

Nephrogenic adenoma : a case report.

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Nephrogenic adenoma is an unusual lesion, occurring in the urothelium-lined organs. The authors report a case of nephrogenic adenoma associated with previous history of surgery and trauma. The clinical features of bladder mass and vesico-vaginal fistula mimicked cancer. This condition should be kept in mind, especially when previous history of some chronic irritative condition or previously injured epithelium has been noted. Although the lesion appears to be benign, close follow-up is recommended.

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*Nephrogenic adenoma เป็นพยาธิสภาพที่พบไม่บ่อยในอวัยวะที่บุด้วยเยื่อบุทางเดินปัสสาวะ ผู้เขียนได้
รายงานผู้ป่วยด้วยพยาธิสภาพนี้ 1 ราย โดยมีประวัติได้รับบาดเจ็บและรับการผ่าตัดในกระเพาะปัสสาวะนำมาก่อน
ก่อนเนื้องอกในกระเพาะปัสสาวะ และภาวะการมีปัสสาวะรั่วทางช่องคลอดจากการที่มีทางติดต่อระหว่างผนังช่องคลอด
กับกระเพาะปัสสาวะทำให้คิดถึงภาวะจากมะเร็งชนิดร้าย พยาธิสภาพนี้ควรคิดถึงไว้ด้วย โดยเฉพาะเมื่อมีประวัติของ
การระคายเคืองเรื้อรัง หรือเยื่อบุทางเดินปัสสาวะได้รับอันตรายมาก่อน แม้ว่าจะมีการดำเนินโรคแบบเนื้องอกไม่ร้ายแรง
ผู้เขียนเห็นว่าน่าจะมีการติดตามผู้ป่วยอย่างใกล้ชิดหลังให้การรักษา*

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Nephrogenic adenoma is an uncommon lesion, which has been reported in the various parts of the urothelium-lined organs⁽¹⁻⁶⁾ as well as in the ileal conduit⁽⁷⁾. Signs and symptoms depend on its location, including hematuria, dysuria, frequency and urgency, suprapubic pain and flank pain⁽⁶⁾. We report herein a case of nephrogenic adenoma with an unfamiliar complaint of urinary incontinence and bladder mass mimicking malignant tumor.

Case Report

A 20-year-old Thai woman had a history of car accident resulting in fracture of the pelvis and bladder injury. Three years after surgical management, she developed urinary incontinence and vesico-vaginal fistula. The further studies revealed a 7.0 cm. nodular mass occupying the trigone of the urinary bladder with involvement of bilateral uretero-vesical junctions. The cystourethroscopy demonstrated marked edema and inflammation of mucosa obscuring both ureteric orifices

Carcinoma of the urinary bladder with vesico-vaginal fistula was suspected and total cystectomy with ileal conduit was performed.

Pathological examination

An isolated shaggy exophytic mass, measuring 8.0×7.0×4.0 cm. was seen occupying the trigonal area. The tumor and adjacent mucosa were congested and edematous. It had a glistening grayish brown cut surface with few discrete foci of hemorrhage.

Microscopically the tumor mass showed papillary and tubular appearance situated in the mucosa and submucosa (Fig. 1&2). The epithelium ranged from cuboidal to columnar, arranged in single layers (Fig. 2).

The papillary stalk was rather broad consisting of edematous lamina propria with prominent vascularity. Numerous inflammatory cells consisting of lymphocytes, plasma cells, scanty eosinophils and neutrophils were noted infiltrating the stroma of intervening tubules. In addition, scattering foci of recent hemorrhage were present. The adjacent submucosa and underlying urinary bladder wall showed decreased amount of muscle fibers with scattering fibrosis. The similar inflammatory reaction was also observed in the aforementioned urinary bladder wall.

Discussion

Over 270 cases of nephrogenic adenoma have been documented from 1949 to 1987⁽⁶⁾. The lesion is commonly related to a previous injury to the epithelium and chronic irritative conditions, including previous surgery, trauma, infection and stones^(1,8). Electron microscopy⁽⁹⁻¹¹⁾ and membrane surface lectin⁽¹²⁾ examinations currently points to metaplastic change as its pathogenesis, particularly of the urothelium. However, a recent case of nephrogenic adenoma, arising in an ileal conduit, was also believed to have been result of metaplastic reaction of the intestinal epithelium⁽⁷⁾. This concept reflects its non-neoplastic nature, unlike adenoma in other organs such as adenoma of the gall bladder. Our case represents a classical one, since it is associated with previous history of surgery and trauma. Thus, the pathogenesis is most likely to be the metaplastic response.

