

Adrenal Adenoma–Hemangioma: A Unique Collision Tumor

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ABSTRACT

Collision tumors are distinct and rare groups of neoplasms characterized by occurrence of two histologically different neoplasms at one site as a single lesion without significant tissue admixture. They are described in different parts of the body including adrenal glands. Collision tumors reported in relation to adrenal gland are mainly combinations of adenoma and metastasis. Here we present a case of incidentally detected adrenal collision tumor with adenoma and hemangioma as components which is a very rare and unique occurrence.

Keywords: Adrenal tumor, Adrenalectomy, Adenoma, Hemangioma, Incidentaloma.

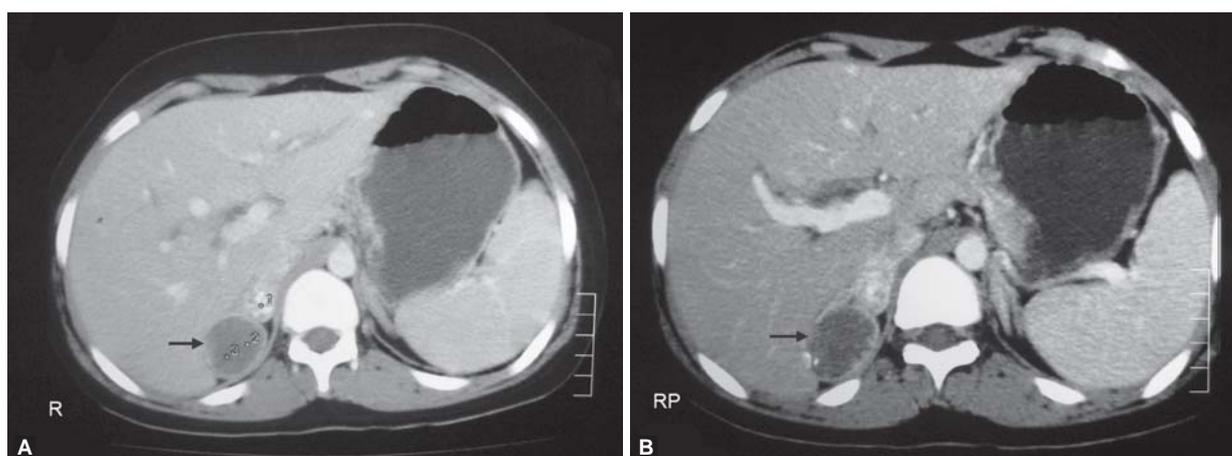
INTRODUCTION

Adrenal collision tumors are rare clinical entities in which two distinct types of neoplasms coexist in a single mass with a clear demarcation between the two. Most of the adrenal collision tumors described are combinations of adenoma and metastasis. Here, we discuss a case of adrenal adenoma—hemangioma collision tumor which is probably the second reported case of such kind.¹

CASE REPORT

A 17-year-old male patient presented with abdominal pain and jaundice of 2 weeks duration. The patient had no other associated symptoms. His general physical examination and examination of abdomen were unremarkable. Laboratory

investigations revealed an obstructive type of jaundice with serum total bilirubin of 10.6 mg%, conjugated bilirubin 6.6 mg% and serum alkaline phosphatase 535 iu/liter. All other laboratory investigations were normal. An extensive hematological work-up was done to rule out hemolytic anemias, but the findings were negative. An ultrasound abdomen showed cholecystolithiasis with choledocholithiasis producing biliary obstruction. Incidentally, a well-defined, predominantly hyperechoic, lobulated mass lesion involving right adrenal gland was noted. Subsequent contrast-enhanced CT scan of the abdomen revealed calculi at the distal common bile duct with cholecystolithiasis. Right adrenal gland showed well-defined, hypodense lesions with foci of discrete calcifications and tiny foci of fat (Figs 1A and B). Based on close scrutiny of cross-sectional and reconstructed images,



