Nephrogenic Adenoma of the Urethra Presenting as Hematuria

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Introduction

Nephrogenic adenoma (NA) is a form of nephrogenic metaplasia with formation of benign epithelial tumours. NA arise from the urothelium as a result of reaction to urothelial injury like trauma, indwelling catheters, previous surgery, renal transplants or calculi. The exact aetiology of this lesion however still remains uncertain [1].

In view of its propensity to cause hematuria, NA needs to be considered and treated especially in the relevant background of previous surgical trauma in the urinary tract. We highlight the presentation and management of this lesion in the following case report.

Case Report

A 13-year-old Asian boy presented with 2 months history of terminal hematuria and occasional dull pain in the right flank. At 3 years of age, he underwent correction of coronal

hypospadias. There was no past history of urinary tract infections, trauma, sexual activity, history of bleeding tendencies or evidence of previous streptococcal infection. Clinical examination did not reveal any abnormality. The anti-streptolysin O titres, full blood count and baseline blood investigations were within the normal range.

An ultrasound of the urinary tract was unremarkable and hence was discharged with a diagnosis of possible posterior urethritis.

However, he came back after 11 months, with the persistence of terminal hematuria and accompanying dysuria. Sonographic evaluation showed a normal study of the urinary tract. In view of recurrent symptomatic hematuria, a diagnostic cystoscopy was performed. An inflamed posterior urethra with a very thin and long lesion (2 × 1 cm) was identified on cystoscopy, which was excised to determine its nature.

Histology revealed the lesion to be a NA (Fig. 1). At 1-year follow-up, he was free of hematuria and dysuria.

Discussion

NA is most frequently observed in the bladder. In the urethra, its occurrence is reported to be 15 % [2]. Most of the data available in the literature are from the adult population and no definite percentage occurrence has been reported in the children.

Patients with NA generally present with non-specific symptoms such as flank pain, hematuria, urgency and dysuria. Diagnosis is established by histopathological examination. Tubular and microcystic patterns are the most common, although papillary structures may also be seen [2].

Interestingly, in our case, the boy presented as late as 10 years after hypospadias repair and with intermittent hematuria. This association of hypospadias repair and late-onset hematuria has not been reported in the literature.

It is widely agreed that children with gross hematuria with normal radiographic studies require cystoscopy when

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Fig. 1 Image showing the histological appearance of the nephrogenic adenoma in the urethra (H & E stain, $\times 40$)

hematuria cannot be attributed to urethritis [3]. In line with this, we undertook cystoscopy in our case which helped to establish both diagnosis and treatment.

Occasionally, NA extends into the underlying fibromuscular stroma, which could lead to misdiagnoses. In adults, this could be mistaken for a prostatic or clear cell adenocarcinoma of the urethra. In children however, the differentials include a rhabdomyosarcoma or transitional cell carcinoma [4]. In addition to the usual multifocal nature of NA, the presence of an overlying urothelium and the typical architectural pattern in areas of the lesion immediately below the urothelial lining are helpful distinguishing features [5].

There is no evidence to suggest that NA is premalignant, despite the similar cytogenetic abnormalities urothelial carcinoma [6]. Cheng et al. followed up patients with atypical or conventional nephrogenic metaplasia. The study showed no further progress to carcinoma [7].

In most cases, transurethral resection of the lesion is the treatment of choice. However, there have been case reports with conservative management [8]. Transurethral resection of the lesion has been recommended as the treatment but with a high recurrence rate of up to 37 % [9]. Cystoscopy for follow-

up is to be used judiciously as the trauma during scopy, by itself, can be a stimulus for recurrence of NA.

Conclusion

In children presenting with hematuria, common causes have to be excluded, with the possibility of NA if no other cause is identified. A previous stimulus like post-hypospadias repair has a strong positive correlation with the occurrence of NA. Cystoscopic excision of urethral NA is recommended with long-term follow-up.

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