Neonatal bilateral adrenal hemorrhage: case presentation

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Abstract
Adrenal hemorrhage is a relatively uncommon condition (0.2%–0.55%) during the neonatal period that may lead to acute adrenal crisis, shock, and death, unless promptly recognized and treated appropriately. The precarious blood supply of the adrenal gland makes it vulnerable to hemorrhage. The differential diagnosis includes multicystic kidney, hydronephrosis, neuroblastoma and nephroblastoma. The cause of adrenal hemorrhage has been associated with traumatic birth and hypoxia, shock, septicemia, and intravascular coagulation The clinical features may include anemia, jaundice, abdominal mass, painful swelling with bluish discoloration of the scrotum, acute adrenal crisis or shock. The large majority of patients with adrenal hemorrhage do not have clinically obvious signs of adrenal insufficiency. The diagnosis is usually made incidentally at imaging performed for another reason. Since patients with unilateral or bilateral adrenal hemorrhages are usually treated conservatively, noninvasive diagnostic techniques are preferable. This report presents a case of neonatal bilateral adrenal hemorrhages diagnosed ultrasonographically.

Key words
Adrenal hemorrhage, hyperbilirubinemia, adrenal mass, abdominal ultrasound diagnosis

■ INTRODUCTION

Adrenal hemorrhage (AH) is a relatively uncommon condition (0.2%--0.55%) during the neonatal period that may lead to acute adrenal crisis, shock, and death, unless promptly recognized and treated appropriately.(1) Most abdominal masses in the neonatal period are retroperitoneal. The differential diagnosis includes multicystic kidney, hydronephrosis, neuroblastoma and nephroblastoma. Adrenal abscesses rarely occur, but hemorrhages are less rare. The adrenal gland is disproportionately large in neonates, with rapid involution early in childhood. The posterior location gives them a protected position within the rib cage. The precarious blood supply of the adrenal gland makes it very vulnerable to hemorrhage. The adrenal gland is supplied by more than two dozen branches from three main adrenal arteries, forming a subcapsular plexus that drains into the medullary sinusoids. The gland is drained by comparatively few venules, resulting in a relative 'vascular dam'.(2)

The cause of adrenal hemorrhage remains uncertain but it has been associated with traumatic births. It may be the result of difficult labor or delivery, particularly in infants of diabetic mothers or infants who are large for their gestational age. Other factors implicated in the development of adrenal hemorrhage include hypoxia, shock, septicemia, and intravascular coagulation. Clinical features may include anemia, jaundice, abdominal mass, painful swelling with bluish discoloration of the scrotum, acute adrenal crisis or shock.(3) The large majority of patients with AH do not have clinically obvious signs of adrenal insufficiency. The diagnosis is usually made incidentally at imaging performed for another reason. Since patients with unilateral or bilateral adrenal hemorrhages are usually treated conservatively, noninvasive diagnostic techniques are preferable.
This report presents a case of neonatal bilateral adrenal hemorrhages diagnosed ultrasonographically.

■ CASE REPORT

A six-day old male neonate was admitted to our hospital due to jaundice from the second day of life. The neonate was born by spontaneous vaginal delivery at Western Regional Hospital, Belize, to a 23 year old mother with obstetric history of G4*P4† Ab0‡ and gestational diabetes. The neonate had an APGAR of 9 at the first minute and 9 at 5 minutes and a gestational age of 38 weeks by the Capurro method; birth weight was 4110g.

On admission, the neonate was alert and breastfeeding well. He had no history of fever, showed significant jaundice, but no palm nor plantar impregnation.

