

Case report

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Preterm Prelabour Rupture of Membranes and Vernix Caseosa Peritonitis: A Case Report

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SUMMARY

Introduction: Vernix caseosa peritonitis (VCP) is a rare postpartum complication rarely recognized by clinicians despite an increased incidence of cesarean sections.

Case report: A 23-year-old patient, who had not had any prior medical examination during pregnancy and with preterm premature rupture of the membranes (PPROM), gave birth by cesarean section. On the 4th day after the cesarean section, the patient developed fever and abdominal pain. Laboratory parameters showing inflammation were increased. Two bacteria were found in the swab of the lochia: *Escherichia coli* and *Proteus mirabilis*. After relaparotomy was performed inside the abdominal cavity, hemorrhagic-purulent contents and multiple cheesy fibrin deposits on the intestines were visible. After peritoneal lavage and triple antibiotic therapy prescribed in duration of seven days, the patient completely recovered. Histopathological analysis showed a mixture of inflammatory cells concentrated around non-nuclear cells with wrinkled edges, which primarily corresponded to the inflammatory-altered vernix.

Conclusion: Early recognition of vernix caseosa peritonitis is very important because it can prevent the resection of the abdominal organs that are altered with inflammation.

Keywords: vernix caseosa peritonitis, postpartum complications, PPRM

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INTRODUCTION

Preterm prelabour rupture of membranes (PPROM) is defined as a rupture of membranes before the onset of labor and before 37 weeks of gestation (1).

Vernix caseosa peritonitis (VCP) is a rare postpartum complication (only 30 cases have been described in the available literature - PubMed search: Vernix caseosa peritonitis) that clinicians rarely recognize despite an increased incidence of cesarean sections (2). The typical presentation of VCP includes severe abdominal pain, pyrexia and peritonism after cesarean section (3). This entity was noted as a complication even with vaginal delivery in two published cases (2, 4). The pathogenesis of VCP has not been sufficiently elucidated but it is known to occur as a response to the abnormal presence of even a small amount of amniotic fluid containing vernix (5). The definitive diagnosis of this postpartum complication is made according to the histopathological findings, but in the initial phase of the disease, radiological methods such as ultrasound can be of great help in the differential diagnosis (6).

CASE REPORT

A 23-year-old patient, a foreign citizen, with a rupture of the amniotic membrane and dilatation of the cervix of about 2 cm, was admitted to the Department of Gynecology of Clinical Center in Kosov-

ska Mitrovica. In spite of the fact that the patient did not provide adequate anamnestic data on the last menstruation cycle, it was estimated that the patient was approximately in the 36th week of pregnancy, stating that the pregnancy had not been monitored by ultrasound and microbiological testing. The patient denied any chronic and hereditary diseases. General condition of this female patient was as follows: BMI -26, blood pressure - 115/70, pulse - 65, temperature - 36.5 degrees C, smoker, without edema, varicose veins, eupneic. Patient denied chronic diseases and allergies and was in good general condition.

Laboratory analysis was within the reference values. CTG record was reactive, fetal heart rate was about 130. On admission, therapy with erythromycin was prescribed and vaginal swab was taken. Ultrasound finding corresponded to the 35th week of gestation. After 24 hours of stimulation with synthetic oxytocin and antispasmodics, the dilatation of cervix failed to progress and so the fetal heart rate decreased (fetal bradycardia about 100). Therefore, we decided to perform a caesarean section. The newborn (female, 2710 g, 53 cm long, head circumference 33) had an App score 7/8. Amniotic fluid was clear. In the postoperative course, the patient was treated with dual antibiotic therapy (Gentamicin and Clindamycin). Two days later, the result of the vaginal swab arrived, showing the presence of *Escherichia coli* and *Proteus mirabilis*, which are sensitive to the already prescribed postoperative antibiotic therapy.



