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Novel, congenital iliac arterial anatomy: Absent common iliac arteries and left internal iliac artery

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Congenital anomalies of the iliac arterial system are rare and can be associated with ischemia. With an increase in vascular imaging and interventions, such anomalies are likely to be encountered with greater frequency. We present the case of a 25-year-old female who was incidentally found to have absence of the common iliac arteries bilaterally and the left internal iliac artery, a constellation not previously reported in the literature. We present relevant imaging findings, review embryonic vascular development, and discuss potential clinical implications.

Case report

A 25-year-old female presented to the emergency department following unintended ingestion of a callus remover containing potassium hydroxide. The patient complained of burning chest pain and abdominal pain associated with nausea and vomiting. The workup included esophagogastroduodenoscopy (EGD) and contrast-enhanced computed tomography (CT) scans of the chest, abdomen, and pelvis. Imaging revealed unexpected unique arterial vasculature, with absence of the common iliac arteries bilaterally and the left internal iliac artery (Figs. 1-2). Prominent collaterals were noted, including enlarged iliolumbar arteries, lumbar arteries, and the median sacral artery, which contributed to the reconstitution of flow into the external iliac arteries (Fig. 2A).

Given the nondynamic nature of CT image acquisition (as compared to catheter angiography), some inference was necessary to reconstruct the proposed pathway of collateralization based on vessel calibers, anastomoses, and normal anatomy. On the right, there was absence of the common



Figure 1. Axial, contrast-enhanced CT image through the level of the upper pelvis. There is absence of the common iliac arteries bilaterally. Note, however, the presence of both the right and left common iliac veins (white arrows). An enlarged median sacral artery (curved white arrow) provides a means of collateral flow. An enlarged superior hemorrhoidal artery with the paired vein is also noted (black arrowheads).

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iliac artery with an enlarged right iliolumbar artery supplying the majority of flow into the external and internal iliac arteries through the lumbar arteries, as well as collateral supply by sacral branches of the median sacral artery (Fig. 2B). On the left, there was absence of both the common and internal iliac arteries with an enlarged left iliolumbar artery supplying flow into the external iliac artery and pelvic visceral branches as well as collateral supply by the superior hemorrhoidal artery (Fig. 2C).

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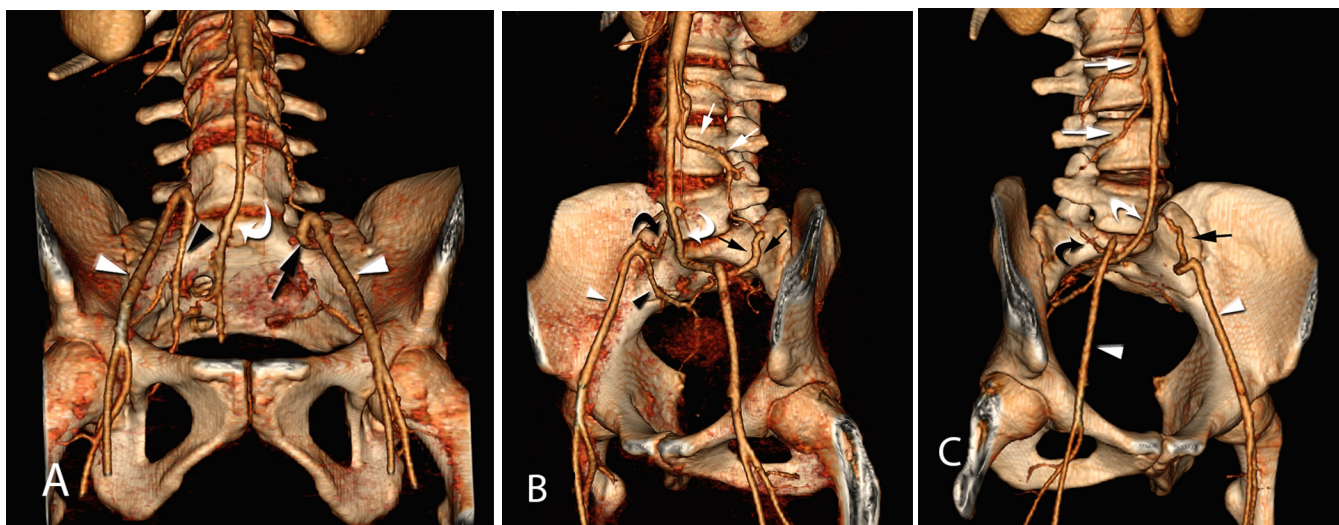


