

Spontaneous Non-traumatic Upper Urinary Tract Rupture in Pregnancy: Case Report and Literature Review

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Abstract

Background: Hydroureteronephrosis is one of the most common causes of flank pain in pregnancy, which on rare occasions can progress to rupture of the urinary tract.

Case presentation: A 31 year old previously healthy primigravida presented to the Emergency Department complaining of severe left flank pain. A provisional diagnosis of renal colic secondary to a possible migrant renal calculus was established based on an initial bedside Ultrasound (US), with no abnormalities, and moderate microscopic haematuria on urinalysis. A Magnetic Resonance urogram was performed and demonstrated perinephric fluid considered related to a pelvicalyceal rupture on the left side, with no evidence of calculi. A left percutaneous nephrostomy was performed, however 48 hours later she developed similar symptoms on the right side. A serial US exam revealed resolution of the hydronephrosis on the left side with persistent and progressing hydronephrosis on the right but no obvious rupture. A right percutaneous nephrostomy, then provided relief of symptoms. She made an uneventful recovery, was discharged home five days later and gave birth at 36 weeks to a healthy baby.

Conclusion: Spontaneous rupture of the upper urinary tract is a rare but significant complication of pregnancy that should be considered in the pregnant patients thought to be experiencing renal colic.

Keywords: Hydronephrosis; Spontaneous urinary tract rupture; Renal colic; Flank pain pregnancy; Nephrostomy; Kidney; Ureter

Background

Loin/flank pain in pregnancy is a common presentation to the Emergency Department (ED). There are a wide range of causes: medical, surgical, urological, obstetrical and gynaecological, but the predominant cause is acute hydronephrosis [1]. Hydroureteronephrosis is frequently associated with pregnancy and on rare occasions can progress to rupture of the urinary tract. This paper describes the case of a young female who developed an initial rupture of the left ureter, progressing following admission to develop symptoms on the right side. Also we have included a narrative review of the literature on this subject. To date, we believe, this is the first case report of a patient with bilateral symptomatic upper urinary tract hydroureteronephrosis.

Case Presentation

A 31 year old white European primigravida (28 weeks gestational age) with an uncomplicated pregnancy presented to the Emergency Department (ED) complaining of severe progressive left flank pain. The symptoms had commenced approximately 2 hours prior, whilst travelling by train, with gradual onset of colic pain of her left groin, worsening and ascending to the left flank and renal angle. Her past medical history included an episode of left pyelonephritis four years previous, bilateral breasts reduction two years ago and mild eczema which improved with progression of her pregnancy. She had no allergies and was not on any current medication. There was no family history of significance. On examination she was alert and oriented (GCS 15/15), not pale or cyanotic, anxious, restless, and complaining of severe pain (10/10 on numeric pain rating scale). Her body temperature was normal (36.6°C), with mild tachycardia (110 bpm), BP 140/105 mmHg, respiratory rate 17 breaths/minute, SaO₂ 96% on room air and a capillary refill time < 2 sec. Percussion tenderness was elicited of the left loin. Her uterus had a normal height for the gestational age; there was no vaginal discharge or bleeding. Foetal movements were present with a heart rate of 120 bpm. The remainder of the clinical exam was unremarkable. An 18 G Intravenous (IV) cannula was inserted and

baseline bloods investigations drawn for full blood count, group and screen, urea and creatinine, electrolytes, glucose, liver function test's, "C" Reactive Protein (CRP) and Venous Blood Gas (VBG). She was initially managed with opioid analgesia (pethidine), intravenous paracetamol and maintenance fluids (0.9% NaCl). Shortly after her pain score subsided to 3-4/10.

No free fluid or other abnormalities were revealed by an ED Ultrasound (US). A bed side urinalysis revealed a moderate amount of blood, but negative for nitrites and leukocytes. Her VBG and glucose were normal.

Based on the clinical presentation and microscopic haematuria a provisional diagnosis of renal colic secondary to a possible migrant renal calculus was established.

Laboratory blood results revealed an elevated WBC ($14.5 \times 10^9/L$) with mild neutrophilia ($11.8 \times 10^9/L$) and elevated CRP (8.3 mg/L). Urea, creatinine, LFT's and electrolytes were normal. Prophylaxis with IV amoxicillin-clavulanic acid was commenced.

A formal renal ultrasound examination was requested of the Department of Radiology, which revealed bilateral dilatation of the renal collecting system (deemed physiological), predominantly on the right, with no sonographic evidence of renal calculi (Figure 1). The ureters were not visualized and the spleen size was at the upper limit

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(13 cm) of measurement with normal parenchymal echotexture. No structural abnormalities were identified.

