Case Report

A solitary bronchial squamous cell papilloma with increased 18-fluorodeoxyglucose uptake and high serum levels of squamous cell carcinoma antigen

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Introduction

Squamous cell papillomas are the most common benign tumors of the larynx (1). A solitary bronchial squamous cell papilloma (BSCP) was first reported in 1954 (2). BSCP is a benign tumor originating from squamous cell epithelium cells, and it accounts for 0.38% of all lung tumors (3). There are two primary clinical features of bronchial papillomas. First, BSCP has a tendency to spread to multiple sites within the bronchial tree. Second, previous studies suggest that this type of benign tumor has the potential for malignant transformation (2). The malignant potential of BSCPs ranges from 8% to 40% (4). Therefore, the standard treatment of solitary BSCPs is endoscopic or surgical resection. In this report, we describe a very rare case of solitary BSCP with high serum levels of squamous cell carcinoma antigen (SCC-Ag) and a high maximum standard uptake value (SUVmax), which was difficult to distinguish between benign tumor and squamous cell carcinoma.

Case presentation

A 54-year-old, non-smoking woman presented to the hospital with hemoptysis. She had no health problems or medical history. A chest computed tomography (CT) scan showed a lung mass which was the diameter of 2.6 cm in the right lower lobe (*Figure 1*). Serum SCC-Ag was found to be 6.4 ng/mL, and 18-fluorodeoxyglucose positron emission tomography (FDG-PET) revealed the maximum standardized uptake value (SUVmax) to be 9.05 (*Figure 2*). A bronchoscopy showed normal findings of the bronchus, and no diagnosis regarding the tumor by trans-bronchial lung biopsy could be determined. FDG-PET and brain magnetic resonance imaging (MRI) revealed no distant or lymph node metastasis. We suspected the tumor to be squamous cell carcinoma of the right lower lobe of the lung. We performed right lower lung lobectomy and systematic lymph node dissection, and frozen sections of the tumor showed variant squamous cells. Histopathological findings of the resected specimens revealed a solitary squamous cell papilloma in the right lower bronchus (*Figures 3*,*4*) and DNA examination of the tumor was negative for human papillomavirus (HPV). At 27 months post-surgery, serum levels of SCC-Ag decreased to 0.5 ng/mL and there was no evidence of recurrence.

Discussion

Solitary bronchial papillomas are exceedingly rare, accounting for 7–8% of all benign lung tumors (3). Bronchial papillomas have 3 clinical presentations: multiple papillomas, inflammatory polyps, or solitary papilloma. Multiple papillomas are usually observed in children and affect the larynx, trachea, and bronchi. Inflammatory polyps arise from chronically inflamed bronchial mucosa. Solitary papillomas are the rarest type, and they usually present as an endobronchial mass in the segmental bronchi. The majority of patients with solitary papillomas are smokers, males, and in the sixth decade of life (5,6). However, this patient was a 54-year-old female that did not smoke. Solitary papillomas

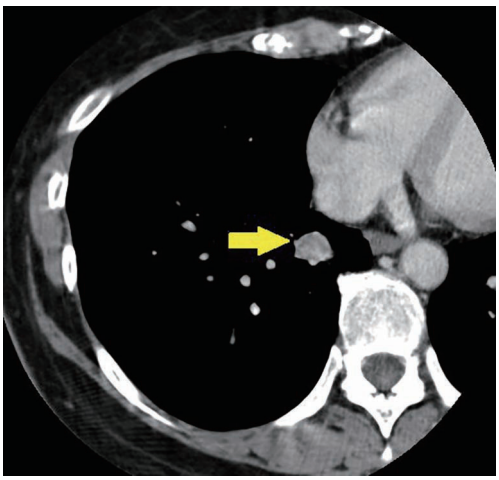

Figure 1 Chest enhanced CT showing a tumor in the right lower lobe of the lung (arrow). The tumor showed enhancement and measured 2.6 cm in diameter. CT, computed tomography.

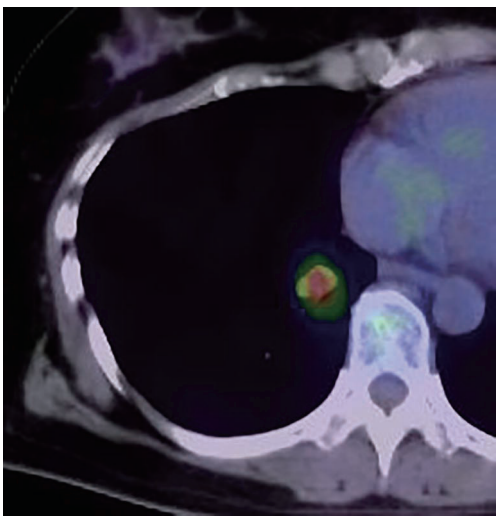

Figure 2 FDG-PET scan showing an SUVmax of 9.05 in the tumor. FDG-PET, 18-fluorodeoxyglucose positron emission tomography.

