

Superior mediastinal typical carcinoid detected by ^{99m}Tc -MIBI SPECT/CT imaging

A case report

Chenghui Lu, MD*, Zenghua Wang, PhD, Guoqiang Wang, MD, Xufu Wang, PhD, Xinfeng Liu, MD

Abstract

Rationale: This is a rare case of mediastinal typical carcinoid tumor with elevated parathyroid hormone (PTH) and hypercalcemia detected by ^{99m}Tc -methoxy-isobutyl-isonitrile (^{99m}Tc -MIBI) single photon emission computed tomography/computed tomography (SPECT/CT) imaging.

Patient concerns: A 65-year-old male patient presented with hypercalcemia and elevated PTH.

Diagnoses: The preoperative diagnosis was ectopic parathyroid adenoma in the right superior mediastinum.

Interventions: We performed endoscopic removal of the mass in the superior mediastinum.

Outcomes: Histopathology and immunohistochemistry revealed typical carcinoid.

Lessons: The differential diagnosis of mediastinal tumors with elevated PTH detected by ^{99m}Tc -MIBI dual-phase imaging should include ectopic parathyroid adenoma as well as carcinoid.

Abbreviations: 5-HT = 5-hydroxytryptamine, ^{99m}Tc -MIBI = ^{99m}Tc -methoxy-isobutyl-isonitrile, ACTH = adrenocorticotropic hormone, APUD = amine precursor uptake decarboxylation, CgA = chromograninA, CT = computed tomography, iPTH = intact parathyroid hormone, PTH = parathyroid hormone, SPECT/CT = single photon emission computed tomography/computed tomography.

Keywords: carcinoid, mediastinum, parathyroid hormone, scintigraphy

1. Introduction

Carcinoid is a rare well-differentiated tumor that originates from amine precursor uptake decarboxylation (APUD) cell system.^[1,2] Typical and atypical carcinoid tumors are distinguished on the basis of morphology, mitosis count, and the presence or absence of necrosis.^[3,4] Typical carcinoid has a less aggressive behavior; it rarely relapses after complete surgical resection, and nodal involvement and distant metastases are rare.^[5,6] Carcinoids occur most frequently (90% of cases) in the gastrointestinal tract.^[6] The second most common location is the respiratory tract (Figs. 1 and 2).

Other anatomic locations are extremely rare.^[7,8] There were less reports about mediastinal carcinoid.^[9] Here, we report a rare

case of mediastinal typical carcinoid tumor in a patient with elevated parathyroid hormone (PTH) and hypercalcemia.

2. Case report

A 65-year-old man complained about increased foam in urine. He had been diagnosed with chronic renal insufficiency for more than 5 months. Laboratory tests showed that his blood urea nitrogen (16.8 mmol/L; reference range, 3.6–9.5 mmol/L), serum creatinine (283.1 $\mu\text{mol/L}$; reference range, 31–133 $\mu\text{mol/L}$), serum calcium (3.61 mmol/L; reference range, 2.11–2.52 mmol/L), intact PTH (iPTH 1780 ng/L; reference range, 15.00–65.00 ng/L), serum alkaline phosphatase (276.9 IU/L; reference range, 45–125 IU/L) concentrations were all elevated. The primary diagnosis after subsequent radionuclide examination was ectopic parathyroid adenoma. Surgical excision was performed, and the pathology showed typical carcinoid. His intact PTH went down to 230.6 ng/L 4 weeks after operation. And serum calcium declined to normal (2.13 mmol/L).

The ethics committee of the Affiliated hospital of Qingdao University did not require ethical approval for case report. Written informed consent was obtained from the patient for the publication of this case report.

3. Discussion

We report a rare case of mediastinal typical carcinoid tumor in a patient with elevated PTH and hypercalcemia. The mechanism of the elevation of PTH and hypercalcemia may be as follows: As carcinoid tumors originate in the cells of APUD system, they can express different peptides and biogenic amines,^[1] such as 5-hydroxytryptamine (5-HT), bradykinin, adrenocorticotropic

Editor: Orazio Schillaci.

The authors report no conflicts of interest.

Department of Nuclear Medicine, The Affiliated Hospital of Qingdao University, Qingdao, Shandong, China.

* Correspondence: Chenghui Lu, Department of Nuclear Medicine, The Affiliated Hospital of Qingdao University, Qingdao, Shandong 266000, P.R. China (e-mail: luchenghui_1988@126.com).

Copyright © 2017 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial License 4.0 (CCBY-NC), where it is permissible to download, share, remix, transform, and buildup the work provided it is properly cited. The work cannot be used commercially without permission from the journal.