A Magnetic Resonance urogram was performed which demonstrated mild bilateral hydronephrosis and proximal hydroureter, with compression of the distal ureters by the gravid uterus. Perinephric fluid was identified considered related to pelviccaliceal rupture, on the left, with no evidence of calculi (Figure 2).

She was referred to the Department of Urology for further management. She was commenced on dexamethasone in the eventuality she may enter in preterm labour. A left percutaneous nephrostomy, under US guidance and minimal fluoroscopy was performed the following day under procedural sedation with resolution of symptoms. She subsequently developed similar right side symptoms forty eight hours later. A serial US exam revealed resolution of the hydronephrosis on the left side with persistent and progressing hydronephrosis on the right but no obvious rupture (Figure 3). A right percutaneous nephrostomy then provided relief of symptoms. She made an uneventful recovery and was discharged home five days later. The nephrostomies tubes (Figure 4) remained in situ for the remainder of her pregnancy.

Following discharge and return to her natal country she experienced several episodes of urinary tract infection requiring admission and IV antibiotics. She gave birth at 36 weeks of gestation to a 2.4 kg baby girl via vacuum assisted vaginal delivery. No epidural anaesthesia was performed due to risk of infection. The nephrostomies were removed one week after the birth of her daughter. She experienced an episode of bladder hypotonia and urinary retention, which mandated admission and conservative management. Five weeks later, the mother is free of any urinary symptoms and continues under the care of the Department of Urology. The baby is well and developing normal.

Discussions and Literature Review

Upper urinary tract dilatation in pregnancy is a phenomenon described for more than 200 years [2]. Nearly all pregnant women will have by term detectable asymptomatic hydronephrosis [3] but only a small proportion will progress to anuria, urinary tract rupture or renal failure. It was also noticed an enlargement of the kidneys associated with an increase of the interstitial fluid and vascular volume

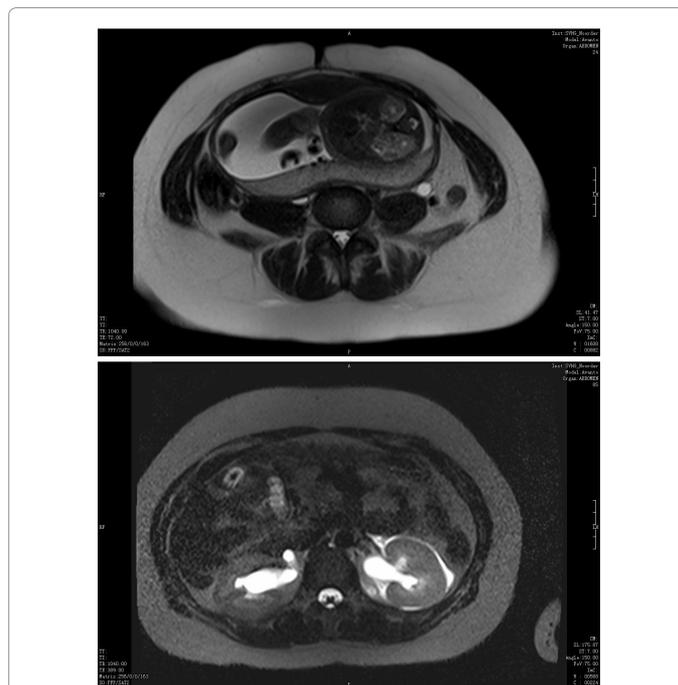


Figure 2: A Magnetic Resonance urogram was performed which demonstrated mild bilateral hydronephrosis and proximal hydroureter, with compression of the distal ureters by the gravid uterus. Perinephric fluid was identified considered related to pelviccaliceal rupture, on the left, with no evidence of calculi.



Figure 3: A serial US exam revealed resolution of the hydronephrosis on the left side with persistent and progressing hydronephrosis on the right but no obvious rupture.

