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Case Report

Giant retroperitoneal liposarcoma

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Abstract...

Retroperitoneal liposarcomas are rare malignant neoplasms. They usually grow asymptomatically until they are large enough to compress the surrounding organ. Giant retroperitoneal liposarcoma with a diameter of more than 50 cm and a mass of more than 12 kg is extremely rare.

Clinical case: Male K., 57 years old, upon admission complained of heaviness in breathing, pain in the lumbar region, weight loss (6 kg in 3 months). The patient was examined for pruritus in the clinic at the place of residence. Ultrasound of the abdominal cavity revealed a large mass in the abdominal cavity. Further, MSCT of the abdominal cavity revealed a lesion emanating from the retroperitoneal space with compression of the abdominal cavity organs and small pelvis. Additional examination at A.V. Vishnevsky National Medical Research Center of Surgery (Moscow) identified giant retroperitoneal liposarcoma cT2bN0M0. Complete resection is the predominant treatment for retroperitoneal liposarcoma in order to avoid recurrence, therefore, after preoperative preparation of the patient, surgery was performed to remove the retroperitoneal tumor. However, the size and location of the tumor made it necessary, after mobilization of the mass, to divide it into lobes and remove it in parts using coagulation and ligation of tubular structures. Morphological diagnosis: highly differentiated liposarcoma of sclerosing and lipoma-like variants of the structure (M 8851/3; Grade 1).

Conclusion: Giant retroperitoneal liposarcoma is an extremely rare tumor with a high recurrence rate depending on a number of factors such as histological type and grade, metastasis, and completeness of tumor resection. In this case, we performed a total resection without combined resection of the surrounding organs (minimum injuries were decapsulation of the kidney upper pole and removal of a small tumor from the small intestine mesentery). In addition, we will continue to closely monitor our patient for relapse.

Keywords: Giant retroperitoneal liposarcoma; Ultrasound; MSCT; Morphological features; Surgical treatment; Prognosis.

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Introduction

For the first time in the literature, a retroperitoneal tumor found at autopsy was described by Benivieni in 1507 [1]. The term "retroperitoneal sarcoma" was proposed in 1829 by Lobstein, who believed that these tumors originate from the vertebrae [2]. The history of the treatment of patients with non-organ retroperitoneal lesions began in 1824, when Lizars first successfully removed a retroperitoneal lipoma [3].

Inorganic retroperitoneal lesions account for 0.03-1.0% of all oncological diseases [40-7]. Liposarcoma is one of the most common soft tissue tumors in adults. According to the different researchers [8,9], its frequency varies from 9.8 to 16% of the total number of malignant neoplasms of soft tissues. Along with this, liposarcoma in children is considered one of the rarest tumors [10]. Under the general name "liposarcoma", several types of tumors are combined, which are histologically, biologically, cytogenetically and molecularly different from each other [11-13] (Table 1). Neoplasms of this group can affect any organ, but most often they are localized in the upper or lower extremities (56%), trunk (19%), retroperitoneal space (15%) and in the head and neck (9%) [14,15]. Among these tumors, there are both non-metastasizing highly differentiated variants (highly differentiated liposarcoma) and highly malignant often metastatic ones (round cell and pleomorphic liposarcomas).

Table 1: Features of different liposarcomas types.

Liposarcoma type	Age, years	Localization	Cytogenetic abnormalities	Clinical features
Highly differentiated liposarcoma	50-70	Limbs (75%), etroperitoneum	Giant and ring-shaped chromosomes	High frequency of local recurrences, absence of metastasis, dedifferentiation in 5-15% of cases
Dedifferentiated liposarcoma	50-70	Retroperitoneal space (75%)	Giant and ring-shaped chromosomes and other anomalies t (12; 16)	High rate of local recurrence, metastasis
Myxoid/roundcell liposarcoma	24-45	Limbs (75%)		Local recurrence and metastasis (due to the round-cell component)

The source of inorganic retroperitoneal fatty tumors (lipomas and liposarcomas) is most often perirenal adipose tissue [16]. A feature of these lesions are numerous fatty processes extending from the main focus to the side and even lying apart, at a distance from it, which causes a high frequency of recurrence [17,18].

