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A Case Report of Hemolymphangioma of Mesentery in Adults

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List of Abbreviations:

CDK4: Cyclin-dependent Kinase 4**;** MDM2: Mouse Double MMinute 2**;** Ki-67: Antigen Kiel-67**;** CA125: Carbohydrate Antigen 125; CT: Computed Tomography**;** HMB45: Human Melanoma Black 45**;** SMA: Spinal Muscular Atrophy**;** CD31: Platelet Endothelial Cell Adhesion Molecule-1**;** CD34: Cluster of Differentiation 34**;** VEGF; Vascular Endothelial Growth Factor**;** AFP: Alpha-Fetoprotein**;** CEA: Carcinoembryonic Antigen**;** CA199: Carbohydrate Antigen 199**;** SD: Stable Disease

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Abstract .1

A 34-year-old young female patient was admitted to hospital with abdominal discomfort and intermittent fever for more than 1 month. Puncture pathological result: except 1. Vascular-derived tumours (especially lymphangiomas); 2. Angiomyolipoma. Abdominal examination: mild tenderness in the whole abdomen, no rebound pain and muscle tension, liver and spleen not palpable below the costal margin, Murphy's sign was negative, a huge mass about 10*20cm in size could be palpated on the right abdomen, the boundary was unclear, tenderness upon palpation, .and the abdominal percussion was solid sound

Case Report .2

A 34-year-old young female patient was admitted to hospital with abdominal discomfort and intermittent fever for more than 1 month. Multiple puncture biopsies were performed in the other hospital, and the pathological results showed that fibrous adipocytes were examined, and there were numerous irregular vascular-like structures, focal smooth muscle tissue hyperplasia, and focal lymphocyte infiltration, which were consistent with mesenchymal lesions, except [1]. Vascular-derived tumors (especially lymphangiomas) [2]. Angiomyolipoma. Immunohistochemical results: CDK4 (-), MDM2(+/-), S-100 (-), Ki-67 (about 5%+). Previous good health, no family history. Abdominal examination: mild tenderness in the whole abdomen, no rebound pain and muscle tension, liver and spleen not palpable below the costal margin, Murphy's sign was negative, a huge mass about 10*20cm in size could be palpated on the right abdomen, the boundary was unclear, tenderness upon palpation, and the abdominal percussion was solid sound. Blood routine: hemoglobin 76g/L, blood biochemistry: total bilirubin 24.7μmol/L, direct bilirubin 10.5μmol/L, tumour marker: CA125:106 [3]. U/ml, other blood tests and urine analysis were normal. Abdominal enhanced CT showed an irregular mass in the right midabdominal mesentery with a maximum cross-sectional area of 11.2*20.9cm. CT enhanced scan did not show significant lesion enhancement. Radiologic diagnosis: Abdominal cavity mass, lymphangioma considered possible. Preoperative diagnosis: retroperitoneal tumour (Figure 1). The patient underwent an exploratory laparotomy. Intraoperative exploration: There was a huge mass in the abdominal cavity, from the hilar of the liver to the entrance of the right pelvic cavity. The right boundary was located at the anterior axillary line, and the left boundary reached the medial medial line of the left clavicle, with a size of 40cm*30cm*20cm (Figure 2). The tumour originated from the root of the small intestine mesentery, wrapped around the pancreatic head, duodenum and mesentery vessels, and pushed the duodenum and Trochii ligament to the right lower abdomen. The right half colon was pushed to the upper abdomen, and no space was found in the entire colon and upper rectum. The tumour compresses the gallbladder duct and causes gallbladder enlargement, about 10cm*4cm*4cm, with high surface tension. Duodenal bulb and descending part were obstructed by compression, and lumen was narrow. The small intestine 150cm to 110cm away from the ileocecal part was caused by vascular invasion caused by tumour. Because the tumour enveloped the superior mesenteric artery, radical resection was impossible, so it was decided to perform partial resection of mesenteric mass + partial resection

of small intestine + cholecystectomy + gastrojejunostomy + enter enterostomy. Histological examination after surgery revealed partial resection of mesenteric masses: more dilated lymphatic vessels in mature adipose tissue with thin tube walls and lymph fluid, scattered and small clusters of lymphocytes around the tube walls, and a small number of moderately large malformed muscular vessels with a size of about 9*6*4cm.Immunohistochemical results [5]: HMB45 (-), MelanA (-), SMA (vascular +), Ki-67 (<3%+), MDM2 (-), CDK4 (+), P16 (individual +), CD31 (+), D2-40 (minority +), CD34 (+), VEGF (weak +). (Figure 3,4). The patient recovered well after surgery without any related complications. The antiangiogenic drug everolimus is being treated. 5 months after the operation, the tumour did not increase .[significantly and SD was evaluated [2

