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Persistent Sciatic Artery and its role in limb salvage in oncological surgery

AIM: *The Persistent Sciatic Artery (PSA) is a rare congenital anomaly due to missed involution of embryo-fetal sciatic artery, which is the main blood supply to lower limb during embryonic development until superficial femoral artery (SFA) is formed. The PSA is frequently related to complications in adults like aneurysm and embolism. Here we present a case in which the discovery of a complete PSA resulted limb saving. In case of oncologic or trauma surgery, when no other options are available, the PSA can help in management of reconstructive surgery.*
CASE REPORT: *A case of PSA was discovered during management of a patient affected by a soft tissue sarcoma of the lower limb. Tumor resection needed the femoral neurovascular bundle demolition to ensure radical surgery and subsequent vascular reconstruction, which failed due to complications.*
RESULTS: *Despite failure reconstruction, a misdiagnosed type IIa PSA, replacing the role of the SFA, saved the lower limb from ischemia and subsequent amputation. Functional reconstruction was thus achieved with almost total recovery of lower limb function.*
DISCUSSION AND CONCLUSIONS: *In oncological and trauma surgery we recommend investigate the whole lower limb vascularization, from the pelvis to the foot, suspecting the PSA existence. Indeed, although it is always preferable to reconstruct the SFA system despite a complete PSA is present, due to its frequent complications, the PSA can represent a limb saving option.*

KEY WORDS: Computerized tomography angiography, Persistent sciatic artery, PSA, Superficial femoral artery, SFA fibromyxoid sarcoma

Introduction

The persistent sciatic artery (PSA) is a rare congenital anomaly, due to persistence of primitive sciatic artery that is present during embryonic development as a branch of the umbilical artery, which is the precursor of the internal iliac artery (IIA). The PSA normally obliterates before birth, replaced by the superficial femoral artery (SFA) which develops as a continuation of the external iliac artery (EIA), and its remnants contribute to develop permanent arteries. Its persistence is often associated with aneurysm along its course and consequential related illness, such as potentially limb- and life-threatening embolism or peripheral nerve disorder, due to intrapelvic or extrapelvic compressive action of the aneurysmatic bulk to the nearby sciatic nerve or to its roots originating from L5 and S1. Most of the diagnoses are subsequent to aneurysm related symptoms or incidental reporting due to cadaveric dissection for anatomical purpose or to radiological exam, such as angiography, CTA (Computerized Tomography Angio-graphy) or MRA (Magnetic Resonance Angiography), lead for other major conditions (traumas, tumors, wounds), with an asymptomatic incidental finding of the PSA.

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As described by several authors as Zuckerkandl [1], Senior [2] and Cazenave-Mahé [3], bud of sciatic artery appears during embryonic development, between 4th and 5th week (5mm stage), arising as a branch of the dorsal root of the umbilical artery, becoming the major vascular supply to the primitive lower limb, until 6th week (14mm stage) when it is progressively replaced by the SFA, and it undergoes atrophy at 8th week (22mm stage). Its remnants contribute to develop superior and inferior gluteal, femoris profunda, popliteal, peroneal and dorsalis pedis arteries together with the SFA system.

Case Report

A 32-year-old male presented to Plastic Surgery Department because of a subcutaneous mass discovered within right femoral triangle boundaries. The patient was investigated and staged with CTA and MRI scan of pelvis and upper two thirds of the thigh. Imaging revealed a 55mm Ø mass occurring within the quadriceps femoris muscle, attached to the rectus femoris, sartorius, ileopsoas and pectineus muscles, to the common femoral artery and to the femoral nerve. Presence of the PSA was misdiagnosed, because exams were focused on the mass and the SFA system had a usual pattern in the upper two thirds of the thigh. A percutaneous biopsy revealed a low-grade fibromyxoid sarcoma (cT2N0M0, G1, Stage IB, AJCC 8th ed, 2017) [4].

At physical examination, patient presented a right femoral mass with a rubbery, elastic consistence, non-mobile on the underlying surfaces, without any femoral nerve compression related symptoms, despite the intimate relationship with it.

