Case Reports

Solitary Common Iliac Artery Inflammatory Aneurysm in a Healthy Woman: Case Report and Review of the Literature

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Inflammatory aneurysms represent only 3–10% of all aortoiliac aneurysms and tend to be more common in men. We report a case of a solitary inflammatory aneurysm of the right common iliac artery in a healthy young woman. The patient presented with persistent abdominal and right flank pain. She had no risk factors for vascular disease, except mild hypertension and a strong family history of aneurysm disease. Her work-up demonstrated a 3.0 cm right common iliac artery aneurysm with intramural thrombus, focal calcification, and perianeurysmal inflammation without evidence of systemic atherosclerosis. There was right hydroureteronephrosis secondary to ureteral compression by the inflammatory aneurysm. She underwent open right common iliac artery aneurysmorraphy with polytetrafluoroethylene interposition graft and concomitant ureterolysis without complication. She remains asymptomatic more than 1 year postoperatively with no evidence of additional aneurysm disease, resolution of her hydroureteronephrosis, and normal kidney function. We report a rare case of a solitary inflammatory aneurysm of the right common iliac artery in a healthy young woman, with a review of the current literature on inflammatory aneurysms.

Inflammatory abdominal aortic aneurysms (AAAs) are defined as aneurysm disease with an intense, surrounding host inflammatory response seen as thick perianeurysmal fibrosis with inflammatory cell infiltration and adhesions to surrounding structures.[1] Inflammatory aneurysms represent only 3–10% of all aortoiliac aneurysms and tend to have a male predominance.[2-4] The presence of an isolated common iliac artery inflammatory aneurysm has been reported in only two previous cases.[5] In both cases, the aneurysm occurred in the presence of systemic atherosclerotic disease, typical of inflammatory aneurysms. We report a case of a solitary right common iliac artery inflammatory aneurysm in a young, otherwise healthy woman, in the absence of any evidence of systemic atherosclerotic disease or significant risk factors, except for a strong family history of aortoiliac aneurysm disease.

CASE REPORT

The patient was a 49-year-old woman who presented to the Emergency Department with a 1-week history of right flank and right lower quadrant abdominal pain. She had been treated by her primary-care physician during the previous week for a urinary tract infection, but the pain persisted. The pain was intermittent and not exacerbated or

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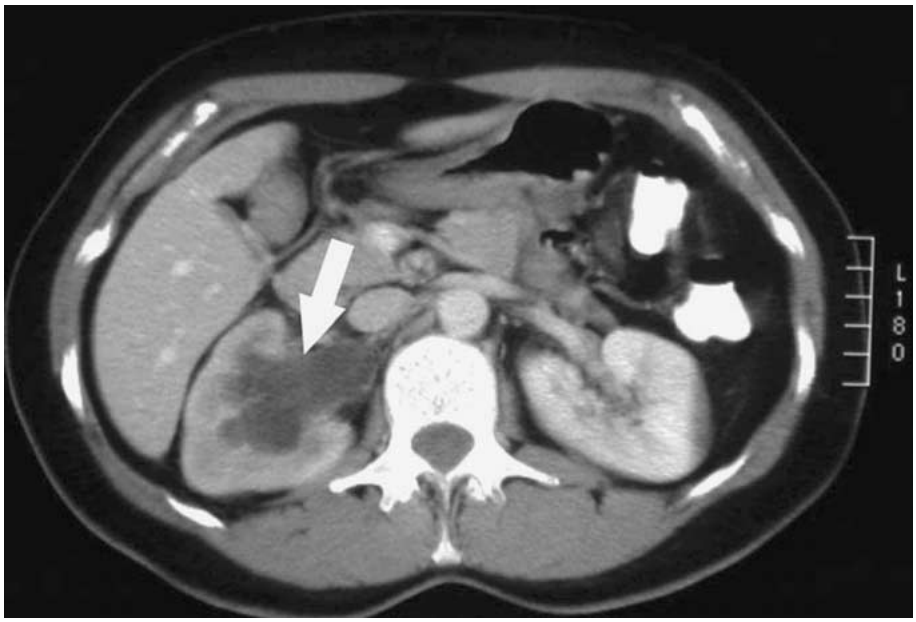

Fig. 1. Preoperative CT scan demonstrating severe right hydroureteronephrosis (*arrow*).

