Adductor canal compression syndrome in an 18-year-old female patient leading to acute critical limb ischemia: A case report

Yi Zhou, Evan J. Ryer, Robert P. Garvin, Jeremy L. Irvan, James R. Elmore

Department of Vascular and Endovascular Surgery, Geisinger Medical Center, Danville, PA, United States

Article history:
Received 31 March 2017
Received in revised form 12 June 2017
Accepted 12 June 2017
Available online 17 June 2017

Keywords:
Adductor canal compression syndrome
Acute limb ischemia
Thrombolysis
Mechanical thrombectomy
Femoral artery reconstruction

ABSTRACT

BACKGROUND: Adductor canal compression syndrome is a rare non-atherosclerotic cause of arterial occlusion and limb ischemia.

PRESENTATION OF CASE: The patient is an 18-year-old healthy female who presented to the emergency department with acute left lower extremity ischemia. Her symptoms began as sudden onset mild foot pain approximately two months ago. Over the 72 h prior to presentation, she developed severe pain, pallor, paralysis, loss of pedal pulses, paresthesia, and poikilothermia. Due to her advanced ischemia, she was taken immediately to the operating room for angiography and intervention. Initial angiography demonstrated distal superficial femoral and popliteal artery occlusions along with lack of tibial or pedal artery blood flow. She underwent percutaneous mechanical thrombectomy and initiation of catheter directed thrombolysis. After 48 h of catheter directed thrombolysis and repeat mechanical thrombectomy, computed tomography (CT) was performed and demonstrated external compression of the superficial femoral artery in the adductor canal and residual chronic thrombus. Echocardiography and CT of the thoracic aorta was also performed, and were negative, therefore excluding other potential sources of arterial embolism. She next underwent surgical exploration, division of an anomalous musculotendinous band compressing the left superficial femoral artery and thromboendarterectomy of the distal left superficial femoral artery. The patient recovered well without any post-operative complications and could return to her daily activities 3 weeks following surgery.

CONCLUSION: Knowledge of rare non-atherosclerotic vascular disorders, such as adductor canal compression syndrome, is paramount when treating patients who present with limb ischemia and lack traditional risk factors.

© 2017 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

1. Introduction

Adductor canal compression syndrome is a rare, non-atherosclerotic cause of lower extremity arterial insufficiency. This disorder primarily affects young, healthy, physically active patients and was initially described by Eduardo Palma in early 1950's [1-3]. The pathophysiology of this condition involves chronic compression of the superficial femoral artery within the adductor canal that ultimately leads to vessel injury, in-situ thrombosis, limb ischemia and potential limb loss [4-6]. The precise source of the external compression varies from case-to-case but published reports have described "embryologic" fibrous bands [5,7], an anomalous musculotendinous band arising from the adductor magnus muscle [4] and hypertrophied adductor magnus or vastus medialis muscles [8]. Treatment is always surgical with some combination of musculotendinous release followed by an arterial revascularization procedure. Here, we report successful limb salvage in a healthy 18-year-old female who presented with advanced acute limb ischemia due to adductor canal compression syndrome.

