

Case Report

Reoperation for complicated tracheoesophageal fistula after surgery of a tracheal lymphoma

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Abstract: A 52-year-old woman with tracheal lesions underwent tracheal resection and carina reconstruction directly because histopathologic diagnosis was not obtained before surgery. Postoperative pathological analysis confirmed a diagnosis of mucosa-associated lymphoid tissue (MALT) lymphoma. Subsequently, she suffered serious complications of tracheal stenosis, bronchial stenosis, and tracheoesophageal fistula (TEF), which failed to improve with conservative treatment. A reoperation using esophageal double patch technique was successfully performed. MALT lymphoma of the trachea is extremely rare. This case emphasizes the importance of considering MALT lymphoma in the differential diagnosis of tracheal tumors, and obtaining a histopathologic diagnosis before treatment. The esophageal double patch technique can be a choice in treating complicated TEF.

Keywords: Tracheal neoplasms, lymphoma, surgery, reoperation, tracheoesophageal fistula

Introduction

Tumors of the trachea are uncommon. The most common histologic types are squamous cell carcinoma and adenoid cystic carcinoma, for which surgery is the first choice of treatment [1]. Mucosa-associated lymphoid tissue (MALT) lymphoma most commonly occurs in the stomach, salivary glands, orbital soft tissue, thyroid, and lungs. Primary MALT lymphoma of the trachea is extremely rare, and there are only a few case reports in the literature [2, 3]. There is no standard treatment modality for MALT lymphoma of the trachea due to its rarity. The reported treatment methods include chemotherapy, radiotherapy, cryotherapy and laser therapy. It is crucial to obtain a pathological diagnosis before treatment, although difficult sometimes [2].

Tracheal stenosis (TS) or bronchial stenosis (BS) is a common complication after tracheal resection and reconstruction [4]. Reoperation is indicated for patients if dilation fails to provide sustainable relief. However, reoperation is challenging, especially when TS or BS is accompanied by tracheoesophageal fistula (TEF) [5, 6]. Here we report a successful case of reoper-

ation in a patient complicated by TS, BS, and TEF, occurring after surgery of a tracheal MALT lymphoma, and with emphasis on the experience and lessons.

Case report

A 52-year-old woman, with cough for one month, was admitted to our hospital. Bronchoscopy showed multiple nodular masses in the trachea, from the carina to 4 cm above the carina (**Figure 1A, 1B**). Chest computed tomography (CT) revealed multiple soft tissue nodules in the lower part of the trachea (**Figure 1C, 1D**). Histopathologic diagnosis was not obtained before surgery due to the following reasons: i) the initial bronchoscopic biopsy of the tracheal lesions was not successfully performed due to bleeding; ii) the patient refused to undergo a re-biopsy because of worrying about the risk of massive bleeding; iii) we did not have a rigid bronchoscope to deal with serious bleeding when performing biopsy, especially for lesions located in the lower part of trachea. iv) we took it for granted that the tracheal lesions might be the common histologic types for which surgery was the preferred treatment. The patient then underwent tracheal resection of a length of 5

