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Case Report

Partial biochemical response of adrenal artery embolization for pheochromocytoma: A case report and review of the literature ☆

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ABSTRACT

We present the case of a 22-year-old male with a left adrenal pheochromocytoma, initially diagnosed during a workup for thoracic pain. The patient's tumor was refractory to medical management, and surgical resection was ruled out due to high cardiovascular risk, stemming from cyanotic congenital heart disease, aortic aneurysm, and factor VII deficiency. The patient underwent adrenal artery embolization (AAE) as a salvage treatment. Following the procedure, there was an initial reduction in hypertensive crises and biochemical markers, with plasma normetanephrine levels decreasing from 1490 pg/mL to 313 pg/mL. However, over subsequent months, the patient experienced symptom recurrence, and biochemical relapse occurred, with normetanephrine levels rising to 742 pg/mL by 3 months postprocedure. This case highlights the potential for AAE to provide short-term symptom relief and biochemical response; nevertheless, it may not be an effective long-term curative option. More collaborative and prospective studies are needed to assess its success and efficacy.

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Introduction

Pheochromocytoma is a catecholamine-secreting tumor typically arising from the adrenal medulla chromaffin cells. The excessive secretion of adrenaline and noradrenaline by chromaffin cells leads to a distinctive set of symptoms, including hypertension, headache, sweating, and panic attack-like symptoms. Patients may be severely ill at the time of diagnosis, mainly due to chronic hypertension and, if left untreated, can cause severe or life-threatening cardiovascular complications [1]. Following the diagnosis, most patients undergo surgical resection of the tumor after medical preparation. Nevertheless, surgical resection may not be always feasible. In such cases, alternative approaches, such as percutaneous ablation and adrenal artery embolization (AAE) may be considered. AAE is a minimally invasive therapy that has been used successfully in the palliative management of adrenal tumors, suppression of excess adrenal hormone production, in the treatment of ruptured adrenal tumors and traumatic adrenal injuries [2,3]. Limited data exists on its feasibility and outcomes in the treatment of pheochromocytomas. We report herein a case of a left adrenal pheochromocytoma treated using AAE that initially demonstrated a favorable course but subsequently exhibited symptom recurrence and biochemical relapse during follow-up.

Case report

A 22-year-old male presented to our institution with symptoms of a slow-growing left adrenal pheochromocytoma that was refractory to medical management (Fig. 1). The pheochromocytoma was incidentally diagnosed during a computed tomography angiogram (CTA) performed to investigate episodes of thoracic pain. The CTA revealed a 30-mm left adrenal mass, highly suggestive of pheochromocytoma, which was later confirmed through hormonal studies. Initial laboratory results showed elevated plasma metanephrine levels of 46 pg/mL and plasma normetanephrine levels of 740 pg/mL, both of which increased over the next 9 months to 98 pg/mL and 1490 pg/mL, respectively. Follow-up imaging demonstrated the lesion's growth to 38 mm.

The past medical history of the patient included cyanotic congenital heart disease with pulmonary atresia, ventricular septal defect, and a 58-millimeter ascending aorta aneurysm. Additionally, the patient had a mild factor VII deficiency. Progressively, the symptoms reported by the patient included dizziness and photopsia. There were also episodic increases in systolic blood pressure (up to 180 mmHg) and epistaxis. Surgical resection was ruled out during multidisciplinary discussion due to the high surgical risk. AAE was then proposed as an alternative intervention.

The procedure was performed in a monoplane angio-suite (ARTIS-Q, Siemens Healthineers®). Ultrasound-guided right femoral access was obtained using a 5 French 12 cm vascular sheath (Terumo®). Access into the left adrenal artery was obtained using a 5 French SOS Omni Selective catheter (Angiodynamics®) and a Progreat 2.4 French microcatheter (Terumo®). Subsequent angiographic runs demonstrated the main supply to the tumor from the middle branches of the left adrenal artery. Feeding arteries were identified and successfully embolized using 200±75μm HydroPearl microspheres (Terumo®) and 2.5mm EmboCube^{TM} Embolization Gelatin (Merit Medical ®). Postembolization angiographic run demonstrated proper tumor devascularization (Fig. 2). The patient presented mild left flank pain immediately after the procedure. This was successfully controlled with NSAIDs. No other symptoms were reported, and the patient was discharged the next day.

