
Solitary pulmonary adenofibroma in a middle-aged man with bladder cancer

Sir,

Pulmonary adenofibroma is a rare benign tumor of the lung that is composed of a glandular epithelial and a stromal component. On histology, this tumor resembles an adenofibroma of the breast and the female genital tract.^[1] A limited number of cases of this benign entity have been reported in the literature.^[2] We report a case of pulmonary adenofibroma in a 59-year-old male patient that was found incidentally on chest computed tomography (CT) during the staging work-up of bladder cancer.

A 59-year-old male patient was referred to the urology clinic complaining of intermittent gross hematuria and urinary frequency for the past 4 months before presentation. He also had a dry cough, exertional dyspnea,

and no hemoptysis. The remaining review of systems was negative. His past medical history is significant for hypertension with no other medical illnesses. The patient has 20 pack years smoking history and quit smoking 24 years ago. On physical examination, the vital signs were within normal range, and no significant physical findings were found. On laboratory examination, Urinalysis showed moderate microscopic hematuria and mild proteinuria, but no signs of infection, the remaining laboratory studies were unremarkable. The patient then had a CT scan of the pelvis, which showed a bladder mass measuring 5.1 cm × 4.2 cm. A cystoscopy was performed and showed a 4 cm exophytic papillary tumor; a biopsy was taken and confirmed the diagnosis of transitional cell carcinoma.

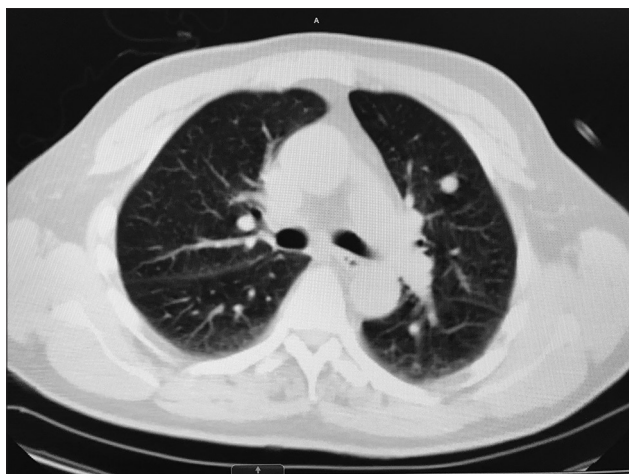


Figure 1: A computed tomography scan revealing an 8.7 mm nodule in the left upper lobe with well circumscribed with rounded borders

Following the diagnosis of bladder cancer, a chest and abdomen CT scan was ordered for staging purposes and revealed an 8.7 mm nodule in the left upper lobe; the lesion is well circumscribed with rounded borders [Figure 1]. This was followed by a CT-guided biopsy of the lesion, which; on microscopic examination; showed benign alveolar tissue along a fibroepithelial neoplasm [Figure 2]. The stroma was myxoid to mildly fibrotic. The intervening glands were small and benign with an intact basal layer that is P61 positive. The stroma showed no positivity for smooth muscle actin or desmin. Thus, the diagnosis of pulmonary adenofibroma was made. The lesion is to be surgically resected after the patient's bladder cancer is treated.

Pulmonary adenofibroma is a rare biphasic tumor of the lung that was first reported by Scarff and Gowar in 1944.^[3] They suggested that its origin might be the same as lung cartilaginous hamartoma. Butler and Kleinerman described it as a neoplastic finding in 1963.^[4] In 1993, Suster and Moran implied that this tumor is an immature type of hamartoma.^[5] Cavazza *et al.*, 2003 described it as a solitary fibrous pseudopapillary tumor of the lung.^[6] To the best of our knowledge, the exact etiology and histogenesis of the tumor remain unclear.^[7-10]

Histological description of the lesion was consistent in the literature with the epithelial constituent being composed of complex glands lined by simple cuboidal to columnar epithelium that did not show evidence of necrosis,^[7-15] and the stromal constituent being composed of bland spindle cell fibroblastic proliferation.

Immunohistochemical analysis was first carried out by Cavazza *et al.*, 2003.^[6] They demonstrated that the stromal cells were strongly positive for vimentin and CD34, focally positive for BCL2 and CD99, and negative for cytokeratin, epithelial membrane antigen (EMA), Thyroid transcription factor-1 (TTF-1), calretinin, smooth muscle actin, desmin, and S100 protein, and that the epithelial cells were immunoreactive for cytokeratin,

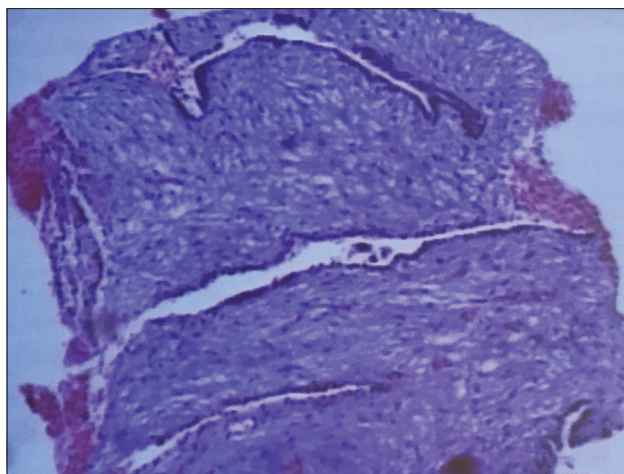


Figure 2: Microscopic examination of lung biopsy specimen revealing benign alveolar tissue along a fibroepithelial neoplasm with myxoid to mildly fibrotic stroma

EMA and TTF-1. Other studies confirmed the previous findings, and further demonstrated that the stromal constituent is positive for CD34. Fusco *et al.*, 2017^[16] further demonstrated that the stromal constituent showed over-expression of the estrogen receptor in 71% of the cases. They also showed a highly recurrent NAB2-STAT6 fusion variant (exon 4-exon 2) in the stromal constituent of the lesions they analyzed.

