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Case Report

Spontaneous isolated iliac artery dissection in a young male: Case report and review of literature ☆☆☆

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ABSTRACT

Spontaneous isolated dissection of the iliac artery (SID-IA) is a rare vascular condition typically associated with trauma or connective tissue disorders. We present a unique case of a 39-year-old male without known risk factors or trauma history who presented with lower abdominal pain. Diagnostic imaging revealed SID-IA involving the right external iliac artery with thrombus formation. Despite negative findings for connective tissue disorders, the patient underwent successful endovascular stenting following initial medical management. Vigilance in diagnosis and prompt intervention are crucial in managing SID-IA to prevent complications such as limb ischemia and aneurysm formation. This case emphasizes the importance of considering SID-IA in young patients presenting with abdominal pain, even in the absence of traditional risk factors, and highlights evolving treatment options for this rare condition.

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Introduction

SID-IA is a rare and unusual medical condition. It is often associated with traumatic causes including trauma, [1] extreme physical activity, [2] pregnancy, [3] and iliac catheterization [4] as well as nontraumatic causes including Marfan syndrome, [5] Ehlers-Danlos syndrome, [6] fibromuscular dysplasia, [7,8] and cystic medial degeneration [9]. Rupture is the most frequent complication, occurring particularly in cases of collagen disorders, and can lead to fatal outcomes [8]. Most commonly, these dissections result in occlusion of the iliac artery, [10] leading to lower limb ischemia, and in some instances, the occlusive lesion may lead to thrombus formation and subsequent lower extremity microemboli [11]. While most reported cases are linked to connective tissue disorders, we present a unique instance of a 39-year-old man with SID-IA, without a known connective tissue disorder or history of trauma.

Case presentation

A 39-year-old healthy male presented with complaints of lower abdominal pain concentrated over the right groin area, radiating to the back, accompanied by vomiting, and 2 episodes of diarrhea. He had no comorbidities or history of trauma. On examination, the patient was afebrile, normotensive, with minimal tenderness detected in the right iliac fossa. No discoloration or edema was noted in the lower extremities. The patient's past medical history was unremarkable, with no significant family history. The patient's collagen connective tissue work-up was negative.

Initial laboratory investigations on day 5 revealed a prolonged partial thromboplastin time (PTT), weakly positive results in antinuclear antibody (ANA) immunoassay and positive c-antineutrophil cytoplasmic antibody (c-ANCA) along with a normal range of anti-cyclic citrullinated peptide (anti-CCP) antibodies (Table 1). Imaging via abdominal and pelvic ultrasound revealed dissection in the right external iliac artery with a thrombus in the right common iliac artery. Blood flow was seen in both lumens, with the true lumen appearing larger and the false lumen smaller. A CT scan of the chest, abdomen, and pelvis with angiography on day 6 indicated arterial dissection in the right external iliac artery mildly extending into the right common iliac artery and internal iliac artery (Figs. 1 and 2A). Additionally, it revealed an acute thrombus within the false lumen of the right proximal external iliac artery, causing a 70 percent occlusion (Fig. 3). A subsequent color Doppler study of the right lower limb on day 7 showed a 47 percent diameter reduction due to an increased throm-



Fig. 1 – Cross-sectional image of the computed tomography scan showing dissection of the right external iliac artery.

bus size compared to the prior 39 percent. Within the thrombus, an internal flap extended to the external iliac artery after the division of the common femoral artery into the superficial and deep femoral arteries (Figs. 4A and B). The veins of the right lower limb exhibited spontaneous color flow with normal phasic variation and no evidence of deep vein thrombosis was observed.

Differential diagnoses considered included causes of acute abdominal pain notably acute appendicitis, mesenteric ischemia, abdominal aortic aneurysm, and spontaneous isolated external iliac artery dissection. Given the patient's young age, absence of comorbidities, and the findings of arterial dissection and thrombus, a spontaneous isolated external iliac artery dissection was considered.

The patient was managed with initial high-dose dual antiplatelet therapy, statin, and injection heparin 5000 units stat followed by heparin 1000 units/HF intravenously. The patient was shifted to the intensive care unit (ICU) for further management. Despite being symptomatic, the dissection was not ruptured. Following evaluation, endovascular treatment was pursued, similar to managing a type B aortic dissection. Peripheral angioplasty was performed on day 7 through the left

Table 1 – Results of the laboratory investigations.

