Case Report

A Case of Spontaneous Hemoperitoneum in Labor.

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Abstract

Background

Intrapartum hemoperitoneum is a life-threatening condition for both mother and fetus, commonly associated with uterine rupture and abnormal placentation. Infrequently, according to the literature, the condition may result from spontaneous rupture of utero ovarian vessels or varices, rupture of feeding vessels in pregnancy-associated myomas, deciduous, disruption of endometriotic implants, spleen or liver rupture, rupture of aneurismatic vessels.

Spontaneous, unprovoked, hemoperitoneum (SHiP) is an extremely rarer complication, of unknown etiology, first described only at the beginning of the 20th century and amount, by the year 2017, to a total of slightly more than 200 cases.

Symptoms, if present, initially resemble other obstetric and abdominal conditions, and may be difficult to interpret since obvious causality is lacking: no delay in emergency surgery represents the life-saving approach.

Case presentation

We present the case of a primigravida, without evidence of antenatal risk factors, who was admitted in initial labor at a term of pregnancy and had excellent progression until full dilatation. Due to fetal bradycardia during the active 2nd stage and no progress despite attempted instrumental delivery, an emergency cesarean section was prepared. Upon entering the abdomen, an unexpected hemoperitoneum of 1000 MLS was observed. A systematic exploration of the uterus did not reveal any frank uterine rupture, as initially suspected, whilst apparent was the disruption of the serosa of the right posterolateral uterine wall, with obvious bleeding from the anastomotic arteriovenous branches of the uterine vessels.

Successful hemostasis was achieved using a continuous suture. Mother and fetus were both discharged in good health on due time.

Conclusion

Prolonged fetal bradycardia without obvious obstetrical reasons, probably from maternal hypovolemia, and a failed instrumental delivery, prompted a life-saving laparotomy.
Hemoperitoneum had developed from laceration of the serosa on the posterior uterine wall with bleeding from arteriovenous anastomotic vessels. Possible etiology and similar cases are discussed given the literature available.

Keywords:

Pregnancy complications, SHiP, hemoperitoneum, uterine rupture, uterine vessels, endometriosis.

Introduction

Up to the mid-fifties of the last century, SHiP was reported in not more than 75 cases **(**1**)** in the English literature. Challenges in differential diagnosis, encompassing relatively more common cases of uterine rupture, placenta abruption, liver subcapsular hemorrhage, splenic rupture, and the presence of oftentimes deceitful and rapidly evolving symptoms and delayed management, burdened the condition with a maternal mortality of around 50% **(**1**,**2**)**.

In the second half of the century, whilst maternal mortality dropped to 4%, thanks to improved knowledge and multidisciplinary approach, still, the rate of fetal demise accounted for 31 %, in nearly half cases from maternal hypovolemia and associated fetal distress. **(**3**,**4**)**

Case presentation

A 35 years old primigravida presented to the labor ward at a gestational age of 39 weeks +3 days with regular contractions, cervix 1 cm dilated and effaced, intact membranes, normal CTG. Her Hb was 11.6 on admission.

History

She had regularly attended routine antenatal care since the 17th week when she first came for consultation in the hospital with the complaint of abdominal cramps. Remarkable events in the history were anemia since childhood, a bowel polypectomy without reported complications, and recurrent abdominal pain.

At booking, her Hb was 10.8 g/dl, in line with the condition reported by the patient, of preexisting anemia. Later on, she also reported a delay of one year in achieving pregnancy, which had raised elsewhere the suspicion of possible endometriosis, although never undergoing further evaluation, since eventually falling pregnant spontaneously.

Antenatal follow-up was unremarkable: recurrent episodes of abdominal pain 'on the right side', especially in late pregnancy, attributed by the patient to fetal movements, remained unanswered, since unremarkable routine scans, near term NSTs and stable values of FBCs, although not responsive to therapy. An episode of hemorrhoidal thrombosis was successfully treated at 34 weeks without further complications.

In the ward

After receiving an epidural, excellent and fast progress in labor was observed and in 4 hours she spontaneously reached 8 cm dilatation, with the station of the head at the spines, and 'early decelerations' on the CTG.

The obstetrician was called and on arrival after 40 minutes, on vaginal examination, ascertained:
full dilatation, station of presenting part 0 /+1, visible at the outlet under a 'trial' of pushing, rotation still incomplete (>45^ Left Occiput Anterior), fingers of the right hand of the fetus palpable on the homolateral ear (compound presentation).

Deep early decelerations were still present at each contraction and the patient was complaining of unspecified abdominal pain: expedite delivery was deemed possible and the patient was positioned in a lithotomy position and requested to start actively pushing.

Soon afterward, whilst noticeably ineffective appeared the maternal efforts and her alertness to follow instructions, the onset of deep bradycardia prompted an emergency trial of instrumental (vacuum) delivery. After 3 failed attempts with the kiwi cup, due to no progress and persistent bradycardia, a Category 1 Cesarean section was prepared.

