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TRACHEAL MALIGNANCY PRESENTING AS TRACHEO OESOPHAGEAL FISTULA

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ABSTRACT

Malignant tracheo oesophageal fistula though rare, occurs most commonly in association with oesophageal malignancy. Incidence of primary tracheal malignant lesions are extremely rare. Two most common tracheal malignancies known are squamous cell carcinoma involving the lower trachea and adenoid cystic carcinoma involving the upper trachea. Tracheo oesophageal fistulous communication has been described as one of the uncommon complications of tracheal malignancy according to literature. Here we report a rare case of primary tracheal malignancy with invasion of oesophagus causing tracheo oesophageal fistula.

KEYWORDS: Primary tracheal malignancy, acquired tracheo oesophageal fistula, squamous cell carcinoma.

INTRODUCTION

Tracheo oesophageal fistulas that present in adulthood are usually due to malignancy or iatrogenic.[1]

Primary malignant tumours of the trachea are rare, representing only 0.1% to 0.4% of all malignant disease. Squamous cell carcinoma (SCC) and adenoid cystic carcinoma are the most common histological subtypes, making up approximately two-thirds of primary tracheal neoplasms.[2]

They usually present with respiratory symptoms depending on the level and degree of luminal obstruction. When associated with malignant trachea oesophageal fistula far more grave presentation results. “Ono’s sign” i.e. cough spells with swallowing efforts, with liquids and particulate food matter in sputum is a salient clinical finding. Patients also develop signs of chronic aspiration including cough, fever, halitosis, dysphagia, odynophagia, chest pain, and recurrent pneumonia.[3]

The etiopathogenesis of acquired trachea oesophageal fistulas can be broadly categorized into malignant and non-malignant. The pathogenesis of malignant oesophagorespiratory fistulas has historically been attributed to direct tumour invasion, tissue necrosis and erosion of tissue barrier between oesophagus and airway at varied levels. Cancer primarily arising in either membranous tracheal wall can invade into the oesophagus and vice versa, with the adjacent areolar tissue providing microenvironment for tumour growth. In addition a large oesophageal tumour mass can mechanically impinge on the thin membranous tracheal wall which is only 4 mm in average thickness. With time this leads to ulceration ad necrosis and eventual fistula formation in a devitalized tissue.[3]

Chest radiography is often the initial imaging study performed for patients presenting with respiratory symptoms. Tracheal and proximal bronchial air columns should be closely inspected in such cases. Studies have shown that chest radiograph identifies only 18%-28% of tracheal tumour.[4,5] Chest radiography can also sometimes show focal lobular or rounded opacity within the tracheal or bronchial air column.[6]

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.[7]

Barium oesophagogram plays an important role in the diagnosis of a tracheo oesophageal fistula under high clinical suspicion for adults. Esophagoscopy and bronchoscopy can demonstrate the fistula and can make provision for therapeutic interventional techniques, such as stenting.[8]

Multidetector CT with multiplanar reformations and volumetric rendering images are recommended for a precise pretherapeutic assessment of extent. They also provide increased resolution, and decreased motion artifact, has the potential to detect adult tracheoesophageal fistulas of any etiology. On, CT they appear as sessile, eccentrically located soft tissue mass, usually affecting posterior and lateral wall with mediastinal extension in 30-40%.[9]

Treatment of these fistulas is difficult, considering that most of these patients have an inoperable tumour at the time of diagnosis. Therefore, supportive and palliative treatment forms the basis of improving the quality of life. Direct surgical fistula closure, bypass or fistula resection do not yield good results, and endoscopic stenting or endoprosthesis are palliative options in this situation.[10]

Virtual PET/CT bronchoscopy has been widely employed to improve detection and characterization of airway neoplasms and early detection of recurrent disease following treatment.

