Clinical Case Report

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# Ectopic immature renal tissue in gubernaculum associated with undescended testis of a 1-year-old child

## A case report

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**Abstract**

**Rationale:** Ectopic immature renal tissue (EIRT) is extremely rare in congenital malformations. Moreover, the fundamental pathogenesis of EIRT is still unclear and controversial.

**Patient concerns:** The right scrotum of a 1-year-old man was found empty for a period of 1 month. B-ultrasonography revealed normal bilateral kidneys and a hypoechoic nodule in the right groin.

**Diagnoses:** Based on B-ultrasonography, surgery and pathological examination, we concluded a case of abnormally located and EIRT in the inguinal canal.

**Interventions:** After pathological diagnosis, the patient was not treated with drugs.

**Outcomes:** One year after the operation, the patient recovered.

**Lessons:** EIRT in gubernaculum is extremely rare. Because of the potential risk of malignant transformation, it is necessary to diagnose and treat it early.

**Abbreviations:** CD = cluster differentiation, CgA = chromogranin A, CK = cytokeratin, EIRT = ectopic immature renal tissue, HE = hematoxylin-eosin staining, Ki67 = nuclcar- associated antigen Ki- 67, PLAP = placental alkaline phosphatase, Syn = synuclein, WT-1 = Wilm's tumor protein-1.

**Keywords:** ectopic immature renal tissue, inguinal canal, metanephric tissue, nephroblastoma, teratoma

### 1. Introduction

Ectopic immature renal tissue (EIRT) is a metanephric nephrogenic rest found at an extrarenal site.[1] EIRT is also known as ectopic, heterotopic, or extrarenal nephrogenic rest or extrarenal nephroblastomatosis.[2] EIRT occurs primarily due to the failure of kidneys to occupy the normal position in the waist. EIRT is often associated with other malformations, such as skeletal abnormalities, heart valve insufficiency, undescended testicle, and hypospadias in the urogenital system. EIRT occurs mostly in the pelvic cavity resulting in the formation of pelvic kidney.[3,4] We report a case of 1-year-old man who was diagnosed with EIRT in his inguinal canal. The case illustrates the scope for further research into EIRT including the embryonic and pathological aspects.

### 2. Case report

The right scrotum of a 1-year-old man was found empty for a period of 1 month. He had a healthy medical history and normal prenatal examination. His parents were healthy, without any family history of genetic disorders. A detailed examination upon admission to the hospital revealed an empty right scrotum; the state of native kidney and renal function are all normal; a nonpalpable testicle, and a palpable tumor in the right inguinal region, measuring $1.2 \times 0.5$ cm in size. The tumor was hard and was neither retracted into the abdominal cavity, nor pushed into the scrotum. However, a palpable testis was noticed in the left scrotum, with a fairly developed penis. B-ultrasonography revealed normal bilateral kidneys and a hypoechoic nodule in the right groin with dimensions of $11 \times 5$ mm. The nodule was identified as an undescended testicle on the right side (Fig. 1). A slightly soft right testis, with dimensions of $1 \times 0.5$ cm, and mildly separated from epididymis was found during surgery. The ductus deferens was developed fairly and the gubernaculum testis adhered to aponeurosis of oblique externus abdominis. About 2 cm away from the testis, a solid mass measuring $0.8 \times 0.5$ cm in size was detected on the gubernaculum testis, with an unclear border. Informed content of patient was obtained. This study was content informed and approved by the local Human Research Ethics Committee in Affiliated Hospital of Nantong University.

