SAGE Open Medicine
Volume 12: 1–6

DOI: 10.1177/20503121241243245
journals.sagepub.com/home/smo
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Spontaneous haemoperitoneum in pregnancy in women with endometriosis: Diagnostic challenges and management strategies

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Abstract

Spontaneous haemoperitoneum in pregnancy is a rare but potentially life-threatening condition that requires prompt recognition and management to avert a catastrophic outcome for the mother and baby. The main aim of this review is to summarise the current knowledge on this topic, including its incidence and clinical presentation, highlight the diagnostic challenges, and suggest management strategies. To achieve our objective, we examine the existing literature to provide insights into this complex clinical presentation to enhance our understanding and, in so doing, contribute to the existing body of knowledge on the subject of spontaneous haemoperitoneum in pregnancy.

Keywords

Endometriosis, management strategies, spontaneous haemoperitoneum in pregnancy, multidisciplinary care, pregnancy, spontaneous haemoperitoneum

Date received: 23 September 2023; accepted: 14 March 2024

Introduction

Spontaneous haemoperitoneum is uncommon during pregnancy, and its association with endometriosis further adds to the complexity of the condition.[1,2] SHiP is spontaneous, non-traumatic intraperitoneal bleeding in pregnancy and up to 42 days after delivery requiring surgical intervention or embolisation, not due to uterine rupture, ruptured ectopic pregnancies, or caesarean section-related bleeding.[2,3] SHiP has been described in the setting of ruptured uterine arteries, varicose veins or aneurysms of the splenic artery, but causes due to endometriosis are increasingly reported in the literature.[3–10] Other reported causes of SHiP include spontaneous rupture of the liver, the right renal hamartoma, rupture of the external iliac vessel branch and unscarred uterine rupture due to placenta accreta spectrum.[11] The exact pathogenesis of SHiP in women with endometriosis remains an area of ongoing scientific research. SHiP in endometriosis may emanate from ruptured endometrioma, utero-ovarian vessel bleeding or bleeding from endometriotic implants eroding pelvic blood vessels.[4–6] Diagnosis of SHiP may be challenging, mainly because of its ability to mimic different acute gynaecological and surgical pathologies; this may lead to adverse maternal outcomes, including maternal deaths in severe cases and perinatal morbidity and mortality.[2–7] This article aims to review the existing literature, identify potential etiological factors, highlight the diagnostic challenges and suggest management strategies.

Search strategy

A literature search was completed using the topic modelling method. To identify relevant articles, we created a search

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question titled 'Current management strategies of spontaneous haemoperitoneum in pregnancy (SHiP)'. We searched different databases, namely Google Scholar, Scopus, PubMed, Web of Science, Research Gate, Cochrane Library, protocols and guidelines using the following keywords and phrases: spontaneous haemoperitoneum, haemoperitoneum in pregnancy, haemoperitoneum and endometriosis, haemoperitoneum and assisted reproduction, haemoperitoneum in pregnancy and management challenges, and endometriosis. We identified papers published in peer-reviewed journals, including retrospective and prospective studies, case reports, case series, systematic reviews and meta-analyses. A total of 54 peer-reviewed articles were identified, and after excluding non-recent publications, we also excluded papers that do not address the review's objectives and papers published in languages other than English; a total of 27 articles were selected and cited in this review article.

Incidence

Spontaneous haemoperitoneum during pregnancy is rare, with limited reported cases in the literature. The exact incidence of this condition in patients with severe endometriosis remains unknown, but a conservative estimate of 1:10,000 is quoted in the literature.[12] The SHiP has been an old problem in obstetrics, with the first case report published in 1957 by Doyle et al.,[13] but it continues to pose challenges in contemporary practice. The incidence of this condition will surely increase as more women with endometriosis who were previously considered sub-fertile are increasingly conceiving through in vitro fertilisation (IVF) and naturally,[5] which underscores the need for greater awareness among clinicians and vigilance in patients with significant risk factors for SHiP.

In the United Kingdom, a national survey led by the United Kingdom Obstetric Surveillance System since 2016 is ongoing to determine the true incidence of SHiP; data collection is continuing, and results are expected to be published soon.[3] A national survey completed over 2 years in the Netherlands reported an incidence of 4.9/100,000 births.[2] Like other cases in the literature, the Netherlands survey revealed an initial misdiagnosis in most of their case series. A large multicentre prospective cohort study[8] completed in Italy reported an incidence of 0.04/1000 births. Bazurini et al.[1] reported a case series of six patients over 13 years with an estimated prevalence of 1.7/10,000 births. This literature review highlights the limited available evidence on this topic and the need for further research to enhance our understanding of the underlying mechanisms, risk factors and optimal management strategies.

