Case Report

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An unusual case of hematemesis and epistaxis caused by a pheochromocytoma

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Abstract

Pheochromocytoma is a rare catecholamine-secreting neoplasm that is the cause of hypertension in <0.2% of patients with hypertension. We encountered an unusual case of pheochromocytoma involving hematemesis and epistaxis episodes with accompanying hypertensive attacks. Venous ectasia was detected in the esophagus. Abdominal magnetic resonance imaging revealed an adenoma in the left adrenal region. The present case illustrates that pheochromocytoma can mimic different clinical conditions.

Keywords

Pheochromocytoma, hematemesis, epistaxis episodes, hypertension, venous ectasia, magnetic resonance imaging

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Introduction

Pheochromocytoma is a rare catecholamine-secreting neoplasm with an estimated annual incidence of 0.8 per 100.000 person-years. It is the cause of hypertension in <0.2% of patients.[1,2] Early and correct diagnosis is essential for effective treatment, which usually involves surgical resection and is curative in most cases. The manifestations of pheochromocytoma are various, and the tumor can mimic a variety of conditions, often resulting in misdiagnosis.[1] We herein report an unusual case of pheochromocytoma involving hematemesis and epistaxis episodes with accompanying hypertensive attacks. This case is being reported to alert clinicians to the fact that a thorough investigation should be performed in patients suspected to have a pheochromocytoma to avoid an incorrect diagnosis and delayed treatment.

Case report

The patient described in this report provided written and verbal consent to report the details of his case. Our ethics review committee waived the requirement for study approval based on the study design (single case report).

A 39-year-old man with hematemesis was hospitalized for diagnostic purposes. He had a 3-year history of epistaxis and hematemesis but no other symptoms such as palpitations, sweating, headache, or esophageal varices. The patient had experienced both epistaxis and hematemesis episodes once a month for 2 years, then once a week for the last year. He was not taking any medication during that time. During every bleeding attack, the amount of bleeding was approximately 30 mL; the blood tended to gush and was normal in color. His blood pressure was always high (200/100 mmHg) at the time of the bleeding episodes during hospitalization; he also developed paroxysms of hypertension but had no portal hypertension. His systolic and diastolic blood pressure during hospitalization varied from 120 to 200 mmHg and 70 to 100 mmHg, respectively. The patient reported mild fatigue during the attacks.

Physical examination revealed no signs of excessive production of adrenocortical steroids, as in Cushing's syndrome. The result of a 1-mg low-dose dexamethasone suppression test was 1.6 μg/dL (reference range, <1.8 μg/dL). No postural drop in blood pressure occurred. His pulse was regular and the rate was normal, ranging from 60 to 100 beats/min except during the episodic hypertension. At those times, his pulse rate was >100 beats/min. His hemoglobin concentration, platelet count, and international normalized ratio were normal at 15.4 g/dL (reference range, 11.1–17.17 g/dL), 318,000/mm^3 (reference range, 140,000–360,000/mm^3), and 1.07 (reference range, 0.8–1.2), respectively. Other laboratory parameters were also normal, including urea (40 mg/dL; reference range, 10–50 mg/dL), creatinine (0.68 mg/dL; reference range, 0.6–1.2 mg/dL), serum renin (2.83 ng/mL/h; reference range while lying down, 0.51–2.64 ng/mL/h; reference range while standing, 0.98–4.18 ng/mL/h), and aldosterone (5.72 ng/dL; reference range, 3.5–30 ng/dL). Esophagogastroduodenoscopy was performed because of recurrence of the hematemesis attacks. A vascular formation (venous ectasia) was detected in the esophagus. No other abnormalities were found on computed tomography angiography of the thorax and aortic arch. Renal color Doppler ultrasonography was performed to exclude renal artery stenosis, and the findings were normal.

