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Treatment of Tracheoesophageal Fistula With Occluder Device Assisted by Three-Dimensional Printing

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Tracheoesophageal fistula is a pathologic communication between the trachea and the esophagus that can cause severe complications and progress rapidly, potentially leading to death in a short period. This report describes a case of malignant tracheoesophageal fistula treated with an atrial septal defect occluder under digital subtraction angiography guidance using 3-dimensional printing assistance. Postoperative computed tomography and bronchoscopy showed good recovery, significant relief of infection symptoms, and marked improvement in airway irritation symptoms.

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Tracheoesophageal fistula (TEF) is a pathologic communication between the trachea and the esophagus that is manifested clinically as recurrent lung infections and choking during meals. It progresses rapidly and can lead to death in a short period. Approximately 75% of malignant TEF cases are caused by esophageal tumors.[1] For malignant TEF, palliative treatment, including conservative treatment and stent placement, is often used.[2] Atrial septal defect (ASD) occluders are typically used to treat congenital heart disease but are now gradually being applied to treat TEF.[3] Applying 3-dimensional (3D) printing technology for rational preoperative planning can achieve individualized and precise treatment of patients with malignant TEF, helping to select appropriate ASD occluder models, improving surgical success rates, and reducing the incidence of complications. In this report, we describe a case at our center of malignant TEF treated with an ASD occluder under digital subtraction angiography guidance using 3D printing assistance.

A 56-year-old man was admitted because of an intractable TEF. Before admission, the patient had undergone esophagectomy for squamous cell carcinoma of the esophagus. Seven days after operation, the patient had chest tightness and high fever with a peak temperature of 39 °C. Computed tomography (CT) imaging revealed an anastomotic fistula and bilateral pneumonia. Subsequent examination with CT (Figures 1A-1C) and bronchoscopy (Figure 1D) indicated the presence of a fistula located 8.5 cm from the sternal notch and 6 cm from the vocal cords.

Given the large size of the fistula and the severity of the lung infection, it was decided to close the fistula with an ASD occluder. This was accomplished by advancing a vertebral artery catheter and hydrophilic guidewire through the oral cavity, into the esophagus, and across the fistula. Injection of contrast material revealed extrusion of the contrast agent into the trachea. By a combination of catheter and guidewire, an 8F sheath was advanced across the fistula, and a 6-mm ASD occluder was deployed after proper positioning. Follow-up imaging with injection of contrast material and DynaCT (Siemens) scanning revealed excellent positioning of the occluder. However, the patient suddenly experienced severe coughing, and the occluder was expectorated. Postoperative analysis revealed that there was an error in the measurement of the fistula size by CT and bronchoscopy, which led to selection of an occluder that was too small. The occluder caused discomfort and coughing, resulting in changes in the size of the fistula and eventual occluder detachment. A 3D reconstruction of the patient's imaging information was performed, and a 1:1 model was designed using 3D printing technology to replan the surgical approach. In vitro simulation was used to seal the fistula with an ASD occluder, and a 10-mm ASD occluder was selected (Figure 2). Three days after the second surgery, follow-up CT (Figures 3A-3C) and bronchoscopy (Figure 3D) showed a good recovery, with significant improvement in infection and airway irritation symptoms. The patient's quality of life was significantly improved and the inflammation in both

