



Multifocal Pancreatic and Liver PEComas Mimic Metastatic Tumors: Case Report and Literature Review

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Abstract

Background: PEComa is a rare type of mesenchymal tumor. Its morphology includes several types such as angiomyolipoma, lymphangiomyomatosis and pulmonary clear-cell “sugar” tumor with similar immunophenotype. Multifocal PEComa is an extremely rare lesion, which can cause confuse both in diagnosis and treatment. In some cases, it may be diagnosed as a metastatic tumor, and radical treatment is adopted. We reported here a rare case of simultaneous PEComa of the liver and pancreas. At the same time, we reviewed all PEComa cases from 2000 to 2021, found 12 cases of multifocal PEComa, analyzed the clinicopathological characteristics and explored the possible pathogenesis of such lesions.

Case Report: A 28-year-old woman presented with a lesion in the head of the pancreas and liver 5/6 segment respectively, initially suspected of a pancreatic tumor with liver metastasis. A liver mass resection and a pancreatic mass biopsy were performed at the first operation. Both lesions were diagnosed as epithelioid angiomyolipoma. Then the patient underwent a pancreatic segmental resection. After the operation, the patient did not receive any adjuvant treatment and was followed up for 25 months without tumor recurrence or metastasis.

Conclusion: Multifocal PEComas present a chronic clinical course and have a good prognosis. Surgical resection is the main treatment. The occurrence of sporadic multifocal PEComa may be related to the benign blood dissemination of tumor cells. In the diagnosis, in addition to considering metastatic tumors, multifocal lesions also need to be differentiated from multifocal PEComa; in the treatment, radical treatment should not be performed for such type of disease; in the long-term monitoring of the disease, it is necessary to consider the possibility of a second or a greater number of PEComa lesions even many years later.

Keywords: Perivascular epithelioid cell tumor (PEComa); Multifocal; Pancreas; Liver

Abbreviations

PEComa: Perivascular Epithelioid Cell neoplasm; AML: Angiomyolipoma; LAM: Lymphangiomyomatosis; CCST: Clear Cell Sugar Tumor; PEComa-NOS: PEComa-Not Otherwise Specified; CE-CT: Contrast-Enhanced Computed Tomography; TSC: Tuberous Sclerosis Complex; NGS: Next-Generation Sequencing; M: Metachronous; S: Simultaneous

Introduction

Perivascular Epithelioid Cell neoplasm (PEComa) is a group of mesenchymal tumors with different morphologies but similar immunophenotypes. Its families include Angiomyolipoma (AML), Lymphangiomyomatosis (LAM), pulmonary Clear Cell Sugar Tumor (CCST) or other tumor types with perivascular epithelioid cell differentiation (PEComa-not otherwise specified, PEComa-NOS) [1]. Most PEComas are considered to be benign tumors unless the pathological morphology reaches the criteria of malignancy [2]. Usually, PEComa manifests as a solitary lesion. Multifocal PEComa is extremely rare, especially in sporadic cases not related to Tuberous Sclerosis Complex (TSC), which usually confuses clinical diagnosis and management. We reported here a case of multifocal PEComas in the pancreas and liver in a young patient, which was suspected as a pancreatic tumor with liver metastasis before surgery.

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Case Presentation

A 28-year-old woman was referred to our hospital for a lesion on the head of the pancreas. She had no obvious clinical symptoms and no personal or family history of other diseases. Abdominal Contrast-Enhanced Computed Tomography (CE-CT) demonstrated a 1.9-cm-sized low-density lesion at the pancreatic head, which was enhanced in the arterial phase and decreased during the portal and delayed phases (Figure 1). The lesion had an irregular shape with no relation to the pancreatic duct system. Additionally, it was found that a 1.1 cm × 1.0 cm sized low-density lesion was located in the segment 5/6 junction area of the liver (Figure 2). No enlarged lymph nodes were noted in the abdominal pelvic cavity or groin. The patient had no personal or family medical history. Based on these findings, possible diagnoses, such as a solid pseudopapillary tumor or neuroendocrine tumor of pancreas with a metastasis to the liver, were considered.

