



# **Intramural Esophageal Bronchogenic Cyst**

## **Mimicking Malignity and Support of PET-CT in The Diagnosis**

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### **Abstract**

Bronchogenic cysts are congenital lesions caused by the abnormal budding of the trachea-bronchial tree in the embryogenic period. Intramural esophageal bronchogenic cysts occur by the abnormal budding seen between mucosa and muscle layers of the esophagus which is an extremely rare localization. Intramural esophageal bronchogenic cysts is asymptomatic until it reaches to a large size. A complicated intramural esophageal bronchogenic cysts can mimic malignity as in our case. The treatment is surgical resection.

**Key Words:** Esophagus, bronchogenic cysts, intramural, PET-CT.

**Maligniteyi Taklit Eden Özofagusun İntramural Bronkojenik Kisti ve Tanida PET-CT'nin Desteği**

### **Özet**

Bronkojenik Kist'ler; embriyojenik hayatı trakea-bronşial ağaçta olan anormal tomurcuklanma sonucu ortaya çıkan kongenital lezyonlardır. Özofagus'un İntramural Bronkojenik Kistleri, bu anormal tomurcuklanmanın, özofagus mukoza ve kas tabakası arasında olması sonucu, meydana gelen oldukça nadir bir lokalizasyondur. Özofagusun İntramural Bronkojenik Kistleri büyük boyutlara ulaşınca kadar asyptomatik kalırlar. Komplike olmuş intramural özofageal bronkojenik kistleri, bizim vakamızda olduğu gibi maligniteyi taklit edebilir. Tedavisi cerrahidir.

**Anahtar kelimeler:** Özofagus, bronkojenik kist, intramural, PET-CT

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## INTRODUCTION

Bronchogenic Cysts (BC) are congenital lesions resulting from the abnormal development of the foregut and tracheobronchial tree. Tracheobronchial tree develops from the ventral part of the foregut, and these cysts occur as a result of the abnormal budding of tracheobronchial tree (1). BC are generally mediastinal and intraparenchymal localized, but they may be seen anywhere with development of abnormal budding of tracheobronchial tree during the fetal development. One of the extremely rare localization of BC is between the mucosa and muscle layer of the esophagus which is called the intramural esophageal bronchogenic cyst (IEBC) (1,2). IEBC don't present symptoms, unless they reach to a large size. It is not recognized on the chest X-ray radiographies, unless they reach a large size since the trace of the esophagus is behind the heart. In this article, we present a case of the IEBC in our region in which esophagus carcinoma is endemic, which was radiologically mimicking malignity and has been only few published reports in the English literature and the facilitating support of PET-CT in this case.

## CASE

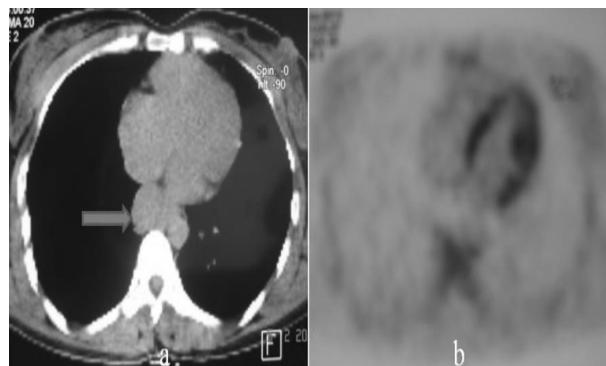
On the endoscopy performed on a 32-years-old female patient due to her dyspeptic complaints, external compression findings were defined at 34 cm of the esophagus. On the endoscopic evaluation, mucosa was seen to be intact in this localization. Chest X-ray graphy was normal and a mass lesion image which is 3,4x2,9x3,1 cm in diameter was monitored at the 1/3 distal of the esophagus (Figure-1a). We routinely use PET-CT for preoperative evaluation of the esophagus malignity in our region in which esophagus carcinoma is endemic. In this case also on PET-CT evaluation carried out to rule out the malignity, lesion was seen not to involve FDG (Figure-1b). With these findings, surgery was planned in the case accepted as the esophagus benign lesion, the lesion was reached with a right postero-lateral thoracotomy and when esophageal longitudinal muscle plane was passed, the lesion was seen to be an intramural localization cystic lesion localized between the mucosa and muscle layers of the esophagus. When the cyst wall was opened, a yellowish and mucoid fluid was aspirated from it (Figure-2a). The cyst was not associated with the esophagus lumen (Figure-2b). The cystic wall was

completely dissected from the esophageal mucosa and resected (Figure-2b,c). On histopathologic evaluation, it was reported as a bronchogenic cyst which respiratory type pseudostratified columnar epithelium and cartilage are apparent (Figure-2d).

## DISCUSSION

Laryngotracheal groove occurs in the embryogenic foregut in the end of the gestational 3rd week. Esophagus develops from the dorsal part of the foregut, while the respiratory system develops from its ventral part. Left and right lungs occur budding from the elongated trachea the end of the 4th week (3,4). Fetal esophagus and trachea are initially laid with ciliated epithelium, and the ciliated epithelium is replaced with squamous epithelium the gestational 17th week. BC are a development caused by the tracheobronchial budding in an abnormal localization in fetal, infant and until adulthood periods (1,3,4). These are often around the trachea-bronchial tree and mediastinal localized unilocular lesions. They are intrathoracically localized close to main bronchi, trachea, esophagus and pericardium. Furthermore, subdiaphragmatic, intrapericardial, intracardiac, cervical and subcutaneous localized bronchogenic cysts have been reported (1,4,6). One of the very rarely seen localization is intramural esophagus (2,7,10). On histopathological evaluation of BC, the definitive diagnosis is established by the inner wall laid with columnar epithelium and presence of cartilage tissue. Inside the cyst is filled with mucoid material (1,2,4).