A multidisciplinary team, after evaluating the clinical and radiological presentation, decided for immediate mass resection and direct reconstruction, without additional radiotherapy [5].

Surgical wide resection (with intent to obtain negative margins) was considered the primary definitive treatment for a stage IB (T2-4, N0, M0, low grade) tumor, if the margins had been greater than 1 cm or the fascial plane

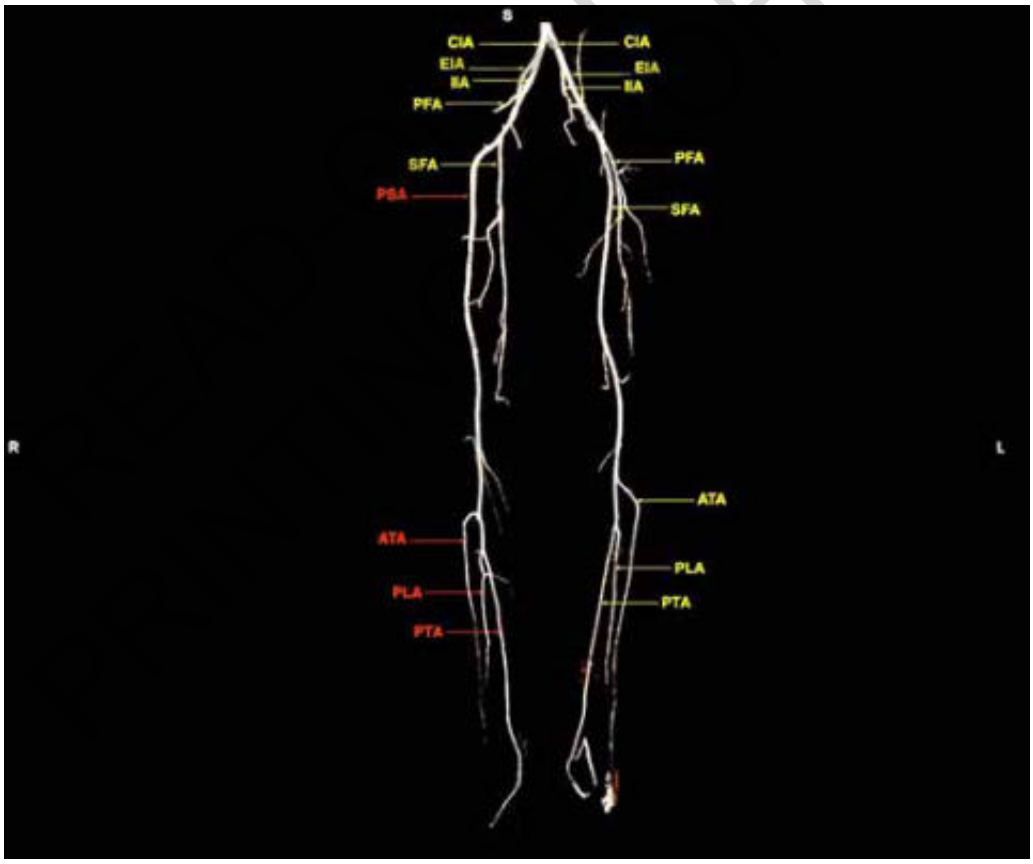

Fig. 1: Postoperative CTA 3D reconstruction. On the right limb, course of PSA, arising from IIA, and its branches can be observed, with ATA, PLA and PTA derived from distal portion of PSA. Arterial bypass of SFA is still patent, while PFA has been ligated after tumor resection. On the left limb, usual vascular pattern of SFA system can be observed, compared to contralateral anomalous PSA pattern.
Legend: CTA = Computerized Tomography Angiography, CIA = common iliac artery, EIA = external iliac artery, IIA = internal iliac artery, SFA = superficial femoral artery, PFA = profunda femoris artery, PSA = persistent sciatic artery, ATA = anterior tibial artery, PTA = posterior tibial artery, PLA = peroneal artery.