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* G- gravida (number of pregnancies), P†- para (number of live births) and Ab‡- abortions

Initial laboratory findings were as follows:

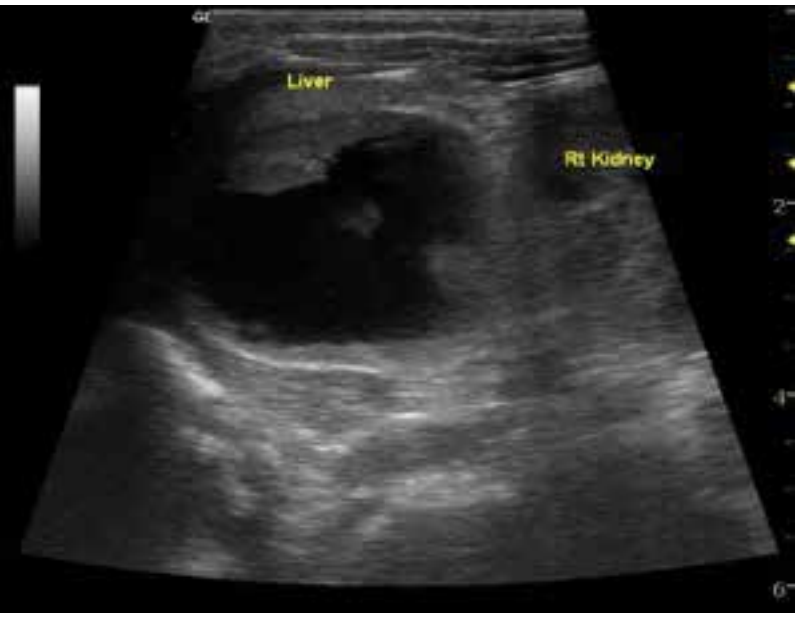
RBS	67mg/dL	Total bilirubin	36mg/dL
WBC	27 500	Indirect bilirubin	4mg/dL
Hb	18.6	SGOT	80mg/dL
Hct	54.4%	SGPT	80mg/dL
Polymorphonuclear	65%	Creatinine	0.4
Linphocytes	25.2%	K+	4.57
Monocytes	8%	NA+	140
Platelets	132 000	PT	14sec
CRP	NA	PTT	35sec

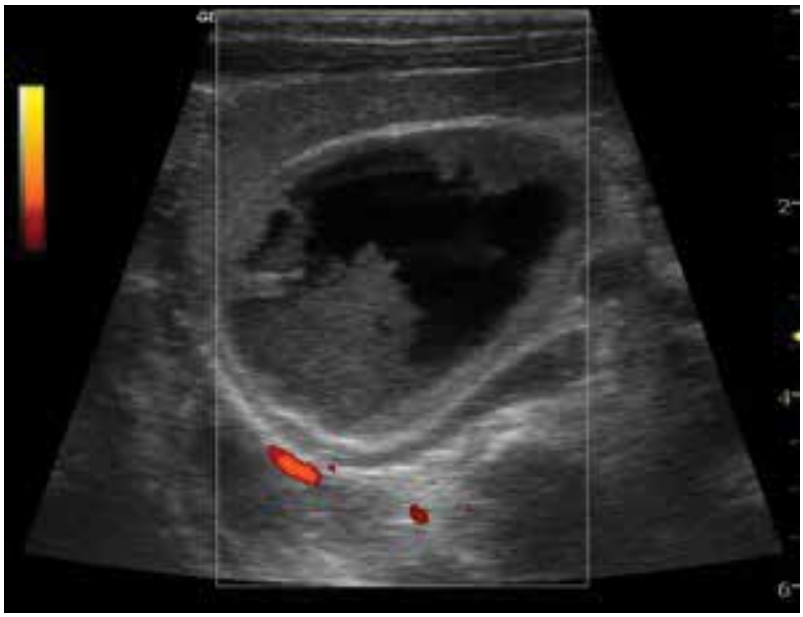
RBS- random blood sugar, WBC-white blood cell count, Hb-hemoglobin, CRP- C reactive protein, NA - not available, SGOT- serum glutamic oxaloacetic transaminase, SGPT- serum glutamic piruvic transaminase, K+ - potassium, Na+- sodium, PT- prothrombin time, PTT-partial thromboplastin time

