Case report

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Primary adenoid cystic carcinoma in the peripheral lung: a cytological, histopathological and immunohistochemical report of two cases

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Abstract

Primary adenoid cystic carcinoma (ACC) of the peripheral lung is a rare entity. Here we report two cases of primary ACC. Case 1 is an 84-year-old male with a past-medical history of cecal cancer presented with a 10 mm left upper lung nodule. Case 2 is a 40-year-old female who presented with 30 mm right upper lobe. Intra-operative (Case 1) and pre-operative (Case 2) histopathologic and cytologic diagnoses were consistent with a primary peripheral lung ACC. An upper lobectomy±mediastinal lymph node dissection was performed and immunohistochemical staining with thyroid transcription factor (TTF)-1, c-KIT and MYB on the excision specimen confirmed our diagnosis.

Keywords: Adenoid cystic carcinoma, lung, cytology, immunohistochemistry, case reports

Introduction

Adenoid cystic carcinoma (ACC) is a biphasic tumor consisting of epithelial and myoepithelial cells. ACC is a rare, slow-growing low-grade malignancy which usually arises within salivary glands and tends to demonstrate local recurrence and frequent distant metastasis.

ACC also develops in several sites other than the salivary gland. Primary lung ACC is extremely rare and accounts for approximately 0.04%-0.2% of all lung cancers [1,2]. Primary lung ACC is categorized as a salivary-type lung cancer which mostly occurs in the trachea, carina, or in a main stem bronchus (70.7%) [3]. An even rarer event is primary ACC in the peripheral lung within the parenchyma [4]. Only a few reports have so far described the cytologic findings of peripheral lung ACC [4,5].

We recently diagnosed two cases of this rare malignancy via intra-operative frozen section and touch imprint in Case 1 and pre-operatively via biopsy and brush cytology (Case 2). Our diagnosis was confirmed by immunohistochemical stains.

Case presentation

Case 1

An 84-year-old Japanese male patient was admitted to our hospital for evaluation of an incidental left upper nodule. The patient had undergone surgery for cecal adenocarcinoma 33 month prior to this admission and the nodule was discovered on a post-operative chest computerized tomography (CT) examination (Figure 1). Although there were no abnormal positron emission tomography (PET) findings, the nodule size had increased over the past 3 years from 4 mm to 10 mm. Clinically, a metastasis from his colon was suspected and therefore, surgical treatment was offered.

Past medical history was significant for a 1300 smoking index. The patient was otherwise asymptomatic on admission and had no co-existing diseases. His vital signs, physical examination, and laboratory findings, including serum tumor markers [CEA, 1.9 ng/ml (<5 ng/ml in normal); and ProGRP, 69.8 pg/ml (≤80 pg/ml in normal)] were all within normal limits. The

patient underwent video-assisted thoracoscopic surgery to perform a partial resection of the left lung (**Figure 2**). There were no regional or mediastinal lymph node enlargements to suggest metastasis. Intraoperative frozen section (**Figure 3a**) and cytologic examination (**Figure 4a**) with touch imprint smears was performed and the diagnosis of peripheral lung ACC was made. A complete upper lobectomy was performed.

Figure 1. CT examination of Case 1 shows a round shape and a well circumscribed mass (arrow) in the left upper lobe without any lymph node swelling.

Figure 2. A close-up photo of the whole mount section that contains a resected tumor (11x10x10 mm) stained with H&E in the left upper lobe (Case 1). Note: a tumor is present beneath the pleura. A whole mount section shows a cellular and solid tumor with well-defined demarcation. It is adjacent to the visceral pleura devoid of any bronchial transition. H&E stain, whole mount section, scale bar, 5 mm.

Figure 3. Histopathology of the resected tumor (Case 1). Frozen section (a) during the operation and permanent section (b) show the typical pathological features of ACC. Both show cribriform patterns of tumor cells. H&E stain, scale bars, (a) 500 μ m and (b) 100 μ m.

The patient was considered to have been stable on follow-up for 36 months.

Case 2

A 40-year-old Japanese woman was referred to our hospital for evaluation of a right upper lobe lung mass, which had been found incidentally on chest X-ray examination for symptoms of an acute upper respiratory infection. She did not present with any symptoms related to bronchial obstruction and had no previous medical problems. The patient had no history of smoking or recent weight loss. Her initial laboratory values, including tumor markers [CEA 0.6 ng/ml; CYFRA, 1.3 ng/ml (<3.5 ng/ml in normal)] were unremarkable, and the findings of a pulmonary function test were normal. A CT scan of the chest revealed a 30 mm, relatively well-defined mass near B3a in her right upper lobe (**Figure 5**).

