

## SHORT REPORT

# Leiomyoma of the Ureter in a Child\* (the Youngest Reported Case) (Case Report)

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Benign and malignant primary tumours of the ureter are uncommon and represent only 1% of the upper urinary tract (1-3).

Benign ureteral tumours are histologically classified for their epithelial or urothelial and mesodermal or stromal origin (1). Benign mesodermal tumours are the less common type of ureter tumours (4). With the exception of a 4-year-old boy, the highest incidence is in the 4<sup>th</sup> and 5<sup>th</sup> decades of life, the average being 44.3 years old (1,4).

Leiomyomas are tumours of smooth muscle origin which are referred to as fibroids. The genitourinary tract, especially the ureter, is an uncommon localisation for them (1,4,5).

A case of leiomyoma of the ureter in a 2-month-old boy is reported because of its rarity and benignity, and also this is one of the youngest cases recorded in the literature.

### Case Report

During a routine control of a 2-month-old boy, relapsing, recurrent urinary tract infection was observed. An excretory urogram (IVP) and ultrasonography revealed grade III right hydronephrosis and ureteropelvic

junction obstruction. The left kidney had a normal appearance and function.

With the presumptive diagnosis of right hydronephrosis secondary to a congenital ureteropelvic junction obstruction, the patient underwent right renal exploration and right pyeloureteroplasty was performed 1 month later. Partial ureterectomy and ureteroneocystostomy were performed because of obstruction of the uretero vesical junction and partial ureteral resection material was sent to the pathology laboratory.

Gross examination showed a pink-grey material having an open lumen, 2 cm long and measuring 1 cm in its greatest diameter and 2 cm at the narrowest localisation.

There was a polypoid mass protruding into the dilated ureteral lumen, which measured 0.5 cm in diameter. Of the same colour as the periphery with a clear border, a firm, stalked round polypoid mass protruded into the dilated ureteral lumen, which measured 0.5 cm in diameter. The sections were stained with Hematoxylin-Eosin (H-E) and evaluated with a light microscopy after routine procedure.

Microscopic examination verified the polypoid mass as a tumour formed by interlacing bundles of smooth muscle

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which progressed to different sites intersecting each other and forming a vortex (Figure 1). No pathological feature was observed in the ureteral wall.

The highly cellular tumour was composed of spindle and uniform cells clasped together firmly, and some of the neoplastic spindle cells had a rather large, rounded or plump elongated nucleus.

The portion of tumour which protruded into the ureteral lumen had a smooth surface covered by either flattened or normal appearing urothelium. Mitoses and necrosis were not observed. Pathologically the tumour was diagnosed as a benign leiomyoma. Positive immunohistochemical staining with both desmin and smooth muscle actin supported the pathological diagnosis (Figure 2).

Primary tumours of the ureter are rare (6). Mesodermal tumours occur more uncommonly than do epithelial tumours (1). Benign tumours of the ureter usually form a polypoid mass protruding into the ureteral lumen (1). The benign mesodermal tumours arising from the wall of the ureter include fibroepithelioma and leiomyoma (1,2,4).

In 144 cases of primary benign ureteral tumours reported by Scott (2,3) only about one-third of these

were tumours of nonepithelial mesodermal origin, and only 3 of them were leiomyoma. Only 8 cases have been reported since 1955 and one of them was in a child (4).

The highest incidence of the benign ureteral tumours is in the 4<sup>th</sup> and 5<sup>th</sup> decades of life.

Primary ureteral tumours occur more frequently in the left ureter and involve the distal third of the ureter in 60 to 75% of cases (3).

Yashi et al. reported that in 5 children the tumours were found in the right ureter, while only 3 involved the left ureter (4). Sex distribution only slightly favours the male subject and the tumours generally form a mass (1,3).

In this case the tumour formed a polypoid mass which connected with a tight peduncula to the wall in the distal region of the right ureter.

Most of the cases with ureteral leiomyoma present with symptoms of ureteral obstruction and  $\alpha$  filling defect of the ureter may help the diagnosis (1,4).

