

Synchronous occurrence of intrahepatic, intraductal cholangiocarcinoma and rectal cancer

Rita Slim^{1*}; bernard el khoury¹; Carla Tohme²; Joseph Khazen³; William Nehme⁴; Roger Noun⁴; Fadi Nasr⁵

¹Gastroenterology department, Hotel-Dieu de France university hospital, Saint Joseph university, Beirut, Lebanon.

²Radiology department, Hotel-Dieu de France university hospital, Saint Joseph university, Beirut, Lebanon.

³Pathology department, Hotel-Dieu de France university hospital, Saint Joseph university, Beirut, Lebanon.

⁴Surgery department, Hotel-Dieu de France university hospital, Saint Joseph university, Beirut, Lebanon.

⁵Oncology department, Hotel-Dieu de France university hospital, Saint Joseph university, Beirut, Lebanon.

Received Date : Mar 30, 2022
Accepted Date : Apr 19, 2022
Published Date : May 04, 2022
Archived : www.jcmimagescasereports.org
Copyright : © Rita Slim 2022

***Corresponding Author:** Rita Slim, Gastroenterology department, Hotel-Dieu de France university hospital, Saint Joseph university, Beirut, Lebanon. Tel: 009613622343
Email: rslimkaram@hotmail.com

Abstract

Liver metastasis from colorectal cancer is a common finding, but synchronous occurrence of intrahepatic cholangiocarcinoma (ICC) and colorectal cancer is extremely rare. We report a case of a 68-year-old man, diagnosed with rectal cancer and in whom, the staging CT scan showed a hepatic metastasis with intra-ductal hilar lesion. Biopsy of the hepatic lesion revealed an infiltrating cholangiocarcinoma. And the final diagnosis was a klatskin tumor coinciding with a rectal carcinoma. The possibility of multiple primary cancers should always be suspected in every oncologic patient even if the cancer is known to have metastatic lesions.

Introduction

Colorectal cancer (CRC) is the third most common cancer of all cancers with metastasis being the major cause of death in the majority of patients [1]. Metastatic rectal cancer often spreads to lungs, peritoneum, bones or the liver where it takes mostly the form of parenchymal mass and rarely the form of intraductal lesions making the diagnosis with intraductal cholangiocarcinoma (ICC) difficult. Some features are described as suggestive of intraductal metastasis rather than ICC [2]. Immunohistochemical studies post biopsy remain the gold standard to differentiate between the two [2]. The digestive system is the most common site of multiple primary tumors which is still a rare condition even if multiple cases have been reported in the literature. In our case, a cholangiocarcinoma presented as intra-ductal enhancing lesion and necrotic secondary lesion in patient known to have rectal cancer.

Case presentation

A 68-year-old man was diagnosed with rectal adenocarcinoma (**Figure 1**). The abdomino-pelvic MRI done for the staging of the rectal tumor, showed in addition to the lower rectal lesion an intraductal enhancing lesion at the confluence of the hepatic ducts extending to the origin of the right hepatic duct with a necrotic secondary lesion in segment II (**Figure 2**). A

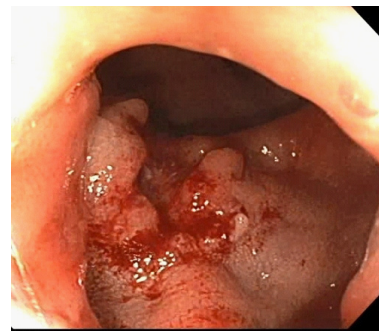


Figure 1: A 4 centimeter ulcerated lesion, 2 centimeters above the anal margin.

biopsy of the liver lesion showed moderately differentiated adenocarcinoma with a phenotype consistent with a primary cholangiocarcinoma. Therefore, the diagnosis of two primary tumors was made and the patient received the appropriate chemotherapy for both tumors and radiotherapy for the rectal tumor. On follow-up, the right extension of the intraductal lesion regressed and an extended left hepatectomy (**Figure 3**) was performed to be followed by a proctocolectomy. The pathological examination of the resected liver showed a non-encapsulated, firm intrahepatic mass with a prominent polypoid growth inside a dilated bile duct. On microscopic examination, the tumor is composed of large cells with marked

Citation: Rita Slim. Synchronous occurrence of intrahepatic, intraductal cholangiocarcinoma and rectal cancer. *J Clin Med Img Case Rep.* 2022; 2(3): 1134.

atypia and pleomorphism lining malignant small ducts and slits, strongly expressing the cytokeratin 7, confirming the diagnosis of cholangiocarcinoma (**Figure 4**).

