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Case report

Adrenal intravascular papillary endothelial hyperplasia: A case report and literature review



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ABSTRACT

A 65-year-old Taiwanese man was admitted owing to the incidental finding of a left adrenal mass during a survey of gastrointestinal tract bleeding. Retroperitoneoscopic left adrenalectomy was performed smoothly. Pathology examination revealed intravascular papillary endothelial hyperplasia (IPEH). IPEH is a benign reactive intravascular process and must be distinguished from angiosarcoma carefully by histological characteristics. IPEH is mainly located in the head, neck, and upper extremities. By contrast, the incidence of intra-abdominal IPEH is extremely low and is often associated with gastrointestinal tract bleeding. Thus far, there are only four documented cases of adrenal IPEH. All cases underwent adrenalectomy and had no recurrence to date. Although the cases of adrenal IPEH are rare, we should still consider it as a differential diagnosis of adrenal mass accompanied with unexplained gastrointestinal tract bleeding.

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1. Introduction

Intravascular papillary endothelial hyperplasia (IPEH), also named Masson's hemangioma, was first discovered by Pierre Masson in 1923. IPEH is now believed to be a benign reactive intravascular process.¹ We present a case of adrenal IPEH, which was found incidentally during a survey of gastrointestinal tract bleeding. Patient consent for research was sought and granted. Furthermore, in this study we discuss the classification, differential diagnosis, and characteristics of IPEH and review all the published case reports of adrenal IPEH.

2. Case report

A 65-year-old Taiwanese man with a past history of type 2 diabetes mellitus was referred to our clinic for the evaluation of a left adrenal mass found incidentally during an image survey of gastrointestinal tract bleeding. The initial presentation was only an intermittent bloody stool without abdominal pain, weight loss, or recent trauma. Abdominal computed tomography revealed a 5.8-cm-sized heterogeneous mass with tiny internal calcification and poor contrast enhancement over the left adrenal region (Fig. 1A). Moreover, multiple bilateral renal cysts were noted. Laboratory

examinations were within normal limits, including serum potassium level (K, 3.8 mEq/L). Also, there was no hypertension, palpitation, or Cushing-like appearance. Under the impression of a nonfunctional adrenal tumor, retroperitoneoscopic left adrenalectomy was performed smoothly and the patient had an uneventful postoperative recovery.

Pathology report showed a 4.5 cm × 3.8 cm × 3.5 cm-sized reddish and yellowish solid tumor (Fig. 1B). Microscopically, the section of tumor revealed fibrin and a blood clot in the central area with papillary hyperplasia of endothelial cells, calcification, and recanalization (Fig. 1C). The residual adrenal cortical tissue was compressed to the peripheral area. The pathological findings were compatible with IPEH.

3. Discussion

IPEH is characterized by endothelial cells lining and proliferating around the thrombus or hematoma during venous stasis.¹ IPEH can be classified into three subtypes with distinct proportions: (1) pure form (55.8%, *de novo* process without comorbidity); (2) mixed form (39.9%, with preexisting vascular anomaly such as hemangioma or pyogenic granuloma); and (3) extravascular form (4.3%, with trauma-induced hematoma).^{2,3} It is important to distinguish IPEH from angiosarcoma because they are very similar in histology. When compared with angiosarcoma, IPEH is confined to the vascular lumen without obvious tumor necrosis and marked pleomorphism or mitosis.² The radiological feature of IPEH is nonspecific and described as a well circumscribed, heterogeneous

