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CASE REPORT

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Unveiling the uncommon: hypoplasia of external iliac artery–a case report and literature review

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Abstract

The absence or dysplasia of the iliac artery(IA) is an exceedingly rare condition, with limited cases documented in the literature. In this report, we present a case of hypoplasia of the right external iliac artery (EIA) in a 69-year-old male patient. The patient presented with right lower abdominal pain attributed to an aneurysm of the right internal iliac artery (IIA), yet notably, there was no evidence of lower limb ischemia at the time of consultation. Computed tomography angiography (CTA) of the aorta revealed a slender and occluded right EIA. Additionally, aneurysms were identified in the abdominal aorta (AA), the common iliac artery (CIA), and the right IIA, with collateral circulation involving the deep femoral artery and internal pathways. We performed aortoiliac aneurysm repair with a bifurcated synthetic graft on the patient's AA and iliac artery (IA), successfully excising the abdominal aorta aneurysms (AAA) and the CIA aneurysm. Postoperatively, thrombosis of the internal iliac aneurysm was observed, and the patient experienced a resolution of pain symptoms in the right lower abdomen. This paper delineates the vascular variations and treatment strategies employed and provides a review of the existing literature on IA malformations.

Keywords External iliac artery atresia, Aneurysm, Obturator artery, Corona mortis, Artificial vascular replacement

Introduction

The occurrence of vascular variations in the IA is uncommon, and the precise incidence rate remains undetermined. Notably, the study conducted by Greebe J et al. identified six cases of iliofemoral artery abnormalities among a cohort of 8,000 patients [1]. Variations in the IA can manifest in a range of clinical symptoms or, in some instances, may be asymptomatic. The literature on IA variations is predominantly extensive yet limited in scope, with cases of isolated EIA dysplasia being particularly uncommon. In 2003, Teruyuki Koyama and colleagues documented a case of hypoplasia of the left EIA, which was identified following the exacerbation of ischemia in the left lower limb. Notably, the collateral circulation was not well-defined prior to surgical intervention [2]. We present a case study involving an aneurysm of the AA and IIA, attributed to the hypoplasia of the right EIA. CTA distinctly illustrates the source of blood supply to the patient's right femoral artery.

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Case report

A 69-year-old male patient presented with a two-month history of persistent pain in the right lower abdomen, with no accompanying symptoms of intermittent claudication, lower limb pain, or numbness. His medical history is notable for smoking, alcohol consumption, and hypertension, with no reported history of hyperlipidemia or diabetes. The patient initially underwent a computed tomography (CT) examination at a local hospital. Due to the presence of a right iliac aneurysm, the patient sought treatment at our facility. Upon admission, a comprehensive physical examination was conducted, revealing normal pulsation in the bilateral femoral arteries. The left dorsalis pedis artery exhibited normal pulsation, whereas the right dorsalis pedis artery demonstrated weak pulsation. Blood pressure measurements indicated a reading of 170/102 mmHg in the left lower limb and 102/63 mmHg in the right lower limb. Subsequently, a CTA of the aorta was performed. The examination revealed the presence of aneurysms in the patient's AA (36.1 mm) and right IIA (40.1 mm). Additionally, the right EIA was found to be slender, calcified, and occluded, while the diameter of bilateral common iliac arteries is enlarged (left:18.3 mm, right:19.8 mm). Furthermore, the ectatic right deep femoral artery(19.8 mm)was observed to communicate with the right IIA via the obturator artery and corona mortis. The right common femoral artery (CFA) was noted to have a connection with the right inferior epigastric artery. Abdominal pain is attributed to the fact that the abdominal aneurysm was symptomatic. Since an AAA becomes symptomatic, impending rupture is a concern despite the aneurysm size, and repair is warranted as soon as possible under optimal conditions (ideally during working hours) after a rapid assessment and optimization [3]. Given the association between aneurysm formation and the occlusion of the right EIA, and considering the slender nature of the EIA which renders it unsuitable for endovascular treatment, we have undertaken the replacement of the AA and bilateral IA with artificial blood vessels (ePTFE, 18×9 mm in diameter). The aortic and iliac aneurysm were opened, the AA was cut off below the renal arteries and a Y-shaped synthetic graft was placed. The right distal anastomosis was performed with the right CFA and at the left side with the bifurcation of the