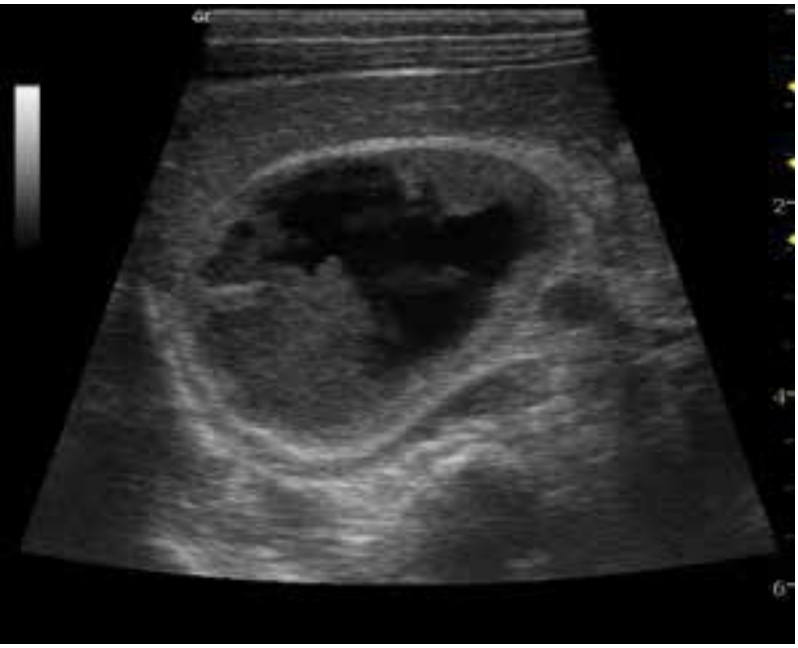
The abdomen showed no palpable mass, no hepatosplenomegaly. The rest was unremarkable.

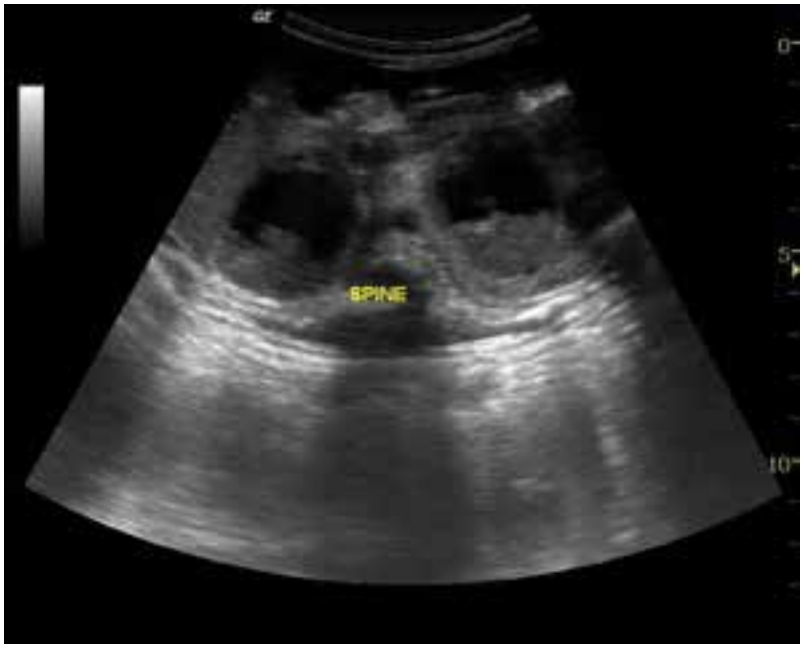
The child's temperature was 36.7 °C; respiratory rate (RR), 42/min; heart rate (HR), 128/min and oxygen saturation, 96%. Both the mother's and the neonate's blood group and Rh were the same: O+.

Due to high bilirubin level, a total exchange transfusion was









performed without any complications.

Diagnosis

Since there was no evident explanation for the hyperbilirubinemia (jaundice) such as ABO/Rh incompatibility or sepsis, an abdominal ultrasound (US) was indicated. The abdominal ultrasound findings reported bilateral neonatal hematomas.

