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CYSTADENOMA OF THE EXTRAHEPATIC BILIARY TRACT AND PANCREATIC TAIL CYST RUPTURE AMONG SURGICAL CONDITIONS OF PREGNANT WOMEN

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ABSTRACT

Objective: Evaluation of the results of surgical interventionsforemergency surgical conditionsin pregnancy. **Material and methods:** Retrospective analysis of medical records of 11 women hospitalized in 2012-2017, who underwent surgery during pregnancy for emergency surgical conditions. The analysis included the patients' age, gestational age at the time of surgical treatment, type of treatment, histopathological results of the material obtained as a result of surgical procedures, and obstetric outcomes. **Results:** Emergency surgical interventions (11 cases, 0.108%) selected from10,160 deliveries during the 6-year-period. Appendicitis (7 cases, 63.6%) was the most frequent condition requiring surgical intervention.Other conditions were: cholecystitis (2 cases, 18.2%), giant cyst of the extrahepatic biliary ducts(1 case, 9.1%), and rupture of the pancreatic tail cyst concomitant with acute pancreatitis and peritonitis (1 case, 9.1%). Laparotomy and laparoscopy were performed in 10and 1 woman respectively. Pretermdeliveries related to the surgery were recorded twice (18.2%). Reoperation was necessary in one of three (27.3%) cases complicated by peritonitis. **Conclusions:** Surgical intervention due to surgical emergenciesin pregnancy poses a risk of complications threatening the health and life of the pregnant woman and obstetric failure such as preterm delivery.

KEYWORDS: pregnancy / appendicitis / cholecystitis / cystadenoma of extrahepatic bile ducts / pancreatic cyst rupture.

INTRODUCTION

Undertaking surgical intervention during pregnancy, whenever delay is possible, is recommended in the second trimester. This is associated, *inter alia*, with the completion of fetalorganogenesis. Acute appendicitis (AA) and cholelithiasis are the most common surgical conditions requiring intervention in pregnant women, of them appendicitis poses a significant clinical challenge.

The literature data present different frequency of AAin pregnant women, ranging 0.04- 0.95%[1-4], different in subsequent trimesters of pregnancy, i.e. 24.5-47.6%, 40-51%, 4.8-24.5% in the 1st, 2nd, and 3rd trimester respectively.[2-4] Pregnancy-related physiological differences may account for delayed diagnosis and implementation of prompt treatment. Some symptoms of appendicitis may be misinterpreted as accompanying the pregnancy. In addition, the abdominal anatomy altered by the pregnant uterus poses diagnostic difficulty.

Both the diagnosis and decision to qualify for surgery are predominantly clinical, andbased on laboratory and imaging results. The classic criteria include the diagnostic sequence of colicky central abdominal pain followed by vomiting with migration of the pain to the right iliac fossa, i.e. a triad described by Murphy. To support clinical diagnosis of appendicitis, the judicious use of leucocytosis which is raised up to several thousand / µL (with the shift to the right), and CRP are recommended. Abdominal ultrasound examination often reveals the presence of free fluidin the peritoneal cavity.[4,5]

The cases of AAare treated by surgery. Appendectomyshould be undertaken within 24 hours after admission to hospital.[6,7] Maternal and fetalconditionworsen dramatically along with the severity of appendicitis. Fetal loss in simple appendicitis occurs in 1.5% cases, with generalized peritonitis (6%), and if the appendix perforates it amounts up to 36%.[8] In addition to the risk of intrauterine fetal death, perforation of the appendix increases the risk of preterm delivery, while adhesions in the smaller pelvis can complicate future female fertility.[9] Considering the significant risk, if the appendixperforates in a pregnant patient, any delay in intervention should be avoided. The technique of appendectomyin the pregnant patient depends on the gestational age and available surgical knowledge. If perforation of the appendix is suspected, immediateappendectomy by laparotomy should be

performed.[8] Even if the appendix looks normal, it should be removed. Up to 35% of laparotomies are negative.[10]

The use ofmagnetic resonance imaging (MRI) can eliminate that uncertainty. When clinical examination findings and ultrasound scan results are inconclusive, MRI provides a useful alternative to exclude or confirmthe suspicion of AA during pregnancy. MRI scans taken in pregnant women with the suspicion of appendicitis are 91% sensitive and 98% specific.[11,12]

