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Hypoplastic Dysplastic Kidney with a Vaginal Ectopic Ureter Identified by Technetium-99m-DMSA Scintigraphy

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Ectopic insertion of a ureter draining a hypoplastic dysplastic kidney is a significant cause of urinary incontinence in girls. In this case, such a kidney was detected with ^{99m}Tc-DMSA scintigraphy but not by intravenous pyelography. Scintigraphy facilitated further delineation of the anatomy with CT prior to nephrectomy. Based on this case and a literature review, we suggest that ^{99m}Tc-DMSA scintigraphy be performed early when evaluating girls with urinary incontinence.

Key Words: technetium-99m-dimercaptosuccinic acid; kidney; urinary incontinence; ectopic ureter

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CASE REPORT

An 8-yr-old girl with urinary incontinence was referred for renal cortical scintigraphy due to a suspected ectopic hypoplastic right kidney with an ectopic ureteral orifice. The patient's urologic history was otherwise remarkable only for urosepsis at age 3 wk. Diagnostic evaluation at that time revealed left vesicoureteric reflux (VUR) and a left megaureter. Ultrasonography and intravenous pyelography (IVP) showed a solitary left kidney with pelvicalyceal dilatation. At cystoscopy, a right ureteral orifice was not identified. Left ureteral reimplantation was performed, and a follow-up voiding cystourethrogram indicated that VUR was no longer present. Subsequent to this, and after toilet training at a normal age, she regularly had damp underwear every day and night. Before referral to our institution, work-up had included four additional IVP studies, which demonstrated only a normal appearing left kidney, and urodynamic evaluation, which showed normal uroflow, normal urethral pressure profile and adequate bladder compliance. Bladder training exercises, biofeedback and oxybuty-

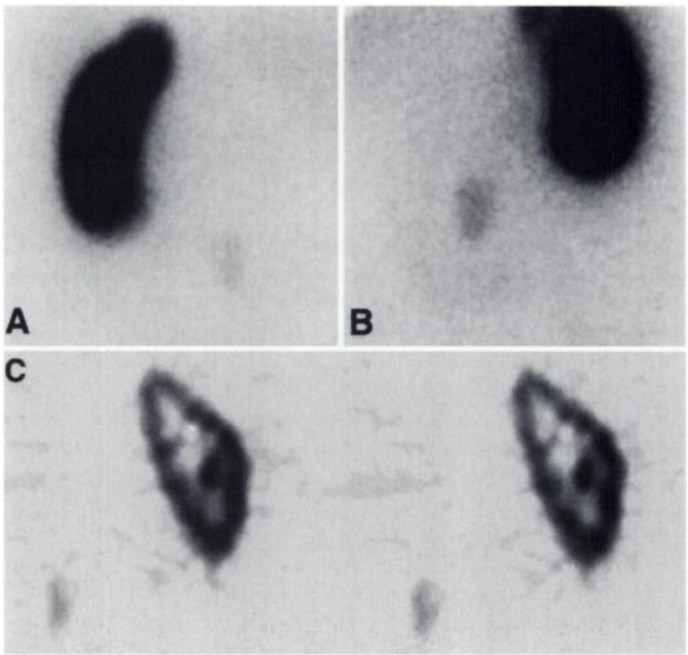

FIGURE 1. Planar images: (A) posterior; (B) anterior; (C) coronal SPECT reveal tracer localization in a small ectopic right kidney.

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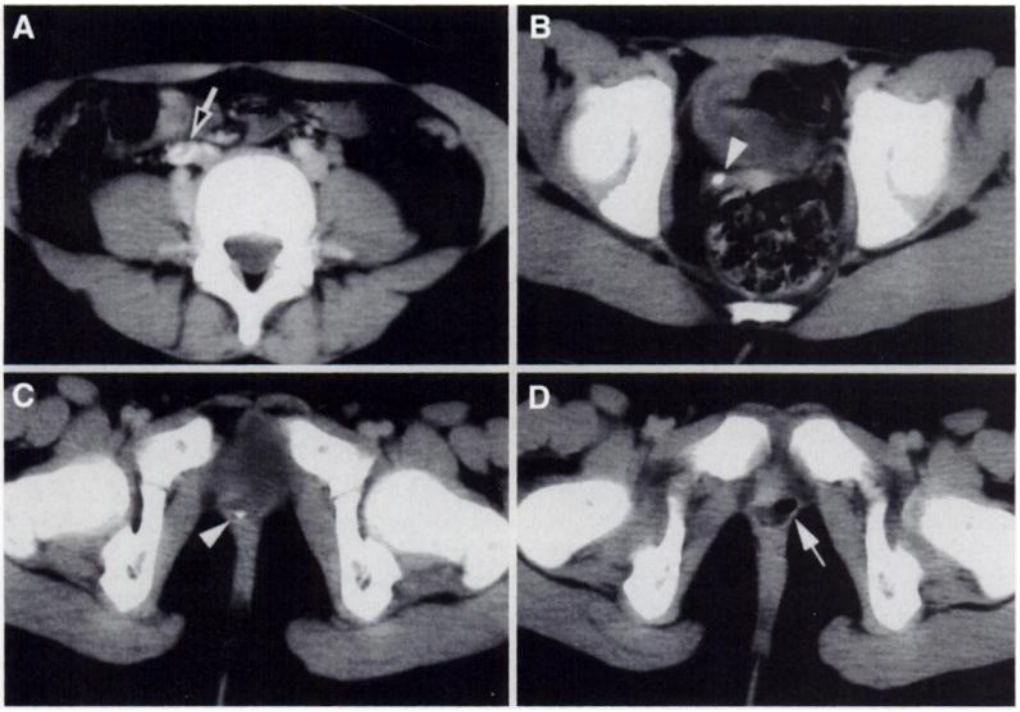

