



Secondary primary common bile duct neuroendocrine tumor 10 years after the diagnosis of rectal adenocarcinoma: A case report

Saba Ebrahimiyan MD, Sakineh Soleimani Varaki MD

Corresponding author: Saba Ebrahimiyan MD. General Surgeon, Babol University of Medical Sciences, Iran

Abstract

Development of a second primary cancer in patients under follow-up because of metastasis is rare. We presented a 58-year-old man with neuroendocrine cancer of vater ampulla and distal common bile duct as a second primary cancer. The patient had been diagnosed with rectal adenocarcinoma and lung metastasis who underwent total mesorectal excision, lung lobectomy, and adjuvant therapy with an interval of 5 years. This article emphasizes on the importance of early detection of second primary cancer and treating it as the primary one.

Keywords: Secondary cancer, bile duct neuroendocrine; Rectal adenocarcinoma

Background

The possibility of secondary cancer is usually ignored in patients with cancer disease. Knowing the clinical presentations of second cancer in patients with colorectal carcinoma is important for an early diagnosis and treatment [1]. Neuroendocrine tumors are mostly associated with synchronous or metachronous second primary cancers [2]. There are a few cases of synchronous colorectal cancer (CRC) and gastrointestinal neuroendocrine tumors reported in the literature [3-5]. However, development of a primary pancreatic neuroendocrine tumor after complete resolution of the colorectal carcinoma has not been reported previously.

In this paper, we reported a patient with rectal adenocarcinoma who presented with neuroendocrine pancreatic tumor as a second primary cancer after receiving surgical and chemotherapeutical treatments for lung metastatic colorectal malignancy.

Case presentation

The patient was a 58-year-old man with no family history of cancer who presented with icterus and pruritus 5 years after being diagnosed with colorectal metastatic lung disease. He had no other complaints.

On past medical history, he had a history of a ten-centimeter

ulcerative mass above the anal verge diagnosed as stage two rectal adenocarcinoma 10 years ago, followed by total mesorectal excision surgery and chemotherapy (Figure 1). Five years later he was diagnosed with metastatic lung cancer for which he underwent right lower lobectomy and chemotherapy (Figure 2).

After a complete evaluation and physical examination, the patient underwent endoscopic ultrasound and retrograde cholangiopancreatography (ERCP) with periampullary tumor suspicion. Following ultrasound, severe stenosis of the common

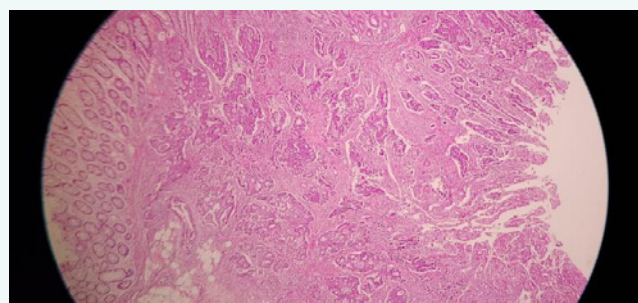


Figure 1 Rectal adenocarcinoma.

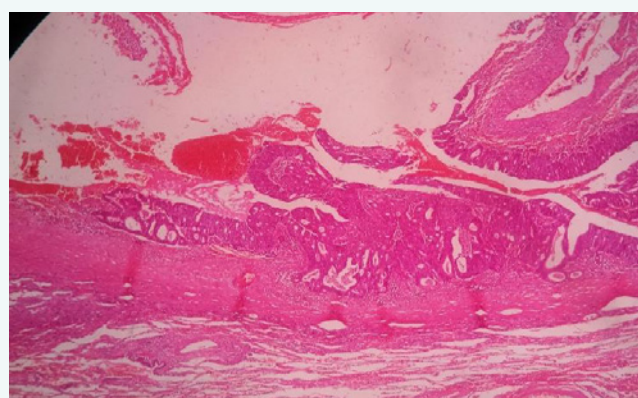


Figure 2 Lung metastasis.

Submitted: 08 August 2022 | **Accepted:** 13 September, 2022 | **Published:** 16 September, 2022

***Corresponding author:** Saba Ebrahimiyan MD. General Surgeon, Babol University of Medical Sciences, Iran

Copyright: © 2022 Ebrahimiyan S, et al. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Citation: Ebrahimiyan S, Varaki SS (2022) Secondary primary common bile duct neuroendocrine tumor 10 years after the diagnosis of rectal adenocarcinoma: A case report. J Surg Oncol Clin Res 4: 3.



bile duct, dilation of the proximal common bile duct, intrahepatic bile duct, and main pancreatic duct, as well as a 16-millimeter hypochoic lesion was detected. The MRCP and ERCP revealed 10-millimeter severe stenosis of the proximal common bile duct. Following ERCP, a plastic stent was inserted, and a fine-needle aspiration biopsy was taken indicating malignancy (Figure 3). The patient underwent Whipple surgery 10 days later after all biochemistry elements including bilirubin and hepatic enzymes became normal. The tumor was positive for synaptophysin, chromogranin and negative for CDX2, and PAX8. The morphologic and immunohistochemically findings were in favor of a well-differentiated neuroendocrine tumor (Figure 4).

