Acıbadem Üniversitesi Sağlık Bilimleri Dergisi
Cilt: 6 • Sayı: 1 • Ocak 2015

Hoarseness Secondary to Cardiovascular Disease: Cardiovocal Sydrome in a Patient With Pulmonary Artery Aneurysm and Giant Atria

Serkan Duyuler[1], Pınar Türker Bayır[2], Ahmet Korkmaz[3], Yeşim Güray[4], Erdemli Özcan[5]

[1]Acıbadem Ankara Hastanesi, Kardiyoloji Bölümü, Ankara, Türkiye
[2]Ankara Numune Eğitim ve Araştırma Hastanesi, Kardiyoloji Bölümü, Ankara, Türkiye
[3]Çankırı Devlet Hastanesi, Kardiyoloji Bölümü, Çankırı, Türkiye
[4]Türkiye Yüksek İhtisas Eğitim ve Araştırma Hastanesi, Kardiyoloji Bölümü, Ankara, Türkiye
[5]Acıbadem Ankara Hastanesi, Anestezi Bölümü, Ankara, Türkiye

ABSTRACT

Cardiovocal syndrome or Ortner syndrome is the hoarseness secondary to recurrent laryngeal nerve palsy due to compression enlarged cardiovascular structures. Dilated left atrium with mitral valve disease is a well-known cause for this rare syndrome; however several cardiovascular conditions also contribute to the pathogenesis. Data suggest that, recurrent laryngeal nerve seems to be compressed in the window between enlarged hypertensive pulmonary artery, aorta and ligamentum arteriosum not solely by enlarged left atrium. In this context, we presented a case of cardiovocal syndrome in a patient with pulmonary artery aneurysm, giant atria and corrected atrial septal defect. The patient was admitted for gradual hoarseness for two years and laryngoscopy revealed left vocal cord paralysis. Cardiovascular examination with transthoracic echocardiography showed pulmonary artery aneurysm with giant atria which is compatible with cardiovocal syndrome.

Key words: hoarseness, cardiovocal syndrome, pulmonary artery, aneurysm

KARDIYOVASKÜLER HASTALIĞA BAĞLI SES KISIKLIĞI: PULMONER ARTER ANEVRIZMASI VE DEV ATRIYUMLARI OLAN BIR HASTADA KARDIYOVOKAL SENDROM

ÖZET

Kardiyovokal sendrom veya Ortner sendromu genişlemiş kardiyak yapıların sıkıştırmasına sonucu gelişen sol laringeal rekürren sinir palsisine bağlı ses kısıklığıdır. Mitral kapak hastalığı ile olan genişlemiş sol atriyum bu nadir sendromun iyi bilinen sebebidir ancak başka kardiyovasküler durumlarda patogeneze katkida bulunabilmektedir. Mevcut veriler sadece genişlemiş sol atriyumun değil, sol rekürren laringeal sinirin genişlemiş hipertansif pulmoner arter, aorta ve ligamentum arteriosum arasında sıkıştığını göstermektedir. Bu bağlamda, pulmoner arter anevrizması ve dev atriyumları ve düzeltilmiş atriyal septal defekti olan bir hastada kardiyovokal sendrom olgusunu sunduk. Hasta iki yıldır kademeli olarak artan ses kısıklığı ile başvurdu ve laringoskopide sol vokal kord paralizi tespit edildi. Transtorasik ekokardiyografi ile yapılan kardiyovasküler incelemede, kardiyovokal sendrom ile uyumlu olarak pulmoner arter anevrizması ve dev atriyumlar mevcuttu.

Anahtar sözcükler: ses kısıklığı, kardiyovokal sendrom, pulmoner arter, anevrizma

Ortner's syndrome or cardiovocal syndrome, which was first described in 1897 by Norbert Ortner, refers to recurrent laryngeal nerve palsy secondary to cardiovascular disease (1). The most common historical cause is a dilated left atrium due to mitral stenosis, however several case reports suggested that cardiac hoarseness can be caused by some other clinical situations with dilated cardiac structures such as aortic or pulmonary artery aneurysms. True pulmonary artery aneurysms are rare and usually associated intrinsic weakness of the arterial wall which is related with increased hemodynamic shear stresses. Here, we presented a cardiovocal syndrome in a patient with pulmonary artery aneurysm, giant atria and corrected atrial septal defect.

