87899_Auto_Edited.docx

WORD COUNT 2335
TIME SUBMITTED 06-DEC-2023 09:27AM
PAPER ID 105074038

Name of Journal: *World Journal of Clinical Cases*
Manuscript NO: 87899
Manuscript Type: CASE REPORT

Intraperitoneal hyaline vascular Castleman disease: Three case reports

Intraperitoneal hyaline vascular Castleman disease

Jiawei Gao, Zheyi Shi, Zhaobi Zhu, Xiangrong Xu, Wei Chen

Abstract

BACKGROUND

Castleman disease (CD) was first reported in 1954. It is a rare non-malignant lymphoproliferative disease and its etiology is unclear. As the clinical manifestations of CD are different, there are difficulties in diagnosis and treatment Therefore, for patients with CD, it is important to establish the diagnosis in order to choose the appropriate treatment.

CASE SUMMARY

In this report, three patients with intraperitoneal CD treated in our center from January 2018 to June 2023 were reviewed, and the clinical and paraclinical examinations, diagnosis and treatment were analyzed, and all three patients were diagnosed with CD by routine histopathological and immunohistochemical examinations.

CONCLUSION

CD is a complex and rare disease. Because there are no special clinical symptoms and laboratory abnormalities, the diagnosis often depends on routine pathological and immunohistochemical findings.

Key Words: Castleman disease; Intraperitoneal mass; Lymph node

Gao J, Shi Z, Zhu Z, Xu X, Chen W. Intraperitoneal hyaline vascular Castleman disease: A report of three cases. *World J Clin Cases* 2023; In press

Core Tip: Castleman disease (CD) is a rare and complex disease. As the clinical signs and laboratory results are not specific, clinical diagnosis is difficult, which often depends on imaging and pathology examinations. This report summarizes the diagnosis and treatment of three cases of CD and reviews the related literature to explore the diagnosis and treatment process of CD in order to improve the clinical management of this disease.

INTRODUCTION

Castleman disease (CD) is a group of rare disorders with characteristic histopathological features. Monocentric CD (UCD) is a benign localized lymphoproliferative disease, which was first reported in 1954 and described as mediastinal localized lymphoproliferative disease [1]. The clinical subtypes of CD include monocentric type and multicentric type, and histological subtypes include transparent vessels, plasma cells and mixed type[2-3]. Overall, monocentric CD has a good prognosis, requiring only local surgical resection and no additional treatment. Patients usually survive without recurrence following treatment. However, at present, there is still no standard treatment for multicentric CD due to its potential invasiveness and it has poor prognosis, and some lesions may develop into malignant tumors[4]. Comprehensive treatment includes monoclonal antibody immunotherapy, combination of radiotherapy and chemotherapy, and due to its rich blood supply, anti-angiogenic drugs also play an important role in treatment.

CASE PRESENTATION

Chief complaints

Case 1: A 37-year-old male was admitted to hospital due to "a mass in the pancreas found one week ago".
Case 2: A 38-year-old female was admitted to hospital due to " an abdominal mass found for one month".
Case 3: A 71-year-old female, was admitted to hospital due to "a gallbladder mass found 2 mo ago".

History of present illness

Case 1、Case 2 and Case 3: During the course of the disease, the patient had no paroxysmal palpitations, headache or other positive symptoms.

History of past illness

Case 1 and Case 2: The patient had no previous history of hypertension or other chronic diseases, and had no obvious abnormal tumor markers after admission.
Case 3: Blood pressure control was satisfactory, and the patient had a history of hypertension for > 3 years.

Personal and family history

Case 1、Case 2 and Case 3: There is no personal and family history.

Physical examination

Case 1、Case 2 and Case 3: Cardiopulmonary and abdominal examination showed no abnormalities and no positive signs.

Laboratory examinations

Case 1: Endocrine tests showed the following: adrenocorticotropin-cortisol rhythm was normal, renin-angiotensin II-aldosterone recumbent position test and 24-h urinary aldosterone were normal. Blood 3-methoxynorepinephrine, blood 3-methoxyepinephrine, 24-h urine catecholamine and vanillymandelic acid were normal,

and there was no evidence of cor pulmonale, primary aldosteronism or pheochromocytoma. The human immunodeficiency virus (HIV) test was also negative.

