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Lumbosciatic Pain Revealing a Cystic Lymphangioma of the Pancreas

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Abstract **Case Report**

The cystic lymphangioma of the pancreas is an exceptional benign vascular malformation especially by its location, its diagnosis is based on imagery and confirmation is made by anatomopathological analysis. The only treatment is the most complete surgical excision possible to prevent recurrences which are frequent. We report a case of a young 26-year-old patient who presented with an atypical symptomatology made up of left lumbosciatic pain as well as abdominal pain. Abdominal MRI showed the presence of a large cystic mass and the anatomopathological examination of the surgical specimen after complete excision confirmed the diagnosis of cystic lymphangioma of the pancreas.

Keywords: Cystic lymphangioma, pancreas, abdominal MRI, Lombosciatic pain.

INTRODUCTION

Cystic lymphangioma is a benign condition, of probable malformative origin, frequently appearing at a young age. The majority of cystic lymphangiomas appear at the cervico-axillary level [1]. Abdominal location is very rare and represents less than 5% in adults and is preferentially located in the mesentery and the retroperitoneum; pancreatic involvement is exceptional [2]. The lack of clinical, biological and radiological specificity means that the diagnosis is histological made on a resection specimen [2]. Complications are numerous and are generally related to location and volume, hence the importance of rapid diagnosis of this abdominal mass in order to avoid very serious abdominal complications (mesenteric ischemia, volvulus, intestinal pain, etc.).

OBSERVATION

This is a 26-year-old male patient, with no particular history, whose onset of symptoms dates back 3 months with the appearance of left lumbosciatalgia resistant to the usual analgesic and anti-inflammatory treatment. The evolution was marked by the appearance of intermittent crampoid type abdominal pain and a feeling of heaviness, especially in the hypochondrium and the left flank. An abdomino-pelvic CT had objectified two abdomino-pelvic peritoneal masses with doubt on a clogged rupture at the level of the ACE, the possible diagnoses were either a cystic mesothelioma of the peritoneum or a cystic peritoneal lymphangioma. Complementary MRI reveals a voluminous intra-abdominal cystic mass, well circumscribed and finely partitioned, probably originating at the level of the back cavity of the omentums and developing forwards in the peritoneal cavity, this mass evokes either a false cyst of the pancreas, i.e. a benign peritoneal mesothelioma. However, other etiologies, in particular a cystic lymphangioma or a dedifferentiated necrotic tumor, cannot be formally ruled out.

The clinical examination finds a conscious patient, well oriented in time and space, a GSC at 15/15, no sensory-motor deficit, a positive Lasègue sign, the conjunctivae were normally colored, a mass index body at 22.04 kg /m2. The abdomen was supple, no tenderness or defense on palpation, no palpable mass, no spleno-hepatomegaly, no collateral circulation.

Cardiovascular examination: heart rate was 70 bpm, blood pressure 121/67, cardiac auscultation revealed no murmurs and peripheral pulses were present.

On the respiratory level, the patient was eupneic with a respiratory rate of 18 c / min, a symmetrical chest expansion, well received vocal

vibrations and well perceived vesicular murmurs, no rales on auscultation and pulsed oxygen saturation was 99 % to ambient air.

The pre-anaesthetic consultation classifies the patient as ASA I, no dyspnea, no exertional angina, functional capacity was greater than 4 METs, a flexible cervical spine, no signs of intubation or difficult mask ventilation with good mouth opening, a thyro-chin distance greater than 6.5 cm and a Mallampati of 1.

On the biological assessment, the hemoglobin was at 15.6g/dl, the platelets at 194000/µl, the leukocytes at 6400/µl, the CRP negative at 1.4mg/l, a natremia at 140mmol/l, a correct serum potassium at 5.2 mmom/l, normal renal function with Urea at 0.29 g/l and creatinine at 9 mg/l, normal liver function tests without cytolysis or biological cholestasis.

Echinococcus serology is negative

CA19-9 and ACE tumor markers were normal (CA19-9: 4.02U/ml and ACE: 3.42ng/ml). The ECG was unremarkable with a regular sinus rhythm, a heart rate of 55 bpm, no arrhythmias or conduction disturbances. A PR space and QT interval within the norms.