The patient experienced a gradual reduction in the frequency and severity of hypertensive crises over the following days, with blood pressure values of 90-100/50-60 mmHg being reported over consecutive days. Moreover, photopsia completely disappeared. Laboratory testing on day 3 after treatment demonstrated a significant reduction in plasma normetanephrine levels (baseline value: 1490 pg/mL; posprocedure value: 313 pg/mL). Despite the initially favorable findings, normetanephrine levels increased again, reaching 435 pg/mL on day 10, 628 pg/mL on day 18, 742 pg/mL at

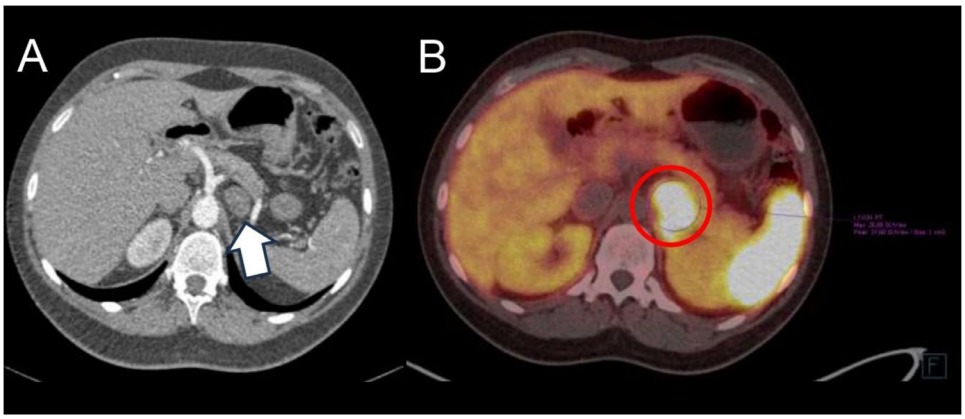

Fig. 1 – Pretreatment imaging. (A) CT on arterial phase depicting a slow-growing pheochromocytoma (arrow). (B) PET-CT with 68Ga-DOTATOC performed showing an intense radiotracer update within the known pheochromocytoma in the left adrenal gland (red circle).

