Clinical Case Report

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A rare case report of bilateral common and internal iliac arterial fibromuscular dysplasia

Coexisted dissection, aneurysm, and stenosis

Luyun Fan, MD, Wenjun Ma, MD, PhD, Huimin Zhang, MD*, Jun Cai, MD, PhD*

Abstract

Rationale: Iliac arterial fibromuscular dysplasia (FMD) was rarely reported and its demographic, clinical, and imaging features have not been precisely described resulting in uncertain therapeutic methods.

Patient concerns: A 31-year-old man was referred because of 3-month-ago onset hypertension, low serum potassium, and a small-sized right kidney with normal renal artery under ultrasound examination. This patient was suspected of primary aldosteronism, whereas spirolactone was poorly effective.

Diagnosis: Contrast-enhanced computed tomographic angiography (CTA) and three-dimensional reconstruction of the whole aorta discovered an aneurysm from the right common iliac artery (CIA) to the internal iliac artery, consistent with a left CIA dissection and a remarkable right renal artery aneurysm before a stenosis. Iliac and renal arteries FMD were then confirmed through digital subtraction angiography (DSA).

Intervention: Percutaneous transluminal angioplasty (PTA) of right renal artery was operated and a stent was deployed in left CIA.

Outcomes: This patient was normotensive, asymptomatic, and free from recurrence without any antihypertensive agents at an 8-month follow-up.

Lessons: To our knowledge, this is the first bilateral common and internal iliac arterial FMD case in China, with unique asymptomatic dissection, aneurysm, and renovascular hypertension. Screening for secondary hypertension in young population and for iliac or renal arterial FMD is therefore suggested with CTA and reconstruction from neck to pelvis and MRA in those with intracranial disorders. Among youth FMD, the potential of PTRA in renovascular hypertension out of antihypertensive drugs and stent in dissection is novelly indicated.

Abbreviations: CIA = common iliac artery, CTA = computed tomographic angiography, DSA = digital subtraction angiography, EIA = external iliac artery, FMD = fibromuscular dysplasia, GFR = glomerular filtration rate, IIA = internal iliac artery, LE = lower extremity, MRA = magnetic resonance angiography, PTA = percutaneous transluminal angioplasty, PTRA = percutaneous transluminal renal angioplasty.

Keywords: aneurysm, dissection, fibromuscular dysplasia, iliac artery, renovascular hypertension

1. Introduction

Fibromuscular dysplasia (FMD) is a rare non-atherosclerotic and non-inflammatory vascular disease predominantly involving women age 20 to 60 years of unknown etiology. FMD is reported in affecting various arterial beds particularly renal and carotid arteries, with angiographic manifestations including stenosis, beading, aneurysm, dissection, and tortuosity.[1] The clinical presentations of FMD vary from an asymptomatic condition, hypertension to a multisystem disease resembling vasculitis, determined largely by the distribution, type and severity of involved lesions.[2] However, iliac arterial FMD was limited reported in case reports or series, and the demographic, clinical, and imaging features of iliac arterial FMD have not been precisely described.[3–16] We herein report the first man case of bilateral common and internal iliac arterial FMD in China, with renal artery involvement discovered by computed resonance angiography (CTA) and contaminant stenosis, aneurysm, and dissection, and then perform a literature review on iliac arterial FMD.

2. Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

3. Case report

A 31-year-old man was referred because of 3-month-ago onset hypertension, with a clinic blood pressure of 207/170 mmHg, a

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State Key Laboratory of Cardiovascular Disease, National Center for Cardiovascular Diseases, Fuwai Hospital, Chinese Academy of Medical Sciences and Peking Union Medical College, Beijing, China.

** Correspondence: Huimin Zhang, Department of Hypertension, State Key Laboratory of Cardiovascular Disease, National Center for Cardiovascular Diseases, Fuwai Hospital, Chinese Academy of Medical Sciences and Peking Union Medical College, Beijing 100037, China (e-mail: fwzhanghuimin@126.com); Jun Cai, Department of Hypertension, State Key, Laboratory of Cardiovascular Disease, National Center for Cardiovascular Diseases, Fuwai Hospital, Chinese, Academy of Medical Sciences and Peking Union Medical College, Beijing 100037, China (e-mail: caijun@fuwaihospital.org).*

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serum potassium level of 3.2 mmol/L, and an abdominal ultrasound that revealed a small-size right kidney (8.3 cm × 5.0 cm × 4.9 cm) with abnormal thin cortex but normal bilateral renal arteries. His past medical history, life history, and family history were unremarkable. He was initially suspected of primary aldosteronism and prescribed with spirolactone 40 mg 3 times a day. However, his blood pressure still fluctuated around 150/110 mmHg. Spirolactone was alternated by benidipine 8 mg per day for future tests.