Usually, retroperitoneal fatty tumors reach a significant size, since they grow unhindered in the loose tissue of the retroperitoneal space for a long time, without causing any complaints or disorders in the patient's internal organs [6,19]. Retroperitoneal liposarcomas usually occur in adults aged 40–60 years with a slight male predominance [20].

We present our own clinical observation of a giant retroperitoneal liposarcoma in a patient who did not present any complaints until the appearance of skin itching.

Clinical case

Male K., 57 years old, upon admission complained of heaviness in breathing, pain in the lumbar region, weight loss (6 kg in 3 months).

Anamnesis: The patient was examined for pruritus in the clinic at the place of residence. Abdominal cavity ultrasound (03/28/22): the study is difficult due to a large neoplasm. Multislice Computed Tomography (MSCT) of the abdominal organs (native) (04.04.22): MSCT-signs of the lesion of the retroperitoneal space on the left (retroperitoneal lipoma?) with compression of the abdominal cavity and small pelvis. A tumor biopsy was performed at the local hospital. Histological conclusion: material with pronounced necrotic changes. The revealed changes do not allow for a differential diagnosis between atypical lipomatoid tumor and liposarcoma. The patient was hospitalized for additional examination and surgical treatment at A.V. Vishnevsky National Medical Research Center of Surgery (hospitalization 06/08/22).

Initial examination: Height: 171, weight: 77 kg, BMI: 26. Body temperature: 36.4°C. The general condition is satisfactory. Consciousness is clear, the patient is contact, adequate, oriented in place, time and self. The skin is moderately pale, clean. In the lungs, vesicular breathing, no wheezing. Heart sounds are muffled, rhythmic. Heart rate 72 beats/min. AD 110/70 mm Hg. The tongue is moist and clean. Physiological functions are normal.

Surgical status: A dense lesion is palpated over the entire surface of the abdomen, with percussion a dull sound is determined.

The following studies were performed as an additional examination.

Ultrasound: Free fluid was not detected In the abdominal cavity, retroperitoneal space, as well as in the small pelvis.

In the retroperitoneal space, from the level of the left dome of the diaphragm, filling the entire abdominal cavity and spreading into the small pelvis, significantly squeezing the bladder, a giant lesion is determined, somewhat larger located in the left sections of the abdominal cavity and retroperitoneal space (approximate dimensions 53.0 x 22, 0 x 37.0 cm), The lesion has a solid structure of isoechoic density (corresponds to adipose tissue) with numerous hyperechoic fibrous bands in the structure, and also has minor pleomorphic areas in the structure. Arterial (low resistance) and venous collaterals are located along the course of these fibrous cords, as well as in some places along the contour of the tumor (Figure 1a).

The liver, pancreas and spleen are clearly pushed up and back by the lesion described above. Various sections of the intestine are pushed into the right sections of the abdomen. The right kidney is displaced downward and backward, the left kidney is rotated and located anterior to the aorta. The aorta and the inferior vena cava can be traced along the posterior contour

of the lesion, pushed to the spine, their lumen is stained during color mapping (Figure 1b). A biphasic, somewhat turbulent blood flow is recorded along the inferior vena cava (with LBF up to 0.17 m/s), and the magistral blood flow is along the aorta (LBF up to 0.38 m/s). The lumbar arteries, which are involved in its blood supply, depart from the aorta and enter the lesion structure (Figure 1b). The right iliac vessels are pushed a little backward by the lesion, the left iliac vessels are flattened on the lesion and squeezed by it. The lumen of the iliac vessels is completely stained in color doppler imaging (Figure 1c).