The most common reasons to qualifypregnant women for cholecystectomy include acute cholecystitis (AC), acute pancreatitis (AP), choledocholithiasis, and recurrent cholecystic colic. In 90% of cases, AC develops due to urolithiasis.In the remaining 10% cases, AC is a consequence of cholestasis or frequent infection. Those are pregnancy-induced conditions, and include delayed gallbladder emptying, increased bile saturation with cholesterol (caused by elevated blood cholesterol), and a reduced pool of bile salts.[13,14] An increased risk of urolithiasis and cholecystitis were often found in women previously diagnosed with intrahepatic cholestasis of pregnancy.[15]

In pregnancy, AC may constitute >30% of overall indications for cholecystectomy.[16] Cholelithiasisoccurs in about 7% of primapara, and 19% of multipara.[17] Some of them develop symptoms of cholelithiasis. Repetitive biliary colic was the most common indication for cholecystectomy in pregnant women.[18] Conservative management is not always effectivein such cases. Recurrence was recorded in 70-80% of pregnant women. The risk of recurrence turned out to be dependent on the period of pregnancy when the onset of first symptoms occurred, i.e. 92%, 64%, and 44% in the first, second and third trimester of pregnancy, respectively.[19,20,21] According to the literature data, the percentage of pregnant patients conservatively treated for cholelithiasis was13-35%[16,19], and25-50% of pregnant women with cholelithiasis treated conservativelyrequired surgery.[16.22] The possibility of miscarriage is estimated at 16%[23,24]

Biliary cystic adenomas (BCAs) are rare benigntumorslikely to develop into malignant epithelial neoplasms. They account for < 5% of liver cyst-like lesions.[25] Most (80-85%) BCAs are located in the intrahepatic bile ducts.[26,27] Recently, some authors have reported that the actual incidence of BCA is unknown.[28] Actually, the occurrence of cystadenoma in the extrahepatic bile ducts and gallbladder is defined as very rare.[29, 30] A case report onconcomitant occurrence of intrahepatic and extrahepatic cystadenoma (in the common bile duct) described itas casuistic.[31]

The etiology of BCA is not always conclusive. Congenital BCA probably arisesduring disordered embryogenesis at the junction of the distal bile and pancreatic ducts. In contrast, acquired bile duct cysts can be a complication to acute pancreatitis. These lesions are thought to have formed from the ectopic remains of the primitive digestive tract, embryonic bile ducts, ovarian cells, or the intrahepatic perivascular glands.[32,33] The origin of gonadal cystadenoma is supported by immunohistochemical findings (presence in stromal cells, ovarian stroma (os), estrogen and progesterone receptors). Another evidence is the form of BCA phenotypically similar to gonads and detected in the adjacent organs during embryogenesis (between 5 and 8 hbd), i.e. the liver, pancreas, spleen.[34-36] BCA is diagnosed in app.90% of women.[37-40] Higher incidence of BCA in females is explained by different histological structure of the cyst walls.Pathomorphologically, BCA is an enlargement rather than a cyst (a cyst by definition, is a closed space lined with the epithelium) of the intrahepatic and / or extrahepatic bile ducts. In this case, there is a layer of undifferentiated mesenchymeal cells with the ovarian-like stroma in the 3-layer cyst wall beneath the epithelial lining.[41] The presence of ovarian stroma in the cyst wall is associated with its more frequent detection in women on hormonal therapy and during pregnancy[42,43]

According to Todani et al. bile duct cysts are classified into 5 types. Extrahepatic biliary cysts, defined as rare, are classified as type IVb.[44] The clinical picture of BCA varies. For many patients, the condition is asymptomatic at the time of diagnosis. Non-specific symptoms observed are pain (90%), abdominal bloating, nausea and vomiting, satiety, weight loss.[40,42] Cholestatic jaundice is mentioned as the most common symptom in patients with extrahepatic cystadenoma.[45] Cyst rupture and hemorrhage are very rare complications to extrahepatic cysts. These complications may occur during pregnancy and childbirth as they tend to exacerbate during this period.[46]

