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Case Series

Internal Medicine Section

Acute Upper Airway Obstruction as Atypical Manifestation of Uncommon Aetiologies- A Case Series

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ABSTRACT

Acute upper airway obstruction, an emergent situation, can sometimes put the clinician in a dilemma with no positive history or clinical findings to aid in the diagnosis. A myriad of conditions causes upper airway obstruction. Priority is to secure the airway and then seek out the underlying cause. Computed tomography scan is an important imaging modality for reaching a definitive diagnosis and might at times, reveal unexpected findings. In this case series, authors report three cases of acute upper airway obstruction which were atypical manifestation of diseases. Case 1 (82-year-old male) is about Lemierre's syndrome with compromised airway in the absence of abscess formation. Case 2 (47-year-old male) depicts an atypical presentation of Boerhaave syndrome as retropharyngeal accumulation of air and pneumomediastinum. Case 3 (67-year-old male) is about retropharyngeal haematoma in a patient with anticoagulant therapy without any definite history of trauma. Treatment with reversal of anticoagulation was successful in the case 3. It is important for the clinician to be aware of the unusual reasons for airway compromise, as early suspicion can translate into better patient survival. Airway should be secured by either intubation or tracheostomy. Many a time, radiological and blood investigations help to arrive at a definite diagnosis.

Keywords: Anticoagulants, Boerhaave syndrome, Esophageal perforation, Haematoma, Lemierre's syndrome, Venous thrombosis

INTRODUCTION

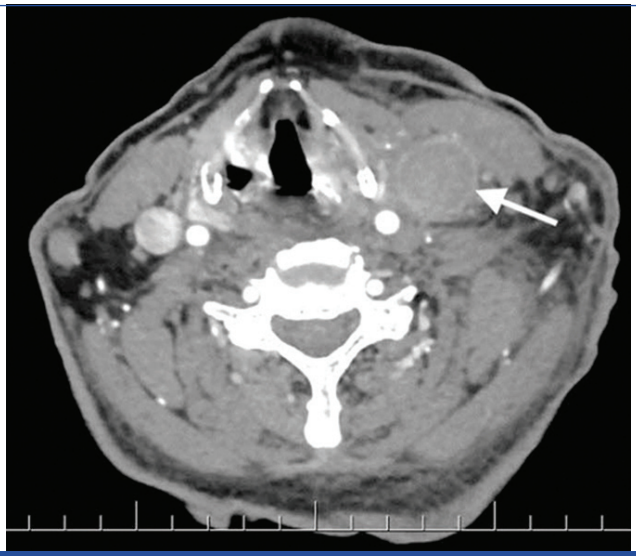
Airway emergencies must be recognised and dealt with promptly. Common causes of acute airway obstruction in adults include anaphylaxis, angioneurotic oedema, trauma, infections, inhalational injury, vocal cord paralysis [1]. Intubation is anticipated to be difficult. Occasionally prompt, disease specific management can avert intubation such as adrenaline shots in anaphylaxis. Treatment of the primary cause such as antibiotics in infections and abscess drainage is essential. While knowledge about the common reasons for acute airway obstruction is essential, awareness about the rare, atypical manifestations would increase the chances of patient survival. Authors report three cases of acute upper airway obstruction requiring intensive care, which are atypical manifestations of diseases. These conditions, Lemierre's and Boerhaave syndrome, itself are uncommon and require specific interventions. Authors aim to bring the spotlight on these to instill a high index of suspicion.

CASE SERIES

Case 1

A 82-year-old male with no co-morbidities presented with fever, sore throat, cough for four days, swelling, pain in the left side of the neck and odynophagia for two days. His symptoms quickly progressed to dysphagia with pooling of secretions and airway compromise requiring intubation. Laboratory results were unremarkable except for total White Blood Cells (WBC) count of 18000/mm^3 (polymorphs 86% with toxic granules, lymphocyte 16%) and raised C-reactive Protein (CRP). Coagulation studies were normal. Blood cultures were negative. Contrast Enhanced Computed Tomography (CECT) scan neck showed left Internal Jugular Vein (IJV) thrombosis with surrounding oedema and mass effect with deviation of trachea. Thrombus extended into the left subclavian and axillary veins. Cervical and mediastinal lymphadenopathy was present [Table/Fig-1]. Other causes for IJV thrombosis like history of IJV cannulation, trauma, malignancy, or other systemic disorders could not be elucidated during further evaluation. Computed Tomography (CT) scan of brain, neck, thorax, abdomen, and tumour biomarkers, all were negative. No metastatic focus of infection was identified.