CASE REPORT

A 45 year old female presented with history of dysphonia for 2 months, dysphagia, recurrent aspiration and cough with purulent sputum for 2 weeks duration with significant weight loss. Chest radiograph showed mild superior mediastinal widening and widened left paratracheal stripe [Fig.1]. Oesophagoscopy showed oedematous cricoarytenoid fold. No other mucosal abnormalities/extrinsic compression was seen in the cricopharynx. Contrast enhanced multidetector CT showed circumferential tracheal wall thickening with irregular projections into the tracheal lumen predominantly along the posterior and left lateral aspect [Fig.2] with an adjacent heterogeneously enhancing soft tissue density lesion in the trachea oesophageal groove suggestive of infiltrated lymph node [Fig.3] and communication with the upper thoracic oesophageal lumen at the level of D2 vertebra [Fig. 4a &4b]]. The diagnosis of a primary tracheal malignancy with an enlarged tracheooesophageal lymph node eroding into oesophageal lumen and formation of tracheo oesophageal fistula was made. Flexible bronchoscopy revealed multiple nodular growth arising from all walls of trachea from 3rd ring onwards, extending for about 5cm downwards [Fig. 5]. Biopsy was taken from the lesion. Histopathologic examination revealed a tumour composed of atypical squamous cells with enlarged nuclei with angulated contours and prominent nucleoli and increased mitosis; well formed keratin pearls were also seen confirmative of well differentiated squamous cell carcinoma.[Fig.6].

The patient underwent bronchoscopic stenting of tracheal lumen using Nitinol Self Expandable Metallic stent [Fig.7]. Immediate post procedural chest Xray showed stent insitu [Fig.8]. Patient was symptomatically better after 4 cycles of chemotherapy.

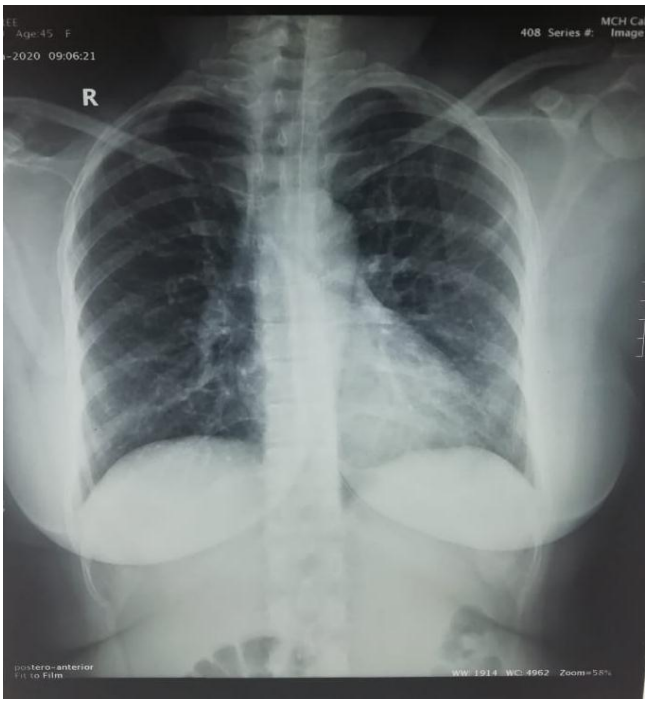

Fig. 1: Chest radiograph shows mild superior mediastinal widening and widened left paratracheal stripe.