In addition to preoperative clinical diagnosis and laboratory tests, imaging diagnostics is decisive.Large extrahepatic biliary cysts may be available for palpation through the abdominal wall. Some authors think that imaging diagnosticsfacilitates preoperative diagnosis ofBCA suspicion in up to 89% of cases, and in the case of pregnancy, ultrasound and MRI are used.[28,47] In BCAs, the treatment of bile duct lesions involves complete surgical removal whenever it is feasible. Surgical treatment is indicated even if the patient has no typical symptoms.[48] It is necessary because of the risk of malignant transformation of BCA into BCAC (up to 20%). A high recurrence rate has also been reported after restricted surgery or partial resection.[28,34,38,45,49]

The incidence of acute pancreatitis in pregnancy is app. 1 in 1,000 to 1 in 10,000 pregnancies.[50] In over 50% of cases, the condition is diagnosed in the third trimester, and is more common with progressive gestational age.[51 APwas associated with cholelithiasis more often (in >65% cases) in pregnant women compared with the

general population, while hyperlipidemia and alcohol abuse are less common causes.[52, 53] Chinese authors evaluated the incidence, causes, clinical features, and treatment outcomesin 36 women with acute pancreatitis in pregnancy (APIP), delivery, and the puerperium. In the second and third trimesters of pregnancyit occurred in 25% and 61%, respectively.

The reported main causes of AP are hypertriglyceridemia (39%) and bile duct diseases (19%). Severe acute pancreatitis was significantly more common in patients with hypertriglyceridemia than without hypertriglyceridemia, respectively 79 and 27%.[54] Geng Wet al2011[55] found the cause of APIP was unclear in 11.1% of patients, and in this group idiopathic APIP was diagnosed. Other authors believe the majority of this group may comprise cases of unrecognized biliary microlithiasis.[56]

The diagnosis of acute pancreatitis (AP) in pregnant women is based on the clinical picture and the results of laboratory and imaging tests. The most common symptoms of pancreatic disease are abdominal pain, nausea, vomiting, and loss of appetite.[14] Atlanta classification by Banks PA et al2012[57], of AP and its complications distinguishes three clinical grades of severity: mild, moderately severe, and severe. Diagnosis of the severity is clinical, on the basis of local or general complications. On the other hand, in the diagnosis of transient or permanent organ failure - cardiovascular, respiratory, and kidney - a modified Marshall scale is used.[58] However, the diagnosis of severe AP bases on the Ranson scale (criteria). The assessment is performed twice (considering different criteria) immediately after admission to hospital and after 48 hours.[59]

MATERIAL AND METHODS

In the period from 2012 to 2017 in the Gynecology and Obstetrics Department of R. Sz. S. in Radom 10,160 deliveries took place, including 6,196 (61.0%) vaginal deliveries, and 3,964 (39.0%) by Caesarean section (cc). A 6-year retrospective analysis covered medical records of 11 women who underwent surgery during pregnancy due to surgical emergencies. In these patients, the following were assessed: age of patients, gestational age at the time of treatment, treatment mode, histopathological results of the material obtained during surgery, and obstetric results.

RESULTS

Acute surgical conditions (11 cases) constituted 0.108% among the overall number of 10,160 deliveries in our department.The surgical emergencies included appendicitis 7 cases (63.6%, the most frequent), cholecystitis 2 (18.2%) cases, giant cyst of extrahepatic biliary tract 1 (9.1%) case, and rupture of the pancreatic tail cyst complicated by acute pancreatitis and peritonitis 1 (9.1%) case.

Laparotomy and laparoscopy were performed in 10 and 1 woman respectively.

The women were operated on in particular trimesters of pregnancy: 1 (9.1%), 4 (36.4%), and 6 (54.5%) women, respectively in the first, second and third trimesters of pregnancy. The list of operations in each trimester of pregnancy is presented in Table 2.In three (27.3%) cases, surgical operations during pregnancy were complicated by peritonitis, twice by AA, and in one by severe form of AP (Table 1). Reoperation was necessary in one case after appendectomy (Table 1). In the examined group, 3 (27.3%) women after surgery foracute appendicitisdelivered naturally. In the group of 8 (72.7%) patients who delivered by cc, three deliveries (27.3%) were performed concomitantly with the surgery (Table 1-2).Pretermdeliveries were reported in 2 (18.2%) cases (Table2).

Table 2 presents information on the obstetric history of patients, gestational age during surgery and on the day of delivery, and assessment of the newborn's conditionby Apgar score. Regardless of the trimester of pregnancy in which operations were carried out, none of the 11 babies experienced a delay in psychophysical developmentwithin 7 months to 2 years. This information was obtained during interviews with parents, and outpatient medical records.

