Case Report

Presentation of Metastatic Cholangiocellular Cancer with Orbital Metastasis

Metastatik Kolanjioselüler Kanserin Orbital Metastaz ile Prezentasyonu

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ABSTRACT
Cholangiocarcinomas are cancers arising from epithelial cells of the intrahepatic and extrahepatic bile ducts. Most are locally advanced lesions when present. They usually present with jaundice, abdominal pain, and abnormal liver biochemical tests.

Proptosis developed in the right eye of a 49-year-old female patient who was diagnosed with carcinoma metastasis in the right femur and we examined for the primary one. Lacrimal gland biopsy of the patient was found to be compatible with cholangiocarcinoma metastasis. Neuroophthalmologic complications in cholangiocarcinoma are very rare and 10 cases have been reported in the literature.

Keywords: orbital metastasis, CNS metastasis, cholangiocarcinoma, exophthalmos

ÖZET
Kolanjiokarsinomlar intrahepatik ve ekstrahepatik safra kanallarının epitel hücrelerinden kaynaklanan kanserlerdir. Çoğu prezente olduğunda lokal olarak ilerlemiş lezyonlardır. Genellikle sarılık, abdominal ağrı ve anormal karaciğer biyokimyasal testleri ile prezente olurlar.

Sağ femurda karsinom metastazı saptanıp primaryne yonelik tetkik ettigimiz 49 yaşındaki kadın hastanın sağ gözünde proptozis gelişti. Hastanın lakrimal bez biyopsisi kolanjiokarsinom metastazı ile uyumlu saptandı. Kolanjiokarsinomda nörooptalmolojik komplikasyonlar oldukça nadir olup literatürde 10 vaka bildirimiştir.

Anahtar Kelimeler: orbital metastaz, SSS metastazı, kolanjiokarsinom, eksoftalmus

Introduction
Metastatic orbital masses make up less than 5% of orbital tumors [1]. Most frequent metastatic cancers of the orbital are breast (52%), prostate (12%) and lung cancers (8%) [2]. Only ten cases of neuro-ophthalmologic and oculcar presentations of cholangio-
carcinoma have been reported in the literature [3-6].

Case Presentation
A 49-year-old female patient who admitted to orthopedics with low back and hip pain underwent a biopsy of the right femoral proximal after the bilateral T1A and T2A
images in the patient’s hip MRI showed diffused lesions suspected of metastasis on bone surfaces. As a result of the biopsy which revealed carcinoma metastasis, the patient admitted to the oncology outpatient clinic. In the computer tomography scans of the patient multiple metastatic mass lesions of 24x11 mm in size were detected in the liver parenchyma at the level of segment 7. A biopsy of the liver was performed on the patient. While waiting for the biopsy result, proptosis and a limitation of eye movement developed in the right eye of the patient. An orbital MR was performed on the patient. The patient's cranial and orbital MRI images are shown in figure 1a-1b-1c-1d. In the orbital and brain MRI images before and after contrast, soft tissue mass lesions in the lateral vicinity of the right orbita in the
right lateral rectus muscle which erase extraconal periorbital fat plans, with their central showing necrotic heterogeneous contrast involvement were observed. Exophthalmos was present in the ipsilateral glob.Pathologic contrast involvement was also seen in the subcutaneous soft tissue muscle plans in orbital neighborhood. In addition, irregular dural thickening and contrast were observed in the intracranial area adjacent to the right orbit, which was evaluated as dural involvement. In T1 and FLAIR images after contrast, dural involvement was observed not only in orbital proximity but also in the opposite cerebral hemisphere. Diffusion MRI images showed no restriction of diffusion in the lesions. With these findings, the lesions were primarily considered in favor of metastatic involvements. But in MRI images, because of ventilation losses in the right paranasal sinuses compatible with sinusitis and rapid progression of the patient's MRI findings, it was thought in the differential diagnosis, that the infection should be excluded and biopsy from the lacrimal gland was performed on the patient. While waiting for the biopsy result, empirical vancomycin, meropenem and amphotericin B were initiated.

The result of the liver biopsy of the patient was reported as cholangiocellular carcinoma.

The result of biopsy from the lacrimal gland was consistent with cholangiocellular carcinoma metastasis. Radiotherapy was planned for the eye and the whole brain of the patient. After radiotherapy, it was decided to start gemcitabine and cisplatin.

Discussion

The presentation of our case was not compatible with typical presentation of cholangiocarcinoma. Initially bone metastases were detected and metastasis of orbital and central nervous system developed during the examination process. CNS metastasis and orbital metastasis are rather rare in cholangiocarcinoma [7-9]. There are 26 CNS metastasis cases in the literature. The brain was reported as the site of metastasis in 1.6% of patients with stage IV intrahepatic cholangiocarcinoma in a 2010-2015 review of US population. Orbital metastasis is also very rare and our case is the eleventh case in the literature. One case of a clival mass and sixth cranial nerve palsy, one case of metastasis to the medial rectus muscle and diplopia, two cases of metastasis to the occipital lobe and homonymous hemianopia, and one case of a hypercoagulable state-related stroke and homonymous hemianopia make up the five previously reported neuro-ophthalmologic presentations of cholangiocarcinoma[3]. A combination hepatocellular carcinoma/cholangiocarcinoma that metastasized to the retina and vitreous was reported in one case, and there have also been two instances of cholangiocarcinoma that metastasized to the orbit and caused eye pain[5]. While, a case presenting with skin and orbital metastasis was reported in 2020 in Japan [10], in the same year metastatic cholangiocarcinoma presenting with 6th cranial nerve paralysis was reported from Miami [6].

REFERENCES


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