After admission, a blood pressure of 120/90 mmHg on both arms and a heart rate of 87 beats per minute were measured. Cardiac and abdominal examinations were unremarkable. The upper and lower extremity arterial pulses were palpable and symmetric bilaterally. Ambulatory blood pressure monitoring presented an average 24-hour blood pressure of 130/90 mmHg. Routine laboratory tests including blood chemistry (such as creatinine and potassium), liver function test, C-reactive protein, and immunological evaluation such as rheumatoid factor, erythrocyte sedimentation rate, were within normal ranges. Screening tests related to primary aldosteronism and Cushing Syndrome presented negative results except for a high serum renin level of 62.3 uIU/mL (normal, 4.4–46.1 uIU/mL). Renal scintigraphy showed normal left renal perfusion but a small-size right kidney, while both kidneys were determined with decreasing glomerular filtration rate (GFR, left kidney, 33.6 mL/min, right kidney, 20.4 mL/min). Contrast-enhanced CTA and three-dimensional reconstruction of the whole aorta showed an aneurysm formation involving the right common iliac artery (CIA) and extending into the internal iliac artery, which was consistent with a left CIA dissection and an aneurysm continued

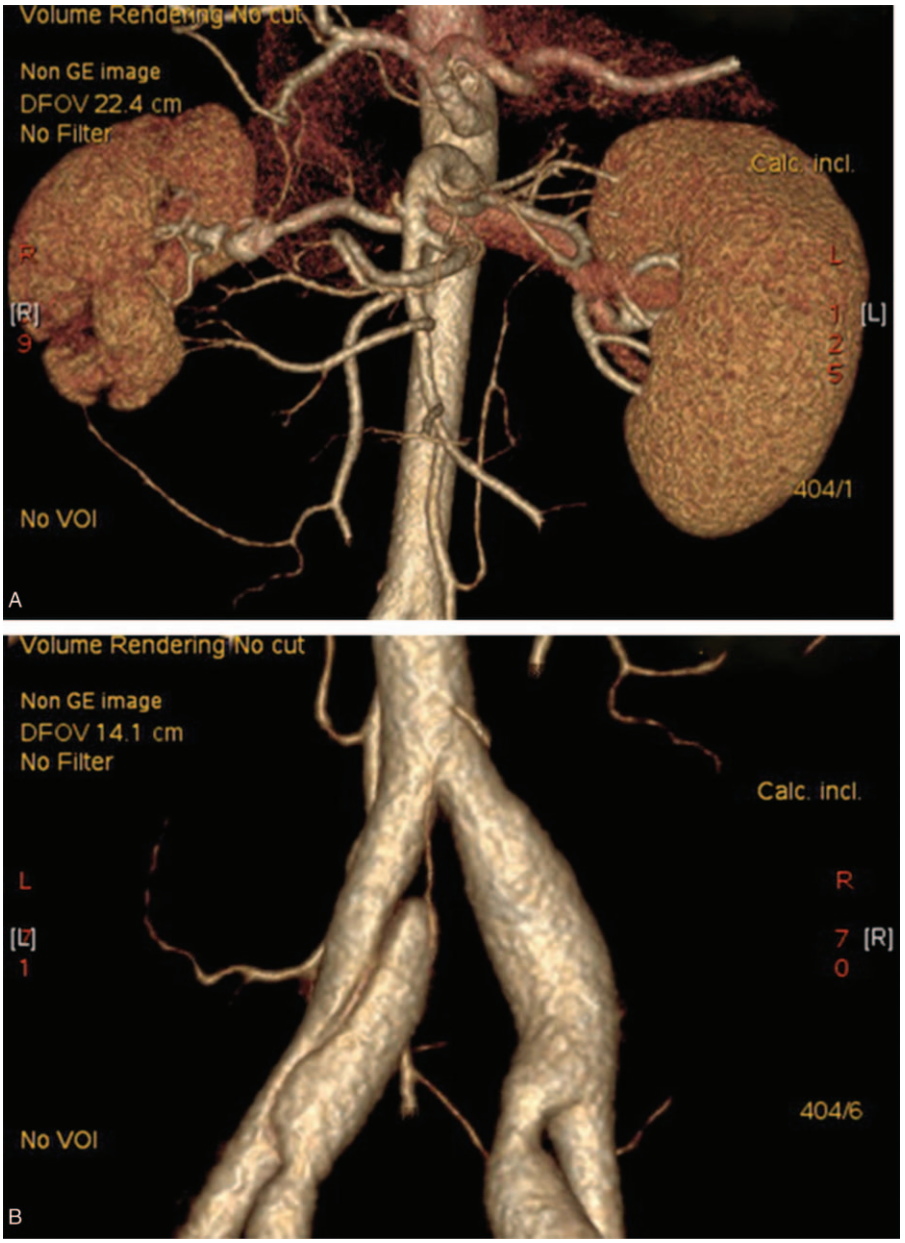

