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# 妊娠肾上腺囊肿伴出血报道1例并文献分析

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## 摘　要

**肾上腺囊肿是一种良性病变，在临床上并不常见。一般无明显临床症状和体征。通常在影像学诊断、手术或尸解中意外发现。妊娠期合并肾上腺囊肿比较罕见，在没有出现并发症时，常表现为盆腔包块，确定囊肿起源时有时具有挑战性，这给临床医生带来了诊断上和治疗上的困难。一名26岁孕妇在孕14周零4天出现腹部胀痛1月余入院。超声显示患者右上腹约有20 cm的囊实性包块。肿瘤标志物及其它实验室检查均正常。MRI检查显示右上腹腹膜后囊实性巨大肿块，合并出血。计划进行剖腹手术。在剖腹手术中，观察到左侧上腹部腹膜后直径约20 cm囊性为主包块，子宫和双侧附件未见明显异常，囊肿起源于右肾上腺并切除。**

## 关键词

**肾上腺囊肿，妊娠，出血，病例报告**

# Pregnancy Adrenal Cyst with Hemorrhage: A Case Report and Literature Analysis

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## Abstract

**Objective: Adrenal cysts are benign lesions that are not common in clinical practice, generally no**

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**obvious clinical symptoms and signs. It is usually found by accident during radiographic diagnosis, surgery, or autopsy. Adrenal cysts during pregnancy are rare and often present as pelvic masses in the absence of complications. Determining the origin of cysts is sometimes challenging, which poses diagnostic and therapeutic difficulties for clinicians. A 26-year-old pregnant woman developed abdominal distension at 14 weeks and 4 days of gestation more than 1 month after admission. Ultrasound revealed a cysticular mass of about 20 cm in the right upper abdomen. Tumor markers and other laboratory tests were normal. MRI examination revealed a large posterior peritoneal cystic mass with hemorrhage. A laparotomy is planned. During laparotomy, a cystic mass with a diameter of about 20 cm was observed in the left epigastric retroperitoneal mass. No obvious abnormalities were observed in the uterus and bilateral adnexa. The cyst originated from the right adrenal gland and was excised.**

## Keywords

**Adrenal Cyst, Pregancy, Bleeding, Case Report**



## 1. 背景介绍

肾上腺囊肿是一种少见病之一，文献报道发病率约为 0.06%~0.018% [1]。大多数是良性的，属于无功能，不容易被发现。近几年随着肿瘤筛查的开展和影像学技术的进步，肾上腺囊肿检出率似乎在增长。当囊肿快速肿大、出血、感染、囊肿内容物破裂或压迫邻近器官时出现临床症状。我们报告了一例巨大肾上腺囊肿，在妊娠早期由于囊内出血出现急腹症并成功切除，术后出现一些并发症并及时处理，顺利出院。

## 2. 病例报告

一名 26 岁的初产妇急诊入院，孕 14 周零 4 天，病人主诉腹胀伴“岔气”样疼痛 1 月余。在检查中，腹部触诊子宫如孕 3 个半月大小，腹部超声提示右上腹探及一大小约为 201 × 140 mm 囊实性包块，边界欠清，囊性部分内透声可，实性部分内回声均，内未见明显血流信号，肝脏及肾脏受压移位。提示：右上腹囊实性包块。腹部磁共振检查右上腹部腹膜后囊实性巨大肿块，提示合并出血。实验室指标和肿瘤标志物均未见明显异常。术前充分评估患者，积极请相关科室会诊，计划进行剖腹探查，告知患者手术风险。在普外科和泌尿外科帮助下，术中发现左侧上腹部腹膜后直径约为 20 cm 囊性为主的包块，位于肾脏水平至肝下，上方达膈肌，内侧与下腔静脉长约 10 cm 关系密切，见肿瘤内侧及外侧均有黄色似肾上腺组织，考虑肾上腺肿瘤可能性大，穿刺抽吸暗红色囊内液体约 4000 毫升余，切除肿瘤及右肾中上份肾周筋膜及脂肪囊，放置引流管 2 根。手术顺利，术后第四天拔除引流管。术中渗血 4000 毫升，输血浆 780 毫升，悬浮红细胞 12 U，冷沉淀 10 U。考虑到手术创面大、术中渗血多和妊娠期高凝状态，术后转入 ICU。术后第一天出现腹腔感染、中度贫血、失血性肝损伤、心肌缺血缺氧性损伤。白细胞 18.02 ($10^9$/L)，中性粒细胞比率 87.6%，血红蛋白 78.0 g/L，肌钙蛋白 T 0.046 ng/ml，降钙素原 0.41 ng/ml，非结合胆红素 23.4 umol/L，结合胆红素 62.6 umol/L，总胆红素 62.6 umol/L，AST 493.0 U/L，ALT 154.5 U/L。依据临床经验给予抗感染治疗、纠正电解质紊乱。营养科评估，NRS2002 营养风险筛查提示：6 分。建议急

