

# Case Report

## Endoscopic treatment of tracheobronchial tree fistulas using atrial septal defect occluders: preliminary results\*

Tratamento endoscópico de fistulas da árvore traqueobrônquica com dispositivos para a correção de defeitos do septo interatrial: resultados preliminares

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### Abstract

Fistulas in the tracheobronchial tree (bronchopleural and tracheoesophageal fistulas) have a multifactorial etiology and present a variable incidence in the literature. In general, the related morbidity and mortality are high. Once such a fistula has been diagnosed, surgical closure is formally indicated. However, the clinical status of affected patients is usually unfavorable, which precludes the use of additional, extensive surgical interventions. In addition, attempts at endoscopic closure of these fistulas have seldom been successful, especially when the fistula is large in diameter. We report the cases of three patients submitted to endoscopic closure of fistulas, two of which were larger than 10 mm in diameter, by means of the insertion of atrial septal defect occluders. The procedure was minimally invasive, and the initial results were positive. The results indicate that this is a promising technique for the resolution of tracheobronchial tree fistulas.

**Keywords:** Bronchial fistula; Tracheoesophageal fistula; Bronchoscopy; Respiratory therapy.

### Resumo

As fistulas da árvore traqueobrônquica, sejam elas broncopleurais ou traqueoesofágicas, apresentam etiologia multifatorial, com incidência variável na literatura. Em geral, apresentam alta morbidade e mortalidade, com indicação formal de correção cirúrgica. Entretanto, a condição clínica dos pacientes muitas vezes não permite uma reintervenção cirúrgica de grande porte. Além disso, as tentativas de fechamento endoscópico raramente têm sucesso, principalmente em fistulas de grande diâmetro. Relatamos os casos de três pacientes submetidos ao fechamento endoscópico de fistulas, sendo duas maiores que 10 mm, com a aplicação de dispositivos oclusores utilizados na cardiologia intervencionista, de forma minimamente invasiva e com resultados iniciais positivos. Esses dados sinalizam que essa pode ser uma técnica promissora na resolução de fistulas da árvore traqueobrônquica.

**Descritores:** Fistula brônquica; Fístula traqueoesofágica; Broncoscopia; Terapia respiratória.

### Introduction

Fistulas in the tracheobronchial tree have a multifactorial etiology and present a variable incidence in the literature. The related morbidity and mortality are high. Bronchopleural fistulas usually result from surgical procedures involving pulmonary resection; the incidence of these

fistulas ranges from 0 to 28%,<sup>(1,2)</sup> and the mortality rate ranges from 16 to 72%.<sup>(3)</sup> Non-neoplastic tracheoesophageal fistulas can be congenital,<sup>(4,5)</sup> iatrogenic<sup>(6)</sup> or related to thoracic trauma. Once such a fistula has been diagnosed, surgical closure is formally indicated.

\* Study carried out at the University of São Paulo School of Medicine *Hospital das Clínicas*, São Paulo, Brazil.

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However, the clinical status of affected patients is usually unfavorable, which precludes the use of additional, extensive surgical interventions.

In an attempt to avoid surgical closure, various endoscopic techniques have been described. However, endoscopic closure of these fistulas has seldom been successful, especially when the fistula is large in diameter.<sup>(7-12)</sup>

In the three cases described next, we report our initial experience in performing endoscopic closure of fistulas, one of which was a chronic tracheoesophageal fistula and two of which were complete bronchial stump fistulas, by means of the insertion of atrial septal defect occluders. This study was approved by the Research Ethics Committee of the University of São Paulo School of Medicine Heart Institute.

## Case reports

### Case 1

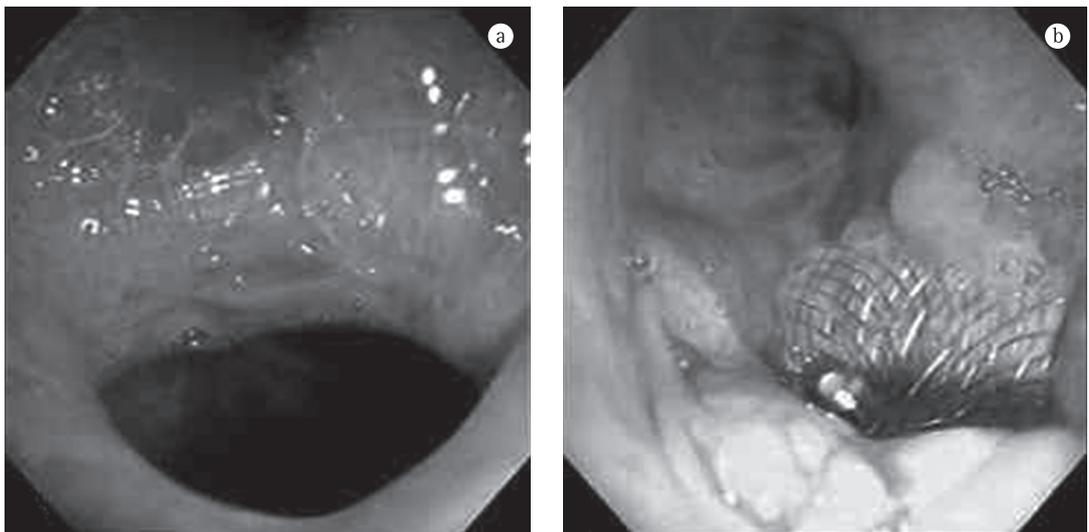
A 53-year-old male patient presented with hepatitis C, abdominal aortic aneurysm, carotid aneurysm, systemic arterial hypertension and grade II heart failure. The patient was a smoker (70 pack-years) and had a history of treated pulmonary tuberculosis and right upper lobe aspergilloma with hemoptysis.