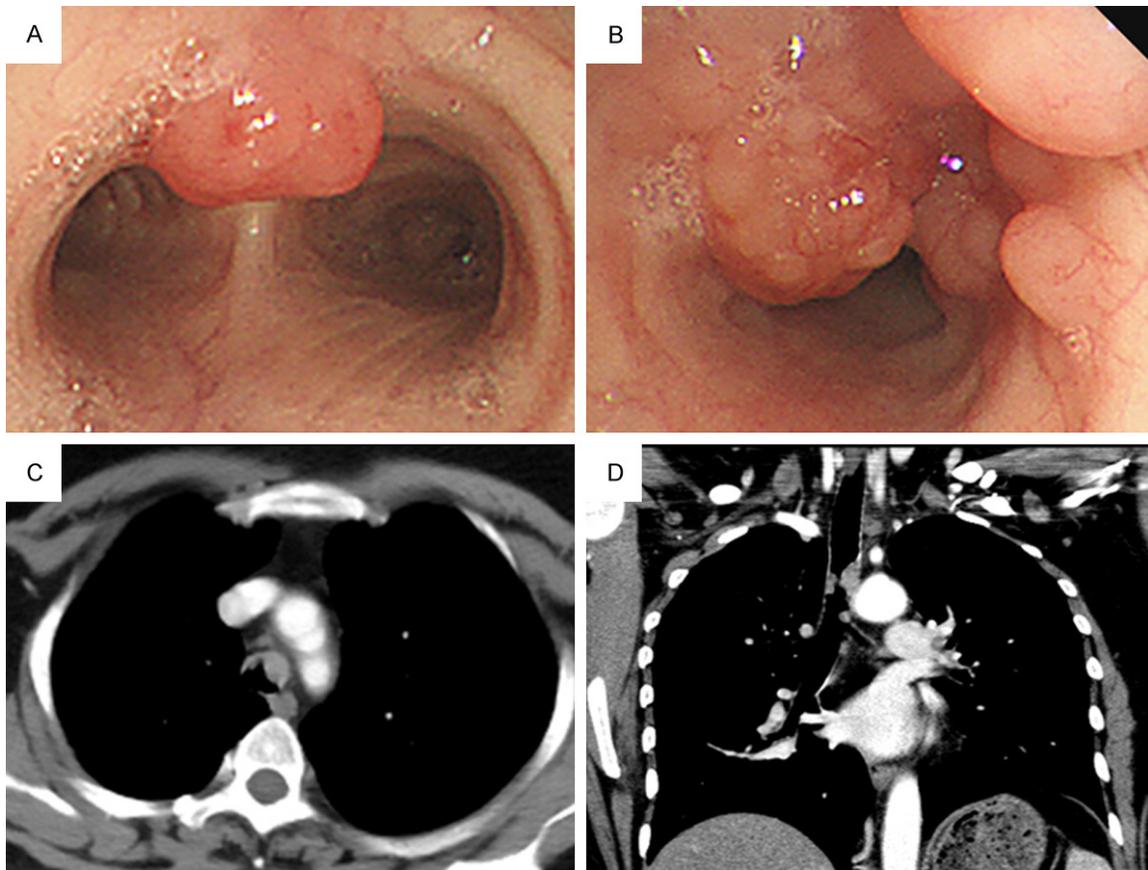


Figure 1. Tracheal tumor before resection. A, B: Bronchoscopy showed multiple nodular masses in the trachea. C, D: Chest CT revealed multiple soft tissue nodules in the lower part of the trachea.

cm, and carina reconstruction through a right posterolateral thoracotomy. Postoperative pathological examination confirmed the diagnosis of MALT lymphoma, without metastasis in the lymph nodes. The tracheal and bronchial stumps were negative.

Six weeks after the surgery, the patient presented with difficulty in breathing and expectoration. Bronchoscopy showed TS of ~0.7 cm diameter, located 2 cm above the new carina, and in addition stenosis of ~0.5 cm at the new left main bronchial opening (**Figure 2A, 2B**). Biopsies were performed to exclude the stenosis caused by tumor recurrence. The patient's symptoms showed no significant improvement after being treated by various methods, including repeated balloon dilatation, covered expandable metallic stent implantation, and radiation with a dose of 15 Gy (3.0 Gy/fraction). Chemotherapy was not used because of the poor general condition of the patient. Six months after the surgery, the patient was readmit-

ted to the hospital due to vomiting and severe cough. Bronchoscopy and chest CT found a TEF, 0.6 cm × 0.7 cm in size, located 1 cm above the new carina (**Figure 2C, 2D**). The patient experienced no significant improvement in symptoms after the following conservative therapeutic strategies: fasting, continuous gastrointestinal decompression, nutritional support, and covered tracheal stent implantation. Therefore, when the patient could breathe spontaneously and her condition warranted an operation [7], reoperation was indicated although there were enormous challenges.

The patient finally underwent a single-stage reoperation via a right thoracotomy, laparotomy and cervical incision. The surgical procedures included TEF repair with two esophageal patches (**Figure 3A-C**), left main BS resection with an end-to-end reconstruction, and esophagogastric bypass in the neck by a posterior mediastinal route. Before the surgery, intraoperative balloon dilatation was applied for the treatment

