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RESPIRATORY CASE REPORTS

Treatment of Bronchopleural Fistula with Watanabe Spigots

Bronkoplevral Fistülün Watanabe Tıkaçları ile Tedavisi

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Abstract

Bronchopleural fistula (BPF) is a known complication of necrotizing pneumonia that is treated primarily with pleural drainage and antimicrobial therapy, while surgical treatments are generally reserved for those who do not respond to conservative management. Endobronchial spigots – a potentially less invasive approach to the treatment of bronchial occlusion – can be utilized when the culprit bronchi can be isolated and successfully blocked. We describe here the case of a 44-year-old female with a persistent right lower lobe bronchopleural fistula complicating necrotizing MRSa pneumonia, despite pleural drainage and directed antimicrobial therapy. The use of an endobronchial spigot for the bronchial occlusion of two bronchopulmonary segments led to an immediate reduction in the size of a large pleural cavity, contributing to significant symptomatic and biochemical improvement. The treatment can thus be considered an alternate cost-effective minimally invasive approach to the management of non-resolving bronchopleural fistula.

Keywords: *Bronchopleural fistula, bronchoscopy, interventional pulmonology, Watanabe spigot.*

Öz

Bronkoplevral fistül (BPF), nekrotizan pnömoninin bilinen bir komplikasyonudur. BPF için ilk tedavi, plevral drenaj ve antimikrobiyal tedaviyi içerir. Cerrahi tedaviler konservatif tedavinin başarısız olduğu durumlarda kullanılmaktadır. Daha az invazif bir alternatif tedavi seçeneği olan endobronşiyal tıkaçlar fistülün problematik bronş düzeyinde oklüzyonunu sağlamak amaçlı kullanılabilmektedir. Bu olgu sunumunda MRSa pnömonisine sekonder gelişen ve plevral drenaj ve antimikrobiyal tedaviye rağmen devam eden sağ alt lob bronkoplevral fistülü olan 44 yaşında bir kadını sunuyoruz. İki bronş segmenti spigot ile oklüde edilerek, hastada klinik ve laboratuvar düzelme ile birlikte plevral boşluğun boyutunda önemli ölçüde bir azalma sağlandı. Bu yöntem iyileşmeyen bronkofistüllerin tedavisi için alternatif, uygun maliyetli ve minimal invazif bir yaklaşım olabilir.

Anahtar Kelimeler: *Bronkoplevral fistül, bronkoskopi, girişimsel göğüs hastalıkları, Watanabe spigot.*

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A bronchopleural fistula (BPF) is an aberrant connection between the main stem, lobar or segmental bronchus and the pleural space resulting from an infectious, iatrogenic or structural etiology, and is associated with several risk factors (Table 1) (1-3). Depending on the etiology, presentation can range from subacute progressive symptoms of infection, including fever, purulent sputum, dyspnea and persistent air leak, to features of acute tension pneumothorax with respiratory failure (2).

Radiography may reveal intrapleural air, air-fluid levels, pneumothorax, pneumomediastinum and underlying pathologies such as pulmonary consolidation, as well as evidence of thoracic surgery and possibly the fistula itself (4,5). Bronchoscopy can allow the direct visualization of the defect, or the site of the fistula may be identified through other techniques, such as segmental balloon occlusion, demonstrating the cessation of a persistent air leak when a particular bronchus is blocked.

The first-line management of BPF when an infectious cause is suspected involves the insertion of an intercostal catheter (ICC) and antimicrobial therapy. Minimal suction, or a simple underwater seal drain, should be used to minimize air leakage through the BPF, as Pierson et al. (7) report that smaller air leaks provide a better prognosis. (6) Surgical repairs should be performed early (within 14 days) in patients with surgery-related BPF. Autologous blood patch pleurodesis and bronchoscopic therapies such as one-way valves, spigots, glues and other instilled agents are seeing increased use for the sealing of persistent BPFs.

CASE

A 44-year-old female with asplenia endured a prolonged 11-week hospitalization secondary to Influenza A complicated by methicillin-resistant Staphylococcus aureus (MRSA) necrotizing pneumonia and empyema, resulting in a bronchopleural fistula (BPF). The patient had a recent history of gastrointestinal spirochete infection and had lost 25 kg in weight over 6 months, a 10-pack/year smoking history, cannabis use via a water pipe and previous heavy alcohol consumption.

