Journal of Cancer Research and Practice 5 (2018) 112–115

Contents lists available at ScienceDirect

Journal of Cancer Research and Practice

journal homepage: http://www.journals.elsevier.com/journal-of-cancer-research-and-practice

Case Report

Multifocal synchronous angiomyolipomas in a kidney and lungs

Jia-Uei Wong [a, d], Chih-Ming Lin [b], Yu-Chieh Wang [c, *]

[a] Division of General Surgery, Department of Surgery, Cathay General Hospital, Taipei, Taiwan
[b] Division of Urology, Department of Surgery, Cathay General Hospital, Taipei, Taiwan
[c] Department of Surgery, HsinChu Cathay General Hospital, HsinChu, Taiwan
[d] School of Medicine, College of Medicine, Fu-Jen Catholic University, Taiwan

ARTICLE INFO

Article history:
Received 7 December 2017
Received in revised form
26 February 2018
Accepted 2 March 2018
Available online 3 March 2018

Keywords:
Angiomyolipoma
Tuberous sclerosis
Nephrectomy
Therapeutic

ABSTRACT

Angiomyolipoma (AML) is a benign tumor composed of vascular, adipose, and muscle tissues. AML commonly occurs in the kidneys, and is associated with tuberous sclerosis. Different sizes of AML have been reported in the literature. In general, a giant or huge AML usually refers to a tumor size greater than 10 cm in diameter. Several studies have suggested that a giant AML is associated with an increased risk of complications. Herein, we report a case of a giant AML in a 56-year-old patient with abdominal bloating for 6 months. Ultrasound and computed tomography examinations revealed a giant renal AML as well as multiple pulmonary tumors, and thus the patient was scheduled for nephrectomy and pulmonary wedge resection. The resected mass measured 24.6 × 18.4 × 27.3 cm. This report also reviews all previously described cases of large AMLs.

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1. Introduction

Renal angiomyolipoma (AML) is a benign tumor consisting of fat cells, smooth muscle, and vascular tissue. Because recurrence and metastasis of AML are rare, conservative therapy is recommended.[1,2] Most small renal AMLs are asymptomatic and can be treated by observation; however, large AMLs have a higher risk of spontaneous rupture and require aggressive management. Herein, we report the case of a 56-year-old female patient with a giant right renal tumor and multiple visceral angiomyolipomas. She underwent nephrectomy and pulmonary wedge resection due to suspected malignancy, and a pathology study confirmed the diagnosis of multifocal synchronous AML.

2. Case report

A 56-year-old female with no relevant medical or surgical history presented to our Gastroenterology Department complaining of consistent abdominal bloating for 6 months and weight loss of 10 kg. Ascites was noted during a physical examination. Abdominal sonography revealed a giant and irregular hypoechoic renal mass about 20 cm in diameter. An abdominal computed tomography scan showed a large right retroperitoneum mass lesion along with a prominent fatty component and intratumoral vasculature. Two pulmonary tumors were also noted. The first was 2.1 cm in size and located at the right lower lobe, and the second was 5.4 cm in size and located at the left lower lobe (Fig. 1). The diagnosis was possibly retroperitoneal liposarcoma or other type of sarcoma, but less likely renal or liver AML. Laparotomy was then suggested. Before the operation, we consulted a urologist with regards to the placement of a right side double-J stent to prevent ureter injury during surgery. We performed a right thoraco-abdominal incision and radical nephrectomy with en bloc excision on July 29, 2015. For the lung tumor, we performed wedge resection of right lung segment 9 simultaneously in the same incision (Fig. 2). The postoperative course was uneventful. She had no renal insufficiency, atelectasis, or intra-abdominal infection during the postoperative course, and she was discharged. Two months later, she received video-assisted thoracoscopic surgery for wedge resection of left lung segment 10. A genetic test was ordered due to the suspicion of tuberous sclerosis, however the result was negative. One year after surgery, she was asymptomatic and in good health.

The gross specimen, measuring 31.3 × 26.4 × 14.9 cm, was well-defined, homogeneously yellow and composed of solid and soft parts. Immunohistochemistry analysis showed no features of lymphangioleiomyomatosis of adjacent parenchyma. CDK4 and MDM2 stains were negative. HMB-45, desmin and SMA stains were

* Corresponding author. Division of General Surgery, Cathay General Hospital, 280 Renai Rd. Sec.4, Taipei.
E-mail address: yujie@cgh.org.tw (Y.-C. Wang).
Peer review under responsibility of Taiwan Oncology Society.

