Case Report

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Ilio-Iliac Arteriovenous Fistula with May-Thurner Syndrome: A Case Report

May-Thurner 증후군과 동반된 장골동맥-장골정맥루: 증례 보고

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An ilio-iliac arteriovenous fistula (AVF) is rare. Common factors leading to ilio-iliac AVF include congenital malformations, iatrogeny, and trauma. There is limited documentation in the literature of cases involving ilio-iliac AVF with May-Thurner syndrome. Here, we present a case of an ilio-iliac AVF with May-Thurner syndrome in an 80-year-old male. CT and angiography confirmed extensive ilio-iliac AVF. Successful endovascular procedures for ilio-iliac AVF were performed using several variable-sized coils and 1400–2000 μm gelatin particles. After embolization, follow-up abdominopelvic CT revealed an improvement in edema in the left leg.

Index terms Arteriovenous Fistula; May-Thurner Syndrome; Endovascular Procedures

INTRODUCTION

Ilio-iliac arteriovenous fistula (AVF) is an extremely rare disease, with only a few cases reported in the literature. To the best of our knowledge, no cases of ilio-iliac AVF have been reported in Korean radiology journals. Well-known causes of ilio-iliac AVF are congenital malformations, iatrogeny, and trauma (1).

May-Thurner syndrome commonly refers to the compression of the left common iliac vein by the right common iliac artery, with or without venous thrombosis (2). Only a few studies have reported a relationship between May-Thurner syndrome and ilio-iliac AVF (3-7). Herein, we report a case of an ilio-iliac AVF with May-Thurner syndrome that was successfully treat-

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ed with endovascular embolization.

CASE REPORT

An 80-year-old male patient presented with swelling and pain in the left leg. He was admitted with cellulitis in the right arm. The patient had a history of hypertension, diabetes mellitus, and cerebral infarction. Laboratory tests revealed a slight increase in D-dimer levels to 1.81 mg/L. Abdominopelvic CT with a deep venous thrombosis protocol was performed, and compression of the left proximal common iliac vein by the right common iliac artery with atrophic changes in the left femoral vein and multiple collateral veins were observed.

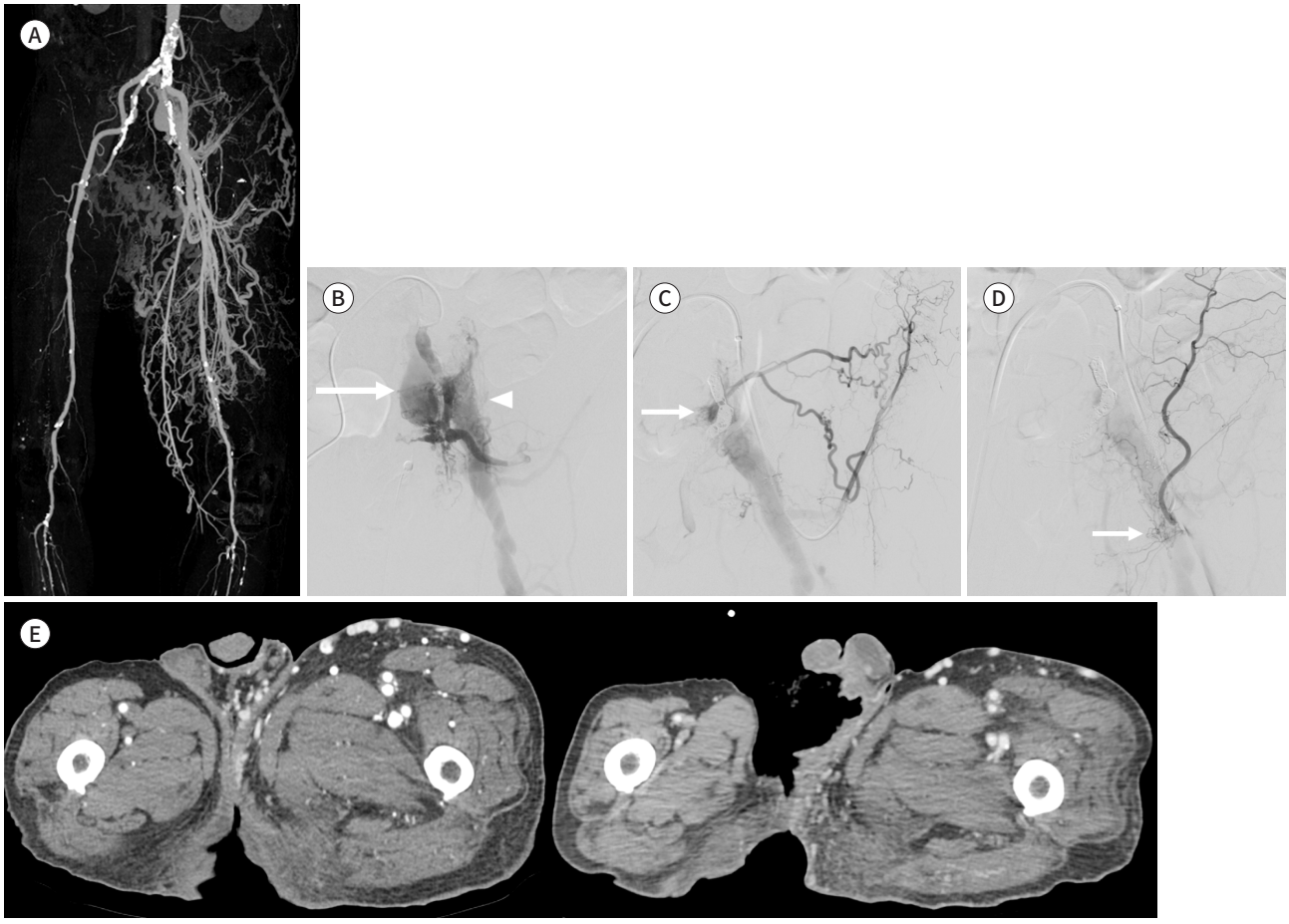
After the abdominopelvic CT, metallic stenting of the left proximal common iliac vein was performed. Therefore, preoperative left extremity venography was performed, which revealed strong reverse flow in the left common femoral vein with drainage through the right-sided veins. We suspected vascular malformations, such as AVF, and performed lower-extremity CT angiography. CT angiography confirmed the diagnosis of ilio-iliac AVF, with findings of early draining in the left iliac and femoral veins with extensive collaterals (Fig. 1A).

