An unusual case of malignant esophago-pulmonary fistula secondary to esophageal carcinoma

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ABSTRACT

Esophageal cancer is an aggressive cancer and has a poor survival rate. An esophago-pulmonary fistula is a rare complication of this cancer. This fistula causes pulmonary abscess formation and death in most patients. We are reporting a case of esophago-pulmonary fistula in a 55-year-old male patient who presented to us with complaints of fever and cough with expectoration. Chest radiographs initially showed right upper lobe consolidation which further progressed to cavitation and later on contrast-enhanced multi-detector computed tomography scan was used to diagnose this condition. An esophago-respiratory fistula is a rare complication of esophageal cancer, and esophago-pulmonary fistula is an even rarer subtype of this complication. It is essential to diagnose this condition early to avoid development of further complications and death.

Key words: Esophageal carcinoma, Esophago-pulmonary fistula, Esophago-respiratory fistula, Multi-detector computed tomography

E sophageal cancer is the eight most common cancer in the world with a poor prognosis and an increasing worldwide incidence [1]. Esophago-respiratory fistula is congenital or acquired communications between the respiratory tract and esophagus and an acquired fistula can be formed as a complication of this disease. They are further subdivided into esophago-tracheal, esophago-bronchial, and esophago-pulmonary types most of the fistula, which occurs in this type of cancer are either of esophagotracheal or esophago-bronchial fistulae types, esophago-pulmonary fistulae are a less common complication of this cancer [2].

We present a case of esophago-pulmonary fistula secondary to esophageal carcinoma. These fistulae usually cause the formation of a pulmonary abscess. If these patients are left untreated, it leads to the progressive worsening of their condition and may result in death within a short period of time [3].

CASE REPORT

A male patient of age 55 years, a farmer by occupation came to the department of pulmonology with the chief complaints of a cough with yellowish expectoration, right-sided chest pain, and fever since 1 month. He was a heavy smoker and alcoholic since past 15 years.

Chest radiograph had been taken outside a week back, which showed a right-upper lobe consolidation and was treated with intravenous antibiotics, which showed no improvement in the patient's condition. Repeat chest radiograph showed a right upper lobe cavitatory lesion and an increase in consolidation. Blood reports showed total counts 19000 cells/mm³ with a predominance of neutrophils. All other blood investigations were within normal limits. Meanwhile, the patient was treated with higher antibiotics. Sputum acid-fast bacilli were negative. The sputum culture report showed growth of *Escherichia coli* which was sensitive only to tigecycline. As repeat chest radiograph showed no resolution, the patient was referred to the department of radiodiagnosis to undergo a contrast-enhanced multi-detector computed tomography (MDCT) scan of the thorax.

Contrast-enhanced MDCT scan of the thorax was performed on Siemens Somatom perspective 64 slice MDCT scanner using 50 ml of i.v contrast injected at 3 ml/s. The MDCT scan showed a large thick walled cavity with air-fluid levels and thick septations measuring 9 cm×10 cm×14.5 cm (APxTRxCC) involving the apical and posterior segments of the right upper lobe and superior segment of the right lower lobe (Fig. 1). The wall showed mild enhancement post-contrast.

Consolidation was seen in the rest of the right upper lobe and superior segment of the right lower lobe. A hypodense lesion was seen posterior to the trachea measuring $5.6 \text{ cm} \times 6 \text{ cm} \times 8.7 \text{ cm}$ (APxTRxCC) extending from the lower border of the thyroid to the superior mediastinum. The lesion was seen displacing the trachea and esophagus to the left. Post-contrast showed peripheral enhancement suggestive of a necrotic lymph node. Esophagus showed irregular wall thickening from first to sixth thoracic vertebral level (Fig. 2). There were multiple air pockets within the mediastinum suggestive pneumomediastinum (Fig. 3).

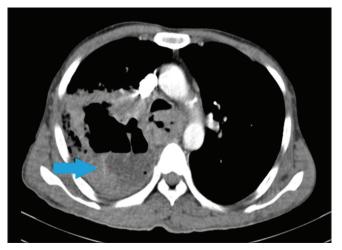


Figure 1: Axial contrast-enhanced (intravenous) multi-detector computed tomography scan image of thorax showing thick walled cavity with air-fluid level in the right upper lobe (arrow)



Figure 2: Sagittal contrast (oral) multi-detector computed tomography scan image of thorax showing irregular filling of the upper thoracic esophagus secondary to irregular wall thickening (arrow)

The patient was given oral contrast to assess the esophagus and a repeat MDCT scan was done. It showed irregular thickening of the esophagus for a length of 8.3 cm (first to sixth thoracic vertebral level) with a maximum anterior wall thickness of 13 mm and irregular filling of the lumen with oral contrast. Extravasation of contrast was noted into multiple tracts in the mediastinum. The contrast was seen passing into the lung cavity with pooling of contrast within the abscess, suggestive of esophago-pulmonary fistula (Fig. 4). There was no contrast within the trachea-bronchial tree. Endoscopy was performed and it detected a mass lesion in the upper thoracic esophagus with fistula. A biopsy was done, which showed a poorly differentiated carcinoma of the esophagus. The patient was referred to the higher center for further management of the esophageal carcinoma and stent placement.