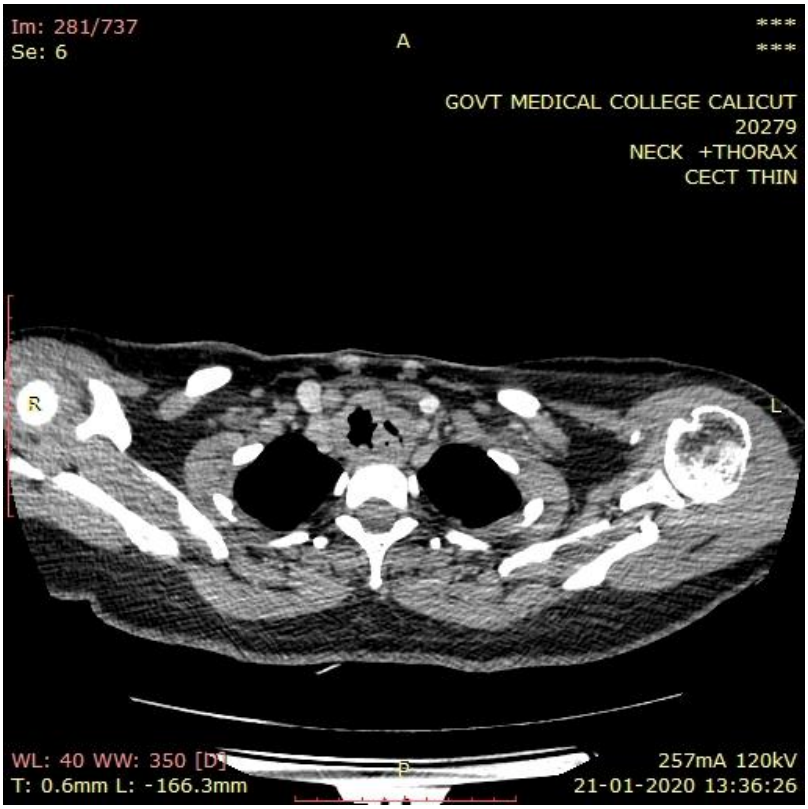

Fig. 2: CECT section showing circumferential tracheal wall thickening with irregular projections into lumen.

