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Case Report

Clinical and radiological features of parapancreatic hyaline-vascular Castleman disease: a case report and literature review

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Abstract: Parapancreatic Castleman disease is a rare angiofollicular lymph node hyperplasia that are likely to imitate other parapancreatic lesions on radiological images, often occurs in Asian countries. First, we retrospectively analyzed the radiological appearance and clinical data of a patient in our hospital, and then we conducted a systematic review of all previous parapancreatic reports. Although rare, parapancreatic hyaline-vascular Castleman disease should be considered when a parapancreatic lesion observed in a patient which is asymptomatic, appears as a well-defined solid or cystic solid mass with slight to intense enhancement patterns in radiological images.

Keywords: Castleman disease, pancreas, computed tomography, magnetic resonance imaging, pathology

Introduction

Castleman disease (CD) is an uncommon lymphoproliferative disorder of unknown origin that usually affects both lymph nodes and non-nodal tissue, initially described as a pathological entity in 1954 and later defined by Castleman in 1956 [1, 2]. Histopathologically, CD can be classified into hyaline-vascular (HV) type, plasma cell (PC) type, and mixed (HV and PC) type. It can also be divided into localized (unicentric) or multicentric disease based on clinical features [3]. It is most commonly seen in the mediastinum but may occur as well in other sites such as neck, axilla, mesentery and retroperitoneum [4, 5]. Pancreatic localization is rare and is often indistinguishable from other parapancreatic diseases both clinically and radiographically. Here, we describe a case of CD in parapancreatic tissue and review the clinical and radiological features concerning this rare disease.

Patients and methods

Institutional review board approval was obtained for this study, and the informed consent requirement was waived due to the retrospective study design. Clinical data of patients diagnosed with parapancreatic CD were collected retrospectively from January 2012 to July 2017 in our institution. The patient's laboratory data were collected and documented prior to treatment.

Literature review

A literature search was conducted in English for case report of parapancreatic CD. We searched PubMed, the Springer, Ovid and Google Scholar from January 1, 1992 to July 1, 2017. The key words were listed as follows: (pancreas or peripancreas or parapancreas or abdomen) and (Castleman disease). The data of available case reports were all included and analyzed. For each case report, first author, publication year,

