

## CASE REPORT

# Hybrid approach to management for a patient with culture negative infective endocarditis with profunda femoris mycotic pseudoaneurysm: A case report and review of the literature

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## Key Clinical Message

Mycotic pseudoaneurysms can be a serious and life threatening complication of left sided infective endocarditis. They most commonly affect the major axial vessels. Profunda femoris artery (PFA) aneurysms are rare and present in only 0.5% of all peripheral aneurysms, regardless of the underlying etiology. We present a case of a patient who underwent mitral valve repair for severe mitral regurgitation secondary to culture negative IE which was complicated by multiple mycotic pseudoaneurysm. The PFA pseudoaneurysm which was affected and was complicated with a large hematoma compressing the femoral nerve. This was managed by a staged hybrid approach. Endovascular stenting was performed first to seal the pseudoaneurysm and facilitate open surgical repair using a reversed interposition saphenous vein graft. To the best of our knowledge, this is the first reported case of a PFA mycotic aneurysm (MA) being managed by a hybrid approach using endo-vascular and open surgical repair. MAs and pseudoaneurysms are complex and life threatening conditions requiring meticulous planning for optimal management. Endovascular stenting can be considered as an alternative to surgical management in certain cases or as a bridge to definitive open surgical repair depending on anatomical location and associated complications.

## KEYWORDS

infective endocarditis, mitral valve repair, mycotic aneurysm, profunda femoris artery, pseudoaneurysms

## 1 | BACKGROUND

The first introduced clinical aspects of infectious endocarditis (IE) came from the French clinician Jean Fernel in the

16th century.<sup>1</sup> When the cause of IE was identified as microorganisms, appropriate diagnostic and antibiotic treatment were initiated, this led to a decrease in the incidence of complications. One of these complications is mycotic

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aneurysms (MAs), a term first described by Sir Osler in 1885 in a 30-year-old man with four aneurysms in the aortic arch.<sup>2</sup> These are mainly caused by septic emboli originating from vegetations in the cardiac valves that occlude the vasa-vasorum of peripheral arteries leading to weakness in the arterial wall and subsequent aneurysm formation. Profunda femoris artery (PFA) aneurysms are rare, representing only 0.5% of cases of all aneurysms, regardless of the underlying etiology.<sup>3</sup> We present a case of a male patient with culture negative mitral valve IE with multiple septic emboli and a PFA mycotic pseudoaneurysm.

