

Relapsing Ganglion Cyst of the Hip: A Rare Cause of Disabling Sciatica

Quisto Gangliónico da Anca Recidivante: Uma Causa Rara de Ciática Incapacitante

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Abstract

Ganglion cysts of the hip are uncommon, and often asymptomatic. We report a case of a patient suffering from disabling sciatic nerve compression by a rare and relapsing ganglion cyst of the hip. The 49-year old man had a 3-year history of right lower limb neuropathic pain, and functional impairment, refractory to cycles of oral medication. On examination, he had gluteal tenderness, with no palpable mass, foot drop, absence of Achilles reflex, and steppage gait. Previous investigation suggested diagnosis of refractory sciatica due to lumbar herniary compression. After an unsuccessful bout of conservative treatment, hip magnetic resonance revealed a probable neurogenic cyst compressing the sciatic nerve. He underwent arthroscopic removal of the lesion, with pain relief, but symptoms reappeared 4 weeks later. Subsequent magnetic resonance confirmed relapsing of the cyst, and since then he is experiencing progressive disability. Sciatica is most commonly caused by herniated discs or lumbar spinal stenosis. When conservative treatment fails, other etiologies must be investigated even when there is previous presumptive diagnosis.

Keywords: Ganglion Cysts/complications; Hip Joint; Sciatica/etiology; Sciatica/rehabilitation

Resumo

Os quistos gangliónicos da anca constituem uma entidade incomum e são frequentemente assintomáticos. Relatamos

o caso de um paciente que sofre de compressão ciática provocada por raro quisto gangliónico recidivante da anca. Um doente de 49 anos apresentava história de 3 anos de dor neuropática no membro inferior direito e incapacidade funcional, refratárias a vários ciclos de medicação oral. O exame objetivo evidenciou tensão à palpação da região glútea direita, sem massa identificável, pé pendente, ausência do reflexo de Aquiles e marcha em *steppage*. Os exames complementares previamente realizados sugeriam o diagnóstico de ciática refratária por compressão herniária lombar. Após ciclo infrutífero de tratamento conservador, a ressonância magnética da anca revelou um provável quisto neurogénico a comprimir o nervo ciático. O doente foi, então, submetido a remoção artroscópica da lesão, com alívio da dor, mas os sintomas reapareceram 4 semanas mais tarde. Uma nova ressonância magnética confirmou recidiva da lesão e, desde então, o paciente tem manifestado incapacidade progressiva. A dor ciática é mais frequentemente causada por hérnias discais ou estenose do canal lombar. Quando o tratamento conservador falha, devemos suspeitar de e investigar outras etiologias.

Palavras-chave: Articulação da Anca; Ciática/etiologia; Ciática/reabilitação; Quistos Gangliónicos/complicações

Introduction

Intraneural ganglion cysts are non-neoplastic mucinous cysts present within the epineurium of peripheral nerves.

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The most common location is in the common peroneal nerve, accounting for approximately 60% of cases. The proximal sciatic nerve is only involved in 1.4% of the cases, making intraneural ganglion cysts in this location a very rare condition.¹

The pathogenesis of these lesions has been controversial throughout the years. Several hypotheses have been developed, namely recurrent trauma, intra-neural hemorrhage, mucoid degeneration, and *de novo* formation from hemartomatous cell rests. The most accepted theory today, the “unified articular (synovial) theory”, advocates that an articular branch of a nerve connects to a synovial joint through a labral/capsular defect, serving as a conduit for cystic fluid to pass this joint defect possibly by a one-way valve mechanism. Patients with abnormal joints or joint capsules (previously traumatized, degenerated, or congenitally weakened) may be predisposed to the development of these intraneural cysts. Under pressure, the articular branch may become markedly enlarged. The fluid appears to take the path of least resistance, typically dissecting proximally through the epineurium up the articular branch to the main nerve trunk. Within the parent nerve, the cyst is formed eccentrically within the epineurium, resulting in lateral displacement and even compression of the nerve fascicles.²

Intraneural ganglia are relatively rare cysts located within the epineurium of peripheral nerves.¹ Apart from being rare, intraneural ganglion cysts of the hip are usually small, and asymptomatic.³ When they are large enough, they may become palpable or produce symptoms of neurovascular compromise, which warrants surgical treatment.^{4,5}

Most symptomatic cysts described in the literature extend anteriorly to the hip joint and present as a femoral hernia or as symptoms caused by compression of femoral neurovascular bundles.^{6,7}

We could only find three reports of sciatic pain resulting from compression by a posterior ganglion cyst of the hip,^{4,5,8} but to the best of our knowledge no report of a relapsing lesion is available to date.

