Received: 19 July 2024 | Accepted: 16 October 2024

DOI: 10.1002/rcr2.70055

CASE REPORT

Respirology Case Reports OPEN ACCESS APSR WILEY

Synchronous bilateral typical pulmonary carcinoid tumours diagnosed by robotic navigation bronchoscopy: A unique case

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Associate Editor: Panwen Tian

Abstract

Pulmonary carcinoids are uncommon malignant neoplasms, believed to derive from specialized neuroendocrine cells known as Kulchitsky cells. We evaluated a 69-year-old female presenting symptoms consistent with carcinoid syndrome, such as intermittent flushing and diarrhoea, along with complaints of shortness of breath and cough. Imaging revealed bilateral lung nodules, confirmed by biopsy to be carcinoid tumours. The treatment of choice for carcinoid tumours is complete surgical resection. Nonetheless, individualized management plans are crafted based on the tumour's location and the patient's respiratory function as these present challenges to anatomical resection of tumours.

KEYWORDS
bronchoscopy and interventional techniques, histology/cytology, lung cancer, rare lung diseases, thoracic surgery

INTRODUCTION

Pulmonary carcinoids are uncommon malignant neoplasms, believed to derive from specialized neuroendocrine cells known as Kulchitsky cells.[1] They account for 1%–2% of all lung neoplasms,[2] and of all carcinoid tumours, approximately 25% are located in the respiratory tract.[1] Synchronous tumours are pathologically and histologically different tumours that exist at the same time. Diagnosis will pose a challenge as the presence of multiple tumours at the same time can be mistaken to be metastatic lesions[3] Herein we present a rare case of synchronous bilateral typical pulmonary carcinoid tumours in a 69-year-old female.

CASE REPORT

A 69-year-old female, with no smoking habits and a medical history significant for controlled hypertension treated by hydrochlorothiazide, visited our clinic for a follow-up after being discharged from the hospital due to complaints of shortness of breath that was diagnosed as an asthma exacerbation. During her hospital stay, incidental nodules were detected on chest imaging. She was discharged on a combination of inhaled corticosteroids and beta-agonists. Before her hospitalization, she reported intermittent cough with whitish sputum and progressively worsening shortness of breath over 6 months, denying symptoms like hemoptysis, fever, night sweats, or weight loss. The patient also disclosed experiencing symptoms of intermittent flushing and loose stools. On auscultation, expiratory wheezes were noted with no additional significant findings on physical examination. Upon reviewing the patient's CT scans, we noted bilateral pulmonary nodules, with the left perihilar nodule in the left upper lobe measuring 2.3 × 2.4 cm and the right middle lobe perihilar nodule measuring 2.8 × 1.7 cm (Figure 1). Robotic-assisted navigation bronchoscopy was performed to establish a diagnosis, and pathology results from both nodules indicated a well-differentiated neuroendocrine tumour consistent with a typical carcinoid tumour. Overall, the findings are consistent with a well-differentiated neuroendocrine neoplasm. No significant increase in mitosis (<2 mitoses/2 mm^2) is seen. No necrosis is seen. Differential diagnoses include carcinoid and atypical carcinoid. Given the bland morphology and shallow proliferative index, this likely represents a typical carcinoid tumour (Figure 2). No mediastinal lymphadenopathy was noted. The patient also did PET scans which showed middle lobe (1.3 × 2.8 cm; mean SUV of 2.5) and left upper lobe (2.2 × 2.2 cm; mean SUV of 4.8) lung nodules. No adenopathy or abnormal FDG uptake of skeletal structures, pelvic sidewalls, abdominal organs, inguinal, mesenteric, or para-aortic lymph

