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The *WJCC* is now indexed in PubMed, PubMed Central, Science Citation Index Expanded (also known as SciSearch®), and Journal Citation Reports/Science Edition. The 2019 Edition of Journal Citation Reports cites the 2018 impact factor for *WJCC* as 1.153 (5-year impact factor: N/A), ranking *WJCC* as 99 among 160 journals in Medicine, General and Internal (quartile in category Q3).

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Responsible Electronic Editor: *Yan-Xia Xing*
 Proofing Production Department Director: *Yun-Xiaojuan Wu*
 Responsible Editorial Office Director: *Jin-Lei Wang*

NAME OF JOURNAL
World Journal of Clinical Cases

ISSN
 ISSN 2307-8960 (online)

LAUNCH DATE
 April 16, 2013

FREQUENCY
 Semimonthly

EDITORS-IN-CHIEF
 Dennis A Bloomfield, Bao-Gan Peng, Sandro Vento

EDITORIAL BOARD MEMBERS
<https://www.wjgnet.com/2307-8960/editorialboard.htm>

PUBLICATION DATE
 June 6, 2020

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STEPS FOR SUBMITTING MANUSCRIPTS
<https://www.wjgnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION
<https://www.f6publishing.com>

Hemophagocytic syndrome as a complication of acute pancreatitis: A case report

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Supported by the National Natural Science Foundation of China, No. 81800467, No. 81974062, and No. 81770637.

Informed consent statement: Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Conflict-of-interest statement: The authors declare that they have no conflict of interest.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

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Abstract

BACKGROUND

Haemophagocytic syndrome (HPS) is rarely seen in patients with acute pancreatitis (AP). HPS as a complication of AP in patients without any previous history has not been elucidated.

CASE SUMMARY

A 46-year-old man was admitted for symptom of persistent abdominal pain, nausea, and vomiting for 2 d after heavy drinking. During hospital stay, he suddenly developed skin rash and a secondary fever. The laboratory findings revealed progressive pancytopenia, abnormal hepatic tests, and elevation of serum triglyceride, ferritin, and lactate dehydrogenase levels. However, apparent bacterial or viral infections were not detected. He was also possibly related to autoimmune diseases because of positive expression of various autoimmune antibodies and no remarkable past history. Finally, the bone marrow examination showed a histiocytic reactive growth and prominent hemophagocytosis, which resulted in a diagnosis of HPS. Unexpectedly, the patient responded well to the immunosuppressive therapy.

CONCLUSION

HPS is a very rare extrapancreatic manifestation of AP. The diagnosis relies on bone marrow examination and immunosuppressive therapy is effective. For AP with skin changes, the possibility of HPS should be considered during clinical work.

Key words: Haemophagocytic syndrome; Acute pancreatitis; Immunosuppressive therapy

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Manuscript source: Unsolicited manuscript

Received: January 21, 2020

Peer-review started: February 14, 2020

First decision: April 9, 2020

Revised: April 17, 2020

Accepted: May 13, 2020

Article in press: May 13, 2020

Published online: June 6, 2020

P-Reviewer: Can G, Schietroma M

S-Editor: Tang JZ

L-Editor: Wang TQ

E-Editor: Qi LL



Core tip: Haemophagocytic syndrome is a rare extrapancreatic manifestation of acute pancreatitis. We herein report the first such case to share our experience with its diagnosis and treatment. The patient responded well to the immunosuppressive therapy. Early diagnosis and treatment are crucial.

Citation: Han CQ, Xie XR, Zhang Q, Ding Z, Hou XH. Hemophagocytic syndrome as a complication of acute pancreatitis: A case report. *World J Clin Cases* 2020; 8(11): 2364-2373

URL: <https://www.wjgnet.com/2307-8960/full/v8/i11/2364.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v8.i11.2364>

INTRODUCTION

Haemophagocytic syndrome (HPS) is an uncommon disorder that may be characterized by fever, peripheral blood pancytopenia, liver dysfunction, coagulation abnormalities, and hepatosplenomegaly^[1,2]. It may be triggered by genetic disorders, malignant neoplasms, viral infections, and autoimmune disorders^[3-5]. Acute pancreatitis (AP) and HPS are sporadically observed in the course of systemic lupus erythematosus (SLE) or fulminant ulcerative colitis secondary to autoimmune diseases^[2,6,7]. We herein report such a case to share our experience with its diagnosis and treatment.

CASE PRESENTATION

Chief complaints

A 46-year-old man was admitted for symptom of persistent abdominal pain, nausea, and vomiting for 2 d after heavy drinking and then transferred to our hospital.

History of present illness

The patient's symptoms started with mild abdominal pain, which had worsened in the last 2 h.