DISCUSSION

Esophago-respiratory fistulae are congenital or acquired communications between the respiratory tract and esophagus.



Figure 3: Coronal contrast (oral) multi-detector computed tomography scan image of thorax showing irregular filling of the upper thoracic esophagus with multiple air pockets in the mediastinum suggestive of pneumomediastinum (arrow)



Figure 4: (a and b) Axial and coronal contrast (oral) multi-detector computed tomography scan images of thorax showing extravasation of oral contrast into the lung cavity (arrows), suggestive of esophagopulmonary fistula

Acquired esophago-respiratory fistulae are usually rare and can arise due to malignancy, prolonged intubation, corrosive ingestion, granulomatous mediastinal infections, previous surgery on the esophagus or trachea-bronchial tree, or trauma [4]. They are further subdivided into esophago-tracheal, esophago-bronchial, and esophago-pulmonary types. Esophago-tracheal fistula is seen in 52-57% of the patients with esophago-respiratory fistula and is the most common subtype. Esophago-bronchial fistula is seen in 37-40% of the patients while esophago-pulmonary is the least common [3]. Esophago-pulmonary fistula is rarely reported and accounts for only 3-11% of all esophago-respiratory fistulae [3]. Malignant esophagorespiratory fistula is seen in 4.9% of the patients with esophageal carcinoma [5]. Esophageal cancer is the most common malignancy reported to cause an esophago-pulmonary fistula. Other tumors such as lung, tracheal, or primary metastatic lymphoma are also known to cause this condition [4]. The previous radiotherapy has also lead to the formation of such a fistula.

Esophago-respiratory fistula needs to be treated rapidly. They can progress to multiple fistulae formation, pneumonia, and lung abscess formation. If they are left untreated, it may lead to pulmonary sepsis and acute respiratory distress syndrome secondary to chronic aspiration [6]. This can cause the death of the patient. Hence, early diagnosis and treatment are of utmost importance in the prognosis of the patients with this condition. Various modalities used to diagnose esophago-pulmonary fistula include contrast radiography, contrast-enhanced MDCT scan, esophagoscopy, and bronchoscopy. Contrast-enhanced MDCT thorax with oral and intravenous contrast is an excellent modality to diagnose this condition. It provides excellent anatomical detail and can assess the trachea-bronchial tree as well as the lung parenchyma very well. The flow of the oral contrast from the esophagus into the lung parenchyma can be demonstrated. It can also demonstrate the level and extent of the esophageal carcinoma and associated lymph nodal or lung metastasis. MDCT also helps in multi-planar reconstruction of the MDCT images which can better assess this condition. It also employs volume rendering techniques for three-dimensional image reconstruction which is helpful in interventions [4].

Esophago-respiratory fistulas are treated by palliative methods including stenting, bypass, or exclusion. Surgical repair with excision of the fistulous tract may also be done. The main aims of treatment are to prevent the development of complications such as pulmonary sepsis, to prolong the patient survival and to alleviate patient discomfort. The patient can also undergo radiotherapy or chemotherapy. Median survival rate in an untreated patient may only be 1-6 weeks [7]. Endoscopic esophageal stenting using coated self-expanding metal stents (SEMS) is currently the most widely used palliative treatment modality [8]. A covered SEMS may be a palliative option in these patients and helps to improve the patient's quality of life. In a study published in 2009, of the 14 patients of esophago-pulmonary fistula who underwent palliative treatment with endoscopic stent placement all died within a mean observation period of 65 days [3]. If the patient is stable, surgical resection of the gangrenous lung parenchyma can be done.

Our case is unusual in that the fistula crossed anatomical boundaries in the superior mediastinum with the fistula communicating directly with a cavity in the right upper lobe. Carcinoma of the esophagus can show such flagrant disregard for anatomical boundaries. Our patient presented with respiratory complaints although the primary cause of the complaints was the esophageal carcinoma. The respiratory symptoms were due to the formation of the abscess as a complication of the esophagopulmonary fistula. Hence, it is imperative to investigative further such patients who are not responding to treatment. Our patient was referred to the higher center for further management of the esophageal carcinoma and stent placement.

CONCLUSION

Esophageal carcinoma is aggressive cancer and can lead to the formation of esophago-respiratory fistula as a complication. Early diagnosis and management are essential to prevent the development of complications such as lung abscess and pneumonia and to avoid the death of the patient. Contrast-enhanced MDCT scan plays an indispensable role in the diagnosis of this condition. Majority of the patients are managed by stenting to relieve the patient's symptoms and to prevent the development of further complications.

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