Garin *et al.*
Bulletin of the National Research Centre (2024) 48:63
https://doi.org/10.1186/s42269-024-01216-z

Bulletin of the National Research Centre

CASE REPORT

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Severe fetal distress and acute maternal pancreatitis secondary to severe hypertriglyceridemia: a case report

Charlotte Garin[1*], Agathe Bridier-Petit[1], Sophie Ménard[1] and Clotilde Lamy[1]

Abstract

Background Acute pancreatitis (AP) during pregnancy is a rare condition and a potential cause of maternal–fetal mortality. Its diagnosis can be challenging. Hypertriglyceridemia (HTG) is one of the causes. We describe a case of severe fetal distress in the third trimester of pregnancy, associated with severe HTG complicated by AP.

Case presentation A 35-year-old pregnant patient at 36-week gestation presented to the emergency department with acute epigastric pain. After excluding preeclampsia and initially challenging laboratory analyses, a diagnosis of AP was made based on a lipase level 12 times the normal range. The etiology was HTG at 40 times the normal level. Despite initial conservative management, the patient deteriorated rapidly clinically, and severe fetal distress (SFD) necessitated an emergency cesarean section. Lactescent blood appearance was observed intraoperatively. We suspect that the severe and rapid HTG may have caused acute fetal hypoxia.

Conclusions AP during pregnancy is a rare condition, with significant maternal–fetal mortality. Early diagnosis remains challenging. A multidisciplinary approach is necessary for optimal management, with special consideration for rapid fetal delivery in cases of AP secondary to HTG due to the increased risk of fetal mortality.

Keywords Pancreatitis, Pregnancy, Hypertriglyceridemia, Severe fetal Distress, Case report

Background

Acute pancreatitis (AP) during pregnancy is a rare condition and a potential cause of maternal–fetal mortality. Its diagnosis proves to be a challenge. Hypertriglyceridemia (HTG) is one of the causes. We describe a case of severe fetal distress (SFD) in the third trimester of pregnancy, associated with severe HTG complicated by AP.

Case presentation

This is a 35-year-old patient, gravida 4, para 2, at 36 weeks of gestation. Her first two pregnancies were uncomplicated, with full-term vaginal deliveries. Her medical history includes a normal body mass index of 23 kg/m^2, hypertension treated with an angiotensin-converting enzyme inhibitor, discontinued at the beginning of pregnancy. Her diet was unremarkable, and she did not drink alcohol or take any other medication. She also underwent a dilatation and curettage for miscarriage. Family history includes hypercholesterolemia and hypertension. During this pregnancy, she was diagnosed with and managed for gestational diabetes with diet. According to our protocol, lipid profile screening was not performed. In the antenatal care, her blood pressure was always within the normal range and its glycemic profile has always been correct, meaning well-balanced gestational diabetes. Screening ultrasounds at 13, 21, and 31 weeks of gestation were reassuring, showing normal fetal growth. A noninvasive prenatal testing (NIPT) for trisomies 13, 18, and 21 was low risk.

*Correspondence:
Charlotte Garin
Charlottegarin08@gmail.com
[1] Department of Obstetrics, Hôpital Universitaire de Bruxelles, Brussels, Belgium

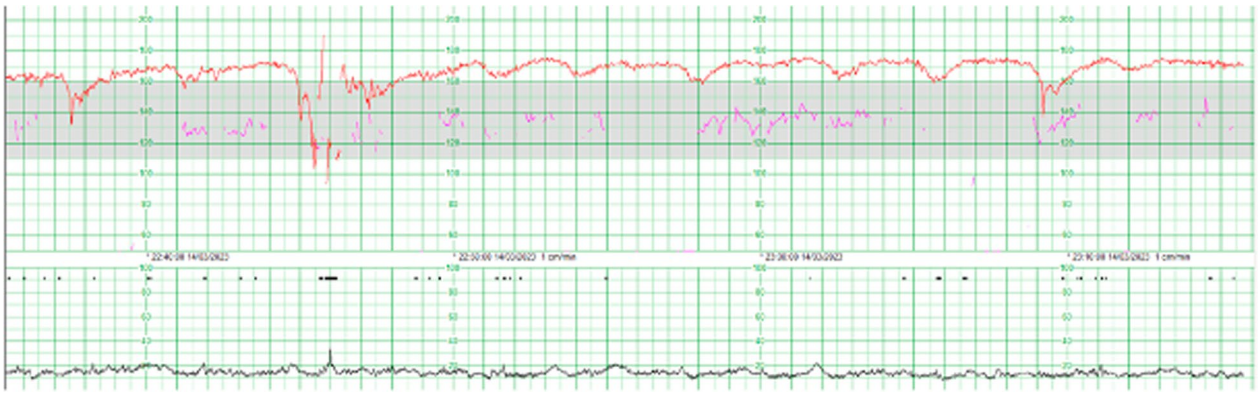

Fig. 1 Fetal monitoring showing base line elevation, decreased variability, and occasional decelerations

