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Case Report

Atypical spindle cell lipomatous tumor in the pelvis causing sciatic nerve compression: A case report ☆

Loïc De Roose, MD[a,], Tina Decorte, MD[a], Bart Eeckhaut, MD[b], An Hofman, MD[b], Jo Van Dorpe, MD, PhD[c], Luc Vanden Bossche, MD, PhD[a]*

[a] *Department of Physical and Rehabilitation Medicine, Ghent University Hospital, Corneel Heymanslaan 10, Ghent 9000, Belgium*

[b] *Department of Physical and Rehabilitation Medicine, Vitaz, Moerlandstraat 1, Sint-Niklaas 9100, Belgium*

[c] *Department of Pathology, Ghent University Hospital, Corneel Heymanslaan 10, Ghent 9000, Belgium*

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ABSTRACT

A 60-year-old woman with chronic lower back pain and deep gluteal pain with radicular symptoms was diagnosed with an atypical spindle cell lipomatous tumor in the pelvis compressing the sacral plexus and sciatic nerve. She underwent successful surgical excision of the tumor, resulting in significant symptom relief. This case highlights the importance of considering rare tumors in the pelvis, such as atypical spindle cell lipomatous tumors, in the differential diagnosis of radicular pain to avoid misdiagnosis.

Introduction

An atypical spindle cell lipomatous tumor is a distinctive adipocyte tumor that mostly originates in subcutaneous tissue of the extremities or trunk, typically in middle-age adults [1–3]. The term "atypical spindle cell lipomatous tumor" represents a relatively new name for the entity previously referred as "spindle cell liposarcoma" [1,2] and at the time of first description regarded as a variant of a "well-differentiated liposarcoma" [2,3]. This new label is justified by the distinctively indolent clinical course, in contrast to the more aggressive nature of a well-differentiated liposarcoma [2].

Intramuscular atypical spindle cell lipomatous tumors located in the pelvis are rare and the diagnosis may be complicated by the presence of atypical symptoms [1,3].

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* Corresponding author.
E-mail address: loic.deroose@uzgent.be (L. De Roose).

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Furthermore, misdiagnosis can lead to inappropriate treatment, with unsatisfactory outcomes. This case report describes a large atypical spindle cell lipomatous tumor in the pelvis compressing the sacral plexus and sciatic nerve, mimicking a radicular syndrome. This case highlights the importance of considering pelvic tumors in the differential diagnosis of patients presenting with radicular pain.

Case presentation

A 60-year-old woman presented to the Department of Physical and Rehabilitation Medicine with complaints of chronic lower back pain and deep gluteal pain on the right side. Over several months, she experienced progressively worsening radiating pain in the right leg, accompanied by neurogenic claudication. These symptoms significantly impacted her daily activities, with prolonged walking and stair climbing exacerbating pain and causing rapid leg fatigue, necessitating frequent rest periods. Her medical history included hypertension and hypercholesterolemia, with no prior relevant treatments or surgeries. There was no significant family or social history.

Clinical examination revealed no postural abnormalities in the axial skeleton, with no signs of scoliosis. Lumbar spine and hip range of motion were within normal limits. Provocative tests for sacroiliac joint dysfunction were negative. The straight leg raise test, along with other radicular irritation tests, also yielded negative results. Muscle strength, sensation, and reflexes were all within normal limits. However, during unilateral stance on the right leg, a slight drop of the left gluteal mass was observed, indicating a positive Trendelenburg sign on the right side. There was no tenderness upon palpation of the lumbar, gluteal, or trochanteric regions.

Lumbar spine magnetic resonance imaging (MRI) revealed a mild degenerative disc disease at L5-S1 without disc herniation (Fig. 1). Subsequently, a multidisciplinary lower back rehabilitation program was initiated, which significantly improved the lower back pain but had no effect on the radiating pain to the right leg and claudication symptoms.

