

## CASE REPORT

# Asymptomatic solitary papilloma of the bronchus: review of occurrence in Japan

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*Asymptomatic solitary papilloma of the bronchus: review of occurrence in Japan. H. Miura, T. Tsuchida, N. Kawate, C. Konaka, H. Kato, Y. Ebihara. ©ERS Journals Ltd 1993.*

**ABSTRACT:** A case of solitary papilloma of the bronchus is described, and 15 other cases occurring in Japan are reviewed. A 57 year old asymptomatic male was referred to our hospital because many squamous metaplastic cells with moderate atypia were observed in sputum cytology, twice in 3 months. The chest X-ray showed no abnormal findings. Endoscopic examination revealed a polypoid tumour at the bifurcation of the right B<sup>6</sup>a and B<sup>6</sup>b. The tumour was removed completely by endoscopic biopsy, and histological findings showed squamous papilloma. There has been no evidence of recurrence for 3 yrs.

We are not aware of a previously reported case of bronchial papilloma without symptoms or abnormal shadow on chest X-ray. Considering the fact that malignant change was reported in only one case in Japan, and that there is a possibility of cancer subsequently developing at another location in the lung which might also require resection, lobectomy should be avoided if the tumour can be removed completely through endoscopy. If the lesion is limited to a small area in the bronchus, conservative treatment such as photodynamic therapy and/or yttrium aluminium garnet (YAG) laser vaporization might be sufficient to obtain a complete cure.

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Solitary papilloma of the bronchus is a rare tumour, generally discovered when the patient has symptoms, such as continuous cough or bloody sputum, or shows an abnormal shadow on chest X-ray. We report a case of solitary papilloma of the bronchus without any symptom or abnormal shadow on chest X-ray. We also studied the 15 reported cases of solitary papilloma of the bronchus in Japan.

### Case report

A 57 year old male was referred to our hospital because sputum cytology performed in an annual check had showed atypical squamous metaplastic cells that needed more detailed examinations. He had no symptoms. There were no remarkable findings in the physical examination. He had undergone surgical treatment for sinusitis 12 yrs previously, but had no contributory family history. The sputum cytology specimen showed many round-to-oval-shaped metaplastic cells, that had thick cytoplasm and oval-shaped nuclei with smooth margins. The nuclear chromatin was not increased, and nucleoli were not prominent. A few cells were degenerated and had pyknotic nuclei (fig. 1). Similar findings were observed twice in three months. Chest roentgenograms showed no remarkable change.

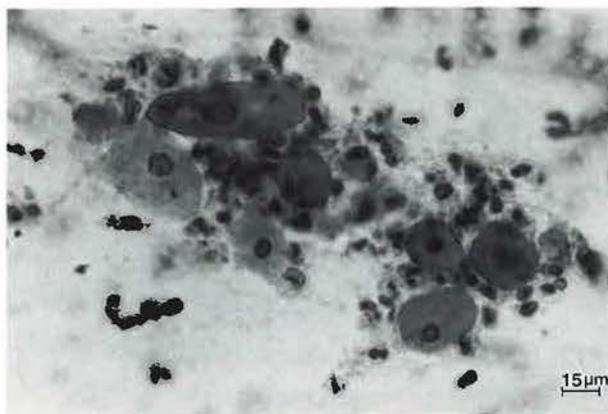


Fig. 1. - Sputum cytology specimen. Round-to-oval-shaped metaplastic cells are observed. Nuclear/cytoplasmic ratio is low. Thick cytoplasm is stained by orange G or light green. Nuclei margins are regular. Nucleoli are not prominent. (Papanicolaou  $\times 400$ ).

Endoscopic examination revealed a polypoid tumour at the bifurcation of the right B<sup>6</sup>a and B<sup>6</sup>b. The tumour had a milky-white, smooth, cauliflower-like shape (fig. 2). There were no tumorous lesions in other bronchi. Endoscopic biopsy removed the tumour completely.



Fig. 2. — Bronchoscopic findings. A polypoid tumour is observed at the bifurcation of the right B<sup>6a</sup> and B<sup>6b</sup>.

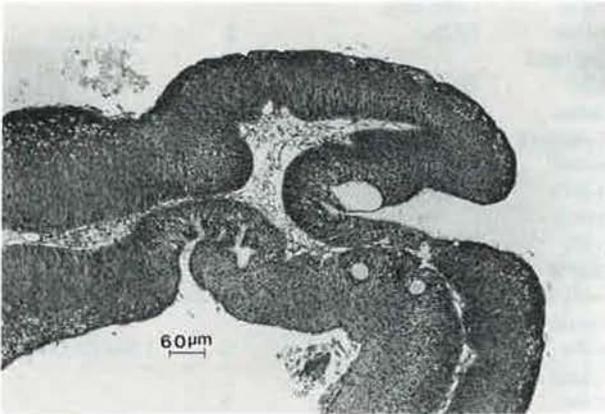


Fig. 3. — Pathological findings. Fibrous stalk of papillary structures, covered by stratified squamous epithelium. (Haematoxylin and eosin  $\times 100$ ).

