Annals of Diagnostic Pathology 18 (2014) 238–243

Contents lists available at ScienceDirect

Annals of Diagnostic Pathology

Pulmonary adenofibroma: clinicopathological study of 3 cases of a rare benign lung lesion and review of the literature

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ARTICLE INFO

Keywords:
Pulmonary
Lung
Adenofibroma
Hamartoma
Solitary fibrous tumor
Immunohistochemistry (IHC)

ABSTRACT

Pulmonary adenofibroma is a rare benign biphasic tumor of the lung composed of epithelial and stromal components. We report 3 cases of this unusual lesion of lung in a male (25 years old) and 2 female (40 and 55 years old) patients. Breathlessness on exertion and mild left-sided chest pain of 1 month's duration were the main concerns in 2 patients, whereas the third had cough and hemoptysis for 3 months. Chest radiograph and computed tomography scan revealed a well-circumscribed, subpleural homogenous mass in left lower chest fields in 2 cases and solid-cystic lesion in left upper lobe in the third patient. All 3 patients underwent lobectomy, following biopsy in 2 cases. Histology revealed a well-circumscribed lesion composed of complex glandlike spaces lined by cuboidal to columnar epithelium surrounded by a hyalinized spindle-cell fibroblastic proliferation reminiscent of adenofibroma of the female genital tract or fibroadenoma of the breast. Immunohistochemical examination supported the diagnosis of a benign pulmonary adenofibroma. All 3 patients were are alive and doing well with no evidence of recurrent or metastatic disease. Diagnosis on biopsy can be challenging and may be misinterpreted as well-differentiated adenocarcinoma with extensive fibrosis or low-grade sarcoma. Frozen-section consultation will be a valuable adjunct in planning for limited lung resection of this benign lung lesion. Although we described 3 cases of pulmonary adenofibroma, still this is the largest published series of this rare entity till date. The possible histogenesis and various differential diagnoses are discussed along with literature review.

1. Introduction

Pulmonary adenofibroma is a rare benign biphasic tumor composed of epithelial and stromal components. Histologically, it resembles adenofibroma of female genital tract and fibroadenoma of the breast. Rarity of this lesion can be endorsed by the fact that only a handful of case reports are available in the literature (Table) [1–7]. Clinical manifestations are usually nonspecific; and radiologically, it appears as a solitary coin lesion. Diagnosis is established only by histopathological examination, and it needs to be distinguished from other benign and malignant entities with biphasic growth pattern [3,6–8].

We hereby report the clinicopathological and immunohistochemical (IHC) features of 3 cases of pulmonary adenofibroma. To the best of our knowledge, this is the largest published series of this rare entity so far and the first ever documented cases of pulmonary adenofibroma from India. The possible histogenesis and various differential diagnoses are discussed along with literature review.

2. Clinical history

2.1. Case 1

A 25-year-old, nonsmoker man complained of breathlessness on exertion, with mild discomfort and pain over left lower chest and low-grade fever for the last 15 days. The chest radiograph revealed a single radiopaque coin lesion in the left lower chest field (Fig. 1a). The computed tomographic (CT) scan showed a well-circumscribed 4.5 × 3.5–cm homogenous soft tissue nodular mass close to the pleural surface in the left lower lobe of lung (Fig. 1b).The patient did not have any significant medical history. All the laboratory parameters were within normal limits. Further clinical and radiological evaluation failed to disclose tumor elsewhere. A CT-guided needle core biopsy revealed tiny fragment of hyalinized and fibrotic stromal tissue only. On imaging, the possibility of carcinoid tumor was considered; and the patient underwent left lower lobectomy. No additional treatment was given. The patient is doing well 5 years postsurgery with no evidence of recurrent disease.

2.2. Case 2

A 40-year-old, nonsmoker woman presented with the chief concerns of cough and hemoptysis for 3 months. Results of her

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http://dx.doi.org/10.1016/j.anndiagpath.2014.04.002
1092-9134/

Table
Cases of pulmonary adenofibroma: literature review

Authors/year	Age (y)/sex	Concerns	Laterality	Location	T size	Type of surgery	IHC profile			Histological diagnosis	Follow-up
							Epithelial component (pos)	Stromal component (pos)	Stromal component (neg)		
Scarf et al (1944)	66/M	Incidental (on autopsy)	Left	Upper lobe, peripheral	1 cm	NA	ND	ND	ND	Fibroadenoma of lung	NA
Butler et al (1969)	NA	NA	NA	NA	NA	NA	NA	NA	NA	Pulmonary hamartoma	NA
Suster (1993)	54/F	Incidental	Right	Upper lobe	1 cm	Lobectomy	CK, EMA	Vimentin	Desmin, SMA, S-100P, CK, EMA	Pulmonary adenofibroma	Alive, FOD 8 y postsurg
	56/M	Incidental	Left	Upper lobe	2 cm	Lobectomy	CK, EMA	Vimentin	Desmin, SMA, S-100P,	Pulmonary adenofibroma	Died unrelated to tumor 5 y postop
Cavazza (2003)	62/M	Asymptomatic	Right	Lower lobe, peripheral	0.8 cm	Excision of nodule	CK, EMA, &TTF1.	Vimentin and CD34 focally positive for Bcl2 & CD99	CK, EMA, TTF1, calretinin, SMA, desmin, & S100	Pseudopapillary variant of SFT	Alive, FOD 18 mo postop
Sironi et al (2005)	68/M	NA	Left	Subpleural, lower lobe	8.8 × 5.7 × 5 cm	NA	TTF-1	CD34 and CD99	NA	Adenofibromatous/ papillary variant of SFT	NA
Vitkovski (2012)	29/F	Midline chest pain	Left	Middle lobe, away from pleura	NM	Wedge resection with VATS	CK & TTF-1	Vimentin, desmin, & weakly positive for SMA-1	CD34, Bcl-2	Pulmonary adenofibroma	Alive, FOD 7 mo postop
Wang et al (2013)	55/M	Left chest discomfort & vague pain	Left	Lower lobe sub pleural,	2 × 1.5 cm	Wedge resection with VATS	CK, EMA &TTF-1	Vimentin & CD34	NA	Pulmonary adenofibroma	Alive, FOD 16 mo postsurg
Kumar et al case 1 (2014)	25/M	Breathlessness on exertion, mild chest pain	Left	Lower lobe, subpleural	4.5 × 3.5 cm	Left lower lobectomy	CK, EMA & TTF-1	Vimentin & CD34, focal desmin & SMA.	S-100 protein, Bcl-2	Pulmonary adenofibroma	Alive, FOD 5 y postop
Kumar et al case 2 (2014)	40/F	Cough and hemoptysis	Left	Lower lobe, sub pleural	5 × 4.3 × 3.8 cm	Left lower lobectomy	CK-7 & TTF1 & Napsin A	SMA	S-100 protein, Bcl-2, CD34, HMB45	Pulmonary adenofibroma	Alive, FOD 12 mo postop
Kumar et al case 3 (2014)	55/F	Breathlessness on exertion	Left	Upper lobe, sub pleural	2.2 × 2 × 1.9 cm	Wedge resection with VATS	CK-7 & TTF1 & Napsin A	SMA	S-100 protein, Bcl-2, CD34, HMB45	Pulmonary adenofibroma	Alive, FOD 6 mo postop

FOD, free of disease.