CLINICAL CASE

ADRENAL SURGICAL APPROACH IN A WOMAN WITH SYNCHRONOUS BILATERAL ADRENAL TUMORS

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Abstract

Bilateral adrenal tumours are very rare and display various endocrine profiles. The unilateral approach is indicated if a nonsecretor contralateral pattern is suspected in order to avoid adrenal insufficiency.We present a 48-year-old woman with a history of bradimenorrhea at the age of 44. The gynecological evaluation was negative but two adrenal tumours were found. CT revealed a right adrenal tumour of 2 cm and a left one of 2.4 cm. Low-normal ACTH and high serum chromogranin A were confirmed. After one year, the left tumour increased to 4 cm so laparoscopic transabdominal left adrenalectomy (with 3 trocars) was successfully performed without any incident. No conversion was required. Neither intraoperative nor postoperative complications occurred. The patient was hospitalised 5 days and the wounds healed normally. The pathological report revealed a benign corticoadenoma. The endocrine evaluation after one week showed that after surgery ACTH de-suppressed to normal and chromogranin A normalised. The right adrenal tumour was followed-up for another year and the diameters were stationary. The patient was still hypertensive and she was offered adequate therapy (which we considered essential for high blood pressure). Life-long periodical check-up is necessary.Unilateral adrenalectomy in patients with both side tumours allows an adequate pathological report and avoids unnecessary contralateral surgery if an incidentaloma is revealed, thus allowing the preservation of adrenal function.

Keywords*: bilateral adrenal tumors, adrenalectomy, incidentaloma*

Introduction

Laparoscopic surgery represents an elegant option in approaching the adrenal tumors regardless of their endocrine profile. The procedure is safe and minimally invasive with a good outcome [1]. Both the functioning and nonfunctioning adrenal masses may be approached, the main issues being related to the patient's comorbidities, the endocrine profile and the associated surgical risk, as well as the tumour sizes [2]. The suspected malignancy represents a more complex aspect: the suspicion is based on size and radiological features [3,4].

An initial laparoscopic procedure is recommended in tumors less than 4 centimeters (cm) diameter but the need to conversion is frequently seen in larger tumors [5]. A diameter

larger than 10 cm associating a high risk profile of malignancy requires the open procedure, which is encouraged from the beginning [3-5]. The adrenal-preserving surgery is a controversial issue. The most useful situation is bilateral pheochromocytoma with a genetic background, as multiple endocrine neoplasia type 2A and non-expected malignancy [6,7]. The data regarding bilateral tumours in patient with adrenal Cushing's syndrome are still a matter of debate [8]. We are presenting a case with unilateral laparoscopic adrenalectomy that was performed in a woman with bilateral adrenal tumours.

Case presentation

A 48-year-old non-smoker woman has a history of bradimenorrhea at the age of 44. The gynecological evaluation did not find any specific pathology except for an adrenal tumor at abdominal ultrasound. She was referred for an endocrine assessment. On admission, the medical history reveals mild arterial hypertension while the family medical background is negative.

The computed tomography (CT) revealed bilateral adrenal tumors of 1.9 by 1.5 by 2 cm (right) and 2.4 by 1.6 by 2.4 cm (left). The endocrine proved a low-normal ACTH (Adrenocorticotropic Hormone) level but a suppressed morning plasma cortisol, after low dose of dexametasone suppression test, so no clear Cushing's syndrome criteria were found(Table 1). High chromogranin A was found. She was followed-up for one year and the evaluation revealed a higher dimension of the left adrenal mass (of 4 cm maximum diameter) (Figure 1).

Parameter	Level	Normal ranges	Units
Neuron specific enolase	4.79	0-12	ng/mL
Morning plasma cortisol (baseline)	15.75	6.7-22.6	μg/dL
Morning plasma cortisol (after overnight 1 mg of dexametasone)	1.21	Less than 1.8	μg/dL
Calcitonin	1	1-11.8	ng/mL
ACTH (AdrenoCorticotrop Hormone)	**9.42**	3-66	pg/mL
Chromogranin A	**274**	20-125	ng/mL
Plasma metanephrines	26	10-90	pg/mL
Plasma normetanephrines	124	15-180	pg/mL
Serum calcitonin	2.53	1-4.8	pg/mL

Tabel 1 - Endocrine parameters in a female patient with bilateral adrenal tumours

