CASE REPORT

The clot thickens: an incompletely ligated left atrial appendage

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Summary

Our patient presented with known mechanical mitral valve endocarditis documented by 2D transesophageal echocardiogram (TOE) from a recent hospitalization at an outside facility. On admission to our center, there was no prior knowledge of an incompletely ligated left atrial appendage (LAA) according to patient- or family-reported history, review of outside records or the outside facility's 2D TOE report. A 3D TOE performed at our center to assess her pathology, since a month had passed from her prior hospitalization, revealed a LAA ligation with evidence of communication to the left atrium and with clot present in the appendage. This case report highlights the common finding of incomplete closure of the LAA following surgical ligation, thus making it inadequate for stroke prevention in patients with atrial fibrillation, and that 3D TOE plays a valuable role in assessing the durability of LAA ligation.

Key Words

- 3D transesophageal echocardiography
- bacterial endocarditis
- left atrial appendage thrombus
- rheumatic heart disease

Learning points:

- 3D transesophageal echocardiography (TOE) is a valuable tool in assessing the durability of left atrial appendage (LAA) ligation given the superior image granularity as compared with 2D TOE.
- LAA ligation may not be adequate for stroke prevention in patients with atrial fibrillation as incomplete closure is common following surgical ligation.
- LAA occlusion should be considered in these cases.

Background

We describe a case of a patient with mechanical mitral valve endocarditis, native severe symptomatic aortic stenosis and incomplete left atrial appendage (LAA) ligation that was discovered on a 3D transesophageal echocardiogram (TOE) and which had been previously unrecognized by a 2D TOE. This case highlights that incomplete LAA closure is common following surgical ligation, and thus, should not be presumed to be adequate for stroke prevention in patients with atrial fibrillation. This may further prompt consideration of LAA closure devices in these patients. Additionally, we have found the application of 3D TOE to be valuable in the identification of incomplete LAA ligation as demonstrated by the fact that this was previously identified by neither transthoracic echocardiography nor 2D TOE performed on this patient during a prior hospitalization at an outside facility.

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Case presentation

The patient is a 68-year-old female with a history of rheumatic heart disease, persistent atrial fibrillation on systemic oral anticoagulation and mechanical mitral valve replacement in 2004. Three months prior to her presentation at our center, she suffered a transient ischemic attack (TIA) and had also been treated for group B *Streptococcus* bacteremia. Then a month prior, she again had recurrent neurologic deficits and was admitted to a hospital in Oregon. She underwent an MRI brain, which showed multiple scattered infarcts. As part of her work up, transthoracic echocardiogram and 2D TOE were performed. She was diagnosed with infective endocarditis of her mechanical mitral valve. Additionally, the 2D TOE revealed the presence of moderate-to-severe native aortic valve stenosis and visualized the LAA with the report commenting there was no thrombus; yet, no remarks regarding LAA ligation were made. She was treated with antibiotics, and surgical intervention was considered. In preparation for such an intervention, cardiac catheterization performed demonstrated 65–70% ostial left anterior descending and 90% mid-circumflex stenosis. Based on cardiology, cardiothoracic surgery and infectious disease (ID) team consultation at the outside facility, a continued trial of antibiotics was recommended, and the patient was discharged home. Following this, the patient moved to Kansas City due to social considerations. She presented to our tertiary care hospital within days of her move with complaints of fatigue and dyspnea.

On admission, her vital signs included a temperature of 97.8°F, heart rate 41 b.p.m, blood pressure of 132/61, respiratory rate of 20 breaths/min and oxygen saturation of 100% on 2 L of oxygen via nasal cannula. Her examination was significant for jugular venous distention to the angle of her mandible and positive hepatojugular reflux. Her lungs had faint bibasilar crackles. She had an irregularly irregular rhythm, a 3/6 crescendo decrescendo murmur best appreciated at the right upper sternal border with radiation to the left clavicle, mechanical S1 and S2 appreciated at the apex. Her abdomen was soft and non-tender. Her extremities were warm and well perfused with 1+ pitting edema bilaterally. Her neurologic examination revealed a slight left facial droop; otherwise, her strength and sensation were intact.

