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Pulmonary Adenofibroma; A Rare Finding

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Abstract Pulmonary adenofibroma is a rare lung benign tumor resembling adenofibroma of the breast and genital tract. Few cases have been reported with this entity. We report a case of pulmonary adenofibroma in a 65-year-old female, which was found incidentally through the work up for her gastrointestinal problems. Chest radiograph and computed tomography scan disclosed a well circumscribed pulmonary mass at left lower lung lobe. Surgical excision was done and the specimen studied macroscopically and microscopically. Histological findings revealed a biphasic lesion showing a leaflike fibroepithelial pattern with stromal and epithelial components. Immunohistochemical studies showed positivity for pan cytokeratin (CK), CD34, vimentin, smooth muscle actin (SMA), but negative for S100 and calretinin. The patient had neither symptoms nor recurrence through one and half year follows up.

Keywords: *Pulmonary, Lung, Adenofibroma, benign, tumor, immunohistochemistry (IHC)*

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1. Introduction

Pulmonary adenofibroma is a rare benign tumor of lung with biphasic appearance resulting from the intimate admixture of epithelium and stroma. To our knowledge, handful of cases of this diagnose have been reported [1-7]. Its rarity in clinicopathological reporting and the importance of differentiating from other types of benign and malignant tumors, which involve the lung, is challenging. We report a case of pulmonary adenofibroma, which is found incidentally in evaluating patient's constipation. To the best of our knowledge, this is the first documented case of pulmonary adenofibroma in Iran.

2. Case Report

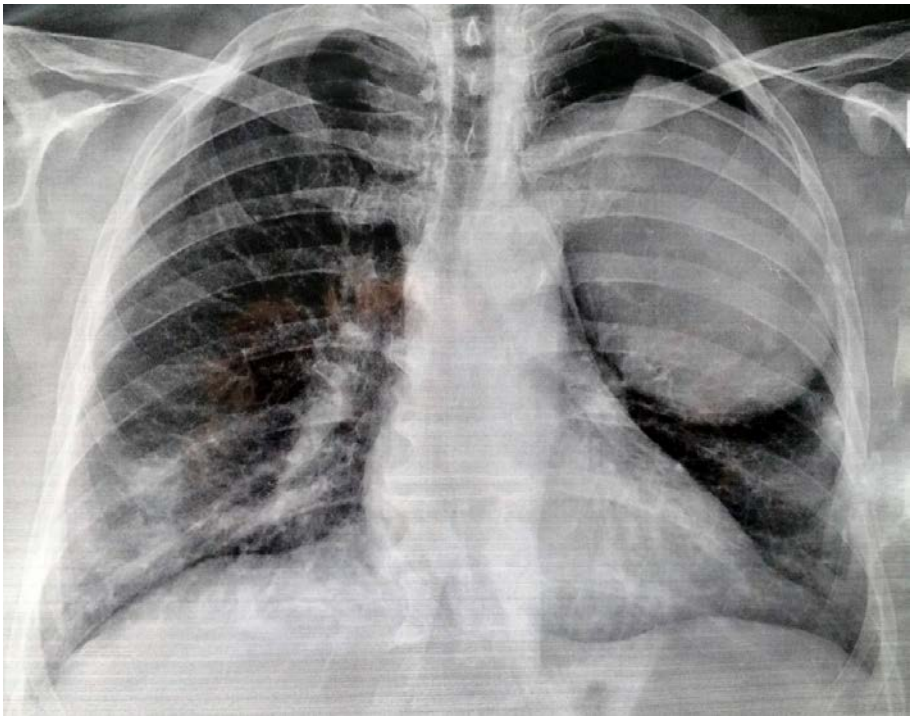

Figure 1. chest radiograph showing left lung lobe mass

A 65-year-old nonsmoker woman complained of vague abdominal pain and constipation for last six months. Her medical history, physical examinations and laboratory findings were not notable. Upper cuts of abdominal computed tomographic (CT) scan, which was done to obtain a clue for patient's discomfort, incidentally revealed signs of pulmonary mass. The patient had undergone chest radiography and chest CT-scan, both showed well-circumscribed soft tissue nodular mass, measuring 9 * 8 cm, located in left lower lobe of lung (Figure 1 and Figure 2). The patient underwent left lower lobectomy. On pathologic findings, grossly, it was a grayish-white well circumscribed subpleural mass, measuring 9.5 * 7.5 * 5 cm (Figure 3). Histological studies showed an unencapsulated biphasic lesion consisted of a leaflike fibroepithelial pattern with stromal and epithelial components. The epithelial component composed of gland like structures with simple columnar to cuboidal lining and the stromal component composed of spindle cell fibroblastic proliferation surrounding the glandular part (Figure 4, Figure 5). Immunohistochemical analysis demonstrated that epithelial component stained positivity for pancytokeratin (CK), whereas the stromal component was positive for SMA, vimentin and CD34 (Figure 6, Figure 7, Figure 8, Figure 9). Immunohistochemistry results for S100 and calretinin were negative (Figure 10, Figure 11). Thus the diagnosis of pulmonary adenofibroma was made. Patient is well, without evidence of disease after one and half year.

