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Case Report

A Rare Case of Contralateral Ciliated Muconodular Papillary Tumor after Limited Surgery

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Abstract

Ciliated muconodular papillary tumor (CMPT) was a rare tumor with low-grade malignancy. The histopathogenesis of CMPT remains unclear. Three typical components are ciliated columnar cells, goblet cells, and basal cells. Limited resection is sufficient for CMPT. No recurrences or metastases have been reported. Herein, we reported a 73-year-old man who presented with right lower lobe partial sold nodule. He received partial resection for the right lower lobe tumor. CMPT was confirmed. However, contralateral left lower lobe ground-glass opacity (GGO) was found 3 months later. He received partial resection for the left lower lobe GGO. CMPT was diagnosed again. Double primary CMPTs were recognized because morphologic patterns were different. At his 2-year follow-up, there was no local recurrence or metastasis.

Keywords: Ciliated muconodular papillary tumor, lung tumor, surgery

Introduction

Ishikawa first described a rare ciliated papillary tumor of the lung in 2002.[1] This newly defined peripheral peribronchiolar lung tumor was termed ciliated muconodular papillary tumor (CMPT).[1-10] Typically, ciliated columnar cells, goblet cells (mucous cells), and basal cells are the characterized components with abundant intra-alveolar mucins.[2,3,5-9] However, the histopathogenesis of CMPT remains unclear.[2,6-8] Some genetic mutations or rearrangements were reported that indicate that CMPT is a neoplastic tumor instead of a reactive disease.[3-6] Most of the reported CMPT patients were East Asians, and only a few Western patients were reported.[2,3] Low-grade malignancy and benign clinical course after limited partial resection are current understandings in CMPT according to the few reported cases.[7-9] No recurrence after the operation was reported.[2-5,7-10] Herein, we first described one CMPT patient receiving partial resection where the contralateral lung

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CMPT was found 3 months later. This study was approved by the Institutional Review Board of Cheng Hsin General Hospital, and the approval number was (451)103A-28. Informed consent was waived due to the retrospective design.

Case Report

Our case

A 73-year-old Han Chinese man was a nonsmoker with a history of hypertension. He received a health examination at our hospital with nonspecific symptoms. One small 1.5 cm partial solid lung nodule in the right lower lobe was accidentally found by chest computed tomography (CT) on 2015/12/16 [Figure 1]. The nodule was persistently exhibited when following up with a chest CT on 2016/03/14. Tumor markers, carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA 19-9), were within the normal limits. He received video-assisted thoracoscopy surgery (VATS) right lower lobe partial resection and mediastinal lymph node dissection on 2016/05/09. CMPT was confirmed because of cuboidal to low columnar mucin-containing cells and mucous lakes. No mitotic figures were noted. Most of the lining mucous cells had a basal cell layer [Figure 2]. Mediastinal lymph nodes had no tumor cell invasion. Epidermal growth factor receptor (EGFR) mutation analysis was wild type. Anaplastic lymphoma kinase (ALK) immunostaining was also negative.

He was regularly followed up at our clinic. One new small ground-glass opacity (GGO) lesion had been noted in the left lower lobe superior segment by chest CT since 2016/08/08 [Figure 3a]. Chest CT on 2017/03/27 revealed the increasing size of the GGO lesion [Figure 3b]. Initially, malignancy was suspected, and resection was suggested according to the enlargement of the lesion. CEA and CA19-9 levels were within the normal limits. He underwent VATS left lower lobe partial resection on 2017/05/15. CMPT was confirmed again [Figure 4]. EGFR mutation analysis was still wild type. ALK immunostaining was negative as in the previous result. Since then, chest CT has been followed up every 3–4 months. For 2 years after the last surgery, he remained relapse free until now.

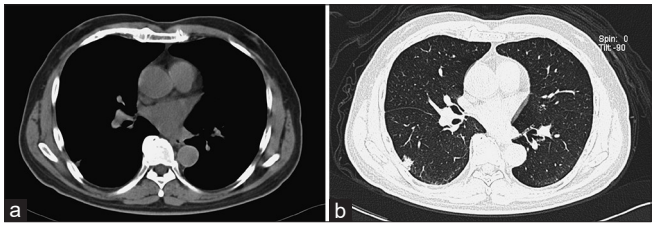

Figure 1: Computed tomography images of the chest. (a) The mediastinal window revealed one small partial solid nodule in the right lower lobe lung field. (b) Lung window revealed a 1.5 cm pulmonary infiltration patchy lesion in the right lower lobe lung field

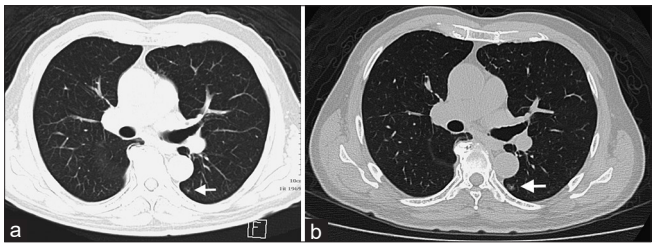

Figure 3: Lung window images of chest computed tomography. (a) One small ground-glass opacity over the left lower lobe superior segment. (b)Increasing ground-glass opacity was noted 7 months later

