Primary epithelial-myoepithelial carcinoma of the lung with cavitary lesion: A case report

MASATAKA MORI[1], TAKESHI HANAGIRI[1], RYOICHI NAKANISHI[2], SHUHEI ASHIKARI[1], MANABU YASUDA[1] and FUMIHIRO TANAKA[3]

[1]Department of Thoracic Surgery, Shin-Kokura Hospital, Federation of National Public Service, Personnel Mutual Aid Associations, Kitakyushu, Fukuoka 803-8505; [2]Department of Oncology, Immunology and Surgery, Nagoya City University Graduate School of Medical Sciences, Nagoya, Aichi 467-8601; [3]Second Department of Surgery, University of Occupational and Environmental Health, Kitakyushu, Fukuoka 807-8556, Japan

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Abstract. Epithelial-myoepithelial carcinoma (EMC) typically arises in the salivary glands, whereas EMC of the lung is an extremely rare histological form that originates from the bronchial glands. Although cavitation in primary lung cancer is not uncommon, to the best of our knowledge, a case of EMC with a cavitary lesion has not been reported to date. We herein describe a case of cavity-forming pulmonary EMC. A 72-year-old man was referred to our department due to a thickened cystic wall discovered in the upper lobe of the left lung and underwent thoracoscopic left upper lobectomy. Microscopically, the tumor was characterized by biphasic architecture, with glands surrounded by myoepithelial cells. The pathological diagnosis was EMC. The patient has remained in good health for 2 years postoperatively, without any evidence of recurrence. As regards the mechanism of cavity formation, it was hypothesized that the bronchial gland in the primary cystic lesion had been present 3 years prior to the development of the EMC, and grew to become a cavitary lesion. Therefore, although the mechanism of cavity formation remains to be elucidated, EMC of the lung may include a cavitary lesion.

Introduction

Epithelial-myoepithelial carcinoma (EMC) is a rare malignant tumor that typically arises in the salivary glands. This tumor displays a typical biphasic pattern, with a central ductular structure surrounded by clear cells of myoepithelial origin. EMC accounts for ~1% of all salivary gland tumors (1,2). EMC may also arise in other locations, albeit less often, such as the minor salivary glands or the upper and lower respiratory tract (3). EMC of the lung is an extremely rare histological form that originates in the bronchial glands. Although EMC of the salivary gland is considered to originate from the intercalated duct (4), pulmonary EMC appears to originate from the ductal structure of the bronchial gland, which is one of the lung counterparts to the intercalated duct (5), and accounts for ~0.1% of all primary lung carcinomas (6). To the best of our knowledge, a case of pulmonary EMC with a cavitary lesion has never been reported to date. We herein report the case of a patient with cavity-forming pulmonary EMC treated by thoracoscopic surgery.

Correspondence to: Dr Masataka Mori, Department of Thoracic Surgery, Shin-Kokura Hospital, Federation of National Public Service, Personnel Mutual Aid Associations, 1-3-1 Kanada, Kokurakita Ward, Kitakyushu, Fukuoka 803-8505, Japan
E-mail: masataka-m@med.uoeh-u.ac.jp

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Case report

A 72-year-old man was referred to the Department of Thoracic Surgery, Shin-Kokura Hospital (Kitakyushu, Japan) in March 2014 due to thickening of the cystic wall in the left upper lung field. The cystic lesion had first been identified on routine medical checkup (annual chest X-ray) 3 years earlier (Fig. 1A and B). The patient did not have any respiratory symptoms, such as cough, hemoptysis or dyspnea. The laboratory findings were unremarkable. A computed tomography (CT) scan of the chest revealed an irregularly shaped lung tumor, 35 mm in greatest diameter, with a cavitary lesion (Fig. 1C). There was no associated enlargement of the hilar or mediastinal lymph nodes. Brain magnetic resonance imaging examination, bone scintigraphy and body CT scan with contrast enhancement detected no distant metastasis or lymphadenopathy. The diagnosis following transbronchial lung biopsy was non-small-cell lung carcinoma; thus, thoracoscopic left upper lobectomy was performed.

Macroscopically, the tumor was gray-white in color, 30x25x20 mm in size, with a central cavitary lesion (Fig. 2A). Microscopic examination demonstrated a biphasic architecture, with glands surrounded by myoepithelial cells (Fig. 2B). The inner layers were composed of ductal cells with eosinophilic cytoplasm, whereas the outer layers were composed of

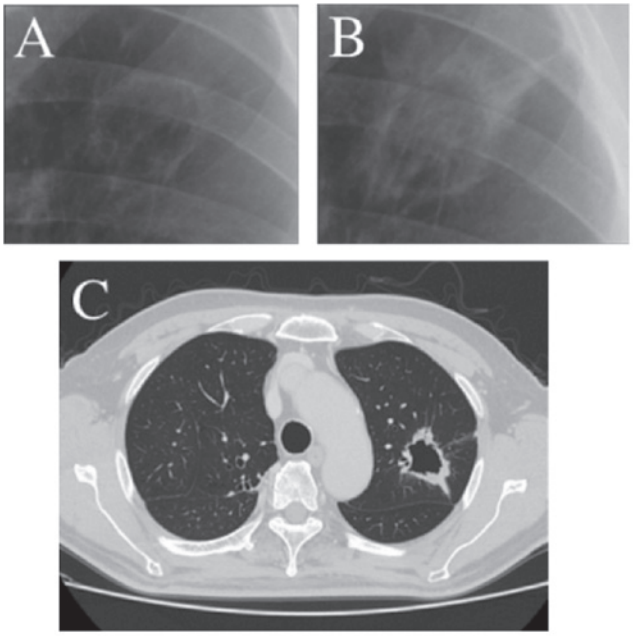

Figure 1. Chest X-ray findings. (A and B) The wall of the cystic lesion in the left lung upper field (B) was thicker compared with that 3 years earlier (A). (C) Computed tomography scan of the lesion. A solid, irregularly marginated tumor surrounding a cavitary lesion was identified in the upper lobe of the left lung. The greatest diameter of the tumor was 35 mm and the thickness of the cavity wall was 9 mm.

