

More than what meets the eye in COVID-19 critical illness: A case report of bilateral femoral neuropathy due to psoas hematomas

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ABSTRACT

يعد اعتلال العصب الفخذي نادراً، خاصةً الذي يحدث بسبب تجمع دم في كلا العضلات الفطنية والحرقفية. نقدم حالة من الاعتلال العصبي الفخذي الثنائي بسبب الأورام الدموية التي تطورت أثناء مرض كوفيد – 19. أصيب مريض يبلغ من العمر 41 عاماً بالتهاب الرئوي الناجم عن فيروس كورونا، وتدهورت حالته بسرعة. أدى انخفاض مستوى الهيموجلوبين إلى إجراء دراسات تصويرية أثناء إقامته في وحدة العناية المركزة. تم تحديد الأورام الدموية الثنائية كمصدر للنزيف. بعد ذلك اشتكى المريض من ضعف في الأطراف العلوية والسفلية وتنميل في الطرف السفلي. واعتبر أنه يعاني من مرض اعتلال الأعصاب بعد إقامته في العناية المركزة وتمت إحالته إلى التأهيل. اقترح اختبار التوصيل العصبي وجود اعتلال عصبي فخذي ثنائي بسبب الضغط الناتج عن الأورام الدموية التي تطورت أثناء إقامته في وحدة العناية المركزة. يمكن أن تكون عواقب الأورام الدموية كارثية، تتراوح من الصدمة النزفية إلى الضعف الشديد، لذلك نسلط الضوء على أهمية معرفة هذا الكيان.

Bilateral femoral neuropathy is rare, especially that caused by bilateral compressive iliopsoas, psoas, or iliacus muscle hematomas. We present a case of bilateral femoral neuropathy due to spontaneous psoas hematomas developed during COVID-19 critical illness. A 41-year-old patient developed COVID-19 pneumonia, and his condition deteriorated rapidly. A decrease in the hemoglobin level prompted imaging studies during his intensive care unit (ICU) stay. Bilateral psoas hematomas were identified as the source of bleeding. Thereafter, the patient complained of weakness in both upper and lower limbs and numbness in the lower limb. He was considered to have critical illness neuropathy and was referred to rehabilitation. Electrodiagnostic testing suggested bilateral femoral neuropathy because of compression due to hematomas developed during the course of his ICU stay. The consequences of iliopsoas hematomas occurring in the critically ill can be catastrophic, ranging from hemorrhagic shock to severe weakness, highlighting the importance of recognizing this entity.

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A wide range of neurological complications associated with COVID-19 affecting both central and peripheral nervous systems have been reported. Unilateral femoral mononeuropathy is an uncommon condition. Bilateral femoral neuropathy is rare, especially that caused by bilateral compressive iliopsoas, psoas, or iliacus muscle hematomas. In this report, we present the case of a 41-year-old patient with COVID-19-associated acute respiratory distress syndrome (ARDS), who developed bilateral femoral neuropathy as a consequence of bilateral psoas hematomas. We additionally report the findings of a literature review search for bilateral femoral neuropathy cases in association with psoas, iliacus, or iliopsoas hematomas. The management of such cases remains largely unstandardized and understudied.

Case Report. Patient information. A 41-year-old man, a known case of diabetes mellitus type 2, bronchial asthma, and obesity, presented with cough, shortness of breath, and fever. His COVID-19 positive status was confirmed via nasopharyngeal polymerase chain reaction testing. The patient developed ARDS during his hospital stay, and transferring him to the intensive care unit (ICU) was deemed necessary. His condition continued to deteriorate; hence, he was mechanically

ventilated and placed on venovenous extracorporeal membrane oxygenation (ECMO) 3 days into his ICU stay. His ICU course was both complicated and lengthy. He developed partial thrombosis of the proximal part of the right internal jugular vein, which was the ECMO access point at that time. Therefore, the access was switched to the left side (left internal jugular and right common femoral veins). A decrease in the hemoglobin level from 9.0 to 7.5 g/L prompted imaging investigations. Computed tomography (CT) scan of the abdomen and pelvis demonstrated the source of the bleeding: right iliopsoas and left psoas hematomas (Figures 1-2). At the time of the bleeding, heparin infusion was being administered. The patient was treated conservatively, and his hemoglobin level subsequently stabilized.

On discharge from the general hospital, although higher brain function was preserved, upper and lower limb weakness and loss of sensation in the ventral side of both lower limbs were evident. Dabigatran 110 mg twice daily was added to his home medications.