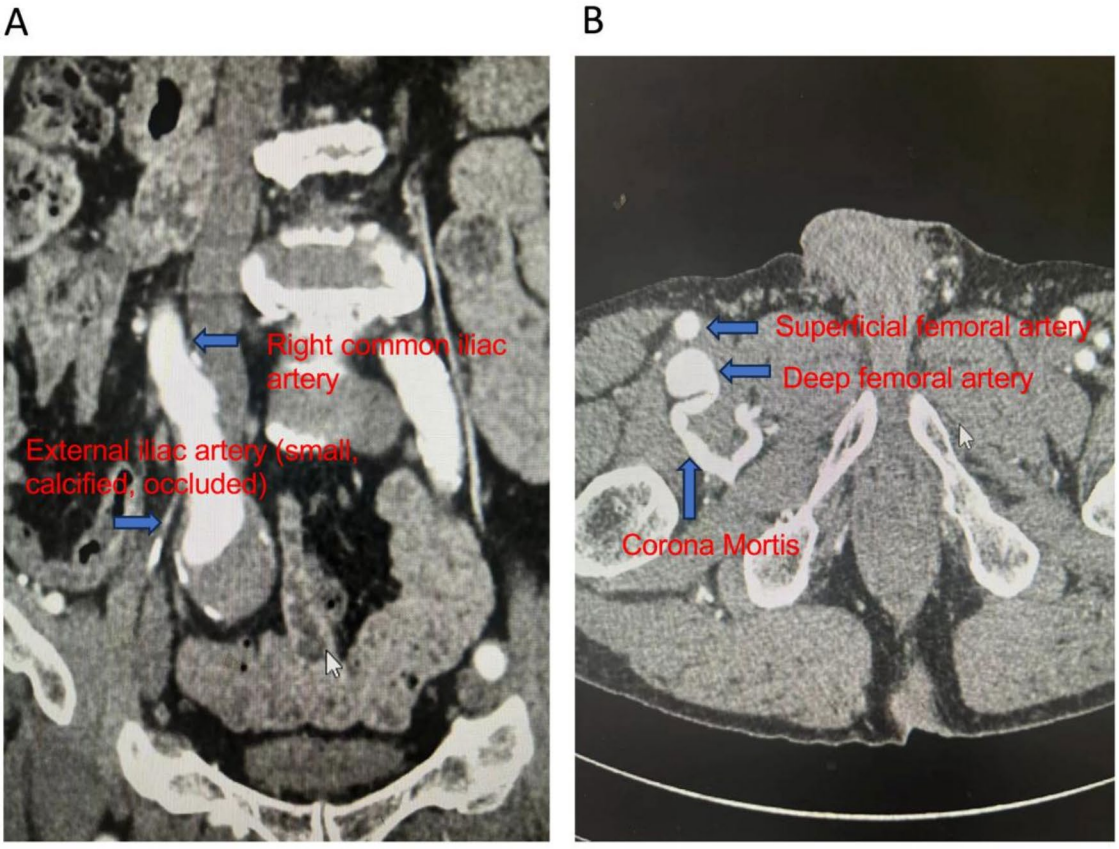

Fig. 1 **A**. Preoperative CTA showed that the right EIA was slender, calcified and occluded. **B**. Collateral circulation involving the enlarged profunda femoris artery is also depicted