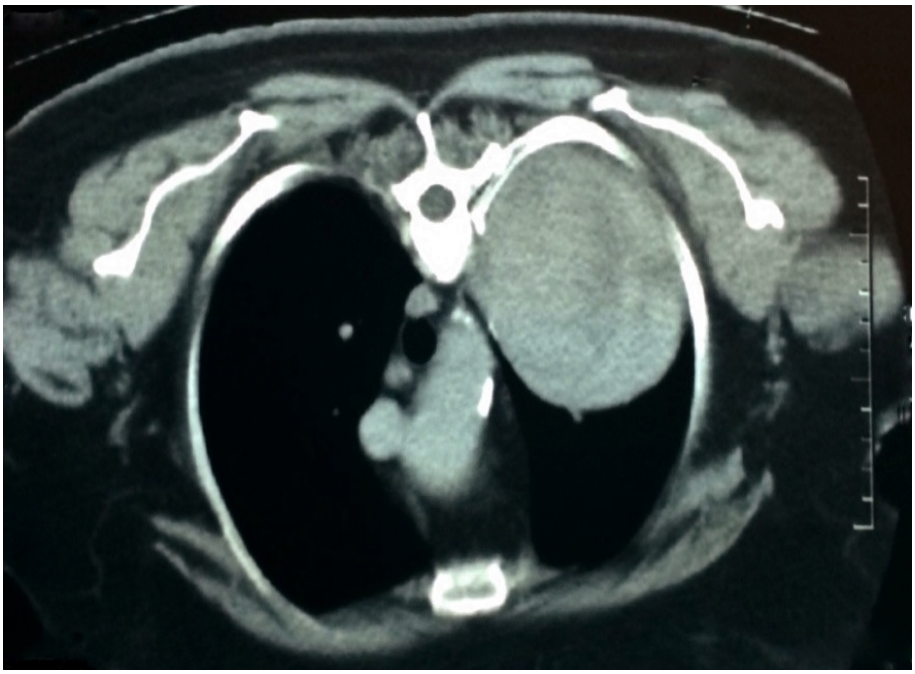

Figure 2. CT-scan disclosing left lung lower lobe mass

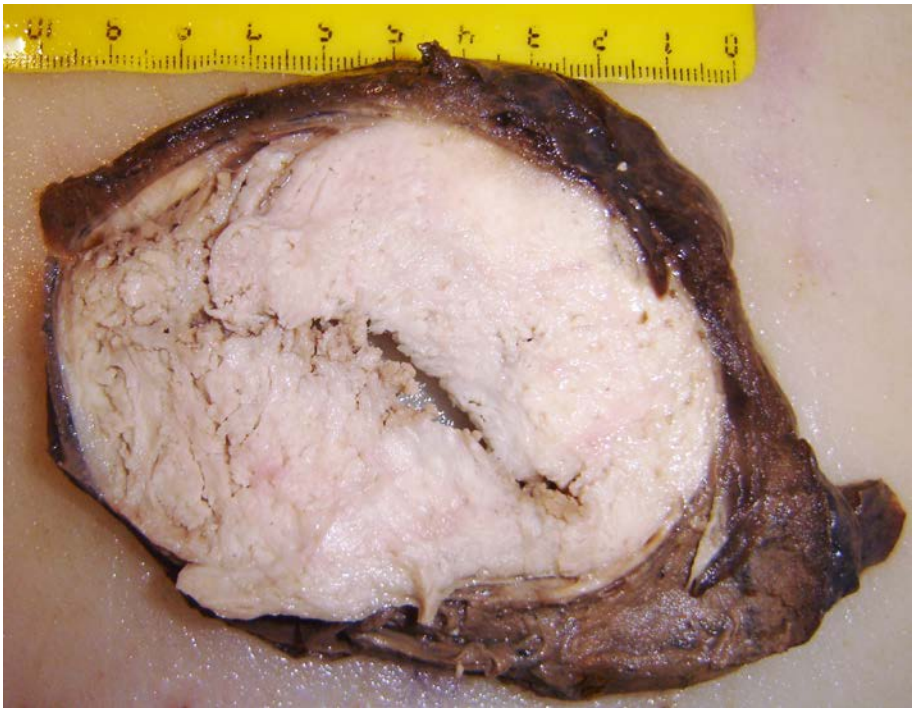

Figure 3. grayish-white well-circumscribed homogeneous mass

