One step thoracotomy approach for atrial-esophageal fistula repair without cardiopulmonary bypass

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Case Report

Keywords: atrial-esophageal fistula, thoracotomy, cardiopulmonary bypass

Posted Date: October 5th, 2022

DOI: https://doi.org/10.21203/rs.3.rs-2106375/v1

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Abstract
Although atrial-esophageal fistula is an extremely rare disease, it is a life-threatening complication after catheter ablation for atrial fibrillation. There is no consensus on management or repair for atrial-esophageal fistula which has a high mortality rate. Here, we describe a lateral thoracotomy approach focused on simplifying the repair procedure for atrial-esophageal fistula with two cases.

Two male seniors in their 60s and 70s, respectively, who had undergone catheter ablation for atrial fibrillation presented to the emergency department with fever and mental decrease. Blood culture confirmed Streptococcal bacteremia. Brain magnetic resonance imaging showed multiple cerebral infarcts. After demonstrating atrial-esophageal fistula with computed tomography of chest, an emergency operation was planned.

The two patients were placed in the right down decubitus position with single lung ventilation. A lateral thoracotomy was performed on the left 5th intercostal space. Multiple pledgeted 4-0 prolene sutures for ligation were paced on the fistula of the pericardial side. From the esophageal side, a fistulectomy was performed, followed by an additional primary repair for the esophageal defect. Follow-up endoscopy demonstrated a well-repaired fistula site. The two patients were discharged without recurrence.

Our experience suggests that one step repair for atrial-esophageal fistula via lateral thoracotomy might be feasible in selected patients.

Introduction
Catheter ablation for atrial fibrillation (AF) is widely utilized and considered safe with 6% of complications [1]. Although atrial-esophageal fistula (AEF) is an uncommon disease, it is a life-threatening complication after catheter ablation for AF [2, 3]. The onset of AEF occurs several days to 2 months after catheter ablation. Its symptoms are diverse, including neurologic changes [2]. Because of its rarity, there is no agreed gold standard for repairing AEF which has a high mortality rate up to 80% [4]. Herein, we illustrate a lateral thoracotomy approach focusing on a simple repair for AEF with two cases.

Case Presentation
Case 1
A 61-year-old man presented to the emergency department with a history of 3 days of fever. He has a background of catheter ablation using radiofrequency energy for AF one month ago, end stage renal disease requiring hemodialysis, and pace maker insertion. After admission to the hospital for evaluation of fever, his mental decrease was noted. Brain magnetic resonance imaging (MRI) showed multifocal embolic infarct. *Streptococcus salivarius* and *Streptococcus mitis/oralis* were cultured in blood. A repeated chest computed tomography (CT) demonstrated AEF (Fig. 1A).
Surgical strategy was urgently planned. After endotracheal general anesthesia, the patient was placed in the right down decubitus position. Single lung ventilation was initiated. A thoracotomy was performed on the left 5th intercostal space. After left lung retraction, pericardium was opened, revealing severe adhesion. Access to the fistula through pericardial space was attempted, but was abandoned due to excessive adhesion. Attention was then turned to outside the pericardium. After opening mediastinal pleura on left hilum, the fistula site was founded along the esophagus (Fig. 1E). We confirmed the 3 mm fistula with esophageal traction cautiously. From posterior pericardial reflection, three times of pledgeted 4–0 prolene sutures for ligation were paced on the fistula of the pericardial side gently. Fistula ligation and resection were performed, followed by an additional primary repair for esophageal defect. The repair procedure was finished with massive irrigation and chest drain tube insertion.

Postoperatively, the patient was treated for sequelae of an initial stroke and pneumonia. After one week without oral nutrition, we confirmed a well-repaired fistula site without residual lesion on endoscopy (Fig. 1B). He was discharged on postoperative day 31 with a tracheal tube.

**Case 2**

A 70-year-old man had radiofrequency catheter ablation for AF. Fifty days following the procedure, he was admitted with cognitive decline. Multiple cerebral infarct lesions were found on brain MRI. Blood cultures for fever revealed multiple *Streptococcus* spp. A CT scan of the chest was suggestive of an AEF (Fig. 1C).

