

## CASE REPORT

# Giant Hemorrhagic Adrenal Pseudocyst in Pregnancy

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## ABSTRACT

Adrenal cysts are a rare diagnosis, particularly in pregnancy. We report the case of a 32-year-old pregnant woman with an adrenal pseudocyst in her 28th week of pregnancy. This represents the 13th case of an adrenal pseudocyst in pregnancy reported in the literature. The patient presented to our facility complaining of abdominal pain, believing she could be in preterm labor. After further evaluation and surgery, she was diagnosed with a left adrenal pseudocyst. In this report, we will not only describe our case but also discuss the relevance of the findings to what is already present in the literature. Our case report is an example of multidisciplinary approach to a pregnant patient with an abdominal/pelvic mass. Inter-professional collaboration allowed successful surgical intervention that resulted in an excellent outcome for the mother and term delivery of child.

**Keywords:** Adrenal pseudocyst, Abdominal pain, Pregnancy, Preterm labor, Ultrasound, Color Doppler ultrasound.

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## INTRODUCTION

Adrenal cysts are rare findings and quite foreign to the literature. As of early 2012, there have been less than 100 cases of hemorrhagic pseudocysts reported with the giant variety being even rarer.<sup>1</sup> Doran wrote in 1908 that Greiselius of Vienna removed the first documented adrenal cyst in 1670.<sup>2</sup>

There are 4 different types of adrenal cysts—parasitic cysts (7%), epithelial cysts (9%), pseudocysts (39%) and

endothelial cysts (45%). The latter two are vascular in etiology. The pathogenesis remains somewhat unclear but is thought to be secondary to repeated episodes of trauma, infection or bleeding.<sup>3</sup> Adrenal cysts fall under the umbrella of adrenal incidentalomas; however, adrenal cysts only comprise 6% of all worked-up adrenal incidentalomas.<sup>4</sup>

We report the case of a 32-year-old woman in her 28th week of pregnancy that presented with what was originally thought to be an adnexal mass and was taken to the operating room for definitive diagnosis and exploration. Upon definitive diagnosis, confirmation of a giant hemorrhagic adrenal pseudocyst was made, yielding the 13th reported case of a giant hemorrhagic adrenal pseudocyst in pregnancy.<sup>5</sup>

## CASE REPORT

A 32-year-old G6 P3023 in her 28th week of pregnancy with one prenatal visit at 20 weeks in Mexico was admitted to the labor and delivery unit at our facility complaining of abdominal pain. The patient had a past medical history significant for HPV, herpes and chlamydia. She had a past surgical history of cervical cryotherapy secondary to HPV as well as two dilation and curettages secondary to incomplete abortions.

At presentation, she was taking no medications and evaluation for gonorrhea/chlamydia was negative. At our facility she was diagnosed with gestational diabetes. An ultrasound of the abdomen/pelvis was performed to evaluate fetal well-being and revealed a well-delineated 23 × 17 × 15 cm complex abdominal mass that appeared to originate from the left adnexa (Fig. 1). The mass was qualified as multilocular—solid, with no evidence of papillary protrusions. Posterior enhanced through-transmission was seen, signifying the predominant cystic nature of the lesion. Within the mass there were numerous septations measuring from 1 to 10 mm, which contributed to reticular appearance of the mass. Color Doppler showed no flow in these septations and/or solid parts. There was no free fluid in the *cul-de-sac*. The gynecologic oncology team was consulted and the decision was made to take the patient to the operating room due to the risk that based on morphologic assessment, this mass could be malignant. Because of lack of perfusion on Doppler, adnexal torsion was considered in differential diagnosis. After antenatal corticosteroids were administered, an exploratory laparotomy was performed. After freeing adhesions, the mass was found to be free of any