Laboratory investigation	Result	Normal range
Partial Thromboplastin Time (PTT)	>180 seconds	25-35 seconds
Anti-CCP antibodies	<0.50 U/mL	<0.50 U/mL
ANA immunoassay	Weakly positive	Negative
c-ANCA	Weakly positive	Negative

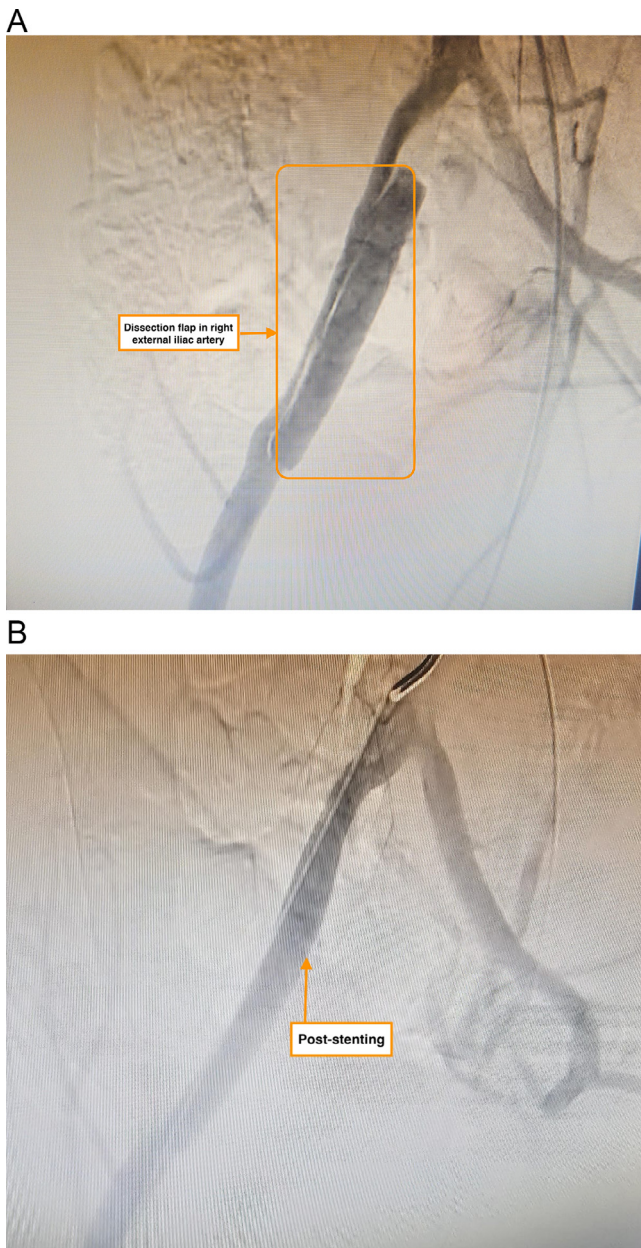


Fig. 2 – Angiography showing: (A) Arterial dissection in the right external iliac artery. (B) Favorable outcome poststenting.

femoral route, upgrading from a 6F to an 8F sheath. The true lumen was traversed using the fielder FC and subsequently swapped to an amplatz wire using a Rubicon catheter. Shoots were taken from above to confirm the placement of the stent via the right radial artery. Stenting of the right external iliac artery was performed using Covera plus 8.0 × 80 cm, with a favorable outcome observed on reshoot angiography (Fig. 2B). Postoperation, intravenous heparin INS was discontinued, and dual antiplatelet therapy with statins was initiated and continued.

The patient did not experience reperfusion compartment syndrome after the procedure and was discharged on the third postprocedural day with palpable peripheral pulsation.

He was prescribed a daily dose of 100 mg acetylsalicylic acid (aspirin) and scheduled for annual follow-up with a Doppler ultrasound check-up. Upon follow-up, as of the most recent examination, the patient remains hemodynamically and vitally stable, with no significant fluctuations in physical exam findings or laboratory parameters.

Discussion

SID-IA without involvement of the aorta, as seen in this case, is an extremely uncommon occurrence. Typically, most spontaneous cases are associated with connective tissue disorders or trauma. Clinical manifestations can vary, ranging from sudden onset acute ischemia to chronic, unilateral ischemia. Symptoms may also be transient [12,13].