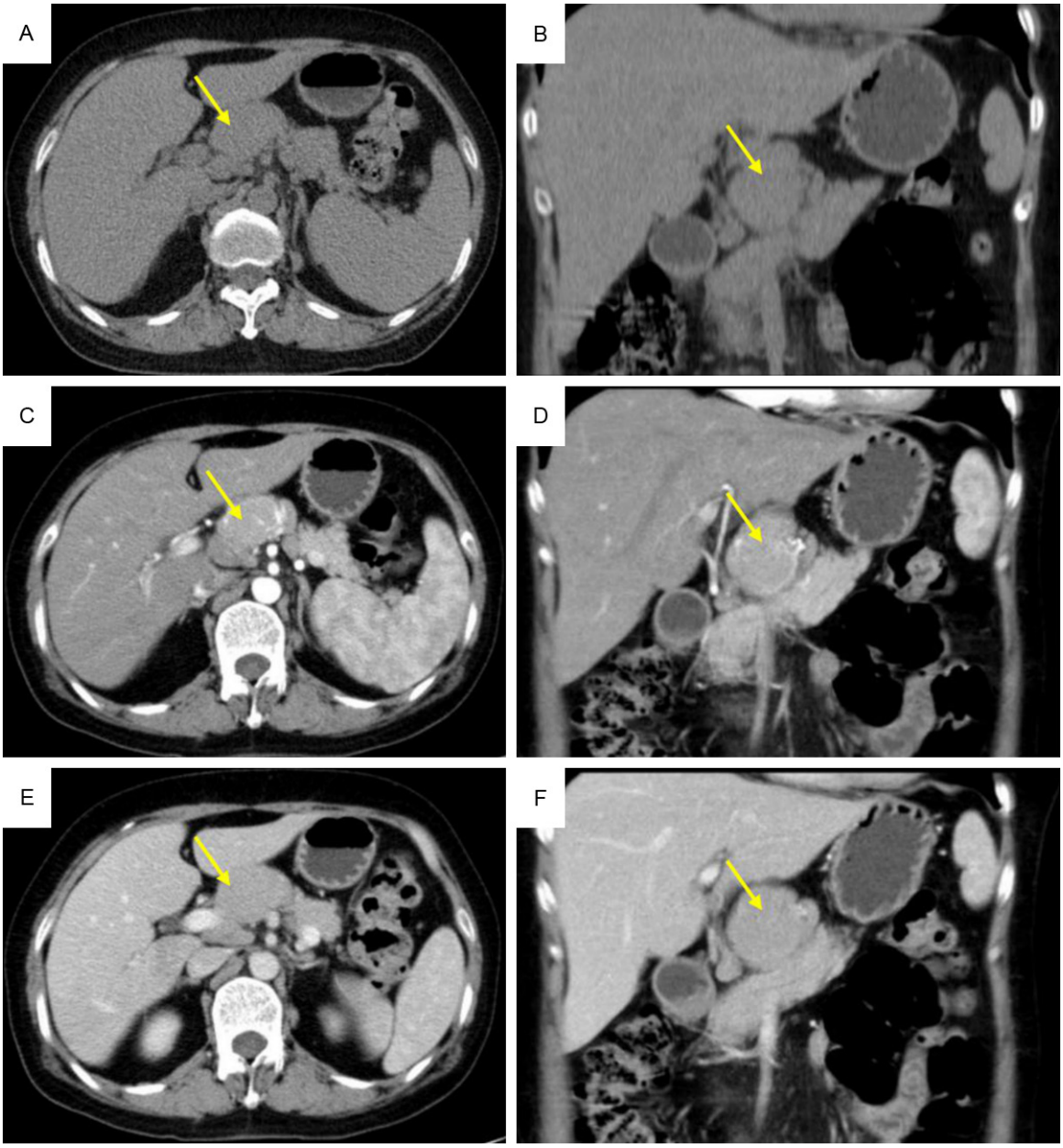

Figure 1. CT scans obtained before (A. Axial), (B. Coronal) and after (C. Axial), (D. Coronal) arterial phase; (E. Axial), (F. Coronal) venous phase injection of contrast medium show a well-circumscribed, round mass (yellow arrows) that appears intensely enhanced and the enhancement of blood vessels in the arterial phase with moderate washout in the venous phase.

patient's age, gender, symptom, site, size, gross morphology, calcification, contrast-enhanced manifestations, pathologic type, treatment, prognosis and regional distribution sites were listed.

Results

Case report

A 57 years old female with dull epigastric pain for two years was admitted to our hospital for further investigation. This patient had a history of cholecystectomy and blood transfusion once due to anemia three years ago. She had no family history of malignancy or inherited disease. Her physical examination showed upper right middle abdominal tenderness. Abdominal unenhanced and contrast-enhanced CT demonstrated a well-circumscribed solid mass at the right side of pancreatic head (yellow arrows) that appears intensely enhanced and the enhancement of blood vessels in the arterial

Table 1. Laboratory tests of our patient

Biochemical test	Result	Reference value
Fibrinogen	6.96	2.00-4.00 g/L
White blood cell count	23.5	0-15.8/ul
Epithelial cell count	35.8	0-17.2/ul
Percentage of mononuclear cells	11.20	3-8%
Width of RDW-CV	17.00	11.6-14%
Alkaline phosphatase	192.40	36-110/L
Glycosylated hemoglobin	16.97	12-16.2%
Globulin	47.10	20-45 g/L

Abbreviations: RDW-CV, Red blood cell distribution width.

phase with moderate washout in the venous phase (**Figure 1A-F**). Laboratory tests are presented in **Table 1**. There was no serological indication of active HIV, B and C hepatitis and tumor markers were negative. The patient underwent exploratory laparotomy. Histological analysis revealed CD of the HV type. Macroscopic examination showed preserved lymphoid tissue with follicles at various degrees of maturation and diffuse hyaline involution like an 'onion' appearance (**Figure 2**). Postoperative course was uneventful and the patient was discharged on 9th day after surgery. She is alive and free of recurrence at three years follow-up.

Literature review

From 1992 to 2017, 38 parapancreatic CD cases with complete information were published in English literature (**Table 2**). In addition to our case, this study documented and analyzed 39 parapancreatic CD cases. The flow chart of cases in the literature was described in **Figure 3**.

