Amyloidosis in adrenal incidentaloma: Finding by laparoscopic adrenalectomy

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Case Report

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Abstract: An adrenal incidentaloma (AI) is an asymptomatic adrenal mass at least 1 cm in diameter detected on imaging studies performed for non-adrenal related disease. Amyloidosis is a rare disease and its presentation as an adrenal gland incidentaloma without systemic disease is exceptional. We present a 62-year-old woman with history of blunt abdominal trauma secondary to a vehicle accident 18 months ago. During her clinical assessment, an abdominal CT-scan was performed showing an AI with malignancy-suspicious features. Laparoscopic right adrenalectomy was performed. Histopathological examination exhibited amyloid infiltration in the hematoxylin and eosin (HE) staining, and it was confirmed by a positive Congo Red (CR) staining and a positive kappa and lambda chains immunohistochemistry.

Key Words: Adrenal incidentaloma, laparoscopic adrenalectomy.

Introduction

An adrenal incidentaloma (AI) is an asymptomatic adrenal mass at least 1 cm in diameter detected on imaging studies performed for non-adrenal related disease. As imaging techniques evolve, the incidence of AI has increased in the last decades. Autopsy studies suggest a prevalence of clinically unapparent adrenal masses of nearly 2% and radiological studies report a frequency of approximately 3% in the age of 50 years, which increases up to 10% in the elderly.[1] Laparoscopic adrenalectomy is trend in the operative management of AI.[2] Only a few cases in the current scientific literature have been described regarding histopathological findings of amyloidosis in AI.[3,4]

Case report

A 62-year-old female came to the endocrine surgery outpatient clinic for evaluation of an AI identified by and abdominal CT- scan performed due to an abdominal blunt trauma associated with a vehicle-accident 18 months ago. Past medical history included episodes of sinus tachycardia associated with predominantly nocturnal diaphoresis and arterial hypertension in the last 24 months, controlled with atenolol 25 mg twice a day. CT-scan showed a hypodense right adrenal mass of 12 x 36.9 x 13.4 mm and lobulated edges (Figures 1-A, and 1-B). Non-contrast phase displayed a 37 HU attenuation with a significant enhancement in arterial phase and an absolute washout of 27% (Figure 1-C).
Biochemical work-up showed: WBC count 5.11 x 10^3/µL, hemoglobin 14.3 g/dL, hematocrit 42.9%, platelets 279 x 10^3/µl, INR 1, serum glucose 101 mg/dL, serum creatinine 1.0 mg/dL, serum sodium 141 mEq/L and serum potassium 4.4 mEq/L. 24 hour-urine metanephrines and serum catecholamines between the normal range.
A diagnosis of a non-functional right AI with imaging features suspicious of malignancy was stablished; laparoscopic right adrenalectomy was offered to the patient, which she consented. Surgery went uneventful; with the following intraoperative findings: lobulated right adrenal with unusual adherence to the liver and an adrenal vein of 12 mm length. The patient was discharged without complications in postoperative day 2.
Histopathological examination exhibited amyloid infiltration in the hematoxylin and eosin (HE) staining (Figure 2-A), confirmed with a positive Congo Red staining (Figure 2-B) and a positive kappa and lambda chains immunohistochemistry (Figure 3-A, and 3-B). Based on the previous findings, further clinical assessment was performed by the Hematology department with a negative bone marrow and periumbilical fat tissue biopsies, including Congo Red staining looking for amyloid infiltration.

