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Case Report

Diffuse Aspiration Bronchiolitis secondary to Plummer Vinson Syndrome – A rare yet preventable pulmonary complication

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Abstract

Chronic micro aspiration to the lungs can lead to a wide spectrum of manifestations ranging from inflammatory changes to irreversible damage in the form of fibrosis, bronchiectasis, and cavitation leading to Diffuse Aspiration Bronchiolitis (DAB). A significant number of DAB patients goes undiagnosed without clinical suspicion because of atypical presentation. In this report, DAB secondary to oesophageal webs, as part of Plummer-Vinson syndrome is described. Plummer-Vinson syndrome (PVS) is a triad constituting dysphagia, oesophageal web, and iron deficiency anaemia. Even though DAB has been described in patients with predisposing factors like neurological disorders, Gastro Oesophageal Reflux disease (GERD) etc. Plummer Vinson Syndrome has never been reported as a cause in previous literature.

A 56-year-old lady, known case of Plummer Vinson syndrome presented with history of cough and expectoration of purulent sputum for last 10 days. On examination, patient had pallor and bilateral basal crepitations were heard on auscultation. On evaluation, her HRCT thorax showed bilateral lower lobe bronchiectasis with tree in bud opacities and left lower lobe cavitation. Sputum examination was negative for AFB staining and culture revealed growth of Klebsiella pneumoniae. Bronchoscopy showed thick purulent secretions from both lungs. She had microcytic hypochromic anaemia with low iron stores. A diagnosis of Diffuse Aspiration pneumonitis (DAB) secondary to oesophageal web has been made. Patient was treated with antibiotics as per the susceptibility pattern, chest physiotherapy and postural drainage and other symptomatic and supportive measures. Oesophageal web dilatation was done. Iron deficiency anaemia was corrected using parenteral iron therapy followed by oral iron. A proper patient education and counselling was done about the prophylactic measures to be adopted to prevent chronic aspiration.

Diffuse Aspiration Bronchiolitis (DAB) is an underrecognized clinical entity which occurs secondary to a variety of predisposing conditions. Oesophageal dysfunction is one of the most common predisposing factors which in turn can be due to multiple causative factors. Plummer Vinson syndrome is one such rare cause. It is important to evaluate and identify iron deficiency anaemia and early DAB as a complication in such patients. This can help in adopting essential prophylactic measures like correction of iron deficiency and endoscopic dilatation of oesophageal web. This can prevent chronic micro aspiration and the irreversible lung damage which can cause long term pulmonary morbidity.

Keywords: Aspiration, Bronchiolitis, Oesophageal web

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1. Introduction

Aspiration pneumonia occurs usually as an acute process secondary to aspiration of gastric contents, particulate matter, food particles etc. A more insidious process of chronic micro aspiration (chronic occult aspiration) can also happen leading to a chronic condition called diffuse aspiration bronchiolitis (DAB). There are many causes of DAB of which the most common are Gastroesophageal reflux disease (GERD), dysphagia, neurological diseases and drug abuse.[1] In this report, we describe about a patient diagnosed with DAB secondary to oesophageal webs, as part of Plummer-Vinson syndrome. Plummer-Vinson syndrome is a triad constituting dysphagia, oesophageal webs and iron deficiency anaemia.[2,3] The structural and functional abnormalities of oesophagus can lead to chronic micro aspiration as seen in this patient, when oesophageal webs cause prolonged dysphagia and DAB. This scenario has been rarely reported in the current available literature.

In our patient, diagnosis of oesophageal web was done before 6 years and she has developed chronic micro

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aspiration which led to recurrent inflammation and bronchiolitis eventually leading to structural damage of lungs. Awareness about this condition by clinicians with close monitoring and constitution of early prophylactic measures can avoid the risk of aspiration in such patients. Our case report thus aims to give valuable information about this preventable complication due to oesophageal webs.

2. Case Presentation

A 56 year old lady with a h/o Plummer Vinson syndrome diagnosed in 2017, presented to us with complains of cough for 10 days. She was having dysphagia since last 6 years with difficulty in swallowing solids. She was evaluated by medical gastro enterologist during the onset of dysphagia and she underwent barium swallow study (**Figure 1**, **Figure 2**) and upper GI endoscopy (**Figure 3**) and got diagnosed to have oesophageal webs in the crico-pharyngeal region of oesophagus. During that evaluation, patient was found to have anaemia due to iron deficiency. A final diagnosis of Plummer Vinson syndrome was thus made in view of the clinical triad: iron deficiency anaemia, dysphagia, and oesophageal webs. Oesophageal web dilatation was done during the time of diagnosis and advice for correction of iron deficiency was given, but patient was lost to follow up. After two years of diagnosis, she got recurrent episodes of cough with expectoration and was managed symptomatically every time. She also gave history of recurrent episodes of haemoptysis in the form of blood streaking sputum. As the major underlying predisposing factor of iron deficiency was left uncorrected, the patient continued to have chronic micro aspiration leading to irreversible lung damage.