Aetiopathogenesis and clinical presentation

The precise aetiology of spontaneous haemoperitoneum in such cases remains unclear; it is thought that endometriotic implants may undergo structural changes and become prone to bleeding during pregnancy.[9–13] Different theories have been proposed, including rupture of endometriotic cysts, decidual haemorrhage and hormonal and vascular changes associated with pregnancy.[2–16] Current theory presumes that decidualisation, chronic inflammation and pre-existing adhesion of endometriosis play a role in SHiP development. During pregnancy, the uterus has an increased blood supply, and utero-ovarian vessels become physiologically hypertrophic. Endogenous progesterone during pregnancy enhances endometriotic implant decidualisation and penetration into vessels.[6,10] Chronic inflammation of the endometriotic lesion leads to adhesion formation, while pregnancy-related hormonal changes cause the surrounding tissues and vessels to become friable. As the uterus enlarges, the formed adhesion creates traction on surrounding tissue, making the affected tissue or vessels prone to bleeding when the uterus grows rapidly in the second and third trimesters[4–13] which may explain why SHiP seldom happens in the first trimester.[1,14] Although the origin of bleeding remains unknown in a few cases, even during laparotomy, connective tissue diseases should be considered in the etiological analysis.[17]

SHiP typically presents with acute-onset abdominal pain,[10–13] often accompanied by signs of hemodynamic instability. Patients may exhibit diffuse abdominal tenderness, guarding and rebound tenderness, which may mislead the unwary clinician, leading to delay in diagnosis and increased potential for adverse outcomes.[4–10] Some patients also present with signs of foetal distress identified on the cardiotocograph or ultrasound scan. Arteries and superficial veins, or varicosities on the posterior surface of the uterus or parametria, are often the sites of bleeding.[5] There is evidence to show a preponderance of SHiP emanating from endometriosis involving the left adnexal greater than the right.[6,9,14] However, there is no clear explanation for this occurrence in women presenting with features of SHiP; a previous history of endometriosis before conception should raise suspicion for SHiP. Endometriosis is the most common risk factor or etiologic cause in the literature, but there is no correlation between the stage of endometriosis and SHiP in the majority.[1,4,6,10,15] With IVF, more women with endometriosis are conceiving, and therefore, IVF is increasingly reported as a common factor in patients diagnosed with SHiP.[1,6] To determine the prevalence of SHiP among 362 endometriosis-affected women conceiving through IVF, Benaglia et al.[18] found a rate of 0.3%, and they concluded that SHiP is rare in the general population of endometriosis-affected women undergoing IVF.

Diagnostic challenges

Diagnosing SHiP poses a significant challenge due to its rarity and non-specific clinical presentation.[1] Diagnostic challenges arise due to the overlap of symptoms with other common pregnancy-related conditions, such as placental abruption and uterine rupture, and this means that many

patients with SHiP have been misdiagnosed.[4–6,10] Both obstetric and non-obstetric causes have been reported to cause SHiP. However, the aetiology is usually confirmed during surgical intervention, and in some cases, it is almost impossible to be sure of the diagnosis in advance.[5,10] Imaging modalities such as ultrasound, computed tomography (CT) and magnetic resonance imaging (MRI) play crucial roles in identifying the source of bleeding and assessing the foetal condition. However, false-negative results can occur, necessitating a high index of suspicion and a multidisciplinary approach. In some patients, a combination of diagnostic modalities might be required if the clinical diagnosis is equivocal. Ship management can be pretty challenging, mainly when it occurs remotely from term, and this should involve a multidisciplinary team.[7] Culdocentesis may aid in clinical diagnosis, but the limitations of such a diagnostic approach limit its application. Radiological diagnosis is preferred in developed countries and centres with good diagnostic imaging facilities. However, culdocentesis or abdominal paracentesis may be done in resource-constrained environments to check for haemoperitoneum, but there could be false-positive results from aspirating a blood vessel. Correct preoperative diagnosis of SHiP based on a clinical presentation is difficult because of its non-specific symptoms at presentation, rarity and hence frequently misdiagnosed.[15] In suspicion, without apparent clinical evidence of SHiP, posterior culdocentesis could be performed. CT scans and MRIs could also be necessary to rule out differentials like vascular lesions.[11] Urgent laparoscopy may be required when there is a high index of suspicion regarding the cause of SHiP to identify the source of the bleeding and guarantee hemostasis.[19] This may be quite beneficial in early pregnancy. Reported cases of spontenous haemoperitoneum in pregnancy are shown in Table 1.