Surgery was urgently performed. Surgical approach was the same as case 1. Through left thoracotomy on the 5th intercostal space, inspection of the pericardial space showed calcified adhesion. Through mediastinal space, esophagus was exposed and pulled gently. The 3 mm fistula on posterior aspect of the paricardium was isolated and ligated with several pledgeted 4–0 prolene sutures carefully. After fistulectomy, primary esophageal repair was done. With massive irrigation, a chest tube was then inserted. The wound was closed in a layered fashion.

The patient's postoperative course was focused on respiratory care with tracheostomy and rehabilitation for sequelae of cerebral infarction. Endoscopy revealed no specific findings after one week of fasting period (Fig. 1D). He was discharged on postoperative day 23 with a tracheal tube.

**Discussion**

Early diagnosis and surgical intervention are important in AEF. There are several treatment options for AEF, including esophageal stenting, intra-cardiac repair, extra-cardiac repair, and esophageal repair [2, 3, 5, 6].

Intra-cardiac repair of AEF needs sternotomy and cardiopulmonary bypass. Two-stage approach using intra-cardiac repair and esophageal repair has several disadvantages. It not only needs a cardiopulmonary bypass, but also needs position change of the patient [7]. Another hybrid technique [8]
including intra-cardiac repair and an endoscopic clipping has a risk for possibility of failure of the clipping procedure and the need for thoracic operation in case of refusulization.

Cases with single step repairs of AEF using thoracotomy approach with or without cardiopulmonary bypass have also been reported [9, 10]. Our cases had a treatment strategy similar to the method reported by Khandhar et al. [10]. However, we did not use intercostal muscle flaps or any stapler devices.

The single step of lateral thoracotomy for AEF has several advantages in patients who are judged to be free of left atrial active bleeding to the pericardial space. This lateral thoracotomy method can reduce operating time and eliminate the need for a cardiopulmonary bypass and a surgical position change. An AEF can be visually and reliably removed in one step. Even if the problem on the left atrium side remains, there is a room for further approach by open heart surgery. Like our two cases who are less likely to have active bleeding of the left atrial wall, we can check intra thoracic finding first and solve the fistula lesion. Even in those with multiple cerebral infarcts, it becomes burdensome to use cardiopulmonary bypass worrying about cerebral hemorrhagic change.

**Conclusion**

We encountered two AEF cases which are quite rare. Long-term follow-up data between surgical approaches for AEF are lacking. Our experience suggests that one step repair for AEF via lateral thoracotomy might be feasible in selected patients, especially in those with concomitant cerebral infarct.

**Declarations**

**Acknowledgements**

Not applicable

**Ethical Approval and consent to participate**

Ethics approval was obtained from the Institutional Ethics Committee. Consent was obtained from the two patient's families for participation in the study.

**Consent for publication**

Consent was obtained from the two patient's families for the publication of this report and any accompanying images.

**Competing interests**

The authors declare that they have no competing interests.

**Authors' contributions**
JL and DYK : study design, research and manuscript writing. JY, SBH, YHK and HWK : data collection. All authors read and approved the final manuscript.

Funding

Not applicable.

Availability of data and materials

As this paper is a letter to the editor, all generated or analyzed data are included in this article.

References


Figures
Figure 1

Perioperative findings about atrial-esophageal fistula in two patients. (A) CT scan of chest showing abnormal linear air densities in left atrium demonstrating an AEF in case 1. (B) An endoscopic finding with a well-repaired previous fistula site in case 1. (C) CT scan of chest revealing air bubbles at mediastinum suggestive of an AEF in case 2. (D) An endoscopic finding was read as unremarkable for prior AEF site covered with whitish exudate scar in case 2. (E) Intra-operative findings showing a well exposed, ligated AEF (white arrow, white arrowhead) site by left thoracotomy approach in case 1.