

Gallbladder sludge in a pregnant woman as the cause of severe complicated hemorrhagic-necrotizing pancreatitis with a spectacular manifestation

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ABSTRACT

Pancreatitis is an obvious but rare event in pregnancy. From mild disease to multiorgan failure and sepsis, acute pancreatitis has numerous causes and often an unpredictable outcome. The authors present a case of a 22-year-old pregnant woman with severe pancreatitis due to biliary sludge. The unusual clinical manifestation of pancreatitis in our patient is worth emphasizing: massive bleeding from the upper alimentary tract and two concomitant pancreatic fistulas. The bleeding

was a manifestation of pancreatic juice-induced injury to the splenic artery, whereas the fistulas were a consequence of disconnected duct syndrome and superficial necrosis of the pancreatic head. After two and a half years of treatment, the patient was on a regular oral diet with supplementation of pancreatic enzymes, and showed normal glycaemia levels. She returned to full physical activity.

Keywords: Biliary sludge, hemorrhage, pancreatic fistula, pregnancy, severe pancreatitis

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INTRODUCTION

Acute pancreatitis is a consequence of intrafollicular activation of digestive enzymes, and results in anti-systemic release of pro-inflammatory cytokines [1]. Although the disease is usually mild, approximately 20% of affected individuals may develop its severe form with pancreatic necrosis and progressive multiorgan failure. This leads to a septic state in approximately 40-70% of the patients and secondary multiorgan failure [2]. Complications of acute pancreatitis constitute an indication for surgical treatment; as a result of surgical intervention, 100% mortality may be reduced to 24-39%, or even down to 6-12% [3]. Cholelithiasis one of the most common causes of acute pancreatitis, and pregnancy predisposes to the formation of cholesterol deposits due to changes in the lipid composition of bile and impaired gallbladder motility [4]. Acute pancreatitis is a well-known, although rare, complication of pregnancy. Its incidence is estimated at 3 per 10,000 [5], and relative risk of acute pancreatitis in pregnant women is 1.43 [6]. We report a case of a young female patient, who presented with non-specific symptoms of acute pancreatitis during her pregnancy, and developed unusually severe complications post-*puerperium*.

CASE PRESENTATION

A 22-year-old woman, mother of two children, had a history of episodic severe epigastric pain with nausea and vomiting, starting from the 4th month of her second pregnancy. No significant abnormalities were documented on physical examination, and abdominal ultrasound did not reveal any pathologies, apart from a "gallbladder sludge." Despite the reported ailments, the patient was treated in an outpatient setting and was diagnosed with catarrhal gastritis and neurotic disorders. However, the pain did not resolve post-partum; due to progressive exacerbation, the patient was hospitalized at the local hospital six months later and eventually diagnosed with acute pancreatitis. Diagnostic imaging confirmed the presence of gallbladder sludge without evident gallstones. As the status of the woman deteriorated throughout a 1-month inpatient treatment period, she was transferred to the 2nd Department of General and Gastroenterological Surgery in Bialystok. On admission she complained of acute pain, and presented with palpable irregular resistance in the epigastrium and left-sided pleural effusion. The patient had neither a fever nor an abnormal lipid profile, but showed elevated acute phase parameters: leukocyte count (13.22×10^3), CRP (117 mg/l), and fibrinogen concentration (601 mg/dl). The clinical status of the woman corresponded to a 4-point Apache II score and

Balthazar grade D. She was treated conservatively with carbapenems and parenteral nutrition. After 30 days of the treatment, her status improved and she was discharged home.