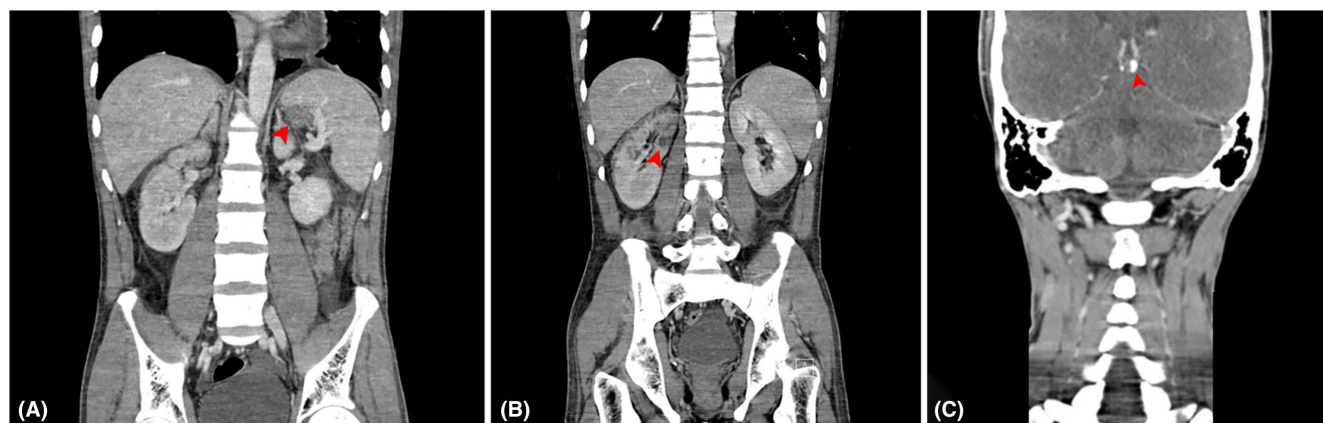
## 2 | CASE PRESENTATION

### 2.1 | Case history and examination

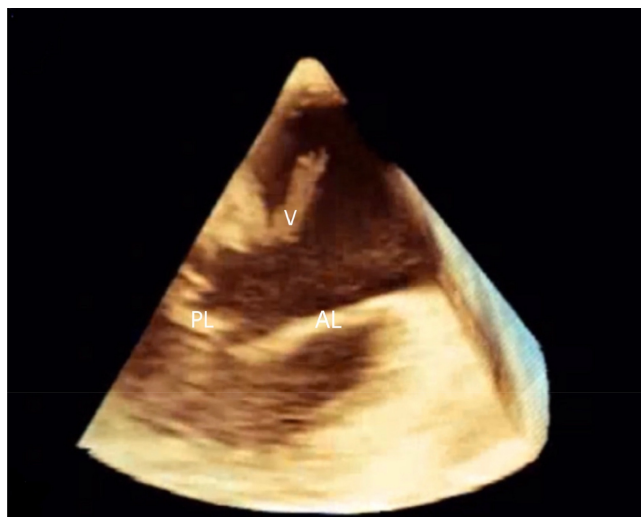
A 27-year-old male patient with no chronic diseases, prior hospitalization or prior surgical Interventions, presented to the hospital complaining of a 1-week history of fever. He had no other associated symptoms. He was hemodynamically stable, with multiple spikes of fever reaching 40.2°C. His physical exam revealed a pansystolic murmur best heard at the apex. A full septic workup was performed; his blood cultures were drawn as two sets from two different venipuncture sites within 30 min of each other and they were negative, other labs were significant for leukocytosis of ( $38 \times 10^9$ ), elevated C-reactive protein (92 mg/L) and erythrocyte sedimentation rate (35 mm/h). Due to the high inflammatory markers, he was started on ceftriaxone 1 g IV daily and vancomycin 1 g IV every 12 h after the blood cultures were drawn. Anidulafungin 200 mg IV daily was added a week after due to the patient still developing spikes of fever reaching 38.9°C. A chest and abdominal computed tomography (CT) scan revealed a small (1.2 cm) right renal artery aneurysm with cortical infarctions and a subcapsular

splenic infarction (Figure 1). A trans-esophageal echocardiogram revealed a mobile mitral valve vegetation located above the posterior leaflet and mild mitral valvular regurgitation (Figure 2), therefore, the diagnosis of culture negative infective endocarditis was made. Repeated echocardiogram showed progression of the mitral regurgitation to severe, the decision was then made to proceed with surgical repair.

A successful mitral valve repair was performed with debridement of infected posterior leaflet tissue, primary leaflet repair and limited suture annuloplasty. He was then shifted to the surgical intensive care unit (SICU) where he had a scabrous postoperative course. He had three episodes of cardiac arrest post extubation. The first arrest was the third day postoperatively where the patient developed rapid ventricular fibrillation, requiring cardiopulmonary resuscitation for 4 min a total of two cycles then a shock was delivered and patient returned to sinus rhythm and epinephrine was given via an internal jugular central venous line that was inserted in the operating room prior. After that a full arrest workup was sent including an ECG which showed that the arrest happened as a result of torsades des pointes requiring electrical cardioversion. This was secondary to long QT syndrome as a complication of the antifungal medication which was stopped by the infectious diseases team and labs showed that the patient had low magnesium reaching 0.5 meq/L. The arrhythmia team was also connected and recommended that the patient stop all medications that cause an increase in QT segment length, correct the electrolyte abnormalities and start the patient on bisoprolol. The second cardiac arrest occurred later that day and was also due to ventricular fibrillation and CPR was done for one cycle and was shocked again with 200 J. The third arrested was the following day and also was due to ventricular fibrillation and he was unresponsive with CPR commencing for one cycle and a single epinephrine dose was given followed by a cardioversion



**FIGURE 1** Coronal computed tomography scan: (A) Demonstrates a finding of a right sided subcapsular splenic infarction. (B) Demonstrates a finding of multiple cortical renal infarctions. (C) Demonstrates a finding of a mycotic aneurysm of the left cortical branch of the middle cerebral artery in the parietal lobe.