History of past illness

He had no previous history except for smoking.

Physical examination

The physical examination on admission revealed a discontinuous fever (38 °C) (Figure 1B) and upper abdomen tenderness. His skin was normal.

Laboratory examinations

The laboratory data showed significantly increased serum levels of amylase (1454.9 U/L), lipase (1705.28 U/L), white blood cell count (17 G/L), positive C-reactive protein (4.8 mg/mL), and erythrocyte sedimentation rate (58 mm/h) (Figure 1D-F), slightly elevated glucose (7.71 mmol/L) and D-dimer (1.63 mg/L), slightly reduced calcium (1.88 mmol/L), and hypoalbuminemia (31.0 g/L). The coagulation, hepatic, and renal functions were all within normal limits. The detailed laboratory values are described in Table 1.

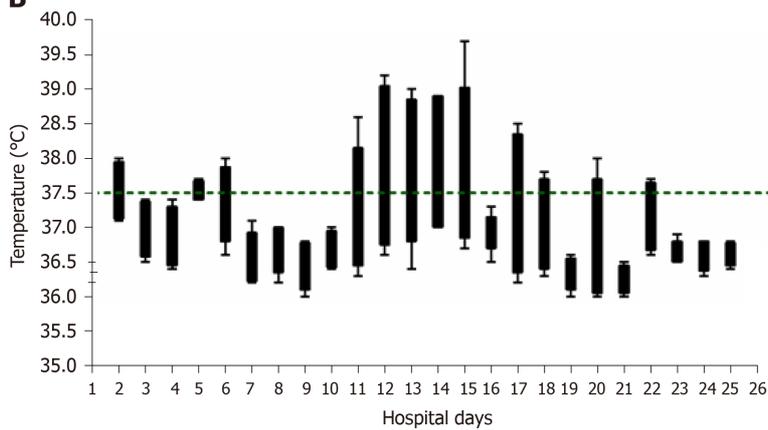
Imaging examinations

Pancreatic enlargement and peripancreatic seepage on computerized tomography confirmed the diagnosis of AP (Figure 2). Then, the patient was given enteral nutrition *via* a jejunal nutrition tube (Figure 1C) and treated with pantoprazole sodium (40 mg per 8 h), octreotide acetate (0.6 mg/q12h), and anti-inflammatory drugs (ornidazole 0.5 g/q8h, meropenem 0.5 g/q8h, *etc.*). Traditional Chinese medicine catharsis was performed for the prevention of intestinal function failure (Figure 1A). The symptoms of the patient gradually disappeared. He had no febrile and each index was close to the baseline level (Figure 1D-F). Bilateral pleural effusion was absorbed better than before (Figure 3). Surprisingly, from the hospital days 11 to 17, the patient suddenly developed a secondary fever and the highest temperature reached 39.7 °C (Figure 1B). We strengthened the antibiotic treatment, including antifungal therapy, although blood culture did not find any evidence of bacterial or fungal infection. Unfortunately, although the temperature was controlled, his general condition

