## ONCOLOGY

# CASTLEMAN DISEASE. A RARE CLINICAL CASE OF RETROPERITONEAL TUMOR LOCALIZATION IN AN ELDERLY PATIENT

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## ABSTRACT

*Castleman disease is an extremely rare benign disease of the lymphatic system with an estimated incidence of 1–9 cases per 1,000,000. Its etiology remains unknown; interleukin 6 (IL-6) plays an important role in pathogenesis. Castleman disease has two clinical forms: localized (up to 90 % of cases) with a favorable prognosis, treated predominantly by surgical method; generalized (up to 10 % of cases) with less favorable prognosis, treated by pharmacological therapy. The diagnosis is rarely established at the preoperative stage.*

***The aim.*** *To present a clinical case of diagnosis and treatment of Castleman tumor of a rare topical localization.*

***Results.*** *A 66-year-old patient was admitted at the Abdominal Oncology Department of the G.E. Ostroverkhov Kursk Oncology Scientific and Clinical Center. Diagnosis: Retroperitoneal mass on the right found at the preventive examination; no peripheral lymphadenopathy was detected on ultrasound and computed tomography (CT). After the examination, a preliminary diagnosis was made: Gastrointestinal stromal tumor (GIST) of the small intestine mesentery.*

*Based on the results of the case conference, the decision was taken to perform a surgery – laparoscopic removal of the tumor under endotracheal anesthesia. For surgical approach, a fan-shaped arrangement of ports was chosen. The surgery had no complications. Intraoperative blood loss was 50.0 ml. The total operating time was 98 minutes.*

*According to the results of the histological study, the following diagnosis was made: Castleman disease, unicentric form, hyaline-vascular variant.*

*En bloc surgery is the standard method for the treatment of localized forms of the Castleman disease. In all cases, long-term follow-up shows a long relapse-free period in almost all patients.*

*During follow-up examinations (ultrasound of the abdominal cavity and retroperitoneal space, CT of the abdominal cavity with contrast enhancement, CT of the chest), no disease recurrence was detected during the year of observation.*

*Castleman disease is a rare non-clonal lymphoproliferative disease of unknown etiology. A rare case of its retroperitoneal localization indicates that in cases with an uncertain nature of the peritoneal mass, Castleman disease should be included in the differential diagnostic search.*

***Key words:*** *Castleman disease, retroperitoneal lymphadenopathy, immunodeficiency, hyaline-vascular variant, interleukin 6, computed tomography, laparoscopy*

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# БОЛЕЗНЬ КАСТЛЕМАНА. РЕДКИЙ КЛИНИЧЕСКИЙ СЛУЧАЙ ЗАБРЮШИННОЙ ЛОКАЛИЗАЦИИ ОПУХОЛИ У ПАЦИЕНТКИ ПОЖИЛОГО ВОЗРАСТА

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## РЕЗЮМЕ

*Болезнь Кастлемана относится к крайне редким доброкачественным заболеваниям лимфатической системы с приблизительной заболеваемостью 1–9 случаев на 1 000 000. Этиология заболевания остаётся неизвестной; в патогенезе важную роль играет интерлейкин 6 (IL-6). Заболевание имеет две клинические формы: локализованная (до 90 % случаев) – имеет благоприятный прогноз, лечение преимущественно хирургическое; генерализованная (до 10 % случаев) – прогноз менее благоприятный, применяются медикаментозные методы лечения. Диагноз редко устанавливается на дооперационном этапе.*

***Цель**. Представить клинический случай диагностики и лечения опухоли Кастлемана редкой топической локализации.*

***Результаты.** В отделение абдоминальной онкологии ОБУЗ «Курский онкологический научно-клинический центр им. Г.Е. Островерхова» поступила пациентка 66 лет с диагнозом: Объёмное образование забрюшинного пространства справа, выявленное в ходе профилактического осмотра; по данным ультразвукового исследования (УЗИ) и компьютерной томографии (КТ) перефирической лимфаденопатии выявлено не было. После обследования пациентке был выставлен предварительный диагноз: Гастроинтестинальная стромальная опухоль (GIST) брыжейки тонкой кишки.*

