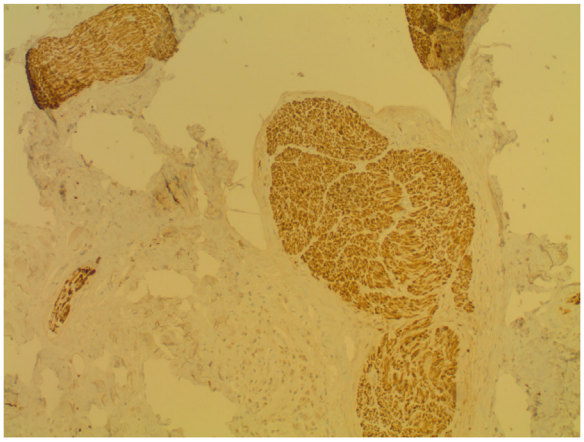

Fig. 4. Immunohistochemical stain positive for S100.

4. Systematic review

After reviewing the articles, 25 met our selection criteria. The full text, English version of seven articles were not available and they were excluded. Sixteen of the selected articles were individual case reports [2–17] and two were case series [1,18]. The PRISMA flowchart in Fig. 5 summarizes the selection process. From the selected papers, there were 27 cases altogether. With inclusion of the case discussed above, the total number of cases in this review is 28 (Table 1).

The age of the patients at the time of diagnosis ranged from 26 years to 75 years with a mean age of 53.3 years. The median age of the patients is 55.5 years. Most of the cases reported were diagnosed in patients 50 years or older (21 out of 28, 75%) (Table 2). In fact, most cases were diagnosed in the 6th decade (42.9%), followed by those in the seventh decade of life (25%). The male gender constituted 53.6% of the cases.

Dysphagia was the most common presenting symptom (53.6%). Shortness of breath was documented in two patients and both had upper esophageal lesions. Of the 28 cases, 4 (14.3%) had coexisting diagnosis of neurofibromatosis. Most cases were solitary neurofibromas (67.9%). Of the 4 with multiple lesions, 1 had coexisting Von Recklinghausen's disease. Most of the reported cases were in the upper esophagus (39.3%) with an equal distribution between the mid and distal esophagus. Of note, eleven patients had distance from the incisors documented for the location of the tumor within the esophagus.

The most common diagnostic approach utilized was esophagogastroduodenoscopy (57.1%), and the most adopted imaging

Table 1
Reported cases of esophageal neurofibroma.

