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Xanthogranulomatous pyelonephritis infiltrating the liver

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Abstract We report for the first time a patient with a caliceal diverticulum that was detected in early childhood, who in adolescence developed xanthogranulomatous pyelonephritis to the diverticulum and surrounding kidney and infiltrated to the liver. The condition was treated by nonradical organ-sparing surgery and prolonged antibiotic therapy.

Xanthogranulomatous pyelonephritis (XGP) is a rare and severe, chronic variant of pyelonephritis characterized by destruction of the renal parenchyma. Renal parenchyma is replaced with a chronic inflammatory infiltrate and lipid-laden macrophages causing poor renal function or nonfunctional enlarged tumorlike kidney. Xanthogranulomatous pyelonephritis is associated with chronic urinary obstruction and urinary tract infection. Usually nephrectomy is required for the treatment [1], but in focal cases, partial nephrectomy has been successful [2,3]. Because of infiltrative behavior of XPG, resection of affected tissue such as intestine is often required to manage the condition [2].

Caliceal diverticula are rare transitional epithelium-lined cavities within renal parenchyma and they usually communicate with collecting system through a narrow infundibulum [4].

Here we report a patient who had XGP in conjunction with caliceal diverticulum. The process infiltrated the liver. The condition was treated by nonradical organ-sparing surgery and prolonged antibiotic therapy. Cases of XGP that infiltrated liver have not been published previously.

1. Case report

The patient had urinary ultrasonography at the age of 10 years because of prolonged nighttime enuresis. A cystic lesion with a diameter of 1 cm was detected in the upper pole of the right kidney. Intravenous pyelography revealed caliceal diverticulum (Fig. 1). No follow-up was considered necessary.

At the age of 15 years, the patient was admitted because of intermittent abdominal and right flank pain, diarrhea, and fever during the last 3 months. Physical examination was unremarkable. Laboratory examinations revealed normal white blood cell count and sedimentation rate. Urine was clean. The patient did not have a history of symptomatic urinary tract infections. Abdominal ultrasound demonstrated a mass of 10 × 9 × 6 cm involving upper pole of the right kidney and the right lobe of the liver. The mass involved a cyst with a diameter of 5 cm in the upper pole of the kidney. A magnetic resonance imaging (MRI) demonstrated a tumor sized 6 × 6 × 3 cm between the upper pole of the right kidney and the posterior right liver lobe with some infiltration to both organs (Fig. 2).

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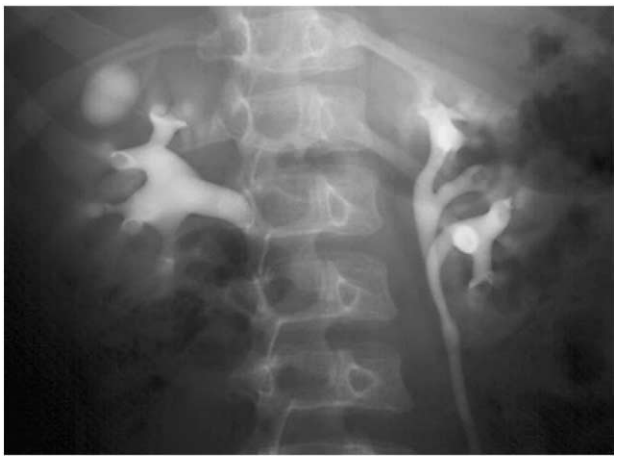

Fig. 1 Caliceal diverticulum in the upper pole of the right kidney.

At laparotomy, a retroperitoneal mass infiltrating to the upper pole of the right kidney and the right lobe of the liver was found. Frozen section biopsy specimens revealed XGP. The tumor was excised nonradically from the liver together with the upper pole of the kidney. Some marginal XGP tissue was left in the liver because more extensive liver resection was considered risky and unnecessary in this kind of infectious condition. No stones were detected.

Bacterial culture from the tumor showed growth of *Escherichia coli,* which was sensitive for all tested antibiotics. The postoperative management consisted of a 1 week course of intravenous antibiotics (cefuroxime, 1500 mg 3 times a day, and clindamycin, 600 mg 4 times a day) followed by oral kefalexin, 500 mg 3 times a day for 3 weeks. After that, the patient continued oral trimetophrin, 100 mg twice a day for 3 months.

The first follow-up ultrasonography 4 weeks postoperatively revealed a 58 × 19-mm echogenic mass (apparently postoperative hematoma) in the operative area. In dimercapto-succinic acid scintigraphy, the function distribution was 43% on the right and 57% on the left. Magnetic resonance imaging 3 months postoperatively demonstrated still abnormal heterogeneous mass (21 × 20 mm) at the operative area. In the last MRI control, 15 months postoperatively, no residual disease was detected, but the upper pole of the right kidney was somewhat deformed.

2. Discussion

Caliceal diverticula are rare incidental findings with the incidence of 3.3/1000 [4]. About one third of the caliceal diverticula are symptomatic, mostly associated with calculi [4]. The natural history of caliceal diverticula is not known; however, no follow-up program has been considered necessary [4]. Similarly, in our patient, no follow-up was organized after detection of an incidental caliceal diverticulum.

