

Noblestitch Failure After Percutaneous Patent Foramen Ovale Closure in a Case Of Platypnea-Orthodeoxia Syndrome: Is this Device Suitable for All Patients?

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Introduction

Patent foramen ovale (PFO) is a common congenital cardiac lesion corresponding to a normal fetal interatrial communication that typically closes after birth with the fusion of the atrial septum primum to the septum secundum.¹ When the foramen ovale persists, it is usually asymptomatic and benign, with no need for treatment.² In rare cases, it can be implicated in the pathogenesis of some medical conditions, such as platypnea-orthodeoxia syndrome (POS). In this context, PFO closure may be indicated.²

Several occluder devices, traditionally based on a double-disc design, have shown that percutaneous PFO closure is possible.³ Despite the proven efficacy, their use is not technically feasible for some patients. In addition, they have the potential to cause some complications.⁴ NobleStitch EL (NS) is an alternative percutaneous PFO closure technique available in Europe, European Commission (EC) marked for cardiovascular suturing and PFO closure, and in the US, with Food and Drug Administration (FDA) clearance for vascular and cardiovascular suturing. It consists of a “deviceless” system with two dedicated suture delivery catheters.^{5,6} To date, the experience with the NS device is limited, and we still lack information about its failure determinants, which could help select patients.⁶⁻⁸

Herein, we describe the case of a failed percutaneous PFO closure with NS that could add to the evidence that interatrial septum (IAS) anatomy may play a key role in the success of the procedure.

Clinical Case

We report the case of a 46-year-old man with a history of hypertension and acromegaly in the context of a pituitary adenoma, surgically removed in June 2020, and secondary

panhypopituitarism developed after the procedure. He went to the emergency department complaining of 6-week exertional dyspnea. On examination, he was polypneic but hemodynamically stable, with no cardiac murmurs or adventitious lung sounds. Blood gases showed a severe type 1 respiratory failure, with a PaO₂ of 33 mmHg in room air. Electrocardiogram (ECG) revealed no relevant anomalies in sinus rhythm. His blood test results had no significant abnormalities, with hemoglobin of 14 g/dL and no elevation of inflammatory parameters. A chest and abdominal computed tomography (CT) angiogram (Figure 1A) ruled out the diagnosis of pulmonary thromboembolism and interstitial lung disease. The results described an abnormality in the normal liver morphology, with atrophy of the fourth segment, and kyphoscoliosis, determining a deformity of the anterior right costal grid. The oxygen saturation obtained in pulse oximetry was lower while the patient was standing or sitting. POS was then established. The patient was admitted for further study.

A transthoracic echocardiogram (TTE – Figure 1B) showed left ventricular hypertrophy, aneurysmatic IAS, and mild aortic root ectasia of 39 mm. Additionally, it revealed an extrinsic compression of the right atrium by the ascending thoracic aorta and the liver. No other relevant findings were identified, including the stigma of pulmonary hypertension. Transesophageal echocardiography (TEE – Figure 1C; Video 1) corroborated the presence of PFO, which showed a spontaneous bidirectional flow. Right-sided catheterization confirmed that right chamber pressures were not increased; Qp/Qs in recumbent position was: 1.08.

Since the patient was clearly symptomatic, the decision was to proceed with percutaneous PFO closure. Given the marked right atrium deformation, he was considered a poor candidate for the placement of a traditional umbrella-like occluder device due to the high risk of wall erosion. The alternative chosen was the NS technique guided by fluoroscopy (Figure 2A) and TEE. Two sutures were placed due to the presence of a significant residual shunt after the first one (Figures 2B, 2C, and 2D; Video 2). After 3 days, the patient complained of recurrent dyspnea while standing. Re-evaluation TTE showed the persistence of patency of the foramen ovale, and TEE revealed two linear echogenic structures compatible with previously implanted devices at the septum secundum, but detached from the septum primum (Figure 3A and 3B; Video 3). The patient was subsequently referred to and accepted for surgical PFO closure with a bovine pericardium patch, which was performed without complications. Postoperative TEE showed no residual shunt. The patient was discharged after 6 days. At 6-month follow-up, he was asymptomatic, reporting a significant improvement in quality of life.

Keywords

Heart Defects, Congenital; Foramen Ovale, Patent; Platypnea-Orthodeoxia Syndrome; NobleStitch/methods; Atrial Septum.

