The right apical tracheal bronchus with bronchial leiomyoma
A case report
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Abstract
Rationale: Tracheal bronchus is an aberrant bronchus that arises directly from the carina of the trachea. It was rare, with only 0.1% to 2% on the right and 0.3% to 1% on the left side of the lateral wall. Tracheal bronchus is usually asymptomatic and can be accidentally detected during bronchoscopy or computed tomography (CT).
Patient concerns: A 62-year-old woman presented with recurrent cough and sputum for more than 1 month.
Diagnosis: Bronchoscopy and three-dimensional-CT confirmed that the bronchial leiomyoma blocked the opening of the right apical segmental tracheal bronchus.
Interventions: After 2 bronchoscopy treatments, the patient still complained of. cough and sputum. Finally, the patient underwent thoracotomy for a right apical segmentectomy and tracheoplasty.
Outcomes: The patient was safely discharged and no longer experienced cough. or sputum.
Lessons: Surgery can reliably remove bronchial leiomyomas. When crossing. the tracheal bronchus, it is important to be cautious about the pulmonary. In such situations, bronchoscopy and three-dimensional-CT are beneficial for surgery.
Abbreviations: 3D = 3-dimensional, B1 = right apical segmental bronchus, CT = computed tomography, TB = tracheal bronchus.
Keywords: bronchial leiomyoma, segmentectomy, tracheal bronchus, tracheoplasty

1. Introduction
Tracheal bronchus (TB) is a rare congenital abnormality that arises directly from the tracheal carina. Its presence has been reported as 0.1% to 2% on the right and 0.3% to 1% on the left side of the lateral wall.[1,2] TB is usually benign, asymptomatic, and does not require treatment. It can be accidentally detected during bronchoscopy or computed tomography (CT). TB may cause localized stenosis, resulting in recurrent local wheezing and infection. However, similar symptoms may not elicit by TB but other reasons. Here, we report a case of right-sided TB ventilating the apical segment, accompanied by bronchial leiomyoma at the opening.

2. Case presentation
A 62-year-old woman (weight, 75 kg; height, 154 cm) was admitted because of recurrent cough and sputum that had lasted more than 1 month. The patient had no history of smoking. Chest CT revealed a mass located in the bronchus of the right upper lobe, which had grown into the endotracheal lumen and led to obstructive pneumonia (Fig. 1). Subsequently, she underwent bronchoscopy, which showed a spherical mass located 2 cm above the carina and at the 3 o'clock direction of the trachea, and the lumen was narrowed by approximately 50% (Fig. 2A). To relieve the intolerable cough, she underwent partial mass removal using an electrocautery snare through
bronchoscopy (Fig. 2C). Although the opening of the right apical segmental bronchus (B1) was revealed after the snare, the bronchus was still blocked by the remaining mass. The resected lesion was pathologically diagnosed as bronchial leiomyoma (Fig. 3). One week after the first bronchoscopy, the patient underwent CT again, which showed a suspicious filling defect in the B1, accompanied by distal atelectasis (Fig. 4). The patient underwent a second bronchoscopy, which showed that the neoplasm completely blocked the orifice of B1 (Fig. 2D), and the opening of B1 was exposed after the snare and clamping (Fig. 2F). However, the patient still complained of cough and sputum after 2 bronchoscopy treatments. Because it was difficult to remove the whole mass through bronchoscopy due to space limitations and the wide base of the leiomyoma, she was transferred to our department to undergo surgery.

Pre-operative CT showed that the leiomyoma was not completely resected (Fig. 5). A posterolateral thoracotomy incision was made at the level of 5th rib to expose the upper thoracic cavity. After hilar dissection and arch of the azygos vein division, we confirmed that the airway of the right apical segment originated directly above the trachea, 2 cm from the bronchoscopy (Fig. 2C). Although the opening of the right apical segmental bronchus (B1) was revealed after the snare, the bronchus was still blocked by the remaining mass. The resected lesion was pathologically diagnosed as bronchial leiomyoma (Fig. 3). One week after the first bronchoscopy, the patient underwent CT again, which showed a suspicious filling defect in the B1, accompanied by distal atelectasis (Fig. 4). The patient underwent a second bronchoscopy, which showed that the neoplasm completely blocked the orifice of B1 (Fig. 2D), and the opening of B1 was exposed after the snare and clamping (Fig. 2F). However, the patient still complained of cough and sputum after 2 bronchoscopy treatments. Because it was difficult to remove the whole mass through bronchoscopy due to space limitations and the wide base of the leiomyoma, she was transferred to our department to undergo surgery.

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carina. Segments with variable right apical segmental artery and right apical segmental vein could be cut using a stapling device. The demarcation line was confirmed using the inflated–deflated method. After cutting and exploring the opening of B1, we found that the tumor had grown into the endotracheal lumen (Fig. 6). Part of the lower trachea and B1 were removed, and tracheoplasty was performed with a continuous suture (3-0 polypropylene) (Fig. 7). Fast-frozen pathology revealed a negative surgical margin. The patient was extubated on postoperative day 6 without any complications (Fig. 8). At the same time, she no longer experienced cough or sputum. The patient was discharged from the hospital on postoperative day 8.