Then the patient underwent an operation involving "fluorescence laparoscopic liver tumor resection + pancreatic tumor biopsy" to explore the tumor. In the intraoperative frozen tissue pathological examination, a well-defined 1.2 cm × 0.8 cm solitary nodule was noted in the liver segmental resection. The cut surface of the tumor was gray-yellow and soft, and the boundary was relatively clear. The remainder of the liver appeared noncirrhotic. Microscopic examination showed that the tumor cells were epithelioid or spindle-shaped and arranged in a nested pattern. The cytoplasm of the tumor cells was transparent and eosinophilic. No necrosis and mitotic figures were observed. During operation, a pancreatic puncture biopsy was performed; the pancreatic tumor had morphology similar to that of the hepatic tumor. Due to the similar morphology of the tumors at the two sites, it was difficult to determine whether they were metastatic or double primary tumors during surgery. Therefore, it was recommended to suspend the pancreatic surgery and wait for the postoperative pathological diagnosis to decide the next steps of management.

The morphology of the paraffin-embedded tissue showed a small number of fat cells and obvious thin-walled sinusoidal vascular structures in the tumor of the liver (Figure 3A, 3B). The tumor cells were epithelioid or spindle-shaped. Epithelioid cells had obvious nucleoli and central concentrated cytoplasm, which were distributed in the perivascular region in a radial pattern (Figure 3C). Some foci displayed more "myoid" spindle cells arranged in whorled and interlacing fascicles. Additionally, no necrosis or mitotic figures were observed. The same features were noted in the pancreatic tumor (Figure 4A). The immunohistochemical assay showed that tumor cells in the liver were positive for HMB-45, Melan-A, and



Figure 1: the lesion in the pancreas: CT showed that there was a low-density area in the head of the pancreas, the largest cross-section was about 1.9 cm × 1.8 cm, and the lesion was closely related to the head of the pancreas and the proper hepatic artery.

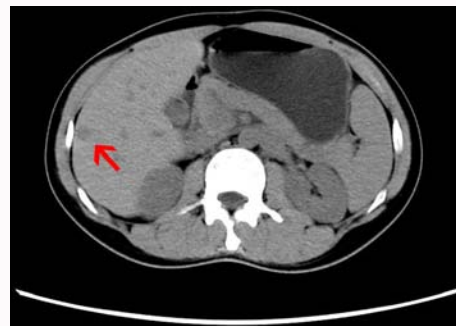


Figure 2: The lesion in the liver: There was a low-density area at the segment 5/6 junction of the liver; the size was about 1.1 cm × 1.0 cm.

