

CASE REPORT

INTERMEDIATE

CLINICAL CASE

# Rare Mechanical Complication of Percutaneous PFO Closure Leading to Recurrent Strokes in a Teenage Boy



Zaran Butt, MB BCH BAO, Sandra Quinn, BSc, MB BCH BAO, PhD, Brendan McAdam, MB BCH BAO

## ABSTRACT

An 18-year-old male subject presented with recurrent strokes 3 years after percutaneous patent foramen ovale (PFO) closure with an PFO occluder device for presumed PFO-related stroke. A transesophageal echocardiogram revealed apparent device displacement during dynamic cardiac cycle. This case highlights an unusual but important delayed complication of transcatheter PFO closure. (**Level of Difficulty: Intermediate.**) (J Am Coll Cardiol Case Rep 2021;3:1456-1458) © 2021 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 previously healthy 15-year-old schoolboy presented to the emergency department with a 2-day history of progressively severe occipital headache, ataxia, and vomiting. He had no focal neurologic deficits, neck stiffness, or photophobia. He also had no fevers or

chills, and no recent foreign travel or recent scuba-diving experiences.

Examination revealed a regular heart rate of 88 beats/min, blood pressure of 154/88 mm Hg, respiratory rate of 18 breaths/min, and oxygen saturation of 95% on room air. The patient was afebrile. Glasgow Coma score was 15. Bilateral limb and cranial nerve examinations were unremarkable. Heart sounds S1 and S2 were noted with no additional sounds.

## LEARNING OBJECTIVES

- To appreciate that serious complications of transcatheter PFO closure can be delayed in presentation.
- To understand the need for meticulous investigation to rule out hypercoagulable states or other concurrent etiologies for ischemic events in the setting of a PFO before considering device implantation.
- To recognize that a thorough discussion of the risks and benefits of percutaneous PFO closure with patients is important, particularly in younger patients.

## PAST MEDICAL HISTORY

The patient had no medical or surgical history and took no regular medications. There was no family history of cardiovascular disease. He was a nonsmoker and denied alcohol or recreational drug use.

## DIFFERENTIAL DIAGNOSIS

The initial differential diagnosis was broad, including labyrinthitis, ischemic stroke, migraine, and primary brain tumor.

From the Department of Cardiology, Beaumont Hospital, Dublin, Ireland.

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](#).

Manuscript received June 29, 2021; accepted July 1, 2021.

## INVESTIGATIONS

A computed tomography scan of the brain revealed a left posterior inferior cerebellar artery territory infarct. Full blood cell count, renal profile, C-reactive protein, erythrocyte sedimentation rate, and coagulation studies were within normal limits.

Admission electrocardiogram revealed normal sinus rhythm. Results of a 72-hour Holter monitor were normal, and there was no evidence of paroxysmal atrial fibrillation. Magnetic resonance angiography showed patent carotid and vertebral arteries with no evidence of dissection.

Transesophageal echocardiogram (TOE) revealed an aneurysmal intra-atrial septum with patent foramen ovale (PFO) with diameter of 5 mm visualized on bubble study at rest. No thrombus or vegetations were noted. Heart valves, chambers, and right and left ventricular systolic function were preserved.

Vasculitis screen was positive for anti-nuclear antibody, but the remaining panel (double-stranded DNA, anti-Ro, anti-La, anti-SM, anti-RNP, anti-MPO, anti-PR3, and C3/C4) was negative. Thrombophilia screen (lupus anticoagulant, protein C/S, anti-thrombin III assay, and fibrinogen) was also negative.

## MANAGEMENT

The patient was treated with aspirin for delayed presentation of cryptogenic stroke. After multidisciplinary discussion, percutaneous PFO closure was deemed most appropriate. A 25-mm device was implanted without immediate complication. Post-procedure TOE showed the device in good position, with no visual shunt, and results of the bubble study were negative. The patient was discharged on dual antiplatelet therapy for 6 months. TOE at 6 months showed no residual shunting on bubble study. He was maintained on daily aspirin.

The patient remained well until presentation 3 years later with painless blurring of vision beginning suddenly while on a high-velocity amusement ride. Computed tomography brain scan showed a left-sided central retinal artery occlusion, confirmed with fluorescein angiography. TOE revealed color flow around the device but no definitive residual shunting on bubble study. Warfarin was initiated and a repeat TOE was planned in 3 months' time.

The patient re-presented 1 month later, however, with recurrent episodes of transient right-sided visual disturbance lasting 10 to 15 min with spontaneous resolution, occurring up to 6 times a day since discharge. He described "fuzziness" within his visual field but no vision loss. Fundoscopy revealed a

healthy optic disc. Magnetic resonance imaging of the brain showed evidence of a new infarct in the left putamen region (**Figure 1**).

TOE revealed a large mobile echodensity consistent with thrombus on the posterior aspect of the left atrial (LA) wall, where the posterior leg of the device appeared to be in contact with the posterior LA wall during dynamic cardiac cycle (**Videos 1, 2, and 3**).

There was no residual shunting by bubble study and no apparent device-related thrombus. There was no evidence of paroxysmal atrial fibrillation on repeat 72-hour Holter. Given this finding despite anticoagulation, the patient was considered at high risk for recurrent stroke, prompting referral for surgical removal of the PFO device and thrombus, as well as pericardial patch repair.