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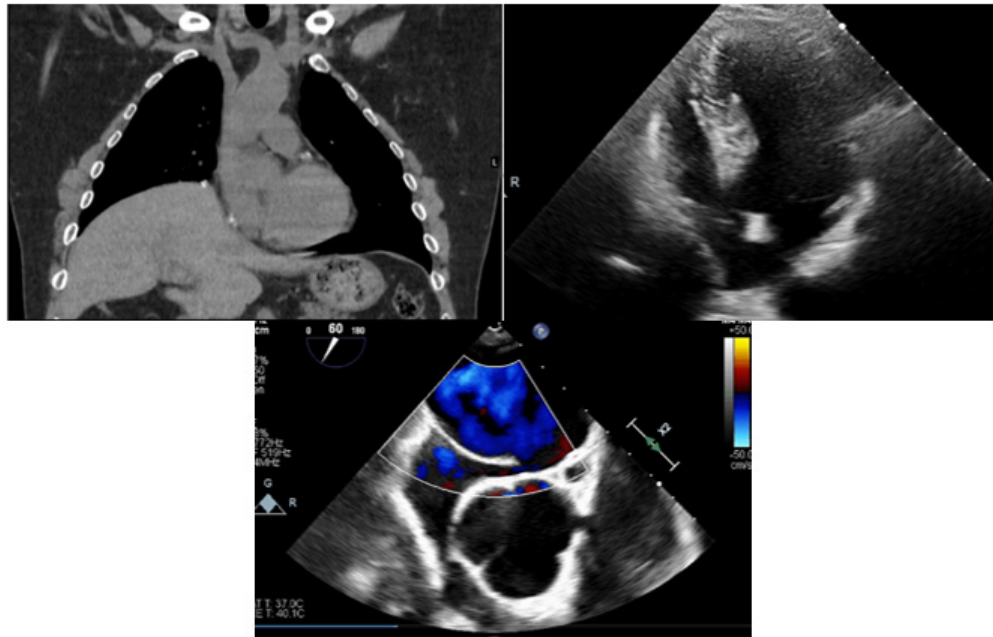
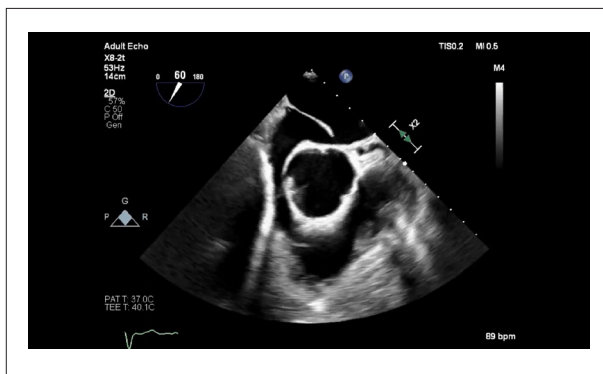


Figure 1 – Pre-procedure percutaneous images. A: CT showing thoracic deformation with close relationship between right atrium and the liver; B: TTE showing right atrium compression; C: TEE mid-esophageal short axis view at 60° showing right atrium compression and PFO.



Video 1 – Pre-procedure TEE showing right atrium compression and PFO with spontaneous bidirectional flow.

Link: <http://abcimaging.org/supplementary-material/2023/3601/ABC-309-video01.mp4>

Discussion

POS is a rare syndrome caused by a right-to-left shunt (RLS) found in the heart or lungs. Most cases originate from an intracardiac shunt, which occurs when a structural component, such as interatrial communication, coexists with a functional component that favors an RLS in the upright position.⁹ This can happen with conditions that affect the position and anatomy of the heart, dictating a transient increase in pressure in the right atrium and/or favoring the flow direction from the inferior vena cava directly through the septal defect, such as: aortic dilatation (one of the most common), diaphragmatic paralysis, post-pneumonectomy, kyphoscoliosis, prominent Eustachian

valve or Chiari network, lipomatous hypertrophy of the IAS, or pericardial disease.¹⁰

In our patient, thromboembolic disease, arteriovenous malformations, and interstitial lung disease were excluded by chest CT as plausible causes of POS. In the absence of abnormal liver function tests, hepatopulmonary syndrome seemed also unlikely. The PFO described in TEE prevailed as the only reliable cause for the syndrome manifestation. In our case, aortic ectasia, thoracic deformation, and liver distortion seemed to be the structural cause interfering with the heart, favoring the appearance of intracardiac shunt.