*По результатам консилиума было принято решение выполнить оперативное вмешательство – лапароскопическое удаление опухоли под эндотрахеальным наркозом. Для оперативного доступа была выбрана веерообразная расстановка портов. Оперативное вмешательство прошло без осложнений. Интраоперационная кровопотеря составила 50,0 мл. Общее время операции составило 98 минут.*

*По результатам гистологического заключения был выставлен диагноз: Болезнь Кастлемана, уницентричная форма, гиалиново-сосудистый вариант. Хирургическое вмешательство «en bloc» является стандартом лечения локализованных форм болезни, во всех случаях долгосрочного наблюдения показывает длительный безрецидивный период практически у всех пациентов. При контрольных обследованиях (УЗИ брюшной полости и забрюшинного пространства, КТ брюшной полости с контрастным усилением, КТ грудной клетки) в течение года наблюдения рецидива заболевания не выявлено.*

*Таким образом, болезнь Кастлемана представляет собой редкое неклональное лимфопролиферативное заболевание неизвестной этиологии. Редкий случай забрюшинной локализации указывает на то, что в случаях с неопределённым характером объёмного поражения брюшиной полости в дифференциально-диагностический поиск должная входить болезнь Кастлемана.*

***Ключевые слова:** болезнь Кастлемана, забрюшинная лимфаденопатия, иммунодефицит, гиалиново-сосудистый вариант, интерлейкин 6, компьютерная томография, лапароскопия*

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## INTRODUCTION

Castleman disease (angiofollicular nodular hyperplasia) is a rare benign lymphoproliferative disease. This pathology was first described in 1954 by Benjamin Castleman as a localized enlargement of mediastinal lymph nodes with a characteristic increase in the number of follicles, involution of their central part, and a significant vascular network with endothelial hyperplasia [1].

According to the literature, the incidence is 1–9 cases per 1,000,000 population. There are no precise incidence data; the average incidence is 1 case per 150,000 population. The incidence among male and female population is approximately the same, but precise data are not available. The age group of 40 years or less is more common among patients diagnosed with Castleman disease [2]. Precise data on the incidence of the disease in the Russian Federation are not available.

Three main morphological variants of the tumor were described: hyaline-vascular, plasma cell and mixed [3]. Currently, a 4$^{th}$ variant, HHV-8-associated, is distinguished. Hyaline-vascular variant is characterized by disorder of lymph node architectonics due to hyperplasia of lymphatic follicles, reduction of their size, atrophy and hyalinization of the central zones, wide mantle part, interfollicular hypervascularization; externally, this variant resembles an «onion skinning». Mitoses, as a rule, are absent. A vessel runs to the center of the hyalinized follicle, increased vascularization is noted in the interfollicular zones (the most frequent type of vessel is a postcapillary venule with hyperplastic endotheliocytes), single or grouped lymphocytes, plasmacytes, plasmacytic monocytes are found between the vessels [4]. In the plasma cell variant, the structure of lymph nodes is disturbed due to hyperplastic follicles with a narrow mantle zone of lymphocytes. In the interfollicular zones and cerebral layer there are wide areas of mature and immature plasma cells, sinuses are dilated [5].

Thus, the occurrence of hyaline-vascular variant is 80–90 %, of plasma cell variant – 8–10 % [6]. The literature also identifies a mixed variant (about 5 % of cases). Recently, the HHV-8-associated variant has been identified as new data on the influence of human herpesvirus type 8 on the development of Castleman disease have been obtained [7].

The clinical classification is based on the number of affected lymph nodes or groups of lymph nodes. Thus, two forms of the disease are distinguished: localized (unicentric) (one region or lymph node is affected) and generalized (multicentric) (several regions are affected) [8]. The unicentric form is the most common one (80–90 %). Thoracic lymph nodes are most commonly affected (about 70 %), particularly in the mediastinum. Abdominal and retroperitoneal lymph nodes are the next most frequently affected (approximately 10 %). The rarest localizations are the tonsils, orbit, lymphatic tissue of the nasopharynx and tongue (no more than 8 %). The generalized form has a more significant clinical picture and a less favorable prognosis. In the multicentric form, most cases manifest in patients 50–60 years of age, and some cases have secondary renal and pulmonary involvement, paraneoplastic vesicular disease, and peripheral neuropathy. Laboratory findings include anemia, thrombocytosis, hypergammaglobulinemia, and elevated C-reactive protein [9–12].