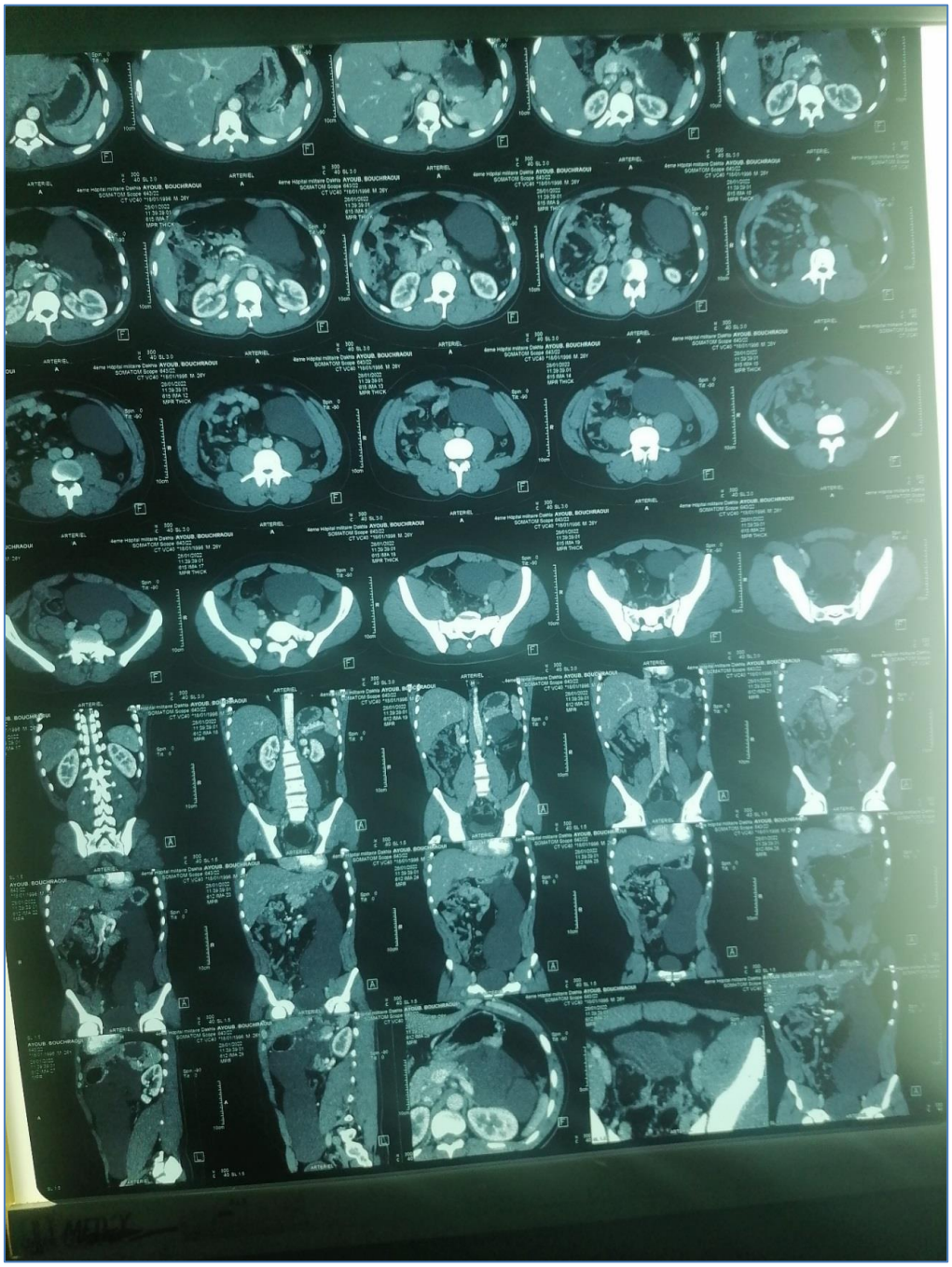

Fig-1: Abdomino-pelvic CT revealing the intra-peritoneal abdomino-pelvic mass

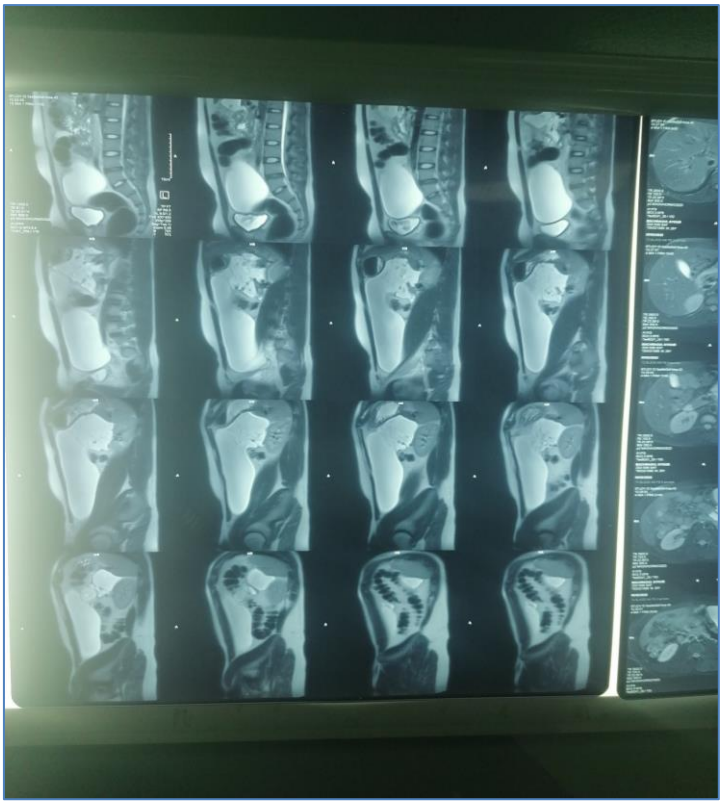

Fig-2: Abdominal MRI showing a large intraperitoneal cystic mass

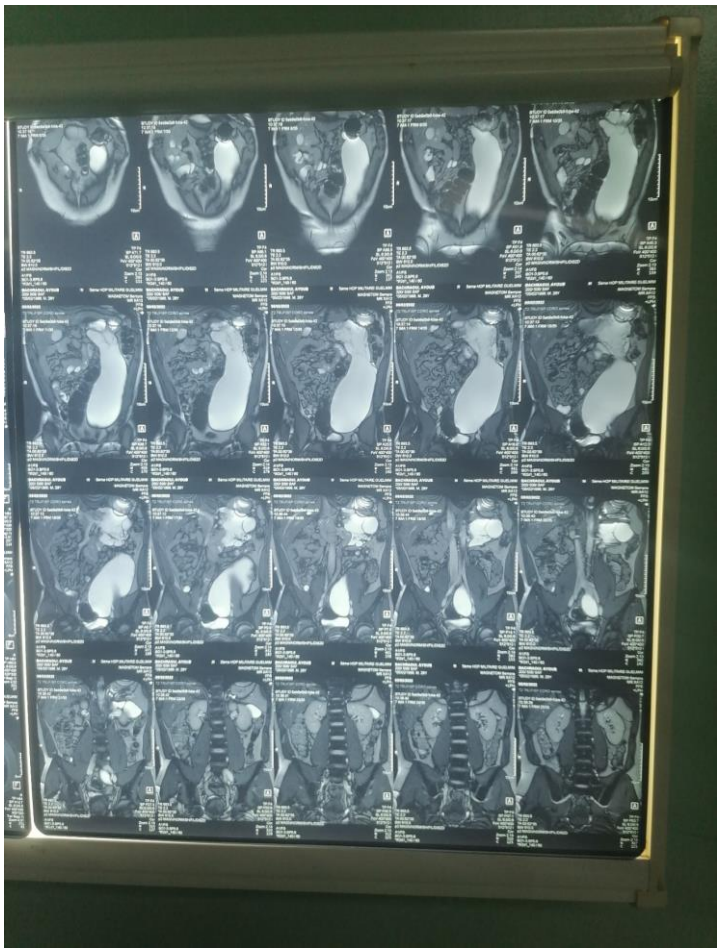

Fig-3: Abdominal MRI revealing the large cystic mass of pancreatic origin

The surgical indication was taken in this patient in front of the large volume of the cyst in order to avoid the evolution towards numerous mechanical, infectious and hemorrhagic abdominal complications as well as the dreaded mesenteric ischemia.