Among the pregnancies terminated by delivery, there were 7 cases of AA(0.069%).The diagnosis of AA was confirmed in all 7 cases (Table 1).The most decisive of laboratory testswas increased WBC mean $18.3x10^3$ / µl (range 13.3 - $21.8x10^3$ / µl. SD $3.1x10^3$ / µl).In subsequent trimesters of pregnancy, AA was noted in the first trimester 1 (14.2%) case, and inthe secondand third 3 (42.9%) cases in each.

Reoperation was necessary in one woman on 7^{th}day after appendectomy. Inter-loop abscesses were evacuated. However, on 14^{th}day from the primary surgery, secondary suturing of the postoperative wound was performed (Table 1).In one case (12.8%), 4 days after appendectomy (laparoscopic approach – LA) it was necessary to terminate the pregnancy by cc on 30^{th}hbddue to the intrauterine infection of the fetus. In twocases (28.6%) AA was complicated by peritonitis.

Cholecystectomy is the second to appendectomy most frequently performed surgery in pregnant women.[60] In our material, there were two cases of cholecystectomy (0.019%)among the pregnancies completed by childbirth. Emergent cholecystectomy in pregnancy is required due to urolithiasis and cholecystitis (Table 1). Details of the occurrence of these conditions, the impact on the course and mode of pregnancy termination, and the condition of newborns after delivery are presented in the tables 1 -2.

The article presents two rare surgical conditions in pregnancy recorded in our ward in the discussed period.

1. Cyst of extrahepatic bile ducts
2. Rupture of the pancreatic tail cyst with comorbid severe form of acute pancreatitis.

Case 1

The patient, GG, a 31-year-old pregnant woman,was referred by the District Hospital and admitted on 18 November, 2013. Referral notes: II Pregnancy, 17 hbd, history of caesarean section (4 years ago), presented with epigastric tumor.

On admission: body weight 92 kg, BMI 33.8. The patient presented with pain in the right epigastric region lasting 4 weeks. The pain in the area exacerbated two days prior to admission, she did not vomit.

Physical examination revealed the fundal height P/2, uterine tone normal, the size of the fetus corresponding to 17 hbd. Palpation found an epigastric tumor on the right side reaching X/N (half the distance between the xiphoid process and the navel), painless on palpation.

Abdominal ultrasound examination findings: Pregnant uterus. In the right epigastrium, enlargedfluid-filled encapsulated tank with septa, at least 25mm in diameter. Inside, several hyperechoic structures not related to its wall (they moved with the change of the patient's body position). There were no reflections within the tank that could correspond to gas. Non-characteristic picture – possibly suggesting a large amount of fluid content in the colon orthe stomach. Further diagnostics was recommended (MRIexaminationconsidered).

The tank pressed upon the liver displacing it to the left of the midline. The liver adhered tightly to the spleen. The liver tests wereunremarkable. The gallbladder without deposits. The intrahepatic bile ducts unwidened. The pancreas unavailable for examination. The spleen enlarged, 17- cm long with normalechostructure. The kidneys with normal echostructure. The bladder empty.

Laboratory tests found: anemia and proteinuria.

Blood tests: WBC 9.6 K / ul, RBC 3.79 M / ul, HGB 11.1 g / dl, HCT 32.0%, PLT 149 K / ul.

Urine test: Proteinuria 1233.4 mg / dl, and leukocytes 30-40-50hpf, fresh erythrocytes 2-4-6hpf, vitreouscasts 0-1hpf, fine-granularcasts 0-1-2 hpf. Liverfunction test normal, bilirubin normal.

Surgical consultation. The right epigastrium and mesogastriumoccupied by a large mass, painless and movable. Considering ultrasound examination results,the clinical picture could correspond to a pedunculated adnexal cyst, likelyoriginating from the pancreas. It was recommended to test urine diastase. After exclusion of the cyst originating from the appendages as the cause of the problem, the patient was transferred to the Department of General Surgery.

The patient was consultedby a specialist in infectious diseases.

On abdominal ultrasound scan the massof a cyst type, uncommunicating with the liver. Due to proteinuria the patient was consulted by a nephrologist and it was decided to extend imaging diagnostics and to test for tumormarkers: AFP, CEA, Ca 19-9, and Ca 125.