## DISCUSSION

This case highlights a most unusual complication related to implantation of a septal occluder device in a young patient for assumed paradoxical embolism in the setting of a complex PFO and in the absence of other attributable causes. After successful closure, the patient continued to experience recurrent ischemic neurologic events despite post-closure TOE showing no device-related thrombus and a negative result on bubble study, and despite antiplatelet therapy per international guidance (1) and subsequent anticoagulation. Delayed complications after percutaneous PFO closure are extremely rare (2).

## ABBREVIATIONS AND ACRONYMS

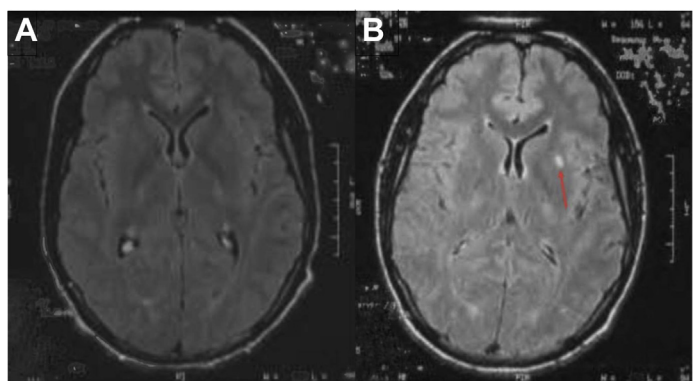
**CRAO** = central retinal artery occlusion

**LA** = left atrial

**PFO** = patent foramen ovale

**TOE** = transesophageal echocardiogram

**FIGURE 1** Comparison of Magnetic Resonance Imaging Brain Imaging From 2006 and 2009



Note the new focus of increased signal on fluid-attenuated inversion recovery imaging in the left putamen deep to the Sylvian fissure (**red arrow**) in 2009 compared with prior imaging, likely representing a new small infarct. (**A**) Imaging from 2006. (**B**) Imaging from 2009.

We believe that the mechanism of the recurrent thromboembolic events was due to local mechanical disruption of the endothelial lining of the posterior LA wall from the posterior limb of the device on the LA side, which in our opinion was compounded by dynamic cardiac motion. There was no residual shunting across the device or thrombus on the device. This phenomenon may also have potentially led to erosion through the posterior left atrium, which has been seldom described in the literature (3).

### FOLLOW-UP

Warfarin was discontinued 15 months' post-operatively. The patient has remained clinically well in the 9 years since surgical repair, on aspirin monotherapy, with no further ischemic neurologic events, and interval magnetic resonance imaging of the brain showed no new lesions.

Of note, the patient required high doses of warfarin with supplemental heparin to maintain a therapeutic international normalized ratio. Given the difficulty in achieving therapeutic international normalized ratio levels, he has also since been investigated for prothrombotic disorders. Total homocysteine level was measured at 54 mmol/L (range: 0-18 mmol/L), methionine was 40 mmol/L (range: 6-40 mmol/L), folic acid was 3.2 ng/mL (range: 3.0-17.6 ng/mL), and vitamin B<sub>12</sub> was 217 pg/mL (reference range: 189-883 pg/mL). Results of a skin biopsy confirmed homozygosity for the thermolabile variant of the methyl-entetrahydrofolate reductase (MTHFR) gene. He was

initiated on high-dose folic acid and a low-protein diet, which led to interval improvement in total homocysteine (10 mmol/L).

### CONCLUSIONS

Detailed assessment for rare complications of percutaneous PFO closure as illustrated by this case is essential. This should include detailed post-procedure TOE to not only assess the device in situ but also to carefully inspect surrounding structures and tissues, as well as perform bubble studies with the Valsalva maneuver. Thorough pre-procedural assessment is also vital to exclude hypercoagulable states and to identify patients at higher risk of potential complications (4). This case serves to remind clinical teams to have thorough discussions of the risk and benefits of percutaneous closure of intra-atrial septal defects, especially in younger patients.

**ACKNOWLEDGMENTS** The first author wishes to express his sincere gratitude to Dr Sandra Quinn and Professor Brendan McAdam, in helping to prepare this work.

### FUNDING SUPPORT AND AUTHOR DISCLOSURES

The authors have reported that they have no relationships relevant to the contents of this paper to disclose.

**ADDRESS FOR CORRESPONDENCE:** Dr Zaran Butt, Beaumont Hospital, Beaumont Road, Dublin DO9V2N0, Ireland. E-mail: [zaran.butt@ucdconnect.ie](mailto:zaran.butt@ucdconnect.ie).

### REFERENCES

1. Pristipino C, Sievert H, D'Ascenzo F, et al. European position paper on the management of patients with patent foramen ovale. General approach and left circulation thromboembolism. *EuroIntervention*. 2019;14(13):1389-1402.
2. Abaci A, Unlu S, Alsancak Y, Kaya U, Sezenoz B. Short and long term complications of device closure of atrial septal defect and patent foramen ovale: meta-analysis of 28,142 patients from 203 studies. *Catheter Cardiovasc Interv*. 2013;82(7):1123-1138.
3. Ivens E, Hamilton-Craig C, Aroney C, Clarke A, Jalali H, Burstow DJ. Early and late cardiac perforation by Amplatzer atrial septal defect and patent foramen ovale devices. *J Am Soc Echocardiogr*. 2009;22(9):1067-1070.
4. Zajarias A, Thanigaraj S, Lasala J, Perez J. Predictors and clinical outcomes of residual shunt in patients undergoing percutaneous transcatheter closure of patent foramen ovale. *J Invasive Cardiol*. 2006;18(11):533-537.

**KEY WORDS** atrial septal defect, congenital heart defect, echocardiography, imaging, occlude, stroke

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