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CORONARY, PERIPHERAL, AND STRUCTURAL INTERVENTIONS

CASE REPORT: CLINICAL CASE SERIES

Diaphragmatic Paralysis After Heterotopic Bicaval Stenting



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ABSTRACT

Heterotopic bicaval stenting offers an alternative therapeutic approach for indirectly addressing the systemic effects of tricuspid regurgitation (TR) in patients with severe symptomatic TR, despite optimal medical therapy. In this report, we describe 2 cases of procedure-related unilateral diaphragmatic paralysis, a clinically important complication not previously documented in the literature. (JACC Case Rep 2024;29:102422) © 2024 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/).

CASE 1

HISTORY OF PRESENTATION. An 81-year-old woman with a medical history of long-standing permanent atrial fibrillation (AF), tricuspid regurgitation (TR), and chronic kidney disease presented to the outpatient clinic with a report of worsening peripheral edema and dyspnea, despite optimal medical therapy (OMT) during the previous 3 months.

INVESTIGATIONS AND MANAGEMENT. Transthoracic and transesophageal echocardiography showed

LEARNING OBJECTIVES

- To understand the role of heterotopic bicaval stenting in the management of patients with symptomatic severe TR.
- To recognize potential device-related complications of heterotopic bicaval stenting.
- To be able to diagnose unilateral diaphragmatic paralysis as a potential complication of heterotopic bicaval stenting.

severe eccentric TR, with a vena contracta width of 9.6 mm and a tricuspid annulus diameter of 31 mm (Video 1). Both left and right ventricular function were within normal limits, and no other significant valvular heart disease was detected. Right-sided heart catheterization revealed mild precapillary pulmonary hypertension with a mean pulmonary artery pressure (mPAP) of 28 mm Hg and a pulmonary capillary wedge pressure of 12 mm Hg. Cardiac output, assessed by thermodilution, was reduced to 2.8 L/min. Following deliberation by the heart team, taking into account the patient's age and comorbidities, as well as anatomical aspects of the TR, which were deemed unsuitable for edge-to-edge repair, transcatheter heterotopic bicaval stenting emerged as the preferred therapeutic approach. The implantation of bicaval stents using the TricValve system (P+F Products) proceeded smoothly without intraoperative complications. However, a postoperative chest radiograph (Figure 1C) revealed a noticeably elevated right diaphragm on deep inspiration (2 intercostal spaces) that was not present on the preoperative

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The authors attest they are in compliance with human studies committees and animal welfare regulations of the authors' institutions and Food and Drug Administration guidelines, including patient consent where appropriate. For more information, visit the Author Center.

ABBREVIATIONS AND ACRONYMS

AF = atrial fibrillation

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mPAP = mean pulmonary artery pressure

OMT = optimal medical therapy

TR = tricuspid regurgitation

chest radiograph (Figures 1A and 1B), thus indicating paralysis of the right diaphragm. The patient, however, was asymptomatic, her clinical course uneventful, and she was discharged 2 days after the procedure.

FOLLOW-UP. Three months after discharge, the patient visited the outpatient clinic with

a report of persistent dyspnea and ankle swelling. A new chest radiograph was performed as part of the outpatient follow-up, and it showed persistence of the diaphragmatic paralysis (Figures 1D and 1E). The diagnosis was subsequently confirmed by fluoroscopic examination (Figure 1F, Video 2).

CASE 2

HISTORY OF PRESENTATION. An 83-year-old woman with a long-standing medical history of permanent AF and severe TR visited the outpatient clinic with a report of progressive ankle swelling and dyspnea despite OMT.

INVESTIGATIONS AND MANAGEMENT. Transthoracic and transesophageal echocardiography (Video 3) revealed a severely dilated right atrium, severe TR, and normal right and left ventricular function. The right ventricle was mildly dilated. Right-sided heart catheterization revealed mild precapillary pulmonary



Preoperative (A) posteroanterior and (B) lateral views. (C) Anteroposterior view immediately after the procedure. (D) Posteroanterior and (E) lateral views at 3-month follow-up. (F) Fluoroscopic evaluation at 3-month follow-up. Horizontal lines indicate the space between the left and right hemidiaphragm. Arrows mark the heterotopic bicaval stents.