The treatment for these patients consists of closing the atrial septal defect.² This option should be considered after ruling out other alternative causes of POS; when it has a significant impact on the patient's functional capacity; and there are no increased right chamber pressures.² Our patient met all these criteria.

Whenever possible, percutaneous closure is preferred. Double-disc occluder devices are the most used in these procedures;³ however, they were not considered a good option for our patient, given the atypical anatomy of his right atrium, which did not have enough space to accommodate the device. Instead, we opted to use the NS device, which would allow us to overcome this limitation associated with classical devices; yet, the procedure was not successful.

To date, the experience with the NS device is limited, with only one published registry,⁶ which seems to confirm the feasibility, safety, and efficacy of the procedure, with a documented closure rate (defined by RLS grade ≤ 1) of 89% and a full closure rate of

Case Report

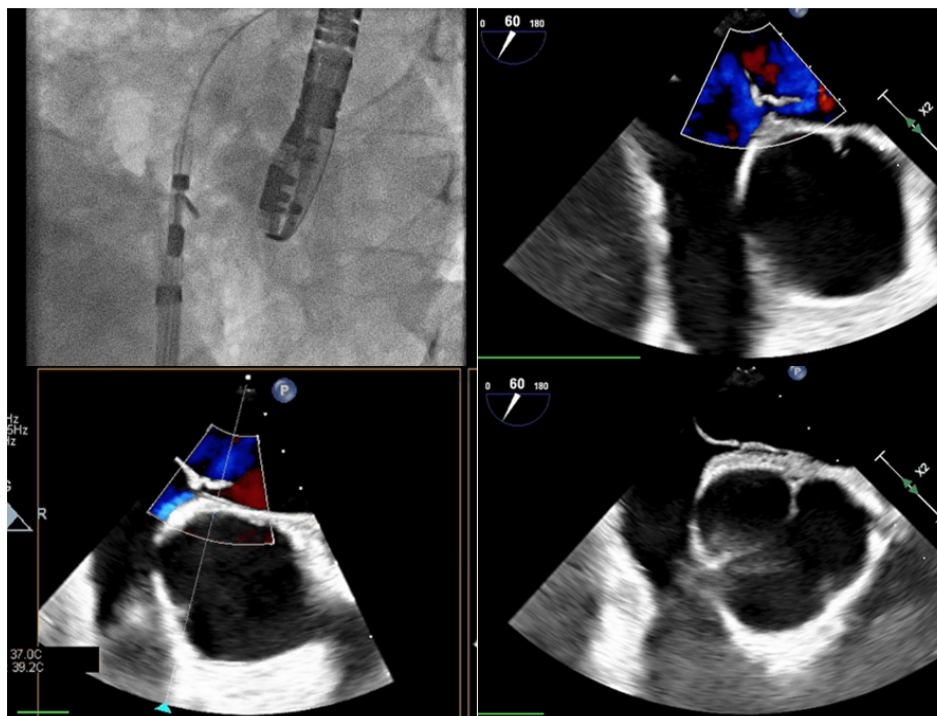
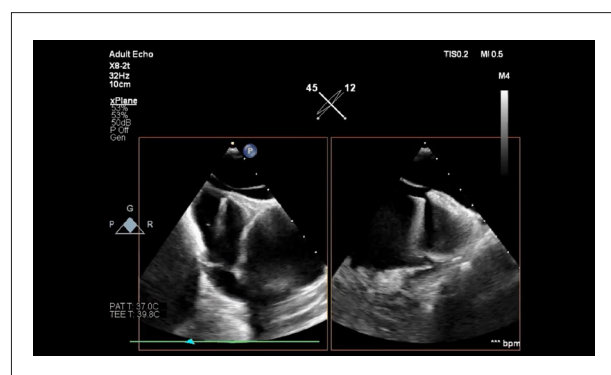


Figure 2 – Peri-procedure percutaneous images. A: Fluoroscopic image with a suture delivery catheter of the NobleStitch system; B and C: TEE mid-esophageal short axis view at 60° revealing significant residual shunt after placement of the first suture; D: TEE using the same view with the two sutures in place.



