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CASE REPORT

INTERMEDIATE

CLINICAL CASE

Multidisciplinary Approach to Isolated Pulmonary Vein Compression by an Enlarging Vertebral Osteophyte

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ABSTRACT

Compression of mediastinal structures by vertebral osteophytes is rare. We report a case of pulmonary vein compression by a vertebral osteophyte that failed stenting. A minimally invasive approach to osteophyte removal with subsequent re-expansion angioplasty yielded an optimal outcome, negating the need for cardiopulmonary bypass, stent removal, and pulmonary venoplasty. (**Level of Difficulty: Intermediate.**) (J Am Coll Cardiol Case Rep 2022;4:145–149) © 2022 The Authors. Published by Elsevier on behalf of the American College of Cardiology Foundation. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

HISTORY OF PRESENTATION

A 75-year-old man presented with worsening dyspnea on exertion and decreased exercise tolerance over the course of a year. He denied chest pain, syncope, edema, orthopnea, or paroxysmal nocturnal dyspnea. A computed tomography (CT) scan of his chest showed evidence of a bicuspid aortic valve with associated aortopathy (43 mm at sinuses), pectus excavatum with a Haller Index of 4.9 (Figure 1A), and significant stenosis of his right inferior pulmonary vein (RIPV) (lumen measuring 9 × 4 mm, down from 18 × 9 mm previously) with gradual evolution in size of a corresponding vertebral osteophyte at the T_8 level (Figures 1B and 1C).

LEARNING OBJECTIVES

- To identify extrinsic vertebral osteophyte compression as a potential cause of cardiac structure compression.
- To understand the optimal treatment paradigm requiring osteophyte excision and adjuvant endovascular therapy to provide full long-term expansion of a stenotic pulmonary vein.

PAST MEDICAL HISTORY

His pertinent history was notable for paroxysmal atrial fibrillation/flutter on anticoagulation and treated with multiple ablations (including a cryo-opulmonary vein isolation and cavotricuspid isthmus ablation done 1 year ago and a repeat pulmonary vein isolation as well as flutter ablation done 6 months ago), bicuspid aortic valve, chronic obstructive pulmonary disease, hyperlipidemia, pectus excavatum, and a previous left upper lobectomy for multiple non-necrotizing pulmonary nodules 15 years ago.

