Hypertensive Crisis During Excision of Retroperitoneal Mass in Patients with Abdominal Aortic Aneurysm
- A Case Report -

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INTRODUCTION

Paragangliomas are rare catecholamine-secreting tumors that arise from the chromaffin cells of the sympathetic ganglia and are known as extra-adrenal pheochromocytomas. (1-3) The excessive production of catecholamine can cause life-threatening cardiovascular complications such as severe hypertension, cardiac arrhythmia, cardiomyopathy, myocardial infarction, and pulmonary edema. (2) However, as most extra-adrenal paragangliomas are hard to diagnose preoperatively, (3) some of them can only be noticed after surgical manipulations that stimulate the release of catecholamines from the tumor. These situations can lead to an endocrine emergency with an unpredictable course and hemodynamic instability during surgery, particularly in patients who have not been appropriately prepared for the surgery. We report a case of preoperatively undiagnosed paraganglioma that caused a hypertensive crisis during the surgical removal of a retroperitoneal mass and the repair of an infra-renal abdominal aortic aneurysm (AAA) and left common iliac artery aneurysm.

CASE REPORT

A 68-year-old male patient was scheduled for the surgical removal of a retroperitoneal mass and the repair of an infra-renal abdominal aortic aneurysm (AAA) and left common iliac artery aneurysm. The abdominal computed tomography (CT) scan showed a slightly heterogeneous and calcified round retroperitoneal mass of 4.0×3.6×3.5 cm in the left para-aortic space (Fig. 1) and infra-renal aneurysmal dilatation (Fig. 2). The aneurysmal dilatation measured 3×3 cm, with mural calcification. The scan also revealed an aneurysmal change of the left common iliac artery of 5.1 cm of diameter. The patient did not report any symptoms, including sweating, headache, palpitation, or postural hypertension. He had been on medication for ten years for hypertension, coronary artery occlusive disease (3 vessels), a cerebrovascular accident, and
Fig. 1. Axial computed tomography for solid nodular lesion. (Axial computed tomography scan shows solid nodular lesion with small calcification of 4.0 × 3.6 × 3.5 cm in left para-aortic space.)

Fig. 2. A: Axial computed tomography for infra-renal abdominal aortic change (Axial computed tomography scan shows infra-renal abdominal aortic change. The aneurysmal dilatation measured 3 × 3 cm, with mural calcification.), B: Axial computed tomography for left common iliac arterial aneurismal change (Axial computed tomography scan shows left common iliac arterial aneurismal change with mural calcification. The size of aneurismal change increases to 5.1 cm.)

diabetes mellitus, for which his blood pressure and glucose level were well-controlled. As the patient wanted to have the retroperitoneal mass and the increment of the left common iliac artery removed as the aneurysm was progressing, a decision was made to operate on him in order to repair the AAA and perform a concomitant resection of the retroperitoneal mass instead of a stent-graft insertion.

When the patient arrived in the operating room, his initial blood pressure (BP) was 110/80 mmHg and his heart rate (HR) 92/min. For continuous BP monitoring, an arterial catheter was inserted in the left radial artery with the local anesthesia. The anesthesia was induced with propofol 160 mg and remifentanil 0.2 μg/kg/min, and the tracheal intubation was facilitated with rocuronium 50 mg. The anesthesia was maintained with a continuous infusion of remifentanil (0.05-0.2 μg/kg/min) and sevoflurane (0.8-1.2 MAC) in order to keep the bispectral index score between 40 and 60. After induction of the anesthesia, a central venous catheter was inserted in the internal jugular vein for monitoring of the central venous pressure. No significant hemodynamic disturbances occurred during the induction, intubation, or surgical incision.

During the surgery, the surgeon found the retroperitoneal mass, which lay in the para-aortic space and adhered to the AAA. When the retroperitoneal mass was manipulated, the patient’s BP suddenly increased to 220/120 mmHg, with a pulse rate of 110 (beats/min). The depth of the anesthesia was deepened by increasing the concentration of sevoflurane from 1.5vol% to 2.5vol% and that of remifentanil from 0.05-1 μg/kg/min to 0.3 μg/kg/min. However, the blood
pressure continued to rise. The systolic blood pressure reached 180 mmHg and the systolic blood pressure 100 mmHg. The hypertension and tachycardia were treated with intermittent boluses of nicardipine (2.5 mg of nicardipine in five doses of 500 μg each) and labetalol (10 mg of labetalol in two doses of 5 mg each), with an additional continuous infusion of nitroglycerin (20 μg /min). Although the BP decreased to 143/90 mmHg, there was still an increase in the systolic BP (above 180 mmHg) and the HR (100-120 /min) whenever the surgeon manipulated the mass directly. Despite the increase in the anesthetic depth and the anti-hypertensive therapy, the BP continued to fluctuate widely between 170-100 mmHg/60-80 mmHg and was difficult to control due to the frequent swings between hypertension and hypotension. Therefore, there were high suspicions of an unrecognized paraganglioma.

Following the tumor removal, the BP decreased to as low as 60/40 mmHg, and the HR to 75/min. In order to improve the patient’s hemodynamic parameters, we administrated 500 ml of volume expander and 750 ml of crystalloid, and initiated a continuous norepinephrine infusion (0.15-0.2 μg/kg/min) with intermittent ephedrine (16 mg of ephedrine in two doses of 8 mg each) and phenylephrine (100 μg of phenylephrine in two doses of 50 μg each) injections. As the infra-renal abdominal aortic aneurysmal change was more minimal than expected, the surgeon decided to insert an endovascular stent-graft in the left common iliac artery aneurysm instead of performing surgical repair.