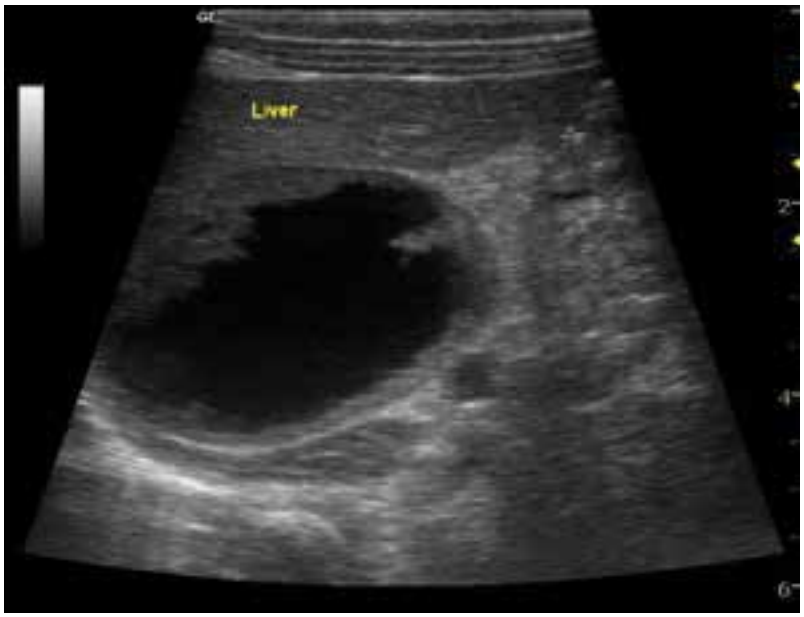
Ultrasound diagnostic images showed bilateral adrenal masses composed by mixed echogenicity with an anechoic center and peripheral echogenic areas. Doppler negative.