性期能量按 20~25 kcal/kg·d，以肠内营养联合肠外营养的方式进行营养支持。输注红细胞 2 U 纠正贫血。同时患者出现巩膜和结膜黄染，请相关科室会诊，给予谷胱甘肽等药物保肝降酶处理。复查实验室检查出现低白蛋白，白蛋白 25.8 g/L，总蛋白 45.2 g/L，给予白蛋白纠正低蛋白血症，继续给予红细胞 2 U 纠正贫血。术后第三天转入普通病房。肿块的病理显示(肾上腺)符合囊肿伴出血，直径约 22 cm，重 750 g，大部已破，切开多房，内含暗红色液体，囊壁大部附血块样物，壁厚 0.2~1.6 cm。术后第十天，患者顺利出院，术后未发现激素异常。患者由于考虑到手术时间长、术中出血多和灌注不足对胎儿有很大创伤及使用药物担心胎儿致畸，选择在出院后一周行引产手术。

## 3. 讨论

肾上腺囊肿是一种少见的异质性病变，可以在任何年龄段被诊断，但最常见的年龄段为 30~60 岁，男女比例为 1:3，多数属于无功能性的[2]。囊肿大小差异很大，直径从几毫米到几十厘米不等，可能是单房性或多房性，通常是单侧的，大约有 8%~15%的病例为双侧[3]。目前，仍然采用 Foster 等[4]将肾上腺囊肿分为 4 种类型：1) 内皮性囊肿，约占 45%，其中包括血管瘤样囊肿和淋巴管瘤样囊肿，其中以淋巴管瘤样囊肿最为多见；2) 假性囊肿，约占 39%，囊壁由纤维组织构成，无被覆内皮和上皮细胞，主要由肾上腺出血导致；3) 上皮性囊肿，约占 9%，内皮被覆柱状上皮；4) 寄生虫性囊肿，约占 7%，多由包虫感染，常伴子囊且囊壁厚伴钙化。我们在 Medline (通过 Pubmed 界面访问)、科学网等，使用关键词“肾上腺囊肿”、“肾上腺假性囊肿”和“妊娠”，对已经发表过的关于妊娠期肾上腺假性囊肿的英文文献进行系统回顾，我们发现有 14 例病例报告了妊娠期合并肾上腺假性囊肿[5]。我们的患者成为第 15 例。报道妊娠期肾上腺假性囊肿年龄(平均 29.4 岁，范围 20~41 岁)，与我们患者年龄接近；囊肿直径均大于 10 cm (平均大小约 20.7 cm，范围 10~40 cm)，也显示出相似大小；囊肿出现破裂出血(12/15 例，80%)，本例病理提示囊肿伴出血，通常囊肿更常见于妊娠中期(10/15 例，67%)，少见于妊娠早期(4/15 例，27%)妊娠晚期(1/15 例，7%)，患者孕 14 周零 4 天，处于妊娠早期；囊肿通常起源于右侧肾上腺(11/15 例，73%)，与本例相似，这些都进行手术完整切除，其中 2 例患者曾尝试将经皮引流作为一线治疗，但均未成功，随后需要进行完全切除[6] [7]。有 1 例患者是在期待妊娠结束后行腹腔镜下囊肿完全切除[8]。15 例中有 13 例报告了产科结局。其中正常分娩 7 例[6]-[12]，4 例患者选择终止妊娠：其中 3 例患者因担心胎儿致畸而选择流产[11] [13] [14]，这与本例报道一样，因担心手术及药物对胎儿影响大选择在出院后 1 周流产，1 例患者因母体反复出现抗凝血治疗的肺栓塞而终止治疗[13]，1 例患者在孕 34 周因先兆子痫导致死产[5]，1 例发生早产[15]。在这 15 例病例中，均未在正常产检中发现肾上腺假性囊肿，突显孕妇在正常产检甚至术前难以发现肾上腺疾病。在妊娠期，超声是诊断肾上腺囊肿的重要辅助手段，但诊断价值有限[16]。MRI 对于腹部肿块有着重要的诊断价值。在怀孕期间，由于雌激素水平高，肾上腺囊肿可能表现出快速增长和囊壁结缔组织疏松。因此，妊娠期出现急性上腹痛和侧面疼痛或失血性休克的表现时，应警惕肾上腺假性囊肿或发生破裂出血[10] [11]。还应该注意的是，妊娠本身和分娩过程可能会导致肾上腺假性囊肿的破裂出血[17]。Ghandur Mnaymneh 等人[18]指出，肾上腺囊生长模仿恶性肿瘤行为，倾向于粘附周围组织器官生长，常完全切除病侧肾上腺。本例患者，我们在术中发现囊肿内侧与下腔静脉长约 10 cm 关系密切，下极与肾脏关系密切，这增加手术难度，使得术中出血较多及术后出现一些并发症。我们在术中及时给予输血及术后对症治疗，使患者平稳恢复和顺利出院。患者考虑到手术时间长、术中出血多和灌注不足对胎儿会有很大创伤选择出院后流产。总之，妊娠期出血性肾上腺假性囊肿的治疗仍然是一项具有挑战性、仍有争议的外科任务。我们报告了一例妊娠期肾上腺假性囊肿合并囊内出血的病例。手术切除是治疗这种罕见疾病最有效的方法，我们针对患者术后并发症，及时给予对症治疗，帮助患者顺利出院(见表 1)。