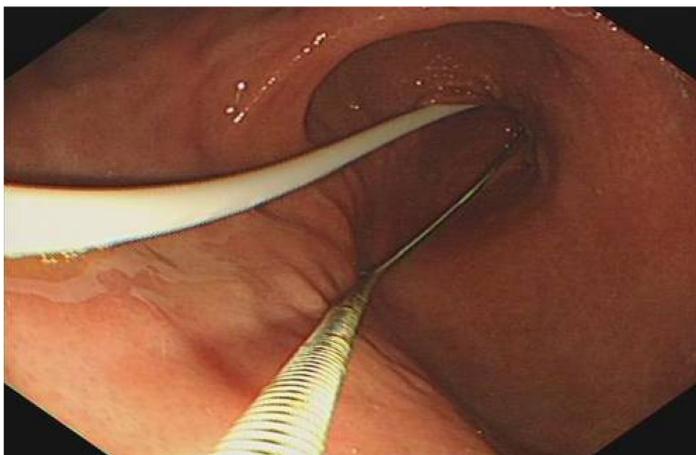
A

Hospital days		1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24	25	26
Fasting		+	+	+	+	+	+	+	+	+	+	+	+	+	-	-	-	-	-	-	-	-	-	-	-	-	-
Jejunum nutrition tube		-	+	+	+	+	+	+	+	+	+	+	+	+	-	-	-	-	-	-	-	-	-	-	-	-	-
Traditional Chinese medicine catharsis		+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	-	-	-	-	-	-	-	-
Nutrition support therapy		+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	-	-	-	-	-	-	-	-
Acid suppression	Pantoprazole Sodium 40 mg q8h	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	-	-	-	-	-	-	-	-
	Pantoprazole Sodium 40 mg qd	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	+	+	+	-	-	-	-	-
	Octreotide acetate 0.6 mg q12h	+	+	+	+	+	+	+	+	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-
	Octreotide acetate 0.1 mg q8h	-	-	-	-	-	-	-	+	+	+	+	+	+	+	+	+	+	+	-	-	-	-	-	-	-	-
Anti-inflammation	Ornidazole 0.5 g qd	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	-	-	-	-	-	-	-	-	-
	Meropenem 0.5 g q8h	+	+	+	+	+	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-
	Meropenem 1.0 g q8h	-	-	-	-	+	+	+	+	+	+	+	+	+	-	-	-	-	-	-	-	-	-	-	-	-	-
	Teicoplanin 0.4 g qd	-	-	-	+	+	+	+	+	+	+	+	-	-	+	+	+	+	+	+	-	-	-	-	-	-	-
	Teicoplanin 0.2 g qd	-	-	-	-	-	-	-	-	-	-	-	-	+	+	-	-	-	-	-	-	-	-	-	-	-	-
	Moxifloxacin hydrochloride 0.4 g qd	-	-	-	-	-	-	-	-	-	-	-	-	-	-	+	+	-	-	-	-	-	-	-	-	-	-
	Fluconazole 0.4 g qd	-	-	-	-	-	-	-	-	-	-	-	-	-	-	+	+	+	+	+	+	+	+	-	-	-	-
	Cefoperazone sodium 3 g q8h	-	-	-	-	-	-	-	-	-	-	-	-	-	-	+	+	+	+	+	+	+	+	-	-	-	-
Hepatic protection	Albumin 10 g qd	-	-	+	+	+	+	+	+	+	+	+	+	+	+	+	-	-	-	-	-	-	-	-	-	-	-
	Furosemide 20mg once	+	+	+	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-
	Salvia miltiorrhiza ligustrazine 10 mL qd	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	-	-	-	-	-	-	-	-	-	-
Antiallergic	Magnesium isoglycyrrhizinate 0.15 g qd	-	-	-	-	-	-	-	-	-	-	-	-	-	-	+	+	+	+	+	-	-	-	-	-	-	-
	Reduced glutathione 1.8 g qd	-	-	-	-	-	-	-	-	-	-	-	-	-	-	+	+	+	+	+	-	+	-	-	-	-	-
Defervescence	Hexadecadrol 5 mg qd	-	-	-	-	-	-	-	-	-	-	-	-	-	-	+	+	+	+	-	+	-	-	-	-	-	-
	Calcium gluconate 20 mL qd	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	+	+	+	+	+	+	-	-	-	-	-
	Vitamin C 3 g qd	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	+	+	+	+	+	+	-	-	-	-	-
Defervescence	Diclofenac sodium 1/3 once	-	-	+	+	+	-	-	-	-	-	+	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-
	Physical cooling once	-	-	+	+	-	-	-	-	-	-	-	+	+	-	-	-	-	-	-	-	-	-	-	-	-	-