The anterior course of iliac artery dissections often progresses to aneurysms of the common iliac artery, with the potential for local compression of nearby structures such as the urinary or digestive tract before rupture [13,14]. Rapid diagnosis and immediate treatment are crucial to prevent adverse outcomes. Careful assessment of limb color, temperature, and pulse is essential, with noninvasive methods such as Doppler studies aiding in diagnosis.

In Fig. 3, computed tomographic reconstruction revealed arterial dissection in the right external iliac artery, extending slightly into the right common iliac and internal iliac arteries. The presence of an acute thrombus within the false lumen, as shown in the angiography, caused approximately 70% occlusion of the artery. This finding highlights the potential severity of SID-IA, where the dissection can lead to significant luminal compromise and ischemic symptoms.

Fig. 4 further highlights the pathology by demonstrating the dissecting flap dividing the proximal external iliac artery into a true lumen and a false lumen. The echogenic thrombus within the false lumen, as highlighted in the Doppler scan, reinforces the risk of progressive ischemia and the potential for catastrophic outcomes if left untreated. This imaging clearly visualizes the dissected artery's structural changes and the thrombus formation within the false lumen, contributing to the patient's ischemic symptoms.

In this case, the patient had no history of arterial intervention, trauma, or excessive physical activity, ruling out iatrogenic and traumatic causes as the etiology of the dissection. Additionally, there were no signs or symptoms suggestive of connective tissue disorders, and both pathologic and immunologic examinations were nondiagnostic.

Various strategies have been proposed for managing SID-IA, including conservative therapy and endovascular or surgical intervention [15]. The choice of strategy and timing is generally guided by the clinical presentation. Complicated dissections, such as those presenting with acute limb ischemia, progressive dissection, or arterial rupture, necessitate urgent intervention [16,17]. However, there are no established guidelines for treating acute ischemia caused by iliac dissection, and the optimal therapeutic approaches remain under debate, often extrapolated from evidence related to aortic dissection. Open surgery, which involves replacing the dissected aorta with a graft and removing the intimal tear, carries a high risk

Table 2 – A review of literature (case reports), detailing patient characteristics, the affected artery, and interventions in patients presenting with spontaneous isolated dissection of the iliac artery without prior trauma or connective tissue disorder.

Study	Age/Sex	Artery involved	Intervention
Fernandez et al. [20]	56/M	Common iliac artery	Right aortofemoral bypass
Hirai et al. [21]	36/M	Left external iliac artery	Endovascular stent placement
Thalhammer et al. [22]	60/F	External iliac artery	Conservative management
Savolainen et al. [23]	42/M	Common iliac Artery	Dacron graft
Engin et al. [24]	41/M	Bilateral external iliac arteries	Aortobifemoral bypass
Fukui et al. [25]	49/M	Bilateral external iliac arteries	Iliofemoral vein graft bypass
Novotny et al. [16]	38/M	Common iliac artery	Self-expandable stent

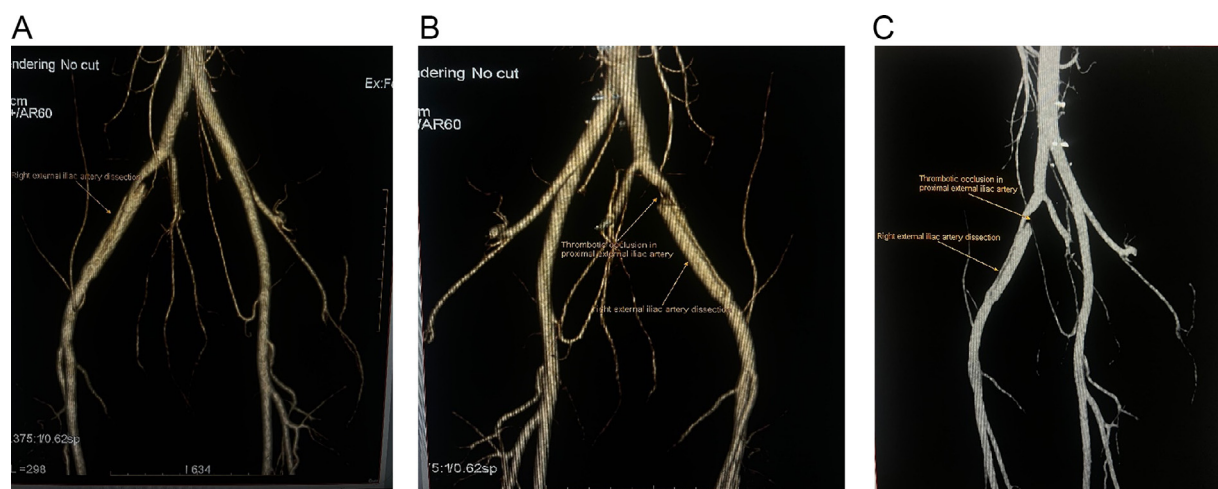


Fig. 3 – Computed tomographic reconstruction showing: (A) Arterial dissection in the right external iliac artery, mildly extending into the right common iliac and internal iliac arteries. (B and C) Acute thrombus within the false lumen of the right proximal external iliac artery, causing approximately 70% occlusion.

