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# Surgical strategy in abnormally increased Fluorine-18 fluorodeoxyglucose uptake in an asymptomatic lower esophageal submucosal tumor – Report of a case

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## ABSTRACT

**INTRODUCTION:** Leiomyoma is the most common benign tumor of the esophagus (67–80%), it represents 0.4–1% of all esophageal tumors.

**PRESENTATION OF CASE:** An incidentally discovered gastro-esophageal submucosal tumor was found to have increased fluorine-18-fluorodeoxyglucose (FDG) uptake on positron emission computed tomography (PET/CT). After laparoscopic surgical exploration and local enucleation the tumor turned out to be a benign esophageal leiomyoma.

**DISCUSSION:** There are few reports of esophageal leiomyomas with a positive uptake on (PET/CT) and even fewer adopting our combination of a minimally invasive approach and frozen section examination as a management plan. Our approach avoided excessive morbid surgical resections and underestimation of a malignant disease.

**CONCLUSION:** We report this case hoping to expand the existing literature on the topic and to highlight the limitations of PET/CT in guiding the diagnosis and subsequently the management of esophageal submucosal tumors.

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## 1. Introduction

Leiomyoma is the most common benign tumor of the esophagus (67–80%); it represents 0.4–1% of all esophageal tumors.<sup>1</sup>

Its differential diagnosis should be established with gastrointestinal stromal tumors (GIST), and other benign tumors. The criteria to resect or not a leiomyoma is based on the symptoms it produces, especially dysphagia, and on the need to exclude malignancy. Positron emission computed tomography (PET/CT) using fluorine-18-fluorodeoxyglucose (FDG) provides information on tissue metabolic activity and can help in the differential diagnosis.

We report a case of esophageal submucosal tumor that showed a markedly increased FDG uptake on initial preoperative workup PET/CT. Based on this information we opted for a surgical minimally invasive exploration.

## 2. Presentation of case

A 54 year old man was admitted to another hospital for abdominal trauma following a motor vehicle accident. His past medical history includes hepatic steatosis secondary to excess alcohol intake but no history of tobacco smoking.

On admission a thoraco-abdominal CT showed several fractured ribs, an incidental gastro-esophageal tumor and a ruptured spleen for which the patient underwent a splenectomy via open midline laparotomy.

The tumor was described as a bulky partially calcified mass measuring 45 × 52 × 29 mm, poorly vascularized and seemingly extrinsic to the esophageal lumen coming in contact with the anterior pericardium (Fig. 1). Radiological diagnosis was that of a likely malignant process. Multiple celiac lymph nodes were also noted, the largest measuring 16 mm.

Following the patient's postoperative recovery, a follow-up CT showed the same unchanged celiac lymph nodes and an increase in size of 4 mm of the previously described tumor.

Further investigations included an upper endoscopy confirming the submucosal location of the tumor. An endoscopic ultrasonography (Fig. 2) disclosed a heterogeneous submucosal tumor with well-defined margins in the muscularis mucosa located at

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**Fig. 1.** Admission CT-scan showing the gastro-esophageal calcified mass (white arrow) along with the perisplenic collection (red arrow).

35–40cm from the dental arcade. This mass was hypoechoic with multiple hyperechoic foci inside of it. A differential diagnosis of degenerated GIST versus leiomyoma/leiomyosarcoma or post-traumatic hematoma was made. No abnormal lymph nodes were detected. After undergoing the above mentioned investigations namely the endoscopic ultrasound and both abdomino-pelvic tomodensitometries the patient was referred to our institute for further management.

A PET/CT scan (Fig. 3) showed an abnormally increased FDG uptake (SUV = 6.1) in the tumor with no other metabolically active lesion detected. Both the elevated SUV and the tomodensitometric appearance were more in favor of a malignant process. A Fine Needle Aspiration (FNA) was performed after the positive PET/CT result to rule out a malignancy and was inconclusive.



**Fig. 2.** Endoscopic ultrasound showing the submucosal tumor with well-defined margins in the muscularis mucosa (black arrow).

To avoid both, missing a malignant disease and unnecessary large surgical procedures, we opted for a minimally invasive surgical exploration guided by frozen section. The different resection possibilities, tumorectomy or partial esogastrectomy with Merendino reconstruction or radical esophagogastrectomy, were comprehensively explained to the patient.

