Clinical Features and Management of Endobronchial Hamartoma

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Research article

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Abstract

Background: Endobronchial hamartoma(EH) is a benign endobronchial tumor with low incidence. The objective of this study was to explore the clinical features and management of patients with endobronchial hamartoma, improve the diagnosis and treatment.

Methods: Retrospective study on the clinical data of 13 cases of endobronchial hamartoma confirmed by pathology in The First Affiliated Hospital of Soochow University from July 2000 to October 2019.

Results: The 13 patients, 11 males and 2 females, aged 48-74 years, mean(63 ± 9)years, were diagnosed with EH. Three of the 13 patients with EH are asymptomatic, others had atypical symptoms. CT showed obstructive pneumonia in 10 of the 13 patients. Among the 13 patients, visible endobronchial tumor were present in 7 patients on CT, and only 3 cases were diagnosed with endobronchial tumor by radiologists. All cases received bronchoscopy. Seven patients were treated via flexible bronchoscopy, using electrocautery, argon plasma coagulation(APC) and cryotherapy. Six patients received pulmonary lobectomy via thoracotomy. Follow-up CT scan was performed in 7 of 13 patients(mean 47 months, range 17-69 months), which revealed no recurrence in all 7 patients.

Conclusions: In clinical work, EH sometimes may be neglected in CT images. Bronchoscopy plays an important role in diagnosis. Pathological study is required to confirm the diagnosis. The management of endobronchial hamartoma should be individualized, bronchoscopic treatment is the first choice for EH. But in some instances, surgery should be considered.

Background

Pulmonary hamartoma is the most common lung benign tumor, accounting for about 75 % of lung benign tumor[1]. Most pulmonary hamartomas are parenchymal hamartomas, and endobronchial hamartomas are very rare. In the largest review series published(N=215), only 1.4% of hamartomas were endobronchial, with the remainder located on the parenchyma[2]. EH sometimes may cause airway obstruction, different degrees of obstructive pneumonia and even irreversible lung damage. The treatment of EH involves surgical treatment and bronchoscopic intervention. Here we present a series of patients with EH to raise awareness of this disease.

Methods

Retrospective study on the clinical data of 13 cases of endobronchial hamartoma confirmed by pathology. Research of patients with EH in this study concluded the following contents: the patient's age, sex, clinical symptoms, accompanied diseases, image features include chest radiograph and CT, diagnosis given by radiologists, distributions of EH, bronchoscopic features, treatment involves bronchoscopic intervention and surgery, and follow-up.

Results

301 cases of pulmonary hamartoma confirmed by pathology in The First Affiliated Hospital of Soochow University from July 2000 to October 2019 were retrospectively reviewed, including 13(13/301, 4%) cases of endobronchial hamartoma. The patients included 11 males and 2 females, aged 48-74 years, with an average age of (63±9) years. All 13 cases of EH were single lesion.

Among the 13 patients, eight patients were found incidentally during health examination or examination for another reasons, including CT and bronchoscopy. Three of the 13 patients with EH are asymptomatic. For the remaining 10 patients had atypical symptoms: cough in 6 cases, chest tightness in 4 cases, chest pain in 1 case, fever in 2 cases and hemoptysis in 2 cases.

There were 3 patients having underlying respiratory diseases: one had chronic obstructive pulmonary disease(COPD), one had allergic alveoli, and one had interstitial lung disease and died of respiratory failure during hospitalization finally. Other basic diseases include hypertension in 5 cases, diabetes in 2 cases and renal carcinoma in 1 case. Six patients had a history of smoking and one had a history of exposing to dusts.

The location of EH

The location of these tumors are as follows: one was found in the right upper lobe bronchus, one in the right middle lobe bronchus, three in the right lower lobe bronchus, one in the right intermediate bronchus, three in the left upper lobe bronchus, two in the left lower lobe bronchus and two in the left main bronchus.

Image features

The following image features of 13 cases are obtained from image and image report records.

Chest radiograph findings were abnormal in 3 of the 11 patients(excepting the 2 patients with allergic alveolitis and Interstitial lung disease), main performance is obstructive pneumonia.

Chest CT showed obstructive pneumonia in 10 of the 13 patients. However, in our study, many cases may be poorly demonstrated or may not be seen at all on chest CT. After our careful review of CT, among 13 patients with EH, visible endobronchial tumor were present in 7 patients, retrospectively. Among these 7 cases, however, before the diagnosis of EH, only 3 cases were diagnosed with endobronchial tumor by radiologists. In addition, two case was misdiagnosed as lung cancer according to CT.

