

A PAINFUL, ERYTHEMATOUS, NODULAR LESION IN BUCCAL REGION

Case Report

BUKKAL BÖLGEDE AĞRILI, ERİTEMATÖZ, NODÜLER BİR LEZYON

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ABSTRACT

Cholangiocarcinoma is the cancer of the bile duct epithelium and usually presents with painless jaundice and weight loss. Cutaneous metastases have rarely been reported in cholangiocarcinoma. Most of the reports of cutaneous metastases of cholangiocarcinoma are those seen around the percutaneous biliary drainage in the abdominal skin due to implantation of tumor cells during the procedure. Here we report a 41-year old male patient who was diagnosed with cholangiocarcinoma. The presented case is the first case of cholangiocarcinoma with distant cutaneous metastases to the face. Distant skin metastases of cholangiocarcinoma must be in the differential diagnosis if atypical cutaneous lesions appears in patients with known cholangiocarcinoma.

Key Words: *Cholangiocarcinoma;
skin;metastasis;buccal.*

ÖZET

Kolanjiyokarsinom safra kanalı epitelinde köken alan ağrısız sarılık ve kilo kaybı ile seyreden bir kanser türüdür. Kolanjiyokarsinomun deri metastazları az sıklıkta görülmektedir. Bildirilen olguların çoğu genellikle perkütan yolla bilayer drenaj katateri etrafında abdominal bölge derisine tümör ekilimi yolu ile olmaktadır. Burada kolanjiyokarsinom tanısı konan 41 yaşında bir erkek hasta sunulmaktadır. Bu olgu yüze metastaz yapan ilk kolanjiyokarsinom olgusudur. Kolanjiyokarsinom tanısı ile takip edilen hastalarda atipik yerleşimli lezyonların kolanjiyokarsinomun uzak deri metastazı olabileceği akıllarda bulundurulmalıdır.

Anahtar kelimeler: *Kolanjiyokarsinom;
deri; metastaz; bukkal.*

INTRODUCTION

Cholangiocarcinoma is the cancer of the bile duct epithelium and usually presents with painless jaundice and weight loss. It primarily occurs in male patients older than 50 years.⁽¹⁾ It typically metastasizes to regional lymph nodes and to distant sites in liver.⁽²⁾ The response rate to chemotherapy alone is less than 15%.⁽³⁾

Here we report 41-year old male patient diagnosed cholangiocarcinoma with the metastasis to buccal region while continuing of gemcitabin-cisplatin chemotherapy regimen.

CASE REPORT

A 41-year old male patient was admitted to our hospital with the complaint of right upper quadrant abdominal pain. The CT scan of the abdomen revealed a contrast-enhanced multi-loculated lesion with diameter of 3,5 x 5,5 cm, involving segments IVA and VIII of the liver. There were several other millimetric foci in the liver with similar patterns. Multiple lymphadenopathies (LAPs) in left paraaortic, left gastric, celiac, and mesenteric regions were also detected. The differential diagnoses were amoebic or pyogenic abscess but amoebic serology was negative. The patient underwent diagnostic and therapeutic laparotomy. Segmentectomy and celiac lymph node biopsy were done. Frozen section during the operation revealed carcinoma and surgery was stopped. The pathological examination revealed poorly differentiated adenocarcinoma consistent with intrahepatic cholangiocarcinoma. Tumor cells were positive for pancytokeratin, CEA, CK 7, EMA and negative for CD 10, CD 15, CD 20 and AFP. Positron emission tomography/computerized tomography (PET/CT) detected metastases to second lumbar vertebra, left iliac bone and right ischial tuberosity, in addition to liver metastases and intra-abdominal LAPs. The patient received 9 cycles of gemcitabin-

cisplatin chemotherapy regimen. A partial response was achieved after cycle 3. After cycle 8, the patient complained of a painful, erythematous, nodular lesion of 8 mm in his left cheek. As he also had poor oral hygiene and gingivitis, this lesion was thought to be of infectious origin and antibiotic therapy with amoxicillin/clavulanate was initiated. However, the nodular lesion reached diameter of 2 cm and ulcerated after cycle 9 (**Figure 1**).



Figure1. Two cm nodular lesion in buccal skin.

The lesion was biopsied and the pathological examination revealed poorly differentiated adenocarcinoma (**Figure 2**).

