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## CASE REPORT

# Resolution of asthmatic symptoms following successful endoscopic resection of tracheal mucoepidermoid carcinoma

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## SUMMARY

Mucoepidermoid carcinoma (MEC) is a rare tumour of the trachea accounting for up to 0.2% of reported primary lung malignancy. We report a case of a 54-year-old man, ex-smoker, whose presentation mimicked adult onset asthma with cough and wheezing, which did not respond to conventional treatment. He had occasional haemoptysis and weight loss in which CT scan performed for malignancy screening showed a protruding mass in the distal trachea causing endobronchial obstruction. Bronchoscopic intervention was performed to relieve the obstruction that resulted in resolution of asthmatic symptoms. Histological diagnosis confirmed MEC. This case emphasised the importance of a high index of suspicion in an unusual presentation of a common disease and the pivotal role of bronchoscopic intervention in malignant central airway obstruction.

## BACKGROUND

Mucoepidermoid carcinoma (MEC) is a rare tumour which originates from the mucus and serous glands of upper airway.<sup>1</sup> The incidence being reported as 0.1%–0.2% of primary lung cancer and accounted for 1%–5% of tracheobronchial adenoma.<sup>1</sup> Clinical presentation can be non-specific with cough, dyspnoea and wheeze, which could be misdiagnosed or mimics asthma as per our case presentation.

## CASE PRESENTATION

A 54-year-old man, an ex-smoker (stopped 10 years ago), was referred to a tertiary centre for poorly controlled asthma, which was diagnosed 2 months prior to presentation. Further history revealed he had chronic cough with occasional haemoptysis, wheezing and significant weight loss of 10 kg of 3-month duration. No family history of malignancy was found. Physical examinations were unremarkable apart from the presence of inspiratory rhonchi with prolonged expiratory phase.

## INVESTIGATIONS

Tuberculosis work-up was negative and chest X-ray (CXR) (figure 1) was normal. CT thorax (figure 2) was performed in view of red flag symptoms of malignancy, revealed a lobulated heterogeneous-enhancing soft tissue lesion at T3/T4 level measuring 1.2 × 1.1 × 1.3 cm with no clear plane from trachea

wall and no infiltration into the surrounding structures. In view of the CT findings, a flexible bronchoscopy was done and revealed a broad-based fungating mass obstructing 90% of the tracheal lumen; 8 cm from the vocal cord and 2 cm from main carina. No biopsy was attempted at this point. Spirometry with flow loop volume was initially planned but unfortunately was not performed in view of clinical deterioration.

## DIFFERENTIAL DIAGNOSIS

Primary tracheal carcinoma.

Primary/metastatic endobronchial carcinoma.

Tracheobronchial tuberculosis.

Tracheobronchial fungal infection.

## TREATMENT

Patient was admitted to ward for an elective rigid bronchoscopy. However, while in the ward, he developed worsening upper airway obstruction. Emergency rigid bronchoscopy was performed followed by electrocautery snaring to remove bulk of the tumour (figure 3A). Residual tumour was removed via rigid forceps (figure 3B) followed by application of argon plasma coagulation at the base of the lesion (figure 3C). However, complete excision of the tumour was unattainable.

Histological report revealed malignant cells of clear cell, intermediate cell, epidermoid cell and

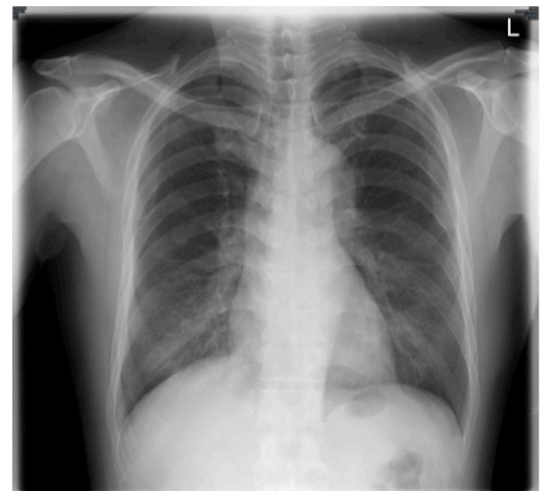


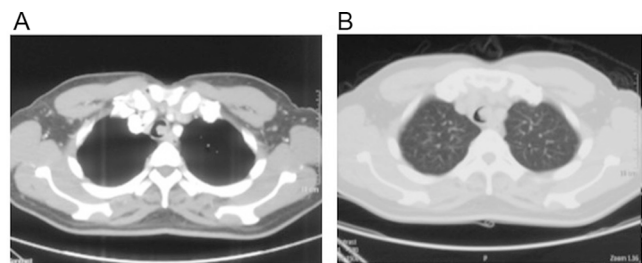
Figure 1 Chest X-ray showing normal findings.



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**Figure 2** CT thorax mediastinal window (A) and lung window (B) showing tracheal obstruction by a lobulated heterogeneous enhancing soft tissue lesion at T3/T4 level measuring 1.2×1.1×1.3 cm.

mucous cell type, arranged mainly in solid sheets and lobules with multicystic component. The cells are moderately pleomorphic with low proliferative index. A diagnosis of MEC of intermediate grade by Modified Healey grading was made (figure 4A,B).