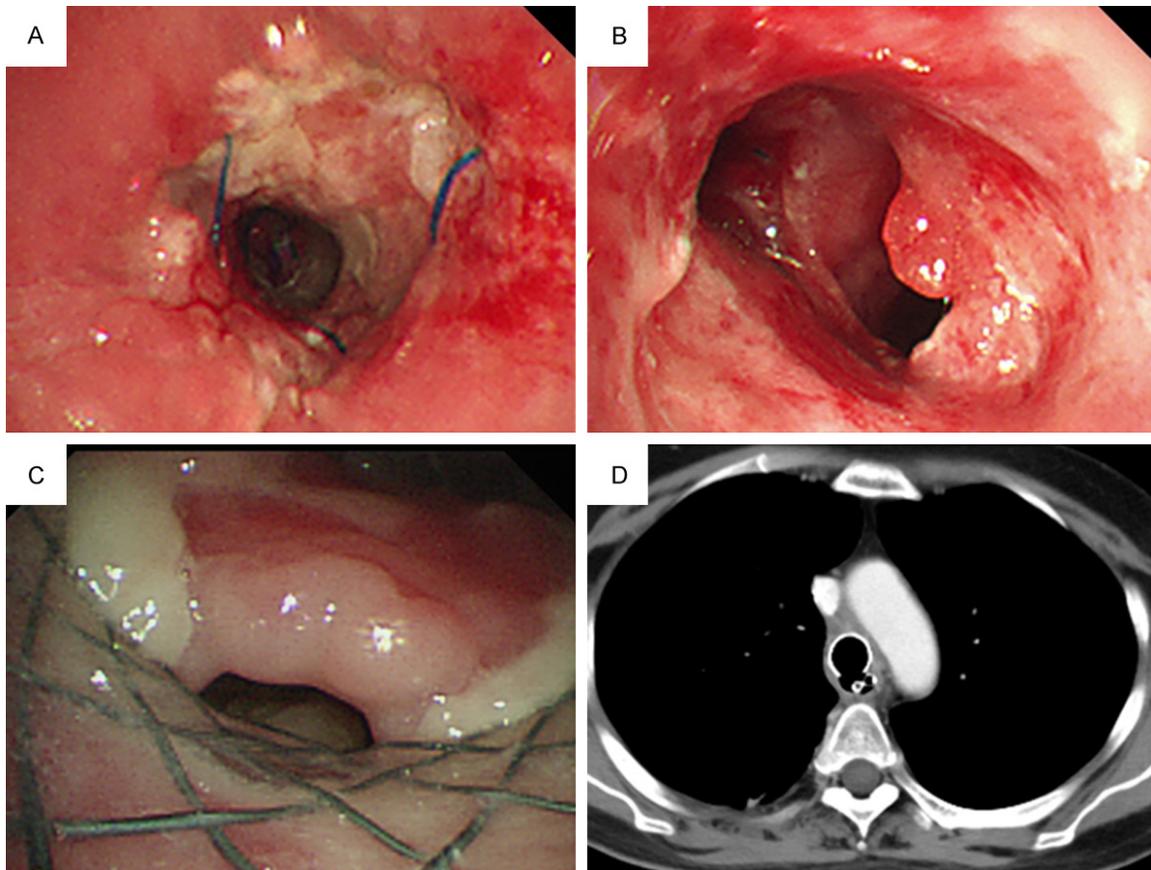


Figure 2. Complications after tracheal resection. (A) Bronchoscopy showed TS with a diameter of ~0.7 cm, (B) left main bronchial opening stenosis with a diameter of ~0.5 cm, and (C) TEF with a size of 0.6 cm × 0.7 cm. (D) Chest CT revealed TEF with tracheal stent and nasogastric tube.

of TS due to difficult intubation. However, during the intraoperative exploration, TS was found inapparent, and the TS scar could not be accurately identified. Thus, the originally scheduled plan of TS resection and reconstruction was canceled. The postoperative course of the patient was eventful. Bronchoscopy indicated that the left vocal cord was fixed and the TEF repair portion narrowed with breath movement (**Figure 3D**). Fortunately, these conditions gradually improved after placement of a self-expanding tracheal stent. The patient currently lives a normal life and has shown no sign of tumor recurrence.

Discussion

Primary MALT lymphoma of the trachea is extremely rare, and the major therapeutic strategies maybe chemotherapy and radiotherapy. If we successfully obtained the histopathologic diagnosis of the tracheal lesions, the original

surgery and the following complications might be avoided. This was the biggest lesson we have learned from this case. In addition, we recommended an effective and safe method-placement of a tracheal stent through bronchoscopy first and then performing a biopsy, to obtain the histopathologic diagnosis for this case through literature review [3].

TS or BS concurrent with TEF is clinically rare, mainly resulting from prolonged artificial pulmonary ventilation [5, 6]. In this case, the most possible cause of TEF was that the time of tracheal stent placement was too long, especially, together with radiation. Therefore, the stent should have been removed earlier and this is one lesson to be learned. Reoperation has been particularly difficult and risky for patients with TS, BS, and TEF, especially for those who had already undergone tracheal resection and carina reconstruction. To the best of our knowledge, surgical treatment for the combination of

