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Laparoscopic Bilateral Partial Adrenalectomy for Adrenocortical Adenomas Causing Cushing's Syndrome: Report of a Case

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Abstract
Laparoscopic total adrenalectomy has become a standard technique for small adrenal tumors; however, bilateral adrenalectomy results in postoperative adrenal insufficiency, necessitating lifelong steroid replacement. To preserve adrenocortical function in a 41-year-old woman with bilateral adrenocortical adenoma (BAA) causing Cushing's syndrome, we performed laparoscopic bilateral partial adrenalectomy. We based our preoperative diagnosis of bilateral adrenocortical tumors causing Cushing's syndrome on the results of endocrinological investigations and imaging findings. Thus, we performed lateral transperitoneal laparoscopic bilateral partial adrenalectomy, preserving the adrenal glands, which were normal. Pathological examination of both tumors confirmed the diagnosis of adrenocortical adenoma. The patient had no postoperative complications, and her adrenocortical function was normal without steroid replacement at her 10-month follow-up. This report shows that Cushing's syndrome resulting from bilateral adenomas can be effectively treated by laparoscopic bilateral partial adrenalectomy as a minimally invasive, adrenocortical-preserving operation.

Key words Bilateral adrenocortical adenoma · Cushing's syndrome · Laparoscopic partial adrenalectomy

Introduction

Laparoscopic partial and unilateral total adrenalectomy is commonly performed for small adrenal tumors; however, bilateral total adrenalectomy causes acute adrenal insufficiency (Addisonian crisis), necessitating lifelong steroid replacement. To our knowledge, there is only one report of bilateral adrenocortical adenoma (BAA) causing Cushing's syndrome.[1] We performed laparoscopic bilateral partial adrenalectomy, a novel operation, designed originally for pheochromocytoma,[1–5] to treat BAA causing Cushing's syndrome. We describe this novel operation, which we performed using a lateral transperitoneal approach.

Case Report

A 41-year-old woman was admitted to our hospital with nausea, diarrhea, and abdominal pain. She gave a history of having gained 10 kg over the preceding 2 years. The following clinical measurements were recorded on admission: height, 155 cm; weight, 63 kg, blood pressure, 170/110 mmHg; pulse rate, 70 beats/min and regular. Central obesity, abdominal striae cutis, and a buffalo hump were apparent. Complete blood count and serum biochemistry values were within normal limits, but endocrine studies revealed high urinary 17-hydroxycorticosteroid, low plasma adrenocorticotropic hormone (ACTH), and high plasma cortisol with no circadian variation. On ACTH stimulation testing, the cortisol levels rose with time (Table 1). Abdominal computed tomography (CT) showed bilateral adrenal masses, with a maximam diameter of 2.0 cm on the right and 2.4 cm on the left. Both masses appeared smooth and well circumscribed, and the mass on the left contained an area of calcification (Fig. 1). Adrenal scintigraphy revealed tracer accumulation in both adrenal glands (Fig. 2). Based on these findings, we made a preoperative diagnosis of bilateral adrenal tumors causing Cushing's syndrome.

To preserve adrenal function, we performed laparoscopic bilateral partial adrenalectomy via a lat-

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Table 1. Clinical and endocrinological findings on admission

Plasma level		Urinary level	
Renin activity	20.0 ng/ml/h	17-OHCS	16.5 mg/day
Aldosterone	34.7 pg/ml	17-KS	13.0 mg/day
E2	28.10 pg/ml	11-OHCS	29.2 μg/day
LH	2.28 mIU/ml	A	8.974 μg/day
FSH	12.92 mIU/ml	NA	100.796 μg/day
A	0.031 ng/ml	DA	0.803 μg/day
NA	0.537 ng/ml	5-HIAA	3.0 mg/day
DA	0.337 ng/ml	VMA	5.4 mg/day
TSH	0.138 μU/ml	HVA	3.3 mg/day
FT3	2.94 pg/ml		
FT4	1.64 ng/dl		

Daily profile of cortisol and ACTH				
Clock	08:00	12:00	16:00	23:00
Cortisol (μg/dl)	27.85	32.52	38.69	27.74
ACTH (pg/ml)	<4	<4	<4	<4
ACTH tolerance test				
Time (min)	0	30	60	
Cortisol (μg/dl)	25.74	42.04	54.05	