https://doi.org/10.1016/j.jcrpr.2018.03.002
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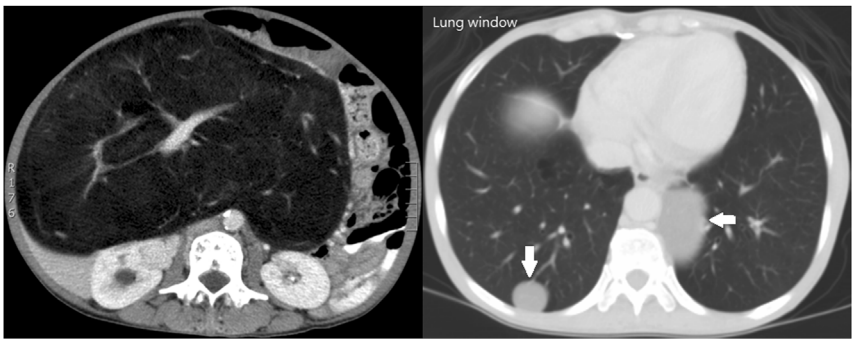

Fig. 1. A CT scan showed tumors in the chest and abdominal cavity simultaneously.

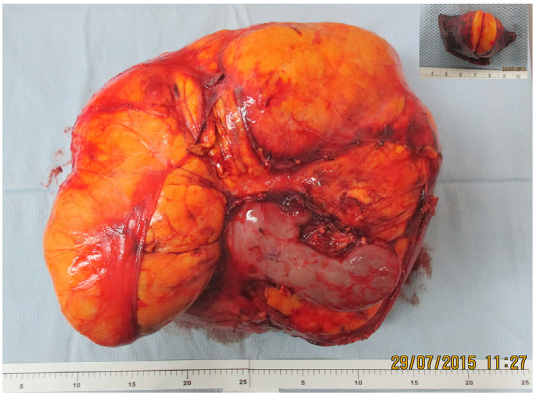

Fig. 2. The huge mass was harvested by radical nephrectomy with en bloc tumor resection under thoraco-abdominal incision. The right lung tumor was also resected during the same operation.

positive with SMA showing the strongest reactivity (Fig. 3). The morphology of the lung tumor was similar to that of the renal counterpart. A lung pathology test also revealed a diagnosis of pulmonary AML. The surgical margin was free (2 mm). The findings confirmed the diagnosis of multifocal synchronous AMLs in both kidney and lungs rather than a metastatic lesion from one another.

3. Discussion

AML belongs to a family of perivascular epithelioid cell tumors (PEComas). According to the WHO, these tumors are histologically, ultra-structurally, and immunohistochemically distinctive perivascular epithelioid cells.[3] AMLs are strongly associated with tuberous sclerosis (TS), a genetic disease that is caused by mutations in either the TSC1 or TSC2 gene. Therefore, AML is usually classified as two variants, one of which is associated with tuberous sclerosis complex (TSC), and the other is sporadic. Most AMLs grow in bilateral kidneys of TSC patients, and can easily be mistaken as renal cell carcinomas.[4,5]

With regards to the management of renal AML, we followed the guidelines provided by Oesterling et al., in 1986.[6] These guidelines suggest that AMLs that are more than 4 cm in diameter can usually be treated by embolization or tumor resection. Furthermore, in order to preserve renal function, partial nephrectomy or enucleation is suggested. Large AMLs can be treated by embolization,

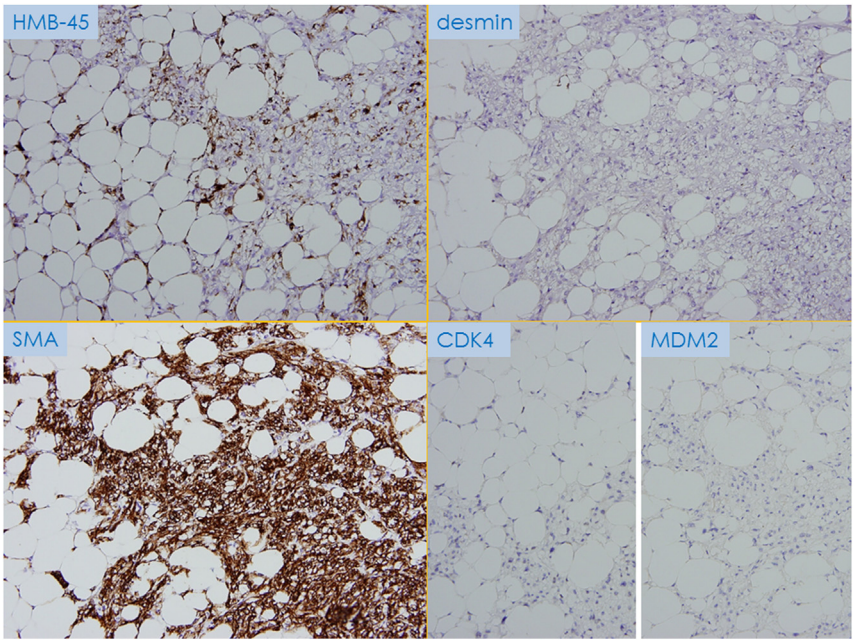

Fig. 3. Immunohistochemistry staining showed that the pathology of the tumor was angiomyolipoma.

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