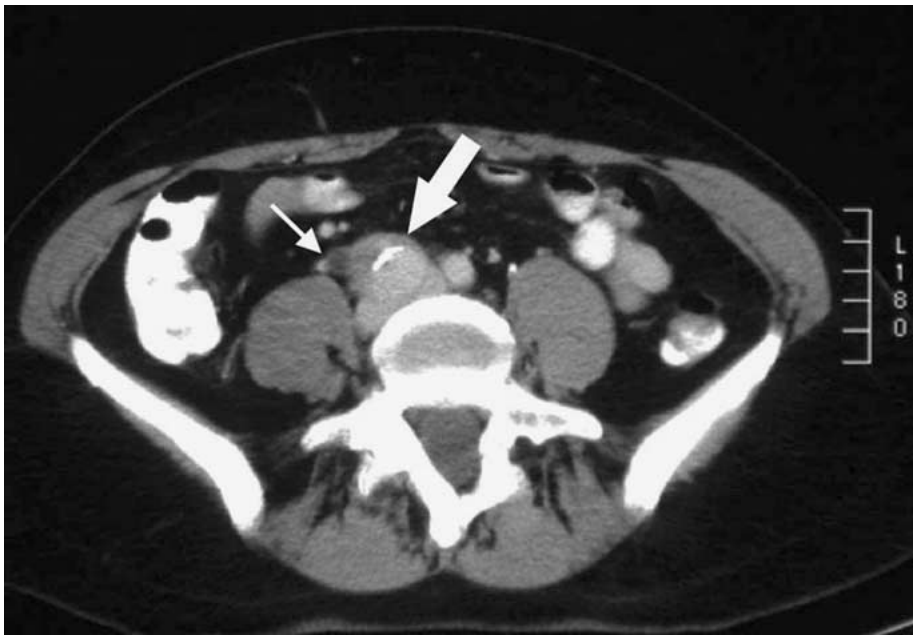

Fig. 2. Preoperative CT scan demonstrating 3 cm right common iliac artery aneurysm with focal calcification (*large arrow*) and hydroureteronephrosis (*small arrow*).

attenuated by any factors. She denied gastrointestinal symptoms, menstrual irregularities, renal lithiasis, or any history of abdominal trauma. Her past medical history was significant for cholelithiasis, for which she underwent laparoscopic cholecystectomy, and a left inguinal hernia repair. She took no medications and had no smoking history. Her family history was significant for an isolated right iliac artery aneurysm in her father and AAA in a maternal uncle. On physical examination, she was afebrile, her pulse was 108, and her blood pressure was 153/66. She had normal cardiac and pulmonary exams, and her abdomen was soft and nondistended. There was minimal tenderness to deep palpation diffusely without evidence of peritonitis. There were no palpable, pulsatile abdominal masses or bruits, and bowel sounds were present. Her pelvic exam was without abnormality. She had normal brachial, radial, femoral, popliteal, and pedal pulses bilaterally. Laboratory values were significant for a leukocytosis of 10.8, a hematocrit of 38.1%, and a creatinine of 1.4 mg/dL. She was