3. Discussion

TB, also called Bronchus suis or “pig bronchus,” was first reported by Anatomist Sandifort in 1785. The aberrant bronchus raised mostly within 2 cm above the carina and the maximum distance was up to 6 cm; the diameter ranges from 0.5 to 1.0 cm, and the length ranges from 0.6 to 2.0 cm. TB includes displaced (most common), rudimentary, supernumerary, and anomalous right upper lobe TB. Our patient was classified as having displaced right apical TB. Inada and Kishimoto and Qi et al described a similar displaced TB ventilating the apical and the apical and posterior segmental bronchi. TB can be regularly diagnosed by bronchoscopy or chest CT, but CT cannot detect the variation because of leiomyoma in this case. Bronchoscopy confirmed TB by the presence of an ectopic opening from the tracheal wall. Approximately 25% of patients with TB exhibit symptoms of pneumonia, chronic bronchitis, and bronchiectasis. The main reason for this is the inadequate local drainage of the involved bronchus. Generally, asymptomatic or mildly symptomatic patients do not require treatment, patient and parent education, appropriate vaccination, early and active management of pulmonary infections and pulmonary physiotherapy may help the affected patients. If the patient presents with persistent or recurrent pulmonary infections or severely impaired exercise tolerance and pulmonary function, surgery including resection of the involved bronchus or lobe and tracheoplasty can be avoided. This patient had recurrent cough and sputum due to pneumonia for more than a month. Considering the duration of symptoms, it might not be caused by TB and could be relevant to the endobronchial obstruction of the leiomyoma.

There are few reports on benign tumors in TB, although leiomyomas are rare. Primary bronchial leiomyomas are thought
to develop from the smooth muscle of the bronchi or bronchioles, constituting approximately 2% of benign lung tumors. Bronchial leiomyomas can present as parenchymal or endobronchial lesions with non-specific respiratory symptoms. To the best of our knowledge, only 1 case of leiomyoma of an accessory TB has been reported. Therefore, it is difficult to determine the susceptibility of bronchial leiomyomas to TB because of the rarity of the combination. Bronchoscopy is normally safe and useful for the treatment of tracheobronchial leiomyoma. Several fiberoptic bronchoscopy techniques, such

Figure 6. Airway exploration was performed after incising the opening of B1. B1 = right apical segmental bronchus.

Figure 7. (A) The right apical segmental bronchus and part of the lower trachea were removed. (B) Tracheoplasty was performed with a continuous suture after shaping the incision as diamond.
as neodymium-yttrium aluminum-garnet laser or electrosurgery snare, are effective for pedunculated polyps. However, if leiomyomas are widespread, therapy may result in incomplete resection or recurrence. Park et al[12] reported that surgery can achieve complete resection and satisfactory outcomes in patients with bronchial leiomyoma. Therefore, we decided to perform surgery for the residual tumor.

We finally chose thoracotomy because of the anatomical variation and the location of the leiomyoma. TB is usually associated with anomalous venous drainage,[13] and awareness of the travel of the pulmonary blood vessels and bronchi will promote the success of surgery. We reconstructed 3-dimensional (3D) imaging for better pre-operative planning. In this case, the variable bronchus could not be performed on 3D-CT because of the position of the leiomyoma. We marked the displaced TB based on what we observed during the operation (Fig. 9A). 3D-CT showed that right apical segmental artery and right posterior segmental artery jointly arising from the main pulmonary artery

Figure 8. Postoperative chest radiograph showed full expansion of the lung.

Figure 9. (A) We marked the tracheal bronchus (yellow) on 3-dimensional image after surgery. (B) 3D-CT showed the relationships among the bronchus (green), variable pulmonary vein (blue), and pulmonary artery (red). 3D = 3-dimensional, CT = computed tomography.
and part of the right superior pulmonary vein was traveling behind the right main pulmonary artery (Fig. 9B). In addition, some studies have indicated that there is a risk of recurrence due to incomplete resection of tracheal or bronchial leiomyomas.\[12,14\] It is necessary to explore the nearby airways to achieve R0 resection when the pedicle of the leiomyoma is broad.

4. Conclusion

Surgery can be used to reliably remove bronchial leiomyomas. However, when accompanied by a TB, it is important to be cautious about pulmonary vessel variations. Bronchoscopy and pre-operative 3D reconstruction planning may contribute to surgical success.

Author contributions

Conceptualization: Yu Zhang.
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Investigation: Haitao Ma.
Methodology: Yu Zhang.
Project administration: Haitao Ma.
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Software: Yu Zhang.
Supervision: Haitao Ma.
Validation: Yu Zhang.
Visualization: Yu Zhang.
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Writing – review & editing: Haitao Ma.

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