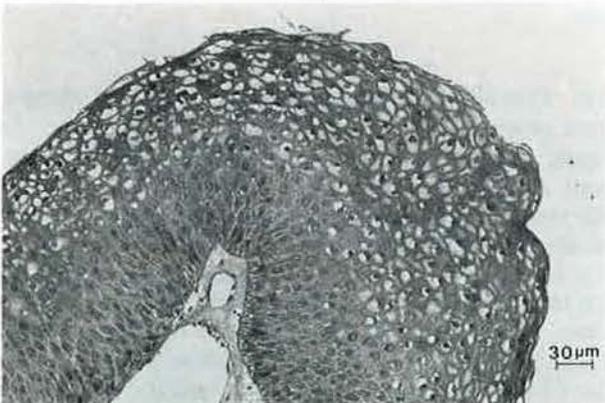


Fig. 4. — Pathological findings. Koilocytosis can be seen in the squamous epithelium. There are no mitoses. Basement membrane is preserved. (Haematoxylin and eosin  $\times 200$ ).

The histopathological findings showed a fibrous stalk of papillary structures, covered by stratified squamous epithelium. Koilocytosis could be seen in the squamous epithelium (figs. 3 and 4). Mitotic figures were rare. Malignant degeneration was not found. The bronchoscopic findings, 3 weeks after the total biopsy, revealed no residual tumour. This case was followed for 3 yrs,

but there was no evidence of recurrence, or growth of another tumour.

### Discussion

DRENNAN and DOUGLAS [1] divided papillomas of the bronchus into three groups: 1) multiple papillomatosis; 2) inflammatory polyps; and 3) solitary papilloma. The first group is usually seen in children, and affects the larynx, trachea and bronchi. Bronchial papillomatosis was observed in 2.1–8.0% of cases of laryngeal papillomatosis [2, 3]. Such cases have seldom been reported in Japan. The cause remains unknown, although a viral cause is strongly suggested [4]. The second group has single or multiple papillary growths, arising in the mucosa of patients with chronic respiratory infections. Histologically, the polypoid lesions are covered with ciliated columnar epithelium, and have a fibrous tissue core with inflammatory cell infiltration. This group should not be considered as neoplasms. The third group, solitary papilloma, is a pedunculated tumour developing from normal bronchial epithelium in older people. MAXWELL *et al.* [5] divided this group into two categories. One is defined as solitary papillomas covered by columnar or cuboidal epithelium, with or without squamous metaplasia, but showing little or no evidence of inflammation. The other consists of solitary squamous papillomas, which have well-formed connective tissue stroma, covered by stratified squamous epithelium. Although this classification is useful when considering the papillomatous findings in the respiratory tract, it is controversial whether or not these groups of solitary papillomas should be distinguished, because most of the solitary papillomas have some degree of squamous metaplasia.

Solitary bronchial papilloma is rare, only 16 cases, including our case, having been reported in Japan (table 1). Age distribution was 22–80 yrs (mean: 54 yrs). Highest incidence was seen in the 6th decade. The male/female ratio was 1.7, male cases being slightly more frequent. There were 11 symptomatic cases. Cough was the most common symptom, followed by bloody sputum. Among the five asymptomatic cases, four were detected by abnormal shadows on chest X-ray. Concerning X-ray findings, tumour shadows were observed in six cases, including three peripheral type. In five cases, secondary changes such as atelectasis or an infiltrative shadow, were found. There were four cases which showed no abnormal findings. It was interesting that our case showed no abnormal findings in the chest X-ray and was without symptoms. Only findings of sputum cytology, which showed the presence of many squamous metaplastic cells with moderate atypia, were clues for detection. The cytology of solitary papilloma of the bronchus was only reported by ROGLIC *et al.* [6]. However, the cytological features of this tumour included regular squamous cells, round basal cells without atypia or nuclear irregularity, and suggested only benign tumour. We agree with their conclusion that the definitive diagnosis can be established only by histological examination.

Table 1. - Reported cases of solitary bronchial papilloma in Japan [13]