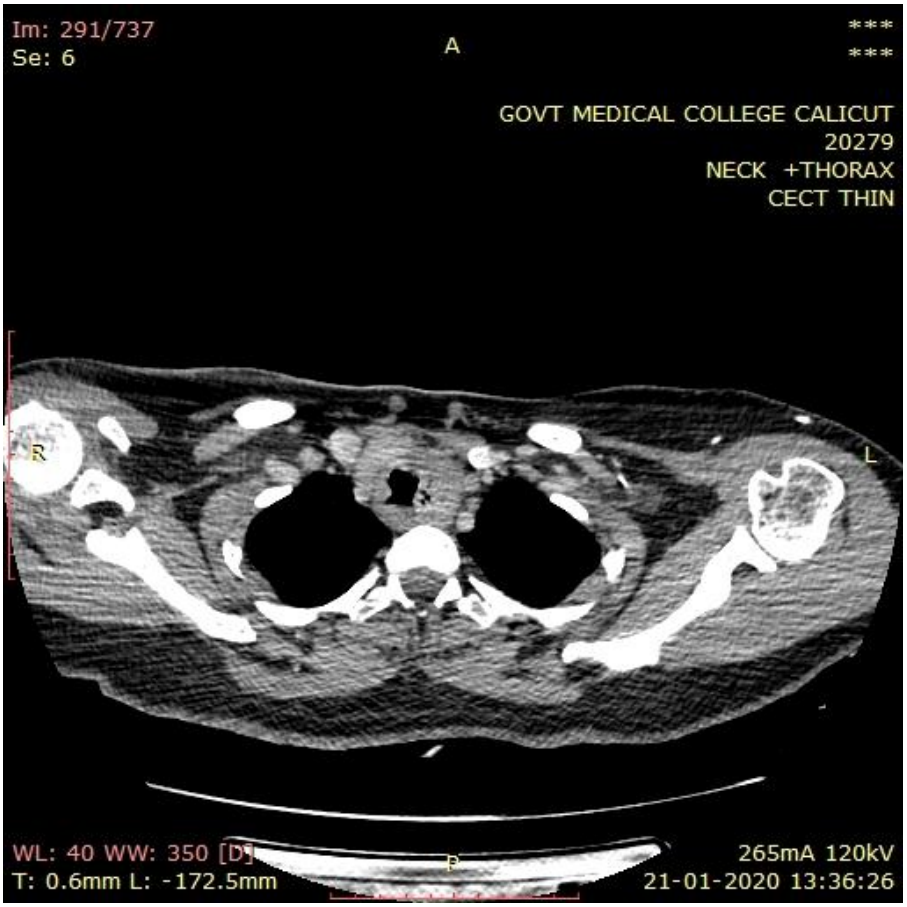

Fig. 3: CECT section showing trachea oesophageal groove lymph node.

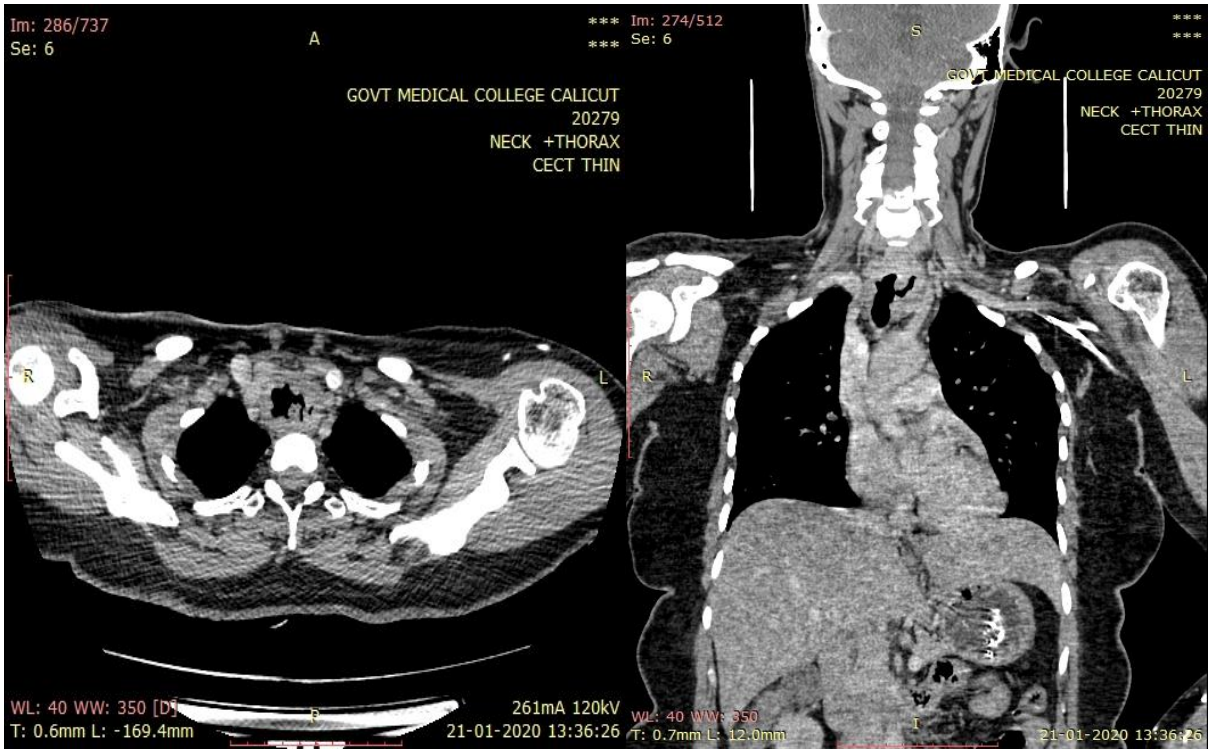

Fig. 4a & 4b. CECT axial and coronal section showing tracheo oesophageal fistula formation.