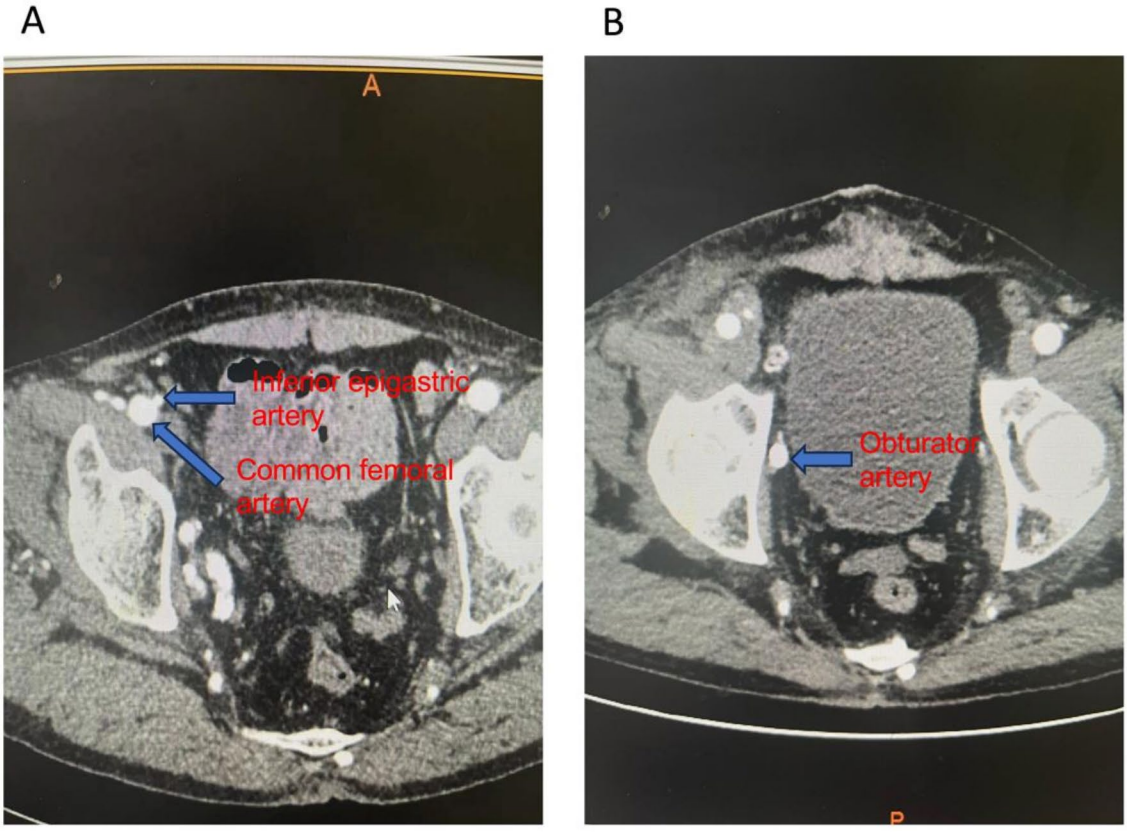

Fig. 2 **A**. Preoperative CTA indicates communication with CFA and inferior epigastric artery. **B**. Preoperative CTA showed the obturator artery supplying blood to the deep femoral artery through Coronas Mortis

left CIA. We tied off the opening of the right IIA and the proximal end of the CFA. To prevent ischemia of the right IIA, the communicating vessels between the IIA and the deep femoral artery are preserved. Following the surgical procedure, the patient's symptoms of abdominal pain resolved, and a CTA of the aorta was conducted on the sixth postoperative day. The graft was patent; there was no anastomotic stenosis, and thrombosis in the right internal iliac aneurysm was confirmed (Figs. 1-4).

Literature review

The CIA originates at the level of the fourth lumbar vertebra and subsequently bifurcates into the external and internal iliac arteries approximately 5 cm distally. There is a paucity of literature regarding variations in the IA, particularly concerning the dysplasia of the EIA. The embryonic development of the IA has been documented in the relevant literature [4]. IA variations are predominantly documented through case reports. A review of the existing literature reveals the following classification of IA variations. The primary type of IA malformation involves an abnormal origin or trajectory. Two reports describe cases where the CIA is absent, resulting in both the bilateral external and internal iliac arteries originating directly from the AA. This scenario represents a variation in the orifice of the IIA [5, 6]. The second deformity of the IA is its absence. Tsutomu Doita et al. reported a case in which the left CIA and the EIA were absent, as determined through physical examination. In this patient, the femoral artery received its blood supply from the median sacral artery [7]. In 1992, a case was documented involving the congenital absence of the left EIA and IIA. The left EIA was anomalously connected to the left kidney, resulting in ischemia of the left lower limb following nephrectomy [8]. This case further indicates a potential association between IA malformation and urinary system malformation. Additionally, several other studies have documented instances of absent IA, wherein collateral circulation is facilitated through alternative pelvic vessels [9, 10]. The third type of deformity affecting the IA is characterized by dysplasia or atresia. In 1965, a case of IA dysplasia was documented. Similar to our case, this patient exhibited slender and occluded blood vessels; however, this patient