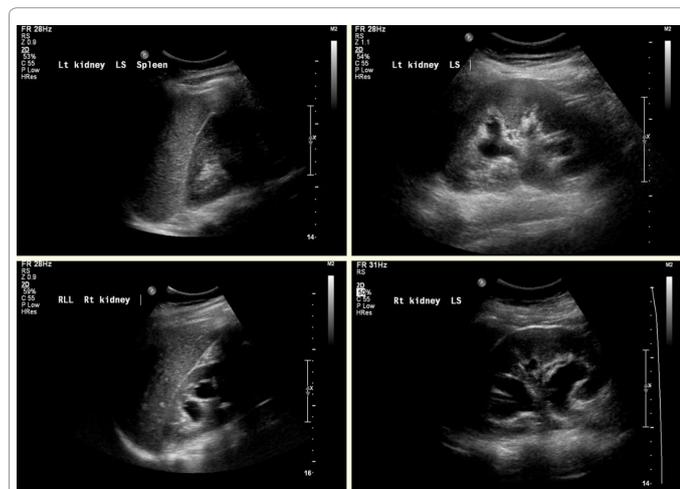


Figure 1: A formal renal ultrasound examination was requested of the Department of Radiology, which revealed bilateral dilatation of the renal collecting system (deemed physiological), predominantly on the right, with no sonographic evidence of renal calculi.

[4]. Hydronephrosis may develop from the 6th - 10th week of gestation [5] and according to Gillenwater, rupture of the collecting system can occur from week 18 of gestation up to day 1 post delivery [6].

Spontaneous rupture of the kidney/urinary tract is defined as a rupture of a urinary system that occurs in the absence of surgery, instrumentation or trauma [7]. According to Middleton et al. and Meyers et al. the patients with pregnancy associated ruptures may fall into three categories: (1) patients with no condition other than pregnancy, (2) patients with non-tumoral structural disease of the urinary tract and (3) patients with rupture of a renal tumour [8,9]. Most of these ruptures are associated with different underlying conditions of the kidneys or upper urinary collecting system [2,10-14] such as infections, scars, congenital structural abnormalities, malignant or benign tumors (i.e. angiomyolipoma), or cysts. For our case there was a positive history of infection four years previous. Anecdotally, Tang et al. reported a case of iatrogenic rupture that became symptomatic on day two following a caesarean section, secondary to accidental placement of sutures through the left bladder wall obstructing the left ureteral flow [15].

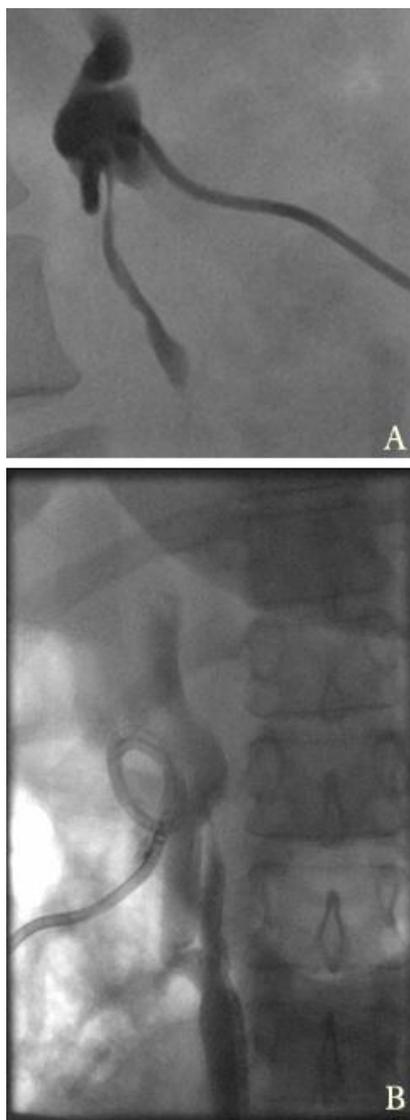


Figure 4: The nephrostomy tubes remained in situ for the remainder of her pregnancy.

Spontaneous rupture with no underlying condition is a phenomenon observed more frequently on the right side [2,10-13]. A possible explanation for this refers to the compression of the right ureter caused by the physiological dextrorotation of the gravid uterus or by the engorged right ovarian artery [16] or vein coupled with the hormonal ureteral atony [17]. This suggests a mixed mechanism, mechanical and endocrine, of urinary tract distension in pregnancy. The uterine dextrorotation debuts around 20th week of gestation and persists up to two weeks after delivery [18]. The pelvic brim is regarded as the site of compression, sustained by the fact that in pregnant patients with pelvic transplanted kidneys, the physiological hydronephrosis is not seen [5]. Therefore the pelvi-ureteric distension seen in pregnancy involves only the upper urinary tract above the pelvic brim [10]. However Fried showed that 60% of the females involved in their study had hydronephrosis in the first trimester before the uterine distension occurred [19]. As shown by other studies, pelvic pathologic processes associated with pregnancy (i.e. fallopian tube torsion) can lead to compression of the ureters, hydronephrosis and possible rupture.

