WITHDRAWN: Lung Involvement Multicentric Castleman of Hyaline-vascular Variant: 3 cases

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Research

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The full text of this preprint has been withdrawn by the authors while they make corrections to the work. Therefore, the authors do not wish this work to be cited as a reference. Questions should be directed to the corresponding author.

Abstract

Background

Castleman disease is clinically divided into unicentric Castleman disease (UCD), with hyaline-vascular Variant (HV) as major pathological pattern, and multicentric Castleman disease (MCD), with plasma cell type (PC) most commonly. Lung involvement is a serious complication of CD and usually leads to death.

Method

We retrospectively analyzed the medical records of 3 patients admitted to The First Affiliated Hospital of Guangxi Medical University during January 1, 2007 to September 30, 2020.

Result

2 male and 1 female with age ranging from 16 years old to 63 years old were admitted. The involved areas varied a lot. Respiratory symptoms were seen in 3 cases, along with fever, weight loss and splenomegaly. Dry and wet rales were found in all patients. All patients had hypoxemia and obstructive ventilation dysfunction. Computed tomography (CT) mainly showed bronchiectasis and mediastinal lymph node enlargement. All 3 cases were complicated with PNP. 1 case failed chemotherapy after local mass excision, 1 case remitted after chemotherapy but the lung lesion was irreversible, and 1 case was untreated and soon die of respiratory failure.

Conclusion

Cases of HV-MCD with pulmonary affected were characterized by small airway lesions and poor prognosis. Respiratory symptoms along with systemic symptoms were common.

Background

CD, previous named as giant lymphadenopathy, angiolymphatic hamartoma, or angiofollic lymph node hyperplasia, the etiology remains unknown and share some clinical and pathological features with virus infection, tumor and some autoimmune disease[1]. HV is the most common pathological type of UCD while PC is the most common type for MCD. HV-UCD is always asymptomatic or presented by compression symptoms because of enlarged lymph nodes compressed adjacent tissues. Damage to skin and mucous membranes which result to oral ulcers or rashes exist when complicated by PNP. As to PC-MCD, it can be manifested as lymph node enlargement in multiple regions, recurrent systemic symptoms (weight loss, fever, fatigue, anemia, edema, hypoproteinemia) and can involve with several important organs, such as lung, liver, spleen, kidney, etc. There have been a lot of coverages about the above two variants and lung involvement is a serious complication, which induce shortness of breath, lung shadow, lung function involvement and other Respiratory symptoms, and finally death due to respiratory failure.

HV-MCD is a relatively rare clinical type, especially those cases with pulmonary involvement, and case has been reported in the literature mentioned the details of respiratory lesion was rare[2] so far. However, the details of pulmonary involvement were not descripted in those reported cases. Thus, to improve the understanding of the clinical characteristics of this rare type, the clinical data of 3 patients were systematically analyzed retrospectively.

Case Report

Case 1 is a 16-year-old boy. He was admitted to hospital because of "abdominal mass accompanied by fever, oral ulcer and shortness of breath for one and a half years and worsened for three months". In January 2008, the patient found an abdominal mass without obvious inducement, accompanied by oral ulcer, fever, shortness of breath, cough, expectoration, abdominal pain, abdominal distension, no fatigue, night sweats, poor tolerance, no discomfort such as dry mouth, dry eyes, joint pain, and no special treatment was conducted. In late March 2009, the patient developed shortness of breath, obvious during activities, and no chest tightness or chest pain. Then admitted to hospital on 2 June 2009. Physical examination on admission: splenomegaly, a mass could be palpated in the abdomen, lymph nodes of the size of soybeans could be palpated in both groins, and rales of dry and wet could be heard in both lungs. Blood routine: WBC: 10.31×10E9/L, HB119g /L, PLT: 286×10E9/L, N%: 0.67, L%: 0.251; Albumin: 27.5g/ L, Globulin: 26.5g/ L, A/G:1.02; CRP: 1.8mg/ L; IgM: 2.52g/ L, IgG: 11.69g/ L, IgA: 1.73g/ L; Autoantibodies: histone (+/-), keratin antibody (+), anti-nuclear antibody (+), anti-DS-DNA (+/-), anti-cardiolipin antibody (+); T lymphocyte subsets: total T cells: 78.2%, CD4+ cells: 39.4%, CD8+ cells: 33.4%, CD4/CD8 ratio: 1.18; Blood gas analysis: pH :7.434, PO2:78.4mmHg, PCO2:42.7mmHg, HCO3:28; Plain CT scan of chest and abdomen: 1. Huge space occupying lesion in the right posterior abdominal cavity. 2. splenomegaly. 3. Emphysema, infectious lesions in the anterior segment of the right upper lobe.4. Endogenous gas in the mediastinum and posterior chest wall. (Figure 1). Pulmonary function examination: FEV1%: 23.3%, FVC%: 67.2%, FEV1/FVC: 28.96%, suggesting: 1. Severe obstructive ventilation dysfunction. 2. Peripheral resistance, total airway resistance Peripheral elastic resistance increased. Bronchial dilation test: absolute value of FEV1 increased by 70ml. Right posterior abdominal mass resection was performed during hospitalization. Postoperative pathology showed Castleman's disease of lymph node HV. Oral mucosal biopsy showed slight epithelial hyperplasia, basal cell liquefaction, no spinous layer release, vascular hyperplasia in lamina propria, lymphocyte infiltration. Accompanied by ulcer formation, ulcer surface has vascular hyperplasia, lymphocytic infiltrate. Diagnosis: 1. Multicentric transparent vascular Castleman's disease (left neck, right supraclavicular, bilateral axilla, inguinal lymph nodes); 2. Paraneoplastic pemphigus; 3. Bronchiolitis obliterans. No significant improvement was observed after the treatment of methylstrongone, aminophylline and bronchodilator. The symptoms were not improved after the treatment of COP regimen combined with interferon and CHOP regimen. He was subsequently loss to follow-up.