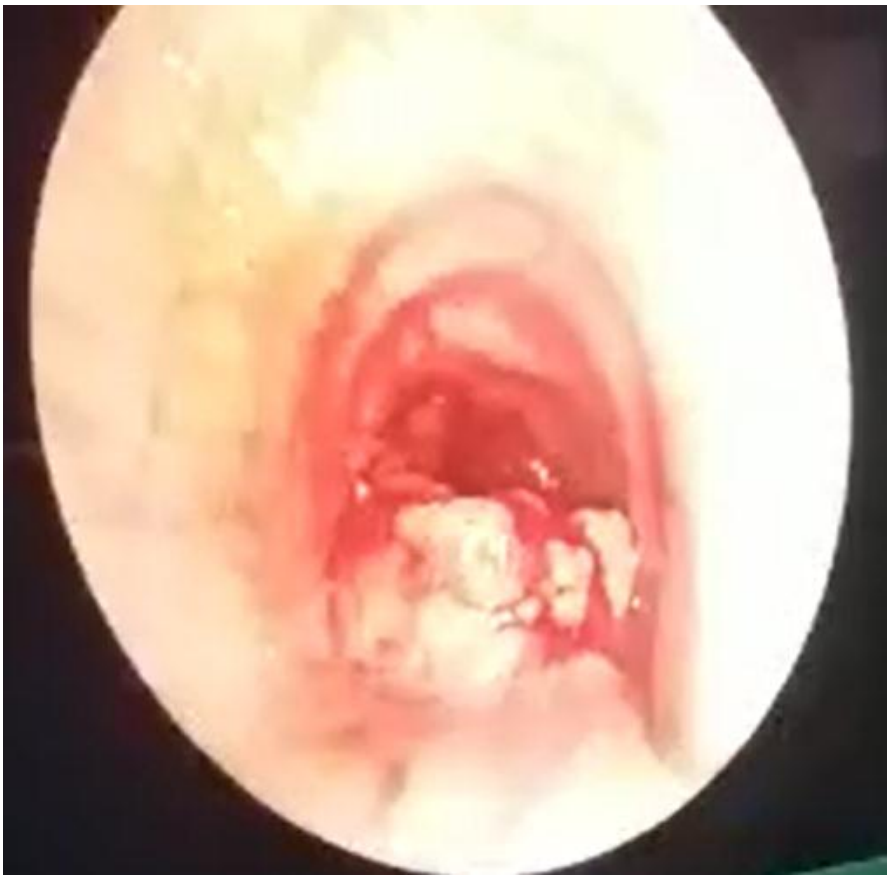

Fig. 5: Bronchoscopy showing the proliferative mass in the tracheal lumen.