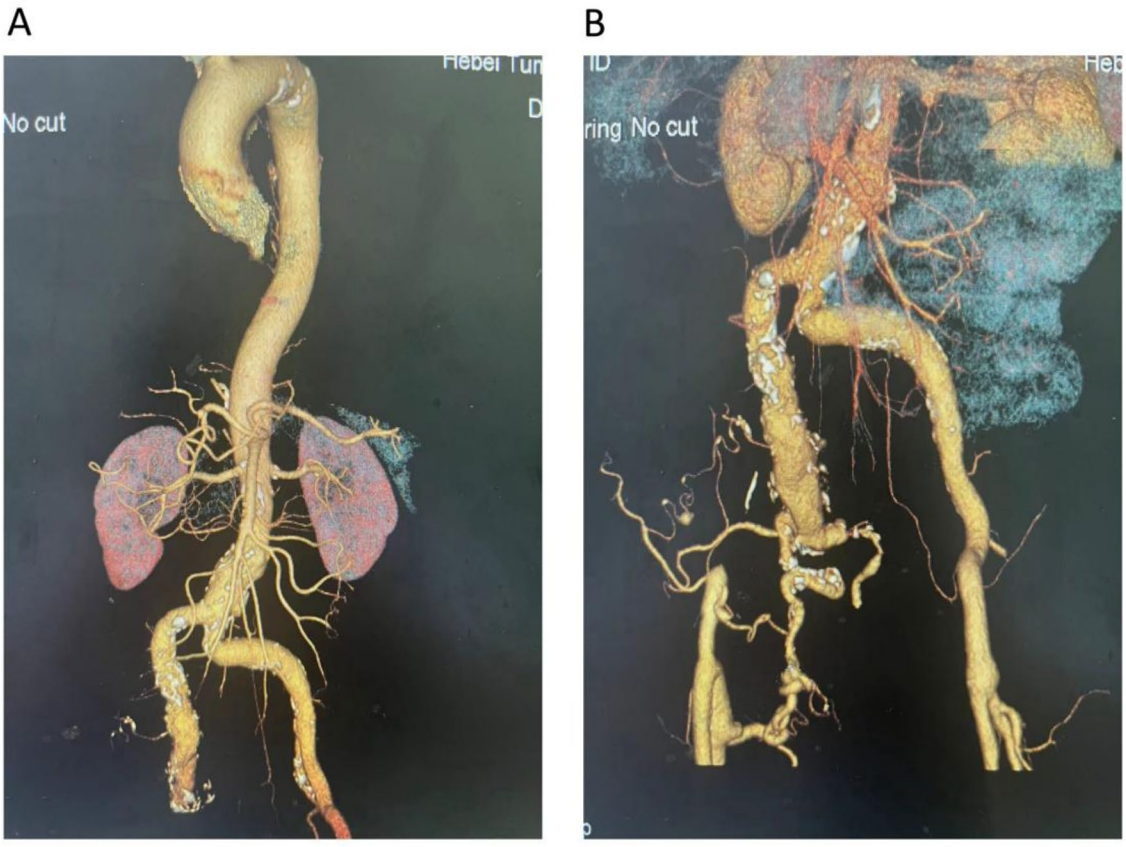

Fig. 3 **A**. Preoperative CTA three-dimensional reconstruction of blood vessels revealed AAA and right internal iliac aneurysm. **B**. Preoperative CTA three-dimensional reconstruction of blood vessels showed that the diameter of deep femoral artery increased, and the deep femoral artery communicated with IIA

also presented with symptoms of lower limb ischemia [11].

The variation of the simple EIA is infrequently documented in the literature, and limited resources are available for investigation. In 2022, a case was reported involving a high bifurcation position of the deep femoral artery [12], but this does not affect the blood supply. A further case involves a child aged 18 months who presents with bilateral absence of the EIA. However, there is significant collateral vascular reconstruction observed above the level of the popliteal arteries. The IIA and its branches appear normal. The iliofemoral branch is visible originating from the superior mesenteric artery, inferior mesenteric artery, renal artery, CIA, and IIA. Notably, bilateral deep femoral arteries and sciatic arteries are absent [13]. A 37-year-old man seems to be like the case we provided. CT showed that the right CFA communicated with the dilated IIA, but no EIA was observed [14]. However, the imaging data of this patient is limited, and it seems that no collateral circulation has been established, and the EIA and IIA cannot be identified. Table 1 presents various characteristics of cases documented in the literature.