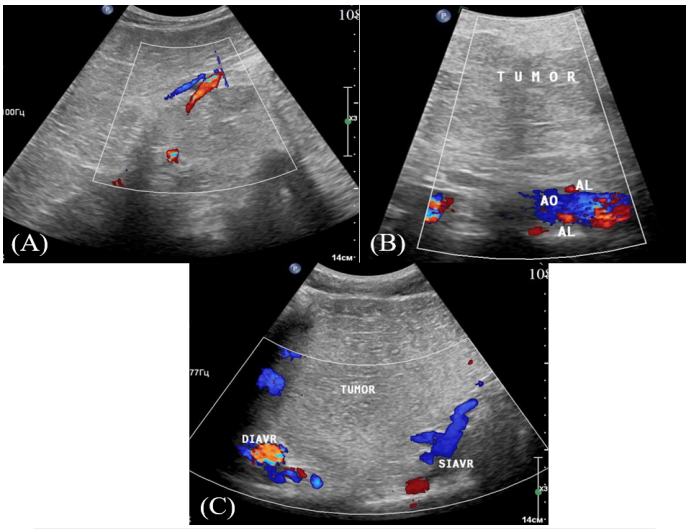

Figure 1: Ultrasound images of retroperitoneal liposarcoma in the color doppler mapping: **(a)** - the structure of the tumor is shown: **(b)** - the mass (TUMOR) compresses the aorta (AO), the lumbar arteries (AL), which are involved in the blood supply of the tumor, are identified; **(c)** – relationship of the tumor (TUMOR) with the branches of the iliac vessels.

The liver is not enlarged in size: the anterior-posterior size of the right lobe is 135.0 mm, the left lobe is 59.6 mm, the contours are clear, even, the parenchyma of increased echogenicity, there is a slight attenuation of the echo signal to the periphery. The vascular pattern of the liver is evenly expressed, however, the speed indicators are evenly reduced. The dimensions of the gallbladder are 70.7 x 20.0 mm, homogeneous contents are determined in the lumen, the structure of increased echogenicity is determined near the wall, not displaced by a change in body position, up to 2.0 mm in size, the walls are not thickened. Intrahepatic and extrahepatic bile ducts are not dilated.

Pancreas: Pushed back by the lesion, not visualized.

The area of the spleen is 42 cm^2, it has clear, even contours, the structure of the parenchyma is homogeneous. The vascular pattern of the spleen is evenly expressed, however, the speed indicators are evenly reduced.

No enlarged regional lymph nodes were found.

The right kidney is visualized with dimensions of 109.0 x 44.3 mm, with clear, even contours, cortico-medullary differentiation is clearly seen, the parenchymal layer is quite pronounced - 13.7 mm, calyx and pelvis are not expanded. Concrements were not found. A thin-walled cyst up to 10 mm in size is determined. The left kidney is visualized with dimensions of 105.0 x 56.6 mm, with clear, even contours, cortico-medullary differentiation is clearly seen, the parenchymal layer is quite pronounced - 17.4 mm, calyx and pelvis are not expanded. Concrements were not found.

Conclusion: Inorganic retroperitoneal lesion, ultrasound picture corresponds to liposarcoma. Pronounced extravasal compression of the aorta, inferior vena cava, renal and iliac vessels. Polyp of the gallbladder. Diffuse changes in the liver parenchyma.

MSCT: On the scans included in the study area, focal and infiltrative changes in the basal parts of the lungs are not detected. No fluid was found in the pleural cavities. Atherosclerosis of the coronary arteries.

Gas, free/encapsulated fluid in the abdominal cavity was not detected.

In the retroperitoneal space, from the level of the left dome of the diaphragm to the upper wall of the bladder, there is a giant lesion with approximate dimensions of 51 x 22 x 38 cm, fat density, with numerous partitions in the structure, accumulating a contrast agent for the delayed phase of the study, and liquid layers (Figure 2). The lesion occupies almost the entire abdominal cavity; due to this arrangement, the duodenum and small intestine are pushed into the right lateral region, the liver is posterior, and the left kidney is rotated. In the thickness of the lesion are the sigmoid, ascending colon, splenic flexure of the colon with the vessels feeding them. There were no signs of invasion of parenchymal organs or vessels.