2. Presentation of case

The patient is an 18-year-old female who presented to our emergency department with acute left lower extremity ischemia. Her past medical history was unremarkable except for oral contraceptive use for irregular menstruation. Her symptoms began as sudden onset moderate foot pain approximately two months prior to presentation. For her foot pain, she was evaluated in the outpatient setting by her primary care physician. It was thought that her symptoms were due to an overuse musculoskeletal injury related to participation in a competitive high school marching band. She continued along with her activities and controlled her symptoms with ibuprofen as needed. Over the 72 h immediately prior to her ED visit, she developed severe pain, pallor, loss of pedal pulses, paresthesia, and poikilothermia. On physical examination in the emergency department, her temperature, pulse and
blood pressure were normal. Pulse examination revealed normal right lower extremity pulses and a normal left common femoral artery pulse. Left popliteal and pedal artery pulses were absent. Moreover, her left foot and distal leg were pale and cool to the touch. She also had diminished sensation on both the dorsal and plantar aspect of her forefoot. Due to her advanced ischemia, she was immediately anticoagulated with intravenous heparin and taken to the operating room for angiography and intervention. After satisfactory anesthesia, we accessed her right common femoral artery under ultrasound guidance and placed a 4 French sheath. We next obtained contralateral femoral access through a series of standard maneuvers and performed left lower extremity angiography. Initial angiography demonstrated thrombosis of the distal superficial femoral artery at the level of the adductor canal (Fig. 1A). More distal angiographic images demonstrated subacute distal embolization with minimal reconstitution of the anterior tibial artery (Fig. 1B) and no arterial flow to the foot (Fig. 1C). In preparation for our therapeutic interventions, our 4 French sheath was upsized to a 45 cm long 6 French sheath (Pinnacle Destination Sheath, Terumo Medical Corporation, Somerset, NJ). Using a 0.035 inch stiff angled hydrophilic coated Glidewire and Navicross support catheter (Terumo Medical Corporation, Somerset, NJ), we could traverse the thrombosed segments. We next exchanged our guidewire for a Hi-Torque Versacore guidewire (Abbott Vascular, Santa Clara CA) and performed mechanical thrombectomy with an Angiojet Solent Omni device (Boston Scientific, Marlborough, MA). Due to residual thrombus on follow-up angiography, a Fountain infusion catheter (Merit Medical Systems, South Jordan UT) was placed and catheter directed thrombolysis began with instillation of Alteplase (recombinant tissue plasminogen activator). The patient returned to the operating room 24 h later. At this second procedure, there was notable improvement in the superficial femoral and popliteal arteries but significant residual thrombus in the tibial and pedal vessels. Mechanical thrombectomy of left peroneal, anterior tibial and dorsalis pedis arteries was performed with both an Angiojet Solent Distal device (Boston Scientific, Marlborough, MA) as well as with the Penumbra Indigo aspiration system (Penumbra, Inc. Alameda, California). Once again, catheter directed thrombolysis was continued due to residual thrombus. After an additional 24 h of catheter directed thrombolysis, the patient returned for repeat angiography. Angiography (Fig. 2A) demonstrated a much-improved result but residual thrombosis within the left superficial femoral artery at the level of the adductor canal. Thrombolytic therapy was stopped. Catheter and sheath were removed. The next day following cessation of thrombolytic therapy, the patient underwent transesophageal echocardiography (TEE) and computed tomography angiography (CTA) of the chest, abdomen and lower extremities were performed. TEE and CTA of the chest and abdomen were normal. CTA of the lower extremities demonstrated external compression of the superficial femoral artery in the adductor canal along with the residual chronic thrombus within the superficial femoral artery at this level (Fig. 2B). The following day, the patient was taken to the operating room for her last surgery. A standard medial thigh approach was used (Fig. 3A). The area of arterial occlusion was identified and adjacent to this focal occlusion there was an impinging anomalous musculotendinous band compressing the left superficial femoral artery (Fig. 3B). This band appeared to arise from the adductor magnus muscle. The anomalous musculotendinous band was divided using electrocautery (Fig. 4A). At this point, the patient was systemically heparinized. Proximal distal control was secured with silastic vessel loops. A longitudinal arteriotomy was made. Thromboendarterectomy of the distal left superficial femoral and above knee popliteal artery was performed (Fig. 4B & C). After flushing and backbleeding, a bovine pericardial patch was sewn in place using a 6-0 Prolene suture (Fig. 4D). Hemostasis was obtained and a standard surgical closure performed. The patient recovered well without any postoperative complications and could return to her daily activities.
3 weeks following surgery. Post-operative arterial duplex ultrasound (Fig. 5A–C) and lower extremity Doppler examination with ankle brachial indices (Fig. 5D) demonstrate normal lower extremity arterial perfusion following our extensive efforts. Evaluation of the right superficial femoral artery revealed no evidence of external compression. Please note this case report follows the CARE guidelines and has also been reported in line with the SCARE criteria. [9,10]

3. Discussion

While thrombotic occlusion of the superficial femoral artery at the adductor hiatus is extremely common in patients with traditional atherosclerotic risk factors, occlusion due to external compression in patients with absence of atherosclerosis is extremely rare and is termed adductor canal compression syndrome. Although described over 60 years ago, available data regarding this disorder is limited to case reports. The syndrome is generally defined as compression of the superficial femoral artery in the adductor canal, an aponeurotic tunnel bordered by the vastus medialis (anterolaterally) and the adductor longus and magnus muscles (posteriorly). This source of the external compression varies from case-to-case but may be due to “embryologic” fibrous bands [5,7], an anomalous musculotendinous band arising from the adductor magnus muscle [4] and hypertrophied adductor magnus or vastus medialis muscles [8]. Symptoms usually begin as exercise induced claudication but can progress to thrombosis and acute limb ischemia. Patients are typically younger than peripheral arterial occlusive disease patients and lack traditional risk factors. In a recent review article [11], Sapienza et al. reports that most patients are male, with a mean age of 45 years, involved in a running sport and are in excellent overall health. Indeed, a female patient suffering from this disorder is rare and we are only aware of one other case report involving a female patient [5]. Further highlighting the rarity of an effected female, Palma, the first to report this disorder in 1950, went as far as to hypothesize that females could not be affected by this disorder due to a more oblique femur and a larger adductor canal [1]. This case contradicts Palma’s assertion and adds to the existing literature by detailing a modern therapeutic approach to an affected female with a successful outcome. Work-up of this suspected diagnosis begins with a history and physical examination focusing on peripheral pulses along with non-invasive vascular labs (ankle brachial indices and arterial duplex ultrasound) but may
Fig. 3. Intra-operative views showing approach to (A) and isolation of hypertrophied adductor magnus muscle compressing the superficial femoral artery in the adductor canal (B).