The surgical intervention was done by general anesthesia; induction by 3 µg/kg of Fentanyl (i.e. 225 µg), 2.5 mg/kg of Propofol (190 mg) and 0.6 mg/kg of Rocuronium i.e. 45 mg, easy orotracheal intubation (Cormack-Lehane grade 1) by a probe number 7; maintenance of anesthesia was provided by Sevoflurane 1.7 MAC. Non-invasive monitoring of blood pressure, pulsed oxygen saturation by pulse oximeter, 5-brain electrocardioscope and capnogram. The most complete surgical excision of this cyst was performed via an enlarged midline laparotomy.

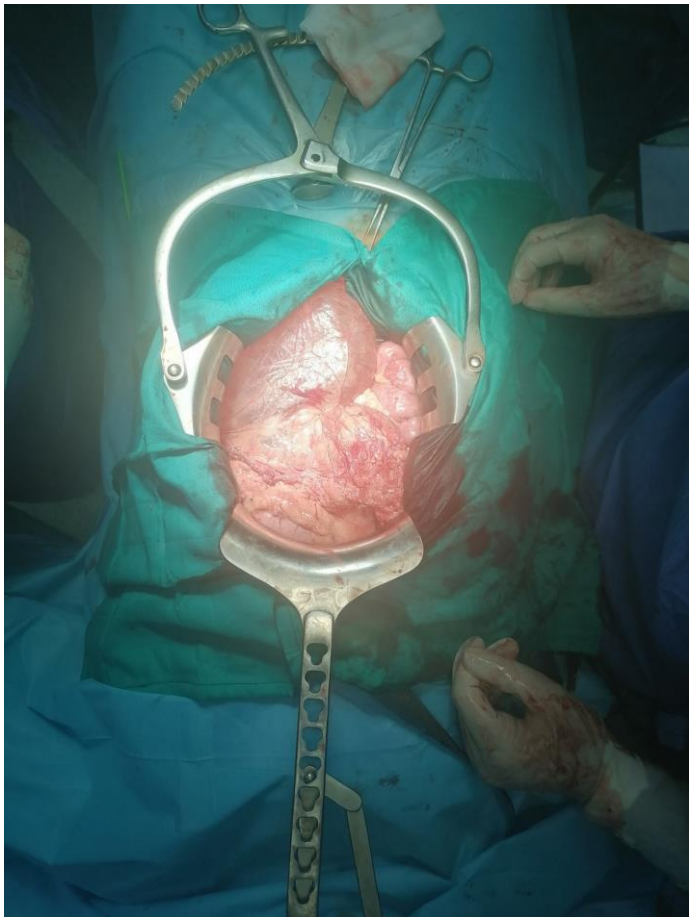

Fig-4: Cystic lymphangioma of the pancreas at the time of laparotomy

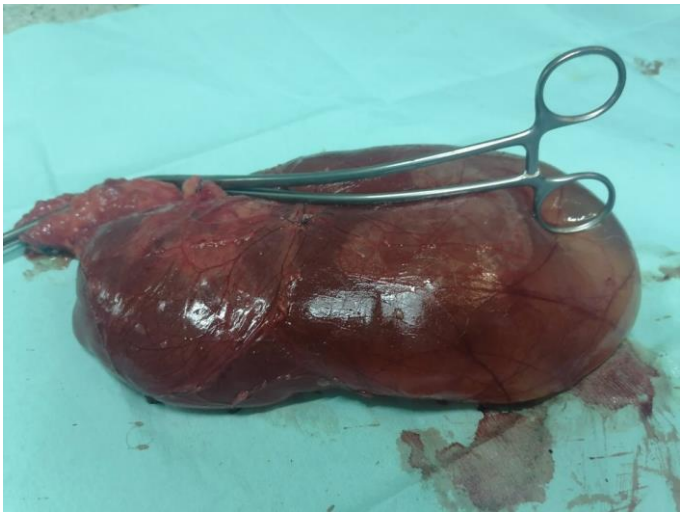

Fig-5: The pancreatic cyst after complete excision

Histological results reveal a cystic mass measuring 14 x 6 x 5 cm attached to pancreatic tissue. The cyst has serous contents, smooth surface and thin wall. The histological study shows a cystic formation made up of numerous lymphatic structures. The wall is thin lined with endothelial cells without cytonuclear atypia and the lumen harbors the lymph associated with lymphocytes and thus confirming the diagnosis and eliminating the signs in favor of malignancy.