Embolization of the ilio-iliac AVF was performed using several variable-sized coils (Interlock coils, Boston Scientific, Marlborough, MA, USA) and 1400–2000 μm gelatin particles (EG-gel, Engain, Seongnam, South Korea). First, the right common femoral artery was punctured under local anesthesia. Left internal iliac artery angiography was performed using 6 French (Fr) vascular sheaths and 5 Fr catheters (Cobra 2, Cook Medical, Bloomington, USA). Angiography revealed numerous AVFs arising from the left internal iliac artery (Fig. 1B). Therefore, we performed main trunk embolization of the left internal iliac artery using a microcatheter (Renegade, Boston Scientific, MA, USA) and Interlock microcoils of 10 mm × 30 cm (four coils), 12 mm × 30 cm (two coils), 14 mm × 20 cm (two coils), and 14 mm × 30 cm (five coils). After embolization of the main trunk of the left internal iliac artery, angiography revealed collateral vessels arising from the left circumflex iliac artery supplying the incompletely packed main trunk of the left internal iliac artery (Fig. 1C). We embolized the collateral vessels of the left circumflex iliac artery using Interlock microcoils of various sizes, including 2 mm × 4 cm (two coils), 3 mm × 6 cm, 5 mm × 8 cm, 5 mm × 15 cm (two coils), and 6 mm × 20 cm (two coils), along with a 1400–2000 μm gelatin particle (EG-gel, Engain). Multiple AVFs originating from the left inferior epigastric artery were observed on a left inferior epigastric artery angiogram (Fig. 1D). Therefore, we embolized the AVFs of the left inferior epigastric artery using a 4 mm × 8 cm interlock microcoil and 1400–2000 μm gelatin particles (EG-gel, Engain).

We could not embolize all AVFs because of the presence of numerous miniscule AVFs. On the final angiogram, many AVFs were embolized; however, residual ilio-iliac AVFs remained. No immediate complications were observed.

After the procedure, left common iliac vein stenting was scheduled for the May-Thurner syndrome; however, the patient refused to undergo the procedure. After two months, the patient was readmitted, and significant improvement in the swelling of the left leg was observed. In addition, improvement in the left leg edema was evident on a follow-up abdominopelvic CT scan after two months (Fig. 1E).

Fig. 1. Ilio-iliac arteriovenous fistula with May-Thurner syndrome in an 80-year-old male.
A. CT angiography MIP image reveals early draining of the left iliac and femoral veins, with extensive AVFs arising from the left internal iliac artery.
B. Left internal iliac artery angiogram. A few seconds after contrast administration, early draining of the left internal iliac vein (arrow) and left external iliac vein (arrowhead) is observed, indicating an ilio-iliac AVF.
C. Collateral vessels arising from the left circumflex iliac artery supplying the incompletely packed main trunk (arrow) of the left internal iliac artery are observed.
D. Left inferior epigastric artery angiogram. An angiogram showing multiple AVFs (arrow) originating from the left inferior epigastric artery.
E. Pre-embolization CT scan (left) and post embolization CT scan (right, two months later). Note improvement of left leg edema on post embolization CT scan.
AVF = arteriovenous fistula, MIP = maximum intensity projection



DISCUSSION

Most reported cases of ilio-iliac AVF are associated with congenital defects or iatrogenic trauma.