2.1. Clinical findings

On general examination, patient was emaciated, pallor present with nails showing koilonychia. On respiratory system examination, auscultatory finding of bilateral basal coarse crackles were present. Other system examination was within normal limits.

2.2. Diagnostic assessment

Complete hemogram showed Hb of 10.1, Total leukocyte count was 12000. Iron studies showed low iron stores with microcytic hypochromic anaemia. Bronchoscopy was done which showed thick purulent secretions bilaterally, more from superior segment of left lower lobe (**Figure 3**).Chest Xray and HRCT thorax was taken showing evidence of bilateral lower lobe bronchiectasis and left lower lobe cavity with tree in bud opacities (**Figure 4**, **Figure 5**) In Bronchial washings, AFB Smear, CB NAAT was done which did not detect Mycobacterium tuberculosis. Bacterial culture yielded growth of Klebsiella pneumoniae.

2.3. Therapeutic interventions

Patient was treated with antibiotics as per the susceptibility pattern, chest physiotherapy and postural drainage and other symptomatic and supportive measures. Oesophageal web dilatation was done. Iron deficiency anaemia was corrected using parenteral iron therapy followed by oral iron. A proper patient education and counselling was done about the prophylactic measures to be adopted to prevent further chronic aspiration.

2.4. Follow up and Outcomes

On follow up, patient became symptomatically better, radiologically consolidatory and tree in bud opacities got cleared. In view of bronchiectasis and cavitary changes, she has been advised to do bronchial hygiene techniques regularly.

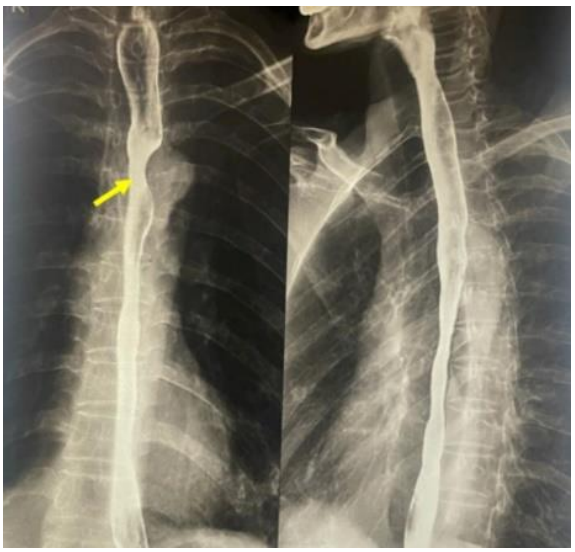

Figure 1: Barium swallow study showing oesophageal web (lateral view)

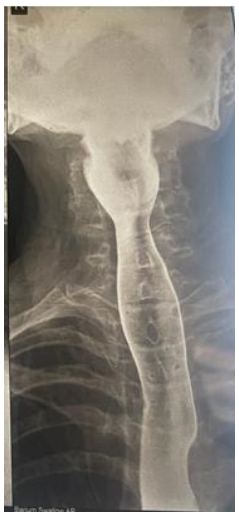

Figure 2: Barium swallow study showing oesophageal web (Anterior view)