Among 7 visible endobronchial tumors on CT, areas of focal fat were observed in 4 lesions. Calcification can be seen in 3 lesions of all 13 cases. Contrast-enhanced CT was performed in 3 cases, which showed no enhancement in 1 case, slight enhancement in 1 case, obvious heterogeneous enhancement in 1 case. (Figure 1)

Bronchoscopic features

All cases received bronchoscopy. We recorded 10 cases of bronchoscopy images, as follows.

Bronchoscopy showed mass with visible stalk during bronchoscopy in 5 of 10 cases. But we believe that mass with stalk is much more than 5 cases, because some EH obstruct the airway severely, we may not be able to see the stalk of EH of the posterior airway after the obstructed site. Seven cases showed Smooth or regular surface under bronchoscopy, three cases showed irregular surface. For mucosa characteristics: Surface with a lot of necrosis tissues in 1 case, pale mucosa in 2 cases, increased redness mucosa in 2 cases and similar to normal mucosa were present in 5 patients, of which 1 case rich in blood vessels.(Figure 2)

Treatment

Bronchoscopic treatment

Seven patients were treated with bronchoscopic techniques, all of them were treated via flexible bronchoscopy.

Three patients received bronchoscopic treatments more than once: One case(N1) failed in the first interventional treatment with snare via flexible bronchoscope due to difficulty in exposing the stalk of the mass, and then switched to argon plasma coagulation(APC) therapy. Residual tumors was found in the second bronchoscopy, APC was performed the second time. Four months later, bronchoscopy showed that the EH was completely removed, but hyperemia of the mucosa. CT examination showed no recurrence 16 months later.

One case(N2)'s EH was removed partially with snare via flexible bronchoscopy the first time. So a second intervention, rigid bronchoscopy was performed initially, but we found it was difficult to insert rigid bronchoscopy due to patient's narrow oropharynx. Then we switched to flexible bronchoscopy via laryngeal mask airway(LMA). Using APC combined with cryotherapy, the mass was removed completely, taken out by wire basket.

One case(N3) received bronchoscopic intervention 4 times. The mass was cauterized with APC the first time, APC and cryotherapy were used at the second time due to recurrence, and cryotherapy was used the third time. Scar stenosis was found during the fourth bronchoscopy, then dilated by endoscopic biopsy forceps.

For the remaining 4 patients, both of them were treated with snare electrocautery via flexible bronchoscopy, and then mass root was treated with APC or cryotherapy, or combination of the two.

There were no major complications in these 7 cases, except a little bleeding in some patients and one patient who developed slight pneumomediastinum and pneumothorax, successfully recovered with simple oxygen therapy.

Surgical treatment

Six patients received pulmonary lobectomy via thoracotomy. All patients who underwent surgery received bronchoscope biopsy and brush biopsy in the initial bronchoscopy. However, no definite diagnosis of EH was given.

One of them appeared intolerance and sharp increased blood pressure during bronchoscopy, though under adequate local anesthesia with lidocaine. In addition, bronchoscopy showed the mass with a lot of necrotic, which was highly suspected of malignancy. So we gave up the interventional treatment through bronchoscope, and switched to surgery.

For the remaining 5 patients, CT scan showed moderate or severe irreversible pulmonary damage which require pulmonary lobectomy or enlargement of hilar or mediastinal lymph node, which cannot rule out malignancy lymphatic metastasis. The enlargement of hilar and mediastinal lymph node finally confirmed as chronic inflammation by surgical pathology.

Chylothorax occurred in 1 case, and hydrothorax became clear and significantly reduced after 5 days of drainage. No other complications were observed.

The clinical characteristics of 13 cases of endobronchial hamartoma are summarized in Table 1.

Follow-up

Follow-up CT scan was performed in 7 of 13 patients(mean 47 months, range 17-69 months), which revealed no recurrence in all 7 patients. Unfortunately, follow-up bronchoscopy was not performed except two cases(N1, N3) mentioned above, which spanned a longer time period for bronchoscopy(4 months and 5 months in total).

Discussion

Pulmonary hamartoma can be divided into parenchymal type and endobronchial type according to its location, the latter is rare. Parenchymal hamartoma usually has no symptoms, but endobronchial hamartoma often causes atypical symptoms, including cough, sputum, fever, hemoptysis, chest pain, chest tightness[3]. The clinical symptoms of endobronchial hamartoma usually relate to its location and the severity of the obstruction.