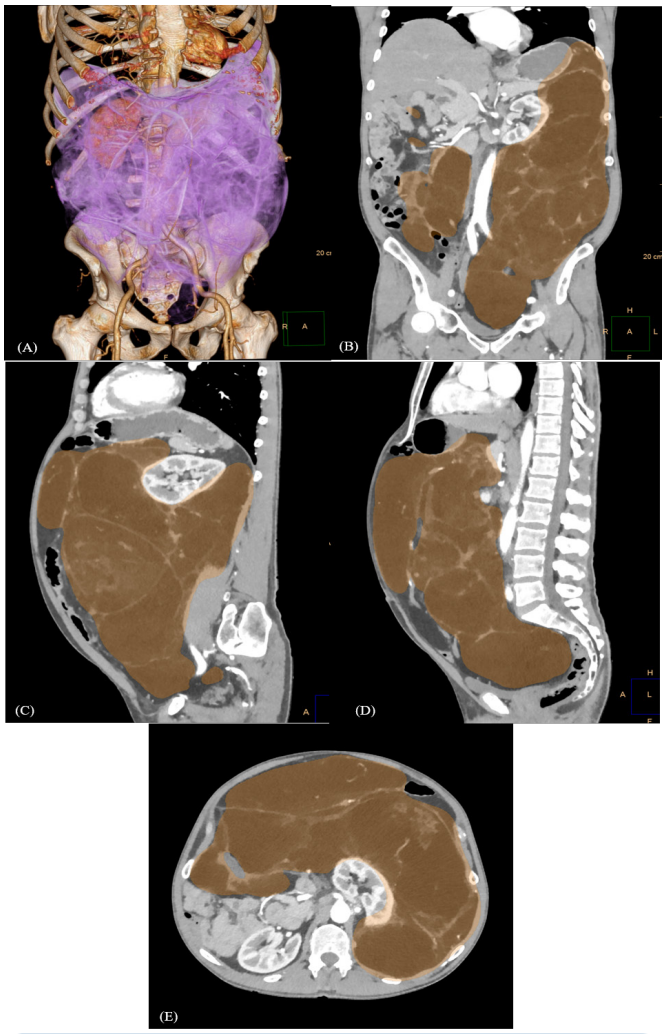

Figure 2: MSCT images of retroperitoneal liposarcoma: **(a)** – three-dimensional reconstruction; **(b)** – coronal projection; **(c,d)** – sagittal projections at different levels; **(e)** - axial projection.

The liver is not enlarged, proportional, the contours of the liver are clear, even, the craniocaudal size of the right liver lobe is 10 cm. The density in the native phase is up to 50 HU. Small cyst up to 4 mm in size in SV of the liver. Intrahepatic bile ducts are not dilated. The gallbladder is up to 30 mm in transverse size, the walls are not thickened. The contents are homogeneous, radiopaque stones in the lumen of the gallbladder and common choledochus were not detected. Choledoch up to 8 mm, not expanded.

Portal vein - 12 mm. Splenic vein - 6 mm. Superior mesenteric vein - 10 mm.

The stomach is not straightened. Part of the cardial part of the stomach prolapses into the posterior mediastinum. Duodenal, small, large intestine without visible pathological changes.

The pancreas has a lobed structure, with clear contours, the volume of the parenchyma is preserved. No lesions were found in the pancreatic parenchyma. MPD is filiform.

The spleen is not enlarged, SI=165, the structure and nature of contrasting are normal. No lesions were found in the parenchyma.

The adrenal glands are not changed. The kidneys are of normal shape, the contours are clear, uneven, the volume of the parenchyma is preserved. The left kidney is rotated. Simple cyst of the right kidney up to 10 mm in size. A solid lesion is adjacent to the lower pole of the left kidney, increased in size compared to the previous study (up to 19x24 mm), accumulating contrast (average density in the native phase 10 HU, arterial phase - 22 HU, venous phase - 43 HU, delayed phase - 88 H.U.). Calyx and pelvis, ureters are not dilated. There are no radiopaque calculi in the calyx and pelvis, ureters.

Hyperplasia of the prostate, calcifications in the prostate structure.

The diameter of the infrarenal abdominal aorta is up to 18 mm. The anatomy of the celiac-mesenteric basin is typical.

Lymph nodes at the scan level are not enlarged.

When analyzing images in the "bone window", no data for the presence of bone-destructive changes were obtained.

Conclusion. MSCT picture of a giant retroperitoneal mass (liposarcoma) without significant dynamics compared to the study dated April 29, 2022. Small cyst SV of the liver. Simple cyst of the right kidney (Bosniak 1). Hyperplasia of the prostate. Atherosclerosis of the coronary arteries.

Complete blood count, biochemical blood test and coagulogram without deviations.

Duplex scanning of the veins of the lower extremities: No pathological changes were detected.

Duplex scanning of the carotid and vertebral arteries in the extracranial region.