Case 2: A 51-year-old man was admitted with "cervical lymph node enlargement with exertive shortness of breath for 1 month". On October 10, 2011, he went to a local hospital due to "inguinal hernia". After

physical examination, cervical lymph node enlargement was found. PET/CT examination showed multiple nodules under the left neck, bilateral clavicle region and mediastinum, with uneven increase in metabolism. Pathological examination of the left cervical lymph node showed HV of giant lymph node hyperplasia (Castleman's disease). The patient was gradually accompanied by weight loss, shortness of breath, blood sputum, right chest pain and oral ulcer. No fever, night sweats, abdominal pain, diarrhea, rash or joint pain were found. No special treatment was given, and the mass in the left neck was larger than before. He went to our hospital for further diagnosis and treatment on November 8, 2011. On admission, physical examination showed that there were several lymph nodes of soybean size in the left neck and supraclavicular fossa. Dry and wet rales can be heard in both lungs. Blood routine examination at admission: WBC: 7.6×10E9/L, N%: 0.548, L%: 0.208, Hb:146g/ L; Albumin: 44.5g/ L, Globulin: 27.5g/ L, A/G:1.8; IgM: 1.693g/ L; PLT: 286×10E9/L. Renal function: Creatinine 100μmol/L, endogenous creatinine clearance rate 57ml/min; T cell subsets: CD4+T cells: 28.10%, CD8+T cells: 41.20%, CD4/CD8:0.68, total T cells: 75.50%. IgG, IgA, liver function and urine routine examination showed no abnormality. Blood gas analysis: pH :7.352, PO2:120mmHg, PCO2:46.5mmHg, HCO3- : 24.1 (oxygen absorption, 2L/min). Chest CT: Multiple mediastinal lymphadenopathies (Figure 2). Diagnosis: Multicenter clear vascular Castleman's disease (left lower neck, bilateral clavicular region, mediastinal lymph node). After four cycles of chemotherapy in CHOP regimen, and no significant reduction of mediastinal lymph nodes was observed. The fifth and sixth cycles of chemotherapy were changed to FCD regimen. On December 8, 2011, chest CT was conducted and result revealed: Mediastinal multiple lymph node enlargement was slightly smaller than before. In January 2013, he began to develop oral ulcer, which was diagnosed as "pemphigus" by the affiliated stemmatological hospital. He was treated with 2 capsules of compound hydrocortisone three times a day, but there was no obvious improvement of oral ulcer. He stopped taking hormones in November 2013 and gradually developed discomfort such as shortness of breath after activity. So, he returned to the hospital for treatment due to cough, sputum and shortness of breath after activity, during hospitalization, lung function examination was performed and result as follows: FEV1%:18.8%, FVC%:48.6%, FEV1/FVC:31.5%, VC%:46.9%, volume of dispersion: 0.49%, rate of dispersion: 0.33, RV/TLC: 78.01. Conclusion: 1. Severe obstructive ventilate dysfunction; 2. Severe peripheral airway obstruction; 3. Severe diffuse dysfunction; bronchial dilation test: 20 minutes after inhalation of Ventolin, FEV1 increased by 16.9%, and the absolute value of FEV1 only increased by 90ml. Chest CT: Patchy, cable-like high-density and ground-glass shadows were seen in both lungs, and multiple enlarged lymph node shadows were seen in the mediastinum. Diagnosis: 1. Castleman's disease 2. Paraneoplastic pemphigus 3. Interstitial pneumonia. The patient was given piperacillin sodium tazobactam, levofloxacin and fluconazole for anti-infection. After discharge, the patient was voluntarily switched to itraconazole but still with mild symptoms. Later, he was admitted to hospital because of "cough, sputum, shortness of breath" and other symptoms, no significant improvement was found but gradually worsened. The reexamination of chest CT indicated that mediastinal lymph nodes disappeared, but the lung lesion is irreversible with cavitation and always visited hospital for respiratory infection.