of periprocedural complications, including bleeding, neurological issues, or multiorgan failure, affecting 40% to 80% of patients undergoing this procedure. Additionally, open surgery is associated with a significant mortality rate, with in-hospital mortality ranging from 25% to 50%. In contrast, endovascular techniques offer a less invasive treatment option, with lower reported rates of periprocedural complications and in-hospital mortality (ranging from 2.6% to 9.8%) [17]. Endovascular repair has thus become the preferred treatment option for complicated acute type B aortic dissection, [15,17] and has also been successfully used in SID-IA with low periprocedural mortality and morbidity [15,16].

The primary goals of treatment for iliac artery dissection are to halt the progression of the dissection, maintain blood flow distally through the true lumen, and prevent artery rupture. Early surgical intervention is often recommended, especially since there have been reported cases of rupture occurring months after the onset of dissection [18]. Endovascular management, such as percutaneous transluminal angioplasty with endovascular stenting, has shown success in patients with iliac artery dissection [19].

Table 2 provides a compelling overview of similar cases reported in the literature, highlighting the diversity in patient characteristics and interventions utilized. Our case managed with endovascular stenting, aligns with the evolving treatment strategies for SID-IA. Comparison with other cases reveals a spectrum of approaches. This diversity emphasizes the complexity of managing SID-IA and the importance of individualized treatment plans.

Patients with SID-IA are also prone to aneurysm development, highlighting the importance of annual check-ups for screening and monitoring. The urgency of intervention depends on the presentation, with acute limb ischemia or signs of rupture necessitating emergency endovascular or open repair. Endovascular treatment is generally associated with high technical and clinical success rates and low mortality and morbidity compared to open repair, making it a preferred option for complicated cases like SID-IA without signs of rupture [15,16].

Despite favorable outcomes with treatment, the possibility of fatal dissection remains. Continued vigilance, careful follow-up, and prompt diagnosis and treatment are essential