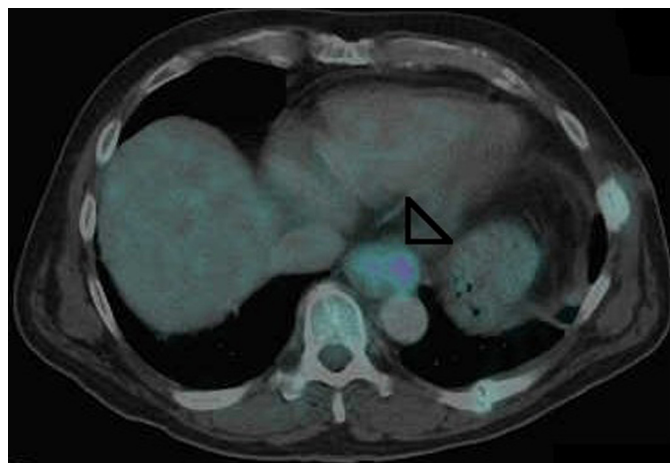
The operation was started with the patient in the supine position. Laparoscopic exploration of the abdominal cavity showed no gross anomalies except the adhesions from the previous splenectomy.

Liberation of the abdominal esophagus and progressive dissection of the thoracic esophagus revealed a multi-lobulated lesion adherent to the esophagus. This tumor was progressively dissected from the esophageal muscular layers under direct endoscopic control.

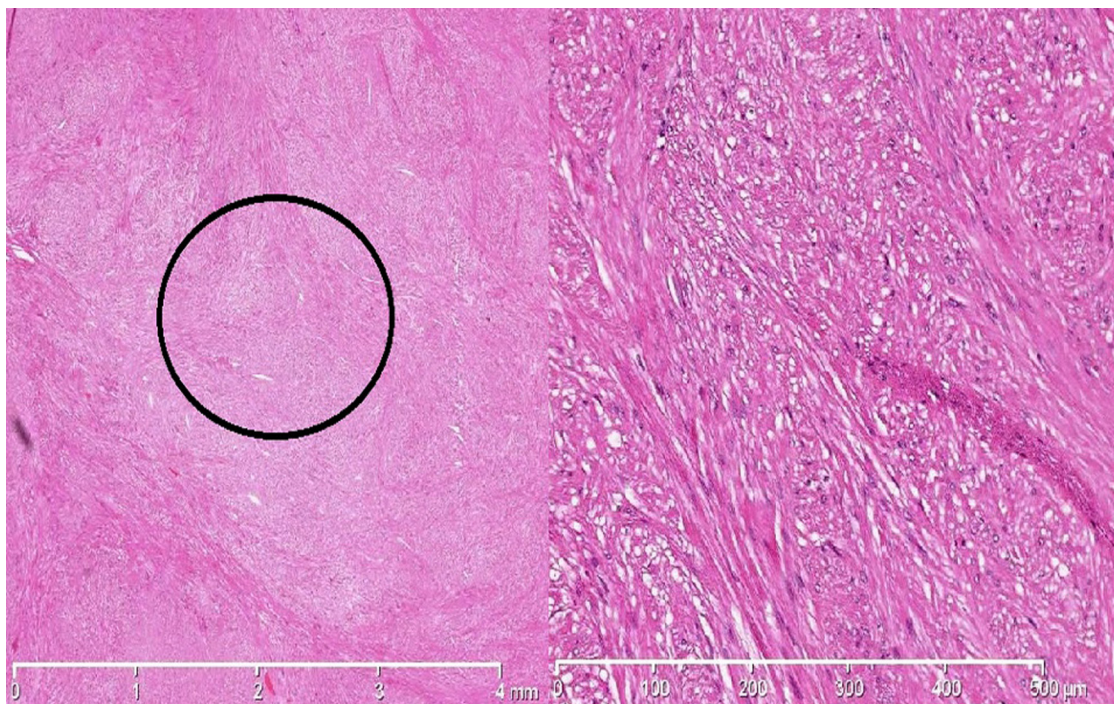
The mass was then extracted in an endo-bag via a small mid-line laparotomy using the superior part of the existing splenectomy incision, intraoperative histo-pathological examination revealed a benign esophageal leiomyoma. A Nissen fundoplication was performed to cover the esophageal sub-mucosa.

