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Contents

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REVIEW

- 3621 Autoimmunity as the comet tail of COVID-19 pandemic
Talotta R, Robertson E
- 3645 Gender medicine: Lessons from COVID-19 and other medical conditions for designing health policy
Machluf Y, Chaïter Y, Tal O

MINIREVIEWS

- 3669 Complexities of diagnosis and management of COVID-19 in autoimmune diseases: Potential benefits and detriments of immunosuppression
Georgiev T, Angelov AK

ORIGINAL ARTICLE

Retrospective Study

- 3691 Emergency surgery in COVID-19 outbreak: Has anything changed? Single center experience
D'Urbano F, Fabbri N, Koleva Radica M, Rossin E, Carcoforo P
- 3697 Somatostatin receptor scintigraphy in the follow up of neuroendocrine neoplasms of appendix
Saponjski J, Macut D, Sobic-Saranovic D, Ognjanovic S, Bozic Antic I, Pavlovic D, Artiko V
- 3708 Efficacy of stool multiplex polymerase chain reaction assay in adult patients with acute infectious diarrhea
Ahn JS, Seo SI, Kim J, Kim T, Kang JG, Kim HS, Shin WG, Jang MK, Kim HY
- 3718 Comparison of gemcitabine plus nab-paclitaxel and FOLFIRINOX in metastatic pancreatic cancer
Han SY, Kim DU, Seol YM, Kim S, Lee NK, Hong SB, Seo HI
- 3730 Shear wave elastography may be sensitive and more precise than transient elastography in predicting significant fibrosis
Yao TT, Pan J, Qian JD, Cheng H, Wang Y, Wang GQ
- 3743 Radioactive ¹²⁵I seed implantation for locally advanced pancreatic cancer: A retrospective analysis of 50 cases
Li CG, Zhou ZP, Jia YZ, Tan XL, Song YY
- 3751 Active surveillance in metastatic pancreatic neuroendocrine tumors: A 20-year single-institutional experience
Gao HL, Wang WQ, Xu HX, Wu CT, Li H, Ni QX, Yu XJ, Liu L
- 3763 Clinical efficacy of tocilizumab treatment in severe and critical COVID-19 patients
Zeng J, Xie MH, Yang J, Chao SW, Xu EL

- 3774** Phosphatidylinositol-3,4,5-trisphosphate dependent Rac exchange factor 1 is a diagnostic and prognostic biomarker for hepatocellular carcinoma

Cai Y, Zheng Q, Yao DJ

Observational Study

- 3786** Awareness and attitude of fecal microbiota transplantation through transendoscopic enteral tubing among inflammatory bowel disease patients

Zhong M, Sun Y, Wang HG, Marcella C, Cui BT, Miao YL, Zhang FM

CASE REPORT

- 3797** Cauda equina arachnoiditis – a rare manifestation of West Nile virus neuroinvasive disease: A case report

Santini M, Zupetic I, Viskovic K, Krznaric J, Kutlesa M, Krajinovic V, Polak VL, Savic V, Tabain I, Barbic L, Bogdanic M, Stevanovic V, Mrzljak A, Vilbic-Cavlek T

REVIEW

- 3804** Portal gas in neonates; is it always surgical? A case report

Altokhais TI

CASE REPORT

- 3808** Large lingual heterotopic gastrointestinal cyst in a newborn: A case report

Lee AD, Harada K, Tanaka S, Yokota Y, Mima T, Enomoto A, Kogo M

- 3814** Osteochondral lesion of talus with gout tophi deposition: A case report

Kim T, Choi YR

- 3821** Traumatic neuroma of remnant cystic duct mimicking duodenal subepithelial tumor: A case report

Kim DH, Park JH, Cho JK, Yang JW, Kim TH, Jeong SH, Kim YH, Lee YJ, Hong SC, Jung EJ, Ju YT, Jeong CY, Kim JY

