RETROPERITONEOSCOPIC NEPHRECTOMY AS A SECOND-LINE TREATMENT AFTER TRANSARTERIAL EMBOLIZATION FOR SYMPTOMATIC AUTOSOMAL DOMINANT POLYCYSTIC KIDNEY DISEASE

Shusuke Akamatsu², Hidefumi Kinoshita³, Yosuke Shimizu¹, Koji Yoshimura¹, Noriyuki Ito¹, Toshiyuki Kamoto¹ and Osamu Ogawa¹

¹The Department of Urology, Kyoto University Graduate School of Medicine

²The Department of Urology, Nishi-Kobe Medical Center

³The Department of Urology and Andrology, Kansai Medical University

Patients with autosomal dominant polycystic kidney disease (ADPKD) often suffer from abdominal symptoms. Although laparoscopic nephrectomy has been reported as a minimally invasive therapy, it is still technically demanding due to the large size of the kidneys. Transarterial embolization (TAE) is one of the alternatives, but there are only limited reports on its application in ADPKD. We describe a case in which bilateral nephrectomy was performed as a second-line treatment after TAE. One kidney was removed because a small feeding arterial branch was not completely embolized. The other kidney was removed due to infection. Retroperitoneoscopic nephrectomy was a good choice as a second-line modality in the case without infection because the volume of the kidney was reduced even with incomplete TAE, and adhesion after TAE was minimal. TAE is an effective choice in ADPKD patients without infection as a first-line treatment even when complete embolization is difficult, since nephrectomy after TAE is technically easier than removal of a fresh ADPKD kidney.

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Key words: Autosomal dominant polycystic kidney disease, Transarterial embolization, Retroperitoneoscopic nephrectomy

INTRODUCTION

Transarterial embolization (TAE) is reported to be a sufficiently effective alternative modality for treatment of autosomal dominant polycystic kidney disease (ADPKD) with abdominal symptoms in anuric patients. However, its indication has not been clarified. We report the case of a patient who ultimately necessitated bilateral nephrectomy as a second-line therapy.

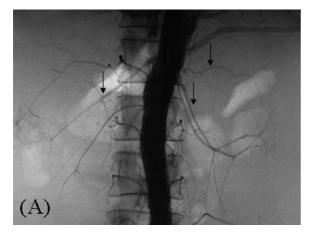
CASE REPORT

A 66-year-old woman was referred to our hospital in September 2001 with symptoms of abdominal fullness and recurrent infection. She had been diagnosed with ADPKD in 1980, and had been on hemodialysis since 1997. Her kidneys continued to enlarge progressively, and she had suffered from abdominal symptoms such as constipation and appetite loss, and frequent infection. After informed consent, TAE of bilateral renal arteries with coil and gelatin sponge was performed. In the left kidney there were two renal arterial branches each feeding the upper and lower poles. The upper branch was slightly too narrow and kinked for a coil to be placed, and it was embolized with a gelatin sponge only (Fig. 1). The procedure was performed without any complications except mild flank pain lasting for a few

days.

Computed tomography (CT) six months later showed that the size of her right kidney had shrunk from 13×13 cm to 6×8 cm in diameter. On the other hand, the size of her left kidney had only shrunk from 14×12 cm to 11×9 cm at the upper pole, which was far less than the change seen with her right kidney.

She was free of any symptoms for two years after TAE, but gross hematuria and left back pain recurred in April 2003. There was no sign of infection. At that time, CT demonstrated that the cysts in the right kidney had almost disappeared. However, those of the left kidney still remained, although the size of the kidney had reduced by half (Fig. 2). Medical management failed and retroperitoneoscopic left nephrectomy performed as a second-line therapy. The patient was placed in the decubitus position and 4 trocars were placed (Fig. 3). Despite the presence of the renal cysts, retroperitoneum could be sufficiently dilated using a PDB balloon (Origin Medsystems, Menlo Park, Calif), and pneumoretroperitoneum was created. As with the standard retroperitoneoscopic nephrectomy procedure, renal vessels were easily identified and divided. The presence of the renal cysts did not interfere with the approach to the renal vessels. Then, the kidney was dissected from the peritoneum anteriorly. Cyst walls



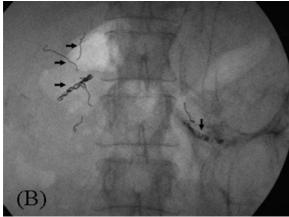
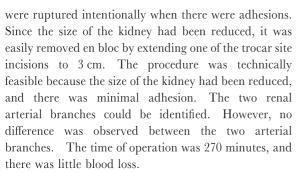
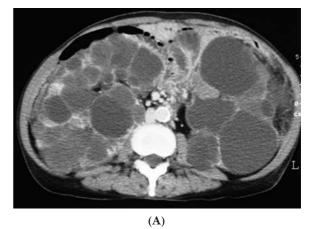


Fig. 1. (A) Aortography before embolization. Arrows point to the renal arteries. (B) Aortography after embolization. Arrowheads indicate the coils used. Notice that the artery feeding the upper pole of the left kidney is not embolized by a coil.



One year later, she had recurrent infection to the right renal cysts. Retroperitoneoscopic right nephrectomy was performed. The procedure was the same as that for the left nephrectomy. This time, although renal arteries and veins were easily identified and divided, the kidney was strongly adherent to the surrounding tissue and it was technically more difficult to remove than the left kidney. Adhesion was especially strong between the cysts and the peritoneum. The time of operation was 315 minutes, and there was little blood loss. The patient has been free of any symptoms since the nephrectomy.