Discussion

When an AI is discovered, assessment should focus on determining the functional status of the tumor, the risk for malignancy and if there are associated symptoms reported by the patient. [5] Laparoscopic adrenalectomy is the gold standard for the treatment of most adrenal masses, as has been proved to reduce postoperative pain, length of stay,

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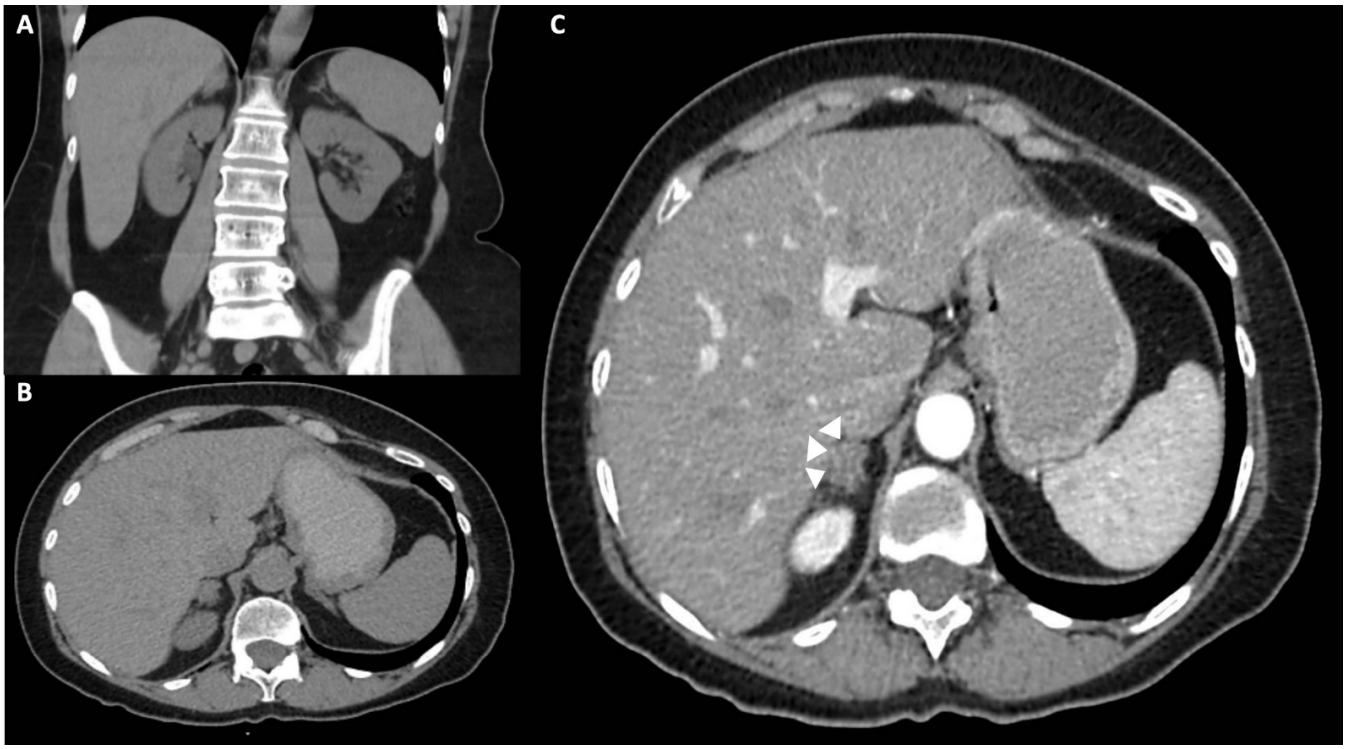

Figure 1. A-B Non-contrast coronal and axial CT scan slides show a 12x36.9x13.4mm hypodense right adrenal mass with lobulated edges. **C** Contrasted axial CT scan slide shows enhancement and 27% absolute washout of the adrenal incidentaloma (white arrow).

and intraoperative blood loss, as well as faster recovery time.[1] In our case, we performed a laparoscopic right adrenalectomy due to suspicion of malignancy for the imaging features in the CT scan with the incidental histopathological finding of amyloidosis, which was confirmed with Congo Red staining.