Figure 1. Ultrasound picture of subfascial fluid collection

The patient developed fever on the 4th day after the caesarean section (up to 39^o C) and intense productive cough. Due to the ongoing covid-19 epidemic, the patient was tested for covid-19. The patient believed that her cough was caused by cigarette use. The result of a test for covid-19 was negative. Laboratory analyzes showed the presence of leukocytosis - 32.1 with granulocytosis - 91.8, CRP - 305, procalcitonin - 4.28. The lochia swab contained the same bacteria as on the admission. A third antibiotic was included into the therapy according to the antibiogram (Metronidazole). Hyperthermia persisted and abdominal pain occurred. Clinically, the abdomen was hard, tense with the presence of muscular defenses. Small pelvis ultrasounds showed a sub-fascial collection in the diameter of about 58 x 23 mm, which may differentially correspond to hematoma or thickened omentum (Figure 1). For all the above reasons, we opted for extended relaparotomy median inferior. Liquid and hemorrhagic-purulent contents in the abdominal cavity (sent for microbiological examination) were found when the abdomen was cut open, with the omentum firmly attached to the uterus, edematous and very bloody, which was the reason for partial omentectomy. The incision site was intact. Multiple cheesy fibrin deposits found on the intestines and in the abdominal cavity were easily removed and were sent to histopathological examination. Peritoneal lavage was performed.

After the surgery, the patient was admitted to the intensive care unit, under 24-hour monitoring system of the vital parameters, on triple antibiotic therapy (Amikacin, Ceftriaxone, Ciprofloxacin). Parameters of the inflammation were monitored on a daily basis showing a decrease in the laboratory values. The microbiological finding of the abdominal cavity fluid showed the presence of *Escherichia coli* and *Proteus mirabilis* in large numbers (as in the vaginal swab).

The patient was discharged 7 days after the surgery, with a healthy baby, feeling well. The baby was prescribed dual antibiotic therapy for seven days (Gentamicin and Ampicillin) because of leukocytosis.

DISCUSSION

The major risk factors for the existence of PPROM among other things include: history of PPROM, nutritional deficiencies, low body mass index, low socioeconomic status, cigarette smoking

and infection. The patient described in this case report had a lot of these risk factors (1). The risk of gaining intra-amniotic infection was too high (positive vaginal swabs, PPROM, prolonged labor, multiple digital exams > 3, nulliparity and cigarettes use are more likely to require oxytocin augmentation) (7).

The spillage of amniotic fluid into peritoneal cavity due to cesarean section is almost inevitable, and it is usually insignificant in terms of symptomatology and it may rarely progress to peritonitis. The exact mechanism leading to the development of such a complication is unknown (2). It is believed that the presence of two bacteria in vaginal swab could have caused an infection of the amniotic fluid and consequently lead to peritoneum, which was proved by the positive culture of fluid in the abdominal cavity. Positive abdominal fluid culture was found in only two of the published cases. (5) All published cases described pregnant women in 37 - 43 gestational week (2 - 6), except for one case described by Schwartz et al. in 1985, where gestational age was 35 weeks as in the case described in this paper. This was the first case where a culture of the lochia was reported to be positive.

The first symptoms in the case of the patient mentioned above appeared four days after cesarean section. In the available literature, it is stated that the longest period from the cesarean section to the onset of symptoms was 5 weeks and the shortest period was 3 hours (2, 5). In six published cases, authors found peritoneal nodules, which was not found in the patient admitted to the Department of Gynecology of Clinical Center in Kosovska Mitrovica (2 - 3, 6). In 18 patients, described in the available literature, appendectomy, cholecystectomy and hysterectomy were performed and in three cases colon resection was performed (5). In this case, partial omentectomy was performed as was described by George et al. in 1995. Histopathological findings were similar in all cases, including the one described in this paper, and they all pointed to the presence of anucleate squamous cells and foreign body giant cell reaction (2 - 6). After laparotomy or laparoscopy with antibiotic therapy prescribed, all of the patients completely recovered, excluding two cases where one patient developed recurrent vernix caseosa abscesses, whereas in the other patient, abdominal pain appeared 7 months later (3).