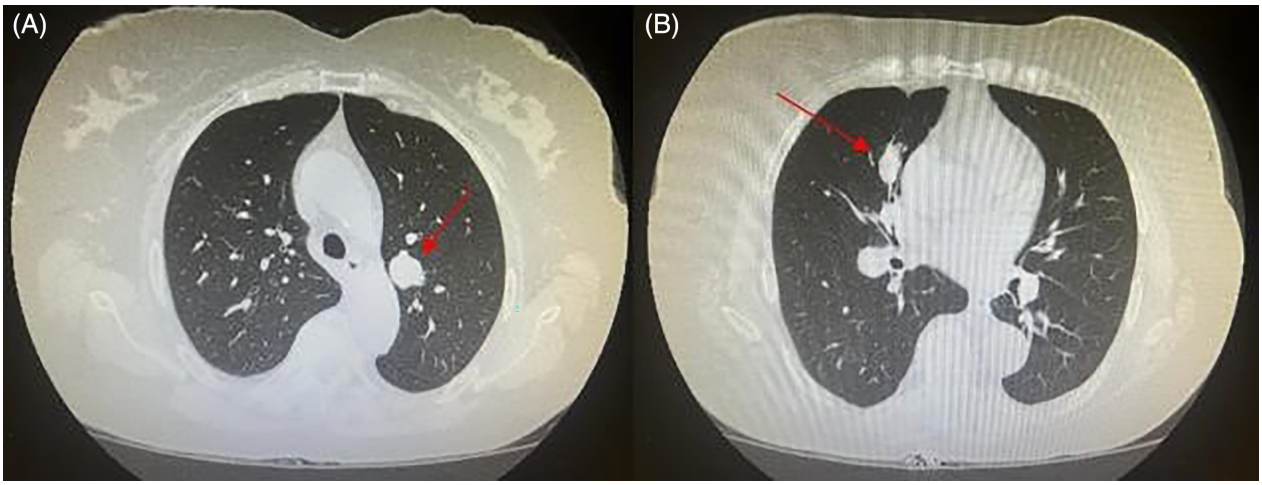

FIGURE 1 (A) Left perihilar nodule in the left upper lobe measuring 2.3 × 2.4 cm, the nodule has well defined margins. (B) Right middle lobe perihilar nodule measuring 2.8 × 1.7 cm.

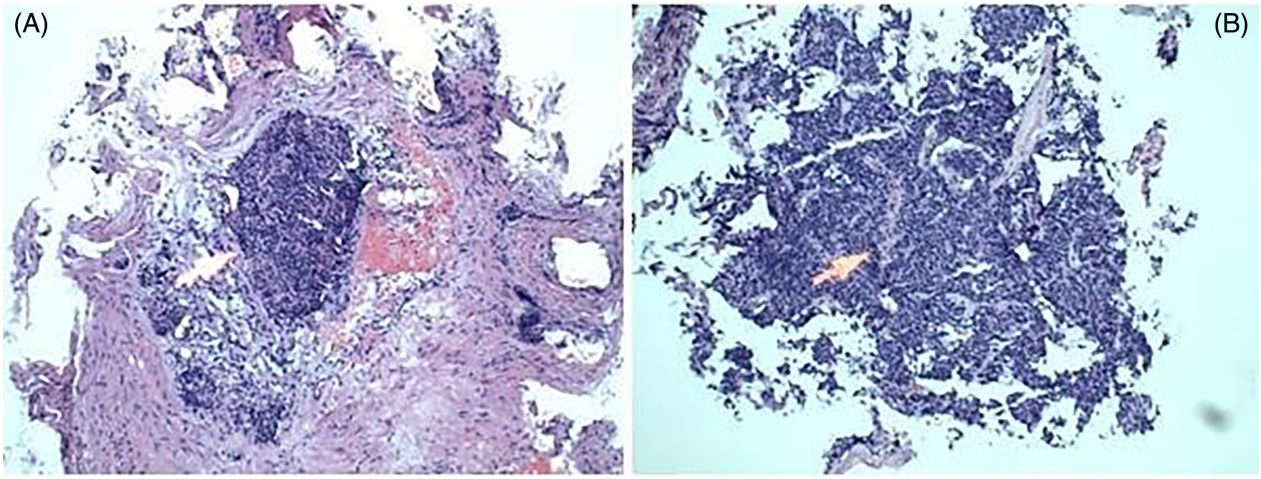

FIGURE 2 (A) Biopsy from left upper lobe lung tumour showing well-differentiated neuroendocrine tumour, consistent with carcinoid tumour, no significant increase in mitosis. (B) Biopsy from right middle lobe lung tumour showing well-differentiated neuroendocrine tumour, consistent with carcinoid tumour, no significant increase in mitosis (<2 mitoses/2mm^2).

nodes. Our patient was sent to cardiothoracic surgery for possible resection.

