

Superselective Transcatheter Arterial Embolization for Large Unruptured Renal Angiomyolipoma in Lymphangioliomyomatosis

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ABSTRACT

Purpose: To retrospectively evaluate therapeutic performance and complications of superselective transcatheter arterial embolization (TAE) for unruptured renal angiomyolipoma (AML) in patients with lymphangioliomyomatosis (LAM) and to investigate the correlation between percentage reduction in tumor volume and intratumoral fat content.

Materials and Methods: Superselective TAE was performed in 14 consecutive patients with 15 large unruptured renal AMLs associated with LAM (mean age, 38 y; range, 21–57 y). Patients had renal AML with aneurysms ≥ 5 mm in diameter, tumor-related abdominal symptoms, or both. In all cases, embolization of 1 tumor was achieved in a single session by using multiporous gelatin sponge particles (GSPs) with additional metallic coils in all but 1 case. Tumor volume and fat content percentage were measured on CT or MR imaging before and after superselective TAE (median, 11 months; range, 6–14 months).

Results: Residual tumor staining declined by $> 90\%$ after all TAE sessions except 2, with embolization of all treated aneurysms. No nontarget embolization or severe complications were encountered. Mean percentage reduction in tumor volume after superselective TAE was 69% (range, 21%–95%). Percentage tumor volume reduction was negatively correlated with fat content before embolization ($P < .01$).

Conclusions: Superselective TAE with multiporous GSPs and metallic coils for large unruptured renal AML in patients with LAM is useful for reducing tumor volume and treating intratumoral aneurysms without major pulmonary or renal complications. Extent of tumor reduction may be influenced by fat content before embolization.

ABBREVIATIONS

AML = angiomyolipoma, GSP = gelatin sponge particle, LAM = lymphangioliomyomatosis, TAE = transcatheter arterial embolization, TSC = tuberous sclerosis complex

Lymphangioliomyomatosis (LAM) is a rare disease affecting women in their 30s at a rate of approximately 1 or 2 people per 1 million (1). It occurs in 2.3%–3.9% of patients with tuberous sclerosis complex (TSC) (2–5). LAM is classified into 2 types: TSC-associated LAM (TSC-LAM) and sporadic LAM. The latter appears clinically

unassociated with TSC, but both types are caused by mutation of the TSC gene; in both types, LAM cells invade the lungs, lymph nodes, or both. Pulmonary sequelae include dyspnea, cough, chylous pleural effusion, and pneumothorax. Renal angiomyolipoma (AML) is found in 47%–54% of patients with LAM (6,7) at a rate of 100% in TSC-LAM and 50% in sporadic LAM (8). Both TSC-LAM-related AML and sporadic LAM-related AML carry similar risks of bleeding and the need for interventions such as transcatheter arterial embolization (TAE) or nephrectomy (8). Renal AMLs are composed of mature adipose tissue, thick-walled blood vessels, and sheets of smooth muscle (9). Tumor-related symptoms and signs, such as pain, discomfort, abdominal fullness, and hemorrhage, are major problems in renal AML.

TAE for unruptured AML is indicated in patients who have ≥ 1 aneurysms ≥ 5 mm in diameter or abdominal symptoms

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Table 1. Demographics, Clinical Characteristics, and Angiographic Results of Patients