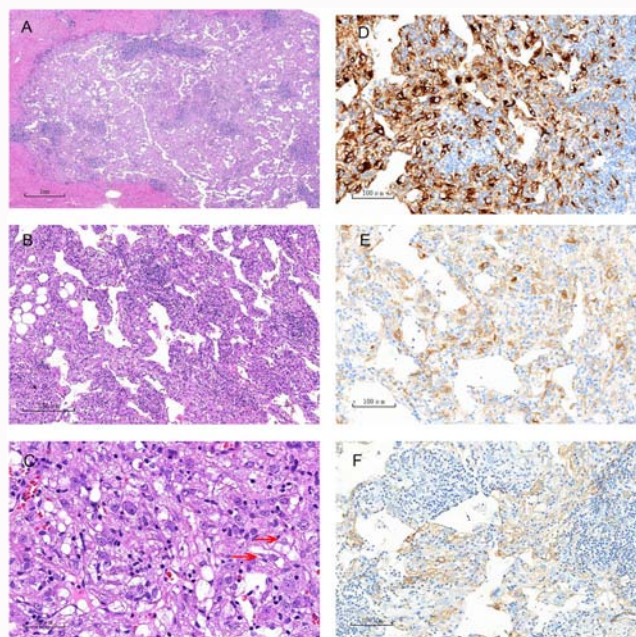


Figure 3: Histological morphology (H&E) and immunohistochemical staining of the lesion in the liver. A) A well-defined intrahepatic nodule. B) The tumor tissue was rich in blood vessels and had a small amount of adipose tissue. C) The tumor was mainly composed of epithelioid cells, among which a small number of short spindle cells and spiderweb-like cells (shown by arrows) were seen. D) HMB45 immunohistochemical staining showed that most of the tumor cells were positively expressed. E) Melan-A immunohistochemical staining showed positive expression in some tumor cells. F) SMA immunohistochemical staining showed weak positive expression in a small number of tumor cells.

SMA (Figures 3D-3F), but negative for AE1/AE3, CK18, CK7, CK20, Hepatocytes, Arg-1 and GPC3. Ki-67 staining revealed a low proliferation index (5%+). The tumor cells in the pancreas were positive for HMB45 (because the tumor component was small, no additional immunohistochemical assay was performed in pancreatic tumors). Therefore, both liver and pancreatic tumors were diagnosed as angiomyolipoma with an epithelioid pattern. Half a month later, the patient underwent "laparoscopic resection of pancreatic tumor + release of intestinal adhesions". The gross examination of the resected pancreatic specimen showed a 2 cm × 2 cm × 1.5 cm solid mass located in the pancreatic body and neck, which was soft, grayish-yellow with clear borders. The histomorphology was similar to the hepatic tumor except for more adipose components (Figure 4B, 4C). No cell pleomorphism, necrosis or mitotic figures, vascular tumor thrombus or nerve invasion were observed. The border of

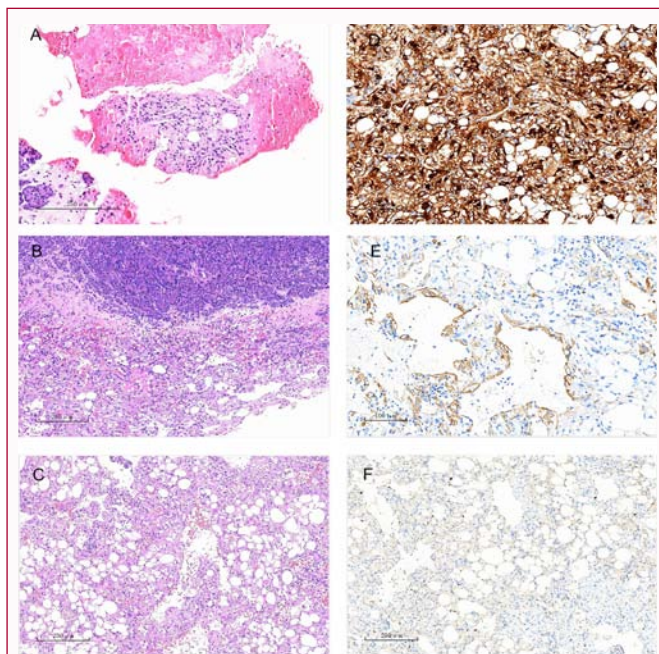


Figure 4: Histological morphology (H&E) and immunohistochemical staining of lesion in the pancreas. A) Intraoperative puncture biopsy of pancreatic tumor. B) There was a lesion in the pancreatectomy specimen, the morphology of which was consistent with the tumor in the liver. C) Compared with liver tumors, pancreatic tumor tissue contained more adipose components. D) HMB45 immunohistochemical staining showed the same diffuse and strong positive expression. E) SMA immunohistochemical staining showed positive in the perivascular area. F) Ki67 staining showed a low tumor proliferation index.

the tumor was not infiltrative. Its immunophenotype was the same as that of the liver tumor, and Ki-67 staining also revealed a low proliferation index (2%+) (Figures 4D-4F). According to the criteria for malignancy of PEComa described by Folpe et al. [2], there was no malignant indication in two lesions. We suggested that the tumors were multifocal epithelioid angiomyolipoma originating from the liver and pancreas rather than tumors with distant metastasis.