There are no established risk factors for the localized variant, and the disease is idiopathic. At the same time, the main risk factor for multicentric variant of the lesion is an immunodeficiency state of varying severity, and in some cases, the presence of human herpes virus type 8 (HHV-8) [13].

Therefore, the presence of immunodeficiency caused by HIV infection would be a predisposing factor for the occurrence of Castleman disease. In the vast majority of cases, HIV-positive patients will be HHV-8-positive, whereas the rate of HHV-8 detection among HIV-negative patients ranges from 2 % to 50 %. Although HHV-8 status is directly related to viral load in the population, patients in this group have an increased risk of transformation into HHV-8-positive plasmablastic lymphoma and the occurrence of Kaposi's sarcoma [14–16].

Although the etiopathogenesis of Castleman disease is still unclear, indolent chronic inflammation, immunodeficiency and/or autoimmune conditions are considered as likely factors. In 1993, S. Akira et al. first pointed out the role of interleukin 6 (IL-6). The effect of IL-6 overproduction and dysregulation on stimulation of acute phase protein production by hepatocytes, stimulation of B-cells, macrophages, fibroblasts, direct and indirect stimulation of neoangiogenesis and endothelial cell proliferation has been established. Therefore, anti-IL6-immunotherapy is used as one of the treatment options [17].

In the vast majority of cases, the localized form of the disease is treated surgically. This report presents a clinical case of a rare retroperitoneal paraduodenal localization of Castleman disease, its diagnosis and treatment.

## CASE HISTORY

A 66-year-old female patient was admitted at the Abdominal Oncology Department of the G.E. Ostroverkhov Kursk Oncology Scientific and Clinical Center. Diagnosis: Retroperitoneal mass on the right. She had no complaints. She was referred to the department by the medical and preventive institution at her place of residence due to a retroperitoneal mass lesion detected during a preventive examination, which coincides with reports of a prolonged asymptomatic course of the process [18, 19]. General condition on admission was satisfactory, no bad habits, body mass index – 31.24 kg/m$^2$. She reported that a preventive ultrasound of the abdominal cavity revealed a right-sided mass lesion up to 6.0 cm. In 2012, she underwent a surgical intervention – "open" appendectomy, without complications. No plasma or hemotransfusions were performed. Laboratory tests are within reference values. The tests for Wassermann reaction (WR), hepatitis B surface antigen (HBS Ag), antibodies to hepatitis C virus (HCV), HIV were negative. Associated disease: hypertension.

The patient was examined at our center. Chronic atrophic gastritis was detected during fibrogastrodu-

odenoscopy. Fibrocolonoscopy revealed no organic pathology. No thrombosis or reflux on ultrasound examination of the lower limb veins. Mammography: fibrotic breast changes (BIRADS-2). No gynecological pathology was detected during the examination. No coronavirus COVID-19 RNA was detected.

Ultrasound of peripheral, abdominal, retroperitoneal lymph nodes showed no lymphadenopathy. Changes on pelvic ultrasound are within the age-related normal range.

Abdominal ultrasound: in the area of the head of the pancreas there is a round-shaped hyperechogenic mass lesion with a clear even contour, 5.2 × 3.7 × 4.4 cm in size, well vascularized. Computed tomography (CT) of the chest organs revealed fibrotic changes. Subsequent computed tomography of the abdominal cavity organs with intravenous contrast (Ultravist 100 ml): in the root of the mesentery on the right side, in front of the lower horizontal part of the duodenum, a space-occupying mass with a rather clear contour (5.4 × 3.7 × 4.6 cm), soft tissue density, contrast-accumulating, with a vascular network along the contour, without clear organ affiliation was detected (Fig. 1). The findings may be consistent with gastrointestinal stromal tumor (GIST) of the small intestine mesentery. The differential diagnosis of abdominal and retroperitoneal localizations of Castleman disease is most often made with GIST or neuroendocrine tumors [18].