Medicine (2017) 96:52(e9457)

Received: 16 July 2017 / Received in final form: 5 December 2017 / Accepted: 6 December 2017

<http://dx.doi.org/10.1097/MD.00000000000009457>

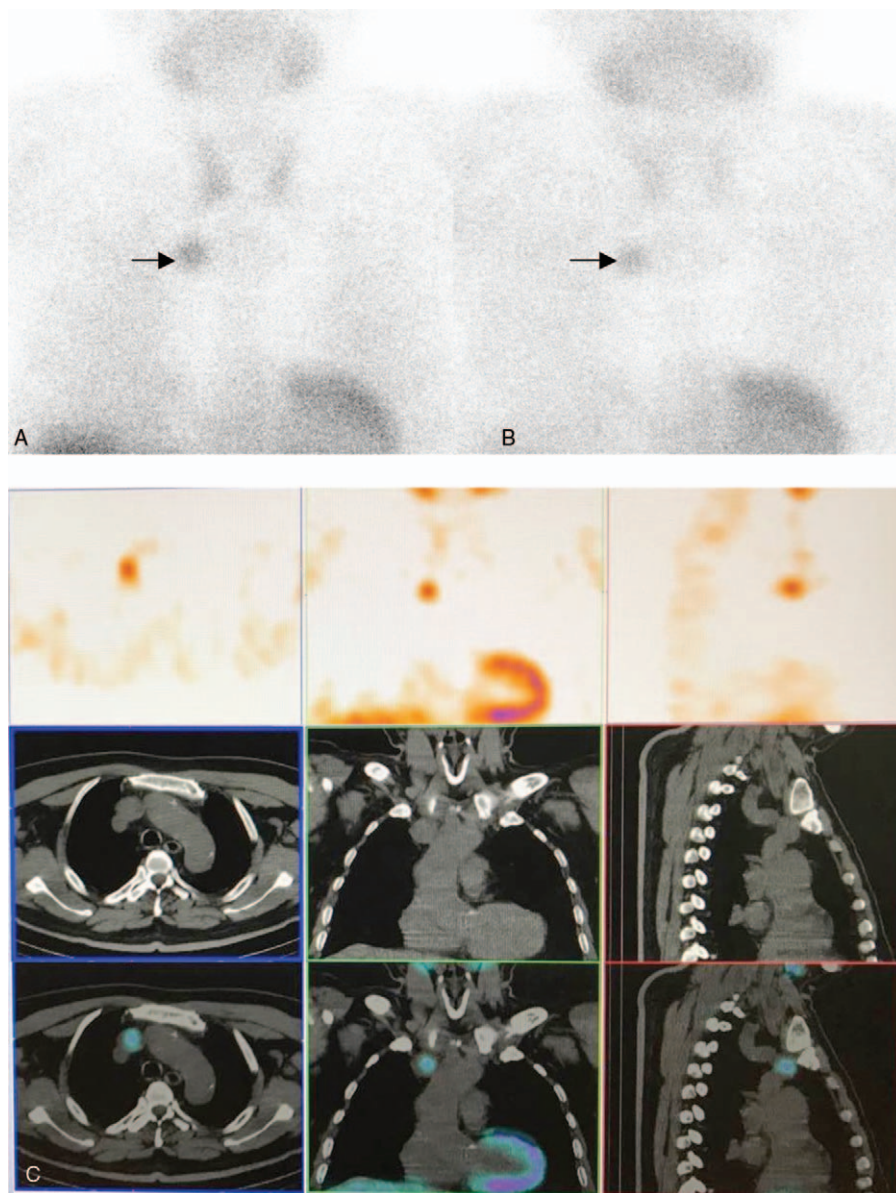


Figure 1. The scintigram on the early image (15 min) showed an increased focal accumulation of the radiotracer in the right superior mediastinum (black arrow) (A). Scintigram on the delayed image (120 min) revealed an abnormal focus of radiotracer uptake in the right superior mediastinum (black arrow) (B). SPECT/CT further demonstrated a soft tissue mass with increased radiotracer uptake in the right superior mediastinum (C). Enhanced computed tomography(CT) scan showed an oval heterogeneous mass (2.6 cm × 2.5 cm × 1.8 cm) with mild enhancement in superior mediastinum between left and right brachiocephalic vein. A solid nodule was detected in operation. It had close relationship with adjacent vessels, soft, brown in the cut surface, and measured 3.5 cm × 2.5 cm × 1.0 cm.

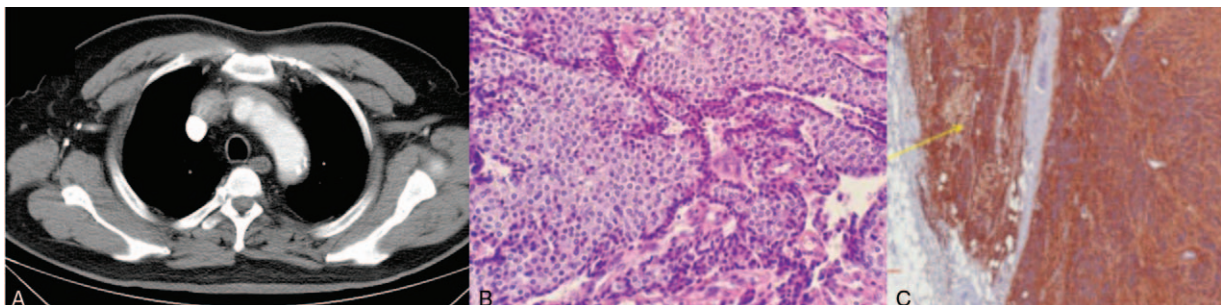


Figure 2. (A) Histopathology (hematoxylin and eosin, × 200) revealed a regular arrangement of tumor cells with consistent size. There was 1 abnormal mitosis in every 10 high power field (B). Immunohistochemistry demonstrated chromograninA (CgA)-positive cells in the carcinoid tissue (C).