To complete the preoperative diagnostics medical consultants ordered MRI scheduled on 20 November, 2013. However, as the symptoms exacerbated it was decided to perform surgery.

Course of surgery. The operation was performed in the Gynecology and Obstetrics Ward by a team of surgeons (including a specialist in vascular surgery) and gynecologists.

The abdominal cavity was opened from the upper right medial incision. A disk-shaped cyst ca. 40-mm-thick was found, adhering closely to the right lobe of the liver. The lesion corresponding to the extrahepatic bile duct cystadenomawas separated (not without difficulty) from the liver surface. During cyst removal, iatrogenic incidental damage to the portal vein occurred. The damage was repaired by a specialist in vascular surgery.

A Redon drain was inserted into the lodgeleft by the cyst. The patient was transfused intra- and perioperatively 4U of packed red blood cells and 2U of plasma. On the second day after the surgery, the patient was transferred to the General Surgery Department for further treatment. Until the 20thday after surgery,bile-streaked serosa was draining through the Redon systemfrom the subhepatic space.

The patient was discharged on 22nd day after the operation with the pregnancy intact.

Histopathological results

- liver cyst fluid - Protein fluid with traces of lymphocytes,
- hepatic cyst - *Cystadenoma biliarehepatis.* Thick-walled multi-chamber cyst, 150x130x50mm, partially filled with brown clots, the mucosa wassmooth.

Childbirth by cc took place on April 20, 2014 (after completed 38 hbd). The newborn, male, body weight 4130g, length 57 cm, Apgar score 10.

Case 2

The pregnant woman, 33 hbd, first pregnancy was admitted on June 19, 2017,presenting with pain in the epigastric region and the left hypochondrium.

Laboratory tests of 19.06.2017. WBC 18.9x10^3 / µl, RBC 3.64x10^6 / µl, Hb 11.7 g / dl, HCT 32.4%, d-dimers 5418.0 ng / ml, Plt170x10^3 / µl, fibrinogen 4.95 g / dl, CRP 0.70 mg / dl, total protein 6.30 g / dl.

Urine test. Protein 25.00 mg / dl, leukocytes 3-4 hpf, fresh single erythrocytes in hpf. Urine culture. Asymptomatic bacteriuria (2 types of Gram (+) granulosa). Vaginal and cervical swabs identified *Enterococcus faecalis*.

The next day, abdominal pains dramatically exacerbated. Abdominal ultrasound examination detected likely presence of a deposit in the right renal calyx, with slight urinary retention in the pyelocaliceal system. Free fluid in the hepatorenal recess and around the spleen was also detected.

Laboratory tests. WBC 22.39x10^3 / µl, CRP 8.10 mg / dl, blood serum amylase 14 U / l (normal range 28-100), urine amylase 20754 U / l (norm <460).

Based on the clinical picture, laboratory test results, and diagnostic imaging ordered bythe surgical consultant, the diagnosis of acute pancreatitis and peritonitis was made, and the patient was qualified for cc and prompt surgery.

The course of surgery. Laparotomy was performed by the lower median incision between the pubic symphysis and the navel which exposed app. 1,000-1,500 ml of yellowish fluid in the peritoneal cavity. The sample was taken for bacteriological examination. On the larger omentum, numerous foci of Balcer's necrosis (‘soap’) and a significant amount of fibrin on the peritoneal wall and mesentery were seen. The vesico-uterine recess was incised and the bladder slid downwards. The uterine muscle was cut transversely in the lower section. A female fetus was brought forth by the head from the clean amniotic fluid. The newborn was in severe condition: Apgar score 2/5, body weight 1920 g, length 44 cm. After the separation of the umbilical cord, the patient was administered 5 U of oxytocin to the uterine muscle. The entireafterbirth was removed manually. The uterine cavity was instrumentally checked. The wound angles were secured with two single sutures on both sides. The uterine muscle was sutured with a continuous two-layer suture. Additional hemostatic sutures wereinserted to the uterine muscle and Pabal administered intravenously. Adnexalcondition was normal. Hemostasis was normal. The surgeon found edema and inflammation of the pancreas caused by the ruptured pancreatic tail cyst. Theabdominal organs were examined for hemostasis. Double peritoneal lavage was performed. Two safety drains were inserted into the area of the pancreatic tail cyst and Douglas Bay. The abdominal cavity was closed by single stitches and the wound sterile-dressed. Urine outflow by Foley catheter clean. The uterus shrinking was uneventful.