- 3828** Autoimmune hepatitis in a patient with immunoglobulin A nephropathy: A case report

Jeon YH, Kim DW, Lee SJ, Park YJ, Kim HJ, Han M, Kim IY, Lee DW, Song SH, Lee SB, Seong EY

- 3835** Diagnosis of an actively bleeding brachial artery hematoma by contrast-enhanced ultrasound: A case report

Ma JJ, Zhang B

- 3841** Lung adenocarcinoma harboring rare epidermal growth factor receptor L858R and V834L mutations treated with icotinib: A case report

Zhai SS, Yu H, Gu TT, Li YX, Lei Y, Zhang HY, Zhen TH, Gao YG

- 3847** Gastroduodenitis associated with ulcerative colitis: A case report

Yang Y, Li CQ, Chen WJ, Ma ZH, Liu G

- 3853** Majocchi's granuloma caused by *Trichophyton rubrum* after facial injection with hyaluronic acid: A case report

Liu J, Xin WQ, Liu LT, Chen CF, Wu L, Hu XP

- 3859** Novel deletion mutation in Bruton's tyrosine kinase results in X-linked agammaglobulinemia: A case report
Hu XM, Yuan K, Chen H, Chen C, Fang YL, Zhu JF, Liang L, Wang CL
- 3867** Multidisciplinary treatment of life-threatening hemoptysis and paraplegia of choriocarcinoma with pulmonary, hepatic and spinal metastases: A case report
Lin YY, Sun Y, Jiang Y, Song BZ, Ke LJ
- 3875** Diagnostic value of ultrasound in the spontaneous rupture of renal angiomyolipoma during pregnancy: A case report
Zhang T, Xue S, Wang ZM, Duan XM, Wang DX
- 3881** Gallbladder sarcomatoid carcinoma: Seven case reports
Qin Q, Liu M, Wang X
- 3890** Surgical strategy used in multilevel cervical disc replacement and cervical hybrid surgery: Four case reports
Wang XF, Meng Y, Liu H, Hong Y, Wang BY
- 3903** Diagnosis and treatment of an elderly patient with 2019-nCoV pneumonia and acute exacerbation of chronic obstructive pulmonary disease in Gansu Province: A case report
He TP, Wang DL, Zhao J, Jiang XY, He J, Feng JK, Yuan Y
- 3911** Diagnosis and treatment of mixed infection of hepatic cystic and alveolar *echinococcosis*: Four case reports
A JD, Chai JP, Wang H, Gao W, Peng Z, Zhao SY, A XR

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The primary aim of *World Journal of Clinical Cases* (WJCC, *World J Clin Cases*) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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Diagnostic value of ultrasound in the spontaneous rupture of renal angiomyolipoma during pregnancy: A case report

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Abstract

BACKGROUND

Spontaneous rupture and hemorrhage of renal angiomyolipoma (RAML) is a life-threatening clinical emergency. When it occurs during pregnancy, it is compared to a "bomb explosion," which makes the diagnosis and treatment more challenging. An ultrasound examination is a quick and safe examination with the benefit of no radiation exposure, which is always preferred for pregnant women. Currently, cases of spontaneous rupture and hemorrhage of RAML during pregnancy are rare, as is the diagnostic value and characteristics of ultrasound. The lack of understanding of the condition among ultrasound doctors makes it prone to misdiagnosis. In this study, we present the case of a pregnant woman who was preliminarily diagnosed with spontaneous rupture and hemorrhage of the left RAML using ultrasound and discuss the ultrasound characteristics.

CASE SUMMARY

A 38-year-old woman in her 19th wk of pregnancy (G2P1) was referred to our clinic for a sudden, persistent pain on the left side of the waist. She had not undergone any previous related abdominal examination. Ultrasound of the urinary system revealed a giant nonhomogenous lump in the left kidney area. The diagnosis was considered spontaneous rupture and hemorrhage of the left RAML in pregnancy *via* ultrasound. Her left-side waist pain continued to be intense. Subsequently, she underwent computed tomography, which led to the same diagnosis. Based on many factors, the patient underwent left nephrectomy after the induction of labor. The pathological result was the rupture and hemorrhage of a vascular leiomyoma lipoma.

