

Case Report

Malignant psoas syndrome: first pediatric case report in Mexico

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ABSTRACT

Malignant psoas syndrome (MPS) is a rare, painful condition caused by malignant infiltration of the psoas muscle and adjacent nerves, causing fixed flexion of the leg over the hip. Unfortunately, it is often underdiagnosed. We presented the case of an 8-year-old girl with a clinical history of lumbar embryonal rhabdomyosarcoma and high-grade sarcoma presenting as MPS. To the best of our knowledge, this is the first pediatric case report described in Mexico.

Keywords: Malignant psoas syndrome, Rhabdomyosarcoma, Psoas sign

INTRODUCTION

MPS is a painful, tumor related condition first described by Stevens et al in 1990. It is defined as a painful condition which is often underdiagnosed and difficult to treat.¹

Clinical findings include proximal lumbosacral plexopathy with painful flexion of the hip and a positive psoas muscle- stretch test. In order to achieve diagnosis, clinical suspicion must be supported by either a CT or histopathological analysis of the region confirming tumoral infiltration (Table 1).²

MPS can arise by either direct muscle invasion or metastasis. The associated neuropathy can have multiple presentations depending on the compromised structures and the degree of involvement.

Pain is usually continuous and can be nociceptive due to muscle inflammation and spasm, neuropathic due to

lumbar plexus injury, or both, making it hard to diagnose and manage.³ We presented the first case report of a pediatric patient in Mexico and the experience that we achieve.

Table 1: Diagnostic criteria for malignant psoas syndrome.²

Diagnostic criteria
Clinical evidence of proximal lumbosacral plexopathy (L1- L4)
Painful fixed flexion of the ipsilateral hip with positive psoas test
Evidence of tumoral infiltration by either CT or biopsy

CASE REPORT

This was the case of an eight-year-old girl with a history of a lumbar embryonal rhabdomyosarcoma in 2016 which

was treated with an excisional biopsy followed by nine cycle of chemotherapy and two cycles of radiotherapy.

Periodic controls were made with MRI and PET-CT until 2020 due to the COVID-19 pandemic. Follow-up was restarted in June 2021. Her parents reported a one month increase of volume at the lumbar region and lumbar pain with right gluteal irradiation, compromising limb extension and walking.

Subsequently, the pediatric oncology department reported a volume increase in lumbar region, extending to the right gluteus and ipsilateral limb with inability for extension adding pain, irradiated from the ipsilateral hip to the knee.

Hospitalization extension adding pain, irradiated from the ipsilateral hip to the knee. Hospitalization was indicated for study and pain management, using acetaminophen and ketorolac. Tru-cut biopsy was performed finding a high-grade sarcoma. Simple and contrasted nuclear magnetic resonance (Figure 1) reported the presence of an expansive osteolytic lesion dependent on right iliac bone, characterized by a soft tissue component occupying the right pelvic fossae and gluteal region, with diameters greater than 13.4×12 cm in the axial plane, causing a mass effect with compression and displacement of the hollow pelvic organs, vascular tracts and surrounding muscular structures. This lesion has a heterogeneous signal of predominance isointense in T1, hyperintense in T2 concerning the striated muscle, and intense heterogeneous reinforcement of predominance in the periphery.

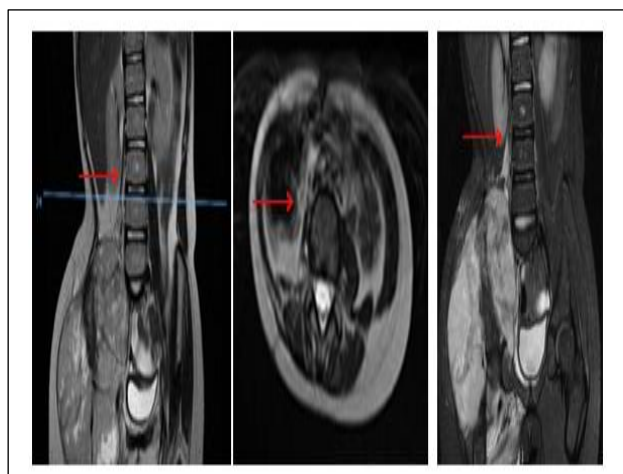


Figure 1: MRI of the lumbosacral spine, showing involvement of the right psoas major.

Algology service was consulted due poor pain control in the lower right limb, despite NSAIDs and acetaminophen. The patient expressed a limitation for the extension of the right leg due to pain and poor response to the first line therapy. Due to clinical findings (Figure 2) and malignant involvement of the right psoas muscle, confirmed radiologically by MRI, the patient was diagnosed with malignant psoas syndrome.

Our intervention started with tramadol 1 mg/kg/dose every 8 hours with rescues of 10 mg IV in case of pain, a maximum of five times in 24 hours. Simultaneously, the intake acetaminophen, ibuprofen at usually dose plus gabapentin 300 mg/day with schedule. In the next days we achieve a better pain control (basal VAS 0/10, VRS absent, with occasionally 5/10 at right leg extension).