Postoperative material. The placenta was sent for histopathological examination. Theperitoneal fluid was cultured for aerobic and anaerobic bacteria. Antibiotic sensitivity test was done.

The patient was transferred to the General Surgery Ward where she stayed until July 7, 2017.

Laboratory tests detected the following
26 June, 2017WBC 16.2x10^3 / µl, CRP: 13.40 mg / dl,
30June, 2017 WBC 18.6x10^3 / µl,
3 July, 2017 CRP 5.40 mg / dl, Lipase 209 U / L
6July, 2017 CRP 2.20 mg / dl, Lipase 336 U / L, Blood serum amylase341 U / l.
7 July, 2017WBC: 8.1x10^3 / µl, RBC 3.06x10^6 / µl, Hb 9.3 g / dl, HCT: 29.3%, Plt 479x103 / µl, Fibrinogen 4.95 g / dl, CRP 1.70 mg/ dl, Lipase 125 U / L, Alkaline phosphatase 663 U / l (normal range 35-104).

Chest x-ray of 27 June, 2017 revealed fluid on the left of pleural cavity reaching the anterior segment of VI rib, and a trace of fluid in the right diaphragmatic recess. Repeated x-ray of5 July, 2017 found fluid in the leftcostodiaphragmatic recess, and a trace fluid in the right costodiaphragmatic recess, CT scan of 29 June, 2017 revealed a slight enlargement of the pancreas (swollen?) with discreet, segmentally blurred margins, enhanced quite homogenously with the contrast, surrounded by a liquid, without any visible encapsulation. In addition, three deposits in the lower calyx of the right kidney were detected, the largest 10 mm.

After conservative treatment and stabilization of the general condition, the patient was discharged from the Department of General Surgery with the diagnosis of severe acute pancreatitis, rupture of the pancreatic tail cyst, and diffuse peritonitis. She was in the third trimester of her first pregnancy, completed by caesarean section. The hospitalization in the Gynecology and Obstetrics Department continued until 11 July, 2017.

Currently, the patient does not report any complaints related to the past disease. The baby’sdevelopment isuneventful.

RESULTS AND DISCUSSION

In the literature, suspicion of acute appendicitis and qualification for surgery is estimated as 1/600 to 1/1000 pregnancies. However, the diagnosis is confirmed in 1/1800 to 1/1500 pregnancies.[3,61,62] Some authors, reported the occurrence of AA and surgery during pregnancy as 1 in 635 to 1 in 500 pregnancies per year (0.04% to 0.20%).[63.64]

In the presented material, the frequency of surgical procedures was 0.07%, and 1/1451 recorded surgeries for appendicitis were within the range presented in the literature.

In 7 cases discussed above, all operations were carried out within 24 hours after the admission to the Department. In one case, hospitalization was delayed for two days due to the reasons on the patient's side, which resulted in perioperative complications, i.e. peritonitis and puerperal sepsis. Such complications associated with the delayed implementation of surgical treatment, also due to conservative treatment,are mentioned in the literature.[65,66] It was found that the greater the delay in surgical treatment, the greater the risk of maternal and fetal morbidity.[67] In the literature, the incidence of AA in subsequent trimesters is defined within quite wide limits: 1st 24.5-47%; 2nd 40-51%; and 3rd 4.8-24.5.[2,3,4] AA is more common in 2ndtrimester of pregnancy compared to1st or 3rdtrimester.[63,68] Ueberrueck T et al 2004[2], Jenkins TMet al2003[69], and Choi JJet al 2011[64] noted the following AA frequency respectively.

- in the second trimester of pregnancy 53% and 48.3%,
- in the first trimester 23% and 26.9%,
- in the third trimester 24% - 27.6%.

