Congenital absence of left common and external iliac arteries

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ABSTRACT

Congenital atresia of the common and external iliac arteries is an extremely rare vascular anomaly, although often associated with limb ischemia and genitourinary malformations. We have presented a rare case of the congenital absence of the left common and external iliac arteries, with no limb ischemic symptoms or organ anomalies present. (J Vasc Surg Cases Innov Tech 2022;8:16-8.)

Keywords: Congenital anomaly; Iliac artery; Persistent sciatic artery

Congenital vascular malformation of the iliofemoral arteries is less common than that of the thoracic and abdominal aorta and is usually discovered incidentally or because of chronic lower limb ischemia.[1] Greebe[2] identified no more than six cases by angiography in a series of 8000 patients who had had symptoms related to limb ischemia, including intermittent claudication or leg pain. Furthermore, few reports of the complete absence of a common iliac artery have been presented. We have described a very rare case of the congenital absence of the unilateral common iliac artery. Our patient provided written informed consent for the report of her case and imaging studies.

CASE REPORT

A 44-year-old woman with no significant medical history, including no tobacco use or previous trauma, had been referred to our department for examination after a low ankle brachial index (ABI) was observed in the findings obtained as a part of a comprehensive health checkup. She had no symptoms related to limb ischemia, and the patient had reported no history of lower extremity pain nor any limitations to activities such as running or prolonged walking because of pain or fatigue. The left and right side ABI was 0.84 and 1.24, respectively. The left and right toe brachial index was 0.69 and 1.09, respectively. The left femoral pulse and dorsalis pedis pulse were both palpable, although weaker than those on the right side. The circumference and length of the lower extremities on both sides were equal. Blood tests showed that the white blood cell count and C-reactive protein level were within normal limits. Enhanced computed tomography was performed, which revealed the complete absence of the left common iliac artery (CIA) and external iliac artery (EIA; Fig, *A* and *B*). Also, the median sacral artery reconstituted the left internal iliac artery, and the left internal iliac artery supplied most of the flow to the left femoral artery (FA; Fig, *C*). Moreover, the area of stenosis in the left popliteal artery was >50%. The left anterior tibial artery and peroneal artery were less visible than those on the right, and a collateral vessel from the left superficial FA to posterior tibial artery was well developed (Fig, *C* and *D*). The abdominal organs were normal. The patient had no ischemic symptoms; thus, we decided to perform follow-up examinations based on the symptoms. During a 6-year observation period, the ABI and TBI remained stable, and no ischemic symptoms were noted.

DISCUSSION

In the early embryo, the CIAs result from the fifth lumbar arteries at the level of the fourth lumbar vertebra. Next, the EIA arises from the CIA and bifurcates into the inferior epigastric artery and FA. The FA annexes the foot plexus of the sciatic artery and its origin, and the distal parts of the sciatic stem are appropriated by the FA near its origin from the EIA, giving rise to the anterior tibial artery, which connects with the planter arch distally.[3] When considering this developmental process of the lower extremity arteries, it is reasonable that a persistent sciatic artery (PSA) often provides the blood supply to the lower extremity as a collateral vessel in such cases of the congenital absence of the CIA or EIA.

A search found only 12 cases of congenital CIA absence reported from 1964 to 2021 (Table).[1,4-14] The congenital absence of the CIA had been diagnosed incidentally for nearly all those patients, and many had had no history of limb ischemic symptoms. This was probably because the collateral vessels develop well in the embryo and

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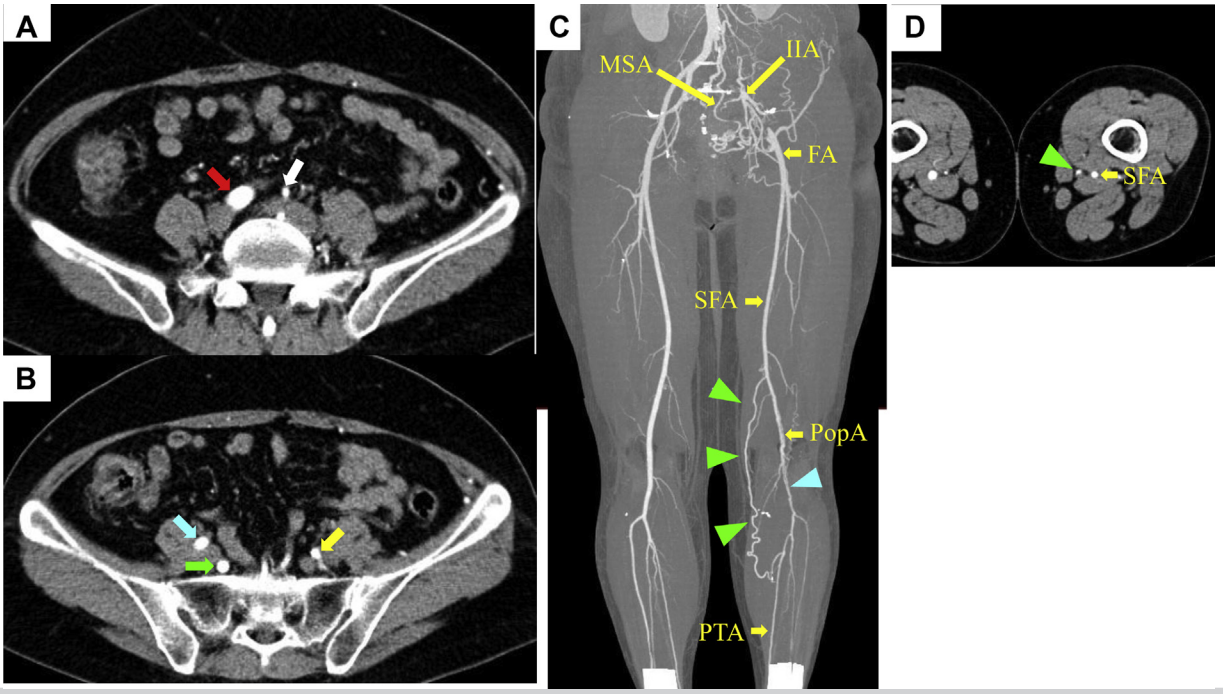