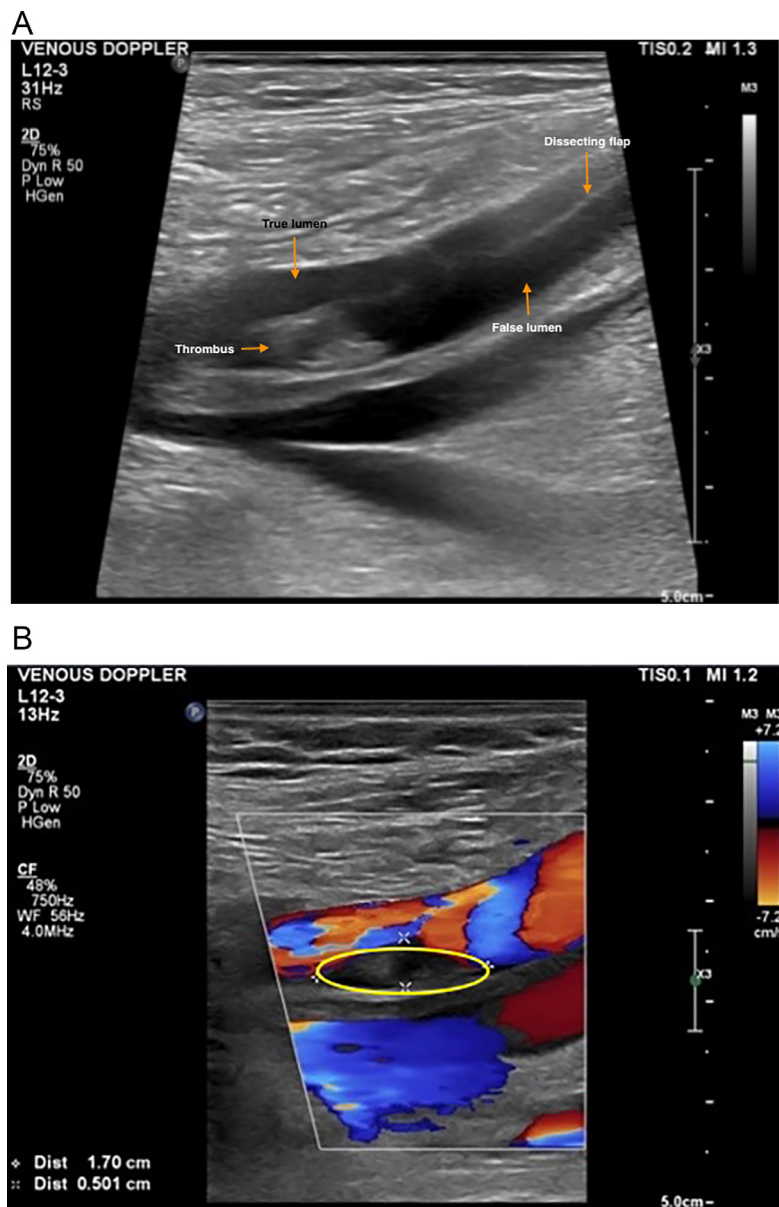


Fig. 4 – Doppler scan showing: (A) Dissecting flap (arrows) dividing the proximal external iliac artery into an upper true lumen and a lower false lumen. (B) Echogenic thrombus (encircled in yellow) within the false lumen of the proximal external iliac artery.

to mitigate the risk of catastrophic consequences in such vascular occurrences.

Conclusion

This case highlights the need for heightened clinical suspicion in young patients presenting with abdominal pain, even in the absence of known risk factors. Successful management with antiplatelet therapy, statins, and endovascular stenting highlights the evolving treatment options for SID-IA. Clinicians

should maintain a high index of suspicion for SID-IA, ensuring prompt diagnosis and intervention to prevent complications. Continued research is warranted to refine treatment strategies and improve outcomes in such rare cases.

Patient consent

Written informed consent was obtained from the patient for their anonymized information to be published in this article (case report).

