Case Report

Outcomes of Arterial Embolization of Adrenal Tumor in Siriraj Hospital: Case Report

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Objective: *To evaluate the treatment outcome of arterial embolization in adrenal tumor.*
Material and Method: *Clinical records of patients with adrenal tumor who underwent adrenal arterial embolization with pre- and post-embolization computed tomography (CT) imaging at Siriraj Hospital, between 2007 and 2013 were retrospectively reviewed.*
Results: *Five sessions of embolization were performed in three patients. The superior adrenal artery was occluded in two patients, the middle adrenal artery in two sessions in one patient, and inferior adrenal artery in two patients. Adrenal arterial embolization successfully reduced tumor vascularity in all patients. The pain relief in all patients was the effect of embolization and complete cessation of retroperitoneal bleeding from ruptured myelolipoma in one patient. There are no late complications such as pleural effusion, adrenal insufficiency, or abscess.*
Conclusion: *Adrenal arterial embolization is a good choice for adjunctive or palliative treatment of unresectable adrenal tumor in particular conditions such as palliative pain control, ruptured adrenal tumor with retroperitoneal hemorrhage, or reduction of tumor vascularity. Even if embolization is not able to reduce tumor size, it is an effective initial treatment before further management, without serious complication.*

Keywords: *Adrenal arterial embolization, Outcome*

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Adrenal tumor is infrequently found in clinical practice. The most common adrenal tumor is benign adenoma, which rarely need surgical intervention unless patients have symptoms or complications from the tumor. Treatment of choice for adrenal tumor is surgery, followed by medical treatment or chemotherapy. In particular cases, surgery cannot be performed at the time of presentation due to advance staging, tumor hemorrhage or unstable vital sign therefore adrenal artery embolization can be a less invasive and safe alternative adjunctive approach. This technique attained successful palliative adrenal tumor control, suppression of excess adrenal hormone production, and maintenance of hemostasis in particular circumstances: ruptured adrenal tumors, traumatic adrenal injury, and adrenal artery aneurysm occlusion[1]. The purpose of the present study was to evaluate the treatment outcome of arterial embolization in adrenal tumor in the authors' institution.

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Material and Method

The present study was approved by the Institution Review Board. All patients with adrenal tumor who underwent adrenal arterial embolization with either pre- and post-embolization computed tomography (CT) or ultrasonography imaging at Siriraj Hospital, between January 2008 and October 2013, were searched by radiological reports. Three patients were enrolled in the study. Clinical records, laboratory data, imaging, treatment, and pathological findings were retrospectively reviewed.

Results

Patients #1 and #2 were initially presented with abdominal pain, while #3 manifested with spontaneous adrenal hemorrhage associated to adrenal tumor. Demographic data and clinical details were shown in Table 1.

Patient #1 (Fig. 1)

The computer tomography (CT) revealed a large well-defined hypervascular mass with multiple spot calcification and central necrosis occupied right suprarenal fossa. In addition, two well-defined hypodensity masses with central low-density occupied

Table 1. Clinical presentation in the case series

No.	Clinical presentation	Side and tumor size	Additional findings	Embolization	Final diagnosis	Additional treatment	Follow-up (months after embolization)
1	35-year-old female; back pain and weight loss for 5 months	Right; 12x12x7 cm	Multiple liver metastasis	1 session	Adrenocortical carcinoma	Right adrenalectomy and adjuvant chemotherapy	Alive (36months)
2	55-year-old female; palpable mass with abdominal pain for 2 months	Right; 18x16x18.5 cm	-	3 sessions	Cavernous hemangioma	Right partial adrenalectomy with right nephrectomy	Alive (26 months)
3	45-year-old male; retroperitoneal hemorrhage	Right; 9.2x8.1x8.4 cm Left; 9.6x11.6x8.6 cm	Ruptured left adrenal tumor	1 session	Myelolipoma	Left adrenalectomy	Alive (25 months)

Laboratory finding: No associated functioning adrenal tumor was identified in all patients.

segment 3 and 4B of liver were identified. Adrenal cortical carcinoma with liver metastasis was considered in this patient.