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CONCLUSION

Ultrasound examination plays an important role in the diagnosis of the spontaneous rupture and hemorrhage of RAML during pregnancy.

Key words: Angiomyolipoma; Kidney; Rupture; Pregnancy; Ultrasonography; Case report

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Core tip: The diagnosis and treatment of spontaneous rupture and hemorrhage of renal angiomyolipoma during pregnancy are challenging because it is a life-threatening clinical emergency. We present the case of a pregnant woman who was preliminarily diagnosed with the condition using ultrasound and discuss the ultrasound characteristics. Ultrasound is a quick and safe examination with no radiation exposure and is preferred for pregnant women. Although pathology remains the gold standard for diagnosis, ultrasound can facilitate a preliminary diagnosis, leading to differential diagnosis to reduce misdiagnosis, prompting further diagnosis and treatment. Therefore, ultrasound plays a crucial role in the diagnosis of this condition.

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INTRODUCTION

Renal angiomyolipoma (RAML), a common benign tumor, is composed of mature blood vessels, smooth muscle, and fatty tissue. Some scholars believe that RAML consists solely of tumors and may originate from epithelioid cells around the blood vessels^[1]. Its incidence in the general population is estimated to be 0.13%, and about 80% of affected patients are women^[2,3]. Spontaneous rupture and hemorrhage is a life-threatening primary complication of RAML. Pregnancy is a risk factor for increasing the size, leading to rupture of RAML. Because computed tomography (CT) exposes the patient to radiation and magnetic resonance imaging (MRI) is time-consuming, these imaging methods are not always advised for pregnant women. Ultrasound is a quick and convenient method that does not predispose the patient to radiation; therefore, it is preferred for pregnant women. However, because ultrasound doctors lack understanding of the disease, it is more prone to misdiagnosis.

CASE PRESENTATION

Chief complaints

A 38-year-old woman in her 19th wk of pregnancy (G2P1) was referred to our clinic for a sudden persistent pain on the left side of the waist on July 28, 2017.

History of present illness

The patient's physical examination revealed tenderness over the left kidney area. Her blood pressure was 120/85 mmHg, heart rate was 86 beats/min, and body temperature was 36.8 °C. The patient had no significant medical history. She had not undergone any related abdominal examination previously.

History of past illness

The patient had no significant medical history. She had not undergone any related abdominal examination previously.

Physical examination

The patient was hospitalized and given conservative treatment, but her left-side waist pain continued to be intense. Because the size of the tumor was so large, and the fetal

heart rate was unstable, the patient decided to undergo left nephrectomy after the induction of labor.

Laboratory examinations

Laboratory tests indicated that the patient's hemoglobin level was 80 g/L, and the hematocrit was 0.242 L/L. On the 2nd d, hemoglobin was 95 g/L, and the hematocrit was 0.286 L/L.

Imaging examinations

Ultrasound examination of the urinary system (LOGIQ E9, GE) revealed a giant nonhomogenous lump in the left kidney area, which had caused the left kidney to move to the midabdomen. The size of the lump was approximately 159 mm × 100 mm, and the border was faintly visible. The lump showed a "striped sign" in which the outer part was hypoechoic with a strong stripe echo (Figure 1A), and the inner part near the left kidney was hyperechoic (Figure 1B). A stripe-shaped echoless zone was seen around the lump (Figure 1C) (arrow). Color Doppler flow image showed some spot-like blood flow signals around the lump (Figure 1D). A hyperechoic nodule was seen in the right kidney with a size of 30 mm × 25 mm. There was a fetus echo in the uterus. Preoperative CT showed a large, mixed-density mass in the left kidney (Figure 2). The density of the area adjacent to the kidney was low, and the area far from the kidney showed high density.