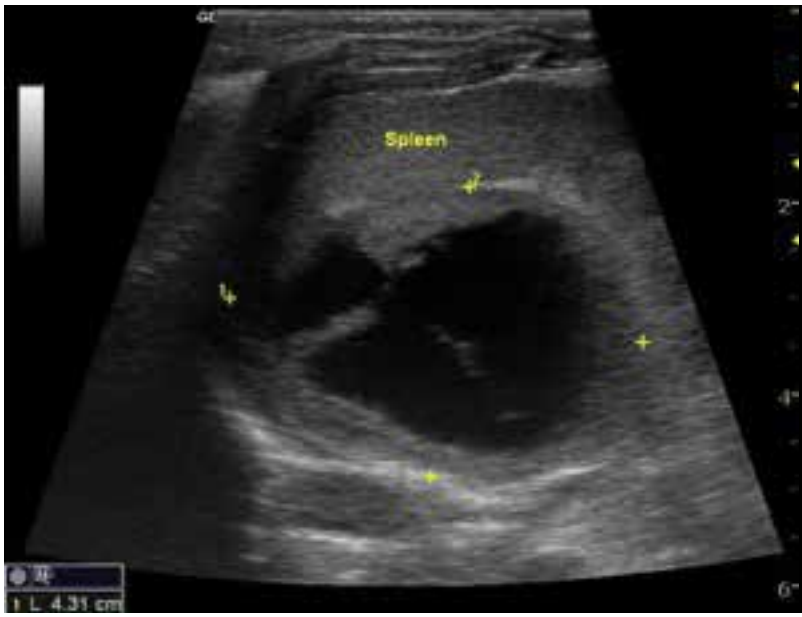
Clinical progress

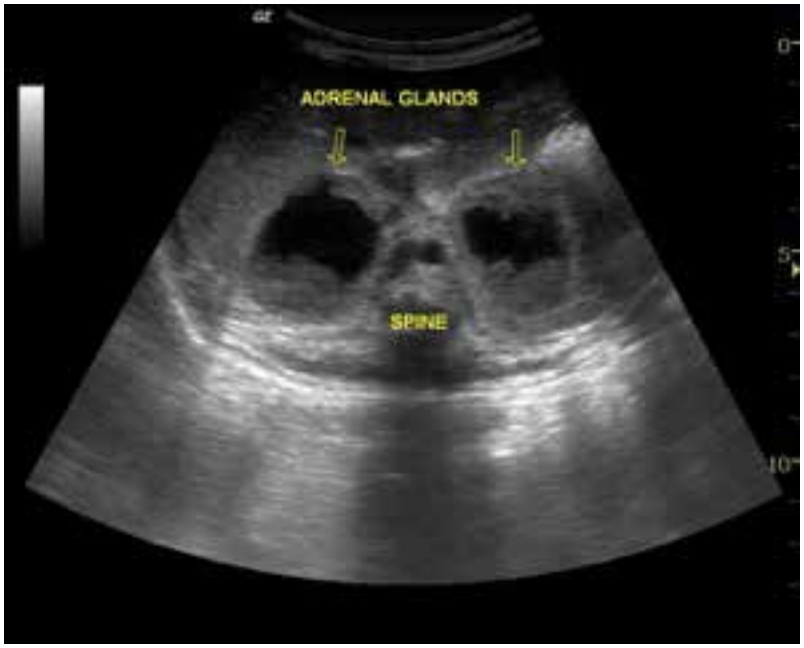
The neonate continued under phototherapy after the exchange transfusion. After transfusion, bilirubin and transaminase levels decreased: total bilirubin 23mg/dL, indirect bilirubin 2.0mg/dL, SGOT 50 mg/dL and SGPT 20mg/dL.

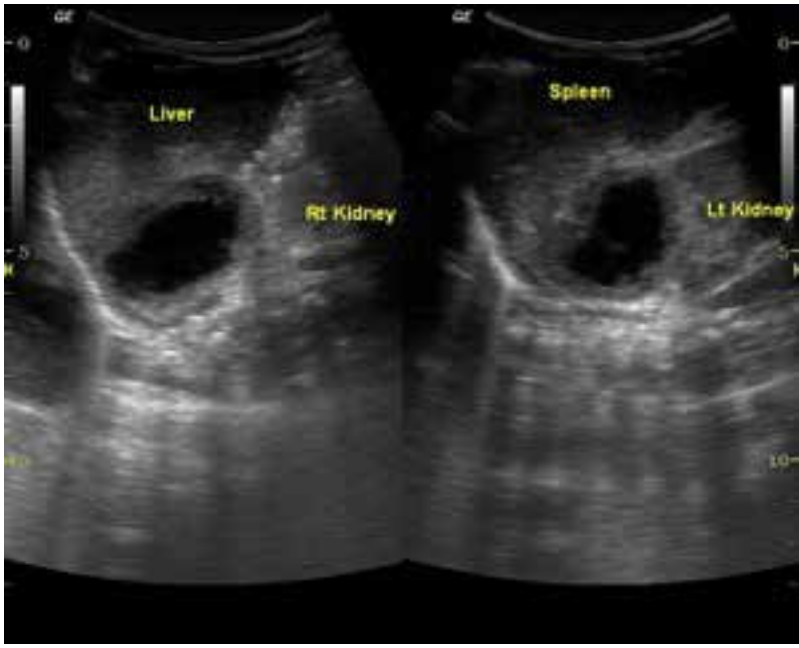
The neonate was hemodynamically stable with no signs of adrenal insufficiency throughout his hospital stay. He was discharged after three days with a total bilirubin level of 18.43mg/dL and indirect at 1.3mg/dL with mild jaundice.

Two weeks after discharge he had a total bilirubin level of 9.7mg/dL and his follow up US at 4 weeks reported that the bilateral adrenal hematomas had decreased in size and appearance.