Emergency surgery. The ailments recurred two weeks after the discharge. The patient was re-admitted to the Clinic with signs of severe dehydration. Severe pain induced a constrained body position, and a 20-cm resistance zone could be found on palpation of the central epigastrium. The patient still had no fever; she scored 5 on the Apache II scale, and enlarging peripancreatic collections of fluid, corresponding to Balthazar grade D pancreatitis, were documented on computed tomography (CT) scans. The CT severity index (CTSI) amounted to 5 points, and intraperitoneal pressure reached 23 mmHg. The peripancreatic cyst was drained with a "pigtail" catheter, with evacuation of ca. 1200 ml ichorous content. The patient received piperacillin-tazobactam (Tazocin) along with total parenteral nutrition. Her status improved, and the volume of fluid collected from the "pigtail" catheter ranged between 200 ml and 900 ml per day. However, massive upper gastrointestinal bleeding with rapidly progressing hypovolemic shock occurred on the 18th day of the drainage. Endoscopy revealed that the "pigtail" catheter perforated the stomach and caused massive arterial bleeding from the posterior gastric wall. Due to failure of the endoscopic intervention and progressive hemorrhagic shock, the patient was operated. Intraoperative examination showed bleeding from the splenic artery perforated with the catheter. The bleeding was stopped by ligation, and resection of the cyst was performed along with a necrosectomy. Also, the gallbladder was removed as the presence of gallstones was documented intraoperatively. Due to postoperative cardiorespiratory failure, the patient was mechanically ventilated in an ICU (Intensive Care Unit) setting for 30 days. After readmission to our Clinic, we observed normal healing of the surgical wound and restoration of physiological intestinal passage. Unfortunately, we also found trace amounts of pancreatic juice in the drainage fluid from the peripancreatic space. The pancreaticocutaneous fistula secreted 300-400 ml fluid per day and did not close despite the administration of somatostatin. As a result of intensive rehabilitation, the status of the patient improved so she could be discharged home. Since the active fistula still required catheter drainage, its surgical treatment was recommended for a later time.

Treatment of the pancreatic fistula. The MRI (magnetic resonance imaging) performed after re-admission to the Clinic two months later showed loss of pancreatic parenchyma and discontinuity of the pancreatic duct at proximal 1/3 tail of the

pancreas, along with a 7-cm cyst in the splenic hilum [Fig.1]. Therefore, the patient was subjected

to left-sided pancreatectomy and splenectomy [Fig. 2].

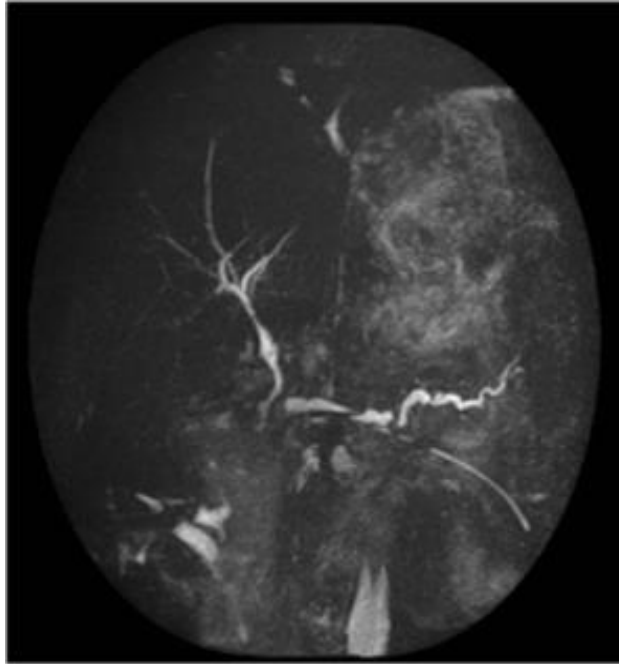


Figure 1. Magnetic resonance cholangiopancreatography. Discontinuity of the pancreatic duct (disconnected duct syndrome). Contrasting agent leaks through a catheter



Figure 2. Pancreatosplenectomy. Histopathological examination revealed the presence of a blood-filled cyst (6.7 x 4 cm) in the splenic hilum

The pancreaticocutaneous fistula remained active post-surgery, and secreted 100-200 ml pancreatic juice per day. Moreover, a parenchymal injury was found in the head of the pancreas, as another location of a pancreatic fistula. The patient was subjected to selective endoscopic sphincterotomy, and appropriate integrity of the pancreatic stump was confirmed intraoperatively.

