

## Ectopic Ureterocele in Adults with a Comparison of the Anomaly in Children

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**Abstract.** Ectopic ureteroceles, while not uncommon in children, have been reported only rarely in adults. We present five adults with ectopic ureteroceles with emphasis on the varied clinical and radiographic manifestations. These findings were compared with those in 32 children with ectopic ureterocele.

It was found that the clinical presentation differed in adults and children, but the radiological findings were similar. The diagnosis was in some cases delayed for many years. The anomaly could not be detected by imaging means in two of five adults and eight of 32 children, and was found only at surgery.

**Key words:** Ectopic ureterocele, adults — Ectopic ureterocele, children — Ureter, abnormality, imaging.

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An ectopic ureterocele is a congenital anomaly defined as the cystic ballooning of the distal end of a ureter ending ectopically in the bladder neck or the urethra. This ureter usually drains the upper pole of a duplex kidney.

An ectopic ureterocele, according to its size and location, is often obstructive and causes urinary tract infection in the majority of cases [1, 2]. It is therefore usually detected in infancy or early childhood. Now-

adays with the increasing use of prenatal ultrasound, a dilated fluid-filled collecting system and sometimes even the ureterocele [3] can be demonstrated in utero. After the infant is born the anomaly can be delineated better, again by ultrasonography, and aided by other imaging studies.

Ectopic ureteroceles detected in adults are very rare. We present five such cases with emphasis on the clinical presentation and the radiological diagnosis. These data in adults will be compared with those of children with the same condition, diagnosed in our hospital.

### Patients and Methods

We reviewed the records and imaging studies of five adult patients with ectopic ureterocele and tabulated the findings. We also reviewed the clinical and radiological data of all children with ectopic ureterocele, who were diagnosed and/or treated in this institution in the 30-year period from 1960–1990. We then compared these two patient populations to see whether the anomaly causes identical symptoms and radiological findings in adults and children.

### Results

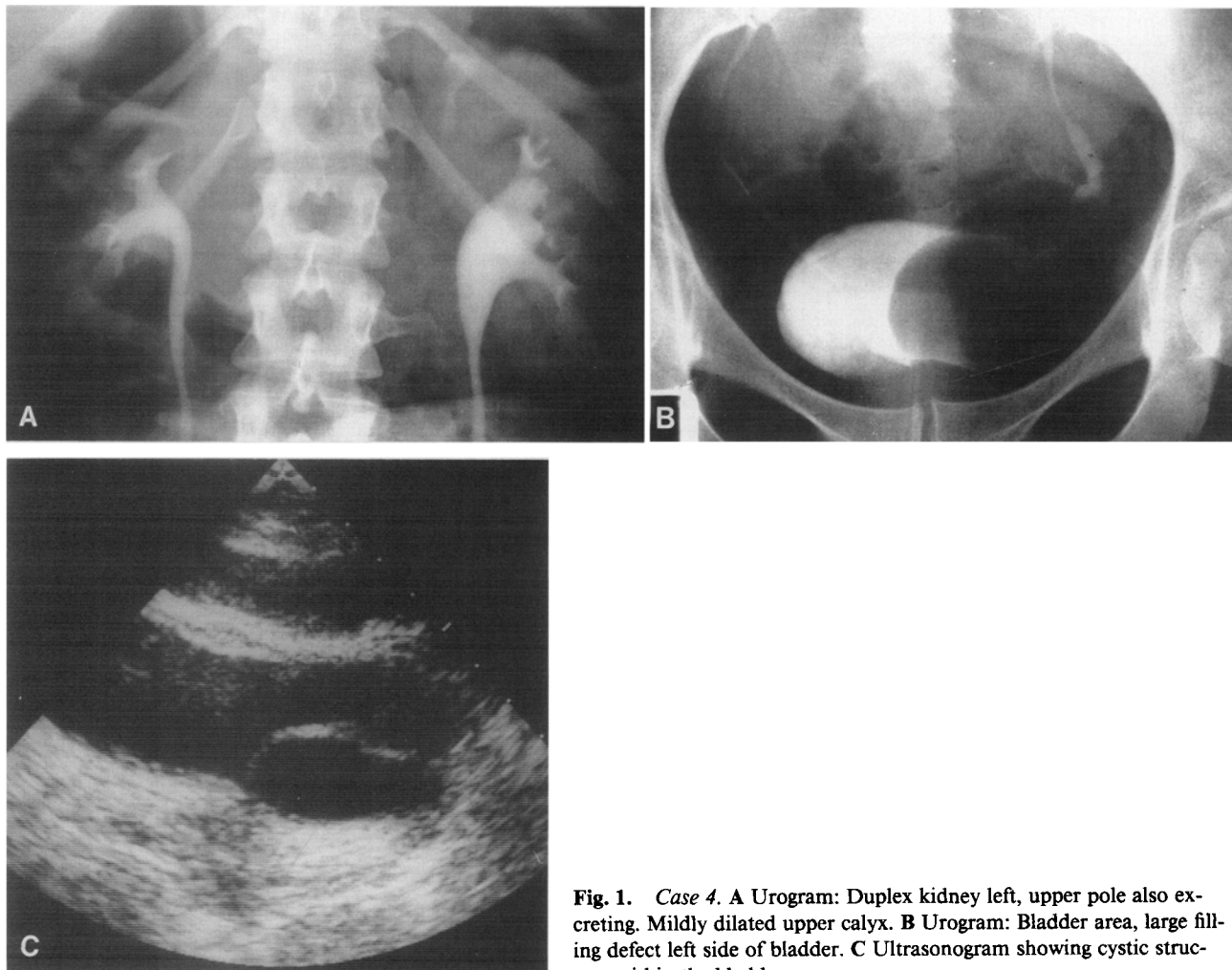
The relevant clinical and radiological data of the five adult patients with ectopic ureterocele are shown in Table 1. There were three men and two women, their ages ranging from 21–39 years. Urinary tract infection was not a feature of the clinical presentation. Leukocytes were detected in the urine of only one man. All three male patients had hematuria (in one case calculi were present in the ureterocele, a possible cause for the hematuria). In the two women,

