DOI: 10.24938/kutfd.1318745 Kırıkkale Üniversitesi Tıp Fakültesi Dergisi 2023;25(2):361-364
Case Report Olgu Sunumu

MUCOEPIDERMOID CARCINOMA OF THE LIVER: A RARE, DIAGNOSTICALLY CHALLENGING CASE

Zorlayıcı ve Nadir Bir Vaka: Karaciğerin Mukoepidermoid Karsinomu

Arzu Hazal AYDIN[1] Seda YAMAK[2] Göktürk GÜRSOY [3]
İkbal OVALI ERDOĞDU[3] Yusuf BAYRAM ÖZOĞUL[3]

[1] *Department of Medical Pathology, Aksaray University Aksaray Training and Research Hospital, AKSARAY, TÜRKİYE*

[2] *Department of Medical Pathology, Ankara City Hospital, ANKARA, TÜRKİYE*

[3] *Department of General Surgery, Ankara City Hospital, ANKARA, TÜRKİYE*

ABSTRACT

Mucoepidermoid carcinoma is the most common malignant tumor of the salivary gland. It rarely occurs in other organs. Less than 20 cases of mucoepidermoid carcinoma of liver have been reported in the literature.

A 70-year-old woman with a liver mass is reported in this case report. After liver segmentectomy operation, the specimen was examined microscopically, a tumor consisting of epidermoid, mucinous, and intermediate cells was observed. It was interpreted as mucoepidermoid carcinoma. As no other focus was detected from comprehensive scanning of the patient, it was diagnosed as primary mucoepidermoid carcinoma of liver. Due to mimicking other tumors such as adenosquamous carcinoma and squamous cell carcinoma; cautious examination and differential diagnosis are important.

Keywords: *Mucoepidermoid carcinoma, liver, intrahepatic cholangiocarcinoma*

ÖZ

Mukoepidermoid karsinom tükürük bezinin en sık malign tümörü olup diğer organlarda nadiren görülür. Literatürde karaciğerde yerleşim gösterdiği bildirilen 20'den az vaka vardır. Biz de nadir bulunan karaciğerin primer mukoepidermoid karsinomu vakasını literatür eşliğinde sunduk. Karaciğerde kitle tespit edilen 70 yaşında kadın hastanın karaciğer segmentektomi spesimeni incelendi. Mikroskopik olarak epidermoid, müsinöz ve intermediate olmak üzere üç tip hücrenin karışık popülasyonundan oluşan tümör izlendi. Kapsamlı tarama sonrası karaciğer dışı olası odak tespit edilmeyen hastada tümör, karaciğerin primer mukoepidermoid tümörü olarak tanı aldı.

Mukoepidermoid karsinom karaciğerde nadir görülmesi, skuamöz hücreli karsinom ve adenoskuamöz hücreli karsinom gibi tümörleri taklit etmesi nedeniyle dikkatli ve kapsamlı mikroskopik muayenenin ve ayırıcı tanının önemli olduğu malignitedir.

Anahtar Kelimeler: *Mukoepidermoid karsinom, karaciğer, intrahepatik kolanjiokarsinom*

Correspondence / Yazışma Adresi: **Dr. Arzu Hazal AYDIN**
Department of Medical Pathology, Aksaray University Aksaray Training and Research Hospital, AKSARAY, TÜRKİYE
Phone / Tel: +905053480670 **E-mail** / E-posta: arzu.hazal.aydin@gmail.com
Received / Geliş Tarihi: 26.06.2023 **Accepted** / Kabul Tarihi: 22.07.2023

KÜ Tıp Fak Derg 2023;25(2):361-364
Doi: 10.24938/kutfd.1318745

INTRODUCTION

Mucoepidermoid carcinoma is a malignant epithelial neoplasm consisting of a mixture of mucinous, epidermoid, and intermediate cell types. It is most common malignant tumor in salivary gland in both children and adults (1,2). It occurs usually in major and minor salivary glands (1). It arises rarely in other organs such as lung, breast, thyroid gland (3). In gastrointestinal tract it occurs extremely rare. In literature some unusual locations have been reported including esophagus, liver, anal canal (3). We reported a case of primary mucoepidermoid carcinoma of the liver.