Pre-operative adrenal arterial embolization was operated prior to surgical resection in order to reduce tumor size and mass effect. Her angiography

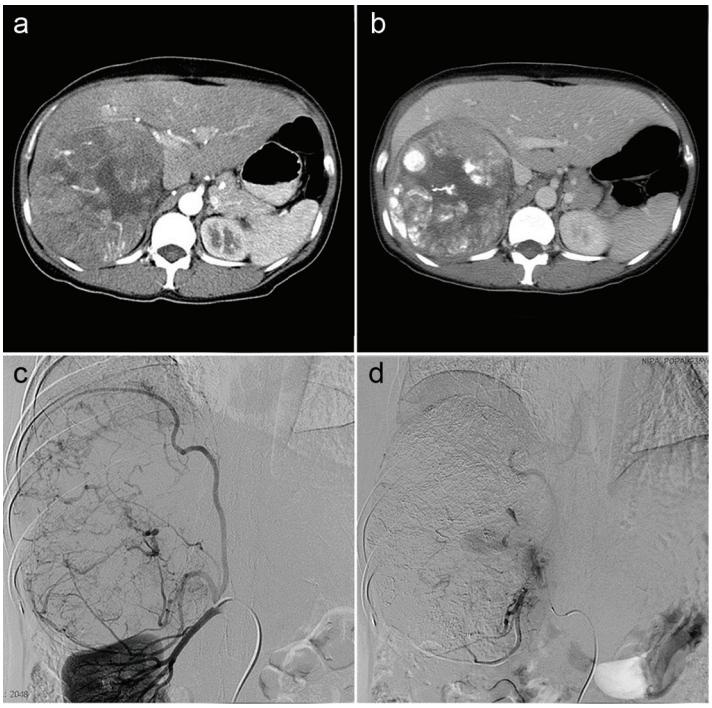

Fig. 1 a) Pre-embolization contrast enhanced CT scan shows a huge well-defined hypervascular mass with multiple spot calcification and central necrosis occupied right suprarenal fossa. b) Post-embolization contrast enhanced CT scan performed at 2 months after right adrenal embolization shows no significant change in size of the right adrenal mass with lipiodol staining. c) Angiogram prior to embolization shows a huge hypervascularized with neovascularized mass at right upper quadrant of abdomen fed from right inferior phrenic and right inferior adrenal artery. d) Post-embolization angiogram shows decreased tumor vascularization and decreased flow at right inferior phrenic artery and right inferior adrenal artery.

revealed a large hypervascularized mass with neovascularization at right upper quadrant of abdomen. This mass was fed from right inferior phrenic and right inferior adrenal artery. The finding indicated that this mass originated from right adrenal gland, therefore, selection into right inferior phrenic artery and right inferior adrenal artery were performed using a 2.7-French microcatheter. Subsequently, chemoembolization was given using mixture of 20 mg of Mitomycin C and 500 mg of 5-Fluorouracil with Lipiodol, followed by Gelfoam. Post-embolization angiography showed a lipiodol-stained mass with markedly decreased tumor vascularization. The less flow of blood into right inferior phrenic artery and right inferior adrenal artery was observed.

Patient #2 (Fig. 2)

A 55-year-old woman with a large well-defined heterogeneous mass at right suprarenal region causing pressure effect on multiple adjacent organs underwent exploratory laparotomy with tumor biopsy from another hospital. The pathological report revealed frank blood without tumor tissue. Thereafter, she was referred to the authors' hospital.

This patient underwent angiography with embolization to reduce tumor size and vascularity. First angiogram revealed a huge hypervascularized-neovascularized mass at right-sided abdomen with tumor staining, in which mainly fed by right superior adrenal artery. Superior portion of the mass was partially fed by right hepatic artery. Selection into inferior branch of right superior adrenal artery was performed with a 2.7-French microcatheter, followed by embolization with 250 to 355 micron-Polyvinyl alcohol (Ivalon) and 355 to 500 micron-Ivalon. Post-embolization angiogram showed more than 50% decreasing in tumor vascularity.