FINAL DIAGNOSIS

The final diagnosis of the present case was rupture and hemorrhage of the left RAML, right kidney hamartoma, and pregnancy.

TREATMENT

The patient underwent left nephrectomy after the induction of labor. During the operation the renal artery spurted after the left nephrectomy, the blood pressure dropped to 80/50 mmHg, and the blood loss reached 1000 mL. Four units of red blood cell suspension were transfused after emergency hemostasis, and the blood pressure rose to 100/60 mmHg. After the operation, gastrointestinal decompression and intravenous nutrition were given to the patient. The gastric tube was removed on the 2nd d after the patient exhausted. Antibiotics were continuously applied for 5 d, and the patient was discharged on the 6th d after the operation.

OUTCOME AND FOLLOW-UP

The pathological result was rupture and hemorrhage of the vascular leiomyoma lipoma. We instructed the patient to review regularly every 6 mo, but the patient has not reviewed in our hospital.

DISCUSSION

RAML, a common benign tumor, is composed of mature blood vessels, smooth muscle, and fatty tissue. Spontaneous rupture and hemorrhage is one of the main complications of RAML, which may be related to the lack of elastic fibers in the blood vessel wall or the sudden increase in renal intratumor and intravascular pressure. It is generally considered that tumors with a diameter of > 4 cm are more likely to rupture. However, there are tumors smaller than 4 cm that can still rupture^[4]. Thus, an assessment based only on tumor size is not accurate. Aneurysm formation, tuberous sclerosis complex (TSC), pregnancy coagulopathy, trauma, and hormone levels are also closely associated with rupture^[2]. About 80% of patients with TSC can develop angiomyolipoma^[2], and approximately 20% of patients have TSC^[5]. The TSC-related renal hamartomas tend to be multiple, large, and bilateral, and these lesions are more likely than isolated lesions to rupture^[6].