CASE REPORT

Our patient was a 70-year-old female who has a history of cholecystectomy. She did not have any comorbidity except from diabetes mellitus and Chronic Obstructive Pulmonary Disease. A mass of approximately 65x55 mm in size, located in the liver segment 5, was detected in dynamic computed tomography (CT) in an external center and it was evaluated as a possible primary liver malignancy. The patient consulted our hospital for further examination. In repeated abdomen CT the liver was described as slightly larger than normal, its surface was detected as smooth and homogeneous. A heterogeneous hypodense mass located in the segment 5 was revealed. It had mild peripheral enhancement, and its contour was irregular. Main hepatic duct was 20 mm at its largest part and, while the common hepatic duct was 9 mm in diameter and ended narrowing. On the F18-FDG PET/CT imaging, F-18 FDG uptake with an SUV max of 7.34, was observed in the liver segment 5. Any other pathologic F-18 FDG uptake was not detected. In addition, esophagoduodenoscopy and colonoscopy was performed. However, no pathologic lesion was identified.

Later, the patient was taken to liver segmentectomy operation with the decision of the council. In the frozen material sent to our department during the operation, malignant epithelial cells were detected in addition to the component resembling abscess content consisting of dense polymorphonuclear leukocytes, and the result was reported as malignant. Tumor adhesions to the colon mesentery were detected during the operation and segmental colon resection was added to the procedure.

In macroscopic examination of the resection specimen, a lesion with ill-defined border was detected (Figure 1). The tumor which was multilobulated with cream cut surface was 7x6 cm in diameter. It had gray gelatinous foci accompanying necrotic and hemorrhagic areas. There was no finding of cirrhosis or chronic liver disease at non-neoplastic liver tissue that could be seen in a little area.

Figure 1: A tumor located in liver with multilobulated and necrotic cut surface extending irregularly to peripheral tissue

The lesion was composed of tumor cells that coalesce as glandular structures and solid nests in some areas as well. In addition, macro and micro cystic areas were present in tumor at varying proportion. In higher magnification intracytoplasmic mucin was detected in some of the tumor cells. Some of them were well-delimited with polygonal shape and had abundant eosinophilic cytoplasm. Among these cells, numerous dyskeratotic cells were seen. Except these two profiles there was different cell population described as intermediate cells. All of these cells were intermingled (Figure 2).

In immunohistochemical study, diffuse and strong CK7 expression was detected in tumor cells. There was p63 positivity in squamous cells. In some cells intracytoplasmic mucin was immunohistochemically demonstrated with MUC5AC.

KÜ Tıp Fak Derg 2023;25(2):361-364
Doi: 10.24938/kutfd.1318745

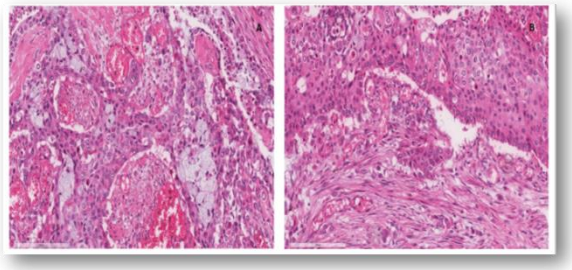

Figure 2: Mixture of three cell types: Epidermoid, mucinous, and intermediate. Hematoxilen & Eosin stain, 20x (A). Numerous dyskeratotic cells in tumor. Hematoxilen & Eosin stain, 20x (B).

DISCUSSION

On non-cirrhotic background there was a tumor consisting of the mixture of squamous, mucinous, and intermediate cells at varying degree. It had different histomorphologic appearance far from the usual features of hepatocellular carcinoma (HCC) and intrahepatic cholangiocarcinoma (ICC) which are the first two mostly seen primary tumors of the liver.

In the differential diagnosis adenosquamous carcinoma, squamous cell carcinoma, combined HCC and ICC, metastatic mucoepidermoid carcinoma and mucoepidermoid subtype of ICC were considered. Squamous cell carcinoma was excluded, due to the presence of mucin. Adenosquamous carcinoma and combined HCC and ICC were also excluded because of the existence of intermediate cells and not having conventional HCC component, respectively.

According to all findings the tumor was compatible with mucoepidermoid carcinoma. Careful screening and examination of the patient was recommended to exclude the possibility of metastases to the liver from any location.