After four-month of lost follow-up, she came back with a slight increase in size of right adrenal mass observed by abdominal CT scan. Subsequently, the second angioembolization was done five months after the first session. At this point of time, new detectable tumor feeders from right middle adrenal artery and some feeders from right renal artery were seen.

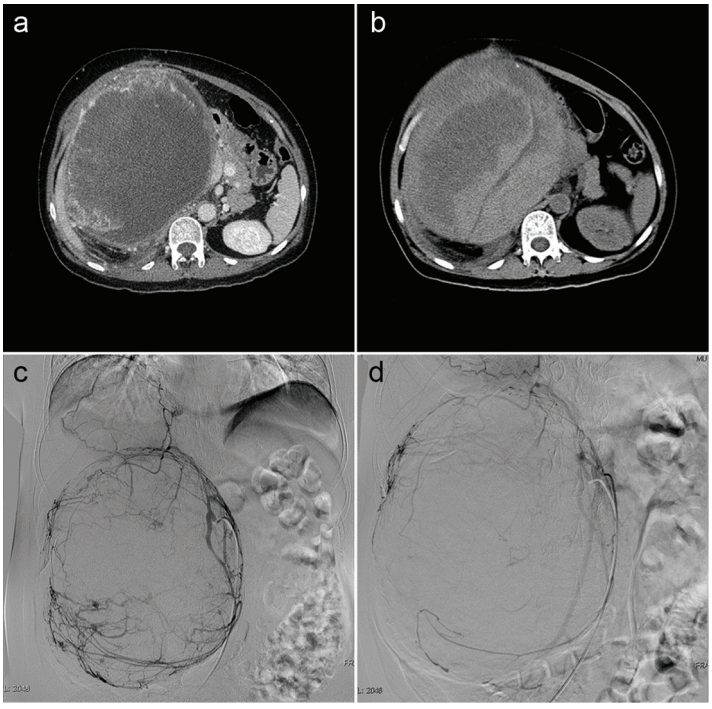

Fig. 2 a) Contrast enhanced CT scan shows a huge well-defined hypervascular mass with multiple spot calcification and central necrosis occupied right suprarenal fossa. b) Emergency non-contrast CT whole abdomen performed at 12 hours after right adrenal embolization shows a large well defined heterogenous hyperdensity mass at right suprarenal region with large intratumoral hemorrhage. c) Pre-embolization Angiogram at right inferior phrenic artery shows a large hypervascularized-neovascularized mass at right sided abdomen with tumor staning. d) Post-embolization angiogram at right inferior phrenic artery shows decreased vascularity of the tumor more than 50%.

Angioembolization of right middle adrenal artery was conducted using 150 to 250 micron-Ivalon. Post-embolization showed nearly obliteration of arterial feeders to this mass.

The third angioembolization was performed three months after the second session, using 150 to 250 micron and 355 to 500-micron-Ivalon into inferior branches of middle adrenal artery. Post-embolization revealed nearly obliteration of arterial feeder from inferior branches of right middle adrenal artery, and the remaining of small arterial feeders from superior branch of right middle adrenal artery.

Patient #3 (Fig. 3)

A 45-year-old man presented with stable bilateral myelolipoma of both adrenal glands. His first abdominal CT scan revealed the enlargement of both adrenal glands, contained large amount of fat with soft tissue density in some portions. Both adrenal tumors had no significant change in size in follow-up by either abdominal CT scan or ultrasonography for two years.

Afterwards, he developed sudden epigastric pain and anemia. The emergency abdominal CT scan from another hospital showed ruptured left adrenal tumor with retroperitoneal hemorrhage. Emergency aderenal angiography with embolization was performed to stop tumor bleeding. The angiographies revealed a large tumor staining at left suprarenal region, fed by branches of the left inferior adrenal artery. Detectable contrast extravasation with a small aneurysm from left inferior adrenal artery was embolized with Ivalon (150-250 micron) via 2.7-French microcatheter. Post-embolization angiography at left renal artery showed a significant lessening of feeding flow into left adrenal myelolipoma with minimal residual feeder branch at medial side of the tumor. Right adrenal tumor showed an arterial feeder from the right inferior phrenic artery without extravasation or aneurysm.

