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CARDIAC DYSPHONIA –UNUSUAL CASE SERIES WITH REVIEW

Otorhinolaryngology

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ABSTRACT

Background: Dysphonia is a common symptom routinely encountered in patients in Otorhinolaryngology department. We are reporting three cases of dysphonia secondary to cardiovascular pathology, better known as Ortner's or cardiovocal syndrome. We are publishing these cases because of the rare and interesting associations. Very few case reports have been published till date. **Case presentation:** The first case described is a 24 year old man diagnosed with severe pulmonary hypertension causing pressure over the recurrent laryngeal nerve leading on to cardiovocal syndrome.The other cases reported are in elderly men with Ortner's syndrome secondary to aortic aneurysm. **Conclusion:** This case series emphasize on the multidisciplinary approach needed in diagnosis.

KEYWORDS

Ortner's syndrome, Cardiovocal , Hoarseness , Rheumatic heart disease

BACKGROUND

The eponym "Ortner "is after Nobert Ortner , an Austrian physician who reported 3 cases of hoarseness of voice with mitral stenosis in 1897. He attributed the cause as compression of left recurrent laryngeal nerve by an enlarged left atrium[1].This theory has been modified by various authors. In 1911, Fetterolf and Norris postulated from a cadaveric study that the recurrent laryngeal nerve gets compressed between the pulmonary artery and aortic arch. In 1934, King et al published that left recurrent laryngeal nerve palsy was due to left ventricular failure[2]. Stocker and Enterline used the term 'Cardiovocal" for the first time in some English journals in 1958[3].They published that dilated pulmonary artery was the prime cause of palsy.

Case Presentation-1

24 year old man came to our Otorhinolaryngology outpatient department with complaints of gradually progressive change in voice for a week. Patient noted a breathy voice.He couldn't talk loudly.He also had difficulty in swallowing solids. He had no history of fever, night sweats, loss of weight or appetite. He was not a smoker. No past history of surgeries, trauma,tuberculosis,hypertension, diabetes mellitus.

Patient was thin built. On examination he had no stridor or cyanosis. Neck examination did not reveal significant lymphadenopathy or goitre. Videolaryngoscopy done was suggestive of left vocal fold palsy. Left true vocal fold was immobile in paramedian position with bowing (Figure 1a).Stroboscopy done showed loss of mucosal wave in left vocal fold and asymmetry between the vocal folds. GRBAS score suggestive of breathy voice secondary to laryngeal cause.

To look for a cause beyond the larynx , the patient was asked to get a high resolution computed tomography from skull base to mediastinum. Radiologist reported mitral valve calcifications with dilated left atrium and ventricle. Left pulmonary artery was dilated and measured around 35mm(Figure 2).At the laryngeal level, sail sign was noted with medialization of left posterior vocal fold margin and ipsilateral dilatation of left ventricle(Figure 1b). Bilateral lung parenchyma was normal.

Upon evaluation by cardiologist, auscultation revealed mid diastolic murmur in the mitral area. Heaving of the parasternal region was noted.2D echocardiography reconfirmed the dilated left atrium,severe mitral stenosis with calcified mitral valve probably due to rheumatic heart disease. The right ventricle systolic pressure was raised to 60.7mmHg and peak tricuspid velocity TR Vmax was 3.4m/sec. Severe pulmonary artery hypertension was elucidated.

Patient was advised to undergo mitral valve replacement.He is delaying the procedure at present due to financial constraints.He is being followed up by Cardiac surgery team.

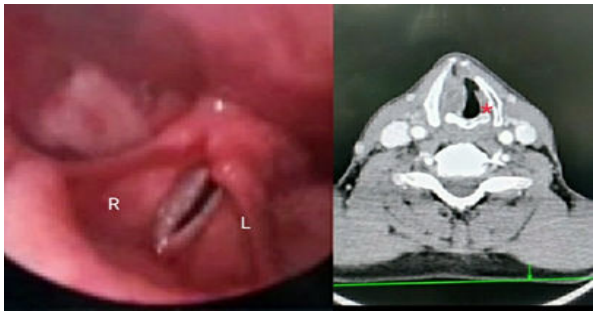

Figure 1a & 1b showing videolaryngoscopic image of left vocal fold in paramedian position with bowing(R-Right and L-Left) and High resolution computed tomographic axial image showing sail sign(red *) respectively.

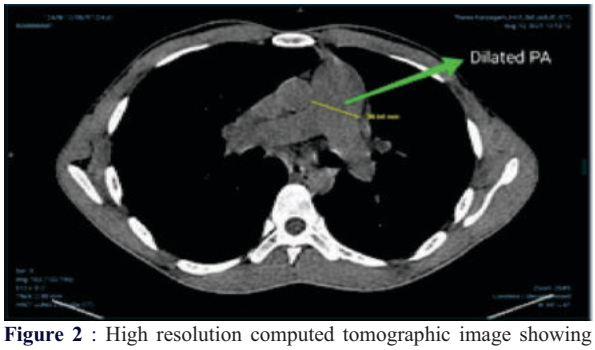

Figure 2 : High resolution computed tomographic image showing dilated left pulmonary artery.

Case Presentation- 2

62 year old man presented with complaints of weak, breathy voice which was gradually progressive.Change in voice was present throughout the day and he gives history of vocal fatigue. He had history of intermittent chest pain with radiation to abdomen and back for the past one year. He felt dyspneic while walking and complained of palpitations with light headedness. He was diagnosed with systemic hypertension one year back and was on irregular medications. He has been a chronic smoker for the past 30 years.

Clinical examination revealed pandigital clubbing .His blood pressure was 160/100mmhg in the right upper limb with radial pulse of normal rhythm. Video laryngoscopy showed left vocal fold palsy in paramedian position.