Fig. Enhanced computed tomography (*CT*) scans. **A,** Axial view showing the right common iliac artery (CIA; *red arrow*), inferior mesenteric artery (*white arrow*), and absence of left CIA. **B,** Axial view showing the right external iliac artery (EIA; *blue arrow*), right internal iliac artery (*IIA*; *green arrow*), left IIA (*yellow arrow*), and absence of the left EIA. **C,** CT angiography, coronal view, showing that the median sacral artery (*MSA*) reconstituted the left IIA and that the left IIA supplied most of the flow into the left femoral artery (FA). Stenosis was present in the left popliteal artery (*PopA*; *blue arrow*). The left anterior tibial and peroneal arteries were less visible than those on the right, and a collateral vessel (*yellow arrow*) was well developed from the left superficial FA (*SFA*) to the posterior tibial artery (*PTA*). **D,** Enhanced CT, axial view, showing the collateral vessel (*green arrowhead*) came from the SFA.

provide the blood supply to the lower extremity in cases of congenital iliofemoral atresia. Therefore, most of the patients were followed up without intervention, although one patient had undergone bypass surgery because of progressively worsening intermittent claudication (Table).[5] Two cases of common iliac atresia with a genitourinary malformation have also been reported (Table). The development of the kidneys begins in the pelvis, after which the organs migrate cranially to their final position on the posterior abdominal wall. The pelvic kidneys derive their arterial blood supply from the iliac system, and complex genitourinary malformations have been associated with iliofemoral anomalies.[8]

Congenital malformations of the EIA can be classified into three types: anomalies related to the origin or course of the artery; hypoplasia or atresia with compensation by a PSA; and isolated hypoplasia or atresia, which will result in chronic ischemia of the lower limb.[15] In cases with a PSA congenital malformation, the prevalence of aneurysms and arteriosclerosis has been high.[15,16] In EIA aplasia or hypoplasia cases, limb ischemia or claudication will often occur; thus, care must be taken regarding their possible presence.

When an iliofemoral anomaly is observed, it is essential to inform the patient and also to confirm whether other organ anomalies are present. Ischemic symptoms are likely to appear if the collateral circulation is damaged; thus, a careful preoperative assessment is required before performing surgery or a catheter-based intervention. Moreover, when limb ischemia occurs or if ischemic symptoms become progressively worse, surgical intervention such as a bypass procedure should be considered.

CONCLUSIONS

A CIA or EIA anomaly is often associated with limb ischemia or genitourinary malformations; thus, care must be taken in such cases.

Table. Common iliac atresia cases reported[a]

Investigator	Age, years; gender	Laterality	Other arterial anomalies	Ischemic symptoms	Other organ anomalies	Treatment	Diagnostic modality
Mansfield et al,[4] 1964	Unknown	B	Unknown	Unknown	Unknown	Unknown	Unknown
Dumanian et al,[5] 1965	44; M	L	L EIA, IIA, CFA atresia	Yes	No	Ao-FA bypass	Survey for intermitted claudication
Oduro et al,[6] 1992	Unknown	L	L EIA arose from RA	LYes	Unknown	Unknown	Survey for intermitted claudication
Llauger et al,[7] 1995	32; M	R	No	No	No	No	Found incidentally
Donnette et al,[8] 2015	21; F	R	No	No	No	No	Found incidentally
Patel et al,[9] 2013	Neonate; M	L	L EIA atresia	Yes	No	No	Survey for limb ischemic symptoms
Christopher et al,[10] 2015	25; F	B	L IIA atresia	Yes	No	No	Found incidentally
Clifton et al,[1] 2015	24; M	R	R EIA atresia	No	VUR	No	Preoperative scan
Radhakrishnan et al,[11] 2015	34; M	R	R EIA, IIA atresia	No	No	No	During surgery
Palkhi et al,[12] 2015	28; F	L	L IIA atresia	No	VUR	No	During surgery
Pham et al,[13] 2021	65; M	B	No	No	No	No	Preoperative scan
George et al,[14] 2021	66; M	B	No	Yes	No	No	Preoperative scan
Present patient, 2021	44; F	L	No	No	No	No	Found incidentally

Ao, Aorta; *B,* bilateral; *CFA,* common femoral artery; *EIA,* external iliac artery; *F,* female; *FA,* femoral artery; *IIA,* internal iliac artery; *L,* left; *M,* male; *R,* right; *RA,* renal artery; *VUR,* vesicoureteral reflux.
[a]A search found only 12 cases of congenital common iliac atresia reported from 1964 to 2021, nearly all of which had been found incidentally. One patient had undergone bypass surgery to treat progressively worsening intermittent claudication. Two cases were associated with genitourinary malformations.

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