Clinical Findings. The patient was referred as a case of critical illness neuropathy for rehabilitation. He was admitted to the inpatient rehab floor 3 months after the discharge. On examination, upper limb tone and power were back to baseline. However, his lower limbs were hypotonic. Hip flexion was 1/5 on the left side and 2/5 on the right side. Both knee flexors and extensors were weak, with powers of 2/5 and 0/5, respectively. His ankle power was 4/5 bilaterally. Knee and ankle reflexes were absent in both limbs. Furthermore, he had reduced sensation in the distribution of the femoral and saphenous nerves.

Diagnostic assessment. Given the above findings, the patient was suspected to have femoral neuropathy in both limbs in addition to the diagnosis of critical illness neuropathy. Therefore, an electrodiagnostic study was performed. The findings were conclusive for mild sensory motor axonal polyneuropathy and bilateral femoral neuropathy with severe denervation in both quadricepses. The iliopsoas on the left side exhibited significant weakness, but needle electromyography was not performed as the patient was on anticoagulants. As he was retrospectively diagnosed, a follow-up CT scan was carried out 8 months after the bleeding, which showed complete resolution of the hematomas.

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Figure 1 - The CT of the pelvis, which demonstrates bilateral psoas hematomas, more significant on the left and right iliac hematoma (transverse plane).



Figure 2 - The CT of the abdomen and pelvis, which demonstrates bilateral psoas hematomas, more extensive on the left as well as right iliac hematoma (coronal plane).

Therapeutic intervention. A comprehensive inpatient rehabilitation program was offered, including occupational and physical therapy, for 5 months.

Follow-up and outcomes. One year after the injury to the femoral nerve, the patient was independent for all activities of daily living and was able to walk with a walking aid (walker, quadripod) under supervision



Figure 3 -Timeline of patient's illness and recovery

Table 1 - Cases of bilateral femoral neuropathy associated with bilateral iliopsoas, psoas, or iliacus hematomas reported in the literature*

Author et al./ year	Gender/ age	Background of the case	Hematoma location and size	Timing of hematoma
Macauley P et al./ 2017 ²	F/ 64 yr	PE and DVT on enoxaparin; history of hypertension, type 2 DM, and COPD	Bilateral psoas hematomas larger on the Rt, without extravasation	Day 10 of admission for PE/ DVT
Basheer A et al./ 2013 ¹	F/ 63 yr	PE on IV heparin and a single dose of clopidogrel, followed next day by warfarin with heparin bridging	Large, bilateral iliopsoas muscle hematomas	Day 7 of presentation with PE
Wada Y, Yanagihara C, and Nishimura Y/ 2005 ⁵	F/ 85 yr	TIA on heparin infusion and warfarin; hypertrophic cardiomyopathy	Bilateral iliopsoas hematomas larger on the Rt with seeping of the contrast material into the hematomas	After 3 days of heparin and warfarin treatment
Jamjoom ZA et al./ 1993 ⁶	F/ 19 yr	DVT on heparin, with warfarin added 10 days later; history of estrogen and progesterone therapy for menstrual disturbances	Two large hematomas in both iliacus muscles; Sizes on US: Lt 19 x 62 mm, Rt 27 x 65 mm	After 3 wk of warfarin treatment
Chevalier X and Larget-Piet B/ 1992 ⁷	F/ 75 yr	Myocardial ischemia on heparin; history of MI	Giant hematoma of Lt psoas muscle and blood collection in Rt psoas muscle	N/A
Niakan E et al./ 1991 ⁸	M/ 54 yr	Acute PE secondary to DVT on heparin and warfarin	Bilateral iliopsoas hematoma; Rt larger than Lt.	N/A
Barontini F and Macucci M/ 1986 ⁹	M/ 65 yr	Previous MI on antiaggregant therapy (Teklid) and anticoagulant (Calciparina); history of gastrectomy and operation for L4-L5 disc hernia	Small hemorrhagic areas of both iliac muscles	More than 2.5 yr after starting Teklid and 3 mon after starting Calciparina
Storen EJ/ 1978 ¹⁰	F/ 50 yr	DVT on heparin infusion, with warfarin added after 3 days	Bilateral iliacus hematoma	5 days after starting heparin

*The mechanism of hematoma in all cases was anticoagulation. N/A: not available; F: female; M: male, yr: year; wk: week; mon: month; Rt: right; Lt: left; PE: pulmonary embolism; DVT: deep vein thrombosis; DM: diabetes mellitus; COPD: chronic obstructive pulmonary disease; IV: intravenous; TIA: transient ischemic attack; MI: myocardial infarction; US: ultrasound; MCV: motor conduction velocity; TAE: transcatheter arterial embolization

Table 1 - Cases of bilateral femoral neuropathy associated with bilateral iliopsoas, psoas, or iliacus hematomas reported in the literature*

