Surgical management of a large peritoneal pseudocyst causing acute kidney injury secondary to abdominal compartment syndrome in a rare case of congenital absence of omentum during pregnancy

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Abstract

Complete congenital absence of the omentum is very rare with only one previously reported case. We present a unique case of the management of a pregnant woman with a large pelvic pseudocyst caused by complications related to congenital absence of omentum, resulting in acute kidney injury, likely secondary to acute compartment syndrome. This case highlights the importance of considering acute compartment syndrome in critically unwell pregnant women and reiterates the need to measure intra-abdominal pressure when clinically indicated. Given that pregnancy is in itself a state of intra-abdominal hypertension, obstetricians should maintain a high index of suspicion in the context of additional risk factors.

Keywords

High-risk pregnancy, nephrology, ultrasound, intensive care medicine, physiology

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Introduction

Peritoneal pseudocysts are benign fluid-filled structures that arise following the accumulation of fluid released from ovarian tissue that would otherwise be absorbed by peritoneum. When the structural integrity of peritoneum is disturbed by the presence of adhesions, peritoneal pseudocysts can occur.¹ Complete congenital absence of the omentum is rare. The only previous case report from 1958 described a case of double omental herniation secondary to 'almost complete absence of both greater and lesser omenta'.²

We present a unique case of the management of a pregnant woman with a large pelvic pseudocyst caused by complications related to congenital absence of omentum, resulting in acute renal failure, likely secondary to acute compartment syndrome (ACS).

Case report

A 34-year-old nulliparous woman, presented at 26 weeks' gestation with right-sided abdominal pain. She had previously undergone two laparotomies, the first due to sub-acute bowel obstruction when she was 19 years old, where complete absence of omentum was confirmed. The second was carried out due to the formation of an infected pseudocyst, which was drained and marsupialised. It was reportedly not possible to remove the cyst in its entirety as it was inseparable from small bowel, mesentery, caecum, sigmoid and parts of the colon. During follow-up, she remained clinically well but ultrasound revealed a stable right-sided pseudocyst measuring 5.6 cm in diameter maximally.

She conceived through in vitro fertilisation, necessitated due to tubal factor infertility, and presented at 26 weeks' gestation with right-sided abdominal pain. Ultrasound revealed that the pseudocyst had increased in size, measuring 14 cm in diameter. Conservative management was planned, but she re-presented at 27 weeks with worsening pain and vomiting. She had a leucocytosis ($14.2 \times 10^9/l$) and an elevated C-reactive protein (CRP; 69 mg/l). Intravenous cefuroxime and metronidazole were commenced, and intramuscular steroids for fetal lung maturation were given. She remained stable until 28 weeks'

gestation when she developed worsening pain, distension, an increased CRP (182 mg/l) and deteriorating renal function (creatinine 94 mmol/l). Magnetic resonance imaging confirmed a $14 \times 12 \times 18$ cm intraperitoneal cyst in addition to multiple dilated loops of small bowel and bilateral mild hydronephrosis. An ultrasound-guided drainage of the cyst was attempted but proved unsuccessful.

She subsequently developed progressively worsening acute kidney injury (AKI) (creatinine 248 mmol/l; estimated glomerular filtration rate $19 \text{ ml/min/1.73 m}^2$) despite aggressive intravascular filling. In view of this, the decision was made for operative delivery with drainage of the cyst at 29⁺² weeks' gestation. A midline laparotomy was performed. The bladder was densely adherent to the inferior aspect of the peritoneal cyst up to level of umbilicus. Superiorly, the transverse colon was adherent to the cyst and the underside of linea alba. The cyst was opened and evacuated. A midline classical incision was made to the uterus and a male infant was born by breech extraction, weighing 1370 g. Apgar scores were 8, 9 and 10 at 1, 5 and 10 min, respectively, and the arterial cord pH was 7.05 with a base excess of -11.6 mEq/L . The uterus was closed in three layers. A 5 cm defect in the bladder dome was identified and repaired in two layers. The cyst wall proved to be histologically benign, and no bacterial growth was identified.

Post operatively, her renal function dramatically improved and within 24 h, her creatinine had decreased to 129 mmol/l. By the fourth post-operative day, her renal function had normalised, and she had an uneventful recovery. Blood tests including anti-glomerular basement membrane antibodies, immunoglobulins, myeloperoxidase, Proteinase-3 and paraprotein were all normal, as was urinary Bence Jones Protein.

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Table 1. Aetiology of ACS.

Physiology	Primary ACS			Secondary ACS
	Increased intraluminal contents	Reduced abdominal wall compliance	Increased intra-abdominal contents	Capillary leak and/ excessive fluid accumulation
Examples	Intestinal obstruction, ileus, gastric paresis	Acute respiratory failure, burns, adhesions, major trauma, prone positioning	Pregnancy, abdomino-pelvic mass, ascites, haemoperitoneum, pneumoperitoneum	Sepsis, oliguria, hypotension, acidosis, massive fluid resuscitation

ACS: acute compartment syndrome.

