Clinical Review

The Royal Society of Medicine

Journal of the Royal Society of Medicine Open;
9(1) 1–9
DOI: 10.1177/2054270417746059

Maternal morbidity and mortality associated with retroperitoneal haematomas in pregnancy

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Summary

Retroperitoneal haematomas in obstetrics are uncommon. The causes and pathogenesis of retroperitoneal haematomas lack clarity and the aim of this review is to recognise retroperitoneal haematomas as a separate entity from commonly seen vaginal and pelvic haematomas. It is time to raise awareness among obstetricians to recognise retroperitoneal haematomas as an important cause of maternal morbidity and mortality which requires high clinical suspicion and multidisciplinary input. As retroperitoneal haematomas are rare but can cause serious threat to maternal wellbeing, resources should be directed towards their management. Existing guidelines of maternal collapse and morbidity during pregnancy and puerperium need to include retroperitoneal haematomas as one of the important causes of maternal shock or morbidity. New learning pathways should be opted for to increase awareness of retroperitoneal haematomas among obstetricians enabling them to reflect on their implications while managing retroperitoneal haematomas. Management of retroperitoneal haematomas is complex and continues to improve with advancements in the investigative strategies, treatment options and multidisciplinary involvement.

Keywords

retroperitoneal, haematoma, concealed

Background

The incidence of retroperitoneal haematomas in obstetrics is not well known. However, literature review has revealed that the incidence of puerperal haematomas varies from 1:309 to 1:1500 deliveries.[1] Considering the overlapping confusion of terminology between puerperal haematomas and retroperitoneal haematomas, the true incidence of retroperitoneal haematomas in pregnancy seems difficult to ascertain. It is because puerperal haematomas mainly account for vaginal, vulval and supravaginal and broad ligament haematomas which are not always retroperitoneal.

There has been less clarity about the aetiological factors of retroperitoneal haematomas as evident by the fact that the RCOG guideline 56[2] only mentions two causes of retroperitoneal haematoma, namely the splenic artery rupture and hepatic rupture, but does not cover other causes of retroperitoneal haemorrhage. Since the year 2007, around 21 case reports of retroperitoneal haematomas in obstetrics resulting in maternal collapse or maternal morbidity have been published (Table 1).

We aim to bridge the gaps in the understanding and management of retroperitoneal haematomas in obstetrics. We argue that retroperitoneal haematomas are rare and a separate entity from commonly considered pelvic haematomas, and we have tried to simplify the proposed classification in Figure 1.

Method

Relevant literature was identified through a search of the online databases (MEDLINE, EMBASE and scholar database) using a number of keywords (e.g. haematoma, retroperitoneal, broad ligament, paravaginal, concealed, supravaginal, infravaginal) either alone or in combination. A range of publications was retrieved, including case reports, RCOG guidelines, congress abstracts and books, and a subset of 30 relevant articles was used for our reference list from 1945 till 2016. Information and evidence have been compiled from these to provide the reader with a comprehensive review on the entity which is never explored intensively as a recognised cause of maternal morbidity and mortality in obstetrics.

Aetiology

Retroperitoneal haematomas can occur during pregnancy and puerperium. Retroperitoneal haematomas can develop spontaneously in the absence of any mechanical cause or result from trauma.

Pathophysiology of spontaneous retroperitoneal haematomas

The spontaneous retroperitoneal haematomas can develop in the absence of any trauma during

Table 1. Presentation, diagnosis and management of retroperitoneal haematomas cases in obstetrics from year 2007 to 2016.