Another major concern is the question of its malignant potential. It should be noted that the vesico-vaginal fistula occurring with urinary bladder mass is often a manifestation of cancer. Furthermore after repeated fulguration, acini of nephrogenic adenoma have been seen among muscle fibers. Moreover, the lesion has been associated in the urinary bladder with concurrent carcinoma in some cases⁽⁶⁾. However its development into frank carcinoma has not been definitely documented.⁽⁶⁾ The bladder wall in our case was weakened, possibly due to inflammation and focal injury with fibrosis of muscle fibers and subsequent fistula formation. Additionally, the lesion was limited to the mucosa without any invasion or atypia. Despite frequent recurrences, no metastasis has been noted after years of observation⁽¹³⁾.

Yet, as an unusual response to injury of the urothelium, particularly in this young patient, close follow-up is recommended. Although cystectomy and augmentation cytoplasty has been used, as in our case, most are well controlled with endoscopic fulguration⁽¹⁴⁾.

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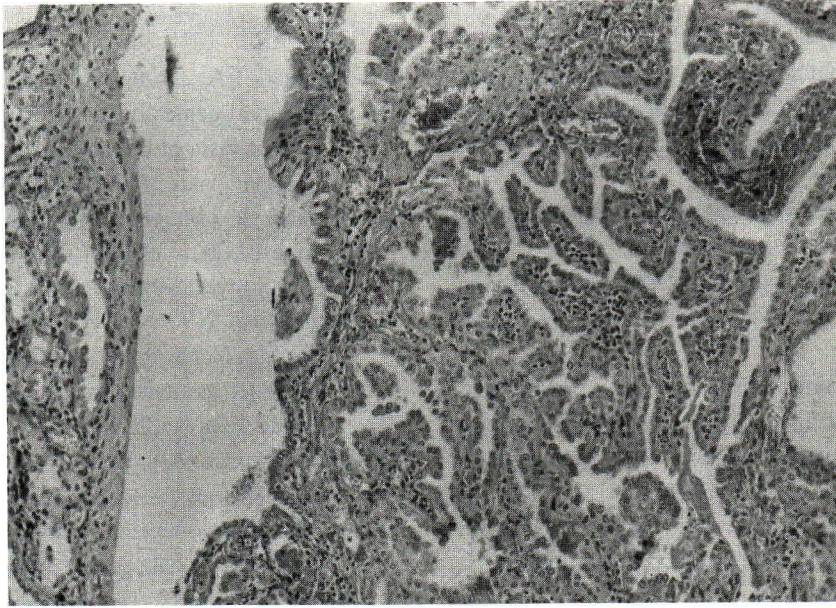


Figure 1 : Illustration showing epithelium lining in papillary and tubular arrangement situated in the mucosa and submucosa (H & E \times 100)

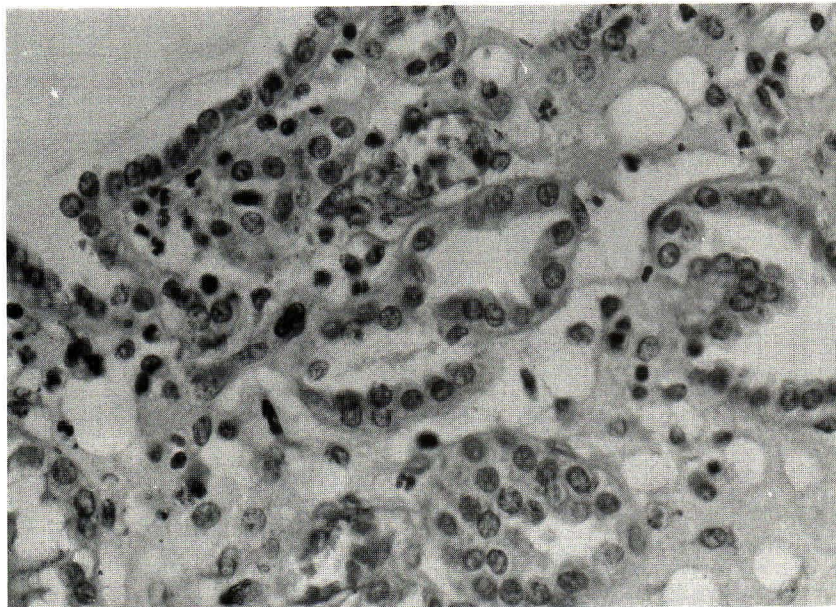


Figure 2 : Higher magnification reveals single layer of cuboidal epithelium and edematous stroma with inflammatory cells infiltrations consisting of lymphocytes, plasma cells and neutrophils (H & E \times 400)

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