The patient underwent right upper lobectomy and developed a bronchial stump fistula. Numerous attempts at closure were unsuccessful,

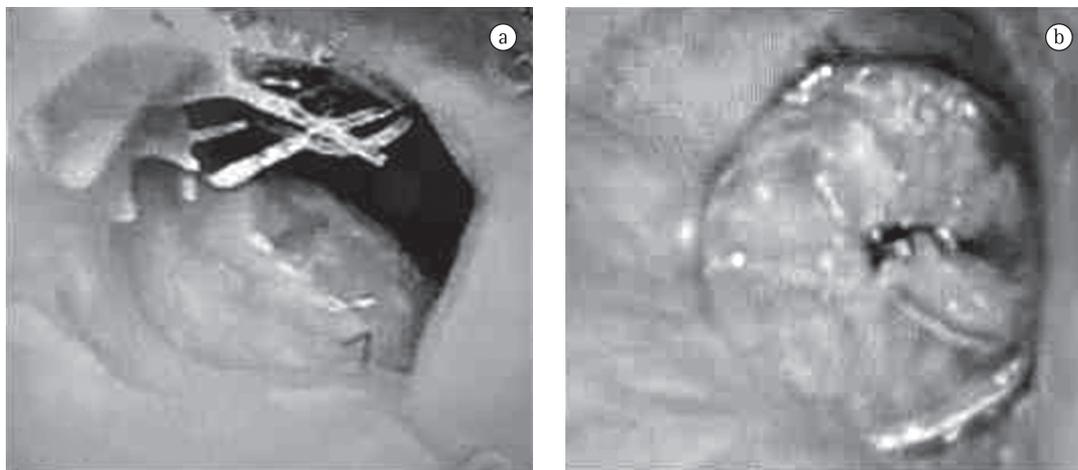
and open drainage of the residual pleural cavity was maintained. Bronchoscopy revealed that the fistula measured 12 mm, and a 15-mm atrial septal defect occluder (Occlutech Figulla; Occlutech GmbH, Jena, Germany) was therefore chosen. We began catheterization of the fistula using a polytetrafluoroethylene-coated guide wire and then advanced the occluder-sheath set to the bronchial stump, the distal disc being released in the pleural cavity and the proximal disc being released in the bronchial tree. There was an immediate reduction in air leak, and the patient was discharged from the hospital after 12 h. The evaluation performed after a follow-up period of 180 days revealed that granulation tissue had closed the fistula almost completely (Figure 1).

### Case 2

A 69-year-old male patient presented with hypertension and atypical carcinoid tumor. The patient was a smoker (50 pack-years). The patient underwent right pneumonectomy. The patient developed a complete bronchial stump fistula of approximately 18 mm with severe air leak. An additional surgical intervention was ruled out due to surgical risk. A 20-mm atrial septal defect occluder (GORE-Helix; Gore, Flagstaff, AZ, USA) was therefore placed under general anesthesia, closing the fistula completely. There was clinical stabilization of the profile, and the patient was



**Figure 1** – In a), bronchopleural fistula after right upper lobectomy. In b), appearance of the site 180 days after the device had been placed.



**Figure 2** - In a), complete stump fistula after right pneumonectomy. In b), appearance of the site immediately after the device had been placed.

