



## Case Report

# Upper Lobectomy for Lung Cancer with True Tracheal Bronchus: A Unique Presentation

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### ABSTRACT

Tracheal bronchus is an aberrant bronchus usually originating from the right lateral wall of the trachea, with an incidence ranging from 0.1% to 5% and usually within 2.0cm above the carina. The incidence of lung cancer with bronchial anomaly is very rare. Only nine cases of lung cancer developing from the tracheal bronchus have been reported in literature. Histological examination showed squamous cell carcinoma in only three of them, and we present a fourth case, in a 57-year-old man. Interestingly, our patient's anomaly included both an absence of the right upper bronchus and the fact that the right upper lobe was ventilated by the true tracheal bronchus. This is the first documented case in the world of a squamous carcinoma originating in the true tracheal bronchus. Post-surgical histological stage was T2aNOMO (stage IB). The patient is in a good condition 48 months after the operation and has no evidence of recurrence.

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## Lobectomía superior por cáncer de pulmón con bronquio traqueal verdadero: una presentación inusual

### RESUMEN

El bronquio traqueal es un bronquio aberrante que se origina generalmente en la pared lateral derecha de la tráquea, a menos de 2 cm por encima de la carina, y su incidencia oscila entre el 0,1 y el 5%. La incidencia de cáncer de pulmón con anomalías bronquiales es poco común. En la bibliografía especializada sólo se recogen 9 casos de este tipo de cáncer desarrollado en un bronquio traqueal. El examen anatomopatológico mostraba carcinoma epidermoide en sólo 3 de ellos, y aquí presentamos un cuarto caso que corresponde a un varón de 57 años de edad. Curiosamente, la anomalía de nuestro paciente incluía la ausencia de un bronquio superior derecho normal, además de que el lóbulo superior derecho ventilaba a través del bronquio traqueal. Éste es el primer caso documentado en todo el mundo de carcinoma escamoso con origen en el bronquio traqueal. El estadio anatomopatológico posquirúrgico fue T2aNOMO (estadio IB). El paciente se encuentra en buen estado de salud 48 meses después de la operación y no presenta indicios de recurrencia.

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## Introduction

Anatomically, the normal right upper lung lobe bronchus divides into three segments, forming three segmental bronchi corresponding to the anterior, posterior and apical segments.<sup>1-3</sup> It was Sandifort

who, in 1785, described the tracheal bronchus as a bronchus of the right upper lobe which originated in the trachea.<sup>4-7</sup> The tracheal bronchus is classified as "displaced" or "supernumerary" and, if it acts as a blind sac, it is defined as a "tracheal diverticulum."<sup>1,8-10</sup> It can be located at any point between the cricoid and the carina cartilage, but generally it is found about 2cm above the latter.<sup>6,7,11</sup> It ranges from 0.5 to 1cm in diameter and it is from 0.6 to 2cm long.<sup>5,12</sup> The tracheal bronchus is a rare deformity, the incidence of which fluctuates from 0.1 to 5%.<sup>1,3,8,12-14</sup> It is common in sheep, pigs and cattle, camels, goats and giraffes, but it is only accidentally found in humans.<sup>2,11,14</sup> The

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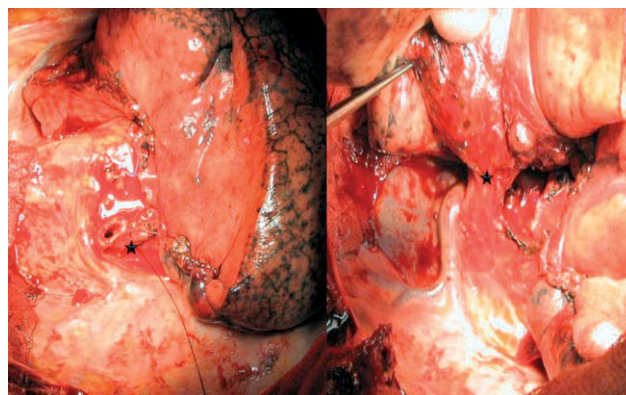
trachea begins to divide during the fourth week of life in the womb and the major ramification of the tracheal tree is formed by about the 16th week of gestation. Consequently, any tracheobronchial malformation occurs in these early stages of the life of the embryo.<sup>1</sup> Abnormal ramification of the tracheobronchial tree is the result of a morphogenic defect which is produced before the end of the second month of gestation.<sup>12</sup> The tracheal bronchus may be linked with other congenital abnormalities; for example, abnormalities of the ribs, pectus excavatum, oesophageal atresia, tracheoesophageal fistula, tracheal stenosis, cystic adenoid malformation, abnormalities of the lower vertebrae, Down's syndrome and Klippel Feil syndrome.<sup>5,9,13,15</sup> Lung cancer is rarely associated with bronchial abnormalities.<sup>2,6,7,9,12,14,16</sup> There are only 9 cases of lung cancer which originated in a tracheal bronchus in the literature specialised in this field.<sup>2,3,12,14,17,18</sup> The anatomopathological analysis revealed squamous cell carcinoma in only 3 of them and here we present a fourth case, in a 57-year-old male. Interestingly, our patient's anomaly included both an absence of the right upper bronchus and the fact that the right upper lobe was ventilated by the true tracheal bronchus. This paper describes the first documented case in the world of squamous cell carcinoma originating in a tracheal bronchus.

### Clinical Observation

A 57-year-old male was referred to our hospital for assessment with symptoms of haemoptysis, a cough and purulent sputum. A simple chest X-ray showed consolidation in the region of the right upper lung. Computed tomography of the thorax revealed a 40x44mm cavitated mass in the right upper lobe, close to the right border of the trachea, as well as a tracheal bronchus with a displaced opening. There were no signs of mediastinal lymphadenopathy in the CT image (fig. 1).