In conclusion, variations in the IA are infrequent. While they can be broadly categorized, the specific characteristics of these variations and the formation of collateral circulation differ in each case. Consequently, these variations may result in diverse symptoms, necessitating the selection of treatment methods tailored to the individual circumstances of each patient.

Discussion

Although variations in the IA are uncommon, there have been documented cases over an extended period. These reports provide insights into the characteristics of such cases, warranting consideration of the following three points. Firstly, the variation characteristics of the IA are noteworthy. In this particular case, computed tomography angiography (CTA) revealed the presence of the EIA; however, it was occluded, with no blood flow traversing it. Additionally, the entire segment of the EIA was observed to be slender with localized calcification,

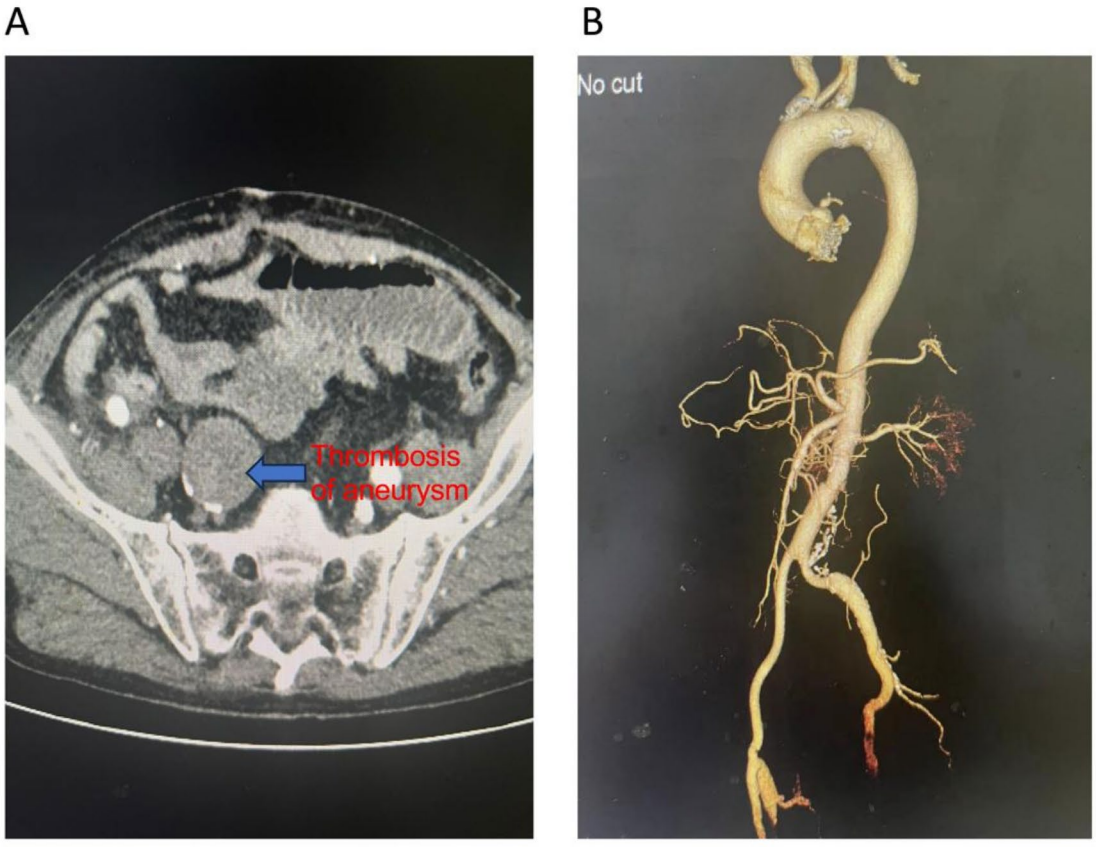

Fig. 4 **A**. Three-dimensional reconstruction of CTA after operation indicates thrombosis of right internal iliac aneurysm. **B**. CTA three-dimensional reconstruction of blood vessels after operation

and there was an absence of intraluminal thrombosis or fibromuscular dysplasia. The patient's right EIA is suspected to have congenital dysplasia, with its early-stage patency remaining uncertain. There have been several reported cases of congenital atresia of the EIA [15, 16], however, there is no more research on incidence statistics