An overview of 30 published cases with clinical and histopathological details is given in Table 1.

Table 1. Characteristics of 30 reported cases of VCP [5,8], modified and supplemented with new data

Author	Year	Cases	Age	Parity	GW and reason for SC	Day after SC	Abdo p/t	Fever	Imaging	Microbiology	CE	FC	Other procedures	FBGC	ASC
Krumerman and Pouliot	1976	1	26	2	NI	28	Yes	No	No	PFC negative	Yes	No	Appendicetomy	Yes	Yes
Herz et al.	1982	2	21	1	NI	8	Yes	No	USS	NR PFC- PV	Yes	No	None Hysterectomy	Yes	Yes
			21	0		14	Yes	Yes	No		NI	No		Yes	Yes
Freedman et al.	1982	2	21	2	NI	9	Yes	Yes	No	NR	Yes	No	Appendicetomy Hysterectomy	Yes	Yes
			20	1		17 h	Yes	Yes	No		Yes	No		Yes	Yes
Schwartz et al.	1985	1	31	0	33w/BP	NI	Yes	No	USS	NI	NI	NI	Appendicetomy	Yes	Yes
Boothby et al.	1985	1	17	0	NI	35	Yes	Yes	USS	NR	Yes	No	Colon resection	Yes	Yes
George et al.	1995	2	32	NI	NI/FM	8	Yes	No	USS	NI NI	Yes	No	Colon resection Appendicetomy and omentectomy	No	Yes
			28	NI	NI/FM	10	Yes	Yes	CT		Yes	Yes		Yes	Yes
Zellers and Balaj	1996	2	17	NI	NI/BP NI/FD	12	Yes	Yes	CT	NI NI	Yes	No	Appendicetomy Appendicetomy	Yes	Yes
			23	NI		5	Yes	Yes	USS		Yes	Yes		Yes	Yes
Nunez et al.	1996	1	33	0	41w/FVD	3	Yes	Yes	USS	NI	Yes	Yes	Cecum and ascending colostomy	Yes	Yes
Mahmoud et al.	1997	1	25	1	NI	3	Yes	Yes	CT	NI	Yes	Yes	Appendicetomy	No	Yes
Wright at al.	1998	2	NI	NI	NI	NI	NI	NI	NI	NI	NI	NI	NI	NI	NI
Tawfik et al.	1998	1	28	3	NI	4	Yes	Yes	CT	Blood cult.neg.	Yes	Yes	None	Yes	Yes
Cumming et al.	2001	1	32	0	38w/FVD	6	Yes	Yes	USS	Gram-negative	Yes	No	Gallblader resection	Yes	Yes
Selo-Ojeme et al.	2007	1	32	0	NI	7	Yes	Yes	CT	Gram-positive	Yes	Yes	None	NR	NR
Stuart et al.	2009	3	40	0	39w/BP	5	Yes	Yes	CT/USS	NR UK- UK-	Yes	No	Omental biopsy Biopsy of the mass Two laparotomies	Yes	Yes
			32	1	39W/BP	3	Yes	Yes	CT/NMR		NI	Yes		Yes	NI
			43	NI	NI/FVD	4	Yes	Yes	CT		Yes	No		Yes	Yes
Wisanto et al.	2010	1	31	2	NI	3w	Yes	No	CT	NR	Yes	No	Biopsy of nodules	Yes	No
Myers et al.	2011	1	38	NI	NI/ESC	3w	No	No	CT/NMR	NR	No	No	Fine needle biopsy	Yes	Yes
Bailey et al.	2012	1	26	0	37w/BP	3	Yes	Yes	CT	NR	Yes	No	Biopsy of VC plaque	Yes	No
Chambers et al.	2012	1	30	0	40/fvd	5w	Yes	Yes	CT	NR	Yes	Yes	Appendicetomy	Yes	No
Sadath et al.	2013	1	28	2	41w/VD	3 h	Yes	Yes	USS/CT	NR	No	No	Biopsy of nodules	Yes	No
Val Bernal et al.	2015	2	33	1	43w/FVD	9	Yes	No	NR	NR NR	Yes	No	Appendicetomy Appendicetomy	Yes	Yes
			29	0	41W/BP	7	Yes	Yes	NR		Yes	No		Yes	Yes
Vieillefosse et al.	2018	1	38	0	NI/FM	5	Yes	Yes	CT/NMR	NR	No	Yes	Biopsy of skin and nodules	No	Yes
Cathelain et al	2019	1	30	2	41w/VD	5	Yes	Yes	CT	NR	Yes	Yes	Appendicetomy	Yes	No