In order to progress to rupture the hydrostatic pressure needs to exceed the holding capacity of the calyceal-renal capsular junction [9] and the protective mechanism of spontaneous extravasation of the urine through the calyceal fornices. This phenomenon was described in excretory urography, performed with external compression, as pyelotubular, pyelosinus and pyelolymphatic backflow, and resolves spontaneously with no sequel [20]. The increased pressure within the pelvicalyceal system generates stretching and decreased elasticity of the capsule, and soft tissue attenuation [5]. There is a higher probability for a rupture to take place in an area where the normal structure was altered by infections, tumors or scarring [21]. As described by Huang et al. in pregnant women with pre-existing hydronephrosis, actions associated with sudden increase of the hydrostatic pressure in the urinary tract, like IV fluid boluses for pregnancy related surgical procedures (i.e. placement of a McDonald cerclage under spinal anaesthesia), can favour rupture [13].

The spontaneous rupture of the urinary system involves the collecting system or the kidney parenchyma but in most of the cases the site of rupture is the renal pelvis or the calyceal system [2].

The real incidence rate of spontaneous non-traumatic upper urinary tract rupture associated with pregnancy is not known. In 2010, Matsubara et al. conducted a literature review for non-traumatic urinary tract rupture during pregnancy, retrieving a total of 31 cases [22]. The main population of 25 cases were reported by Wolff et al. analyzing a period of 50 years (1947 to 1995) [10]. Matsubara et al. [22] added 6 new cases to this, covering a period of 10 years (2000 to 2010). Most of the cases (18 cases) had a documented underlying condition in the past or at the time of rupture, such as urinary tract infections, abscess, stones or tumors. Using the Medline database we have reviewed the literature, covering the period 1995 to 2011, and retrieved four more cases [5,23-25] which were added to those reported by Matsubara et al. (Table 1); we have added another, through this case report, raising the total number of cases in the literature to 36. With our review we have found only 11 cases of rupture occurring on left side (Table 1). By our knowledge, this case is the first case reported in the literature, with rupture on the left side and progressive symptoms on the right as well.

The typical clinical presentation of the rupture is a sudden onset of pain on the side involved. Sometimes it may be associated with pyrexia, suggesting an infected urinoma, abscess or chemical peritonitis. In rare situations, if the collecting system rupture is associated with a parenchymal tear, a palpable mass is generated by the leaking urine and blood can be felt on examination of the flank [5]. According to Wolff et al. parenchymal ruptures are associated with sudden haematuria, loin pain, loin mass and or hypotension, while a calyceal rupture usually presents with loin pain with or without haematuria [10]. If the rupture occurs on the right side, it can create confusion with acute appendicitis leading to unnecessary appendectomy. Hwang et al. reported the case of a patient presenting with one week history of right flank pain, mild leukocytosis, clear urine and moderate dilatation of the right calices, renal pelvis and proximal ureter (deemed to be physiological) who underwent appendectomy [11]. The pathological report of the appendix infirmed the inflammatory process and three days post-operative the patient developed severe right flank pain. A second US scan followed by an MRI scan confirmed the diagnosis of kidney rupture with associated urinoma, managed with retrograde ureteral catheterization. Other differential diagnoses to be considered are: nephrolithiasis, acute hydramnios, cholecystitis, placental abruption, red degeneration of uterine fibroids, renal colic and twisted fallopian tube [2,26]. The initial clinical presentation may cause difficulty differentiating between a

Author (publication year)	No of cases reported/reviewed	Side	Underling condition of the kidneys or urinary tract
Wolff et al. [10]*	25	Left – 7 cases Right – 18 cases	Yes (17 cases)
Chelli et al. [23]	1	Left	Unknown – lack of post-partum investigations
Hwang et al. [11]	1	Right	No
Delakas et al. [25]	1	-	-
Nabi et al. [5]	1	Right	No
Satoh et al. [12]	1	Right	No
Huang et al. [13]	1	Right	Yes - query calculus
Sidra et al. [2]	1	Right	Not mentioned
Lo et al. [14]	1	Left	No
Matsubara et al. [22]	1	Left	No
Klasen et al. [24]	1	Left	Yes – calculus

*Covering the period 1947 to 1995

Table 1: Literature synopsis for spontaneous upper urinary tract/kidney rupture in pregnancy.

spontaneous urinary tract rupture and a preterm labour [14]. If there is concern of the latter, an experienced gynaecologist should be consulted and dexamethasone (to accelerate foetal lung maturity) should be considered as soon as possible.