Discussion

ACS is defined as a sustained intra-abdominal pressure (IAP) >20 mmHg, associated with new organ dysfunction.³ It is thought to contribute to renal failure through both pre-renal and renal mechanisms including renal vein compression, increased renal vascular resistance and the release of inflammatory cytokines, catecholamines, renin and angiotensin.⁴ ACS has been increasingly recognised as a potentially life-threatening complication in severely ill patients with a reported mortality rate of up to 68%.⁵ However, the condition is both poorly understood and rarely considered in pregnancy. The effect upon the fetus is also not well established, but it is likely sudden increases in IAP may cause fetal compromise due to reduced uterine perfusion. Whilst this has not been shown in a human model, data can be extrapolated from the ewe model during laparoscopy, where an IAP of 20 mmHg resulted in a reduction in placental blood supply by 61%.⁶

The evolution of ACS is associated with multiple risk factors as depicted in Table 1. Primary ACS occurs as a result of pathology in the abdomino-pelvic region, whereas secondary ACS results from pathology elsewhere in the body, causing capillary leak and/or excessive fluid accumulation within the abdomen.⁷

Intra-bladder pressure measurements are considered the gold standard for measuring IAP in non-pregnant subjects. A recent study examined the reproducibility of IAP measurements in pregnant women using the Foley Manometer Low Volume (FoleyManometer LV, Holtech Medical, Charlottenlund, Denmark).⁸ They found the measurements highly reliable and reproducible, and confirmed pregnancy is a state of intra-abdominal hypertension (IAH), which is relieved significantly upon delivery. During pregnancy, the abdominal wall is gradually stretched, and the progestogenic hormonal influence increases abdominal wall compliance. These compensatory physiological adaptations allow pregnant women to withstand pregnancy induced IAH, whereby such pressures in non-pregnant woman may cause significant harm.⁹

In this case, it is likely a sequence of interrelated events overcame these protective mechanisms to contribute to the development of ACS. The combination of a large pelvic pseudocyst during pregnancy, along with the subsequent development of subacute bowel obstruction would all raise IAP. This would be compounded by reduced abdominal cavity compliance due to the dense intra-abdominal adhesions that arose as a consequence to the congenital absence of omentum. The onset of AKI signifies the likely onset of ACS, although in the absence of recorded pressures this cannot be confirmed. Aggressive fluid resuscitation likely aggravated the already fulminant primary ACS with an additional secondary component, demonstrated by further deterioration in renal function.

ACS was only considered retrospectively in this case, and as such, the IAP was not measured. It is, however, of the authors' opinion that ACS is most likely, as a diagnosis of exclusion. A pre-renal cause is doubtful as she was not clinically dehydrated and deteriorated with aggressive intravascular filling. The screen for primary renal disease was negative, and she did not receive nephrotoxic drugs. An obstructive picture was considered, but the mild degree of hydronephrosis seen

on imaging was in keeping with normal physiological adaptation to pregnancy, not an obstructive cause significant enough to cause severe AKI. The deterioration in renal function was felt unlikely to be related to preeclampsia, as her blood pressure remained normal, there was an absence of significant proteinuria and liver function and platelets remained normal throughout.

Expert recommendation is that IAP is measured in any patient admitted to intensive care, or, with evidence of clinical deterioration with more than two risk factors for IAH, as demonstrated in this case.⁷ If a sustained IAP >12 mmHg is identified in non-pregnant women, the algorithm for managing IAH should be followed including treatment to reduce causative factors.⁷ Sustained IAP >20 mmHg with new organ failure constitutes ACS, which would necessitate definitive surgical treatment with abdominal decompression. Unfortunately, literature contains scarce case reports of ACS during pregnancy, which is often only recognised in critically unwell pregnant women.^{10,11} Furthermore, the lack of normal values for IAP in pregnancy makes recommendations for management problematic. Clinically recognising ACS can also be challenging due to diagnostic similarity with preeclampsia including non-specific symptoms such as abdominal pain and oliguria.¹² It has also been suggested that IAH may play an integral role in the aetiology of preeclampsia, initiating a multisystem cascade of events as a consequence to occluded venous return caused by raised IAP.¹³ This limits abdominal and pelvic organ perfusion including the kidneys and placenta.¹³ Subsequent renal hypoperfusion activates the renin-angiotensin system, which ultimately results in systemic hypertension whilst placental hypoperfusion in turn causes placental ischaemia and thus may impact fetal growth and wellbeing.

Management of pseudocysts in pregnancy should be individualised and include a multidisciplinary team comprising of obstetricians, gynaecologists, general surgeons and radiologists. Conservative treatment is mostly commonly indicated, with frequent monitoring of maternal and fetal wellbeing, both clinically and radiologically with ultrasonography. In symptomatic patients or in pseudocysts of large or rapidly increasing size, ultrasound-guided drainage should be undertaken with laparoscopic or open surgical management reserved for cases not amenable to minimally invasive drainage or in critical cases as described in this case. Surgical management should include complete pseudocyst excision where possible, to reduce the potential for recurrence in the future.

This case highlights the importance of considering ACS in critically unwell pregnant women and reiterates the need to measure the IAP when clinically indicated. Pregnancy is in itself a risk factor for ACS and despite protective physiological adaptations, obstetricians should maintain a high index of suspicion in the context of additional risk factors. Appropriate training is necessary to raise awareness of this potentially life threatening, largely underdiagnosed condition.

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Guarantor

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BJ conceived the original idea and wrote the article. TH helped write the article and helped revise the final draft. JT contributed to case report design and participated in final draft revision.

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