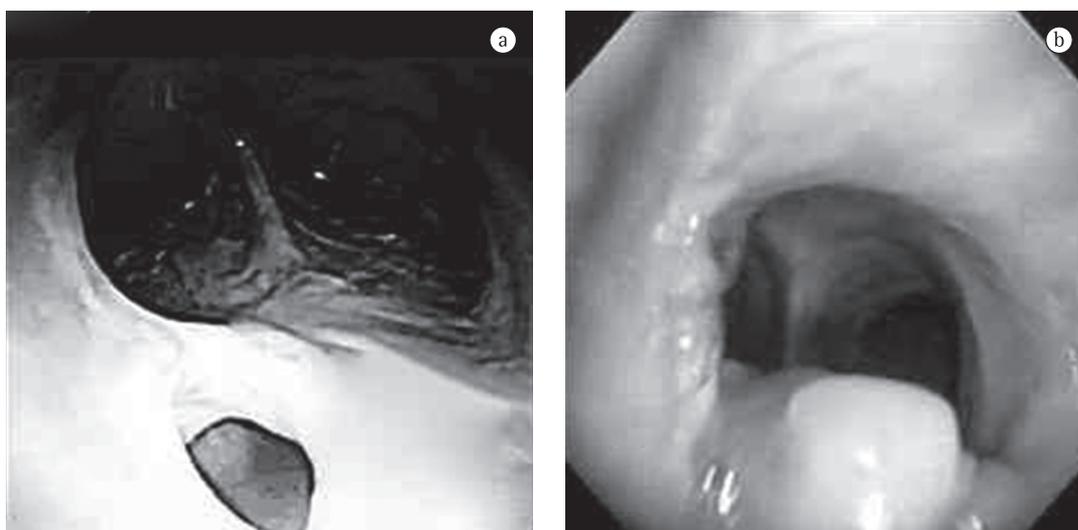
discharged from the hospital 5 days after the procedure.

The evaluation performed after a follow-up period of 30 days revealed that 40% of the occluder had been covered by epithelium. On day 45, the device migrated to the pleural cavity. The patient, however, remained oligo-symptomatic. Bronchoscopy showed granulation tissue that reduced the diameter of the fistula to 7 mm. Another bronchoscopy, performed on day 75, revealed granulation tissue and repair with fibrosis, which reduced the diameter of the fistula to 4 mm and suggested that the healing process was still in progress despite the absence of the occluder (Figure 2).

### Case 3

A 73-year-old male patient presented with malnutrition, a history of severe acute pancreatitis and a diagnosis of distal tracheoesophageal fistula after prolonged intubation. The patient also presented persistent cough, worsened by the ingestion of solids and liquids and by the change of decubitus. The patient had refused the initial indication for surgical closure, an option that was then discarded due to the high surgical risk.

Bronchoscopy revealed a tracheoesophageal fistula of approximately 5 mm in the distal trachea. We chose to place a 20-mm GORE-



**Figure 3** - In a), tracheoesophageal fistula of 5 mm. In b), complete resolution 7 days after the device had been removed.

Helix atrial septal defect occluder. After having been released, however, the device did not lock properly into place, which resulted in flapping of its tracheal face during exhalation and led to accumulation of secretion. We decided to substitute the 20-mm occluder for a 15-mm occluder. The 20-mm occluder locked into place properly, and the profile improved significantly. The patient was discharged from the hospital after 5 days, without cough (even during the ingestion of liquids). The evaluation performed after a follow-up period of 120 days revealed that the esophagus was normal and that the disc placed in the esophagus had been completely incorporated. Bronchoscopy showed epithelialization of the tracheal disc, which protruded partially toward the tracheal lumen. We decided to remove the occluder. Local repair and complete closure occurred after 7 days (Figure 3).

## Discussion

One of the forms of endoscopic closure of fistulas in the tracheobronchial tree is the apposition of different materials with the purpose of forming a closing plug. However, it is difficult to securely affix these materials at the site of the fistula, and their displacement can, in addition to reopening the fistula, release a foreign body into the airway.

Endoscopic techniques have been used to inject different substances (alcohol, tetracycline, hypertonic glucose) in the submucosa, with the purpose of inducing inflammation and closing the fistula. Part of the problem with these techniques is that the aggression is not modulated and can result in ischemia and necrosis and therefore worsen the problem.

A recent study described the closure of a 5-mm bronchopleural fistula by means of a device used in the treatment of heart septal defects.<sup>(13)</sup>

Our objective is to use this type of device, the biocompatibility of which has been confirmed by its extensive use in cardiac interventions.

The structural design of this type of device favors its permanence, anchored in the fistula, and allows the closure of fistulas of large diameter, even of those that were previously untreatable through endoscopy. The device can act as a substrate for an organized inflammatory response, accompanied by granulation tissue formation and epithelialization of the device.

We therefore used two devices that are employed in cardiac interventions and that are also widely used clinically.

The Occlutech Figulla device is composed of two nitinol discs that are sequentially released from the applicator. Thus, the distal disc can be placed at the extremity of the stump in the pleural cavity, whereas the proximal disc is released in the bronchial lumen; the connecting waist between the two discs becomes firmly attached to the fistula and avoids the displacement of the device.<sup>(13)</sup>

The GORE-Helix device consists of a single nitinol wire attached to a continuous tape of polytetrafluoroethylene that forms two hybrid discs in the shape of a spiral that, when released, attach themselves to each side of the fistula.<sup>(14)</sup> Because the GORE-Helix device is more malleable than is the Occlutech Figulla device, we believed the former would be more appropriate to close the tracheoesophageal fistula for two reasons: first, because the two organs involved presented motility; and second, because we were concerned that a rigid device might widen the fistula.<sup>(14)</sup>

If our initial results are corroborated by future studies, this might be a promising technique for the endoscopic treatment of fistulas in the tracheobronchial tree.

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