Plasmapheresis Treatment for a Pregnant Woman with Extremely Severe Hyperlipidemic Pancreatitis: A Case Report

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Abstract

Background: The etiology and pathogenesis of hyperlipidemic pancreatitis are not yet fully understood, and the relationship between the severity and different causes of this disease is still not clear. Now the treatment of hyperlipidemic pancreatitis is mainly to reduce the elevated serum triglyceride and control the secondary risk factors. For severe hyperlipidemic pancreatitis, which can deteriorate in a very fast speed, more efficient and effective therapeutic methods are needed.

Case presentation: The patient was a 32-year-old Chinese woman in her 32-week gestation. She was previously healthy, and came to have a sudden onset of acute pancreatitis induced by hyperlipidemia. Her situation deteriorated rapidly, and she simultaneously developed fetal death, respiratory failure, acute renal injury, and severe infections in the lungs, abdominal cavity and the wound. Besides traditional therapies, we used plasmapheresis treatment and successfully saved her life.

Conclusion: In recent years, hyperlipidemic pancreatitis and hyperlipidemic pancreatitis during pregnancy seem to happen more frequently than before. For the extremely severe hyperlipidemic pancreatitis, plasmapheresis seems to be a safe and efficient therapeutic approach, and is often needed to save the patients’ lives. However, evidence for evaluating the efficiency and safety of plasmapheresis in the treatment of hyperlipidemic pancreatitis is still scarce, and we think high-quality, large-scale RCTs in this field are needed for further studies. In addition, by reporting this case, we want to remind general readers to pay attention to adopting a healthy lifestyle, and to encourage researchers and healthcare professionals to continue this research.

Keywords: Plasmapheresis; Treatment; Pregnancy; Hyperlipidemic pancreatitis

Abbreviations: HLP: Hyperlipidemic Pancreatitis; HTG: Hypertriglyceridemia; TG: Triglyceride; FFA: Free Fatty Acids; WBC: White Blood Cell; HGB: Hemoglobin; PLT: Platelets; ALT: Alanine Transpeptidase; Urea: Urea nitrogen; Cr: Creatinine; AMY: Amylase; LIP: Lipase; TC: Total Cholesterol; CRP: C Reactive Protein; PCT: Procalcitoni; CT: Computed Tomography; CVVH: Continuous Veno-Venous Hemofiltration; AP: Acute Pancreatitis; CRRT: Continuous Renal Replacement Therapy

Introduction

The hyperlipidemic pancreatitis (HLP) is pancreatitis caused by an uncommon reason of an underlying lipid abnormality, especially by Hypertriglyceridemia (HTG). The etiology and pathogenesis of HLP are not yet fully elucidated. The basic mechanisms of HLP are due to an elevated level of serum triglyceride (TG). The free fatty acids (FFA) generated by TG may cause pancreatic injury. In typical cases, patients with hypertriglyceridemia that cause an episode of AP usually have preexisting abnormalities in lipoprotein metabolism or have a familial hyperlipidemia, called the primary factor. Meanwhile, some secondary factors, such as poorly controlled diabetes, alcohol abuse, dietary facts, and some drug usages, are often needed. Pregnancy is also a secondary factor. The HLP occurs more in late pregnancy due to increased estrogen levels inducing an overproduction of TG. A hyperlipidemic pancreatitis during pregnancy can be extremely severe and it has high mortality for both the mother and the fetus. Some small-scale, unblinded clinical trials came to the conclusion that plasmapheresis is a safe and efficient approach for treating severe HLP. However, evidence for evaluating the efficiency and safety of plasmapheresis in the treatment of HLP is still scarce, and better-designed studies are needed to clarify this issue.

Case Presentation

Seven months ago, a 32-year-old pregnant woman in her 32-week gestation came to our hospital complaining of severe pain in her upper abdomen for 7 days, and dyspnea and oliguria for 6 days. Seven days before, after the ingestion of a heavy meal, she suddenly suffered a severe, persistent abdominal distending pain, complicated by nausea and vomiting. Then, she was transferred to a nearby hospital immediately. The blood data showed an elevated level of amylase (579 U/L), and an ultrasound revealed acute pancreatitis and a dead fetus in uterus. An immediate cesarean section was performed, and she was safely extubated after the operation. The next day, the patient showed symptoms of abdominal distention and dyspnea. Her urine volume had decreased to less than 400ml/day, and her serum creatinine had increased to 400 μmol/L. She then was intubated and mechanically ventilated. After examination of her serum lipid, the results showed chylemia, and that the TG was beyond the limits of detection, so it could not be evaluated. A diagnostic of hyperlipidemic pancreatitis was made, and an interrupted plasmapheresis (8-10 hours/day) and 3 plasma exchanges were performed on her. Repeated laboratory results revealed decreasing levels of TG (2 days after plasmapheresis was 6.32 mmol/L, thereafter, 4.00 mmol/L). Since her condition had no obvious improvement, she was transferred to the emergency of our hospital and was soon admitted to the intensive care unit. She was previously healthy, and once had a measurement of systolic pressure of 170mmHg on her 12th week of gestation, which could be controlled to about

