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# Hepatoid Carcinoma of Adrenal Gland: A Case Report

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***Authors' contributions***

*This work was carried out in collaboration between all authors. Author JA examined the patient. Authors NG and MM performed the literature search and prepared the text. Author MK performed the macroscopic examination of the surgical specimen. All authors read and approved the final manuscript.*

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***Case Study***

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## ABSTRACT

**Aims:** Hepatoid carcinoma is a rare tumor that mimics the morphologic and immunohistochemical features of hepatocellular carcinoma. An exceptional occurrence in adrenal gland has been recorded in the literature. The aim of this work is to study the morphologic immunohistochemical differential diagnosis and prognosis of adrenal hepatoid carcinoma.
**Presentation of Case:** We present a rare case of hepatoid carcinoma of left adrenal gland in 70-year-old-men. Histologically, the adrenal mass was composed of cells with enlarged nuclei, proeminent nucleoli and eosinophilic cytoplasm, arranged in a pseudoglandular and thick trabecular pattern with bile plugs, which resembled hepatocellular carcinoma. The main differential diagnosis was adrenal metastasis from hepatocellular carcinoma. The adrenal gland origin of the hepatoid carcinoma was verified by the absence of liver mass on clinical and imagery grounds.
**Conclusion:** We present the first case of adrenal hepatoid carcinoma in Tunisia. The main differential diagnosis was adrenal metastasis from hepatocellular carcinoma, which was eliminated by the absence of liver tumor.

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*Keywords: Hepatoid; adenocarcinoma; adrenal gland; histopathology; liver; hepatocellular carcinoma.*

## 1. INTRODUCTION

Hepatoid carcinoma (HC) is a rare tumor that mimics the morphologic and immunohistochemical features of hepatocellular carcinoma [1]. HC can originate from different organs [2–10]. The stomach is the most common site of this tumor. To the best of our knowledge, only two cases of HC of the adrenal gland have been reported in the English literature [11,12]. We present a case of HC in adrenal gland which represents the first report in Tunisia.

## 2. CASE REPORT

A 70-year-old man presented with abdominal pain and a left abdominal mass. A computed tomography scan of the abdomen showed 11.5 × 7.8 × 7 cm sized large heterogeneous mass of left adrenal gland. The liver was normal without mass or signs of cirrhosis. The levels of serum alpha-fetoprotein (AFP) were not assessed. Serological tests for hepatitis B virus surface antigen and hepatitis C virus antibody were negative. Angiography showed hypervascular tumor over the upper pole of the left kidney. Thoracoabdominal left-adrenalectomy was performed. Gross examination revealed a green solid unencapsulated tumor measuring 12 x 8 x 6 cm with focal hemorrhage and necrosis (Fig. 1).

The histologic study showed pseudoglandular and thick trabecular pattern with bile plugs. These structures were separated by sinusoid-like blood spaces. Tumor cells have abundant eosinophilic cytoplasm, enlarged nuclei with prominent nucleoli and mitosis. This tumor component was not associated with other type of neoplasm. It was surrounded by non neoplastic adrenal parenchyma (Fig. 2).

Bile plugs were stained green in Perls coloration. Immunohistochemically, the HC was diffusely stained for Hepatocyte-cell antibody but not for alpha fetoprotein (AFP), alpha inhibin, cytokeratin, keratin 19 and chromogranin. The immunostainig for CD31 revealed changes of “capillarization” (Fig. 3). The diagnosis of HC of left adrenal gland was reported. Three months postoperatively, the patient is still alive without any further therapeutic intervention.

**Fig. 1. Macroscopic findings: A green solid unencapsulated tumor with focal hemorrhage and necrosis**

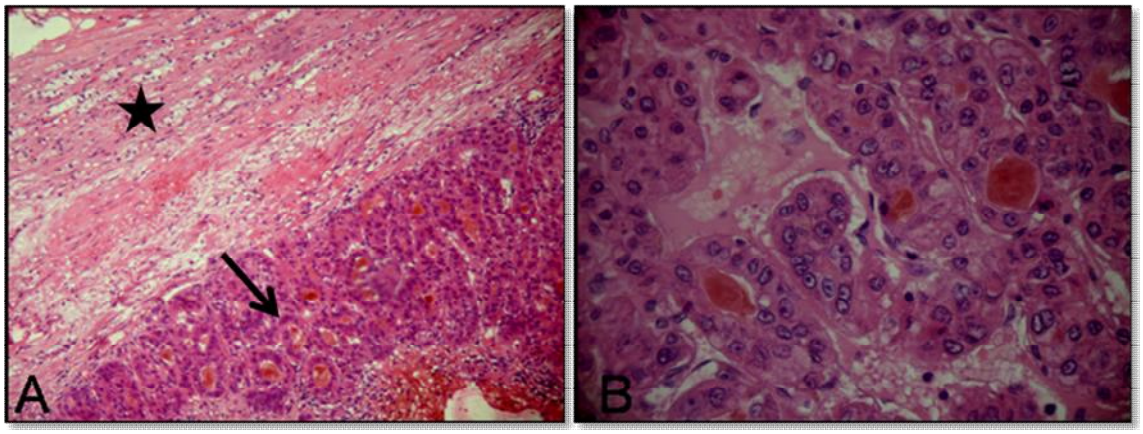


**Fig. 2. A: Tumor with a pseudoglandular and trabecular pattern and bile plugs (arrow), non neoplastic adrenal tissue surround tumor (★) (HEx100). B: Tumor cells with abundant cytoplasm, enlarged nuclei and prominent nucleoli (HEx400)**