The average age of all patients, HV-CD, PC-CD, Mixed-CD of patients was 49.8, 49.5, 40.7 and 58.7 years (range, 27-74 years; 22 men and 16 women). Nine (21.4%) of the patients were asymptomatic. For all patients, the incidences of abdominal pain, epigastric pain, and weight loss were 16.6%, 16.6% and 11.8% respectively. Except for ten patients not mentioning in the literatures, other patients had undergone laboratory tests. 30% of the patients were in the normal level, 10% had erythrocyte sedimentation rate elevated. Most patients had no other history disease. Most of the patients distributed in China (21.1%), Japan (15.9%), USA (13.2%) and Italy (10.6%), the disease also can be seen in other regions such as India and Turkey (**Table 3**).

Histological classification

The patients in the literature were diagnosed with CD by hematoxylin-eosin (HE) staining. A large quantity of patients (65.8%) were diagnosed HV type, others were PC type (18.4%), the other were mixed type besides one undecided case.

CT findings

After the literature review, the total computed tomography (CT) findings of the 38 cases were summarized in **Table 2**. The tumors were located at parapancreatic region, such as head, neck, body and peripancreas. Ten tumors were located in the pancreatic head and twenty were found in the peripancreas. The average diameter of the tumors was 4.7 cm (range: 1.2-12 cm). Half of the lesions involved in this study had maximum diameters greater than 4 cm. Most tumors were well-circumscribed oval masses with homogeneously internal contents. On contrast-enhanced images, the mass mostly presented as moderate or intensive enhancement in the arterial phase and moderate washout in the venous phase. Some parapancreatic CD (28.9%; 11/38) contained punctate or focal calcifications on the CT images.

MRI findings

Among the literature collected 38 cases, eight cases performed MRI examination. Three lesions manifested as homogenous hypointensity on T1 weighted imaging (T1WI) and isointensity or hyperintensity on T2 weighted imaging (T2WI) besides four unknown descriptions. One case presented as hypointensity on T1WI and hyperintensity on T2WI with central punctuate hypointensity.

Treatments and prognosis

All patients in the literature were given different surgical procedures and uneventful postoperative course, except one case was given high dose of corticosteroids due to his illness implicated in multiple tissues.

Discussion

Etiology and pathogenesis

The etiology and the physiopathology of CD remains unclear till now, but several theories

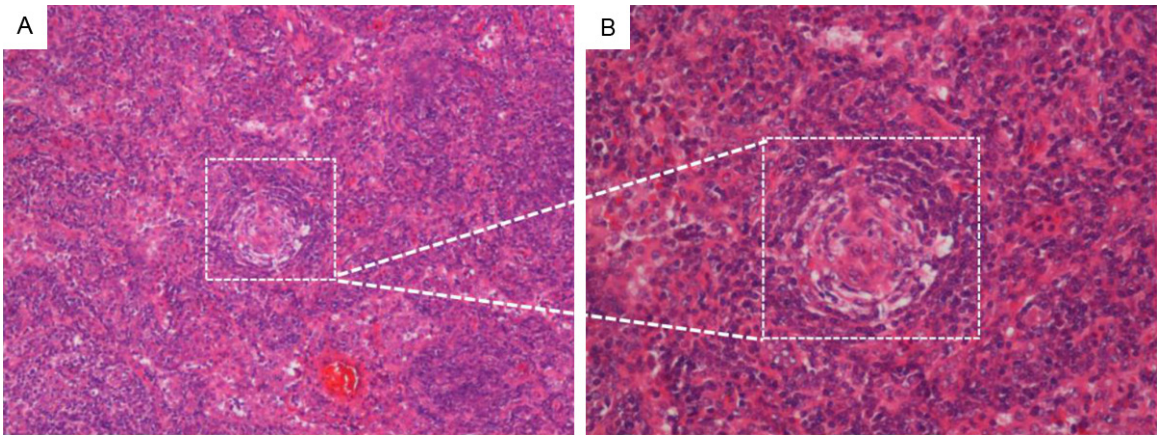

Figure 2. A. Typical histologic features of Castleman disease with hyaline vascular hematoxylin-eosin stained photomicrograph shows typical paracortical expansion with mixed inflammatory cells, including mature lymphocytes and a prominent proliferation of blood vessels (original magnification: ×100); B. High-power photomicrograph (original magnification: ×200) of one area shows a germinal center with the classic "onionskin" appearance.