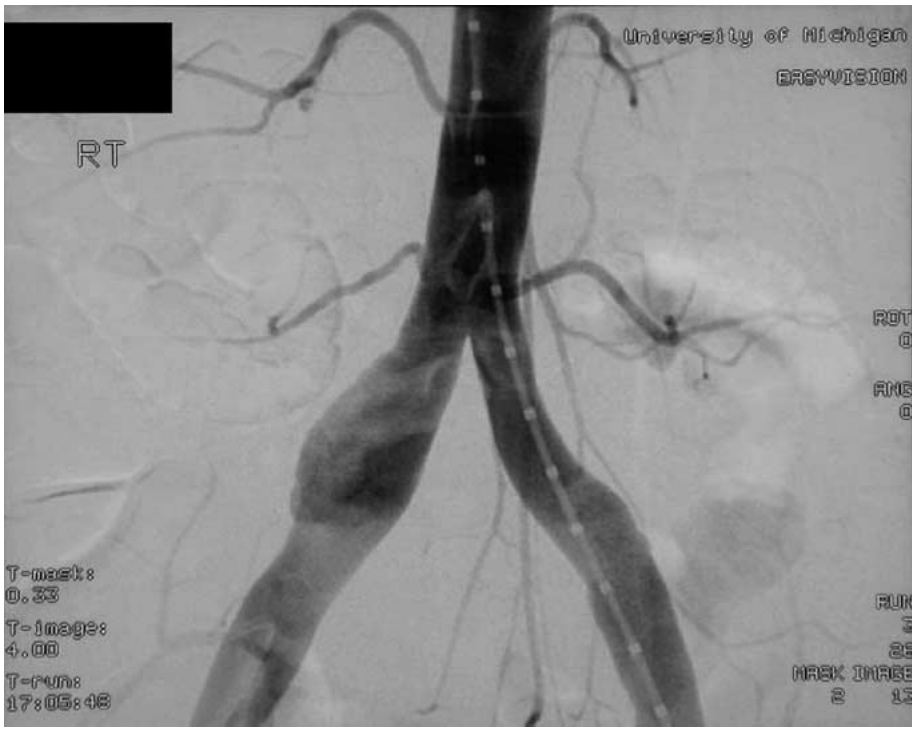

Fig. 3. Preoperative angiogram demonstrating right common iliac artery aneurysm without evidence of rupture.

also noted to have microscopic hematuria and pyuria without infectious organisms by Gram stain and urine culture.

A computed tomographic (CT) scan of her abdomen and pelvis was obtained. The study demonstrated severe right hydroureteronephrosis (Fig. 1) with a solitary, 3.0 cm right common iliac artery aneurysm with intramural thrombus, focal calcification, and periarterial inflammation. There was no evidence of rupture (Fig. 2), synchronous aneurysm disease, or atherosclerosis in her abdominal aorta or elsewhere in her iliac system. She underwent angiography for further evaluation and potential endovascular repair. This redemonstrated an isolated right common iliac artery aneurysm with no evidence of rupture, which was not amenable to endovascular repair secondary to a short proximal common iliac artery neck (Fig. 3). Given this finding, along with the patient's young age and good health, it was decided that the most durable repair would be traditional open aneurysm repair with a polytetrafluoroethylene (PTFE) interposition graft. She underwent preoperative right ureteral stent placement to facilitate intraoperative identification of the ureter. A transperitoneal repair was performed through an infraumbilical, transverse incision. The distal aorta and entire iliac system were carefully examined, and no other abnormalities were noted. Intraoperatively, the aneurysm appeared white and glistening with thick, perianeurysmal fibrosis and was densely adherent to the common iliac vein and right ureter. Ureterolysis was performed. The proximal right common iliac, internal iliac, and distal external iliac arteries were encircled and clamped and the aneurysm sac was opened. There was a focal, smooth calcific plaque within the arterial wall, which was easily enucleated. The aneurysm wall was >1 cm thick without gross evidence of infection. Aneurysmorraphy was performed with a 10 mm PTFE interposition graft from the proximal common iliac artery neck to the confluence of the internal and external iliac arteries. We attempted to close the aneurysm wall over the graft but were unable to. Therefore, the thickened retroperitoneal layer was interposed between the graft and the ureter to provide a tissue partition between the two structures.

Her postoperative course was uncomplicated. No organisms grew from her blood or urine cultures. Arterial wall pathology demonstrated focal calcific atherosclerosis although a full-thickness specimen of the arterial wall was not analyzed. There was no histological evidence of an infectious etiology. She was discharged home on postoperative day 4. Her creatinine decreased to 1.2 mg/dL at the time of discharge. The ureteral stent was removed on an outpatient basis on postoperative day 15. An intravenous pyelogram obtained 4 months postoperatively demonstrated complete resolution of

the obstruction with only mild residual fullness of the right renal pelvis and complete resolution of her ureteral dilation (Fig. 4). She continues to do well more than 1 year postoperatively with normal ankle-brachial indices and no evidence of additional aneurysm disease by CT scan.

DISCUSSION

The term *inflammatory aneurysm* was initially used by Walker et al. in 1972 to describe aortic aneurysm disease with significant perianeurysmal inflammation and adhesions to surrounding structures.[1] This subtype of aneurysm has been reported to account for approximately 3–10% of aortic aneurysms in predominantly males.[2-4] Diagnosis is suggested by CT scan as the perianeurysmal fibrosis is readily apparent. In approximately 20–30% of patients, hydronephrosis or renal failure is present at the time of diagnosis secondary to the inflammatory process involving one or both ureters.[3,4] Rarely, bowel obstruction is observed as the duodenum becomes encased by inflammation.[6] Fever, weight loss, and an elevated erythrocyte sedimentation rate (ESR) in the presence of an aneurysm suggest the diagnosis of an inflammatory aneurysm.[1,3,7] Unfortunately, no ESR or C-reactive protein levels were determined in this patient.