ACTH, adrenocorticotropic hormone; OHCS, hydroxycorticosteroid

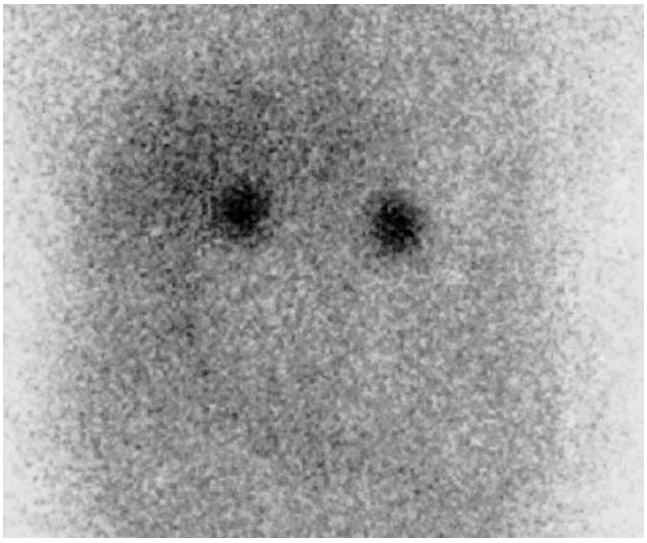

Fig. 2. Adrenal scintigraphy revealed tracer accumulation in both adrenal glands

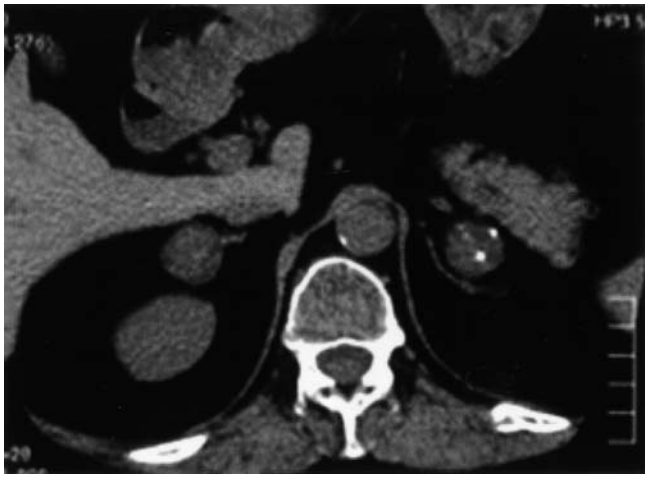

Fig. 1. Abdominal computed tomography showed bilateral adrenal masses, with a maximum diameter of 2.0 cm on the right and 2.4 cm on the left. Both tumors appeared well circumscribed and smooth. The left tumor contained a calcified area

eral transperitoneal approach. The patient was positioned in the right lateral decubitus position and the operating table was angled at the center. We inserted the first 12-mm port in the left midclavicular line and the peritoneal space was insufflated to 8 mmHg using CO_2. Another 12-mm port and a 5-mm port were then inserted in the anterior axillary line and posterior axillary line, respectively. After identifying the spleen, we dissected the underlying retroperitoneum to expose the left kidney. The adipose tissue between the spleen and the kidney was then retracted to expose the tumor, and the tissue surrounding the tumor was trimmed. To control slight hemorrhage from the adipose tissue and to irrigate the area, an additional 5-mm port was placed between the anterior axillary line and the posterior axillary line. We ligated the central vein with three clips and separated the left tumor from the code-like left adrenal gland by using the laparosonic coagulating shears (LCS) attachment of a harmonic scalpel. Finally, we resected the tumor and part of the normal adrenal gland on the left side. The patient was then repositioned in the left lateral decubitus position and the operating table was angled in the opposite direction. We inserted the 12-mm ports in the right midclavicular line and anterior axillary line, and the 5-mm ports in the middle and posterior axillary lines. A cherry dissector was used to transect adhesions between the edge of the liver and the abdominal wall. We exposed the right kidney and identified the tumor within the suprarenal adipose tissue. The code-like adrenal gland was retracted and the surrounding adipose tissue was trimmed. Finally, we resected the right tumor and the tenuous blood vessel from the code-like adrenal gland, using the LCS (Fig. 3), while preserving the normal adrenal gland. The total operating time was 340 min, being 200 min for the left adrenal gland and 110 min for the right, with 30 min for repositioning. Blood loss was minimal. The resected right tumor measured 22 × 20 × 18 mm and weighed 5.4 g, and the left tumor measured 24 × 22 × 16 mm and weighed 9.1 g. Both tumors were well encapsulated with thin fibrous tissue, and exhibited no nodularity. The cut surface was yellowish-brown (Fig. 4). The partially resected area of the left adrenal gland was atrophic. Hematoxylin–eosin staining revealed that both tumors

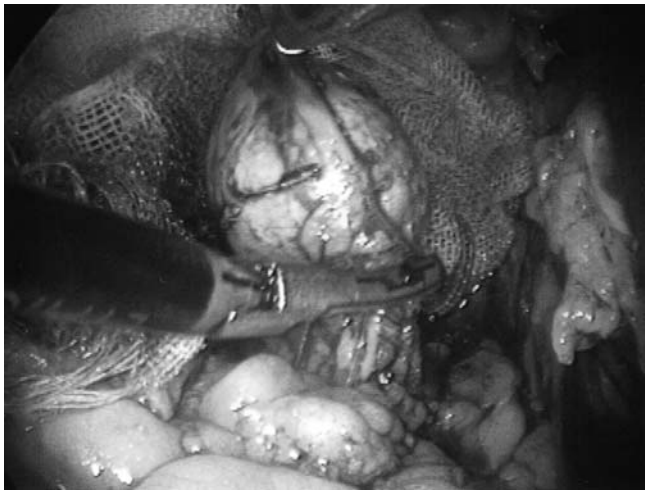

Fig. 3. Intraoperative laparoscopic view showing resection of the right adrenal tumor from the remnant adrenal tissue using the laparoscopic coagulation shears