Right: A. carotis communis (ACC) walls are thickened, the size of the intima-media complex is 1.1 mm. In the middle third of ACC along the anterior wall, the average echogenicity of the atherosclerotic plaque (ASP) is determined, the stenosis is 25%. In the bifurcation with the transition to the mouth of the a. carotis externa (ACE) along the posterolateral wall, the average echogenicity ASP is determined, the stenosis is 25-30%. At the mouth of the a. carotis interna (ACI) along the anterior wall, calcified ASP is determined, stenosis 20%.

Left: ACC walls are thickened, the size of the intima-media complex is 1.0 mm. In the bifurcation with the transition to the mouth of ACI a semi-concentric medium echogenicity with calcium ASP is determined, stenosis of 25-30%. At the mouth of ACI along the posterior wall is determined by the average echogenicity of the local ASP, stenosis 25%. ACE is completely passable.

Conclusion. Atherosclerosis of the carotid arteries. Tandem stenosis of the left ACI 25-30%. Stenosis of the left ACC 25%, ACI 20%, ACE 25-30%.

Esophagogastroduodenoscopy: Hiatal hernia. Reflux esophagitis. Chronic gastritis.

Echocardiography: No pathological changes detected, ejection fraction 68%.

According to the preoperative examination, the ***diagnosis*** was made: Liposarcoma of the retroperitoneal space cT2bN0M0. Hyperplasia of the prostate. Hypertension stage 3, degree III, risk of cardiovascular complications 4.

The identified giant retroperitoneal lesion extends into the small pelvis with severe compression of the bladder. In this connection, in the preoperative period, there is a risk of developing a violation of the patency of the ureters due to compression by the tumor from the outside. In the early postoperative period, there is also a risk of compression of the ureters due to compression by edematous tissues, as well as the lesion of local cicatricial processes involving the ureters and the development of narrowing or stricture. Against the background of the development of stagnation of urine, an infection penetrates the kidneys, which can cause urosepsis and significantly worsen the prognosis of surgical treatment. To exclude such difficulties and complications, it was decided to perform cystoscopy at the preoperative stage with stenting of the ureters, which will prevent the development of their compression, will allow maintaining normal urine outflow and kidney function, and thereby improve the prognosis of surgical intervention.

The following treatment tactics were determined: Stage I - cystoscopy and installation of ureteral catheters on the right and left; Stage II - (on the same day) surgical intervention in the amount of removal of the lesion of the retroperitoneal space.

After preoperative preparation of the patient, surgery was performed.

Surgery: There is no effusion in the abdominal cavity during revision. The peritoneum is shiny and smooth. There are no distant metastases in the visualized parts of the abdominal cavity and in the liver. Lesion almost completely occupies the abdominal cavity. The tumor is visually represented by adipose tissue, which has a lobed structure, soft-elastic consistency with local foci of dense-elastic consistency (Figure 3). The size of the lesion is more than 50 cm in diameter. The descending colon passes along the anterior surface of the lesion, tightly fused with it.

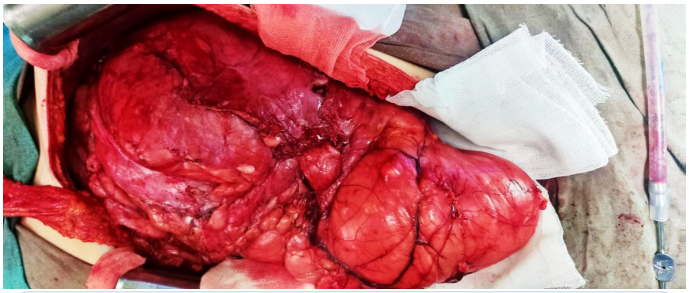

Figure 3: General view of the tumor in the surgical field.

The tumor was mobilized with its division into lobes and removal in parts using coagulation and ligation of tubular structures. When the mass was mobilized, the ureters with catheters inside were visualized. When the tumor was mobilized in the left hypochondrium, the upper pole of the kidney was decapsulated. Bleeding from the kidney was stopped by flashing the bleeding area of the kidney with Vicryl 2/0 with tamponade of adipose tissue (no more than 1 cm long). General hemostasis was performed by bipolar coagulation and the application of a hemostatic sponge.

Conducted a second revision of the abdominal cavity. On the mesentery of the small intestine, a whitish lesion of a densely elastic consistency with a diameter of up to 0.8 cm is determined. The lesion is resected and sent for histological examination. Hemostasis. Drainage of the abdominal cavity through the right counter-opening with 2 silicone drains.