The etiology of inflammatory aneurysms remains unknown. There is evidence to suggest that it may be multifactorial with genetic, autoimmune, infectious, and environmental factors playing a role. Inflammatory aneurysms have a strong association with smoking and hypertension in males in the seventh decade of life.[2-4] They may belong to a subtype of atherosclerotic aneurysm with an exaggerated immune response to atherosclerotic plaques or a disease entity within the spectrum of retroperitoneal fibrosis. Nitecki et al.[7] reported a possible genetic predisposition for inflammatory aneurysm disease. They noted that 17% of patients with inflammatory aneurysms had a family history of AAA. Subsequent studies suggest that the association may be linked to the HLA-DR B1 locus.[2] The patient reported here was a female with no history of smoking, diabetes, or vascular disease. A genetic predilection likely played a major role in the etiology of her aneurysm. Indeed, her father had had an isolated right common iliac artery aneurysm, although it is unclear whether it was inflammatory in nature, and her maternal uncle had been treated for AAA. Her family history and mild hypertension seem to be her only risk factors, lending significant support to the notion of a genetic predisposition for inflammatory aneurysms.

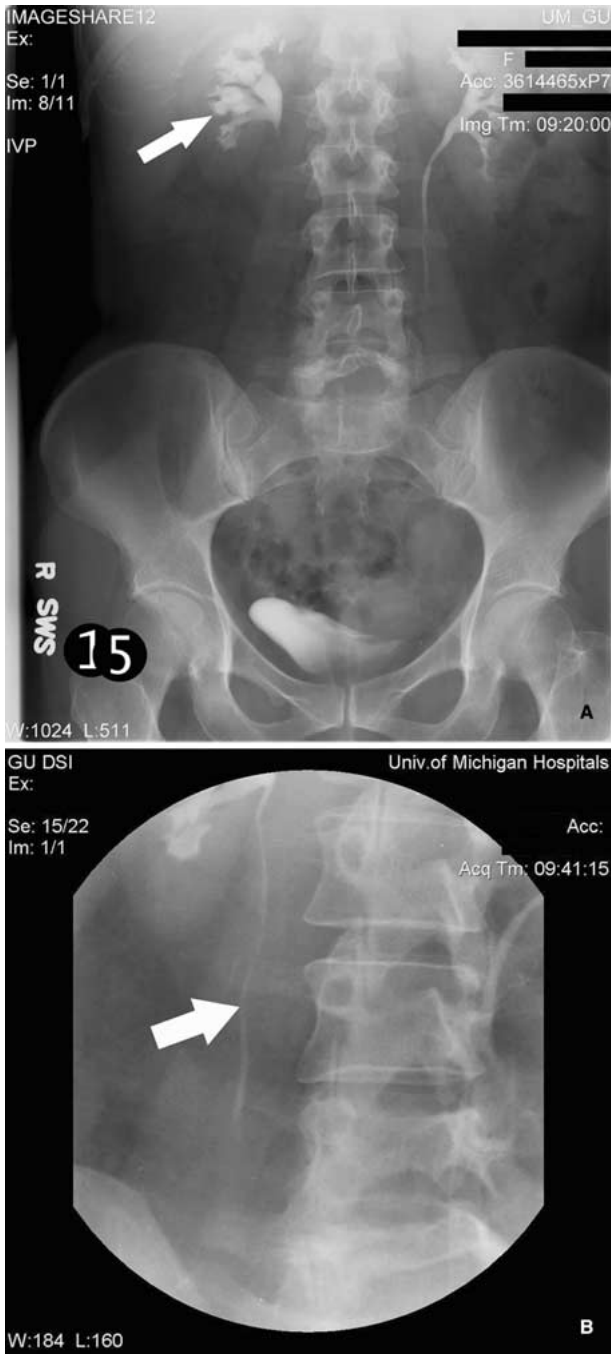

Fig. 4. A Postoperative intravenous pyelogram demonstrating mild residual right renal pelvic fullness (*arrow*). **B** Postoperative intravenous pyelogram demonstrating resolution of right hydroureteronephrosis (*arrow*).

