Clinical Case Report

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Simultaneous occurrence of primary aldosteronism due to aldosteronoma and ectopic meningioma in the adrenal gland

A case report

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Abstract

Rationale: Primary aldosteronism due to aldosteronoma is the most common form of secondary hypertension, with an estimated prevalence of 4% of hypertensive patients in primary care and around 10% of referred patients. Diagnosis is a clinical challenge with simultaneous occurrence of primary ectopic meningioma in the adrenal gland. To our knowledge this is the first reported case of simultaneous occurrence of aldosteronomas and ectopic meningioma in the adrenal gland based on literatures.

Patient concerns: A 30-year-old man presented with resistant hypertension for one year. The computed tomographic scans were suggestive of left adrenal gland hyperplasia.

Intervention: The patient underwent partial unilateral laparoscopic adrenalectomy.

Diagnosis: The histopathological examination of the resected sample confirmed primary ectopic meningioma in adrenal gland and aldosterone producing adenoma (APA). The saline load test, captopril test, and plasma aldosterone/renin ratio were indicative of primary aldosteronism (PA).

Outcomes: The patient had controlled blood pressure postoperatively.

Lessons: The patient was diagnosed with PA due to APA and nonfunctional primary ectopic meningioma in the adrenal gland which is very rare and dealt with unilateral laparoscopic adrenalectomy.

Abbreviations: ACTH = adrenocorticotropic hormone, APA = aldosterone producing adenoma, ARR = angiotensin to renin ratio, CT = computed tomography, PA = primary aldosteronism, WHO = World Health Organisation.

Keywords: aldosterone producing adenoma, partial laparoscopic adrenalectomy, primary aldosteronism, primary ectopic meningioma in the adrenal gland, resistant hypertension

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DR, KS, and MS have equally contributed to this study.

Ethics approval and consent to participate: Written informed consent was obtained from this patient. The Ethics Committees of the First Affiliated Hospital of Nanjing Medical University approved the study.

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1. Introduction

Primary aldosteronism (PA) has an estimated prevalence of 4% of hypertensive patients in primary care and around 10% of referred patients. It is the most common form of secondary hypertension.[1] PA, with a prevalence of 14% to 21% is particularly common in patients with resistant hypertension.[2] Resistant hypertension is defined as systolic blood pressure of ≥140 mm Hg, diastolic blood pressure of ≥90 mm Hg, or an elevation of both, including the use of at least 3 antihypertensive medications from different drug classes, preferably including a diuretic.[3] The overproduction of aldosterone independently from the renin-angiotensin system is a characteristic of PA.[4] Around 40% of PA cases are accountable by aldosteronomas, also known as aldosterone producing adenomas (APAs). APAs are small benign tumors of around 1 to 3 cm.[5] Moreover, primary ectopic meningiomas are exceedingly rare, especially in the adrenal gland as none was found before based on literatures.[6] Such ectopic meningiomas may pose diagnostic difficulties for clinicians. We hereby report a case of simultaneous occurrence of primary aldosteronism (PA) due to aldosteronoma and primary ectopic meningioma in the adrenal gland, which is very rare based on literatures.

2. Case presentation

A 30-year-old male patient came to our hospital with a history of resistant hypertension for one year. Despite being on 3 antihypertensive drugs, his blood pressure was at 170/115 mm Hg. He had no history of hypokalaemia and the plasma aldosterone/renin ratio (ARR) was 75. Laboratory examination tests showed: plasma ACTH: 40.1 (800 hours), 22.9 (1600 hours), 6.02 (0 hour) pg/mL. Plasma cortisol: 438 (800 hours), 274 (1600 hours), 121 (0 hour) nmol/L. Before the saline load test: aldosterone (supine) 122.90 ng/L, rennin activity (supine) 0.13 μg/L/h, angiotensin I 0.16 μg/L, angiotensin II (supine) 52.29 pg/mL, potassium 3.6 mmol/L. After the saline load test: aldosterone (supine) 97.84 ng/L, rennin activity (supine) 0.16 μg/L/h, angiotensinI0.12 μg/L, angiotensin II (supine) 51.60 pg/mL potassium 3.9 mmol/L; Captopril test: 0 hour: aldosterone (standing) 94.04 ng/L, renin activity (standing) 0.29 μg/L/h (low), angiotensin I 10.27 μg/L, angiotensin II (standing) 57.34 pg/mL, cortisol (8 hours) 29.0 nmol/L. 1 hour aldosterone (standing) 94.44 ng/L, renin activity (standing) 0.33 μg/L/h (low), angiotensin I 0.22 μg/L, angiotensin II (standing) 55.19 pg/mL, cortisol (8 hours) 29.0 nmol/L. 2 hours aldosterone (standing) 112.70 ng/L, rennin activity (standing) 0.18 μg/L/h (low), angiotensin I 0.12 μg/L, angiotensin II (standing) 76.61 pg/mL, cortisol (8 hours) 29.0 nmol/L. The computed tomographic (CT) scan showed left adrenal gland hyperplasia with nodular protrusions (Fig. 1). Unilateral partial laparoscopic adrenalectomy was performed and a resected specimen of 1.0×1.1 cm was sent for pathological examination which showed cortical hyperplasia with microadenoma and focal spindle cell hyperplasia. Moreover, the immunohistochemical analysis showed: spindle cell CD34 (−), Ki-67 (scattered +), s-100 (−), SMA (−), Desmin (−), syn (−), PR (+), EMA (+) which were consistent with small meningioma-like nodules (Fig. 2). The patient was therefore, diagnosed with PA due to APA and ectopic meningioma of the adrenal gland. His postoperative blood pressure became approximately 123/61 mm Hg. On follow-up of 1-year duration, the patient has a good control over his blood pressure of around 124/80 mm Hg, with only one beta-blocker drug.