### OUTCOME AND FOLLOW-UP

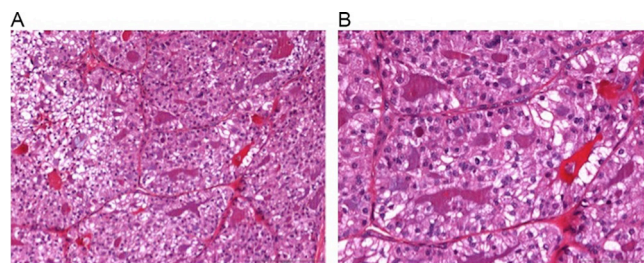
Post procedure, patient was admitted in intensive care unit and was extubated after 2 days. He had an uneventful recovery and was discharged after completion of 1 week of antibiotic for postobstructive pneumonia. The 'asthmatic' symptoms resolved following our intervention. Patient was subsequently referred to oncology due to incomplete excision during bronchoscopy for endobronchial brachytherapy; however, he refused treatment and subsequently lost to follow-up.

### DISCUSSION

Primary tracheal tumours are rare where in adults are overwhelmingly malignant as opposed to be benign nature in paediatric population.<sup>2</sup> MEC accounts for only 0.1%–0.2% of primary tracheal tumours and was first reported in 1952.<sup>1</sup> It is asymptomatic in 25% of patients as the tumour is small or located behind the carina and would only be incidentally found on CXR.<sup>3</sup> However, the presence of endobronchial exophytic mass may present with obstructive symptoms (cough, wheeze, stridor) and haemoptysis.<sup>3,4</sup> The presence of wheezing may lead to misdiagnosis of asthma as in our patient. Previous reported cases have shown that MEC can mimics asthmatic symptoms even in children and may also be misdiagnosed as invasive thyroid cancer.<sup>5,6</sup>

As in any patients with tracheal mass, spirometry with flow loop volume may show variable extrathoracic obstruction which may improve following intervention. Unfortunately in our patient spirometry was not feasible as his condition deteriorated rapidly needing emergency bronchoscopic intervention.

As for imaging, CT scan is a valuable tool in detecting endobronchial tumours as its detection on CXR can be challenging. It is also useful in evaluating the extent of tumour involvement beyond airway, regional lymphadenopathy and pulmonary



**Figure 4** Solid sheets of tumour with intervening thin fibrous septae forming lobules with multicystic component (A) using H&E stain (×100). The tumour cells are composed of clear, intermediate and mucous cell type (B) using H&E stain (×200).

nodules, which are important in determining stage and prognosis of disease.

The gold standard of diagnosing MEC would be bronchoscopy with adequate biopsy as preoperative misdiagnosis as squamous cell carcinoma, adenocarcinoma, bronchial adenoma or unknown mass was reported before.<sup>3</sup> Immunostaining with cytokeratin 7 (CK7) antibody and thyroid transcription factor 1 has been used to distinguish MEC with primary lung adenocarcinoma where CK7 is usually positive in the former.<sup>7</sup>

Prognosis of the disease depends on the grade of the tumour and possibility of complete resection.<sup>4</sup> In a low and intermediate grade tumour, a complete resection may not require subsequent adjuvant chemotherapy or radiotherapy.<sup>3</sup> However, a high-grade tumour carries an estimated 5-year survival of 31%.<sup>3</sup> Song *et al*, in a retrospective study on 32 patients with MEC, found significantly better survival in patients with histologically low-grade tumours than in patients with high-grade tumours.<sup>8</sup>

Currently, there are no randomised controlled trials in determining the most efficacious treatment modalities of tracheal tumour; however, surgical resection coupled with or without radiotherapy is considered the treatment of choice.<sup>8,9</sup> Surgical should be considered if the involved tracheal segment can be resected and reconstructed with primary anastomosis.<sup>10</sup> However, its role may be limited due to anatomical limitations, especially if located near the carina; metastatic disease or poor medical conditions and their contributions mainly revolved around palliative management.<sup>8,10</sup>

Bronchoscopic evaluation is the key step in the treatment of patient with malignant airway obstruction as it not only allows diagnostic confirmation and staging when considering surgery, but also provides significant palliative relief in unresectable lesion, relieve life-threatening obstruction and provide a pathway for other definitive intervention, for example, surgery, radiation or chemotherapy.<sup>8</sup> Reports by Hosakawa *et al* and El-Sameed *et al* demonstrate the effectiveness of bronchoscopic interventions in managing central obstruction due to tracheal tumour.<sup>11,12</sup>



**Figure 3** Bronchoscopic resection of the tumour using electrocautery snaring (A), removal of residual tumour with rigid forceps (B) and application of argon plasma coagulation at the base of the lesion (C).

In conclusion, MEC is a rare tumour of the trachea, which may present with obstructive airway symptoms mimicking asthma. This case highlights the importance of high index of suspicion coupled with diligent use of imaging and bronchoscopy that could prevent the delay in diagnosing this condition. Although surgical resection may provide definitive management, bronchoscopic intervention plays a pivotal role especially in providing diagnosis, staging, immediate relief and even cure in malignant central airway obstruction.

### Learning points

- ▶ Patients with asthmatic symptoms who failed to respond to medications warrants further investigations, for example, CT thorax or bronchoscopy to look for alternative diagnosis.
- ▶ Mucoepidermoid carcinoma is a rare tumour that may present with obstructive airway symptoms leading to misdiagnosis of asthma.
- ▶ Bronchoscopic intervention has a significant role in management of malignant airway obstruction.

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