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**Table 1.** Clinical and radiological findings in five adults with ectopic ureterocele

Case no.	Age (yr)	Sex	Clinical presentation	Excretory urography	Cystourethrography	Comment
1	21	M	Hematuria Acute urinary retention	Bilateral duplex kidney Left upper system dilated Left ectopic ureterocele containing calculi	—	Ectopic ureterocele proven at surgery
2	26	M	Hematuria Leukocyturia	Hypertrophic left kidney Small contracted right kidney with clubbed calyces	Vesicoureteral reflux grade III to the right kidney	Unsuspected ectopic ureterocele with duplication at surgery
3	26	M	Left flank pain Microhematuria	Duplex right kidney Small left kidney with dilated calyces very dilated distal ureter	Vesicoureteral reflux grade IV to the left kidney	Unsuspected ectopic ureterocele with duplication at surgery
4	28	F	Left flank pain several hours after intercourse	Duplex left kidney Upper calyx dilated Large filling defect Left side of bladder	Normal	Anomaly confirmed at surgery
5	39	F	Abdominal pain	Bilateral duplex kidney Left upper collecting system + left ureter dilated Filling defect in bladder	Reflux to ectopic ureterocele	CT showed identical findings Anomaly confirmed at surgery



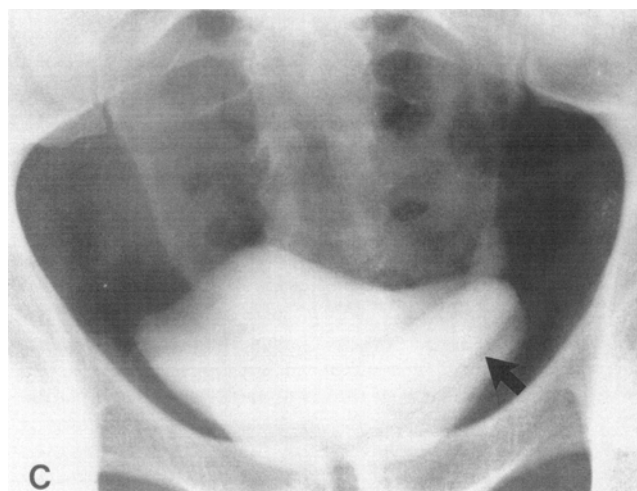
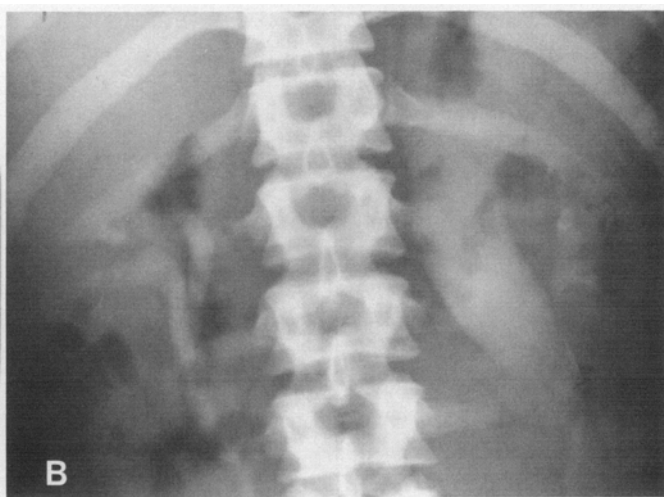
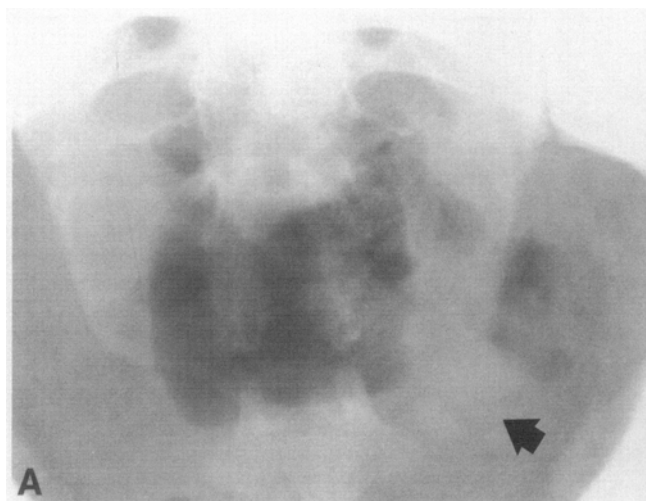
**Fig. 1.** Case 4. **A** Urogram: Duplex kidney left, upper pole also excreting. Mildly dilated upper calyx. **B** Urogram: Bladder area, large filling defect left side of bladder. **C** Ultrasonogram showing cystic structure within the bladder.

**Table 2.** Age at diagnosis of 32 children with ectopic ureterocele

	F	M	Total