In this article, we report the diagnosis of a rare case of sciatic nerve compression by a ganglion cyst located at the level of the greater sciatic foramen, that relapsed after surgery, and is causing pain and progressive disability.

Case Report

A 49-year-old male safety engineer was referred to a Physical and Rehabilitation Medicine (PRM) consultation for a 3-year history of insidious right hip pain, lower limb

intermittent paresthesia and functional impairment, refractory to non-steroidal anti-inflammatory drugs (NSAIDs), and tramadol 150 mg prn. He complained of increasing and burning right thigh and gluteal pain, radiating posteriorly down the lower limb until the posterolateral aspect of the calf, and intermittent paresthesia in the same territory. He rated his pain 6/10 in the Numeric Pain Rating Scale. Symptoms worsened with standing, bending or climbing stairs, and were only partially relieved by bed rest. Sitting, stretching, and pain medication were unable to produce any significant relief. He denied changes in bowel or bladder function, and any constitutional symptoms. Over the previous 6 months, symptoms had progressively worsened, with decreasing lower limb strength and development of foot drop, and at the time of the first consultation he was only able to walk with one crutch, but with frequent falling. His past medical history was unremarkable, namely he denied any prior trauma or hip injury.

On physical examination, he had foot drop and *steppage* gait. Inspection of muscle bulk showed asymmetry in the lower extremities with atrophy of the right thigh, and leg muscles. On palpation tenderness was present on the right buttock (gluteus medius), with no palpable mass. Active and passive hip range of motion was mildly limited bilaterally, but marked pain was elicited on right hip internal and external rotations. Lasègue and Bragard’s tests were both negative bilaterally and did not generate posterior hip pain neither. On strength examination, he presented Medical Research Council (MRC) Scale for Muscle Strength level 3/5 in knee extensors, ankle plantar flexors, and ankle dorsal flexors in the right side, and had normal strength in all other muscle groups tested in the bilateral upper and lower extremities. The right Achilles reflex was absent. The remaining neurovascular examination was unremarkable. Sensation was preserved, and other reflexes were symmetrical in both lower limbs. Peripheral pulses were all palpable and symmetrical, with normal skin color.

He had already been through several investigations in our hospital before the first contact with the PRM department. Results of laboratory tests, including complete blood count, renal function tests, serum electrolytes, and C-reactive protein, were all normal. Plain radiographic examination showed Tönnis grade 1 hip osteoarthritis. Lumbar computerized tomography (CT) scan revealed degenerative changes with narrowing of the right lateral recess at the level of L5-S1 secondary to bulging of the disc, although an unequivocal compression of the right S1 root exit could not be confirmed (Fig. 1).

At this stage, having made the diagnosis of a refractory right sciatic pain, we discussed treatment options with the patient, namely immediate referral to decompressive surgery