Case 3: A 63-year-old female was admitted with "left chest and left lower abdominal pain for more than 1 month". She felt paroxysmal dull pain in left chest and left lower abdomen with no obvious cause early in

January 2012, with oral ulcer, fearless cold, fever, nausea, vomiting, diarrhea, no chest tightness, shortness of breath and other discomfort, pain gradually aggravated, and affected sleep. Thus, she went to the local hospital, abdominal ultrasound was conducted but no obvious abnormity was seen. Chest CT found: Patchy high-density shadow can be seen in both lungs, soft tissue mass shadow can be seen protruding into the left thoracic cavity near the spine of the left lower lung, irregular thickening and envelopment of the left pleura, lymph node shadow of mediastinal tracheal bulge. After that, a series of symptoms, namely cough with a little white foam-like sputum, appetite decreased, fatigue occurred. For further diagnosis and treatment, she was admitted to our hospital. Physical examination on admission: right supraclavicular lymph node enlargement. Complete blood routine examination: WBC 9.1×10E9/L, Hemoglobin 103g/ L, PLT 523×10E9/L; Albumin: 29.2g/ L, Globulin: 41.5g/ L, A/G:0.7; CRP: 31.13mg/L; IgM: 1.16g/ L, IgG: 20.27g/ L, IgA: 4.03g/ L; Autoantibodies: Anti-RO-52 antibody (+), and the remaining index were negative. No abnormalities of renal function, routine urine and complement were observed. Pulmonary function examination: FEV1%:69.4%, FVC%:77.6%, FEV1/FVC:73.32%, VC%:77.1%, volume of dispersion: 1.62%, dispersion rate: 1.46%, RV/TLC: 46.18. Conclusion:1. Mixed ventilatory dysfunction with mild limitation. 2. Mild peripheral airway obstruction. Chest CT: Patchy high-density shadow in both lungs, irregular thickening of the left pleura with envelopment, and lymph node shadow in mediastinal trachea. Fiber bronchoscopy showed chronic bronchitis, a small amount of lung tissue, clear alveoli, no tumor. B-mode ultrasound: Multiple enlarged lymph nodes in the left neck and right supraclavicular, multiple hypoechoic masses in bilateral axilla and inguinal region (lymph node sonography). Percutaneous lung penetration of the lower left lung mass: Microscopic observation showed fibrous connective tissue, hyalinosis and small focal-like chronic inflammatory cell infiltration, no histological evidence of granuloma or carcinoma. The microscopic examination of lymph nodes in the left neck showed structural destruction of lymph nodes, atrophy of lymphatic follicles, hyperplasia of interstitial fibrous tissue with hyalinosis, and hyalinosis in the vascular wall, which was consistent with giant lymph node hyperplasia (clear vascular type). Diagnosis: 1. Multicentric Castleman's disease of HV (left neck, right supraclavicular, bilateral axilla, inguinal lymph nodes); 2. Paraneoplastic pemphigus; 3, bronchitis obliterans. After receiving anti-inflammatory, analgesic and other symptomatic supportive treatment, it was suggested to consult the thoracic surgery department to evaluate whether surgical treatment could be performed, but the patient refused and asked to be discharged when the condition did not improve and soon died of respiratory failure.