Figure 1. Contrast-enhanced computed tomography scan and three-dimensional reconstruction of the whole aorta. (A) Aneurysm involving the right common and internal iliac artery, and left common iliac artery dissection. (B) Aneurysm continued with stenosis of distal segment of right renal artery and small-sized right kidney.

with a stenosis among the distal segment of right renal artery (Fig. 1). Small size and thin cortex of the right kidney, and left adrenal plump were also observed. Other arteries including supra-arch artery were proved with no abnormality.

He was therefore diagnosed with highly suspicious FMD and scheduled for angiographic evaluation. We performed intra-arterial digital subtraction angiography (DSA) above the suprarenal abdominal aorta, demonstrating FMD with an 80% stenosis of distal right renal artery in a 10 mm length and a proximal aneurysm (Fig. 2A). Dissection in middle and distal segments of left CIA and vessel dilation from right CIA to internal iliac artery were simultaneously confirmed (Fig. 2B). Given the early-onset age, the short duration of hypertension and the presence of aneurysm, balloon angioplasty of right renal artery was performed and a stent was deployed in left CIA. Completion DSA demonstrated a right renal artery with brisk flow of contrast and disappearance of dissection in left CIA (Fig. 3). Postoperative recovery was uneventful with an even 24-hour blood pressure of 125/87 mmHg. At 8-month follow-up, he was in good life quality and his blood pressure remained well-controlled without any antihypertensive medications. Periodic imaging was recommended.