Histology. The tumor was presented in the form of 11 nodes ranging in size from 5 x 5 x 2 cm to 30 x 25 x 17 cm, with a total weight of 12 kg. On microscopic examination, all tumor nodes have a similar structure and are represented by highly differentiated lipoma-like and sclerosing liposarcoma (Fig. 4) (M 8851/3; Grade 1).

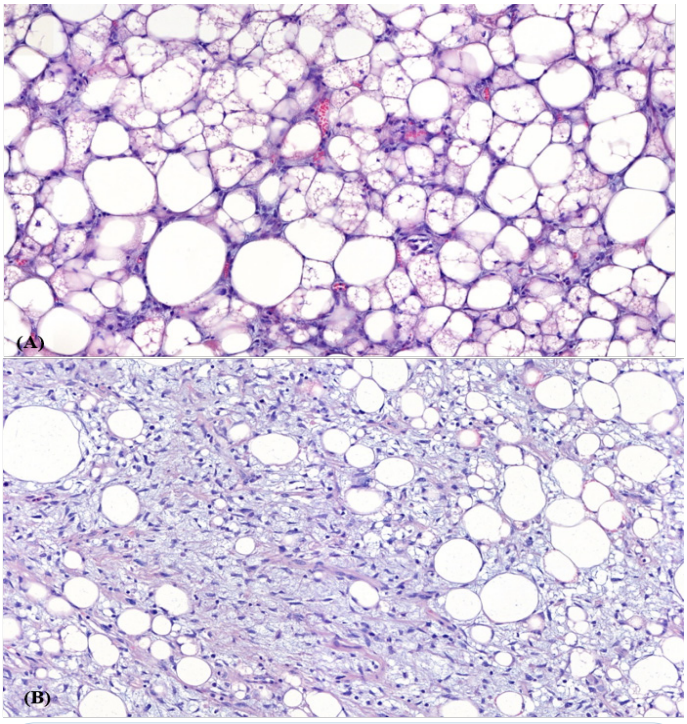

Figure 4: Micrographs, hematoxylin and eosin, magnification x200: **(a)** - highly differentiated liposarcoma of a lipoma-like variant of the structure; **(b)** - highly differentiated liposarcoma of the sclerosing variant of the structure.

Conclusion. Morphological picture of highly differentiated liposarcoma of sclerosing and lipoma-like variants of the structure (M 8851/3; Grade 1).

For the purpose of observation in the early postoperative period, the patient was transferred to the intensive care unit. On the third day, the patient was transferred to the ward of the specialized department and activated. Prevention of infectious, thrombotic complications, analgesic, antibacterial, physio- and anti-inflammatory therapy, dressings with antiseptic solutions were carried out.

Dynamic ultrasound monitoring of the abdominal cavity and retroperitoneal space in the postoperative period showed the presence of a small amount of free fluid, which did not require intervention, but dynamically decreased. Also, after removal of the tumor, restoration of blood flow in the aorta and inferior vena cava was noted, and, as a result, a significant increase in intraorganic blood flow in the liver and spleen. Indicators of intraorganic blood flow in the liver and spleen returned to normal on the 8th day after surgery.

Wound without signs of inflammation, heals by primary intention. The ureteral catheters were removed on the 2nd day. The drainage from the left lateral canal was removed on the 6th day. Drainage from the small pelvis was removed on the 13th day.

On the 9th day after surgery, the patient noted an increase in the left testicle and the appearance of discomfort during urination. An ***ultrasound*** was performed. The left testicle is with dimensions of 48.4 x 32.8 x 28.3 mm, its membranes are clearly visible, the structure is homogeneous. The presence of a moderate amount of clear fluid surrounding the testis outward along the extraperitoneal field is visualized (Fig. 5). The epididymis head is with dimensions of 14.2 x 10.3 mm; three small cysts are determined in its structure, 5.1 mm, 2.6 and 2.0 mm in size. The appendage can be traced in all departments (body, tail), the strengthening of the vascular pattern of the appendage attracts attention. *Conclusion.* The revealed changes in the left testicle (ultrasound signs of edema) are most likely due to the surgery in the area of the inguinal canal. Cysts of the epididymis of the left testicle.