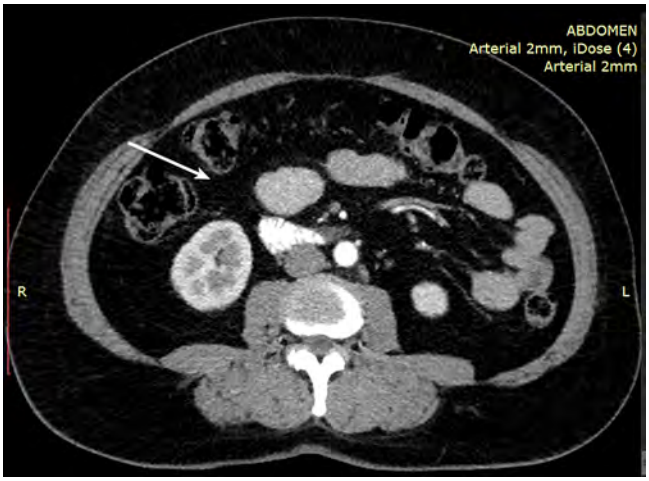


***FIG. 1.***
*CT scan of the abdominal cavity with intravenous contrast, axial projection, arterial phase: round-shaped mass lesion, adjacent to the duodenum*

The CT scan revealed precise topical localization of the tumor, no signs of invasion of surrounding structures, and vascular anatomy – both anatomical landmarks and vessels of the tumor (Fig. 2). There were no signs of dissemination during the study, in particular, there were no focal liver mass lesions, which was confirmed by CT scans in different projections (Fig. 3). However, CT findings for Castleman disease are nonspecific, and due to the rare occurrence of retroperitoneal localization, GIST is most commonly identified. The mesenteric unicentric form of the disease, usually appears on CT as a mass lesion of soft tissue density, without satellite nodes [20, 21].

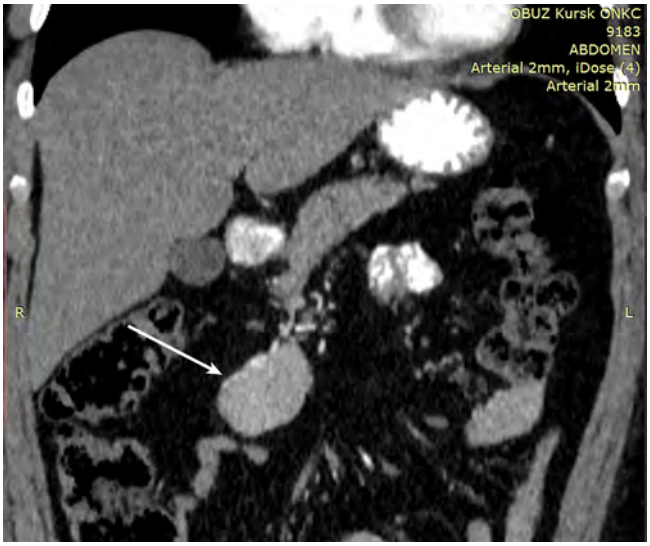


***FIG. 2.***
*CT scan of the abdominal cavity with intravenous contrast, coronary projection, arterial phase: no proved invasion of the duodenum, pancreas, vessels of the small intestine mesentery detected*

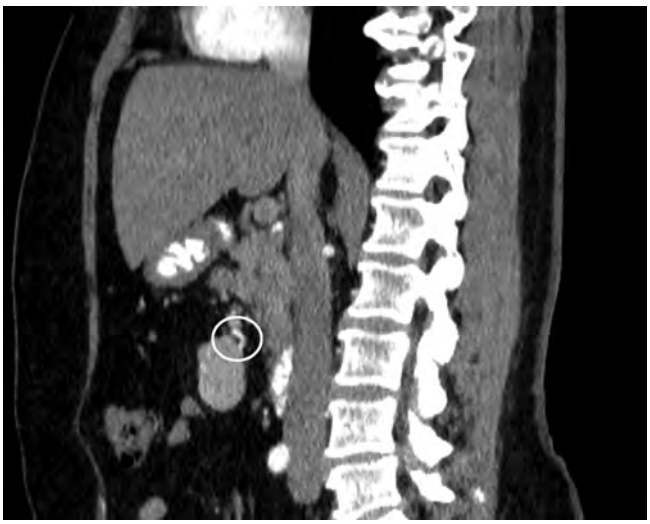

***FIG. 3.***
*CT scan of the abdominal cavity with intravenous contrast, sagittal projection, arterial phase: an arterial vessel along the lateral contour enters the mass lesion in the upper contour*

Pancreaticoduodenal endosonography data were concordant with abdominal CT data. After examination, the patient was given a provisional diagnosis of GIST of the small intestine mesentery.