Discussion

Neuroendocrine tumor is a rare tumor, despite its incidence has been increasing over the past two decades because of improved knowledge and diagnostic techniques and is now estimated to occur in about 5.25 individuals per 100,000[6].

CRC is now the third cause of cancer death in the world, and its incidence is slowly rising in developing countries. Known as colorectal adenocarcinoma, CRC is created from the glandular and epithelial cells of the large intestine. The CRC emerges when certain cells of the epithelium gain a series of genetic or epigenetic mutations [7].

The overall survival of patients with advanced colorectal

carcinoma has been improved with multidisciplinary treatments. However, one of the late outcomes in CRC survivors is the higher risk of second primary malignancies which has become more important in clinical practice[8]. Young age and lower stages of malignancy are important factors associated with a more favorable outcome including complete resolution of malignancy and a normal life expectancy [11]. As in our patient, the cancer was diagnosed at a young age and low stage, normal life expectancy and favorable outcome were predictable, as well as late metastasis and secondary primary cancer in other organs.

Neuroendocrine tumors of the extrahepatic bile ducts are rare, with less than 200 reported cases in the literature since 1961[12]. The neuroendocrine tumor is emerged mainly from enterochromaffin or Kulchitsky cells and has invariant pathological characteristics that typically match the site of origin [9] and may develop in almost any organ. Most neuroendocrine tumors occur in the gastrointestinal tract, pancreas, and bronchopulmonary system [10].

The endocrine nature of the extrahepatic bile duct neuroendocrine tumors cannot usually be diagnosed before surgery because there is no serum marker or hormonal changes associated with the disease. Despite advances in technology and the availability of different diagnostic imaging, as in our patient, the diagnosis is possible using ERCP and biopsy before surgery [13].

Extrahepatic bile duct neuroendocrine tumors grow slowly, and aggressive surgical resection is the cornerstone of a curative treatment [14]. As in this patient, complete surgical resection with free margins was done and follow-up continued.

In conclusion, we reported the development of a rare case of neuroendocrine tumor after complete survival from colorectal metastatic lung disease. Neuroendocrine tumors, although rare, should be considered as a second primary cancer in patients with colorectal adenocarcinoma.

Consent: Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

We declare none of the authors are employed by a governmental agency that has a primary function other than research and education. The paper was done at Babol University of Medical Sciences which is for research and education purposes. None of the authors are submitting the paper as an official representative or on behalf of the government.

References

1. Kaohsiung J, Clinical characteristics of patients with sporadic CRC and primary cancers of other organs. *Med Sci.* 2006 Nov;22(11):547-53. doi: 10.1016/S1607-551X(09)70351-1.
2. Jung-Yu Kan, Jan-Sing Hsieh, Yong-Sang Pan, Wen-Ming Wang, Fang-Ming Chen, Chang-Ming Jan, Yu-Sheng Huang, Tsung-Jen Huang, Jaw-Yuan Wang, Rupert Prommegger, Christian Ensinger, Philipp Steiner, Tonja Sauper, Christoph Profanter, Raimund Margreiter. Neuroendocrine tumors and second primary malignancy-a relationship with clinical impact? *Anticancer Res.* Mar-Apr 2004;24(2C):1049-51.

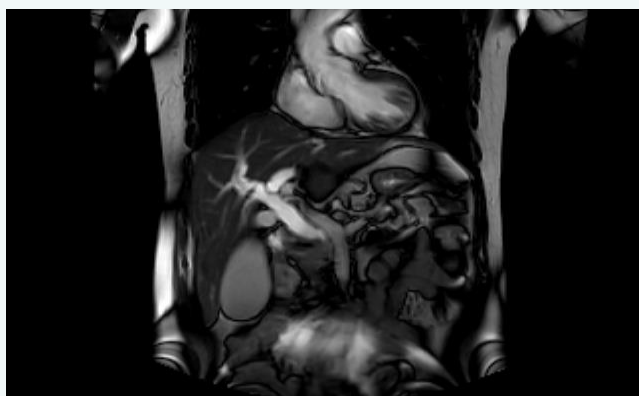


Figure 3 MRCP and dilated intrahepatic bile duct.

