# PAPILLARY ADENOMA OF THE LUNG

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The report presents a very rare case of papillary adenoma of the lung in a 61-year old man, described for the first time in the Polish literature.

## Introduction

Papillary adenoma of the lung is a rare benign tumour, commonly located in the peripheral part of the lung, but there have been described a few cases situated more centrally, communicating with the small bronchi [1, 2]. So far, approximately 20 cases of this tumour have been published in the English literature [3]. The first authors to describe the original two cases were Spencer et al. [4] in 1980. Ultrastructural investigations have provided information that epithelial cells in the tumour are predominantly pneumocytes II, which have osmophilic lamellar bodies [2, 5-10]. These cells cover the papillae in a single layer. Interspersed with pneumocytes, there are a few ciliated cylindrical cells and Clara cells [9, 10]. The cytoplasm of the tumour may show surfactant protein A or Clara cell antigen, nuclei TTF1 [3, 6]. The size of the tumour is within the range of 1-4 cm and the tumours are clinically asymptomatic, usually do not grow and are occasionally found in chest X-ray. Fantone and coworkers [7] observed a tumour stable for 10 years. After surgery, such tumours do not show recurrence or metastases. Some authors have considered possible malignant potential of this tumour [6, 10].

## Case report

A clinically asymptomatic, 61-year old man presented with a tumour located in the lower left lung lobe visualized in chest X-ray. He was subjected to a partial excision of the lower lobe along with the tumour. The initial surgical diagnosis was a solitary coin lesion of the lung. The material fixed in formalin was referred to the Chair of Pathomorphology, Jagiello-

nian University Medical College in Krakow. The tumour, 1.5 cm in diameter, was partially cystic, non-encapsulated, grey-white and spongy. The tumour and the adjacent tissues were embedded in paraffin and slides were HE stained and used for immunohistochemical staining: cytokeratin, TTF1, protein surfactant A and Ki67.

Histologically, the tumour was well demarcated, non-encapsulated and it compressed the adjacent lung tissue and partially grew into the lumen of the small bronchi. The appearance of the tumour was papillary, with a single layer of cuboidal cells with round or oval nuclei covering the papillae, which had loose fibrovascular cores (Fig. 1). Some nuclei had inclusions. Mitotic figures and necrosis were absent. On the border of the tumour, a few papillae were covered by ciliated cylindrical cells forming a continuation of the bronchial epithelium. In some fibrovascular cores, there were several lymphocytes and plasma cells (Fig. 2).

Immunohistochemically, the tumour epithelial cells were positive for cytokeratin (Fig. 3), surfactant protein A (Fig. 4) and TTF1. Only single nuclei in the entire tumour were positive for the Ki67 reaction.

Postoperatively, the patient has been followed up since 2002 and continues to do well with no recurrences or metastases.

#### Discussion

This neoplasm can be observed in patients at any age, but more often in men. It is usually a single tumour and radical surgery is sufficient as therapy. Kurotaki *et al.* [11] described one case with multiple papillary tumours in a 13-year old

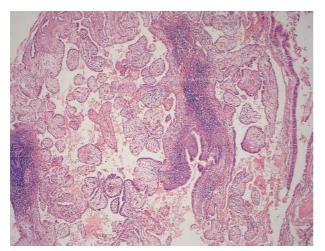


Fig. 1. Papillary adenoma located in the vicinity of the larger bronchi

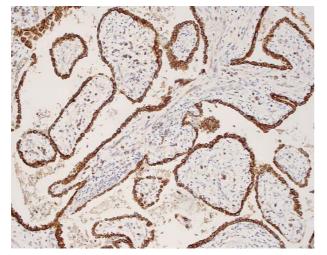
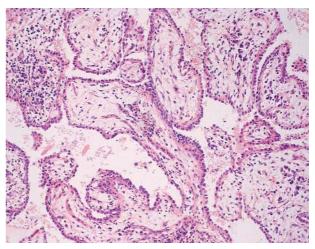


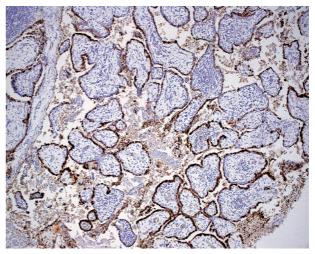
Fig. 3. Positive cytokeratin staining in epithelial cells

boy with von Recklinghausen disease. Differential diagnosis takes into consideration sclerosing haemangioma, alveolar adenoma, bronchiolo-alveolar carcinoma, atypical adenomatous hyperplasia, papillary carcinoid, hamartoma of the lung and also metastatic papillary carcinomas.

Sclerosing haemangiomas have numerous papillary structures, but also solid nests, sclerotic and haemorrhagic parts. With the exception of pneumocytes in sclerosing haemangioma, we can find various precursors cells, round and oval, situated in the central part of papillae. But it is possible that the same papillary adenomas described in the literature were regarded as monomorphic sclerosing haemangiomas. Kurotaki et al. [11] and Mori et al. [10] believe that these tumours may constitute an early stage variant of sclerosing haemangioma.



**Fig. 2.** Papillary structures of the tumour covered by a single layer of cuboidal epithelial cells with loose fibrovascular cores. A few lymphocytes and plasma cells are present in some cores



**Fig. 4.** Surfactant A protein present in cytoplasm of pneumocytes which cover the papillae

Alveolar adenoma (pneumocytoma) is a multicystic tumour lined by pneumocytes and it does not show any papillary structures. Bronchiolo-alveolar carcinoma is a malignant tumour with cellular atypia, mitotic figures, a lepidic growth pattern, irregular borders and sometimes necrosis; in numerous cases, we also observe multiple tumours. Atypical adenomatous hyperplasia also has a lepidic growth pattern and a greater number of dysplastic cells. The rare papillary carcinoids have granular cytoplasm and are positive in staining for chromogranin and synaptophysin.

Metastatic papillary carcinomas show atypia, chaotic architecture and mitotic figures. Sometimes we can observe psammoma bodies in the papillary structures in metastatic carcinomas, which are never seen in papillary adenomas [3, 12, 13]. The presence of multiple tumours and clinical data allow exclusion of metastatic cancer.

In differential diagnosis I also took into consideration pulmonary hamartomas, which show papillary structures and chondroid tissue on the periphery. But in the presented case, the cartilages were fragments of the bronchial wall in the vicinity of the tumour.

It is interesting to know that similar tumours can occur in laboratory animals (mice and rats), either spontaneously or in chemically induced carcinogenesis [5].

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