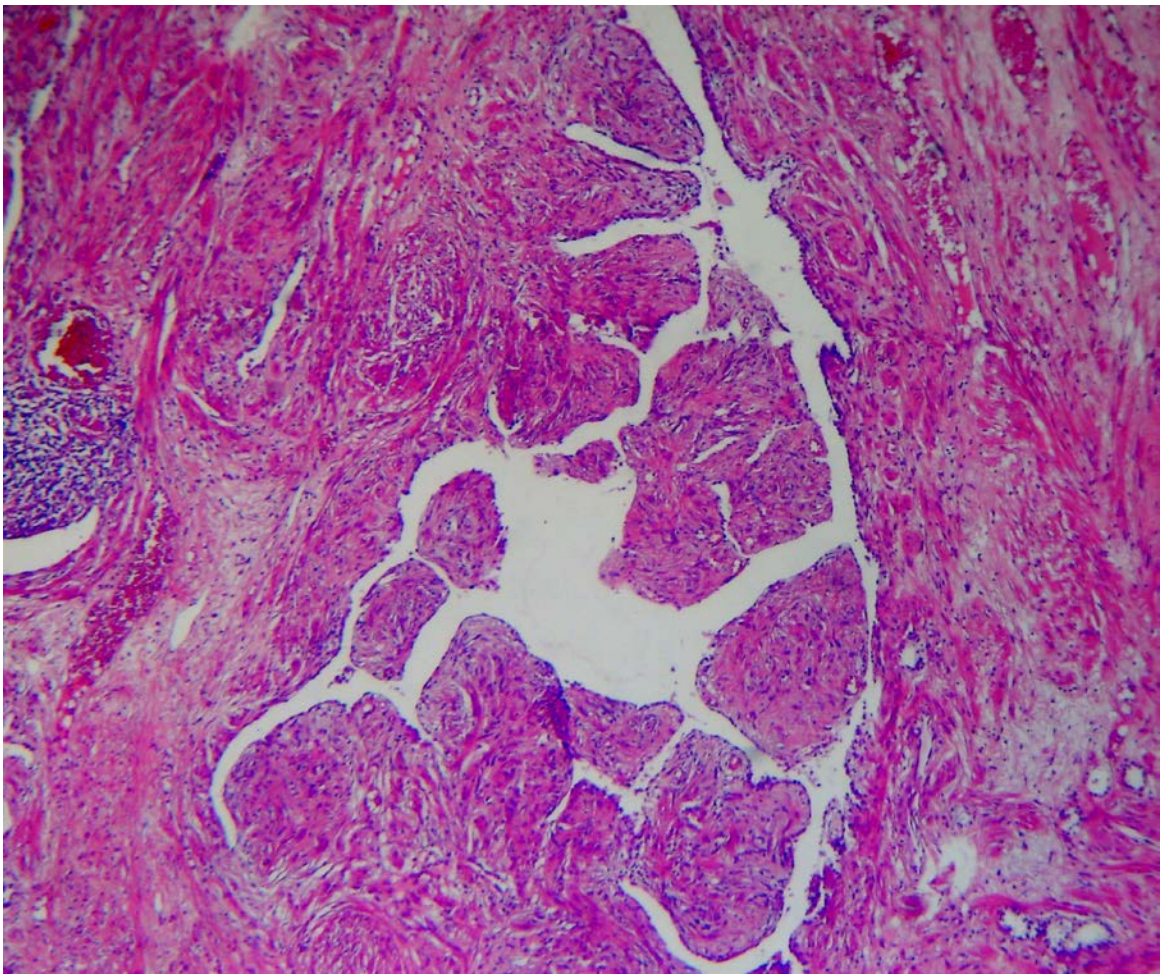

Figure 4. H & E staining, low power(*40) showing leaflike growth patern

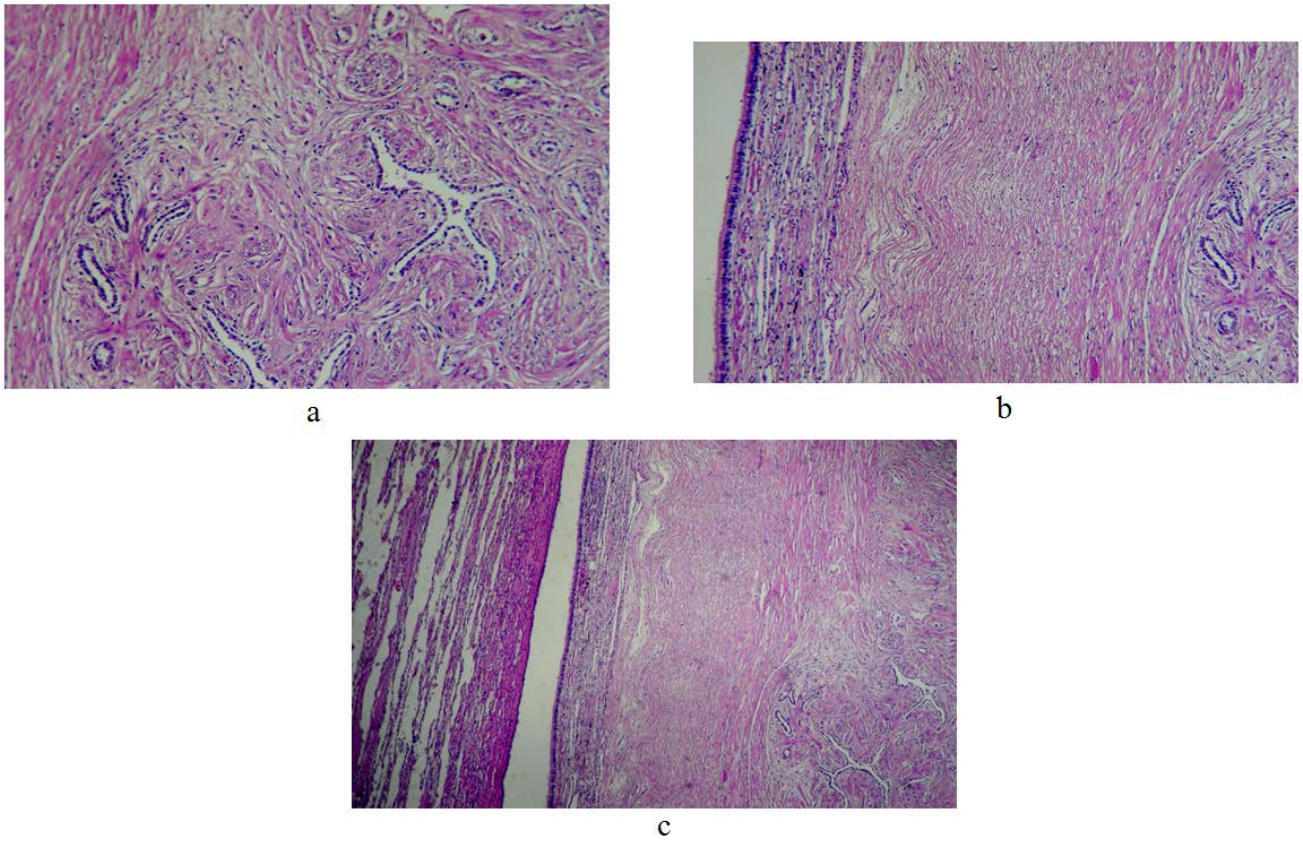

Figure 5. a & b, H & E staining, high power(*400), showing biphasic appearance, composed of epithelial (glandular) and stromal components. 5c,showing pulmonary normal tissue at the left side and biphasic tumoral appearance at the right side

