Case Report

Contrast-Enhanced CT-guided Biopsy for IgG4-Related Disease of the Ureter: a Case Report

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Abstract

The patient was a 44-year-old man with gross hematuria and left lumbago. Computed tomography (CT) showed thickening of the left ureteral wall. Serum immunoglobulin (Ig)G4 level was high (348 mg/dl). To definitively diagnose IgG4-related disease, we initially planned CT-guided biopsy of the ureter. However, biopsy seemed to pose a risk of ureteral perforation. Iodinated contrast medium was therefore administered during the procedure. While contrast agent was discharged into the ureter, the biopsy needle was carefully advanced under CT fluoroscopy in a direction to avoid the visualized ureteral lumen. Tissue was successfully collected from the ureteral wall without causing any complications and a pathological diagnosis was reached. CT-guided biopsy was thus effective for IgG4-related disease of the ureter in this case.

Key words: IgG4-related disease, ureter, CT-guided biopsy

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Introduction

In 2001, Hamano et al. reported autoimmune pancreatitis with high serum concentrations of immunoglobulin (Ig)G4 [1]. Diseases in which swelling or thickening of various other organs occurred with high serum IgG4 concentrations have since been reported [1-4]. Consolidation of these diseases into the entity “IgG4-related disease” was proposed, and diagnostic criteria were established in 2011. Definitive diagnosis requires histopathological evidence [5, 6]. Three criteria must be satisfied for a definitive diagnosis [5, 6]: clinical examination showing characteristic diffuse/localized swelling or masses in single or multiple organs; hematological examination showing elevated serum IgG4 concentrations (≥135 mg/dL); and histopathological examination showing both marked lymphocyte and plasmacyte infiltration and fibrosis and infiltration of IgG4-positive plasma cells (ratio of IgG4/IgG-positive cells >40%; >10 IgG4-positive plasma cells/high-power field) [AE1]. IgG4-related disease of the ureter is a rare disease, with only 13 cases reported to date [7-10].

We report the case of a patient from whom tissue was collected by contrast-enhanced computed tomography (CT)-guided biopsy for suspected IgG4-related disease of the ureter, and a definitive diagnosis was made.

Case

A 44-year-old man was referred to our hospital with a history of gross hematuria and left lumbago. The symptoms had disappeared, and he showed no abnormal findings on physical examinations or urinalysis on admission. [EIC1] Blood testing showed: white blood cell count, 9.9 × 10³/μl; C-reactive protein level, 0.20 ng/ml; IgG, 1875 mg/dl; and IgG4, 348 mg/dl. CT (Fig. 1) showed thickening of the left ureteral wall with a maximum outer diameter of 2 cm and mild dilatation of the renal pelvis (grade 1 hydronephrosis).
First, the biopsy needle was advanced up to just before the thickened ureteral wall and high serum IgG4 concentration (≥135 mg/dl) were met, but the third criterion from pathological examination needed to be met for definitive diagnosis [5, 6].

CT-guided biopsy was performed with the aim of achieving histopathological diagnosis. This procedure was performed under hydroxyzine sedation and local anesthesia using lidocaine. The patient was placed in a prone position, and CT fluoroscopy (Somatom Sensation 16 ICT; Siemens Medical Solutions, Erlangen, Germany) was used to guide the puncture. A semiautomatic 18-G spring-loaded cutting needle (FineCore; Toray Medical, Chiba, Japan) was used. First, the biopsy needle was advanced up to just before the thickened ureter (Fig. 2a). Next, with the purpose of identifying the ureter lumen, iodinated water-soluble contrast medium (iopamidol, Oyalomin®370 Injection; Fuji Pharma, Tokyo, Japan) was injected into an upper extremity vein at a rate of 3 ml/s (total dose, 100 ml), and CT was performed 5 min after the injection. Contrast agent was discharged into the ureter, and during the time the ureter lumen was identi-
Histopathological diagnosis is needed in the definitive diagnosis of IgG4-related disease. Specimens for pathological testing of ureter lesions are generally collected from urine cytology and ureteroscopy. Nearly all ureter lesions occur in the epithelium, and CT-guided biopsy is rarely used. Incidently, as seen with the present patient, ureteral wall findings were normal in observations with a ureteroscope in past reports of IgG4-related disease of the ureter, and transurethral collection of pathological tissue was not considered feasible ([8, 9]). In each of the 13 reported cases of this disease, histological diagnosis was made from surgically resected ureters. The reasons for these resections were that a ureteral tumor was diagnosed or could not be excluded [7-10]. [AE3]

CT guided biopsy is feasible when the thickened wall form a mass with around 2 cm in diameter [9], and it would be useful for pathological diagnosis of IgG4-related disease of the ureter without surgical resection. [EIC3] [EIC4] CT guided biopsy has a risk of injury of ureter. We performed CT guided biopsy under intravenous injection of contrast medium which can identify the ureter lumen, and therefore, a pathological specimen from the thickened wall could be obtained while avoiding the ureter injury. [EIC5]

Despite of the use of contrast-enhanced CT during biopsy, there is a potential risk of injury of ureter and/or adjacent organs especially for cases of anomalous course or types of ureters. Therefore, the course and type of ureter and adjacent vessels should be carefully evaluated before the biopsy procedure, and placement of a ureteral catheter is required when the ureter injury occurs. [EIC6]

In conclusion, we have reported a case in which contrast-enhanced CT-guided needle biopsy was useful in sampling the tissue needed for a definitive diagnosis of IgG4-related disease of the ureter.

Conflict of interest: The authors declare that they have no conflicts of interest to report.
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