intact. Postoperative RT would have been recommended for final surgical margins of 1 cm or less [5].
Surrounding involved structures like rectus femoris muscle, sartorius muscle, vastus lateralis muscle, adductor longus muscle, superficial femoral and profunda femoris arteries and veins and femoral nerve were ablated together with the neoplasm. Surgical resection was considered radical because of margins greater than 1cm.
Vascular reconstruction consisted in performing end-to-end anastomosis with autologous bypass using saphenous vein graft between proximal and distal SFA stumps and end-to-end anastomosis with synthetic venous bypass using PTFE Propaten (W. L. Gore & Associates, Inc., Flagstaff, AZ, USA) prosthesis between deep femoral and common femoral veins. Afterwards, functional reconstruction of the femoral nerve was performed with double sural nerve graft and coverage of the surgical site with ipsilateral musculocutaneous VRAM (vertical rectus abdominis muscle) flap, pedicled on inferior epigastric artery. Antibiotic and anticoagulant therapy was administrated as prophylaxis of thrombosis and infections.
Subsequent fever onset with positive hemoculture for multi-sensible E. cloacae suggested a complete lower limb CTA control (Fig. 1) that revealed occlusion of the venous synthetic bypass, but patent SFA bypass and the PSA existence which was misdiagnosed at preoperative evaluation. Revision surgery was then performed with the removal of venous prosthesis and VRAM flap repositioning.
Afterwards, despite fever remission and good perfusion of the right lower limb, a massive internal hemorrhage with acute anemization forced to return the patient to the operating room. Surgical findings evidenced infected arterial bypass (later confirmed at microbiological samples) with multiple laceration of the vessel graft, but no distal lower limb ischemia was developed, due to persistence of sciatic artery, discovered at previous CTA scan, that the patient presented unilaterally, nourishing the distal lower limb. Ligation of the SFA was then performed with no ischemic consequences.
No further complications were noticed after surgery. Patient was discharged 34 days after first surgery, with no signs of infection, no ischemia of the lower right limb, palpable, symmetric and regular pulses of lower extremities, and with a good and stable wound coverage. Rehabilitation allowed to regain thigh-to-hip flexion until 60° of range of motion at six months follow-up, due to femoral nerve reconstruction and saving of vastus medialis muscle. Two years follow-up did not reveal tumor recurrence or PSA aneurysm development.

Discussion

First described by Green in 1832 [6] and subsequently by Manno [7], Pernkopf [8] and Finerty [9], the PSA is found in 0.03-0.06% of the population with no gender differences [10]. Bower distinguished complete and incomplete PSA, depending on the degree of involution of the PSA and on the SFA hypoplasia [11]. Indeed, this classification of the PSA is based on the main blood provider of the lower limb: incomplete PSA if the main blood provider is the SFA, complete PSA if the latter provides to the nourishing of the lower limb, representing the majority of cases (80%) [10]. Pillet et al. [12] described four categories of PSA, based on the partial or total absence of the SFA. Type I PSA presents a complete PSA development with a normal SFA. Type II PSA presents a complete PSA but an SFA that does not reach the popliteal artery (type IIa) or is completely absent (type IIb). Type III and IV PSA are characterized by an incomplete PSA, more specifically the upper half of the artery (type III) or the lower half of the artery (type IV) and a normal development of the SFA and its related system. Gauffre et al. [13] recently added a Type V PSA originating from the median sacral artery.
The PSA has more often a unilateral presentation and up to a quarter of patients have a bilateral anomaly (22-25%). [14]
A clear etiological factor in persistence of sciatic artery is not identified, but several authors state that vascular pattern of lower limb develops due to the action of vasculogenesis promoting factors (VEGF, angiopoietin-1, TGF-b, PDGF-B, Heparin binding-EGF, etc.), inhibiting signaling factors (angiopoietin-2, angiostatin, etc.) and mechanical factors that act on the remnants of the anterior femoral system and posteriorly located sciatic artery. Persistence of sciatic artery could therefore be the result of the prevalence of triggers which determine the maintenance of a primitive vascular pattern, likewise as amphibians [15].
The PSA is usually symptomatic (25-58%), because of formation of aneurysm and possible complications as sciatic nerve compression, thrombosis, embolism and related ischemia of the corresponding lower limb. Mechanism of aneurysm formation is explained by compression of the PSA against near structures as piriformis muscle, hip bones, sacrospinal ligament or flexion movement of the hip joint. A reduction of elastic fibers in PSA wall has been found either [10,14].
Presence of the PSA is usually discovered after execution of imaging test as arteriography [16], as primary diagnostic technique, or CTA or MRA scan, which are progressively replacing traditional angiography, thanks to the additional information related to surrounding PSA structures and their reduced invasive approach [10].
When incidentally discovered and with no associated complications, the PSA is commonly treated with lifestyle modifications, such as avoiding repeated trauma of the artery (reduced seated time, reducing pressure on the PSA area even if in supine position while sleeping, etc.). Otherwise, complicated or symptomatic PSA needs of further treatment, depending on underlying pathology,