At 36 weeks of gestation, the patient presented to the emergency department with acute epigastric pain, chest tightness, and dyspnea, along with some uterine contractions but no fluid leakage or vaginal bleeding. She had no edema of the lower or upper limbs or face. Her blood pressure on admission was 151/96 mmHg, raising suspicion of severe preeclampsia, for which magnesium sulfate (MgSO4) treatment was initiated. Her oxygen saturation was 100%, her heart rate 100 beats per minute and she was apyretic. Ultrasound showed a vertex fetus with normal vitality, and the cardiotocograph was reassuring with occasional contractions. Initial blood tests were inconclusive due to lactescent plasma, but subsequent tests showed elevated lipase at 730 UI/L (12 times normal) and normal liver enzymes, leading to the diagnosis of AP secondary to severe HTG, confirmed by triglyceride (TG) levels of 79.3 mmol/L (40 times normal). Hemoglobin was 118 g/L and platelets 182×10^9/L. At the same time, we received the urinary protein creatinine ratio of 0.25, enabling us to exclude preeclampsia and discontinue MgSO4 treatment. We suspect that the origin of the HTG, in the absence of any other risk factors, might have been familial. The patient was admitted to a high-risk pregnancy unit for maternal–fetal monitoring, hydration, low-fat diet, and pain management with paracetamol. However, her pain management became challenging and required switching to morphine. Fetal monitoring initially showed some abnormalities, including progressive baseline elevation, fetal tachycardia for 2 h, decreased variability, loss of accelerations, and occasional decelerations (Fig. 1). Despite morphine, the patient's pain persisted. A subsequent blood test, 10 h later, showed further elevation of lipase to 2837 UI/L (47 times normal). Due to worsening fetal distress, an emergency cesarean section under general anesthesia was decided 14 h after the admission of the patient (Fig. 2). At this point, the maternal heart rate was 150 beats per minute, with normal oxygen saturation. During the procedure, milky blood was noted. 18 min after the decision of cesarean section, the patient delivered a 2880 g girl with an umbilical cord arterial pH of 6.85 and Apgar scores of 1–4-8 (at 1, 5, and 10 min, respectively). The newborn was admitted to the neonatal intensive care unit

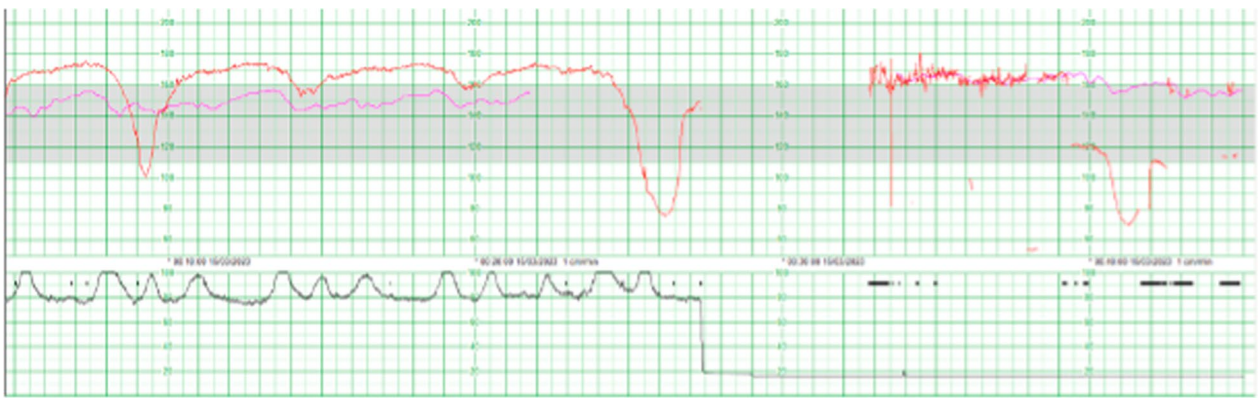

Fig. 2 Fetal monitoring with severe fetal distress

for asphyxia and successfully treated with therapeutic hypothermia. Her condition improved favorably.