Case report

A 62- year-old female patient admitted with hoarseness. She was suffering gradual increase in hoarseness for two years and she had been hospitalized several times with cardiac decompensation in last six months. She also had exertional dyspnea and peripheral edema. She was a non-smoker, in medical history; she had undergone atrial septal defect (ASD) repair and mitral valve replacement for rheumatic mitral stenosis 7 years ago. Laryngoscopy revealed

Gönderilme Tarihi: 24 March 2014 • **Revizyon Tarihi:** 19 June 2014 • **Kabul Tarihi:** 31 July 2014
İletişim: Serkan Duyuler • **E-Posta:** serkanduyuler@yahoo.com

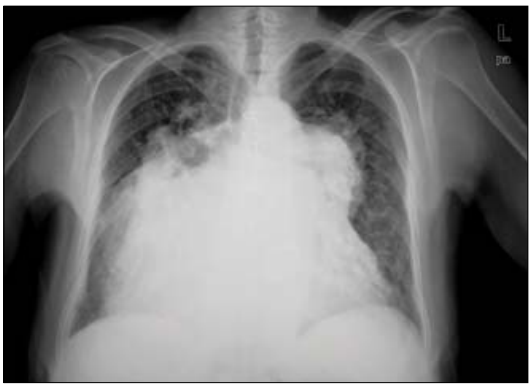

Figure 1. Chest X-ray depicting dilated left atrium, right chambers and pulmonary conus.

a left vocal cord paralysis. Since she had cardiac operation history and cardiac complaints, she was referred for cardiologic evaluation. She was orthopneic and jugular veins were distended. Auscultation revealed rales in basal and mid zones of the lung. Mechanical valve sounds, tricuspid and pulmonary regurgitation murmurs were audible. Pitting edema over tibia was present. Figure 1 shows chest X- ray of the patient. Transthoracic echocardiography confirmed a 6.9 cm pulmonary artery aneurysm (PAA) which was previously reported as only moderately dilated in perioperative evaluation (Figure 2A,2B). Left (65x110 mm) and right (58x105 mm) atria were severely dilated which were similar with perioperative evaluation according to postoperative discharge report (Figure 3A). Left ventricle was normal in size but was D-shaped secondary to high right ventricular pressure. On interatrial septum, thickening compatible with septal repair was noted (Figure 3B). Pulmonary arterial pressure was estimated 70 mmHg. Hoarseness was thought to be secondary to enlarged cardiac structures compressing on laryngeal recurrent nerve and diagnosed as cardiovocal syndrome. Patient was relieved by IV diuretic treatment; however hoarseness persisted almost with no change. No further intervention was planned due to patient's desire and high operational risk.

Discussion

Recurrent laryngeal nerve paralysis has been categorized as non-surgical paralysis, surgical paralysis or a combination of the two according to otorhinolaryngologic approach (2). Besides, cardiac surgery itself may be reason for recurrent laryngeal nerve paralysis. Surgical manipulations, duration of surgery, cardiopulmonary bypass and tracheal intubation are likely to be related to incidence of laryngeal nerve palsy (3). Cardiac causes in addition to mitral stenosis that may lead to this problem, such as patent ductus arteriosus, aneurysm of the aortic arch, aneurysm of the pulmonary artery, Eisenmengers syndrome, pulmonary hypertension, atrial and ventricular septal defect have also been described. Contrary to common thought vocal cord paralysis seems to be triggered by compression of the left recurrent laryngeal nerve in the window area between enlarged hypertensive pulmonary artery, the aorta, and the ligamentum arteriosum and not by dilated left atrium (4) (see Figure 4).

Pulmonary artery aneurysm defined as a pathologic dilatation to more than 1.5 times the normal artery, is a rare entity (5). Uncorrected congenital heart diseases such as patent ductus arteriosus, ventricular septal defect and ASD causing both pressure and volume overload on pulmonary circulation are the leading risk factors for development of high pressure PAA (6,7). As we witnessed in our case, after a certain point, correction of underlying cardiac abnormalities such as ASD and valvular disease may not avert the dilation process going downhill. Also chance of recovery from hoarseness is closely related with degree and duration of injury. Surgery for definitive treatment of cardiac condition may liberalize the compressed recurrent laryngeal nerve.