Her course of ceftriaxone was continued on admission, and the ID and cardiothoracic surgery (CTS) teams were consulted. The ID team changed her antibiotics to penicillin G and gentamycin with plans for 2 weeks of combination for synergy and then 4 additional weeks of penicillin G alone. CTS requested repeat imaging to reevaluate the mitral valve and direct her surgical management (aortic valve replacement, mitral valve replacement and coronary artery bypass graft vs transcatheter aortic valve replacement and percutaneous coronary intervention).

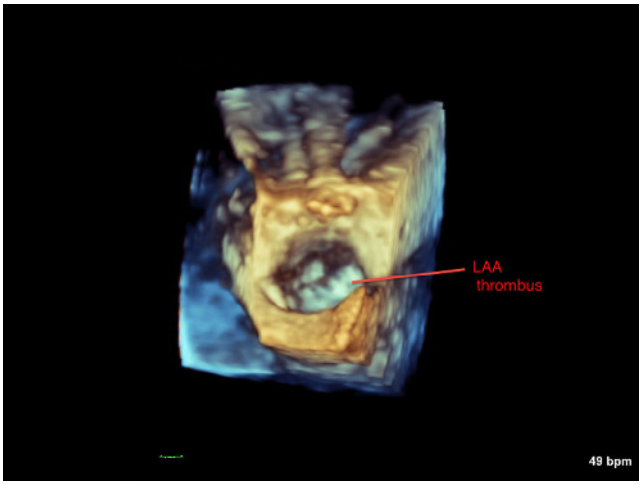

Figure 1
3D image with view looking into LAA with arrow pointing to thrombus. LAA, left atrial appendage.

Investigation

To further interrogate her mitral valve for the presence of any residual infection, as well as to assess her aortic valve, a 3D TOE was performed the day after admission. The 3D TOE images demonstrated left atrial spontaneous echo contrast, or 'smoke', coming from the patient's LAA that had been ligated during her index mitral valve replacement surgery in 2004 (Video 1). Echocardiographically, her LAA had a classic windsock appearance and had been excluded from the left atrium by suturing; yet, the position of the sutures left a remnant LAA, which remained in communication with the left atrium. There was also evidence of a thrombus in her LAA (Fig. 1).

Video 1
Video looking into the left atrial appendage (LAA); 'smoke' can be seen coming out of the LAA indicating its patency and communication with the left atrium. View Video 1 at http://movie-usa.glencoesoftware.com/video/10.1530/ERP-17-0076/video-1.

Next, a heavily calcified aortic valve with moderate-to-severe stenosis with a mean gradient across the aortic

valve of 29 mmHg, maximum velocity of 3.5 m/s, aortic valve area of 0.9 cm² and dimensionless index of 0.31 was visualized. Lastly, the 3D TOE images demonstrate a 1.4 × 0.6 cm echodensity adherent to the posterior margin of the sewing ring of the mechanical mitral valve with prolapse into the posterior leaflet (Fig. 2; Videos 2, 3, 4 and 5). She also had a mild perivalvular leak at the posterior margin of the sewing ring, probably related to erosion from the infection (Figs 2 and 3).

Video 2
3D view of the mechanical mitral valve. View Video 2 at http://movie-usa.glencoesoftware.com/video/10.1530/ERP-17-0076/video-2.

Video 3
Video of the mechanical mitral valve in which the vegetation can be seen moving in the right upper corner of the image and the color Doppler shows the perivalvular leak to the left of the vegetation. View Video 3 at http://movie-usa.glencoesoftware.com/video/10.1530/ERP-17-0076/video-3.

Video 4
Video of the mechanical mitral valve in which the vegetation can be seen moving as the mitral valve opens and closes (right upper corner of clip). View Video 4 at http://movie-usa.glencoesoftware.com/video/10.1530/ERP-17-0076/video-4.

Video 5
Video of the mechanical mitral valve with vegetation moving to the left of it. To the right of the valve, the communication between the left atrium and LAA can be seen. View Video 5 at http://movie-usa.glencoesoftware.com/video/10.1530/ERP-17-0076/video-5.