Studies have suggested a relationship between AAA, giant cell arteritis, and Takayasu's disease.[7,8] Haug et al.[9] described an association of inflammatory aneurysms with various autoimmune diseases, including rheumatoid arthritis, systemic lupus erythematosus, and giant cell

arteritis. Over an 11-year period, they noted that 19% of patients with inflammatory AAA had an associated autoimmune disease, while no patients with noninflammatory AAA demonstrated an association. Takayasu's arteritis, or "pulseless disease," is a chronic inflammatory disease which affects the aorta and its large branches. Typical vascular lesions consist of multiple, long stenoses and rare calcifications; however, associated inflammatory-type aneurysms have been described. Patients tend to be young females and frequently present with diminished peripheral pulses, cerebrovascular insufficiency, or renovascular hypertension.[10,11] Pathology generally demonstrates inflammatory infiltrates followed by fibrosis and destruction of elastic tissue.[12] Although our pathology specimen did not fully assess the entire thickness of the aneurysm wall, it did demonstrate focal calcification and atherosclerosis. Although the histopathology could not confirm or refute the diagnosis, focal calcification within an isolated, thick-walled aneurysmal segment of the iliac artery with dense perianeurysmal adhesions is consistent with an inflammatory aneurysm. The gross appearance of the aneurysm combined with her family history further support the diagnosis of an isolated, atherosclerotic inflammatory aneurysm. The lack of other signs and symptoms commonly associated with Takayasu's arteritis or an infectious etiology makes these diagnoses unlikely.

Behçet's disease is a chronic, relapsing disease of unknown etiology that has also been associated with inflammatory aneurysms. It affects predominantly young males and occurs more frequently in Middle Eastern and Asian countries. Behçet's disease is characterized by aphthous and genital ulcers, uveitis, and a positive pathergy test but can involve other organ systems as well.[13] Vascular involvement typically presents as venous thrombophlebitis and inflammation of the arterial media and adventitia, with inflammatory aneurysms as part of the spectrum.[14,15]

Idiopathic retroperitoneal fibrosis, also known as Ormond's disease, affects the soft tissues of the retroperitoneum and is characterized by compression and encasement of the adjacent tissues. The fibrosis may extend from the mediastinum to the pelvis. The ureters are the structures most frequently affected, which is manifested as hydronephrosis or renal failure.[16] Marcolongo et al.[17] demonstrated that immunosuppressive therapy may be effective for the treatment of retroperitoneal fibrosis. Similarly, several authors have advocated steroid therapy to decrease inflammation before undergoing inflammatory aneurysm repair.[18,19] However, aneurysm repair is still required given the risk of rupture.[3,4] There is no consensus about performing operative ureterolysis since perianeurysmal inflammation appears to resolve after aneurysm repair.[20] In this case, however, the ureter traversed the area, requiring operative repair. Therefore, ureterolysis was mandatory and was performed without incident.

Isolated inflammatory aneurysms of the iliac arteries are rare. There have been two reported cases of solitary inflammatory iliac aneurysms.[5] In both cases, the patients were older males with evidence of systemic atherosclerosis typical of inflammatory aneurysms. Each also had unilateral hydroureteronephrosis. Other risk factors were not discussed. A single case each of isolated superior mesenteric artery and splenic artery inflammatory aneurysms have also been reported.[21,22] All were repaired by open aneurysmectomy. The patient reported here had no evidence of additional aneurysm disease or systemic atherosclerosis.

Inflammatory aneurysms have been successfully repaired using both open and endovascular techniques. Indeed, there may be a higher operative complication rate with open repair given the dense perianeurysmal fibrosis and adhesions to adjacent organs.[3,4,23] Spontaneous regression of inflammation has been reported after endovascular exclusion, making this a promising approach for inflammatory aneurysms.[24,25] In this case involving an otherwise healthy patient with a short aneurysm neck, the most durable repair was believed to be achieved using an open technique.

CONCLUSIONS

An isolated, unilateral common iliac artery inflammatory aneurysm is a rare entity. The etiology of inflammatory aneurysm disease remains unknown; however, it is likely multifactorial. The major risk factors in the present patient appeared to be only a family history of isolated iliac aneurysm and mild hypertension. The differential diagnosis of atypical abdominal pain in a healthy woman is wide and varied. In patients with a family history significant for aneurysm disease, a detailed vascular examination should be performed.

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