Some authors reported higher frequency ofAA in women during the third trimester of pregnancy. They believe that this is due to the reluctance of a pregnant woman to undergo surgery during pregnancy, which delays diagnosis and prompt treatment.[65] In the presented material, AA was recorded in the trimester 1st 14.2%, 2nd42.9%, and 3rd 42.9% (Table 1).Among 7 pregnant women operated on for AA, 2 (28.6%) developed peritonitis. In the first of thecasesdiscussed above, appendectomy was performed in 37hbd of the first pregnancy, concomitantly to caesarean section. The operation was carried out within 24 hours on the next day after the admission. Two days earlier, despite the symptoms indicating AA, the patient did not consent to hospitalization. In the secondcase, however, there was a need for reoperation after appendectomy in 18hbd of the third pregnancy (Table 1).

In another case, a woman in the 29 hbd of the fourth pregnancy was reoperatedon the 3rd day after laparoscopic appendectomy for intrauterine infection of the fetus and ongoing preterm delivery terminated by caesarean section (Table 1).

Such complications are also mentioned in the literature.[70]

Acute cholecystitis and cholelithiasis are among the most common causes of cholecystectomy in pregnancy. These conditions were the reason of surgery in two cases presented in this article (Table 1). Their occurrence in pregnancy increased the risk of premature delivery, maternal and neonatal morbidity.[71] We did not record such complications (Table 1-2).

Cystadenomas of the bile ducts are often located intrahepatically.[72] Extrahepatic biliary cystadenoma is a benign but potentially malignant lesion. Fewer than 100 cases were reported in the literature[73] The occurrence of cystic adenomas of the extrahepatic bile ducts is estimated at < 10% and is described as very rare.[29,30,72] Extrahepatic biliary cystadenoma is found mainly (96.3%) among women. The most common site is the common hepatic duct (32%).[72] The unusual increase in extrahepatic biliary cystadenoma in pregnancy is often related to the use contraceptives and the female sex hormone receptors in this organ.[38] It was also recorded in our material.

The most frequently observed symptoms include obstructive jaundice (85%) and abdominal pain (50%).[72,73] In the above case, extrahepatic biliary cystadenoma was adjacent to the right hepatic lobe. However, jaundice did not occur, though mentioned in the literature as the most common clinical symptom of this condition. Until 20th day after the surgery, bile-streaked serosa was Redon-drained from the subhepatic space.

Anatomical structural variations and lesions in the extrahepatic bile ducts pose a great challenge for the operator. Despite careful surgery within the Calot triangle, portal vein damage could not have been avoided. Asit was a major surgery and no absolute indications existed, cholecystectomy was abandoned. The patient was followed up and recent abdominal CTof 2 January, 2020 found no complications related to the operation performed six years ago.

The review of available English-language literature found no reports on the occurrence and surgical management of extrahepatic biliary cystadenoma in pregnancy. Surgical procedures for pancreatic diseases are among the most difficult in abdominal surgery.[74] Maternal and fetal deaths due to acute pancreatitisare recorded.[75] In acute pancreatitis, not only fetal death is a threat, but also preterm delivery and prematurity. Termination of pregnancy may be an optional modality for the treatment of acute pancreatitis in pregnancy.[76] Treatment of AP may also be conservative. However, surgical cause-dependent treatment must be considered in patients in whom conservative treatment is ineffective. In the case of rupture of the pancreatic cyst and severe peritonitis, drainage of the peritoneal cavity (around the ruptured cyst) is minimally invasive.[14] Surgical treatment (external or internal drainage of the cyst and pancreatic resection) can be performed by laparotomy or laparoscopy.[77-82] APis an indication to terminate pregnancy, and caesarean section is preferable then.[83]

In the treatment of acute pancreatitis, especially at an early stage, fluid resuscitation and enteral nutrition are essential.In the presented case, it was a severe form AP therefore termination of pregnancy by cc was necessary. Caesarean section and drainage of the ruptured cyst around the pancreatic tail were implemented. Both are recommended in the literature as decisive for the successful continuation of pregnancy and the neonate's health.

Hernandez A. et al. 2007[84], examined 21 pregnant patients with AP and recorded four premature deliveries and one miscarriage. However, they did not observe the deaths of mothers.Tang SJ et al. 2010[85] reported 0.97% maternal mortality among pregnant women. The 20th-century publications reported on mortality in pregnant women with severe APin up to 15% cases.[86, 87] In our material the reported incidence of AP (one case) in pregnancy and the gestational age (third trimester) were similar to those reported in the literature.[50, 51] In the case discussed above, termination of pregnancy by cc was indispensable caused by severity of the patient's condition.

Table 1: Surgeries for emergency surgical conditions in pregnancy (N=11).