Although rupture and hemorrhage of RAML during pregnancy is rare, this

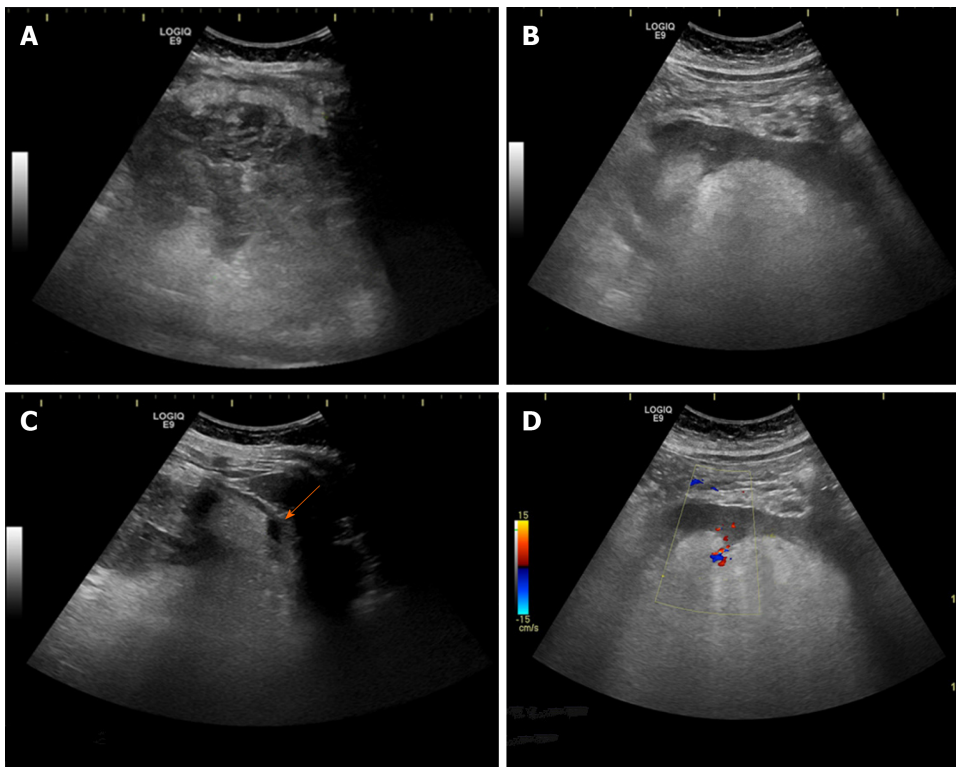


Figure 1 Ultrasound images of a giant nonhomogenous lump in the left kidney area. A: The part of the lump located in the left kidney area was hypoechoic with a strong stripe echo; B: The lump caused the left kidney to move to the middle abdomen. The inner part of the lump near the left kidney was hyperechoic; C: A stripe-shaped echoless zone was seen around the lump (arrow); D: The color Doppler flow image showed some spot-like blood flow signals around the lump.

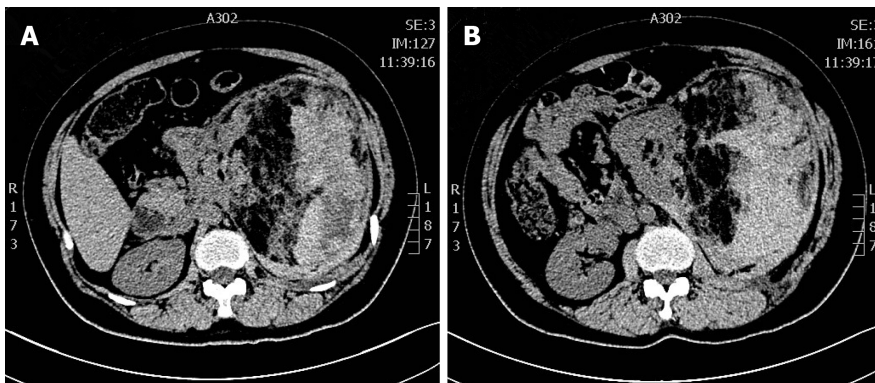


Figure 2 Computed tomography exhibited a large, mixed-density mass in the left kidney. The density of the area adjacent to the kidney was low (A), and the area far from the kidney showed high density (B).

condition has received increasing attention because of the difficulty of diagnosis and treatment. The rupture of RAML can occur at various stages of pregnancy. One study reported that the mean gestational age of patients was 27.7 wk, and the average tumor size was 10.1 cm at the time of diagnosis^[7]. Pregnancy also plays a role in the growth and rupture of RAML, but the mechanism is not clear. Studies have shown that about 25% of patients have estrogen and progesterone receptors in tumor specimens, and estrogen levels increase during pregnancy, which may accelerate tumor growth^[8]. In addition, the increase in circulating blood volume during pregnancy, which causes an increase in blood pressure, coupled with the rise of intra-abdominal pressure, may contribute to the growth and rupture of RAML.

In the clinical setting, patients may present with severe pain on one side of the waist, and the huge mass may be palpated or can manifest as gross hematuria. As we all know, ultrasound is a quick and safe examination with no radiation exposure, and it is always preferred for pregnant women. Because a typical RAML is rich in fat,

ultrasound examination is sensitive and can show a hyperechoic nodule. For the low-fat RAMLs, ultrasound, which shows various forms of echo mode, is not very sensitive or accurate^[9]. At this time, an ultrasound must be combined with other imaging methods, such as CT and MRI.