Considering that the patient was young and had multiple PEComa lesions, a thorough examination of the patient was performed. No other clinical symptoms related to tuberous sclerosis complex were found. Next-Generation Sequencing (NGS) showed no TSC1/TSC2 germline mutations. After the surgical operation, the patient did not receive any adjuvant therapy. After 25 months of follow-up, she was in good condition, and no recurrence or metastasis was found.

Discussion

Multifocal PEComas are extremely rare lesions. We reviewed PUBMED's English literature from 2000 to 2021 and collected a total of 12 cases (Table 1). Our case is the second case of multiple PEComas involving the liver and pancreas. Because of its rarity, some of these cases had caused clinical diagnosis difficulties or even misdiagnosis, and the opinions of the authors varied about whether they are multiple primary tumors or metastatic tumors. To explore the nature of the lesion, we analyze the clinicopathological characteristics of these cases. Of the total of 12 patients, there were 4 male patients (33%) and 8 female patients (67%). The age was range from 38 to 68, the median age was 47. The most common organs involved in multi-organ cases were the kidney and lung, and the latter was often involved in both lung fields. Of all, 2 cases involved more than 2 organs (case #8: kidney,

spleen and bilateral lung; case #10: liver, left kidney and bilateral lung). Moreover, most cases had multiple foci in one organ. The lung was the organ most frequently affected by multiple foci, followed by liver and kidney. Angiomyolipoma (AML) was the most common type in multiple PEComa. There was no evidence of pathological malignancy in all cases, except for focal cell pleomorphism (case #4), and epithelioid AML (case #12). Some authors believe that pure epithelioid PEComa has malignant potential, and is often related to younger patients, larger tumor sizes and disease progression [3]. While the diagnostic criteria for malignant PEComa summarized by Folpe et al. [4] based on soft tissue and gynecologic origin did not include epithelioid morphology. In fact, PEComa has no widely accepted diagnostic criteria for malignancy so far. Most cases were treated surgically, and no recurrence or metastasis during the follow-up period. One case (case #1) was treated with mTOR inhibitor sirolimus 2 mg/d, in which the multiple AML lesions in the lung and kidney were reduced in 8 months, while the Lymphangiomyomatosis (LAM) lesions in the lung were unchanged. In some cases, multiple small lesions had not undergone any clinical treatment, but did not cause clinical symptoms for a long time. Overall, the prognosis of multifocal PEComas was good. Although some cases did not provide follow-up data, the tumors in most cases remained stable for a long time, and the longest follow-up time recorded was 12 years.

There are different opinions on the pathogenesis of multifocal PEComa, and the exploration of its cause determines the clinical treatment of such lesions. Although the clinical course of PEComa is benign or inert, some authors believe that because of the lack of clear criteria for the diagnosis of PEComa malignancy, multifocal PEComa cases, especially those unrelated to Tuberous Sclerosis Complex (TSC), are potentially malignant and cannot be ruled out as metastatic disease [5]. Other authors believe that these are multicentric lesions [6], but the cause is unknown.

10% to 20% of PEComa cases are associated with Tuberous Sclerosis Complex (TSC), which is due to germline mutations of the TSC1 (9q3.4) and TSC2 (16p13.3) genes [7]. PEComa related to TSC often occurs in young patients with bilateral or multifocal origin and a fast growth rate. According to the clinical diagnostic criteria of TSC proposed by the 2012 International Tuberous Sclerosis Complex Consensus Conference [8], the occurrence of ≥ 2 angiomyolipoma or lymphangiomyomatosis is one of the major features, respectively. Therefore, for multifocal angiomyolipoma or lymphangiomyomatosis cases, it is best to check for other TSC-related clinical evidence or perform genetic testing to assist in the diagnosis of TSC. Among the 12 collected cases, 2 cases were reported as clinically definite TSC (case #4 and #6). The two cases were both bilateral renal angiomyolipoma and multiple lymphangiomyomatosis in the lung. Two other cases (case #1 and #9) showed no mutations in the TSC1/2 gene based on genetic analysis, just like our case, and no other TSC-related clinical manifestations were mentioned in the report, indicating that multifocal PEComas were not always related to TSC. PEComas unrelated to germline mutations in the TSC1/2 gene are sporadic cases. Some of these cases also have somatic mutations in the TSC1/2 gene [9], and some have other genetic changes [10].

