Cholangiocarcinoma: Presentation with Parotideal Metastasisand Later Hepatocolonic Fistulization

Kolanjiyokarsinom: Parotis Metastazıyla Prezentasyon ve Sonrasında Hepatokolonik Fistülizasyon Gelişim

Mutlu Doğan[1], Gökmen Umut Erdem[1], Öznur Bal[1], Devrim Tuğba Ünal[2], Murat Vural[3], Efnan Algın[1], Nurullah Zengin[1]

[1]Ankara Numune Training and Research Hospital, Department of Medical Oncology
[2]Ankara Numune Training and Research Hospital, Department of Pathology
[3]Ankara Numune Training and Research Hospital, Department of Radiology

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ÖZET

Parotis metastazı gastrointestinal kanserlerde, özellikle pankreatikobiliyer malignitelerde nadirdir. Parotis metastazı ile ilişkili semptomlar atipik prezentasyona neden olabilir. Hepatokolonikf istülizasyon da kolanjiyo karsinomda nadirdir. 62 yaşında parotis metastazı ile atipik prezentasyonla başvuran ve daha sonra hepatokolonik fistülizayon gelişen hasta literatür eşliğinde sunulmuştur.
Anahtar Kelimeler: kolanjiyo karsinom, parotis metastazı, hepatokolonik fistülizasyon

ABSTRACT

Parotidealmetastasis in gastrointestinal cancers, especially in pancreaticobiliary malignancies is rare. The symptoms related with parotideal metastasis might lead to atypical presentation. Hepatocolonic fistulization is also not common in cholangio carcinoma. A 62-year old cholangio carcinoma with atypical presentation with parotideal metastasis and later hepatocolonic fistulization is reported with review of the literature.
Keywords: cholangiocarcinoma, parotideal metastasis, hepatocolonic fistulization

Introduction

Parotideal metastasis is rare in pancreaticobiliary cancer. The patients with parotideal metastasis may present with the symptoms related to the parotideal metastasis. Hepatocolonic fistulization is also rare in cholangiocarcinoma. In here, we report a 62-year old cholangiocarcinoma patient with parotideal metastasis.

Case

A 62-year old male admitted to the hospital with a right parotideal mass. His pathology revealed adenocarcinoma metastasis. The parotideal histopathology had large tumor cells with vesicular nucleus and eosinophilic cytoplasm formed gland like structure in a desmoplastic stroma (fig 1a). These tumor cells were negative for actin, CEA, GCDFP and CK5/6. He had a hepatic mass (8.5x8cm) on segments 2 & 3 extending to segments 5 & 6 on computerized tomography (CT) which gave rise to tought of cholangiocarcinoma radiologically. He had also intrahepatic biliary dilatation with multiple paraaortic and paracaval lymphadenopathies (LAPs) on CT and bilaterally cervical LAPs. The parotideal and jugular LAP (SUVmax: 10,4), hepatic (SUVmax: 16) and intraabdominal LAPs (SUVmax: 6,4-12,6) had high 18FDG uptake on 18-fluorodeoxyglucose positron emission tomography (18FDG-PET-CT). The tru-cut biopsy of the hepatic lesion revealed intrahepatic cholangiocarcinoma (CC) (fig 1b, 1c). There were atypical larger pleomorphic tumor cells with large eosinophilic cytoplasm, vesicular nuclei and prominent

Adress for correspondence: Ankara Numune Training and Research Hospital, Ankara - Turkey
e-mail: **mutludogan1@yahoo.com**
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nucleoli among dysplastic hepatocytes in the liver biopsy (fig 1b). The tumor cells were negative for TTF-1 and Heppar whereas they were positive for CK7, CK8, CK18, CK19 and polyclonal CEA (fig 1c). The parotideal and liver biopsies were reevaluated together concluding similar morphology (fig 1a, 1c). He was given gemcitabine and cisplatin for metastatic disease. The necrotic component of the primary lesion increased with colonic fistulization besides no other changes after 3 cycles of chemotherapy (fig 2a, 2b). Then, he was followed-up with best supportive care because of clinical progression and poor performance status.