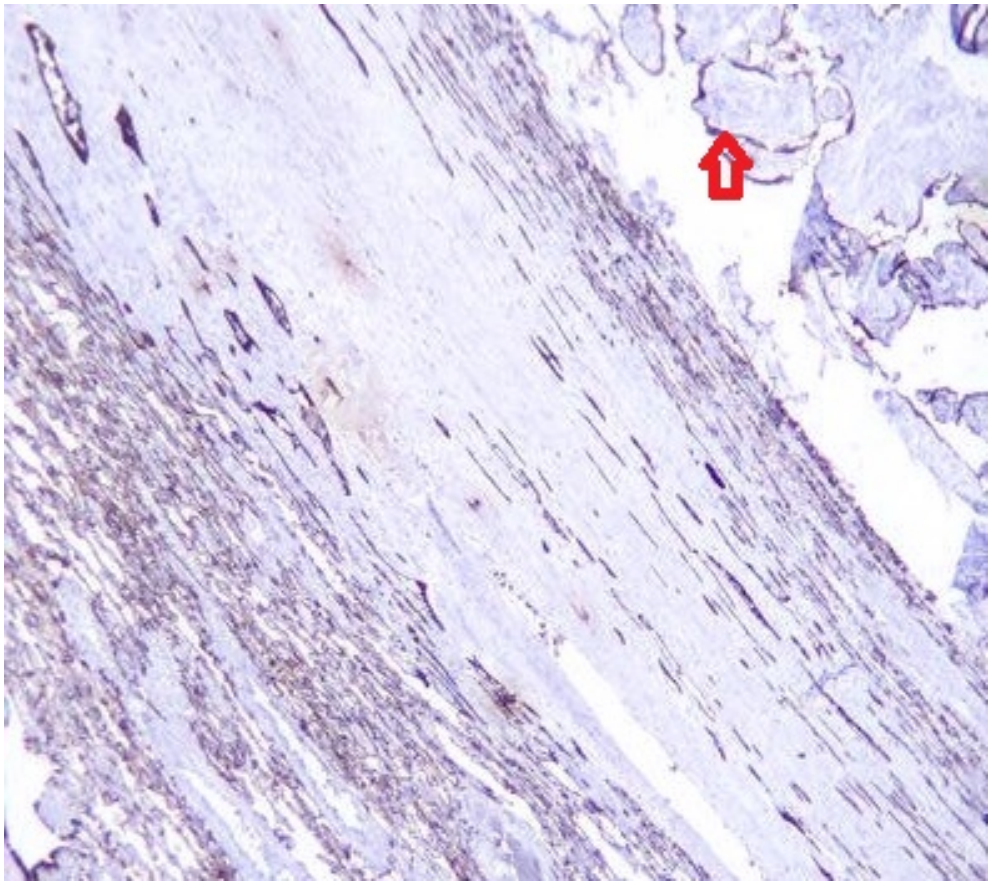

Figure 6. IHC staining for CK show immunopositivity for epithelial component(red arrow)

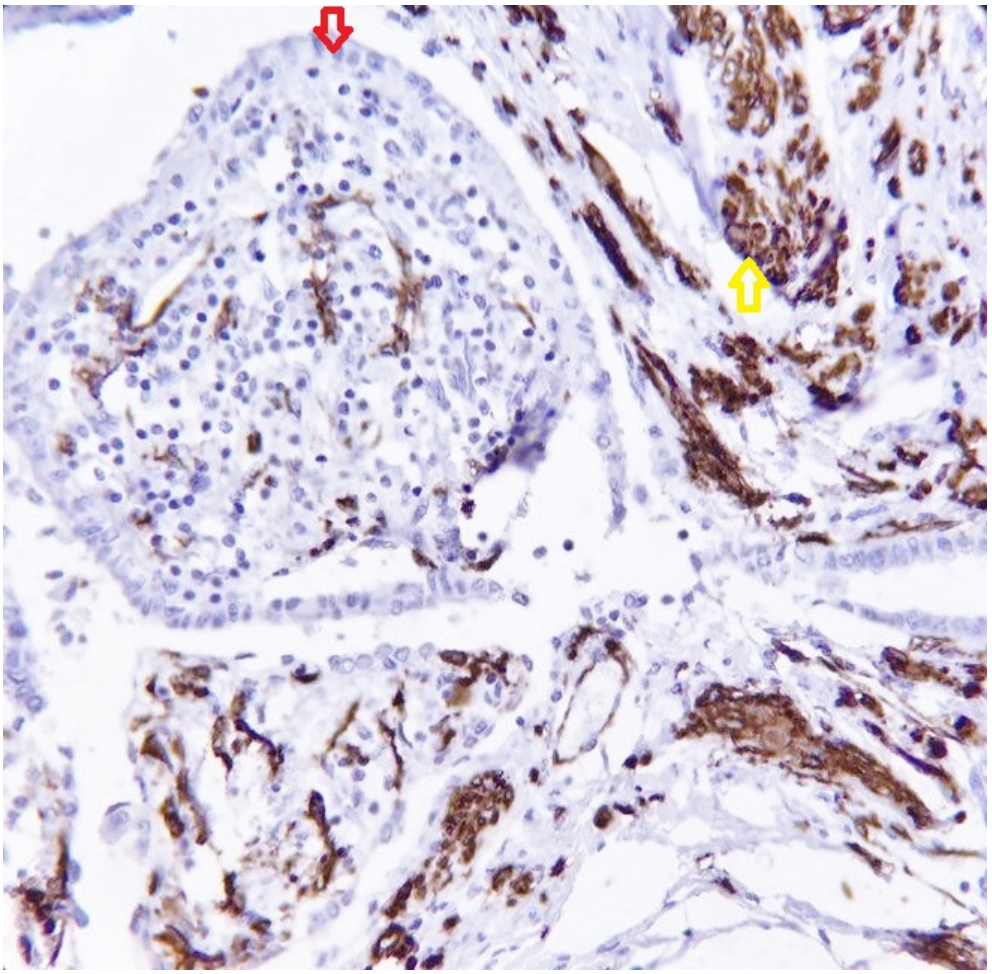

Figure 7. IHC staining for SMA, show negative staining for epithelial component(red arrow) and positive staining for stromal component(yellow arrow)

