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Adrenal hemangioma: A rare presentation of bleeding incidentaloma: Case report

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ABSTRACT

INTRODUCTION: Adrenal incidentalomas are tumors found accidentally by imaging. Among the incidentalomas, hemangiomas are quite rare and in certain cases their surgical intervention should never be overlooked.

PRESENTATION OF CASE: We present a case of 70 years old Lebanese female with an adrenal tumor presented as syncope found to have anemia on presentation and a bleeding 17 cm adrenal tumor on imaging. Patient had workup to rule out functioning adrenal tumors and decision to excise the tumor was taken after failure of embolization. Pathology report denied malignancy despite of the huge size and was in favor of hemangioma.

CONCLUSION: Adrenal hemangiomas are rare and they rarely present as syncope. Attention to such a life-threatening condition should be sustained. Embolization is primarily implied but one should never neglect the failure rate and the need for surgical intervention.

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1. Introduction

Adrenal gland tumors are among the commonest unanticipated tumors encountered [1]. Incidentalomas are lesions more than 1 cm discovered incidentally [1]. Classically there are two clinical concerns for incidentalomas whether they are functioning or having malignant potential. Indeed, most of the times incidentalomas are nonfunctioning adrenal adenomas [1]. Incidentalomas as their name imply rarely complicate, however they do complicate by bleeding. Precisely, AH (Adrenal Hemangiomas) are rare, nonfunctioning and benign vascular tumors [3]. They correspond for

0.01% of adrenal tumors [3]. Decision to operate is a variable of size, secretion and image findings [1,2]. However, in the suspicion of AH surgical indication should not be overlooked lest retroperitoneal bleeding occurs [4]. Here we present a case of AI with unusual presentation of syncope in addition to its enormous size compared to other reports [3,4].

This case was reported in accordance with the SCARE criteria [5].

2. Case description

A case of 70 years old Lebanese female presented to the emergency department for syncope. Patient was previously healthy, takes no medications and undergone no surgeries. She complains of syncope with no chest pain, dyspnea or weakness. Her vital signs were significant for tachycardia of 110 and physical exam was remarkable for diffuse abdominal tenderness. Workup in ER was significant for anemia with a hemoglobin of 8 g/dl. Patient undergone CT (Computed Tomography) scan which showed a 17 × 11 × 10 cm retroperitoneal bleeding mass originating from the left adrenal gland (Fig. 1).

Abbreviations: AH, adrenal hemangioma; CT, computed tomography.