The clinical presentation in the review of the literature was variable, ranging from asymptomatic incidental finding,^[3-6] to chest pain,^[1,2,7-16] cough,^[8] and hemoptysis.^[2,8] It has been reported in both genders with no obvious gender predilection. The age of presentation ranged from 25 to 66, being more common in people in their fifth and sixth decades.^[1] No risk factors have been suggested by literature review.

Radiologically, the lesion is consistently described in the literature as a well-defined homogenous lesion, ranging in size between 0.8 cm^[6] and 9.5 cm.^[1] It can involve both the upper and lower lobes and was reported more in the left lung than the right lung.^[2]

In the previous studies, the lesion was always managed surgically, by either open thoracotomy or video-assisted thoracic surgery.^[2] To the best of our knowledge, there was no recurrence of the tumor in any of the studies with variable durations of follow-up, the longest being 8 years postsurgery.^[5] We do not report the management or the prognosis as the patient is currently being planned for bladder resection surgery. He is also planned for resection of the lung adenofibroma after the management of bladder cancer.

As a conclusion, it is important to recognize this rare entity and differentiate it from other benign and malignant tumors that have an epithelial and stromal component including hamartomas, carcinosarcoma, spindle cell carcinoma, pleuropulmonary blastoma, and metastasis from other soft tissue and visceral sarcomas.^[7,12]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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REFERENCES

1. Esmaili H, Azimpouran M, Shokohi B, Mostafidi E, Karbasi M. Pulmonary adenofibroma; a rare finding. *Am J Med Case Rep* 2016;4:100-7.
2. Braham E, Helal I, Mlika M, Kilani T, El Mezni F. Pulmonary adenofibroma: Report of a rare case with amyloid deposit. *J Cytol Histol* 2014;6:295.
3. Scarff RW, Gowar FJ. Fibroadenoma of the lung. *J Pathol Bacteriol* 1944;56:257-9.
4. Butler C, Kleinerman J. Pulmonary hamartoma. *Arch Pathol* 1969;88:584-92.
5. Suster S, Moran CA. Pulmonary adenofibroma: Report of two cases of an unusual type of hamartomatous lesion of the lung. *Histopathology* 1993;23:547-51.
6. Cavazza A, Rossi G, De Marco L, Putrino I, Pellegrino S, Piana S. Solitary fibrous pseudopapillary tumor of the lung: Pulmonary fibroadenoma and adenofibroma revisited. *Pathologica* 2003;95:162-6.
7. Vitkovski T, Zeltsman D, Esposito M, Morgenstern N. Pulmonary adenofibroma: Cytologic and clinicopathologic features of a rare benign primary lung lesion. *Diagn Cytopathol* 2013;41:991-6.
8. Kumar R, Desai S, Pai T, Pramesh CS, Jambhekar NA. Pulmonary adenofibroma: Clinicopathological study of 3 cases of a rare benign lung

9. lesion and review of the literature. *Ann Diagn Pathol* 2014;18:238-43.
9. Schirosi L, Lantuejoul S, Cavazza A, Murer B, Yves Brichon P, Migaldi M, et al. Pleuro-pulmonary solitary fibrous tumors: A clinicopathologic, immunohistochemical, and molecular study of 88 cases confirming the prognostic value of de Perrot staging system and p53 expression, and evaluating the role of c-kit, BRAF, PDGFRs (alpha/beta), c-met, and EGFR. *Am J Surg Pathol* 2008;32:1627-42.
10. Rao N, Colby TV, Falconieri G, Cohen H, Moran CA, Suster S. Intrapulmonary solitary fibrous tumors: Clinicopathologic and immunohistochemical study of 24 cases. *Am J Surg Pathol* 2013;37:155-66.
11. Sironi M, Rho B, Spinelli M. Adenofibromatous pattern in a solitary fibrous tumor of the lung. *Int J Surg Pathol* 2005;13:79.
12. Wang Y, Xiao HL, Jia Y, Chen JH, He Y, Tan QY, et al. Pulmonary adenofibroma in a middle-aged man: Report of a case. *Surg Today* 2013;43:690-3.
13. Varoli F, Vergani C, Caminiti R, Francese M, Gerosa C, Bongini M, et al. Management of solitary pulmonary nodule. *Eur J Cardiothorac Surg* 2008;33:461-5.
14. Nicholson AG, Tomaszefski JF, Popper H Jr. Hamartoma. In: Travis WD, Brambilla E, Muller-Hermelink HK, Harris CC, editors. *World Health Organization Classification of Tumours. Pathology and Genetics of Tumours of the Lung, Pleura, Thymus and Heart*. Lyon: IARC Press; 2004. p. 113-4.
15. Akin O, Coskun M. Biliary adenofibroma with malignant transformation and pulmonary metastases: CT findings. *AJR Am J Roentgenol* 2002;179:280-1.
16. Fusco N, Guerini-Rocco E, Augello C, Terrasi A, Ercoli G, Fumagalli C, et al. Recurrent NAB2-STAT6 gene fusions and oestrogen receptor - A expression in pulmonary adenofibromas. *Histopathology* 2017;70:906-917.

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