

# Nephrogenic Adenoma Presented Nine Years Later After Previous Surgery: A 10 Years-Old Child Case Report

## İlk Cerrahiden 9 Yıl Sonra Gelişen Nefrojenik Adenoma: 10 Yaşında Çocuk Olgu Sunumu

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**ABSTRACT** Nephrogenic adenoma is very rare benign metaplastic lesion of the urinary tract. It is mostly seen in adults but approximately 10% of the nephrogenic adenomas was seen in children. In the literature about 30 cases of nephrogenic adenomas have been reported in children. Its etiology is uncertain, but chronic irritation of urethelial mucosa is related with pathogenesis. Patients generally complain of nonspecific symptoms such as hematuria, dysuria and bladder irritability. In this report we share a case of nephrogenic adenoma which was detected incidentally in a child. To our knowledge, this is the only case in children detected without any symptom or complaint.

**Key Words:** Urinary bladder neoplasms; metaplasia; child

**ÖZET** Nefrojenik adenom, üriner sistemin oldukça nadir benign metaplastik lezyonudur. Çoğunlukla erişkinlerde görülür ama yaklaşık %10 olguda çocuklarda izlenmiştir. Literatürde yaklaşık 30 çocuk nefrojenik adenom olgusu bildirilmiştir. Etyolojisi belli değildir ama üreteliyal mukozanın kronik irritasyonu ile patogeneze ilişkilendirilmiştir. Hastalar genellikle hematüri, dizüri ve mesane irritabilitesi gibi nonspesifik semptomlarla kliniğe başvururlar. Bu yazıda çocukta tesadüfen saptanan nefrojenik adenom olgumuzu paylaşıyoruz. Bildiğimiz kadarı ile herhangi bir semptom veya yakınması olmayan ilk çocuk olgusudur.

**Anahtar Kelimeler:** Mesane tümörleri; metaplazi; çocuk

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**N**ephrogenic adenoma (NA) is an uncommon, benign metaplastic lesion of the urinary tract that more commonly occurs in adults with a male preponderance of 2:1.<sup>1</sup> Although most common in adults, approximately 10% of NA were seen in children and there is a significant predominance of girls compared with boys (5:1) in contrast to adults.<sup>2</sup> This lesion was first described by Davis in 1949.<sup>3</sup> The term “nephrogenic adenoma” was introduced one year later by Friedman and Kuhlenbeck who noted resemblance of this lesion to the renal tubule.<sup>4</sup> Its etiology is uncertain, but it has been linked to chronic irritating factors, such as trauma, urological surgery, renal transplantation, infection, radiation, foreign bodies, kidney stones, chemical agents and prolonged instrumentation of the urinary tract. Associated symptoms are the usual ones encountered in cases of bladder pathology, such as hematuria, urgency, frequency or dysuria mimicking an infection of the lower urinary tract. Diagnosis is based on histopathologic examination. The NA treatment generally consists of transurethral resection and fulguration of the lesion.

In this report we share a case of NA which was detected incidentally in a child. To our knowledge, this is the only case in children detected without any symptom or complaint.

## CASE REPORT

A 10 year-old boy referred to our clinic for mass measuring 15x12 mm which was placed on anterior wall of the bladder, showed by ultrasonography (Figure 1). At one year of age, the patient underwent right ureteral reimplantation for reflux. He had no complaint and bladder mass was detected on routine check incidentally. On physical examination, there was no abnormality. His biochemical examination, complete blood count and urine analysis were completely normal. Cystoscopic examination showed a 2 cm papillary tumoral lesion on the right wall of the bladder, which was resected. Cystoscopical and radiological diagnosis was that of a malignant urothelial tumor, possibly a sarcoma. The pathology report of the transurethral resection was consistent with NA and immunohistochemical workup of the specimen with CK 7 and CA 125 were positive and CK 20 was negative (Figure 2). We preferred for a periodical evaluation cystoscopy combined with ultrasound. At 1 year of follow-up the patient is asymptomatic and there is no sign of recurrence. Written informed consent for publication was given by the patients family.

## DISCUSSION

NA is a rare and benign lesion in pediatric population. Patients usually complain of hematuria, dysuria and frequency. Pathogenesis of the disease is still unknown, however, most authors agree that patients have been exposed to some kind of chronic irritation or inflammation due to some predisposing factors, such as previous surgery (60%), urinary stones (14%), trauma (9%), radiation, urinary catheterization and repeated instrumentation for diagnostic or therapeutic purposes.<sup>5,6</sup> The most frequent predisposing factor in the pediatric patients for the development of this tumor is surgical injury such as ureteric reimplantation. Our case also had a history of uretero-neocystostomy due to reflux nine years ago.

NAs are mostly seen in the urinary bladder 80%; but urethra or ureter can also be involved in 12% and 8% respectively.<sup>7</sup> NA appears as 55% papillary grown pattern, 35% sessile lesions and 10% polypoid. On cystoscopy, the sessile lesions appear friable and velvety, mimicking urothelial carcinoma in situ. About 60% of tumours are 1 cm or less, 30% between 1 and 4 cm, 10% over 4 cm and 20% of lesions are multiple.<sup>8</sup> Diagnosis primarily includes sonography of the urinary tract, cystoscopy, and biopsy. The present case was asymptomatic and diagnosed incidentally by ultrasound.

