Surgical Revascularization for Pediatric Renovascular Hypertension Caused by Fibromuscular Dysplasia

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A 4-year male was referred to our hospital for high fever. Incidentally, abnormally high blood pressure was detected. A thorough examination revealed severe stenosis at the origin of two left renal arteries and elevation of plasma renin activity as well as aldosterone level. Some lesions of previous asymptomatic brain bleeding were also revealed. Instead of using prosthetic materials, we transected renal arteries and directly anastomosed them to abdominal aorta, expecting subsequent growth of native vessels. The postoperative course was uneventful. The plasma renin activity and aldosterone level as well as the dose of antihypertensive drug decreased significantly after the operation.

Keywords: pediatric renovascular hypertension, surgery, fibromuscular dysplasia

Introduction

Pediatric renovascular occlusive disease is a rare cause of hypertension in children. If untreated, it can lead to serious complications, such as hemorrhagic shock, hypertensive encephalopathy with impaired mental development and left ventricular hypertrophy with severe diastolic dysfunction.

The optimal treatment for pediatric renovascular hypertension has not been clear. For refractory hypertension against medical treatment, endovascular and surgical treatment have been performed. Renal artery stenosis is a common cause of surgically correctable hypertension in children. Before the advent of endovascular treatment, surgical revascularization was the primary therapy for patients. However, as this is a rare clinical condition, there have been few reports demonstrating a large scale of surgical outcomes in this population.1,2) There have been also few reports of successful endovascular treatment for pediatric population.3) Although endovascular treatment can be less invasive than surgical treatment, its long-term durability is unknown and endovascular prostheses cannot change in size along with normal vessel growth.

In this paper, we report a case of incidentally found pediatric renovascular hypertension, in which surgical treatment provided an excellent outcome.

Case Report

The patient was a 4-year old Japanese male. He complained of high fever for 5 days associated with redness of his conjunctiva and tongue. He was referred to our hospital under a suspicion of Kawasaki disease. Physical findings at admission were as follows: height: 106 cm, weight: 17 kg, blood pressure: 231/112 mmHg, body temperature: 40.1°C. His consciousness was alert. Systolic murmur was heard by chest auscultation.
The fever became insignificant soon after the admission. However, the abnormal high blood pressure remained. After starting intravenous nicardipine, a thorough examination was performed to detect the cause of abnormal high blood pressure. As a result, three-dimensional computed tomography showed two left renal arteries which had severe stenosis at their origin with poststenotic dilatation (Fig. 1). Plasma renin activity elevated to 33.3 ng/ml/hr (normal: 0.8–2.0) and aldosterone level also elevated to 574 pg/ml (normal: 36–240). The diagnosis of pediatric renovascular hypertension was made. Serum creatinine was 0.42 mg/dl. Transthoracic echocardiography revealed left ventricular hypertrophy with preserved left ventricular function. Head magnetic resonance imaging showed some small lesions of previous brain bleeding, which were considered to be caused by abnormal hypertension.

Discussion was made about the optimal treatment for him among pediatricians, cardiologists and vascular surgeons. Although some successful reports of endovascular treatment for renovascular hypertension have been demonstrated, its long-term outcome has not been clear and endovascular prostheses cannot change in size along with normal vessel growth. Moreover, from the anatomical perspective, we thought we could surgically correct the stenosis of renal arteries.

The patient was positioned in the right lateral decubitus position. The incision was made obliquely and the oblique muscles of the abdomen were divided and the retroperitoneal space was entered. After exposure of left renal arteries, heparin was administered. Right renal artery could not be detected during the operation. Aortic clamp was made both at proximal and distal side of renal arteries. We transected left renal arteries and infused 20 ml of Ringer’s lactate solution with mannitol (12.5 g/L) and methylprednisolone (125 mg/L) cooled to 4°C, which are reported to be protective for renal function.4) We created two small holes in the aorta with an aortic puncher for coronary artery and anastomosed left renal arteries directly with 7-0 monofilament, non-absorbable continuous suture (Fig. 2). The left renal ischemic time was 29 min.

The postoperative course was uneventful. The patient was extubated a few hours postoperatively. Continuous intravenous nicardipine was required for a few days after operation. However, the dose of antihypertensive drug decreased significantly from preoperative dose. Postoperative max serum creatinine was 0.39 mg/dl which was almost equivalent with preoperative value of 0.42 mg/dl. Plasma renin activity decreased to 3.3 ng/ml/hr from 33.3 ng/ml/hr and serum aldosterone also decreased to 224 pg/ml from 574 pg/ml. He was discharged from our hospital.
wise, the patient had been asymptomatic, however, if untreated, pediatric renovascular hypertension can lead to serious complications. In fact, the patient already had a sign of left ventricular hypertrophy as well as previous silent brain bleeding, which were thought to be caused by persistent abnormal hypertension. An optimal treatment was mandatory to avoid life-threatening complications.

For refractory hypertension against medical treatment, endovascular or surgical treatment has been applied. Although long-term outcomes of endovascular treatment in adults for fibromuscular dysplasia were reported, its role in pediatric population remains controversial. There is a report of early success of endovascular treatment for pediatric renovascular hypertension. However, its long-term durability in children is unknown. Moreover, as endovascular prostheses cannot change in size along with normal vessel growth, size mismatch might be revealed at long-term period.

Before the advent of endovascular treatment, surgical revascularization was the primary therapy. However, large clinical experiences with pediatric renovascular hypertension are uncommon. Thus, the appropriate surgical treatment in this population is ill defined. In terms of Japanese population, there have been few surgical case reports for pediatric renovascular hypertension.

15 days after operation with low dose of antihypertensive drugs.

Pathological findings showed an intimal type of fibromuscular dysplasia (Fig. 3).

**Discussion**

Renal artery occlusive disease is an important but very uncommon cause of hypertension in children. However, renal artery stenosis is a common cause of surgically correctable hypertension in children. Stanley, et al. reported that most of the patients who underwent surgical revascularization for pediatric renovascular hypertension had complex medial and perimedial dysplastic disease complicated with secondary intimal fibroplasias, while a few of them had inflammatory or other ill-defined stenoses. Basically, renovascular fibromuscular dysplasia tends to affect women between 15 and 50 years of age. In most cases, patients have been asymptomatic for many years, and fibromuscular dysplasia is discovered incidentally during the investigation of another problem.

In this report, we demonstrated a case of pediatric renovascular hypertension which was found incidentally during the admission for another reason. The cause of fever was eventually unknown. We think it was related to upper respiratory infection. Otherwise, the patient had been asymptomatic, however, if untreated, pediatric renovascular hypertension can lead to serious complications. In fact, the patient already had a sign of left ventricular hypertrophy as well as previous silent brain bleeding, which were thought to be caused by persistent abnormal hypertension. An optimal treatment was mandatory to avoid life-threatening complications.

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Surgery for Pediatric Renovascular Hypertension

The right kidney had lost any function before the operation and it did not have any effect on the patient's blood pressure and renal function. We have followed the patient after about 1 year after the operation. The patient has been asymptomatic and blood pressure is well controlled with oral antihypertensive medications. Pathological study showed an intimal type of fibromuscular dysplasia, which was the most common type of surgically treated pediatric renovascular hypertension. 1)

Conclusion

We experienced a successful case of surgical revascularization for pediatric renovascular occlusive disease. When adequate direct anastomosis is expected to achieve, surgical revascularization can provide an excellent as well as a radical treatment in this population.

Disclosure Statement

The authors have no conflicts of interest to disclose.

References