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ASYMPTOMATIC ADRENAL MEDULLARY HYPERPLASIA DETECTED WITH INTRAOPERATIVE HYPERTENSION : A CASE REPORT

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We report a rare case of asymptomatic adrenal medullary hyperplasia detected by chance with intraoperative hypertension during surgery for ipsilateral renal cell carcinoma. A 41-year-old male visited our hospital with a complaint of left flank pain. He had normal blood pressure and plasma catecholamine level was within normal limits. Ultrasonogram and CT scan revealed a left renal tumor but did not showed any abnormal masses in the left adrenal gland. The clinical diagnosis was renal cell carcinoma and we performed left total nephrectomy. In the process of operative manipulation, however, the blood pressure and pulse rate of this patient showed a marked increase. Pathological examination of the extirpated kidney revealed renal cell carcinoma, while the resected adrenal gland was diagnosed as adrenal medullary hyperplasia.

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Key words : Adrenal medullary hyperplasia, Renal cell carcinoma

INTRODUCTION

Adrenal medullary hyperplasia (AMH) is relatively rare and accurate diagnosis before surgery is often difficult. We present herein a case of AMH detected by chance with intraoperative significant hypertension and tachycardia during tatal nephrectomy.

CASE REPORT

A 41-year-old male visited our hospital with a complaint of left flank pain. He had normal blood pressure and routine hematological study and plasma catecholamine level were all within normal limits. Ultrasonogram and CT scan revealed a left renal mass measuring 7.7×7.5 cm and a slight swelling of the left adrenal gland but its shape was within normal limits (Fig.1). The angiogram showed no abnormal masses in the left adrenal gland except a hypervascular mass in the upper pole of the left kidney.

The clinical diagnosis was renal cell carcinoma and we performed left total nephrectomy. However, during the process of removing the kidney with the adrenal gland en bloc, the blood pressure of the patient rose over 220 mmHg systolic and pulse rate also increased over 130 per minute. Therefore, we had continued drip infusion of antihypertensive drugs until the operation was over. After the extirpation of the left kidney and adrenal gland both blood pressure and pulse rate of the patient normalized immediately.

Pathological examination of the extirpated kidney showed a renal cell carcinoma (clear cell carcinoma, G1, INFα, pT2), while the resected adrenal gland (5.5×3.5 cm in size) had no mass lesion macroscopically. The cut surface of the adrenal gland was milky-white to yellow with admixed changes and cystic degeneration was partially revealed. Microscopically adrenal medulla was clearly separated from adrenal cortex and expanded diffusely but did not have either nodular formation or encapsulation. The ratio of medullary to cortical area had increased to be about 1 : 3 (normal ratio is 1 : 8 to 1 : 14.3[1,2]) (Fig. 2A). Individual cells of medullary tissure were enlarged and consisted of spheroidal nucleus and granular cytoplasm (Fig. 2B). The catecholamine values of the content of cystic degeneration were extremely high(epinephrine 66,044 ng/ml, norepinephrine 9,134 ng/ml, dopamine 123 ng/ml). Based on these findings this case was diagnosed as AMH.

DISCUSSION

AMH is a disease that resembles pheochromocytoma in the clinical feature, but in most cases, the formation of any masses in the adrenal gland is absent on radiographic or pathological examinations. In most cases AMH has been reported that the patient admitted a hospital with complaints of poorly-controlled blood pressure or hypertensive encephalopathy and were operated with suspicion of small pheochromocytoma due to a high blood catecholamine level but were consequently diagnosed as AMH in the pathological examination[1)]

Only about twenty cases of AMH have been reported in Japan and, to our knowledge, this is the first case of AMH which was not suspected until the operative procedure and detected by chance with intraoperative hypertension. Medullary cells of this disease are similar to those of pheochromocytoma microscopically and the diffuse or rarely nodular[3)] expansion of adrenal medulla and increase of medulla/cortex ratio are the diagnostic criteria[4)]