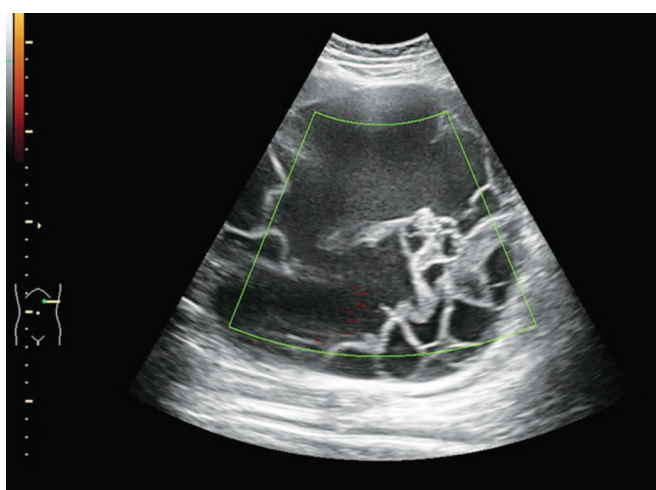
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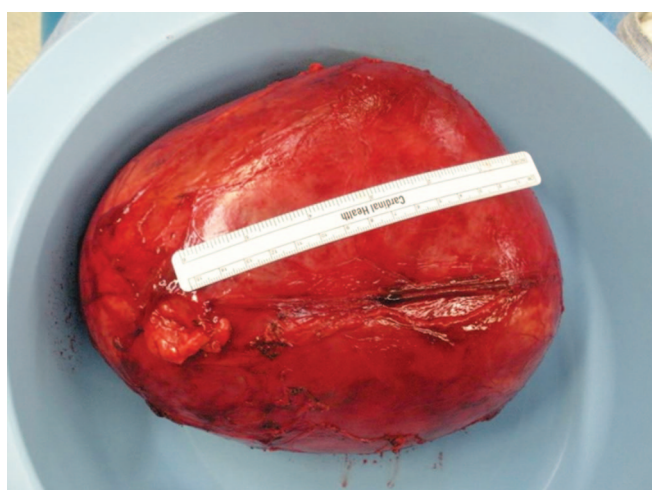
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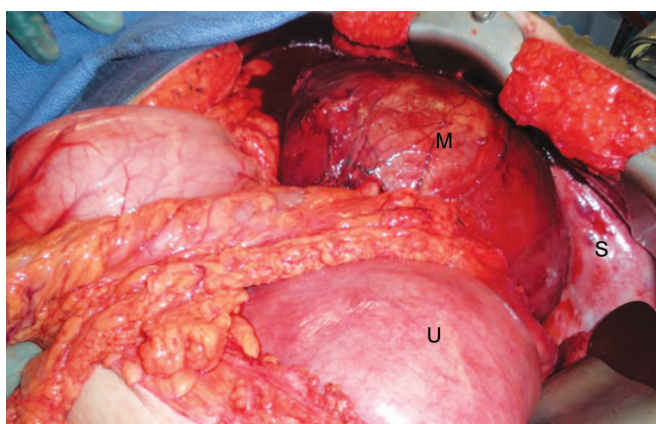
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**Fig. 1:** Power Doppler ultrasound showing complex abdominal mass. Note thickened septations and solid parts



**Fig. 3:** Left adrenal mass—measuring 17 × 15 × 12.5 cm and weighing 3 kg



**Fig. 2:** Intraoperative view of the abdomen with exposed adrenal pseudocyst/mass (M), gravid uterus (U) and spleen (S)

attachments to the pelvic anatomy and appeared to originate from the retroperitoneum. An intraoperative consult was placed for the general surgery team.

The surgeon noted that the mass was adherent to the posterior capsule of the spleen yielding the suspicion that this mass could be originating from the lower pancreas. The lesser sac was explored and the gastrocolic ligament was opened, demonstrating that the mass was originating from another retroperitoneal structure. Further dissection revealed that the lesion was well-encapsulated and full of fluid and originated from the left flank. The mass was also adherent to the diaphragm. This attachment was freed and the base of the tumor was now determined to originate from the left adrenal gland (Fig. 2). The mass was intimately involved with the majority of the adrenal gland so the entire left adrenal gland was removed with the mass. The left kidney appeared anatomically normal.