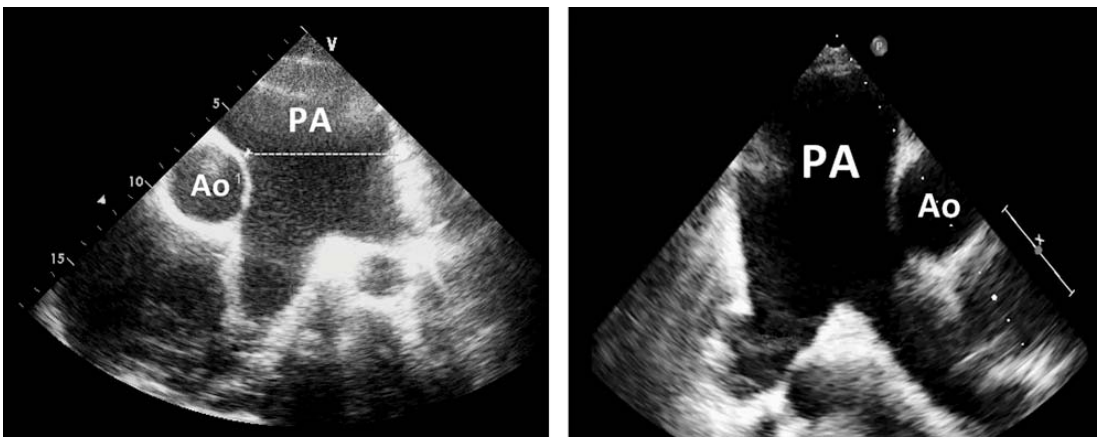

Figure 2. **A.** Parasternal short axis view showing dilated main pulmonary artery. **B.** modified parasternal view showing pulmonary artery and braches. Ao: aorta, PA: pulmonary artery

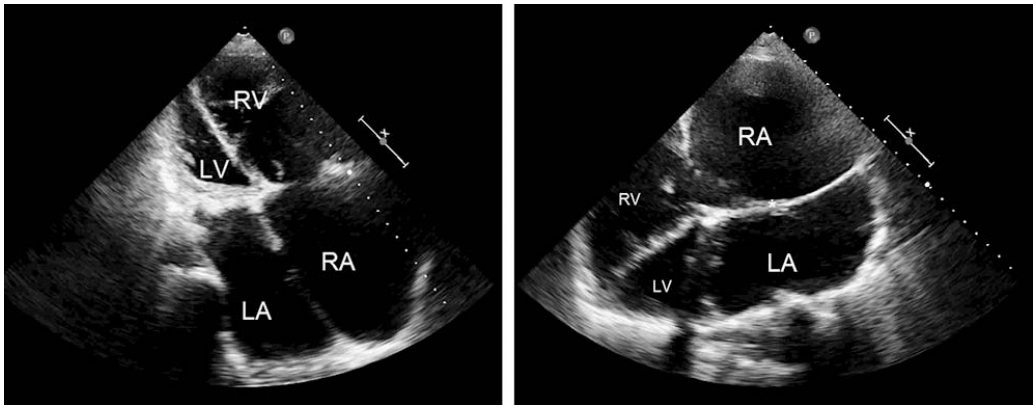

Figure 3. **A.** Apical 4 chambers view depicting dilated atria and right ventricle. **B.** parasternal view showing interatrial septum thickening. **LA:** left atrium, **LV:** left ventricle, **RA:** right atrium, **RV:** right ventricle.