Although the etiology of primary benign ureteral tumours is speculative, infection, chronic irritation, obstruction trauma and carcinogenics have been implicated. Symptoms are non-specific and the classic

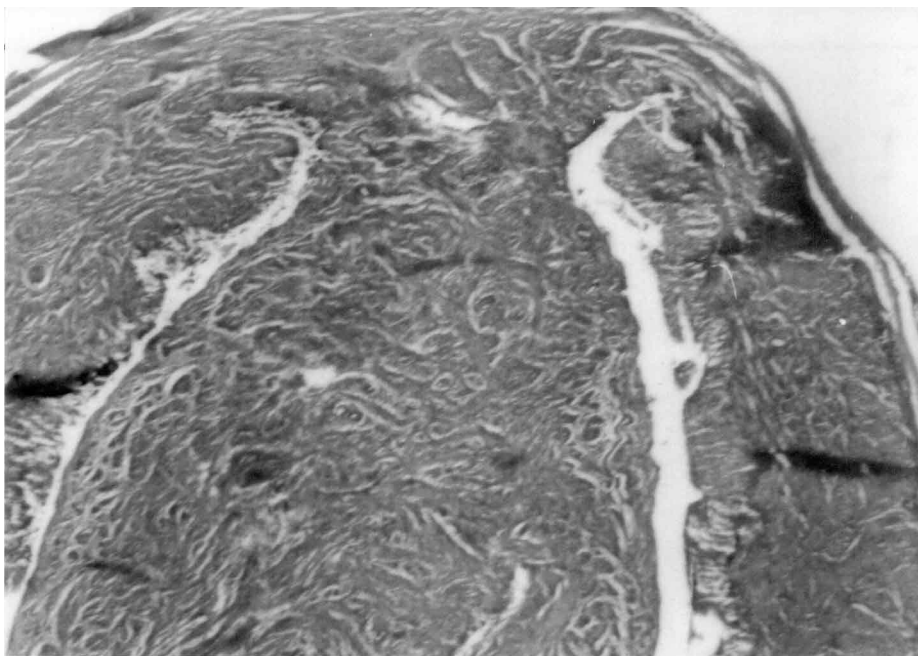


Figure 1. Tumour formed by interlacing bundles of smooth muscle which progressed to different sites and intersected each other and formed a vortex (H-E X 100).

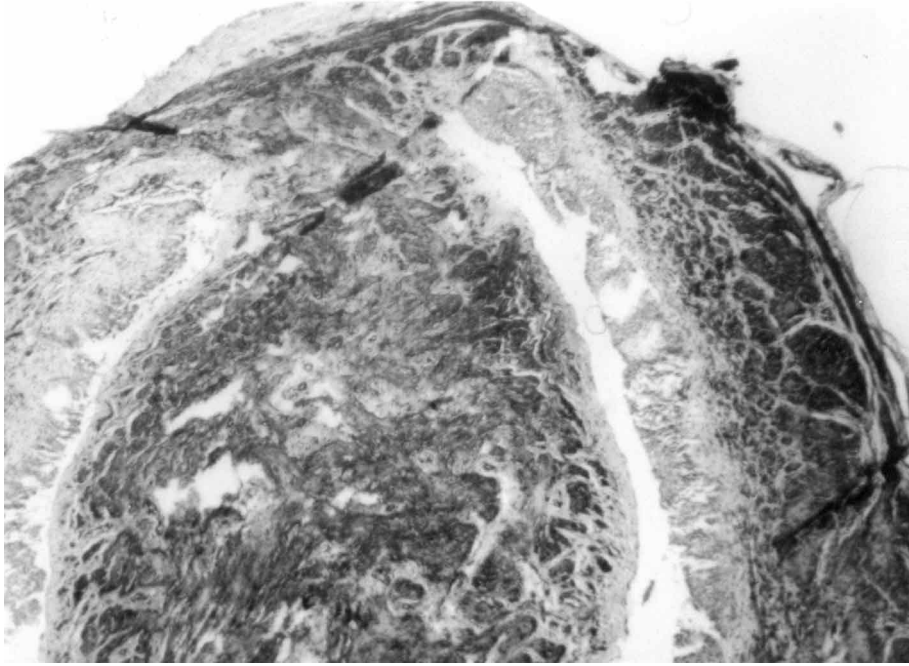


Figure 2. Positive immunohistochemical staining with desmin (Desmin X 100).

triad consisting of hematuria, pain and hydronephrotic mass occurs uncommonly (1).

In this case relapsing recurrent urinary tract infection was observed and neither hematuria nor a mass revealed radiologically or by palpation were determined. Pain could not be evaluated because of age.

The preoperative diagnosis of ureteral tumour is difficult. Retrograde pyelography has been the most successful diagnostic procedure (1). IVP, cystoscopy and selective angiography are also used but a definitive diagnosis can be made by histopathological evaluation. (3).

In this case diagnosis of hydronephrosis secondary to a ureteropelvic junction obstruction was the indication to operaten.

The most common treatment for benign or malignant ureteral tumours had been nephroureterectomy but in recent years, only partial ureterectomy is proposed (1,4).

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