Lastly, this patient underwent left adrenalectomy two years after emergency angio-embolization. No complication from surgery was reported.

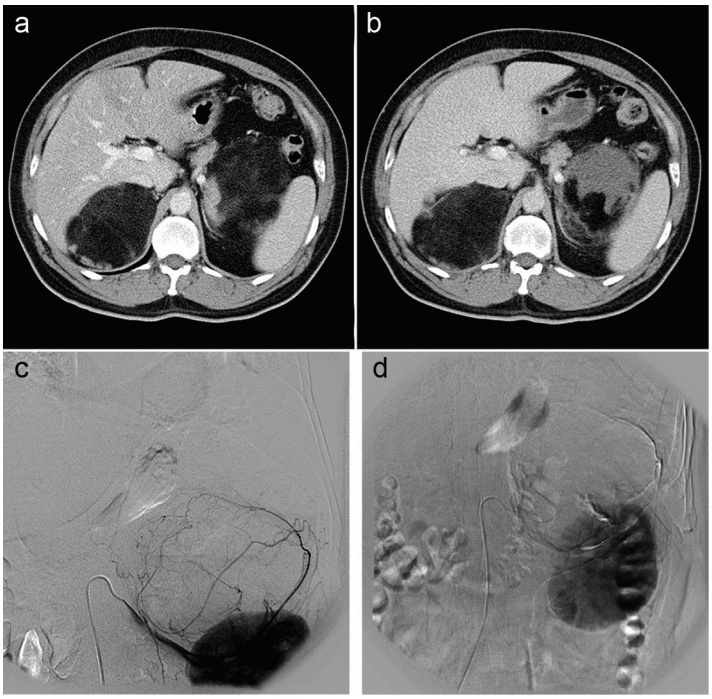

Fig. 3 a) Pre-embolization abdominal CT scan shows enlarge both adrenal glands which contained large amount of fat with soft tissue density in some portions. b) Post left adrenal embolization CT scan shows is increased density in some portion of the adrenal myelolipoma without significant change in tumor size. c) Pre-embolization angiogram shows a large tumoral staining at left suprarenal region as correlated with history of myeolipoma. The tumor is fed by branches of left inferior adrenal artery arising from left renal artery. d) Post-embolization angiogram at left renal artery shows significant decreased feeding flow into left adrenal myelolipoma with minimal residual feeder branch at medial side of the tumor.

Outcomes

Five sessions of embolization were performed in three patients. The superior adrenal artery was occluded in two patients, while the middle adrenal artery was done in two sessions in one patient, and inferior adrenal artery was in two patients.

Adrenal arterial embolization successfully reduced tumor vascularity in all patients. Post-embolization showed a complete cessation of retroperitoneal bleeding from ruptured myelolipoma in patient no.3 and had effective pain relief in all three patients. Two patients were treated by embolization with polyvinyl alcohol (Ivalon) and one patient was with chemotherapy, lipiodol and gelfoam. There was no significant tumor size reduction after embolization in any patients after follow-up imaging with abdominal CT scan. Nevertheless, there is an increased density in some parts of the adrenal myelolipoma after embolization, probably tumor infarction.

There were no mortality results from adrenal arterial embolization. The patient #1 and #3 had fever and right upper quadrant abdominal pain after embolization, which was relieved by paracetamol and recovered within 72 hours. The patient #2 underwent three sessions of embolization and then developed severe abdominal pain with rapid reduction of hematocrit on the day of first session. An emergency abdominal CT scan showed large intratumoral hemorrhage. This patient required an additional five units of packed red cell transfusion and was discharged from the hospital.

In the second and third sessions of embolization, she had mild abdominal tightness after the intervention without late complication such as pleural effusion, adrenal insufficiency, or abscess.