Case 2: There were no obvious abnormalities in hemoglobin 77 g/L, routine biochemical and urine testing, blood coagulation function, tumor markers and HIV testing.

Case 3: Laboratory examinations showed that there were no obvious abnormalities in routine biochemical, blood and urine tests, and tumor markers were normal. Endocrine tests showed the following: The adrenocorticotropin-cortisol rhythm was normal, renin-angiotensin II-aldosterone decubitus test showed no abnormalities, and HIV testing was negative.

Imaging examinations

+ADw-html+AD4APA-p+AD4-Case 1: A contrast-enhanced computed tomography (CT) showed a soft tissue lesion in front of the pancreas with a quasi-circular density, and a diameter of approximately 22 mm, smooth edges, and obvious uniform and continuous enhancement. Similar nodules were seen at the edge of the mass, and there were also several small abdominal lymph nodes with an average diameter of 5 mm. Giant lymph node hyperplasia / ectopic pancreas was considered. In order to further confirm the diagnosis, a painless ultrasound gastroscope biopsy was performed. A linear 7.5 MHz ultrasound gastroscope was used to explore the gastric cavity and duodenum. During the procedure, enlarged lymph nodes above the head of the pancreas were seen, showing a triangular shape, hilar structure could be seen inside, and no obvious blood flow signal was seen in the periphery of the lymph nodes. One cross-section was approximately 20 x 15 mm, the lymphatic edge was sharp, showing uniform hypoechoic changes, and 3 Lymph nodes were punctured (Figure 1).+ADw-/p+AD4-

+ADw-p+AD4-Case 2: Before admission, a head and chest plain CT scan revealed a low density focus in the left lobe of the liver, and she was admitted to the hospital for further examination. Enhanced CT suggested multiple masses in the abdominal and

pelvic cavities, the largest mass was found in the pelvic cavity which had a clear boundary and irregular shape, and a small nodular calcification was seen around the focus. Moderate enhancement was seen, and the enhancement pattern of other abnormal soft tissue nodules was similar, and the possibility of stromal tumor was considered (Figure 2).+ADw-/p+AD4-

+ADw-p+AD4-Case 3: The patient underwent B-ultrasound examination more than 2 mo previously, which indicated gallbladder enlargement and thickening of the gallbladder wall. Enhanced magnetic resonance imaging of the upper abdomen was performed, which indicated an abnormal signal focus in the lower part of the pancreas that was circular, approximately 25 x 24 mm in size, the edge was smooth and the boundary was clear, and the possibility of giant lymph node hyperplasia was considered. Cholecystitis and thickening of the gallbladder wall did not exclude the possibility of malignancy. The patient was then admitted to hospital to complete the preoperative examination. Enhanced CT indicated a round soft-tissue density focus below the pancreas, with obvious enhancement in the arterial phase and slightly decreased enhancement in the venous phase and delayed phase, with a length of approximately 2 5 mm. The possibility of giant lymph node hyperplasia was considered and as the gallbladder wall was not uniformly thickened, the possibility of local malignancy was also considered (Figure 3).+ADw-/p+AD4APA-/html+AD4-

FINAL DIAGNOSIS

Case 1: Routine postoperative pathology showed that a large number of small round cells were stretched and lymphoproliferative lesions were considered. Immunohistochemical diagnosis showed hyaline vascular CD due to the presence of AE1/AE3 (-), CD10 (-), CD20 (+), CD3 (+), CD45R0 (+), CD79 α (+), LCA (+), Ki-67 (+, 15%), CD5 (+), and Bc1-2 (+) (Figure 4).