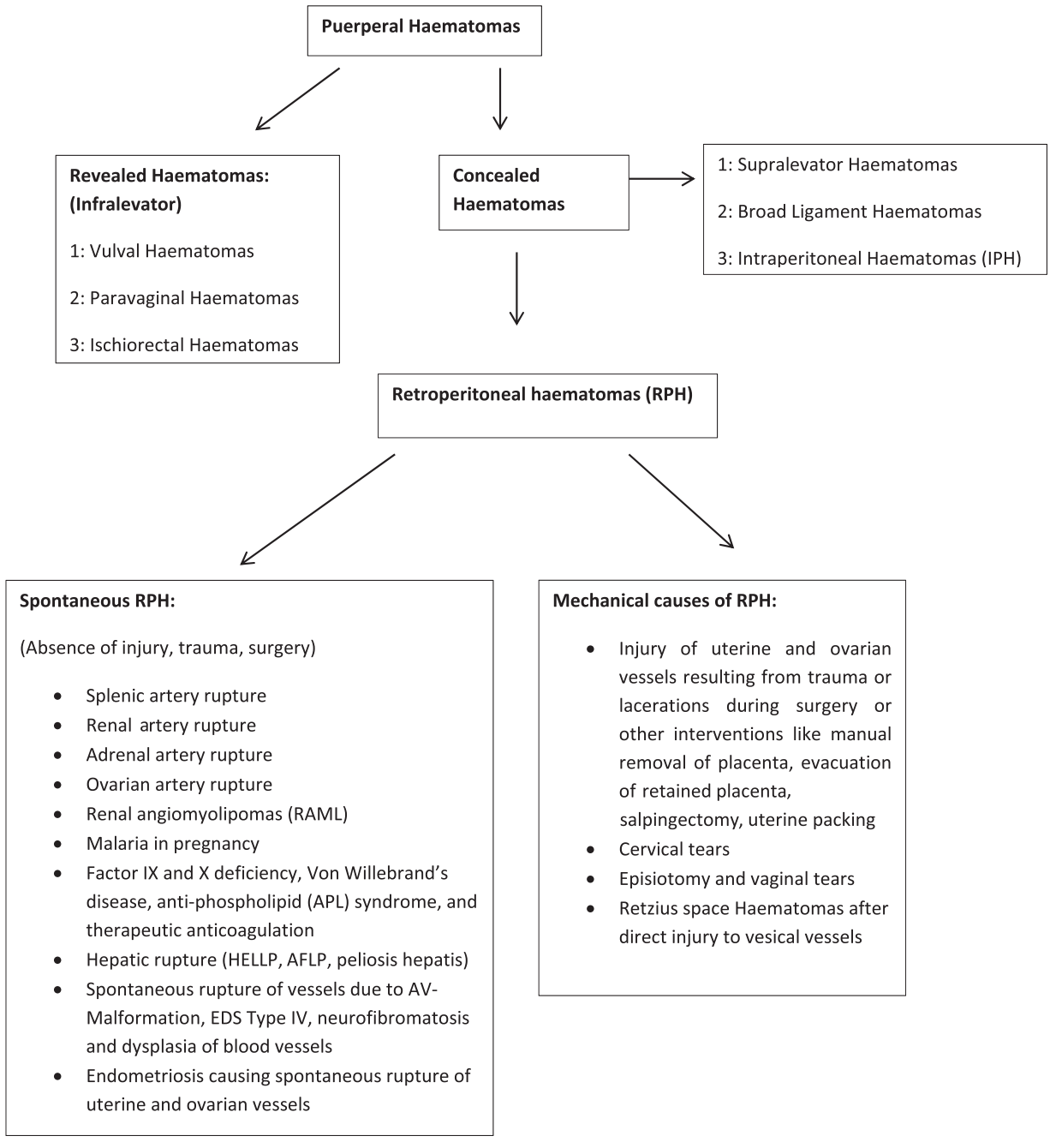
Diagnosis	Presentation	Management
Spontaneous retroperitoneal haematoma: Three cases after normal delivery.[3–5]	Unstable within 3–72 h post vaginal delivery	Depending upon the condition of the patient, the cases were managed conservatively[3] without surgical exploration, embolisation of right internal iliac artery[5] and laparotomy and internal iliac ligation respectively.[4]
RPH identified after secondary PPH: Case 4: RPH due to tear of upper vaginal wall and injury to vaginal artery after episiotomy[6] Case 5: RPH due to uterine rupture identified on laparotomy[7]	Two days' history of left sided perineal and abdominal pain[6] and shock in multipara[7]	Postpartum hysterectomy and repair of pelvic floor including vaginal wall.[6] Postpartum hysterectomy with unilateral salpingo-oophorectomy and pelvic packing.[7]
Case 6: Post caesarean section, 18 cm RH extending from left pararenal space to pelvis[8] Case 7: Post caesarean section, massive RH extending from right kidney to groin due to AV malformation[9] Case 8: Retroperitoneal haematoma diagnosis confirmed during caesarean section[10]	Shortness of breath, abdominal and right iliac fossa pain, septic shock with anuria[8] femoral neuropathy (9) and fetal bradycardia[10]	Cases reported in Chao et al.[8] and Bisseling et al.[9] were managed conservatively. In the case reported by Bolla et al.,[10] intra-abdominal palpation (prompted by ultrasound findings before caesarean section) confirmed an extensive retroperitoneal mass near the right kidney. Interventional radiologist confirmed bleeding from right adrenal artery and successfully achieved haemostasis after coiling.
Cases reported of spontaneous adrenal haemorrhage: Case 9: at 24 weeks[11] Case 10: at 33 weeks[12] Case 11: at 36 weeks[13] Case 12: at 37 weeks[14] Case 13: 12 h post-delivery[15]	Abdominal pain, tachypnoeic,[11,12] chest pain[13,14] and shock[15]	Management varies as: Factor VIII administration and retroperitoneal space packing[13] • Adrenalectomy[12,13] • Intra-arterial ablation of the renal artery[14] • Conservative management[11,15] but cases of adrenal haemorrhage due to thrombosis of adrenal veins may warrant anticoagulation, and then replacement with glucocorticoids and mineralocorticoids may be required depending on adrenal function.[11]
Case 14: Spontaneous retroperitoneal haematoma in context of disseminated intravascular coagulation.[16]	Disseminated intravascular coagulation complicating delivery	Correction of disseminated intravascular coagulation and haemostasis but no surgical management done