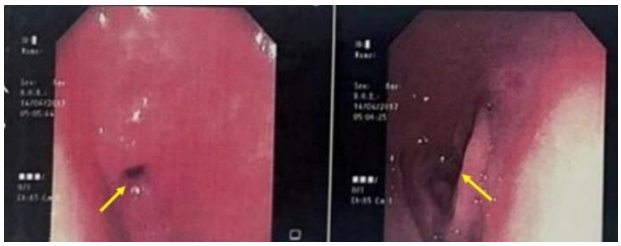

Figure 3: Upper GI endoscopy showing oesophageal web

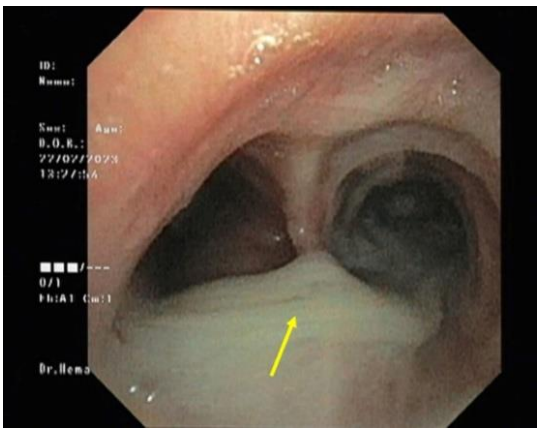

Figure 4: Broncoscopy showing thick purulent secretions in tracheobronchial tree

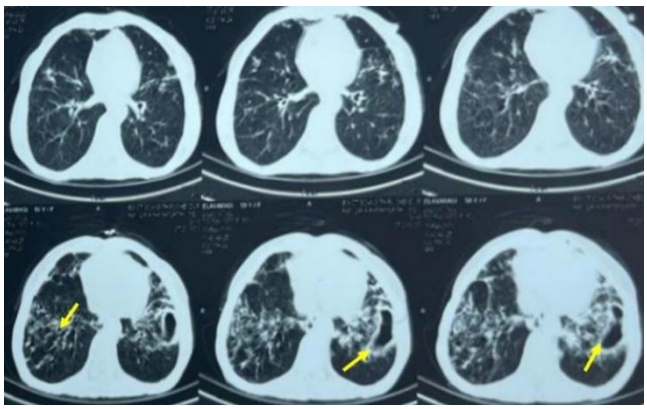

Figure 5: HRCT thorax showing left lower lobe cavity and tree in bud opacities in both lower lobes

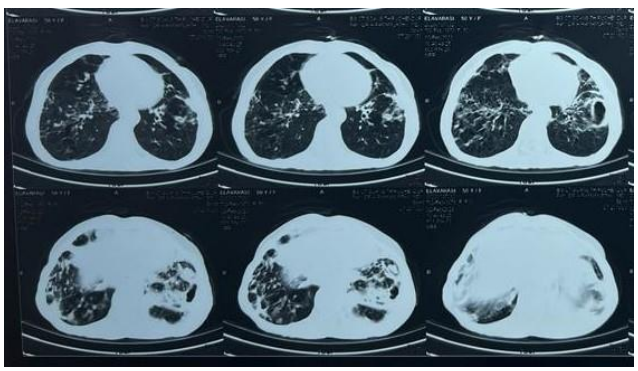

Figure 6: HRCT thorax showing consolidation in bilateral lower lobes

3. Discussion

The term Diffuse Aspiration Bronchiolitis was first defined by Takeshi Matsuse et al in 1996 after studying 4880 consecutive autopsies among which 800 were diagnosed with clinicopathological features of DAB.[4] Out of these 800 patients, Oropharyngeal dysphagia was identified as predisposing factor in half of the patients diagnosed with DAB. In our patient also dysphagia was the most prominent symptom, which persisted for 6 years causing recurrent micro aspirations.

In one of the largest series on DAB, the commonest presenting symptoms were cough and sputum expectoration (80-90%).[1] Radiologically micronodules, tree in bud opacities suggestive of bronchiolitis is seen as in our patient. Our patient also had bronchiectasis also evident in CT scan mentioned in 35% of patients in literature. These radiological findings point towards bronchiolo-centric inflammation secondary to chronic aspiration.

The pathogenesis of iron deficiency causing oesophageal web is unclear, but many mechanisms including iron deficiency, malnutrition, genetic predisposition, autoimmune disorders has been postulated.[6] Iron deficiency causes reduction of iron-dependent oxidative enzymes which results in gradual degradation of the muscles of the pharynx leading onto mucosal atrophy and development of webs.[7]

In a retrospective review of 25 patients with chronic occult micro aspiration defined pathologically on lung biopsy, 40% of patients had associated oesophageal dysfunction in the form of web or stenosis.[5] The mechanism behind this is inefficient passage of food from pharynx to stomach which increases the time of exposure around glottic area which promotes aspiration. According to the chronicity of symptoms as well as the severity, the involvement of lung varies resulting in a spectrum ranging from bronchiolar inflammation to fibrosis and bronchiectasiss.[8,9] Our patient has features of inflammation as well as fibrosis, cavitation and bronchiectasis suggesting the severity of the disease.

Even though the association of oesophageal webs with DAB has been well described, coexisting iron deficiency anaemia constituting Plummer Vinson syndrome has not been reported previously in literature.[8] In our patient, the underlying condition has been corrected and oesophageal dysfunction has improved, but the chronic micro aspirations which has led to irreversible lung damage with significant morbidity.

4. Conclusion

Diffuse Aspiration Bronchiolitis (DAB) is an underrecognized clinical entity which occurs secondary to a variety of predisposing conditions. The lung involvement can range from mild inflammatory changes to irreversible fibrosis and bronchiectasis. Oesophageal dysfunction is one of the most common predisposing factors which in turn can be due to multiple causative factors. Plummer Vinson

syndrome is one such rare cause. It is important to evaluate and identify iron deficiency anaemia and early DAB as a complication in such patients. This can help in adopting essential prophylactic measures and thus prevent the irreversible lung damage which can cause long term pulmonary morbidity.

5. Patient Consent

A written informed consent was taken before publication of the study.

6. Source of Funding

None.

7. Conflict of Interest

None.

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