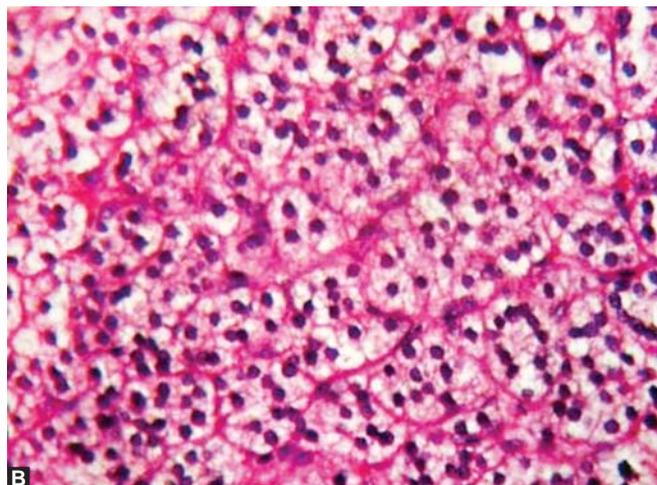
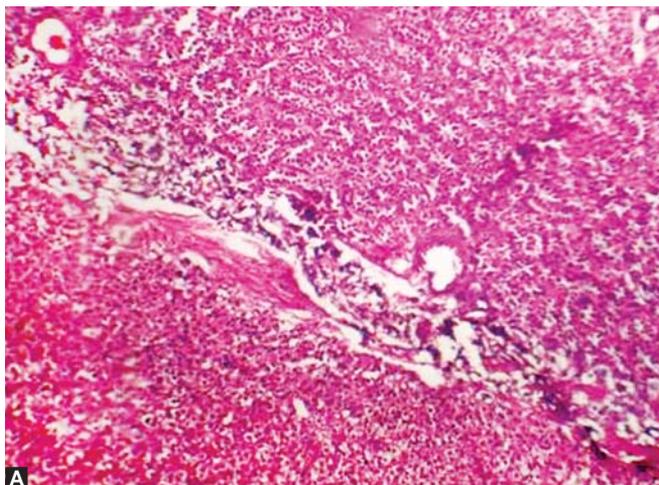
Figs 1A and B: Plain (A) and Contrast enhanced (B) CT scan (axial sections) showing collision tumor of the right adrenal gland

the lesions were interpreted as close yet discrete with one of the lesions showing initial peripheral and progressive centripetal enhancement; the possibility of adrenal collision tumor with one representing hemangioma and the other being adenoma/ganglioneuroma was considered. A further MRI study was not done due to financial constraints expressed by the patient's family. Confirmation of the adrenal incidentaloma prompted a detailed endocrinological work-up and hormonal assays, which were normal. Endoscopic retrograde cholangiographic stone removal was attempted but failed due to impacted stones. An open cholecystectomy and transduodenal CBD exploration and removal of CBD stones was done along with right adrenalectomy (Fig. 2).

Histopathological examination of the resected specimen revealed a collision tumor in the right adrenal gland composed of a hemangioma of size $5 \times 4 \times 2.5$ cm (with thin-walled endothelium lined spaces filled with blood and with areas of hemorrhage) and an adrenal cortical adenoma showing myelolipomatous changes, calcification and ossification of size $2.5 \times 2 \times 1$ cm (Figs 3A to 4B). There was a clear intervening fibrous septum between the two. Gallbladder showed evidence of chronic calculous cholecystitis.



Fig. 2: Vertically cut opened gross specimen demonstrating hemangioma in the upper half and adenoma in the lower half



Figs 3A and B: H & E stain 100 × (A) and 400 × (B) light micrograph of the adrenal adenoma

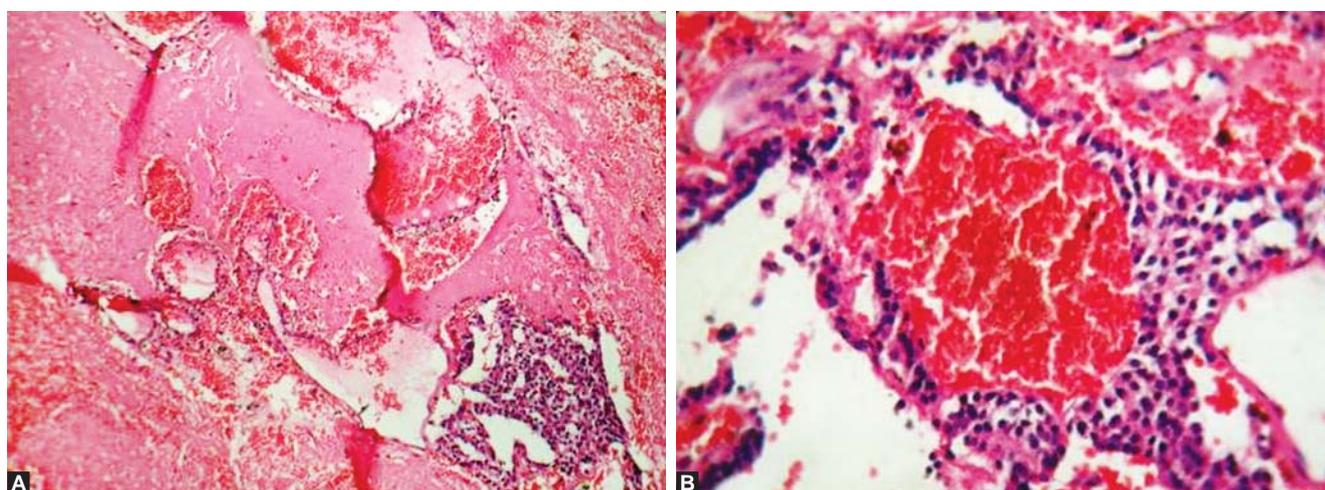
Postoperative recovery of the patient was uneventful and he was discharged on postoperative day ten.