In our case, as there was no patient history of surgery, iatrogenic trauma, or aneurysm, the definitive cause of the ilio-iliac AVF could not be determined. Notably, our patient had May-Thurner syndrome, a well-known cause of compression of the left common iliac vein via the right common iliac artery, with or without deep venous thrombosis (DVT). Although cases of AVF with May-Thurner syndrome are rarely documented, some researchers have reported the occurrence of spontaneous AVF following DVT (6-9). Considering that May-Thurner syndrome

can occur in combination with DVT, there may be a correlation between AVF and May-Thurner syndrome. However, the etiologies of these diseases remain unknown. Furthermore, Che et al. (5) reported three cases similar to ours over a period of two years. Therefore, we suspect that ilio-iliac AVF with May-Thurner syndrome might not be infrequent but rather neglected.

Endovascular treatment can be the first-line therapy for ilio-iliac AVFs because it is safer and more efficient than open surgeries. A previous meta-analysis revealed that perioperative mortality was absent and technical success was achieved in 94% of cases, in contrast to a significantly higher mortality rate of 34% in open surgeries (10). In this case, because of the presence of numerous AVFs and a complex collateral system, we could not perform complete embolization of all visible AVFs. Despite this, we occluded many major and fine AVFs and observed improvements in patient symptoms, such as swelling and pain.

Our single-case review showed that in cases of ilio-iliac AVF with uncertain etiologies, May-Thurner syndrome can be considered a possibility, although its definite etiology remains largely unknown. Furthermore, despite experiencing residual ilio-iliac AVFs after embolization, the patient showed improvement in symptoms. Therefore, in the case of ilio-iliac AVF, endovascular treatment is safe and efficient, with promising outcomes.

Author Contributions

Supervision, Y.J.W., K.H.J., J.S.K.; writing—original draft, K.T.H.; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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REFERENCES

1. Morrow C, Lewinstein C, Ben-Menachem Y. Spontaneous iliac arteriovenous fistula. *J Vasc Surg* 1987;6:524-527
2. Brinegar KN, Sheth RA, Khademhosseini A, Bautista J, Oklu R. Iliac vein compression syndrome: clinical, imaging and pathologic findings. *World J Radiol* 2015;7:375-381
3. Coelho A, Brandão P, Lobo M, Lojo I, Canedo A. Ilio-iliac arteriovenous fistulae-an unusual diagnosis with an even more unusual clinical presentation. *Ann Vasc Surg* 2018;50:298.e1-298.e5
4. Huynh N, Gates L, Scoutt L, Sumpio B, Sarac T, Ochoa Chaar CI. May-Thurner syndrome and iliac arteriovenous fistula in an elderly woman. *J Vasc Surg Cases Innov Tech* 2016;2:46-49
5. Che H, Li L, Song F, Yang M. Is May-Thurner syndrome combined with left iliac arteriovenous fistularare or neglected?: cases report and literature review. *Ann Vasc Surg* 2018;51:328.e13-328.e18
6. Link DP, Garza AS, Monsky W. Acquired peripheral arteriovenous malformations in patients with venous thrombosis: report of two cases. *J Vasc Interv Radiol* 2010;21:387-391
7. Mihmanli I, Kantarci F, Ulus SO, Bozlar U, Kadioglu A, Yildirim D. Lower extremity venous pathology mimicking deep vein thrombosis: 2 case reports. *Ultraschall Med* 2007;28:421-425
8. Labropoulos N, Bhatti AF, Amaral S, Leon L, Borge M, Rodriguez H, et al. Neovascularization in acute venous thrombosis. *J Vasc Surg* 2005;42:515-518
9. Ziello JE, Jovin IS, Huang Y. Hypoxia-inducible factor (HIF)-1 regulatory pathway and its potential for therapeutic intervention in malignancy and ischemia. *Yale J Biol Med* 2007;80:51-60
10. Nakad G, AbiChedid G, Osman R. Endovascular treatment of major abdominal arteriovenous fistulas: a systematic review. *Vasc Endovascular Surg* 2014;48:388-395

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May-Thurner 증후군과 동반된 장골동맥-장골정맥루: 증례 보고

김태현 · 연재우* · 김혁중 · 장석기

장골동맥-장골정맥루는 드문 질환으로 일으키는 주요 요인으로는 선천성 이상 발생, 의인성, 외상 등이 있다. 그중에서도 May-Thurner 증후군과 동반된 장골동맥-장골정맥루에 대한 문헌은 매우 드물다. 이에 따라, 저자들은 80세 남성에서 발생한 May-Thurner 증후군과 동반된 장골동맥-장골정맥루의 증례를 보고하고자 한다. 컴퓨터단층촬영 및 혈관조영술을 통해 매우 넓은 범위의 장골동맥-장골정맥루를 확인하였고 이에 대해 다양한 크기의 색전용 코일 및 1400–2000 μm 크기의 젤라틴 입자를 사용하여 색전술을 시행하였다. 색전술 이후, 추적을 위해 시행한 복부-골반 컴퓨터단층촬영에서 왼쪽 다리의 부종이 개선됨을 확인하였다.

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