

# Bronchial Sialadenoma Papilliferum: A Very Rare Cause of Hemoptysis

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## Abstract

**Purpose:** This case is only the third case of the Sialadenoma papilliferum of the bronchial system. This is a extremely rare tumor of the bronchial system. This report highlights once again the histo-pathological difficulties of diagnosing such a rare tumor.

**Patients and methods:** A 53 year old woman with a 3 weeks history of a productive cough associated with hemoptysis presented to the Community hospital Stuttgart (Teaching Hospital of the University of Tübingen). A thoracic CT revealed a solid mass in the right lower lobe with 10 mm diameter. In the community hospital a bronchoscopic biopsy was suspicious for an adenocarcinoma of the lung.

**Result:** The patient was transferred to our institution for thoracic surgery and a right lower bilobectomy with semicircular intrapericardial vessel resection and total nodal resection was performed. By immune-histochemical analysis, the removed tumor (size 10 mm) revealed to be a benign adenoma from the seromucosal bronchial glands, which is a very rare benign tumor of the Sialadenoma papilliferum type. All of the removed lymph nodes were analyzed and showed no signs of malignancy.

**Conclusion:** At present there have been reported only two cases of the pulmonary Sialadenoma papilliferum in the literature. This case report represents the first case of pulmonary Sialadenoma papilliferum in Germany and western Europe. The biologic behavior of this tumor still remains unknown.

## Case Report

A 53 year old woman was brought to the Community hospital of Stuttgart, one of the teaching hospitals of University Tübingen in Stuttgart, for hemoptysis with a 3 weeks history of a productive cough associated with small volume hemoptysis. The patient was a long term smoker with a daily consumption of at least 30 cigarettes per day. Physical examination revealed an expiratory wheeze under forced expiration. Laboratory examination revealed leucocytosis with an elevated level of C-reactive protein. The ECG was unremarkable without any pathological findings. Pulmonary function tests revealed no obstructive or restrictive ventilatory defect. In the community hospital a thoracic computed tomography (CT) showed a solid mass with a diameter of 10 mm in the right lower lobe (Figure 1).

An additional 18F-FDG-Positron emission tomography (PET-CT) showed no distant metastases. At the community hospital bronchoscopy was performed and it showed a mass emanating from the membranous portion of the bronchial wall of the right lower lung lobe. Histological analyses of the biopsy specimen were regarded as suspicious for an adenocarcinoma of bronchiolo-alveolar origin.

The patient was transferred to our hospital for thoracic surgery and a lower bilobectomy with semicircular intrapericardial vessel and total nodal resection was performed. Recovery was uncomplicated. After surgery, the leucocytosis and elevated C-reactive protein serum levels returned to normal values spontaneously.

The resected specimen showed, in the vicinity of the resection margin of the right lower lobe, an exophytic mass extending into and in part occluding the bronchial lumen. Histologically, the tumor showed coarse lobulation and was composed of cystic and papillary

structures (Figure 2A) that well delineated from the neighboring tissues, in which prominent chronic inflammation is obvious (HE x100).

Residing adjacent to the bronchial chondroid in the basal parts of



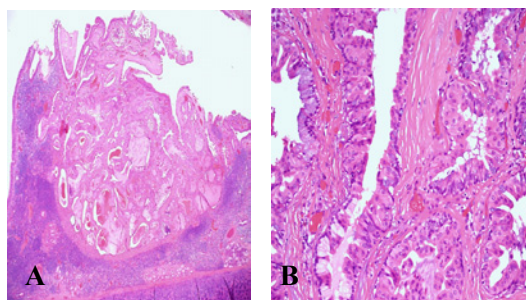
Figure 1:

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**Figure 2:**

the tumor, cystic structures with mucus retention were present, while complex papillary projections prevailed in the superficial parts. The papillae were lined by a single or double row of cells, with features of bronchial epithelium or consisting of slightly enlarged cells with eosinophilic and sometimes granular cytoplasm and round nuclei.

The cysts sometimes featured goblet or mucus-containing cells (Figure 2B). Higher magnification illustrates that the papillary structures are coated by a double row of in part columnar cells with oncocyctic cytoplasm (HE x 400).

Between the glands and papillae, either fibrous tissue was present or, alternatively, mild inflammation could be seen. Immunohistochemistry revealed the epithelial cells to be positive for CK7 and CK5/6 and in part strongly reactive also for protein S100. Smooth-muscle actin was negative, but stained some basal myoepithelial-like cells. CK 20 and TTF-1 were not expressed. A diagnosis of sialadenoma papilliferum of primary bronchial origin was rendered. The remainder of the specimen including the lymph nodes removed failed to show any signs of malignancy. Because of the diagnosis of a benign tumor no further therapy was indicated. Follow up for 2 years was unremarkable.

## Discussion

Sialadenoma papilliferum is a tumor believed to be of salivary gland origin. The great majority of cases arise in the oral cavity. Similar cases are described in the skin as papillary syringocystadenomas. The first case of primary bronchial Sialadenoma papilliferum was reported by Bobos and co-workers (Bobos et al., 2003) with features very similar to the case presented here. Especially, they described an exophytic lesion made up of complex, branching papillary structures with identical morphology and immunophenotype as our case localized in the apical bronchus of the right lower lobe. Their patient was reported to do well over a follow up-time of 8 months following surgical resection. The second case of primary bronchial Sialadenoma papilliferum in a 75 year old man was presented by Honda and co-workers (Honda et al., 2009).

Salivary gland type tumors of the bronchial system include a variety of neoplasms with identical histological features found in tumors of major and minor salivary glands, such as adenoid cystic carcinoma, acinic cell tumors or myoepithelial or epithelial-myoepithelial tumors (Colby et al., 1994; Moran, 1995; Moran et al. 1994). In a certain contrast to the aforementioned tumors, Sialadenoma papilliferum is extremely rare in the bronchial tree, although it has been described in a variety of extra-pulmonary locations, such as the palate, buccal mucosa, the gingival, tonsillar, mandibular and retro molar pad, lip, parotid gland and submandibular gland next to similar tumors arising in the skin (Bobos et al., 2003; Freedman and Lumerman, 1978;

Fantasia et al., 1996; Brannon et al., 2001; Cleary and Batsakis, 1990; Ellis and Auclair, 1996). The pathological features of this neoplasm have been well described. Sialadenoma papilliferum is unique among salivary gland tumors because it generally manifests as an exophytic papillary excrescence of the mucosa rather than a submucosal or intraglandular mass. Salivary gland- type tumors of the bronchi are uncommon and usually present as a polypoid endobronchial lesions (Colby et al., 1994; Moran, 1995). The tumor types mainly include benign (mucus gland adenoma, myoepithelioma, oncocytoma, pleomorphic adenoma) and malignant (acinic cell carcinoma, adenoid cystic carcinoma, adenosquamous carcinoma with amyloid like stroma, carcinoma ex pleomorphic adenoma, epithelial myoepithelial carcinoma, mucoepidermoid carcinoma, sebaceous carcinoma) neoplasms (Bobos et al., 2003; Colby et al., 1994; Moran et al., 1994; Borczuk et al., 2002; Fulford et al., 2001; Moran et al., 1994; Travis et al., 1990).

In conclusion, we here present the only third case reported to date of a primary bronchial Sialadenoma papilliferum, the initial clinical manifestation of which was hemoptysis. This case report highlights some of the histopathological difficulties in the diagnosis of such a rare tumor. Owing to its overall rarity, the long-term biologic behavior of bronchial Sialadenoma papilliferum still remains unknown.

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