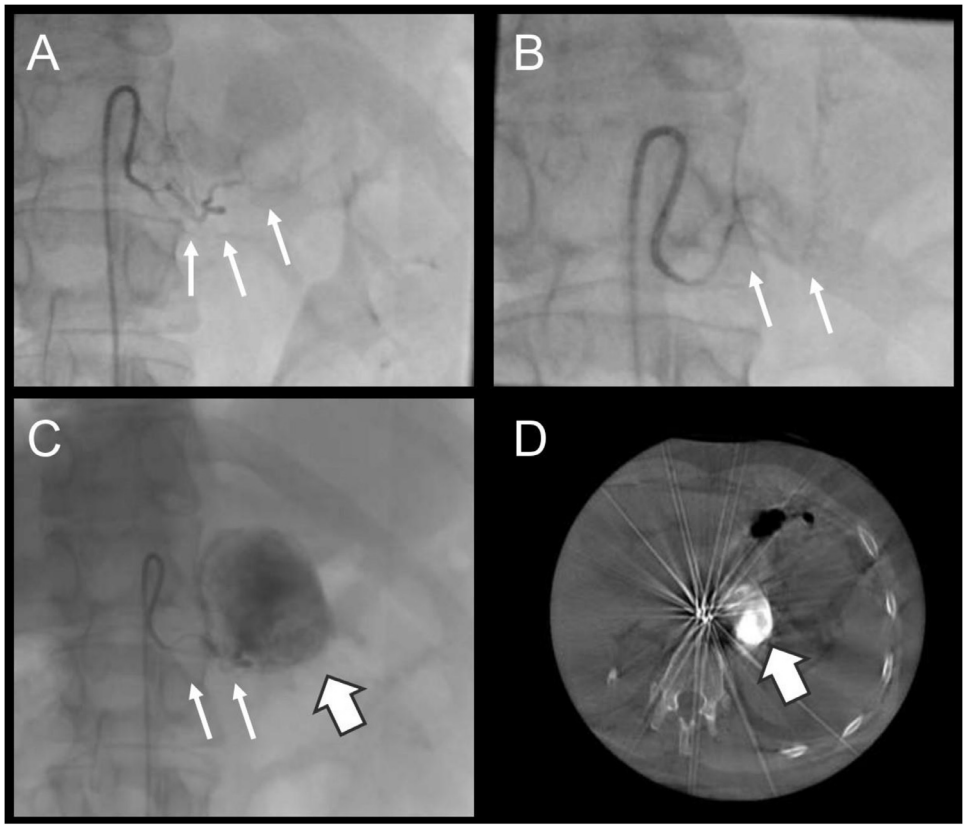

Fig. 2 – Adrenal artery embolization procedure. Digital subtraction angiography before (A), during (B), and after treatment (C) demonstrated catheterization and subsequent embolization of the left adrenal artery (small white arrows) and the tumor (arrow) (D). Postadrenal artery embolization noncontrast enhanced cone-beam CT confirming complete embolization of the left adrenal pheochromocytoma (arrow).

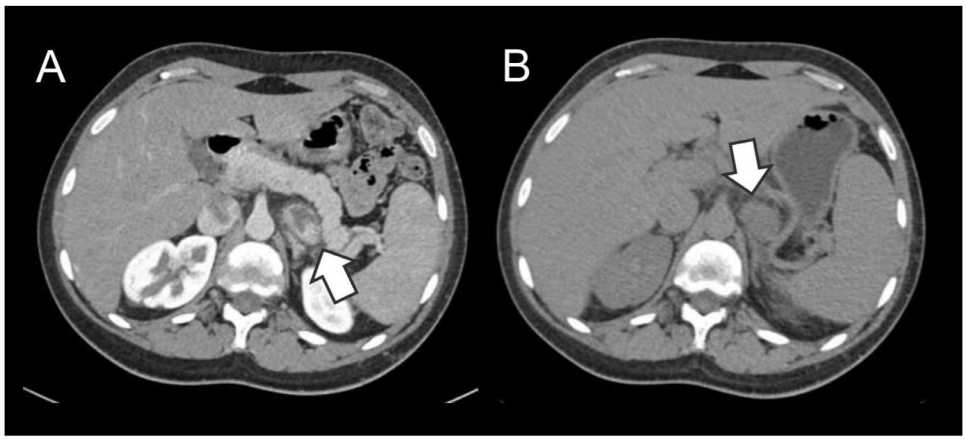

Fig. 3 – Follow-up CT at 2 and 10 months after treatment. (A) Contrast-enhanced CT on late arterial phase at 2 months after treatment showed no changes in vascularization nor size (arrow). (B) At 10 months after treatment, unenhanced CT depicted tumor size stability (arrow).

3 months, 708 pg/mL at 6 months, and 466 pg/mL at ten months post-treatment. Additionally, photopsia and dizziness recurred one month after AAE and persisted over time. Follow-up CT scans acquired at 2- and 10-months post-treatment did not show significant changes in size (Fig. 3) and depicted persisting tumor hypervascularization. Only slight trabeculation of the fat adjacent to the adrenal gland was observed, attributable to postembolization changes.

Discussion

Most symptomatic pheochromocytoma are preferentially treated with surgical resection with medical therapy used for preoperative preparation [4–6]. However, similiar to the case presented, a significant percentage of patients are not candidates for surgery, primarily due to high surgical or anesthetic risk, and remain unresponsive to medical management. Currently, no invasive alternatives to solely medical treatment are established for this group of patients. Two minimally invasive treatments have been described for symptomatic pheochromocytomas: percutaneous ablation and AAE. Ablation has more robust evidence supporting its technical and therapeutic [7–9]. However, in this case, AAE was considered a salvage treatment due to the availability of the technique at our department's and the lack of prior experience with adrenal ablations.