Preoperative (A) posteroanterior and (B) lateral views. (C) Postoperative (C) posteroanterior and (D) lateral views. (E) Posteroanterior and (F) lateral views 3 days after the procedure. Horizontal lines indicate the space between the left and right hemidiaphragm. Arrows mark the Heterotopic bicaval stents.

hypertension (mPAP, 27 mm Hg). The valve was considered suboptimal for an edge-to-edge repair. Accordingly, heterotopic bicaval stenting using the TricValve system was performed without intraoperative complications. However, postoperatively the patient reported increased dyspnea, and a chest radiograph (**Figures 2C and 2D**) displayed a prominently elevated right diaphragm, a finding absent in the preoperative chest radiograph (**Figures 2A and 2B**), thus indicating the possibility of diaphragmatic paralysis.

FOLLOW-UP. The patient's dyspnea improved significantly during the next days of hospitalization, and a chest radiograph before discharge (Figures 2E and 2F) revealed an only slightly elevated right diaphragm compared with the left diaphragm, therefore indicating full recovery of the diaphragmatic paralysis.

DISCUSSION

Symptomatic TR poses a notable risk to prognosis if it is left untreated.¹ In recognizing its clinical significance, several transcatheter therapeutic strategies have emerged as alternatives to high-risk TR surgery.^{2,3} Many of these approaches center on edge-toedge repair or orthotopic replacement, with their success contingent on anatomical suitability.2 A more recent development is heterotopic bicaval stenting, offering an alternative method for indirectly addressing the systemic effects of TR.⁴ Although this procedure has demonstrated relative safety, some procedure-related complications have been reported. The most frequently described complications include vascular complications, bleeding events, transient shoulder pain, the need for conversion to surgery, and device embolization (Table 1).4,5

TABLE 1 Device-Related Complications of Heterotopic Bicaval Stenting	
Device-Related Complications	Incidence, %
Transient shoulder pain	28.5 ⁴
Major bleeding	12-17.1 ^{4,5}
Device embolization	0-8 ^{4,5}
Conversion to surgery	4 ⁵
Procedure-related death	0 ^{4,5}

Here we describe 2 cases of procedure-related unilateral diaphragmatic paralysis, a clinically significant complication not previously documented in the literature. Although previous reports highlighted the occurrence of transient shoulder pain following heterotopic bicaval stenting as relatively common (believed to stem from phrenic nerve compression induced by the inferior vena cava prosthesis), our cases present a new complication.⁴ It is hypothesized that a similar pathophysiological mechanism may underlie the unilateral diaphragmatic palsy observed in our patients. Although this complication resolved transiently in 1 patient, there was no improvement observed at the 3-month follow-up in the other patient. In the 2 cases, no technical difficulties or issues were detected during the procedure, and deployment of the device was smooth at the intended target location.

Prosthesis oversizing may be a possible explanation for this complication, thus highlighting the importance of a preoperative computed tomography work-up for selecting a valve size that precisely matches the patient's anatomy. However, in our cases, both patients underwent thorough preoperative evaluations (Supplemental Figures 1 and 2), and the valve sizes were chosen with careful consideration of the individual anatomies of their superior and inferior venae cavae. Thus, oversizing is unlikely to explain the occurrence of diaphragmatic paralysis in our patients. Furthermore, implementation of electromyographic monitoring techniques during the procedure may help prevent potential harm to the phrenic nerve. These techniques have shown good outcomes in electrophysiologic procedures such as pulmonary vein isolation through cryoablation or radiofrequency ablation.⁶ However, these techniques have not been tested in the context of heterotopic bicaval stenting.

The diagnosis of unilateral diaphragmatic palsy can typically be made using a simple chest radiograph in 90% of cases, and in cases of uncertainty, confirmation may be obtained through fluoroscopic imaging.⁷ Given the possibility of an initial asymptomatic presentation, the true incidence of this condition may be higher because some cases could go unrecognized without routine screening. Thus, we propose that routine chest radiographs following heterotopic bicaval stenting could be beneficial for early detection. This approach would enable prompt identification of diaphragmatic paralysis and facilitate timely initiation of appropriate follow-up diagnostics as needed.

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KEY WORDS complication, tricuspid valve, valve replacement

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