Treatment and outcome

Though the patient completed a course of intravenous antibiotics per ID team guidance, given the persistence of endocarditis and the embolic phenomena, our patient underwent replacement of her mechanical mitral valve with a Biocor porcine valve on hospital day 7. Based on the operative report, the vegetation was only visualized once the mechanical valve was removed. A piece of the vegetation was sent for microbiologic studies, and on Gram stain, no organisms and only rare polymorphonuclear leukocytes were seen. Additionally, the operative report did not state that there was any attempt made to confirm the presence of a communication between the ligated LAA and left atrium nor whether any further intervention was performed on the LAA.

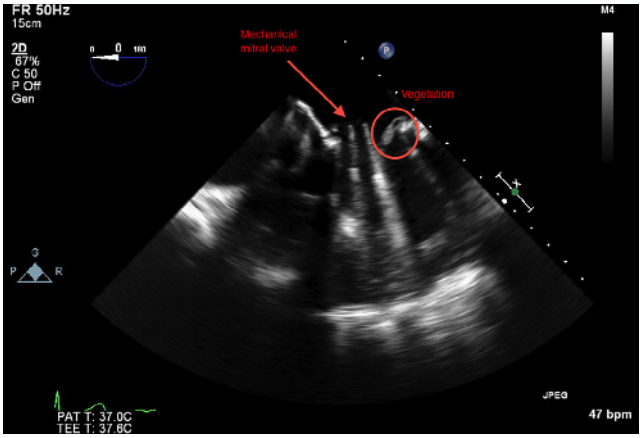

Figure 2
Arrow points to the mechanical mitral valve and a red circle marks the 1.4 × 0.6 cm vegetation.

Her post-operative course was complicated by mixed cardiogenic and vasodilatory shock, acute renal failure requiring continuous renal replacement therapy and acute ischemic hepatitis. After initial stabilization, on post-operative day 5, she again developed shock unresponsive to inotropes, vasopressors or volume resuscitation. Family did not wish for any further aggressive measures, and she expired on post-operative day 5.

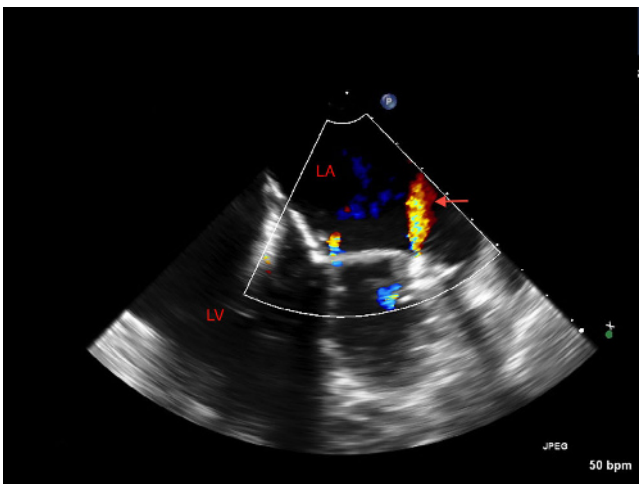

Figure 3
Arrow points to regurgitation of the mechanical mitral valve. LA, left atrium; LV, left ventricle.

Discussion

In our patient, the presence of spontaneous echo contrast arising from the LAA suggests that despite the previous ligation, the appendage was still in communication with her left atrium. Our findings support earlier work by Katz *et al.*, in demonstrating that surgical ligation is frequently incomplete allowing for stagnant blood flow within the appendage to communicate with left atrium (1). In a retrospective study by Kanderian *et al.*, surgical ligation of the LAA was successful in only 55 of 137 patients (40%), indicating the high prevalence of incomplete surgical ligation (2). Reports of incomplete LAA closure have ranged from 10 to 80%; however, in two studies specifically investigating surgical LAA ligation in association with mitral valve surgery, as in our patient's case, the reported incidences of incomplete surgical closure of the LAA were 35 and 36% (1, 3, 4). The presence of a mitral valvular annuloplasty ring or prosthesis has been associated with increased risk of incomplete surgical closure of the LAA due to failure to extend sutures to the most distal edge of the LAA to avoid these structures (3). Thus, this patient's mitral valve prosthesis may have contributed to difficulty in completely ligating the LAA.