The study of Henske, E. P. suggested that PEComa cells could migrate with blood, rather than metastasis [11]. From the clinicopathological characteristics of these multifocal PEComa cases, several points support the opinion: 1), although PEComas exhibit a wide anatomical distribution, the most prevalent locations are

Table 1: List of multifocal PEComa cases reported in the literature.

NO.	Reference	Sex	Age	TSC*	Onset interval**	1 st tumor (Max diameter) (Time of occurrence)	2 nd or more tumor (Max diameter) (Time of occurrence)	Pathological description	Clinical management	Prognosis
1	Sun X [12]	F	38	No (gene assay verified)	M	Right renal: AML (17 years before)	Bilateral lung: multiple LAM, AML; Left renal: AML	N/A	mTOR inhibitor: sirolimus therapy	The renal and pulmonary AML shrank, while pulmonary LAMs were unchanged
2	Neri S [13]	M	38	Unknown	M	Liver: AML (7.5 cm) (5 years before)	Right lung: AML (1.8 cm)	No malignant phenotype	Surgical resection	no recurrence
3	Bhardwaj N [14]	M	61	Unknown	S	Pancreas: solitary AML	Liver: multiple AML	N/A	Surgical resection	Intrahepatic lesions progressed
4	Nasir K and Ahmad A [15]	F	48	Yes	M	Bilateral renal: AML (right: 16 cm; left: 22 cm)	Bilateral Lung: LAM (1.5 cm) (2 month after hemodialysis for bilateral nephrectomy)	Renal AML: pleomorphism focally	Bilateral nephrectomy	The pulmonary tumors were stable
5	Hino H [16]	F	52	Unknown	M	Renal: AML (15 years before)	Lung: multiple AML	No malignant phenotype	Surgical resection	Stable
6	Chang MH [17]	F	39	Yes	S	Bilateral renal: AML	Lung: LAM, MMPH, and clear-cell micronodules	No malignant phenotype	Surgical resection	N/A
7	Gleeson F Cl [18]	F	44	Unknown	S	Right renal: multiple AML (2.6 cm)	Pancreas: multiple AML (0.6-1.2 cm)	No malignant phenotype	Surgical resection	N/A
8	Kasuno K et al. [19]	F	57	Unknown	M	Renal: AML (26 years before)	Bilateral lung: multiple AML (0.5-1.5 cm); Spleen: AML	No malignant phenotype	Surgical resection	No recurrence (followed up for 12 years)
9	Saito M [20]	F	57	No (gene assay verified)	M	Liver: AML (6 cm)	Bilateral lung: multiple AML (0.2-1.0 cm) (5 years later)	No malignant phenotype	Surgical resection	Stable (followed up for 3 years)
10	Kim NR, et al. [21]	M	47	Presumptive TSC (based on clinical presentation)	S	Liver: multiple AML (2-11 cm)	Left kidney: AML (1 cm); Bilateral Lung: multiple LAM	No malignant phenotype	Surgical resection	Lost follow up
11	Dimmler A, et al. [5]	F	68	Unknown	M	Uterine: PEComa (4 cm)	Lung: Multiple PEComa (0.3-2.0 cm) (7 years later)	No malignant phenotype	Surgical resection	No recurrence
12	Mai KT, et al. [22]	M	47	Unknown	M	Kidney: epithelioid AML (15 cm x 12 cm)	Liver: multiple epithelioid AML (up to 5 cm) (9 month later)	90% of epithelioid morphology	Surgical resection	N/A