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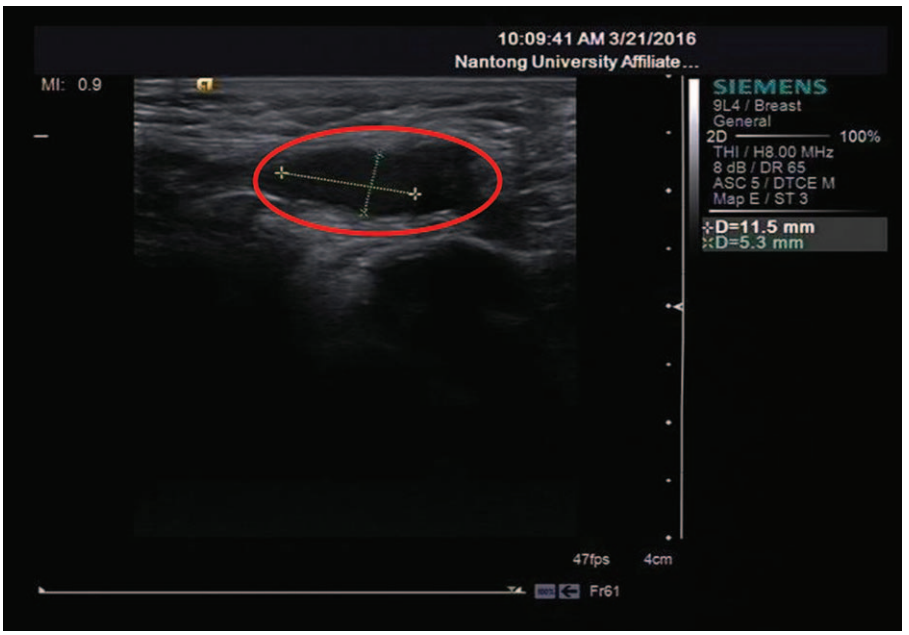


**Figure 1.** B-ultrasonography showed a hypoechoic nodule in the right groin.

Pathological examination confirmed a solid tumor mass of gray–white tissue measuring $2 \times 1.5 \times 1$ cm without a clear coating, and containing a white-to-tan color cut surface with proper hardness. Microscopically, a few small tube-like structures were found in the glomerulus. The size of cells was not pleomorphic and the mitotic count was zero. The cells contained a small psammoma body. No teratomas or blastemas were found (Fig. 2). Immunohistochemical assays showed positive expression of Wilm's tumor protein-1, cytokeratin 7, cluster differentiation 10, paired box gene 8, Vimentin and 2% of nuclear-associated antigen Ki- 67, and tested negative for cytokeratin 20, synuclein, chromogranin A, cluster differentiation 99, cluster differentiation 117, and placental alkaline phosphatase (Fig. 3).

Based on all the test results, we concluded a case of abnormally located and EIRT in the inguinal canal.

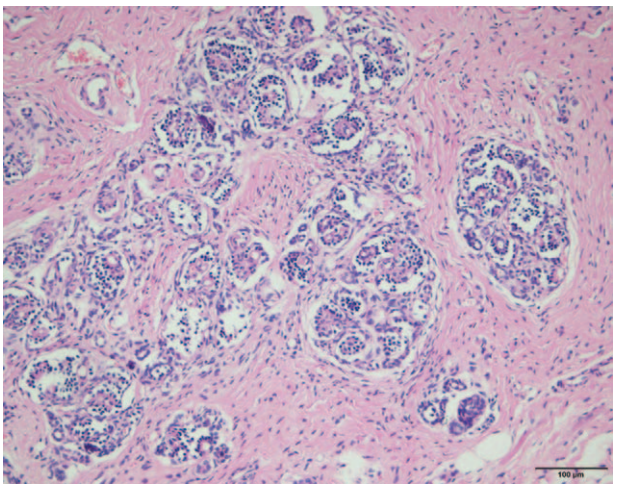

**Figure 2.** This section shows immature glomeruli and uriniferous tubules in the fibrous tissue. (HE ×200).

## 3. Discussion

The kidneys originate in the metanephric tissues of the sacral region.[5] During later embryonic developmental stages, the kidneys usually climb to the waist and develop separately from the abdominal wall. Failure to climb into the waist region results in the development of renal ectopia.[6] Ectopic kidneys are often associated with other malformations, such as skeletal abnormalities, heart valve insufficiency, undescended testicle, and hypospadias in the urogenital system.[7] Normally, during the 7th to 8th month of intrauterine development, the testis is surrounded a double-layered peritoneum, and descends into the scrotum through the inguinal canal. In this case, the patient carried an undescended testicle. We believe that the development of ectopic kidney in the inguinal canal is due to incomplete separation of the mesonephric and paramesonephric ducts from the gonads, resulting in remnants of partially developed kidney tissues in the gubernaculum testis. Such cases are rare. However, ectopic kidney tissues in the undescended testicle are related to nephroblastoma outside the kidney.[8] Nephroblastoma originates in the metanephric blastema. However, the origin of extrarenal nephroblastoma is still controversial. A few studies suggest that renal ectopia may be anterior portion of nephroblastoma.[9] Other studies suggest that extrarenal nephroblastoma is derived from metanephric blastema or residual mesonephros.[10] On the other hand, a few researchers also consider teratoma-containing kidney tissues as originating in extrarenal nephroblastoma.[9]