1.- Sequence of surgeries 2.- Date of surgery 3.-Hbd on the day of surgery Hbd on the day of delivery: 4.- vaginal delivery, 5.- Caesarean section [cc].					**Appendicitis. Inter-loop abscess. Peritonitis**			**Cholecystitis**	**Extrahepatic biliary cystadenoma**	**Acute pancreatitis. Pancreatic tail cyst rupture. Peritonitis**	**Clinical and histopathological diagnosis**
					L-tomy Right lower paramedian incision	**L-tomy Transverse incision**	**L-scopy**	**Laparotomy Kocher incision**	**Laparotomy Right upper paramedian incision**	**Laparotomy Midline incision**	
1.	**2.**	**3.**	**4.**	**5.**	**Appendectomy**			**Cholecystectomy. Drainage**	**Cystadnectomy. Suture of damaged portal vein. Drainage**	**Drainage of ruptured cyst lodge. Drainage of peritoneal cavity**	
1	22. 03./2012	37		37		X					*Appendicitis gangraenosa*
2	26. 08./2013	37		37				X			*Cholecystitis ulceroso-phlegmonosa p. gangraenosa*
3	20. 11./2013	17		39					X[x]		*Cystadenoma biliare hepatis*
4	10. 04./2015	20	38		X						*Appendicitis chronica*
5	30. 06./2015	28		39	X						*Appendicitias gangraenos*
6	02. 09./2015	28		37				X			*Cholecystitis chronic exacerbata*
7	26. 10./2016	29		30			X				*Appendicitis acuta ulcerosa*
8	30. 03./2017	7		38	X						*Appendicitis chronica*
9	20.06./2017	33		33						X	*Rupture of the pancreatic tail cyst*
10	27.06./2017	18	41		X[xx]						*Appendicitis ulceroso-phlegmono*
11	05. 12./2017	23	37		X						*Appendicitis ulceroso-phlegmono*

X[x] - Laparotomy: right upper paramedial incision
X [xx] – Relaparotomy: lower midian incision

Table 2. Surgeries in pregnancy. Obstetric history. Gestational age on the day of surgery and delivery. Neonate's condition (N=11).

No	Date of surgery	Patient's age	Parity	Caesarean section	Miscarriage	Trimester. Gestational age on the day of surgery Hbd	Delivery. Gestational age on the day of delivery Hbd	Gender	Neonate's body weight	Apgar score
1.	22.03./12	26	0	0	0	I -37	I -37	F	3900	10
2.	26.08./13	26	0	0	0	I -37	I -37	M	2500	10
3.	20.11/13	31	1	1	0	II - 17	II - 39	M	4130	10
4.	10.04/15	31	2	0	0	III - 20	III - 38	F	2430	10
5.	30.06/15	33	0	0	1	II - 28	I - 39	F	3350	10
6.	02.09/15	29	0	0	2	III - 28	I - 37	F	3300	10
7.	26.10/16	27	3	0	0	IV - 29	IV -30	M	1740	8
8.	30.03/17	41	2	0	0	III - 07	III - 38	M	3400	10
9.	20.06/17	20	0	0	0	I - 33	I - 33	F	1920	2
10.	27.06/17	30	1	0	1	III - 18	II - 41	F	3430	10
11.	05.12/17	25	1	0	0	II - 23	II - 37	M	3380	10

Table 3. Patients' age, gestational age on the day of surgery and delivery. Neonate's body weight (N=11).

Parameter	Mean	Range	SD
Age	29	41 - 20	5.37
Gestational age on the day of surgery	25.18	37 - 7	9.23
Gestational age on the day of delivery	36.91	41 - 30	3.02

CONCLUSIONS

The diagnosis and treatment of acute abdominal conditions diagnosed in pregnancy should include the followingprocedings.

- patient's detailed medical history,
- a thorough examination of the pregnant woman,
- a thorough obstetric assessment
- basic and, if necessary, extended laboratory diagnostics,
- only necessary diagnostic imaging.

Diagnostic investigations should significantly facilitate the diagnosis and not delay the implementation of prompt treatment.The suggested proceedings should aim at preserving the health and life of the mother and the fetus and should be guided by judicial medical thinking based on clinical experience and common sense. In such specific conditions, the type of surgery performed depends on the surgeon's preferences and experience.

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