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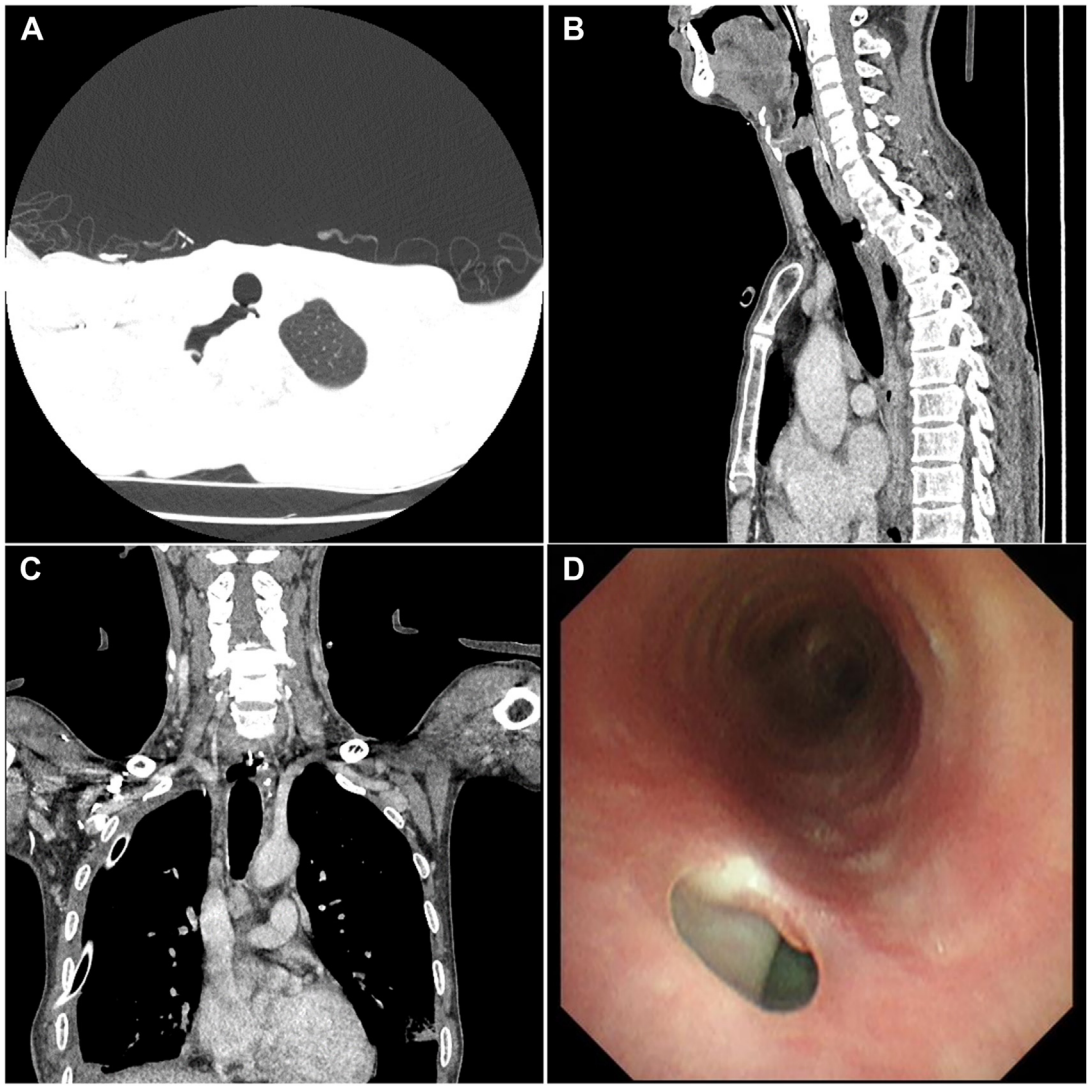

FIGURE 1 (A-C) Chest computed tomography images displaying the tracheoesophageal fistula, pleural effusion, and pneumothorax in lung window, sagittal view, and coronal view, respectively. (D) Bronchoscopy image showing the fistula located 8.5 cm from the sternum and 6 cm from the vocal cords.

lungs basically disappeared when the CT examination was performed again 2 months after the surgery.

COMMENT

TEF is a pathologic condition in which there is an abnormal communication between the trachea and the esophagus. Based on the cause, TEF can be divided into benign and malignant types; approximately 50% of benign TEF cases result from tracheostomy and mechanical ventilation, whereas esophageal tumors account for about 75% of malignant TEF cases.[1] The formation of the fistula leads to the reflux of gastric irritants into the trachea, causing recurrent lung infections. If left untreated, the fistula can be chronic and severely affect the patient's quality of life, with a high death rate.

For benign TEF, surgical intervention can achieve a curative effect. Palliative care is the mainstay for treatment of malignant TEF, which includes conservative management and stent placement.[2] Palliative care can alleviate various symptoms caused by TEF to a certain extent, improving the patient's quality of life. Stent placement is suitable for most patients with malignant TEF, with some requiring the use of both tracheal and esophageal stents to achieve complete occlusion.[4] However, long-term stent placement can lead to local granulation tissue hyperplasia, sputum retention,