Reimplementation of somatostatin resulted in decreased leakage from the pancreatic fistula. The patient was discharged home in good general status, with the fistula secreting up to 50 ml pancreatic juice per day, and low activity of pancreatic amylase. The catheters were periodically replaced in an outpatient setting. After eight months, the patient was rehospitalized again due to the falling

out of a catheter placed within the pancreatic fistula. We continued conservative treatment with external drainage of the fistula, which eventually resulted in its spontaneous closure. The patient was discharged home in good general status, on a regular oral diet with supplementation of pancreatic enzymes, and with normal glycaemia levels.

DISCUSSION

Non-specific pain observed during pregnancy and puerperium in an alcohol abstinent woman, only mild clinical symptoms and lack of evident cholelithiasis, resulted in a delayed diagnosis of acute pancreatitis. It is incomprehensible that at the beginning of the disease, despite continuing patient complains, additional lab tests were not performed. No level of amylase was assayed, nor was the Ranson score calculated. That is why this case is presented. Only intensification of the same complains forced personnel to perform additional tests and enabled making the right diagnosis. It is obvious that early diagnosis and management of acute pancreatitis, especially in the case of acute biliary pancreatitis gives much better treatment results [7].

Cholelithiasis is the most common cause of acute pancreatitis in pregnant women [8]; however, it should be emphasized that the absence of cholesterol deposits does not necessarily preclude the presence of this condition. In one study, the presence of gallbladder sludge was documented in 23 (74%) out of the 31 patients treated due to idiopathic pancreatitis; the sludge was visualized ultrasonographically in nearly half of the subjects (n=11, 48%) [9]. Also, pregnancy is associated with increased risk for gallbladder sludge and gallstone formation.

The authors of one study found ultrasonographic evidence for gallbladder sludge and gallstones in 15% and 6% out of 242 women in the first trimester, respectively. However, repeated ultrasonographic examination of the same group at the end of pregnancy documented the presence of newly-formed gallbladder sludge and gallstones in 30% and 2% of the women, respectively [10]. Gallbladder sludge and gallstones are asymptomatic in most pregnant women, and usually resolve spontaneously postpartum [11]. In 61% of women partaking in one of the studies mentioned above, gallbladder sludge detected during pregnancy was no longer visible 5 months postpartum, and 28% of the patients presenting with gallstones during their pregnancies did not show any deposits 9.7 months after childbirth [12].

It is estimated that approximately half of pregnant women treated conservatively for calculous cholecystitis will experience another episode of this condition antepartum [13]. A severe form of acute pancreatitis usually develops in the

third trimester. Also, Swisher et al. showed that pregnant women with acute pancreatitis and concomitant cholelithiasis presented with significantly lower Ranson's scores [14]. However, despite the better general status of this subset, these authors did not observe a significant decrease in the percentage of patients requiring surgical intervention, or improvement of maternal and fetal outcomes. Untreated biliary pancreatitis is associated with a 37% risk for maternal mortality and a 38% risk for malformation in fetuses beyond 20 weeks of gestation [15].

CONCLUSIONS

In the presented study, patient delivery was not affected by pancreatitis. She gave birth to a healthy daughter. Having no complaints, she is still the outpatient of the gastroenterological surgery department. Correct diagnosis at the beginning of the disease could have decreased the severity of acute pancreatitis. The authors emphasize that biliary sludge can be an important cause of severe acute pancreatitis in pregnant women. According to our experience, we strongly suggest conservative and supportive management of acute pancreatitis in pregnancy, when it is possible. Cholecystectomy should be delayed until the postpartum period, if the patient remains clinically stable.

Conflicts of interest

All authors disclose any financial benefits and have no competing interest.

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