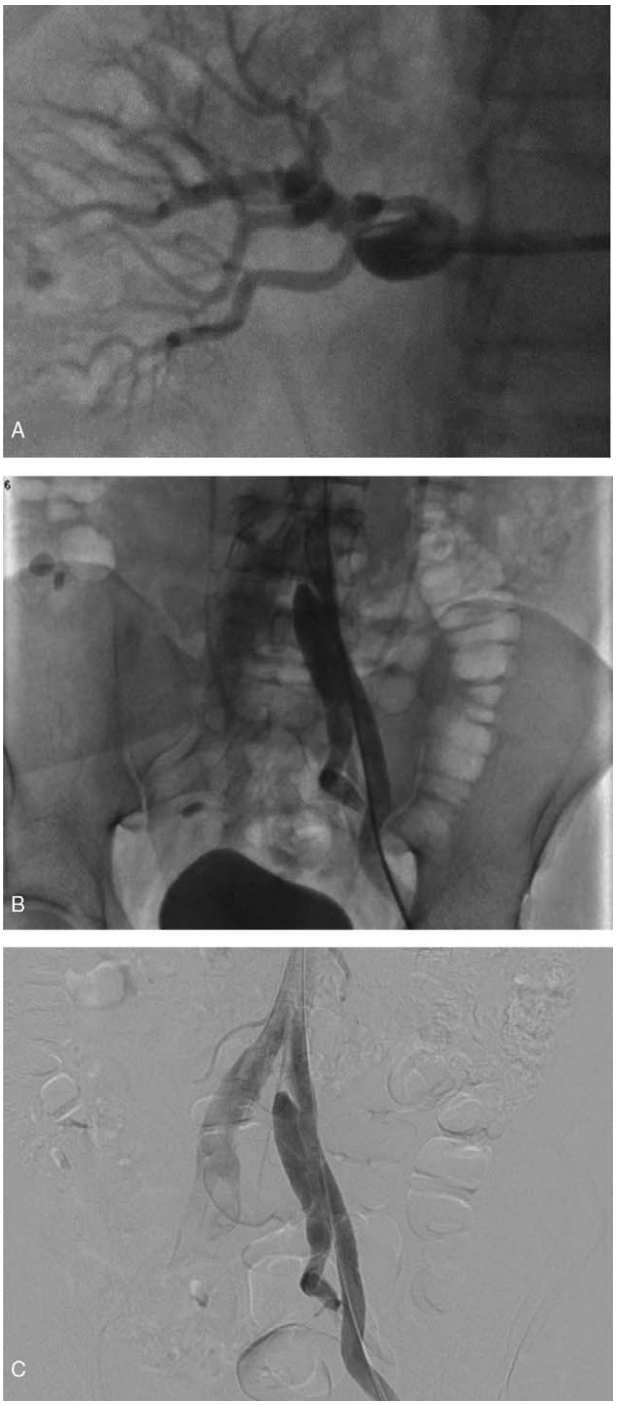

Figure 2. (A) Right renal angiography showing an 80% stenosis of distal segment of main renal artery in a 10 mm length and a proximal aneurysm. (B, C) Iliac angiographic images presenting dissection in middle and distal segments of left CIA and vessel dilation of right CIA to internal iliac artery. CIA = common iliac artery.

4. Literature review

A literature review was performed using PubMed, Web of Science, and Embase database. Original articles in English published since 1990 with full-text and detailed demographic, clinical, imaging and therapeutic information regarding iliac arterial FMD were included. A total of 104 cases on iliac arterial FMD were reported (Table 1).[3–16] The mean age at diagnosis was 52.2 ± 12.6 years, and woman accounted for 86.5%. In

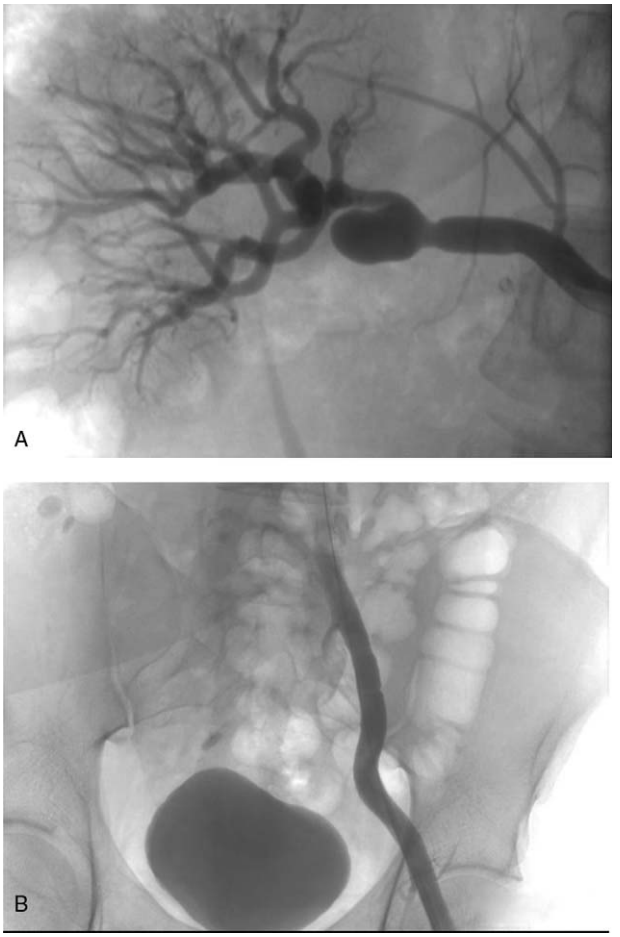

Figure 3. (A) Right renal angiography after angioplasty showing a wide renal artery with diminishing aneurysm. (B) Iliac angiographic images after stenting presenting absence of dissection.