The patient with adrenocortical carcinoma and two liver metastases underwent adrenal embolization, following right adrenalectomy with left hepatectomy and palliative chemotherapy. One year after completion of palliative chemotherapy, she had neither symptom nor evidence of tumor progression until the last day of study (36 months after adrenal embolization). The patient #2 with cavernous hemangioma and #3 with myelolipoma had no detectable progression or recurrent disease at least 25 months after embolization.

Discussion

Adrenal arterial embolization is a minimal invasive procedure that can be used as an alternative or an adjunctive therapy to surgical treatment of adrenal tumor. The present study showed good results in using adrenal embolization for reducing tumor vascularity, pain relief, and treatment of retroperitoneal hemorrhage from bleeding tumor, especially in unstable patient. All three patients have significant reduction in tumor vascularity after embolization. However, adrenal embolization cannot reduce the tumor size in the present study that may be due to large tumor size and several blood supplies.

In previous study by O'Keeffe et al[2], three of the five patients with adrenal metastases treated with arterial embolization experienced effective pain relief. In addition, tumor bulk was stabilized or decreased in three of the four patients who underwent follow-up CT[2]. Compared to the authors' study, follow-up abdominal CT scans in all three patients showed stability in size of the tumors after embolization. All cases had effective pain relief and tumor vascularity reduction.

There are no large studies to support using embolization in unresectable adrenocortical carcinoma patient in purpose of prolonging survival time beyond palliation[3]. In the authors' study, a huge adrenocortical carcinoma patient with liver metastasis had the survival time at least 26 months after adrenal embolization following adrenalectomy and palliative chemotherapy. However, the survival time depends on many factors such as staging, adequate surgical resection, and treatment for recurrent or metastatic diseases[4].

Cavernous hemangioma of the adrenal gland is a rare non-functioning benign neoplasm[5]. The majority of patients underwent surgical resection owing to the impossibility of excluding malignancy in lesions larger than 6 cm, related symptoms in patients with large masses or the risk of spontaneous tumor rupture. A patient in the present study with a huge adrenal cavernous hemangioma received three sessions of embolization to reduce tumor size and vascularity but only vascularity reduction was achieved. Nevertheless, tumor removal after embolization was conducted without massive blood loss.

Myelolipomas are benign tumors composed of fat and bone marrow elements. Although most myelolipomas are asymptomatic and discovered incidentally, they can develop hemorrhage and account for approximately 10% of reported hemorrhagic adrenal masses[6]. Surgical removal of the tumor in acute retroperitoneal hemorrhage should be avoided due to improper oncologic resection and unstable condition of patients. Aim of adrenal embolization in the present study is homeostatic stabilization before

definitive adrenalectomy. Finally, the patient with bilateral myelolipoma underwent unilateral adrenalectomy following embolization to avoid adrenal insufficiency.

Adrenal gland has a small caliber and complex vascular supply. It is mostly supplied by three groups of arteries: the superior, middle, and inferior adrenals arise from the inferior phrenic, aorta, and renal arteries, respectively[7]. In a large adrenal tumor, arterial supply outside adrenal artery can be seen. Due to hypervascularization and neovascularization of the tumor, superselective catheterization into adrenal arteries is better to achieved. The present study used 2.7-French microcatheter for selection into adrenal arteries and adrenal tumor feeders then performed angiography and embolization. Embolization with flow-control technique was used in all patients following post-embolization angiography.

The choice of embolic agent depended on patient disease and target of embolization. Polyvinyl alcohol (Ivalon) was used in arterial embolization of abdominal neoplasms for tumor devascularization. The post-embolization syndrome was the same as that following Gelfoam embolization[8]. The present study used polyvinyl alcohol (Ivalon) for permanent occlusion of distal arterial supply and prevention of latter recanalization of tumors in two cases. Chemoembolization with Mitomycin C, 5-Fluorouracil and Lipiodol, followed by Gelfoam were used in one case with adrenocortical carcinoma for treatment of malignant tumor.