Postoperatively, the patient was admitted to the intensive care unit for a week. An abdominal CT scan showed AP with a modified Balthazar score of E without gland necrosis (severity index of 4/10). Serial blood tests showed gradual improvement in lipase and TG levels. However, she developed multiple necrotic-hemorrhagic collections with bacteremia, necessitating radiological and surgical drainage and long-term antibiotic therapy. The fluid collections were predominantly peri-pancreatic, spanning from the root of the mesentery to the greater omentum, primarily within the retroperitoneum. Additionally, there was a collection adjacent to the cesarean scar. After 4 months of hospitalization, the patient was discharged home with close outpatient gastroenterological and endocrinological follow-up and antihypertensive treatment.

Conclusions

During pregnancy, AP is a rare condition, with an incidence that varies and is reported between 1/1,000 and 1/10,000 (Haiyan et al. 2022; Gupta et al. 2022; Cruciat et al. 2020; Ducarme et al. 2014; Papadakis et al. 2011; Ramin et al. 1995). These variations can be attributed to different diagnostic criteria and geographical factors (Kingsnorth and O'Reilly 2006). Furthermore, recent years have seen an increase in the frequency of AP during pregnancy, a phenomenon that can be attributed notably to the global rise in obesity incidence and poor dietary lifestyle habits (Kingsnorth and O'Reilly 2006; Mądro 2022).

AP is more frequently described in the third trimester of pregnancy. Ramin et al. reported the prevalence of AP as 19% in the first trimester, 26% in the second, 53% in the third, and 2% postpartum (Ramin et al. 1995).

The most common etiology is gallstone disease, described in studies between 65 and 70%. Subsequently, alcohol abuse and HTG (approximately 4–5%) are the next two most common causes (Ducarme et al. 2014; Papadakis et al. 2011; Serpytis et al. 2012). Other rarer etiologies include hyperparathyroidism, pancreatic tumors, trauma, infectious agents (HIV), autoimmune, and hereditary causes (Kingsnorth and O'Reilly 2006).

However, a retrospective analysis of 54 cases of acute gravid pancreatitis reports HTG as the leading cause, with an incidence of 41% in a cohort of Chinese patients (Tang et al. 2018).

TG levels physiologically increase during pregnancy under the influence of estrogen, peaking at 2–4 times the normal level in the third trimester. They usually remain below 2.8 mmol/L and rarely exceed 3.8 mmol/L. Lipids are essential for fetal development and organogenesis (Gupta et al. 2022). This physiological increase notably explains the higher incidence of AP in the third trimester.

When TG levels rise, they can become harmful to the pancreas. According to recent research, a widely reported threshold is 11.3 mmol/L, beyond which the risk of developing AP is high (Haiyan et al. 2022; Cruciat et al. 2020; Ducarme et al. 2014; Papadakis et al. 2011; Mądro 2022; Serpytis et al. 2012; Tang et al. 2018; Vandenbroucke et al. 2009; Cain et al. 2015; Shi et al. 2021).

To our knowledge, only two similar clinical cases to ours have been described. The teams observed lactescent blood appearances with TG levels at 95.4 and 127.7 mmol/L, and one of the two teams also reported that the laboratory initially could not perform the analyses due to technical issues (Vandenbroucke et al. 2009; Exbrayat et al. 2007) In our case, the patient had TG levels of 79.3 mmol/L upon admission, with a lactescent serum. Results were obtained from a second blood draw, as the first one was interpretable.

This illustrates, among other factors, the complexity of diagnosing AP during pregnancy. In the third trimester, when faced with acute upper abdominal pain, a differential diagnosis must be undertaken, and preeclampsia should be ruled out. In our case, MgSO4 treatment was initiated due to the clinical presentation before blood test results were available.

The diagnosis of AP is established according to the Atlanta classification criteria, which require the presence of at least two of the following three elements: clinical epigastric pain, suggestive imaging (CT scan, ultrasound, or MRI), and a lipase level exceeding three times the normal range. The diagnostic criteria are the same as outside of pregnancy (Gupta et al. 2022; Mądro 2022; Tang et al. 2018; Cain et al. 2015; Shi et al. 2021).

However, common symptoms of pancreatitis (nausea, vomiting, abdominal discomfort, etc.) are also frequently described during normal pregnancies. Clinical examination of the abdomen can be challenging due to the displacement of intra-abdominal organs by the uterus. Classic clinical manifestations of peritonitis may be attenuated in pregnant patients due to stretching and displacement of the anterior abdominal wall, thus moving away from the inflammatory area (Serpytis et al. 2012).

Managing the differential diagnosis also proves to be a considerable challenge due to the number of potential pathologies (Pallavee 2015) However, AP represents a priority to be rapidly excluded, given its association with significant maternal and fetal mortality. Early detection and appropriate management of AP during pregnancy are essential to minimize risks.