Symptoms related to femoral neuropathy	Timing of neuropathy presentation	Positive examination findings on neuropathy presentation	Management	Outcome
Inability to move bilateral lower extremities	Day 16 of admission for PE/ DVT	Inability to elevate legs against gravity or to extend knees; absent leg reflexes	Conservative management	Discharged to a rehabilitation center 4 wk after neuropathy diagnosis with motor strength 4/5 in bilateral lower extremities
Moderate weakness in proximal lower extremities	Day 7 of hospitalization	Strength 3/5 in bilateral iliopsoas and 2/5 in quadriceps; hyperalgesia and numbness over the L2-L4 dermatomes, bilaterally	Conservative management followed by surgical decompression	At 3- and 6-month follow-up visits, full strength in lower extremities, but continued mild dysesthesias in anterolateral thighs bilaterally
Severe pain in anterior aspect of Rt thigh, followed next day by similar, less intense symptoms on Rt side	N/A	Reduced sensation to pinpricks in the distribution of Rt saphenous nerve; next day bilateral femoral nerve palsy more severe on the Rt than on the Lt	Conservative management followed by TAE	On discharge, 3 wk after TAE, femoral nerve function had returned to normal apart from a mild residual weakness in Rt quadriceps muscle
Numbness of anterior aspect of both thighs with both legs held flexed at hip joints	Second day of pain onset	Bilateral incomplete femoral nerve palsy, Rt more than Lt	Conservative management followed by bilateral surgical decompression	On discharge, 3 wk after surgery, femoral nerve functions had returned to normal apart from mild residual weakness in Lt quadriceps muscle
Sudden Lt thigh pain, followed by complete deficit of the entire quadriceps	Several hours after hematoma symptom onset	Quadriceps score of zero and complete abolition of Lt patellar reflex; strength of Rt quadriceps and Rt patellar reflex were also diminished	Emergency surgery	Poor postoperative recovery; seven months later, Lt quadriceps deficit score retained between 3 and 4, which considerably interfered with walking
Paresthesias over anterior aspect of Rt thigh, radiating to medial and anterior portions of lower leg; Rt hip flexed; developed similar, less intense symptoms with on Lt side after 48 hr	N/A	Weakness of Rt quadriceps femoris; decreased sensation to pinprick in Rt saphenous nerve distribution; knee jerk: Rt 1+ and Lt 2+; ankle jerk: 1+ bilaterally; flexor plantar responses	Surgical evacuation of Rt hematoma and conservative management of Lt hematoma	All symptoms in both legs resolved within a few days, except for mild paresthesias on the Rt.
Pain in lower limbs, followed by inability to get up	Around the same time as hematoma presentation	Impossibility of extension of legs; remarkable reduction of flexion and adduction of thighs; absent patellar reflexes; hypoesthesia on anterior surface of thighs and middle surface of shins; findings were bilateral, but greater on Lt side	Conservative management	More than 3 mon after admission, walking was possible without support; motor deficit in lower limbs was 4th degree on Lt and 2nd degree on Rt; sensation in femoral nerve territory was remarkably improved
Anesthesia on medial aspect of both thighs	48 hr after hematoma symptom onset	Bilateral flaccid paralysis of femoral nerve; absence of patellar reflex bilaterally	Immediate operation with bilateral hematoma evacuation; warfarin treatment was maintained	At 10 wk after the operation, pt was able to walk unsupported and bend knees to near sitting position; skin sensibility returned to near normal

*The mechanism of hematoma in all cases was anticoagulation. N/A: not available; F: female; M: male, yr: year; wk: week; mon: month; Rt: right; Lt: left; PE: pulmonary embolism; DVT: deep vein thrombosis; DM: diabetes mellitus; COPD: chronic obstructive pulmonary disease; IV: intravenous; TIA: transient ischemic attack; MI: myocardial infarction; US: ultrasound; MCV: motor conduction velocity; TAE: transcatheter arterial embolization

owing to safety concerns. As his knees continued to buckle, a ground reaction ankle foot orthosis was prescribed to aid his gait. The motor power was 2+/5 in his right quadriceps and 2/5 in his left side; his hip flexors also improved, with a power of 3/5.

Discussion. This case report presents a patient with bilateral femoral neuropathy secondary to

compression by bilateral psoas hematomas associated with COVID-19.

Owing to its long course, the femoral nerve is susceptible to compression, which is most common within the psoas muscle, iliopsoas groove, and inguinal ligament.^{1,2} Other causes for femoral neuropathy include surgical procedures involving the abdomen, pelvis, inguinal area, and hip, such as hip arthroplasties.

