

## CASE REPORTS

# Evaluation of the Lower Urinary Tract Function in Caudal Duplication (Dipygus) Anomaly

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● Caudal duplication is a rare anomaly with less than 30 reported cases. For those patients who also have double bladders, there are not enough data regarding the function of the lower urinary tract. A boy with caudal duplication anomaly was evaluated fluoroscopically and urodynamically. The results of the evaluation showed that the bladders were filling and emptying synchronously with normal and almost identical detrusor pressures. The child did not require either or both bladders to be resected. Because the level of the duplication in dipygus cases varies, thorough evaluation of the lower urinary tract, including urodynamics, should be considered for every case.

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**INDEX WORDS:** Caudal duplication, bladder anomalies, lower urinary tract function, urodynamics.

**C**AUDAL DUPLICATION is a rare anomaly with less than 30 reported cases in the literature.<sup>1</sup> Classically, patients present with two pelvises, four legs, intestinal and anal duplications, double genitalia, and an omphalocele. For those who also have double bladders, there are not satisfactory data in the literature regarding the function of the lower urinary tract. A boy with caudal duplication anomaly who underwent surgery early in the neonatal period was evaluated urodynamically and fluoroscopically to see if his double bladders were functioning normally in concordance.

### CASE REPORT

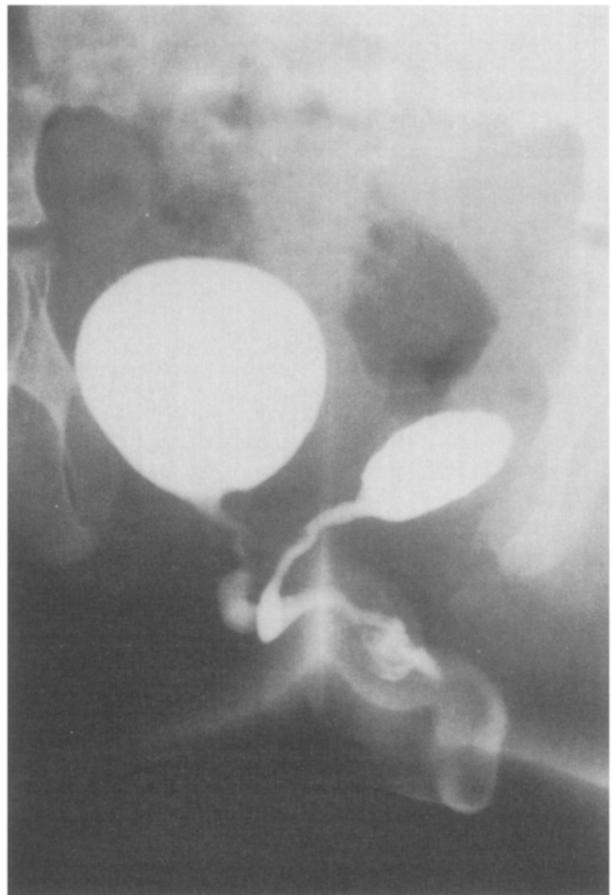
A boy born with a caudal duplication anomaly underwent surgery in the neonatal period. His extra pair of legs and a pelvis had been removed by disarticulation, an ileal duplication was resected, and left orchiopexy was performed. Follow-up until 17 months of age showed no complications. The boy was reevaluated at the age of 4. He did not have any urinary tract infections since birth. He did not complain of voiding difficulties, incontinence, or enuresis, and he was currently voiding five to seven times a day, with acceptable urine volumes and voiding pattern. The motor and sensitive function of the lower extremities and perineum were

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normal. The x-rays of the distal vertebral column, and sacrum looked normal. Because there was no sign of neurological impairment a magnetic resonance imaging (MRI) study was not considered. The boy had two normally functioning kidneys and ureters draining separately into two totally isolated small bladders, which were documented with intravenous urogram. A retrograde urethrogram and voiding cystogram showed two bladders located laterally on each side of the pelvis, and two separate urethras joining into one in the proximal penile level, which was confirmed by cystoscopy (Fig 1). During fluoroscopic examination, the left bladder was smaller than the right in capacity, and had a low-grade vesicoureteric reflux, but the bladders contracted and emptied simultaneously, with no apparent residual urine or retrograde flow. For urodynamic assessment, an initial free flow was obtained, which looked normal. Then two double lumen pressure catheters were



**Fig 1. Voiding cystourethrogram shows two bladders, and urethras joining in the penile level.**

inserted through the single distal urethra, and were placed separately in the bladders under fluoroscopy. Using an eight-channel polygraph (Synectics Poly URO System, Synectics AB Stockholm, Sweden) the two bladders were filled using the same infusion rates (10% of the expected bladder capacity in milliliters according to age), while the pressures, including abdominal, were recorded during bladder filling and voiding. All of the recorded pressures were normal during both phases, and pressures were almost identical for both bladders, with a difference of less than 5 cm H<sub>2</sub>O at any time. The sensation looked normal. There were a few nonsignificant low-amplitude detrusor contractions during filling, which were observed synchronously in both bladders. The two bladders emptied without significant residual urine.

#### DISCUSSION

Duplication of the bladder is common in most cases of caudal duplication. Morphological variations in this rare abnormality have been clearly documented by contrast radiographic studies, and these variations have led to a number of complicated surgical procedures performed in an attempt to correct these exceptional malformations.<sup>2</sup> However, functional evaluation of the lower urinary tract including urodynamics may provide valuable information and modify the management of major urologic anomalies.

The results of the evaluation may be the evidence

for an identical parasympathetic innervation of the two separate bladders existing in this case of caudal duplication anomaly. In the 28 somites stage of the embryo, splitting of the primitive streak results in the production of abnormalities of the lower part of the body with duplication of the bowel and cloaca.<sup>3</sup> According to the clinical status and the results of the evaluation the child is unlikely to require either or both of his bladders resected. Apart from his low-grade unilateral vesicoureteric reflux, this 4-year-old boy, with normal upper urinary tract function and without any specific complaint suggesting lower urinary tract malfunction, might be followed-up without having to perform these invasive studies. However, documentation of even the normally functioning double bladders in such an odd urologic situation have been academically instructive. Because the level of duplication in dipygus cases varies, the findings of this case do not preclude any other case of caudal duplication with two bladders functioning autonomously. Therefore, every particular case of dipygus, or bladder duplication deserves thorough evaluation of the urinary tract, including urodynamics.

#### REFERENCES

1. Ulman I, Rowe MI, Erdener A: Four-legged humans and six-legged animals: An historical perspective. The International Society for the History of Paediatric Surgery. Fourth Biennial Meeting, July 21, 1992, Leeds, UK (abstr 4)
2. Dominguez R, Rott J, Castillo M, et al: Caudal duplication syndrome. *Am J Dis Child* 147:1048-1052, 1993
3. Pena A, Oliveras M, Palomaque L: Genito-urinary and intestinal duplication. *Br J Urol* 32:285-289, 1960