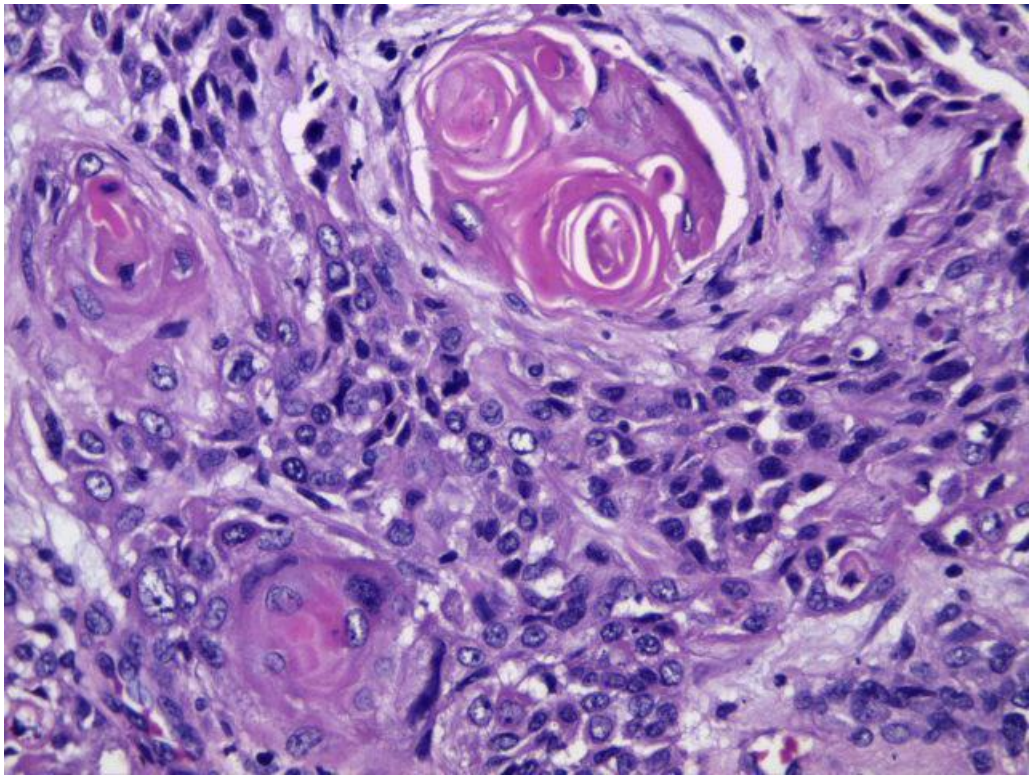

Fig. 6: Histopathologic examination showing tumour composed of atypical squamous cells with enlarged nuclei and prominent nucleoli and well formed keratin pearls suggestive of well differentiated squamous cell carcinoma.

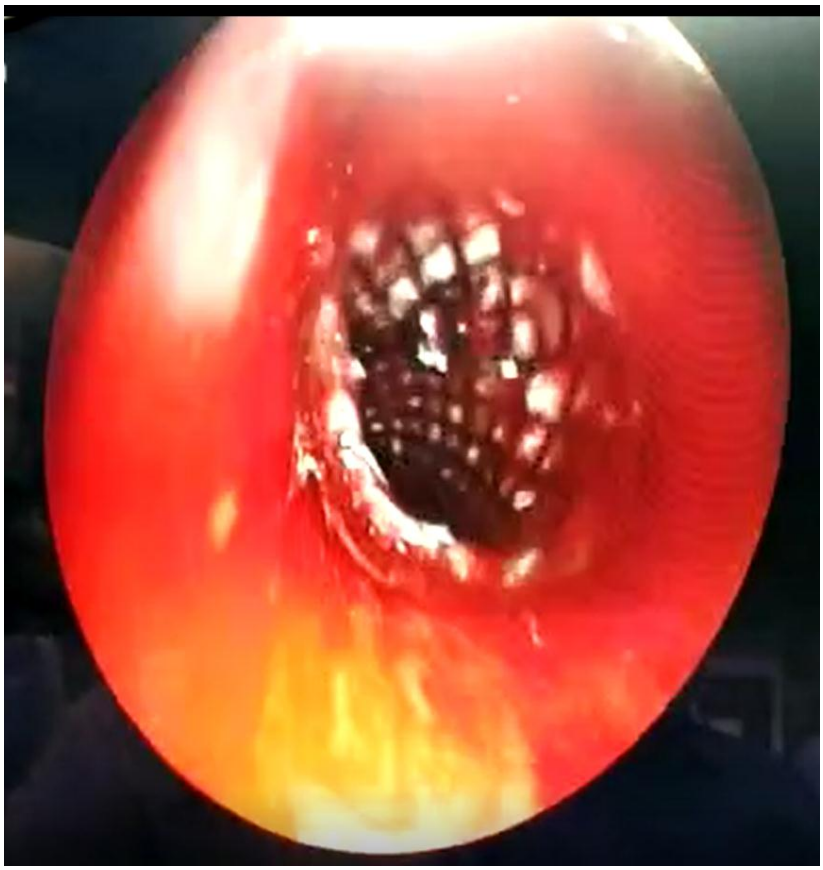

Fig. 7: Bronchoscopic image showing Nitinol stent insitu.