Patient was diagnosed as atypical manifestation of Lemierre's syndrome, septic thrombophlebitis of left IJV. Broad spectrum antibiotic for gram negative and anaerobic organisms was given for six weeks along with therapeutic anticoagulation. The 2D echocardiography was negative for clot or vegetations. Weaning from ventilator was prolonged. He was tracheostomised and decannulated one month later. On follow-up, he was healthy.

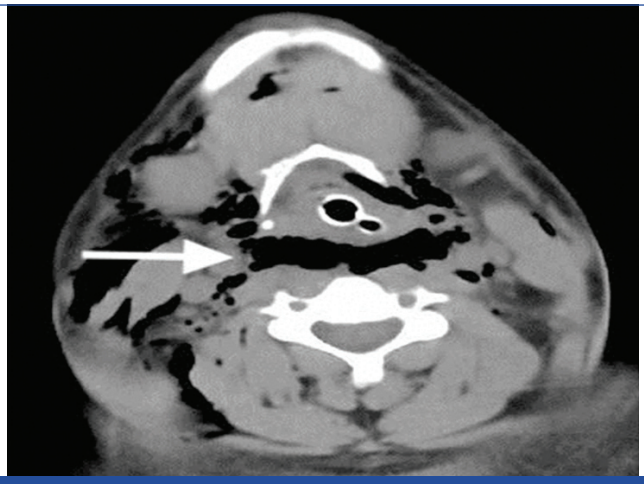


[Table/Fig-1]: CECT scan neck: Left internal jugular vein distended with hyperdense non enhancing thrombus (Case 1).

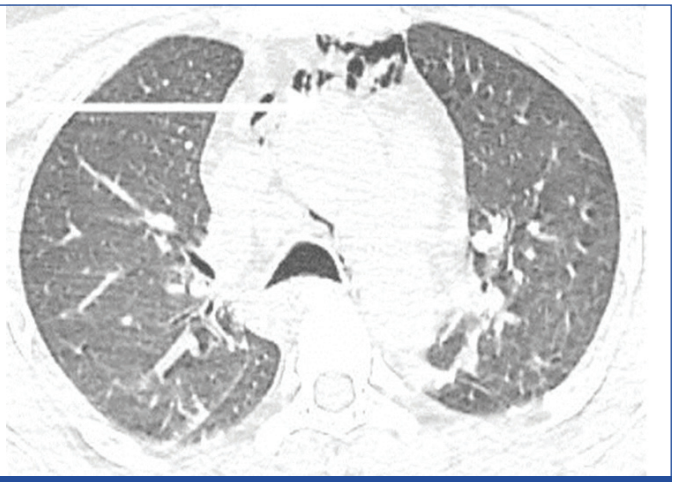
Case 2

A 47-year-old male with history of alcoholism, diabetes mellitus, hypertension, Chronic Kidney Disease (CKD) presented with throat discomfort, oliguria and breathlessness. He was dialysed for acute on CKD. He rapidly developed stridor and desaturation and was intubated. On examination, breath sounds were normal bilaterally. No

obvious swelling of the throat was noticed. Laryngoscopy showed bulging of the posterior wall of oropharynx without oedema of vocal cords or epiglottis. Chest X-ray (CXR) was normal. The CECT neck and thorax was done to rule out possible deep space abscess. It showed extensive air involving the retropharyngeal space extending from the level of clivus to D3 vertebra along with pneumomediastinum but no pneumothorax or pleural effusion [Table/Fig-2,3]. No contrast leaked into mediastinum or lungs, given through a nasogastric tube retracted to the level of carina. No abscess was found in the retropharyngeal or prevertebral regions. No foreign body or tumours were found. An upper gastrointestinal fluoroscopy was done subsequently, which also did not reveal contrast leak.