Procedures that involve catheterization of the femoral artery or vein is another iatrogenic cause. Penetrating or blunt trauma and hip or pelvic fractures can also cause femoral neuropathy. Although diabetes mellitus can sometimes result in isolated femoral neuropathy, involvement of spinal roots, lumbosacral plexus, or other peripheral nerves is also evident on careful clinical or electrodiagnostic assessment in most cases.^{3,4} In our case, we propose the bilateral psoas hematomas to be the main culprit for the patient's bilateral femoral neuropathy. Other contributing factors include right femoral vein cannulation for ECMO, diabetes mellitus, and prone positioning.

Iliopsoas hematomas are uncommon, and most cases are caused by anticoagulation/antiplatelet therapy or hemophilia. In the majority of reported cases, anticoagulation was within the therapeutic range, which applied for both unilateral and bilateral cases (Table 1). Some cases due to trauma in patients with no bleeding disorders have also been reported.¹ With the increasing use of anticoagulants in clinical scenarios, iliopsoas hematomas are expected to be increasingly encountered.

This case is noteworthy as most cases of iliopsoas hematoma-associated femoral neuropathy are unilateral. There are 8 reported cases with bilateral femoral neuropathy-associated hematoma in the literature (Table 1). Additionally, we highlight the importance of identifying this condition promptly to avoid delays in diagnosis. Our patient was diagnosed in the rehabilitation setting (Figure 3). The ICU course was retrospectively reviewed to pinpoint the cause. Thankfully, the hematoma resolved spontaneously although the patient was maintained on dabigatran. Earlier diagnosis of this compressive neuropathy might have offered the benefit of discontinuing anticoagulants as soon as it was feasible. Whether stopping the anticoagulants could have hastened the resolution, reduced the duration of compression, and improved the prognosis is an essential consideration in future cases. Owing to the rarity of the condition and lack of evidence, the ideal plan is an individualized management approach and shared decision-making with the patient. The association of this condition with COVID-19 complicated the case. The hypercoagulable state imposed by the infection increased the need for anticoagulants. As evident from Table 1, our patient did not have a robust recovery. The masking of a critical illness and the consequent delay in diagnosis likely worsened the outcome. Another factor that might have affected the outcome is the association with COVID-19.

The presentation of the iliopsoas hematoma starts with groin and hip pain, radiating to the anterior thigh and lumbar region, with hip flexion and external rotation owing to iliacus muscle spasm. Patients can also develop neuropathy, as in our case, and/or massive bleeding and shock. Femoral neuropathy manifests initially in the form of anteromedial thigh and leg pain, followed by anesthesia at the exact locations. Motor weakness and wasting of the quadriceps can also occur, with a decrease in or absence of knee jerk. Moreover, variable hip flexion weakness may develop.¹ As evident from the CT scan, the left psoas hematoma was more extensive and more proximal, which explained the more severe deficits on this side in the present case.

Diagnosing iliopsoas hematomas is best done using magnetic resonance imaging owing to its high sensitivity and specificity. However, CT is routinely used as it is more readily accessible, faster, and has good sensitivity.¹ Electrodiagnostic studies are valuable in confirming the diagnosis of femoral neuropathy, excluding other neural involvement, and determining the prognosis of femoral neuropathy.

The mainstay of femoral neuropathy treatment is physiotherapy unless the cause of compression can be removed. Early rehabilitation is likely to improve the outcomes and shorten the duration of recovery (Table 1). Initial actions for treating iliopsoas hematomas involve resuscitative measures as well as reversal of coagulopathy if present. The definitive treatment for iliopsoas hematoma in association with femoral neuropathy remains controversial, and the choice between conservative and operative management is not straightforward. Suggested indications for intervention include severe femoral neuropathy, refractory hemodynamic instability, abdominal compartment syndrome, and large hematomas. The disadvantage of surgery is that the removal of the hematoma may increase the bleeding by eliminating the tamponade effect. Moreover, in some cases, abdominal packing is the only surgical option. In addition, many of the patients are poor surgical candidates. Thus, interventional radiology with intra-arterial embolization or stent-grafting is emerging as a preferred treatment modality, especially in case of active bleeding. Poor surgical candidates may also benefit from ultrasound- or CT-guided percutaneous decompression.²

Conclusion. Much attention has been devoted to the thrombotic complications of COVID-19; however, hemorrhagic complications can also be life-threatening and are associated with lifelong disability. Hence, care should be taken to detect and possibly actively manage hematomas, mainly if they occur around the course of

nerves. Knowledge of treatment options is critical, and the therapy should be individualized on a case-to-case basis.

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