

## Case Report

## Mixed squamous cell and glandular papilloma of the lung increased in twelve years -A case report

Kotaro Kawagishi<sup>\*</sup>, Yuya Kogita, Yuko Kagawa, Yukiyasu Takeuchi, Meinoshin Okumura

Department of General Thoracic Surgery, National Hospital Organization Osaka Toneyama Medical Center, 5-1-1 Toneyama, Toyonaka-shi, Osaka, 560-8552, Japan

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## ABSTRACT

We report the case of a 55-year-old man presented with a nodule in the right middle lung on computed tomography. The size of the nodule had increased at 12 years after the first visit. Consequently, the patient was diagnosed with a Mixed squamous cell and glandular papilloma (MSGP) by surgical resection. To our knowledge, there are no previous studies that MSGP in the lung which had been observed for 12 years before surgery. This study reports the case of a patient who underwent surgical resection of a MSGP.

## 1. Introduction

Solitary endobronchial papilloma (SEP) is a rare benign neoplasm that accounts for < 0.5% of all lung neoplasm. Mixed squamous cell and glandular papilloma (MSGP) of the lung is one of three separate categories of SEP. MSGP is the clinical characteristics remain unclear. We present here a case of an MSGP that had been followed for 12 years preoperatively and had increased in size.

## 2. Case report

A 55-year-old man nonsmoker presented with a nodulous shadow in the right middle lung field on a checkup chest X-ray. A chest computed tomography (CT) revealed a nodule was 13 × 13mm in size of on the right middle lobe. It was circular with well-defined margins. The patient had a history of oral cancer 30 years earlier, but had stopped attending the hospital. His last follow-up chest CT taken 12 years ago showed a 3 × 3 mm nodule. Thus, the nodule was growing slowly and we planned to rule out a malignant tumor (Fig. 1).

Partial resection of the right middle lobe was performed by thoracoscopic surgery. On gross examination, the lesion was a yellowish consolidated mass beneath the pleura, and the intraoperative pathological diagnosis was not a malignant finding. So only a partial resection was performed.

Histologically, the lesion was featured by mixed squamous and glandular epithelium covering fibrovascular cores, displaying a papillary growth pattern (Fig. 2A). The glandular epithelium was predominantly pseudo-stratified ciliated or non-ciliated columnar epithelium and massive mucous columnar epithelium, squamous differentiation components were also seen. Immunohistochemical staining showed the squamous component was positive for p40 and thyroid transcription factor 1 (TTF-1), and the glandular cells were focally positive for TTF-1 (Fig. 2B and C). Both components were positive for cytokeratin and CAM5.2 (Fig. 2D). The final diag-

<sup>\*</sup> Corresponding author. Department of General Thoracic Surgery, National Hospital Organization Osaka Toneyama Medical Center, 1-1-5, Toneyama Toyonaka, Osaka, 560-8552, Japan.

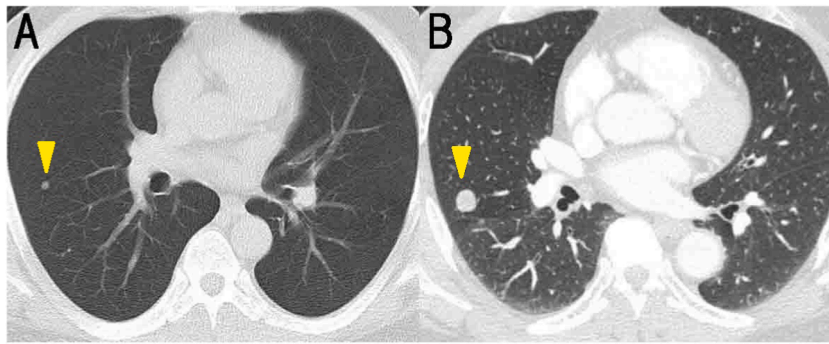
E-mail address: [kawagishi.kotaro.px@mail.hosp.go.jp](mailto:kawagishi.kotaro.px@mail.hosp.go.jp) (K. Kawagishi).