FIGURE 2. The right kidney, which is opacified after contrast administration, is denoted by an arrow (A). Opacification of excreted contrast enables visualization of the right ureter including (B) its point of vaginal insertion; (C) arrowheads. Anatomic definition resulting from a small amount of (D) air (arrow); and contrast within the vagina allows confirmation that the structure into which the right ureter drains is the vagina.

nin (5 mg PO, TID) had been unsuccessful in alleviating her incontinence.

Four hours after intravenous administration of 67.6 MBq ^{99m}Tc-DMSA, anterior and posterior high-resolution planar and SPECT images were obtained. A small right kidney and a normal appearing left kidney were seen (Fig. 1). Differential ^{99m}Tc-DMSA uptake was 1% by the right kidney and 99% by the left kidney. Correlated with a physical examination and the use of a ^{99m}Tc point source marker, the right kidney was noted to be at the level of the umbilicus. After intravenous administration of 50 cc Ioversol-68%, thin-section CT at this level showed a 2 × 1-cm right-sided kidney in the retroperitoneum adjacent to the lower lumbar spine (Fig. 2A). Contrast within the right ureter allowed for delineation of its course posterior to the bladder and into the vagina (Fig. 2 B,C,D). The patient was taken to surgery where, after cysto/vaginoscopy demonstrated a small ureteral orifice just to the right of midline and inferolateral to the normal urethral meatus (Fig. 3), a small right kidney and its ureter (Fig. 4) were removed laparoscopically. Histologic sections showed the kidney to be dysplastic (Fig. 5). Postoperatively, the patient has done well, with complete resolution of her incontinence.

DISCUSSION

Continuous urinary incontinence in young girls is usually due to an ectopic insertion of the ureter (*1*). To cause urinary incontinence, the ureter must insert below the external urethral sphincter in positions such as the vagina, perineum or introitus. Insertion of a ureter into the introitus, as was present in the case reported here, results from a more lateral than normal origin of the ureteric bud from the mesonephric duct. Consequently, the ureter remains connected to Gartner's duct. This mesonephric duct remnant courses from the hymen along the lateral vaginal wall to the adnexa (*2*).

A ureter with an insertion that is so markedly displaced as to cause incontinence typically drains a kidney, or segment of a kidney, that is hypoplastic and dysplastic. This association is believed to reflect failure of an abnormal ureteral bud to induce differentiation of the metanephric blastema into functional renal tissue (*3*).

When an ectopic ureter draining a dysplastic upper segment of a duplex kidney is responsible for incontinence, IVP confirms the diagnosis by showing displacement of the ipsilateral lower pole. Conversely, when a hypoplastic dysplastic kidney with a single ectopic ureter produces incontinence, IVP is not likely to

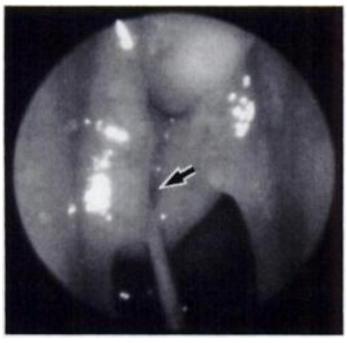

FIGURE 3. Insertion of probe into the ectopic ureteral orifice after detection by vaginoscopy.

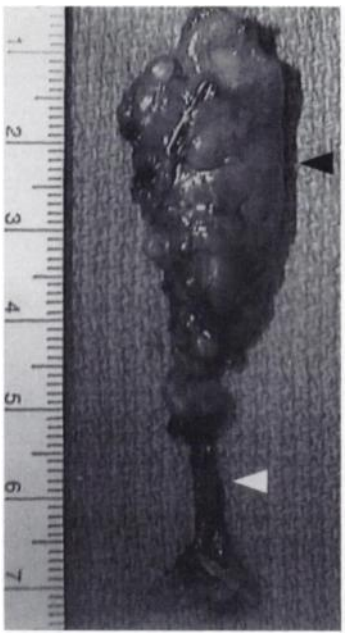

FIGURE 4. Surgical specimen consists of a small kidney (black arrowhead) and its ureter (white arrowhead).