(continued)

Table 1. Continued.

Diagnosis	Presentation	Management
Case 15: RPH with obstructive uropathy post vaginal delivery[17]	Vaginal pain, bilateral flank pain and difficulty micturition 12 h post vaginal delivery	Ligation of the hypogastric artery and decompression of the ureters bilaterally
RPH due to ruptured Renal angiomyolipoma (RAML): Case 16: Ruptured RAML at 18-week gestation[18] Case 17: Ruptured RAML in third trimester[19]	Abdominal pain[19] low back pain and microscopic haematuria[18]	• Managed conservatively from 18 week till 35-week gestation with clinical monitoring and follow up MRI scan to confirm non-expanding haematoma and finally delivered with caesarean section at 35-week gestation[18] • Laparotomy and nephrectomy undertaken two days post caesarean section[19]
Cases 18 and 19: Retroperitoneal haematoma in Wunderlich syndrome (rupture of renal angiomyolipoma with shock)[20,21]	Abdominal pain at 6 week[21] and in other case at 33-week gestation[20]	• Exploratory laparotomy with nephrectomy and evacuation of haematoma[20] • Embolisation of ruptured cystic lesion, partial nephrectomy and evacuation of haematoma through retroperitoneal approach[21]
Case 20: Retzius' space RPH[22]	Abdominal distension, pain, suprapubic bulge and unstable condition admitted to hospital after normal vaginal delivery.	Laparotomy for bilateral uterine artery ligation and bilateral internal iliac artery ligation.
Case 21: RPH diagnosed at post-mortem[23]	Sudden episode of shortness of breath, epigastric pain and cardiorespiratory arrest at 36-week gestation in hospital while awaiting caesarean section for placenta previa in a developing country.	Findings at post-mortem: • RPH in the pelvis extending from mesenteric base, left pararenal space and paracolic gutter behind rectum.