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ABBREVIATIONS AND ACRONYMS

CT = computed tomography

RIPV = right inferior pulmonary vein

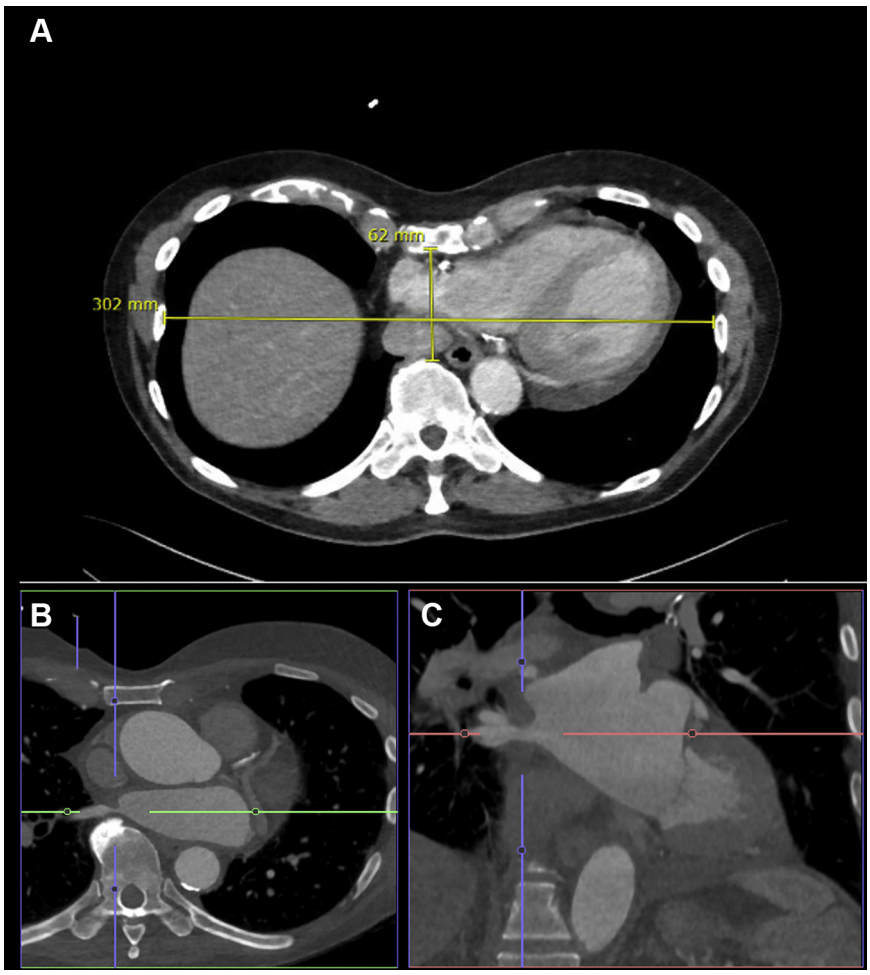
DIFFERENTIAL DIAGNOSIS

Multiple factors could contribute to the patient's dyspnea, including recurrent atrial fibrillation/flutter, possible aortic stenosis in setting of known bicuspid aortic valve, chronic obstructive pulmonary disease, depressed lung reserve given remote history of lobectomy, and worsening RIPV stenosis.

INVESTIGATIONS

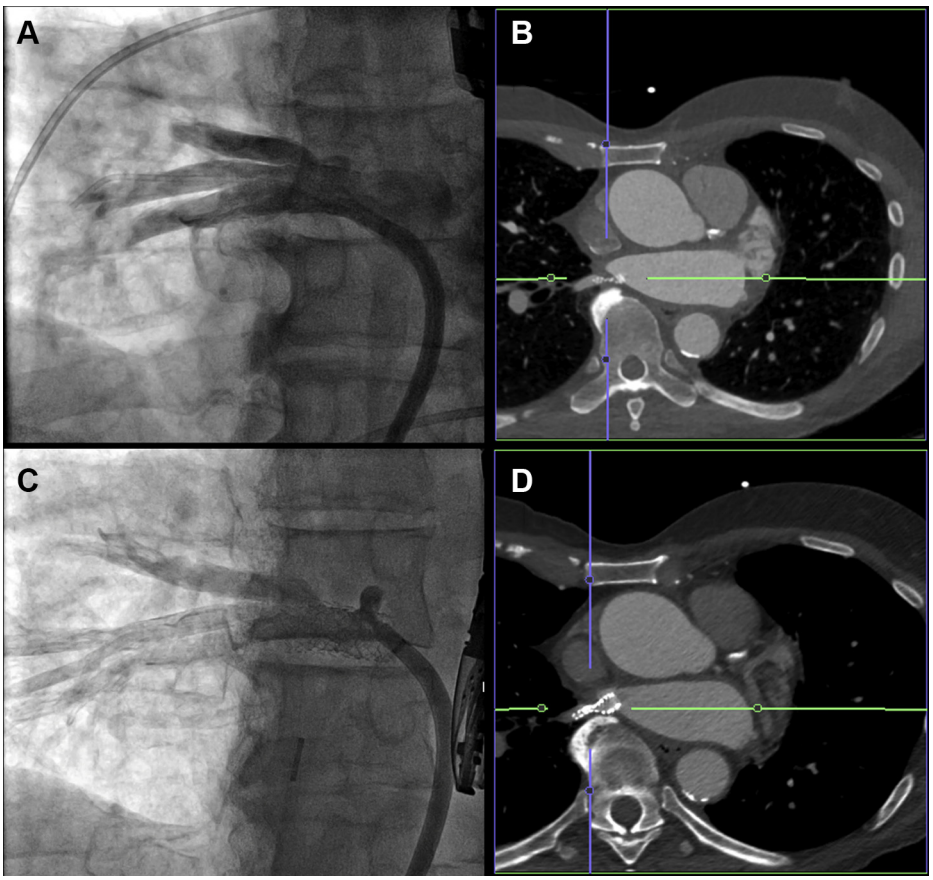
The patient was in sinus rhythm and noted to have no instances of arrhythmias on transtelephonic monitoring since his most recent ablation. A quantitative lung perfusion scan was done showing 41.8% and 58.2% pulmonary perfusion to the left and right lung, respectively. Transthoracic echo demonstrated normal left ventricular function (60%), mild aortic regurgitation, a bicuspid aortic valve with right to left fusion, and a peak/mean gradient of 14/7 mm Hg. The patient underwent heart catheterization, which showed mildly elevated right-sided pressures, cardiac index of 2.85, and a gradient across the right pulmonary arterial wedge pressure and the direct left atrial pressure. Angiography of the RIPV demonstrated an eccentric lesion with 80% narrowing and an 8 mm Hg gradient across the RIPV by direct measurement (Video 1). A bare-metal stent (10 × 19 mm Omnilink, Abbott) was placed with no residual stenosis