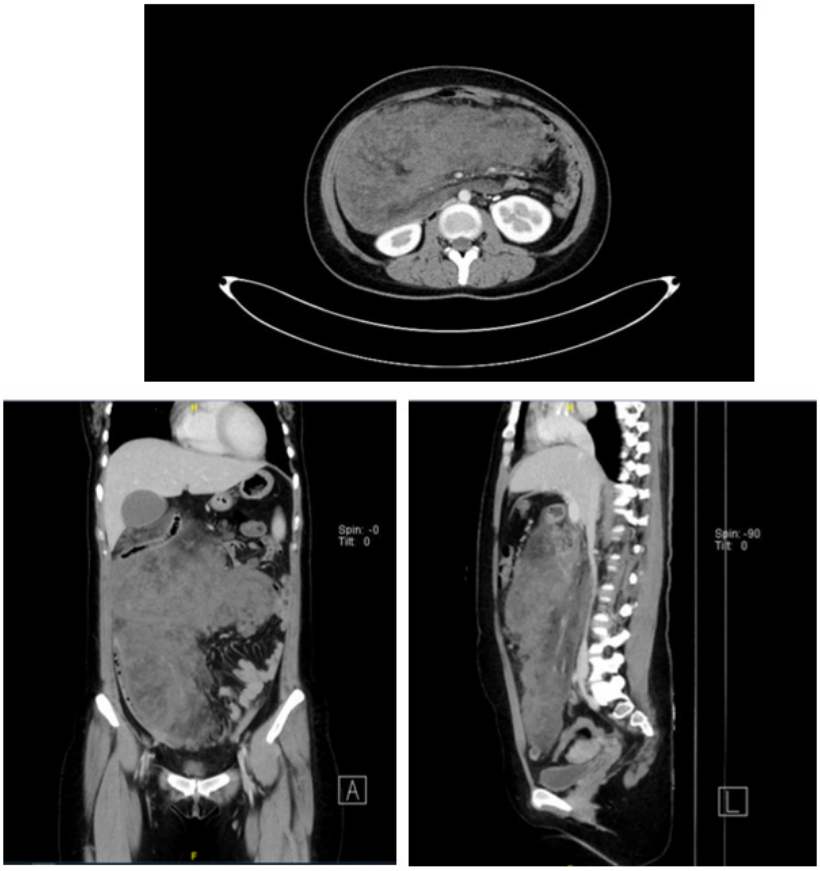


Figure 1: Abdominal enhancement CT revealing an irregular mass. The maximum cross-sectional area of the mass was approximately 11.2 cm × .20.9 cm, The contrast-enhanced CT scans show no obvious enhancement