Figure 2. A. Frontal, three-dimensional (3D) volume-rendered image of the abdomen and pelvis. There is absence of common iliac arteries bilaterally with termination of the abdominal aorta into an enlarged median sacral artery (white curved arrow). Note the presence of the right internal iliac artery (black arrowhead). There is an enlarged left iliolumbar artery (black arrow). There is reconstitution of flow in the bilateral external iliac arteries. B. Left anterior oblique 3D volume-rendered image of the abdomen and pelvis. Note the right iliolumbar artery (black curved arrow) and right internal iliac artery (black arrowhead) with reconstitution of the right external iliac artery. There is a large left fourth lumbar artery (white arrows,) with anastomosis to a large left iliolumbar artery (black arrows). Note again a large median sacral artery (curved white arrow). C. Right anterior oblique 3D volume-rendered image of the abdomen and pelvis. There are prominent lumbar arteries (white arrows), large right (curved black arrow) and left (black arrow) iliolumbar arteries, and a prominent median sacral artery, all providing flow to the reconstituted external iliac arteries bilaterally.

A refocused history and physical examination of the patient revealed additional relevant clinical information. The patient reported a longstanding history of exercise-induced lower-extremity pain since childhood. The patient also reported intermittent lower-extremity paresthesias, numbness, and cyanosis. There was no history of diabetes, tobacco use, prior trauma, or instrumentation. On physical examination, there were palpable but diminished posterior tibial and dorsalis pedis pulses bilaterally without evidence of cyanosis, muscle atrophy, or limb deformity.

Discussion

Vasculogenesis and angiogenesis begin early in the third week of fetal development. In vasculogenesis, there is de novo formation of endothelial-lined cavities that fuse to form vascular channels. From these channels, new vessels form via budding and spread into the adjacent mesenchyme, a process called angiogenesis. These processes are regulated by vascular endothelial growth factor (VEGF) and other growth factors. The abdominal aorta is formed in the fourth week by fusion of the paired dorsal aorta (1). There are four paired dorsal segmental arteries of the abdominal aorta, which form the upper lumbar arteries. The fifth lumbar arteries give rise to the bilateral common iliac arteries at the level of the fourth lumbar vertebra. The median sacral artery persists as a remnant of the dorsal aorta.

In comparison to the thoracic aorta, the abdominal aorta is infrequently associated with congenital abnormalities. The incidence of these anomalies is unknown, but in a report of over eight thousand symptomatic patients who underwent angiography, only six patients were noted to have aberrations of the iliofemoral system (2). Reported anomalies of the distal aorta and iliofemoral system include unilateral absence of an external iliac artery (3-7), unilateral aplasia of the common iliac artery (8), congenital absence of the bilateral internal iliac arteries (9), and bilateral aplasia of the external iliac arteries (10). However, absence of both common iliac arteries has been described only twice in the literature, and only at the time of autopsy. The two postmortem case reports described termination of the distal aorta into four branches, two internal and two external iliac arteries (11-12). Neither case reported associated anomalies of the venous system. Our case presents a unique variant, to our knowledge not yet reported, in a living patient who presented for unrelated concerns.

Tamisier et al grouped congenital malformations of the external iliac artery into three categories (13). The first category included anomalies of origin or course of the artery. The second group comprised hypoplastic or atretic arteries with compensation by a persistent sciatic artery (PSA). The last category encompassed hypoplastic or atretic arteries without compensation by a PSA, leading to limb ischemia and claudication.

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When abnormalities of the distal aorta occur, an extensive collateral network often forms. With occlusion of the common iliac artery, two types of collateral networks develop (14). The first group, the parietal intersystemic pathway, provides flow to the external and internal iliac arteries via the internal thoracic artery, superior and inferior epigastric arteries, fourth lumbar artery, and iliolumbar artery. The second collection of collaterals is termed the visceral intersystemic pathway. This system supplies the internal iliac artery through branches of mesenteric arteries and the hemorrhoidal plexus. Our patient demonstrated collateralization through lumbar and iliolumbar arteries, the median sacral artery, and the superior hemorrhoidal artery.

Although aberrations of the iliac arterial system are uncommon, some authors have suggested that absence of these arteries should be considered in the setting of intermittent claudication. In patients with symptoms of chronic occlusive disease, deliberate evaluation of the collaterals is necessary to prevent their surgical interruption and associated ischemic complications. Disruption of this collateral network can occur, for example, during anterior abdominal wall instrumentation, use of flaps in reconstructive surgery, or lumbosacral orthopedic surgery. Additionally, the internal thoracic artery may be diverted during coronary artery bypass surgery.

Discovery of anomalies of the iliac arterial system is increasing with the expanded use of vascular imaging and intervention. Our patient's heretofore undescribed anomaly was discovered incidentally, but may have been detected with a more comprehensive vascular investigation as part of a workup of her longstanding symptoms. We present this case for its rarity and to highlight the importance of considering such vascular anomalies, especially before surgical or transcatheter interventions.

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