

## CASE REPORT

## Endobronchial enchondroma: Unusual bronchial tumor

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**Abstract**

Bronchial chondromas are very rare pulmonary benign tumors. Despite their rarity, clinicians should be aware of the possibility of endobronchial tumors in patients with unexplained respiratory symptoms. Treatment modalities for the complete excision of the tumor should be initiated to prevent further complications based on the individual tumor situation.

**KEYWORDS**

benign tumor, bronchoscopy, bronchus, chondroma, lung neoplasms

**1 | INTRODUCTION**

Despite most endobronchial tumors obstructing bronchi presenting as bronchogenic carcinoma, benign bronchial tumors can additionally cause bronchial obstruction.<sup>1</sup> Benign bronchial tumors are usually asymptomatic and are found incidentally. However, if the tumors cause bronchial narrowing or obstruction, some patients might suffer from respiratory symptoms as a result of further complications of the bronchi and lungs.<sup>2,3</sup> Here, we present a case of bronchial enchondroma, a sporadic benign bronchial tumor incidentally found by a health screening in a patient suffering from a chronic cough, which was improved after tumor excision.

**2 | CASE HISTORY**

A 61-year-old man was evaluated in the pulmonary clinic due to a right hilar mass discovered on a chest X-ray from a national health screening program in South Korea. He was a 30 pack-year current smoker. He had a mild, dry cough with scanty sputum persisting for more than six months at his visit, but there was no history of fever, hemoptysis, dyspnea, chest pain, weight loss, night sweats, or any other systemic symptoms. He was an office worker, and his past and family histories were unremarkable.

Initial vital signs were stable: Blood pressure, 138/86 mmHg, heart rate, 72/min, respiration rate, 16/min, and temperature, 36.5°C. He displayed a slightly

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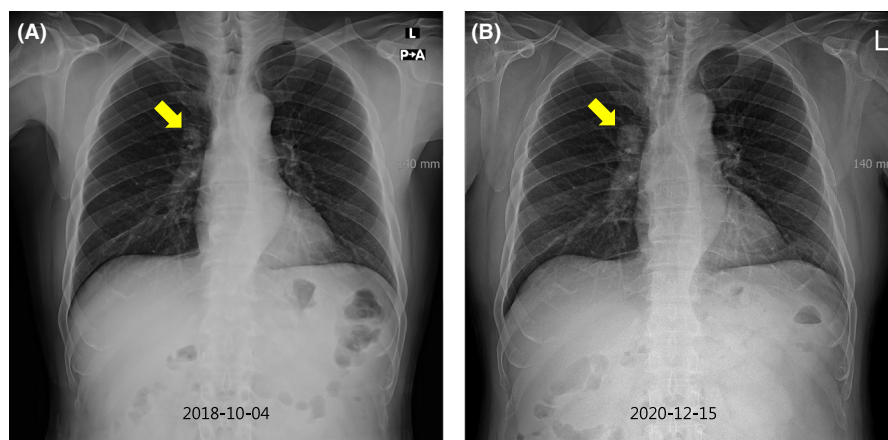
reduced breath sound intensity on the right upper posterior chest on chest auscultation, and other systemic physical examinations were normal. A simple chest X-ray revealed a well-marginated round mass in the right hilum, which had increased in size when compared with the previous chest X-ray taken two years prior (Figure 1). Additionally, a contrast-enhanced chest computed tomography was performed and revealed a 3.5 cm high-density mass obstructing the right upper posterior segmental bronchus (Figure 2). The bronchoscopic examination uncovered a pedunculated red mass completely obstructing the right upper posterior segmental bronchus. A pinkish, hyaline-like core material with a rigid surface was observed after peeling off the surface mucosa during the biopsy procedure (Figure 3).

