CASE REPORT

Case of Cardio-vocal Syndrome with Dysphagia Aortica: A Rare Entity

Manwinder Singh Walia[1], Ruchika Bhagat[2]

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Abstract

Hoarseness is a commonly encountered symptom with several underlying causes ranging from idiopathic to potentially severe diseases. Cardio-vocal syndrome/Ortner syndrome (OS) is a rare entity that presents a challenge due to its tendency to evade detection over prolonged periods. Ortner syndrome, characterized by swallowing difficulty due to extrinsic compression of the esophagus by an atherosclerotic thoracic aorta, further adds to the complexity of the diagnosis. Here we report a case of OS and dysphagia aortica in a 62-year-old male with a history of chronic smoking and cardiovascular comorbidities. This case emphasizes the significance of considering cardiovascular etiologies in unilateral vocal cord paralysis (UVCP) evaluation.

Keywords: Case report, Cardio-vocal syndrome, Dysphagia aortica, Hoarseness, Ortner syndrome, Unilateral vocal cord paralysis.

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Abbreviations used in this Aritcle

CAD = Coronary artery disease; CECT = Contrast-enhanced computed tomography; COPD = Chronic obstructive pulmonary disease; OS = Ortner syndrome; TAA = Thoracic aortic aneurysms; UVCP = Unilateral vocal cord paralysis.

Introduction

Hoarseness characterized by a weakened or altered voice, is a frequently encountered symptom that can indicate various underlying conditions ranging from benign and reversible to potentially life-threatening malignancies. Unilateral vocal cord paralysis (UVCP) may exhibit a spectrum of symptoms from being asymptomatic to experiencing various degrees of dysphonia such as breathy voice, vocal fatigue, or complete aphonia.[1]

Diagnosing UVCP definitively entails obtaining a detailed clinical history, conducting a meticulous examination, and conducting investigations to pinpoint the root cause. It usually arises from damage to the recurrent laryngeal nerve, which may be idiopathic or attributed to factors such as cancer, trauma, chest pathology, or surgical procedures.

Ortner's syndrome, initially documented in 1897, is an uncommon medical condition known as left recurrent laryngeal nerve paralysis caused by an identifiable cardiovascular disease, also referred to as cardio-vocal syndrome.[2] Cardio-vocal syndrome typically peaks in incidence among older individuals; however, it can occur across all age groups, including infants, as documented in medical literature. The condition seems to have a higher incidence among men, potentially attributable to a greater prevalence of cardiovascular conditions in this demographic.[3]

Dysphagia can be categorized based on its location, either oropharyngeal or esophageal, and it arises from neuromuscular motility disorders as well as intrinsic or extrinsic mechanical factors. Extrinsic compression of the esophagus, though uncommon, can be attributed to various factors including mediastinal masses (such as lymphadenopathy, lung cancer, etc.), postoperative fibrotic changes, and cardiovascular conditions like dysphagia aortic, dysphagia lusoria, an enlarged left atrium, or significant pericardial effusion. Dysphagia aortica is a rare condition characterized by swallowing difficulties due to external pressure on the esophagus by an abnormally enlarged, twisted, or atherosclerotic bulging thoracic aorta.[4]

In this report, we present a unique case of hoarseness secondary to Ortner's syndrome and dysphagia aortica providing insights into the underlying pathoanatomical mechanisms.

Case Description

A 62-year-old male patient presented at the ENT outpatient department with a history of gradually worsening hoarseness of voice for a year. Upon further inquiry, it was revealed that the patient has had dysphagia, particularly with solid foods, for the past 7 months which was also gradually progressing. The patient has been a chronic smoker for the past 30 years, with a history of over 20 pack years. Additionally, he has a medical history of hypertension, chronic obstructive pulmonary disease (COPD), and coronary artery disease (CAD), for which he has been on medication for the past 5 years.

[1,2]Department of ENT, Government Multi Specialty Hospital, Sector 16, Chandigarh, India

Corresponding Author: Ruchika Bhagat, Department of ENT, Government Multi Specialty Hospital, Sector 16, Chandigarh, India, Phone: +91 7009290073, e-mail: ruchikabhagat62@gmail.com