Summary of the 3 case

Two of the three patients with the mediastinum involved, and their clinical manifestations included cough, expectoration, shortness of breath, chest pain and other common symptoms of respiratory diseases. Meanwhile, all the three patients had refractory oral ulcer, systemic symptoms such as emaciation, fever, splenomegaly, and local compression symptoms such as local lymph node enlargement or abdominal pain. With respect to laboratory examination, anemia is more common. At the same time erythrocyte sedimentation rate, CRP, immune globulin elevated, albumin declined. Chest CT mainly showed bronchiectasis and mediastinal lymph node enlargement, with flocculent, patchy and stripe high-density shadow. Blood gas analysis showed hypoxemia and pulmonary function examination

showed obstructive ventilation dysfunction, among which 1 case was mixed ventilation dysfunction with mild restrictive ventilation dysfunction. The bronchial dilation test was normal. Physical examination and other auxiliary examinations confirmed the polycentric diagnosis of CD.

Table 1 General information and clinical manifestations of the 3 cases of HV-MCD

Case	Age Year	Gender	Occupation	Clinical features	Involved regions
P1	16	Male	Student	Fever, weight loss, shortness of breath, cough, sputum, abdominal pain, abdominal distention, oral ulcer, splenomegaly	Oral mucosa, abdominal cavity, bilateral inguinal lymph nodes
P2	51	Male	Teacher	Neck lymph node enlargement, weight loss, shortness of breath, bloody sputum, chest pain, oral ulcer	Bilateral neck, supraclavicular fossa, mediastinum, abdomen
P3	63	Female	Worker	Chest pain, abdominal pain, oral ulcer	Left neck, left supraclavicular fossa, mediastinum

Table 2. Laboratory test results of the 3 cases of HV-MCD

	P1	P2	P3
WBC(3.5-9.5) (×109/L)	10.31	7.6	9.1
HGB(130-150) (g/L)	119	146	103
PLT(125-350) (×109/L)	329	286	523
N%(0.4-0.75)	0.67	0.548	0.696
L%(0.2-0.5	0.251	0.208	0.145
ESR(0-15) (mm/h	-	-	84
CRP(0-5) (mg/L)	1.8	-	31.13
ALB(40-55) (g/L	27.5	44.5	29.2
GLB(20-40) (g/L)	26.5	27.5	41.5
A/G(1.2-2.4)	1.02	1.8	0.7
C3(0.79-1.52) (g/L)	1.15	-	1.35
C4(0.16-0.38) (g/L)	0.34	-	0.37
IgM(0.840-1.32)(g/l	2.52	1.693	1.16
IgG(8-18)(g/l	11.69	normal	20.27
IgA(0.9-4.5) (g/l	1.73	normal	4.03
Creatinine(59-104)(umol/L)	normal	normal	normal
RF(0-12.5) (IU/ML	2.6	-	5.3
Autoantibody	Histone(+/-), AKA(+) ANA(+), anti-ds-DNA(+/-), ACA(+)	-	Ro-52 +
Tumor marker	Normal	-	normal
T cell subsets	Total T: 78.2%, CD4+T:39.4%, CD8+T:33.4%, CD4/CD8: 1.18	TotalT:75.50%, CD4+T:28.10%, CD8+T:41.20%, CD4/CD8:0.68	-

Table 3 Instrumental examination results of the 3 cases of HV-MCD

	P1	P2	P3
ECG	Sinus arrhythmia	Sinus rhythm, T wave changes	Normal ECG
Abdominal ultrasound	Right abdominal mass of substance	Liver inner gallbladder wall thickened, echo enhancement, gallbladder stone.	Normal
Urinary ultrasound	-	The left kidney cyst	Normal
Others	Abdominal CT plain and enhanced: 1. Huge space occupying lesion in the right posterior abdominal cavity. 2. splenomegaly	PET/CT: Multiple nodules under the left neck, bilateral clavicle area, and mediastinum, with uneven metabolism and increased, with high possibility of lymph nodes, gallbladder stones, and small left renal cyst considered.	B ultrasound: Uterorectal depression and liquid dark area, 20×13cm.Suggest pelvic effusion.

