

**Discordant lower urinary tract obstruction in a dizygotic twin pregnancy treated with vesicoamniotic shunt**

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## Discordant lower urinary tract obstruction in a dizygotic twin pregnancy treated with vesicoamniotic shunt

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### Case Report:

A woman with a dizygotic twin pregnancy was gone through routine follow-up. The first-trimester screening test revealed a low-risk pregnancy and bladders were visible. At the detailed scan at 20 weeks gestation, fetuses were noted as male and female, with normal fetal structures and normal amniotic fluid indexes. The scan at 29 weeks gestation revealed that the male fetus had an enlarged bladder and dilated proximal urethra typically of a key-hole bladder (Figure 1) and that the fetal kidneys had grade 3 hydronephrosis. Decreased amniotic fluid volume was observed. A late onset LUTO was suspected, and vesicoamniotic shunt was performed to assess renal functions. Fetal urine biochemical parameters are summarized in Table 1 and considered normal, according to diagnostic indices determined *a priori* (Biard et al. 2005). Before renal function was severely affected, a VAS was placed for LUTO possibly due to posterior urethral valve syndrome (PUVs) after discussion of the situation with parents. After informed consent was obtained from parents, a double pig tailed vesicoamniotic shunt (Rocket Medical) was inserted under ultrasound guidance through the mother's abdomen into the fetal bladder in one attempt on the third day of vesicoamniotic shunt. Before the placement of the catheter, a fetal urine sample was taken as a part of serial urine analysis to decrease the risk of repeated interventions (Table 1). Twenty-four hours after the intervention, the inner tip of the VAS was seen inside the empty bladder (Figure 2). Nine days after the intervention, the right kidney still had a grade 3 hydronephrosis, partial recovery was monitored in the left kidney (grade 2 hydronephrosis), and the bladder was empty with VAS in the correct place. Twenty-

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3 five days after the intervention, vesicouretral reflux (VUR) in the right ureter was suspected  
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5 (Figure 3).  
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8 A caesarean section was performed at 35 weeks gestation due to premature contractions. A  
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10 male infant of 2420 g with Apgar scores of 9 at 1 minute, and 10 at 5 minutes was delivered.  
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12 The external tip of the shunt was on the abdomen and external genitalia of the infant appeared  
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14 normal (Figure 4). The female fetus was completely normal. Two days after the birth a urea  
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16 level of 41.4 mg/dl and a creatinine level of 1.67 mg/dl were observed. Due to the increased  
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18 creatinine level, a possible semi-obstruction of the shunt was suspected. An attempt was made  
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20 to insert an 8 French Foley catheter through the urethra, but it was not successful. At follow-  
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22 up, urea and creatinine levels were higher (86 mg/dl and 3 mg/dl, respectively), and the total  
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24 bilirubin level was 14 mg/dl. Hydration and phototherapy were performed. Soon after the  
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26 therapy, urea and creatinine levels started to decrease. A cystoscopy was performed by a  
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28 pediatric urologist, and no sign of the membrane or atretic or stenotic portion of the urethra  
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30 was detected. Only a right ureteral orifice dilatation was observed, and the bladder shunt was  
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32 removed. After the intervention the baby was able to easily void. Voiding cystourethrography  
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34 showed grade IV vesicoureteral reflux in the right lower pole. No reflux was identified on the  
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36 left. Final ultrasound examination showed mild increase on the bladder trabeculation, both  
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38 distal ureters were dilated and proximal ureters were large and elongated. Right and left  
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40 kidney sizes were; 5x2.5x2 cm and 6x3.8x2.2 cm, respectively, and both had a parenchymal  
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42 thickness of 7 mm, with natural parenchymal echogenities. According to the Society Fetal  
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44 Urology, the right kidney had a grade 3 dilatation and the left kidney had a grade 2 dilatation.  
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46 After voiding, minimal residual urine was detected in the bladder. Renal function was  
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48 evaluated by intravenous Tc-99m DMSA. Differential functions for the left and right kidney  
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50 were 71 % and 29 %, respectively, showing low uptake and a dilated collecting system for the  
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3 right kidney. The latest serum creatinine and urea levels were normal (36 mg/dl and 0.96  
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5 mg/dl, respectively). After one year of follow-up, the baby was normal.  
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### 8 **Discussion:**