Video 2 – Peri-procedure TEE images showing a significant residual shunt after placement of the first suture, which disappeared after the second suture was placed. Link: <http://abcimaging.org/supplementary-material/2023/3601/ABC-309-video02.mp4>

75% at 12 months, results that are comparable with traditional devices. Nevertheless, we still lack experience and information on the determinants of residual RLS, which would help select patients with the most favorable anatomies for a suture-mediated closure technique.

In the previous registry,⁶ the authors did not find a relevant correlation of significant RLS (grade ≥ 2) or technique failure with

the various baseline characteristics investigated. However, they highlighted that the presence of a wide redundant septum seemed to be a prevalent aspect in those patients. After a thorough literature search, we found only two case reports about NS failure. One was due to a septal tear, probably caused by the suture,⁸ and the other was related to a significant residual RLS in an individual who also had an aneurysmatic IAS.⁷

Conclusions

POS is a rare condition that can have a significant impact on the patient's quality of life. When it is due to PFO-related intracardiac shunt, the defect closure is a curative treatment. Different percutaneous closure devices can be used. NS is an upcoming appealing alternative; yet, we still lack knowledge of the characteristics that should be considered in patient selection. Herein, we describe the case of another failed percutaneous PFO closure with NS that could add to the evidence that IAS anatomy may play a key role in the success of the procedure, with its use being less favorable in patients with aneurysmatic IAS.

Author Contributions

Conception and design of the research and writing of the manuscript: Moura AR; acquisition of data and analysis and interpretation of the data: Moura AR, Silva M, Rodrigues A, Silva JC, Ribeiro J, Caeiro D, Casanova J; critical revision of the manuscript for intellectual content: Silva M, Rodrigues A, Carvalho RF.

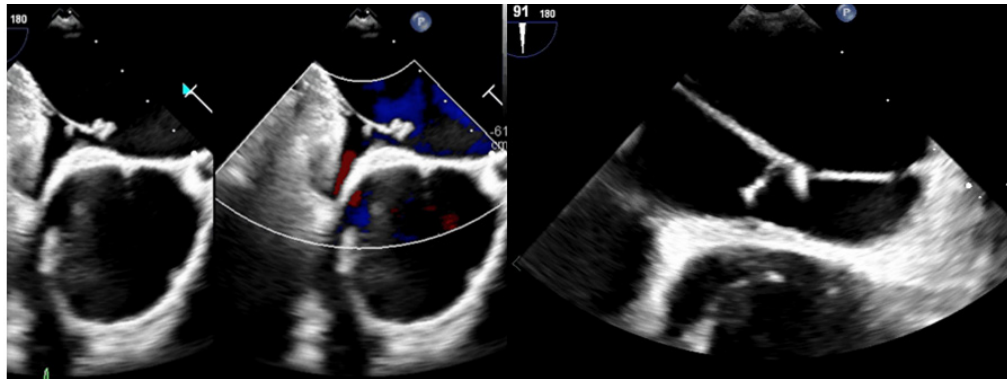
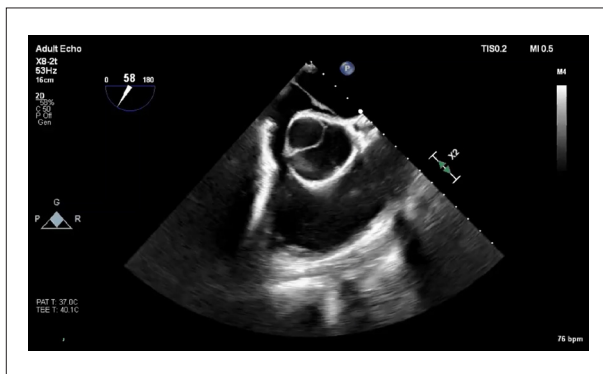


Figure 3 – Post-procedure percutaneous images. TEE mid-esophageal short axis view at 60° with linear echogenic structures compatible with previously implanted devices at the septum secundum but detached from the septum primum and evident interatrial shunt.



Video 3 – Post-procedure TEE showing NS failure with the two previously implanted devices detached from the septum primum.
Link: <http://abcimaging.org/supplementary-material/2023/3601/ABC-309-video03.mp4>

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No potential conflict of interest relevant to this article was reported.

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Study Association

This study is not associated with any thesis or dissertation work.

Ethics Approval and Consent to Participate

This article does not contain any studies with human participants or animals performed by any of the authors.

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