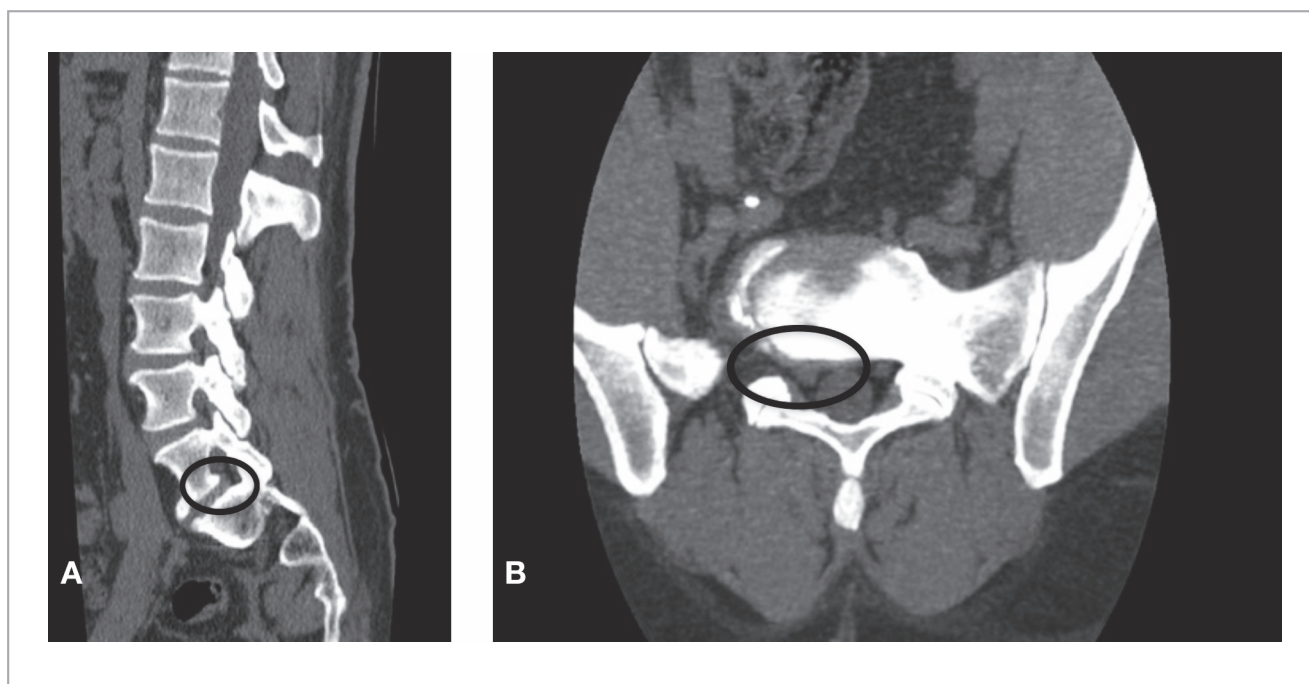


Figure 1 - Lumbar CT at clinical presentation revealing degenerative changes with narrowing of the right lateral recess at the level of L5-S1 secondary to bulging of the disc, with suspected compression of the right S1 root exit. A – Sagittal plane; B – Axial plane.

or conservative rehabilitative treatment in our department. We decided to start with the conservative approach, so we prescribed pregabalin in crescent doses until reaching 900 mg daily and tapentadol 100 mg daily to reduce pain, dynamic ankle-foot orthosis for drop foot (Boxia^o) to improve gait, and physiotherapy and home exercises to improve strength and function. However, after 3 months of treatment, the results remained poor, with progressive gait impairment. Therefore, meanwhile we continued the conservative treatment, we decided to advance in the etiologic investigation, namely extraspinal causes for sciatic pain, by ordering an electromyography test (EMG) of the right lower limb, and magnetic resonance imaging (MRI) of lumbar spine and right hip.

EMG of the right lower limb revealed chronic L4 and L5 radiculopathies. MRI of lumbar spine confirmed CT scan findings, and did not show a clear radicular compression. Then, apart from the already known osteoarthritis, hip MRI (Fig. 2) further revealed a multilocular cystic mass, 3.0 cm in larger size, that was located at the level of the right greater sciatic foramen, and was compressing the sciatic nerve posteriorly in its transition from the pelvis to the hip. This finding was suggestive of a neurogenic sciatic cyst as the cause for this patient refractory sciatica, which prompt us to immediately send the patient to the Orthopedics department. Arthroscopic exploration further confirmed the cyst location at the level of the greater sciatic notch. The

lesion was removed, and neurolysis of sciatic nerve was performed.

In the first postoperative week, pain in the right lower limb had significantly improved, while he maintained paresthesia, neurologic and functional deficits. The patient was discharged, instructed to use bilateral crutches for partial-weight bearing for 3 weeks, and sent back to PRM consultation for further rehabilitation. During this period, he was able to resume indoor ambulation without falling, and only occasionally needed paracetamol p.r.n. to relief pain. However, just one month after surgery, marked pain reappeared. Subsequent MRI performed two months after surgery identified a multilocular cystic mass, 3.5 cm in larger size, located near the sciatic spine, a finding that confirmed relapsing of the sciatic neurogenic cyst (Fig. 2). EMG of the right lower limb performed six months after surgery showed findings of severe lesion of the common trunk of the right sciatic nerve located above the popliteal fossa. We could not find any anatomopathologic examination results in the electronic health record.