1 month	1	2	3
1–12 months	8	4	12
1–2 years	3	—	3
2–5 years	6	3	9
5–10 years	4	1	5
Total	22	10	32

**Table 3.** Main presenting symptoms and signs in 32 children with ectopic ureterocele

	F	M	Total
Urinary tract infection	17	6	23
Prenatal abnormal ultrasonogram	1	3	4
Incontinence	2	—	2
Abdominal mass	1	—	1
Renal failure	1	—	1
Urinary retention	—	1	1
Total	22	10	32



**Fig. 2.** Case 1. **A** Plain film of the pelvis: Several calculi are visualized on the left. **B** Urogram: Bilateral duplication, the left upper collecting system is very dilated and displaces the lower collecting system laterally. **C** Bladder area shows one distal ureter and a longitudinal structure filled with contrast superimposed on the bladder, presumably the ureterocele (arrow).

symptoms had been present for 5–8 years before diagnosis.

As to the radiological findings, the characteristic features of a double collecting system in one kidney and a filling defect in the bladder were present in only one of the patients (Fig. 1, case 4). Ultrasonography showed a characteristic picture of a “cyst within a cyst” in the bladder (Fig. 1C). In another case, a double system with ureterocele and stones was demonstrated (Fig. 2, case 1). Combined studies

[computed tomography (CT), excretory urography (EU), and voiding cystourethrography (VCU)] led to the correct diagnosis in case 5 (Fig. 3). In the two male patients in whom a small kidney with blunted calyces and marked vesicoureteral reflux were demonstrated (Figs. 4 and 5, cases 2 and 3), an erroneous diagnosis of reflux nephropathy was made and only at surgery for ureteral reimplantation was the anomaly discovered.