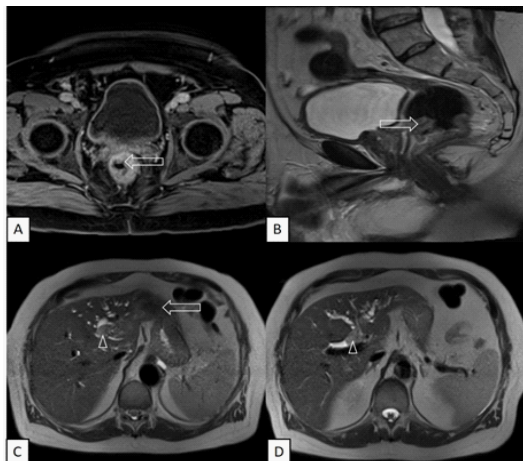


Figure 2: A, B Solid mass of the anterior wall of the lower rectum on axial and sagittal plane, C, D Intraductal enhanced lesion at the confluence of the hepatic ducts (Triangle) with necrotic secondary lesion in segment II (Arrow) on axial MRI plane).

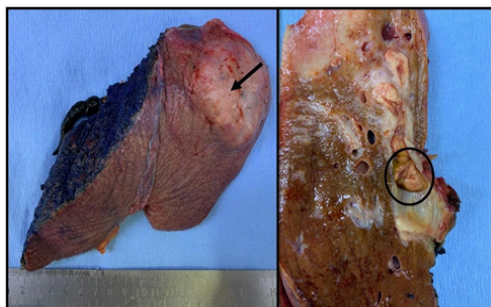


Figure 3: Left hepatectomy showing a large, non-encapsulated, ill-demarcated, firm, white-tan nodular intrahepatic mass (arrow) with a prominent polypoid growth inside a dilated bile duct (circle).

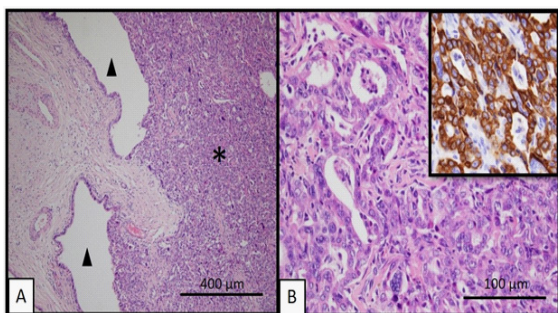


Figure 4: (A) (H&E x100): Microscopy shows that the tumor infiltrates the hepatic parenchyma and has an intraductal component (asterisk), growing inside the lumen of an enlarged bile duct (arrowheads). (B) (H&E x400): The tumor is composed of large cells with marked atypia and pleomorphism lining malignant small ducts and slits. Tumor cells strongly express cytokeratin 7 (insert), and rare cells exhibit faint cytokeratin 20 expression).

Discussion

The presence of multiple synchronous primary malignancies is a rare condition occurring most frequently in the digestive system [3]. The main finding of the present case is that ICC unexpectedly existed in the liver of a patient with rectal cancer without metastatic lesions. CRC eventually develops liver me-

tastases and therefore, hepatic masses are usually diagnosed as metastatic lesions that can infiltrate the biliary ducts and adopt a pattern of intrabiliary growth [3].

It is important to be aware that primary intraductal biliary neoplasms are not the only type of tumor that can appear as an intraductal neoplastic filling defect. In fact, Intraductal ducts metastasis from colorectal cancer is reported in few cases in the literature leading to confusion with primary intrahepatic [4, 5]. Some features are described as suggestive of intraductal metastasis rather than ICC, colorectal cancer as the extrabiliary malignant disease, a parenchymal mass contiguous with the intraductal lesion, and expansile growth of the intraductal lesion [2]. Immunohistochemical studies may be needed to differentiate the two. In fact, the usefulness of immunostaining with CK7 and CK20 is reported to distinguish between intraductal metastasis and ICC. The colorectal epithelium is immunoreactive to anticytokeratin 20 antibody but not to anticytokeratin 7 antibody, which is reactive to biliary epithelium [2]. In our case even though we have findings suggestive of intraductal metastasis, the rectal cancer and the mass contiguous to the intraductal lesion, the immunohistochemical staining of the preoperative liver biopsy as well as the resected liver was positive for cytokeratin 7 and negative for cytokeratin 20, confirming the diagnosis of ICC.