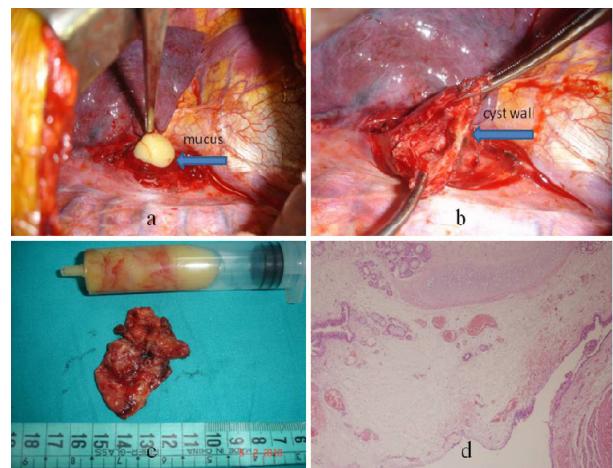
The most common symptoms in IEBC are dysphagia and chest pain, although different symptoms may occur according to the size of the cyst, the mediastinal organs compressed by the cyst and whether it is complicated. Dysphagia occurs only if the esophageal lumen is appreciably compressed by the cyst. Myocardial infarction may occur due to coronary artery compression, dyspnoea by respiratory compression and atelectasis by bronchial compression. While many of the cases may be asymptomatic, dyspepsia and heart burn may present related to the gastrointestinal system. In some cases, oral malodour and hematemesis may rarely occur following the spontaneous rupture of IEBC into the esophagus lumen. The cyst may be complicated, infected and bleeding into the cyst and ulceration in the cystic mucosa may be seen, this cases are symptomatic (1,5,7).



**Figure 1.** a) Image of the lesion mimicking malignity is monitored on the distal of Thorax CT of the case, b) Positive FDG involvement was not found on the PET-CT scans at the same level.

Unless they reach a large size, IEBC cannot be seen on chest X-ray. If the cysts reach a large size, they are seen as smooth contoured mass lesions in the mediastinum. Because content of the cyst is mostly infected, IEBC are smooth contoured and a high density on CT. So they mimic soft-tissue mass with this form (4,6). MRI and multislice CT reveal multiplanar imaging about the lesion. Air-fluid level can be seen if the mediastinal and intraparenchymal bronchogenic cysts are associated with trachea-bronchial tree; however, this is a very rare finding for IEBC, and these radiological findings can be encountered only in case of the rupture of the BC into the esophagus (6). There are some findings on the endoscopy in which the mucosa is intact and include external compression on the esophagus. Endoscopic ultrasound gives an idea about whether the lesion is cystic or solid. The definitive diagnosis is histopathologically established after the lesion is surgically removed. However, EUS-guided fine needle-aspiration or biopsy was emphasized in many studies to obtain a preoperative diagnosis (1,2,4,6). Nevertheless, we think like Annema et al. this method should be avoided because of the risk for mediastinitis occurrence (7).

PET-CT has been demonstrated to be effective in showing the primary tumour in esophageal malignity. It has proven to be an effective guiding preoperative examination for staging, detection of the distant metastases and selection of the operation candidates in the esophageal malignity. It is more sensitive in detection of the distant metastatic lymph nodes rather than proximal ones.



**Figure 2.** a) When the cystic wall was intraoperatively opened, yellowish mucoid fluid is seen discharging from it. b) After the boundaries of the cystic wall were demonstrated, it was seen not to be associated with esophagus mucosa, c) Post-operatively aspirated mucoid fluid and resected cyst wall, d) On the histopathological evaluation, it was reported as the bronchogenic cyst which respiratory type pseudostratified columnar epithelium and cartilage are apparent (H & E X100).

It is more sensitive specially in benign-malignant differentiation of the tumours larger than 1 cm as in our case (8). In our case, negative involvement was seen on PET-CT evaluation. The most common complications encountered in IEBC are infection, ulceration, intracystic bleeding and intraesophageal rupture. Although rarely, fistulisation into respiratory system and malignant degeneration may be seen (1,5,9). In addition, cardiac compression, atelectasis and vena cava superior syndrome are rare conditions emerging after the cyst reaches to a large size (1,2).

Treatment of the IEBC is surgery. The cystotomy and resection of the cystic wall is a general approach applied in the BC surgery via thoracotomy or thoracoscopy. But surgeon have to preserve the esophageal mucosa in this procedure (1,4,9). Endoscopic trans-esophageal needle aspiration and endoscopic mucosal resection are among the reported successful methods for IEBC (1,10). In addition, esophagectomy is one of the treatment options in the suspected malignity and cyst have occurred damages as not to allow primary repair in the esophageal mucosa (4,9).

In this case mimicking esophagus carcinoma, which is endemic in eastern of Turkey, esophagus mucosa was intact and there were external compression findings on the endoscopy. There was not FDG involvement on the PET-CT. Intraoperativ evaluation during surgical resection and ultimately histopathological diagnosis was reported as the intramural esophageal bronchogenic cyst.

Mucosa involvement is almost absolute in esophagus carcinoma. Complicated IEBC are mixed with esophagus carcinoma. In the cases with intact mucosa on the endoscopy, if absence of the FDG involvement is demonstrated with PET-CT, the lesion should be accepted as benign and the surgery should be planned without the need for diagnostic procedures with mediastinitis risk such as EUS-fine-needle aspiration or biopsy. Thoracoscopic resection should be preferred as it is less invasive.

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