Table 4 Results of blood gas analysis, respiratory function and bronchodilation test in 3 cases of HV-MCD

	P1	P2	P3
FEV1%	23.3	18.8	69.4
FVC%	67.2	48.6	77.6
FEV1/FVC	28.96	31.5	73.32
VC%	67.1	46.9	77.1
Bronchial dilation test	The absolute value of FEV1 was increased by 70ml	The absolute value of FEV1 was increased by 90ml	Negatice
Diffusing capacity	-	0.49	1.62
Dispersion rate	-	0.33	1.46
RV/TLC	-	78.01	46.18
PH	7.434	7.352	-
PO2(mmHg)	78.4	120*	-
PCO2(mmHg)	42.7	46.5	-
HCO3-	28	24.1	-

* The concentration of oxygen inhalation was 3L/min

Discussion

The main pathological type of UCD is HV. Except for local tumor compression symptoms, there are generally no systemic symptoms. Most patients with thoracic UCD are usually incidentally found on imaging. Chest CT typically presents isolated, well-bordered, enlarged lymph nodes or localized nodular masses, with no other pulmonary involvement except the masse[3–8]. Laboratory abnormalities of HV-UCD are uncommon, and lung function is generally normal[9, 10]. However, HV-UCD is prone to PNP then leading to oral ulcer and respiratory symptoms such as cough, sputum, shortness of breath and so on when further involved in the lungs. Hypoxemia was found in blood gas analysis. Most of the pulmonary functions were obstructive ventilation dysfunction, and mosaic sign and other signs of obliterans bronchitis could be seen in chest CT. Mimouni et al[11] found 12 patients with HV-UCD in a study of 14 patients diagnosed with PNP, suggesting that PNP may be a signal of the presence of HV-UCD. In his research,10 patients progressed to BO, and 10 patients eventually died. PNP and pulmonary involvement can be partially improved after resection of the primary tumor in some patients. Therefore, for HV-UCD, surgical resection of the local mass is advocated at present.

The most common pathological type of MCD is PC. PC-MCD is often accompanied by systemic symptoms, such as fever, night sweats, fatigue, loss of appetite, weight loss, organ enlargement, diffuse polyadenosis, and edema. And is associated with a number of laboratory abnormalities, including thrombocytopenia, anemia, leukocytosis, hypoproteinemia, hyperglobulinemia, positive autoantibodies, abnormal renal function, and increases in acute phase proteins such as C-reactive protein (CRP), erythrocyte sedimentation rate (ESR), fibrinogen, and interleukin (IL)-6. Lung involvement are more common than HV-UCD cases, with cough, sputum, shortness of breath and other respiratory symptoms. Hypoxemia is common in blood gas analysis, and pulmonary function is mixed ventilation disorder[12–15], suggesting interstitial lung damage and small airway lesions, and the pathological features of BO and lymphocytic interstitial pneumonia have also been confirmed in literature[16]. CT findings include lymphadenopathy, often with diffuse hilar and mediastinal lymphadenopathy, and pulmonary parenchyma changes, including centrilobular nodules, bronchovesicular bundle and septal thickening, cysts, and pleural effusion. Since MCD involves multiple sites, and is a diffuse lesion, the feasibility of surgical resection is low. Two small studies reported that about 50% of MCD patients achieved complete remission after receiving four drugs combined with chemotherapy[17, 18]. Steroids are primarily used to induce remission in acute situations, but lasting remission is rare and therefore not recommended for maintenance therapy. Currently, rituximab has been used in some CD20-positive MCD patients, either alone or after the failure of other treatments, and more than 50% of patients have achieved remission with mild side effects[19–23].

In this study, 3 cases of HV-MCD included were rare cases. Although the masses were not all in the lungs, they all had respiratory symptoms such as shortness of breath, cough and sputum, chest pain, and other symptoms, as well as discomfort at the location of the masses, fever, emaciation, splenomegaly and other systemic symptoms. In addition, all the 3 cases had PNP manifestations such as intractable oral ulcer. Laboratory tests showed elevated IgM or IgG and positive autoimmune antibodies. These changes

are common laboratory abnormalities in PC-MCD. Blood gas analysis indicated hypoxemia, 2 patients with varying degrees of obstructive ventilation dysfunction and 1 patient with restricted mixed ventilation dysfunction. These lung functions suggested small airway lesions and possible interstitial damage. The most common characteristic of CD accompanied by PNP is stomatitis[24, 25], which is usually the first symptom of the disease and persists throughout the course of the disease. Stomatitis is manifested as mucosal erosion and ulceration, which is the most common in HV-UCD. As a result, involving the lung is a prominent characteristic of the 3 cases of HV - MCD patients in this report and characterized by obstructive or mixed ventilation dysfunction, hypoxemia and carbon dioxide increases, as well as fever, emaciation, immune globulin heightens, autoimmune antibody abnormalities and easy to concurrent PNP. Therefore, it is evident that HV-MCD owns the clinical features of HV-UCD and PC-MCD. From the treatment effect aspect, no matter which type of CD, BO is one of the main causes of death, surgical resection or chemotherapy has no obvious effect, and then lead to the death of patients due to respiratory failure, resulting in poor prognosis.