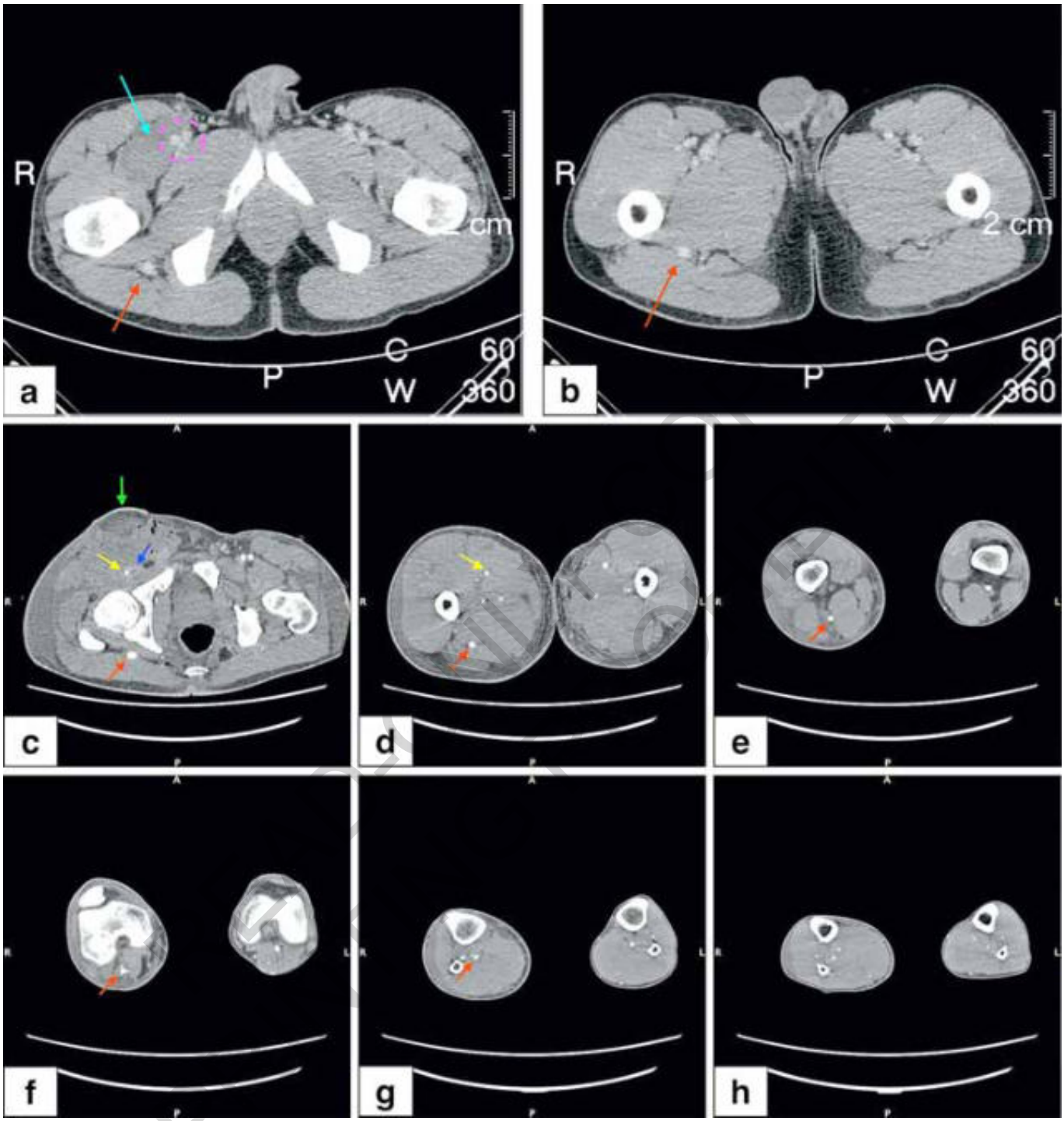

Fig. 2: Preoperative CT scan (A, B) and postoperative CTA scan (C-H). Arrows and circle indicate main elements of the pictures (Red arrow: PSA; Yellow arrow: Native SFA and autologous arterial bypass; Blue arrow: occluded venous synthetic bypass; Cyan arrow: tumor burden; Green arrow: VRAM flap; Dashed circle: femoral neurovascular bundle).
Read in a left-to-right, top-to-bottom order: A) PSA as branch of internal iliac artery, deep to gluteus maximus muscle and superficial to pirifomis, gemellus superior and obturator internus muscles group; B) PSA deep to gluteus maximus muscle at the middle and proximal thirds junction of the thigh; C) PSA as branch of internal iliac artery, deep to gluteus maximus muscle and superficial to pirifomis, gemellus superior and obturator internus muscles group; D) PSA deep to gluteus maximus muscle at the middle and proximal thirds junction of the thigh; E) PSA deep to ischiocrural muscles group in the posterior aspect of the thigh; F) medial and lateral sural arteries arisen from PSA between the two head of gastrocnemious muscle; Fig. G) anterior tibial artery and soleus muscle branch arisen from PSA; H) the three major leg vessels, anterior tibial, posterior tibial and peroneal arteries, arisen from PSA.
Legend: CT = Computerized Tomography, CTA = Computerized Angiography Tomography, PSA = persistent sciatic artery, SFA = Superficial Femoral Artery, VRAM = Vertical Rectus Abdominis Muscle.