In this study, chest radiograph showed abnormalities only in patients with moderate or severe obstructive pneumonia caused by EH. CT can indicate EH by showing endobronchial lesion directly or obstructive pneumonia and atelectasis caused by EH indirectly. However, definite diagnoses of endobronchial mass given by radiologists in our study were not many. We think there are many reasons for missed diagnosis: There were 6 cases of EH in segmental and subsegmental bronchi in our study, which can be easier missed than lesion located in main and lobar bronchus. In addition, it is not easy to distinguish endobronchial lesion on CT when obstructive pneumonia and atelectasis mixing with EH. Fat and calcification density are the characteristics of pulmonary hamartoma. It has been reported that EH contains more fat density[4]. In our study, calcification within the lesion were presented in only 3 cases,

which was more likely to be missed. In imaging diagnosis, radiologists sometimes may neglect examination of airway, which also caused missed diagnosis.

Bronchoscopy is an important diagnostic method for EH, which can directly observe the tumor and evaluate the location, size, shape, base and surface mucosa of the tumor. At bronchoscopy, most EHs appeared as smooth surfaced round masses with stalk[5]. It is difficult to identify with malignant tumor when necrotic tissue exist on the surface of EH. All surgical cases in this study had biopsy and brush biopsy in the initial bronchoscopy before surgery, but no definite diagnoses were made. Because biopsy tissue is too little to provide adequate pathological information, the positive rate of EH routine biopsy via bronchoscope is low, except complete or partial excision via bronchoscopic intervention[6]. However, it plays an important role in confirming endobronchial lesion and providing preliminary information about judgment of benign lesion and strategies for further treatment. In this study, bronchoscopy had important effects on diagnosis of EH, especially in those whose EH cannot be clearly showed on CT. So when endobronchial lesions are suspected on CT, then further bronchoscopy should be considered.

The traditional treatment of EH is surgical treatment. With the development of bronchoscopic interventional techniques, bronchoscopic intervention provide an opportunity of non-operational treatment for EH, has the advantages of simple operation, high security, significant effect, less postoperative complications, fast recovery and low cost. The common interventional techniques of EH treatment include the use of electrocautery, APC, cryotherapy, laser, and balloon dilatation. The above methods have their own indications and advantages, but some limitations[5,6,7,8,9,10].

Electrocautery is an effective therapeutic option for endobronchial tumor. Snare electrocautery is a good option for EH especially, because EH usually have pedicle attached to bronchial wall. For EH with stalk, it is critical to find the location of stem before snare electrocautery[6]. The snare is placed over the lesion and wrapped around the stem. The stalk is then cut while concurrently providing hemostasis[7,8,9,10]. Snare electrocautery can quickly remove the large tumor in the airway at one time, reducing the treatment time and avoiding multiple complications such as perforation of the airway wall, bleeding and airway burn caused by repeated electrocautery, compared with other electrocautery instruments like forceps, knives or rounded probes.

Endobronchial hamartoma has the possibility of recurrence[3]. The key to prevent recurrence is the treatment of residual root, which is usually done by APC or cryotherapy. APC is complementary to other techniques usually. With its superficial effect on tissue, it is safer to remove the tumor in the airway and can also be used for superficial hemostasis, but it can't avoid the disadvantage of granulation tissue proliferation after treatment[7,8,9,10]. Cryotherapy is based on cytotoxic effects of low temperature on living tissue by repeated freeze thaw cycle, which is both immediate and delayed. Because of low water content of cartilage, collagen and poorly vascularized tissue, cryotherapy has less effects on them. So the scarring is minimal and the damage of cartilage is few[8,9,10].

Each technique has its own characteristics. Therefore, in practical applications, various techniques should be combined according to intraoperative situation and experience of operators. However, in one case(N3), even with the combination of APC and cryotherapy, bronchial stenosis caused by scar hyperplasia occurred. We suspected that scar hyperplasia may be related with repeated treatment.

Despite the widespread use of flexible bronchoscope, rigid bronchoscope has many advantages in airway maintenance, controlling massive hemoptysis, shorter intervention time and getting out large tissue, compared with flexible bronchoscope. But rigid bronchoscope can only reach the main bronchi and intermediate bronchus, cannot reach peripheral bronchus, especially superior lobar bronchus.

