Treatment of benign endobronchial tumors: when, how, and why. Insights, experiences, and interventional pulmonology strategies

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Abstract

Benign endobronchial tumors are rare clinical entities characterized by considerable variability in etiology and clinical presentation. The authors report four cases of endobronchial hamartomas treated and followed up from 2018 to 2023. Three of these cases, with identical endobronchial localization in the right lower lobe, were radically treated in flexible bronchoscopy with only the use of biopsy forceps. Another case with a different localization in the left main bronchus was treated with a laser through rigid bronchoscopy. In addition, the authors outline the main interventional pulmonological strategies for the treatment of benign tumors with endobronchial growth based on the existing literature.

Introduction

Benign endobronchial tumors are rare clinical entities characterized by considerable variability in etiology and clinical presentation [1]. They are usually slow-growing and often present with symptoms of bronchial obstruction and compression of local structures, resulting in coughing, wheezing, or chest discomfort [2]. Several benign lung tumors of endobronchial origin have been described in the literature, including hamartomas, lipomas, squamous papillomas, pleomorphic adenomas, papillary adenomas, hemangiomas, neurofibromas, leiomyomas, and papillomatosis.

Among benign lung tumors, pulmonary hamartoma is the most common, with an incidence of 0.025% to 0.32% [3]. It can be asymptomatic in the initial pre-occlusive phase; therefore, more frequently accidentally discovered during a computed tomography (CT) scan [4]. When present, main respiratory and non-respiratory symptoms include fever, cough, hemoptysis, purulent sputum, dyspnea, and chest pain. Recurrent pneumonia secondary to bronchial obstruction can occur [5,6]. CT generally reveals an endobronchial lesion with or without signs of obstructive pneumonia or downstream atelectasis [7]. On bronchoscopic examination, an endobronchial hamartoma appears as a polypoid or pedunculated formation, without signs of submucosal infiltration. It can be well circumscribed, with a smooth and sallow surface. Histology usually detects the coexistence of different kinds of tissue, such as connec-

tive, epithelial, bone, muscular, adipose, and especially cartilaginous tissue [8]. Differential diagnosis includes other benign neoplasms, carcinoids, endobronchial involvement in non-infective systemic granulomatosis (*i.e.*, sarcoidosis, vasculitis), infective granulomatosis (*i.e.*, tuberculosis), endobronchial metastases, and non-small lung cancer [9]. Malignant degeneration of endobronchial hamartomas has been reported, though infrequently [10]. Numerous endoscopic techniques have been described for the treatment of benign endobronchial tumors, using flexible or rigid bronchoscopy: Argon plasma coagulation (APC), Nd-YAG laser therapy, cryotherapy, electrocauterization, or diathermic loops.

Case Report 1

A 58-year-old former smoker was referred for a small endoluminal hypodensity at the right lower lobar bronchus detected during a follow-up high-resolution thoracic CT scan for an organizing pneumonia (Figure 1A). A fiberoptic bronchoscopy was performed using a flexible videobronchoscopy system with a probe diameter of 6.3 mm and a working channel of 2.8 mm. In the border area between the intermediate bronchus and the inferior lobar bronchus of the right lung, a pedunculated encapsulated neoformation was found (Figure 1B). The implantation site was located in the basal-medial segment of the inferior lobar bronchus, without signs of submucosal infiltration. The formation was removed using biopsy forceps introduced through the bronchoscope working channel. The excised material was reddish, lobulated, elastic cartilaginous mucosa tissue with coated and pseudostratified superficial epithelium. The procedure lasted about 20 minutes with a small amount of bleeding controlled by ice-cold saline solution. Final pathological examination reported a bronchial hamartoma mesenchymal lesion with fragments of cartilaginous tissue, partly with fibroadipose stroma, partly coated with atrophic bronchial epithelium (Figure 1C). Follow-up chest CT scan (Figure 1D) and bronchoscopy (Figure 1E) performed 3, 6, 12, 18, 24, and 36 months after removal of the lesion showed good airway patency without any bronchial obstruction or recurrence.