Table 1 Characteristics of cases in references

Investigator	Type of malformation	Gender	Age	Aneurysm	Ischemic symptoms	Other organ anomalies
Teruyuki K et al. [2]	Hypoplasia of left EIA	Male	51	No	Acute ischemic pain in the left leg	No
George JM et al.[4]	Abnormal origin or abnormal path	Male	66	No	Ischemic left foot wound	No
Mansfield AO et al. [5]	Abnormal origin or abnormal path	Female	36	No	Unknow	Unknow
Tsutomu Doita et al. [6]	Absence of IA	Female	44	No	No	No
Oduro GD et al. [7]	Absence of IA	Male	20	No	No	No
Llauger J, et al. [8]	Absence of IA	Male	35	No	No	No
Dabydeen DA et al. [9]	Absence of IA	Female	21	No	No	No
Dumanian AV et al. [10]	IA dysplasia	Male	44	No	Claudication	No
Natsis K et al. [11]	High bifurcation position of deep femoral artery	Female	75	No	Unknow	Unknow
Harikrishnan S et al. [12]	Bilateral aplasia of EIA	Female	6	No	Claudication	Cardiac murmur
Pua U et al. [13]	Congenital anomaly of the EIA	male	37	No	No	No
Howard JM et al. [14]	Congenital Atresia of the EIA	Male	27	No	Claudication	No
Ezzone A et al. [18]	Congenital atresia of the right EIA	Female	38	No	Pain of lower limb	Tetralogy of Fallot

and pathogenesis. Second, the characteristics of collateral circulation in patients exhibiting IA variation are of significant interest. It is widely accepted that abnormalities at the terminal portion of the aorta typically result in the formation of extensive collateral circulation [17]. In the cases we presented, there was limited collateral circulation; however, the IIA supplied blood to the deep femoral artery via its branches. This observation suggests that the occlusion of the EIA may not be chronic. We hypothesize that the branch of the IIA involved is the obturator artery, with the Corona Mortis serving as the anastomosis between the deep femoral artery and the obturator artery. The Corona Mortis is characterized as the vascular connection between the obturator artery and the EIA system [18]. Corona Mortis was not connected to the EIA or inferior epigastric artery in this case, and it was directly connected to the deep femoral artery. In this case, despite the differing characteristics of collateral circulation compared to those of the Corona Mortis, their functions remain analogous. In addition, a typical collateral circulation (cruciate anastomosis) in EIA occlusion is also worthy of attention. The route of blood is through the IIA, to the inferior gluteal artery, to a perforating branch of the deep femoral artery, to the lateral circumflex femoral artery, then to its descending branch into the superior lateral genicular artery and finally into the popliteal artery [19]. Third, these patients typically exhibit no symptoms of lower limb ischemia; however, the development of collateral circulation is uncommon. In the context of pelvic and orthopedic surgeries, there is a heightened risk of compromising the blood supply from the deep femoral artery, potentially resulting in lower limb ischemia, thereby necessitating careful consideration. Additionally, an aneurysm may develop at the proximal end of the occluded artery. Fourth, a comprehensive assessment of the patient was conducted using whole-body computed tomography (CT), echocardiography, and deep vein ultrasound of the lower limbs. In conjunction with a thorough physical examination, these diagnostic procedures revealed no additional abnormalities. It is noteworthy that patients of this nature should be evaluated for potential urinary system malformations. Furthermore, there are documented cases of EIA atresia associated with congenital heart disease, specifically tetralogy of Fallot [20]. In summary, it is imperative for these patients to monitor for the presence of additional malformations.

Conclusion

Simple EIA atresia represents an uncommon form of IA malformation, yet it can result in the development of proximal vascular aneurysms. In such cases, the collateral circulation of the lower extremity arteries may be sustained by a single vessel, often without manifesting symptoms of lower extremity ischemia. It is crucial to consider the characteristics of the collateral circulation and assess for any associated malformations. The replacement of the affected artery with an artificial blood vessel has proven to be an effective treatment modality for patients with this condition.

Author contributions
Chao Li made substantial contributions to the conception. Yu Xu had drafted the work; Yuan Jingbin provided the case.

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Data availability
No datasets were generated or analysed during the current study.

Declarations

Consent for publication
Written informed consent for publication was obtained from all participants.

Competing interests
The authors declare no competing interests.

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