RESIDENT'S CORNER

Bellini duct carcinoma with ovarian metastasis

W. Kassouf, MD, S. Binsaleh, MD, D. D. Cohen, MD, S. Carrier, MD

Division of Urology, Department of Surgery, McGill University Health Center, Montreal, Quebec, Canada

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We report a case of collecting duct carcinoma presenting

as pyelonephritis with hypercalcemia and metastasis to the ovary. Case management and literature review being presented.

Key Words: Bellini duct, collecting duct, renal neoplasm, paraneoplastic syndrome

Introduction

Bellini duct or collecting duct carcinoma, a rare type of renal cell carcinoma, remains poorly understood. This aggressive tumor has consensually been accorded a dismal prognosis and early diagnosis appears to be the only factor that may result in prolonged survival. Its low frequency of about 1% of all renal cell carcinomas limits the input of new clinical or therapeutic data. Almost all patients described in the literature were reported as individual case reports and few groups were able to follow them because the diagnosis of this tumor was based on rigorous histopathological examination.

Case report

A 79 year old female presented to the emergency room with 2-week history of confusion, and left flank pain. The patient was febrile 38.7 °C with a urinalysis that showed nitrites, white and red blood cells. Serum calcium and creatinine were elevated (3.76 mmole/l and 166 umole/l respectively). Patient commenced empirical intravenous antibiotics and scheduled for CT scan of the abdomen and pelvis. CT showed dilated left renal pelvis and hydroureter secondary to a psoas mass consistent with abscess. There was multiple microabscesses in the left kidney, and hilar lymphadenopathy. Patient subsequently underwent a left percutaneous nephrostomy tube insertion and fine

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Address correspondence to Saleh A. Binsaleh, Division of Urology, McGill University, 1212 Pine Avenue West, Montreal, Quebec H3G 1A9 Canada

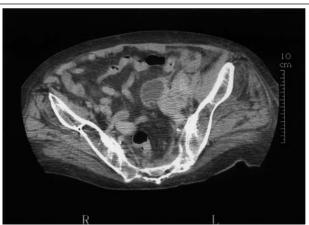
needle aspiration of the psoas mass. Both sites showed purulent fluid that grew E coli that was sensitive to the administered antibiotics. Patient defervesced, but white cell count and serum calcium did not normalize neither did the confusion state improve. With hydration, the creatinine normalized, however the serum calcium remained elevated despite treatment with calcitonin, pamidronate, and intravenous hydration. Work-up for hypercalcemia included a nuclear scan and CT of the neck to attempt to identify a PTH secreting tumor. CT neck showed two parathyroid adenomas that may have contributed to the persistent hypercalcemia. After the 14th day of intravenous antibiotics, patient underwent a sub-total parathyroidectomy. Our team was consulted at that time. Two day post-op, we decided to manage the patient surgically, given that there has been no improvement in the patient's condition, radiologically and clinically. Figures 1 and 2 for the follow-up CT. Intra-op, after a radical nephrectomy was performed, frozen sections were sent on the lymph nodes and psoas mass. Pathology showed high-grade malignant cells in both specimens. There was as well a large mass in the pelvis continuous with cancer. At this time, we decided to close the abdomen and the patient was sent to recovery room. Postoperative recovery was complicated by left deep vein thrombosis, and re-elevation of the serum calcium. Patient was declared palliative and expired post-op day 27. Pathology revealed an invasive collecting duct carcinoma. Autopsy demonstrated collecting duct carcinoma metastatic to the left ovary.

Discussion

Carcinomas arising from the medullary collecting duct or duct of Bellini have been reported as collecting duct carcinoma (CDC) or Bellini's duct carcinoma. They are uncommon tumors. Approximately 100 cases of CDC have been reported previously in the literature, including a large subset from East Asia.^{1,2} Studies have shown that abdominal pain, flank mass, and gross hematuria are the most common presenting symptoms. Constitutional symptoms such as fever may also be seen, but no particular paraneoplastic syndrome has been reported. The tumor typically shows aggressive infiltrative growth where metastatic disease is present at time of presentation in 40% of patients.^{2,3} Our patient presented as acute pyelonephritis complicated by multiple renal abscesses and severe paraneoplastic hypercalcemia. Up to our knowledge, this is the first case report of collecting duct carcinoma with metastasis to the ovaries.

Bellini duct carcinoma has a predilection to the right kidney (82%) for unknown reasons.^{2,3} There is usually a delay in diagnosis that is contributed to by difficulties in radiological diagnosis since characteristic





Figures 1 & 2. Follow-up CT abdomen/pelvis post percutaneous nephrostomy tube drainage and a period of 10 days of intravenous antibiotics.

morphology of a mass centered on the medulla with preservation of renal outline can mimic conditions such as xanthogranulomatous pyelonephritis. Because Bellini duct carcinoma was thought to be an atypical form of transitional cell carcinoma, methotrexate, vinblastine, doxorubicin, and cisplastin were given in some cases, whereas alternative therapy included immunochemotherapy; both of which showed marginal sustained response.⁴⁻⁶

Mejean et al⁷ studied the natural history in 10 patients diagnosed with this renal tumor and found that its clinical evolution rapidly leads to death for the majority of patients, suggesting that the currently applied therapeutic approach, notably surgery, is not adapted, especially when the tumor is large and infiltrates vessels. Henceforth when a large renal tumor has intraparenchymal development on CT, they suggested a fine needle biopsy should be performed systematically to exclude or confirm the diagnosis of collecting duct carcinoma. They also find that in metastatic collecting duct carcinoma radical nephrectomy alone does not appear to be useful except for palliation or in the context of new multicentric chemotherapy trials.

Conclusion

Collecting duct carcinoma of the kidney is a rare variant that is associated with an extremely poor prognosis, often associated with nodal and visceral metastases at presentation. Aggressive multimodality treatment is indicated to achieve best prognosis.

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