have been put forward. One theory submits that reactive lymphoid hyperplasia caused by unknown antigenic stimulation associated with virus. All the infection may lead to oversecretion of interleukin 6 (IL-6) [6]. IL-6 had been suggested to be a critical factor in the evolution and symptoms of CD [3, 7-9]. The other one proposes that it is due to the growth disturbance of the lymphoid tissue (i.e., a vascular lymphoid hamartoma) [6]. In addition, autoimmune phenomena such as autoimmune cytopenias, peripheral neuropathy, systemic lupus erythematosus and Kaposi's sarcoma has become the possible pathogenesis of CD [3, 8]. In the current report, HCV or HBV infections have been implicated in the pathogenesis of the disease [9]. Epstein-Barr virus, Toxoplasma, and Mycobacterium tuberculosis also have been linked to this disorder [3]. However, no genetic or toxic factor has been associated with the disease.

Histological and clinical classification

CD is a relatively rare and benign disorder. The disorder has been described by a number of terms like giant lymph node hyperplasia, lymph node hamartoma, follicular lymophoreticuloma, benign giant lymphoma, angiomatous lymphoid hamartoma, and angiofollicular mediastinal lymph node hyperplasia [3]. Histopathologically, CD presents with three distinct histological variants. The most common histological type is the HV type (about 90% of all cases), characterised by small hyaline-vascular follicles and interfollicular capillary proliferation. PC type (less common type) is characterised by large follicles with intervening sheets of plasma cells. The mixed type is characterized by a combination of both hyaline-vascular and plasma cell morphology [7, 10, 11]. Clinically, CD currently was classified into localized (unicentric) and multicentric types. Localized Castleman's disease (LCD), only affects a group of lymph nodes, is defined as a single, benign lesion which usually affects young people [8, 12]. While multicentric Castleman's disease (MCD) which always affects more than one group of lymph nodes [6]. There were 25 cases of HV type, 7 PC type, and only 5 mixed type in the previous reported 38 cases. And there is one case of MCD in the literature listed in our study. Our patient finally was diagnosed LCD (HV type). Although the patient's age was not typical, other manifestations such as clinical and radiological features were consistent with this type.

Clinical features

Different types of CD have distinct clinical features. LCD usually presents as HV type, however MCD often presents as PC type, therefore, the symptoms of them are basically consistent with histological types [3]. HV type mainly affects adults younger than 35 years, while the PC type affects older people with no sex bias [11, 13, 14]. This study included 22 male and 16 female, with a mean age of 49.8 years (range: 27-74 years). In terms of clinical symptoms, most patients present as asymptomatic.

Table 2. Summary of Castleman disease involving parapancreas reported in the English literature