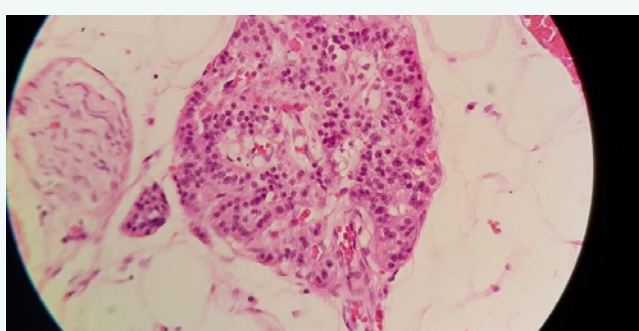


Figure 4 Neuroendocrin tumor.



3. Winn JN, Sathyamurthy A, Kneib JL, et al. Synchronous gastrointestinal carcinoid tumor and colon adenocarcinoma: case reports and literature review. *Am J Case Rep.* 2017;18:626–630. doi: 10.12659/AJCR.903580. [PubMed] [CrossRef] [Google Scholar]
4. Cokmert S, Demir L, Akder SA. Synchronous appearance of a high-grade neuroendocrine carcinoma of the ampulla vater and sigmoid colon adenocarcinoma. *Case Rep Oncol Med.* 2013;2013:930359. [PMC free article] [PubMed] [Google Scholar]
5. Mohapatra S, Ibrarullah M, Mohapatra A, et al. Synchronous adenocarcinoma and neuroendocrine carcinoma of the colon: a case report. *J Surg Case Rep.* 2016;2016(3):rjw042. [PMC free article] [PubMed]
6. Yao JC, Hassan M, Phan A, Dagohoy C, Leary C, Mares JE, et al. One hundred years after “carcinoid”: epidemiology of and prognostic factors for neuroendocrine tumors in 35,825 cases in the United States. *J Clin Oncol.* 2008;26:3063–72.
7. Ewing I, Hurley JJ, Josephides E, Millar A. The molecular genetics of colorectal cancer. *Frontline Gastroenterol.* 2014;5:26–30. [PMC free article] [PubMed] [Google Scholar]
8. Lee, Yu-Ting MD; Liu, Chia-Jen MD; Hu, Yu-Wen MD; Teng, Chung-Jen MD; Tzeng, Cheng-Hwai MD; Yeh, Chiu-Mei MS; Chen, Tzeng-Ji MD, PhD; Lin, Jen-Kou MD, PhD; Lin, Chun-Chi MD; Lan, Yuan-Tzu MD, PhD; Wang, Huann-Sheng MD; Yang, Shung-Haur MD; Jiang, Jeng-Kai MD, PhD; Chen, Wei-Shone MD; Lin, Tzu-Chen MD; Chang, Shih-Ching MD, PhD; Chen, Ming-Huang MD; Teng, Hao-Wei MD, PhD; Liu, Jin-Hwang MD, PhD; Yen, Chueh-Chuan MD, PhD. incidence of Second Primary Malignancies Following Colorectal Cancer. A Distinct Pattern of Occurrence Between Colon and Rectal Cancers and Association of Co-Morbidity with Second Primary Malignancies in a Population-Based Cohort of 98,876 Patients in Taiwan. *Medicine: July 2015 - Volume 94 - Issue 26 - p e1079.*
9. Creutzfeldt W. Carcinoid tumors: development of our knowledge. *World J Surg.* 1996;20:126–31.
10. Modlin IM, Oberg K, Chung DC, Jensen RT, de Herder WW, Thakker RV, et al. Gastroenteropancreatic neuroendocrine tumours. *Lancet Oncol.* 2008;9:61–72.
11. Isabelle Soerjomataram , Melissa S. Y. Thong , Majid Ezzati , Elizabeth B. Lamont , Wilma J. Nusselder , Lonneke V. van de Poll-Franse. Most colorectal cancer survivors live a large proportion of their remaining life in good health . *Cancer Causes Control* (2012) 23:1421–1428-
12. Pilz E. Uber ein karzinoid des ductus choledocus. *Zentralbl. Chir.* 1961;86:1588–1590. [PubMed] [Google Scholar]
13. Nickos M., Theodossis S.P., Georgia K., Ioannis P., Spiros T.P., Ioannis K. Neuroendocrine tumors of extrahepatic biliary tract. *Pathol. Oncol. Res.* 2014;20:765–775.
14. Walter T., Brix-Benmansour H., Lombard-Bohas C., Cadiot G. New treatment strategies in advanced neuroendocrine tumours. *Dig. Liver. Dis.* 2012;44:95–105.