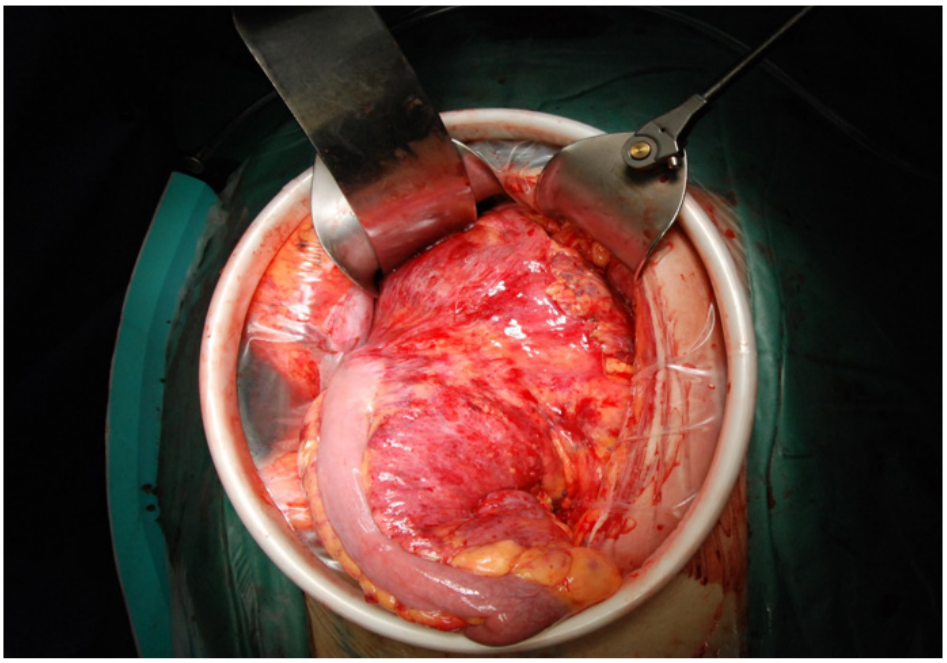
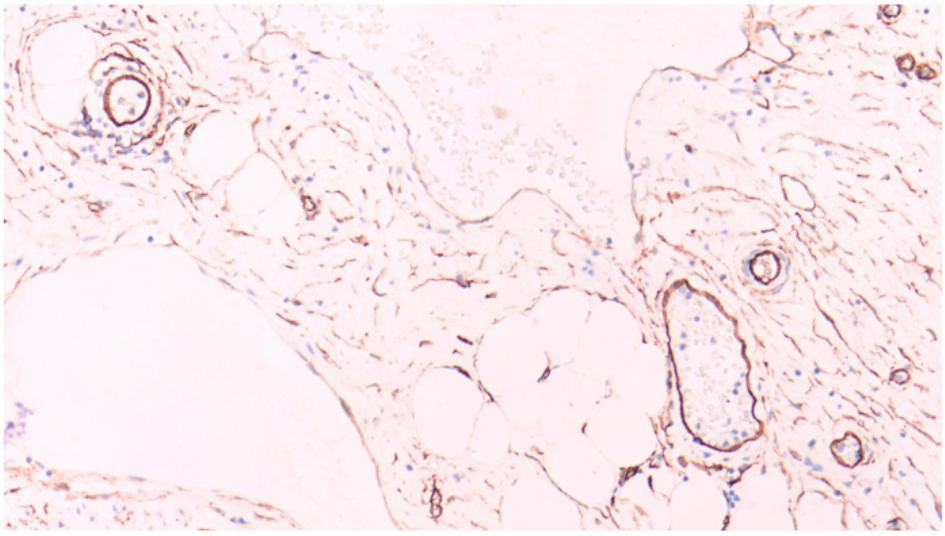
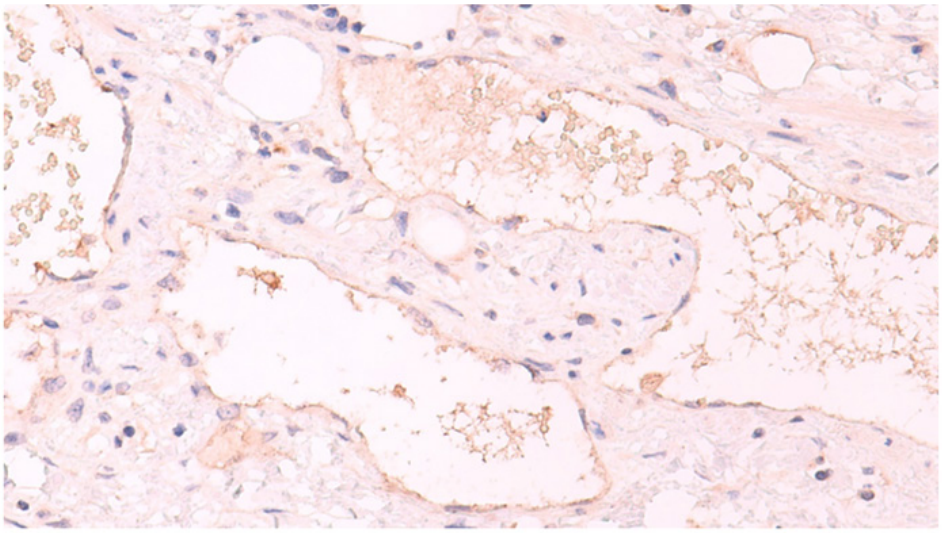


Figure 2: Intraoperative tumour appearance.



（CD34+）

Figure 3: Immunohistochemical: staining is positive for CD34 in the vascular endothelial cells.



(VEGF weak +)

Figure 4: Immunohistochemical: staining is positive for VEGF in the vascular endothelial cells.

