Thoracic aortic aneurysm presenting only as vocal cord paralysis

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ABSTRACT

We present a case of unilateral vocal cord paralysis in a 76-year-old Saudi male presenting with hoarseness of voice caused by thoracic aortic aneurysm. Here, we are illustrating a rare entity of thoracic aortic aneurysm presenting only with hoarseness of voice to draw attention to unusual clinical manifestations of this serious underlying aortic pathology.

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Unilateral vocal cord paralysis (UVCP) occurs from a dysfunction of the recurrent or vagus nerve innervating the larynx, and causes a characteristic breathy voice, this is called neurogenic hoarseness. When this paralysis is properly evaluated and treated, normal speaking voice is typically restored. Normal vocal cord function is reliant on vocal cord glottal closure that results from bilateral adduction of the vocal cords. The UVCP results in glottal incompetence, either partial or complete because of the poor or reduced vocal cord closure resulting in a weak and uncoordinated vocal fold vibration that leads to dysphonia.[1] Patients with UVCP typically present with a sudden onset of breathy, weak, low pitched dysphonia and often UVCP is associated with dysphagia, specifically with liquids, because the resultant glottal incompetence can lead to aspiration. Patients with UVCP often report shortness of breath, or a feeling of running out of air.[2] The most common cause of UVCP remains controversial. From the results of 9 studies, it appears that malignancy is the most common cause of UVCP. The malignant invasion of either the vagus or recurrent laryngeal nerve can occur with skull base tumors, thyroid cancer, lung cancer, esophageal cancer, and metastasis to the mediastinum. Surgical injuries, often touted as the most common cause by some authors, come in second according to the combined results of these studies; these surgeries include thyroid surgery, anterior cervical disc surgery, carotid surgery, or chest surgery. When a clear-cut etiology for the UVCP is not found, it is classified as idiopathic. These cases can be attributed to a viral or inflammatory process, but this is usually a presumptive diagnosis.[3] Patients with thoracic aneurysms are often asymptomatic at the time of presentation. However depending on aneurysm location, chest, or back pain can be a presenting symptom. Ascending and arch aneurysms can erode into the mediastinum and illicit other symptoms.[4] Herein, we report a rare entity of thoracic aortic aneurysm presenting only with UVCP to illustrate all the clinical manifestations of thoracic aneurysm, even rare ones which may be the clue to diagnosis.

Case Report. A 76-year-old Saudi male, known hypertensive on medical treatment and a smoker for the last 40 years, with a history of mild leg claudications, and symptoms of coronary artery disease presented with hoarseness of voice for the last 2 months, and weight loss of 6-8 kg in the last one year. He also gave a history of on and off cough productive of sputum and shortness of breathing on exertion. On examination his blood pressure was 160/90 mm Hg, afebrile, oxygen saturation 91% at room air, venous pressure was raised with mild pedal edema. Chest examination showed a barrel shaped chest, hyper resonant percussion, vesicular breathing with occasional crepitations, and signs of chronic obstructive pulmonary disease (COPD). The abdomen was soft and lax, with no organomegaly. Cardiovascular system examination showed S1+S2 audible with no murmurs. Recent arterial blood gases (on room air) showed pH 7.46, pO_2 134, pCO_2 34, and bicarbonate 23. Sputum cytological examination was negative for malignant cells. Echocardiographic examination showed left ventricular hypertrophy and hypokinesia, and pulmonary arterial hypertension. Chest x-ray posteroanterior view showed marked roundish prominence of the aortic knob suggestive of aortic arch aneurysm, with mild aneurysmal formation towards

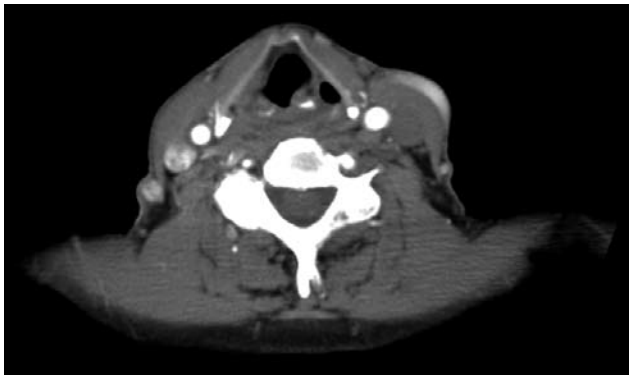

Figure 1 - Neck CT at the level of the larynx showing the left unilateral deformity involving the glottic area suggestive of vocal cord paralysis.

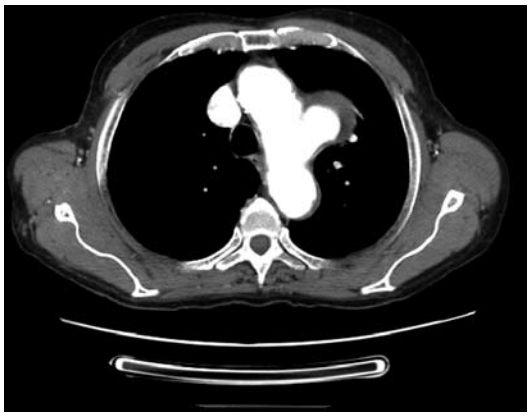

Figure 2 - Computerized tomography axial cuts, mediastinal window at the level of the aortic arch confirming the large saccular aneurysm arising from the distal arch near the aortopulmonary window.