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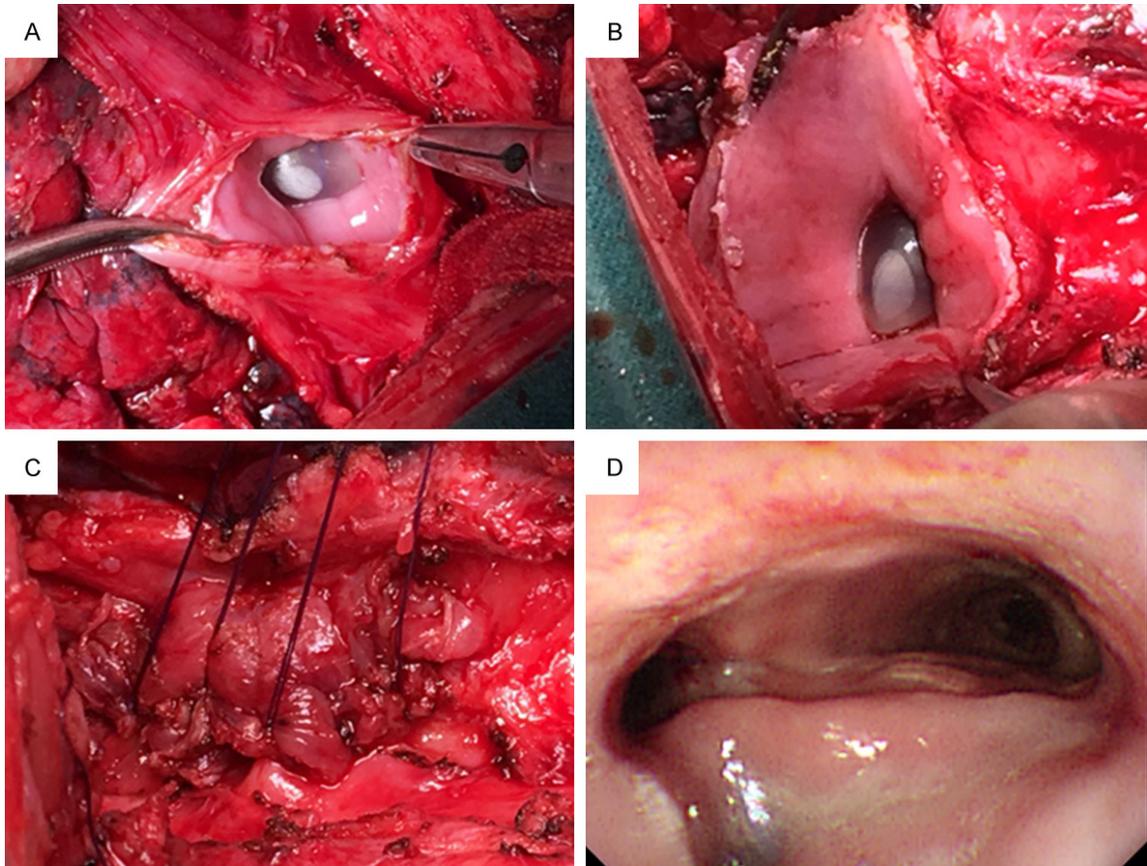


Figure 3. Surgical procedures for repairing TEF. A: The esophagus was opened to expose the TEF defect; B: The esophagus was transected over and under the fistula site to make two esophageal patches; C: The TEF defect was closed with two esophageal patches; D: Bronchoscopy showed that the TEF repair location was relatively narrow.

these 3 complications has not been reported previously.

Common surgical approaches for TS with TEF have been direct closure of the esophageal defect by two layers using interrupted sutures with Vicryl, the segmental resection of the TS, and then end-to-end anastomosis [5, 6]. However, this surgical approach was not suitable for our case, due to the insufficient residual tracheal length to anastomose. In addition, our department had little experience in closing the tracheal defect with a muscle flap. Therefore, we used the esophageal double patch technique [8, 9], by which the TEF was repaired by two esophageal patches, the esophagus was excluded and an esophagogastric bypass was performed. This technique is especially suitable for repairing large, complicated, or recurrent TEFs. The biggest advantage of this technique is that tracheal resection and reconstruction is not required. Therefore, the complexity of the operation and the risk of postoperative

complications are greatly decreased. The disadvantage of this technique is that the esophagus of the patient is damaged. However, it may be worthwhile for the patient in the long term.

The postoperative course of this patient was eventful, because of the tracheal restenosis at the place of repair and paresis of the recurrent laryngeal nerve, which led to dyspnea and weak expectoration. Lessons we learned from this case were that over-relaxation of the esophageal double patch during repair of the TEF and injury of the recurrent laryngeal nerve should be avoided, to prevent postoperative tracheomalacia and glottis insufficiency that may affect breathing and expectoration. In addition, a self-expanding tracheal stent should be considered to prevent secondary tracheomalacia, especially when a large TEF is repaired using esophageal patches [8].

In conclusion, it is crucial to consider MALT lymphoma in the differential diagnosis of tracheal

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tumors, and more importantly, to obtain a histopathologic diagnosis before treatment. The esophageal double patch technique can be a choice in the treatment of complicated TEF.

Disclosure of conflict of interest

None.

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