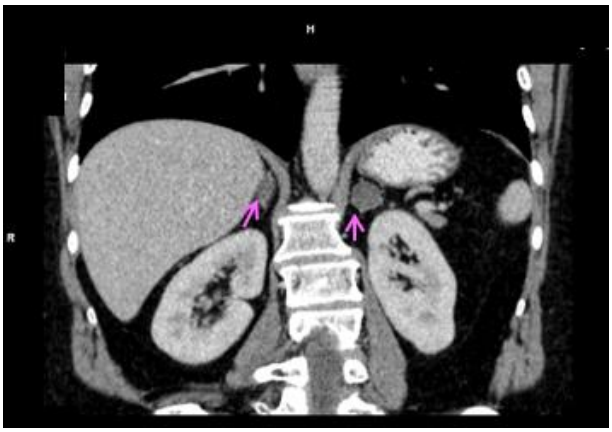

Figure 1 - Abdominal computed tomography (with contrast) aspect (coronal plane): bilateral adrenal tumours (arrows)

The endocrine profile showed low normal value of ACTH but the classical diagnosis of adrenal Cushing's syndrome was still inconsistent. The chromogranin A continued to be high. Based on the increased dimensions of the left adrenal tumour, surgery was recommended.

Laparoscopic transabdominal left adrenalectomy (with 3 trocars) was successfully performed without any incident. No conversion was required. Neither intraoperative nor postoperative complications occurred. The patient was hospitalised 5 days and the wounds healed normally. The pathological report revealed a benign corticoadenoma. The

endocrine evaluation after one week showed that ACTH de-suppressed after surgery up to the normal level of 20pg/mL. The right adrenal tumour was followed-up for another year and the diameters were stationary. The patient was still hypertensive and she was offered adequate therapy (which we considered essential for high blood pressure). Life-long periodical check-up is necessary.

Discussion

We introduce the case of woman within her fifth decade of life diagnosed with synchronous adrenal tumours. The endocrine features showed some activity by augmented chromogranin A and low-normal ACTH despite the fact that no clear diagnosis of Cushing's syndrome could be established. The surgery was an adequate option for a growing tumour and for providing the pathological report and yet preserving the contralateral smaller tumour (probably an adrenal incidentaloma). The normalisation of the endocrine parameters after providing unilateral surgery represents the best argument for choosing the right side to operate (based on initial size and changing diameter during follow-up).

The alternative of adequate identification of the adrenal tumour in case of Cushing's syndrome with bilateral adrenal tumours, is adrenal vein sampling in order to localize a functioning side [9]. This surgical based procedure is difficult to establish and not functional in many centres [10]. We choose not to use it because of high size difference between the two tumours and because clear cortisol hyper-secretion was not identified.

Bilateral adrenal tumours are extremely rare and usually they are bilateral pheoromocytomas, an adrenal secreting tumour with a contra lateral incidentaloma, a bilateral adrenal hyperplasia (with or without adrenal or pituitary Cushing's syndrome), a bilateral metastasis or adrenal cancer with contra lateral metastasis [11]. The genetic background is related to Carney's complex, MEN2A syndrome or new mutations as somatic PRKACA (L206R) mutation as a cause of cortisol-producing adenomas, and somatic mutations in genes coding for ion channels (KCNJ5 and CACNA1D) and ATPases (ATP1A1 and ATP2B3) for primary hyperaldosteronism [12,13]. In this particular case, some hormonal activity was revealed by the higher tumour and the other adrenal had an incidentaloma.

Laparoscopic left adrenalectomy is the standard procedure for approaching adrenal tumours especially if the highest diameter is less than 6 cm [14]. The open technique is limited by a higher risk of complications but offers a better anatomical view which is an important aspect in potential malignancy and it is the indicated procedure in case of local invasion or vascular infiltration [14]. Left adrenal approach is feasible, transabdominally or transperitoneally.

A study comparing the conventional laparoscopic adrenalectomy with single-incision transperitoneal laparoscopic surgery showed that they are both safe if the patients were carefully selected and similar results are displayed [15]. Advanced sealing devices during laparoscopy reduce the surgery time, especially in left adrenalectomy [16]. Regardless of the endocrine profile, laparoscopic adrenalectomy is the gold standard in tumour disease of the adrenals [17]. The clinical evolution is improved after the tumour removal, due to the hormonal improvement. The good outcome is also related to anesthesiological intervention and the surgical team's experience [18,19].

Conclusion

By this woman's case we underline that the unilateral laparoscopic approach is a safe and adequate decision in bilateral adrenal tumours in order to provide the adequate pathological exam and to improve the clinical outcome.

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