Figure 1. Classification of puerperal and retroperitoneal haematomas in obstetrics.



antenatal, intrapartum and postnatal period due to causes including ruptured aneurysms, resulting from anticoagulation therapy, obstetrical coagulopathies, etc. The processes may relate to physiological haemodynamic changes during pregnancy including pelvic vessels engorgement, decreased vascular resistance, increased cardiac output and blood volume. There is some evidence that the elastic layer of blood vessels undergo fibroplasia like changes[24] stimulated by steroid hormones of pregnancy, which can lead to disruption and rarely aneurysm and cause spontaneous haematomas.

Most reported spontaneous vascular rupture causing retroperitoneal haematomas in obstetrics is of splenic artery. Almost 150 cases of splenic artery rupture in pregnancy have been reported since 1945. This is

followed by 42 cases of renal artery aneurysms (RAA), 25 cases of ruptured renal angiomyolipoma (RAML) and 12 cases of ovarian artery rupture. The underlying pathology in many cases was found to be associated with A-V malformations and EDS Type IV, neurofibromatosis and dysplasia.[1]

Relatively commonly seen endometriosis in benign gynaecology is considered as the recognised cause of spontaneous rupture in cases of uterine and ovarian artery rupture.[25] This assumes significance with the fact that recently more women with advanced stage endometriosis are seeking for fertility treatments. Therefore, in many cases of intraperitoneum and retroperitoneal bleed, endometriotic implants are found on the posterior uterine walls, uterosacral ligaments and in the form of retroperitoneal haematomas of lateral pelvic walls involving ureters.

The placenta accreta and percreta are other less known but important causes of retroperitoneal haematomas as the incidence of placenta accreta/percreta is increasing perhaps due to rise in caesarean sections. During delivery, placenta accreta and percreta may result in uterus perforation, internal bleeding and retroperitoneal haematoma and cases have been reported.[26,27]

Hypertensive disorders in pregnancy can be associated with low platelets, coagulopathies HELLP syndrome, AFLP, in extreme cases sub-capsular haemorrhages and liver rupture, contributing to spontaneous retroperitoneal haematomas causes. Spontaneous hepatic rupture is rare, and mostly associated with severe pre-eclampsia, eclampsia and its related complications. An interesting case of hepatic rupture was reported in a 53-year-old woman at 27 weeks' gestation who was conceived by IVF and it was found to be associated with peliosis hepatis.[28]

Although rarely encountered, Retzius space (loose connective tissue and fascia between the bladder and the pubis) haematoma can be caused by injury of Santorini venous plexus by descent and delivery of fetal head. It can extend from paravesical space to broad ligaments and beyond.

Malaria can cause splenic accidents and hence a cause of retroperitoneal haematomas; though non-traumatic, spontaneous splenic rupture in malaria is rare. However, recent revival of epidemics in malaria can cause splenic accidents in pregnant women.[29] The exact mechanism of spontaneous rupture is not established and probably it is due to hyperplasia and stretching of splenic parenchyma, infarcts, haemorrhage and haematomas, tears or fibrosis.

The factor IX and X deficiency, Von Willebrand's disease, anti-phospholipid (APL) syndrome, and therapeutic anticoagulation can be predisposing factors for retroperitoneal haematomas which can be precipitated by mechanical factors of parturition.

Pathophysiology of mechanical factors causing retroperitoneal haematomas

Retroperitoneal haematomas can occur from mechanical factors including trauma from accidents, childbirth and complication of interventions. The most recognised cause of retroperitoneal haematomas is injury of uterine and ovarian vessels resulting from trauma or lacerations during surgery or other interventions like manual removal of placenta, evacuation of retained placenta and uterine packing, etc.[1]

Cervical tears cause laceration of the descending branches of the uterine artery and may lead to the paravaginal haematoma, which can extend through the paravesical space into two layers of broad ligament and beyond forming a retroperitoneal haematomas.