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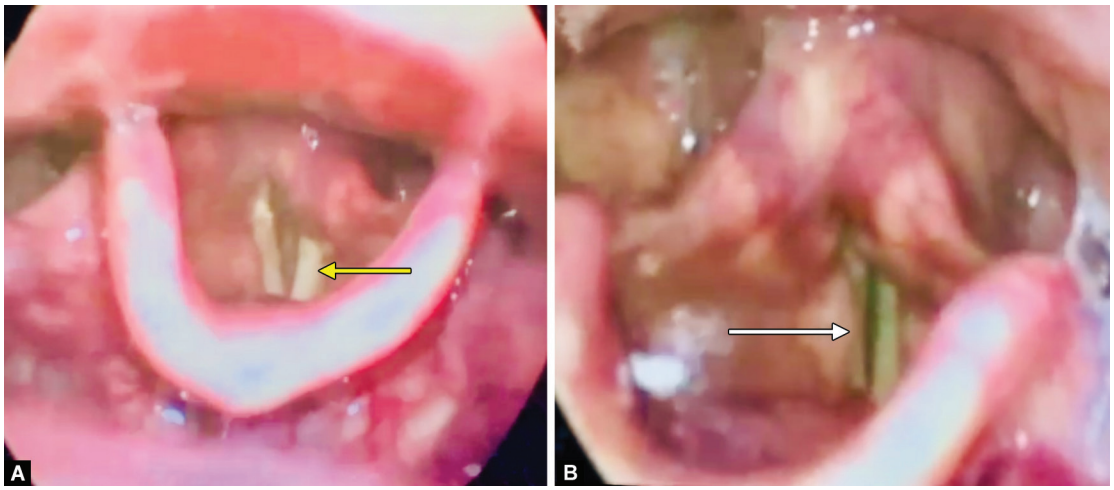
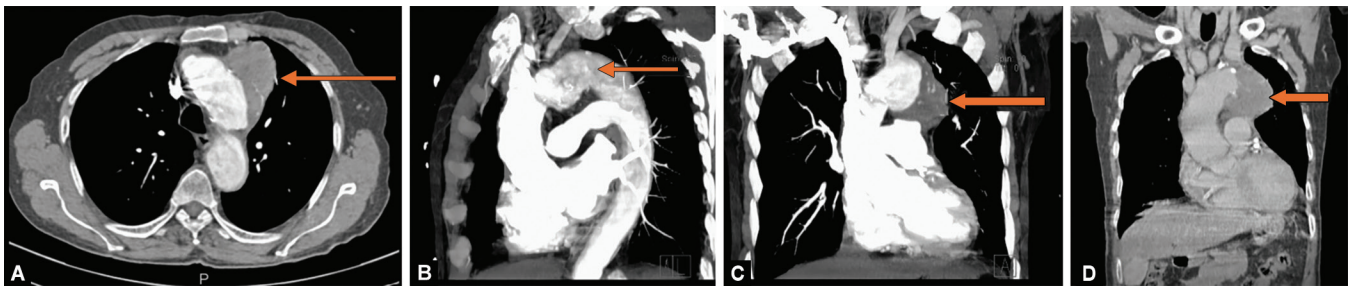


Figs 1A and B: Video laryngoscopy images showing, (A) Non-mobile left vocal cord (yellow arrow); (B) Compensatory movement of right vocal cord during adduction (white arrow)



Figs 2A to D: Contrast-enhanced computed tomography image of chest, (A) Axial; (B) Sagittal; (C) Coronal; (D) Non-enhanced coronal sections well-defined lesion measuring arising from the anterior border of the aortic arch with minimal calcifications representing thrombosed thoracic aortic aneurysm (orange arrow)

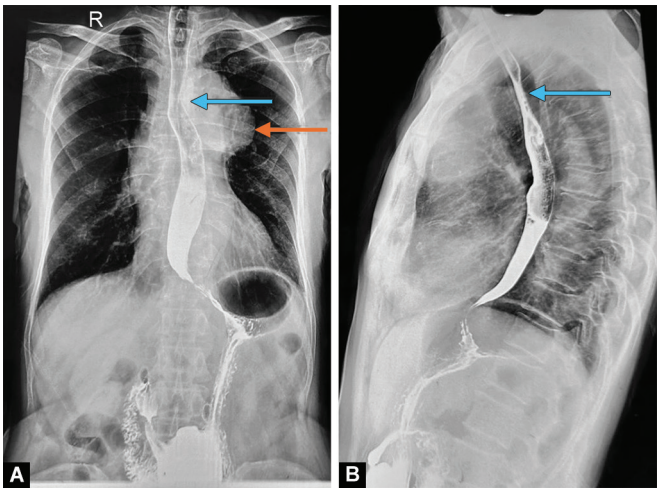
Video laryngoscopy confirmed that there was left-sided vocal cord paralysis with preserved right vocal cord mobility. No other abnormalities were noted on examination of the head and neck region (Fig. 1).

Contrast-enhanced computed tomography (CECT) imaging of the neck and chest was performed to investigate the etiology of vocal cord paralysis in our patient. The CECT scan revealed a well-defined non-enhancing hypodense lesion measuring approximately 5.0 × 4.3 × 5.7 cm arising from the anterior border of the aortic arch with minimal calcifications. A crescentic plaque measuring approximately 11.0 mm in thickness with interrupted intimal calcifications was observed, indicative of a thrombosed aneurysm compressing the left recurrent laryngeal nerve. Additionally, Para septal and centrilobular emphysematous changes were noted bilaterally in the lungs (Fig. 2).

For diagnosing the cause of dysphagia patient was subjected to a barium swallow test that revealed external compression of esophagus by an enlarged aortic arch aneurysm (Fig. 3).

Based on clinical presentation and above-mentioned imaging findings, a diagnosis of cardio-vocal syndrome with dysphagia aortic secondary to thoracic aortic aneurysm compressing the left recurrent laryngeal nerve as well as esophagus was established.