EIRT might occur in the retroperitoneum, sacrococcyx, and inguinal canal.[11] The origin of residual tissues in ectopic kidneys is not clear. The 3 components including mesonephros, metanephros, and teratoma contribute to EIRT. McDougall et al[12] reported a patient with undescended testicle and ectopic renal tissues in his inguinal canal. He believed that these tissues were derived from mesonephros and not metanephros, without any relationship with the ureter. A similar case was also reported by Shono et al,[8] who proposed that the remnants of mesonephric duct intrude into the inguinal canal, and grow

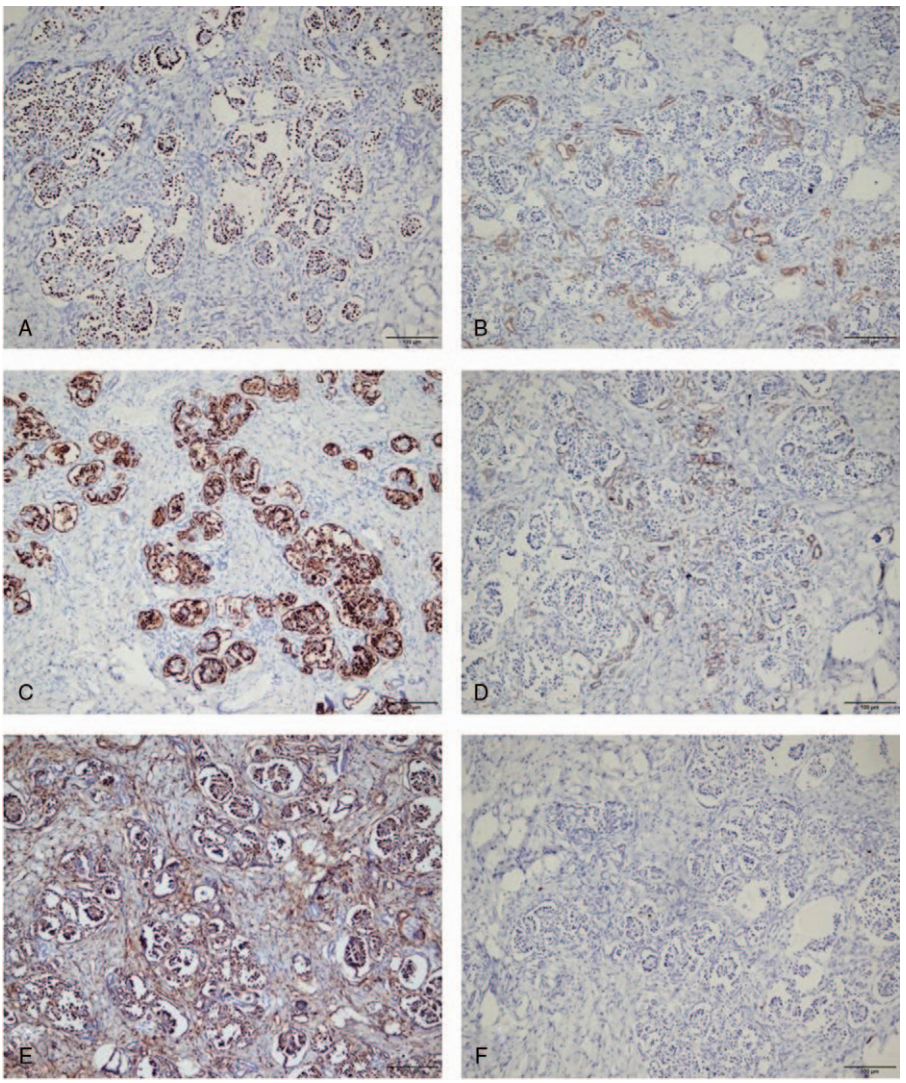


**Figure 3.** (A) Primitive, blastemal cells, and epithelial glomerular elements show nuclear WT1 expression, whereas differentiated tubulesare negative. Original magnification ×200. (B) Immunohistochemistry for CK7 shows cytoplasmic reactivity in tubular structures. Original magnification ×200. (C) Immunohistochemistry for CD10 shows membranal reactivity in tubular structures. Original magnification ×200. (D) Immunohistochemistry for Pax-8 shows nuclear reactivity in tubular structures. Original magnification ×200. (E) Immunohistochemistry for Vimentin shows cytoplasmic reactivity in tubular structures. Original magnification ×200. (F) Cell proliferation index after immunohistochemistry for Ki-67. Original magnification ×200.