Table 1

Reported cases and case series of iliac arteries fybromuscular dysplasia.

Author	Year	Age	Gender	Symptom	Sign	Iliac artery involvement	Other artery involvement	Angiographic changes	Treatment	Outcome
Sauer et al[3]	1990	67	M	Left distal extremity microemboli	None	Left EIA	None	String-of-beads	Resection and Dacron graft	Improved
		55	M	Distal extremity microemboli	None	Bilateral EIA	None	String-of-beads	Resection with end to end anastomosis	Improved
		71	M	Left distal extremity microemboli	None	Left EIA	Bilateral renal	String-of-beads	Resection and Dacron graft	Improved
		56	F	Right thigh and leg claudication	Bruit Pulse deficit	Right EIA Left iliac	Left renal	Dissection	Conservative	Improved
		62	F	Left thigh and calf claudication	Bruit Pulse deficit	Bilateral EIA	Bilateral renal Internal carotid vertebral	Stenosis	Open surgery with dilation	Improved
		45	F	Bilateral claudication	Pulse deficit	Bilateral EIA	Bilateral renal Right superficial femoral	N/A	Open surgery with dilation	Improved
		61	F	Left calf claudication Left malleolar ulcer	None	Bilateral EIA	Right renal	N/A	Aortobifemoral bypass	Improved
		35	F	Bilateral claudication	None	Bilateral EIA	Bilateral renal	N/A	Open surgery with dilation	Improved
Thevenet et al[4]	1992	45	F	Lower limb acute ischemia	Pulse deficit	Bilateral EIA	None	Dissection	Aortobifemoral bypass	Improved
		63	F	Bilateral recumbent pain	None	Bilateral EIA	None	N/A	Bypass	Improved
		48	F	Claudication Pelvic pain	None	Bilateral EIA	None	N/A	Aortoiliac bypass	Improved
		51	M	Claudication	None	Bilateral EIA	Right renal	Dissection	Iliofemoral bypass	Improved
		47	F	Abdominal pain	Bruit	Right EIA	None	Stenosis	Bypass	Improved
		54	F	Claudication Recumbent pain Hypertension	None	Bilateral EIA	Bilateral renal	N/A	Bypass	Improved
		29	F	Iliac fossa pain	None	Left EIA	Left thyrocervical trunk	Dissection Aneurysm Stenosis	Interposition with vein	Improved
		53	M	Claudication	None	Right EIA	None	Dissection	Iliofemoral bypass	Improved
Mandke et al[5]	1993	18	F	Left leg claudication Hypertension	Pulse deficit	Left CIA	Left renal	Stenosis	Nephrectomy Atherectomy	Improved
Atsuta et al[6]	2003	69	F	Abdominal pain	Pulsatile mass	Right CIA Left IIA	None	Aneurysm	Resection and reconstruction with a Y graft	Improved
Honjo et al[7]	2004	30	M	Abdominal pain Shock	None	Left CIA EIA	None	Dissection Rupture	Delayed surgery	Dead
Chin and Ntsekhe[8]	2009	50	F	Asymptomatic Hypertension	Bruits	Bilateral iliac	Bilateral renal	String-of-beads	Conservative	Improved
Akashi et al[9]	2010	49	M	Right leg claudication Right abdominal pain	Bruits Pulse deficits	Right CIA, EIA	None	Dissection	Replacement with Dacron prosthesis	Improved
Sugiura et al[10]	2011	30	M	Sudden abdominal pain	None	Right EIA	Bilateral renal Superior mesenteric	Dissections Stenosis	Conservative	Improved
Okazaki et al[11]	2011	63	F	Left leg claudication, Hypertension	Pulsatile mass Pulse deficit	Bilateral CIA	Internal carotid Superficial femoral Popliteal	Aneurysm Stenosis	Resection and reconstruction with a Y graft, bypass	Improved
		37	M	Sudden back pain	None	Bilateral EIA	None	Dissection Rupture	Interposition with ePTFE	Improved
Rastogi et al[12]	2012	88	F	Right ankle ulcer Hypertension	Pulse deficits	Bilateral EIA	None	String-of-beads	Balloon angioplasty Stent angioplasty	Improved
Ketha et al[13] (14 cases)	2014	56±12	F (86.0%)	Asymptomatic (57%) Claudication (53%) Hypertension (79%)	N/A	EIA (100%) CIA (7.1%) IIA (7.1%)	Renal (85.7%) Carotid (36%) Coronary (7%) Peripheral (14%)	No dissection	Conservative (28.6%) Balloon angioplasty (57.1%) Stent angioplasty (14.3%)	Symptom resolution (8/10;80%)
Olin and Kadian-Dodov[14]	2014	52	F	Right buttock and thigh claudication Hypertension	Bruit	Right EIA	Right renal	Stenosis	Stent angioplasty Balloon angioplasty	Improved
Niizeki et al[15]	2015	60	F	Bilateral claudication	None	Bilateral EIA	None	String-of-beads	Balloon angioplasty	Improved
Brinza et al[16] (62 cases)	2016	52±11.3	F (96.8%)	Asymptomatic (71.0%) Claudication (37.1%) Hypertension (62.9%)	Bruits (88.7%) Pulse deficits (45.2%)	EIA (87.1%) CIA (19.4%) IIA (11.3%)	Renal (80.6%) Carotid (79.0%) Vertebral (43.5%) Mesenteric (25.8%)	Dissection (4.8%)	Conservative (98.4%) Open surgery and interposition graft and angioplasty (1.6%)	N/A
Present case	2017	31	M	Asymptomatic Hypertension	None	Bilateral CIA IIA	Right renal	Dissection Aneurysm Stenosis	Balloon angioplasty Stent angioplasty	Improved