B



C



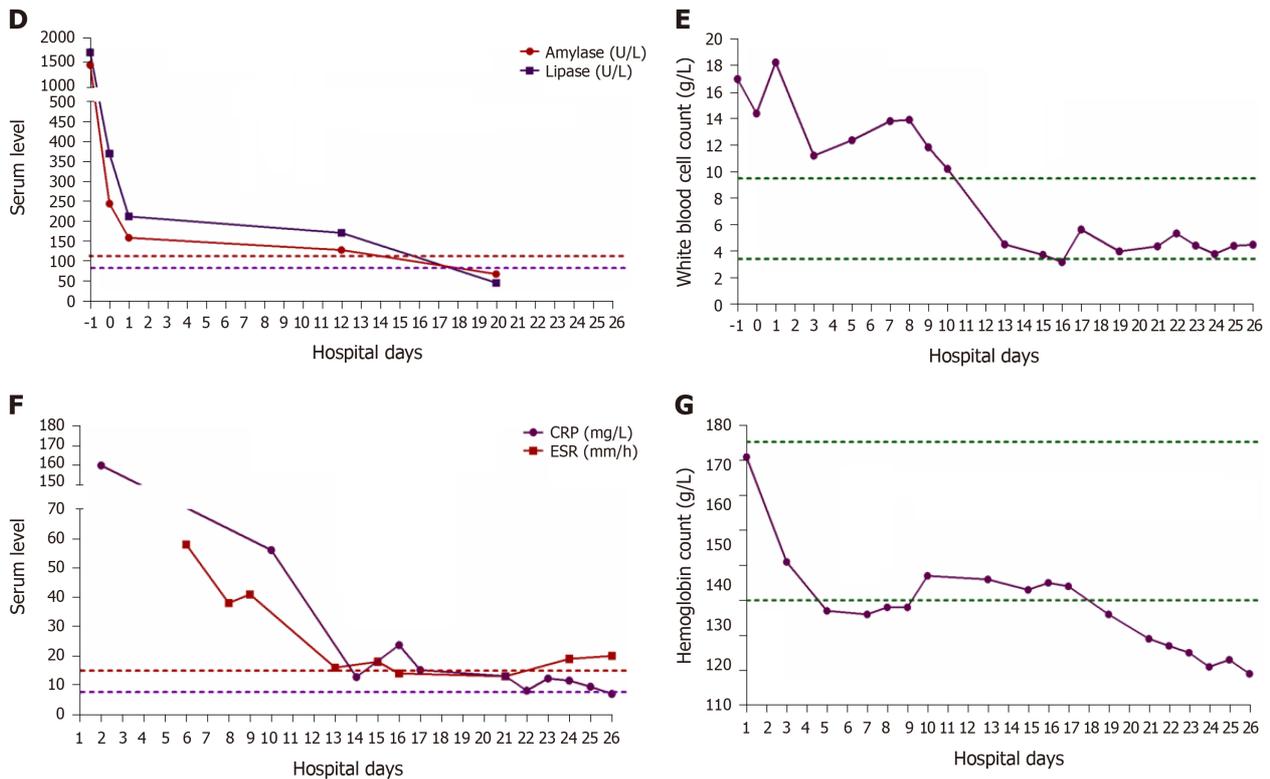


Figure 1 Results of examinations. A: Detailed therapeutic measures taken for the patient with acute pancreatitis during the time of hospitalization. Nutrition support therapy included 5% glucose, compound sodium chloride, hetastarch, mix sugar electrolyte, fructose, compound amino acid peptide, glutamine dipeptide, multi-vitamin, and fish oil fat emulsion; B: Temperature change curve of the patient. The measuring frequency was four times a day; C: Jejunum nutrition tube was placed by gastroscopy; D-G: Changes of amylase, lipase, white blood cell count, C-reactive protein, erythrocyte sedimentation rate, and hemoglobin during hospitalization.

deteriorated. On day 16 after hospitalization, he developed a rash on the trunk, upper limbs, and cruses (Figure 4F), then the indexes of autoimmune disease (e.g., SLE), such as anti-ANA (1:1000), anti-SSA (+), Ro-52-Ab (+), IgG (15.80 g/L), and IgE (471.1 IU/mL), were evaluated. Additionally, he also developed pancytopenia (3.18 g/L of white blood cells, 109 g/L of hemoglobin, and 88 g/L of platelets), hepatic dysfunction (alanine aminotransferase [ALT], 390 U/L; aspartate amino transferase [AST], 172 U/L), and a marked elevation of triglyceride (3.51 mmol/L), ferritin (5850 µg/L), and serum lactic acid dehydrogenase (LDH, 396 U/L) (Figure 4A-E), but his coagulation function was normal without significant abdominal ultrasonography findings.

FINAL DIAGNOSIS

Based on the above findings, a diagnosis of HPS was highly suspected and a peripheral blood smear and bone marrow examination were planned due to the complicated symptoms. Hemophagocytic cells were found in peripheral blood smears (Figure 4G) and the bone marrow examination showed a histiocytic reactive growth and prominent hemophagocytosis (Figure 4H). Thus, HPS as a complication of AP was finally diagnosed.

TREATMENT

The patient was then treated with liver-protecting drugs, antiallergic drugs, and hexadecadrol 5 mg/d for 4 consecutive days. By day 20, his symptoms of pancytopenia, liver function, and LDH and ferritin elevations were improved. On the day that the patient left the hospital (day 26), the laboratory parameters were largely close to the baseline levels again.

Table 1 Laboratory findings for the patient with acute pancreatitis during hospitalization