The patient was referred to the Cardiothoracic Surgery Department for further management of the thoracic aortic aneurysm. Surgical intervention was recommended to relieve the compression on the left recurrent laryngeal nerve and to prevent potential complications associated with the aneurysm.



Figs 3A and B: Barium swallow showing, (A) Coronal; (B) Sagittal sections of the compression in the oesophagus (blue arrow) at the level of thoracic aortic aneurysm (orange arrow)

Discussion

The main source of innervation to the vocal folds originates from branches of the Vagus nerve, specifically the superior and recurrent laryngeal nerves. The term "recurrent laryngeal nerve" reflects its distinctive path beneath the arch of the aorta on the left side and under the subclavian artery on the right side.

Injury to the left recurrent laryngeal nerve can occur at any point along its course. Several potential causes of UVCP are recognized, encompassing idiopathic, iatrogenic, malignancy, and less frequently post-radiation fibrosis or inflammation. In certain instances, due to a narrow gap between the aorta and the pulmonary artery, it gets compressed.

Left-sided vocal cord paralysis occurs 1.75 times more frequently than right-sided paralysis as the left recurrent laryngeal nerve's anatomical course renders it vulnerable to compression, particularly by adjacent vascular structures like the aorta.[5] Unilateral left recurrent laryngeal nerve injury leads to UVCP resulting in hoarse voice. When this is caused by an underlying cardiovascular pathology it is known as Ortner's syndrome or cardio-vocal syndrome, as seen in our case.

In a recent meta-analysis examining Ortner syndrome (OS), authors identified a total of 117 patients that were reported in 92 published articles within English literature over the period of 1955 to 2021. This analysis highlights the potential occurrence of OS across all ages demographics and among individuals with diverse cardiovascular pathologies. They retrieved that initially, mitral stenosis was the primary etiology of OS, i.e., from 1955 to 1990. However, after 1990, the investigation revealed a notable shift in the prevalence and emergence of vascular lesions, particularly thoracic aortic aneurysms (TAA) as the predominant etiological factor.[6,7]

Thoracic aortic aneurysms may grow asymptomatically until they rupture, with a mortality over 90%. As a rare condition, the true incidence of dysphagia aortica is unknown, and most cases are identified incidentally after radiologic investigations. Patients often experience delays in management, leading to potential catastrophic complications of TAA. However, this condition can be managed through conservative or surgical approaches. Therefore, dysphagia aortic should always be considered as a differential diagnosis in cases of dysphagia, warranting heightened clinical suspicion.[8]

Conclusion

Ortner syndrome and dysphagia aortica, while uncommon, pose a diagnostic challenge as they frequently elude detection for a prolonged duration. This rarity of both diseases, coupled with their insidious onset often results in delayed diagnosis and management. Hence, this case highlights the importance of considering cardiovascular etiologies, such as TAA, in the evaluation of UVCP and dysphagia. Prompt diagnosis and early intervention are crucial to prevent associated complications. This case report adds to the growing understanding of Ortner's syndrome and dysphagia aortic. Also, emphasizes the need for a multidisciplinary approach involving otorhinolaryngology and cardiothoracic surgery for optimal management of such cases.

Orcid

Ruchika Bhagat https://orcid.org/0000-0002-2814-9929

References

1. Rubin AD, Sataloff RT. Vocal fold paresis and paralysis. Otolaryngol Clin North Am 2007;40(5):1109–1131. DOI: 10.1016/j.otc.2007.05.012.
2. Thirlwall AS. Ortner's syndrome: A centenary review of unilateral recurrent laryngeal nerve palsy secondary to cardiothoracic disease. J Laryngol Otol 1997;111(9):869–871. DOI: 10.1017/s0022215100138848.
3. Loughran S, Alves C, MacGregor FB. Current aetiology of unilateral vocal fold paralysis in a teaching hospital in the West of Scotland. J Laryngol Otol 2002;116(11):907–910. DOI: 10.1258/00222150260369426.
4. Ghazanfar H, Shehi E, Makker J, et al. The role of imaging modalities in diagnosing dysphagia: A clinical review. Cureus 2021;13(7):e16786. DOI: 10.7759/cureus.16786.
5. Song SW, Jun BC, Cho KJ, et al. CT evaluation of vocal cord paralysis due to thoracic diseases: A 10-year retrospective study. Yonsei Med J 2011;52(5):831–837. DOI: 10.3349/ymj.2011.52.5.831.
6. Verma S, Talwar A, Talwar A, et al. Ortner's syndrome: A systematic review of presentation, diagnosis and management. Intractable Rare Dis Res 2023;12(3):141–147. DOI: 10.5582/irdr.2023.01047.
7. Fife R, Muir A. Laryngeal paralysis associated with mitral stenosis (Ortner's syndrome): Report of two cases. Glasgow Med J 1955;36(5):164–167. PMID: 14380803.
8. Gouveia E Melo R, Silva Duarte G, Lopes A, et al. Incidence and prevalence of thoracic aortic aneurysms: A systematic review and meta-analysis of population-based studies. Semin Thorac Cardiovasc Surg 2022 Spring;34(1):1–16. DOI: 10.1053/j.semtcvs.2021.02.029.