CIA = common iliac artery, EIA = external iliac artery, F = female, IIA = internal iliac artery, M = male, N/A = not applicable.

terms of clinic presentation related to iliac arteries, 54 (51.9%) patients were asymptomatic, while 43 (41.3%) patients presented intermittent claudication or leg pain (an atypical symptom of claudication). Fourteen (13.5%) patients complained of pain in other positions including 3 microembolism, 1 acute ischemia, and 1 shock. Hypertension was observed in 56 (53.8%) patients. Data relevant to physical examination of FMD could be extracted from 90 patients. Sixty-one (67.8%) patients had lower abdominal or iliofemoral bruits and 36 (40.0%) had ≥1 pulse deficits of the lower extremity arteries. Besides, 14 (13.5%) patients had spontaneous dissection of iliac arteries, 2 coexisted with aneurysm, and 2 progressed to rupture with 1 died after delayed surgery. Only 10 had detailed records. The mean age was 41.1 years (range, 30–56), and the female to male ratio was 3/7. The majority of patients (n=9; 90.0%) presented with claudication or pain, undergoing open air surgery, or medications with consequently improved conditions. Up to now, the external iliac artery was the most common location of iliac FMD (n=91; 87.5%), whereas common iliac artery and internal iliac artery represented 18.3% and 9.6% lesions, respectively. Bilateral iliac arterial involvement presented in 60 FMD patients. Other arteries affected were reported in renal artery (n=75; 72.1%), carotid artery (n=56; 53.8%), vertebral artery (n=28; 26.9%), mesenteric artery (n=17; 16.3%), coronary artery (n=1; 1.0%), and other (n=16; 15.4%). Sixty-eight (65.4%) iliac arterial FMD patients were managed conservatively. Twenty-two (21.2%) patients successfully underwent bypass or resection and interposition, while 14 (13.5%) patients experienced angioplasties or stenting with 12 (85.7%) reaching symptom resolution.

5. Discussion

FMD is an idiopathic arteriopathy predominantly involving medium-sized vascular territories. The prevalence of FMD is uncertain and probably underestimated for circumscribed cognition and lacking in systematic screening. In the US FMD Registry, woman accounted for 91% (406/447) registrants, and the clinical phenotype of FMD expanded from traditionally rare cause of renovascular hypertension to now various anatomic distributions and clinical manifestations.[17] Of patients with imaging, a proportion of 79.7% (294/369) was renal artery FMD and 42 LE arteries FMD (primarily iliac artery) confirmed in 70 suspicious cases.[17]