Case 2: Following excision of the mass, rapid pathology suggested lymphoproliferative lesions. Postoperative pathological diagnosis of hyaline vascular CD was made due to the presence of CD20 (+), CD3 (+), CD21 (follicular dendritic +), CD23 (follicular

dendritic +), CD5 (+), CyclinD1(-) (-), Bcl-2 (germinal center -), Bcl-6 (germinal center +), Ki67 (+, about 40%), CD10 (-), MuM1(-), Pax-5 (+), CD30 (-), ALK (-), and CD4 (+) (Figure 5).

Case 3: Following excision of the mass, rapid pathology indicated lymphoproliferative lesions. Postoperative pathology showed clear vascular CD. Immunohistochemical results were as follows: CK20 (+), Pax5 (+), CD3 (+), CD5 (+), CD21 (germinal center +), CD15 (-), CD30 (-), EMA (-), CD10 (germinal center +), Bcl-6 (germinal center +), MuM1 (-), EBER-ISH (-) and Bcl-2 (-) (Figure 6).

TREATMENT

Case 1: Conservative treatment was provided following communication with the patient.

Case 2: Exploratory laparotomy was performed after preoperative preparation, and a mass in the greater omentum on the greater curvature of the stomach was found, which was approximately 5 x 4 cm in size.

Case 3: Radical resection of gallbladder cancer and excision of the abdominal lesion were performed. During the operation, a mass of approximately 3 cm in size was found in the lower part of the pancreas, which was closely adhered to the lower margin of the pancreas. The anatomical mass was completely separated using an ultrasonic knife.

OUTCOME AND FOLLOW-UP

Case 1: The patient's condition was stable and there was no obvious disease progression during the 2-year follow-up.

Case 2: The patient's condition was stable and there was no obvious disease progression during the 2-year follow-up.

Case 3: After the operation, anti-inflammatory, stomach protection and other symptomatic support was provided, and the patient recovered well and was subsequently discharged. At present, CT reexamination 2 mo after surgery showed no signs of recurrence.

DISCUSSION

CD is a rare lymphoproliferative disease characterized by the proliferation and enlargement of lymphoid tissue[5]. The most common site of CD is the mediastinum, accounting for approximately 70% of all CD patients, while the occurrence of CD in the abdomen is uncommon, and only a few cases have been reported[6]. The exact cause of the disease is unknown, but some studies have suggested that it is related to the development of HIV and human herpes virus (HHV)-8.

CD can be divided into monocentric and multicentric subtypes, and the different types of CD are characterized by significant lymphatic architecture changes in all lymph node septa. Monocentric type includes clear vessel type and plasma cell variant type. Multicentric CD is mainly characterized by plasma cell variants, with a few cases showing mixed type. In this report, all three patients underwent routine pathological and immunohistochemical examinations, and all showed clear vascular type CD. Clear vascular CD presents as an affected lymphoid follicle with an enlarged outer layer of concentric rings of small lymphocytes that surround a small germinal center of atrophy or degenerative transformation. Germinal centers usually have penetrating transparent small blood vessels and protruding follicular dendritic cells that may be dilated, destroyed, or have multiple tight connections[7].

In contrast, the germinal center of CD with plasma cell variants is proliferative rather than degenerative, and the interfollicular region of the lymph nodes is vascularized and contains plasma cell sheets with polyclonal characteristics. However, these histopathological findings are not specific to CD and diseases that may cause similar changes in proliferative reactive lymph nodes, such as rheumatoid arthritis and viral lymphadenitis need to be ruled out[8].

The diagnosis of CD mainly depends on histological examination by excision or puncture of the swollen lymph nodes, and includes radiological examination, the most common of which is whole-body CT or CT-18F Fluorodeoxyglucose positron emission tomography examination. Currently, monocentric CD is considered a soft tissue mass