FIGURE 1 Chest Computed Tomography



(A) Axial cut demonstrating severe pectus excavatum in addition to leftward shift of the heart with previous left upper lobectomy. **(B and C)** Right inferior pulmonary vein stenosis on computed tomography chest with axial and coronal cuts, respectively.

FIGURE 2 **Angiogram and Chest Computed Tomography Following Intervention**



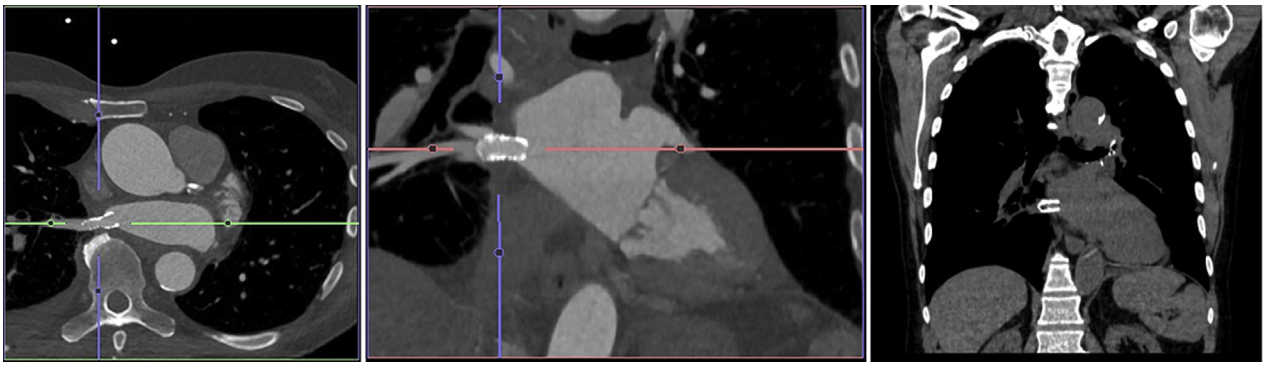
(A) Angiogram showing no residual stenosis of right inferior pulmonary vein following stenting. **(B)** Follow-up computed tomography scan of pulmonary veins demonstrating external stent compression of right inferior pulmonary vein stent. **(C)** Angiogram following second stent placement in right inferior pulmonary vein to provide more radial strength to prevent compression. **(D)** 3-month follow-up computed tomography scan redemonstrating extrinsic compression and narrowing of the peripheral aspect of the right inferior pulmonary vein stents.

(**Figure 2A**). A follow-up CT scan demonstrated external stent compression between the osteophyte and the pectus excavatum and a repeat angiogram and RIPV stenting using a similar bare-metal stent was undertaken with postdilation to 14 atm with the aim of providing more radial strength to prevent external compression (**Figure 2B,** Video 2). Completion angiogram showed no residual stenosis (**Figure 2C**). However, a follow-up CT again demonstrated extrinsic compression and narrowing of the distal aspect of the stents (**Figure 2D**).