The patient herein reported is the first man bilateral iliac arteries FMD in China, presenting asymptomatic but renovascular hypertension. CTA and DSA showed rare features that FMD simultaneously affected right renal artery and bilateral common and internal iliac arteries. Upon our literature review, iliac arterial FMD patients were overwhelmingly woman with a mean age of 52.2±12.6 years, similar to enrolled FMD population in US Registry. Asymptomatic condition and claudication consisted the major clinical phenotypes of iliac arterial FMD, contrast to general FMD population with mere 25(5.6%) asymptomatic patients and 23(5.2%) presenting claudication.[17] Hypertension was diagnosed in majority of general and iliac arterial FMD patients (63.8%; 53.8%).[17] Common and internal iliac artery FMD were only around one thirds of external artery FMD. Other vascular beds, primarily renal arteries, were popularly involved in iliac arterial FMD, convincingly indicating the common hypertension and importance of system screening. A recent study retrospected 360 patients from a FMD registry center, extracting 113 undergoing one-time screening under a specialized CTA protocol of chest, abdomen, and pelvis. New arterial beading, aneurysm, and dissection were surprisingly observed in 49%, 19%, and 3% of patients separately. Reformatted images were found crucial and shaping final assessment of the readers.[18] These conclusions underpin the recommendation of high-resolution CTA screening and reconstruction from neck to pelvis among FMD patients. Magnetic resonance angiography (MRA) without ionizing radiation is suggested for head screening because aneurysms (the most common manifestation of intracranial FMD) can be accurately identified. Besides, our case sparsely coexisted with stenosis, aneurysm, and dissection. One dissection, generally presenting ischemia symptoms, was observed in approximate 6 iliac FMD patients from our review with an inclination to young man population. Recent sub-studies of the US Registry observed dissection in 25.7% FMD patients characterized with lower age (48.4 vs 53.5 years) and man population, underpinning our conclusion.[19–20] Notably, an early recognition for acute presentation of dissection may explain the trends, while our case uniquely reminds of iliac arterial FMD with asymptomatic dissection.

In our case, the renal artery disorder was initially neglected, implying limitation of ultrasound in screening distal renal artery manifestation for operator dependence and slower acquisition. CTA is therefore recommended for asymmetric kidneys suspected of renal artery changes. This patient also presented long-term hypokalemia, resulting in an inclination to primary aldosteronism however with poor spirolactone efficiency. The initial increased aldosterone was actually secondary to renal artery stenosis. Secondary hypertension is consequently recommended to undergo systematic and precise evaluation among the young hypertensive population.

Given therapeutic methods, revascularized renal artery FMD patients were reported often man presenting focal manifestation with a earlyer diagnostic age, higher BP-level hypertension, and higher prevalence of renal asymmetry than patients with conservative therapy.[21] According to the sole systematic review summarizing outcomes of 47 published series with percutaneous transluminal renal angioplasty (PTRA), the combined rate of cure or improvement of hypertension was 86.4% (95% confidence interval [CI], 83.2–89.3) and improved among younger patients with shorter-duration hypertension.[22] Therefore, renal artery angioplasty was suggested for high likelihood of improvement or cure of new-onset renal hypertension, renal artery aneurysm, and renal function loss in our young patient. There were no clear guidelines for iliac arterial FMD or dissections. In terms of our review, 65.4% iliac artery FMD patients were controlled with mere medications and 85.7% of those experiencing angioplasty released from severe symptoms, whereas iliac arterial dissection progressing to rupture resulted in 50% death. Angioplasty with stenting was decided in our patient from the progressive enlargement of the dissected pseudolumen and possible catastrophic consequences of ruptured dissecting aneurysm although asymptomatic, followed by disappearance of the dissection and free from recurrence.

In this article, we report a rare young male patient with iliac and renal arterial FMD with no particular symptoms but new-onset hypertension. This patient is also the first bilateral CIA and IIA FMD in China with both aneurysm and dissection. Iliac arterial FMD is reviewed showing common asymptomatic or claudication symptoms with coexisting hypertension subsequent to renal artery involvement. A system screening among iliac arterial FMD is suggested and CTA is proved to be an optimal screening methodology. Dissection, an infrequent angiographic manifestation of FMD rarely concurrent with aneurysm, inclines

to affect young male patients presenting ischemia or claudication. It is recommended that PTA is reasonable for renovascular hypertension with FMD and stenting seems to be promising for iliac arterial FMD with dissection.

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