Trauma or laceration to vagina involve vaginal vessels which may cause retroperitoneal bleed, posterior and or laterally into broad ligament region. Retroperitoneal haematomas resulting from vaginal trauma can develop post normal or after instrumental delivery due to anatomical disruption as explained above. A commonly seen scenario is a laceration post vaginal delivery extending from posterior vaginal wall to uterosacral ligament and into para rectal space leading to retroperitoneal haematomas. Explanation lies in the way the vagina receives its blood supply from the vaginal arteries and their anastomoses with branches of the uterine, inferior vesical and internal pudendal arteries.

Episiotomy causing injury to internal pudendal and inferior rectal artery can result in ischiorectal fossa and paravaginal haematomas, though not uncommon. Such haematomas may subsequently extend from paravaginal to infralevator and supralevator space finally breaching retroperitoneal space.[30]

The bleeding in Retzius space is a rare occurrence, but is associated with urogynaecology incontinence procedures like TVT. However, in obstetrics, the bleeding into Retzius space is unexpected and encountered even more rarely, possibly after direct injury to vesical vessels. One case has been reported of Retzius haematoma during caesarean section and another was reported of Retzius haematoma case believed to result in retroperitoneal haematoma after spontaneous vaginal delivery without any episiotomy or vaginal tears involving pudendal Santorini plexus.[22]

Spread of blood from one compartment to another compartment is facilitated by the proximity of the paravaginal space with paravesical, parametrial and pararectal spaces in case of injury in the form of lacerations or tears to vagina.

Clinical features, diagnosis and management

Retroperitoneal haematomas can be potentially life-threatening with considerable variability in morbidity depending on the mechanism of retroperitoneal haematomas development, the speed and amount of bleeding.

- Presentation of retroperitoneal haematomas varies from acute to sub-acute and rarely insidious onset and is often recognised from early in the pregnancy until four weeks postnatal.
- The patient may present with signs and symptoms of shock, i.e. tachycardia, hypotension, tachypnoeic, foetal distress, abdominal pain especially upper right or left quadrants pain, nausea, vomiting, interscapular pain, chest pain, and right shoulder pain. Often the clinical picture is sudden in onset and other common features are abdominal pain, accompanied by signs of hypovolemic shock and reduction in haematocrit.
- Vomiting, diarrhoea and epigastric pain may be initial symptoms in splenic artery rupture.
- In gradually developing retroperitoneal haematoma, the early diagnosis is difficult to establish. It manifests only after a significant amount of blood loss has occurred with resultant haemodynamic compromise.
- Vaginal and rectal pain with oedematous vaginal mucosa protruding through introitus may be seen in lower pelvic haematomas.
- Femoral neuropathy,[16] spasms of iliopsoas muscle and sciatica are seen with retroperitoneal haematomas in the iliopsoas region.
- It has been reported that in some cases where retroperitoneal haematoma extend from pararenal space to pelvis, the patient cannot perform hip flexion and knee extension.[8]
- Cullen's sign, which is bruising around umbilicus, occurs due to tracking of haemorrhagic fluid from the retro-peritoneum along the gastrohepatic and falciform ligament to the umbilicus.
- Grey Turner's sign is the blackish discolouration of flanks which may be seen in some cases 24–48 h after the retroperitoneal haematomas development. The cause of this sign is blood tracking from retroperitoneal space along fascial planes to the abdominal wall musculature.