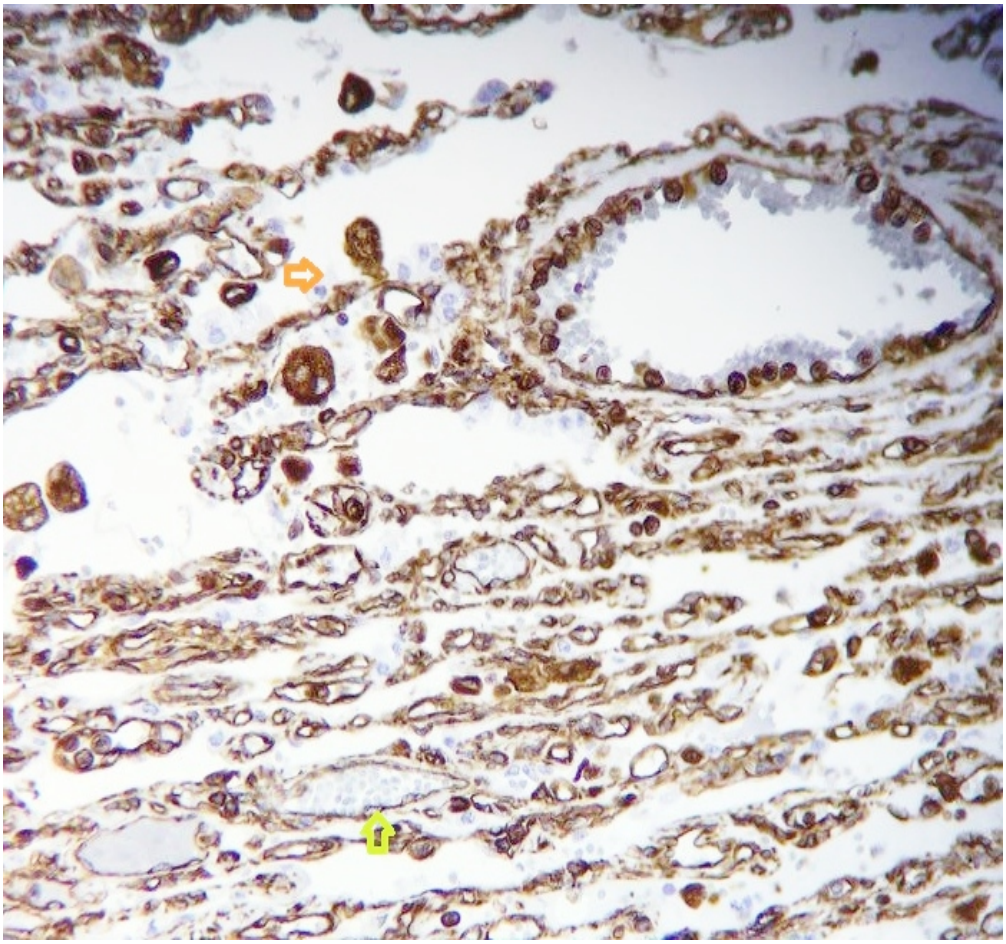

Figure 8. IHC staining for Vimentin, show negative staining for epithelial component and pnuemocytes(red arrow) and positive staining for endothelial cells, fibroblasts and macrophages(yellow arrow).