Day	Index	Value	Range	P/N
1	Calcium	1.88	2.03-2.54 mmol/L	
1	Glucose	7.71	3.9-6.1 mmol/L	
1	Albumin	31.0	35-55 g/L	
1	Creatinine	86.6	44.0-133.0 μmol/L	
1	D-Dimer	1.63	< 0.5 mg/L	
6	PCT	0.28	< 0.5 ng/mL	
7	CA125	320.0	< 35.0 U/mL	
7	CA153	8.2	< 31.3 U/mL	
7	CA199	6.5	< 37.0 U/mL	
14	Anti-ANA	1:1000	< 1:100	
14	IgG	15.80	7.51-15.60 g/L	
14	IgA	2.93	0.82-4.53 g/L	
14	IgM	0.634	0.46-3.04 g/L	
15	Anti-TPO	384.63	< 5.61 IU/mL	
15	Anti-TG	427.53	< 4.11 IU/mL	
15	FT3	2.0	2.63-5.7 pmol/L	
16	C3	0.395	0.79-1.52 g/L	
16	C4	0.052	0.16-0.38 g/L	
16	ASO	49.7	< 200.0 IU/mL	
16	IgG4	0.892	0.03-2.01 g/L	
19	IgE	471.17	1-190 IU/mL	
8	T-SPOT.TB			+
14	Anti-nRNP			-
14	Anti-SSA			+
14	Anti-SSB			-
14	ADA			-
14	DE			-
14	RA-54			-
14	ACA			-
14	AHA			-
14	Ro-52-Ab			+
15	CoxB3-IgM			-
15	CoxB5-IgM			-
15	Evs-RNA			-
15	CMV-IgM			-
15	pANCA			-
15	cANCA			-
15	GBM-Ab			-
15	EB-DNA			-
17	HBV			-
17	HCV			-
17	HIV			-
20	Type A influenza virus			-
20	Type B influenza virus			-
20	Respiratory syncytial virus			-
20	Adenovirus			-
20	Type 1 parainfluenza virus			-
20	Type 2 parainfluenza virus			-
20	Type 3 parainfluenza virus			-

P/N: Positive/negative; PCT: Procalcitonin; Anti-ANA : Anti-nuclear antibody; Anti-TPO: Anti-thyroid peroxidase antibody; Anti-TG: Anti-thyroglobulin antibody; FT3: Free triiodothyronine; C3: Complement 3; ASO: Antistreptolysin; T-SPOT.TB: Tuberculosis T cell spot test; Anti-nRNP: Anti-nuclear ribonucleoprotein;

Anti-SSA: Anti-Sjgren syndrome A antibody; Anti-SSB: Anti-Sjgren syndrome B antibody; ADA: Anti-dsDNA antibody; DE: Anti-ribosomal P protein antibody; RA-54: Anti-nucleosome antibody; ACA: Anti-cardiolipin antibody; AHA: Anti-histone antibody; Ro-52-Ab: Ro-52 antibody; Cox: Coxsackievirus; Evs: Enterovirus; CMV: Cytomegalovirus; pANCA: Anti-antineutrophilic perinuclear antibody; cANCA: Anti-antineutrophilic cytoplasmic antibody; GBM-Ab: Glomerular basement membrane antibody; EB: Epstein-Barr virus; HBV: Hepatitis B virus; HCV: Hepatitis C virus; HIV: Human immunodeficiency virus.

OUTCOME AND FOLLOW-UP

At the 1-mo follow-up visit after discharge, the patient did not take any drugs and had no symptoms or signs without any recurrence.

DISCUSSION

Other abnormal clinical and laboratory findings consistent with the diagnosis of HPS include cerebromeningeal symptoms, jaundice, edema, lymph node enlargement, skin rash, hepatic enzyme abnormalities, hypoproteinemia, and hyponatremia^[8]. In the present case, the symptoms of patient gradually recovered after effective treatment. However, the results of laboratory re-examination were incomprehensibly deteriorative. He developed a secondary fever, rash, pancytopenia, hepatic dysfunction, hyperferritinemia, and elevation of serum triglyceride and LDH levels. The bone marrow examination proved that he had concurrent HPS^[9].

He had no remarkable past medical history, therefore we first considered if HPS was bacteria or virus-associated or malignancy-associated. However, no active infections with viruses as coxsackievirus, enterovirus, cytomegalovirus, hepatitis B virus, Epstein-Barr virus and other respiratory tract infective viruses were observed. Neither prominent hemophagocytosis nor hematological malignancies were detected. There were also no abnormal findings on abdominal CT and tumor markers analysis except slightly elevated CA125. We also suspected concurrent autoimmune diseases because HPS was reported more commonly in patients with pre-existing SLE. However, besides some autoimmune antibodies showing positive expression, no evidence suggested an underlying autoimmune disease.

There was another possibility that HPS was the side effect of pharmaceutical treatment. We indeed used some drugs that could cause rash, pancytopenia, and hepatorenal dysfunction, *e.g.*, antibiotics such as cefoperazone sodium, fluconazole, or teicoplanin and other acid suppression drugs such as pantoprazole sodium. However, all of these are generally well tolerated and therapeutic measures were appropriate^[10-13]. Additionally, some common adverse reactions of these drugs, such as headache, nausea, vomiting, diarrhea, and pruritus did not occur^[14-17]. Therefore, the possibility that it was the side effect of pharmaceutical treatment was ruled out.