| Pt No. | Age yrs | Sex | S.I. | Symptom             | X-ray findings         | Location               | Treatment                               | Histology | Size*          |
|--------|---------|-----|------|---------------------|------------------------|------------------------|---|-----------|----------------|
| 1      | 25      | M   | ?    | Cough<br>Sputum     | Tumour shadow          | Lt-S10<br>Peripheral   | LLL                                     | ?         | Hen's egg      |
| 2      | 22      | M   | ?    | -                   | Tumour shadow          | Rt-B3a<br>Peripheral   | Tumorectomy                             | C?        | Walnut         |
| 3      | 55      | M   | 300  | -                   | Tumour shadow          | Rt-MLB<br>Central      | Rt-pneumonectomy<br>(lung ca. in Rt-S2) | S?        | Pea            |
| 4      | 45      | M   | 750  | Cough               | Tumour shadow          | Lt-B4<br>Central       | Biopsy                                  | C?        |                |
| 5      | 58      | F   | 350  | Bloody<br>sputum    | Atelectasis            | Rt-B4<br>Central       | RML                                     | C         | 0.7x0.7 cm     |
| 6      | 53      | F   | 600  | Haemoptysis         | n.p.                   | Lt-B89<br>Central      | Biopsy                                  | S?        |                |
| 7      | 50      | F   | ?    | Cough               | Atelectasis            | Lt-B6<br>Central       | Lt-pneumonectomy                        | S         |                |
| 8      | 75      | M   | 1000 | Cough<br>Fever      | Infiltrative<br>shadow | Lt-UDB<br>Central      | YAG laser<br>(with cis)                 | S         | Pea            |
| 9      | 60      | F   | 0    | -                   | Tumour shadow          | Rt-B4a<br>Peripheral   | RML                                     | S         | 1.2x1.5 cm     |
| 10     | 66      | F   | 0    | Bloody<br>sputum    | Atelectasis            | Lt-B5<br>Central       | Biopsy                                  | S         |                |
| 11     | 63      | M   | 800  | Stridor<br>Dyspnoea | Lt decrease<br>in size | Lt-main br.<br>Central | Lt main sleeve<br>resection             | C?        |                |
| 12     | 54      | M   | 1200 | Cough<br>Sputum     | n.p.                   | Rt-B45<br>Central      | Biopsy                                  | S         | 0.2 cm         |
| 13     | 25      | M   | 0    | -                   | Tumour shadow          | Lt-B6<br>Central       | Sleeve<br>segmentectomy                 | S         | 4.0x4.0 cm     |
| 14     | 69      | M   | 2820 | Cough<br>Fever      | n.p.                   | Lt-B1+2<br>Central     | Biopsy<br>(triple lung ca.)             | S         |                |
| 15     | 80      | F   | 0    | Chest<br>pain       | n.p.                   | Lt-B6<br>Central       | LLL                                     | S         | 1.5x1.0x1.0 cm |
| 16     | 57      | M   | 700  | -                   | n.p.                   | Rt-B6<br>Central       | Biopsy                                  | S         |                |

S.I.: smoking index; LLL: left lower lobectomy; C: papilloma covered with columnar or cuboidal cells; MLB: middle lobe bronchus; S: squamous papilloma; RML: right middle lobectomy; n.p.: nothing in particular; UDB: upper division bronchus; YAG: yttrium aluminium garnet; \*: the size given is that of the original report.

Most of the tumours were located in segmental or more central bronchi (13 cases), and only three cases were located in subsegmental or more peripheral bronchi. The tumour was located on the left side in 10 cases, and on the right side in six. There was no tendency for the lesion to develop at a particular site. Squamous cell papillomas were observed histologically in 11 cases, and papillomas covered by columnar or cuboidal epithelium in four instances. The question marks in the table mean that there was no clear explanation about histological subtype in the reports, but we classified the cases according to figures and comments in the reports. In the central type lesions, bronchoscopy revealed milky-white or greyish-white, cauliflower-like polypoid tumours, similar to our case. The tumour surface was lustrous and had no necrosis.

The differential diagnosis between this tumour and squamous cell carcinoma was made possible by the bronchoscopic findings. However, since cases with malignant changes have been reported, careful histological examination is necessary. SPENCER *et al.* [9] reported that one of eight solitary papillomas had a focus of carcinoma *in*

*situ*. ROVIARO *et al.* [8] indicated that malignant changes were observed in three of four solitary papillomas. In Japan, only one case has been reported to have malignant change in an autopsy specimen (case No. 8). Moreover, in two cases, squamous cell carcinomas were observed in other bronchi. Case 15 had triple squamous cell carcinomas. Extrinsic carcinogenic factors affecting the bronchus, including smoking, were suspected in such cases. ROVIARO *et al.* [8] suggested smoking to be a cause of malignant changes of benign neoplasia. Among the 13 cases in Japan in which the smoking habits were described, nine cases had a smoking habit, including seven cases of heavy smokers, whose smoking indices exceeded 400. Passive smoking must be taken into consideration in female cases.

Operations were performed in eight cases, including cases which required consideration of malignant changes. ZIMMERMANN *et al.* [9] reported recurrent papilloma of the bronchus after an incomplete endoscopic resection. However, there was no evidence of recurrence at 6 yrs after treatment in case No. 12 at 4 yrs in case No. 14, and at 3 yrs in case No. 16 (our case).

Considering the fact that there was only one case of malignant change reported in Japan, and that there is a possibility of cancer subsequently developing at another location in the lung which might also require resection, lobectomy should be avoided if the tumour can be removed completely through endoscopy. If the lesion is limited to a small area in the bronchus conservative treatment, such as photodynamic therapy [10] and/or yttrium aluminium garnet (YAG) laser [11] vaporization, seems sufficient to obtain a complete cure.

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