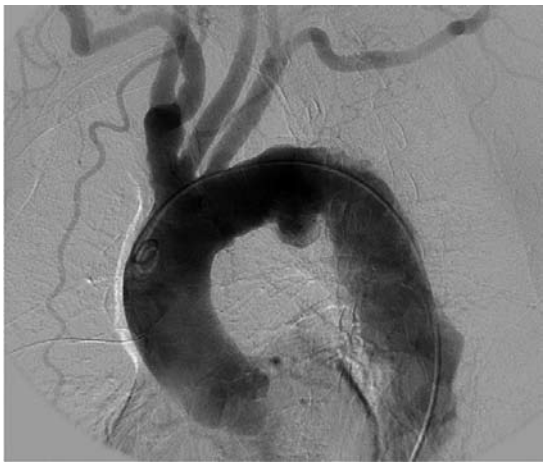

Figure 3 - Arch aortogram clarifies the saccular arch aneurysm arising distal to the origin of the left subclavian artery in addition to the fusiform aneurysmal dilatation of the lower descending aorta.

the lower dorsal aorta. Chest x-ray lateral view showed more clearly the aortic arch aneurysm in addition to the aneurysmal dilatation and tortuosity towards the distal dorsal aorta. A CT scan of the chest and neck was carried out with intravenous contrast, the upper cuts at the level of the larynx (**Figure 1**) showed unilateral deformity involving the left supraglottic and glottic areas with relative thickening, buckling, and asymmetry of the aryepiglottic fold, pyriform sinus, and true vocal cord suggestive of UVCP. Lower down, the cuts at the aortic arch mediastinal window (**Figure 2**) confirmed the large saccular aneurysm arising from the distal arch near the aortopulmonary window measuring approximately 45.8 x 45.2 mm with peripheral eccentric thrombosis. The pulmonary artery and its main branches are prominent due to pulmonary arterial hypertension. The lung window shows the background of extensive centriacinar emphysematous changes and diffuse peri bronchial wall thickening consistent with COPD. The arch aortogram (**Figure 3**) shows the saccular aneurysmal dilatation of the aortic arch seen distal to the origin of the left subclavian artery, in addition to the aneurysmal tortuous dilatation of the lower descending thoracic aorta. The innominate and left common carotid arteries have a common origin (normal variant). He was referred for fiberoptic bronchoscopy that showed left vocal cord paralysis, right vocal cord moving normally, normal trachea, sharp carina, right and left bronchi showing inflammatory changes and hyperemia but no mass lesions, findings that confirmed the suggestion of left vocal cord paralysis caused by pressure of the arch aortic aneurysm on the left recurrent laryngeal nerve and not due to mass lesion. The patient then was referred and followed by the vascular surgeon for assessment of surgical feasibility.

Discussion. Hoarseness of voice is a self-limiting disease usually due to upper respiratory tract infection or vocal abuse. It is symptomatic of laryngeal disease resulting from interference of normal apposition of vocal cords. Evaluation of the history is important as it may explain the etiology of hoarseness. Any recent upper respiratory tract infection, sore throat, fever, chronic sputum, myalgias, or excessive voice use may lead to the diagnosis. Exposure to dust, fire, smoke, irritant fumes, use of tobacco or alcohol, history of neck surgery, intubations or lung tumors, or symptoms of hypothyroidism may elicit the diagnosis.[5] Hoarseness of voice, as the only feature of thoracic aortic aneurysm is a rare entity, occurring in approximately 5% of cases. Ten percent of dissecting thoracic aortic aneurysms are painless. The enlarging lumen of the aneurysm may directly compress on the adjacent nerve, causing neuronal injury of the recurrent laryngeal nerve manifesting in neurological symptoms,[6] as in our case, the patient's

presenting symptom was hoarseness of voice and not chest or back pain. On examination, one needs to note the quality of voice. A breathy voice (as in our case) suggests poor cord apposition; a raspy voice suggests cord thickening due to edema or inflammation; a high shaky or very soft voice suggests inadequate respiratory force. Persistent hoarseness beyond 3 weeks without history of acute infection requires laryngoscopy.[7] In our case, the patient had hoarseness for approximately 2 months without history of acute infection, so was referred for otolaryngologic examination that came back after indirect laryngoscopy as normal, but was proven later by the pulmonologist via bronchoscopy as left vocal cord paralysis.