Caution

1. In acute presentations, retroperitoneal haematomas can cause serious maternal and foetal morbidity or mortality. In gradually deteriorating cases after normal or operative vaginal delivery or caesarean section, there should be an early attention paid to the possible presence or an expansion of retroperitoneal haematomas.
2. Similarly, after any uterine artery embolisation, B-Lynch suture, extension of injury into broad ligament or a complicated caesarean section due to abnormal sited or adherent placentae, it should alert one to the possibility of the presence of retroperitoneal haematomas along with other differentials. As mentioned earlier hypertensive disorders in pregnancy potentially contribute retroperitoneal haematomas due to its association with complications starting from low platelets, coagulopathies, DIC, HELLP syndrome, sub-capsular haemorrhages of liver and, in extreme cases, liver rupture.
3. Recent travel to tropical region should alert one to the possibility of malaria-related splenic accidents in pregnancy[29] and hence a cause of retroperitoneal haematomas. It is prudent to check the previous history of clotting disorders like factor IX and X deficiency, Von Willebrand's disease, antiphospholipid syndrome and anticoagulation, which can be predisposing factors in some cases of retroperitoneal haematomas.

Investigations

Ultrasonography of the abdomen and pelvis is usually the first line of investigation; however, the information gathered is often limited, as it may detect haematoma but it is not precise enough to ascertain the cause. Free fluid in the abdominal or pelvic cavity is an associated finding in most cases, as blood can be tracked into pelvis.

CT scan is highly sensitive but sometimes CT angiography may be required which can reveal the site of the bleed and demarcate the contrast outside the blood vessels.

MRI can be useful in defining the retroperitoneal haematomas but the ones that need to be differentiated from septic thrombophlebitis and abscesses can be helped by undertaking CT imaging as well because the management varies in retroperitoneal haematomas compared to abscesses.

Management

The patients with retroperitoneal haematomas can present either as acutely unwell, rapidly deteriorating patients or symptoms can have insidious onset. Successful management requires a high index of clinical suspicion considering that retroperitoneal haematomas is a rare clinical entity. If not managed by

taking appropriate steps in time, retroperitoneal haematomas may result in high morbidity and mortality.

In both scenarios, the real problem is considering retroperitoneal haematomas as one of the possible causes of a life-threatening clinical picture; therefore, early diagnosis and prompt management in the form of pertinent investigations and treatment plan is vital for a successful outcome. In obstetrics, retroperitoneal haematomas is not commonly seen, and because of confusing clinical features of retroperitoneal haematomas which can overlap with other pathologies, precious time is wasted before diagnosing retroperitoneal haematomas.

Most of the patients with spontaneous or iatrogenic retroperitoneal haematoma can be managed conservatively while being closely monitored, without the need for further intervention. Initial management of retroperitoneal haematomas is targeted at fluid resuscitation, blood transfusion and reversal of coagulopathy. The evidence from non-obstetrical patients revealed that conservative management alone is adequate in most cases of retroperitoneal haematomas where the patient is haemodynamically stable.

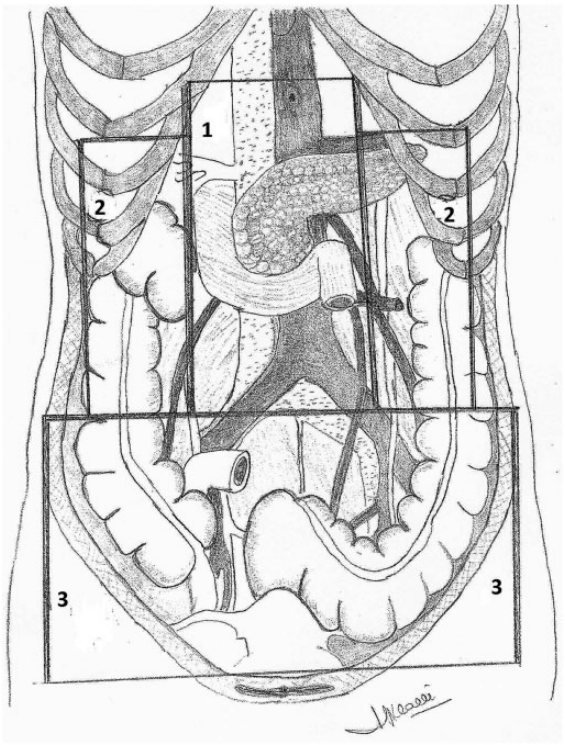
Continued haemodynamic instability is an indication for more definitive intervention either through selective intra-arterial embolisation, stent grafts or abdominal exploration.