ICC sometimes coexists with other cancers such as HCC [6] thyroid cancer, and renal cell carcinoma [7]. To our knowledge, only two cases of synchronous ICC with rectal cancers are described in the literature [8, 9]. Due to the increased life expectancy and improved screening programs, the early detection ratio for multiple primary malignancies is expected to increase. Our case confirms the need to be aware of the possibility of multiple primary cancers, especially in the GI organs even if the patient has a history of cancer that is likely to cause metastatic lesions. Thus, the combination of Capecitabine and Oxaliplatin was chosen specifically in this case to treat both cancers after verifying the absence of metastasis. Accurate diagnosis of metastatic lesions is very important for selection of adjuvant therapy. Staging, therapeutic scheme and eventually prognosis will change dramatically between metastatic disease and synchronous neoplasm.

Declarations: The patient provided informed consent to publish his clinical case as well as the images that were found in his case.

Conflicts of Interest: The authors have no conflicts of interest to declare that are relevant to the content of this article.

Funding: No funding was received to assist with the preparation of this manuscript.

Financial interests: The authors declare they have no financial interests.

References

1. Prashanth Rawla, Tagore Sunkara, Adam Barsouk. Epidemiology of colorectal cancer: incidence, mortality, survival,

and risk factors. *Prz Gastroentero*. 2019; 14(2):89-103. [DOI: 10.5114/pg.2018.81072] [Epub 2019 Jan 6].

2. Lee YJ, Kim SH, Lee JY, Kim MA, Lee JM, Han JK, Choi BI. Differential CT Features of Intraductal Biliary Metastasis and Double Primary Intraductal Polypoid Cholangiocarcinoma in Patients with a history of extrabiliary Malignancy. *Am J Roentgenol*. 2009; 193(4):1061-9. [DOI: 10.2214/AJR.08.2089].

3. Alan I Valderrama-Treviño et al. Hepatic Metastasis from Colorectal Cancer. *Euroasian J Hepatogastroenterol*. 2017; 7(2): 166-175. [DOI: 10.5005/jp-journals-10018-1241].

4. Sasaki S, Nomura Y, Fukutomi S, Shirahama N, Kusano H, Akiba J, Sakai H, et al. Intrahepatic growth type of metastasis from colon cancer, 12 years after curative colectomy: a case report. *BMC Surg*. 2019; 19(1):8. [DOI: 10.1186/s12893-018-0466-4].

5. Yimin Dong, Hitendra Patel and Charmi Patel. Intrahepatic Hepatic Metastasis of Colorectal Carcinoma Mimicking Primary Cholangiocarcinoma: A Case Report and Review of the Literature. *Case Rep Pathol*. 2016; 4704781. [DOI: 10.1155/2016/4704781].

6. Bogdan Pinteau, Luca Di Tommaso, Annarita Destro, Massimo Roncalli. Combined hepatocellular carcinoma - cholangiocarcinoma harboring a metastasis of colon adenocarcinoma. *J Gastrointest Liver Dis*. 2013; 22(3):341-3.

7. BF Levy, A Nisar, ND Karanjia. Cholangiocarcinoma, renal cell carcinoma and parathyroid adenoma found synchronously in a patient on long-term methotrexate. *HPB (Oxford)*. 2006; 8(2):151-3. [DOI: 10.1080/13651820410016705].

8. Akabane S, Ohira M, Kobayashi T, Kuroda S, Tanimine N, Shimizu S, Tahara H, et al. Intrahepatic cholangiocarcinoma coinciding with a liver metastasis from a rectal carcinoma: a case report. *Surg Case Rep*. 2016; 2(1):94. [DOI: 10.1186/s40792-016-0222-x].

9. Guo JQ, Zou JJ, Zhu JD, Jiang C, Shao CX. A case report of rectal adenocarcinoma with intrahepatic cholangiocarcinoma of the liver. *J Int Med Res*. 2019; 47(11):5883-5890. [DOI: 10.1177/0300060519876751].