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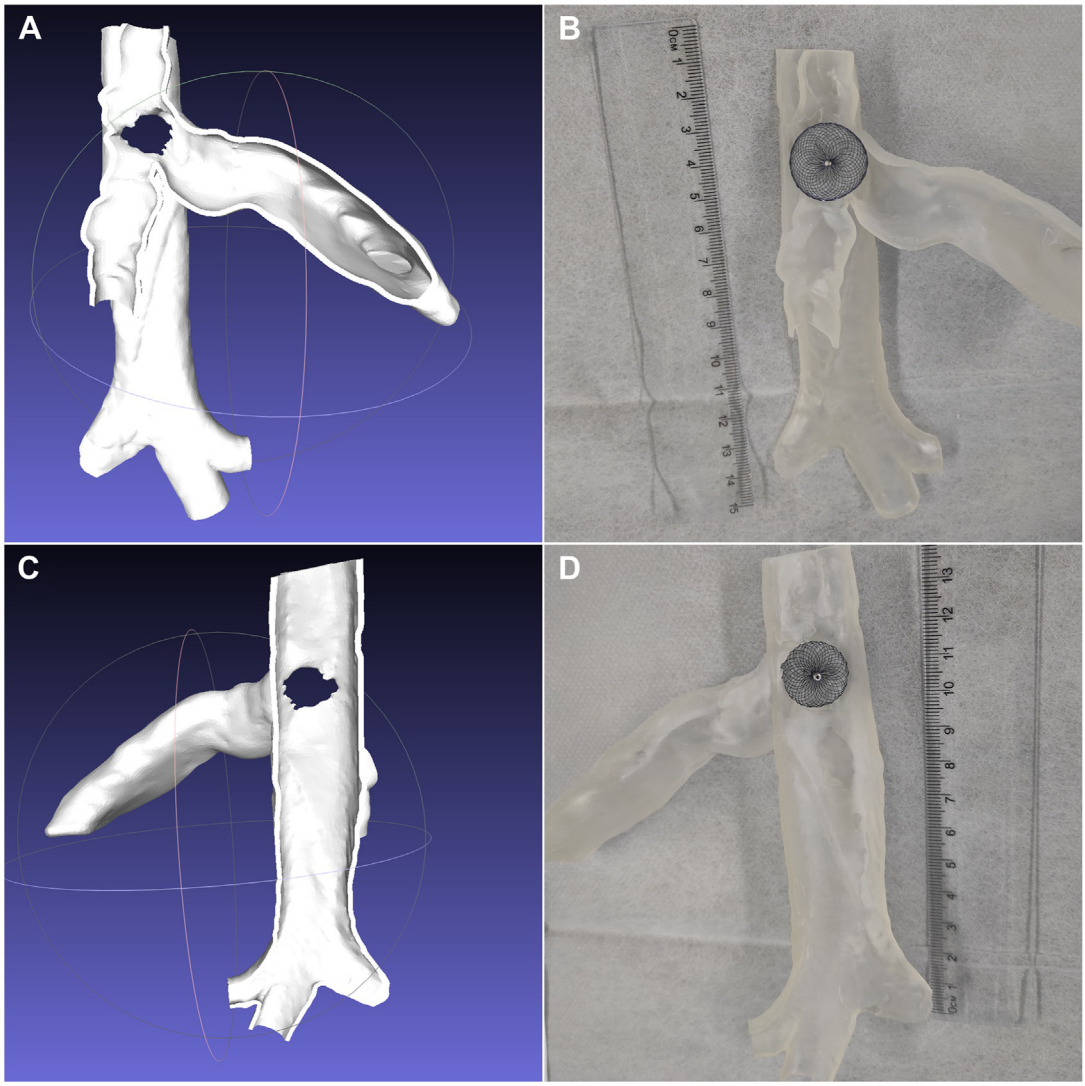

FIGURE 2 (A, B) Three-dimensional printed model of the esophagus. (C, D) Three-dimensional printed model of the trachea.

airway narrowing, exacerbation of lung infections, and even fistula enlargement due to stent tension.[5]

Scordamaglio and colleagues[6] first reported the use of an ASD occluder to treat TEF, achieving good therapeutic effects. However, studies on the use of occluders to treat TEFs are mostly case reports, and there is no uniform standard for occluder size, leading to various potential risks. There have been reports of severe airway obstruction after the use of ASD occluders to treat TEF.[7] Unlike for stents, the selection of occluder models is crucial in treatment. Currently, clinicians often choose larger occluders to achieve complete occlusion, but larger occluders not only have poor adherence to the fistula but also significantly reduce the cross-sectional area of the local airway, leading to airway obstruction. In addition, patients with larger occluders can experience severe foreign body sensation, and long-term relative movement between the occluder and trachea can even form new fistulas. If a more conservative occluder is chosen, it may not achieve complete occlusion, leading to surgical failure, as in the case of this patient's first surgery.