hormone, thus causing carcinoid syndrome, ectopic adrenocorticotropic hormone (ACTH) syndrome.^[3] In this case, no tumor was detected on the parathyroid gland. And when the mediastinal lesion was removed, there was a dramatic decline of iPTH. Then, we could infer that the carcinoid tumor could secrete PTH, which is a low molecular polypeptide, leading to secondary hypercalcemia. Besides, the patient suffered from chronic renal insufficiency, which caused disorder of calcium and phosphate metabolism and secondarily elevated iPTH.

However, after removal of offending tumor in mediastinum, iPTH level was high even after 4 weeks of surgery, albeit, serum calcium level had normalized. This may be because the function of parathyroid was inhibited by the mediastinal mass. So, when the mass was removed, reflexively elevated PTH secreted by parathyroid appeared. Furthermore, the patient was still not recovered from chronic renal insufficiency, which also made a contribution to abnormal PTH level.

Cases about mediastinal carcinoid detected by ^{99m}Tc-MIBI scintigraphy were scarcely reported. Several ^{99m}Tc-MIBI uptake mechanisms are thought to contribute to its accumulation in the carcinoid. On the one hand, it is thought to be due to an increased vascularity and vascular permeability at the site of tumor. On the other hand, it could go back to the chemical characteristics of radiopharmaceuticals, the cationic charge, the lipophilic properties, and the negative transmembrane potentials generated in the cytoplasm and mitochondria of metabolically active cells.^[10]

Carcinoid tumors pose a diagnostic challenge and many are found during surgery. In this case, the most common differential diagnosis will be ectopic parathyroid adenoma.^[11] The exact diagnosis between mediastinal carcinoid with elevated PTH and ectopic parathyroid adenoma depends on pathology. For another, MEN type I could also present with high PTH level. However, there was no evidence to diagnose MEN type I because no abnormality was showed in parathyroid, pancreas, and

pituitary according to laboratory tests, imageological examinations, and history.^[4]

Finally, the deficiency of this paper was that immunostaining did not show whether the resected specimen was positive for PTH. We can infer that the carcinoid tumor secretes PTH by the sharp decline of PTH level after removal of the mediastinal mass.

References

- [1] Creutzfeldt W. Carcinoid tumors: development of our knowledge. *World J Surg* 1996;20:126–31.
- [2] Caplin ME, Baudin E, Ferolla P, et al. Pulmonary neuroendocrine (carcinoid) tumors: European Neuroendocrine Tumor Society expert consensus and recommendations for best practice for typical and atypical pulmonary carcinoids. *Ann Oncol* 2015;26:1604–20.
- [3] Steger C, Steiner HJ, Moser K, et al. A typical thymic carcinoid tumour within a thymolipoma: report of a case and review of combined tumours of the thymus. *BMJ Case Rep* 2010;2010:pil:bcr0420102958.
- [4] Susanne VE, Pascal FHJQ, Babs GT, et al. Classification of low-grade neuroendocrine tumors of midgut and unknown origin. *Hum Pathol* 2002;33:1126–32.
- [5] García-Yuste M, Matilla JM. The significance of histology typical and atypical bronchial carcinoids. *Thorac Surg Clin* 2014;24:293–7.
- [6] Litvak A, Pietanza MC. Bronchial and thymic carcinoid tumors. *Hematol Oncol Clin N Am* 2016;30:83–102.
- [7] Rosado de Christenson ML, Abbott GF, Kirejczyk WM, et al. Thoracic carcinoids: radiologic-pathologic correlation. *Radiographics* 1999;19:707–36.
- [8] Kaifi JT, Kayser G, Ruf J, et al. The diagnosis and treatment of bronchopulmonary carcinoid. *Dtsch Arztebl Int* 2015;112:479–85.
- [9] Sabita J, George K, Pramesh CS. Giant mediastinal carcinoid. *Indian J Med Paediatr Oncol* 2015;36:194.
- [10] Mehrzad B, Jamshid S, Hamid J, et al. ^{99m}Tc-MIBI lung scintigraphy in the assessment of pulmonary involvement in interstitial lung disease and its comparison with pulmonary function tests and high-resolution computed tomography: a preliminary study. *Medicine* 2015;94:e2082.
- [11] Lianxi Li, Libo Chen, Yi Yang, et al. Giant anterior mediastinal parathyroid adenoma. *Clin Nucl Med* 2012;37:889–91.