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CASE REPORT

Pseudoaneurysm of the Internal Iliac Artery: a Rare Cause of Bilateral Lumbosacral Plexopathy

SF Low[1], TY Kew[1], CS Ngiu[2], NA Bakar[1]

Departments of [1]Radiology and [2]Medicine, Universiti Kebangsaan Malaysia Medical Centre, Kuala Lumpur, Malaysia

ABSTRACT

Pseudoaneurysms of the internal iliac artery are uncommon and are usually associated with trauma. Rarely, they expand subclinically for years and cause significant compression of adjacent vital structures. We report an unusual case of delayed onset bilateral lumbosacral plexopathy resulting from direct compression due to a right internal iliac artery pseudoaneurysm. The neurological symptoms of the patient began 2 years after a stab injury to the right gluteal region. To the best of our knowledge, this is the first report of bilateral lumbosacral plexopathy secondary to a solitary internal iliac artery pseudoaneurysm that manifested clinically 2 years after the injury. This report illustrates how a pseudoaneurysm of the internal iliac artery (an extraspinal pathology) has the potential to cause lumbosacral plexopathy, and describes relevant imaging findings.

Key Words: *Aneurysm, false; Iliac artery; Lumbosacral plexus; Wounds and injuries*

中文摘要

假性動脈瘤的髂內動脈：雙側腰骶神經叢病變的罕見病因

劉淑芬、丘天恩、魏財順、NA Bakar

髂內動脈假性動脈瘤很罕見，通常與創傷有關。髂內動脈假性動脈瘤極少呈亞臨床性隱匿擴張數年而顯著壓迫鄰近重要器官。本文報告一宗因右髂內動脈假性動脈瘤直接壓迫誘發的遲發性雙側腰骶神經叢病變的罕見病例。病人在右臀區被刺傷，兩年後開始出現神經系統症狀。據我們所知，這是首宗創傷兩年後才有臨床表現，繼發於孤立髂內動脈假性動脈瘤的雙側腰骶神經叢病變的病例報告。本文闡明髂內動脈的假性動脈瘤（髓外病理學）如何具有導致腰骶部神經叢病變的潛能，並描述了相關的影像學表現。

INTRODUCTION

An aneurysm of the internal iliac artery (IIA) is an uncommon entity and accounts for about 0.008% of all aneurysms in the body.[1] Traumatic pseudoaneurysms of the IIA are considered to be even rarer. Traumatic pseudoaneurysms are usually associated with penetrating injury or blunt trauma,[2] and may be life-threatening. Usually they present with severe haemodynamic instability that warrants immediate surgery or endovascular intervention. Rarely, the

Correspondence: *Dr Soo-Fin Low, Department of Radiology, Universiti Kebangsaan Malaysia Medical Centre, Jalan Yaakob Latiff, 56000 Cheras, Kuala Lumpur, Malaysia.*
Tel: (603) 9145 6149; Fax: (603) 9145 6682; Email: soofinlow@gmail.com

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traumatic pseudoaneurysm remains undetected for years while it gradually expands. Compression and pulsation from IIA pseudoaneurysms may affect adjacent lumbosacral nerve bundles causing lumbosacral plexopathy. We present a rare occurrence of bilateral lumbosacral plexopathy due to an undetected long-standing traumatic pseudoaneurysm of the posterior trunk of the right IIA.

CASE REPORT

A 23-year-old man presented to our institution with bilateral lower limb weakness, numbness, and foot drop in February 2009. He had sustained a stab injury in the right gluteal region 3 years earlier. At that time, there had been considerable blood loss for which he was hospitalised and received a blood transfusion, but he did not undergo any surgical intervention. After 2 weeks he was discharged from hospital, and remained well for 2 years. Five months prior to his latest presentation, he experienced right lower limb pain and numbness. The pain was dull in nature and not relieved by rest. The pain persisted over the ensuing months, but about 2 months later he noticed that the lower limb numbness had become bilateral and there was associated lower limb weakness and foot drop. He also suffered from overflow urinary incontinence and absence of rectal sensation, for which he took rectal enemas regularly and undertook digital evacuation.