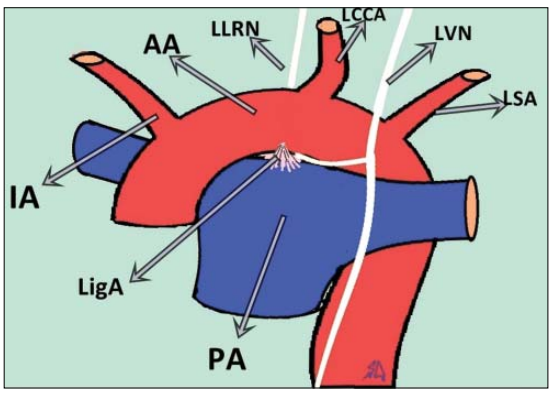

Figure 4. Compression of left laryngeal recurrent nerve between the dilated pulmonary artery, aorta and ligamentum arteriosum. **AA:** aortic arc, **IA**: innominate artery, **LigA:** ligamentum arteriosum, **LCCA:** left common carotid artery, **LLRN:** left laryngeal recurrent nerve, **LSA:** left subclavian artery, **LVN:** left vagus nerve, **PA:** pulmonary artery.

Elimination of compression over the nerve may reverse the hoarseness if the duration is not critically long or nerve is not irreversibly damaged. On the other hand, ischemia related nerve damages are not associated with good prognosis. TTE is very helpful for evaluation of both aneurysm and accompanying cardiac abnormalities. Computed tomography provides good quality images of aneurysm and related thoracic structures (8). But, radiation exposure and using contrast agent in a patient with renal insufficiency who is taking nephrotoxic drugs like diuretics may be problematic. However additional magnetic resonance imaging or computed tomography evaluation is crucial for differential diagnosis of hoarseness and echocardiographic evaluation may not satisfactory for definitive diagnosis.

PAA may compress surrounding structures such as coronary arteries, recurrent laryngeal nerve, and esophagus leading to chest pain, hoarseness and dysphagia respectively (9, 10). As we observed in our case, new onset hoarseness may be a clue for PAA in a patient who already has giant atria at the time of surgery. Additionally, hoarseness may not be overt until severe nerve damage ensued. For this reason, before occurrence of irreversible damage, routine laryngoscopy examination of the vocal fold in heart disease with potential of structural enlargement may be advocated (2). Also, detection of left vocal cord paralysis during laryngoscopy should remind cardiovascular structural enlargement. Conflict of interest: none

References

1. Ortner N. Recurrenslahmung bei mitral stenose. Wien Klin Wochenschr. 1897;10:753–5.
2. Subramaniam V, Herle A, Mohammed N, Thahir M. Ortner's syndrome: case series and literature review. Braz J Otorhinolaryngol. 2011;77:559-62.
3. Kawahito S, Kitahata H, Kimura H, Tanaka K, Oshita S. Recurrent laryngeal nerve palsy after cardiovascular surgery: relationship to the placement of a transesophageal echocardiographic probe. J Cardiothorac Vasc Anesth. 1999;13:528-31.
4. Plastiras SC, Pamboucas C, Zafiriou T, Lazaris N, Toumanidis S. Ortner's syndrome: a multifactorial cardiovocal syndrome. Clin. Cardiol. 2010;33:E99–E100
5. Norman PE, Powell JT. Site Specificity of Aneurysmal Disease. Circulation 2010, 121:560-568
6. Cevik C, Izgi C, Boztosun B. A Rare Consequence of Uncorrected Atrial Septal Defect: Diffuse Pulmonary Artery Aneurysms. Tex Heart Inst J. 2004;31:328-9.
7. Veldtman GR, Dearani JA, Warnes CA. Low pressure giant pulmonary artery aneurysms in the adult: natural history and management strategies. Heart 2003;89:1067–1070
8. Shankarappa RK, Moorthy N, Chandrasekaran D, Nanjappa MC.Giant Pulmonary Artery Aneurysm Secondary to Primary Pulmonary Hypertension. Tex Heart Inst J 2010;37:244–245.
9. Oz F, Emet S, Baykiz D, Oflaz H. Left main coronary artery compression by a giant pulmonary artery aneurysm associated with large atrial septal defect and severe pulmonary hypertension. Anadolu Kardiyol Derg 2011;11:E28-9.
10. Raj V, Gopalan D, Stewart S, Dunning J. Unusual cause of hoarseness of voice: giant pulmonary artery aneurysm. Ann Thorac Surg. 2011;91:285-7.