After a consilium, it was decided to perform surgical intervention – laparoscopic tumor removal under endotracheal anesthesia.

A fan-shaped port arrangement was chosen for surgical approach: 10 mm optical port was located parumbilically below the umbilicus; 10 mm port in the right lateral region along the midclavicular line; 5 mm port – in the right subcostal region; 5 mm port – in the left lateral region along the midclavicular line.

Revision of the abdominal cavity revealed no signs of dissemination, including visual metastatic foci in the liver, pelvic organs without pathology (Fig. 4).

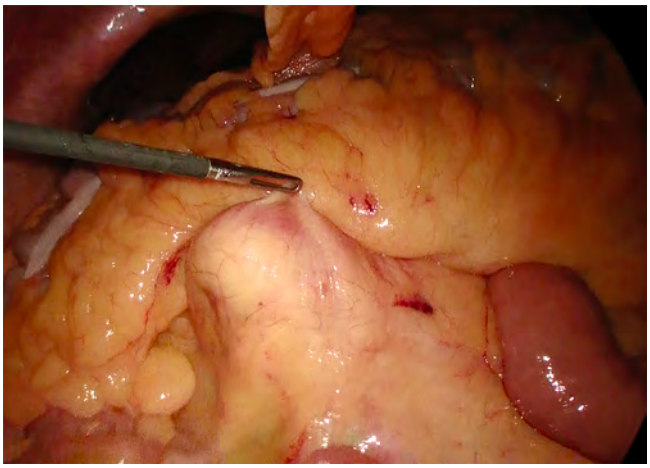

***FIG. 4.***
*Intraoperative photo. Laparoscopic picture: a tumor was found; no carcinomatosis, no ascites; no signs of invasive growth. The lesion is located retroperitoneally, without involving small intestine loops*

The classic fan-shaped port arrangement allows dissection in various planes. After mobilization of the mass lesion, clipping and vessel crossing, tumor removal was performed (Fig. 5, 6). Revision of the abdominal cavity and bed was performed, and hemostasis was controlled (Fig. 7).

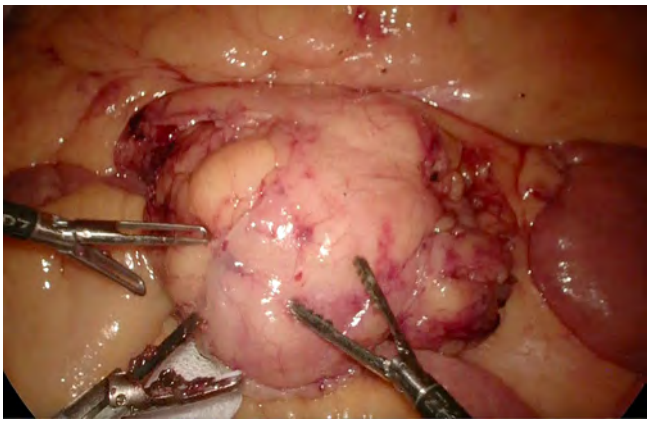

***FIG. 5.***
*Intraoperative photo. The peritoneum was opened along the periphery of the tumor; the tumor is floating; mobilization was performed along the lower contour of the tumor*

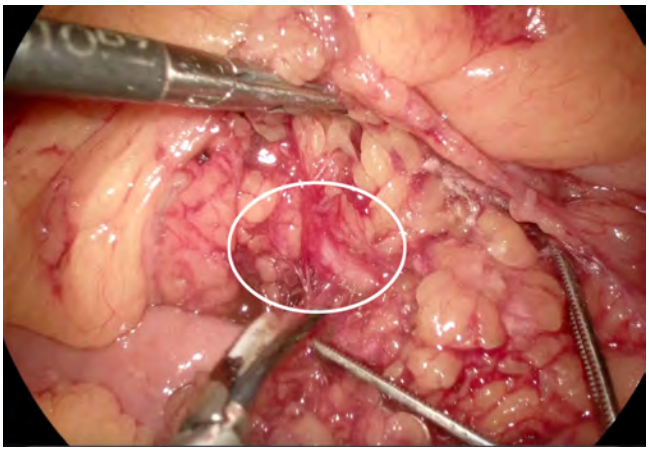

***FIG. 6.***
*Intraoperative photo. A vessel from the superior mesenteric artery system is isolated in the area of upper pole of the tumor, being visualized by CT of the abdominal cavity*