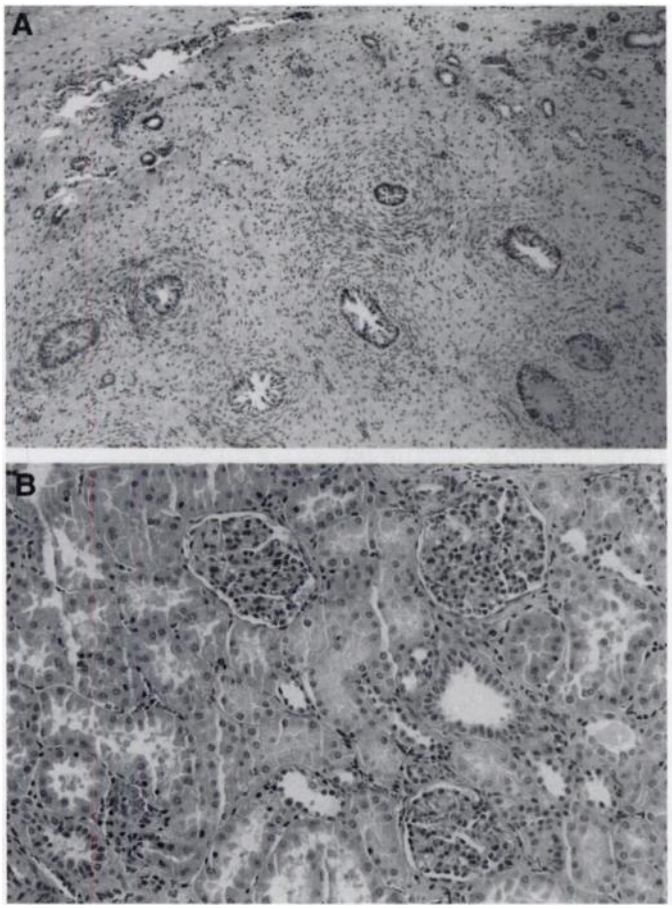

FIGURE 5. (A) Histology shows relatively normal appearing glomeruli and proximal tubules. (B) Primitive ducts surrounded by concentric layers of mesenchymal cells, a specific histologic marker of renal dysplasia, are visualized.

be helpful. With such anatomy, as in this patient, IVP typically reveals only the normal or hypertrophied contralateral kidney. The hypoplastic dysplastic kidney goes undetected (*1,2,4–6*). In a recent report, the ectopic ureter and its kidney were not visualized by IVP in three of four patients where a hypoplastic dysplastic kidney drained through its ureter into the vagina (*1*). Two of these three children had ^{99m}Tc-DMSA scintigraphy; the abnormal kidney was identified in both (*1*). Borer (*5*) and Limbert (*4*) also have reported cases of ^{99m}Tc-DMSA localization in hypoplastic dysplastic kidneys associated with urinary incontinence that were not visualized with IVP. A hypoplastic dysplastic kidney is also unlikely to be detected with ultrasonography (*1,5,6*). Published reports suggest that when ^{99m}Tc-DMSA scintigraphy fails to demonstrate functional renal parenchyma, drainage of a hypoplastic dysplastic kidney as a cause of incontinence is unlikely (*1,4,5*). Weiss et al. (*6*) have, however, described one case where ^{99m}Tc-DMSA scintigraphy, as well as ultrasonography, did not reveal a hypoplastic dysplastic kidney that was subsequently identified by a retrograde study performed during vaginoscopy (*6*). An IVP was not performed in that child.

CONCLUSION

In this case, contrast-enhanced CT was directed by the demonstration of a small ectopic right kidney with ^{99m}Tc-DMSA scintigraphy. CT further delineated the anatomy. Performance of CT without scintigraphic localization of the hypoplastic kidney would require multiple thin-section images through the abdomen and pelvis. Such an examination would carry a high radiation dose and be time-consuming. The combination of ^{99m}Tc-DMSA scintigraphy and CT allowed for appropriate surgical intervention.

We suggest ^{99m}Tc-DMSA scintigraphy be performed early in the diagnostic evaluation of girls with continuous urinary incontinence. In cases where either IVP or ultrasonography demonstrates a single kidney, ^{99m}Tc-DMSA scintigraphy should be regarded as an essential next step in the diagnostic evaluation of girls with continuous urinary incontinence.

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