TITLE:
Ureteral fibroepithelial polyp associated with urolithiasis induced by steroid therapy in a child: a case report

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RIGHT:
URETERAL FIBROEPITHELIAL POLYP ASSOCIATED WITH UROLITHIASIS INDUCED BY STEROID THERAPY IN A CHILD: A CASE REPORT

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A 14-year-old boy complained of left flank pain. He had been given high-dose corticosteroid therapy for chronic inflammatory demyelinating polyneuropathy (CIDP). Retrograde pyelography revealed irregular defects at the left ureteropelvic junction (UPJ), and ureteroscopy demonstrated ureteral polyp. The polyp was removed and histologically diagnosed as fibroepithelial polyp. Hypercalciuria due to the corticosteroids and bedridden was assumed to have been a causative factor in the stone formation. To our knowledge, this is the first report of a ureteral fibroepithelial polyp in children associated with urolithiasis, and associated with CIDP.

Key words: Ureteral polyp, Steroid, Urolithiasis

INTRODUCTION

The ureteral fibroepithelial polyp is a mesodermal benign tumor. It is often accompanied by urolithiasis in adults, but extremely rare in children. We experienced a case of ureteral fibroepithelial polyp associated with urolithiasis in a child. The urolithiasis was considered to have been induced by the corticosteroid administered for the treatment of a neurological disease, chronic inflammatory demyelinating polyneuropathy (CIDP). In addition, the bedridden state caused by neuropathy also induced stone formation.

To our knowledge, this is the first report of the ureteral fibroepithelial polyp in children associated with urolithiasis, and associated with CIDP.

CASE REPORT

A 14-year-old boy who had intermittent left flank pain and spontaneous discharge of calculus was referred to us in December 1998. He had been prescribed high-dose corticosteroids (a total of 3,000 mg or more prednisolone and a total of 13 g methylprednisolone) for the treatment of a neurological disease, CIDP since May 1998.

Patient's condition: The patient, 167 cm in height and weighing 45 kg, had a moon-face appearance, hypertrichosis and central obesity. He was a bedridden invalid because of leg atrophy caused by CIDP.

Laboratory study: Serum Ca and P were within normal limits. Serum HS-PTH was also normal. Acidosis was not found. Urinalysis revealed 5 to 10 red blood cells/hpf, but no white blood cells or bacteria. Hypercalciuria was revealed (total urinary excretion of 216 mg Ca/day) and the urinary Ca-urinary creatinine ratio (UCa/UCr) after an overnight fast was elevated (0.31).

Intravenous pyelography revealed left hydrenephrosis and tiny multiple stones in the left pelvis. Retrograde urography revealed filling defects that moved by flushing with contrast media (Fig. 1). The cytology was negative for malignant cells in the left kidney urine. Our diagnosis was left ureteral polyp and urolithiasis in the left kidney. On December 8,

Fig. 2. Polyps were revealed by ureteroscopy (P: polyp).

Fig. 3. Pathological diagnosis was fibroepithelial polyp.

the polyps were resected using a ureteroscope under epidural anesthesia.

Operative findings: Observations using ureteroscope revealed multiple polyps at the UPJ, and polyps were removed with forceps (Fig. 2). An 8 Fr double J ureteral stent was placed after the procedure.

The polyps were histologically diagnosed as fibroepithelial polyp (Fig. 3).

Postoperative course: Three weeks postoperatively, the ureteral stent was removed. One week later, ESWL was performed for the left kidney stones. The IVP after ESWL revealed mitigation of hydronephrosis. Stone analysis revealed over 98% calcium phosphate. The patient is still receiving therapy for CIPD.

DISCUSSION

Ureteral fibroepithelial polyp rarely occurs in children. Bolton et al. reviewed 1) 140 cases of this polyp in the English literature, which included 30 cases in children (24 male, 6 female). The polyp is often accompanied by urolithiasis in adults, but seldom in children. Reviewed by Bolton et al., urolithiasis was seen in 10 adult cases of the polyp, but in none of the children. Several cases of the polyp have been reported in children, but no cases were associated with urolithiasis to our knowledge.

What caused ureteral polyp in the present case? We considered that urinary calculi were greatly related to the growth of the polyp, and that CIPD contributed to the formation of calculi as follows. Corticosteroids used in the treatment of CIPD have the adverse effect of hypercalciuria. They cause increased urinary excretion of Ca and inhibition of its absorption in the kidney tubules. According to the literature 2-4), the definition of hypercalciuria in children is urinary Ca excretion of over 4 mg/kg/day, or Uca/Ucr ratio at fasting of 0.21-0.23 or more. In the present case, the amount of Ca excreted was 216 mg/day, and the Uca/Ucr ratio was 0.31, which were both higher than normal and indicated hypercalciuria. Oyabu et al. also reported that the incidence of urolithiasis increases when prednisolone is administered at a dose of over 3,000 mg 5) Thus urolithiasis is considered to be related to CIPD.

Another factor is that the patient had been bedridden for the long period due to weakened muscle strength caused by CIPD. We believe that this situation promoted the release of Ca from the bone, and resulted in urolithiasis. The stone analysis in the present case revealed over 98% calcium phosphate. Oyabu et al. reported that most stones induced by corticosteroids were composed of calcium phosphate. This is in agreement with the present case.

To our knowledge, no cases of ureteral polyp in children associated with CIPD have been reported. Therefore, this is the first report of ureteral fibroepithelial polyp associated with urolithiasis, and also is the first report of a case of ureteral fibroepithelial polyp associated with CIPD. In the present case, the administration of steroids is necessary due to the CIPD, and the patient should be followed up closely for recurrence of stones.

CIPD is an autoimmune disease and the demyelinating neuropathy in itself, and it may have been a coincidental condition in this case. The concept of CIPD is new, and it has not been reported as a complication of urinary tract disease previously. However, in the urinary tract, major problems occur due to large doses of corticosteroids in the treatment and bedridden state caused by neuropathy. We emphasize that the physician should pay attention to the possibility of urinary tract complications in the patient administered large doses of corticosteroids or in a bedridden state such as a CIPD patient.

REFERENCES


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和文抄録

ステロイド結石症に合併した小児尿管ポリープの1例

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尿管ポリープは, 小児・若年者においては先天的な要因が多く, 成人例では2次的変化に起因することが多い。われわれは, 慢性炎症性脱髄性多発性神経障害 (Chronic inflammatory demyelinating poly-neuropathy, 以下 CIDP とする) に合併した, 尿路結石症合併小児尿管ポリープの1例を経験した。ポリープは小児例ではあるが多発する結石による炎症性変化と考えた。また結石形成には著名な高カルシウム尿症が関与し, その背景には CIDP 治療のための大量ステロイド投与と, 神経障害による長期臥床という独特な状況が大きく関与していた。小児尿管ポリープに合併したステロイド尿路結石症は過去に報告がなく, またCIDP による尿路合併症の報告としても1例目であった。

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