Magnetic resonance imaging (MRI) of the lumbosacral spine showed a large well-defined deep-seated pelvic lesion in the sacral region (Figure 1). It had a well-defined hypointense peripheral rim in all sequences and a thin hyperintense inner rim on T1-weighted (T1W) images. The content of the lesion was isointense to hyperintense on T1W and mixed hypointense to hyperintense on T2-weighted (T2W) images. The lesion exhibited peripheral rim enhancement post-Gadolinium with a large non-enhancing core. It appeared to be compressing the lumbosacral plexus and eroding the sacral bone. The lesion was medial to the right IIA. These findings were considered highly suggestive of a large organised haematoma, most likely originating from a right IIA pseudoaneurysm. The T2W hypointense component within the lesion was deemed to represent a retracting blood clot. Other differential diagnoses included venous aneurysm, neurogenic tumour from the lumbosacral plexus, and a retroperitoneal cyst. Pelvic angiography was performed via a catheter and confirmed our initial impression. A small pseudoaneurysm of the right IIA with a patent lumen was detected (the image could not be located for review). In the same setting, he underwent embolisation of the pseudoaneurysm and eight coils were deployed

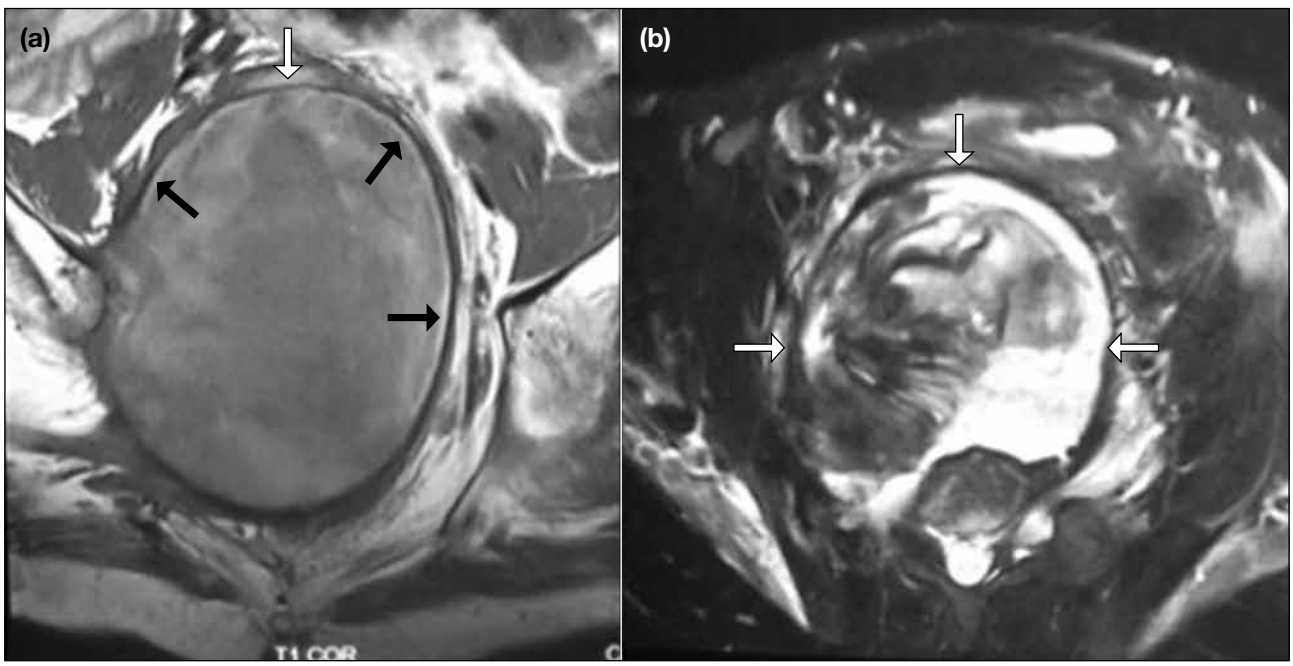

Figure 1. Magnetic resonance imaging: (a) coronal T1-weighted and (b) axial fat-suppressed T2-weighted images of a large pelvic lesion with well-defined hypointense rim (white arrows) and mixed signal intensity within the lesion. The hyperintense inner rim (black arrows) is well demonstrated on T1-weighted images.

to occlude the pseudoaneurysm. After endovascular coiling of the pseudoaneurysm and subsequent physiotherapy, on both sides the weakness in the hips and knees improved, and the lower limb pains and paraesthesia resolved. However, bilateral foot drop and urinary incontinence continued.