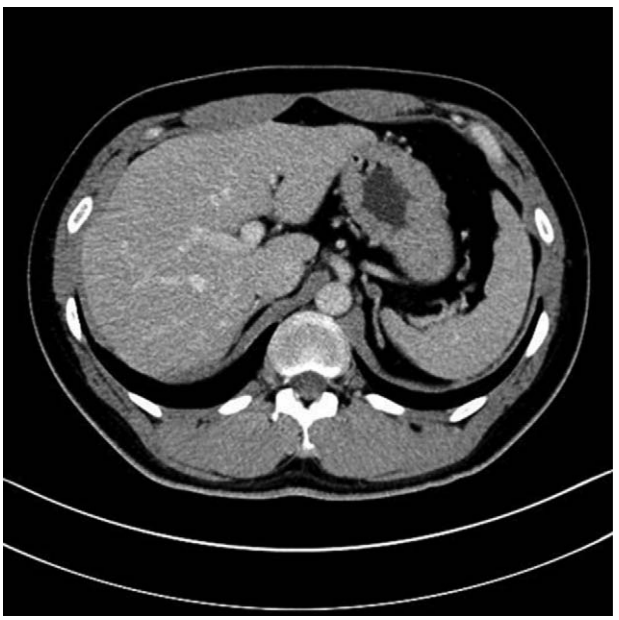

Figure 1. CT scan showing left adrenal gland hyperplasia and nodular protrusions. CT = computed tomography.

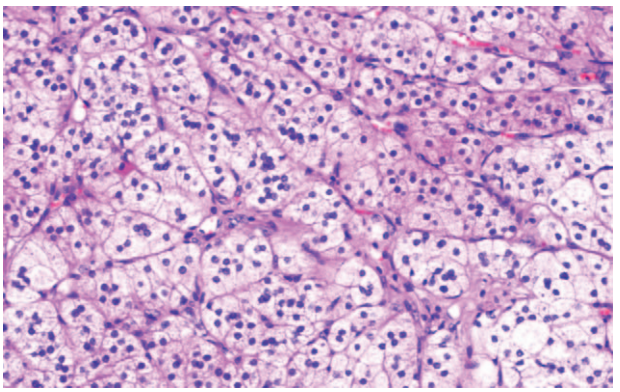

Figure 2. The immunohistochemical appearance of resected sample (magnifying power ×100).

3. Discussion

An autonomous aldosterone production which is inappropriately high for sodium state and mal-regulation by angiotensin II are characteristics of PA. Aldosteronomas also called APAs are responsible for 40% of cases.[5] Adrenal CT must be carried out to exclude adrenocortical carcinoma. It is crucial to distinguish between APAs and bilateral hyperplasia, through adrenal CT, as APAs are treated via adrenalectomy.[5] Usually unilateral laparoscopic adrenalectomy is indicated for patients with APA. Moreover, unilateral adrenalectomy has shown to decrease cardiovascular morbidity caused by excess of aldosterone.[7] Hypertension is cured in about 35 to 80% of patients with APA, after unilateral adrenalectomy.[5]

Furthermore, the aldosterone-to-renin ratio is the most sensitive test that screens for PA. The most commonly adopted plasma aldosterone/renin ration cut-off for PA screening is 30[5] and for our case the value was 75. The absence of hypokalaemia contributes to the rarity of our case.

Besides, the simultaneous occurrence of primary ectopic meningioma in the left adrenal gland was a clinical challenge. Meningiomas are usually clinically inactive.[8] A common feature of meningioma elongated spindle cells on the H-E staining,[9] like our case. The diagnosis of meningioma is further supported by the positive staining for EMA. Expression of proliferation marker such as Ki-67, has generally shown progressive increases in labeling index with a WHO grade from 1.00% to 1.35% for grade I to 1.90% to 9.30% for grade II or atypical meningioma.[10] This data confirm that the classification of meningioma, in this case, is atypical category. A diagnosis of nonfunctional ectopic meningioma was adopted for our patient.

4. Conclusion

Therefore, we conclude that simultaneous occurrence of PA due to APA and primary ectopic meningioma in the adrenal gland represents a clinical challenge. Based on our experience, unilateral laparoscopic adrenalectomy combined with saline load test, captopril test, plasma aldosterone/renin ratio, and histological findings of the resected sample was a good approach which resulted in overall survival ad good quality of life of the

patient. However, the limitation is that it is a single case study and conclusion can only be made based on our experience.

Author contributions

Dadhija Ramlagun, Kamleshsingh Shadhu, Miaomiao Sang, Kai Zhu, Chao Qin, and Min Sun all have made substantial contributions to conception, acquisition of data, analysis, and interpretation of data. All of them have been involved in drafting the manuscript and revising it critically for important intellectual content. All authors read and approved the final manuscript and take public responsibility for appropriate portions of the content and agreed to be accountable for all aspects of work.

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