The specimen was sent to pathology for frozen sections and was noted to be 17 × 15 × 12.5 cm and weighed 3 kg. Frozen section demonstrated a distended adrenal

gland with an adjacent cystic structure with a cavity filled with hemorrhagic material. No tumor masses or papillary projections were found (Fig. 3).

The patient remained stable throughout the surgery and required a splenorrhaphy for a small tear to the splenic capsule during dissection. Fetal heart monitoring throughout the case remained within normal limits. A 19-French Jackson Pratt drain was left in place.

The patient was transferred to the ICU for monitoring and to follow her hemoglobin/hematocrit levels. She remained stable postoperatively and suffered no further complications of her procedure. She had no evidence of adrenal insufficiency and was discharged on postoperative day 7. She delivered an anatomically normal, healthy baby 3 months following surgery.

Histopathologic examination of the specimens submitted failed to identify adrenal neoplasms but did reveal numerous hemosiderin laden macrophages and a single layer of fibroblasts encompassing the cyst. The cyst contained blood and partially organized fibrin. The diagnosis of a giant hemorrhagic adrenal pseudocyst was confirmed.

## DISCUSSION

Adrenal pseudocysts are an exceedingly rare finding and this case represents the rarity of this finding, particularly in pregnancy. The incidence of adrenal cysts has been steadily increasing and this is thought to be due to the increased availability and use of CT scans.<sup>6</sup>

The majority of all adrenal pseudocysts are benign (93%). The risk of malignancy is positively correlated with the size of the pseudocyst, particularly if the pseudocyst is greater than 6 cm in its greatest dimension.<sup>6</sup>

Adrenal pseudocysts are typically asymptomatic; however, if symptomatic the most common presentation is

**Table 1:** Demographics and clinical data of the 13 pregnant patients with adrenal pseudocysts<sup>5,17-26</sup>

<i>First author, Year<sup>ref</sup></i>	<i>Age (yrs)</i>	<i>Gestational age</i>	<i>Size (cm) and/or weight (gm)</i>	<i>Side and type</i>	<i>Treatment</i>	<i>Pregnancy outcome</i>
Karaman, 2011 <sup>5</sup>	40	20 weeks	2 × 15 cm	Left adrenal (hemorrhagic)	Complete cystectomy, left adrenalectomy	Vaginal term delivery
Thomson, 1966 <sup>18</sup>	23	8 weeks	20 × 12 cm	Right adrenal (hemorrhagic)	Complete cystectomy, right adrenalectomy	Vaginal term delivery
Osborne, 1974 <sup>19</sup>	28	17 weeks	15 cm (mean diameter)	Right adrenal (hemorrhagic)	Complete cystectomy, right adrenalectomy, right nephrectomy	Unknown
Costandi, 1975 <sup>20</sup>	32	2nd trimester	12 cm (mean diameter), 360 gm	Right adrenal (non-hemorrhagic)	Complete cystectomy, right adrenalectomy, right nephrectomy	Unknown
Rao, 1976 <sup>21</sup>	27	12 weeks	11,500 gm	Right adrenal (hemorrhagic)	First surgery: exploratory laparotomy, 2nd surgery: complete cystectomy	Vaginal term delivery
Uretzky, 1978 <sup>22</sup>	29	8 weeks	20 cm (mean diameter)	Right adrenal (non-hemorrhagic)	Complete cystectomy, partial right adrenalectomy	Elective termination
Bartlett, 1995 <sup>23</sup>	33	14 weeks	15 × 11 cm, 365 gm	Right adrenal (hemorrhagic)	Unsuccessful percutaneous drainage followed by complete cystectomy with right adrenalectomy	Vaginal term delivery
Trauffer, 1996 <sup>24</sup>	33	14 weeks	20 × 11 × 14 cm	Right adrenal (hemorrhagic)	Unsuccessful percutaneous drainage followed by complete cystectomy with right adrenalectomy	Vaginal term delivery
Tait, 1997 <sup>25</sup>	28	26 weeks	20 × 40 cm	Right adrenal (non-hemorrhagic)	Complete cystectomy	Preterm vaginal delivery
Papaziogas, 2006 <sup>11</sup>	27	28 weeks	11 × 12 cm	Left adrenal (hemorrhagic)	First surgery: exploratory laparotomy, 2nd surgery: complete cystectomy with left adrenalectomy	Cesarean section at term
Sivasankar, 2006 <sup>26</sup>	20	8 weeks	20 cm (mean diameter)	Right adrenal (hemorrhagic)	Complete cystectomy	Elective termination
Sivasankar, 2006 <sup>26</sup>	24	20 weeks	14 × 16 cm	Right adrenal (hemorrhagic)	Complete cystectomy	Preterm vaginal delivery
Present case	32	28 weeks	12.5 × 15 × 17 cm, 3,000 gm	Left adrenal (hemorrhagic)	Complete cystectomy, left adrenalectomy; splenorrhaphy	Vaginal term delivery