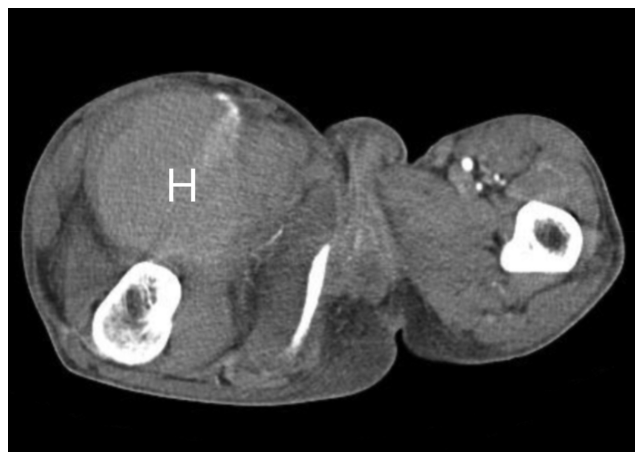
**FIGURE 2** Transesophageal echocardiogram: (midesophageal two chamber view). Demonstrates a finding of a freely mobile vegetation above the posterior mitral valve leaflet. AL, anterior leaflet of the mitral valve; PL, posterior leaflet of the mitral valve; V, vegetation.

shock the patient was awakened but he was gasping for air and had difficulty breathing so he was intubated and was extubated a day after.

## 2.2 | Methods (diagnosis, investigations, and management)

Once he was stable, he was shifted to the surgical step-down unit. Two weeks postoperatively he started complaining of right sided hip pain and decreased mobility. Examination exhibited right hip swelling extending to the thigh with decreased hip extension and flexion. A CT angiogram (CTA) was performed, which revealed a large hematoma measuring at  $10 \times 11$  cm from a communicating right PFA MA and also revealed a left internal iliac artery aneurysm measuring  $2 \times 2.5$  cm (Figure 3).

Vascular surgery were consulted. First, he was shifted to the angiography suite where endo-vascular stenting of the right renal, left internal iliac, and the PFA was performed to seal all the MAs (Figure 4). The next day the patient was taken to the operating room for open surgical repair. The large thigh hematoma was evacuated. The stent graft from the PFA was removed. The vessel was found to be extremely fragile and destructed. It was ligated at its origin. The common femoral artery (CFA) bifurcation was also affected. A trial of end-to-end CFA to superficial femoral artery (SFA) anastomosis was performed to no avail due to the fragility of the tissues. A reversed interposition graft was performed using a healthy segment of the great saphenous vein between the CFA and SFA.



**FIGURE 3** Axial computed tomography angiography (CTA) scan: Demonstrates a finding of large hematoma located in the anterior segment of the right thigh from a mycotic aneurysm arising from the profunda femoris artery indicated by the extravasation of contrast. H, hematoma.