MANAGEMENT

The patient was now referred for surgery, and following a multidisciplinary case assessment, the role of the osteophyte in the setting of pectus excavatum was recognized and the decision made for surgical osteophyte excision and subsequent stent expansion. The right video-assisted thoracoscopic approach used for culprit osteophyte excision is shown in Video 3. A working incision was made, and a small retraction port was inserted for optimal exposure. The pleural soft tissue surrounding the osteophyte was dissected using electrocautery. Once the soft tissue was removed and exposure was obtained, neurosurgery used a long-handled drill for osteophyte removal. Once osteophyte removal was deemed adequate, hemostasis was obtained and copious irrigation was instilled to remove any remaining debris. Standard Marcaine intercostal nerve blocks were done. A small-bore chest tube was

FIGURE 3 Chest Computed Tomography



(Left) A representative axial chest computed tomography image 3 months following surgical and endovascular intervention, showing full stent expansion in the right inferior pulmonary vein with corresponding T_8 osteophyte removal. **(Middle)** A coronal computed tomography image of complete right inferior pulmonary vein expansion at 3 months. **(Right)** 1-year follow-up noncontrast, coronal computed tomography scan demonstrates ongoing stent patency.

placed, and the lung was re-expanded under direct visualization. Several days later, angioplasty with a drug-coated balloon and a 9.0 × 20 mm Charger Balloon (Boston Scientific) was performed with immediate angiography demonstrating excellent flow through the stent (Video 4).

DISCUSSION

Structural impingement of key cardiac structures is a rare cause of dyspnea.[1] Potential causes of intrathoracic structural compression include mediastinal tumors, masses, thrombi, or cystic structures. Vertebral osteophytes are fairly common forms of osteoarthritis characterized by osseous outgrowths along articular surfaces.[2] Complications are rare; the most frequent are myelopathy and radiculopathy caused by mechanical vertebral canal compression and dysphagia secondary to esophageal impingement.[3,4] Cases of external compression of the trachea, bronchi, vertebral arteries, nerves, and the left atrium have been described.[5-7] We report a unique case of symptomatic dyspnea secondary to RIPV compression by a vertebral osteophyte with resultant improvement utilizing a hybrid intervention.

With the diagnosis of pulmonary vein stenosis, a multidisciplinary approach to optimal treatment is recommended, utilizing adjunct imaging to completely delineate the pertinent anatomy.[8,9] The patient's history of both severe pectus as well as a previous lung resection surgery narrowed the mediastinal space and shifted the mediastinum to the left, essentially pulling the RIPV across a sizable vertebral osteophyte. Importantly, endovascular treatment alone with stenting failed as a primary intervention. Even with a second stent in place, the radial force exerted was insufficient to allow full expansion.

Efforts at video-assisted thoracoscopic spine surgery have been reported in the literature with excellent outcomes.[10] The video assisted thoracoscopic surgical exposure in this case allowed for excellent visualization of the T_8 osteophyte and its subsequent removal by the neurosurgical team was accomplished with relative ease using a long-handled drill. After removal of the osteophyte, the stent remained fully expanded. In hindsight, removal of the osteophyte should have been undertaken primarily. It is unlikely that an open operation on cardiopulmonary bypass including stent removal and pulmonary venoplasty would have worked.

FOLLOW-UP

Following osteophyte excision and angioplasty, the RIPV stents remained widely patent on repeat 1-month, 3-month, and 1-year follow-up CT scans (**Figure 3**). The patient reported symptomatic improvement confirmed by corresponding improvement in his cardiopulmonary exercise testing.

CONCLUSIONS

Although rare, vertebral osteophytes have been noted to cause compression of mediastinal structures including the esophagus, trachea, or bronchi, and cardiac structures. We present a unique case of extrinsic pulmonary vein compression by a vertebral osteophyte, with compression strong enough to preclude proper stent expansion. A multidisciplinary

approach entailing interventional cardiology, cardiac and thoracic surgery, along with neurosurgery provided correct diagnosis and was critical to ensure success in this rare case.

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KEY WORDS pulmonary circulation, stenosis, stent, thoracic

APPENDIX For supplemental videos, please see the online version of this paper.