| Tumor | Age, y/Sex | Type [†] | Side | Tumor Size, Largest Diameter, mm | Aneurysms ≥ 5 mm | Symptoms | Devascularization Rate [‡] | No. Metallic Coils | Gelpart, mg | No. Arteries Treated | Days in Hospital after TAE |
|-------|------------|-------------------|------|----------------------------------|------------------|----------------|-------------------------------------|--------------------|-------------|----------------------|----------------------------|
| 1 | 57/F | T (d2) | L | 150 | Single | — | 2 | 9 | 152 | 2 | 7 |
| 2 | 29/F | T (d2) | L | 162 | — | Abdominal pain | 1 | 29 | 142 | 4 | 7 |
| 3 | 35/F | S (d1) | L | 166 | Multiple | — | 1 | 8 | 65 | 3 | 5 |
| 4 | 38/F | S (d1) | L | 144 | Multiple | — | 1 | 23 | 160 | 3 | 10 |
| 5 | 31/F | T (d2) | R | 84 | Multiple | Discomfort | 1 | 1 | 210 | 3 | 7 |
| 6 | 33/F | T (d1) | L | 90 | Multiple | — | 1 | 6 | 128 | 3 | 6 |
| 7 | 37/F | T (d2) | L | 137 | Multiple | — | 1 | 1 | 160 | 3 | 11 |
| 8 | 21/F | T (d1) | L | 92 | Multiple | — | 1 | 2 | 160 | 4 | 8 |
| 9 | 55/F | T (d1) | R | 134 | Multiple | Discomfort | 1 | 21 | 320 | 5 | 8 |
| 10* | 33/F | T (d2) | R | 133 | Multiple | — | 1 | 19 | 200 | 6 | 10 |
| 11 | 33/F | S (d1) | R | 119 | Single | — | 1 | 0 | 160 | 4 | 6 |
| 12 | 34/F | T (d1) | L | 74 | Multiple | — | 1 | 9 | 60 | 6 | 6 |
| 13* | 33/F | T (d2) | L | 67 | Multiple | — | 2 | 19 | 144 | 3 | 10 |
| 14 | 30/F | T (d1) | R | 78 | Multiple | — | 1 | 2 | 16 | 3 | 8 |
| 15 | 25/F | T (d1) | R | 175 | Multiple | Abdominal pain | 1 | 6 | 320 | 6 | 8 |

F = female; L = left; R = right; TAE = transcatheter arterial embolization.

*Tumors 10 and 13 were in the same patient.

[†]T = tuberous sclerosis complex-associated lymphangioleiomyomatosis; S = sporadic lymphangioleiomyomatosis; d1 = definite lymphangioleiomyomatosis (criterion 1); d2 = definite lymphangioleiomyomatosis (criterion 2) (European Respiratory Society criteria).

[‡]1 = > 90%; 2 = 50%–90%.

such as pain, discomfort, and fullness owing to pressure from a large tumor or tumors (10,11). In addition, some studies have considered a tumor diameter of ≥ 4 cm as an indicator for embolization (11–13). To treat AML in patients with LAM, appropriate embolic agents must be chosen to avoid progression of pulmonary dysfunction and nontarget embolization of the normal renal parenchyma, which can cause renal dysfunction. The use of ethanol or small microspheres carries the risk of severe pulmonary complications after arterial embolization (14–16), and nontarget embolization can occur after TAE of the renal artery using ethanol (17,18) or microspheres (19). For these reasons, multiporous gelatin sponge particles (GSPs) were used in the present study. Although careful treatment of both LAM and renal AML is indispensable in these patients, no report has yet focused on the effects and complications of embolization of renal AML in patients with LAM. The influence of fat content before embolization on the effects of TAE has been described previously (20–22). TAE was more effective in tumors with a low fat content before embolization than in tumors with a high fat content before embolization. In this study, the results of TAE in unruptured AML in patients with LAM are reported; percentage fat content before and after embolization was also investigated.

MATERIALS AND METHODS

Patient Selection

Institutional review board approval was obtained for this retrospective study. Informed consent was waived. Patients

with either sporadic LAM or TSC-LAM who had received TAE for unruptured AML were recruited from patients who had received superselective TAE for AML between July 2010 and March 2012 at our hospital. Patients with hemorrhagic AML were excluded. Indications for superselective TAE were an aneurysm or aneurysms ≥ 5 mm in diameter on dynamic contrast-enhanced computed tomography (CT) or magnetic resonance (MR) angiography or abdominal symptoms such as pain, discomfort from compression caused by tumor, and fullness owing to pressure from the renal AML(s), or both. The study included 14 patients with 15 AMLs (Table 1). Three definite criterion 1 sporadic LAMs, 6 definite criterion 1 TSC-LAMs, and 5 definite criterion 2 TSC-LAMs were identified according to European Respiratory Society criteria (1). All patients were women with a mean age of 35 years (range, 21–57 y). One patient had a history of nephrectomy, and 1 patient had received TAE at another hospital. Abdominal pain was reported in 2 patients, and abdominal discomfort was reported in 2 patients. Mean tumor volume on CT or MR imaging before superselective TAE was 400 cm³ (range, 65–938 cm³), and mean maximum tumor length was 120 mm (range, 67–175 mm). In 14 patients, ≥ 1 aneurysms > 5 mm in diameter were detected.

Superselective TAE

All superselective TAE was performed by a single interventional radiologist who had 26 years of experience

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