Case Report 2

A 40-year-old non-smoker man was admitted with a new onset of hemoptysis. Nasopharyngeal swab revealed SARS-CoV-2 infection. He underwent a contrasted CT scan showing an endobronchial polypoid lesion without iodine contrast enhancement in the distal section of the intermediate bronchus (Figure 1F). The lesion was implanted in the lower lobe bronchus and with partial obstruction of the distal canalization. The bronchoscopy was performed with a single-use bronchoscope, with a probe diameter of 5.8 mm and a 2.8 mm working channel. At the border between the intermediate bronchus and the inferior lobar bronchus of the right lung, a circumscribed pedunculated neoformation characterized by a smooth, reddish, poorly vascularized surface with a hard, elastic consistency was observed. No signs of submucosal infiltration were present (Figure 1G). Using biopsy forceps, the lesion was hooked and excised totally, including the implantation site on the mucosa, obtaining macroscopic total removal. The procedure lasted about 15 minutes, and the resulting modest bleeding was stopped mechanically with the disposable bronchoscope. The definitive histological diagnosis was bronchial hamartoma, characterized by a well-circumscribed, uncapsulated nodule of cartilage, respiratory epithelium, and fibrous tissue (Figure 1H). Chest CT (Figure 1I) and bronchoscopy (Figure 1L) performed 3, 6, 18, 24, and 36 months after endoscopic treatment showed no recurrence.

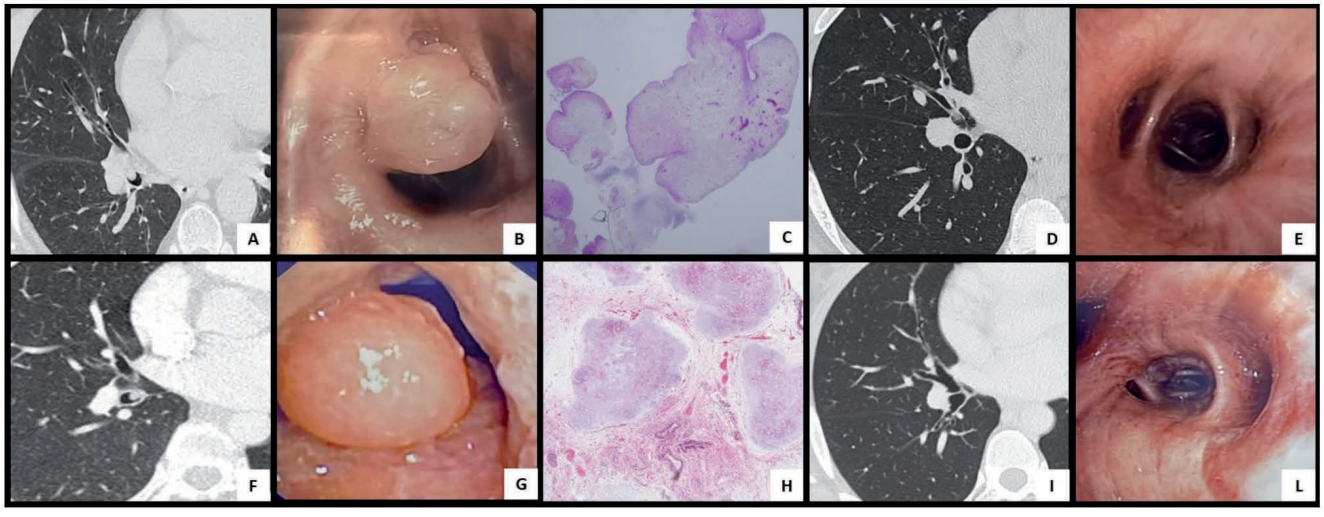

Figure 1. A) Axial scan of high-resolution (HR) chest computed tomography (CT) showing endoluminal hypodensity at the right lower lobar bronchus; B) endoscopic image of a pedunculated encapsulated neoformation in the border area between the intermediate bronchus and the lower lobar bronchus of the right lung; C) histopathological view of bronchial hamartoma with fragments of cartilaginous tissue, partly with fibroadipose stroma, partly coated with atrophic bronchial epithelium; D) axial scan of chest CT; E) endoscopic image 3 months after bronchoscopic treatment showing normal canalization of the intermediate and the right lower bronchus; F) axial scan of HR chest CT an endobronchial polypoid lesion without iodine contrast enhancement in the distal section of the intermediate bronchus; G) endoscopic image of a circumscribed pedunculated neoformation characterized by a smooth, reddish, poorly vascularized surface in the border between the intermediate bronchus and the inferior lobar bronchus of the right lung, a circumscribed pedunculated neoformation characterized by a smooth, reddish, poorly vascularized surface; H) histopathological view of bronchial hamartoma characterized by well-circumscribed, uncapsulated nodule of cartilage, respiratory epithelium and fibrous tissue; I) axial scan of chest CT; L) bronchoscopy performed 18 months after endoscopic treatment showing normal canalization and no recurrence.

