Esophagobronchial fistulae: Diagnosis by MDCT with oral contrast swallow examination of a benign and a malignant cause

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Abstract
We report two cases of esophagobronchial fistulae diagnosed by Multi-detector computed tomography (MDCT) oral contrast swallow examination. It is helpful to supplement the CT study with an oral contrast swallow as it aids in confirmation of a suspected fistula and also demonstrates the fistula tract better. We present the clinical details and the imaging findings on MDCT of two cases of esophagobronchial fistulae – one secondary to chronic chest tuberculosis and the other secondary to a squamous cell carcinoma of the upper esophagus – followed by discussion of the etiology, pathogenesis, and imaging of these fistulae.

Key words: Esophagobronchial; fistula, MDCT; oral contrast swallow

Introduction
Fistulae between the upper respiratory and gastrointestinal tracts are uncommon in adults. Whereas developmental anomaly is the commonest cause in infancy and childhood, the etiology in adults is most frequently secondary to an esophageal malignancy.\[1\]

We report two cases of esophagobronchial fistulae – one secondary to chronic chest tuberculosis and the other secondary to a squamous cell carcinoma of the upper esophagus diagnosed by Multi-detector computed tomography (MDCT). Traditionally, fluoroscopy with oral contrast swallow examination has been the mainstay radiological investigation for the diagnosis of these fistulae.\[2\] However, it can be an inconvenient study with need for multiple projections to adequately demonstrate the fistula and its course. Also, being a luminal study, the cause of the fistula is not adequately evaluated. CT, on the other hand, can detect wall thickening and abnormal enhancement.

It is helpful to supplement the CT study with an oral contrast swallow. Use of the various post-processing features like thick maximum intensity projections (MIPs) and volume rendering techniques (VRTs) enables better detection and depiction of these fistulae. Use of virtual endoscopy also guides the clinician for the conventional endoscopy which would be needed for biopsy and treatment.

Case Reports

Case 1
A 15-year-old male presented with cough, mucopurulent expectoration, and dyspnea since 3 months. There was history of exacerbation of cough after swallowing. This finding of swallow–cough sequence has been referred to
as Ono’s sign.[3] A chest radiograph revealed complete opacification of the right hemithorax with volume loss suggesting complete collapse of the right lung with mediastinal shift to the right [Figure 1]. A plain and intravenous contrast-enhanced CT study with oral contrast swallow was performed on a 128-slice MDCT scanner (Siemens Somatom Definition AS, Erlangen, Germany). The study revealed complete collapse of the right lung with irregular dilated ectatic bronchi in the right lower lobe [Figure 2]. There was stenosis and diffuse narrowing of the right mainstem bronchus with nodularity of the mucosa seen best on the virtual bronchoscopy [Figure 3]. The right main pulmonary artery (MPA) was narrow in caliber. There was a resultant significant shift of the mediastinum and heart to the right. An air-filled tract was noted extending from one of the right lower lobar bronchi toward the posterior mediastinum with ill-defined soft tissue around it. The possibility of an esophagobronchial fistula was suspected, and we did a CT oral contrast swallow study to detect it. Oral contrast swallow study performed with the patient in right decubitus position using diluted non-ionic iodinated contrast medium (1:20 dilution of iohexol with normal saline). It depicted the site and the fistula tract between the right lateral wall of the esophagus and one of the ectatic bronchi in the right lower lobe of the lung. Thick MIP images and VRT processing demonstrated the fistula site and tract [Figures 4 and 5]. Right adrenal gland calcification was also noted. These imaging findings led us to conclude that these changes were most likely the sequelae of chronic tuberculosis. The patient underwent an open right pneumonectomy with repair of the fistula. Histology of the lung specimen revealed distorted bronchioles with diffuse and focal dense infiltration by mononuclear cells
and giant cells. Lymph nodes with prominent germinal centers were also seen.