With the advancement of 3D printing technology, its application in various fields, particularly in medicine, holds significant importance in resolving clinical conundrums.[8] The technology involves reconstructing 3D models based on the precise imaging information of each patient and using specific materials to fabricate a 1:1 scale physical model. This model is beneficial for preoperative diagnosis, surgical planning, and simulation. Employing the physical model for in vitro

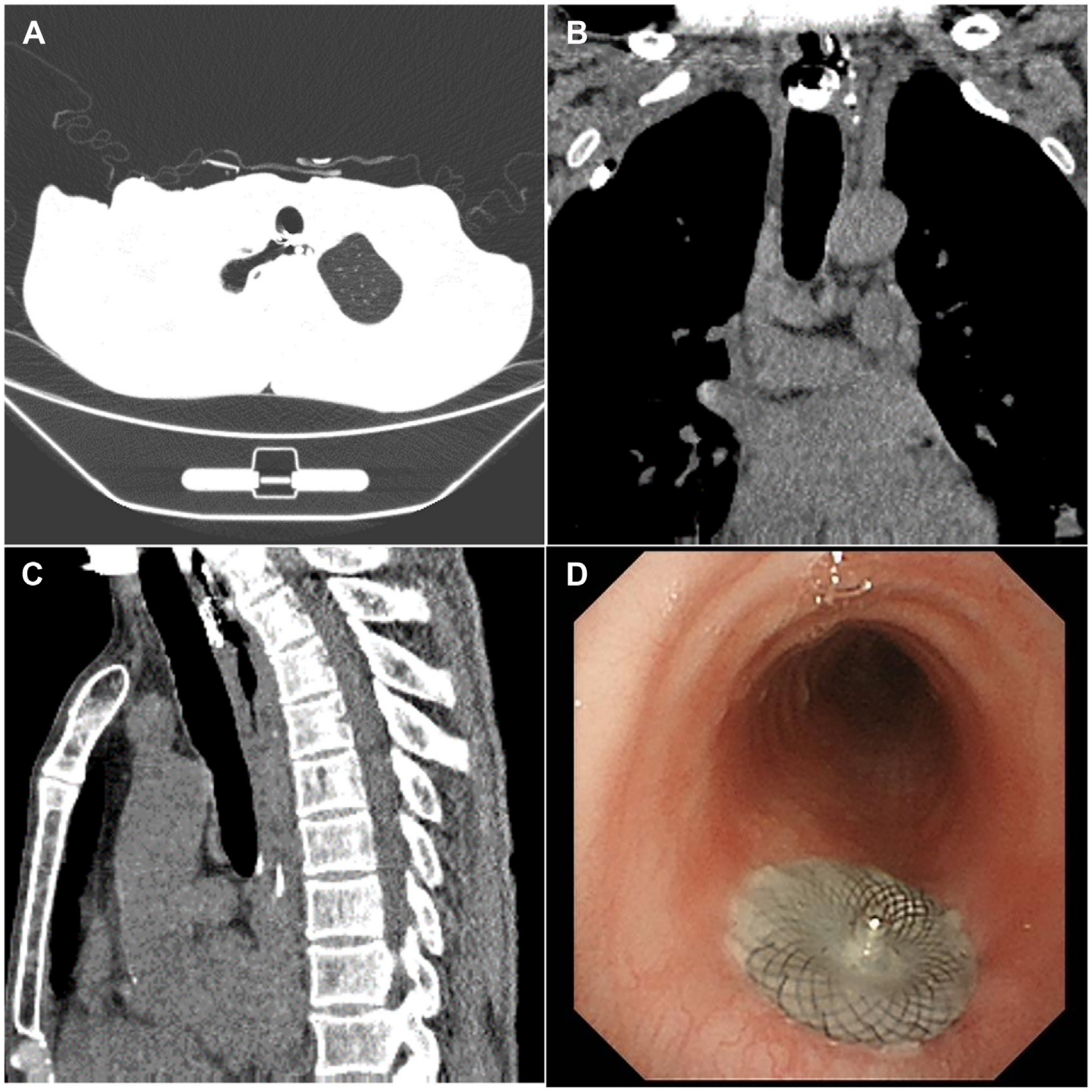

FIGURE 3 (A-C) Chest electronic computed tomography images displaying the satisfactory location of the occluder and reduced lung infection and pleural effusion compared with previous images in lung window, coronal view, and sagittal view, respectively. (D) Bronchoscopy image showing complete occlusion of the fistula and satisfactory placement of the occluder.

simulation before surgery enables the formulation of solutions for potential surgical problems, ultimately improving surgical success rates and reducing complications. In this case, owing to the failure of the initial surgery, we used the patient's latest imaging data for 3D reconstruction and 3D printing of the physical model. We simulated the surgery using the model and selected an appropriate occluder that could guarantee complete blockage while minimizing the occurrence of complications by closely adhering to the tracheal wall. As of the date of this report, the patient has experienced no further discomfort or symptoms of pulmonary infection, including fever.

In conclusion, employing a digital subtraction angiography-guided ASD occluder with the assistance of 3D printing in treating malignant TEF holds a certain degree of feasibility. It assists clinicians in preoperative surgical planning, improves surgical success rates, and reduces related complications.

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PATIENT CONSENT

Obtained.

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