The right vagus nerve passes anterior to the subclavian artery and gives off the right recurrent laryngeal nerve. This loops around the subclavian artery and ascends in the tracheo-oesophageal groove. The left vagus nerve does not give off its recurrent laryngeal nerve until it is in the thorax, where it wraps around the aorta just posterior to the ligamentum arteriosum. It then ascends back towards the larynx in the tracheo-oesophageal groove.[8] Recurrent laryngeal nerve palsy with unilateral paresis of the vocal cords may involve lesions in the chest, skull base, or neck. A radiological evaluation should be carried out as in our case. Lesions in the aorto-pulmonary window may be difficult to identify or characterize on chest x-ray, therefore, contrast enhanced CT scan may be used to further evaluate these lesions. Chest x-ray showed a suspicious marked prominence of the aortic knob, which was confirmed by CT to be arch aneurysm. The differential diagnosis includes enlarged mediastinal lymph nodes, bronchogenic or other foregut cysts, neurogenic or bronchogenic neoplasms, or other causes of compression of the recurrent laryngeal nerve by the dilated atrium.[9]

A rare entity of UVCP is caused by thoracic aortic aneurysm, as in our case. A true aneurysm is currently defined as a localized dilatation of the aorta, 50% over the normal diameter, which includes all 3 layers of the vessel. The 2 major types of aneurysm morphology are fusiform, which is uniform in shape with symmetrical dilatation that involves the entire circumference of the aortic wall; like the one involving the distal dorsal aorta in our case; and saccular, which is more localized and appears as an out pouching of only a portion of the aortic wall, as the one involving the aortic arch in our case. Thoracic aneurysms occur most commonly in the sixth and seventh decades of life, and males are affected approximately 2-4 times more commonly than females, as in our case the patient was a 76 year old male.

Hypertension is an important risk factor, being present in over 60% of patients, our patient was chronically hypertensive. Up to 13% of patients diagnosed with an aortic aneurysm are found to have multiple aneurysms; approximately 20-25% of patients with a large thoracic aortic aneurysm also have an abdominal aortic aneurysm,[9] and these were the features in our case. Patients with thoracic aneurysms are often asymptomatic at the time of presentation. However, sometimes, patients with arch aneurysms can present with one or more of the following: hoarseness due to compression of the left vagus or left recurrent laryngeal nerve; hemidiaphragmatic paralysis due to compression of the phrenic nerve; wheezing, cough, hemoptysis, dyspnea, or pneumonitis if there is compression of the tracheo-bronchial tree; dysphagia due to esophageal compression; or the superior vena cava syndrome. Aneurysm compression of other intrathoracic structures or erosion into adjacent bone may cause chest or back pain. Aneurysmal compression of branch vessels, or the occurrence of embolism to various peripheral arteries due to thrombus within the aneurysm can cause coronary, cerebral, renal, mesenteric, lower extremity, and rarely spinal cord ischemia and resultant symptoms. The most serious complication of thoracic aortic aneurysm is leakage, which may cause pain, or rupture, most often into the left intrapleural or intrapericardial space.[10]

A variety of non-invasive and invasive methods are useful for the diagnosis and evaluation of thoracic aortic aneurysm. A common way in which asymptomatic aneurysms are detected is on routine chest radiography. The aneurysm produces a widening of the mediastinal silhouette, enlargement of the aortic knob or displacement of the trachea from midline, and these were exactly the features in our case. However, smaller aneurysms may not be apparent on chest x-ray. Transthoracic echocardiography is of limited value in thoracic aortic disease because of non-conductance of the signal by lung air, and this was unremarkable in our case regarding the aneurysm. A CT with intravenous contrast is an accurate diagnostic tool in the evaluation of thoracic aneurysmal disease. Helical CT of the thoracic aorta provides a minimally invasive and rapid assessment of the aortic lumen and branch vessels, the aortic wall and the surrounding tissues,[11] and all of these criteria were evaluated in our case by CT. Similarly, Dyer et al[12] found that CT was equivalent to angiography, and this was proven also in our case with the CT results almost matching those of angiography. The CT has 100% sensitivity and 100% negative predictive value. Typically, Dyer uses 1.25 mm slice thickness to get the best details possible, then he reconstructs the data at one mm interval to get the best data sets possible for 3D imaging,[12] as followed in our case. Contrast angiography is the preferred method for evaluation as it provides sharper resolution of luminal characteristics, and is the best method for evaluating branch vessel pathology.

This procedure is invasive with potential nephrotoxicity and cannot discern extraluminal aneurysmal size,[13] a description which correlates well with our case arch aortography. The MRI is the newest diagnostic modality that offers non-invasive angiography with multiplanar image reconstruction and visualization of extraluminal structures. Its disadvantage is lower resolution than traditional contrast angiography.[14] In our case, the diagnosis was already reached accurately without MRI.

In conclusion, in this report, we want to illustrate a rare entity of thoracic aortic aneurysm presenting only with UVCP. Classically, thoracic aortic aneurysms manifest with chest and back pains or stroke symptoms if carotid arteries are involved. Patients who do not present with classic symptoms, as in our case, may be misdiagnosed or diagnosis may be delayed. So, in this report, we want to draw attention and keep in mind all the varieties of clinical manifestations of aortic aneurysms, to have a high index of suspicion in cases of unusual presentations, especially in patients with chronic hypertension and other predisposing conditions and this undoubtedly will pave the way to proper early diagnosis and prompt management before occurrence of serious complications.

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