Because the pain alone and the clinical presentation are non-specific, further investigations are necessary to establish the diagnosis, imaging studies being essential. Plain abdominal X-rays are not useful, unless is a concern of perforation or obstruction. Intravenous urography and contrast-enhanced CT scan (the preferred imaging method for kidney trauma [27]), are readily available and useful but according to some authors should be avoided, especially in early pregnancy, due to the high radiation exposure and lack of safety evidence [5]. The guidelines of the American College of Obstetricians and Gynaecologists state that an X-ray foetal exposure of less than 5 rads does not increase the risk of foetal anomalies or abortion [28]. An IV urogram with only two films (less than 1 rad) can be helpful in identifying the site of rupture without associated harm to the foetus [29]. Despite using a higher dose of radiation a CT scan is still within the foetal radiation safety zone if performed after 18th week of gestation [6]. Urinary tract US and urinary MRI are feasible and safer alternatives. MR imaging is considered safe after the first trimester [13] and is useful in differentiation between urinoma and acute hematoma [11], but unfortunately neither US nor MRI are useful in detecting the site of rupture or leak. Serial US exams with duplex Doppler studies play an important role in detection of the ruptured kidney for those patients with symptomatic hydronephrosis [11]. If there is a high degree of suspicion, serial US exams should be performed regularly especially when there is a symptomatic presentation with minimal benign appearing perinephric collection on initial US exam [5]. MacNeily et al. [30] proposed a method of US differentiation between physiological and pathological dilatation of the renal collecting system in pregnant patients, based on the extension of the dilated ureter in relation to the common iliac artery: a physiological hydronephrosis will extend down only to the level of the artery while in a pathologic obstruction it will extend below the artery. Unfortunately, laboratory tests have limited value, most of the results being non specific. Urinalysis may help differentiating between a parenchymal rupture (more often presenting with gross or microscopic haematuria) and ruptures of the collecting system [2]. In our case there was a moderate amount of microscopic blood on the initial urinalysis confirmed by the laboratory, without parenchymal involvement.

To date there is no consensus on evidence based options of treatment for spontaneous urinary tract rupture in pregnancy. The treatment method may depend on the rupture site and severity [12] and on the clinical situation: rupture of a septic urinary system and rupture of the renal parenchyma. The goals of treatment are to preserve renal

function, relieve pain, allow the site of rupture to heal spontaneously [31] and promote safe progress of the pregnancy to full term and safe delivery with minimal disturbance for the baby. For some cases conservative management may be an option as showed by Rojburt et al. who reported the case of a patient with rupture of the renal pelvis at 33 weeks of gestation who was managed conservatively with twice daily Diclofenac and partial resolution of the urinoma after two weeks [31]. She had a spontaneous vaginal delivery with resolution of the US findings 5 days later. The main two surgical options referred to in the literature so far are: JJ stenting and nephrostomy. In the event of a large amount of extravasated urine and blood, according to some authors explorative surgery should be considered [5,31]. Conservative management may be applied for small perinephric collections [5]. A JJ stent is probably the least invasive effective method for controlling pain and progression of the rupture and also allowing the pregnancy to progress to full term. The advantages of a JJ stent are: pain relief and free drainage, facilitating the closure of the communication between the pelvicaliceal system and perinephric space [5]. Other authors recommend an aggressive approach with rapid kidney exploration, drainage of the perinephric collection and nephrostomy placement [8]. This may be advocated especially in the case of a large rupture where a significant amount of urine and blood leaking into the perirenal space may progress to chemical/infective peritonitis if dissection of the retroperitoneal fascial planes continues [2] forming a retroperitoneal urinoma, haematoma or abscess which may spill into the peritoneal cavity [10]. As suggested by Wolff et al. nephrostomy or nephrectomy may be an option for cases with severe rupture [10]. Nabi et al. suggested an intermediate approach, starting with the placement of a retrograde catheter and careful clinical follow-up of the patient's overall condition, haematocrit and serial US, followed by open exploration in case of deterioration [5]. Despite the use of JJ stents for rupture of urinary tract in pregnancy [2,10-13], the literature is lacking in consistent data to sustain this as a standard method of treatment. When a JJ stent is used the recovery tends to be 'restitutio in integrum' at one month after removal of the stent [11,22]. According to Matsubara et al. there is no data available regarding a patient with JJ stent in situ for spontaneous urinary tract rupture in pregnancy progressing to a safe vaginal delivery at term [22]. In the same paper the authors advanced the idea that vaginal delivery may be possible as long as the patient has a good overall condition with adequate urine drainage [22]. Huang et al. reported the case of a 27 years-old woman who developed a collecting system rupture at 19 weeks of gestation secondary to a fluid bolus during a cervical cerclage, managed with a ureteral catheter, who had a premature birth at 25 weeks [13]. The baby died at the age of 6 weeks due to multiple complications. Nevertheless the delivery option should be analyzed by an experienced multidisciplinary team (urologist, gynaecologist and