The patient presented to our institution again 6 months later because of shooting pains over the medial aspect of his left ankle. Physical examination revealed atrophy of both lower limbs, especially involving the calf muscles. His left hip achieved full power and that of his right hip and both knees was grade 4/5. Power in both ankles and the feet was 0/5. His anal tone was lax. A pelvic radiograph showed a large well-demarcated bony erosion involving the ventral aspect of the sacral bone (Figure 2). Computed tomographic angiography (CTA) of the abdomen was performed to reassess the status of the pseudoaneurysm (Figure 3), and showed a giant thrombosed pseudoaneurysm bounded by fibrous layer and thick granulation tissues (measuring 11 cm at its largest diameter). Absence of contrast pooling within the lumen of the pseudoaneurysm was consistent with total occlusion. Mapping of the course of the IIA using CTA showed the pseudoaneurysm originated from the posterior trunk of the right IIA. There was no opacification of the right superior gluteal artery distal to the pseudoaneurysm, indicating arterial disruption.

Damage to the lumbosacral plexus appeared irreversible, presumably due to chronic compression

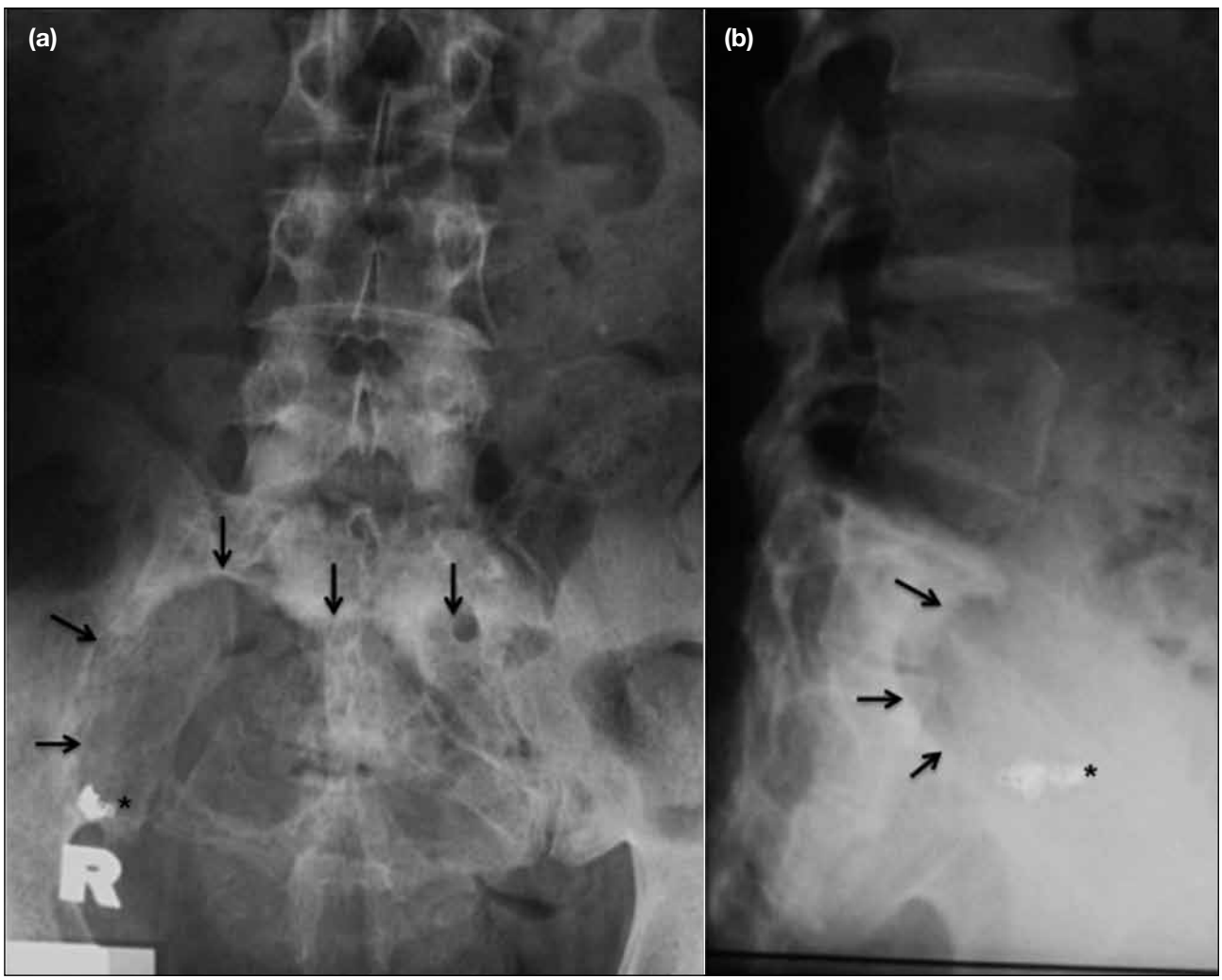

Figure 2. (a) Anteroposterior and (b) lateral views of the lumbosacral plain radiography show a large bony erosion of the ventral aspect of the sacral bone with a narrow transitional zone (arrows), there being no periosteal reaction or matrix of calcification within. The embolic materials (asterisks) are seen at the right antero-inferior aspect of the lytic lesion.