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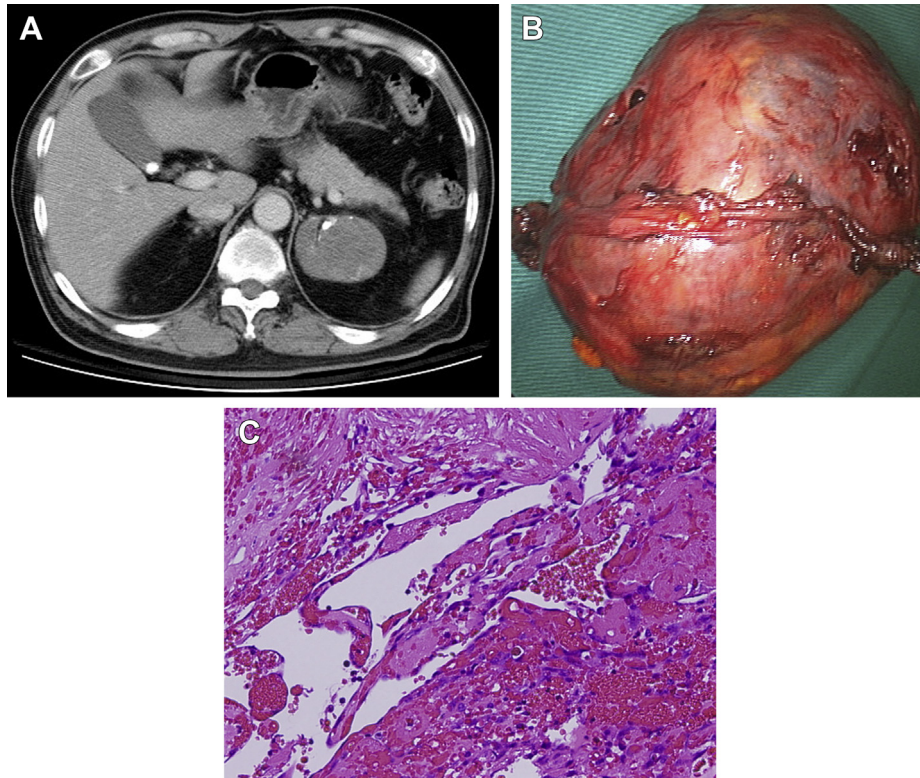


Fig. 1. (A) A 5.8-cm-sized heterogeneous mass with tiny internal calcification and poor contrast enhancement over the left adrenal region. (B) A 4.5 cm × 3.8 cm × 3.5 cm-sized reddish and yellowish solid tumor. (C) Fibrin and blood clot in the central area with papillary hyperplasia of endothelial cells, calcification, and recanalization.

mass with calcification and poor enhancement. Laboratory examination of IPEH is usually unremarkable.⁴

IPEH occurs mostly in the head, neck, and upper extremities.¹ By contrast, the incidence of intra-abdominal IPEH is extremely low. To date, there were around 20 documented cases of intra-abdominal IPEH, which are often related to gastrointestinal tract bleeding.¹ We reviewed the literature from PubMed and three cases of adrenal IPEH were found.^{4–6} In the four cases (including the current case), two cases were men and the other two were women. The mean age was 56 years (range, 49–65 years). All the tumors were pure form without other pre-existing lesions and were unilateral (2 cases were left, 1 case was right, 1 case did not state the laterality). The initial presentations were back pain (2 cases), hematuria (1 case), and gastrointestinal tract bleeding (1 case). The average maximal dimension was 6.13 cm (range, 4.5–8.5 cm). All four cases underwent adrenalectomy without complications and recurrence to date.

In this case, the initial presentation was gastrointestinal tract bleeding, which was thought to be unrelated to adrenal IPEH. Finally, gastrointestinal tract bleeding subsided spontaneously without further intervention and there was no definite diagnosis after serial endoscopic examinations of gastrointestinal tract, including panendoscopy, colonoscopy, and capsule endoscopy. However, we could not rule out the possible existence of IPEH in the gastrointestinal tract.

In conclusion, adrenal IPEH is an extremely rare but benign lesion. Moreover, it should be distinguished from angiosarcoma

carefully, and although rare, it should still be considered as a differential diagnosis of adrenal mass accompanied with unexplained gastrointestinal tract bleeding. Complete surgical resection is the curative treatment of adrenal IPEH if there is no preexisting vascular anomaly.

Conflicts of interest statement

The authors declare that they have no financial or non-financial conflicts of interest related to the subject matter or materials discussed in the manuscript.

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