Historically, maternal–fetal mortality reached 20–50%. However, more recent studies describe perinatal mortality rates between 0 and 18% and maternal

mortality decreased to less than 1% (Ramin et al. 1995). The improvement in maternal mortality is attributed to more precise diagnoses through better complementary examinations (laboratory tests and imaging) and improved therapeutic management. Fetal outcomes have improved due to progress and development in neonatal intensive care (Papadakis et al. 2011; Cain et al. 2015). However, fetal mortality related to pancreatitis remains significant, primarily due to an increased risk of prematurity and intrauterine fetal death (Ducarme et al. 2014). A case of missed diagnosis, masked by suspicion of uterine rupture, has been reported, resulting in fetal demise (Pallavee 2015).

Cases of biliary pancreatitis are associated with better perinatal outcomes compared to non-biliary pancreatitis cases, which are associated with fetal mortality rates described up to 60% (Ducarme et al. 2014; Cain et al. 2015). According to Tang et al., AP associated with HTG is more strongly linked to SFD syndrome compared to pancreatitis of other etiologies. This team describes a fetal loss rate of 20.4%, attributed to likely insufficient maternal–fetal monitoring. Furthermore, as the severity of pancreatitis increases, the incidence of SFD and fetal death also increases (Tang et al. 2018).

Our case highlights the importance of maternal–fetal monitoring, as evidenced by the occurrence of unexpected SFD syndrome. We hypothesize that the continuous and rapid increase in TG up to 40 times the normal level could have led to fetal hypoxia by disrupting placental exchanges, possibly due to increased blood viscosity, thereby compromising adequate fetal oxygenation and resulting in the occurrence of SFD.

There is no international consensus regarding the management of AP during pregnancy due to the rarity of the condition. Treatment of the acute phase remains the same as outside of pregnancy, depending on the etiology and is conservative in nature. Conservative management includes a low-fat diet, adequate hydration, and pain management (Cruciat et al. 2020; Ducarme et al. 2014; Papadakis et al. 2011; Kingsnorth and O'Reilly 2006; Mądro 2022; Serpytis et al. 2012; Vandenbroucke et al. 2009; Cain et al. 2015; Exbrayat et al. 2007).

In cases of AP secondary to HTG, heparin therapy and insulin therapy are therapeutic options. They increase lipoprotein lipase activity, promoting TG degradation (Tang et al. 2018). For rapid TG reduction, plasmapheresis may be performed, although its optimal role is unclear and its use should be considered on a case-by-case basis (Gupta et al. 2022). Additionally, fetal extraction allows for rapid TG reduction and should be considered based on gestational age (Exbrayat et al. 2007).

Therapeutic management should be multidisciplinary and conducted in a specialized center. It is primarily determined by gestational age and disease severity, necessitating an individualized approach for each patient. Medical termination of pregnancy or delivery should be considered based on the severity of the condition. Vaginal delivery is preferred, as cesarean section exposes the patient to an increased risk of infection (Cruciat et al. 2020). However, cesarean section is sometimes the best option in some conditions of maternal or fetal distress, or in the usual obstetrical indications. The main management of HTG remains its prevention and screening. Currently, systematic screening for HTG through TG measurement during pregnancy is not recommended in Belgium. Some teams suggest TG measurement in the first and third trimesters. Patients with documented HTG should adjust their lifestyle by adopting a low-fat diet and engaging in regular physical activity (Gupta et al. 2022; Office de la naissance et de l'enfance 2022).

In conclusion, AP during pregnancy is a rare but potentially serious condition, with HTG being among its most common etiologies. Prevention is key in its management. Early diagnosis of AP remains a challenging task. Close maternal–fetal monitoring is crucial for promptly detecting any potentially severe complications and implementing appropriate therapeutic measures to preserve the health and well-being of both the mother and the fetus. Given the higher risk of fetal mortality in cases of AP due to HTG, expedited fetal extraction should be considered.

Abbreviations

AP	Acute pancreatitis
HTG	Hypertriglyceridemia
SFD	Severe fetal distress
NIPT	Noninvasive prenatal testing
MgSO4	Magnesium sulfate
TG	Triglycerides
HIV	Human immunodeficiency virus

Acknowledgements
Not applicable.

Author contributions
ABP and CG analyzed the data, conducted a scientific literature search, and drafted the article. CL and SM participated in the writing of the article and provided their expert opinions in obstetrics. All authors have read and approved the manuscript.

Availability of data and materials
The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
The ethics committee of the Hôpital Universitaire de Bruxelles (H.U.B.) approved the research on April 11, 2024, with the reference number P2024/217.

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

Competing interests
The authors declare that they have no competing interest.

Received: 22 April 2024 Accepted: 18 May 2024
Published online: 05 June 2024

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