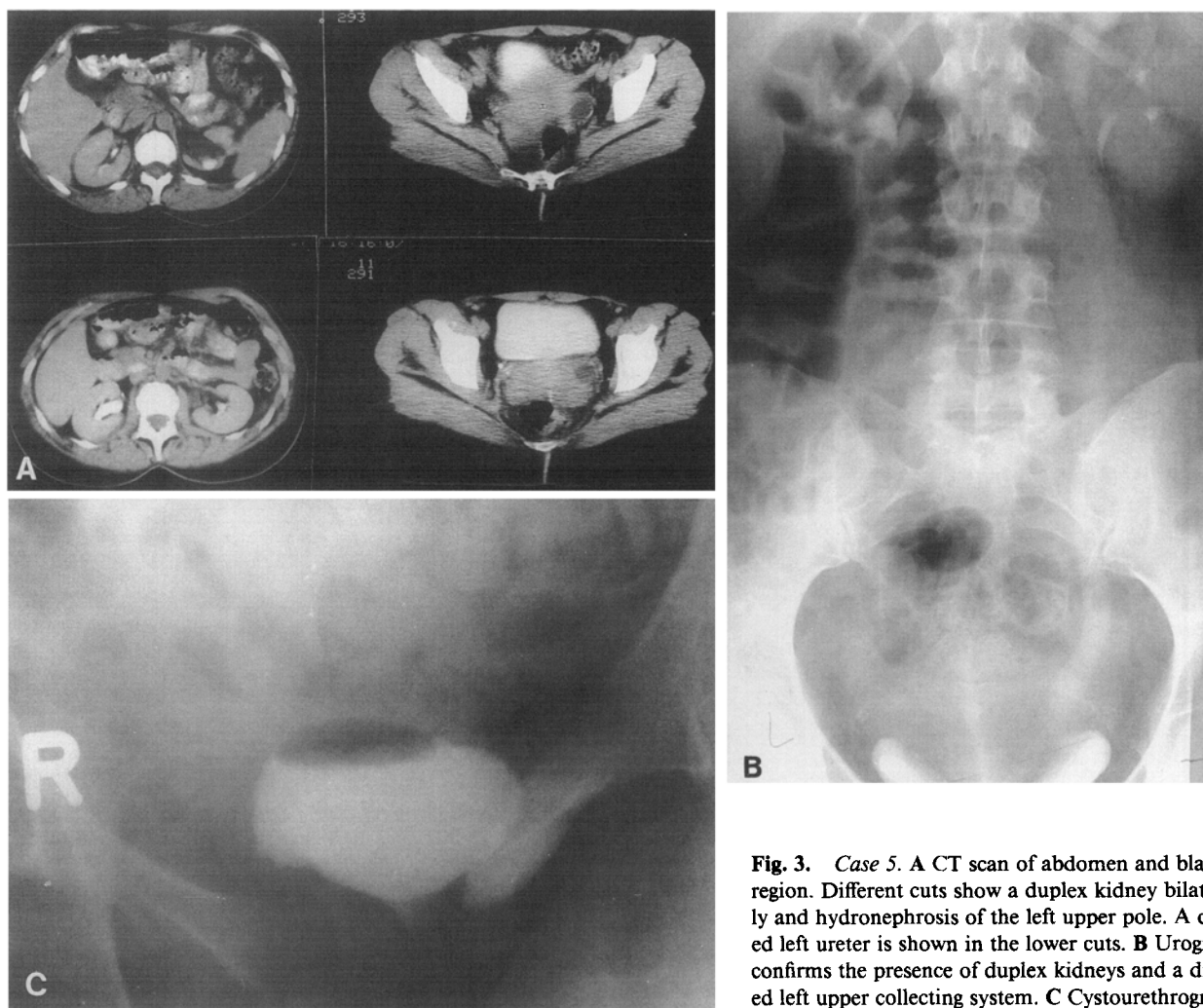
The results of comparative findings in children

**Table 4.** EU in 32 children with ectopic ureterocele (18 left, 13 right, 1 bilateral)

"Drooping lily" appearance	23
Upper pole not excreting 17	
Upper pole excreting 6	
Small kidney with reflux nephropathy	5
Small kidney with dilated collecting system	1
One kidney not excreting	3
Filling defect in bladder	24/32

**Table 5.** Voiding cystogram in 32 children with ectopic ureterocele

Vesicoureteral reflux	18
Left 10	
Right 7	
Bilateral 1	
Filling defect in bladder	14/32

**Fig. 3.** Case 5. A CT scan of abdomen and bladder region. Different cuts show a duplex kidney bilaterally and hydronephrosis of the left upper pole. A dilated left ureter is shown in the lower cuts. B Urogram confirms the presence of duplex kidneys and a dilated left upper collecting system. C Cystourethrogram shows reflux into ectopic ureter.

with ectopic ureterocele are shown in Tables 2–5. There were 32 children: 22 girls and 10 boys. Their ages at diagnosis ranged from 1 week to 10 years. Fifteen of the 32 patients were diagnosed in the first year of life (Table 2). Urinary tract infection was the most common presenting symptom in both boys and girls (Table 3). In four children an abnormal prenatal ultrasonogram and subsequent sonogram complemented by EU and VCU within the first 2 months of life demonstrated the anomaly.