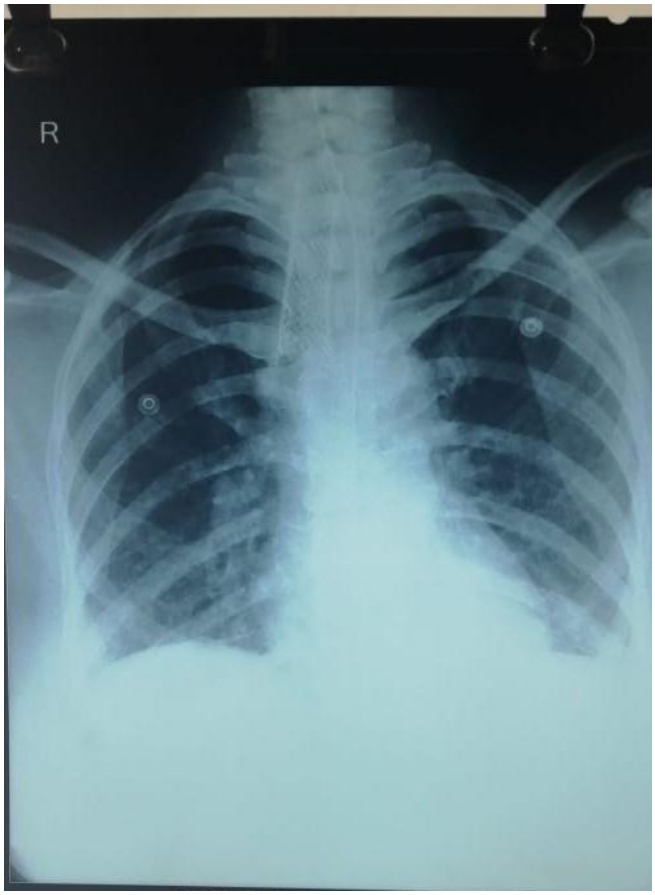

Fig. 8: Post procedure chest radiograph showing tracheal stent insitu.

DISCUSSION

This case presented here aroused interest because of the extreme rarity of a primary tracheal malignancy presenting with tracheo oesophageal fistula which has been described as one of its rare complications.

Most common cause of acquired trachea oesophageal fistula in adults is due to benign etiology. Malignant trachea oesophageal fistula is commonly seen in cases of oesophageal malignancy. Tracheal primary causing tracheo oesophageal fistula is rare. Patients usually present with dysphagia, cough with purulent sputum and symptoms of aspiration of feeds. Most common tracheal primary is squamous cell carcinoma followed by adenoid cystic carcinoma. Acquired tracheooesophageal fistulae can be diagnosed clinically by features such as cough which is brought about by swallowing but can be mild. So, majority of the patients present late in their course of disease when palliative procedures can only help.

Chest radiograph may show features of pneumonia secondary to aspiration and also enlarged lymph nodes if present. Mediastinal widening due to the tracheal primary may be seen as in our case, though mild. Barium swallow can reveal barium into the bronchopulmonary system, but is difficult most of the times as patient invariably develops symptoms of aspiration. Multidetector CECT can reveal tracheal wall thickening, its extent and surrounding changes including lymphadenopathy. Coronal reformatted CT is excellent for demonstrating tracheooesophageal fistula. Oral contrast given at the time of CT may show the presence of contrast within the tracheal lumen if fistula is present. Endoscopic visualisation of fistula in the oesophagus can be useful. Bronchoscopy is essential to demonstrate the exact site of fistulous communication and the interventionist can attempt palliative stenting.

CONCLUSION

Primary tracheal neoplasm can be associated with an acquired tracheoesophageal fistula in adults apart from carcinoma oesophagus. So a strong clinical suspicion with contrast CT is essential for early diagnosis and guiding therapeutic intervention.

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