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130 mmHg through diet and exercise approaches. She had no history of cholelithiasis, diabetes, or smoking, and neither she nor her family had instances of hyperlipidemia.

On admission, she was intubated and mechanically ventilated. Her body temperature was 37.8°C, heart rate was 102 beats/min, respiratory rate was 18 breaths/min, blood pressure was 160/82 mmHg, and arterial oxygen saturation was 98%. Physical examination showed that there were harsh breath sounds, the abdomen was protuberant, and that the wound still had some exudation. There was a subxiphoid tenderness without rebound tenderness or rigidity in her abdomen. Massive vulva and lower extremity edema could also be seen. Laboratory examination revealed white blood cell (WBC) count to be 32.05 × 10^9/L, neutrophil to be 90.9%, hemoglobin (HGB) to be 93 g/L, platelets (PLT) to be 102 × 10^9/L, alanine transpeptidase (ALT) to be 362 U/L, urea nitrogen (Urea) to be 26.23 mmol/L, creatinine (Cr) to be 405 μmol/L, amylase (AMY) to be 134 U/L, lipase (LIP) to be 600 U/L, triglyceride (TG) to be 3.78 mmol/L, total cholesterol (TC) to be 4.38 mmol/L, glucose to be 13.0 mmol/L, C reactive protein (CRP) to be 92.73 mg/L, and procalcitonin (PCT) to be >10 ng/ml. At the same time, the arterial blood gas showed a respiratory failure (PEEP 5 cm H2O, FiO2 40%, PaO2 105 mmHg, PaCO2 31.2 mmHg). Computed tomographies (CT) for chest, abdomen, pelvis and the thin-section pancreas scan were then ordered immediately. They showed pulmonary atelectasis, consolidation, and multiple cord-like shadows in both sides (Figure 1). The pancreas had become swelling and fuzzy, and massive pleural effusion and seroperitoneum could also be seen (Figure 2). The urinary bladder pressure was 25 cm H2O. During the following days, we did repeated bacterial and fungal cultures, and we found Enterococcus faecalis (EFS), Escherichia coli (EC), Candida tropicalis in her sputum, Enterococcus faecium (EFM) in the exudation of the wound, and also Candida tropicalis in her sputum.

The diagnoses of acute pancreatitis with severe case, hyperlipidemia, respiratory failure, pulmonary infection, acute renal injury, infections of the wound and abdominal cavity, and postoperative status of cesarean section were made.

Omeprazole and somatostatin were used intravenously to reduce pancreatic secretion. Due to her high abdominal pressure, we did gastrointestinal decompression and purgation, and performed abdominal paracentesis to reveal the fluid. At the same time, a continuous veno-venous hemofiltration (CVVH) was performed. The volume of dehydration was varied from 150 ml/h to 350 ml/h depending on her abdominal pressure and urine volume. Albumin was used intermittently to increase the plasma colloid osmotic pressure. We gave her anti-infective and antifungal therapy with Tienam (Imipenem/ Cilastatin), Vancomycin (Vancomycin), and fluconazole according to the microbiological findings. On the 6th day after admission, an intestinal feeding tube was put fluoroscopically; thereafter, the enteral nutrition (Peptison) was gradually added. Ten days after admission, her abdominal pressure had gradually reduced to normal level, her renal function had improved, and urine volume had increased. We then stopped CVVH and gradually withdrew the ventilator. On the 23rd day after admission, the tracheal cannula was removed. She was discharged in a stable condition, and was instructed to continue enteral nutrition and to be closely followed up.

Three months after being discharged, the patient was admitted to the emergency room of our hospital again, complaining of a sudden breakdown of the wound and massive turbid fluid pouring out from it. She was febrile, and no dyspnea, abdominal pain, nausea or vomiting was reported. She was obviously more emaciated than before. Physical examinations showed about a 3 cm disruption in the middle of the wound, with large amounts of turbid fluid exuding from it. CT showed multiple pseudocysts forming in the abdominal cavity, with multiple septa among them. The pancreas was atrophying, and there was a lot of seroperitoneum in the abdominal cavity (Figure 3). A drainage tube was inserted into the abdominal cavity under the guidance of CT; after that, several hundred milliliters of fluid were drained out every day. After a comprehensive treatment, 2 months later, her incision was healed and the drainage gradually reduced (Figure 4).