Discussion .3

Hemolymphangioma, also known as Hemangi lymphangioma, is a rare vascular malformation of the venous lymphatic vessels, consisting of hyperplasia or a network of vascular Spaces, including lymphatic vessels, capillaries, veins or arteries, which are lined with bland endothelial cells and a connective tissue matrix in the middle [4]. This is a rare lymphangioma that presents as a mixture of blood and lymphatic vessels. According to Landing and Farber, lymphangiomas can be divided into four subtypes based on the degree of lymphatic dilation: capillary hemangiomas, cavernous hemangiomas, cystic lymphangiomas, and hemolymphangiomas. Its incidence ranges from 1.2 to 2.8 per 1,000 births, and is roughly the same for men and women [5]. Although lymphangiomas can occur at any age and in any part of the body, about 90% of cases occur in children under 2 years of age, most commonly in the head and neck. These lesions are rarely found in adult patients [5]. The formation of these tumours may be explained by venous lymphatic obstruction, membranous proliferative vascular tissue, and systemic circulation [6]. Hemolymphangioma is a rare type of lymphangioma that presents as a mixture of blood vessels and lymphatic vessels. hemolymphangioma usually appears on the surface of the body in infants and children. The incidence of hemolymphangioma in adults is very low, with only a few reports in the literature. Hemolymphangioma in adults mostly occurs in the head and neck, and only a few cases have been reported in the digestive system, such as liver [1] and pancreas [2,3]. We report a 34-year-old female patient with hemolymphangioma of small intestine mesenteric. Hemolymphangioma of the small intestine and its mesentery is very rare in clinical practice. The formation of this tumour is primarily congenital, but the exact mechanism is unknown. We reviewed the literature by searching PubMed for keywords such as “hemolymphangioma” and “small bowel mesentery.” At present, only 2 cases of small intestinal mesen-

teric lymphangioma have been reported [7-8]. In general, hemolymphangioma can be divided into congenital and acquired benign tumours. Impaired venous lymphatic communication between vascular tissue and systemic blood circulation may be the cause of congenital hemolymphangioma. However, some studies have speculated that poor lymphatic drainage due to trauma or prior surgery may be the cause of secondary hemolymphangioma [5]. The clinical manifestations of abdominal lymphangioma vary in location and size. Abdominal pain and abdominal mass are common symptoms in patients with hemolymphangioma of the pancreas [2,3]. Hemolymphangioma of digestive tract is characterized by anaemia and gastrointestinal bleeding [9-11]. Small intestinal mesenteric tumours may cause symptoms such as abdominal pain, intestinal obstruction, and gastrointestinal bleeding when the bowel constricts or the tumour ruptures. Because hemangioma is indistinguishable from other small bowel diseases, preoperative diagnosis is difficult. Routine blood tests and tumour markers such as AFP, CEA, CA199, and CA12-5 usually show normal results. Abdominal CT examination has important clinical value in the diagnosis of abdominal lymphangioma. Because the proportion of blood vessels in hemolymphangioma is different, it shows different enhancement features on imaging. Vascular-rich tumours usually show significant enhancement, which is more pronounced in the venous and delayed stages. The tumour with low vascular ratio is not obvious in enhancement, and it is easy to misdiagnose. In addition, CT is useful in determining the extent and invasion of the mass and in developing surgical strategies [12]. In our study, enhanced CT scans of the abdomen were performed and there was no significant enhancement due to the low proportion of blood vessels in the tumour. The final diagnosis of hemolymphangioma depends on postoperative pathological examination. It was further identified by immunohistochemistry. Both vascular endothelial cells and lymphatic endothelial cells were CD31- and CD34- positive, while D2-40 was only expressed in lymphangiomas and some malignant vascular tumours. Combined with the above indicators, the diagnosis of hemolymphangioma is not difficult. The patient was pathologically diagnosed as vasogenic tumour, and immunohistochemical tests showed positive reactions of CD31(blood vessel +) and D2-40(lymphatic vessel +). Further confirmed the final diagnosis of hemolymphangioma. Abdominal hemolymphangioma is a benign lesion, but the rapid growth of the tumor may compress the intestinal duct and surrounding parenchymal organs, causing acute abdominal symptoms such as abdominal pain, abdominal distension, intestinal obstruction, etc. The tumour itself may also rupture and bleed, leading to infection. Therefore, once the preoperative diagnosis of hemolymphangioma is suspected, the patient should be treated in time. The standard treatment for hemolymphangioma is surgical resection. Other non-surgical treatments, including cryotherapy, laser therapy, radiotherapy and local injection of sclerotherapy, do not show significant advantages compared with surgical treatment [13]. According to relevant literature reports, the recurrence rate of tumours after complete resection is 10-27%, while the recurrence rate of tumours after partial resection is 50-100% [5]. Therefore, for hemolymphangioma that cannot be completely resected, postoperative adjuvant treatment is particularly critical. Hemolymphangioma is a mixture of blood vessels and lymphatic vessels, so some anti-angiogenic drugs can be tried, such as bevacizumab, verlimus tablets, apatinib, etc. Since there is no clear literature on the effectiveness of such drugs, .regular follow-up is necessary for patients after surgery

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