Authors	Year	Age	Sex	NF	Symptoms	Location (distance from incisors)	Workup	Size (cm)	IHC Stains	Treatment
Engelking et al. [12]	1949	39	F	No	Indigestion, Dysphagia	Mid esophagus (26–30 cm)	EGD, Barium Swallow	5 × 3.5 × 3	NR	Enucleation (Open Thoracotomy)
Sturdy [13]	1967	51	F	No	Epigastric pain, dysphagia	Lower esophagus (35 cm)	EGD, Barium Swallow	7.6 × 5.0	NR	Esophagectomy (Open Thoracotomy)
Timm et al. [19]	1975	43	M	No	Dysphagia, GI bleed	Upper Esophagus	NR	NR	NR	Wedge resection
Saitoh [1,21]	1977	26	M	No	Dysphagia	Mid esophagus	NR	4.2 × 4.0 3.0	S100	Enucleation
Goto et al. [1,21]	1982	56	M	No	Abnormal esophageal shadow	Lower esophagus	NR	NR	NR	NR
Oguchi et al. [1,21]	1983	55	M	No	Prolapsed tumor	Upper esophagus	NR	22.5 × 4.5	NR	Enucleation
Inoue et al. [1,21]	1984	50	M	No	Abnormal esophageal shadow	Mid esophagus	NR	0.7 × 0.5	NR	Enucleation
Hisikawa et al. [9]	1984	55	M	No	Epigastric Pain	Mid esophagus	Barium Swallow, EGD, FNA.	2 × 2	NR	Serial follow-up (non-surgical)
Saitoh et al. [6]	1985	64	F	No	Abnormal esophageal shadow, Dysphagia	Mid esophagus	EGD, CXR	4.2 × 4 × 3	S100	Enucleation
Fujiwara et al. [1,21]	1985	75	F	No	GI bleed	NR	NR	NR	NR	NR
Madrid et al. [14]	1986	53	F	No	Dysphagia, Pain Vomiting	Upper esophagus (20 cm)	CXR, EGD EUS Barium Swallow, FNA	8 × 6 × 3 & 2.5 × 2.5 × 2	NR	Enucleation (Open Thoracotomy)
Hara et al. [1,21]	1987	67	F	No	Dysphagia	Mid esophagus	NR	1.7 × 1.5	S100	Enucleation
Sugiyama et al. [1,21]	1989	36	M	No	Abnormal esophageal shadow	Upper esophagus	NR	11.0 × 6.5	NR	Esophagectomy
Ohashi et al. [1,21]	1990	34	M	No	Abnormal esophageal shadow	Upper esophagus	NR	3 × 2.7	S100	Enucleation
Ramirez et al. [1,21,22]	1992	61	F	No	Not reported	Mid esophagus	NR	NR	NR	NR
Fujita et al. [1,21]	1993	48	F	No	Abnormal esophageal shadow	Lower esophagus	NR	6 × 5	S100	Esophagectomy
Lee et al. [2]	1997	58	F	No	Dysphagia, Odynophagia	Upper esophagus (20 cm)	EGD, CT chest, Barium Swallow, FNA	4.0 × 6.0	S100	Enucleation (Right Thoracotomy)
Ishii et al. [15]	2002	35	F	No	Foreign body sensation	Upper esophagus (2 cm from oral end)	EGD Laryngoscopy	Multiple (0.2 - 0.4)	NR	Enucleation
Ganeshan et al. [7]	2005	67	M	Yes	Dysphagia	Lower esophagus (35 cm)	EGD, EUS, CT scan, Barium swallow, FNA	Multiple	NR	Esophagectomy
Sicca et al. [20]	2005	56	M	No	Dysphagia	GE Junction	EGD, EUS, CT scan	Multiple	NR	Esophagectomy
Sica et al. [16]	2006	56	M	Yes	Dysphagia	Lower esophagus (35 cm)	EGD, EUS, CT scan, Barium Swallow, Manometry FNA	NR	NR	Esophagectomy

Table 1 (*Continued*)

Authors	Year	Age	Sex	NF	Symptoms	Location (distance from incisors)	Workup	Size (cm)	IHC Stains	Treatment
Nishikawa et al. [4]	2013	56	F	No	Dysphagia	Mid esophagus (25 cm)	EGD, EUS, MRI FNA	3.4 × 2.8 × 2.2	S100	Enucleation (VATS)
Tanaka et al. [11]	2013	61	M	Yes	Dysphagia	Upper esophagus	EGD, EUS, CT scan, MRI	4.4 × 0.6 × 1.3.	S100 CD 34	Endoscopic Submucosal Dissection
Garcia-Valesquez et al. [17]	2015	51	M	No	Incidental finding	Lower esophagus	CXR, MRI, EGD	4 × 3.2	S100, CD56	Wedge resection
Somnath et al. [18]	2015	50	M	Yes	Neck swelling, Dysphagia, Shortness of breath	Upper esophagus	MRI, Barium Swallow, FNA.	8 × 8 × 6	NR	En bloc resection
Yang et al. [5]	2017	63	M	No	Dysphagia, Chest pain	Upper esophagus (18 cm)	EGD, EUS, Barium Swallow	12 × 3 × 2	PGP 9.5, Vimentin, Nestin, CD56	Enucleation
Booka et al. [1]	2018	73	F	No	Shortness of Breath	Upper esophagus (18 cm–23 cm)	EGD, EUS, MRI, 13 FDG Positron, FNA.	9 × 5 × 5	S100	Esophagectomy
Present case	2020	60	M	No	Dysphagia	Lower esophagus (37 cm)	CT scan, EGD, EUS, MRI, FNA	5.9 × 1.8 × 2.0	S100, CD 34	MIS Esophagectomy

NR = Not Reported, NF = Neurofibromatosis.