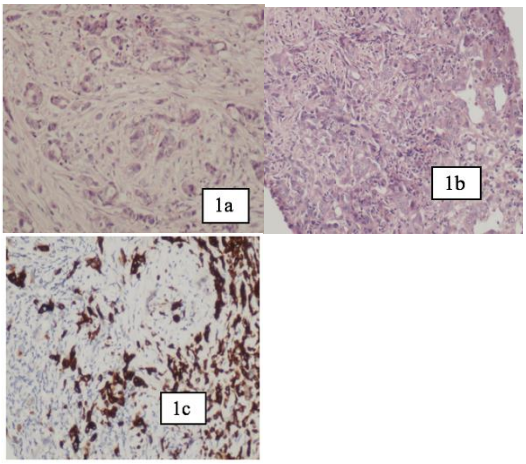

Figure 1: a) paratideal pathology b) liver pathology c) TTF1 negative, CK7, CK8, CK18, CK19 and polyclonal CEA positive biopsy

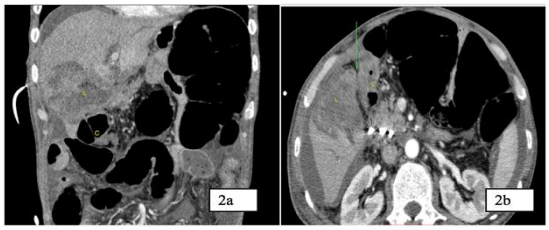

Figure 2: The necrotic component of the primary lesion

Discussion

Parotideal metastasis in solid tumors is not common. The patients with parotideal metastasis may present with a parotideal mass or trismus (1). Parotideal metastasis should be kept in mind while evaluating the patients with parotideal mass although most of them have primary parotideal carcinoma. Cholangiocarcinoma is a rare malignant epithelial tumor of the biliary tract (2). Liver is the most common site for metastasis, however, unexpected sites like parotid were also reported rarely in the literature (1). Most of CC is well-to-moderately differentiated adenocarcinoma with desmoplastic reaction (1). In our patient, the parotideal pathology revealed large tumor cells with vesicular nucleus and eosinophilic cytoplasm formed gland like structure in a desmoplastic stroma (fig 1a). These tumor cells were negative for actin, CEA, GCDFP and CK5/6. There were atypical larger pleomorphic tumor cells with large eosinophilic cytoplasm, vesicular nuclei and prominent nucleoli among dysplastic hepatocytes in the liver biopsy (fig 2a, 2b). The parotideal and liver biopsies were reevaluated together concluding that these had similar morphology. So, the patient was treated as metastatic cholangiocarcinoma. The necrotic component of the hepatic lesion increased giving rise to the thought of benefit from chemotherapy, however this led to a hepatocolonic fistulization which was shown radiologically on CT (fig 2). It was interesting that he had neither cholangitis nor other complications despite hepatocolonic fistulization. Unfortunately, he had clinical worsening in the following weeks and followed up with best supportive care.

Hepatocolonic fistulization is a serious complication of the intrahepatic tumors, especially in those with necrotic component and localized very closer to the colon. Hepatoduodenal fistulization in hepatic hydatid cyst was even reported as an uncommon complication in the literature and hepatoduodenal fistulization in cholangiocarcinoma without septic complication sounds to be more rare (3,4).

In conclusion, atypical metastatic pattern like parotideal metastasis and rare complications like hepatocolonic fistulization might be seen in cholangiocarcinoma.

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Adress for correspondence: Ankara Numune Training and Research Hospital, Ankara - Turkey
e-mail: **mutludogan1@yahoo.com**
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Adress for correspondence: Ankara Numune Training and Research Hospital, Ankara - Turkey
e-mail: **mutludogan1@yahoo.com**
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