## 2.3 | Outcomes and follow-up

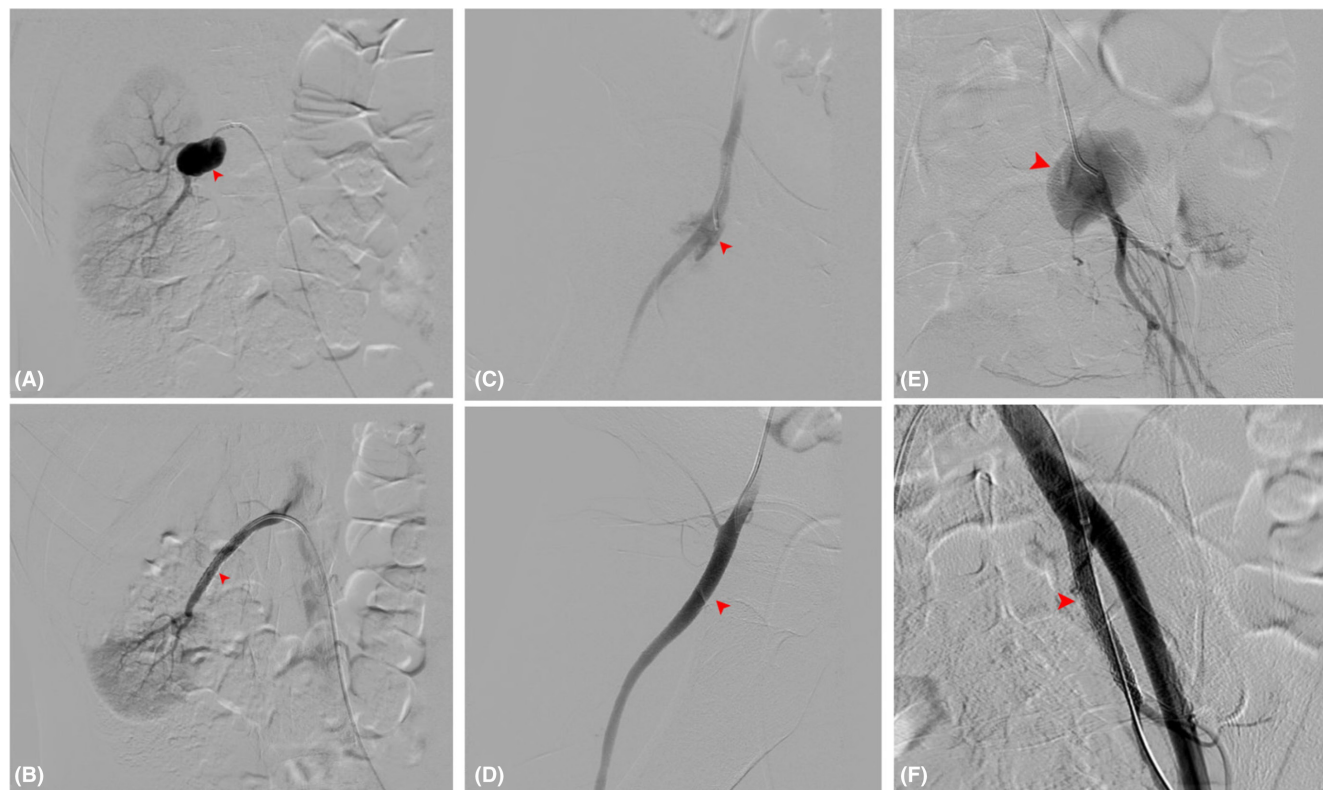
The patient had an uncomplicated SICU course. A peripherally inserted central catheter was inserted for him to continue his antibiotics therapy which was ceftriaxone 1 g IV daily and vancomycin 1 g IV every 12 h for 6 weeks and was discharged home with resolution of his right leg weakness and all neurological symptoms. A CTA was done 9 months postoperatively and showed excellent results (Figure 5).

## 3 | DISCUSSION

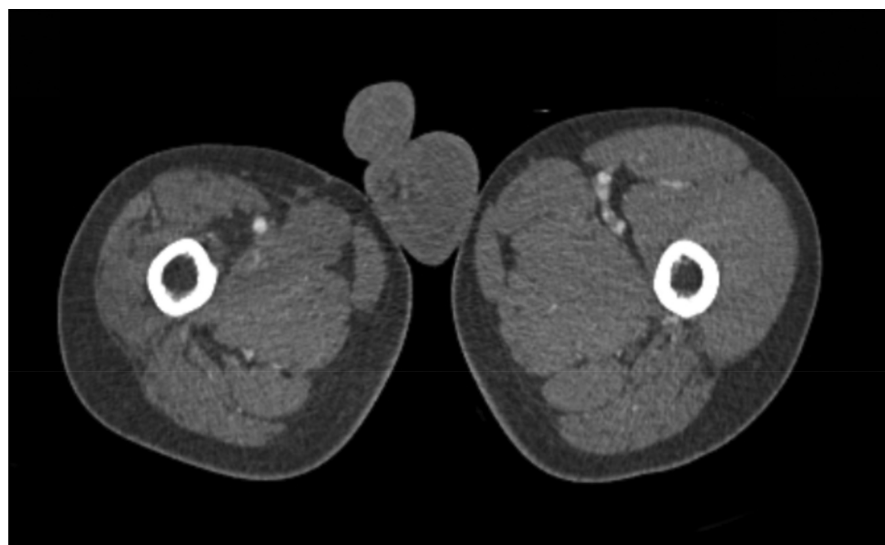
This surgical case of a man with a culture negative IE in a native valve may be representative of a change from the classical epidemiological profile of patients with valves compromised by rheumatic fever or non-infectious endocarditis.