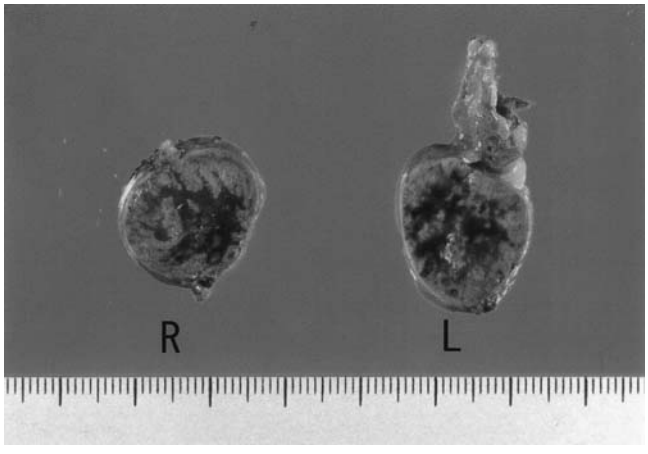

Fig. 4. Resected specimens (right, 22 × 20 × 18 mm, 5.4 g; left, 24 × 22 × 16 mm, 9.1 g). Both tumors were well encapsulated with thin fibrous tissue. The cut surface of both tumors was yellowish-brown

consisted of clear, compact cells. There was no cellular invasion to the capsule, blood vessels, or adjacent tissue. The final pathological diagnosis of both adrenal tumors was adrenocortical adenoma.

The patient's blood pressure normalized after the operation and there were no postoperative complications. Hydrocortisone supplementation was initiated at a dose of 30 mg/day and then tapered off. At her 10-month follow-up, her blood pressure was 122/60 mmHg and stable, and her plasma cortisol and ACTH levels were 0.75 μg/dl and 50.1 pg/ml, respectively. She has not required steroid replacement.

Discussion

To the best of our knowledge, this is only the second report of laparoscopic bilateral partial adrenalectomy being performed for BAA causing Cushing's syndrome.[1] This operation resulted in postoperative preservation of adrenocortical function in our patient. Thus, the advantage of bilateral partial adrenalectomy is that the adrenal insufficiency ensuing from total adrenalectomy and the need for lifelong steroid replacement can be avoided. About 20% of patients who undergo bilateral total adrenalectomy suffer Addisonian crisis for up to 30 years, and Nelson's syndrome develops during the first 7–24 years in 23%.[6]

To select the most appropriate operative procedure, it is important to differentiate Cushing's syndrome caused by BAA from that caused by ACTH-independent macronodular hyperplasia (AIMAH) or primary pigmented nodular adrenocortical disease (PPNAD).[7] Bilateral total adrenalectomy is required for both AIMAH and PPNAD.

Cortisol-producing adenomas (CPA) are generally primary and solitary. On the other hand, BAAs are very rare, with only 25 cases published in the literature since 1977.[1,8] Because primary adrenocortical nodular dysplasia is defined as autonomous adrenocortical hyperplasia, in which plasma corticotropins cannot be detected, it is necessary to accurately distinguish between nodular hyperplasia and multiple adenomas.[9] Endocrinologically, BAA and AIMAH both exhibit adrenal autonomy. Moreover, adrenocortical scintigraphy reveals bilateral tracer uptake and CT shows bilateral adrenal enlargement in both diseases. However, AIMAH is differentiated by multiple nodules in both glands whereas BAA is usually seen as a single nodule in each gland. Morphologic examination is a powerful tool for distinguishing the two disorders. In BAA, the nodules are usually encapsulated and the adjacent non-nodular area is atrophic, whereas in AIMAH the nodules are not encapsulated, and the adjacent area is hypertrophic and sometimes contains small micronodules.[10]

There are four approaches to laparoscopic adrenalectomy, namely, anterior transperitoneal, lateral transperitoneal, lateral retroperitoneal, and posterior retroperitoneal. We used the lateral transperitoneal approach in our patient because it offered good visibility of familiar anatomic landmarks, easy access to other organ systems, the use of gravity to retract the spleen and liver, and a wide exposure to allow easy removal of large adrenal lesions.[11] A good view can also be obtained with low insufflation pressure, assisted by the rib arch, although this method is not suitable for patients who have undergone previous epigastric surgery.

In conclusion, we successfully performed laparoscopic bilateral partial adrenalectomy for BAA causing

Cushing's syndrome. The patient recovered without any postoperative complications and her adrenal function normalized. She still does not require steroid replacement. Thus, laparoscopic bilateral partial adrenalectomy is a minimally invasive and effective adrenocortical-preserving operation for BAA causing Cushing's syndrome.

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