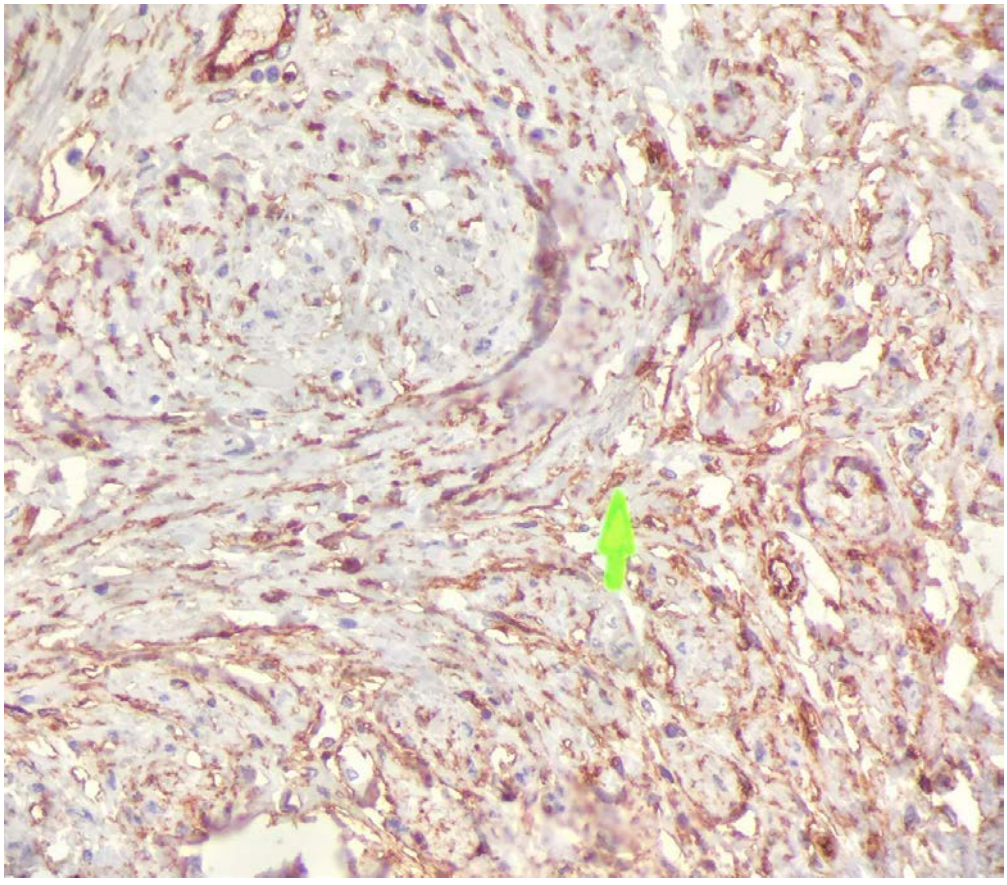

Figure 9. IHC staining for CD34, positive for stromal component(green arrow)

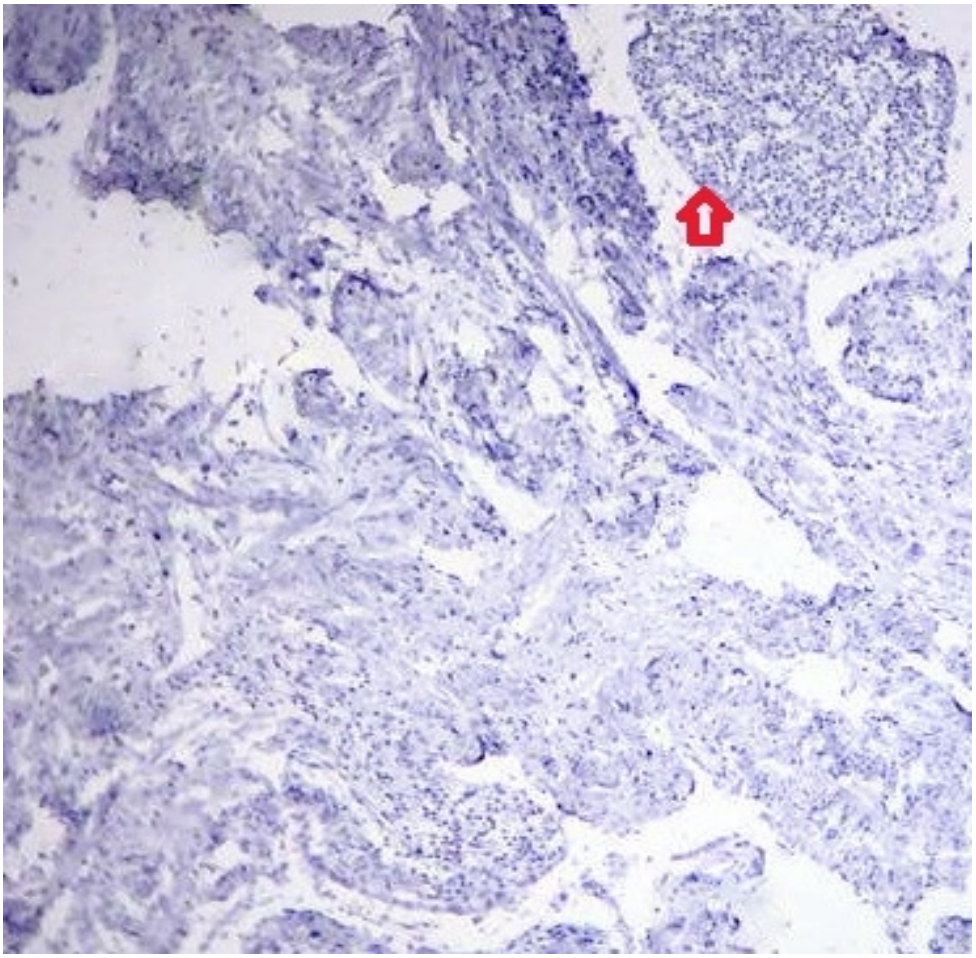

Figure 10. IHC staining for S100, negative(red arrow)