Fig. 4. Intra-operative views showing (A) the superficial femoral artery following division of the hypertrophied adductor magnus muscle, (B) thrombotic occlusion discovered after arteriotomy, (C) specimen following thromboendarterectomy and (D) the superficial femoral artery following patch angioplasty.
also include computed tomography, conventional arteriography or magnetic resonance imaging. Treatment is always surgical and includes resection/release of the external compression in addition to patch angioplasty or bypass of the diseased superficial femoral artery. Patients who present with acute limb ischemia due to this disorder require immediate revascularization to include open surgical catheter thromboembolectomy or percutaneous catheter directed thrombolysis, depending upon available resources and expertise. Lastly, investigation of the contralateral limb for a similar anomaly must be performed [12].

4. Conclusion

Uncommon vascular etiologies must be considered when physically active patients present with ischemic symptoms. Furthermore, knowledge of rare non-atherosclerotic vascular disorders, such as adductor canal compression syndrome, is paramount when treating patients who progress to limb ischemia and lack traditional risk factors.

Conflicts of interest

None

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical approval

NA.

Consent

Written and signed consent was obtained from the patient

Author contribution

YZ, RPG & JLI - data analysis and interpretation, writing the paper, EJR – overall responsibility, data analysis and interpretation, writing the paper, JRE - data analysis and interpretation, writing the paper.

Guarantor

EJR.

References

[1] PALMA EC, Stenosed arteriopathy of the hunter canal and loop of the adductor magnus, An. J. Surg. 83 (6) (1952) 723–733.
[2] PALMA EC, Obliterating disease of the arteries of the lower extremities; syndrome of hunter’s canal and loop of the third adductor, Bol. Soc. Cir. Urug. 22 (1) (1951) 58–78.
[3] PALMA EC, Stenosing arteriopathies of the lower extremity; hunter’s canal and third abductor ring syndrome, Bol. Trab. Acad. Argent Cir. 34 (21) (1950) 771–787.
[4] M.J. Verta Jr., J. Vitello, J. Fuller, Adductor canal compression syndrome, Arch. Surg. 119 (3) (1984) 345–346.
[5] M. Walensi, C. Berg, M. Piotrowski, F.E. Brock, J.N. Hoffmann, Adductor canal compression syndrome in a 46-year-old female patient leading to acute external iliac, femoral, and popliteal artery thrombosis and critical ischemia: a case report, Ann. Vasc. Surg. 38 (310) (2017) 30674, e11-319. e15. 50890-5096 (16 [pii]).
[6] O. Elsan, A. Darwish, C. Edmundson, V. Mills, H. Al-Khaffaf, Non-traumatic lower limb vascular complications in endurance athletes. Review of literature, Eur. J. Vasc. Endovasc. Surg. 28 (1) (2004) 1–8, http://dx.doi.org/10.1016/j.ejvs.2004.02.002.
[7] F. de Oliveira, R.B. de Vasconcelos Fontes, J. da Silva Baptista, W.P. Mayer, S. de Campos Boldrini, E.A. Libert, The connective tissue of the adductor canal-a morphological study in fetal and adult specimens, J. Anat. 214 (3) (2009) 388–395, http://dx.doi.org/10.1111/j.1469-7580.2009.01047.x.
[8] J.G. Mosley, Arterial problems in athletes, Br. J. Surg. 90 (12) (2003) 1461–1469, http://dx.doi.org/10.1002/bjs.4374.

[9] J.J. Gagnier, G. Kenle, D.G. Altman, et al., The CARE guidelines: consensus-based clinical case report guideline development, J. Clin. Epidemiol. 67 (1) (2014) 46–51, http://dx.doi.org/10.1016/j.jclinepi.2013.08.003.

[10] R.A. Agha, A.J. Fowler, A. Saeta, et al., The SCARE statement: consensus-based surgical case report guidelines, Int. J. Surg. 34 (2016) 180–186, S1743-9191(16)30303-X[pii].

[11] P. Sapienza, E. Tartaglia, L. Venturini, P. Gallo, L. di Marzo, Adductor canal compression syndrome: a forgotten disease, Ann. Ital. Chir. 85 (2014) (ePub):S2239253 × 14023020. S2239253 × 14023020 [pii].

[12] A.A. Perlowski, M.R. Jaff, Vascular disorders in athletes, Vasc. Med. 15 (6) (2010) 469–479, http://dx.doi.org/10.1177/1358863X10382944.