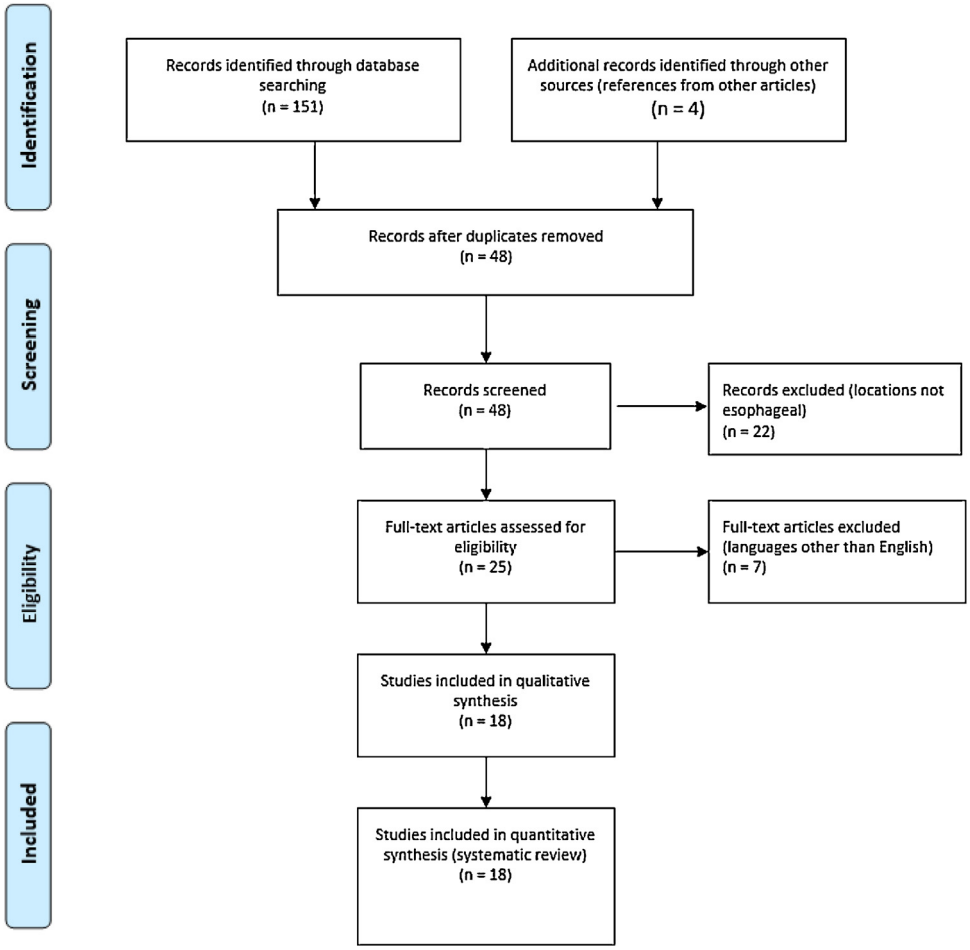

Fig. 5. PRISMA flow diagram showing search algorithm used for systematic review.

modality was the barium swallow (32.1%). Computerized tomographic scan was the next frequently used diagnostic imaging modality. Eleven (39.3%) of the cases had no documentation regarding the diagnostic tests used (Table 2).

The mean diameter of resected tumors was 6.1 cm ± 5.1. Nine of the cases documented preoperative biopsy and 7 were negative or inconclusive. Based on this data, the estimated sensitivity for preoperative percutaneous or endoscopic biopsy is 22.2%. The most assessed immunohistochemical stain was the S100 (39.3%). The surgical specimens for most cases (60.7%) were not subjected to immunohistochemical analysis. Of the specimens reported to have been tested for S100, 10 were positive and only 1 was negative. This gives S100 a 90.9% sensitivity. Other frequently used immunohistochemical stains include CD 34, CD 117 and Desmin. Two out of 6 cases stained for CD34 were positive. Two specimens were stained for CD56 and both were positive. None has been shown to stain positively for CD 117 which is a characteristic stain for gastrointestinal stromal tumors.

Enucleation was the most common modality of treatment (39.3%). Eight patients (28.6%) underwent esophagectomy and only 1 case was managed by observation. Three (10.7%) of the reported cases were treated using minimally invasive techniques – Endoscopic Submucosal Resection (ESMR), Video-Assisted Thoracoscopy (VATS) enucleation and MIS McKeown Esophagectomy (Table 2).