DISCUSSION

Collision tumors are uncommon and represent the coexistence of two adjacent but histologically distinct tumors without histologic admixture. Distinction between collision and composite tumors is made based on the fact that collision tumors are independently existing neoplasms without significant tissue admixture, whereas composite tumors refer to coexisting neoplasms with considerable admixture of the two different cell types. Collision tumors are described in a variety of sites including lungs, bowels, genitourinary tract, meninges and lymph nodes other than adrenal glands.^{2,3} There are only very few reported cases of collision tumors in adrenals and most of them are combinations of adenoma and metastasis. To our knowledge, there is only one reported case of imaging features of adrenal adenoma–hemangioma collision tumor.¹

There are several hypotheses proposed of the possible pathogenesis of collision tumors. One theory is simple occurrence of two different lesions in contiguity by chance. Another hypothesis is that presence of a neoplasm in a particular organ alters the local environment making development of a second neoplasm more likely in that area. Pathology of collision tumors reveals two different types of neoplastic tissues coexisting with a sharp demarcation between the two and without a substantial admixture of histology at the interface. In adrenals frequently one is a benign tissue and the second a malignant metastasis⁴ and very rarely it can be two benign lesions as in our case.

These tumors are usually incidentally detected during work-up for some other disease. In the adrenals detection of incidentalomas has increased with the extensive use of cross-sectional imaging and most of these are adenomas or metastases.⁵ Preoperative diagnosis of individual components of adrenal collision tumors is again another challenge. In the available literature, different modalities have been tried to



Figs 4A and B: H & E stain 100 × (A) and 400 × (B) light micrograph of adrenal hemangioma

diagnose collision tumors. A high-resolution contrast-enhanced CT scan can most often differentiate individual component tissues. A homogenous mass with smooth borders and an attenuation value of less than 10 Hounsfield units on an unenhanced CT and a washout of more than 50% in delayed phase of contrast administration suggest a diagnosis of benign adenoma.⁶ Features of adrenal hemangioma are considered nonspecific with the classical centripetal enhancement being rare. Calcifications have been reported. In our case, the apparent difference in the morphology and enhancement pattern in association with apparent plane of cleavage between the lesions raised the suspicion of collision tumor. If the lesion is indeterminate with CECT, the next option is a chemical shift MRI which is an excellent tool in both anatomical description of the lesions and in characterization of component tissues.⁷ This is done by detecting difference in signal intensity according to the differential lipid content of the tissues. PET/CT scan has also been proven to be a good tool in differentiating benign from malignant components.⁸ Adrenal radiocholesterol scintigraphy is the most recent and noninvasive investigation for distinguishing adrenal cortical adenomas from lesions of extraadrenal origin.⁹ If the lesion is still uncertain with all these investigations then invasive methods like image-guided fine needle aspiration of the lesion or adrenal vein sampling may be tried and adrenalectomy is the last resort. The most important aspect during evaluation is an appropriate biochemical and endocrinological work-up to rule out hyperfunctioning endocrine neoplasms of adrenal origin like pheochromocytoma or Cushing's syndrome.

CONCLUSION

In the present era of advanced cross-sectional imaging, incidence of adrenal incidentalomas has increased. However, most of these tumors have nonspecific imaging features. So careful clinical, biochemical and radiological analysis may help in characterizing these lesions and avoid unwanted treatment.

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