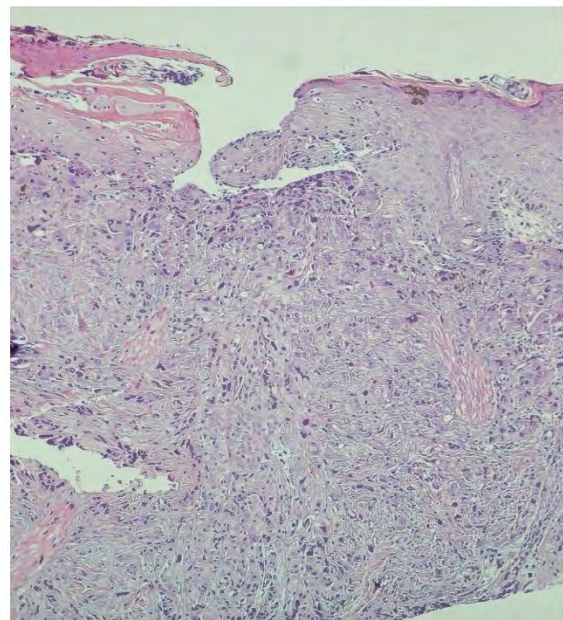


Figure 2. Photomicrograph (H&E 40x view) showing malignant infiltration consisting of bizarre cells with pleomorphic nucleus and prominent nucleoli. Note that these cells tend to form groups in desmoplastic stroma, form lumens and infiltrate whole reticular, superficial and deep dermis under the partially ulcerated epidermis.

The tumor cells were positive for cytokeratin 7, PAS, luminal mucin, CEA, and negative for cytokeratin 20 (**Figure 3**).

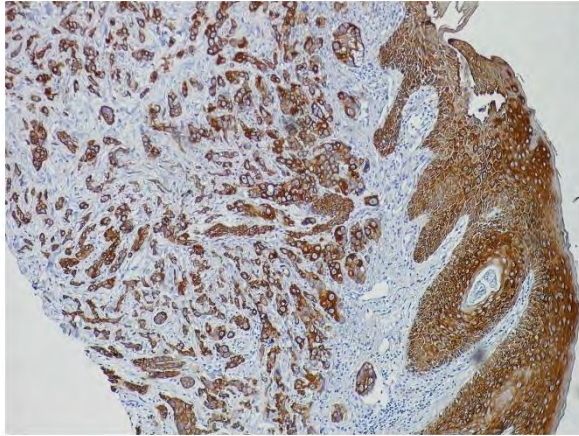


Figure 3. Photomicrograph (Cytokeratin-7 100x view) showing neoplastic cells diffusely stained with cytokeratin-7 antibody while no staining is observed at the epidermal layer.

A month later, he developed a new metastatic subcutaneous nodular lesion on the scalp. The patient then received 3 cycles of capecitabine-oxaliplatin chemotherapy regimen. However, the disease progressed and the patient died 13 months after initial diagnosis and 4 months after cutaneous metastases.

DISCUSSION

Skin metastases of solid tumors are rare and usually a manifestation of widespread disease. Cutaneous metastases have rarely been reported in cholangiocarcinoma. Most of the reports of cutaneous metastases of cholangiocarcinoma are those seen around the percutaneous biliary drainage in the abdominal skin due to implantation of tumor cells during the procedure. (4) Only 11 cases of distant skin metastases have been reported and most of them were on

the scalp. In two cases, metastases were in the chest wall and in one case at knee and chest Wall.(5,9,11,12)

The most common presentation of a metastatic skin lesion is a firm, painless nodule. In our case, the painful and erythematous nature of the nodule led to an initial misdiagnosis of infection. The clinicians should therefore be aware of the possibility of cutaneous metastasis in patients with cholangiocarcinoma, when a new cutaneous nodule is detected. Cutaneous metastases usually indicate advanced disease and poor prognosis(10). They develop during the late course of the disease and are rarely the presenting sign prior to the diagnosis.(5,10).

The presented case is the first case of cholangiocarcinoma with distant cutaneous metastases to the face. Distant skin metastases of cholangiocarcinoma must be in the differential diagnosis if atypical cutaneous lesions appears in patients with known cholangiocarcinoma.

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