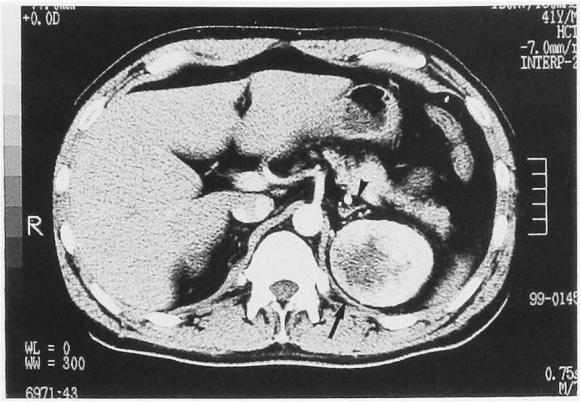

Fig. 1. CT scan showed a left renal tumor (arrow) and a slight swelling of the left adrenal gland but its shape was normal (arrow head).

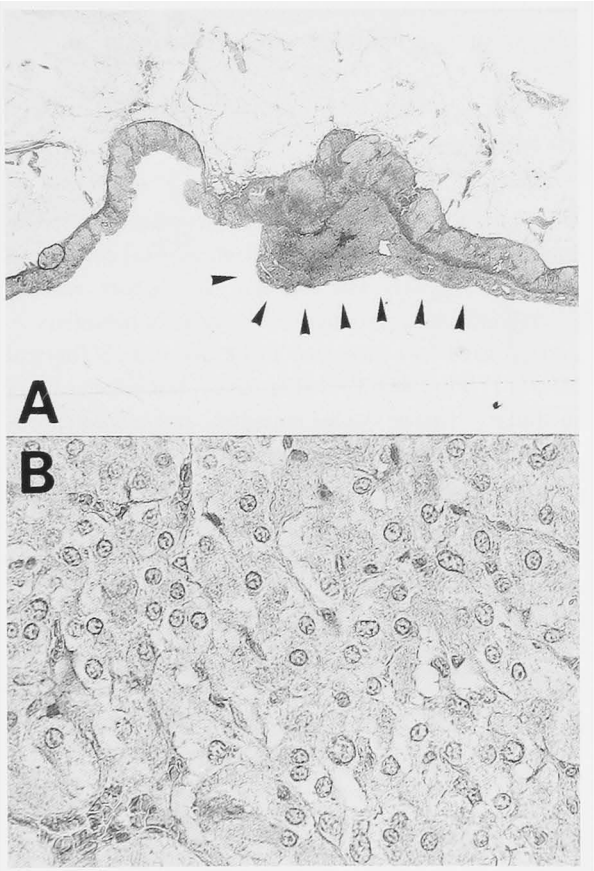

Fig. 2. A, microscopically adrenal medulla expanded diffusely(arrow heads) and did not have either nodular formation or encapsulation. The ratio of medullary to cortical area was about 1 : 3. HE. Reduced from ×4. B, individual cells of medullary tissue were enlarged and consisted of spheroidal nucleus and granular cytoplasm. HE Reduced from ×400.

Since some cases associated with von Recklinghausen disease or multiple endocrine neoplasia, type II (MEN II) have been reported[5], some authors argue that adrenal medullary hyperplasia is a precursor of pheochromocytoma, but the development and the status of this disease have been still controversial. In the case presented here, the patient was asymptomatic until the operative procedure and plasma catecholamine level was normal, and both radiographic and pathological examination revealed no distinct mass lesions in the adrenal glands. These findings were not compatible with those of 'silent' pheochromocytoma, on the other hand, the extirpated adrenal gland microscopically revealed diffuse expansion of adrenal medulla and increase of medulla/cortex ratio which were characteristic of AMH.

In cases, in which blood pressure showed an abnormal increase duging the nephrectomy, we recommend an ipsilateral adrenalectomy because of the possibility of AMH.

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和文抄録

術中高血圧を契機に発見された副腎髄質過形成の1例

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術中高血圧を契機に発見された稀な副腎髄質過形成の1例について報告する．症例は41歳，男性で，左側腹部痛を主訴に来院した．血圧や血中カテコラミン値は正常範囲であった．腹部エコー，CTスキャン，血管造影で左腎腫瘍を認めたが，副腎の形態には異常を認めなかった．左腎細胞癌の診断で根治的腎摘術を施行した．腎摘中に著明な高血圧と頻脈を認めた．病理診断は腎細胞癌であったが，同時に切除された同側副腎に副腎髄質過形成を認めた．

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