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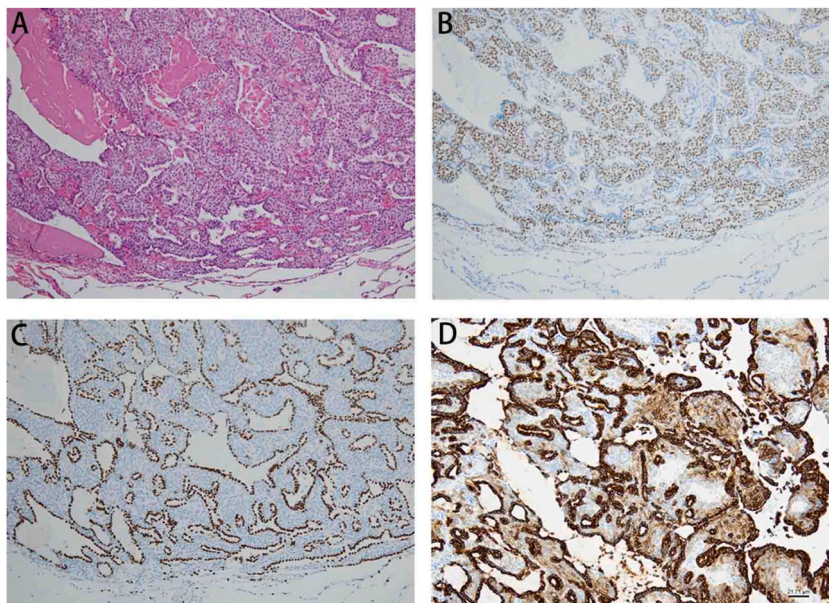
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**Fig. 1.** Chest computed tomography shows  
(A) A nodule located in the right middle lobe measuring  $3 \times 3$  mm in size with a smooth margin. (arrow)  
(B) A nodule was  $13 \times 13$  mm in size, twelve years later. (arrow).



**Fig. 2.** Histopathologic features  
A) Squamous epithelial cells and glandular epithelial cells (H&E100  $\times$ )  
B) P40 positive (100  $\times$ )  
C) TTF-1 positive (100  $\times$ )  
D) CAM5.2 positive (100  $\times$ ).

nosis was MSGP. The postoperative course was uneventful, and the patient showed no recurrence at the final follow-up 6 months after surgery.

### 3. Discussion

Solitary endobronchial papilloma (SEP) is rare benign pulmonary neoplasm, accounts for  $<0.5\%$  of all lung tumors [1]. SEPs are divided into three subtypes in the World Health Organization (WHO) classification: squamous cell papilloma, glandular papilloma, and mixed squamous cell and glandular papilloma (MSGP). Of these types, MSGP is the most uncommon and the clinical characteristics remain unclear [1,2]. SEP usually originates from within the trachea or main-stem bronchi. Therefore, peripheral type MSGP is extremely rare neoplasm. Only 13 cases of peripheral type MSGP have been reported to date, including the present case [3–9].

MSGP is differentially diagnosed from lung tumors such as pulmonary hamartoma, peripheral lung adenocarcinoma, and pulmonary sclerosing hemangioma. According to previous studies, Patients have no specific clinical symptoms. Chest X-ray and chest CT of MSGP showed a well-circumscribed and round nodule; however, these findings were not specific. A study reported that positron emission tomography with 18F-fluorodeoxyglucose (FDG-PET) results in MSGP patients results in mild to high FDG uptake [10].

In this report, we suggest that MGSP may increase slowly over a long period of time. There are no reports of malignant transformation occurring in MSGP, while malignant transformation can occur in the squamous cell component of central type SEP with smoking history, male sex, and human papilloma virus infection identified as possible risk factors for carcinogenesis [2]. Increase in size, up-

take of FDG-PET images, or a contrast effect in contrast-enhanced chest CT are all indicators of the possibility of a malignant tumor, in which case resection for diagnostic and treatment purposes should be planned.

Therefore, we suggest that careful follow-up is an option for peripheral MSGP as a treatment option as long as the patient is asymptomatic and has no obvious issues. On the other hand, we recommend partial resection, segmental lobectomy, or lobectomy if complete resection is necessary, depending on the location and size of the tumor.

Since the number of reported cases of MGSP are low, a further follow-up of new cases with this lesion is needed.

#### 4. Conclusion

We surgically resected a MSGP that had increased in size after a long follow-up period. Some MSGP may slowly increase in size, as in the present case. Furthermore, diagnosis through imaging examinations is difficult, and careful follow-up or biopsy or surgical resection should be considered.

#### Author contributions

Kotaro Kawagishi: Writing - Original Draft,

Yuya Kogita: Validation.

Yuko Kagawa: Validation.

Yukiyasu Takeuchi: Writing - Review & Editing, Supervision.

Meinoshin Okumura: Writing - Review & Editing, Supervision.

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#### Declaration of competing interest

None.

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#### List of abbreviations

MSGP	Mixed squamous cell and glandular papilloma
SEP	Solitary endobronchial papilloma
CT	Computed tomography
TTF-1	Thyroid transcription factor 1
WHO	World Health Organization
FDG-PET	positron emission tomography with 18F-fluorodeoxyglucose

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