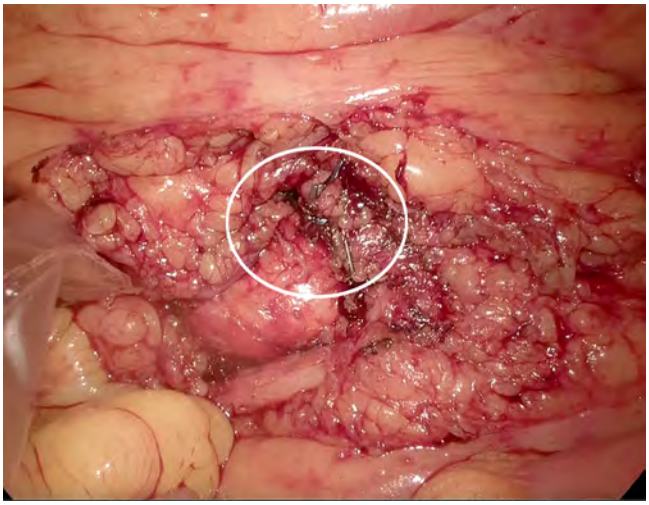

***FIG. 7.***
*Intraoperative photo. Final view: the bed of the removed tumor; clips are put on the vessels (artery, vein)*

## DISCUSSION

The surgery had no complications. Intraoperative blood loss was 50.0 ml. The total operating time was 98 minutes. Due to the absence of bleeding and signs of connection with the pancreas, abdominal drainage was not performed. The container with the gross specimen was extracted through a minilaparotomy incision in the right iliac region with excision of the scar after appendectomy.

Despite the previously established diagnosis (GIST of the small intestine mesentery), our preoperative diagnosis was consistent with M. Ohta et al. who indicated that the preoperative diagnosis never included Castleman disease [22]. The en bloc surgery performed is the standard of treatment for localized forms of the disease. In all cases, long-term follow-up shows a long relapse-free period in almost all patients [23, 24].

Based on the results of the histological study, the following diagnosis was made: Castleman disease, unicentric form, hyaline-vascular variant.

There is increased vascularization of interfollicular zones, mainly due to postcapillary venules, with altered endothelial cells (Fig. 8). Scattered or grouped plasmacytes, small lymphocytes and monocytes are located among the vessels, sometimes eosinophils are included in the interfollicular infiltrate. The histological data obtained were full compliance with those of many investigators [25–29].

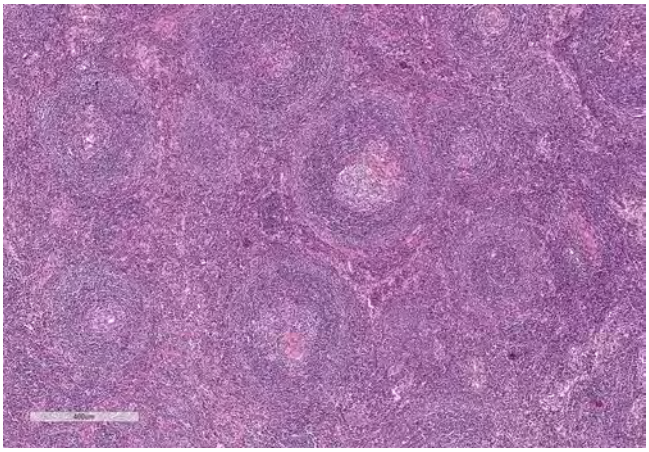

***FIG. 8.***

*Micrograph. Alteration of the histologic structure of the lymph nodes due to an increase in the number of lymphatic follicles and interfollicular zones hypervascularization. The follicles are reduced and have a transformed, atrophic, hyalinized germinal center. Hematoxylin and eosin staining; magnification ×20*

The germinal centers are significantly cell-depleted, consisting of single lymphocytes, follicular dendritic cells, and hyaline deposits (Fig. 9, 10). Immunohistochemical study: cells of interfollicular space consist of $CD68^+$ plasma cells, dendritic cells, endotheliocytes of blood vessels; cells of germinal centre consist of $CD3^+$, $CD21^+$, $CD23^+$ [30].