Case No./ sex/age	Year	Symptoms	Gross morphology	Size (cm)	Site	No enhanced (density/calcification)	No enhanced (T1WI/T2WI)	Contrast-enhanced	Pathologic type	Follow up	Study
1/Female/36	1992	RIF pain	Oval/well-defined	4×3×2	Peripancreatic	Homogeneous/none	None	NDI	HV	Unknown	Suzanne et al [13]
2/Female/50	1992	Asymptomatic	Oval/ill-circumscribed	4×3×3	Head	Homogeneous/central fibrosis	None	Moderate enhancement	HV	Unknown	Yutaka et al [26]
3/Female/50	1994	Fever, fatigue, and WL	Quasi-circular/smoothly marginated	3.5×4	Body and tail	Homogeneous/none	None	Peripheral rim enhancement	Mixed	Unknown	Chaulin et al [30]
4/Male/65	1995	Asymptomatic	Oval/poorly demarcated	10×7×6	Head	Homogeneous/none	None	None	HV	2Y	Hideko et al [36]
5/Male/69	1995	AP	Unknown/demarcated	7×4.5×3.5	Peripancreatic	None	None	None	HV	15M	Hideko et al [36]
6/Female/27	2002	EP	Round/well-circumscribed	5	Head	Inhomogeneous/punctate calcification	None	None	PC	3Y	Donata et al [24]
7/Male/36	2003	Asymptomatic	Circular/well marginated	Unknown	Body and tail	Unknown	Hypointense/isointense to normal parenchyma	Peripheral rim enhancement	PC	1Y	Rafaela et al [5]
8/Female/45	2004	EP	Oval/well-capsulated	7×5×5	Peripancreatic	Homogeneous/none	Hypointense/hyperintense	None	PC	1Y	Nazif et al [20]
9/Female/56	2004	Fatigue, WL and AP	Oval/well-circumscribed	2.5×2.2	Body	Homogeneous/none	None	None	HV	3M	Yilmaz et al [12]
10/Male/53	2005	RC	Oval/well-circumscribed	12×7×4	Tail	Slightly heterogeneous/central calcification	None	None	HV	2Y	Oliver et al [21]
11/Female/49	2005	Asymptomatic	Round/well-circumscribed	3.5	Parapancreatic	Homogeneous/none	None	Intense enhancement	HV	Unknown	Monica et al [10]
12/Male/58	2007	EP	Oval/poorly demarcated	4×3×3	Head	Unknown	None	Enhancement	HV	15D	Hongbei et al [3]
13/Male/54	2007	EP	Oval/poorly demarcated	5.48×5.67	Head	Unknown	None	None	Mixed	Unknown	Justyna et al [2]
14/Female/23	2007	AP	Round/well-circumscribed	8	Body and tail	Homogeneous/none	None	None	HV	1Y	Vonny et al [15]
15/Male/50	2007	AP, LOA, WL	Oval/ill-circumscribed	Unknown	Peripancreatic	Homogeneous/none	None	Homogeneous enhancement	HV	Unknown	Jun et al [35]
16/Male/69	2008	LHP, fever	Oval/well-defined	4×2.5×2.5	Peripancreatic	Inhomogeneous/none	None	Obvious enhancement	Mixed	1Y	Talarico et al [11]
17/Female/50	2008	Asymptomatic	Lobulated/well-demarcated	1.1	Peripancreatic	Homogenous/none	None	Arterial enhancement	HV	Unknown	Kyoung et al [22]
18/Male/62	2009	Unknown	Round/unknown	2.8×3.8	Peripancreatic	Inhomogeneous/none	NDI	Enhancement	Mixed	4M	Adolfo et al [37]
19/Female/31	2010	GF, EP	Oval/well-circumscribed	5.5×4×3.8	Peripancreatic	Inhomogeneous/none	None	None	PC	2Y	Alexandre et al [1]
20/Female/27	2011	Unknown	Oval/well-circumscribed	4.2×4.3	Peripancreatic	Homogeneous/punctate calcification	None	Obvious enhancement	HV	Unknown	Khashab et al [19]
21/Male/64	2012	Asthenia, adynamia, WL, LLMP	Oval/well-defined	5.1×6.1	Head and body	Homogeneous/none	No depiction	None	HV	Unknown	Franz et al [9]
22/Male/43	2012	Asymptomatic	Oval/well-circumscribed	4.2×4.3	Head	Homogeneous/none	None	None	HV	Unknown	Hua et al [8]
23/Female/58	2012	Asymptomatic	Oval/well-circumscribed	4×2.7	Neck	Homogeneous/none	None	Mild enhancement	HV	Unknown	Hua et al [8]