**Table 1.** Demographic and clinical data of 15 pregnant women with adrenal pseudocyst
**表 1.** 15 例肾上腺假性囊肿孕妇的人口学和临床资料

| First author year | Age (years) | Period of gestation | Localization | Size, weight, or volum | Treatment | Outcome of pregancy |
|---|---|---|---|---|---|---|
| Thompson 1996 | 23 | 8 weeks | Right adrenal | 20 × 12 cm, 21 Hemorrha gic cyst | Complete cyst excision with right adrenalectomy | Normal delivery |
| Osborne 1975 | 28 | 17 weeks | Right adrenal | 15 cm, 360 g nonhemorrha gic cyst | Complete cyst excision with right nephrectomy and adrenalectomy | Unkonow |
| Costandi 1975 | 32 | 2nd trimester | Right Adrenal | 12 cm, 360 g nonhemorrha gic cyst | First operation: nephrectomy Second operation: cholecystectomy and complete cyst excision with right adrenalectomy | Unknow |
| Rao 1976 | 27 | 12 weeks | Right adrenal | 11.5 kg Hemorrha gic cyst | First operation: exploratory laparotomy Second operation: complete cyst excision | Normal delivery |
| Uretzky 1978 | 29 | 2nd month | Right adrenal | 20 cm, 21 nonhomorrha gic cyst | Complete cyst excision with partial right adrenalectomy | Therapeutic abortus |
| Bartlett 1995 | 33 | 14 weeks | Right adrenal | 15 × 11 cm, 365 g Hemorrha gic cyst | Unsuccessful percutaneous drainage followed by complete cyst excision with partial right adrenalectomy | Normal delivery |
| Trauffer 1996 | 33 | 14 weeks | Right adrenal | 20 × 11 × 14 cm, Hemorrha gic cyst | Unsuccessful percutaneous drainage followed by complete cyst excision with partial right adrenalectomy | Normal delivery |
| Tait 1997 | 28 | 26 weeks | Right adrenal | 40 × 20 cm, 41 nonhemorrha gic cyst | Complete cyst excision | Preterm spontaneous vaginal delivery |
| Papaziogas 2006 | 27 | 28 weeks | Left adrenal | 12 × 10.9 cm Hemorrha gic cyst | First operation: exploratory laparotomy Second operation: complete cyst excision with left adrenalectomy | Normal delivery with cesarean |
| Sivasankar 2006 | 20 | 8 weeks | Right adrenal | 20 cm, 51 Hemorrha gic cyst | Complete cyst excision | Therapeutic abortus |
| Sivasankar 2006 | 24 | 20 weeks | Right adrenal | 14 × 16 cm, 31 Hemorrha gic cyst | Complete cyst excision | Therapeutic abortus |
| Karaman 2011 | 40 | 20 weeks | Left adrenal | 20 × 15 cm, 31 Hemorrha gic cyst | Complete cyst excision with Left adrenalectomy | Stillbirth in the 34$^{th}$ gestational week |
| Angelico 2013 | 30 | 20 weeks | Left adrenal | 10 × 7 × 10 cm Hemorrha gic cyst | Left adrenal cyst excision with laparoscopy after 3 month delivery | Normal delivery |
| Mandato 2018 | 41 | 22 weeks | Left adrenal | 23.12 × 18.64 cm Hemorrha gic cyst | Complete cyst excision with Left nephrectomy and adrenalectomy | Normal delivery |
| Present case | 26 | 14 weeks | Right adrenal | 22 × 16 × 10 cm 750 g | First operation: exploratory laparotomy Second operation: complete cyst excision with adrenalectomy | Voluntary Abortus |

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