In-phase and out-of-phase T1-weighted axial gradient echo magnetic resonance images demonstrated an adenoma of 26 × 22 mm in diameter in the left adrenal region (Figure 1). Additionally, hypointensities were

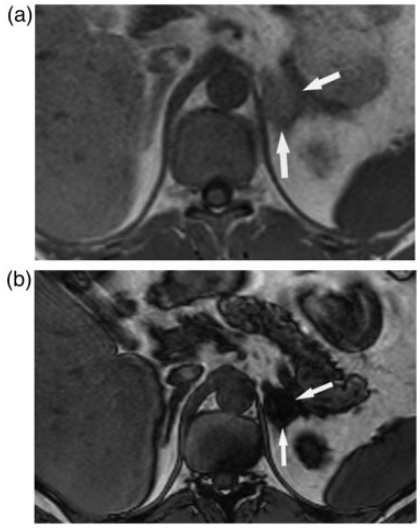

Figure 1. Gradient echo magnetic resonance imaging findings. In-phase (top, a) and out-of-phase (bottom, b) T1-weighted axial gradient echo magnetic resonance images show the lesion (arrows) with significant signal drop-off on the out-of-phase magnetic resonance image due to the high lipid content, which is diagnostic for an adenoma.

present on both T1- and T2-weighted images after gadolinium administration. A radioisotope scanning study was also performed; whole-body indium-111 octreotide single-photon emission computed tomography showed the physiological pattern of a pheochromocytoma (Figure 2). The levels of fractionated metanephrines and catecholamines in a 24-h urine specimen were elevated. These findings are shown in Table 1. The patient began treatment with the α-adrenergic blocker doxazosin (4 mg) followed by the β-blockers metoprolol (25 mg), amlodipine (5 mg), and olmesartan (20 mg) until the surgical operation.

All of the above-described clinical, imaging, and laboratory findings were consistent with the diagnosis of a functional pheochromocytoma of the left adrenal gland.

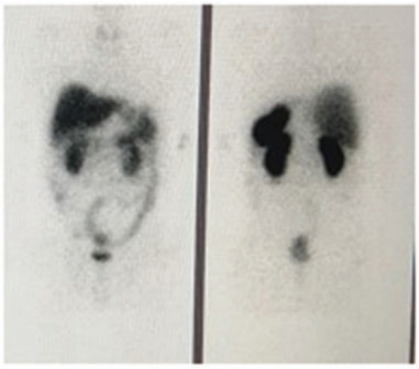

Figure 2. Indium-111 octreotide single-photon emission computed tomography findings.

Table 1. Preoperative and 3-month postoperative laboratory test results

Test	Preoperative	3 months postoperative
24-hour urine collection:		
Metanephrine, μg/24 h	211	105
Epinephrine, μg/24 h	1.5	4
Normetanephrine, μg/24 h	3085	298
Norepinephrine, μg/24 h	129	59
Dopamine, μg/24 h	333	240
Vanillylmandelic acid, mg/24 h	9.9	6.5
Methoxytyramine, μg/24 h	272	124

Reference ranges: Metanephrine, <341 μg/24 h; epinephrine, <22 μg/24 h; normetanephrine, <440 μg/24 h; norepinephrine, <81 μg/24 h; vanillylmandelic acid, <6.7 mg/24 h; methoxytyramine, <247 μg/24 h.

After the patient underwent laparoscopic left adrenalectomy, both the hypertension and ectasia disappeared. The patient declined endoscopic re-evaluation of the esophagus after the adrenalectomy. The hematemesis and epistaxis episodes later ceased, and the patient was discharged

from the hospital on olmesartan medoxomil at 20 mg/day. The patient discontinued the doxazosin and β-blockers. The 24-h urine catecholamine and metabolite levels were normal 3 months after the operation (Table 1).

Discussion

Pheochromocytomas secrete various hormones, especially adrenaline and noradrenaline, that have deleterious effects on the cardiovascular system. High metanephrine and catecholamine levels induce hypertension. Vascular wall changes may occur in patients with pheochromocytomas. Effector tissues such as vascular smooth muscle contain α_1-adrenergic receptors.[3] Stımulation of these receptors causes vasoconstriction and an increase in blood pressure, which may predispose the patient to bleeding from a vascular anomaly as in the present case. Gastrointestinal hemorrhage may rarely occur secondary to multiple varices associated with the tumor mass.[4] According to the previously published literature,[5] pheochromocytoma may be observed together with vascular antral ectasia, as in the present case. After the surgical treatment, both the hypertension and ectasia disappeared. The clinical presentation of pheochromocytoma is not always clear. As mentioned above, a variety of clinical findings may be present. Clinicians should therefore carefully consider all details of a thorough investigation when diagnosing pheochromocytoma.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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