as surgical bypass, endovascular thrombolysis, stenting, or embolization .[17,18]

Our patient presented right sided type IIa [12,13], complete and asymptomatic PSA which saved his lower right limb from ischemia, even if arterial bypass failed, with a normal SFA system on his left lower limb. Complete PSA axis can be seen in 3D CTA reconstruction in Fig. 1. Fig. 2 (A-B) represents the preoperative CT scan with the tumor bulk, where PSA was misdiagnosed and complete PSA course can be observed in postoperative CTA scan (Fig. 2, C-H), compared with normal SFA pattern in contralateral limb.

The right PSA of the patient originated as a branch of the IIA with a parallel apparently normal SFA system in the upper two thirds of the thigh. No aneurysm was detected along its course. The PSA emerged together with the sciatic nerve, the inferior gluteal nerve and the posterior cutaneous femoral nerve from the greater sciatic notch, between the piriformis muscle and the superior gemellus muscle, posterior to the greater trochanter (Fig. 2C).

Afterwards, the PSA continued, proximal to distal direction, in the posterior aspect of the thigh, nourishing the leg and the foot, running, superficially to superior gemellus muscle, then to obturator internus muscle, gemellus inferior muscle and quadratus femoris muscle, deeply to gluteus maximus muscle and laterally to the long head of biceps femoris muscle.