Features of parapancreatic Castleman disease

24/Male/46	2012	AP	Round/well-circumscribed	5×4.5×4.2	Peripancreatic	Homogeneous/none	None	Homogenous enhancement	HV	1Y	Saurabh et al [27]
25/Feamle/38	2012	Asymptomatic	Oval/well-circumscribed	3×3	Neck	Homogeneous/focal calcification	None	Moderate enhancement	PC	6M	Xianhui et al [25]
26/Male/48	2013	EP	Round/well-circumscribed	3.7×3.7	Peripancreatic	Homogeneous/none	None	None	HV	1Y	Filip et al [38]
27/Male/49	2013	Asymptomatic	Round/well-circumscribed	4.5×3.3	Tail	Homogeneous/punctate calcification	None	None	HV	10M	Fu et al [6]
28/Male/39	2013	AP	Unknown	5×3.5	Head	Inhomogeneous/none	None	None	Unknown	Unknown	Fu et al [6]
29/Male/74	2013	Micturation	Unknown	3×2.4	Head	Unknown	NDI	None	PC	26M	Fu et al [6]
30/Female/34	2014	AP, WL	Oval/well-circumscribed	Unknown	Peripancreatic	Homogeneous/foci calcification	None	NDI	PC	2Y	Preethi et al [17]
31/Male/74	2015	Asymptomatic	Oval/poorly demarcated	1.2	Head	Relatively homogeneous/none	None	Slight enhancement	HV	2M	Takaaki et al [4]
32/Female/71	2015	Unknown	Round/sharply demarcated	4×4×3.5	Peripancreatic	None	Hypointense/hyperintense	Moderate enhancement	HV	Unknown	Ang et al [39]
33/Male/40	2016	Unknown	Oval/well-circumscribed	6×4	Peripancreatic	Homogeneous/none	NDI	Enhancement	HV	6M	Andrew et al [40]
34/Male/36	2016	Asymptomatic	Oval/well-circumscribed	7×7.2	Peripancreatic	Homogeneous/patchy calcification	None	Enhancement	HV	20M	Jun et al [41]
35/Male/32	2016	AM	Round/well-circumscribed	6×7	Peripancreatic	Homogeneous/different shaped calcification	None	Intense enhancement	HV	2Y	Leilei et al [18]
36/Male/66	2017	AP	Oval/well-circumscribed	5	Peripancreatic	Homogeneous/punctate calcification	None	None	HV	Unknown	Farnaz et al [16]
37/Male/32	2017	Unknown	Oval/well-circumscribed	14×8	Peripancreatic	Heterogenous/central calcification	None	Distinctive enhancement	HV	4M	Mohammad et al [42]
38/Female/34	2017	EP	Round/well-circumscribed	5×6×3	Peripancreatic	Homogeneous/none	Hypointense/hyperintense	Heterogeneous enhancement	HV	1Y	Nihed et al [34]

Abbreviations: RIF right iliac fossa; WL weight loss; EP epigastric pain; AP abdominal pain; RC renal colic; LOA loss of appetite; LHP left hypocondrium pain; GF gastric fullness; LLMP lower limbs muscle pain; AM abdominal mass; HV hyaline-vascular type; PC plasma cell type; HV-PC hyaline-vascular plasma cell mixed type; NDI no detailed information.

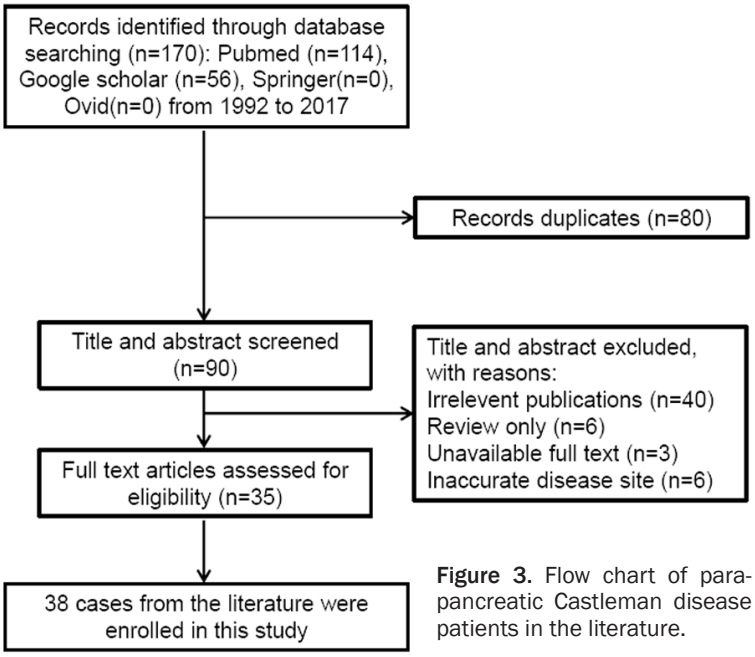

Figure 3. Flow chart of parapancreatic Castleman disease patients in the literature.