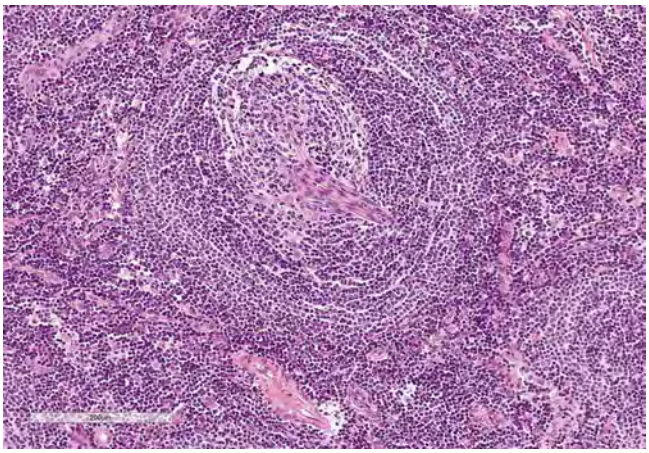

***FIG. 9.***

*Micrograph. Lymphatic follicle. A wide, concentric mantle zone formed by small lymphocytes – "onion skinning". No mitoses detected. Hematoxylin and eosin staining; magnification ×100*

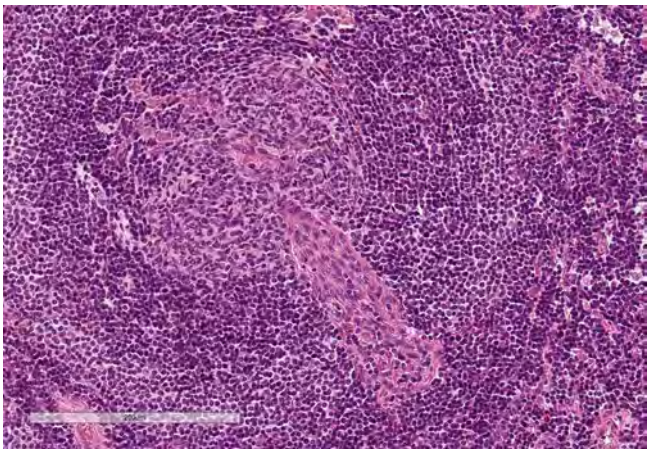

***FIG. 10.***

*Micrograph. Lymphatic follicle. A hyalinized capillary-type vessel passes through the center of the atrophic follicle, the structure resembles a lollipop. Hematoxylin and eosin staining; magnification ×100*

## RESULTS

After the histological report, the patient underwent PCR test to detect HHV-8. The result was negative, which was in compliance with literature data and laboratory and instrumental findings. The patient was discharged from the ward on the 8th day after surgery, referred for consultation to a hematologist: the diagnosis was confirmed, no additional therapy was indicated.

During follow-up examinations (ultrasound of the abdominal cavity and retroperitoneal space, CT of the abdominal cavity with contrast enhancement, CT of the chest), no disease recurrence was detected during the year of observation.

## CONCLUSION

Therefore, Castleman disease is a rare lymphoproliferative disease of unknown etiology pathogenetically associated with IL-6 hyperexpression. It has characteristic histological variants. The diagnosis is often established after morphological examination of the removed surgical specimen. It has a favorable prognosis, especially in the localized form.

The presented case is unusual due to the location of the tumor in the paraduodenal, retroperitoneal, and parapancreatic areas. Laparoscopic removal of the tumor was performed due to the obvious advantages of this surgical approach [31]. Despite the short follow-up time (18 months), there were no signs of disease recurrence, but further dynamic follow-up is necessary due to the rare localization in this case [32].

In cases with indeterminate abdominal and retroperitoneal mass lesions, laparoscopic approach is considered as the last stage of diagnosis and the first stage of treatment.

There is a need to develop CT criteria to suspect Castleman disease in order to include it in the differential di-

agnostic series for abdominal and retroperitoneal mass lesions. Further study of the etiopathogenesis of the disease will make it possible to develop an optimal therapeutic and diagnostic algorithm.

**Conflict of interest**

The authors of this article declare the absence of a conflict of interest.

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