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10 Lower urinary tract obstruction influences 2.2 in 10,000 births (Anumba et al. 2005). The  
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12 most common cause of LUTO is PUVs. Bladder outlet obstruction can cause distended  
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14 bladder, dilated ureters, hydronephrosis and irreversible renal damage. Consequently, prenatal  
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16 intervention should be offered to the parents before the kidneys are severely affected.  
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20 To evaluate the renal function serial urine analysis with vesicocentesis should be performed,  
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22 and the second sampling, 48 to-72 hours after the first, should be considered in determining  
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24 whether a good or bad prognosis is expected. If the fetus is assessed as having a poor  
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26 prognosis, conservative management should be applied because the kidneys are severely  
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28 affected. Fetuses with a good prognosis are candidates for the VAS procedure. Our case, due  
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30 to fetal urine electrolytes and osmolarity, seemed to be in the good outcome group, though the  
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32  $\beta$ 2-microglobulin level was a little higher than expected. As a result, we predicted that the  
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34 case was in the good prognosis group and eligible for the procedure.  
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38 Fetal karyotyping before the procedure is usually recommended because of a chromosomal  
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40 abnormality incidence of 8 to-23 % in obstructive uropathy (Brumfield et al. 1991). Because  
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42 of the normal screening test, the detailed scan, and the extra risk of the cordocentesis  
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44 procedure, the parents did not agree to genetic testing.  
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48 In the present case, despite the evidence of key-hole bladder the pediatric urologist could not  
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50 detect by cystoscopy any sign of atresia or stenosis in the urethra. Membranes were probably,  
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52 ruptured during the first unsuccessful catheterization attempt. Persistence of hydronephrosis  
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54 and right dilated ureters after the VAS procedure suggest that VUR was accompanied by  
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56 PUVs, the coexistence of which occurs 50% of PUVs cases (Hutton, 2005).  
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3 Prenatally diagnosed LUTO has been reported in twins previously (Maruotti et al. 2006).  
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5 Holden and colleagues (1999) followed three dichorionic diamniotic twins discordant for  
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7 LUTO without any intervention. All of the affected twins died soon after birth due to  
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9 pulmonary hypoplasia caused by anhydroamnios; nonetheless, co-twins with a normal  
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11 amniotic sac were normal, so Holden and colleagues suggested therapeutic septostomy for  
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13 this condition. Evidence in agreement with this was reported (Kontopoulos et al. 2005) in  
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15 monoamniotic twin pregnancy where the affected twin did not develop lung hypoplasia due to  
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17 the sufficient amnion in the shared sac, but the amount of amnion was not enough to relieve  
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19 renal functions in the affected twin. In another case series (Lipitz et al. 1993), VAS was  
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21 implemented after vesicocentesis in one monochorionic twins which was complicated by  
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23 chorioamnionitis and vesicocentesis was performed in two dichorionic twins. Again, in cases  
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25 (Sepulveda et al. 2005) discordant for LUTO, no fetal intervention was performed in three  
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27 dichorionic twins and vesicocentesis was performed in two monochorionic twins. As can be  
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29 seen from medical literature, the VAS procedure was rarely executed for LUTO in twins, and  
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31 almost never applied in dichorionic twins.  
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39 In conclusion, although fetal intervention in twin pregnancies is a risky procedure,  
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41 management of late onset LUTO with VAS could give the affected twin a chance for future  
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43 renal functions in relevant cases. However, the risk of preterm birth must be discussed with  
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45 the parents.  
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48 ***Declaration of interest:*** The authors report no conflicts of interest. The authors alone are  
49  
50 responsible for the content and writing of the paper.  
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Table I. Fetal urine biochemistry

Chemical	Measurement		Good prognostic indicator*
	Vesicocentesis	VAS	
Sodium (mEq/L)	73	74	<100
Chloride (mEq/L)	61.8	62.3	<90
Calcium (mg/dL)	1.3	1.23	<8
Osmolarity (mOsm/L)	193	171	<200
$\beta$ 2-microglobulin (mg/L)	-	8	<6

\* Good prognostic indicators edited according to Biard et al 2005.