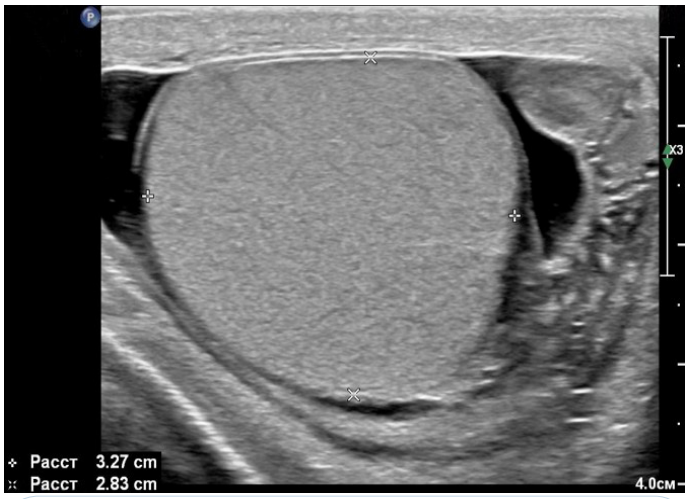

Figure 5: Ultrasound image of a moderate amount of simple fluid surrounding the testis outward along the extraperitoneal field.

He was consulted by a urologist, recommendations were given for medical correction and a follow-up ultrasound in 10 days.

Before discharge on the 14th day after the operation, a ***control MSCT study*** was performed. Postoperative changes in the tissues of the anterior abdominal wall at the site of surgical access. No fluid was found in the pleural cavities. In the area of resection with distribution along the lower edge of the liver, a moderate amount of fluid. Pathological changes in the liver, biliary tract, pancreas and spleen were not detected. Duodenal, small, large intestine without visible pathological changes (Figure 6).

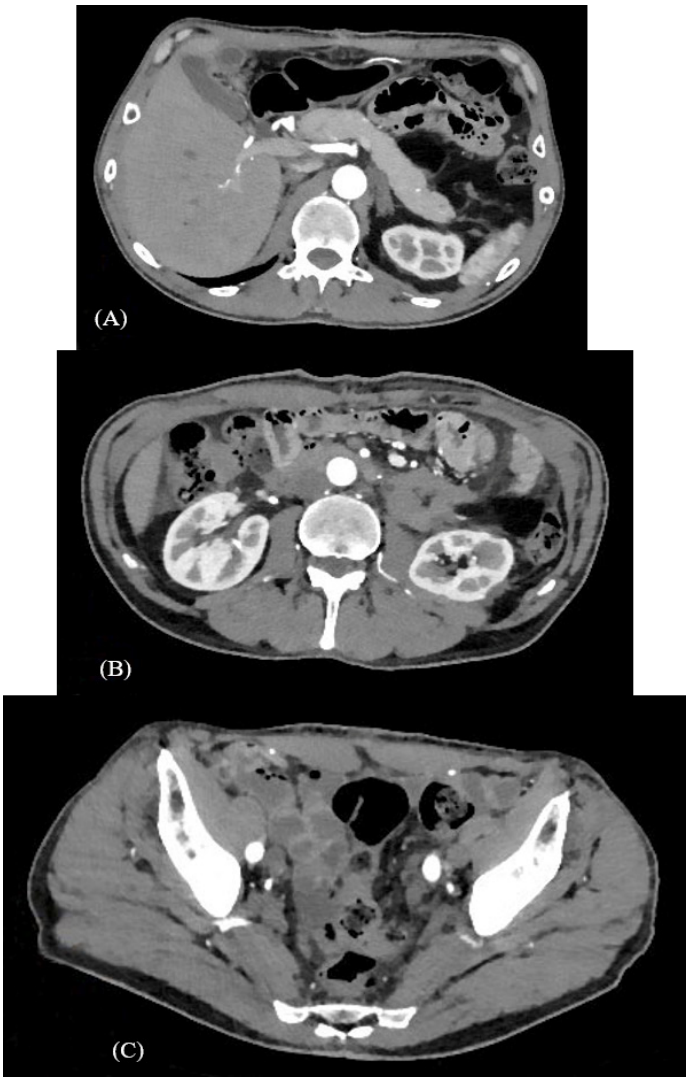

Figure 6: MSCT images, arterial phase, **(a-c)** - in sequential scans of the abdominal cavity and small pelvis,

Conclusion: Condition after removal of the retroperitoneal tumor on June 14, 2022. Postoperative changes in the anterior abdominal wall in the surgical access area. Moderate amount of fluid in the abdomen.