The total operating time was about 165 min. The estimated blood loss was around 300 ml, and the total fluid administration 1,800 ml. At the end of the surgery, the BP was 110/65 mmHg and the HR 75-80/min, although norepinephrine was still infused at 0.02 μg/kg/min. The patient was extubated in the operating room and was transferred to the intensive care unit (ICU) for close monitoring. The norepinephrine injection was discontinued 1h after the surgery and the patient was transferred to the general ward without any hemodynamic disturbances on postoperative day 2. The retroperitoneal mass was histopathologically confirmed as an extra-adrenal pheochromocytoma (paraganglioma) a week later.

**DISCUSSION**

In the present case, the CT findings suggested a simple non-functioning benign tumor or proliferative lymph node in the left para-aortic space. Although the patient had other concomitant underlying diseases, including hypertension, coronary artery occlusive disease, a cerebrovascular accident, and diabetes mellitus, his BP and plasma blood glucose level were well-controlled, at 120/80-140/90 mmHg and 120-200 mg/dl respectively. The patient did not present cardiomyopathy and did not show any symptoms associated with catecholamine-secreting tumors, such as palpitation, headache, or sweating. The size of the tumor had not changed for two years and there was no distant metastasis. Therefore, we overlooked the possibility of a paraganglioma and did not conduct hormonal studies, such as analyses of the 24-hour urine-fractionated metanephrines and catecholamines or of the plasma-fractionated metanephrines.

In fact, patients with a paraganglioma do not always show catecholamine-related symptoms. There is a 26% incidence of headache, 21% of palpitations, 25% of perspiration, 12% of pallor, and 64% of hypertension. Some cases can even be asymptomatic or ambiguous, especially when the patient has a psychiatric disorder, anxiety, facial pallor, weight loss, polyuria, hyperglycemia, secondary erythrocytosis, stroke history, or cardiomyopathy, making a proper diagnosis difficult. For proper diagnosis, if a catecholamine-secreting tumor is suspected, it is useful to collect the 24-hour
urine-fractionated metanephrines and catecholamines. Imaging studies such as CT or MRI are used to localize extra-adrenal paragangliomas, and ¹²³I-metiodobenzylguanidine (MIBG) shows a higher specificity for the confirmation of catecholamine-secreting adrenergic tissue.

Although AAAs with concomitant undiagnosed extra-adrenal pheochromocytomas as in our case are rare, clinicians such as Ehata et al.(6) and Spanos et al. (7) have reported similar cases. The coexistence of paragangliomas or pheochromocytomas with AAAs presents an increased risk of rupture of the aneurysm from the excess of catecholamines and hypertension. These studies reported that for safe operation, the patient must ideally be diagnosed preoperatively and treated with preoperative care for blood pressure control.

Preoperative medication with an α-blocker must be administrated for at least 2 weeks before the operation. β-blockers can be given after administration of the α-blocker, as the use of a β-blocker before administration of a α-blockade may trigger a hypertensive crisis due to the unopposed α-adrenergic receptor stimulation.(8) Fluid replacement is also necessary to prevent volume depletion from chronic vasoconstriction. Sodium nitroprusside, nitroglycerin, and nicardipine hydrochloride can be used to control a sudden hypertensive crisis intraoperatively. Sodium nitroprusside is the most commonly-used, as it is effective for arteriolar dilatation and to suppress the hypertensive response. Nicardipine hydrochloride is also effective for suppression of the secretion of norepinephrine in paragangliomas.

In our case, we did not administer a α-blocker in the preoperative period as we did not suspect the presence of a paraganglioma. Therefore, we did not give β-blockers after the possibility of a paraganglioma was considered. During the operation, we used nicardipine and labetalol. Although the patient’s vital signs became stable after the injection of nicardipine and labetalol, sodium nitroprusside would have been a more adequate management drug than labetalol due to its longer-lasting action. Patients may also require intensive postoperative care. The main postoperative concerns include hemodynamic instability and hypoglycemia. Transient postoperative hypotension is common, as the rebound increase in insulin secretion can cause downregulation of the α-receptors and hypoglycemia.

In conclusion, extra-adrenal paragangliomas are difficult to diagnose in the preoperative patient evaluation as they may show no symptoms. Therefore, the possible presence of paragangliomas should be considered, especially when the CT findings reveal a heterogeneous, calcified, and ill-defined retroperitoneal mass. To prevent cardiac accidents, it is necessary to suspect a potential risk of hypertensive crisis and to respond immediately to manage the risk.

**ABSTRACT**

Paragangliomas are rare tumors that arise from extra-adrenal chromaffine tissues and that secrete catecholamine. The abnormal secretion of catecholamine can cause cardiovascular complications such as severe hypertension, cardiomyopathy, cardiac arrhythmia, and aortic aneurysm rupture. In particular, the risk of mortality and morbidity increases if the patient has not been diagnosed or if the preparation for a possible hypertensive crisis has been insufficient. Moreover, it can lead to high risks of cardiovascular complications if the patient has a concomitant disorder such as aortic aneurysm.

We report a case of preoperatively undiagnosed paraganglioma that caused a hypertensive crisis during the surgical removal of a retroperitoneal mass and the repair of an infra-renal abdominal aortic aneurysm.
(AAA) and left common iliac artery aneurysm.

Key Words: Hypertensive crisis, Paraganglioma, Abdominal aortic aneurysm

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