HPS is a rare extrapancreatic manifestation of AP. A ambiguous association between AP and HPS was first mentioned in 1998 by Kanaji *et al*^[2], who found HPS associated with fulminant ulcerative colitis and concurrent AP. So far, there have been just two reports about HPS associated with AP in patients with SLE disease.

CONCLUSION

We have described the first case of HPS as a complication of AP, which was successfully treated with immunosuppressive therapy. HPS is a severe condition which requires both early diagnosis and treatment, and HPS as one of the extrapancreatic manifestations of AP should be considered in the future.

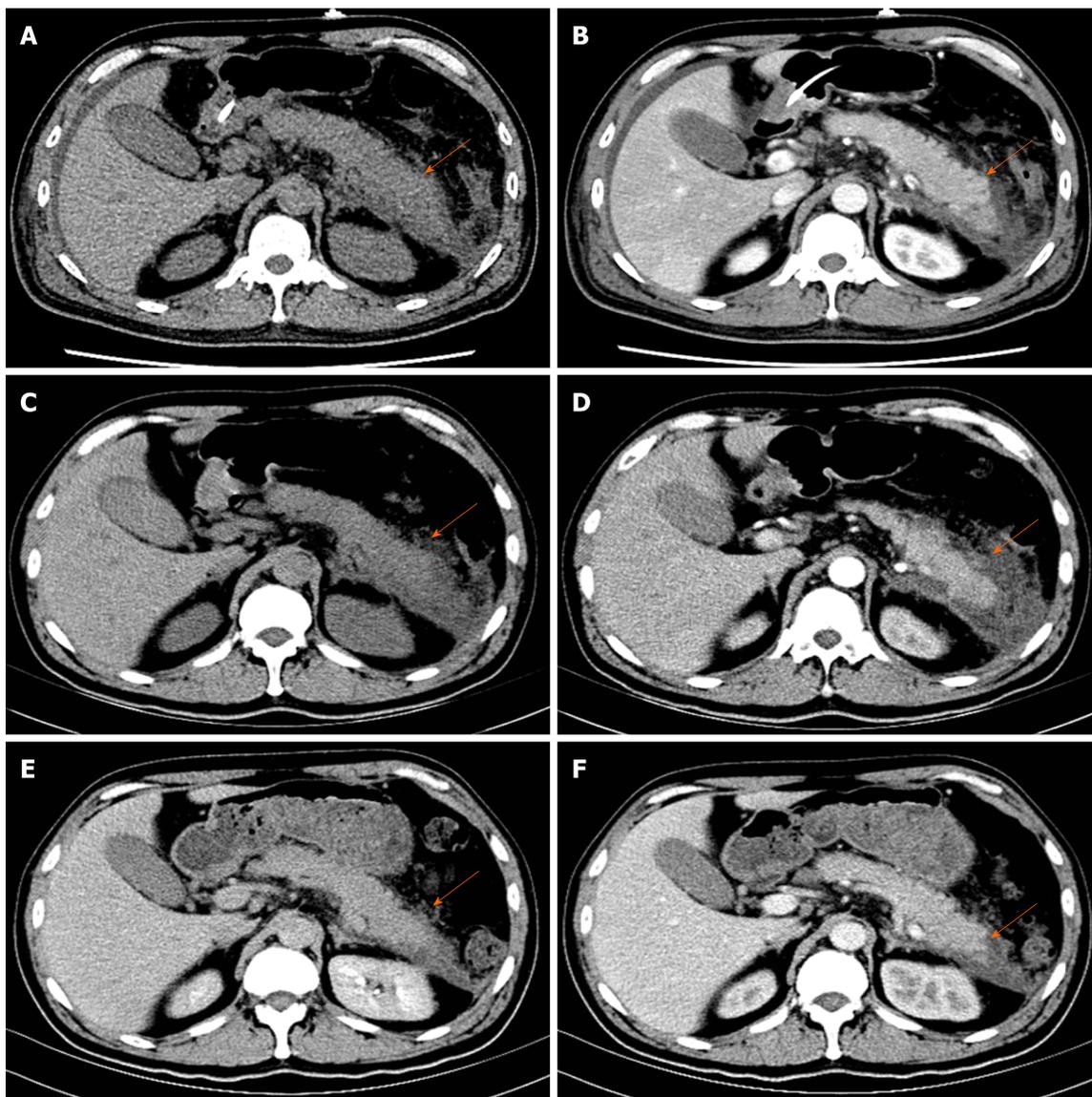


Figure 2 Images of abdominal computed tomography. A and B: Contrast-enhanced computed tomography imaging of the pancreas showed pancreatic swelling, peripancreatic infiltration, and bilateral fascia thickening on the fourth day of hospitalization; C and D: The pancreatic swelling and peripancreatic fluid collections were meliorated on day 13; E and F: The peripancreatic seepage almost recovered on day 23.