DISCUSSION

Carcinoid tumours originate from enterochromaffin cells dispersed throughout the gastrointestinal and bronchopulmonary systems, characterized by their slow growth as neuroendocrine tumours.[4] Pulmonary carcinoids are uncommon malignant tumours and account for 1%–2% of all pulmonary malignancies.[1] These tumours have been traditionally classified as typical or atypical carcinoids based on histologic features, with the former exhibiting a more favourable prognosis than the latter.[5] 25% of people with lung carcinoid tumours may be asymptomatic.[6] However, some patients may present with symptoms such as recurrent pneumonia, coughing, hemoptysis (coughing up blood), and chest pain.[4] As pulmonary carcinoid tumours are essentially carcinoid tumours, certain patients as seen in our case, may display symptoms characteristic of carcinoid syndrome including flushing of the face, diarrhoea, and asthma-like symptoms.[6] Patients with central tumours are more likely to be symptomatic than those with peripheral lesions.[1] Despite sharing the same original cells as other neuroendocrine tumours like small-cell lung cancer and large-cell neuroendocrine carcinoma, typical carcinoid tumours have an excellent prognosis.[2] Typical carcinoids have an 87%–100% 5-year survival, whereas atypical carcinoids have a 40%–83% 5-year survival.[1] Diagnosis of carcinoid tumours may be challenging as they do not have specific radiological signs and therefore could be mistaken as benign lesions or multiple metastases.[2] Both chest x-rays and CT scans commonly reveal well-defined, round, and occasionally slightly lobulated masses in instances of lung carcinoids.[6] Carcinoids are classically

located as nodules close to the hilus or perihilar as seen in our case, and rarely they present peripherally and exceptionally multifocal.[2] Bronchoscopic evaluation provides valuable information for surgical preparation and definitive diagnosis, as transbronchial biopsies are typically diagnostic[1] Carcinoid tumours exhibit growth patterns indicative of neuroendocrine differentiation. The most frequently observed patterns include organoid and trabecular arrangements. Additionally, other patterns such as rosette formation, papillary growth, pseudo glandular growth, and follicular growth may also be present.[6] Positron emission tomography scans generally demonstrate low-level uptake of fluorodeoxyglucose in carcinoid tumours.[1] This technique is useful for distinguishing carcinoids from high-grade neuroendocrine tumours like small-cell or large-cell neuroendocrine tumours.[6]

The treatment of choice for carcinoid tumours is complete surgical resection.[7] The goal of resection is complete excision of the tumour while sparing as much lung tissue as possible. Recent studies have shown that, for peripheral typical carcinoids, it is possible to perform conservative resection with preservation of much of the lung parenchyma, and bronchial resection can be performed for more centrally located tumours.[8] For central tumours, anatomical resection is not possible, and a limited resection, together with a lymphadenectomy may be preferred.[1] Tailor suit management plans are made on a case-to-case basis depending on the location of the tumour as well as respiratory function. Yazıcıoğlu et al. performed wedge resection, and a bilateral lymphadenectomy rather than anatomical resection to their case which has insufficient respiratory function and presented with a nonproductive cough secondary to synchronous bilateral multiple typical pulmonary carcinoid tumours.[1] Yazici et al. also avoided anatomical resection which they acknowledged could heighten the risk for recurrence as nodules were multiple and bilateral.[1] Beshay et al. however, performed right upper lobectomy and left lower lobectomy in staged procedures on their 73-year-old woman with bilateral pulmonary nodules that proved to be carcinoid.[2] Spaggiari et al. have also performed two staged (1 month) thoracotomies radical lymph node dissection to their patient with synchronous bilateral lung carcinoid tumours.[9] Despite being the gold standard of treatment, minimizing the risk of local recurrence, anatomical resection is not suitable for every patient and management must be made on a case to case basis. A careful preoperative evaluation of patients is crucial. Brownlee and his colleagues demonstrated excellent diagnostic yield, safety, and feasibility data even with subcentimeter nodules in the periphery of the lung with the use of robotic bronchoscopy allowing for accurate visualization and biopsies, which therefore provides high chances of accurate diagnosis.

In conclusion, multiple lung nodules on radiological investigations typically indicate but are not mutually exclusive to metastases as synchronous bilateral carcinoid tumours should be kept in mind.[1] In typical carcinoid tumours, especially bilateral cases, as in our patient, anatomical resection may be challenging, and a limited resection, together with a lymphadenectomy may be preferred as ipsilateral lobectomy could result in a significant decline of lung capacity.[1] Therefore, staged resection of tumours could be warranted with quantitative and qualitative lung capacity assessment before and following the initial resection to assess the patient's suitability for surgery.

CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

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How to cite this article: Megahed M, Sharif R, Abdeen Y. Synchronous bilateral typical pulmonary carcinoid tumours diagnosed by robotic navigation bronchoscopy: A unique case. Respirology Case Reports. 2024;12(11):e70055. https://doi.org/10.1002/rcr2.70055