The postoperative course was uneventful with a normal gastrografin swallow done at day 4.

Final histo-pathological result showed macroscopically a 5 × 3 × 5 × 3 cm white homogenous mass with a lobulated aspect. Microscopically, the resected specimen showed intercrossing



**Fig. 3.** FDG-F-18 PET/CT scan showed an abnormally increased FDG uptake (arrow-head) at the level of the lesion described above.

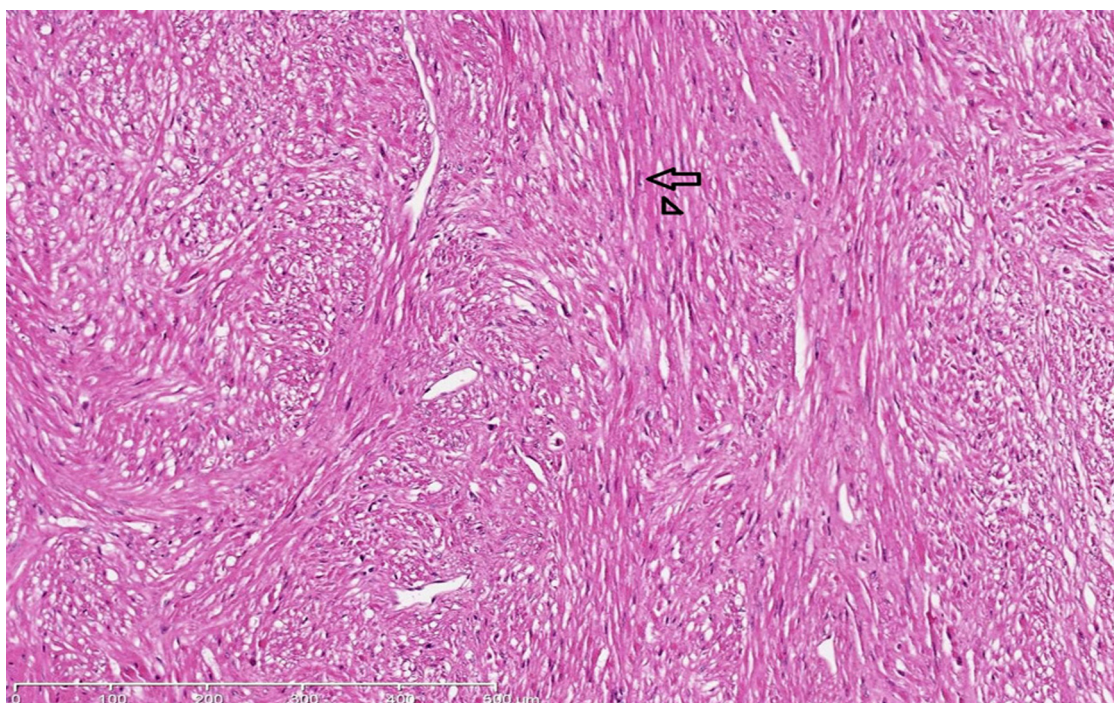


**Fig. 4.** Pathology slides showing nodular hyperplasia formed by the intercrossing smooth muscle cells (black circle).

smooth muscle fibers with no nuclear anomalies and minimal inflammatory focal infiltration; findings characteristic of a leiomyoma (Figs. 4 and 5). Immunohistochemical examination also demonstrated negative reactivity for CD117 (c-kit proto-oncogene product), CD34, MIB-1, and glucose transporter-1 (GLUT-1) along with the absence of mutation in c-kit or platelet-derived growth factor receptor, allowing us to rule out a gastrointestinal stromal tumor.