Ultrasound diagnostic images. Bilateral adrenal masses composed by mixed echogenicity with an anechoic center and peripheral echogenic areas. Doppler negative

■ DISCUSSION

AH is more common in neonates than in older children or adults and is the most common adrenal mass in neonates. This condition varies in presentation, the most frequent being an abdominal mass alone or a mass with jaundice. Some infants are totally asymptomatic. Such hemorrhage sometimes occurs prenatally. Surgical intervention is not indicated or needed if an imaging diagnosis can be made. US has been found to be extremely useful in the diagnosis of adrenal hemorrhage. (1,4)

The exact cause of AH is unknown but factors which have been implicated are:
(a) Stress and trauma at birth,
(b) Anoxia and
(c) Systemic disease thrombocytopenia purpura, hemorrhagic disease of the newborn, septicemia, and congenital syphilis. (4–7)

Trauma appears to be the most common factor. Neonatal adrenal hemorrhage occurs most frequently between the second and seventh days of life. The presenting symptoms vary with the degree of bleeding. Classic AH presents as an abdominal mass, usually right-sided, with hyperbilirubinemia. The right adrenal is involved in 70% of cases. Bilateral involvement is found in 5–10% of cases. Establishing the diagnosis of AH will prevent surgical exploration in most cases, particularly in relatively stable neonates presenting with abdominal mass. (6)

Ultrasound imaging, the most effective modality of diagnosis and follow-up for babies, avoids unnecessary laparotomy. With US the adrenals are easily identified by an experienced observer and infants can be evaluated without radiation exposure or sedation.(2) In neonates, the normal adrenal glands are clearly visualized at US and consist of a hypoechoic cortex and a thin, echogenic medulla.

Gray scale US of the abdomen in infants with an abdominal mass, such as adrenal hemorrhage, is an extremely useful procedure. It can localize the mass and its relationship to surrounding organs, differentiate cystic, solid and mixed lesions, and expedite the diagnostic evaluation.

The pattern of echogenicity of an adrenal hematoma depends on its age:
- an early-stage hematoma appears solid with diffuse or in homogeneous echogenicity
- as liquefaction occurs, the mass demonstrates mixed echogenicity with a central hypoechoic region and eventually becomes completely anechoic and cyst-like
- calcification may be seen in the walls of the hematoma as early as 1-2 weeks after onset and becomes gradually compact as the blood is absorbed
- Color Doppler and power Doppler imaging confirm that the mass is avascular.(2,8)

Other imaging techniques as magnetic resonance imaging (MRI) and computer tomography scans (CT) are helpful for further characterization of the adrenal mass. MR imaging is useful in the diagnosis of coexistent renal vein thrombosis and is often used to determine whether blood is the sole component of the hematoma, a finding that most likely indicates a benign cause. It can also be used to image an adrenal hematoma determining its age. CT is the method of choice for identifying and following adrenal hemorrhage in all patients, but neonates.(2)

Differential diagnosis in neonates:
- Neuroblastoma, often associated with vascularity and sometimes with liver metastases.
- Congenital adrenal hyperplasia, which is bilateral and the glands are enlarged having a cerebriform contour.

(9,10)

■ CONCLUSION

Adrenal hemorrhage should be considered as differential diagnosis of neonatal hyperbilirubinemia, especially in newborns without Rh and ABO incompatibility, hemolytic anemia or jaundice of unexplained cause, particularly associated with traumatic delivery, neonates large for gestational age and infants of diabetic mothers. The latter two factors were present in this case.

Ultrasound should be the diagnostic procedure of choice, as it is non-invasive, safe, does not require infant sedation, is easily performed and inexpensive. Ultrasound can also be used for follow up.