Nowadays, the combination of rigid and flexible bronchoscope intervention technology can compensate for this shortcoming. The difficulty of rigid bronchoscope implantation and anatomic contraindications limit its wide application. Rigid bronchoscopy should be performed under general anesthesia[8,9,11,12]. In this study, a patient failed in inserting rigid bronchoscope due to his narrow oropharynx after general anesthesia. Then we switched to flexible bronchoscopy via laryngeal mask airway(LMA). LMA is a new type of ventilation, with the advantage of simple operation, less airway irritation and less influence on hemodynamics. Flexible bronchoscopy can be performed easily via LMA, which can ensure patients' ventilation function during general anesthesia and improve the safety of bronchoscopic treatment[12,13,14].

Although bronchoscopic treatment is the first choice for EH. But in some instances, surgery is inevitable mainly because of irreversible lung damage, especially difficulty in differentiating benign and malignant lesions. Surgical treatments may include sleeve resection with bronchoplasty, lobectomy, and even pneumonectomy in complicated cases, while normal lung tissues should be reserved as much as possible[15,16,17]. In this study, many cases accompanied with irreversible lung damage, more importantly, some of them were highly suspicious of malignancy, therefore lobectomy take a high proportion of treatment in this study.

Conclusions

In conclusion, in clinical work, ignoring examination of airway in CT images may cause missed diagnosis of endobronchial tumor. Bronchoscopy plays an important role in diagnosis and treatment of EH. When endobronchial lesions are suspected on CT, then further bronchoscopy should be considered. Early detecting of EH may avoid lung damage. The management of endobronchial hamartomas is individualized according to the characteristics of each patient and each hamartoma. And the interventional treatment through bronchoscope is preferred whenever possible. But in some instances, surgery should be considered.

Declarations

Authors' contributions

SB and WL designed the study, SB and XT collected data and wrote the first draft of the manuscript, JW, XZ, YC, XY and WL analyzed the data and edited the draft. All authors have read and approved the manuscript.

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Availability of data and materials

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

Ethics approval and consent to participate

This study is a retrospective study, which only collected the clinical data of patients and removed names of patients, so no ethical approval required.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

Abbreviations

EH: endobronchial hamartoma, CT: computer tomography, LMA: laryngeal mask airway, APC: argon plasma coagulation.

References

- [1] De Cicco C, Bellomi M, Bartolomei M, Carbone G, Pelosi G, Veronesi G, De Pas T, Spaggiari L, Paganelli G. Imaging of lung hamartomas by multidetector computed tomography and positron emission tomography. ANN THORAC SURG 2008, 86(6):1769-1772.
- [2] Gjevre JA, Myers JL, Prakash UB. Pulmonary hamartomas. MAYO CLIN PROC 1996, 71(1):14-20.

- [3] Cosio BG, Villena V, Echave-Sustaeta J, de Miguel E, Alfaro J, Hernandez L, Sotelo T. Endobronchial hamartoma. CHEST 2002, 122(1):202-205.
- [4] Ahn JM, Im JG, Seo JW, Han HS, Yoon HK, Kim WS, Yeon KM. Endobronchial hamartoma: CT findings in three patients. AJR Am J Roentgenol 1994, 163(1):49-50.
- [5] Kim SA, Um SW, Song JU, Jeon K, Koh WJ, Suh GY, Jung MP, Kwon OJ, Park JH, Yi CA et al. Bronchoscopic features and bronchoscopic intervention for endobronchial hamartoma. RESPIROLOGY 2010, 15(1):150-154.
- [6] Liu C, Wang JJ, Zhu YH, Chen C. Successful use of snare electrocautery via flexible fiberoptic bronchoscopy for removal of an endobronchial hamartoma causing chronic lung atelectasis and mimicking malignancy. THER ADV RESPIR DIS 2017, 11(12):435-438.
- [7] Mahajan AK, Ibrahim O, Perez R, Oberg CL, Majid A, Folch E. Electrosurgical and Laser Therapy Tools for the Treatment of Malignant Central Airway Obstructions. CHEST 2019.
- [8] Bilaceroglu S. Endobronchial Ablative Therapies. CLIN CHEST MED 2018, 39(1):139-148.
- [9] Lin CY, Chung FT. Central airway tumors: interventional bronchoscopy in diagnosis and management. J THORAC DIS 2016, 8(10):E1168-E1176.
- [10] Sachdeva A, Pickering EM, Lee HJ. From electrocautery, balloon dilatation, neodymium-doped:yttrium-aluminum-garnet (Nd:YAG) laser to argon plasma coagulation and cryotherapy. J THORAC DIS 2015, 7(Suppl 4):S363-S379.
- [11] Flannery A, Daneshvar C, Dutau H, Breen D. The Art of Rigid Bronchoscopy and Airway Stenting. CLIN CHEST MED 2018, 39(1):149-167.
- [12] Swanson KL. Airway foreign bodies: what's new? Semin Respir Crit Care Med 2004, 25(4):405-411.
- [13] Sarkiss M. Anesthesia for bronchoscopy and interventional pulmonology: from moderate sedation to jet ventilation. CURR OPIN PULM MED 2011, 17(4):274-278.
- [14] McNamee CJ, Meyns B, Pagliero KM. Flexible bronchoscopy via the laryngeal mask: a new technique. THORAX 1991, 46(2):141-142.
- [15] Wang T, Liu Y. Outcomes of surgical treatments of pulmonary hamartoma. J CANCER RES THER 2016, 12(Supplement):116-119.
- [16] Tomos P, Karaiskos T, Lahanas E, Paulopoulos D, Papahristou D, Stauroulias A, Papachristodoulou A. Transverse bronchoplasty of the membranous wall after resection of an endobronchial hamartoma. ANN THORAC SURG 2007, 83(2):703-704.