In Theatre

Upon entry into the abdomen, hemoperitoneum was found unexpectedly and almost 1000 ml of blood was suctioned under the initial suspicion of uterine rupture.

A live fetus in distress was extracted, with a deeply impacted head and compound presentation: Apgar score was initially 5, soon after airways suctioning, improving to 7 and 9 with a vigorous cry. Weight was 3500 gr. The uterus was exteriorized and explored: the source of bleeding was identified on the right border of the posterior wall, as a vertical laceration of the serosa covering the small anastomotic arteriovenous branches of uterine artery and vein, extended from the middle third of the uterine corpus down to the insertion of the uterosacral ligament.

Hemostatic uninterrupted suture with Vicryl 2-0 was successfully placed. After the exploration of the abdominal cavity confirmed the absence of additional bleeding sites, surgery was completed as per standard technique and the abdominal wall closed. Two units of blood were administered due to a Hb value of 7.9 g/dl post-surgically. The hospital course was unremarkable for both mother and baby: both were discharged in stable condition on day 5 th.Histopathology of the placenta showed increased syncytial knotting and fetal membranes with meconium-laden macrophages (correlated with fetal distress). Placenta weight :< 10centile for gestational age. Marginal insertion of the cord.

Discussion

Recognized causes of hemoperitoneum in pregnancy are those associated with rupture of previous uterine scar antenatally and intrapartum, morbidly adherent placentation in postpartum and much more rarely, liver or splenic rupture, rupture of pelvic or abdominal vessels or varicosities of the uterine surface **(**5**,**6**)**, rupture of superficial vessels feeding myomas, rupture of utero- ovarian vessels **(**1**,**2**,**7**,**8**,**9**,**10**,**11**)**. Spontaneous Hemoperitoneum in Pregnancy (SHIP) is a distinct, rare entity, which was first reported in 1909 **(**12**)** and amounts to little more than 200 cases to date in the published

English literature (1,13,14,15,16).

Although occurring predominantly in antenatal (61%) and the postnatal period (21%) it has been described in nearly 18% of cases intrapartum (13) and recurrence (4) during the same or in the following pregnancies has been reported.

The specific features at the presentation - abdominal pain and fetal distress sometimes accompanied by signs of hypovolemia- resembling other conditions not uncommon in labor, represent a challenging diagnosis, given the rapid feto-maternal deterioration and the limited time frame allowed for investigations before deciding on life-saving emergency surgery.

Among all the possible risk factors, since hemorrhage from rupture of subserosal uterine vessels had been more and more frequently observed over the last decades, interest in a possible association with endometriotic lesions was first suggested in 1992 (8). The theory found support in the conclusions of a literature search and two case reviews published between 2009 and 2017 (3,4,13,17) on 131 cases of true SHiP recorded since 1950. A robust association with endometriosis and deciduous was confirmed on histopathology taken from the source of bleeding in the totality of a small case series (13). Vascular disruption of superficial veins or varicosities associated with necrosis, hemorrhagic foci, and deciduous were present in 90% of cases at the broad ligaments, left uterosacral ligament, or posterior side of the uterus.
It should be emphasized, however, that "deciduous," i.e., extra-uterine decidualization of stromal cells, although often associated with, is not synonymous with endometriosis strictly speaking, and can occur anywhere in the peritoneal cavity during pregnancy.

Full pathogenesis is therefore still conjectural. Under investigation the relation between the placental site and the penetrating anastomotic branches of uterine vessels, mechanisms of possible simple 'erosion' of vessels, the progesterone mediated involution and necrosis of ectopic decidua around the uterine vessels, inflammatory adhesions, and whether an increased abdominal pressure, as in late pregnancy, might elicit the rupture of a congested vascular system.

A few remarkable aspects are worth mentioning: the specific presentation and sometimes rapid evolution. Especially when in labor, poor chances of performing instrumental evaluation for confirmation of internal bleeding and its extent, as accounts of hemoperitoneum comprised between 1 to 6 liters suggest. The limited accuracy of ultrasound scanning in the diagnosis of hemorrhage also in case of advanced pregnancy the described high recurrence rate: in the same pregnancy, in the puerperium, in

following pregnancies **(**17**)** the immediate requirement of life-saving emergency surgery combined with the unpredictability of the intraoperative findings.

Conclusions

More widespread acknowledgment of such unusual conditions is foremost for life-saving management. Nevertheless, a better understanding of the pathogenetic mechanisms is advocated, could any preventive measure be established. Of concern appears the possible association with endometriosis, especially in cases of subtle lesions, given the increasing prevalence of the disease combined with improvement of the reproductive techniques supporting those women in achieving pregnancy.

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