*"TSC" represents tuberous sclerosis complex; **"M" represents metachronous and "S" represents simultaneous; AML: angiomyolipoma; LAM: Lymphangiomyomatosis; MMPH: Multifocal Micronodular Pneumocyte Hyperplasia

blood-rich organs, such as the kidney, lung and liver. In multifocal PEComas, we found that the lung was the most involved organ (10 out of 12 collected cases). And in almost all metachronous cases (7 out of 8), the later onset tumor is always pulmonary PEComa (LAM or AML). Organs with abundant blood supply are conducive to the dissemination of tumor cells. 2), multifocal PEComa can involve two or more organs or locations, and most cases (11 out of 12) have multi-foci within one organ. The growth pattern of multiple foci in an organ is also similar to that of metastatic tumors. 3), angiomyolipoma, lymphangiomyomatosis or other types of PEComa are rich in vascular lymphatic vessels and exhibit the characteristics of perivascular growth patterns. It is prone to the spread of tumor cells through the vasculature. 4), these multifocal PEComa cases have a chronic clinical course. The onset of the second tumor in the majority of metachronous multifocal PEComa cases (6 out of 8) was found years later (ranged from 5 to 26 years). Therefore, it seemed the onset of multiple PEComa involved tumor cells spreading by blood, which may be due to the operation performed for the first

tumor or the close relationship with the blood vessel wall. During a long dissemination process, migrating cells encounter blood-rich organs, forming multiple tumors with similar cell morphology and immunophenotype. So, the occurrence of multifocal PEComas is the result of distant migration of "angiotropic" tumor cells, which is a type of "benign" metastasis. However, this hypothesis that is inferred from the clinicopathological characteristics of multifocal PEComa must be supported by further molecular biology research.

Conclusion

Multifocal PEComas, excluding metastatic cases of malignant PEComa, are benign or indolent tumors. The genesis of such tumors may be related to the blood dissemination of tumor cells that ultimately encounter distant organs. It is a benign dissemination process rather than metastasis. Therefore, radical treatment should not be performed for such tumors in clinical management. However, for a large tumor, because of rich in blood supply, it requires early treatment for the risk of bleeding and other symptoms. In addition,

although it is rare, for multifocal lesions occurring in organs such as kidney, lung, liver and pancreas, the diagnosis of multiple PEComas should also be considered. And even cases of PEComa with benign pathological morphology should be monitored for a long time, because a second or a greater number of PEComa lesions may appear many years later.