Case Report 3

A 63-year-old male former smoker (52 pack years) was referred for a small endoluminal neoformation at the right lower lobar bronchus (Figure 2A) during a follow-up high-resolution thoracic CT scan for a previous colon cancer treated with surgery and adjuvant chemotherapy. A fiberoptic bronchoscopy was performed using a flexible videobronchoscopy system with a probe diameter of 6.3 mm and a working channel of 2.8 mm. In the border area between the intermediate bronchus and the inferior lobar bronchus of the right lung, a pedunculated encapsulated neoformation with a smooth surface was found (Figure 2B). No signs of submucosal infiltration were found. The endobronchial neoformation was removed using biopsy forceps introduced through the bronchoscope working channel. The procedure lasted about 20 minutes with a small amount of bleeding controlled by ice-cold saline solution. The definitive histological diagnosis was hamartochondroma (Figure 2C). Follow-up chest CT scan (Figure 2D) and bronchoscopy (Figure 2E) performed 3, 6, 12, 18, and 36 months after lesion removal showed good airway patency without bronchial obstruction and recurrence.

Case Report 4

A 75-year-old male former smoker (30 pack years) was admitted to our hospital with a new onset of pain in the shoulder and left arm. He complained of weight loss and recurrent pneumonia in recent years. He performed a high-resolution thoracic CT scan showing an endobronchial polypoid lesion in the left main bronchus without signs of ilo-mediastinal involvement (Figure 2F). Videobronchoscopy revealed a voluminous multilobated neoformation, with a hard-elastic consistency, richly vascularized surface, and a large implantation site on the right posterolateral wall of the left main bronchus (Figure 2G). The lesion was excised with rigid bronchoscopy using a 20 W Nd-YAG laser spots and biopsy forceps. Thermal necrosis of the large implantation site was performed with 15 and 10 W laser spots. The histological diagnosis was lipomatous hamartoma characterized by mature adipocytes in a fibrous background (Figure 2H). A CT scan follow-up (Figure 2I) and videobronchoscopy (Figure 2L) with narrow band imaging performed at 30 days and 6, 12, 18, 24, and 36 months after treatment showed no recurrence.

Discussion

We report four cases of endobronchial hamartomas treated and followed up from 2018 to 2023. All patients underwent positron emission tomography-CT scan to rule out malignancy of the treated lesions. Despite the low occurrence of benign endobronchial tumors, early diagnosis and treatment are crucial due to the potential for serious complications. There is no standardized consensus or guidelines for the treatment of benign endobronchial tumors. In the current literature, the substrate for the management of benign endobronchial tumors is typically classified into three main groups: non-randomized experiments, observational studies including case series, and retrospective studies [11]. The therapeutic management plan generally depends on three main domains: location, extent, and implantation site of the tumor. The recommended first-line approach is endoscopic treatment through rigid bronchoscopy, laser photocoagulation, or mechanical resection, due to the benign nature of the tumor and the low rate of recurrence after excision [12]. Traditional surgical treatment of bronchotomy or thoracotomy is currently indicated in cases where endoscopic treatment is not feasible. Irreversible pulmonary fibrotic consequences associated with recurrent infections may also require surgical resection [13]. Bronchoscopic management plays a crucial role in removing obstruction and restoring ven-