The patient had to date been treated with empiric antibiotics (moxifloxacin) in conjunction with antiviral oseltamivir, followed by directed therapy for MRSa (cultured from pleural fluid) in the form of a 10-week course of vancomycin. A right-sided intercostal catheter (ICC) was inserted for drainage of the empyema, with a resultant 9-day air leak that resolved spontaneously, and the drain was subsequently removed. A newly diagnosed left ventricular dysfunction was then detected on transthoracic echocardiography with a left ventricular ejection fraction of 18% (Simpson biplane). Notably, the patient had recorded normal ventricular function 1 month earlier.

The condition progressed to complicated pneumonia, despite the use of antibiotics, with a right-sided pleural effusion and empyema, and a subsequent large non-resolving pleural cavity with a bronchopleural fistula identified on thorax CT (Figure 1).

After 7 weeks of non-surgical management of the necrotizing pneumonia and BPF, including a prolonged course of antibiotics and ICC insertion, a large cavity persisted, and there had been minimal clinical improvement (Figure 1), and multidisciplinary team discussions led to the proposal of four treatment options for consideration:

1. Ongoing conservative therapy
2. Insertion of an ICC into the pleural cavity
3. Thoracic surgery with muscle flap repair of the BPF
4. Endoscopic options for the management of the BPF without an active air leak

Initially, a right-sided pulmonary resection with a large pectoralis major flap surgery was planned for the repair of the defect. Surgery was delayed, however, due initially to a mild nosocomial COVID-19 infection, and subsequently in consideration of the high anesthetic risk due to newly diagnosed heart failure, leading to the pursuit of the endoscopic option.

The bronchoscopic insertion of two Watanabe spigots for BPF closure under general anesthetic with muscle paralysis was performed 12 weeks after the initial presentation using a rigid 12mm tracheoscope. These included a 5mm Watanabe spigot (Figure 2) that was advanced into the right lower lobe to occlude the right posterobasal bronchus (RB10), sliced obliquely to facilitate its manipulation into the segment; and a 7mm spigot inserted into the posterolateral bronchus (RB9) (Figure 2). Intravenous vancomycin was continued for 5 days followed by trimethoprim/sulfamethoxazole for a further 2 weeks. The RB10 spigot spontaneously expectorated 72 hours after insertion, and so a larger (6mm) spigot was inserted (also obliquely sliced), with no further complications.

The patient attended a 6-week follow-up, during which a serial chest computed tomography (Figure 3) revealed a marked reduction in the size of the bronchopleural cavity. The symptoms showed a marked improvement, including a resolution of the cough, chest pain, fevers and night sweats, and the patient had gained 10 kg in weight and recorded an albumin increase from 23g/L (nadir during admission) to 40g/L at the time of review. Her functional status improved further, and she became able to consistently walk 12,000 steps per day. Repeat imaging at 6 months following the endobronchial Watanabe spigot insertion (Figure 3) revealed further improvements, with the resolution of the cavity defect, leaving only minor pleural thickening. The spigots were successfully removed 6 months after insertion.

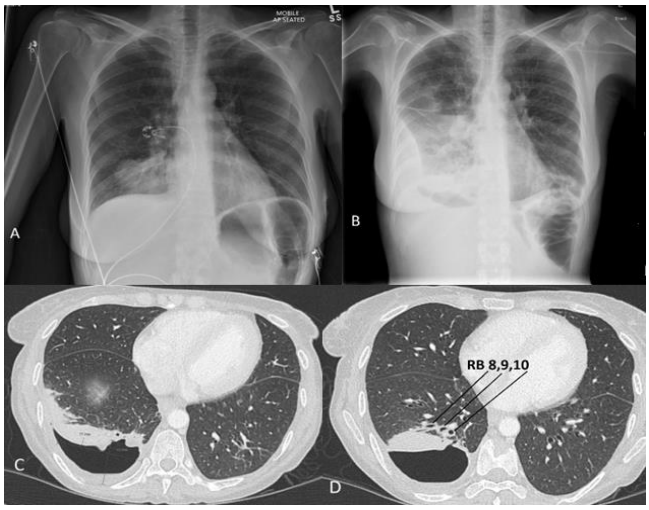

Figure 1: Progression of imaging demonstrating the development of the bronchopleural fistula. A: Initial chest radiograph on presentation. B: Progress radiograph <7 days following initial presentation. C: Computed tomography scan of the chest demonstrating the size of the defect. D: Computed tomography scan of the chest at the level of RB8/9/10