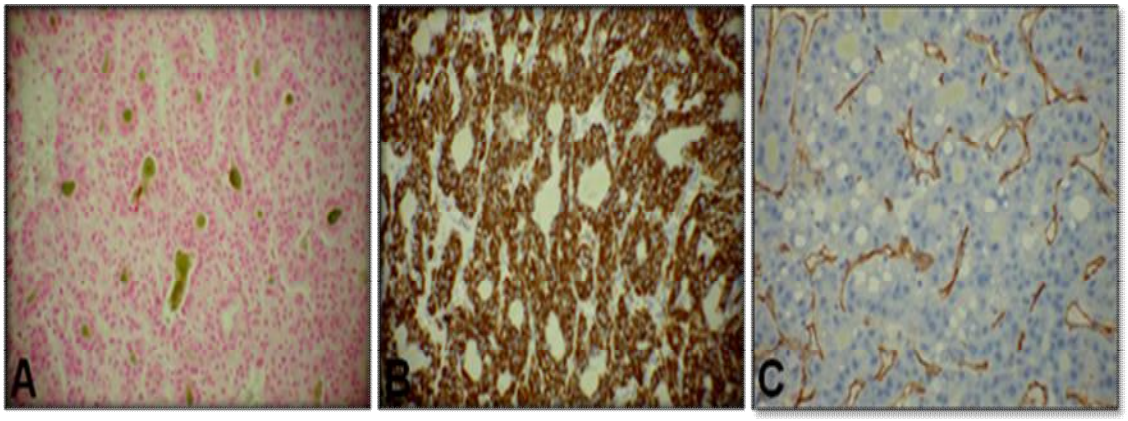


**Fig. 3. A: Green staining of bile plugs in perls coloration (HEx200). B: Intracytoplasmic expression of Hepatocyte-cell (HEx200). C: Immunostainig for CD31: capillarization (HEX200)**

## 3. DISCUSSION

Hepatoid carcinoma (HC) is a rare malignant epithelial neoplasm with significant component of hepatocellular differentiation. It usually occurs in the sixth or seventh decade of life and is more common in men [1]. HC was proposed as a specific type of primary gastric cancer by Ishikura et al. in 1985 [3]. Since then, HC has been described in a variety of anatomic locations including lung, pancreas, and gallbladder [2-10]. The adrenal location is exceptional. Only two cases of HC of the adrenal gland have been reported [11,12]. Hepatoid differentiation can occur in a pure form (like our case) or in association with ductal or a neuroendocrin neoplasm (hepatoid adenocarcinoma) [1]. Typically, an elevated level of serum AFP is detected. Although normal levels, like in our case, have also been reported [13]. Imaging studies are important for the exclusion of primary adrenal HC from metastasis of hepatocellular carcinoma in adrenal gland. The production of bile or fat and the accumulation of cytoplasmic material glycogen affect the macroscopic findings. HC is generally composed of large polygonal cells with abundant eosinophilic cytoplasm, enlarged nuclei and prominent nucleoli [1]. Fatty change and bile are present in well-differentiated HC. Most HC express AFP. The expression of hepatocyt specific antigen (hepatocyt paraffin-1) is more specific [14]. A canalicular pattern of labeling with antibodies to polyclonal CEA and CD10 can help establish the diagnosis in some cases. Adrenal cortical carcinoma is the most common malignant

epithelial tumor of adrenal gland. The architectural and cytologic features of this tumor differ from HC. Immunoreactivity for alpha-inhibin is sensitive but not specific for adrenal cortical carcinoma. In contrast to other epithelial tumors, adrenal cortical carcinoma is positive for cytokeratin and negative for epithelial membrane antigen. A metastasis to adrenal gland need to be excluded usually on clinical grounds before establishing the diagnosis of a HC primary to the adrenal gland [1]. Data on the prognosis of HC are minimal. Patients with little tumor confined to the adrenal and completely excised may pursue an indolent course.

## 4. CONCLUSION

We present the first case of adrenal HC in Tunisia. The main differential diagnosis was adrenal metastasis from hepatocellular carcinoma which was eliminated by the absence of liver mass on clinical and imagery grounds. Tumor size and capsular invasion should be the most important prognostic factor.

## CONSENT

All authors declare that 'written informed consent was obtained from the patient (or other approved parties) for publication of this case report and accompanying images.

## ETHICAL APPROVAL

Not applicable.

## COMPETING INTERESTS

Authors have declared that no competing interests exist.

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