The patient was discharged on the 15th day after surgery in a satisfactory condition under the supervision of a surgeon and a urologist of the place of residence clinic.

The patient was discharged on the 15th day after surgery in a satisfactory condition under the supervision of a surgeon and a urologist of the polyclinic at the place of residence.

The main treatment for liposarcomas is their surgical removal. The ideal operation is a radical resection of the tumor, and it is desirable to remove the tumor in the capsule. Quite often (almost 50% of cases), radiation therapy is used in conjunction with surgery, both preoperatively and during surgery. Most often, radiation therapy is used in the postoperative period - up to 90% [14]. The effectiveness of radiation therapy depends on the location of the tumor. For example, in case of liposarcomas of the extremities, radiation therapy significantly increases the survival rate of patients, while in case of intra-abdominal and retroperitoneal liposarcomas, the use of radiation therapy is limited or even impossible due to the negative effect of radiation on adjacent structures [21]. An effective, but still predominantly experimental method is intraoperative tumor irradiation, especially in cases of local relapses. Chemotherapy using various schemes is rarely used for liposarcomas, both in combination with other methods of treatment, and in isolation [22, 23].

Based on the characteristics of the development and course of retroperitoneal liposarcomas, they often reach large and even gigantic sizes [24-28]. The large size of the tumor should not be considered as a contraindication to surgical removal. Complete surgical removal can possibly be achieved even in giant tumors with careful examination and perioperative management. Patients with giant liposarcomas usually suffer from malnutrition, hypoproteinemia, or anemia as a result of the long existence of the tumor, so preoperative preparation is necessary to improve the patient's condition (compensation of identified cardiovascular changes if necessary, support with enteral or parenteral nutrition). Interdisciplinary collaboration is especially important in the treatment of giant retroperitoneal liposarcomas. In addition, carefully designed surgical tactics and monitoring of vital signs during surgery play an important role.

In the presented clinical case, conditionally radical resection of the tumor was performed. We use the term "conditionally" because, due to the gigantic size of the tumor, it was not possible to remove it as a single array in the capsule, the tumor was divided into lobes and removed in parts.

Factors that worsen the prognosis of the course of liposarcoma are the age of patients over 50 years old, the tumor diameter is more than 10 cm, the presence of tumor growth at the resection margins, local recurrences, histological type (well-differentiated liposarcomas have a better prognosis than dedifferentiated ones) and localization of the primary tumor (extremity tumors have better prognosis compared with retroperitoneal and located in the abdominal cavity, superficially located is better than deep-lying). An important factor is the total histological gradation of the tumor (the higher it is, the worse the prognosis) [22, 23]. Literature data suggest that successful complete resection of retroperitoneal liposarcoma can increase 5-year survival from 16.7 to 58% [29, 30]. Highly differentiated tumors have a more favorable prognosis, which has a 5-year survival rate of 83% to 90% [31].

Based on the presented data, taking into account the fact that Grade 1 highly differentiated liposarcoma was verified, we can hope for a relatively favorable prognosis for the course of the disease in our patient. However, factors such as age (57 years), tumor diameter (more than 50 cm in diameter) and its retroperitoneal localization can worsen the prognosis. Of course, regular dynamic monitoring of the patient's condition is necessary. Studies conducted at our Center on dynamic monitoring of 54 patients after surgical removal of retroperitoneal tumors revealed tumor recurrence in 27.8% of patients, in whom, taking into account repeated and third relapses, 22 tumors were detected. Clinical manifestations had no specific symptoms. Recurrent tumors were detected only during ultrasound monitoring in 22.7% of cases [6].

Conclusion

Giant retroperitoneal liposarcoma is an extremely rare tumor with a high recurrence rate depending on a number of factors such as histological type and grade, metastasis, and completeness of tumor resection. In the presented clinical case, we performed total resection without combined resection of surrounding organs (minimum injuries were decapsulation of the upper pole of the kidney and removal of a small lesion from the mesentery of the small intestine). In addition, we will continue to closely monitor our patient for relapse.

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