No other primary focus was detected by additional imaging methods, so the tumor was diagnosed as mucoepidermoid carcinoma, a rare subtype of ICC.

For histologic grading several systems are used including Katabi, AFIP, Modified Healey and Brandwein (2,4). Tumor is graded histologically as low, intermediate or high by evaluating growth pattern, nuclear pleomorphism, rate of mitosis, existence of necrosis, lymphovascular, perineural and bone invasion. Because of the presence of necrosis and infiltrative borders, having mostly solid component and high mitotic activity (>5/10 BBA), and predominance of intermediate cells as well, the tumor was evaluated as high grade in our case.

Mucoepidermoid carcinoma is a malignant epithelial neoplasm consisted of epidermoid, mucinous, and intermediate cells. It is most common malignant tumor of salivary glands in adults and children (2). It rarely occurs in other organs (3). Mucoepidermoid carcinoma of the liver is an extremely rare neoplasm. The first case of mucoepidermoid carcinoma of the liver was presented by Pianzola and Drut in 1971 (5), and only 19 cases have been presented so far (3). There are different thoughts about its pathogenesis. One of them is that primary MEC may arise from terminal bile duct in association with squamous metaplasia (6). Another theory which suggested by Katsuda et al., is that MEC might originate from a congenital cyst that has not a connection with biliary tract (7). Due to its rarity, its etiology has not been cleared yet. Primary MEC of the liver is an aggressive malignancy with short survival (3,8). Our patient died six months later after the initial diagnosis. Elucidating its pathogenesis might contribute to the development of treatment strategies. Because it is rare in the liver and resembles other tumors such as squamous cell carcinoma, adenosquamous carcinoma; comprehensive macroscopic and microscopic examination of the lesion and excluding any possible metastases are crucial.

Conflict of interest: The authors declare no conflicts of interest.

Researchers' Contribution Rate Statement: Concept/Design: AHA, SY, YBÖ, GG, IOE; Analysis/Interpretation: AHA, SY, YBÖ, GG, IOE; Data Collection: AHA, SY, YBÖ, GG, IOE; Writer:AHA, SY; Critical Review: AHA, SY ; Approver :AHA, SY, YBÖ, GG, IOE

Informed Volunteer/Consent Form: Written informed consent was obtained from the patient for publication of this case report.

KÜ Tıp Fak Derg 2023;25(2):361-364
Doi: 10.24938/kutfd.1318745

Support and Acknowledgment: No financial support was received from any source for this work.

REFERENCES

1. Dahan LS, Giorgi R, Vergez S, Le Taillandier de Gabory L, Costes-Martineau V, Herman P, et al. Mucoepidermoid carcinoma of salivary glands: A French Network of Rare Head and Neck Tumors (REFCOR) prospective study of 292 cases. Eur J Surg Oncol. 2021;47(6):1376-83.
2. Katabi N, Ghossein R, Ali S, Dogan S, Klimstra D, Ganly I. Prognostic features in mucoepidermoid carcinoma of major salivary glands with emphasis on tumour histologic grading. Histopathology. 2014;65(6):793-804.
3. Hou P, Su X, Cao W, Xu L, Zhang R, Huang Z, et al. Whole-exome sequencing reveals the etiology of the rare primary hepatic mucoepidermoid carcinoma. Diagn Pathol. 2021;16(1):29.
4. Brandwein MS, Ivanov K, Wallace DI, Hille JJ, Wang B, Fahmy A, et al. Mucoepidermoid carcinoma: A clinicopathologic study of 80 patients with special reference to histological grading. Am J Surg Pathol. 2001;25(7):835-45.
5. Pianzola LE, Drut R. Mucoepidermoid carcinoma of the liver. Am J Clin Pathol. 1971;56(6):758-61.
6. Koo J, Ho J, Wong J, Ong GB. Mucoepidermoid carcinoma of the bile duct. Ann Surg. 1982;196(2):140-8.
7. Katsuda S, Nakanishi I, Kajikawa K, Takabatake S. Mucoepidermoid carcinoma of the liver. Acta Pathol Jpn. 1984;34(1):153-7.
8. Arakawa Y, Shimada M, Ikegami T, Kubo T, Imura S, Morine Y, et al. Mucoepidermoid carcinoma of the liver: Report of a rare case and review of the literature. Hepatol Res. 2008;8(7):736-42.