[Table/Fig-2]: CECT neck: Extensive air involving the retropharyngeal space (Case 2).



[Table/Fig-3]: CECT thorax: Pneumomediastinum (Case 3).

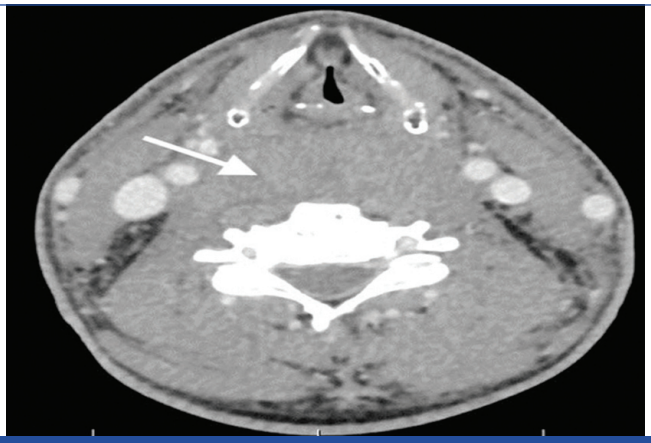
On further enquiry, history of retching and vomiting following episodes of alcohol binges was obtained. He had chest discomfort two days before admission with progressive breathlessness. There was no history of trauma or medical interventions. A diagnosis of spontaneous oesophageal rupture, Boerhaave syndrome was made based on history of vomiting, chest pain and pneumomediastinum. Investigations revealed total WBC 23800/mm^3 (polymorphs 90%, lymphocyte 7%) with raised CRP. Conservative treatment with nil per mouth, broad-spectrum antibiotic piperacillin-tazobactam was given as per gastrosurgery advice with intensive monitoring for sepsis deterioration. On day 13 of admission, he was transferred to another hospital as per family request and lost follow-up.

Case 3

A 67-year-old male, presented with vague pain in the throat and breathing difficulty which started an hour after taking a vegetarian meal. He was diabetic and was on anticoagulants for mitral valve replacement. There was no history of trauma, foreign body ingestion, fever, or neck pain. On examination, SpO_2 was 93%, pulse rate was 88/minute and blood pressure was 150/90 mmHg. He was in stridor and had laboured breathing. Throat examination showed a uniform bulge in the posterior pharyngeal wall with haemorrhagic mucosa. Laryngeal crepitus was absent. Flexible laryngoscopy showed bulging posterior pharyngeal wall with narrowing of glottic space.

A diagnosis of retropharyngeal haematoma, possibly triggered by coarse food matter ingested on the backdrop of anticoagulation therapy was made. Investigations showed haemoglobin 8 gm/dL, International Normalised Ratio (INR) was 8 (prothrombin time test 96 sec, control 12 sec) with normal white cell count, platelet count, liver, and renal function tests.

The CECT neck showed a well defined, hyperdense, non enhancing collection in the retropharyngeal space measuring 2×5 cm and extending craniocaudally from C2-D4 vertebral level [Table/Fig-4]. As intubation had the risk of triggering further bleed and patient's blood gases were in acceptable range, he was admitted for close observation. Anticoagulation was withheld until INR reached the target range. He improved with conservative management. Videolaryngoscopy performed after a week showed resolution of the haematoma.



[Table/Fig-4]: CECT neck: Hyperdense non enhancing collection in the retropharyngeal space (Case 3).