neonatologist) on an individual basis. In some cases the JJ stent may not be effective at all. Nabi et al. reported the case of the 28 year-old, 18 weeks pregnant, female who presented with a two days history of right flank pain and oliguria on a single right functional kidney [5]. A small perinephric collection was identified by US and a JJ stent was inserted with no complications under local anaesthesia. Shortly after the procedure a good urine output was obtained but within the next hours the haemoglobin dropped by 4 points with no visible cause of bleeding. A serial US demonstrated a large perinephric hematoma. A percutaneous drainage attempt failed and was followed by abortion. The open perinephric drainage in the operating room failed to identify a site of rupture. After 2 months the patient had a normal functioning kidney.

In addition to the clinical treatment methods available for this condition, we should also consider the impact that this pathology may have on the future mother. Increased anxiety associated or not with a pre-existent psychiatric background may lead to depression and alter the mother's decision regarding the future course of pregnancy. Psychiatric/psychological support may be considered for these cases. Matsubara et al. reported the case of a 38 year old primigravida (34 weeks of gestation) who requested the termination of pregnancy through caesarean section two days after the insertion of a JJ stent [22]. She delivered a healthy baby boy with a weight of 2500 g and an Apgar score of 7/9 at 1/5 minutes [22,32,33].

Regarding the treatment modality, in order to provide the best care, all cases should be analysed by a multidisciplinary team on an individual basis. In our case the Department of Urology and Radiology in collaboration decided on a minimal-invasive approach: US guided nephrostomy with minimal fluoroscopy under procedural sedation and analgesia. The procedures were uneventful, and well tolerated by the patient, with rapid resolution of symptoms. The patient was discharged, 24 hours after the second nephrostomy was performed. Despite her several episodes of urinary infections (an inherent complication) associated with the nephrostomy tubes she was complaint and able to continue the pregnancy with no complications for 8 further weeks. It is difficult to state if there is a connection between the urological procedure performed and the premature membrane rupture at 36 weeks.

The role of the emergency assessment in these cases is extremely important and may lead to the final diagnosis of rupture, as occurred in our case, due to access to imaging modalities from the ED. The ED assessment should be thorough and centred on the presenting history, past medical history, examination, effective analgesia and fluid resuscitation as necessary. A gynaecological and obstetric history and examination should be performed, to rule out pregnancy related conditions and assess the potential impact on the foetus. Aggressive analgesia should be commenced rapidly to minimize the maternal and foetal distress. In our case opioid and paracetamol analgesia were sufficient to obtain an acceptable degree of comfort for the patient until the final treatment is delivered. NSAIDs should be avoided due to high risk of premature ductus arteriosus closure and pulmonary hypertension. IV antibiotics according to the local protocol should be administered early if there is a suspicion of an infective process. Involvement of the specialized teams, especially the urology and gynaecology team, should be requested as soon as possible if there are signs of foetal distress and threatened preterm labour.

Conclusions

Spontaneous rupture of the upper urinary tract is a rare but significant complication of pregnancy. Emergency Medicine clinicians,

who will be the primary or secondary contact with these patients, should consider this diagnosis in the pregnant patient thought to be experiencing renal colic.

Consent

I, Catalin-Iulian Efrimescu on behalf of the authors' group, declare that the patient presented in this case report freely agreed with the publications of her case. All the information presented in text or images was anonymized so identification of the patients will not be possible. A verbal consent was obtained prior the submission of this paper. Unfortunately, due to the fact that the patient was not a resident in Ireland and after discharge she returned to her natal country it was not possible to obtain written consent. Catalin-Iulian Efrimescu

Competing Interests

The authors declare that they have no competing interests.

Authors' Contributions

CIE wrote the initial version of the article and edited the images presented.

DB and DM supervised the writing of this paper and made major changes after reviewing the first version of the paper.

All the authors read and approved the final manuscript.

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