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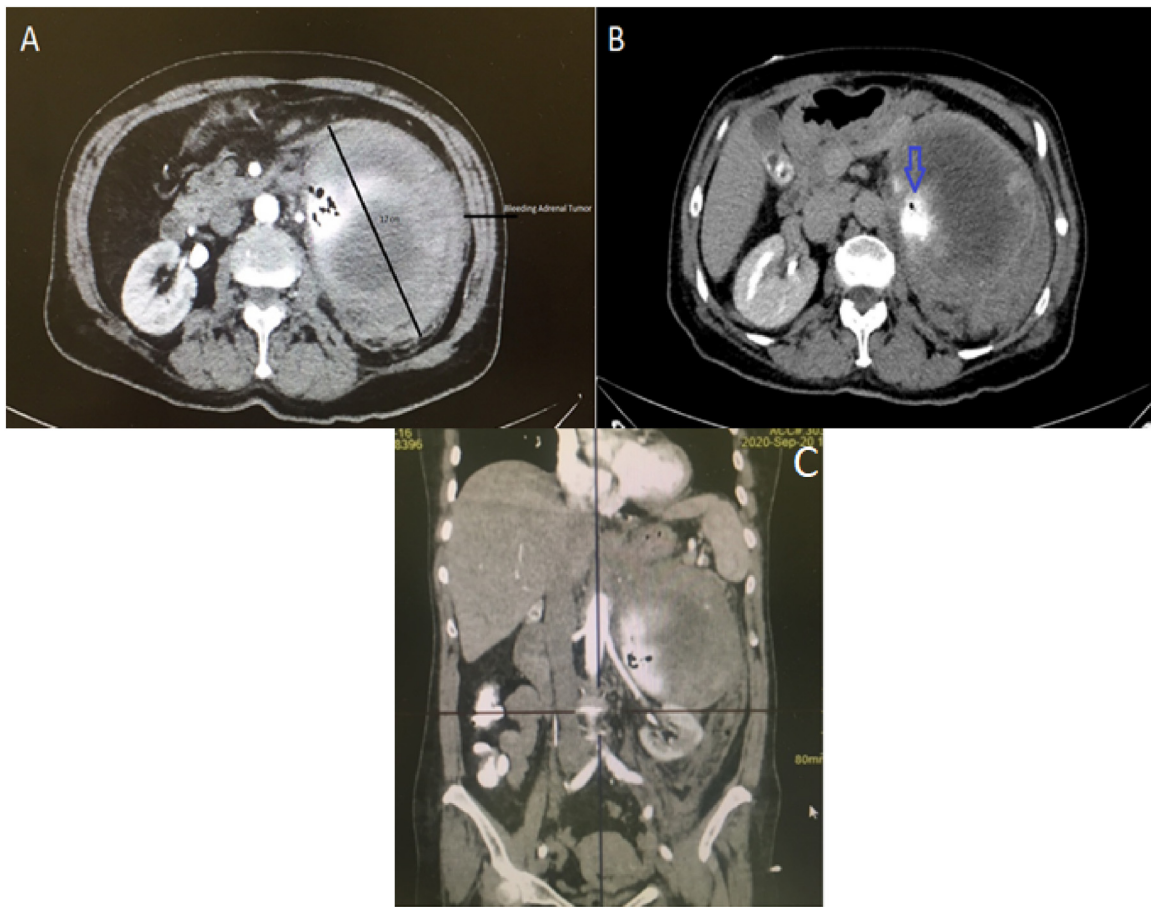


Fig. 1. CT scan of the upper abdomen in the portal (A) and delayed (B) phases. It shows a large left adrenal mass (17 cm) with surrounding hematoma and peripheral nodular enhancement. Note is made of the presence of contrast media and air bubbles from the embolization attempt (Arrow). Figure C shows the coronal section.

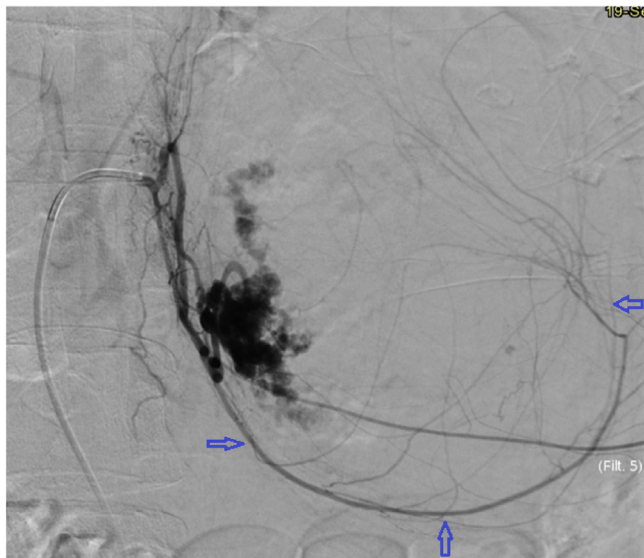


Fig. 2. Digital Subtraction Angiography (DSA) of the left adrenal artery showing a large lesion with centro-medial enlarged tortuous arteries and a peripheral thin capsular vascularity (Arrows).

Eventually, decision for embolization was taken. Interventional radiologist was able to identify the bleeding however an attempt to embolize failed to stop the bleed due to inability to identify the nourishing artery (Fig. 2).