Presence of incomplete LAA closure places the patient at risk (and in our patient's case, a thrombus *was* noted) for development of clot in the left atrium. In the same retrospective study by Kanderian *et al.*, 41% of patients with unsuccessful LAA exclusion were found to have thrombus present and 15% of patients with unsuccessful LAA exclusion had experienced a stroke or TIA at the time of TOE (2). Prior studies have even suggested that incomplete closure of the LAA places a patient at greater risk of thromboembolism compared to either complete closure or a non-ligated LAA (3). In a study by Aryana *et al.*, even though patients with incomplete LAA closure were more likely to be on long-term oral anticoagulation, this group had eight times higher risk of stroke and systemic embolization compared to those with complete LAA closure (4).

In our case, the patient had been maintained on warfarin due to her mechanical mitral valve, and the etiology of her cerebrovascular events were felt to be related to septic emboli; however, she was also at risk of having a thrombotic event related to thrombus development in her LAA. With a high rate of unsuccessful closure of the LAA following these procedures and the suggested increased risk of thromboembolism with an incompletely ligated LAA, select patients may benefit from continued lifelong anticoagulation. Others have suggested that for those with an incompletely ligated LAA who cannot tolerate long-term anticoagulation, alternative strategies for closure of the incompletely excluded LAA should be considered (5).

Unlike surgical ligation of the LAA, use of the Watchman device for LAA closure has data to support its use. In both the PREVAIL trial and a 2.3-year follow-up of the PROTECT AF trial, LAA occlusion with the Watchman device was shown to be non-inferior to warfarin for prevention of stroke and systemic emboli (6, 7). In a substudy of the PROTECT AF trial, even though peri-device flow into the LAA was common up to 12 months following implantation (present to some degree in 32% of 389 patients who had a 12-month TOE follow-up), the presence of this flow was not associated with an increased risk of thromboembolism (8). Therefore, unlike LAA ligation, the Watchman device has been proven to be an effective alternative to long-term warfarin for stroke prevention in patients with non-valvular atrial fibrillation.

Additionally in this case, 3D TOE was the imaging modality used to identify and confirm the LAA clot and spontaneous echo contrast. While previous authors have shown that 3D TOE is superior in measuring the LAA orifice and reliable in providing detailed information about LAA morphology, this case also demonstrates that 3D TOE can be useful in identifying pathology within the LAA (9, 10, 11). Although Wunderlich *et al.* demonstrated the utility of 3D TOE in placement of LAA occlusive devices and the post-procedure surveillance, this case highlights the additional utility of 3D TOE in documentation of persistent communication between the LAA and left atrium in patients who have previously undergone surgical ligation of the LAA with the aim of guiding decisions regarding discontinuation of systemic anticoagulation in patients with atrial fibrillation (12).

Three studies albeit some from the surgical literature suggest that 3D TOE is more accurate than 2D TOE for the assessment of LAA orifice diameter (13, 14, 15). Though guidelines have not incorporated the use of 3D TOE for the assessment of LAA patency following LAA ligation, select surgical literature suggests that 3D TOE is preferred in the assessment of LAA orifice diameter ahead of percutaneous LAA closure device procedure and during procedures for proper selection of device size. In our case, 3D TOE identified the incompletely ligated LAA, while 2D TOE did not identify the finding. The complementive 2D TOE plus 3D TOE offers superior visualization of LAA dimensions and surrounding landmarks.

In conclusion, incomplete closure of the LAA is common following surgical ligation of the LAA and places

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these patients at risk of thromboembolism. 3D TOE can provide greater benefit in identifying these patients in comparison to 2D TOE.

Declaration of interest

The authors declare that there are no conflicts of interest that could be perceived as prejudicing the impartiality of this case report.

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Patient consent

Patient is deceased. We attempted to contact the next of kin on multiple occasions but never received a response.

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