Wang et al. found that specific B cell clone exist in the HV – CD can produce antibodies to identify antigen expressed in epithelium[26], so easy to concurrent PNP, prone to oral ulcer. Antibodies against bronchial epithelial proliferation caused airway occlusion and thus formed occlusive bronchiolitis. After the removal of diseased tissue, antibody concentration decline. So the removal of tumor brough about good prognosis. Some studies have reported that lung lesions of PC-MCD other than mass were confirmed by pathological biopsy as interstitial fibrous thickening, plasma cell infiltration, and alveolar collapse. Reichard et al. found karyotype change of chromosome 7p15 double-allele containing IL-6 locus in PC-CD, which may be related to the imbalance of IL-6 cytokines[27]. Mihara et al. found that IL-6 could promote the migration of inflammatory cells and the production of antibodies by B cells[28]. Therefore, lung lesions of PC-MCD may be caused by interstitial destruction and fibrosis caused by excessive infiltration of neutrophils and plasma cells due to increased IL-6 secretion. It has been suggested that anti-IL-6 therapy plays an important role in the treatment strategy of PC-MCD.

4 cases of HV-MCD involving the lungs were reported previously[2, 29], but chest CT, blood gas analysis and lung function results were not described in detail. HV-MCD is a rare type of CD. All the 3 cases in this report involved the lungs, suggesting that lung involvement of HV-MCD is an important clinical feature. Attention should be paid when hypoxemia, obstructive ventilation dysfunction, lung shadow, accompanied by fever, emaciation, splenomegaly and other systemic symptoms, for example, anemia, hypoalbuminemia, increased immunoglobulin, CRP and other laboratory abnormalities, or refractory oral mucosa, skin lesions were seen in a patient to rule out whether it is caused by CD. Timely examination of lymph nodes, chest and abdomen should be performed. If there is a mass or enlarged lymph node, early biopsy should be performed.

Conclusion

1. HV-MCD is a rare type of CD and pulmonary involvement, namely small airway lesion is an important clinical feature, which leads to death because of respiratory failure. HV-MCD not only presents like

HV-UCD that easy to be complicated with PNP and bronchiolitis obliterans, but also manifested as the lung shadow and is accompanied by systemic symptoms such as fever, emaciation and splenomegaly, as well as laboratory examination abnormalities such as anemia, hypoalbuminemia, elevated immunoglobulin and elevated CRP, which was common to PC-MCD. Though the primary focus disappears after treatment, the small airway lesion is irreversible and affect quality of life. Thus, the prognostic of HV-MCD is poor. Since HV-MCD is a rare disease and easily misdiagnosed by respiratory physician, if a patient with respiratory symptoms, lymphadenectasis, elevated immunoglobulin, hypoxemia, obstructive ventilation function disturbance, small airway lesion and there is no improvement after conventional therapy, general check-up and lymph node biopsy were needed to clarify a diagnosis.

Abbreviations

UCD: unicentric Castleman disease, HV: hyaline-vascular Variant, MCD: multicentric Castleman disease, PC: plasma cell type, CT: Computed tomography, PNP: Paraneoplastic pemphigus, BO: Bronchiolitis obliterans.

Declarations

Ethics approval and consent to participate

Ethics approval and consent to participate This study was approved by the Ethics Committee associated with the Faculty of Medicine at The First Affiliated Hospital of Guangxi Medical University. Written informed consent was provided by the parents of the patient.

Consent for publication

Consent for publication Written informed consent was obtained from the patient's parents for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Competing interests

The authors have no conflicts of interest to declare.

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Author contributions

Jiemei Cen, Jiarui Chen, Ye Qiu, Jianquan Zhang performed the research; Jiemei Cen, Jiarui Chen, Wen Zeng, Jianquan designed the research study; Jiemei Cen, Jiarui Chen, Ye Qiu, Wen Zeng, Jianquan Zhang contributed essential reagents or tools; Jiemei Cen, Jiarui Chen analysed the data; Jiemei Cen, Jiarui Chen wrote the paper.