DISCUSSION

Acute airway obstruction may, at times, be atypical manifestation of diseases. Many a times, imaging studies are ordered to rule out deep space infections of the neck which then subsequently, reveal unexpected aetiological findings as was the case in this series. All three cases reported were uncommon diagnoses and manifested in an atypical fashion with acute airway obstruction. In the first case, once the CECT scan clinched the diagnosis of IJV thrombosis, patient required prolonged antibiotics and mechanical ventilation, resulting in significant morbidity. Initial history and symptoms need not reveal the aetiology as in the second case. The history of chest pain and vomiting came to light only on further directed questions after the diagnosis was suspected based on retropharyngeal air in the CECT. Initial clinical examination and CXR also missed signs of oesophageal perforation. It is therefore important to maintain a high index of suspicion and be aware of the atypical presentations of Boerhaave disease. Third case was difficult in that coagulopathy made any intervention risky. While the first two required emergency intubation, retropharyngeal haematoma could safely be managed with reversal of anticoagulation.

Most common causes of IJV thrombosis are malignancy and central venous catheterisation, less common causes being local infections, trauma, surgery, or hypercoagulable states [2]. IJV thrombosis in the setting of recent oropharyngeal infection, with anaerobic bacteraemia caused primarily by F. necrophorum is diagnosed as Lemierre's syndrome [3]. Upper respiratory tract is the most common primary site of infection, although skin, gastrointestinal and genitourinary infections can also predispose to this condition. Metastatic infections occur in 63-100% of patients, most commonly in the lung followed by the major joints. Life-endangering complications such as

pulmonary embolism, airway swelling, cortical venous thrombosis, septic emboli can occur [4]. Early antibiotic therapy in this case possibly prevented progression of thrombosis to metastatic sites. The full spectrum of Lemierre's syndrome is seldom seen in the present era of advanced diagnostics and antibiotics.

Oesophageal perforation is potentially life-threatening with 13% mortality rate. It may be iatrogenic (46.5%), spontaneous (38%) or due to foreign body (6%) [5]. Boerhaave syndrome, is a spontaneous perforation of the oesophagus that results from a sudden increase in intraesophageal pressure combined with negative intrathoracic pressure such as that associated with vomiting [6]. Although rare, if unrecognised, it can be fatal. Majority of perforations are thoracic (70%). The classic triad of acute epigastric and substernal chest pain, subcutaneous emphysema, and vomiting may not always be present. Atypical presentations of Boerhaave syndrome include hoarse voice, back pain, haematemesis, lung abscess, pericarditis [7]. Patient may appear well with hardly any signs of inflammation before they suddenly decompensate, in cases where the perforation is sealed. The variable clinical presentations may contribute to a delayed diagnosis and poor outcome. The CT with oral contrast and presence of salivary amylase in the pleural fluid may be diagnostic. Treatment may be conservative, endoscopic, and surgical intervention depending on the severity of the perforation and clinical progression to sepsis [8]. If the perforation occurs beyond 24 hours, and the leakage is contained without systemic signs of infection, then conservative treatment is usually advocated.

Haematoma in the retropharyngeal space has the potential risk for airway obstruction. Retropharyngeal haematomas occur most commonly following road traffic accidents, neck trauma and following sharp foreign body ingestion [9]. Anticoagulant with uncontrolled bleeding parameters can trigger bleeding in the retropharyngeal space even with trivial injuries as in the present case series [10]. At times, cervical angiography and emergent transcatheter arterial embolisation of the bleeding vessels might be helpful [11]. Along with discontinuation of the anticoagulant, vitamin K and fresh frozen plasma might be required. Blood dyscrasias can also cause spontaneous retropharyngeal haematoma [12]. The CECT or Magnetic resonance imaging can reveal the extent of bleed. Intubation can potentially aggravate bleeding and may be impossible in the presence of posterior pharyngeal swelling [11]. Often, tracheostomy is advocated to secure airway which again, can be hazardous in patients with deranged bleeding parameters. If saturation is maintained, a wait and watch policy may be resorted to, with plans for a surgical airway, when required. Management of retropharyngeal haematoma with fibreoptic intubation has been reported [10]. Drainage of haematoma was attempted, obviating the need for intubation in another case report of aspirin induced haematoma showing persistent symptoms [13].

CONCLUSION(S)

Awareness about the rare causes of acute airway obstruction and early suspicion is imperative for a positive outcome. The CECT may be diagnostic when clinical symptoms are non specific, and history is not forthcoming.

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