The evolution was favorable one month after the operation: good general condition, disappearance of abdominal pain and lumbosciatalgia with an unremarkable abdominal ultrasound. The patient has been informed of the risk of a recurrence and should contact his surgeon again if the symptoms reappear.

DISCUSSION

Cystic lymphangioma is a benign and rare tumor of the lymphatic vessels: its frequency in the mesentery is estimated at 1/100,000 in adults and 1/20,000 in children [1, 3], while pancreatic localization is exceptional. These malformative vascular tumors correspond to a sequestration of lymphatic tissue secondary to an anomaly in the embryological development of the lymphatic system [4]. The LKP is located in 2/3 of cases in the corporeo-caudal region [2, 5, 6]. The size of the tumor is variable between 2 and 20 cm [2]. The non-specific and polymorphic clinical presentation of cystic lymphangioma is linked to the tumor volume, the variable location and the types of complications it generates (mechanical/ infectious/haemorrhagic) [1, 3]. The unusual location in the pancreas that we observed has no specific clinical signs [5]. Small cysts are often asymptomatic and are discovered incidentally [5]. For cysts over 2 cm in diameter, they can be revealed by chronic abdominal pain, retentional jaundice (head location) or an epigastric mass [2, 4, 6]. There is a particularly rare clinical form, in the form of cystic dissemination mimicking peritoneal carcinomatosis, called peritoneal cystic lymphangiomatosis [7]. In our observation, the revealing signs were intermittent pain in the hypochondrium and the left flank and a symptomatology which is perhaps described for the first time in the literature: left sciatica. Sometimes, the diagnosis is made after complication: rupture, torsion, hemorrhage, infection of the cyst, or vascular, digestive or biliary compression [2, 4]. No sign is specific and it is the imaging that guides the diagnosis.

On ultrasound, lymphangiomas appear as encapsulated hypo- or anechoic uni- or multilocular masses with a thin, well-defined wall [6]. In computed tomography, the tumor is homogeneous liquid, hypodense before and after injection of contrast product [6]. A fatty content characteristic of the presence of chylous fluid can be objectified by a negative density [8]. MRI is currently the reference examination [9]. It makes it possible to better delimit the cranio-caudal extension of the lesion and to diagnose certain complications, in particular intracystic hemorrhage [5]. In case of liquid content, there is a hyposignal in T1, a hypersignal in T2 which is reinforced on late echoes in T2 [5]. A fatty signal in the cyst is characteristic and results in an iso- or hypersignal in T1, a hypersignal in T2 decreasing in late T2 [9]. The partitions and the walls are in general in hyposignal with the 2 sequences T1 and T2. Gadolinium uptake by the wall and septa can however be observed [9].