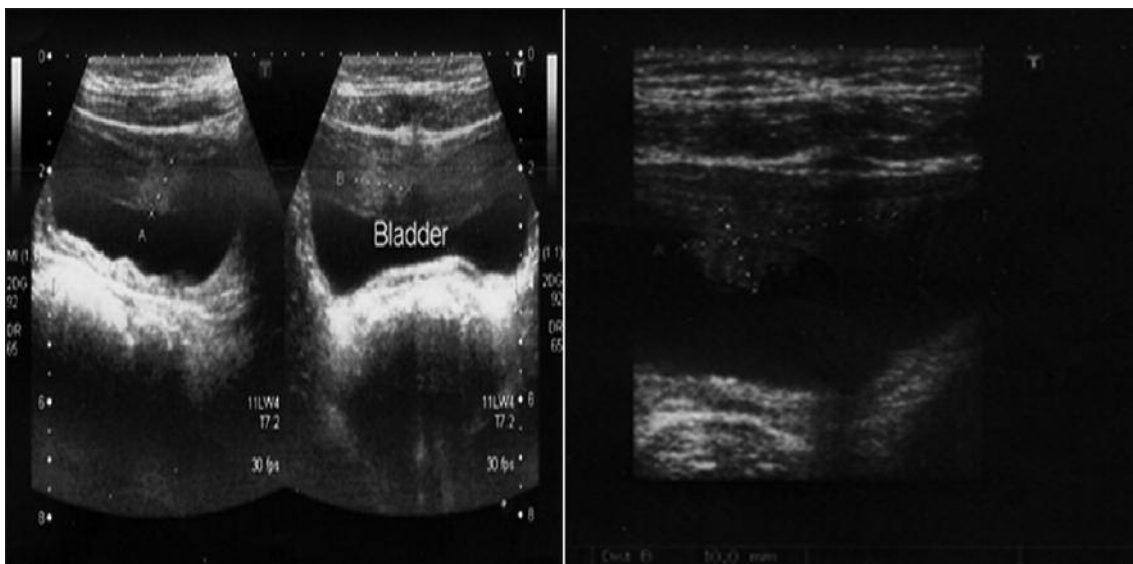
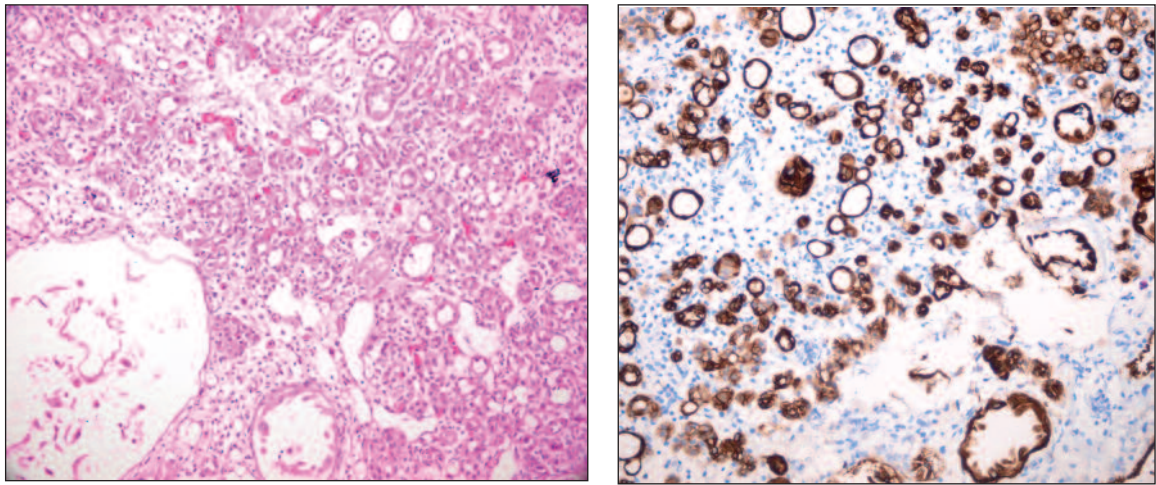


FIGURE 1: Ultrasonography images.



**FIGURE 2:** Pathologic examination of the tissue samples with HE shows that mostly small tubular structure, consisting of occasional dilated cystic tubules and papillary structures with cuboidal and low columnar epithelium lesions. Immunohistochemical studies with CK 7 was showed strong staining in all areas of epithelial lesions (HE, x200).

(See color figure at <http://www.turkiyeklinikleri.com/journal/uroloji-dergisi/1309-632X/>)

It is important to distinguish benign NA from other mimickers that commonly occur in the pediatric population, such as rhabdomyosarcoma, fibroepithelial polyp, papillary/polypoid cystitis, and urothelial neoplasms, in order of frequency.<sup>9</sup> In the present case immunohistochemical examination of the resection revealed positive finding for NA and negative finding for other mimickers.

Treatment is controversial, transurethral resection and fulguration is mostly used for the treatment of NA but also its traumatic effects of mucosa may cause high rate of recurrence.<sup>10</sup> Some authors report the high recurrence rate (37,5% to 75%) of NA during long term follow up of pediatric pa-

tients.<sup>11</sup> Despite the high recurrence rate, there have been no cases of malignant transformation in children reported in the literature.<sup>12</sup> Because of the high probability of NA recurrence, the follow up period has not been exactly established. Our case was followed with 3 months intervals for the first year with urine analysis, ultrasonography and cystoscopy showing no evidence of recurrence.

Overall, although bladder neoplasms are rare in children, NA should be a consideration when a focal lesion is detected in a child with the appropriate previous history, particularly that of bladder surgery. This reports aims at drawing attention to this rare entity in children.

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