The physical examination revealed normal vital signs. The patient showed a blood oxygen saturation level of 99.3% when breathing normal air, an oxygen blood pressure of 90.6mmHg, a carbon dioxide blood pressure of 36.9mmHg and a pH of 7.37. The respiratory sounds were normal and no wheezing, crackling, rales or rhoncus were detected. Neither peripheral lymph nodes nor acropachy were observed. The pulmonary function tests showed a slightly restricted capacity (forced vital capacity: 4.02l; forced expiratory volume in one second: 2.68 l; correlation between both parameters: 66.6%) and moderate chronic obstructive pulmonary disease.

Rigid and optic fibre bronchoscopy revealed a tracheal bronchus originating approximately 2cm above the carina. No tumoral growth was observed in the lumen of the tracheal bronchus. A biopsy of the



**Figure 2.** Photo of the resected upper lobe and the true tracheal bronchus taken during surgery.

tumour and brush cytology were performed and the presence of a squamous cell carcinoma was confirmed. A systematic examination (in other words, a bone scan, brain magnetic resonance and an abdominal ultrasound scan) failed to reveal any indication of metastasis. The tumour was classified and its clinical stage was T2aN0M0 (stage IB). The tumour was completely removed by right upper lobectomy and the tracheal bronchus was resected at its origin from the right lower tracheal wall (fig. 2). Continuous 2/0 propylene suture was used to close the bronchus. A radical mediastinal and hilar lymph node dissection was performed. The final anatomopathological examination showed a moderately differentiated squamous cell carcinoma. The resection margins showed no tumour cells (R0 resection) and the tumour had not penetrated the visceral pleura. All the lymph nodes which were examined showed evidence of reactive hyperplasia but no metastatic tumour cells, so the anatomopathological classification was pT2aN0M0 (IB stage). There were no problems during the post-operative period. The patient, who continues to be in good condition 48 months after surgery, was discharged six days after his operation.

### Discussion

We report a rare case of tracheal bronchus with an squamous cell carcinoma, which was treated successfully. A true tracheal bronchus arises from the right wall of the trachea, about 2cm from the carina, but it can be located anywhere between the cricoid and the carina cartilage.<sup>1,3,7,8,17</sup> Occasionally it may be a supernumerary bronchus which supplies a segment of pulmonary tissue known as the tracheal lobe.<sup>9</sup> If the entire right upper lobe arises from the trachea, the main right bronchus becomes the intermediate bronchus, which supplies the middle and inferior right lobes. This deformity is known as "true tracheal bronchus" or "pig bronchus" because this morphology is normal in pigs.<sup>4,8,9,16</sup> A displaced bronchus constitutes an abnormal positioning of the right upper lobe bronchus or any of its segments, typically the apical segment.<sup>7,9</sup> Sometimes the tracheal bronchus culminates in a blind sac, which is called a tracheal diverticulum.<sup>1,8,9</sup>

Tracheal bronchi generally cause no symptoms.<sup>8,11,12,14</sup> Usually they are found by chance and are of no clinical importance; however, they may be associated with localised pulmonary problems, including chronic atelectasia, recurrent pneumonia, chronic bronchiectasis, a persistent cough, rales, haemoptysis and cysts or localized emphysema.<sup>7,13,14,16</sup> Indeed, in paediatric patients the tracheal bronchus has been linked to recurrent infections, rales, breathing difficulties and thoracic masses.<sup>8,10,16</sup> The presence of a tracheal bronchus can result in accidental intubation of the tracheal bronchus, which could cause pneumothorax and inadequate ventilation.<sup>5,8,9,16</sup>

**Figure 1.** Computed tomography image of a tracheal bronchus. The tracheal bronchus originates approximately 2cm above the main carina and the cavitary mass in the upper lobe.

The tracheal bronchus can become occluded on the endotracheal tube side, which can lead to lobe collapse.<sup>7,10</sup>

The diagnosis of tracheal bronchus can be made by rigid and/or flexible bronchoscopy.<sup>6,8,13</sup> The variant found in our patient was classified as a true tracheal bronchus; in other words, the bronchus divided to form the three regular branches of the right upper lobe bronchus and there was no true right upper lobe bronchus arising from the main right bronchus.<sup>3,8</sup> It is rare that in our patient a normal right upper bronchus was lacking and the true tracheal bronchus which ventilated the right upper lobe was positioned about 2cm above the carina. Rarely is the tracheal bronchus a true supernumerary bronchus, which supplies air to a segment of pulmonary tissue known as the tracheal lobe and which generally has its own blood supply from the pulmonary artery, as in our case.<sup>9</sup>

Lung cancer associated with the tracheal bronchus is very uncommon and there are only 9 cases of surgical resection in the literature specialised in this field.<sup>2,3,12,14,17,18</sup> Histological examination showed squamous cell carcinoma in only three of them, and we present a fourth case. We performed a lobectomy of the right upper lobe and radical lymph node dissection. In the 3 reported cases right upper lobectomy was the therapeutic choice.<sup>2,3,17</sup> Our patient is healthy 48 months after the operation and has no signs of recurrence.

To conclude, certain specialists, such as pneumologists, thoracic surgeons and anaesthesiologists need to know about bronchial variants and abnormalities. Although there is a lack of evidence to establish whether the tracheal bronchus is more susceptible to malignant neoplasias, it is advisable to perform a detailed bronchoscopy and computed tomography before surgery in order to exclude the possibility of neoplasia.

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