With suspicion of the presence of a tumor within the cartilage component (including hamartoma and chondroma), video-assisted thoracoscopic surgery (VATS) was subsequently undergone. The tumor was removed in total and measured  $4.0 \times 2.5$  cm after excision (Figure 4). The histopathology of the excised tumor revealed a bronchial enchondroma composed of lobules of mature hyaline cartilage separated by normal cancellous bone or marrow

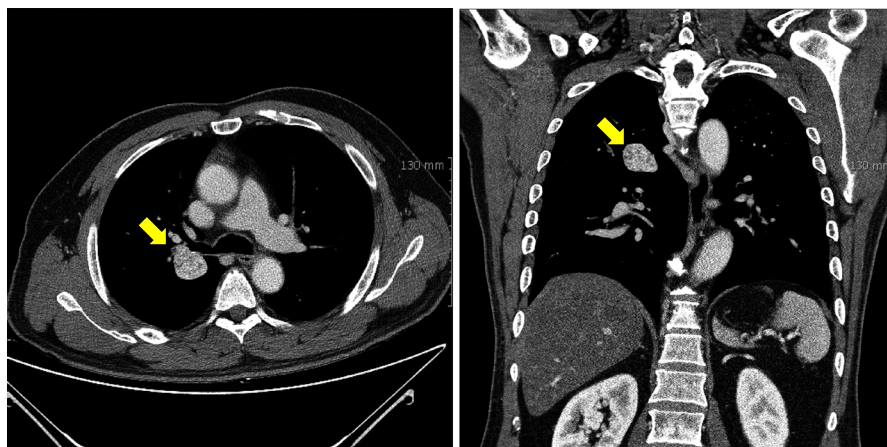
without adipose tissue and the smooth muscle component suggesting hamartoma (Figure 5). In consideration of the possibility of the Carney triad, no specific findings were observed during gastroscopy, and there were no signs and symptoms suggesting paraganglioma through repeated examinations. The patient recovered from surgery, and the dry cough had resolved on his visit to the pulmonary clinic one month post-tumor removal.

### 3 | DISCUSSION

Benign tracheobronchial tumors are sporadic tumors representing only 0.2% of all pulmonary tumors in the United States.<sup>4</sup> The two most common types of benign pulmonary tumors are hamartomas and papillomas. Leiomyomas, lipomas, chondromas, and neurogenic tumors can also present as less frequent benign neoplasms.<sup>1</sup> Most benign pulmonary tumors are slow-growing, asymptomatic, and often remain undiagnosed prior to causing bronchial obstruction or mass effects on the surrounding organs. The diagnosis of these tumors is often challenging as the signs and symptoms are non-specific and a simple chest

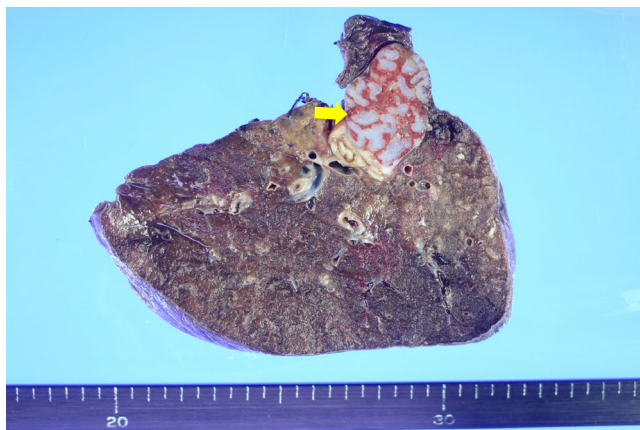
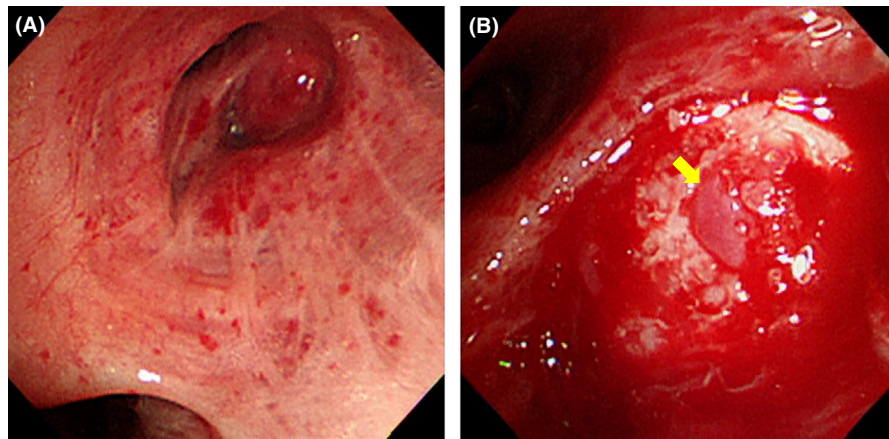