abdominal or flank pain due to compression of surrounding structures. Other symptoms include shortness of breath or fever/chills. The most common sign is a palpable mass on exam or sensation of abdominal fullness. Other documented signs include hypertension, shock or superimposed infection.<sup>3,7,8</sup> Adrenal pseudocysts causing signs of adrenal hypo- or hyperfunction are very uncommon, however.<sup>4</sup> There have been other documented cases in pregnancy of adrenal pseudocysts presenting with an acute abdomen and this is thought to be secondary to hemorrhage occurring into the cyst.<sup>7</sup>

In the obstetric population, the diagnosis of an adrenal pseudocyst is rarely established preoperatively and the provisional diagnoses have included ectopic pregnancy with massive hemorrhage, ovarian vein syndrome, gallbladder

hydros, ovarian cyst undergoing intermittent torsion, cyst of the right lobe of the liver, cyst arising from the right adnexa, bleeding hepatic adenoma or retroperitoneal cyst, bleeding liver tumor and a mesenteric cyst.<sup>8,9</sup>

For diagnosis of adrenal pseudocysts, the gold standard remains computed tomography, although ultrasound and MRI are also utilized. MRI has high sensitivity for even small masses and is superior for tissue characterization.<sup>6</sup>

In our pregnant patient the clinical features included pelvic pain and abdominal mass. Complex appearance of the lesion, mixed echogenicity, thick septations and solid parts originating from disintegrated blood clot gave an appearance of ovarian neoplasm. However, color Doppler revealed no flow in these septations and solid parts, because fibrin strands and blood clot do not contain blood vessels.



Acute onset of abdominal pain may mimic other gynecologic conditions such as adnexal torsion. Absence of ovarian blood flow on Doppler sonography is a good predictor of adnexal torsion, which was considered in the differential diagnosis.<sup>10</sup>

Treatment of adrenal pseudocysts remains surgical removal for all tumors larger than 5 cm, if there is any suspicion of malignancy or if the tumor is hormonally active.<sup>11-16</sup> There is also evidence to recommend surgery if the tumor is at least 4 cm in size. If the tumor is less than 4 cm in size, patients should undergo a repeat CT scan in 3 months and be monitored for at least 18 months.<sup>6</sup>

Table 1 illustrates demographics and clinical data of the 13 pregnant patients with adrenal cysts reported in the literature.<sup>5,10,18-26</sup> Although these tumors are exceedingly rare in pregnancy, a multidisciplinary approach allowed successful surgical intervention that resulted in an excellent outcome for the mother and term delivery of child. A high index of suspicion for retroperitoneal tumors and adrenal pseudocysts should be present in every patient evaluated for adnexal masses, especially during pregnancy.

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