[17] Guo W, Zhao YP, Jiang YG, Wang RW, Ma Z. Surgical treatment and outcome of pulmonary hamartoma: a retrospective study of 20-year experience. J Exp Clin Cancer Res 2008, 27:8.

Table

Table 1 The clinical characteristics of EH patients

Patient's number (N)	sex	Symptoms	Distribution	Display EH clearly on CT	Obstructive pneumonia on CT	Anesthesia methods for bronchoscopy	Bronchoscopic intervention times	Treatment
N1	M	NR ^a	left upper lobe bronchus	yes	mild	local anesthesia	2	1 st APC 2 nd APC
N2	M	chest tightness	intermediate bronchus	yes	mild	1.local anesthesia 2.general anesthesia	2	1 st snare electrocautery 2 nd failed in rigid bronchoscopy switched to flexible bronchoscopy via LMA: APC+ cryotherapy
N3	F	cough, chest tightness	medioanterior basal segmental bronchus	no	mild	local anesthesia	4	1 st APC 2 nd APC+cryotherapy 3 rd cryotherapy 4 th dilated by endoscopic biopsy forceps
N4	F	cough, hemoptysis	right lower lobe basal segment	no	moderate	local anesthesia	0	lobectomy
N5	M	fever	right lower lobe basal segment	yes	moderate	local anesthesia	0	lobectomy
N6	M	NR ^a	left main bronchus	yes	no	combined intravenous anesthesia+ local anesthesia	1	snare electrocautery+cryotherapy
N7	M	chest pain	left lower lobe superior segment	no	severe	local anesthesia	0	lobectomy
N8	M	cough, fever	left upper lobe bronchus	no	moderate	local anesthesia	1	snare electrocautery+APC
N9	M	cough, hemoptysis	left upper lobe bronchus	no	moderate	local anesthesia	0	lobectomy
N10	M	no ^b	left main bronchus	yes	no	local anesthesia	1	snare electrocautery
N11	M	cough, chest tightness	right lower lobe superior segment	yes	no	local anesthesia	1	snare electrocautery+APC+ cryotherapy
N12	M	no ^b	right upper lobe anterior segment	no	severe	local anesthesia	0	Lobectomy
N13	M	cough,	right middle lobe bronchus	no	severe	local anesthesia	0	Lobectomy

^aNR: not record; ^bno: no symptom

Figures

Figure 1

Chest CT of endobronchial hamartoma: (A),(B) CT shows a lesion within the left main bronchus, containing fat density (white arrow). (C) EH(red arrow) within the right lower lobe basal segment, which was likely to be missed. (D) A patient with interstitial lung disease, EH(blue arrow) was within the superior segment of right lower lobe, which more likely to be missed along with pulmonary fibrosis. (E) CT showed obstructive pneumonia mixed with EH in in right lower lobar, and EH was not visible. (F) CT showed severe irreversible pulmonary damage in anterior segment of right superior lobe, and EH was not visible.

Figure 2

Endoscopic appearance of endobronchial hamartoma: (A) EH almost completely blocked the left upper lobe bronchus, which with smooth surface and mucosa similar to normal. (B) Bronchoscopy showed EH with irregular surface. (C) Bronchoscopy showed irregular surface with a lot of necrosis tissues. (D) EH was treated using snare electrocautery via flexible bronchoscopy. (E),(F) Bronchoscopy showed EH partially obstructed the left main bronchus, after treatment bronchoscopy showed EH was removed and hyperemia of the mucosa.