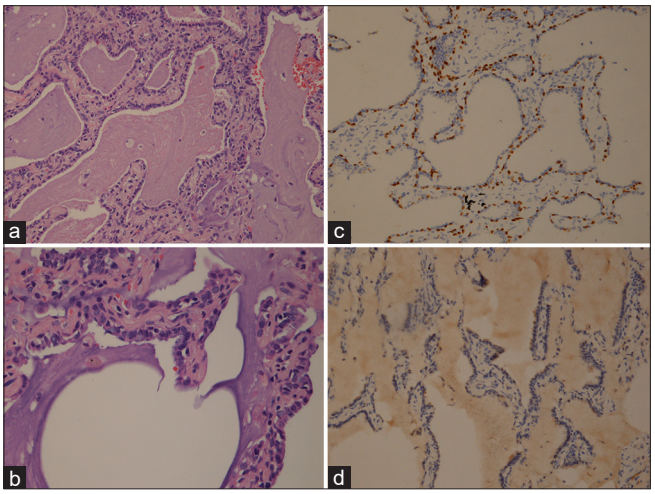

Figure 2: Immunohistochemical analyses of the right lower lobe ciliated muconodular papillary tumor. (a) The ciliated muconodular papillary tumor was composed of ciliated columnar cells, mucous cells, and basal cells by H and E, ×200. (b) Cuboidal to low columnar cells with cilia, mucin-containing lining cells, and mucous lakes by H and E, ×400. (c) Most lining mucous cells have a basal cell layer, which is highlighted by p40 immunostaining, ×200. (d) Cytoplasmic staining was positive in three components of ciliated columnar cells, mucous cells, and basal cells by BRAF V600E, ×200

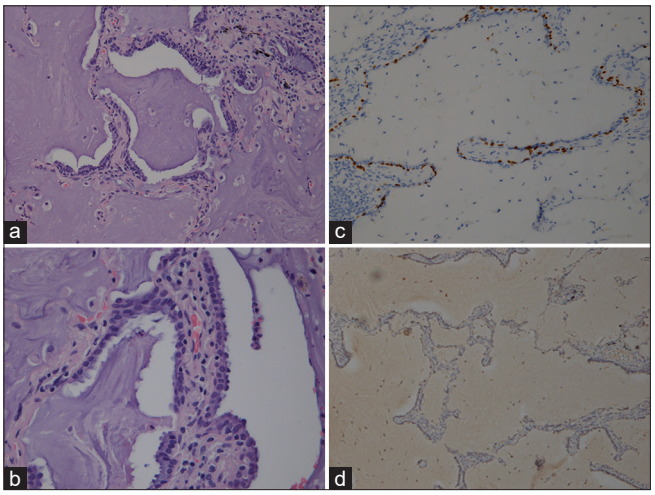

Figure 4: Immunohistochemical analyses of the left lower lobe ciliated muconodular papillary tumor. (a) The ciliated muconodular papillary tumor was composed of ciliated columnar cells, mucous cells, and basal cells by h and e staining, ×200. (b) Ciliated columnar cells, mucin-containing lining cells, and mucous lakes by H and E, ×400. (c) The basal cell layer was highlighted by p40 immunostaining, ×200. (d) Cytoplasmic stain was also positive in three components by BRAF V600E, ×200

Discussion

Conventionally, pulmonary tumor cells with cilia are benign lesions. These benign tumors are rare and localized to the central airway.[7,10] CMPTs have benign features of ciliated cells and the absence of mitotic figures.[2,4,5] However, CMPTs possessing ciliated columnar cells have recently been recognized as low-grade malignancies.[2,3,7] They develop in the peripheral lung field without symptoms.[3-5] Because the number of reported cases is extremely rare, the understanding of CMPTs is still unclear. CMPTs are composed of benign histopathological features, such as ciliated columnar cells, goblet cells, and basal cells.[2-9] However, driver mutations, including V-raf murine sarcoma viral oncogene homolog B (BRAF), EGFR, or ALK, were found in some reported CMPTs.[3-5] The malignant potential was recognized in CMPTs.

Partial resection or sublobar resection might be sufficient for CMPTs due to low-grade malignancy.[7-9] No recurrences or metastases have been reported.[2-5,7-9] Our patient was the first case in which contralateral CMPT occurred 3 months after partial resection of the first CMPT. Although EGFR and ALK were not mutated in either CMPT, we thought they might be double primary CMPTs. First, the image morphologic patterns were quite different. The first one was a partial solid nodule, but the second one was a pure GGO lesion. Second, no metastatic tumor cells were observed in mediastinal lymph nodes or intrapulmonary lymph nodes. Contralateral lung metastasis without mediastinal lymph node involvement is rare. Extrapulmonary metastasis was also absent in this patient. Third, no local recurrences were noted on either side after surgery. We believed that the recurrence or metastasis of CMPT was unlikely. He might be the first case with two primary CMPTs.

As for surgical management, lobectomy was performed in some previously reported cases.[2,3,5] However, partial resection or sublobar resection was performed in other reported cases without recurrences or metastases.[2-5,7-10] Our patient received partial resections for bilateral CMPTs. He regularly followed up by chest CT every 3–4 months. No local recurrences were observed for 3 years after the first surgery for the right lower lobe CMPT and 2 years after second surgery for the left lower lobe CMPT. According to our results, partial resection with a safety margin might be acceptable for double primary CMPTs.

Conclusion

CMPTs are rare low-grade malignant tumors. Partial resection or sublobar resection may be a sufficient treatment. No local recurrences or metastases have been reported. To the best of our knowledge, this patient was the first case in which the other contralateral CMPT was noted after partial resection of CMPT. However, double primary CMPTs may be considered due to different morphologic patterns. Partial resection or sublobar resection may still be acceptable in double primary CMPTs.

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Conflicts of interest

There are no conflicts of interest.

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