with clear and regular edges, uniform density, speckle calcification or accompanied by bleeding and necrosis on CT[9]. On an enhanced scan, the mass shows enhancement in the arterial phase, with continued enhancement in the venous phase and delayed phase. On CT, multicentric CD usually presents as multiple round and low-density masses of similar size, most of which have uniform enhancement, and partial irregular enhancement may occur if the lesion is large[10]. However, the imaging features of CD are difficult to distinguish from other diseases such as neuroendocrine tumors or lymphomas, such as neurogenic tumors, lymph node metastases, and gastrointestinal stromal tumors. In many cases, the preoperative imaging diagnosis is inconsistent with the postoperative histopathological diagnosis. In the current report, it was found that there were indeed difficulties in preoperative diagnosis. A patient was considered to have a stromal tumor after completing preoperative examination. The diagnosis could only be made after the mass was removed intraoperatively and rapid pathology was obtained, and corresponding surgical methods were selected to avoid unreasonable extensive resection. Surgical removal of monocentric CD is currently the gold standard of treatment, and we report two surgical patients with no signs of recurrence. In other reports, long-term follow-up of patients also showed that monocentric CD was cured after surgical resection, and there was no recurrence during the follow-up period of up to 20 years[11].Recurrence after complete resection was rare, and paraneoplastic complications, such as AA amyloidosis, were usually gradually relieved after complete resection. In unresectable monocentric CD, if no adjacent structures are threatened by compression, the patient can be followed up regularly without intervention, as lesion growth may be very slow. If symptoms develop, rituximab with or without steroids may be considered to reduce the size of the lesion. For patients with a reduced mass after treatment, surgical resection is recommended if complete resection is feasible. For patients who cannot undergo complete surgical resection of the lesion after medication, radiotherapy or arterial embolization can be considered[12]. Multicentric CD is rare and has a poor prognosis. It is usually treated with glucocorticoids combined with chemoradiotherapy. At present, there are no clear guidelines for the treatment of CD.

Many drugs have been tried, such as monoclonal immunotherapy against IL-6[13], antiviral drugs used in relation to HIV infection[14], and related chemotherapeutic drugs (such as doxorubicin, vincristine, cyclophosphamide, melphalan and chloramphenicol)[15,16], and even some new targeted drugs have achieved satisfying results in some clinical applications due to their anti-angiogenic effects[17]. However, there is limited clinical practice experience, comprehensive treatment should be given to suitable patients, and it is necessary to closely observe patients after medication.

CONCLUSION

CD is a complex and rare disease, and a standard treatment is still lacking. For clinicians, it is difficult to make a clear diagnosis before surgery due to the lack of specific radiological markers. Therefore, when the diagnosis is unclear, histopathological examination is important to guide treatment. Complete resection is the gold standard for the treatment of monocentric CD, while multicentric CD requires comprehensive treatment depending on the patient's condition, but the overall prognosis is poor.

87899_Auto_Edited.docx

ORIGINALITY REPORT

6%
SIMILARITY INDEX

PRIMARY SOURCES

1	www.ncbi.nlm.nih.gov Internet	56 words — 2%
2	Qi Tan, Qi Li, Yangfei Ma, Zehui Gu, Suxian Chen. "Primary adenosquamous carcinoma of the sigmoid colon: A case report and retrospective analysis", Research Square Platform LLC, 2023 Crossref Posted Content	24 words — 1%
3	Antonino Carbone, Margaret Borok, Blossom Damania, Annunziata Gloghini et al. "Castleman disease", Nature Reviews Disease Primers, 2021 Crossref	14 words — 1%
4	Kristina Norvilaite, Monika Kezeviciute, Diana Ramasauskaite, Audrone Arlauskiene, Daiva Bartkeviciene, Valentinas Uvarovas. "Postpartum pubic symphysis diastasis-conservative and surgical treatment methods, incidence of complications: Two case reports and a review of the literature", World Journal of Clinical Cases, 2020 Crossref	14 words — 1%
5	Lu Zhang, Yu-jun Dong, Hong-ling Peng, Hao Li et al. "A national, multicenter, retrospective study of Castleman disease in China implementing CDCN criteria", The Lancet Regional Health - Western Pacific, 2023 Crossref	14 words — 1%

6	www.omicsonline.org Internet	13 words — 1%
7	link.springer.com Internet	12 words — 1%

EXCLUDE QUOTES	ON	EXCLUDE SOURCES	< 12 WORDS
EXCLUDE BIBLIOGRAPHY	ON	EXCLUDE MATCHES	< 12 WORDS