CS - cesarean section; GW - gestation week; p/t - pain/tenderness; CE - cheesy exudate; FC - fluid collection; FBGC - foreign body giant cell reaction; ASC - anucleate squamous cell; NR - not requested; NI - no information; CT - computed tomography; USS - ultrasound scan; NMR - nuclear magnetic resonance, BP - breech presentation, FVD - failure vaginal delivery; VD - vaginal delivery; FM - fetal macrosomia; PFC - peritoneal fluid culture; FD - fetal distress. PM- Prevotella melaninogenica, UK- negative findings on urin culture, w- week, ESC- elective cesarean section

CONCLUSION

Based on this analysis, during pregnancy that has not been regularly monitored, some undetected pathogens in the vaginal environment and untreated infection can appear, which first causes premature rupture of fetal membranes with prolonged childbirth, probably leading to amniotic fluid infection. Infected amniotic fluid with the vernix spilled into the abdomen during the caesarean section caused the complications described above, which was confirmed by the positive finding of the peritoneal cavity fluid.

This health issue can significantly affect the health of the mother, so its early recognition is of a great importance. The presence of vernix can lead to extensive inflammation and resection of the abdominal organs due to non-recognition of this rare syndrome.

This clinical syndrome should be suspected with the appearance of fever, abdominal pain and suspicious collection of fluid in the abdomen seen during the ultrasound after cesarean section and vaginal birth.

After more detailed diagnostic procedures, it is necessary to perform an exploratory laparotomy or laparoscopy with drainage and lavage of the abdominal cavity, giving the prescribed antibiotic therapy for inflammation, which in this case showed health improvement and satisfying results.

In order to prevent premature rupture of membranes and other complications during and at the end of pregnancy, women should be educated and informed that regular microbiological testing and doctor's appointments are of great importance.

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Prevremeno prsnuće plodovih ovojaka i vernix caseosa peritonitis: prikaz slučaja

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SAŽETAK

Uvod. Vernix caseosa peritonitis (VCP) retka je postporođajna komplikacija, koju kliničari retko prepoznaju usprkos povećanoj incidenciji carskih rezova.

Prikaz slučaja. Dvadesettgodišnja bolesnica, čija trudnoća nije praćena kliničkim pregledima, sa prevremenim prsnućem plodovih ovojaka, porađa se carskim rezom. Četvrti dan nakon urađenog carskog reza, bolesnica razvija povišenu telesnu temperaturu, bol u stomaku i povišene laboratorijske parametre, koji ukazuju na zapaljenje. U brisu lohija nađene su dve bakterije: *Escherichia coli* i *Proteus mirabilis*. Izvršena je relaparotomija, te su pronađeni hemoragično-gnojni sadržaj u trbušnoj duplji i višestruke siraste naslage fibrina na crevima i u trbušnoj duplji. Histopatološka analiza pokazala je mešavinu inflamatornih ćelija koncentrisanih oko anuklearnih ćelija sa naboranim ivicama, što prvenstveno odgovara inflamatorno izmenjenom verniksu. Bolesnica se postoperativno, nakon doziranja trostruke antibiotske terapije u trajanju od sedam dana, oporavlja u potpunosti.

Zaključak. Rana dijagnostika VCP-a veoma je važna, jer se može sprečiti širenje infekcije na abdominalne organe, kao i sprečiti njihova resekcija.

Ključne reči: vernix caseosa peritonitis, postpartalne komplikacije, PPRM