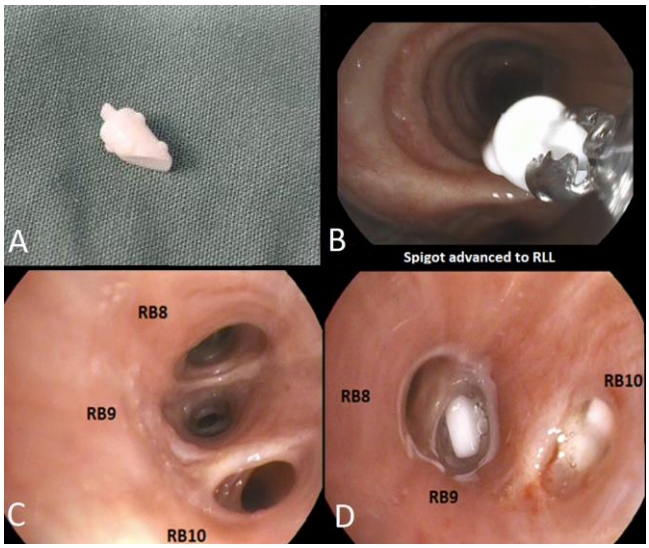

Figure 2: Watanabe Spigot insertion. A. Obliquely sliced Watanabe spigot. B. Spigot grasped by forceps endobronchially C. RB8,9,10 prior to spigot insertion. D. RB 8,9,10 following spigot insertion

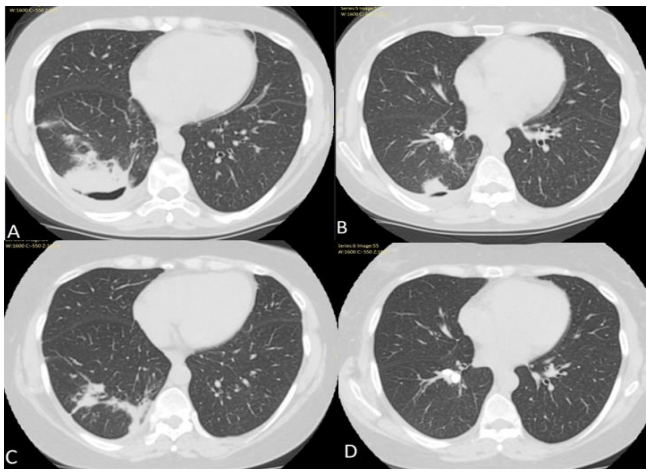

Figure 3: Follow-up imaging: A: Reduction in size of the BPF at 6 weeks (A) and 6 (C) months. B: Watanabe spigots in-situ (RB9 and RB10) at 6 weeks (B) and 6 (D) months

Figure 4: Variable sizes of the spigots available. Image sourced from Novatech online; https://www.novatech.fr/en/ewstm/ewstm/

DISCUSSION

To the best of our knowledge, this is the first registered use of Watanabe spigots for the treatment of a bronchopleural fistula (BPF) in Australia. These minimally invasive, endobronchially inserted spigots can be considered an effective treatment option for those who are unable to undergo standard thoracic surgery for the treatment of BPF. This case describes the clinical benefits and a reduction in the size of a large BPF cavity in a patient six months after insertion.

The use of Watanabe spigots was first described in 2001, after being developed in Japan by Yoichi Watanabe (8). The endobronchial Watanabe spigot was developed with three key features supporting successful bronchial occlusion, including (1) the spigot shape of the device and the studs on its surface that support the fixture of the spigot in place, (2) the presence of graspable segments at either end to facilitate placement and/or removal, and (3) the variety of sizes available to suit different bronchus sizes (Figure 4).

Watanabe investigated the use of bronchial blockades with spigots in 63 cases (9) for the treatment of intractable pneumothorax (in 40 cases), pyothorax with bronchial fistula (12 cases), pulmonary fistula (7 cases) and one case each for bronchial fistula, broncho-biliary fistula, broncho-esophageal fistula and broncho-gastric fistula. The technical success of the bronchial occlusion was reported in 97% of cases, with the total cessation of the air leakage reported in ~40% of the cases, and a marked reduction in ~38% of the cases. It was noted that it took 2–3 days for the air leakage to resolve in some patients following the insertion of the spigots. The average number of spigots inserted in each case was ~4. The complications that developed following spigot insertion included dyspnea (3.3%), pneumonia (3.3%) and fever (1.7%). The spigots were removed 2–4 weeks after being inserted, where possible.