EU and VCU were performed in all the children.

In Table 4 the urographic findings are presented. In five children the affected kidney was small with irregular borders and dilated blunted calyces. In three others the entire kidney did not function. Vesicoureteral reflux was demonstrated in all these kidneys giving the appearance of a kidney with reflux nephropathy. A filling defect in the bladder was demonstrated on the urogram in 24 patients, but on VCU in only 14 of the 32 patients.

Reflux was present in 18 patients, always to the lower pole of the ipsilateral kidney and in one case bilaterally (Table 5).

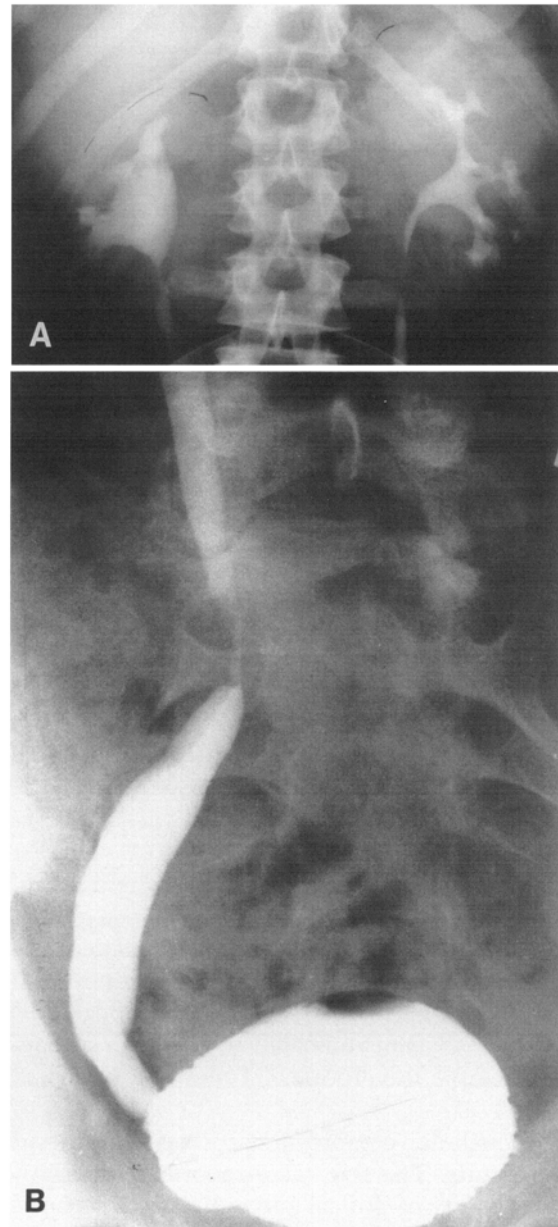
### Discussion

Ectopic ureteroceles have rarely been reported in adults and their diagnosis may be difficult [4, 5]. The clinical presentation differs from that in children, flank pain during voiding being the main complaint reported [5]. Three of our five adult patients suffered from flank and abdominal pain. Hematuria has been mentioned in the literature in one patient [6] and this was present in three adult males in our series, one of whom also had stones in the ureterocele which may have accounted for the hematuria.

Urinary tract infection is the most common finding in children [1, 2] as was corroborated in our pediatric series. The diagnosis can be made on an EU when a radiolucent filling defect is demonstrated in the bladder associated with a duplicated collecting system (cases 4 and 5). The upper pole segment may, however, fail to function. When the upper pole collecting system is dilated but nonvisualized, the lower pole collecting system may show indirect signs of duplication, such as a “drooping lily” appearance [7, 8] as was demonstrated in 23 of the 32 children. The contralateral kidney may be duplicated as well [1] and this was encountered in three of the five adults, but only in four of the 32 children. When the upper pole is small, dysplastic, and nonfunctioning and the lower unit is distorted by reflux, the diagnosis of duplication cannot be established [9] as in our cases 2 and 3. However, when a ureterocele is identified on urography or ultrasonography, a duplex kidney may be suspected [10]. Even when a duplex kidney is visualized the diagnosis may be missed, as the anomaly is so rare in adults. Patient 4 had suffered for 8 years before the diagnosis was established, despite repeat urograms done elsewhere, which on retrospect showed the duplex anomaly with the ectopic ureterocele. Three of the four adult patients, reported by Feldman and Lome [5], suffered for 5–15 years from recurring flank pain and urinary tract infection before the diagnosis was established.