To the best of our knowledge, the severe complication after adrenal embolization is uncommon. Flank pain and fever are the most common complications of adrenal artery embolization, which are self-limited[9,10]. Transient pleural effusion or hiccup from inferior phrenic artery embolization, labile blood pressure, infection, or abscess may also be seen. In addition to intratumoral bleeding, which improved after receiving blood transfusion in one patient, there was no serious complication in the authors' study.

Conclusion

The authors concluded that adrenal arterial embolization is a good choice for adjunctive or palliative treatment of unresectable adrenal tumor in certain conditions such as palliative pain control, ruptured adrenal tumor with retroperitoneal hemorrhage, or reduction of tumor vascularity. Even though embolization could not apparently reduce the tumor size, it is an effective initial treatment before further management without serious complication.

What is already known on this topic?

Adrenal artery embolization is a less invasive and safe alternative approach adjunctive to surgery. This technique is successful for palliative adrenal tumor control, suppression of excess adrenal hormone production, and maintenance of hemostasis in particular circumstances such as ruptured adrenal tumors, traumatic adrenal injury, and adrenal artery aneurysm occlusion.

What this study adds?

There are few studies about adrenal embolization in Thailand. This study was conducted to demonstrate the treatment outcome of arterial embolization in adrenal tumor in the authors' institution.

Potential conflicts of interest

None.

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ผลของการรักษาผู้ป่วยเนื้องอกที่ต่อมหมวกไตด้วยวิธีการอุดหลอดเลือดแดงในโรงพยาบาลศิริราช

กฤษฎี ประภาสะวัต, สมพล รวมเจริญเกียรติ

จุดประสงค์: *เพื่อประเมินผลของการรักษาเนื้องอกของต่อมหมวกไตด้วยวิธีการอุดหลอดเลือดแดง*
วัสดุและวิธีการ: *ทบทวนประวัติการรักษาจากเวชระเบียน ภาพเอกซเรย์คอมพิวเตอร์ และภาพจากการรักษาด้วยวิธีอุดกั้นหลอดเลือดแดงในผู้ป่วยที่มีเนื้องอกที่ต่อมหมวกไต ที่ได้รับการรักษาด้วยวิธีอุดกั้นหลอดเลือดแดงในโรงพยาบาลศิริราช ตั้งแต่ พ.ศ. 2550 ถึง พ.ศ. 2556*
ผลการศึกษา: *ผู้ป่วยที่มีเนื้องอกที่ต่อมหมวกไตทั้งหมด 3 ราย ได้รับการรักษาด้วยวิธีการอุดกั้นหลอดเลือดแดงทั้งหมดรวม 5 ครั้ง โดยอุดหลอดเลือดที่ตำแหน่ง superior adrenal artery ในผู้ป่วย 2 ราย middle adrenal artery 1 ราย และ inferior adrenal artery 2 ราย พบว่า การรักษาด้วยวิธีการอุดหลอดเลือดแดงสามารถลดปริมาณเลือดที่มาเลี้ยงเนื้องอกและลดอาการปวดได้ในผู้ป่วยทั้ง 3 ราย และสามารถหยุดภาวะเลือดออกหลังช่องท้องจากเนื้องอกได้ในผู้ป่วย 1 ราย โดยไม่พบผลข้างเคียงที่ร้ายแรงในผู้ป่วยทุกราย*
สรุป: *การรักษาเนื้องอกที่ต่อมหมวกไตด้วยวิธีอุดกั้นหลอดเลือดแดงเป็นตัวเลือกที่ดีสำหรับการรักษาแบบประคับประคอง เพื่อลดอาการปวดจากเนื้องอก เพื่อรักษากรณีมีเลือดออกจากเนื้องอก หรือ เพื่อลดปริมาณหลอดเลือดที่มาเลี้ยงเนื้องอกก่อนการผ่าตัดต่อไป ถึงแม้ว่าการรักษาด้วยวิธีอุดกั้นหลอดเลือดแดงจะไม่สามารถลดขนาดของเนื้องอกที่ต่อมหมวกไตได้ชัดเจน แต่ก็เป็นวิธีที่มีประสิทธิผลในการรักษาและไม่มีผลข้างเคียงที่ร้ายแรง*