In the meantime, the patient took 4 units of PRBC and had fluid resuscitation. Fortunately, she was hemodynamically stable and her hemoglobin level stabilized at 9 g/dl on the following day. Workup for secreting adrenal tumor was taken however 24 -h urine collection failed to identify increased levels of metanephrines and VMA (Vanillylmandelic acid). Moreover, serum cortisol level was within normal range and aldosterone to renin ratio failed to suffice to diagnose pheochromocytoma, cortisol – secreting tumor or aldosterone – secreting tumor respectively. Therefore, decision to operate and excise the tumor was taken. In the operating room, a midline laparotomy incision was made, opening the retroperitoneal space, then identification of the mass which was hyper-vascularized and adherent to the spleen, vertebral, aorta and the left kidney. Ligation of the left adrenal vein and artery and other collaterals was secured and the mass was excised and sent to pathology (Fig. 3). Patient had an unremarkable post-operative recovery and was discharged on day 5. Moreover, pathology report was in favor of a 17 cm hemangioma with no malignant findings.

3. Discussion

Incidentalomas of the adrenal gland carry a risk for cancer when they have a size above 4 cm and in such a case, they require resection [2]. On the other hand, AH- although benign in nature- have a risk for hemorrhage [4] and their management seems perturbing. In addition, it is sometimes difficult to differentiate AH from adrenal cortical carcinoma on imaging [6] hence surgical indication seems reasonable to prevent retroperitoneal bleed, reduce pressure effect and to exclude malignancy. Precisely, there are 4 types of

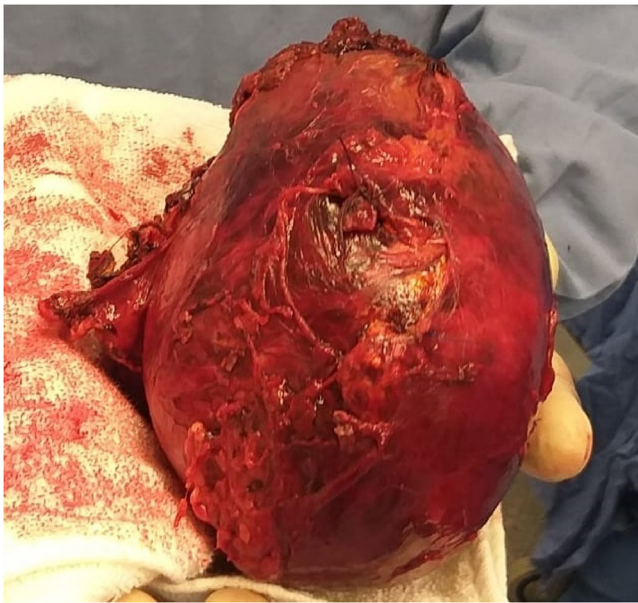


Fig. 3. Intra operative image of the excised left adrenal tumor.

AH: cavernous hemangioma, venous hemangioma, capillary type hemangioma, and mixed hemangioma [7]. Despite the advance in imaging techniques, differentiating the types of adrenal incidentalomas remains challenging [7] and most of the AH are diagnosed post-operatively similar to our case. In cases where malignancy can be excluded pre-operatively, choosing laparoscopic approach is justifiable and it is now considered the standard of treatment for benign adrenal lesions [8].

To our knowledge, our case has the largest size of an AH being reported in the literature with a 17 cm mass compared to previous studies of 12 cm maximally [8]. For instance, we elected for open approach since we couldn't rule out malignancy risk and due to its size. In addition, the presentation of our patient makes this report unique in that the AH bled in the absence of trauma or inciting event causing her syncope.

AH carry a challenging task for interventional radiologist to stop the bleeding due to wide and complex arterial network of the adrenal gland normally [9] not to mention the hypervascularity nature of the AH and the inability to identify the nourishing vessel similar to our case.

In a nutshell, due to the inability to rule out malignancy by pre-operative imaging, the risk for hemorrhage and the failure rate of radiological intervention due to the hypervascularity nature of the tumor, surgical intervention remains the proper treatment of AH.

4. Conclusion

AH is typically discovered incidentally, however our case presented with syncope due to a bleeding AH. In addition, a 17 cm AH is the largest tumor of this kind reported. Due to the challenging preoperative diagnosis, malignancy and the hemorrhage risk we found this report to endorse the need for surgical intervention in a judicious timely manner.

5. Study limitations

This is a case report which lacks solid evidence on the chance of bleeding of AH and the proper management. Further studies with

statistical background should be pursued to answer the questions proposed by this paper.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

The study type is exempt from ethical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

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