REFERENCES

- [1] Lyden SP, Srivastava SD, Waldman DL, Green RM. Common iliac artery dissection after blunt trauma: case report of endovascular repair and literature review. *J Trauma* 2001;50(2):339–42. doi:10.1097/00005373-200102000-00024.
- [2] Ehsan O, Darwish A, Edmundson C, Mills V, Al-Khaffaf H. Non-traumatic lower limb vascular complications in endurance athletes. Review of literature. *Eur J Vasc Endovasc Surg Off J Eur Soc Vasc Surg* 2004;28(1):1–8. doi:10.1016/j.ejvs.2004.02.002.
- [3] Nolte JE, Rutherford RB, Nawaz S, Rosenberger A, Speers WC, Krupski WC. Arterial dissections associated with pregnancy. *J Vasc Surg* 1995;21(3):515–20. doi:10.1016/s0741-5214(95)70296-2.
- [4] Gul M, Aksu HU, Oz K, Bakir I. Rupture of the NovaFlex balloon during transcatheter aortic valve implantation and subsequent dissection of the right iliac arteries. *Eur J Cardio-Thorac Surg Off J Eur Assoc Cardio-Thorac Surg*. 2013;43(2):437–438. doi:10.1093/ejcts/ezs508
- [5] Barker SG, Burnand KG. Retrograde iliac artery dissection in Marfan's syndrome: a case report. *J Cardiovasc Surg (Torino)* 1989;30(6):953–4.
- [6] Abayazeed A, Hayman E, Moghadamfalahi M, Cain D. Vascular type Ehlers-Danlos Syndrome with fatal spontaneous rupture of a right common iliac artery dissection: case report and review of literature. *J Radiol Case Rep* 2014;8(2):63–9. doi:10.3941/jrcr.v8i2.1568.
- [7] Akashi H, Nata S, Kanaya K, Shintani Y, Onitsuka S, Aoyagi S. Spontaneous dissection of the iliac artery in a patient with fibromuscular dysplasia. *Ann Vasc Surg* 2010;24(7):952.e13–952.e16. doi:10.1016/j.avsg.2010.02.047.
- [8] Honjo O, Yamada Y, Kuroko Y, Kushida Y, Une D, Hioki K. Spontaneous dissection and rupture of common iliac artery in a patient with fibromuscular dysplasia: a case report and review of the literature on iliac artery dissections secondary to fibromuscular dysplasia. *J Vasc Surg* 2004;40(5):1032–6. doi:10.1016/j.jvs.2004.08.020.
- [9] Dueppers P, Jankowiak S, Schelzig H, Wagenhäuser MU, Oberhuber A. Spontaneous rupture of an isolated iliac artery dissection in a young man because of cystic medial degeneration Erdheim-Gsell. *Ann Vasc Surg* 2015;29(3):596.e11–596.e13. doi:10.1016/j.avsg.2014.10.041.
- [10] Houston C, Rosenthal D, Lamis PA, Stanton PE. Fibromuscular dysplasia of the external iliac arteries: surgical treatment by graduated internal dilatation technique. *Surgery* 1979;85(6):713–15.
- [11] Mehigan JT, Stoney RJ. Arterial microemboli and fibromuscular dysplasia of the external iliac arteries. *Surgery* 1977;81(4):484–6.
- [12] Declémy S, Kreitmann P, Popoff G, Diaz F. Spontaneous dissecting aneurysm of the common iliac artery. *Ann Vasc Surg* 1991;5(6):549–51. doi:10.1007/BF02015282.
- [13] Long MC. Aneurysms of the iliac arteries. *J Gerontol* 1948;3(2):105–10. doi:10.1093/geronj/3.2.105.
- [14] Kratzer GL, Dilcher RH. Aneurysm of the common iliac artery revealed by proctoscopic examination (case report). *Am J Dig Dis* 1950;17(6):210–11. doi:10.1007/BF03005012.
- [15] Liang Z, Guo W, Du C, Xie Y. Effectiveness of the conservative therapy for spontaneous isolated iliac artery dissection: preliminary results. *Vascular*. 2017;25(6):649–656. doi:10.1177/1708538117710845
- [16] Novotny R, Chlupac J, Beran J, Janousek L, Fronek J. Spontaneous isolated common iliac artery dissection treated with self-expandable stent in a 38-year-old patient: a case report. *EJVES Short Rep* 2019;42:4–6. doi:10.1016/j.ejvssr.2018.11.001.
- [17] Rimbau V, Böckler D, Brunkwall J, Cao P, Chiesa R, Coppi G, et al. Editor's choice - management of descending thoracic aorta diseases: clinical practice guidelines of the European Society for Vascular Surgery (ESVS). *Eur J Vasc Endovasc Surg* 2017;53(1):4–52. doi:10.1016/j.ejvs.2016.06.005.
- [18] Wychulis AR, Kincaid OW, Wallace RB. Primary dissecting aneurysms of peripheral arteries. *Mayo Clin Proc* 1969;44(11):804–10.
- [19] Cook PS, Erdoes LS, Selzer PM, Rivera FJ, Palmaz JC. Dissection of the external iliac artery in highly trained athletes. *J Vasc Surg* 1995;22(2):173–7. doi:10.1016/s0741-5214(95)70113-3.
- [20] Fernández AL, Herreros JM. Spontaneous and isolated dissection of the common iliac artery. *J Cardiovasc Surg (Torino)* 1997;38(4):377–9.
- [21] Hirai S, Hamanaka Y, Mitsui N, Isaka M, Kobayashi T. Spontaneous and isolated dissection of the external iliac artery: a case report. *Ann Thorac Cardiovasc Surg Off J Assoc Thorac Cardiovasc Surg Asia* 2002;8(3):180–2.
- [22] Thalhammer C, Aschwanden M, Blum B, Labs KH, Jaeger KA. Unusual cause of intermittent claudication. *VASA Z Gefasskrankheiten* 2004;33(4):257–9. doi:10.1024/0301-1526.33.4.257.
- [23] Savolainen H, Heller G, Fleischmann A, Widmer MK, Carrel TP, Schmidli J. Spontaneous dissection of common iliac artery: a case report. *Vasc Endovascular Surg* 2004;38(3):263–5. doi:10.1177/153857440403800311.
- [24] Engin C, Calkavur T, Apaydin AZ, Durmaz I. Bilateral spontaneous and isolated dissection of the external iliac arteries: report of a case. *EJVES Extra* 2005;9(2):19–21. doi:10.1016/j.ejvsextra.2005.01.005.
- [25] Fukui S, Chelbi E, Paraskevas N, Soury P, Gigou F, Petit MD, Laurian C. Bilateral dissection of external iliac artery. *Ann Vasc Surg* 2007;21(3):373–5. doi:10.1016/j.avsg.2006.06.016.