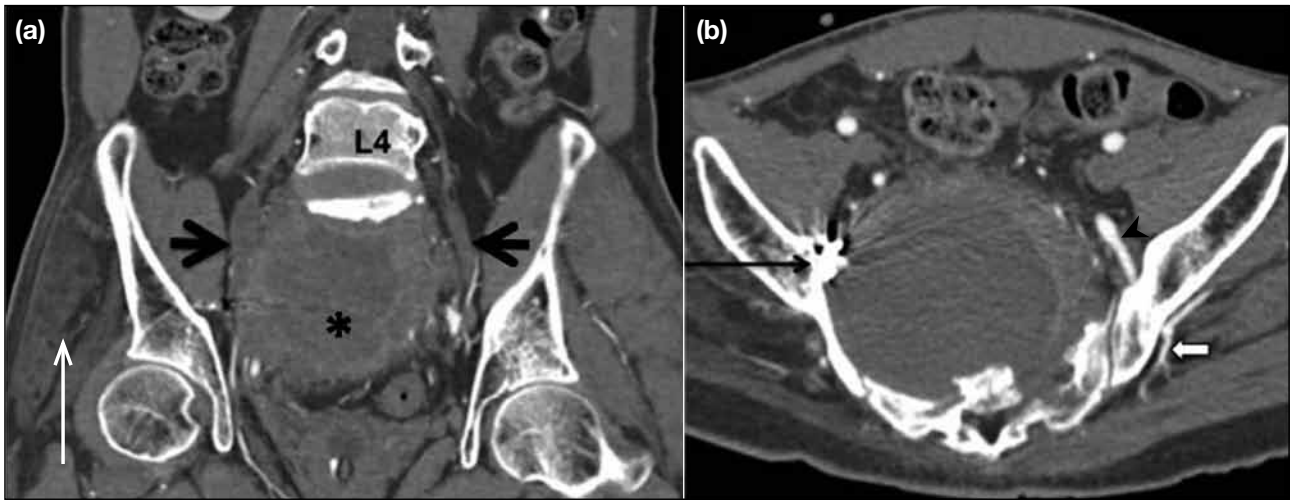

Figure 3. Computed tomographic angiography of the pelvis in the arterial phase. (a) The thrombosed pseudoaneurysm (asterisk) is contained by a thick fibrous layer and surrounding granulation tissues. Bilateral L4 spinal nerves swelling (horizontal black arrows) are evident, especially on the right due to compressive effect from the pseudoaneurysm. There is atrophy and fatty infiltration of the right gluteal muscles (vertical white arrow). (b) There is erosion of the sacrum, neural foramina and right sacro-iliac joint. Embolic material (black arrow) located at the right lateral aspect of the pseudoaneurysm is visible. Normal posterior trunk of the left internal iliac artery (arrowhead) and superior gluteal artery (white arrow) are well-depicted. Non-opacification of the right superior gluteal artery represents arterial disruption at the site of pseudoaneurysm.

by the pseudoaneurysm. The vascular surgeon felt that surgical removal of the thrombosed pseudoaneurysm would not reverse the neurological deficit, nor was the patient keen to have surgery. The patient remains on physiotherapy, is wheelchair bound, and self-manages urinary bladder catheterization and defaecation by recourse to rectal enemas. Occasionally, he experiences shooting pains over the medial aspect of the left ankle.

DISCUSSION

Pseudoaneurysms are also known as false aneurysms or pulsatile haematomatas. Most pseudoaneurysms are secondary to trauma or iatrogenic injury, including those after arterial catheterization. The arterial injury causes full-thickness arterial wall disruption with extravasation of blood into the adjacent tissue, resulting in haematoma formation. The haematoma organises gradually and produces a fibrous layer which constitutes the wall of the pseudoaneurysm. Unlike a traumatic pseudoaneurysm of the thoracic aorta, which is often fatal without emergency surgery, the adjacent connective tissues of the abdomen, pelvis and extremities are able to form a tamponade to contain the haematoma. This phenomenon was seen in the present case. The wall of the pseudoaneurysm was formed by fibrous and granulation tissues with surrounding streakiness from the pelvic fat.