For management in home we change to tramadol ten drops orally every 8 hours and rescues of five drops orally in case of moderate and intense pain, in a maximum dosage of three times in 24 hours and continued with gabapentin 300 mg/day without pain exacerbation.

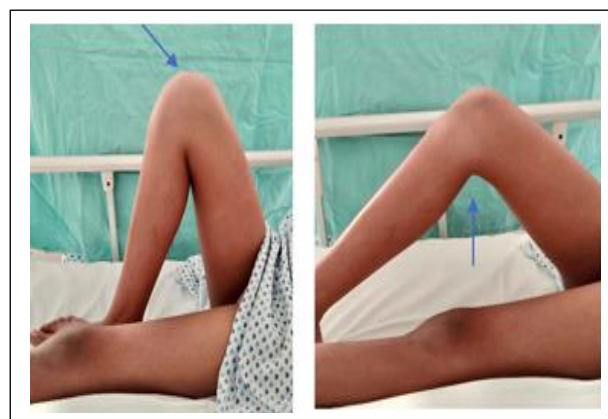


Figure 2: Psoas sign or positive psoas muscle-stretch test.

DISCUSSION

Malignant psoas syndrome can be an underdiagnosed entity and an important cause of pain in the oncologic patient.

To the best of our knowledge this is the first pediatric case documented in Mexico. The most commonly involved nerve structures are L1-L4 (Figure 3).⁴

Approximately 30 cases of MPS have been reported since it was first described.⁴⁻⁶ Most of these corresponded to adult patients; in fact, few pediatric cases are known.⁴ Stevens et al make mention of a 15-year-old boy diagnosed with embryonal rhabdomyosarcoma of the bladder, and Kounami et al documented a 14-year-old girl with a left inguinal tumor, in whom histopathology showed a primary anaplastic large cell lymphoma of the psoas muscle.^{2,5}

MPS is usually found in patients with solid tumors arising primarily from the genitourinary tract, prostate, and colorectal region. However, any primary malignancy or metastasis has the potential to produce this syndrome if the retroperitoneum is involved.⁷

There is no specific treatment consensus for MPS pain management and adverse clinical impact of malignant is further amplified by the average ratio of survival of affected patients (10.7 months; range, 0.5 to 36 month).⁸

Therapeutic options include NSAIDS, steroids, neuromodulators, opioids, local anesthetics and radiotherapy.^{3,8,9}

Optimal treatment of MPS should include early recognition, targeted cancer therapies (comprehended radiation therapy or surgical removal when possible), and evidence-based polymodal pharmacotherapy.⁴

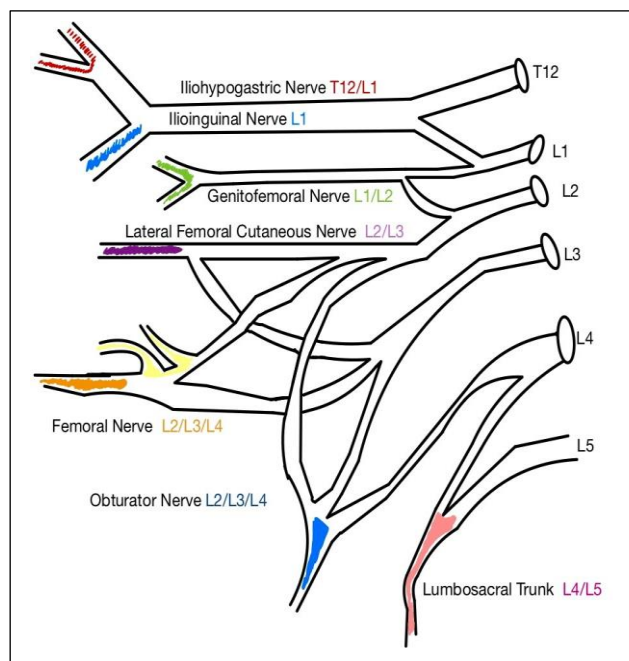


Figure 3: The psoas innervation originates from the lateral aspects of the lumbar spine (T12-L5). These branches run in a common plane within the psoas muscle, and include: iliohypogastric nerve (L1, 60% of cases with T12); ileoinguinal nerve (L1); genitofemoral nerve (L1-L2); lateral femoral cutaneous nerve (L2-L3); femoral nerve (L2-L4); and obturator nerve (L2-L4).¹⁰

CONCLUSION

Pain management is a challenge that really makes a difference in the quality of life of our patients. In some cases, where the cause of pain is shady or with poor response to first line drugs, it's important a meticulously physical examination and a good clinical record with MRI. The pain management have to be optimized in hospitalization and individualized for its ambulatory use.

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