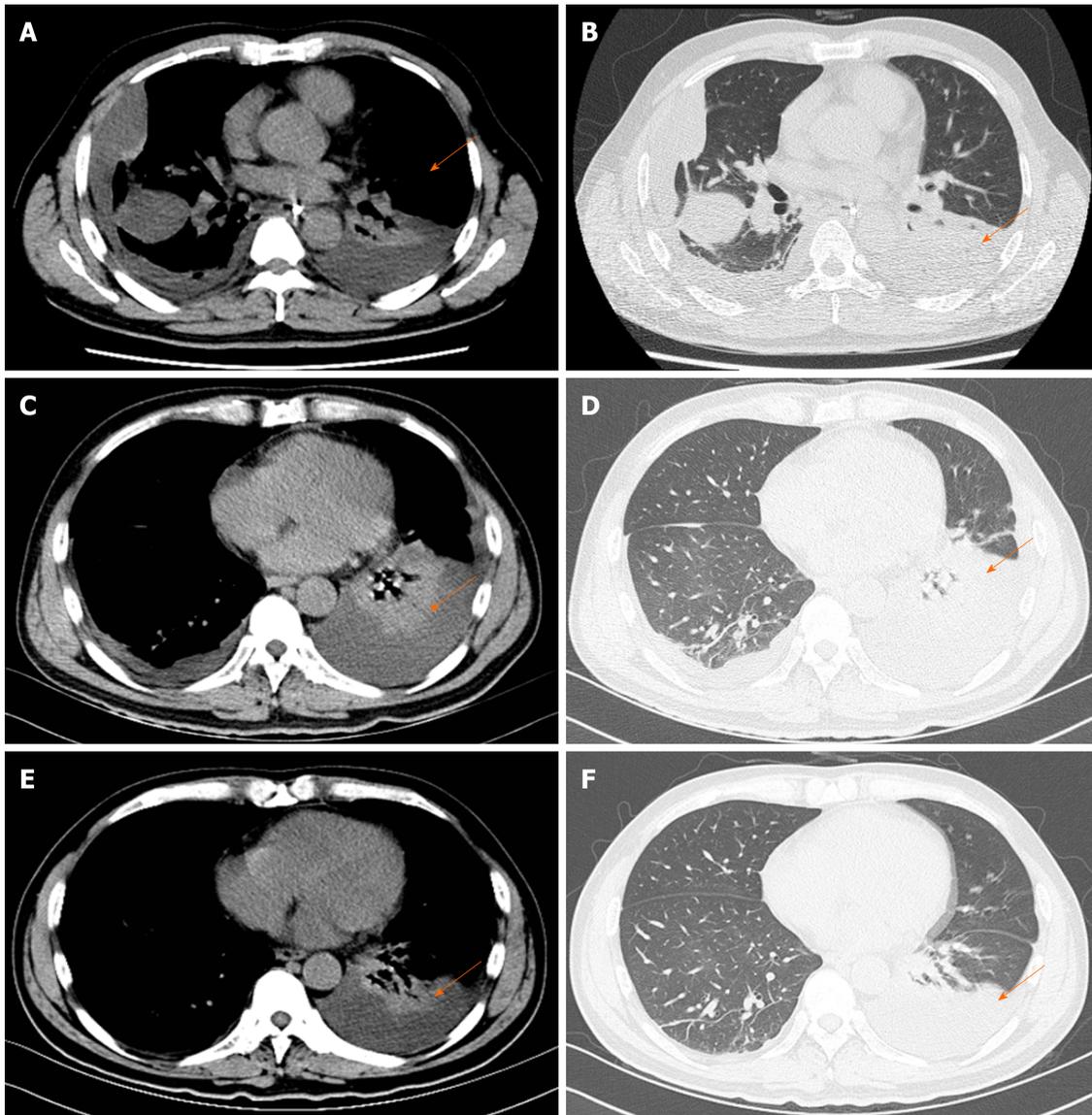


Figure 3 Images of thoracic computed tomography. A and B: On the fourth day, thoracic computed tomography showed bilateral pleural effusion with partially encapsulated effusion. The lungs also showed scattered linear and lamellar high-density shadows, which were considered to be due to an infectious disease; C and D: Thoracic computed tomography indicated that encapsulated effusion was gradually absorbed but the left lung showed segmental atelectasis on day 13; E and F: Bilateral pleural effusion completely disappeared and segmental atelectasis was recovered on day 23.