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References

1. Thway K, Fisher C. PEComa: Morphology and genetics of a complex tumor family. *Ann Diagn Pathol.* 2015;19(5):359-68.
2. Folpe AL, Mentzel T, Lehr HA, Fisher C, Balzer BL, Weiss SW. Perivascular epithelioid cell neoplasms of soft tissue and gynecologic origin: A clinicopathologic study of 26 cases and review of the literature. *Am J Surg Pathol.* 2005;29(12):1558-75.
3. Nese N, Martignoni G, Fletcher CD, Gupta R, Pan CC, Kim H, et al. Pure epithelioid PEComas (so-called epithelioid angiomyolipoma) of the kidney: A clinicopathologic study of 41 cases: Detailed assessment of morphology and risk stratification. *Am J Surg Pathol.* 2011;35(2):161-76.
4. Folpe AL, Kwiatkowski DJ. Perivascular epithelioid cell neoplasms: Pathology and pathogenesis. *Hum Pathol.* 2009;41(1):1-15.
5. Dimmler A, Seitz G, Hohenberger W, Kirchner T, Faller G. Late pulmonary metastasis in uterine PEComa. *J Clin Pathol.* 2003;56(8):627-8.
6. Fadare O, Parkash V, Yilmaz Y, Mariappan MR, Ma L, Hileeto D, et al. Perivascular epithelioid cell tumor (PEComa) of the uterine cervix associated with intraabdominal "PEComatosis": A clinicopathological study with comparative genomic hybridization analysis. *World J Surg Oncol.* 2004;2:35.
7. Henske EP, Józwiak S, Kingswood JC, Sampson JR, Thiele EA. Tuberous sclerosis complex. *Nat Rev Dis Primers.* 2016;2:16035.
8. Northrup H, Krueger DA. Tuberous sclerosis complex diagnostic criteria update: Recommendations of the 2012 International Tuberous Sclerosis Complex Consensus Conference. *Pediatr Neurol.* 2013;49(4):243-54.
9. Dabora SL, Jozwiak S, Franz DN, Roberts PS, Nieto A, Chung J, et al. Mutational analysis in a cohort of 224 tuberous sclerosis patients indicates increased severity of TSC2, compared with TSC1, disease in multiple organs. *Am J Hum Genet.* 2001;68(1):64-80.
10. Vannucchi M, Minervini A, Salvi M, Montironi R, Raspollini MR. TFE3 gene rearrangement in perivascular epithelioid cell neoplasm (PEComa) of the genitourinary tract. *Clin Genitourin Cancer.* 2020;18(6):e692-7.
11. Henske EP. Metastasis of benign tumor cells in tuberous sclerosis complex. *Genes Chromosomes Cancer.* 2003;38(4):376-81.
12. Sun X, Feng R, Zhang Y, Shi J, Xu KF. Coexistence of pulmonary lymphangioliomyomatosis and pulmonary angiomyolipoma. *BMC Pulm Med.* 2016;16(1):120.
13. Neri S, Ishii G, Aokage K, Hishida T, Yoshida J, Nishimura M, et al. Multiple perivascular epithelioid cell tumors: Clear cell tumor of the lung accompanied by angiomyolipoma of the liver. *Ann Thorac Cardiovasc Surg.* 2014;20 Suppl:453-6.
14. Bhardwaj N, Garcea G, Lloyd D. A rare case of multi-focal angiomyolipoma affecting the pancreas and liver. *J Surg Case Rep.* 2012;2012(4):5.
15. Nasir K, Ahmad A. Giant renal angiomyolipomas and pulmonary lymphangiomyomatosis. *Saudi J Kidney Dis Transpl.* 2010;21(2):314-9.
16. Hino H, Ikeda S, Kawano R, Sato F, Tagawa K, Hoshino T, et al. Angiomyolipoma in the lung detected 15 years after a nephrectomy for renal angiomyolipoma. *Ann Thorac Surg.* 2010;89(1):298-300.
17. Chang MH, Yao-Peng H. Lymphangioliomyomatosis, multifocal micronodular pneumocyte hyperplasia, and clear-cell micronodules in a 39-year-old woman with tuberous sclerosis: A case report. *Int J Surg Pathol.* 2010;18(6):522-5.
18. Gleeson FC, de la Mora Levy JG, Zhang L, Levy MJ. The differential broadens. EUS FNA appearance and cytological findings of pancreatic angiomyolipoma. *JOP.* 2008;9(1):67-70.
19. Kasuno K, Ueda S, Tanaka A, Tanaka-Kasuno Y, Kuwahara T. Pulmonary angiomyolipoma recurring 26 years after nephrectomy for angiomyolipoma: Benign clinical course. *Clin Nephrol.* 2004;62(6):469-72.
20. Saito M, Tsukamoto T, Takahashi T, Sai K, Fujii H, Nagashima K. Multifocal angiomyolipoma affecting the liver and lung without tuberous sclerosis. *J Clin Pathol.* 2004;57(2):221-4.
21. Kim NR, Chung MP, Park CK, Lee KS, Han J. Pulmonary lymphangioliomyomatosis and multiple hepatic angiomyolipomas in a man. *Pathol Int.* 2003;53(4):231-5.
22. Mai KT, Yazdi HM, Perkins DG, Thijssen A. Fine needle aspiration biopsy of epithelioid angiomyolipoma. A case report. *Acta Cytol.* 2001;45(2):233-6.