**Case 2**

A 65-year-old male presented with progressive dysphagia to solids over the past 2 months. An ultrasound of the neck revealed few enlarged lymph nodes which on fine needle aspiration (FNA) revealed neoplastic squamous cells. A plain and intravenous contrast-enhanced CT study with CT oral contrast swallow was performed. There was moderate irregular enhancing wall thickening of the upper and mid thoracic esophagus. There was a loss of fat plane between the esophagus and the carina, left mainstem bronchus, and aorta. A defect was seen in the posterior wall of the proximal left mainstem bronchus suggesting formation of a fistula with the esophagus [Figure 6]. A CT oral contrast swallow study performed in prone position with diluted non-ionic contrast (1:20 dilution of iohexol in normal saline) revealed a small fistula tract between the esophagus and the proximal left mainstem bronchus just distal to the carina [Figure 7]. Post-processing of this contrast swallow study with volume rendering depicted the fistulous communication in 3D [Figure 8].

**Discussion**

Esophagobronchial fistulae are uncommon and difficult to diagnose. In the elderly, they are most frequently seen with an intrathoracic malignancy and are most commonly associated with malignancy of the esophagus. In a large case series, esophagorespiratory fistula formation was seen in 12.5% of cases of esophageal malignancies. The malignant tissue spreads to involve the tracheal or bronchial wall. Subsequent ulceration and necrosis of the malignant tissue leads to tissue breakdown and fistula formation. Fistulas may also occur secondary to radiotherapy.

Non-malignant causes are infrequent and include trauma (blunt, penetrating, or iatrogenic), chronic inflammation (chronic infections like tuberculosis and
histoplasmosis, Crohn's disease,[2,24] late presentation of a congenital fistula,[2,25] and rarely poisoning.[2,26] Over the years, iatrogenic trauma has become a commoner cause for such fistulae compared to infections.[9] Tuberculosis as a cause for these fistulae has been infrequently reported in the western literature.[10,11] However, in the Indian subcontinent where the incidence of tuberculosis is high,[12] chest tuberculosis should be high in the list of differentials when a non-malignant fistula is encountered. In nonmalignant conditions, traumatic bronchial or tracheal wall necrosis or necrotizing inflammation is responsible for the fistulization.[13]

Fluoroscopic oral contrast swallow examination with barium is the initial investigation of choice for evaluation of dysphagia and suspected fistulae, even though endoscopy is needed for definite evaluation.[3,14,15] If esophageal perforation is suspected, an iodinated contrast medium should be used as barium extravasation can lead to mediastinitis. If there is no frank leak seen, then barium can be given as it produces better radiographic quality being of higher density than iodine. If respiratory fistula is suspected, barium still may be used as small quantity of barium in the tracheobronchial tree is harmless. However, iodine contrast medium should not be used as it can cause chemical pneumonitis and only non-ionic iodinated contrast medium should be used in such cases.[14] The best initial agent to use in both these scenarios of perforation or fistula is a non-ionic iodinated contrast agent. Fluoroscopy allows for dynamic evaluation of esophageal motility as well as evaluation of its lumen. Even though barium swallow fluoroscopy examination has the advantage of being a real-time study, fistula tracts may not always be detected.[17-19] If detected, depiction of the three-dimensional course of the fistula may be difficult in spite of use of multiple projections. Also, being a luminal study, it would fail to show changes in the wall of the esophagus and in the mediastinum that are shown accurately by CT.

CT has a proven role in the diagnosis of neonatal tracheobronchial fistulae.[20] No large case series has been published about its efficacy in adult fistulae, but few case reports have highlighted its role.[21-23] A routine CT study could miss these fistulae if the fistula tract is collapsed. This drawback can be overcome by performing a CT oral contrast swallow study. A diluted preparation of a non-ionic iodinated contrast agent should be used. The patient should be given a mouthful bolus of the preparation and asked to swallow it promptly on instruction to do so. Since the oral and pharyngeal phases of deglutition take no more than 2 seconds, the acquisition can be triggered immediately after the instruction to the patient to swallow has been given. The patient position may be changed to better opacify the fistula tract as we used prone and right decubitus positions in our cases to better opacify the fistula tracts. A recent study demonstrated that CT contrast swallow was better tolerated and more sensitive than fluoroscopy at detecting post-esophagectomy anastomotic leaks.[2,26]

Post-processing these studies with maximum-intensity projection and with volume rendering allows a three-dimensional evaluation of the fistula tract.

Conclusion

Supplementing oral contrast swallow to the chest CT protocol in cases where fistulae are suspected can improve the diagnostic ability of CT and also better demonstrate these fistulae. Virtual endoluminal images also guide the endoscopists while performing biopsies or therapeutic interventions.

References

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