Adrenocortical Adenoma Arising from Adrenohepatic Fusion: A Mimic of Hepatocellular Carcinoma-Case Report

Ann. Ital. Chir., 2024 95, 1: 13–16
pii: S0003469X24023299

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Aim: We present a case of adrenocortical adenoma originating from the adrenohepatic fusion (AHF) region, accompanied by advanced hepatosteatosis in the liver tissue, and discuss its distinction from hepatocellular carcinoma.
Case Experience: A 68-year-old male patient was admitted to the hospital following a fall from a height. He was referred to our hospital after an incidental discovery of a liver mass during an abdominal ultrasound examination. Subsequently, magnetic resonance imaging (MRI) imaging was conducted, followed by segmental liver resection with right adrenalectomy, and histological analysis of a biopsy from the lesion.
Results: Upon histologic examination, the case was determined to be an adrenocortical adenoma originating from the AHF.
Discussion: Adrenohepatic fusion (AHF) denotes the histological amalgamation of cells from the right adrenal cortex and right hepatic parenchyma. Only a limited number of cases of neoplasia originating from this region have been documented. These rare instances often present a diagnostic challenge, with preoperative imaging frequently misidentifying them as primary malignancies of either hepatic or adrenal origin, potentially leading to unnecessary extensive resections. The integration of immunohistochemical staining alongside clinical and radiological data proves helpful for accurately diagnosing this condition.
Conclusion: Awareness among clinicians, radiologists, and pathologists regarding the tumors that may arise from this region can mitigate the risk of performing extensive resections unnecessarily.

Keywords: adrenocortical adenoma; adrenohepatic fusion; hepatocellular carcinoma

Introduction

Adrenohepatic fusion (AHF) refers to the histological combination of cells from the right adrenal cortex and right hepatic parenchyma [1]. There is a complete or partial loss of the organ capsule between the liver and right adrenal gland at the fusion site [1]. AHF is diagnosed in approximately 10% of large autopsy series cases [2,3,4]. However, only a small number of cases of neoplasia originating from this area have been reported [5]. Adrenocortical adenoma originating in the AHF is often mistakenly diagnosed as hepatocellular carcinoma because of its rarity and similar radiological features with hepatocellular carcinoma [6]. We present a case of adrenocortical adenoma originating from the AHF region, accompanied by advanced hepatosteatosis in the liver tissue. The clinicopathological distinction between hepatocellular carcinoma and adrenocortical adenoma arising from the adrenohepatic fusion site was discussed.

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Case Presentation

A 68-year-old male patient presented to the hospital following a fall from a height. He was referred to our facility after an incidental discovery of a liver mass during an abdominal ultrasound. The patient's medical history includes hypertension, diabetes mellitus, and hepatosteatosis. Physical examination and laboratory test results were within normal limits. A contrast-enhanced abdominal magnetic resonance imaging (MRI) examination revealed a nodular lesion measuring 21 mm in diameter, located beneath the liver capsule near the right adrenal gland. This lesion exhibited a hypointense signal on T1-weighted images and was slightly hyperintense on T2-weighted images. Additionally, the MRI examination identified a nodular lesion localized in the right adrenal isthmus, characterized by well-defined boundaries, fat content, non-contrasting features, and a diameter of 17 mm, suggestive of an adenoma. Hypertrophic changes were observed in the left adrenal isthmus, with an increase in transverse diameter. Evaluation of liver tissue using dynamic contrast-enhanced MRI, with diffusion series incorporated to aid in the differential diagnosis, revealed a lesion in subsegment 7 of the liver. This lesion exhibited a more hyperintense internal structure compared to the surrounding liver parenchyma in the T2-weighted sequence, suggesting a mass lesion with malignant potential (Fig. 1).

Based on these findings, a surgical operation was scheduled to address the mass. Radiological images indicated two lesions, one located beneath the liver capsule and the other in the right adrenal gland. However, during the operation, it was perceived that the single mass originating from the right adrenal gland had invaded the liver. As a result, segmental liver resection and right adrenalectomy were performed. Macroscopic examination revealed a lesion measuring 4.5 × 3.5 × 2.5 cm, exhibiting a yellow-orange color and solid characteristics (Fig. 2).