Considering his status detailed cardiac imaging was done. Computed tomography of the neck with aortogram showed an aneurysm involving parts of the isthmus, thoracic and abdominal portions of aorta.Maximum dilatation of aneurysm noted near the distal portion of aortic arch narrowing the aorto-pulmonary window with possible compression of the left recurrent laryngeal nerve(Figure 3a,3b).

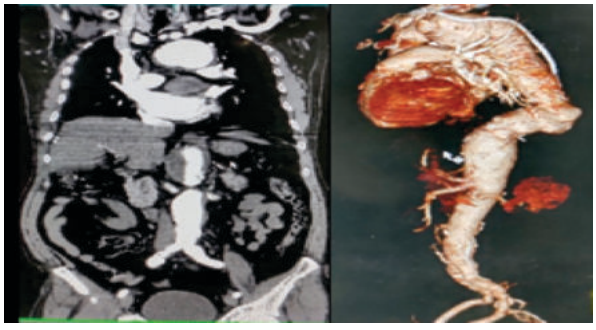

Figure 3a & 3b: Coronal images of 3D Aortogram and contrast enhanced computed tomography showing aneurysm of the aorta with maximum dilatation near the distal portion of aortic arch narrowing the aorto-pulmonary window.

We discussed with the Cardiothoracic surgeon and he advised a further detailed cardiological evaluation .Coronary angiography was done and multiple thrombotic blocks were noted in the coronary blood vessels.He was advised coronary artery bypass graft along with grafting of the aneurysm.Patient and family are denying procedure due to age and the risks concerned.

Case Presenation- 3

69 year old elderly man , known hypertensive and diabetic visited our department with hoarseness of voice for a month.He was a chronic smoker.We performed video laryngoscopy and the left vocal cord was in paramedian position with bowing(Figure 4a).Neck and systemic examination was uneventful. As a first line imaging we referred the patient for contrast enhanced CT from skull base to mediastinum.

Imaging localized atherosclerosis along with a saccular aneurysm in the arch of aorta close to the origin of left subclavian artery (Figure 4b).We attributed the cause of left vocal cord palsy to the compression of left recurrent laryngeal nerve by the saccular aneurysm of arch of aorta.Patient was referred to cardiothoracic surgery for further management.

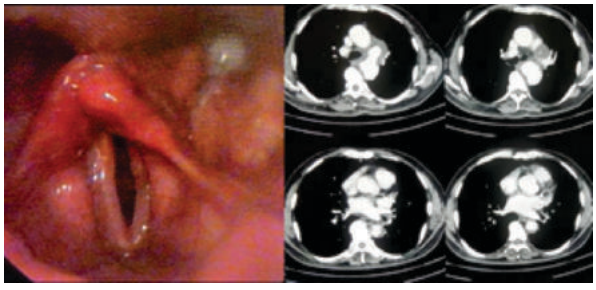

Figure 4a & 4b :
showing Video laryngoscopic image of bowing of left vocal cord in paramedian position and contrast enhanced CT image of dilated arch of aorta.

DISCUSSION

Vagus nerve, true to its name, has a vagabond nature. Vagus nerve wanders as superior and recurrent or inferior laryngeal nerve. In the right side the recurrent laryngeal nerve loops around the right subclavian artery and runs in the tracheoesophageal groove. The left recurrent laryngeal nerve has a longer course and is more prone to injury[4]. It loops around the aortic arch,runs in the narrow aortopulmonary window and ascends towards the neck.

Ortner's syndrome includes any non-malignant, cardiac or intrathoracic process causing recurrent laryngeal nerve palsy by stretch,pull or compression.[1]

We have used the keyword Ortner's syndrome, Cardiovocal syndrome and identified 21 cases reported in Pubmed for the past 5 years.On analyzing the research data over the past five years, the average age of presentation was the 6th decade of life.55 % of the patients were male and 45% of the patients were females. We collected data on 21 cases and almost 15 cases had change in voice due to aneurysm in the thoracic aorta,subclavian or pulmonary artery. Only one case of rheumatic heart disease has been published in the recent years. It was seen in a 60 year old lady who presented with dyspnea and breathy voice. She had a giant left atrium with mitral valve stenosis. Post double valve replacement, rapid reversibility of voice was observed.[5]In case 1, the patient diagnosed with rheumatic heart disease was younger and had complaints of change in voice with dysphagia unlike the other cases which commonly presented with dyspnea. Dysphagia could be due to pressure effect by the dilated left atrium secondary to severe mitral stenosis. In case 2 , patient had extensive saccular aortic aneurysm of the arch,thoracic and abdominal aorta.Owing to impending rupture it was considered a medical emergency. In case 3 patient had a similar saccular aneurysm in the arch of aorta.Only 2 cases of Ortner's syndrome both secondary to thoracic artery aneurysms have been reported from the Indian subcontinent in the past 5 years.[6,7]An Interesting case of an acute aortic dissection presenting with acute dysphonia had been reported in the past and timely intervention had saved that patient[8].Hence it is crucial to broaden our horizons and be prompt in our diagnosis. Successful recovery of voice depends on the cardiovascular causes and its severity.

CONCLUSION

We have published these case reports because of their rare and intriguing association. We had to rule out the common causes of recurrent laryngeal nerve palsy initially by performing videolaryngoscopy. We always do a detailed examination of chest if the cause is not in the larynx to look for tuberculosis which is endemic in our nation.If the cause is still not conclusive we should ideally screen from the skull base to mediastinum to look for causes along the entire course of vagus nerve. Although these patients presented initially with dysphonia to the otorhinolaryngology department, we had to combine with the department of radiology,speech and language pathology,cardiology and cardiothoracic surgery to connect the dots and manage the case effectively. Ortner's syndrome as a differential diagnosis, though rare, should never be forgotten.

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