Table 3. Summary of distribution site, history disease, symptom, laboratory test and treatment of Castleman disease involving parapancreas in the literature

	Number of patients	Percentage (%)
Distribution site		
China	8	21.1
Japan	6	15.9
Usa	5	13.2
Italy	4	10.6
India	3	7.9
Turkey	2	5.3
Brazil	1	2.6
Greece	1	2.6
Czech	1	2.6
Korea	1	2.6
Poland	1	2.6
Germany	1	2.6
Iran	1	2.6
Spain	1	2.6
France	1	2.6
Austrlia	1	2.6
History disease		
None	20	52.6
Others	12	31.6
Hypertensity	4	10.5
Diabetes	2	5.3
Symptom		
Asymptomatic	9	21.4
Abdominal pain	7	16.6

The second symptom often seen is abdominal pain. More than half patients in this study have no other disease. In the reported literatures, HV type usually displays no clinical symptoms, while the PC type commonly includes various systemic symptoms and laboratory abnormalities. Those patients may often present with fever, malaise, sweating, weight loss, anemia, thrombocytosis, hypergammaglobulinemia, and an elevated erythrocyte sedimentation rate. Of these, fatigue, weight loss and abdominal pain were all uncharacteristic symptoms in present patients [12]. In addition, we also found the patients mostly distributed in Asian countries such as China and Japan.

Radiological features

CT findings: CD often occurs in mediastina, but could be extrathoracic sites such as neck, axilla, mesentery, pelvis, pancreas, adrenal gland, and retroperitoneum [13]. On CT images, parapancreatic CD often presents as a well-circumscribed and smoothly marginated solid mass, with or without calcification, which may or may not invade main pancreatic duct and pancreatic parenchyma [6, 8-13, 15-22]. There is one case involved in the pancreatic duct in previous literature, which showed as a dilated main pancreatic duct compressed by the mass [4]. Calcification in CD is uncommon and occurs in 5%-10% of cases [23]. It often presents as punctate, focal and central calcification in location no matter what types of parapancreatic CD [6, 18, 19, 21, 24, 25]. In addition, central fibrosis can also be involved in this disease

Epigastric pain	7	16.6
Weight loss	5	11.8
Asthenia	2	4.8
Abdominal mass	2	4.8
Fever	2	4.8
Fatigue	2	4.8
Right iliac fossa pain	1	2.4
Micturation	1	2.4
Adynamia	1	2.4
Left hypocondrium pain	1	2.4
Loss of appetite	1	2.4
Gastric fullness	1	2.4
Laboratory test		
Normal level	15	30
Unknown	10	20
Others	9	18
Erythrocyte sedimentation rate	5	10
Creactive protein	3	6
Anaemia	2	4
Hypoalbuminemia	2	4
Hypergammaglobulinemia	2	4
Hyperglicemia	2	4
Treatment		
Exploratory laparotomy	12	41.3
Others	4	13.7
Whiple operations	3	10.2
Distal pancreatectomy	2	6.8
Hemipancreatectomy	1	3.5
High dose corticosteroid	1	3.5
Operative exploration	1	3.5
Laparoscopy	1	3.5
Laparotomy and chemotherapy	1	3.5
Laparoscopic resection	1	3.5
Enuclaetion	1	3.5
Pancreaticoduodendectomy	1	3.5

[11, 26]. The CT features in our case has been given in the results.

Most lesions manifest as homogeneous enhancement after injecting contrast agents [10, 16, 18, 27], while larger lesions (>5 cm) demonstrate heterogeneous enhancement because of the occurrence with fibrosis, necrosis, and degeneration [28, 29]. Due to varying degrees of vascularization in different types of CD, the lesions enhance varing with their histological type. The classical enhanced CT appearance of HV-CD is a single enlarged lymph node or localized nodal mass that demonstrates homogeneous intense enhancement [8]. While, the PC-CD manifests as moderate enhancement due to lesser vascularization [30]. McAdams et al have proposed that the degree of enhancement may be variable on account of the method, volume and rate of injected contrast agent regarding the HV-CD [23]. The patient in our hospital demonstrates classic enhanced CT appearance of HV-CD.

MRI findings

In previous reports, most LCD appear as a well-marginated mass, hypointense on T1WI images and hyperintense on T2WI images, with variable but moderate or intense enhancement after administration of gadolinium [5, 31, 32]. It was reported that some cases of localized hyaline vascular type displayed central and linear hypointense on both T1WI and T2WI images, which attributed to calcifications, fibrous septa, or vessels [5, 32, 33]. These manifestations could also be applied to different types of parapancreatic CD [5, 20, 34]. So far, few cases had been reported regards MRI findings, until Rafaela et al firstly reported the pancreatic CD in the 2003 [5]. There still need more researches to be summarized in terms of MRI features of the different CDs.