Management strategies

Spontaneous haemoperitoneum during pregnancy is an infrequent occurrence that demands immediate attention due to its potential maternal and foetal complications.[1–6,12] When this condition arises in patients with severe endometriosis, the challenges of diagnosis and management further escalate. A high index of suspicion and a multidisciplinary approach involving obstetricians, anaesthetists, gynaecologists and surgeons are essential for successful management. Haemodynamic stabilisation through fluid resuscitation and blood transfusion is paramount. Surgical intervention, typically laparotomy, is often required to identify and control the source of bleeding.[1–10] Preservation of the mother's well-being and consideration of the foetal condition guide the decision-making process.

In an emergency with life-threatening bleeding, the most expeditious intervention should be offered to save the patient's life. The cases reported by Aliyu et al.[5] and Kim et al.[16] had a hysterectomy due to massive ongoing bleeding. Tesia performed a bilateral salpingo-oophorectomy in addition to a hysterectomy. Where possible, identification and suture ligation of the bleeding vessel should be the treatment of choice. However, if it becomes necessary to perform a hysterectomy, this should be done without delay to avoid maternal death. The decision about foetal delivery should be balanced based on the clinical situation; where there are signs of foetal deterioration, foetal delivery should be expedited during the laparotomy. It is vital that tissue biopsies are taken during surgery for SHiP and that any tissues removed are sent for histology to confirm the diagnosis and guide future management plans. Maternal mortality due to SHiP has dropped dramatically during the second half of the 20th century to approximately 4%.[5] However, foetal mortality remains high (approximately 31%), especially in earlier gestations, with 44% of the deaths attributable to maternal shock.[5] Overall, there was an excellent maternal outcome in many cases reviewed in the literature.[1–17]

Watchful waiting may be recommended in well-selected cases, especially in those that are asymptomatic, clinically and hemodynamically stable patients in the early trimesters. Also, for those quite remote from the term, conservative management may be instituted after the initial surgical intervention.[26] Despite the low rate of successful post-SHiP pregnancy continuation, patients should be made aware of the possibility of miscarriage, recurrence of SHiP during the index and subsequent pregnancy.[11] Also, the possibility of an emergency surgical intervention should be discussed with the patient. Lier et al.,[9] in their evaluation of the clinical consequences of the spontaneous haemoperitoneum in pregnancy (SHiP) and its association with endometriosis, recommended that expectant treatment be taken into consideration if hypovolemic shock or foetal distress were not present. Fluid resuscitation and expectant management may be explored. Brosen et al.,[27] in their work, concluded that pregnancy-related spontaneous haemoperitoneum must be treated case-by-case. Conservative care until the term is appropriate in preterm instances where the bleeding points are effectively addressed.[27]

Follow-up and future pregnancy

Clear postoperative follow-up plans should be implemented to provide ongoing assessment and psychological support, as the long-term impact of SHiP could be enormous. If the uterus is conserved, there is no contraindication to future conception; the timing and mode of delivery should be individualised, but the ultimate decision should rest with the patient. If a hysterectomy has been performed and the patient desires children, adoption and surrogacy are viable options and should be discussed holistically with the patient.

Conclusion

Spontaneous haemoperitoneum in pregnant patients with severe endometriosis is a rare and challenging condition.

Table 1. Summary of cases of spontaneous haemoperitoneum in pregnancy.