Xanthogranulomatous pyelonephritis is a rare complicated form of pyelonephritis. In the last 10 years, 114 pediatric cases have been documented in the English and European literature [1]. Usually, the disease involves the whole kidney, but in 8%, XGP has been reported to be focal [5]. Synchronous obstruction of the ipsilateral renal unit with concomitant urinary tract infection is the most common etiology [5]. The obstruction is most commonly secondary to underlying stone disease of the renal pelvis or ureter [1]. Xanthogranulomatous pyelonephritis has been reported also in association with caliceal diverticula in children [6].

The final diagnosis of XGP is usually based on histology, usually after nephrectomy. During the last decade; however, increasingly sensitive radiologic investigations (computed tomography, MRI) in addition to clinical suspicion have made preoperative diagnosis of XGP possible [1]. However, in many cases the indication for the operative treatment is a suspicion of a neoplasm [5] such as in our patient.

Although it is well known that XGP can invade the surrounding tissues, we found only one case wherein the disease was reported to be in contact with the liver [7]. In this case, no infiltration to the liver parenchyma was detected unlike in our case. Usually, radical surgery with nephrectomy or partial nephrectomy is regarded necessary to treat XGP [2,3]. However, nonsurgical management of focal XGP has

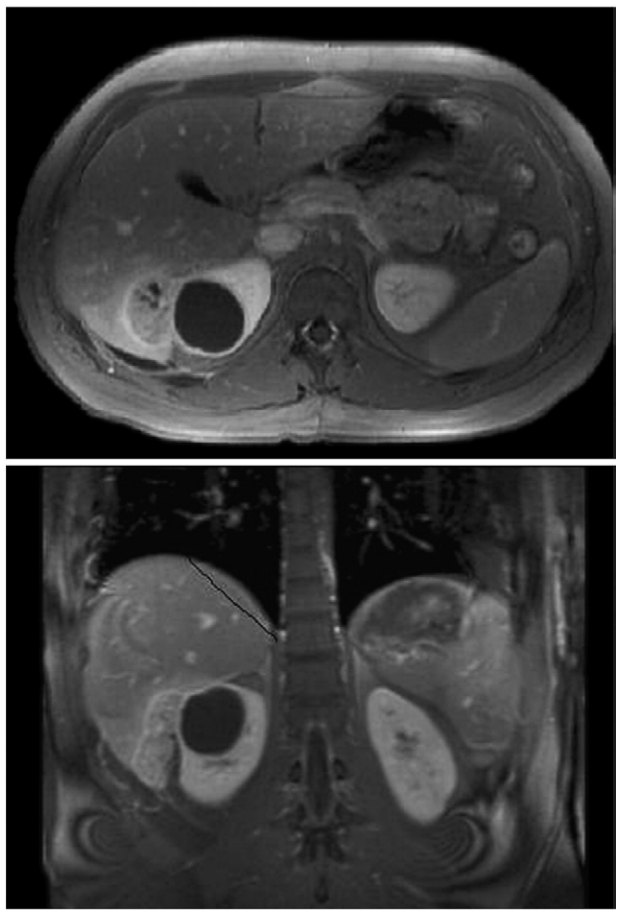

Fig. 2 Xanthogranulomatous pyelonephritis in the upper pole of right kidney and infiltrating the liver.

been described [8]. In our patient, partial nephrectomy including all diseased tissue was performed. The resection of XGP tissue from the liver lobe was, however, not radical. Despite this, prolonged antibiotic treatment successfully cured the condition.

Caliceal diverticulum complicated with XGP is an extremely rare incident. Resection of all diseased tissue is recommended in cases of XGP; however, in some cases, less radical surgery in combination with prolonged antibiotic therapy apparently cure the condition.

References

[1] Zugor V, Schott GE, Labanaris AP. Xanthogranulomatous pyelonephritis in childhood: a critical analysis of 10 cases and of the literature. Urology 2007;70:157-60.

[2] Quinn FM, Dick AC, Corbally MT, et al. Xanthogranulomatous pyelonephritis in childhood. Arch Dis Child 1999;81:483-6.

[3] Chang JW, Chen SJ, Chin TW, et al. Xanthogranulomatous pyelonephritis treated by partial nephrectomy. Pediatr Nephrol 2004; 19:1164-7.

[4] Monga M, Smith R, Ferral H, et al. Percutaneous ablation of caliceal diverticulum: long-term followup. J Urol 2000;163: 28-32.

[5] Samuel M, Duffy P, Capps S, et al. Xanthogranulomatous pyelonephritis in childhood. J Pediatr Surg 2001;36:598-601.

[6] DeMarco RT, Cain MP, Davis MD. Xanthogranulomatous pyelonephritis associated with a congenital caliceal diverticulum. Urology 2001;57:168III-168V.

[7] Esposito G, Savanelli A, de Luca U, et al. Xanthogranulomatous pyelonephritis in childhood: report on an unusual case. Z Kinderchir 1987;42:120-2.

[8] Brown PS, Dodson M, Weinturb PS. Xanthogranulomatous pyelonephritis: report of nonsurgical management of a case and review of the literature. Clin Infect Dis 1996;22:308-14.