We diagnosed the tumor to be peripheral lung ACC via percutaneous transthoracic fine needle aspiration cytology

Figure 4. Imprint smears (a and b) of the resected tumor (Case 1). Moderately cellular smears consist of various sized epithelial cell clusters in the bloody (a) and clean (b) backgrounds (Papanicolaou stain, scale bars, 100 μ m). Based on the cytological findings combined with those of histopathology, we diagnosed the patient to have lung peripheral ACC during the surgery.

(FNAC, **Figures 6a-6d**) and CT-guided biopsy (**Figures 7a and 7b**). The patient underwent right upper lobectomy (**Figure 8**) with mediastinal lymph nodes dissection. The diagnosis of primary peripheral ACC was confirmed histologically. The lymph nodes were negative for metastatic disease. The postoperative course of the patient was uneventful and she is currently doing well and is being followed up on an outpatient basis.

Macroscopic findings

Case 1

Macroscopically, the tumor was located in the periphery of the lung parenchyma and it was not related to the main bronchus (**Figure 2**). The tumor was well-circumscribed and measured 11x10x10 mm. The cut surface of the tumor was homogeneously white. After touch imprint slides were made and fixed in 95% ethanol for Papanicolaou staining, sections of the resected lung specimen were fixed in 10% buffered

Figure 5. Chest CT shows a 30 mm-sized, relatively well-defined mass (arrow) near the B3a in her right upper lobe (Case 2).

Figure 6. FNAC smears stained by the Papanicolaou method (Case 2) are in the high power view. (a): Moderately cellular, mostly consisting of tightly cohesive aggregates of relatively small and uniform tumor cells on a clean background. (b-d): There are microfollicle-like structures within the tumor cell clusters resulting in a cribriform appearance. These findings suggest the presence of lung ACC. Papanicolaou stain, scale bars, 100 μ m.

formalin for the histopathologic diagnosis. The specimen was embedded in paraffin wax, sectioned and stained with hematoxylin-eosin (H&E), Alcian blue, and periodic acid Schiff (PAS). In addition, immunohistochemistry (**Figures 9a-9n**) was performed using several antibodies (Ventana/Roche, Tokyo

Figure 7. H&E stained biopsy specimens (Case 2) show neoplastic cells in a solid pattern (a) and a cribriform arrangement (b). H&E stain, scale bars, 100 μ m.

Japan), including AE1/AE3, α -smooth muscle actin (SMA), MIB-1, MYB, p63, TTF-1 and c-KIT.

Case 2

Examination of the right upper lobectomy specimen showed a 30x24x22 mm well-circumscribed, lobulated, white mass (**Figure 8**) in the periphery of the lung without any connection to the bronchial tree or submucosal extension. The mass was located 7 mm away from the bronchial resection margin. There was no pneumonic infiltration in the surrounding lung parenchyma, and the surgical margin was free from the tumor. Immunohistochemistry (**Figures 9a-9n**) using the antibodies used in Case 1 was performed to confirm the diagnosis.

Microscopic findings

Case 1

Microscopically, two small bronchi were observed at the pe-

Figure 8. Macroscopic photos of the resected tumor in the right upper lobe (Case 2). Note: a lobulated tumor (30x24x22 mm) is present near the B3a.

riphery of the tumor (**Figure 2**). The tumor consisted of two cell types, epithelial and myoepithelioid cells, with predominantly the latter cells (**Figures 3a** and **3b**). They were relatively small and uniform with round or ovoid nuclei. They formed strands or clumps with either cystic or alveolar spaces forming an interlacing cylinder and/or cribriform pattern (**Figures 3a** and **3b**). The center of the cylinder or tubule was positively stained with PAS and Alcian blue.

Case 2

Tubular and solid patterns were also noted in the tumor tissue. Biopsy specimens showed epithelial and myoepithelioid neoplastic cells arranged in a solid pattern (**Figure 7a**). Myoepithelioid tumor cells were predominately observed (**Figures 7a** and **7b**). In the lobectomy specimen, a cribriform architecture was also seen with hyaline material within the lumina (**Figure 7b**). The hyaline material stained positive for Alcian blue. The tumor cells themselves were small to medium in size with slight atypia. All submitted mediastinal lymph nodes were negative for carcinoma.