### 3. Discussion

Esophageal leiomyoma is a rare finding (0.4–1% of all esophageal tumors).<sup>1</sup> Endoscopy typically reveals a submucosal mass with intact mucosa.<sup>2</sup> Preoperative diagnosis of leiomyoma relies on many imaging techniques such as endoscopic ultrasonography that shows a homogeneous and hypoechoic lesion with clear margin.<sup>3–5</sup> CT scan ideally completes the preoperative evaluation of



**Fig. 5.** Magnified view of Fig. 4 showing ovoid nucleus (arrow), and eosinophilic cytoplasm (arrowhead).

**Table 1**  
Cases of esophageal leiomyoma with positive FDG PET/CT uptake, the surgical approach; mini-invasive vs open was also precised.

Cases	Positive PET/CT uptake	Mini-invasive approach	Open approach	Number of cases
Depypere et al. <sup>14</sup>	Yes		YES	2
Miyoshi et al. <sup>15</sup>	Yes	YES		1
Meirelles et al. <sup>16</sup>	Yes		YES	1
Lee et al. <sup>17</sup>	Yes		YES	1
Our case	Yes	YES		1

the patients, revealing in most cases a mass originating from esophagus without lymphadenopathies. In our case, CT-scan showed enlarged celiac lymph nodes.

Preoperative biopsy of the tumor is a debating issue. Some argue that it can render enucleation more difficult also FNA entails the risk of esophageal leak or fistula with a risk of potential mediastinitis.<sup>6–8</sup>

Positron emission tomography (PET) with a labeled glucose analog, F18-fluorodeoxyglucose (FDG) is a valuable diagnostic tool that is widely used to survey malignant and inflammatory disease.<sup>9</sup> FDG-PET is an imaging modality for abnormality of function or metabolism in tissue and organs, which usually precedes anatomic change. FDG accumulation has already been described in cases of uterine leiomyomas<sup>10</sup> and in patients with esophageal and genital leiomyomatosis.<sup>11</sup>

Abnormal FDG accumulation in the esophagus can be explained by many non-oncological inflammatory conditions such as: gastro-esophageal reflux, Barrett esophagus, acanthosis and infection.<sup>12</sup> It would be wise to correlate an incidental FDG uptake with CT-scan findings in the gastro-esophageal junction.<sup>13</sup>

To our knowledge, there have been 4 reports (5 cases) of positive FDG-PET in the case of isolated benign lower esophageal leiomyoma.<sup>14–17</sup> Kentaroh Miyoshi's reported case<sup>15</sup> was enucleated through thoracoscopy and a mini-thoracotomy. The 2 cases reported at Leuven<sup>14</sup> were excised via a left thoracotomy [Table 1].

About 0.2% of leiomyomas show a malignant transformation to leiomyosarcoma.<sup>18</sup> The management of asymptomatic esophageal leiomyomas is non-surgical. However in our case, the suspicion of malignancy following chest CT findings and the FDG uptake on the PET scanner imposed a surgical option. With the finding of esophageal leiomyoma on frozen section exam, laparoscopic enucleation was the most reasonable and least morbid option in our case. The minimally invasive approach versus the open one offers considerable advantages including reduced pain, better respiratory function, shorter hospital stay, faster return to regular activities, and esthetic of the surgical wound.<sup>19,20</sup>

A high index of suspicion is demanded when dealing with positive FDG-PET/CT of the lower esophagus. One should always take into account the macroscopic aspect as well as the results

of the various imaging techniques available such as endoscopy, ultrasonography, CT or MRI, laparoscopy and frozen section examination.

**4. Conclusion**

When dealing with an asymptomatic submucosal esophageal lesion with an abnormally increased uptake on PET/CTFDG scanner, this paper indicates that integrating all results of different imaging techniques does not allow an accurate diagnosis and an exploratory minimally invasive approach combined with a frozen section examination can potentially avoid excessive morbid surgical resections and particularly an underestimation of a malignant disease.

**Conflict of interest**

The authors declare that there is no conflict of interest.

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None

**Ethical approval**

“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”

**Author contributions**

Johnny Haddad is the main author. Fikri Bouazza helped in patient contact. Hassan Baraké helped in article layout. Gabriel Lib-erale did the literature review. Patrick Flamen provided expertise in the field of nuclear medicine, and Issam El Nakadi also helped in patient contact and correction of the manuscript.

**Key learning points**

- Minimal invasive approach combined with a frozen section can avoid excessive surgical resection and underestimation of malignant disease in positive PET/CTFDG submucosal esophageal lesion.

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