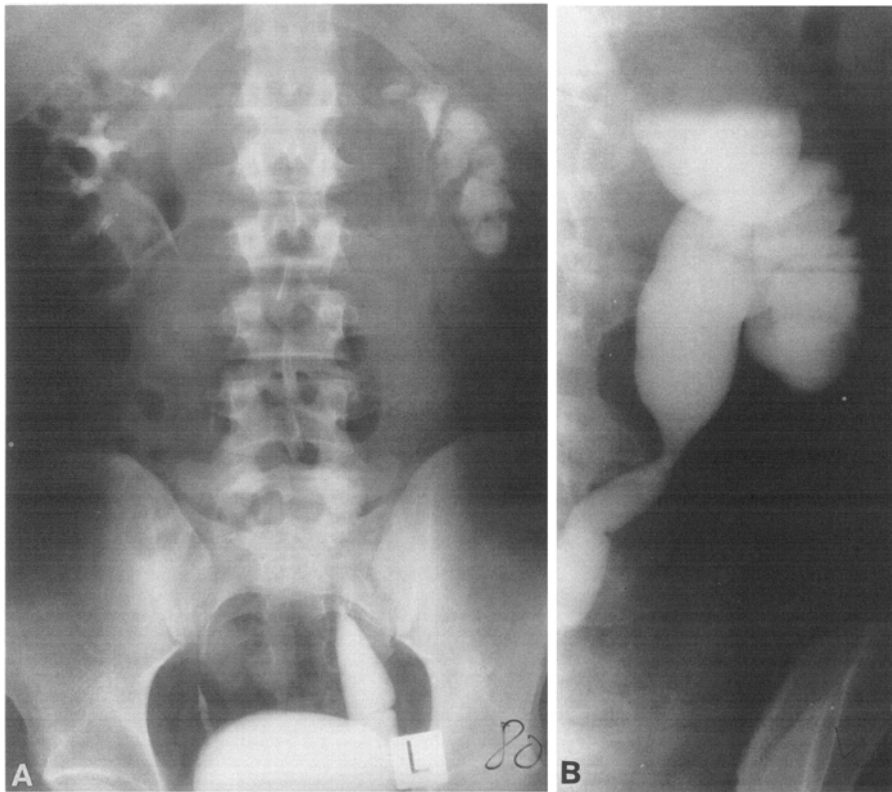
The cystogram may show reflux to the ipsilateral lower pole (two of five adults, 18 of the 32 children). In one adult, reflux was observed from the urethra into the ectopic ureter during voiding (case 5, Fig. 3). This may occur when the ectopic ureterocele has a wide orifice [11].

Nowadays ectopic ureterocele can be diagnosed in many patients by ultrasonography [3, 8, 12]. The



**Fig. 4.** Case 2. **A** Urogram: Hypertrophied left kidney; right small contracted kidney. **B** Cystogram: Marked vesicoureteral reflux into the right collecting system, which proved to be the lower moiety of a duplex kidney.

typical appearance will be that of a “cyst within a cyst” (Fig. 1C) [12]. Only in four children was the diagnosis made on ultrasonography performed in infants in the first few weeks of life as they had a dilated collecting system in utero. The other children in this series were not examined by ultrasonography. It is to be expected that in the future ultrasound will be the main imaging method in detecting the anomaly in utero or perinatally. EU and MCU were added to confirm the diagnosis, to assess



**Fig. 5.** Case 3. **A** Urogram: Double collecting system in right kidney; dilated calyces, left kidney, and dilated left distal ureter. **B** Cystogram demonstrates marked vesicoureteral reflux into a very dilated left collecting system. At surgery this proved to be the lower moiety of a duplex kidney.

the function of the kidney, and to see whether vesicoureteral reflux was present.

CT, performed in one of our adult patients as an initial study, showed bilateral duplex kidneys and a dilated distal left ureter (Fig. 3). Although this study was really not indicated to establish a diagnosis of ectopic ureterocele, CT showed the lesion very clearly.

To conclude, ectopic ureteroceles may occur even in adults. The five cases reported here show the wide variety of clinical and radiological findings in this anomaly. Compared with children with ectopic ureterocele, the adults had a different clinical presentation. The imaging features were, however, similar to those of children.

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