Microscopically, the tumor, devoid of capsule structure, demonstrates nodular proliferation and is distinctly demarcated from the liver by a smooth border. In certain sections, liver tissue is interspersed among the tumoral nodules. Comprising cells resembling those of the adrenal cortex, the tumor forms solid islands and cords, characterized by large, lipid-rich, clear cytoplasm and mildly atypical nuclei (Fig. 3). No evidence of necrosis, mitosis, or lymphovascular invasion was observed. In focal areas, tumor cells exhibit narrower, oncocytic cytoplasm. Additionally, normal adrenal tissue surrounding the tumor, including the medulla, was identified in some sections.

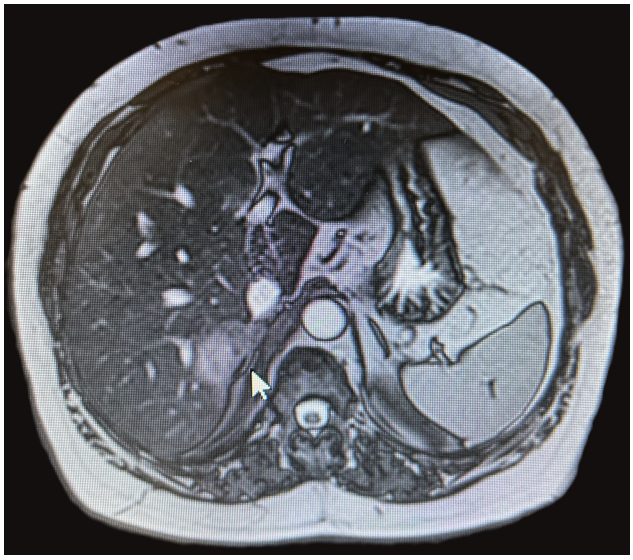

Fig. 1. Magnetic resonance imaging (MRI) images, the lesion is located under the liver capsule and is about the right adrenal in the lower sections (arrow head).

Results

In light of these findings, the case was evaluated as an adrenocortical adenoma developing from the adrenohepatic fusion site.

Discussion

AHF denotes the histological fusion of cells from the right adrenal cortex and right hepatic parenchyma [2]. Two

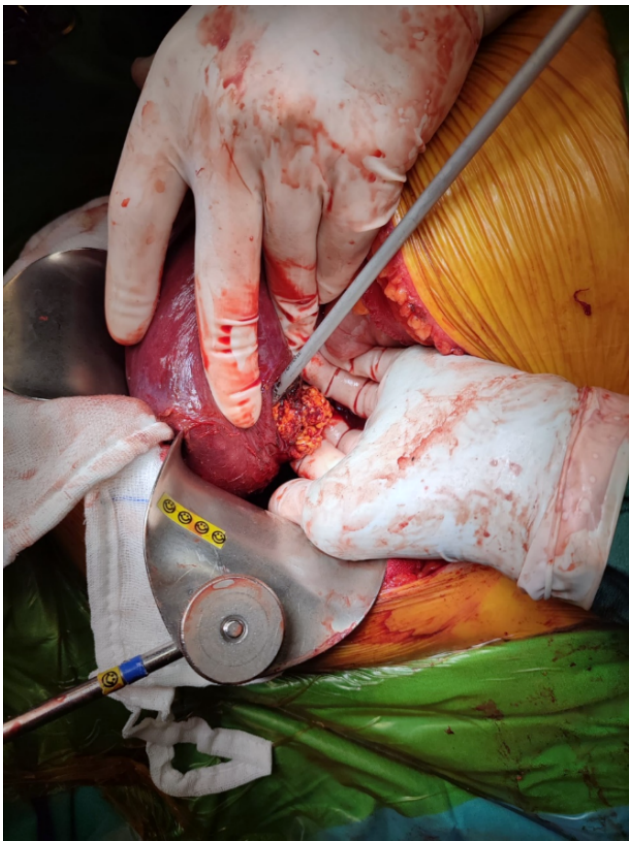

Fig. 2. Perioperative adrenal gland excision.