The unspecific radiological features of parapancreatic CD result in the difficulty of the differential diagnosis. Not only pancreatic carcinoma, but also various neoplastic, inflammatory (tubercolosis or sarcoidosis) and other miscellaneous peritoneal diseases need to be included in the differential diagnosis, so that the pathological diagnosis is often the only way to get the final diagnosis. Here, we give several other diseases similar to the signs of CD regarding our case in **Table 4**.

Table 4. Differential diagnosis of parapancreatic Castleman disease

Disease	Site	Age	Sex	Symptom	Radiologic features	Study
Parapancreatic castleman disease	Parapancreatic site	Occur in different age	No difference in sex	Often presents no symptoms	A well circumscribed and homogeneous solid mass, obviously enhance but lower than pancreas; Nearly isointense to muscle in signal intensity on T1WI, with heterogeneous signal characteristics within the mass on T2WI	
Gastrointestinal stromal tumors	Mostly occurs in stomach; Secondly occurs in small bowel; Rare occurrences in sophagus, colon, rectum, mesentery and omentum	Above 50 years of age, and rarely are found before the age of 40 years	Slightly higher male prevalence	Non-specific and are basically associated with the site and size of the lesion; Abdominal pain, distension, gastrointestinal bleeding, anemia, body weight loss and palpable mass are some of possible signs of the disease	A well circumscribed mass; Small tumors are often of homogeneous density or signal and large tumors tend to show irregular lobulated margins, mucosal ulceration, central necrosis, hemorrhage, cavitation, and heterogeneous enhancement	[43, 44]
Extramedullary hematopoiesis	Liver/spleen, Abdominal viscera, pleura/lymph nodes/adrenal glands/breast/thymu/kidneys/ gastrointestinal tract, intracranial structures and paraspinal regions	Occur in different age	Higher male prevalence	Lack of characteristics; Often shows as the increase of liver, spleen and lymph nodes except original disease	A well-demarcated and heterogeneous density mass, obviously enhanced; Equisignal on T1WI and equisignal/highsignal on T2WI, slightly evenly enhancement; the signal is mixed when bleeding	[45, 46]
Solid-pseudopapillary tumor of pancreas	The head, body and tail of pancreas	Medium age of 23.9 years	Often occurs in young women; Occasionally in men and older women	Clinical symptoms and signs are non-specific; Often found in physical examination	A cystic or solid/cystic solid density mass with clear/unclear border, complete or incomplete capsule's, can be associated with calcification; Solid part of the mass is medium-hypo intensity signal on T1WI, cystic part is medium-high intensity signal on T2WI, Solid part of the mass shows unevenly medium-high enhancement and the degree of enhancement is lower than pancreas	[47]

Treatment and prognosis

Several studies have shown that surgical resection is a reasonable therapeutic option for the LCD with a favorable long term prognosis [7, 9, 20]. Bowne et al reported that surgical excision has been regarded as the best chance of cure for localised disease [11]. Therefore, surgical excision may be the best option for localised disease. There is no effective therapy ready for MCD which commonly viewed as a systemic disease. Surgical resection may play a limited role for cases of MCD. Steroids, single-agents, radiation, or combination chemotherapy and immunotherapy are often applied to the MCD [3]. However, the effect of these curative methods is not very well. Recently the anti-IL-6 receptor antibody therapy has been expected to become an effective treatment for the disease [7]. In our case, a complete surgical excision was made and no evidence of tumor recurrence has been detected in the three years follow-up.

There are some limitations in our study. First, the sample size of this study is relatively small on account of the parapancreatic Castleman disease is rarely seen. Second, the limited MRI data make it difficult to describe the detailed performance of the lesion, which would have helped to explain the similar CT appearance of parapancreatic CD.

In conclusion, we report a rare case of parapancreatic hyaline-vascular Castleman disease and give a literature review to make radiologists and clinicians aware of parapancreatic Castleman disease. In terms of epidemiology, the disease appears to be more common among in Asian countries. It should be considered as HV-CD when manifests a well-capsulated, solid or cystic solid mass, sometimes calcified, commonly without obvious clinical symptoms. Although rare, the disease is still worthy of attention.

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Disclosure of conflict of interest

None.

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