The diagnosis of IE may be made by using the modified Duke criteria, which includes a list of major and minor criteria. If a case fulfills one major and one minor or three minor criteria, IE is considered likely.<sup>4</sup>

Blood culture-negative infective endocarditis (BCNIE) refers to IE in which no causative microorganism can be grown using the usual blood culture methods. It poses considerable diagnostic and therapeutic difficulties. The 2023 guidelines for the management of infective endocarditis by the European Society of Cardiology state that the main cause of BCNIE is the administration of antibiotics prior to blood cultures, and that the antibiotics may be paused to allow bacterial growth and draw another set of cultures if the patient is stable. Depending on local



**FIGURE 4** Arterial angiography: (A) right renal mycotic aneurysm. (B) Stenting of the right renal mycotic aneurysm. (C) Left internal iliac artery mycotic aneurysm. (D) Stenting of the left internal iliac artery mycotic aneurysm. (E) large right profunda femoris artery mycotic pseudoaneurysm. (F) Stenting of right profunda femoris artery mycotic Pseudoaneurysm. The stented arteries demonstrated sealing of the aneurysms with normal blood flow.



**FIGURE 5** Postoperative axial computed tomography angiography (CTA) scan: Demonstrates a finding of resolving of the hematoma on right thigh and normal profunda femoris artery after repair of the mycotic aneurysm.

epidemiology, serological testing for *Coxiella burnetii*, *Bartonella* spp., *Aspergillus* spp., *Mycoplasma pneumoniae*, *Brucella* Spp., and *Legionella pneumophila* should be proposed, followed by specific polymerase chain reaction assays for *Tropheryma whipplei*, *Bartonella* spp., and fungi (*Candida* spp., *Aspergillus* spp.) from blood and the tissue. In addition, 16S and 18S ribosomal ribonucleic acid (rRNA) sequencing from tissue may provide

a microorganism diagnosis in BCNIE. Pathological examination of resected tissue or embolic fragments remains the gold standard for IE diagnosis. Tissue samples excised during surgical valve debridement/resection should be sent in a sterile container for pathological and microbiological investigation, which may facilitate the diagnosis of non-infectious causes of endocarditis, such as neoplastic and autoimmune causes.<sup>4</sup>



The incidence and mortality from the complications of IE have decreased in the recent era of antibiotics and valvular surgery. One of these complications is MAs, which are localized, irreversible vascular dilatations that develop when a septic embolism causes blockage in the vasa-vasorum of a peripheral artery. The first MA described was in 1885 by Sir William Osler where he used the term 'mycotic' to describe the mushroom shaped appearance of the aneurysms and not the microbiological etiology.<sup>1</sup> Gram-negative bacteria different from those of classical IE may be involved, as recent evidence suggest they may be overrepresented in embolising IE, including right-sided IE,<sup>5</sup> and are known to cause MAs in similar vascular infections such as Lemierre syndrome.<sup>6</sup>

Majority of the MAs are located in the axial vessels, such as the aorta, mesenteric artery, brain vessels, or femoral artery. However, involvement of the PFA is noteworthy as they represent only 0.5% of cases of all aneurysms regarding the underlying etiology.<sup>3</sup> In our literature review only five cases were reported involving PFA being of a mycotic origin with only two being in the modern era (which is defined by the era of widespread use of endovascular treatment modalities) (Table 1).<sup>7-11</sup> With the first case being reported on a postmortem examination of a patient who had IE and leg pain in 1890, by Sir Brice Duckworth where a ruptured PFA aneurysm was found.<sup>12</sup>

No established guidelines exist on the management of PFA aneurysms. Of all the cases reported on PFA MAs only a single recent case was managed using an endovascular approach, with all older cases using an open surgical approach.<sup>4,8,9,10,11</sup> Accordingly, few studies have described endovascular approaches either by stenting or embolization for PFA aneurysms as a whole, in both mycotic and non-MAs. In a recent study performed by Khalid et al. In 2014 on a successfully repaired non-mycotic ruptured PFA pseudo-aneurysm using a stent graft similar to our approach, they reported in their review that endo-vascular approach using stent graft was performed on other cases of PFA non-MAs with success and can be an alternative to open surgical approach.<sup>3</sup>

The mainstay treatment of the PFA aneurysms has been an open surgery approach with either ligation of the PFA or reconstruction with a vein or a graft. Depending on the SFA's state, either procedure may be performed. If the SFA is patent with occlusions or aneurysmal tissue, then ligation of the PFA is feasible due to that blood will pass via the popliteal-femoral tract. However, fixing the PFA may be preferable to ligating to preserve blood flow to the lower limbs. Concomitant SFA disease requires the PFA to be reconstructed to prevent ischemia of the lower limb.<sup>13</sup>

The limited reports on endovascular repair of PFA aneurysm report success with endovascular coil embolization and covered stents to exclude the aneurysm.