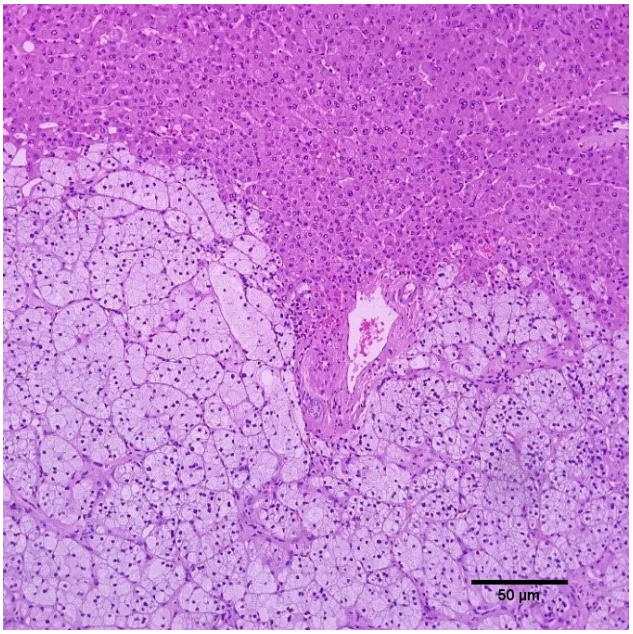

Fig. 3. Tumor cells with large clear cytoplasm on hematoxylin-eosin sections and normal liver parenchyma without surrounding capsule structure (HE×100).

mechanisms have been proposed for the formation of AHF. One theory suggests that a defect in mesenchymal tissue leads to a delay in capsule formation, resulting in AHF with

incomplete differentiation into fetal and subsequently adult-type fat cells. The other mechanism suggests that AHF is an acquired event [7].

AHF, defined by Honma [3] in 1991 as the histological combination of right adrenal cortex cells and right hepatic parenchyma cells, exhibits a detection rate of 9.9% in autopsy series encompassing 636 cases. Despite the seemingly non-rare incidence of AHF, instances of adrenocortical adenoma originating from this region are notably sparse. Adrenocortical adenomas arising from the AHF region lack specific imaging findings [5]. Consequently, these rare cases are often misdiagnosed as primary hepatic or adrenal origin malignancies during preoperative imaging, leading to potentially unnecessary large resections [2].

Preoperative tissue diagnosis assumes paramount importance in averting extensive resections in such scenarios. However, distinguishing histopathologically between adrenocortical adenoma and hepatocellular carcinoma arising from the AHF region poses a significant challenge, as they exhibit similar histological features [6]. In cases where clinical and radiological suspicions arise, differential diagnosis can be facilitated through immunohistochemical studies. Positive staining with immunohistochemical markers such as inhibin, melan-a, and synaptophysin can aid in diagnosing adrenocortical adenoma. However, if the biopsy material predominantly consists of tumor cells with oncocytic cytoplasm, as observed in our case, the differential diagnosis becomes even more challenging. Notably, there are documented instances of oncocytic adrenocortical adenoma originating from this region [8,9].

When encountering a granular mass within the liver, the presence of tumor cells exhibiting eosinophilic cytoplasm typically prompts consideration of hepatocellular carcinoma as the primary diagnosis. However, if the mass is adjacent to the right adrenal gland, and hepatic viral markers are negative while serum alpha-fetoprotein levels remain within normal range, inclusion of adrenocortical adenoma arising from the adrenohepatic fusion site in the differential diagnosis becomes imperative [9,10]. Utilizing immunohistochemical staining in conjunction with clinical and radiological data proves instrumental in accurately diagnosing this condition.

Conclusion

Clinicians, radiologists, and pathologists should be mindful of neoplasms that may arise from the adrenohepatic fusion site. Familiarity with the tumors that can originate from this region is crucial as it can help prevent unnecessary extensive resections.

Availability of Data and Materials

The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions

Study conception and design: RD, ÖS. Data collection and processing: RD, ÖS, PT. Analysis and interpretation of data: RD, ÖS. Writing important parts of the article: RD, ÖS. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript. All authors have participated sufficiently in the work and agreed to be accountable for all aspects of the work.

Ethics Approval and Consent to Participate

Since it is a case report, ethics committee approval was waved by Uludag University. However, informed patient consent was obtained for this study in accordance with the Declaration of Helsinki.

Acknowledgment

Not applicable.

Funding

This research received no external funding.

Conflict of Interest

The authors declare no conflict of interest.

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