**FIGURE 1** A, Right hilar mass (arrow) partially obscured by the right pulmonary trunk is observed. B, Compared with the chest X-ray taken two years before, the right hilar mass increases in size, and the shape is more clearly exposed



**FIGURE 2** Contrast-enhanced chest computed tomography shows about a 3.5 cm sized, well-defined, high-density mass (arrow) in the right upper posterior segment, obstructing the posterior segmental bronchus

**FIGURE 3** A, Round mass with a red-colored, smooth surface completely obstructing the right upper posterior segmental bronchus is observed by the bronchoscopic examination. B, Pinkish, hyaline-like core (arrow) is exposed by the biopsy procedure



**FIGURE 4** Large, twisted bronchus is nearly obstructed by a bronchial enchondroma (arrow) composed of lobules of mature hyaline cartilage separated by normal cancellous bone or marrow

X-ray fails to present remarkable findings. Thus, they are often misdiagnosed as more common causes of chronic cough or dyspnea, such as chronic obstructive bronchitis, asthma, and bronchogenic carcinomas.<sup>2,3,5</sup> As most of the symptoms of bronchial tumors are primarily caused by their growth and complications to adjacent structures, early detection and treatment are clinically significant.

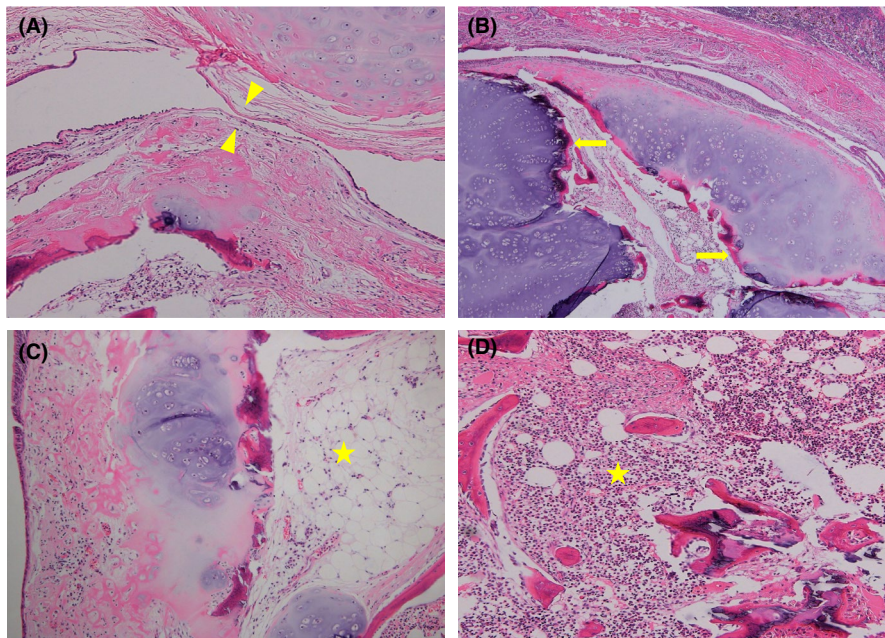
*Bronchial chondromas* are very rare benign tumors arising from bronchial cartilage.<sup>1</sup> As they are often misclassified as hamartomas, the most common type of endobronchial neoplasm, the true incidence of bronchial chondroma is still unknown.<sup>1</sup> In contrast to hamartomas, endobronchial chondromas have a distinct pathology consisting of calcified/ossified cartilaginous components without the adipose tissue, smooth muscle, epithelial, and stromal elements characteristic of hamartomas.<sup>6,7</sup> Well-differentiated chondrosarcomas can resemble chondromas.<sup>4,8</sup> Additionally, chondrosarcomas can be differentiated by the presence of tissue invasion or mitosis<sup>9</sup> and clinicians should evaluate the possibility of lung metastasis from other skeletal systems considering their high incidence.