TABLE 1 Profunda femoris artery mycotic aneurysm reported cases in the modern era.

Authors	Year	Age	Presentation	IE	Culture	Diagnostic test	Size (cm)	Management	Follow-up
Bakr et al.	2019	54	Incidental Finding Then Complained Of a right sided Pulsating swelling.	Yes	Negative	CTA	3.6	Endo-vascular with a covered stent graft	Duplex ultrasound showed the stent graft occluded within 2 years but with no symptoms of limb ischemia or evidence of aneurysm recurrence.
Dixon et al.	2021	47	Incidental finding on ultrasound due to history of Claudication and popliteal	Yes	<i>Staphylococcus warneri</i>	Ultrasound + CTA	1.3	Open surgical approach where the PFA was anastomosed in an end-to-side fashion to the superficial femoral artery.	Patient followed up for 6 months while taking the antibiotic course For IE without recurrence of the aneurism
Alshair et al.	2024	27	After mitral valve repair the patient started complaining of right sided hip pain, decreased mobility and right hip swelling extending to the thigh	Yes	Negative	CTA	Couldn't be measured due to large hematoma formation	A hybrid approach to management was performed. First, endovascular stenting of the aneurysms was performed at the same setting, followed by an open surgical repair of the PFA MA using a reversed interpositioning saphenous vein graft	CTA was done 9 months postoperatively and showed excellent results

Abbreviations: CTA, computed tomography angiography; IE, infective endocarditis.

However, embolization may be inappropriate in previous or current SFA occlusive disease. The literature suggests that PFA stenting is a good alternative.<sup>14</sup> Two published cases reported a ruptured PFA aneurysm of a non mycotic origin that was successfully repaired with the use of stent deployment.<sup>13,15</sup> Endovascular approach was shown to be successful and is less invasive than open surgery.<sup>13,16</sup>

Endovascular PFA aneurysm repair lacks long-term follow-up on patency and possible stent failure. The only reported PFA MA repaired by endovascular approach<sup>10</sup> the procedure was performed because of previous groin surgery and resulting high surgical risk. A follow-up duplex scan after 2 years revealed complete blockage of the stent graft in the absence of symptoms due to the development of collaterals.

In our case the patient was managed by a hybrid approach, where a stent graft was used to stabilize and seal the MA prior to definitive surgical management. After 9 months, a CTA showed a completely patent vein graft with no new aneurysm formation and complete resolution of his symptoms.

To the best of our knowledge, this is the first reported case of a PFA MA associated with culture-negative IE managed by a hybrid approach using endo-vascular stenting followed by open surgical repair using a reversed interpositioning saphenous vein graft.

## 4 | CONCLUSION

PFA are rare in the modern era with lack of literature comparing different types of management methods and lack of long term follow-up. Presentation of MAs can vary depending on the anatomic location, and whether the aneurysm is causing a mass effect on adjacent structure. One should have a high index of suspicion of MA in cases of IE and have a low threshold of ordering CT angiogram to diagnose and manage such a dangerous complication. From our experience we recommend the use of endovascular stenting as a preferred method when there is a previous operation in the groin area leading to adhesions, or as a bridge to surgical management. To better standardize the approach for management of PFA aneurysms, studies have to be performed comparing different endovascular techniques with open surgical approach with a long term follow-up period.

## AUTHOR CONTRIBUTIONS

**Fahad M. Alshair:** Conceptualization; data curation; methodology; writing – original draft; writing – review and editing. **Abdullah H. Baghaffar:** Conceptualization; project administration; supervision; validation; visualization;

writing – original draft; writing – review and editing. **Mazin A. Fatani:** Methodology; supervision; validation; visualization; writing – review and editing. **Reda A. Jamjoom:** Supervision; validation; visualization.

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## CONFLICT OF INTEREST STATEMENT

The authors declare that there are no conflicts or competing interests.

## DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

## ETHICS STATEMENT

Written informed consent was obtained directly from the patient to publish this case report, so ethical approval is not applicable.

## CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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