Operative versus non-operative management approach depends on aetiology, size and the extent coupled with haemodynamic status and the foetal status. Sometimes it is the location and mechanism of injury that guide the decision to explore. Usually the site of retroperitoneal haematomas can give clue to the underling blood vessels involved; the areas can be the midline retroperitoneum (zone 1), the perinephric space (zone 2) and the pelvic retroperitoneum (zone 3) (see Figure 2).

The general principle adopted by surgeons is that injuries of zone 1 mandate exploration for both spontaneous and iatrogenic injuries because of the high likelihood of major vascular injury in this area, Zones 2 and 3 injuries require exploration for unstable patients mainly, whereas for haemodynamically stable ones, where retroperitoneal haematomas are nonexpanding without pressure symptoms, a conservative approach can be chosen. However, when the patient develops pressure symptoms called compartment syndrome resulting from the large retroperitoneal haematoma, it leads to open surgery.

Depending on haemodynamic stability of the patient, availability of management facility and the degree of blood loss, the vascular embolisation can be used as an alternative to open surgery for the management of retroperitoneal haematomas.

Figure 2. Abdominal cavity divided into three zones as a guide while managing retroperitoneal haematomas.



There have been supportive evidence where the use of intra-arterial embolisation is a recommended management undertaken with the help of the angiogram which shows active bleeding sites. The results seemed satisfactory in most of the cohort of cases published so far. However, after successful embolisation, there are reports of developing pressure symptoms or compartment syndrome. External fixation or angiographic embolisation can be employed in cases of blunt trauma to pelvis resulting in fractures. Diagnosis, clinical presentation and management of retroperitoneal haematomas cases reported in literature from 2007 to 2016 have been summarised in Table 1.

Conclusion

Retroperitoneal haematomas are a rare entity and different from commonly seen pelvic haematomas thereby demanding more awareness among obstetricians as they can cause maternal collapse and maternal morbidity by posing a serious threat to maternal and foetal wellbeing. Contrary to the usual concept that retroperitoneal haematomas only occur after caesarean section and also that vaginal and

instrumental deliveries can only cause vaginal, infralevator and supralevator haematomas, this review highlights that retroperitoneal haematomas can also occur after normal vaginal and instrumental deliveries. Also apart from mechanical causes, we mentioned the pathophysiology of unexpected spontaneous causes of retroperitoneal haematomas in pregnancy, which can be easily missed due to rarity in occurrence. retroperitoneal haematomas can occur during antenatal period, intrapartum and postpartum period. Existing guidelines of maternal collapse and morbidity during pregnancy and puerperium need to include retroperitoneal haematomas as one of the important causes of maternal shock or morbidity. New learning pathways should be opted for to increase awareness of retroperitoneal haematomas among obstetricians enabling them to reflect on its implications while managing retroperitoneal haematomas.

Declarations

Competing Interests: None declared.

Funding: None declared.

Ethics approval: Not required as no identifiable patient data.

Guarantor: HK.

Contributorship: JR conceived the idea, performed literature search, wrote the first draft and contributed to revisions. JR is the contact person with the editorial office. HK contributed to text writing, sketching the diagram (Figure 2), proof reading, editing and approved the final paper.

Acknowledgements: This article was presented as an oral presentation – S-155: 25th European Congress of EBCOG on 17 May 2017: Need to recognise retroperitoneal haematomas in pregnancy as important cause of maternal morbidity and mortality.

Provenance: Not commissioned; peer-reviewed by Judy Cohain.

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