*Bronchial chondromas* are occasionally present as a component of the Carney triad, characterized by the simultaneous presence of gastric epithelioid leiomyoblastoma, extra-adrenal paragangliomas, and pulmonary chondroma.<sup>10</sup> In the current case, after the patient's tumor was defined as an endobronchial enchondroma, in consideration of the possibility for the presence of the Carney triad, a gastroscopic examination was performed to evaluate the presence of gastric leiomyoblastoma. We additionally completed a history-taking and physical examination once more to assess sign and symptoms representative of paragangliomas. However, there was no evidence for the presence of the Carney triad. The tumor was eventually diagnosed as an isolated endobronchial enchondroma, which is extremely rare among pulmonary chondromas.

Although there is currently no consensus for the best treatment approach for bronchial chondroma, most treatments are performed through complete excision with bronchoscopy or through surgery, including VATS or open thoracotomy. Therapy decisions should be individually considered for each case based on the tumor's operability, size, type, and location.<sup>10,11</sup> Even if only a small number of cases were reported, no patients died or had severe complications, regardless of the treatment options, and recurrence after treatment was very low. One report revealed that only one patient presented recurrence among 23 treated cases between 1945 and 1980.<sup>9</sup> Furthermore, an additional report recounted a recurrent tracheal chondroma with the sarcomatous transformation.<sup>12,13</sup> Considering the low risk of treatment morbidity, mortality, recurrence, potential risk of malignant transformation, and structural destruction of adjacent organs, treatment options for complete excision should be considered individually based on the tumor situation.

In the current case, the patient visited the pulmonary clinic for the first time and had had a chronic cough for more than six months. It was initially considered to be due to his cigarette smoking habit. However, even if most benign bronchial tumors are asymptomatic, some patients





**FIGURE 5** A, Bronchial enchondroma grows along the bronchial walls, showing diffuse luminal narrowing, and multifocally near obstruction (arrowheads). B, Mature cartilage lobules of this bronchial enchondroma are peripherally encased by bone with endochondral ossification (arrows). C and D, Both components of the enchondroma are normal, cancellous bone with fatty (C, star) hypercellular (D, star) marrows and mature hyaline cartilage lobules, with no other hamartomatous adipose tissue or smooth muscle components

may have symptoms such as intractable cough or hemoptysis, suggesting chronic airway diseases.<sup>2,3,14</sup> After excising the tumor entirely by the lobectomy procedure, his cough disappeared one month after the operation. This case emphasizes the importance of early recognition and treatment of benign bronchial tumors by clinicians.

## 4 | CONCLUSION

Bronchial chondromas are extremely rare pulmonary benign tumors. Early recognition and diagnosis are challenging as most benign bronchial tumors are asymptomatic prior to growing large enough to obstruct the airways or cause complications to adjacent structures. Despite their rarity, clinicians should pay close attention to patients suffering from an unexplained chronic cough, hemoptysis, dyspnea, and recurrent infection as there is the possibility of endobronchial neoplasm. Prompt and proper treatment options for the complete excision of the tumor should be initiated in order to prevent complications and malignant transformation based on the individual tumor situation.

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## CONFLICT OF INTEREST

None declared.

## AUTHOR CONTRIBUTIONS

KR managed the patient and prepared the manuscript. NM examined the pathology and prepared the manuscript.

DK managed the patient and prepared the manuscript collectively.

## ETHICAL APPROVAL

Hereby, I, Dohhyung Kim, consciously assure that for the manuscript, Endobronchial Enchondroma: Unusual Bronchial Tumor, the following is fulfilled: (1) This material is the authors' own original work, which has not been previously published elsewhere. (2) The paper is not currently being considered for publication elsewhere. (3) The paper reflects the authors' own research in a truthful and complete manner. (4) The paper properly credits the meaningful contributions of co-authors and co-researchers. (5) The results are appropriately placed in the context of prior and existing research. (6) All sources used are properly disclosed (correct citation). Literally copying of text must be indicated as such by using quotation marks and giving proper reference. (7) All authors have been personally and actively involved in substantial work leading to the paper and will take public responsibility for its content. I agree with the above statements and declare that this submission follows the policies of Clinical Case Reports as outlined in the Guide for Authors and in the Ethics Statement.

## CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

## DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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