Ventilation/Perfusion scan aids in the diagnosis of diabetes mellitus induced trepopnea due to isolated right phrenic nerve palsy

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ABSTRACT

Dyspnea can rarely be due to diabetes mellitus induced neuropathy. The term “trepopnea” is sparingly used in clinical practice and refers to dyspnea on assuming a particular lateral decubitus position. Trepopnea is rarely described in association with unilateral diaphragmatic paralysis, which in itself is an uncommon cause of respiratory distress. We report a 27-year-old diabetic female who presented with sudden onset of dyspnea. On close interrogation, patient complained of dyspnea that was exaggerated while lying on the left side (left lateral decubitus position). A fluoroscopic sniff test showed a paradoxically moving right diaphragm confirming the diagnosis of unilateral diaphragmatic paralysis attributed to diabetes induced isolated phrenic nerve palsy. This case highlights the importance of ventilation — perfusion imaging in non-pulmonary etiologies and also attaches importance in recognizing trepopnea as an early clinical symptom of diaphragmatic paralysis. This case illustrates that diabetic neuropathy due to isolated phrenic nerve palsy can occur in the absence of peripheral neuropathy and that glycemic control is unrelated to the manifestation or severity of this disease.

Keywords: Diabetes, diaphragmatic palsy, lung perfusion and ventilation imaging, neuropathy, trepopnea

INTRODUCTION

Dyspnea is a common complaint and may be an important manifestation of an underlying cardiac or respiratory pathology. It can rarely be due to a neuropathy. Some of the “pneas” routinely used in the clinical practice are orthopnea, and dyspnea. However, sparingly used ones are trepopnea and platypnea. Trepopnea is an almost forgotten terminology, used to denote a positional breathing difficulty, i.e., appears when the patient assumes a particular lateral decubitus position. Unilateral diaphragmatic paralysis is one of the causes of trepopnea, which in itself is an uncommon isolated presentation in a young diabetic patient.

CASE REPORT

A 27-year-old female with a 3 year history of diabetes mellitus presented with sudden onset dyspnea of 2 weeks duration. Patient is on 5 units of regular human insulin subcutaneously per day and was otherwise asymptomatic. Clinically, patient was conscious, well-oriented with preserved higher functions and intact cranial nerves. No sensory motor deficit was elicited. Patient was well-nourished, afebrile, with a pulse of 84 beats/min, a blood pressure of 120/80 mmHg (right arm; sitting position), a respiratory rate of 20/min and oxygen saturation of 96% on room air. Patient showed no jugular venous distension and cardiovascular examination revealed no abnormality. Trachea was situated in the midline. Patient showed reduced chest expansion and dullness to percussion on the right side and decreased breath sounds in the right posterior lower lung field. Her basic biochemical investigations were negative including D-dimer test. Recent chest X-ray was non-contributory [Figure 1].

Patient was referred to Nuclear medicine to rule out pulmonary embolism given the sudden onset of symptoms. $^{99m}$Tc DTPA (Di ethylene triamine penta acetic acid) aerosol lung ventilation scintigraphy [Figure 2a] was performed followed by $^{99m}$Tc MAA (Macro aggregated albumin) lung perfusion scintigraphy [Figure 2b]. Both sets of images revealed no segmental or sub-segmental ventilation — perfusion (V/Q) defects. However, elevation of right hemidiaphragm was noticed in both scans.
A fluoroscopic sniff test showed paradoxically moving right diaphragm confirming the diagnosis of unilateral diaphragmatic paralysis that was attributed to isolated phrenic neuropathy caused by diabetes mellitus. Patient was treated with parenteral immunoglobulins.

DISCUSSION

Trepopnea originates from the Greek word “trepo” meaning to twist or turn. Initially, it was referred as “rotopnea,” by Francis wood that was later renamed to “trepopnea.” Trauma and iatrogenic etiologies are the most common causes of phrenic nerve palsy. Various other documented causes are viral infections (herpes zoster, human immunovirus), neoplasms such as bronchogenic carcinoma, mediastinal tumors, head and neck tumors, inflammation such as vasculitis and spinal cord injuries. In a significant number of cases, no etiology is found. Of the iatrogenic causes, injury to the phrenic nerve during cardiac surgery is frequent. Diabetic phrenic neuropathy was the only identifiable etiology in our patient and the probable explanation for trepopnea in left decubitus position with right diaphragmatic paralysis may be related to compression of the normal left lung due to the displacement of the mediastinum towards the left side. This would have caused further reduction in the overall lung capacity for gaseous exchange.

The diagnosis of phrenic nerve palsy can be particularly difficult in cases of subtle paradoxical diaphragmatic excursions under fluoroscopy as in our case. Early diagnosis usually relies on a high index of clinical suspicion supported by radiological evidence of elevated hemidiaphragm. However, as diaphragmatic position is asymmetrical, individual variations have to be kept in mind. A difference of approximately two centimeters in height between the left hemidiaphragm and the right or vice versa should raise a suspicion. Chest radiography alone is unreliable and fraught with interpretational errors, due to factors related to positive pressure ventilation, and diffuse lung disease. Similarly, Computed tomography (CT) has no significant role. Supine position and variable inspiratory effort of a patient can significantly alter the height of the diaphragm on CT. Nevertheless, CT may be useful to rule out a compressive pathology of phrenic nerve. Magnetic resonance imaging is particularly suited in the evaluation of superior sulcus tumors. Fluoroscopy Sniff test examination of the diaphragm is confirmatory. In this test, patient is allowed to undertake rapid inspirations while the hemi diaphragm is observed for any mal excursions. In normal subjects, both hemi diaphragms descend with inspiration.

Recently, there has been a renewed interest in identifying trepopnea, a symptom associated with congestive heart failure.

CONCLUSION

Although, dyspnea may be a frequent complaint in diabetics especially in the background of co-existing coronary artery disease, cardiomyopathy, congestive heart failure, eliciting a history of trepopnea is invaluable. This case highlights the use of V/Q scan in identifying non pulmonary causes of dyspnea and guiding the physician to look for diaphragmatic palsy. Careful
observation of V/Q images is essential to identify subtle related and unrelated abnormalities.

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