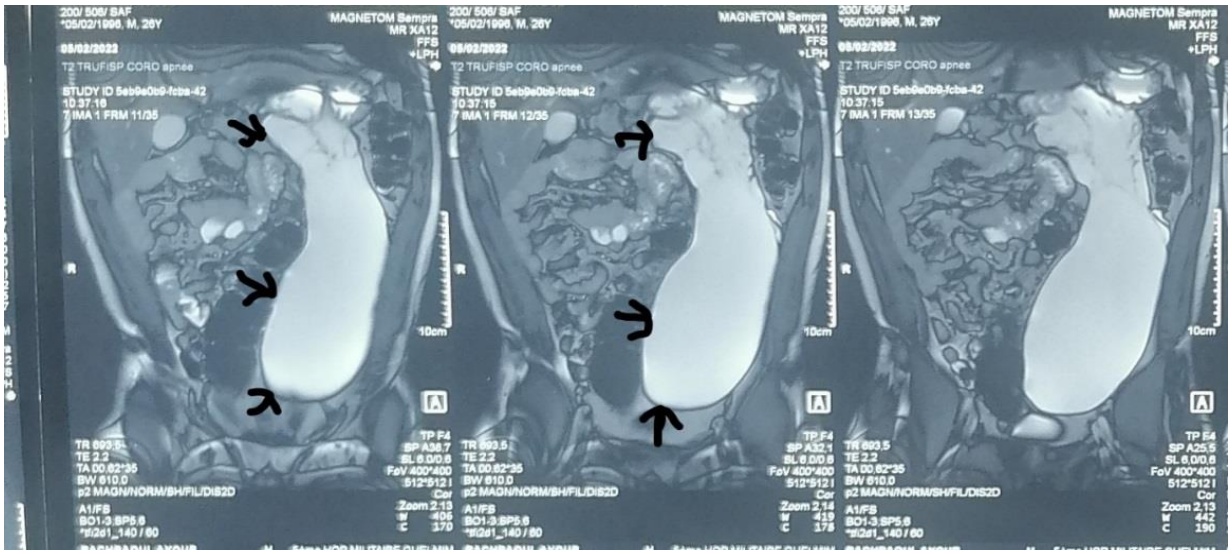

Fig-6: Abdominal MRI of our patient showing cystic lymphangioma originating in the pancreas

Nevertheless, the diagnostic certainty is provided by the anatomopathologic analysis of the tumour. Macroscopically, cystic lymphangioma can be single or polycystic, with oligo-macrocystic, micro-polycystic and mixed forms [7]. Microscopically, three criteria are necessary for diagnosis: 1) it is a cystic formation; 2) septa consist of a connective stroma with lymphoid tissue and smooth muscle; 3) the cyst is lined

with a lymphatic-like endothelial lining (positive for factor D2-40) demonstrating the vascular origin of the tumor[1,7] . Malignant transformation is exceptional [2].

The treatment is surgical [2, 6]. Complete excision of the tumor is the treatment of choice [6]. For the corporeo-caudal location, some authors perform a left pancreatectomy with or without preservation of the spleen [2]. Whatever the surgical technique chosen, prolonged monitoring (ultrasound and abdominal computed tomography) must be systematic in search of a recurrence [9]. The recurrence rate is 40% after incomplete resection and 17% after macroscopically complete excision [1, 5]. Aspiration of the cystic contents with or without injection of a sclerosing product constitutes a therapeutic alternative, especially in the case of easily accessible oligo-macro-cystic lesions, however the recurrence rate is very high.

CONCLUSION

The LPK represents a progressive and rare benign malformative tumor of the lymphatic system, the diagnosis is generally delicate and should be evoked in front of any cystic mass of the pancreas. Diagnostic confirmation is provided by anatomopathological analysis of the tumour. Complete surgical excision is the only treatment associated with prolonged survival and to avoid possible recurrences.

REFERENCES

1. Mabrut, J.Y., Grandjean, J.P., Henry, L. (2002). Les lym-phangiomes kystiques du mésentère et du méso-colon.Prise en charge diagnostique et thérapeutique. *Ann Chir, 127*, 343-349
2. Paal, E., Thompson, L.D., Heffess, C.S. (1998). A clinicopathologic and immunohistochemical study of ten pancreatic lymphangiomas and a review of the literature. *Cancer, 82*; 2150–8.
3. Prabhakaran, K., Patankar, J.Z., Loh, D.L.S.K. (2007). Cysticlymphangioma of the mesentery causing intestinal obs-truction. *Singapore Med J, 48*, 265-266
4. Lyngdoh, T.S., Konsam, R., Th, B. (2008). Giant cystic lymphangioma of pancreas. ANZ J Surg, 78; 673–4
5. Goh, B.K., Tan, Y.M., Ooi, L.L. (2006). Hepatobiliary and pancreatic: cystic lymphangioma of the pancreas. *J Gastroenterol Hepatol, 21*; 618.
6. Colovic, R.B., Grubor, N.M., Micev, M.T. (2008). Cystic lymphangioma of the pancreas. World J Gastroenterol, 14; 6873–5.
7. Bezzola, T., Bülher, L., Chardot, C. (2008). Le traitementchirurgical du lymphangiome kystique abdominal chezl'adulte et chez l'enfant. *J Chir*, 145, 238-243.
8. Jathal, A., Arsenescu, R., Crowe, G. (2005). Diagnosis of pancreatic cystic lymphangioma with EUS-guided FNA: report of a case. *Gastrointest Endosc, 61*; 920–2.
9. Leung, T.K., Lee, C.M., Shen, L.K. (2006). Differential diagnosis of cystic lymphangioma of the pancreas based on imaging features. *J Formos Med Assoc*, 105; 512–7.