Oncology

Spontaneous rupture of a hybrid oncocytic chromophobe tumor: A case report

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A B S T R A C T

The prognosis of Hybrid oncocytic chromophobe tumor (HOCT) is usually excellent, nevertheless, we are reporting a rare case of HOCT that resulted in death from tumor infection and rupture. Bilateral solid and cystic masses were detected in a 55-year-old woman during a computed tomography examination. HOCT was diagnosed following histopathological examination obtained during needle biopsy. Watchful waiting at another hospital was selected as the treatment strategy. Three years later, she was referred to our hospital in shock and died on the 3rd hospital day. The cause of death was thought to be peritonitis secondary to rupture of an infected HOCT.

Introduction

Hybrid oncocytic chromophobe tumor (HOCT) is a renal cell neoplasm characterized by cellular and architectural features of both renal oncocytoma (RO) and chromophobe renal cell carcinoma (CHRCC). The prognosis of HOCT is usually excellent, nevertheless, we are reporting a rare case of HOCT that resulted in death from tumor infection and rupture.

Case presentation

A 55-year-old female underwent a computed tomography (CT) examination during a diagnostic investigation of renal dysfunction. CT revealed bilateral nephromegaly caused by bilateral solid and cystic masses (Fig. 1-A). More than 20 tumors were found in the kidneys, with diameters ranging from 1 cm to 6 cm. Percutaneous needle biopsy of the largest tumor located in the lower pole of the right kidney was performed. The tumor was composed of cells with abundant granular eosinophilic oncocytic cytoplasm. Immunohistochemical staining of the tumor was positive for cytokeratin 7, CAM 5.2, and cluster of differentiation 117 and was negative for vimentin, inhibin, chromogranin, synaptophysin, and cluster of differentiation 10. These results suggested hybrid RO and CHRCC. Because of the patient’s wishes, watchful waiting was selected as the treatment strategy. Three years later, she was referred to our hospital in shock. Laboratory analysis on admission revealed severe liver failure (aspartate aminotransferase of 819 units/L and alanine aminotransferase 533 units/L) and inflammatory responses (white blood cell count 20700/μL and C-reactive protein 21.54 mg/dL). CT revealed enlargement of the largest tumor located in the lower pole of the right kidney (from 6 cm to 10cm), liver swelling, and ascites (Fig. 1-B). Nevertheless, the precise cause of shock could not be determined. We continued intravenous antimicrobial agents and continuous hemodiafiltration, but she died on the 3rd hospital day. With permission from the patient’s family, a postmortem anatomical evaluation was performed. This revealed multiple organ failure, including diffuse hepatic necrosis, alveolitis, pulmonary edema, and bleeding of various organs. There were also findings suggestive of peritonitis. The entire kidneys were occupied with multiple yellow-brown tumors and cysts (Fig. 2). Necrosis, hemorrhage, and abscesses were found in the largest right kidney tumor that was partially ruptured. Microscopically, tumor cells showed clear to eosinophilic cytoplasm, weak nuclear atypia and were positive for cytokeratin 7 (Fig. 3), consistent with CHRCC. There were multiple ROs and CHRCCs in the kidneys, with low amounts of normal kidney tissue. The pathological diagnosis of the tumor was HOCT. Therefore, the cause of death was thought to be peritonitis due to rupture of the infected HOCT. Written informed consent was obtained from the patient for the publication of this case report and any accompanying images.

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Renal oncocytosis is a condition in which both kidneys are involved by diffuse numerous oncocytic nodules. It has been suggested that HOCT may exist on a tumor spectrum between CHRCC and RO. The diagnosis of HOCT has been proposed for tumors with a solid architectural pattern made of eosinophilic cells displaying overlapping CHRCC and RO features. The proportion of cells immunoreactive for CK7 in each HOCT varies, consistent with our case.

HOCT have been described in three conditions: patients with Birt-Hogg-Dube syndrome (BHDS), patients with renal oncocytosis, and patients with no clinical features of either disease (sporadic). BHDS is an autosomal dominant disease characterized by skin fibrofolliculomas, associated with an increased risk of pulmonary cysts, spontaneous pneumothorax, and renal cancer (especially CHRCC, RO, and HOCT). Our patient had no sign of skin fibrofolliculomas and no history of spontaneous pneumothorax. Both kidneys were involved with several nodules. Therefore, we considered HOCT in our patient to be sporadic.

A prior report demonstrated that all sporadic HOCT were limited to the kidney with no infiltration of nearby tissue or distal metastasis, suggesting benign behavior. The prognosis of HOCT appears to be excellent, as no recurrence and metastasis with the mean follow-up 50 months were noted by Waldert et al. Therefore, we selected watchful waiting as the treatment strategy in consideration of the patient’s wishes.

Discussion

Renal oncocytosis is a condition in which both kidneys are involved by diffuse numerous oncocytic nodules. It has been suggested that HOCT may exist on a tumor spectrum between CHRCC and RO. The diagnosis of HOCT has been proposed for tumors with a solid architectural pattern made of eosinophilic cells displaying overlapping CHRCC and RO features. The proportion of cells immunoreactive for CK7 in each HOCT varies, consistent with our case.

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Nevertheless, she died from tumor rupture. Because spontaneous rupture of renal cell carcinoma is rare,\(^5\) we did not expect her death. Rupture of CHRCC occurs in only 4% of renal cell carcinoma. Therefore, this report is the first case in which HOCT was ruptured and followed a rapid course.

A careful history revealed that she had several urinary tract infections during the watchful waiting period. On postmortem anatomical evaluation, the ruptured tumor showed ascending urinary tract infection. In addition, she had been on hemodialysis 3 months prior to admission. Therefore, it might have been necessary to consider earlier nephrectomy.

Conclusion

HOCT seem to behave indolently as no evidence of malignant behavior was documented in prior reports. However, early resection is a safe and reasonable approach, depending on the patient’s condition.

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