Further cases treated with endobronchial Watanabe spigots were reviewed over a 13-year period in a study by Himeji et al. (10) in which all cases had intractable pneumothorax or pyothorax with a bronchial fistula, had failed appropriate drainage for 2 weeks and were unsuitable for thoracic surgery. Of the total 21 cases identified, 10 had intractable pneumothorax, seven had pyothorax with a bronchial fistula, and four had postoperative air

leakage. The successful treatment rates by primary disease were 80% for intractable pneumothorax, 100% for pyothorax with a bronchial fistula and 75% for postoperative air leakage. The limitations of the study include the concurrent use of additional therapies, which included washings, bronchial injection of fibrin glue and pleural adhesion. Similar to Watanabe, the air leak resolved immediately in some cases, but took up to 2 days in others following spigot insertion, and several spigots were often required (mean 6.5 per patient). The complications encountered included spigot migration and a single case of aspergillus infection 4.5 years following the insertion of the endobronchial Watanabe spigots, suggesting that the spigots should be considered for removal from clinically appropriate patients with a good long-term prognosis.

Alternative bronchoscopic treatment modalities for the management of BPF include adhesives, hemostatic agents, sclerosing agents, thermal occlusions, stents and one-way valves. There is a lack of quality data on these techniques, and there have been no randomized controlled trials to date. One-way endobronchial valves (EBVs) were first used for bronchial occlusion in the early 2000s. A one-way endobronchial valve can be inserted via a flexible bronchoscope, and can prevent air from entering the affected segmental bronchus, but allowing the egress of air and mucus. These features of the one-way valve have secured it a pivotal role in endobronchial lung volume reduction procedures. One retrospective study reviewed data from over a 3-year period on the use of EBV for the treatment of BPF in 26 patients (11). Prior to insertion, a Chartis assessment was performed to identify the culprit lobe/segment, and a total of 46 EBVs were inserted, with a median of two valves per procedure. The underlying causes of BPF included post-operative (50%), pneumothorax (15%) and the remaining infective precipitants; non-tuberous mycobacterial disease (19%) and tuberculosis (12%). Prior to valve insertion, 16 patients were fitted with a chest tube. The average duration of ICC drainage was 88 days (range 14–222) prior to EBV. Following valve insertion, the ICC remained in place for an average of 28.2 days (range 2–98). The authors concluded that EBVs succeeded in improving the rate of BPF resolution by 73.1%, while five of the 26 underwent additional procedures to assist with management at different time points, which were not specified. These included the use of silicone plugs, lauryl alcohol, argon laser plasma coagulation, ventricular septal and umbrella occlusion devices. Complications included bronchial bleeding requiring embolization following the removal of the EBV in one patient, while two further patients required thoracic surgical intervention. Previous studies have reported complications associated with EBVs, and a systematic review of EBV for the treatment of persistent air leaks revealed such complications as migration or expectoration of the valves, moderate oxygen desaturation, as well as infection of the related lung (12).

A prospective, randomized unblinded study was conducted investigating the success rates of treatments of persistent air leaks following secondary spontaneous pneumothorax using either selective bronchial autologous blood patches (ABP) with the addition of thrombin, silicone bronchial spigots (BS) or prolonged ICC (13). All of the 150 patients included in the study had persistent air leaks for 7 days following the insertion of an ICC, and had at least one of the following conditions: chronic obstructive pulmonary disease (n=65), pulmonary bullae (n=47), pneumonia (n=24), pulmonary cancer (n=20), bronchiectasis (n=11), asthma (n=9) and/or pulmonary fibrosis (n=6). The size of the pneumothorax was not statistically significant between the three groups; all patients were observed for up to 14 days following intervention. The resolution of the pneumothorax within 14 days was achieved most frequently in the silicon spigot group (84%), followed by the ABP (82%), and least frequently in the ICC alone group (60%). Comparatively, ABP and BS were significantly superior to chest tube drainage alone ($p = 0.008$), and were also statistically superior in terms of the duration of air leak cessation; lung re-expansion and hospital stay vs. chest tube drainage ($P<0.001$ for all). Fever, cough and chest pain complications were similar between the three groups. Temporary hemoptysis of <10mls was significantly more prevalent in the blood patch and silicon spigot groups, occurring in 100% of cases in both groups, compared to 12% in the ICC alone group. Spigot displacement occurred in 8% of cases. As endobronchial Watanabe spigots were not available from China, the silicon spigots used were individually trimmed to create dumbbell-shaped plugs from chin silicone implants.