The ultrasound findings of rupture and hemorrhage of RAML show a strong, lamellar heterogeneous echogenic mass, similar to “onion skin” in the kidney area when it occurs under the renal capsule. It should be distinguished from the nonbleeding hamartomas, which also can appear as mixed. If the tumor ruptures and bleeds out of the renal capsule, there is a stripe echoless zone around the hematoma, and the no-echo area may be more reliable for the diagnosis of the rupture. The Color Doppler flow image can measure the blood flow of the tumor and determine the presence of an aneurysm. Contrast-enhanced ultrasound can identify the presence of hematomas and the existence of active bleeding and aneurysm formation^[10].

Differential diagnosis includes obstetric factors such as abruptio placentae, placenta previa, uterine rupture, and ruptured ectopic pregnancy. In addition, it also needs to be distinguished from bilateral renal cell carcinoma, perinephric abscess, and primary retroperitoneal tumor. Renal cell carcinoma has general characteristics of cancer. The patient with typical renal cell carcinoma will have intermittent, painless, gross hematuria clinically. Most of them are hypoechoic on ultrasound, and the color blood flow is mostly spherical. For the atypical cases, combining enhanced CT or MRI can improve the accuracy of diagnosis.

In summary, although the gold standard for diagnosis is pathology, ultrasound examination can facilitate a preliminary diagnosis with its characteristics. Combining clinical data (female with TSC, related medical history, multiple bilateral hyperechoic nodules, and clinically with acute low back pain) can increase the accuracy of ultrasound diagnosis.

In the ultrasound images of this case, the inside part of the mass near the left kidney was homogeneously hyperechoic, which highly suggested the diagnosis of hamartoma. The lateral part of the mass was hypoechoic with a strong stripe-like echoless area, which suggested the diagnosis of rupture and hemorrhage. In addition, the patient appeared with a sudden persistent pain on the left waist, and the ultrasound showed a right kidney hamartoma, which was more helpful for the diagnosis.

At present, no consensus has been reached on the optimal treatment for this condition. When an RAML is detected during pregnancy, the patient should be informed of the risk of rupture. If the tumor is large (*i.e.* > 4 cm), active surgery or arterial embolization before pregnancy is recommended. Studies have shown that renal artery embolization with less trauma has better nephron retention than surgery^[11], especially for patients with TSC or bilateral multiple hamartomas^[12]. For patients with ruptured hemorrhage during pregnancy, the most appropriate treatment depends on the gestational week and the condition of the pregnant woman. When RAML causes maternal hemodynamic instability, it should be managed actively with surgical treatment or arterial embolization simultaneously with antishock therapy. If the patient's blood pressure is stable, and tumor bleeding is steady with no progression, temporary conservative treatment can be considered^[13]. Alongside close clinical observation, the patient may choose natural birth or pregnancy termination before treating the lesion. After 28 wk of pregnancy, if the fetus's lungs are mature and the mother's condition is severe, a cesarean delivery may be required to terminate the pregnancy. There are some successful cases in the literature of conservative treatment^[13-15], and there is also a case report of a patient who underwent radical nephrectomy during pregnancy while continuing the pregnancy to full term, which of course, is associated with very great challenges^[16].

In addition, for the patients undergoing conservative treatment, ultrasound has an advantage of no fetal radiation damage. For the patients who cannot perform MRI in the emergency department, or who need emergency surgery in critical condition, ultrasound is an important method to make the diagnosis and differential diagnosis.

In this case, the patient's left-side waist pain remained intense after conservative treatment. Taking into consideration the huge size of the tumor and the patient's general condition, there was a great risk of continuing the pregnancy, and she chose to terminate the pregnancy and underwent left nephrectomy.

CONCLUSION

Rupture and hemorrhage of RAML during pregnancy is compared with a “bomb

explosion," which is life-threatening. Ultrasound has many advantages, such as rapid diagnosis and no radiation. Although ultrasound examination lacks specificity, it can lead to a preliminary disease diagnosis, facilitating a relevant differential diagnosis to reduce misdiagnosis, which can prompt further treatment. Therefore, ultrasound examination played an important role in the diagnosis of the condition during pregnancy. Depending on the patient's condition, treatment should be individualized.

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