Figure 9. Immunohistochemistry of the tumor tissue specimens in Case 1 (a, c, e, g, i, k and m) and Case 2 (b, d, f, h, j, l and n). The neoplastic cells were positive for AE1/AE3 (a and b), α -SMA (c and d), p63 (e and f), c-KIT (g and h), MYB (i and j), and TTF-1 (k and l). MIB-1 (m and n). Note: the epithelial tumor cells are positive for AE1/AE3, c-KIT, and TTF-1, while the myoepithelial cells are positive for α -SMA, p63 and MYB. The MIB-1 positive rates are 60% in Case 1 and 40% in Case 2, suggesting that the biological aggressiveness of Case 1 is greater than that of Case 2.

Cytological findings

Case 1

The touch imprint slides showed tumor cells in ball-like (Figure 4b). The cells have nuclei with fine granular chromatin and prominent nucleoli. Variably-sized central cores of homogeneous material were seen filling the cystic spaces.

Case 2

Papanicolaou-stained FNAC smears were moderately cellular, and consisted mostly of tightly cohesive aggregates of relatively small, uniform tumor cells (Figures 6a-6d). The cells were arranged in three-dimensional, tubular and spherical configurations. The tumor cells had uniform ovoid nuclei with increased nuclear to cytoplasmic ratios, and finely granular chromatin. Neoplastic clusters included acellular spheres of dense, homogeneous material that was lightly stained with Papanicolaou stain.

Immunohistochemical studies

The immunohistochemical studies from Case 1 and Case 2 were similar. Tumor cells stained positive for AE1/AE3 (Figures 9a and 9b), α -SMA (Figures 9c and 9d), p63 (Figures 9e and 9f), c-KIT (Figures 9g and 9h), MYB (Figures 9i and 9j) and TTF-1 (Figures 9k and 9l). Case 1 had a MIB-1 (Figures 9m and 9n) index of 60% and Case 2 had a MIB-1 index of 40%.

Discussion

ACC mainly develops in the salivary glands and is rarely in

other tissues. Primary pulmonary ACC most often develops in the lower trachea or mainstem bronchus [6] and accounts for approximately 0.2% of all cases of primary lung cancer [7]. Peripheral lung ACC is even more rare, accounting for approximately 10% of primary pulmonary ACC [8]. Bronchial glands are present from the 1st to the 4th order bronchi and are found in 79% of the 5th order bronchi, and in only 11% of the 6th order bronchi [9]. Therefore, this tumor is most commonly seen in the extra-pulmonary bronchi. Tumor arising in the intra-pulmonary bronchi is extremely rare [10-12]. We present two cases which were located in the peripheral lobe adjacent to the visceral pleura without bronchial tree involvement or submucosal extension.

In a review by Yokouchi et al., have summarized the clinical features of primary peripheral lung ACC reported in the English-language literatures [13]. Primary peripheral lung ACC is more frequently seen in males than females. The average age was 58 years and at presentation, patients are frequently asymptomatic, as was found in our cases. Most of the reported ACC cases have been accurately diagnosed and treated surgically, with favorable clinical outcomes. Only two patients had recurrence after surgery [11,14]. Because primary lung ACC is refractory to radiotherapy and chemotherapy, alternative strategies using specific ACC-targeted drugs are warranted. In this context, phase II clinical trials using imatinib mesylate, a c-KIT tyrosine kinase inhibitor, against salivary ACC have been conducted [15,16] based on the evidence that this tumor

expresses high levels of c-KIT protein [17-19], as shown in our cases. Salivary ACC is highly responsive to imatinib [16] and mutations of exons 9 or 11 are reported to be more important than the c-KIT protein level for the imatinib response [19,20].