of the bronchus can be divided into 3 groups: squamous cell papilloma, glandular papilloma, and mixed-squamous cell and glandular papillomas. Most patients present with obstructive symptoms or hemoptysis (6), which was the case for our patient. Some reports have shown that pulmonary mixed-squamous cell and glandular papilloma (PMSGP) display abnormally high SUVmax on FDG-PET, but there has been no report regarding SUVmax in BSCP (7,8). In addition, Masunaga *et al.* reported that glucose

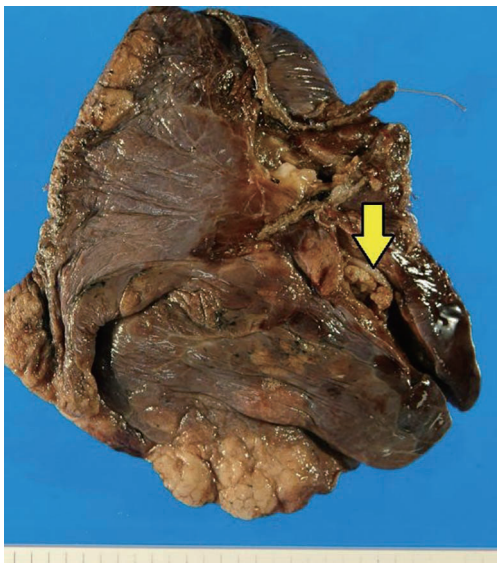

Figure 3 Permanent macroscopic sections show a non-invasive papillary tumor in the right lower lobe of the lung.

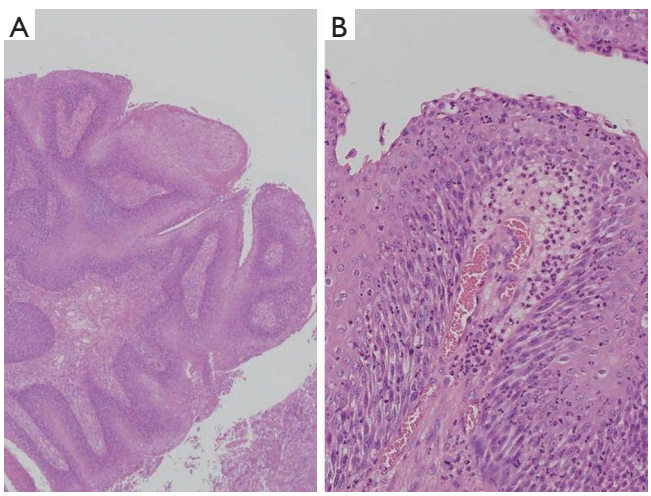

Figure 4 (A) Microscopic examination shows a papillary structure, covered by stratified squamous epithelium (original magnification ×40, hematoxylin and eosin stain); (B) part of the tumor consists of papillary growing with non-atypical squamous epithelial cells (original magnification ×200, hematoxylin and eosin stain).

transporter-1 (GLUT-1) may be associated with high SUVmax in PMSGP (8). On the other hand, there are only a few reports of pulmonary papillomas with elevated tumor markers, such as SCC-Ag or carcinoembryonic antigen (9). However, squamous cell papilloma with high FDG uptake and elevated serum levels of SCC-Ag is a very rare case.

We performed a trans-bronchial lung biopsy, but no definite diagnosis was obtained. Therefore, we performed a right lower lobectomy of the lung with lobe specific node dissection for suspicion of primary lung cancer.

The malignant potential of BSCPs from the literature ranges from 8–40% (3,4). Furthermore, some reports have suggested that the malignant conversion of solitary BSCPs, as well as malignant degeneration of sites where solitary BSCPs have been endoscopically removed. The risk of the malignant conversion of BSCPs increases with smoking status, age greater than 40 years, and infection with HPV serotype 16 or 18 (3,6). Therefore, complete surgical resection is the most common treatment method for solitary BSCPs. We could have performed partial resection or segmentectomy for preservation of pulmonary function, but lobectomy was the only choice for complete resection. In addition, frozen section diagnosis by needle biopsy was not performed due to the risk of massive hemoptysis. It is important to note that solitary BSCPs can present cancer-like findings in terms of FDG uptake and serum levels of SCC-Ag. A correct preoperative diagnosis may allow for a more conservative surgical approach, and the prognosis is good in patients who undergo complete resection of solitary BSCPs (6).

Acknowledgements

None.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Informed Consent: Written informed consent was obtained from the patient for the publication of this case report and any accompanying images.

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