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Figures

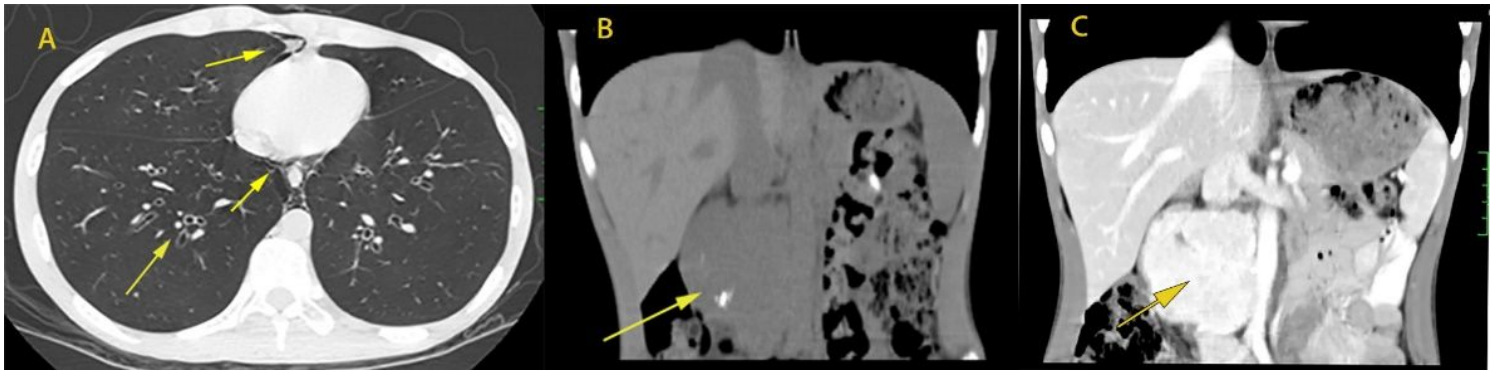

Figure 1

(Case 1) A: Patchy shadow of increased density seen in the anterior segment of the right upper lobe. Endogenous gas was present in the mediastinum and posterior chest wall, and bronchiectasis was seen in both lungs. B(plain scan): the density of the right posterior abdominal mass was uniform; C(enhanced CT): the right posterior abdominal mass was significantly enhanced.

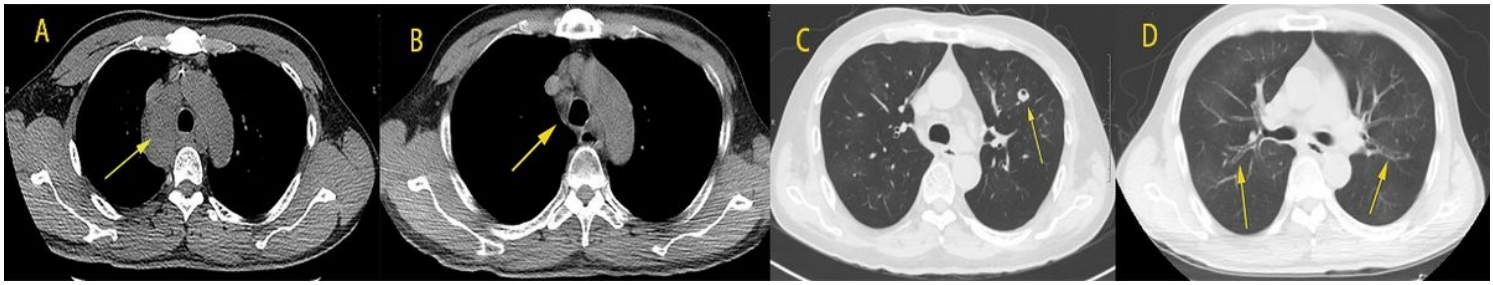

Figure 2

(Case 2). A: (January 2011, before treatment) Mediastinal lymph node enlargement. B:(In 2018, post-treatment review) Mediastinal enlarged lymph nodes disappeared. C :(October 2011) A void can be seen in the left lung; D :(2018) Bronchiectasis is seen in both lungs.