along with the testis and spermatic cord. Saito et al[13] had argued that tissues derived from both mesonephros and metanephros were left in the inguinal canal during the growth of testis and spermatic cord. An extensive review of the literature showed occurrence of EIRT both associated and unassociated with a teratoma.[11] Suspicious or obvious features of EIRT malignancy have in fact been reported mainly within teratomas, and treated with additional surgery and chemotherapy. On the contrary, some patients unassociated with teratoma had recurrence. They were still alive without adjuvant chemotherapy.[14]

These rare cases need to be distinguished from other lesions. For example, in metanephric adenoma, benign tumors are generally seen in the young as well as middle-aged women. Microscopically, the metanephric adenoma is composed of fine tubular structures and papillary glands with limited mesenchyme. The overall morphological characteristics suggest that the metanephric adenoma originates in renal tubular epithelial cells. Sertoli trophoblastic tumor and tumor-like lesions are diagnosed by the presence of tubular structures, suggesting a lining of elongated cells resembling Sertoli cells.

In summary, Patients carrying undescended testicles may also contain EIRT in their inguinal canal. Such cases are very rare. However, EIRT developed toward extrarenal nephroblastoma, especially when they contain metanephric tissues and teratomas. These tissues require clear excision, followed by regular monitoring postoperatively.

## References

[1] Dasgupta S, Ghosh R, Das RN, et al. Sacrococcygeal teratoma with ectopic immature renal tissue. Indian J Pathol Microbiol 2015;58:124–5.
[2] Wu Y, Zhu X, Wang X, et al. Extrarenal nephroblastomatosis in children: a report of two cases. BMC Pediatr 2014;14:1–5.
[3] Ritchey M. Belman AB KLR, Kramer SA. Anomalies of the kidney. Clinical Pediatric Urology 2002;Martin Dunitz Ltd, 537–58.
[4] Bauer SB. Wein AJ, Kavoussi LR, Novick AC, et al. Anomalies of the upper urinary tract. Campbell Walsh Urology 9 Saunders Elsevier, Philadelphia:2007;3278–81.

[5] Ubetagoyena AM, Areses TR, Arruebarrena LD. [Renal position and fusion anomalies]. Anales De Pediatría 2011;75:329–33.
[6] Marte A, Marte G, Pintozzi L. Spectrum of ectopic pelvic kidney in children: a ten-year experience. Austin J Urol 2015;2: 1025.
[7] Somajita C, Partha M, Gouri M. Sheares' method of vaginoplasty: our experience. J Cutan Aesthet Surg 2011;4:118–21.
[8] Shono T, Kai H, Suita S. Ectopic renal tissue in the gubernaculum associated with undescended testis. BJU Int 2002;89:320–1320.
[9] Aterman K, Grantmyre E, Gillis DA. Extrarenal Wilms' tumour: a review and case report. Invest Cell Pathol 1979;2:309–1309.
[10] He LL, Wu HY, Tang YZ. [Extrarenal Wilms' tumor: report of a case]. Zhonghua Bing Li Xue Za Zhi 2017;46:347–8.
[11] Coli A, Angrisani B, Chiarello G, et al. Ectopic immature renal tissue: clues for diagnosis and management. Int J Clin Exp Pathol 2012;5:977–81.
[12] Mcdougall EM, Mikhael BR, Carpenter B. Ectopic renal tissue associated with an undescended testis: a case report. J Urol 1986;135:1018–9.
[13] Saito S, Higa I, Koyama Y, et al. Immature renal tissue in inguinal canal. J Urol 1989;142:106–7.
[14] Desphande AV, Hei ER, Kellie S, et al. PS01 ectopic nephrogenic rests in children: the clinico-surgical implications. J Pediatr Surg 2009;44: e13-13e16-16.