Figure 1. At 29 weeks gestation the dilated proximal urethra and enlarged bladder typical key-hole bladder view of the twin discordant for lower urinary tract obstruction.





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Figure 2. Twenty-four hours after vesicoamniotic shunt (VAS) replacement, empty bladder and the tip of the shunt is visible (arrow).

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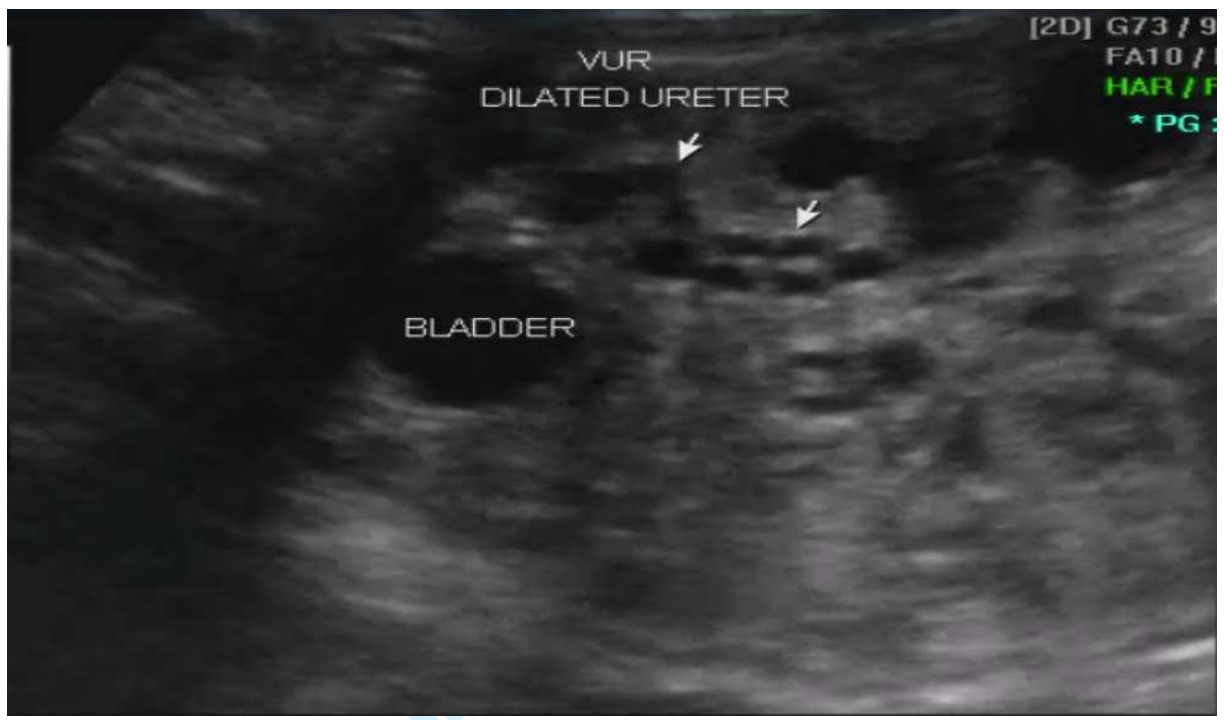


Figure 3. Twenty –five days after the intervention, dilated right ureter reminding vesicoureteral reflux (VUR) (arrows).



Figure 4. Dizygotic twins after birth. The male fetus (on the left) with vesicoamniotic shunt and normal external genitalia.