The patient continues to maintain regular follow-up in both the Orthopedics and PRM departments, and despite a comprehensive and prolonged rehabilitation program, the patient is still experiencing progressive disability.

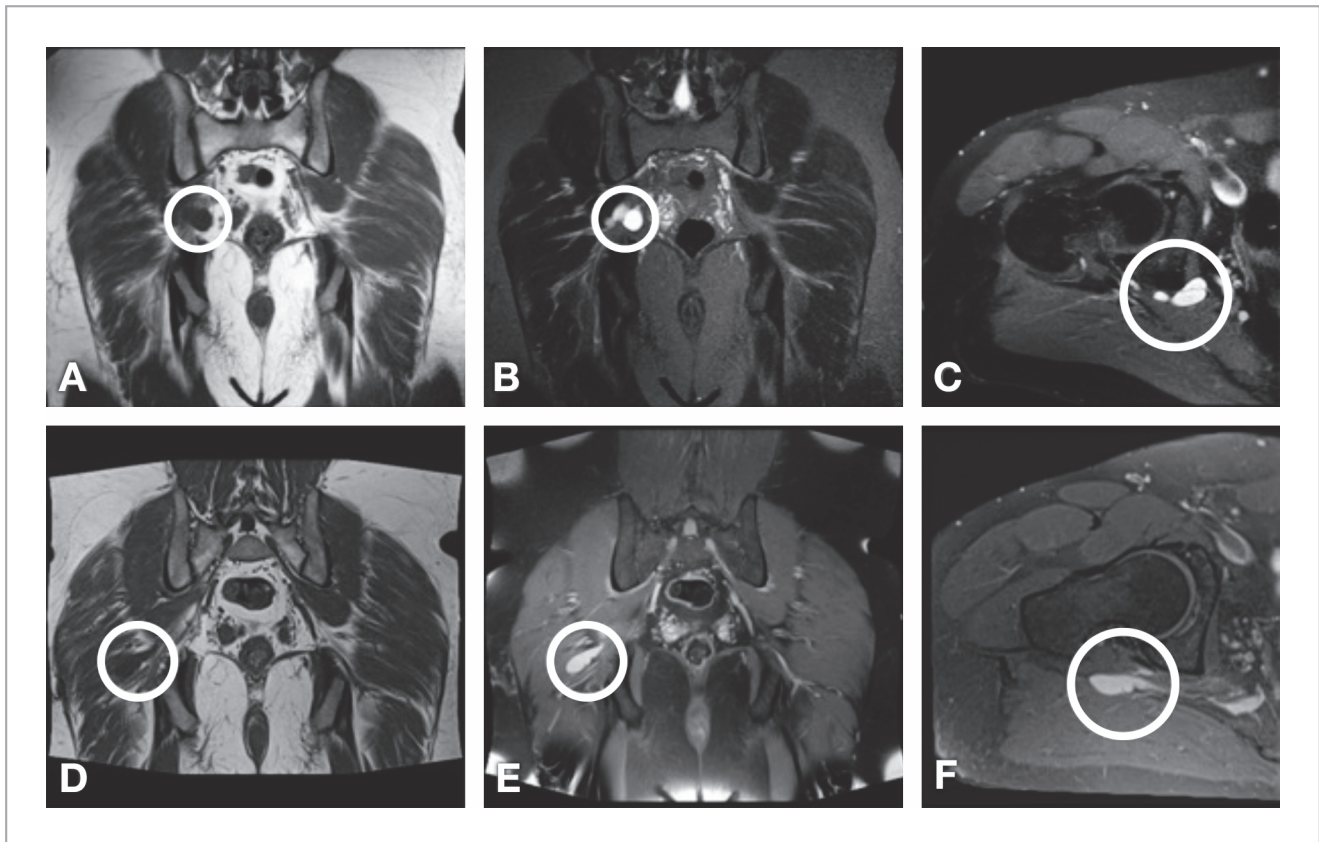


Figure 2 - Preoperative and postoperative right hip MRI.