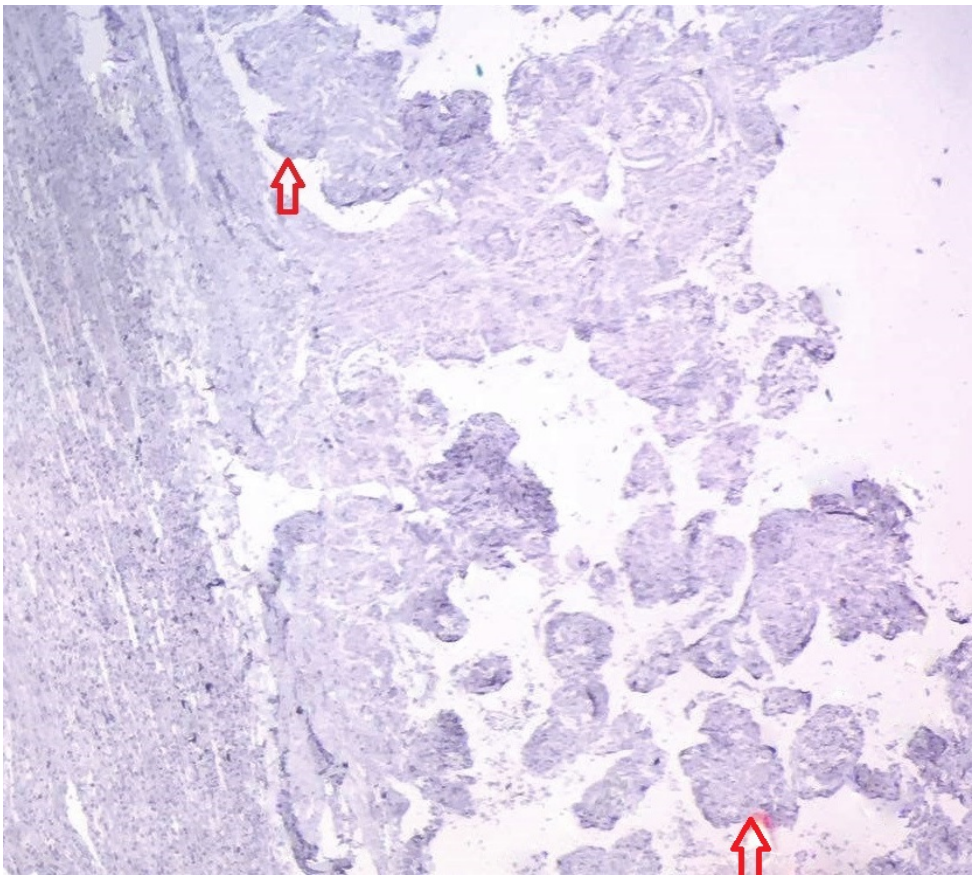

Figure 11. IHC staining for Calretinin, negative(red arrows)

3. Discussion

Pulmonary adenofibroma is an uncommon pathologic entity; its main histogenesis is in doubt. It was first described by Scarff and Gowar in 1944[1], they implied that its origin might be the same as lung cartilaginous hamartoma. In 1969, Butler and Kleinerman described it as neoplastic finding [2], however Suster and Muran in 1993 stated this tumor as an immature form of lung hamartoma [3]. Later in 2005 Cavazza and Rossi interpreted it as Solitary fibrous pseudopapillary tumor of the lung [4].

We reviewed few cases which have been reported as pulmonary fibroadenoma [1-10], there was no specific clinical finding, most of them found incidental. Both genders in any race may be affected. It recognized in adults, mostly in 5^{th} and 6^{th} decade. They didn't have similar lesions in other organs, nor any metastasis after the tumor detected. The tumor had variable size ranging from 0.8 cm to 8.5 cm. Although it is large in size, it can have benign manner without specific clinical findings, as it found incidentally in previously reported cases. To our knowledge our case has the largest size lesion of pulmonary adenofibroma (9.5 cm), which is found incidentally in abdominal CT-scan.

Histologically Pulmonary adenofibroma resembles fibroadenoma of breast and female genital tract, but it is important to distinguish it from other types of lung hamartoma, e.g. pulmonary solitary fibrous tumor, pulmonary blastomas, and metastases from soft tissue tumors. First differential diagnosis is pulmonary hamartoma which is clinically and histologically similar to adenofibroma, but it mainly consists of mesenchymal component like mature hyaline cartilage, which may be calcified or ossified.

Intrapulmonary solitary fibrous tumor is a mesenchymal neoplasm, which is not well recognized as it is also a rare condition [7,8]. IHC profile is worth to notice in which in this case, there was immunopositivity for CK in epithelial component and immunopositivity for CD34, SMA and vimentin in stromal component. Reviewing previous cases disclosed positivity of markers like pancytokeratin, EMA, and TTF-1 in epithelial component in most cases and CD34 and vimentin in stromal component, whereas SMA, desmin and Bcl2 noted to be expressed variably [5-13].

Typically biopsy of pulmonary lesions is the gold standard for diagnosis of many tumors; however in this special disease, histologic similarity with other tumors together with limited reported data about biopsies of such tumors, misdiagnosis by biopsy in our case could be very probable. Meanwhile, due to the large size of the tumor biopsy was not a logical decision.

As histopathologic findings are needed for establishing the diagnosis of pulmonary adenofibroma , VATS seems to be the best diagnostic and therapeutic modality in the management of this lesion[10,11].

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