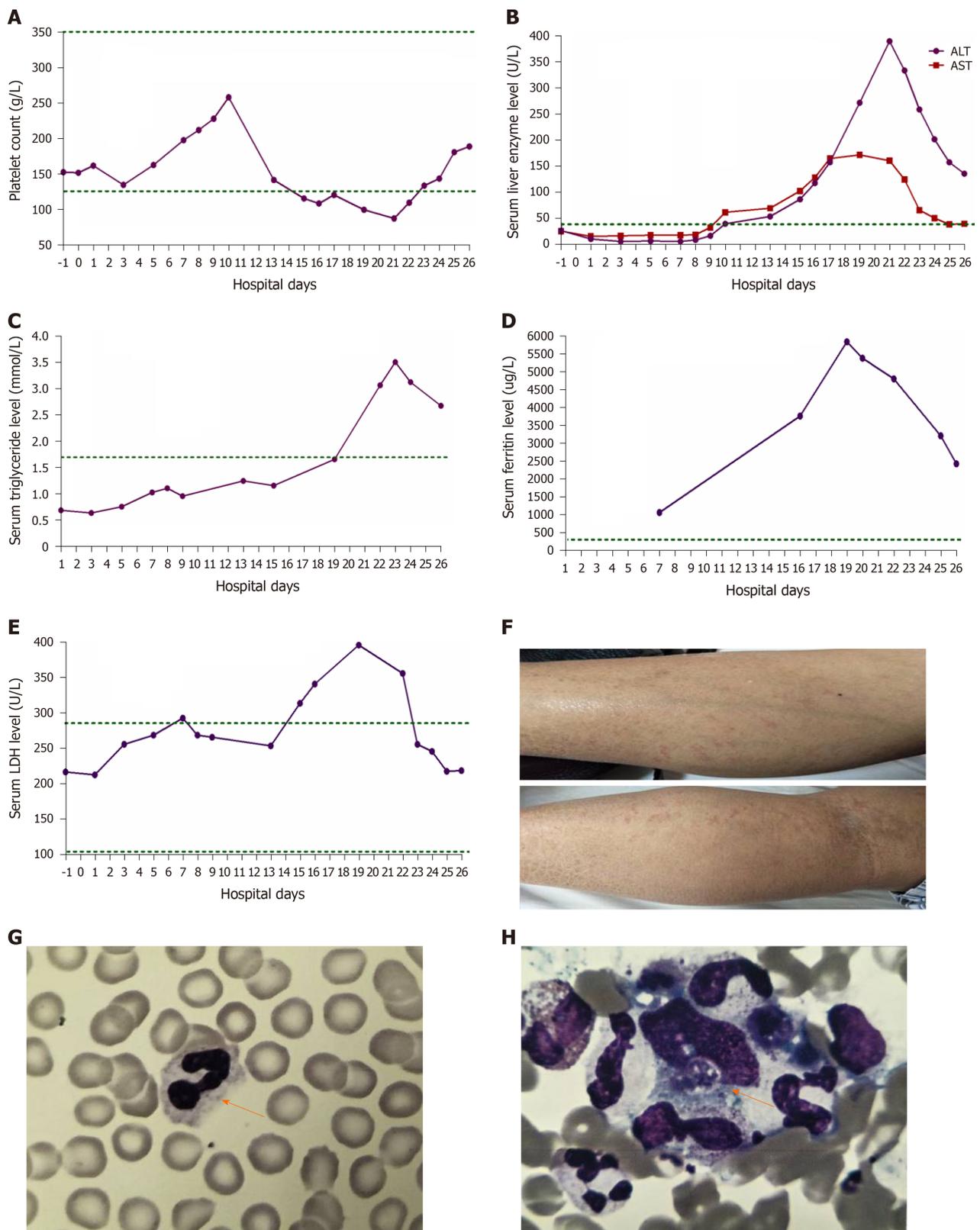


Figure 4 Changes of indexes after effective treatment. A-E: Changes of platelets and serum levels of alanine aminotransferase, aspartate amino transferase, triglyceride, ferritin, and lactic acid dehydrogenase during hospitalization; F: The patient suddenly developed skin rash; G: No obvious abnormality was found in the peripheral blood smear (original magnification, 1×10^3); H: Overactive macrophage phagocytosis, erythrocytes, leucocytes, platelets, and their precursors were also not found in the bone marrow aspiration specimen (original magnification, 1×10^3). ALT: Alanine aminotransferase; AST: Aspartate amino transferase.

ACKNOWLEDGEMENTS

The authors are grateful to the patient for participating in the study.

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