A, B, and C. Preoperative hip MRI showing a multiloculated mass at the level of the greater sciatic foramen compressing the sciatic nerve (circle). The mass was 3 cm in larger size, and had low signal intensity on T1-weighted and high signal intensity on T2-weighted images. A and B – Coronal planes; C – Axial plane.

D, E, and F. Postoperative hip MRI (performed two months after surgery) showing relapsing of the multiloculated mass, with 3.5 cm in larger size (circle). D and E – Coronal planes; F – Axial plane.

Discussion

Sciatic pain is most commonly caused by herniated discs or spinal stenosis. When conservative treatment fails to improve symptoms, other etiologies have to be suspected, namely extraspinal causes. In the hip, causes for sciatic pain include heterotopic ossification, sacroiliitis, piriformis syndrome, deep gluteal syndrome and cysts.^{9,10} Indeed, sciatic pain caused by a large ganglion cyst of the hip is a very rare diagnosis.^{4,5}

Physical examination is very useful in differentiating extraspinal from intraspinal sciatica, since imaging studies may reveal an asymptomatic disc herniation that may lead to misdiagnosing a symptomatic extraspinal lesion.⁵ Our patient had a history of refractory low back and right thigh and gluteal pain radiating down the ipsilateral lower limb. However, Lasègue and Bragard's tests were both negative, which should have raised our attention earlier. Nevertheless, he also had L5-S1 degenerative disc disease documented

on imaging studies, which initially made the diagnosis of intraspinal sciatica more probable. Additionally, we attributed pain on hip rotation to previous osteoarthritis, thus not giving this finding its due importance, as we would later find out.

After failure to improve with a bout of conservative treatment, further investigation led to the diagnosis of an extraspinal source for the sciatic pain, in this case an intraneural ganglion cyst of the sciatic nerve. In accordance to previous reports of sciatica caused by ganglion cysts of the hip,^{4,5} tenderness on palpation at the point of exit of the sciatic nerve was present. However, we could not find any palpable mass, which together with the lumbar CT Scan findings might have delayed the diagnosis, which was established by means of MRI. There are also differences between the results of the first EMG and the second one performed six months after surgery worth discussion. The first result of L4 and L5 radiculopathies was not corroborated by clinical examination findings, since, for instance, the patellar reflex was preserved and sensation

was normal in the respective dermatomes. The results of the postsurgical EMG are much more reliable. Nevertheless, and although the patient was correctly oriented to the Orthopedics department, we should have confirmed the results of the first EMG. This definitely highlights the importance of an accurate and thorough anamnesis and physical examination as the mainstays of the clinical approach and decision.

The pathogenesis of the cyst reported here could not be definitely established. However, according to the “unified articular theory”, it might be related to hip osteoarthritis.² Our patient had radiologic evidence of hip joint degeneration at presentation, and capsular rents to articular nerve branches might have provided the source of synovial fluid to create and fill the cyst.

Regarding the treatment, after being referred to the Orthopedics department, our patient was submitted to arthroscopic cyst resection and sciatic neurolysis. Cyst resection is indeed the most frequent form of treatment,¹ and sciatic neurolysis has been performed in deep gluteal syndrome with good results after two years of follow-up.¹¹ More radical surgical techniques include nerve resection

with or without nerve grafting or nerve transfer. However, there are no reports of the effectivity of these procedures in the management of sciatic intraneural ganglion cysts. Contrarily to the more traditional surgical approaches, and in consonance with the increasing acceptance of the “unified articular theory”, experts are now recommending treating the articular branch connection and/or the joint (cyst origin) to prevent intraneural cyst recurrence, since failure to disconnect the articular branch or treat the joint pathology was found to be a statistically significant risk factor for cyst recurrence.¹ Although this conclusion is not specific of proximal sciatic nerve ganglion cysts, it might have contributed to cyst relapse in the case presented here.

Intraneural ganglion cysts of the hip are rare clinical entities that may cause disabling sciatic pain. A high index of clinical suspicion and a detailed physical exam are very important to distinguish intraspinal from extraspinal causes of sciatica, and to identify these lesions. Treatment is usually surgical aiming to remove the cyst. Crescent evidence also advocates for the importance of identifying and treating potential factors contributing for the genesis of intraneural ganglion cysts in order to avoid recurrence.

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