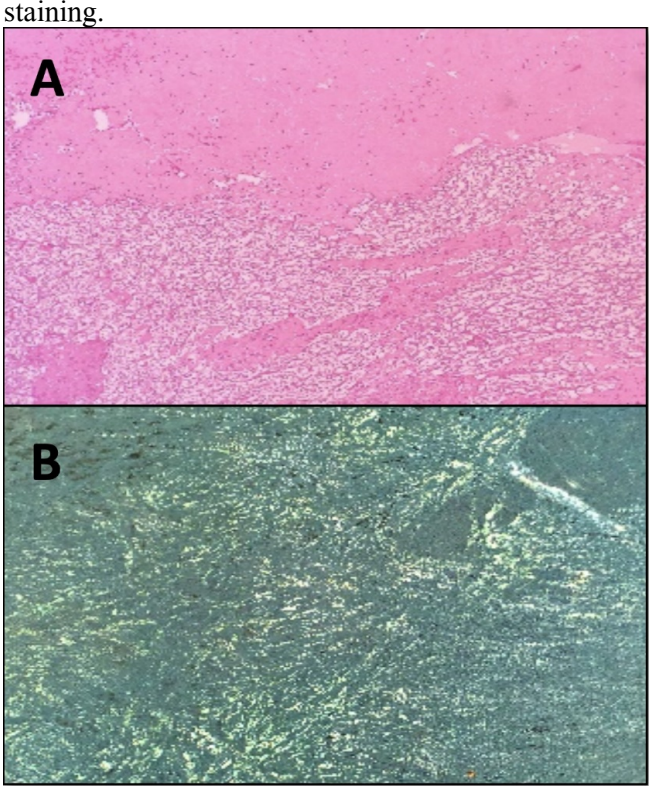

Figure 2. A Hematoxylin and eosin staining photomicrography of amyloid depositions in the adrenal gland cortex (10x). **B** Congo red staining showing apple green birefringence under polarized light (10X).

Systemic amyloidosis is a multi-system disease caused by fibrillary protein deposition with ensuing dysfunction of the affected organ systems. The main types of amyloid include amyloid associated (AA), amyloid light chain (AL), and amyloid transthyretin (ATTR). The most common presentation is cardiac (75-80%) and renal (65%) amyloidosis. [4]

This is a rare presentation of an amyloidosis case in an adrenal incidentaloma. Few cases have been showed in the scientific medical literature, Trevor et al. [3] described a case with leukocyte cell-derived chemotaxin2 (LECT2)-associated amyloidosis in the adrenal gland in a patient with monoclonal gammopathy of undetermined significance (MGUS). Furthermore, Thakur et al.[4] reported a case of amyloidosis presenting as right adrenal mass diagnosed on endoscopic ultrasound-guided fine needle aspiration.

The diagnosis is histological through the identification of amyloid depositions in biopsies of periumbilical fat, bone marrow or even the apparently affected organs.[4] This patient was studied by Clinical Endocrinology and Hematology who ruled out the presence of disease in other organs with the negative results of the bone marrow and periumbilical fat tissue biopsies.

Conclusion

Amyloidosis is a rare disease, the involvement of the adrenal without systemic disease is exceptional. There are no clinical manifestations for its suspected diagnosis and conservative management would possibly be an alternative.

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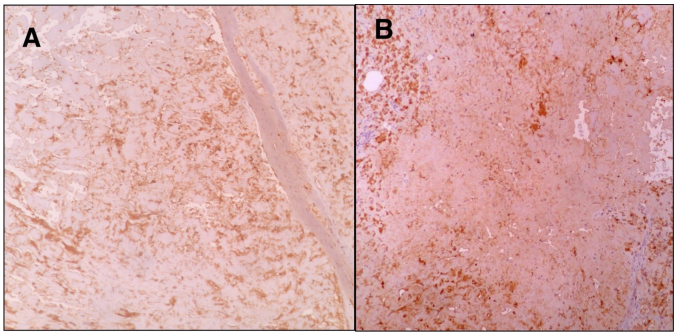

Figure 3. Positive kappa (**A**) and lambda (**B**) chains immunohistochemistry.

Conflicts of interests

There was no conflict of interest during the study, and it was not funded by any organization.

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