On histology, three growth patterns of salivary gland ACC are seen: cribriform (cylindromatous), tubular, and solid patterns [21]. The most common type is the cribriform pattern, which is also called "classic ACC". The immunohistochemical profile is also characterized by the proliferation of myoepithelial (α -SMA and p63) and ductal cells (AE1/AE3), as was observed in our cases. Our cases were positive for MYB and c-KIT, consistent with ACC. Tumor cells were also positive for TTF-1, indicating that the origin was the lung [1,22,23]. In Case 1, the patient had a history of adenocarcinoma of the colon posing an additional diagnostic challenge. Distinguishing primary and metastatic lung ACC based on cytomorphology or histology can be difficult [6]. The lung ACC cells in the present study expressed TTF-1. TTF-1 is a protein expressed in the forebrain, thyroid, and lung [24] and only a few reports [1,22,23] have described the expression of TTF-1 in primary lung ACC.

Few discussions on the cytologic findings of primary peripheral lung ACC have been reported in the literature [4,5,22]. The cytologic findings from imprint smears (Case 1) and percutaneous FNAC (Case 2) helped us to accurately diagnose the solitary and peripheral lung tumors. Bronchial brushings have been reported to be useful for the diagnosis of primary ACCs of the lung arising from the trachea and the main bronchus [25,26]. Similarly, bronchoscopy-guided FNAC may be used to diagnose primary lung ACC arising from the main bronchus [6,27]. Although primary peripheral ACC of the lung rarely occurs, a preoperative accurate diagnosis should be made in cases where appropriate cytologic smears can be obtained for clinicians to provide appropriate treatment for the patient. In our two cases, we were able to diagnose peripheral lung ACC both during and before the operation, because of the typical cytological features of ACC.

Patients with a predominantly tubular growth pattern have a better prognosis than those with a solid growth pattern [28]. Interestingly, the spontaneous regression of lung ACC has also been reported [29]. Although ACC arising from the trachea or bronchus is associated with a high incidence of submucosal extension, such extension has not been described in primary peripheral ACC [8]. Our patients are both currently doing well at 3-year follow-up in Case 1 and at 2-year follow-up in Case 2. Continued follow-up is required as the potential for high-grade transformation of ACC [30] exists.

As little is known regarding the molecular pathways underlying ACC, no specific targeted agents or chemotherapy are in current use. That tumor cells stain immunohistochemically positive for c-KIT and MYB, suggest a potential role of the transmembrane tyrosine kinase receptor, c-KIT and a proto-oncogene for which a fusion transcript, MYB may have in the development of ACC [31] and malignant transformation [32]. The MYB-NFIB fusion transcript has been reported to

be present in a specific subset of ACC and is related to MYB over-expression [33,34]. MYB immunostaining is confined to the myoepithelial cells [34] and this is seen in our cases, although such translocation was not found in our cases. These findings suggest that there are intact regulatory mechanisms in the neoplastic cells that can regulate the levels of the fusion protein. As c-KIT and MYB may play some role in the ACC pathogenesis, c-KIT and MYB may be good targets for future therapeutic management.

Conclusion

We herein described two patients with primary peripheral lung ACC. Although rare, ACC should be considered in the differential diagnosis of a primary lung tumor, even when present in the peripheral lung.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

Authors' contributions	FE	NW	MM	NK	AS	KN	TS1	TS2	TM	TN	TT
Research concept and design	✓	✓	--	--	--	--	--	✓	✓	✓	✓
Collection and/or assembly of data	✓	--	✓	✓	✓	✓	✓	✓	✓	--	--
Data analysis and interpretation	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Writing the article	✓	✓	--	--	--	--	--	--	--	--	✓
Critical revision of the article	✓	✓	--	--	--	--	--	--	--	✓	✓
Final approval of article	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Statistical analysis	--	--	--	--	--	--	--	--	--	--	--

Acknowledgement

This study was supported in part by Grants-in-Aid from the Ministry of Education, Science, Sports and Culture of Japan (No. 90157206).

Publication history

Editor: Jianyu Rao, UCLA Ronald Reagan Medical Center, USA.

EIC: Markus H. Frank, Harvard Medical School, USA.

Received: 11-Mar-2015 Final Revised: 11-Apr-2015

Accepted: 13-Apr-2015 Published: 17-Apr-2015

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Citation:

Etori F, Watanabe N, Matsuyama M, Kawaguchi N, Sekiya A, Nambu K, Sakakima T, Sawa T, Marui T, Naiki T and Tanaka T. **Primary adenoid cystic carcinoma in the peripheral lung: a cytological, histopathological and immunohistochemical report of two cases.** *Pathol Discov.* 2015; **3**:3.
http://dx.doi.org/10.7243/2052-7896-3-3