5. Discussion

Neurofibroma of the esophagus is a rare benign neoplasm usually made up of a combination of neural and connective tissues [1,4]. Reviews of esophageal submucosal tumors (SMTs) have reported a prevalence of about 0.9% [1,4,5]. While most cases of visceral organ neurofibromas are associated with genetic disorders such as Von Recklinghausen's disease, isolated occurrences have been reported [6]. Neurofibromas can be localized, diffuse or plexiform. Of the three types, localized neurofibromas are the most common in the gastrointestinal (GI) tract. In fact, one case of plexiform neurofibroma of the GI tract is reported so far in the literature [1,7]. Solitary esophageal neurofibroma is the most common form of esophageal neurofibroma. It is also pertinent to point out that presenting with multiple esophageal neurofibromas is possible without any underlying or coexisting diagnosis of Von Recklinghausen's disease.

Like most esophageal lesions, esophageal neurofibroma can present with a variety of symptoms but dysphagia constitutes the most common presenting symptom [1]. Dysphagia may be the sole presenting symptom or part of a constellation of complaints. Epigastric or chest pain was another symptom frequently observed in these patients. Possible postulates which may explain the noted symptoms include direct nerve invasion, mass effect, or connective tissue involvement [5].

Regarding the location of the tumor within the esophagus, variation in the anatomic landmarks used to divide the esophagus to

Table 2
Results from the systematic review.

Characteristic	Number of patients (percentage)
Age	
< 50 years	7 (25%)
≥ 50 years	21 (75%)
Mean age- 53.3 years ± 12.1	
Median age- 55.5 years	
Gender	
Male	15 (53.6%)
Female	13 (46.4%)
Symptoms	
Dysphagia	15 (53.6%)
Abnormal imaging	7 (25%)
Chest/Epigastric pain or discomfort	6 (21.4%)
Indigestion	1 (3.6%)
Gastrointestinal bleeding	2 7.1%)
Prolapsed tumor	1 (3.6%)
Odynophagia	1 (3.6%)
Foreign body sensation	1 (3.6%)
Neck swelling	1 (3.6%)
Shortness of breath	2 (7.1%)
Vomiting	1 (3.6%)
Neurofibromatosis	
Yes	4 (14.3%)
No	24 (85.7%)
Location	
Upper Third	11 (39.3%)
Middle Third	8 (28.6%)
Lower Third	8 (28.6%)
Not reported	1 (3.6%)
Work up	
EGD	16 (57.1%)
EUS	10 (35.7%)
Barium Swallow	9 (32.1%)
CT Scan	7 (25%)
Laryngoscopy	1 (3.6%)
MRI	6 (21.4%)
Chest X Ray	3 (10.7%)
FNA	9 (32.1%)
HIDA	1 (3.6%)
Manometry	1 (3.6%)
Not reported	11 (39.3%)
Treatment	
Enucleation	11 (39.3%)
Wedge/En bloc resection	3 (10.7%)
Endoscopic Submucosal Dissection	1 (3.6%)
Esophagectomy	8 (28.6%)
Observation	1 (3.6%)
Not Reported	4 (14.3%)
Mean tumor size	6.1 cm ± 5.1
Number of Tumors	
Single	19 (67.9%)
Multiple	4 (14.3%)
Not reported	5 (17.8%)
FNA results	
Positive	2 (7.1%)
Negative	7 (25.0%)
Not reported	19 (67.9%)
Immunohistochemical Stains	
S100	11 (39.3%)
CD56	2 (7.1%)
PGD 9.5	1 (3.6%)
Nestin	1 (3.6%)
Desmin	5 (17.9%)
Vimentin	1 (3.6%)
CD34	6 (21.4%)
CD117	6 (21.4%)
SMA	3 (10.7%)
NSE	1 (3.6%)
DOG 1	1 (3.6%)
SOX 10	1 (3.6%)
Actin	1 (3.6%)
Not reported	17 (60.7%)

upper, middle, and lower esophagus is recognized as a potential source of bias in this observation. While some authors used distance from the incisors to location within the esophagus as the landmarks, others attributed location to position of the esophageal lesion within the thoracic cavity. A better approach to documentation is the use of distance from the incisors as it allows for uniformity and precision in determining location of the tumors.

Preoperative diagnosis of esophageal neurofibroma was observed to be a constant challenge across all cases in the literature. Radiologic studies like barium swallow, CT scan, MRI and EUS will show the narrowing of esophageal lumen and submucosal location of the tumor. So far, there are no radiologic features unique to esophageal neurofibroma. With differential diagnoses including other SMTs such as leiomyomas, histologic analysis is important for confirmation of diagnosis.