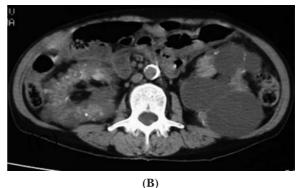


Fig. 2. (A) CT images before embolization shows markedly enlarged bilateral kidneys with multiple cysts. (B) At 20 months after embolization, the size of both kidneys has decreased. However, renal cysts with moderate size still remain in the left kidney.

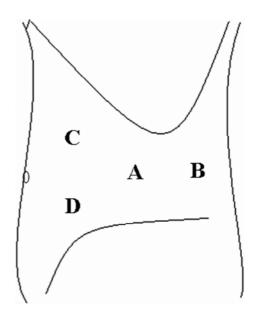


Fig. 3. Trocar placement in retroperitoneoscopic nephrectomy. A, 15 mm camera port; B, 12 mm working port; C and D, 5 mm working ports. Port D was extended to 3 cm when removing the kidney.

DISCUSSION

In a substantial number of cases of ADPKD, patients suffer from such symptoms as recurrent infection of the cysts, hemorrhage, constipation, anorexia, and ileus due to progressive enlargement of the kidneys¹⁾. In these cases, intervention is necessary and nephrectomy with or without renal transplantation is the mainstay of treatment. Recently, many cases of bilateral nephrectomy under laparoscopic technique have been reported. This may be one of the standard modalities as a minimally invasive therapy, but it is sometimes technically demanding²⁾. Laparoscopic marsupialization of renal cysts has been reported as a feasible alternative, especially in patients with preserved renal function. However, the success rate is about 60-70%, and its indication is yet to be determined^{3,4)}. TAE is also recently reported to be a less-invasive and equally effective alternative in anuric patients. Ubara et al. performed TAE on 64 patients with markedly distended abdomen or hematuria due to ADPKD. In all cases, the size of the kidneys had decreased by 12 months after TAE to a median of 53.4%, and no major complications were observed $^{5)}$.

In our case, compressive symptoms from the right renal cysts could be controlled by TAE. The compressive symptoms and hematuria from the left renal cysts recurred. The effect of the gelatin sponge usually lasts for only one to two months. It is suspected that the temporal embolization of the left renal artery feeding the upper pole with a gelatin sponge alone was not sufficient and that the artery re-canalized. We need to emphasize the importance of achieving complete embolization of all small branches with coils to obtain the maximum therapeutic effect. However, even if complete embolization is difficult as in our case, TAE still remains to be the choice in patients who have compressive symptoms since retroperitoneoscopic nephrectomy after TAE is technically easier than removal of a fresh ADPKD kidney due to decreased size.

TAE has been performed in other renal disorders such as angiomyolipoma, renal cell carcinoma and arteriovenous fistula. However, reports on nephrectomy after TAE are rare, and the extent of adhesion after TAE is not understood. In our case, left nephrectomy was

performed two years after TAE and there was no adhesion around the kidney. We can assume that without infection, adhesion never occurs after TAE or even if adhesion occurs, it improves with time. The ideal timing of nephrectomy after TAE is still unknown.

Infection to the right renal cysts could not be controlled by TAE. There are usually two pathways to infection of renal cysts. One is systemic infection via blood and the other is retrograde infection from the urinary tract. It is speculated that TAE is not effective against preventing retrograde infection. TAE is not indicated in patients with infective symptoms. Retroperitoneoscopic nephrectomy is technically more demanding in cases with infection due to adhesion, and the merit of size reduction by TAE is minimal.

We conclude that TAE is a good alternative to the ADPKD patients without infection and that retroperitoneoscopic nephrectomy is a good choice as a second line therapy. Effort should be made to embolize all the arterial branches completely to obtain the maximum therapeutic effect in TAE.

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和文抄録

症候性の常染色体優性嚢胞腎に対して経カテーテル的動脈塞栓療法を 行った後に Second-Line 治療として後腹膜鏡下腎摘術を行った 1 例

赤松 秀輔², 木下 秀文³, 清水 洋祐¹, 吉村 耕治¹ 伊藤 哲之¹, 賀本 敏行¹, 小川 修¹

¹京都大学大学院医学研究科泌尿器科分野,²西神戸医療センター泌尿器科 ³関西医科大学泌尿器科学教室

常染色体優性嚢胞腎(ADPKD)の患者では腹部症状を認めることが多い.腹腔鏡下腎摘術は低侵襲治療として報告されているが,腎が大きいため高度の技術を要する.経カテーテル的動脈塞栓術(TAE)は有効な選択肢の1つであるが,ADPKDに対する報告は限られている.われわれは,ADPKDに対し TAE を行った後に Second-Line 治療として両側の後腹膜鏡下腎摘術を行った1例を報告する.一方の腎は細い栄養血管が完全に塞栓されていなかったために腎摘術を要

した. もう一方の腎は感染のコントロールができず腎 摘術を要した. TAE 後の後腹膜鏡下腎摘術は, 感染を伴わない腎では癒着も少なく, たとえ不完全なTAE であっても腎が小さくなっているために容易であり Second-Line 治療として有効であった. また TAE は, 感染を伴わない症例ではたとえ完全な塞栓が難しくても後の腎摘術が容易になるため, First Line 治療として有効な選択肢であると考えられた.

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