Between the proximal and middle thirds of the thigh, the PSA continued running deeply to gluteus maximus muscle, long head of biceps femoris muscle, semitendinosus muscle and superficially to adductor magnus muscle and to the short head of biceps femoris muscle (Fig. 2, D-E), until the lower third of the thigh, together with the sciatic nerve. The SFA progressively reduced its caliber and completely vanished at distal third of the thigh. Distally, at knee level, the PSA continued posteriorly in popliteal fossa, more superficially than usual SFA derived popliteal artery, giving rise to medial and lateral sural arteries (Fig. 2F), running between the two heads of gastrocnemius, and superficially to popliteus and soleus muscles.

Finally, between the proximal and middle thirds of the leg, the PSA gave rise to anterior tibial artery (ATA) (Fig. 2G) and, afterwards, to the peroneal artery (PLA), continuing as posterior tibial artery (PTA) with the usual anatomic course as in the contralateral limb (Fig. 2H).

Our patient's femoral axis was reconstructed because preoperative evaluation was based on a partial, and without contrast enhancement, imaging investigation, only including the pelvis and the upper two thirds of the thigh, which revealed an efficient and apparently normal SFA, that nourished the anterior compartment of the thigh, but not its uncomplete course and the replacement role of the PSA (PSA type IIa [12,13]), which was misdiagnosed.

However, if the preoperative imaging had revealed the presence of the PSA, an attempt to reconstruct the SFA system would have been reasonable anyway, due to the young age of the patient and the uncertain behavior of the PSA during his life, as the development of aneurysms is frequent and dangerous.

Altogether, the failure of the vascular bypass to save the SFA system, did not affect perfusion and vitality of lower limb, due to efficiency of the PSA system that ensured nourishing of the leg and foot.

Conclusion

Vascular arterial malformation as PSA could represent a threat to lower limb bloodstream and vitality, and sometimes be life-threatening to the patients, due to its frequent complications. Nevertheless, the PSA can be a resource as a further limb-saving option in oncological and trauma surgery when completely replaces the SFA system. Definitely, angiography or CTA scan should always be performed including the whole limb in vascular reconstruction, especially when sparing the SFA system is not a viable choice.

Riassunto

L'arteria ischiatica persistente (PSA) è una rara anomalia congenita causata dalla mancata involuzione dell'arteria ischiatica embrio-fetale, che rappresenta la principale fonte di apporto ematico all'arto inferiore durante lo sviluppo embrionale fino a quando l'arteria femorale superficiale (SFA) si sviluppa. La PSA è frequentemente associata allo sviluppo di complicanze negli adulti come aneurismi ed embolie. Presentiamo un caso in cui la scoperta di una PSA completa è risultata cruciale nel salvataggio di un arto. In caso di chirurgia oncologica o traumatologica, quando non sono disponibili altre opzioni, la PSA può essere d'aiuto nella gestione della chirurgia ricostruttiva.

Un caso di PSA è stato scoperto durante la gestione di un paziente affetto da un sarcoma dei tessuti molli dell'arto inferiore. La resezione tumorale ha necessitato la demolizione del compartimento vascolonervoso femorale per assicurare la radicalità chirurgica e la successiva ricostruzione vascolare, che tuttavia è fallita per via di complicazioni.

Nonostante il fallimento della ricostruzione, una PSA di tipo IIa non diagnosticata, vicariante il ruolo della SFA nella vascolarizzazione dell'arto inferiore, è stata in grado di salvarlo dall'ischemia e dall'amputazione. La ricostruzione funzionale è stata comunque ottenuta con un recupero quasi totale della funzione dell'arto inferiore. Nella chirurgia oncologica e traumatologica raccomandiamo l'esecuzione di accurato esame diagnostico di imaging per lo studio della vascolarizzazione dell'intero arto

inferiore, dalla pelvi al piede, nel sospetto dell'esistenza di una PSA. Infatti, nonostante sia sempre preferibile ricostruire il sistema della SFA anche in presenza di una PSA completa, per via delle sue frequenti complicazioni, la PSA può comunque rappresentare un'opzione di salvataggio dell'arto.

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