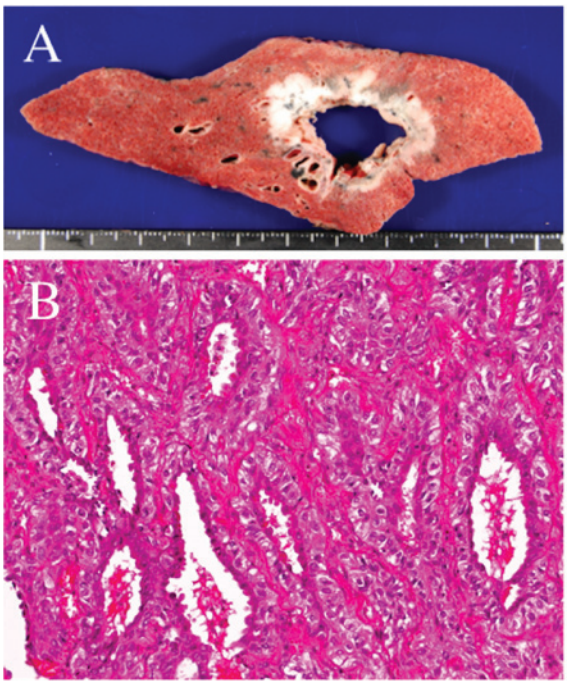

Figure 2. (A) Cut surface of the surgical specimen. The gray-white tumor measured 30x25x20 mm, with irregularly shaped edges surrounding the central cavity. (B) Microscopic findings (hematoxylin and eosin staining; magnification, x400). The tumor was composed of two types of cells: The inner glandular layer with eosinophilic cytoplasm; and the outer myoepithelial layer with clear cytoplasm. The structure was surrounded by the thickened basement membrane.

myoepithelial cells with clear cytoplasm. The pathological diagnosis was EMC. The dissected hilar and mediastinal lymph nodes were free of metastatic disease. The postoperative course was uneventful. Although the patient received no adjuvant therapy and the EMC did not recur; however, rectal cancer was subsequently diagnosed and the patient succumbed to mortality in February, 2018.

Discussion

EMC is known as a salivary gland-type tumor and displays a typical biphasic pattern: A central ductular structure surrounded by clear cells of myoepithelial origin. EMC accounts for ~1% of all salivary gland tumors (1,2). Despite its predilection for the parotid gland, EMC also arises in other locations, such as the minor salivary glands or the upper and lower respiratory tract, albeit less often (3). EMC of the salivary gland is considered to originate from the intercalated duct (4). Pulmonary EMC is considered to originate from the ductal structure of the bronchial gland, which is one of the pulmonary counterparts to the intercalated duct (5). The salivary gland-type tumor of the lung accounts for 0.1% of all primary lung carcinomas (6), among which mucoepidermoid carcinoma is the most frequently observed histological subtype, adenoid cystic carcinoma is the second, and EMC is the third. The frequency of each subtype is reported to be 70, 23 and 7%, respectively (7,8).

EMC tends to be located in the central region of the lung, rather than the periphery (8). Almost all the bronchial glands are located in the central airway (9), which may explain the tumor's propensity to arise in central lung regions. The shape of EMC is mostly round to oval or lobulated (8). The tumor in the present case was located in the peripheral region of the lung, and appeared as a lobulated mass accompanied by a cavitary lesion. To the best of our knowledge, a case of EMC with a cavitary lesion has never been reported to date.

The reported mechanisms of cavitary formation are as follows: i) Central necrosis due to the rapid tumor growth, with nutritional needs exceeding the blood supply; ii) bronchial or alveolar expansion as a result of the ectatic changes of the peripheral part following tumor invasion to more central parts; and iii) infectious diseases, such as abscesses, fungal infections and tuberculosis (10). In the present case, however, the microscopic findings revealed neither necrotic tissues nor expanding bronchi or alveoli in the inner part of the tumor. The bacteriological examination revealed no signs of infection. Therefore, it was hypothesized that a ductal structure of the bronchial gland in the primary cystic lesion that had been detected 3 years earlier was the site of origin of the EMC and grew to become a cavitary lesion.

We herein described a rare case of primary EMC of the lung with a cavitary lesion. Although the mechanism of the cavity formation remains to be elucidated, EMCs of the lung can include cavitary lesions.

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Availability of data and materials

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Authors' contributions

MM and MY conceived and designed this case report. SA collected and interpreted the data. MM and TH wrote the initial draft of the report. TH, RN and FT critically reviewed the manuscript. The final version of the manuscript has been read and approved by all authors.

Ethics approval and consent to participate

Not applicable.

Patient consent for publication

The patient provided consent to the publication of the case details and associated images.

Competing interests

The authors declare that they have no competing interests to disclose.

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