Authors	Maternal age (years)	Parity	Clinical presentations	Gestational age (weeks)	Estimated blood loss (ml)	Histology/size and number of endometriosis mass	Blood transfusion	Management	Foeto-maternal outcome
Mamah et al.[20]	30 plus	Primigravida	Persistent abdominal pain misdiagnosed as acute appendicitis. MRI showed complex tubo-ovarian mass, and subsequent haemodynamic instability. Past medical hx of endometriosis	32 weeks + 5 days	4700 ml	Ovarian endometrioma	Yes. Five units of red cells	Emergency laparotomy, caesarean delivery, left salpingo-oophorectomy, and hysterectomy	Live baby. Good postop recovery
Kato et al.[21]	41	GIP0	Severe abdominal pain, foetal heart rate abnormality and haemoglobin of 7.7 g/dl. Had a history of adenomyosis. Conception was via IVF	28 weeks. She had earlier presented at 12 weeks of gestation	2020 ml	Endometriotic deposits on the posterior uterine wall. Histology not done	No blood transfusion	Emergency caesarean section and haemostasis secured	Live neonate. Mother subsequently discharged on the 15th day postoperatively
Li et al.[22]	32	G3P1	Severe upper and lower abdominal pain, with foetal heart rate abnormality	36 weeks + 2 days	2000 ml	None	Yes	Emergency caesarean section + transfixion of ruptured blood vessels	Live neonate. The patient was admitted to the ICU and discharged subsequently
Li et al.[22]	24	G1P0	Progressive abdominal pain with palpitations, tenesmus, and dizziness. IVF-ET conception. Prior history of endometriosis	27 weeks	3600 ml	None. Stage iv endometriosis	Yes	Emergency caesarean section + double loop transfixion in the uterine anterior wall	Stillbirth. The patient made a good recovery
Huang et al.[10]	35	Primigravida	Generalised lower abdominal pain with subsequent onset of maternal shock	18 weeks	1500 ml	Decidualised endometrial tissue with contiguous infiltration from the peritoneal surface to more than 5 mm deep	Not documented	Emergency laparotomy + hystorotomy and arrest of bleeding from decidualised endometriotic tissue	Stillbirth. Good postop recovery
Aliyu et al.[5]	31	G1P0	Severe abdominal pain, dizziness and restlessness with a subsequent significant drop in haemoglobin from 10.8 to 3.6 g/dl	24 weeks	5000 ml	Histology of previous surgery revealed endometriosis. Endometriotic deposits were noted intraoperatively during the index surgery	Yes. Red cells × 19 units, Fresh frozen plasma × 15 units, Platelet concentrate × 8 pools and 2 units of cryoprecipitate	Laparotomy + hysterectomy	Two female neonates, fresh stillbirths. They recovered well and were discharged on the 14th day postoperatively
Yang et al.[23]	23	Primigravida	Lower abdominal pain, palpitation, dizziness, nausea and abdominal bloating	29 weeks 1 day	1900 ml	None	Yes. 6 units of packed red cells	Exploratory laparotomy, caesarean section and haemostasis	Foetal death
Mayadeo et al.[24]	23	Primigravida	Abdominal pain, tender uterus, abnormal foetal heart on CTG	34 weeks	1400 ml	Histology of placenta and membranes (suspicion of chorioamnionitis) revealed normal findings	Yes. 2 units of packed red cells	Exploratory laparotomy, caesarean section, and haemostasis secured involving superficial serosal veins on the posterior uterine surface	Live female neonate. Good foeto-maternal outcome and were discharged 7th day postoperatively
Brichant et al.[25]	23	G2P1	Labour pains. Developed severe abdominal pain and haemodynamic instability following vaginal delivery. Previous history of surgery for right endometrioma	39 weeks	2000 ml	Right ovarian endometrioma	Not documented	Emergency exploratory laparotomy and right adnexectomy	Live female baby and good foeto-maternal outcome
Brichant et al.[25]	37	GIP0	Presented for induction of labour and subsequently developed hypovolemic shock and foetal distress. Had previous surgery for endometriosis	41 weeks	2000 ml	None. Uterine artery bleeding caused by possible endometriotic adhesion	Not documented	Emergency caesarean section plus uterine artery ligation and secure of haemostasis	Good outcome. Mother discharged 5th day postoperatively

CTG: cardiotocography; g/dl: grams per decilitre, ml: millilitres; IVF: in vitro fertilisation; IVF-ET: in vitro fertilisation-embryo transfer, hx: history.

Every pregnant patient that presents with sudden onset of severe abdominal pain, signs of peritonism and hypovolemic shock should be evaluated for SHiP, having excluded other prevalent causes of haemoperitoneum in pregnancy. This review highlights the importance of accurate diagnosis, multidisciplinary collaboration, and, if indicated, timely surgical intervention in achieving favourable outcomes for the mother and baby. We recommend further research to fully understand the pathophysiology and optimal management strategies for one of the gynaecological imitators, SHiP.

Limitation

The authors acknowledge that this review may be incomplete, and all the published articles on the subject may have yet to be reviewed during our literature search. Given that much research is ongoing on this evolving subject, we hope our article will make a small contribution to the discussion.

Acknowledgements

The authors thank everyone who supported us while writing this review article. We also acknowledge the valuable works of the scholars whose materials we have used for this review.

Author contribution

JE Mamah was involved in the conceptualisation of the study. JE Mamah, CR Otu, CC Okafor and CG Okafor were engaged in article writing, proofreading and overall revision.

Data availability statement

Not applicable.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship and/or publication of this article.

Ethical approval

Not applicable.

Informed consent

Not applicable.

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