The MR appearance of the haematoma varies according to the age of blood product. Over time, the haematoma may demonstrate a hypointense peripheral rim and hyperintense inner rim on T1W images, producing the so-called concentric ring sign.[3] This finding was noted in the present case, which raised suspicions of vascular lesion of arterial or venous in origin. Very rarely, congenital or acquired weakening of the venous wall may lead to venous dilatation and subsequently gives rise to venous aneurysms, even under normal pressure.[4] We nevertheless considered arterial pseudoaneurysm to be the diagnosis, due to the extreme rarity of the alternatives.

The greater sciatic foramen is the major foramen which allows the exit of vessels and nerves from the pelvis. The posterior trunk of the IIA exits through the foramen with the superior gluteal artery being one of its largest branches. In the present case, we postulate that a deep penetrating injury in the right gluteal region had extended into the pelvis through the greater sciatic foramen. The penetrating injury had most probably lacerated the posterior trunk of the IIA, which led to severe haemorhage at the time of the injury. Whilst emergency surgery and angiography were not being performed immediately after the initial injury, adjacent soft tissue and organisation of the haematoma contained

the haemorrhage and led to pseudoaneurysm formation. We presume that the IIA pseudoaneurysm continued to expand slowly while escaping clinical detection. To our knowledge, this is the first reported case of a solitary pseudoaneurysm causing late-onset bilateral lumbosacral plexopathy. In this instance, a thick organising haematoma with small patent lumen of the pseudoaneurysm could explain its slow growth and late onset of symptoms. Thus, the slowly expanding haematoma appeared to have caused chronic erosion and compression to the pelvic bone and lumbosacral plexus.

Limited instances of unilateral lumbosacral plexopathy secondary to a pseudoaneurysm have been previously reported. Aetiologies of pseudoaneurysms involving IIA have included blunt trauma,[2] pelvic fracture,[5] penetrating injury,[6] and iatrogenic injury following renal transplantation.[7] One patient who developed an inferior gluteal artery pseudoaneurysm following transvaginal needle biopsy for endometriosis has also been described.[8]

Surgical exploration of iliac artery branches is extremely challenging.[9] As such, endovascular embolisation of the pseudoaneurysm is the preferred therapy in many centres, particularly for small lesions. In the acute setting, a percutaneous transarterial approach can be utilised to locate the exact site of a pseudoaneurysm and achieve haemostasis. The adequate use of embolic materials has been shown to reduce the rate of complications, improve outcomes, and facilitate early recovery.[2] Regarding larger pseudoaneurysms causing local compressive effects, surgical decompression is often warranted to relieve neurological deficits and facilitate favourable outcomes. In this case however, surgical excision was not performed as the damage to the lumbosacral plexus was deemed irreversible due to chronic compression by the pseudoaneurysm.

Lumbosacral plexopathy is mainly caused by pelvic trauma and obstetric complications, and is an uncommon entity compared with lumbosacral radiculopathy and peripheral neuropathy. Tumours, aneurysms, and idiopathic neuropathies are infrequent aetiologies of lumbosacral plexopathy. Two theories have been proposed for compressive injury of the nerves: mechanical compression and ischaemia.[10] Imaging plays a crucial role in identifying such causes. Thus, imaging can reveal nerve compression including focal flattening or obliteration of a nerve, a swollen nerve proximal to the compression, a hyperintense MRI of a nerve, muscle oedema after acute denervation, and muscle atrophy with fatty infiltration in the chronic stage.[11] Treatment of lumbosacral plexopathy, its clinical course, and its prognosis are largely dependent on the underlying disorder. The potential of nerve recovery is inversely related to the duration of injury. Moreover, the degree of axonal damage and nerve fibre lengths needing repair both influence the process of regeneration. Regrettably, most patients do not achieve complete functional recovery as recognition of the problem is often unduly delayed.

CONCLUSION

Imaging is warranted whenever any structural cause of a plexopathy is suspected. To facilitate the diagnosis of extraspinal plexopathy, attention should be paid to the extraspinal structures during evaluation of lumbosacral computed tomographies or MRIs. Although IIA pseudoaneurysm is a rare cause of lumbosacral plexopathy, if detected early its consequences can be avoided and / or the lumbosacral plexus can be salvaged from further irreversible damage.

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