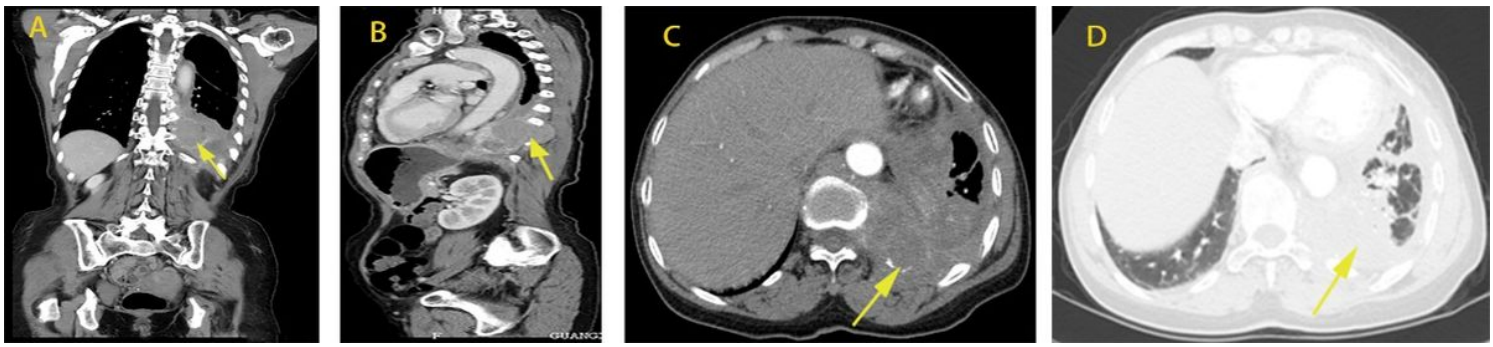

Figure 3

(Case 3). A (coronal view): A mass can be seen near the spine in the left lower thoracic cavity. The capsule is clear, but the diaphragm is not penetrated, accompanied by A small amount of pleural effusion in the left thoracic cavity. B (sagittal): A mass can be seen in the left thoracic cavity, breaking through the pleural cavity with a small amount of pleural effusion. C: A mass can be seen near the spine of the left thoracic cavity, which breaks through the posterior pleural cavity, accompanied by left pleural effusion, and patchy and cable-like exudative shadow of the left lung. D: the enhancement of the left thoracic mass was not obvious

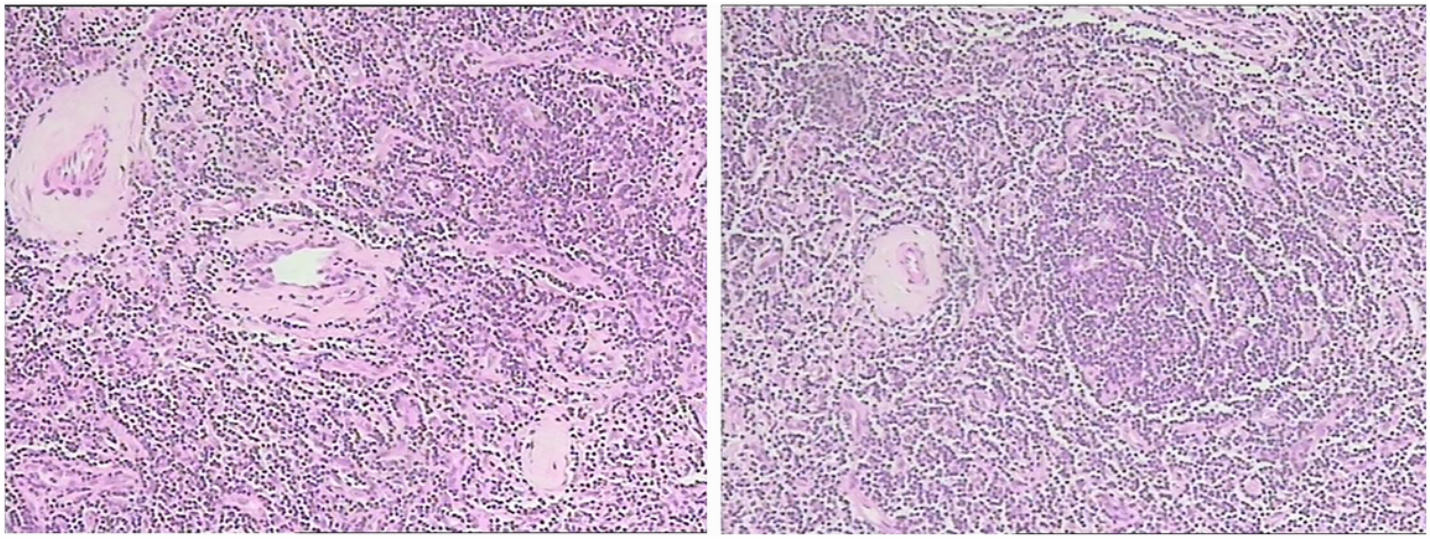

Figure 4

The pathology characteristics of the 3 cases of HV-MCD