Conclusions

The HLP is pancreatitis caused by an uncommon reason of an underlying lipid abnormality, especially by hypertriglycerideremia (HTG). HLP commonly induces an episode of acute pancreatitis (AP) or recurrent pancreatitis. According to recent researches, the incidence of HLP with AP was 3% in the US [1], 1.8% in Japan [2], 6% in Croatia [3], and 10.36% in Beijing, China [4]. In typical cases, patients with HTG that cause an episode of AP usually have preexisting abnormalities in lipoprotein metabolism or have familial hyperlipidemia. Meanwhile, some secondary factors such as poorly controlled diabetes, alcohol abuse, dietary facts, and some drug usages are often needed [5].

Pregnancy is also a risk factor for HLP. The HLP occurs more in late pregnancy, when the serum TG increases to the peak level. The HTG induced AP during pregnancy used to be rare, but has become more frequent. In recent studies, among patients with AP during pregnancy, the proportions of which caused by hyperlipidemia was estimated to be 27.8%-50% [6,7].
In this case, the patient reported not to have a preexisting abnormality in lipoprotein metabolism or familial hyperlipidemia. No diabetes, alcohol intake, or drug usage was reported. Her episode of AP had a sudden onset and rapid deterioration, with severe complications of fetal death, respiratory failure, pulmonary infection, and acute renal injury occurring together. It also had a protracted course, which together gave a fatal attack to the patient and her family. According to present studies, the relationship between the severity and different causes of HLP is still not clear. For severe or very severe cases, both a primary (genetic) factor and a secondary factor are often needed, and the role of genetic factors in HLP is still waiting to be studied [8]. This patient was under a very complicated situation, which might indicate a very poor prognosis. After a comprehensive treatment, she had a good recovery. Plasmapheresis, which includes continuous renal replacement therapy (CRRT), and continuous veno-venous hemofiltration (CVVH), has been performed respectively to rescue the patient. Plasmapheresis as a treatment for HLP has been applied in clinical practice during these years and has proven to be a valid and safe technique [9], for it could make rapid and efficient reductions of inflammatory mediators, and proinflammatory cytokines such as IL-1, IL-6, IL-10 etc., thereby preventing the inflammatory cascade. At the same time, the body fluid volume was precisely controlled, and the blood lipid could be rapidly lowered. According to our experience, plasmapheresis is an efficient therapeutic approach and is often needed for treating severe HTG complicated by AP in addition to other traditional therapies. However, evidence for evaluating the efficiency and safety of plasmapheresis in the treatment of HLP is still scarce. The majority of literature refers to small-scale, unblinded clinical trials with unclear randomizing methods, or some single cases [10-13]. A recent systematic review including 7 trials and 360 cases showed that CVVH can significantly reduce the levels of lipid, IL-6, overall mortality and APACHE II score in comparison with only routine medical treatment. Meanwhile, a significant heterogeneity and limitations due to the quality and quantity of the included studies have been reported [14]. High-quality and large-scale RCTs in this field are needed for further studies.

Studies have manifested that the incidence of HLP increased year by year in China [4,15]. In the Beijing region, a recent multicenter study on etiology of AP revealed that the hypertriglyceridemia induced AP increased at a faster speed than alcoholic AP and has become the second cause of AP in Beijing [4]. It may be attributed to the rising living standards and better recognition of this disease. In China, people’s living standards continue to rise, but many people lack knowledge of rational diet. Studies showed that the prevalence of HTG in Chinese adults was approximately 11.9% in 2002 [16], and till 2014, the rate was estimated to be 17.7% [17]. In particular, pregnant women in China will always become the focus of the family. They usually assume that the more nutrition the mother intakes, the more nourished the infant will be. This may lead to bad consequences. As we can see in this case, it is difficult to predict who is likely to develop an episode of AP, especially for people without hyperlipidemia or other risk factors. Therefore, especially for people without preexisting hyperlipidemia, we highlight the importance of adopting a healthy lifestyle, such as having a rational diet, avoiding alcohol intake, and controlling risk factors to prevent the episode of AP.

**Consent to Publish**

We have obtained consent to publish from the participant and to report the patient’s data.

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**Figure 3:** Contrast abdominal and pelvic CT.

**Figure 4:** Contrast abdominal and pelvic CT. Two months after the drainage tube inserted into the abdominal cavity.
References