Hemorragia suprarrenal bilateral neonatal: presentación de un caso

Resumen

La hemorragia suprarrenal es una condición clínica poco común (0,2%--0,55%) durante el período neonatal, que puede conducir a una aguda crisis adrenal, al shock y la muerte, si no es reconocida rápidamente y tratada adecuadamente. El precario suministro de sangre de la glándula suprarrenal la hace proclive a la hemorragia. El diagnóstico diferencial incluye el riñón multiquístico, la hidronefrosis, el neuroblastoma y el nefroblastoma. La causa de la hemorragia suprarrenal se ha asociado con un parto traumático y la hipoxia, el shock, la sepsis y la coagulación intravascular. Los síntomas clínicos pueden incluir anemia, ictericia, masa abdominal, hinchazón dolorosa con coloración azulada del escroto, insuficiencia suprarrenal aguda o shock. La gran mayoría de los pacientes con hemorragia suprarrenal no tienen signos clínicos evidentes de insuficiencia suprarrenal. El diagnóstico se hace generalmente de manera incidental con una imagen realizada por otro motivo. Dado que los pacientes con hemorragias suprarrenales unilaterales o bilaterales se tratan generalmente de forma conservadora; son preferidas las técnicas de diagnóstico no invasivas. En este trabajo se presenta un caso de hemorragia suprarrenal bilateral neonatal diagnosticada por ultrasonido.

Palabras clave

Hemorragia suprarrenal, hiperbilirrubinemia, masa suprarrenal, diagnóstico por ultrasonido abdominal

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1st Case of Female-to-Male Sexual Transmission of Zika

• Reported FRIDAY, July 15, 2016 (HealthDay News) -- A New York City woman who became infected with the Zika virus on a trip outside the United States passed the infection to her boyfriend during sex, city health officials reported.

It's the first reported case of female-to-male sexual transmission of Zika – before this case, sexual transmission had only been reported as spreading from men to women.

The woman, in her 20s, said she had traveled to a Zika-endemic area and developed headache, cramps, fever, fatigue, rash and other symptoms during the day she flew home and after returning to New York City.

She had unprotected vaginal sex with her partner on the day of her return, and about a week later her male partner came down with symptoms of what also turned out to be Zika infection.

Both individuals recovered from Zika illness, which is usually transient.

Zika is typically transmitted via the bite of the *Aedes aegypti* mosquito, and the greatest danger is when infection occurs in pregnancy. These infections have been tied to thousands of cases in Latin America of a devastating birth defect known as microcephaly, in which babies are born with abnormally small heads and neurological issues. The New York City woman was not pregnant, city health officials said.

Infectious disease experts noted that while mosquitoes are by far the most common means of Zika transmission, sexual transmission can sometimes occur.

The case documented in New York City isn't surprising, said Dr. Marc Siegel, a professor of medicine at NYU Langone Medical Center in New York City.

"Anytime you see male-to-female transmission, there's always the risk of female-to-male transmission – we found that with HIV," said Siegel, who was not part of the research.

He believes that the New York City case is probably not the first female-to-male transmission – just the first such case reported.

However, "if our concern about Zika is the risk of birth defects, female-to-male transmission isn't going to increase that risk, unless the male has multiple partners," Siegel explained.

He also believes that sexual transmission of the virus should not keep people from protecting themselves from mosquitoes – the main source of infection – whenever they travel to Zika-endemic areas.

The report was published July 15 in the U.S. Centers for Disease Control and Prevention's *Morbidity and Mortality Weekly Report*.

In the face of the growing reality of sexual transmission of Zika, U.N. health officials issued updated guidelines earlier this year aimed at helping prevent infection. The advisory urges that women planning to become pregnant wait at least eight weeks before trying to conceive if they or their partner live in – or are returning from – areas where the Zika virus is active. The guidelines had previously recommended a four-week waiting period.

And if the male partner has had symptoms of Zika infection, couples should wait six months before trying to have a baby, the World Health Organization officials added.

To limit any potential spread of Zika virus via mosquitoes, health officials on the federal, state and local level are deploying a three-pronged strategy: improving mosquito control; expanding their ability to test for Zika; and urging the public to protect themselves against mosquitoes.

Women of child-bearing age who live in an active Zika region should protect themselves from mosquito bites by wearing long-sleeved shirts and long pants, using mosquito repellent when outside, and staying indoors as much as possible, according to the CDC.

SOURCES: Marc Siegel, M.D., professor, medicine, NYU Langone Medical Center, New York City; July 15, 2016, *Morbidity and Mortality Weekly Report.*