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CASE REPORT

Pheochromocytoma diagnosed after anticoagulation for atrial fibrillation ablation procedure: A giant in disguise

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KEYWORDS
Pheochromocytoma;
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Abstract Pheochromocytoma is a rare catecholamine-producing tumor, discovered incidentally in 50% of cases. We present the case of a 44-year-old male with a history of paroxysmal palpitations. Baseline ECG, transthoracic echocardiogram and ECG stress test showed no relevant alterations. Paroxysmal atrial fibrillation was detected on 24-hour Holter ECG. After antiarrhythmic therapy, the patient remained symptomatic, and was accordingly referred for electrophysiological study and atrial fibrillation ablation. Anticoagulation was initiated before the procedure. After ablation and still anticoagulated, he complained of hematospermia. The abdominal and pelvic imaging study showed a 10-cm left adrenal mass, predominantly cystic, compatible with pheochromocytoma, which was confirmed after biochemical tests (increased urine metanephrines and plasma catecholamines). Metaiodobenzylguanidine scintigraphy scanning confirmed localized disease in the adrenal gland, excluding other uptake foci. Following appropriate preoperative management, surgical resection of the giant mass was performed successfully and without complications.

PALAVRAS-CHAVE
Feocromocitoma;
Fibrilhação auricular;
Hematoespermia

Feocromocitma diagnosticado após hipocoagulação para ablação de fibrilhação auricular – o gigante disfarçado

Resumo O feocromocitoma é um tumor raro produtor de catecolaminas, que se descobre incidentalmente em 50% dos casos. Os autores apresentam o caso clínico de um homem de 44 anos de idade, com história de palpitações paroxísticas. O ECG basal, o ecocardiograma transtorácico e a prova de esforço não demonstraram alterações de relevo. O Holter de 24 h revelou fibrilhação auricular paroxística. Após terapêutica antiarrítmica, o doente manteve-se sintomático, pelo

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que foi orientado para estudo eletrofisiológico e ablação da fibrilhação auricular, tendo iniciado hipocoagulação oral pré-procedimento. No período pós-ablação, ainda hipocoagulado, referiu hematoespermia. O estudo imagiológico abdomino-pélvico revelou uma massa de 10 cm na glândula suprarrenal esquerda, predominantemente cística, compatível com feocromocitoma, diagnóstico que foi confirmado após testes bioquímicos (elevação das metanefrinas urinárias e das catecolaminas plasmáticas). A cintigrafia corporal com metaiodobenzilguanidina confirmou doença localizada na glândula suprarrenal, excluindo outros focos de captação. Após preparação pré-operatória adequada, procedeu-se à resseção cirúrgica da massa gigante com sucesso e sem complicações.

Introduction

Pheochromocytoma is a rare catecholamine-producing tumor, with an incidence of 2–8 cases per million per year,[1] occurring in about 0.5% of patients with hypertension and suggestive symptoms.[2] Typical symptoms include paroxysmal hypertension, pounding headache, palpitations, diaphoresis and pallor.[3] The diagnosis is based on biochemical tests, with demonstration of catecholamine oversecretion by adrenal chromaffin cells, and imaging studies, which localize the tumor and guide surgical resection.[1,3] Half of all pheochromocytomas are discovered incidentally.[4] Cardiovascular complications include myocardial hypertrophy, heart failure, myocardial infarction and various arrhythmias, of which atrial fibrillation is one of the most common.[5–7]

We report a case of a giant pheochromocytoma diagnosed after imaging study for hematospermia subsequent to oral anticoagulation for an atrial fibrillation ablation procedure.

Case report

A 44-year-old man with a history of tobacco use was referred for cardiological assessment in July 2009 due to a three-year history of paroxysmal palpitations lasting about five minutes, which were associated with a throbbing feeling in the head. No other accompanying symptoms were reported. Physical examination, blood pressure (110/70 mmHg), 12-lead electrocardiogram (ECG), transthoracic echocardiogram, ECG stress testing and standard blood analysis including blood glucose (97 mg/dl) were unremarkable. The 24-hour Holter ECG study revealed sinus rhythm with tachycardic activity during 45% of the record, alternating with several periods of ectopic atrial rhythm and one episode of atrial fibrillation lasting 1 hour and 15 minutes. The patient was initially prescribed antiarrhythmic therapy, but the symptoms did not resolve. Electrophysiological study with catheter ablation for atrial fibrillation was discussed and accepted by the patient. Oral anticoagulation was initiated and the pulmonary veins were isolated successfully in November 2010, although the procedure was complicated by a transient period of arterial hypotension and hypoxemia, requiring surveillance in the cardiac intensive care unit for a short period. The patient recovered well and was discharged with indication to maintain oral anticoagulation for three months. During that period, he returned for his periodic cardiological assessment and complained of hematospermia. After discussion with the urology department, antibiotic therapy with levofloxacin was started and abdominal and pelvic ultrasonography was performed, which revealed a 10-cm mass, predominantly cystic, adjacent to the left kidney upper pole. Abdominal magnetic resonance imaging (MRI) enabled better visualization of the mass and showed a nodular formation on the left adrenal gland with a major axis diameter of 9.8 cm, predominantly cystic, with thickened irregular walls and a solid component in the anterior, medial and inferior portions (maximum thickness 4 cm), with heterogeneous late enhancement after contrast administration and diffusion restriction (Figure 1), findings compatible with a giant cystic pheochromocytoma. Urinary metanephrines and plasma catecholamines (specifically adrenaline and noradrenaline) were markedly elevated (Table 1), confirming the diagnosis. A metaiodobenzylguanidine scintigraphy scan revealed localized disease with an intense uptake focus in the left adrenal gland (Figure 2) and no evidence of other uptake foci. Since the patient had no family history of pheochromocytoma, germline mutations that predispose to catecholamine-secreting tumors were not investigated. After appropriate preoperative treatment with phenoxybenzamine, beta-blockers and intravenous fluid expansion, surgical resection of the neoplasm by traditional open surgery was performed in May 2011. Histopathologic and immunohistochemical evaluation were consistent with pheochromocytoma. The immunohistochemical study was positive for chromogranin A, synaptophisin and S100, findings consistent with a diagnosis of cystic pheochromocytoma.[8] There were no intraoperative or postoperative complications. Urinary and plasma metanephrines normalized after surgery (Table 1). During follow-up (more than two years) the patient remained asymptomatic without evidence of recurrence.

Discussion

A ''rule of 10s'' has been attributed to pheochromocytoma, with approximately 10% extra-adrenal location (i.e., secretory paraganglioma), childhood onset, bilateral tumor formation, recurrence after resection, malignant phenotype and familial occurrence.[3] Recently, however, inherited germ line mutations have been described in up to 25% of patients.[4,9]