Percutaneous ablation has proven successful in treating adrenal neoplasms, including pheochromocytomas. In a case series involving 23 tumors across 22 patients, Wolf et al. demonstrated that radiofrequency (RFA) and microwave ablation (MWA) are both safe and effective for local control of metastatic lesions and functioning adrenal tumors. Tumor progression occurred in 4 of the 23 tumors, but in the 2 patients with pheochromocytomas treated with RFA, no residual disease was detected postprocedure. Both patients were able to discontinue antihypertensive medications, and their clinical symptoms resolved [7]. Similarly, in a preliminary series of 12 patients with 13 adrenal lesions, including one case of pheochromocytoma, Mayo-Smith et al. achieved effective local control, with normalization of blood pressure and discontinuation of antihypertensive medication in the pheochromocytoma patient [8]. Additionally, Mendiratta-Lala et al. reported symptom resolution and biochemical improvement in a series of 13 patients with functional adrenal tumors, including one case of pheochromocytoma, after a mean clinical follow-up of 41.4 months and a mean biochemical follow-up of 21.2 months [9]. Moreover, percutaneous ablation techniques including RFA may have a role in metastatic pheochromocytomas with potential for local disease control and alleviation of metastasis-related symptoms [10,11]. Despite these promising results, no randomized clinical trials comparing AAE and ablation techniques exist to date, and current evidence is based on case series and retrospective studies.

Outcomes of AAE vary depending on the disease. Regarding hormonal suppression, Lai et al., in a series of 182 cases of primary hyperaldosteronism, demonstrated that superselective embolization of adrenal arteries had a beneficial effect on renal function in these patients [12]. Similarly, Hokotate et al. reported a series of 33 cases of aldosteronomas in which superselective embolization of adrenal arteries proved successful in 82% of cases, with no severe complications identified [13]. In the context of reducing vascularization and tumor size, Sormaz et al. described 3 cases of AAE performed on hypervascular adrenal tumors 24 hours before adrenalectomy. These cases included an 8 cm right pheochromocytoma and 2 adrenocortical carcinomas measuring 17 cm and 18 cm. The results of AAE were favorable, achieving a significant reduction in vascularization, which could potentially reduce the risk of intraoperative bleeding [14]. In another case series, O'Keeffe et al. described 9 cases of AAE in patients with adrenal tumors, including 4 adrenocortical carcinomas and 5 cases of metastatic adrenal lesions. The indications for AAE were to reduce tumor size in all patients, suppress tumor function in 3 patients, provide pain relief in 4 patients, and reduce tumor vascularization in one patient. The results indicated that AAE effectively reduced urinary steroid levels in 2 of the 3 patients with Cushing's syndrome, with a return to normal levels within 48 hours after AAE. Additionally, they reported a decrease in tumor size and vascularization in a patient with adrenocortical carcinoma. Among the 5 patients with metastatic adrenal lesions, 3 who had painful metastatic lesions experienced pain relief following AAE [15].

The published literature regarding the use of AAE in patients with pheochromocytoma is limited to case reports and small case series, and the results are varied. In an emergency setting, Ichikawa et al. reported the case of a patient with a hemorrhagic pheochromocytoma who was successfully treated with AAE in terms of bleeding control. Six months postembolization, a 1 cm reduction in tumor size was observed [16]. Similarly, Giurazza et al., Habib et al., and Hanna et al. described cases of bleeding pheochromocytomas where hemorrhage was successfully controlled through AAE [17–19]. Evidence in AAE for hormonal suppression in pheochromocytomas is less robust. Bunuan et al., Horton et al., and O'Halpin et al. achieved variable durations of symptom control, but all these cases reported tumoral recurrence following the procedure [20–22]. There are also case reports that describe successful preoperative embolization, which effectively reduced catecholamine levels, allowing for safer adrenalectomy [22,23], and a case report that used AAE as treatment in combination with radiofrequency ablation [24]. The patient presented in this case report initially responded well to the treatment; however, by day 10, the clinical values and symptoms began to increase, eventually returning to pretreatment levels by ten months post-treatment. This decline may be attributed to intratumoral revascularization, a relatively common phenomenon in hypervascular tumors across various locations. Considering this, a second embolization session could be considered. However, more evidence is required to consider the usefulness of AAE for treating patients in the long term.

In conclusion, AAE is recognized as a viable and safe procedure employed for reducing tumor vascularization, controlling bleeding, and suppressing hormone secretion before surgical resection. Based on this case, it may also have a potential short-term benefit in terms of symptom relief and biochemical response. Nevertheless, it may not be an effective long-term curative treatment. Due to the current lack of robust evidence in such cases, further collaborative and prospective studies are essential to assess predictors of treatment success and efficacy of AAE.

Patient consent

Informed consent for publication was obtained from patient.

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