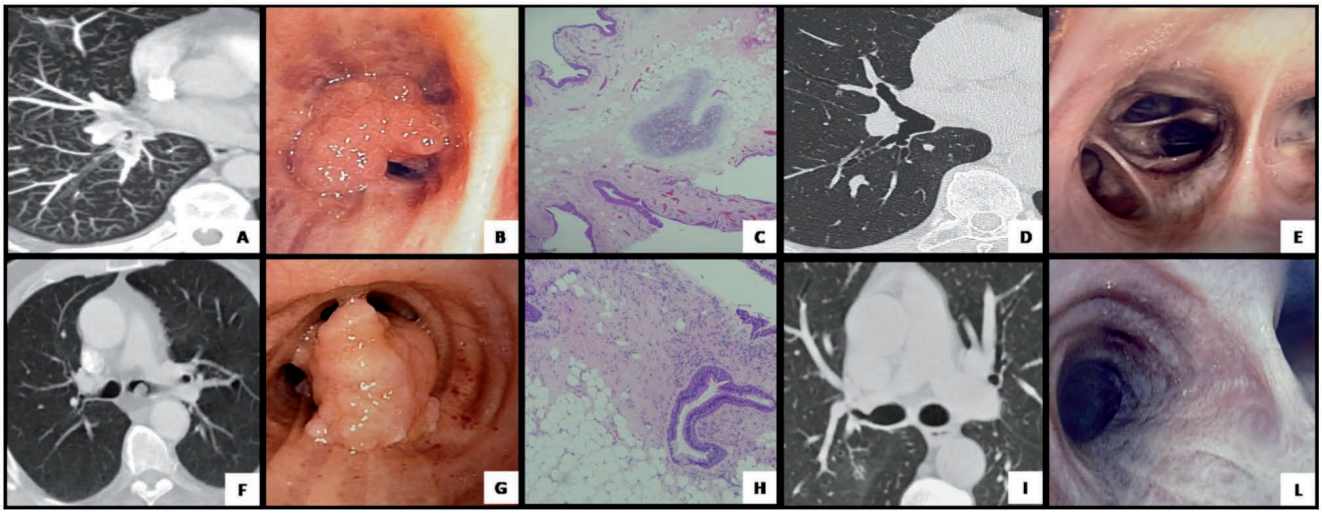

Figure 2. A) Axial scan of high-resolution (HR) chest computed tomography (CT) showing a small endoluminal neoformation at the right lower lobar bronchus; B) endoscopic image showing a pedunculated encapsulated neoformation with a smooth surface at the border area between the intermediate bronchus and the lower lobar bronchus of the right lung; C) histopathological view of hamartochondroma; D) follow-up chest CT scan; E) bronchoscopy performed 6 months after lesion removal showed good airway patency without bronchial obstruction and relapse; F) axial scan of HR chest CT showing an endobronchial polypoid lesion in the left main bronchus without signs of ilo-mediastinal involvement; G) endoscopic image showing a voluminous neoformation multi-lobed with a large implantation site on right posterolateral wall of the left main bronchus; H) histophatological view of a lipomatous hamartoma characterized by mature adipocytes in a fibrous background; I) axial scan of chest CT; L) bronchoscopy performed 30 days after endoscopic treatment showing normal canalization of left main bronchus.

tilation, resulting in improved dyspnea and other obstruction-related symptoms [14]. The endoscopic approach is less invasive with fewer complications and mortality rates, without compromising any future surgical resection in the event of recurrence. Laser treatment with rigid bronchoscopy is considered the gold standard technique. Flexible bronchoscopy with a biopsy forceps supported by Nd-YAG laser or APC is a potential alternative to rigid bronchoscopy. The choice of endoscopic technique is based on several aspects: the extension of the implantation site, the location of the tumor in the bronchial tree, the presence of underlying vascular structures, and specific aspects related to the patient, such as bleeding tendency or neck stiffness [15]. Electrocauterization by flexible bronchoscopy is a possible alternative to rigid bronchoscopy, although it may have potential complications such as profuse bleeding and perforation of the bronchial wall [16]. Cryotherapy has also been reported for the treatment of endobronchial lesions, using flexible or rigid bronchoscopy, though it is mainly used in palliative care for malignant central airway obstruction [17,18]. All these techniques can be used to treat endoluminal tumors that are confined to the subsegmental bronchi, with a small implantation site and without infiltration of the submucosal layer. Tumors with a polypoid structure are easier to treat endoscopically [19]. Endobronchial tumors with a large implantation site usually require surgical resection [11].

In our experience, we have used different endoscopic approaches depending on the characteristics of the endobronchial lesions. The results obtained with the different approaches have been almost identical. In case 4, we used laser treatment through rigid bronchoscopy, considering that the lesion was located in a main bronchus and its implantation site on the posterior wall was large. In the other three patients, the lesion had identical endobronchial localization, the lower lobar bronchus, and a small implantation site; therefore, a less invasive endoscopic approach than rigid bronchoscopy was used. We removed the lesion exclusively with a biopsy forceps using two types of flexible bronchoscope, the traditional multi-use and the disposable one. It was not necessary to use the Nd-YAG laser, APC, cryotherapy, or other methods. In all cases, we obtained the radical removal of the lesion without significant complications during and after the procedure and without disease recurrence at follow-up.

Conclusions

The authors strongly support that benign endobronchial tumors should be referred to centers with good expertise to minimize the risk of complications and maximize the outcome of treatment. Experienced radiologists are essential for accurate and systematic evaluation of the CT scan. Interventional pulmonologists and thoracic surgeons are also essential to safely perform the treatment and manage potential complications such as airway hemorrhage. Although many cases of benign endobronchial tumors treated radically by endoscopic approach are described in the literature, there are currently no randomized trials comparing the different endoscopic techniques with surgery. The authors suggest that in selected cases and in centers with good expertise, the removal of endobronchial benign tumors by flexible bronchoscopy is a valid, safe, less expensive, and less invasive therapeutic approach compared to other endoscopic procedures. The paper's findings indicate that flexible bronchoscopy could be considered as an alternative for removing benign endobronchial tumors in cases where rigid bronchoscopy is not possible and/or available. However, rigid bronchoscopy remains the first choice for both greater safety and better operability.

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