Watanabe spigots differ significantly from EBVs. They are customizable, and can be cut to optimize airway fit. As outlined in our case, we were able to obliquely slice the spigot to ensure an optimal fit and thus successful insertion into the target bronchus. EBVs are not modifiable, however the insertion procedure is technically less complex. Cost-effectiveness is vastly different – EBVs are significantly more expensive per unit, but have the benefit of one-way drainage, allowing the clearance of secretions while potentially minimizing post-obstructive pneumonia.

CONCLUSION

We conclude that the endobronchial insertion of Watanabe spigots, a customizable option for the management of bronchopleural fistula, can result in the successful treatment of fistulae leading to clinical and radiological improvement, and so should be considered in

appropriate patients as a minimally invasive and cost-effective therapy.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - J.W., J.F., D.N., S.S., B.F.; Planning and Design - J.W., J.F., D.N., S.S., B.F.; Supervision - J.W., J.F., D.N., S.S., B.F.; Funding -.; Materials -; Data Collection and/or Processing -; Analysis and/or Interpretation -; Literature Review -; Writing -; Critical Review -

REFERENCES

1. Salik I, Vashisht R, Abramowicz AE. Bronchopleural Fistula. [Updated 2022 May 8]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2022 Jan-. Available from: https://www.ncbi.nlm.nih.gov/books/NBK534765/.
2. Sarkar P, Chandak T, Shah R, Talwar A. Diagnosis and management bronchopleural fistula. Indian J Chest Dis Allied Sci 2010; 52:97-104. [CrossRef]
3. Aliaga F, Grosu HB, Vial MR. Overview of bronchopleural fistula management, with a focus on bronchoscopic treatment. Curr Pulmonol Rep 2022; 11:65-73. [CrossRef]
4. Kim EA, Lee KS, Shim YM, Kim J, Kim K, Kim TS, et al. Radiographic and CT findings in complications following pulmonary resection. Radiographics 2002; 22: 67-86. [CrossRef]
5. Westcott JL, Volpe JP. Peripheral bronchopleural fistula: CT evaluation in 20 patients with pneumonia, empyema, or postoperative air leak. Radiology 1995; 196: 175-81. [CrossRef]
6. Pierson DJ, Horton CA, Bates PW. Persistent bronchopleural air leak during mechanical ventilation: a review of 39 cases. Chest 1986; 90:321-3. [CrossRef]
7. Liberman M, Cassivi SD. Bronchial stump dehiscence: update on prevention and management. Semin Thorac Cardiovasc Surg 2007; 19:366-73. [CrossRef]
8. Watanabe Y, Matsuo K, Tamaoki A, Hiraki S, Moriyama S. Bronchial embolization using an endobronchial Watanabe spigot for intractable pneumothorax and bronchial fistula. J Jpn Soc Bronchol 2001; 23:510-5.
9. Watanabe Y, Matsuo K, Tamaoki A, Komoto R, Hiraki S. Bronchial occlusion with endobronchial Watanabe spigot. J Bronchol 2003; 10:264-7. [CrossRef]
10. Himeji D, Tanaka GI, Fukuyama C, Shiiba R, Yamanaka A, Beppu K. Clinical evaluation of endoscopic bronchial occlusion with an endobronchial Watanabe spigot for the management of intractable pneumothorax, pyothorax with bronchial fistula, and postoperative air leakage. Intern Med 2020; 59:1835-9. [CrossRef]
11. Song X, Gu Y, Wang H, Zhang L. The efficacy of endobronchial valves for the treatment of bronchopleural fistula: a single-arm clinical trial. J Thorac Dis 2022; 14:712-720. [CrossRef]
12. Ding M, Gao YD, Zeng XT, Guo Y, Yang J. Endobronchial one-way valves for treatment of persistent air leaks: a systematic review. Respir Res 2017; 18:186. [CrossRef]
13. Zhang HT, Xie YH, Gu X, Li WP, Zeng YM, Li SY, et al. Management of persistent air leaks using endobronchial autologous blood patch and spigot occlusion: a multicentre randomized controlled trial in China. Respiration. 2019; 97:436-43. [CrossRef]