Preoperative fine needle or core needle biopsies yielded varying results. From the cases reviewed, fine needle aspiration cytology showed extremely low sensitivity. This shows severe limitation of fine needle aspiration cytology in preoperative diagnosis. This may be related to the limited tissue obtained and inability to perform detailed histology or IHC analysis. To address this issue, several technical factors must be considered. The type and size of the needle must be carefully chosen to improve diagnostic accuracy, adequacy of sample size and decrease the number of passes needed. Having an on-site cytopathologist also improves the diagnostic yield of EUS-guided FNA. Core needle biopsy can be used to acquire a histopathology sample which allows preservation of tissue architecture and cellularity of the lesion and may lead to a more definitive diagnosis. When EUS-guided FNA fails, consider bite-on-bite deeper biopsies using jumbo forceps. Endoscopic submucosal resection (ESMR) can also be used to obtain larger and deeper tissue sample with higher diagnostic yield [8]. In addition to histology, immunohistochemistry (IHC) constitutes an extra layer of diagnostic tool to confirm the diagnosis by differentiating neurofibroma from other types of SMTs [1,4,5]. S-100 immunostaining is particularly useful in this regard.

Since most patients were symptomatic on presentation, management has been predominantly surgical [9]. This is because most of the symptoms fail to resolve with non-invasive measures such as dilation, as exemplified by the patient presented in this study who failed serial pneumatic dilations. In the case reported by Hisikawa et al., no surgical treatment was pursued, and the patient was doing well at follow-up [9]. The patient had a histologically confirmed diagnosis via needle biopsy. This suggests that asymptomatic cases can be safely observed. However, the potential for malignant transformation is another driver for surgical intervention [5]. The lifetime risk of malignant transformation is estimated to be 5 percent [10]. The newly reported patient in this study had features which were concerning for malignancy (presence of enlarged lymph nodes and progressive increase in size of the tumor) and these expedited the decision to pursue surgical resection. Most of the cases identified were managed by enucleation. In instances where esophagectomy was employed, concerns for malignancy or difficulty in achieving limited resection were the major reason for radical resection.

This study is limited by the nature of the previous studies used in this review. The articles used were case reports and case series. These are level IV evidence according to the Oxford's levels of evidence [21]. In addition, some of the studies had incomplete data. Another limitation is the variability in documentation of the exact location of the tumor in the esophagus. This inconsistency may have affected the proportion of tumors reported as involving upper esophagus. Despite these challenges, this study has generated significant and relevant data about esophageal neurofibromas with the goal of facilitating better understanding which will translate to prompt diagnosis and appropriate treatment of this rare tumor.

While it is highly desirable to have a level I evidence like a randomized clinical trial to further investigate the management of esophageal neurofibroma, the rarity of cases will preclude this. However, a prospective study or an international, multi-institution/multicenter collaborative registry can also be established to better delineate management strategies for esophageal neurofibroma.

6. Conclusion

While there are more common causes of dysphagia, esophageal neurofibroma should be considered in the differential diagnoses when initial diagnostic work up reveals a subepithelial tumor. We reckon that accurate preoperative histologic diagnosis of esophageal neurofibroma by using the biopsy algorithm described above, in conjunction with immunohistochemical analysis, will favor less aggressive surgical treatment like enucleation, wedge resection and endoscopic submucosal resection for symptomatic patients. It may also promote surveillance of asymptomatic lesions. Minimally invasive approach to surgical resection is feasible and should be considered when the expertise is available.

Declaration of Competing Interest

The authors report no proprietary or commercial interest in any product mentioned or concept discussed in this article.

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Ethical approval

This case report is exempt from ethical approval in our institution.

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.

Registration of research studies

N/A.

Guarantor

Tolutope Oyasiji.

Provenance and peer review

Not commissioned, externally peer-reviewed.

CRediT authorship contribution statement

Sajjaad H. Samat: Data curation, Formal analysis, Methodology, Writing - original draft. **Chibueze Onyemkpa:** Data curation, Methodology, Formal analysis. **Mohammad Torabi:** Data curation, Formal analysis. **Tolutope Oyasiji:** Coceptualization, Data curation, Formal analysis, Methodology, Supervision, Writing - original draft, Writing - review & editing.

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