Afterwards, an electroneuromyography (ENMG) evaluation revealed an A-wave upon F-wave stimulation of the right tibial nerve. Pelvic MRI demonstrated a large, intramuscular mass within the gluteus medius and gluteus minimus muscles, hyperintense on T1 (Fig. 2) and hypointense on T2 with fat saturation (Fig. 3). The maximum dimensions were 7.3 × 15.5 × 16.0 cm (antero-posterior x latero-lateral x cranio-caudal), with significant extension of the lesion into the pelvis, causing compression of the sacral plexus and sciatic nerve.

Contrast-enhanced pelvic MRI revealed no clear contrast enhancement of the mass, nor any thickened septations or nodular zones (Fig. 4).

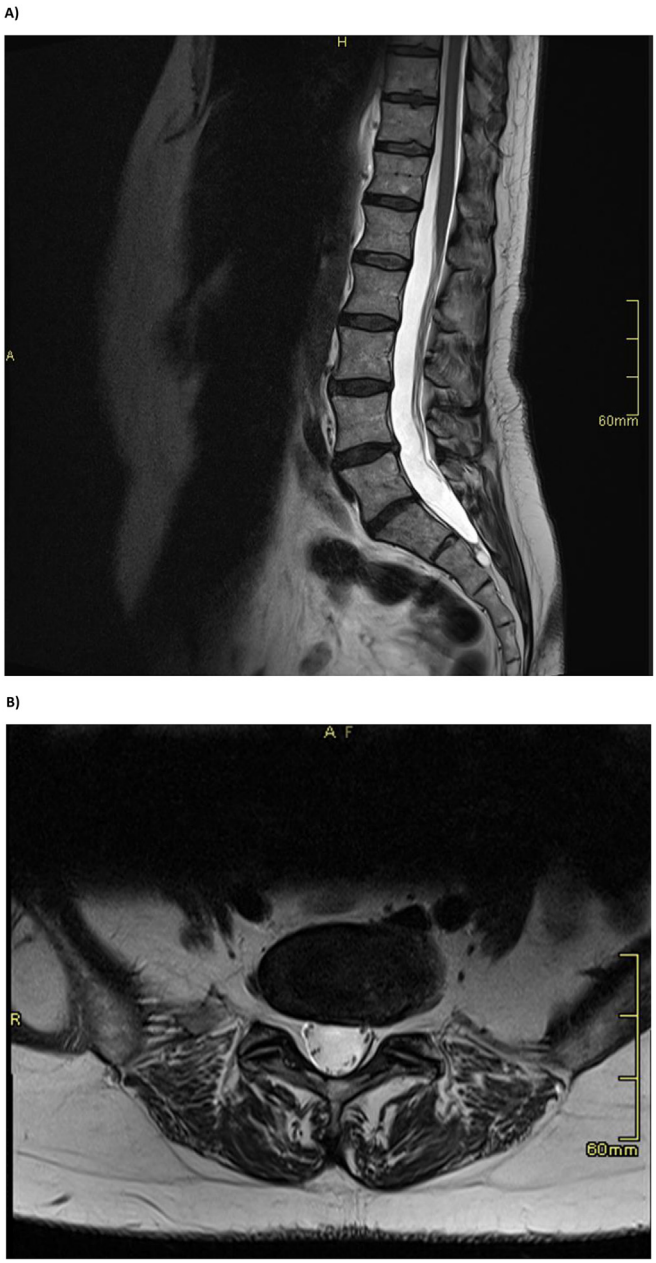

Fig. 1 – Magnetic resonance imaging (MRI) (T2 - sagittal view (A), axial view (B)) of the lumbar spine showing mild discopathy at L5-S1 without disc protrusion or radicular compression.

Treatment and follow-up

A bidirectional resection was performed using a gluteal and extraperitoneal abdominal approach, enabling the tumor to be removed in 1 piece with wide margins. The extracted mass weighed 532 grams/18.77 ounces. There were no complications related to the procedure.

Anatomopathological analysis revealed a morphologically well-differentiated lipomatous lesion with molecular loss of RB1. Given the fibrous septa containing spindle cells with mild atypia (larger and plumper), absence of prominent ropy collagen, size of the lesion, location, and loss of RB1 expression, the best fit was an atypical spindle cell lipomatous tumor.

Postoperatively, the patient reported significant symptom relief within weeks. Subsequent clinical and radiological follow-up was scheduled for 6 months.

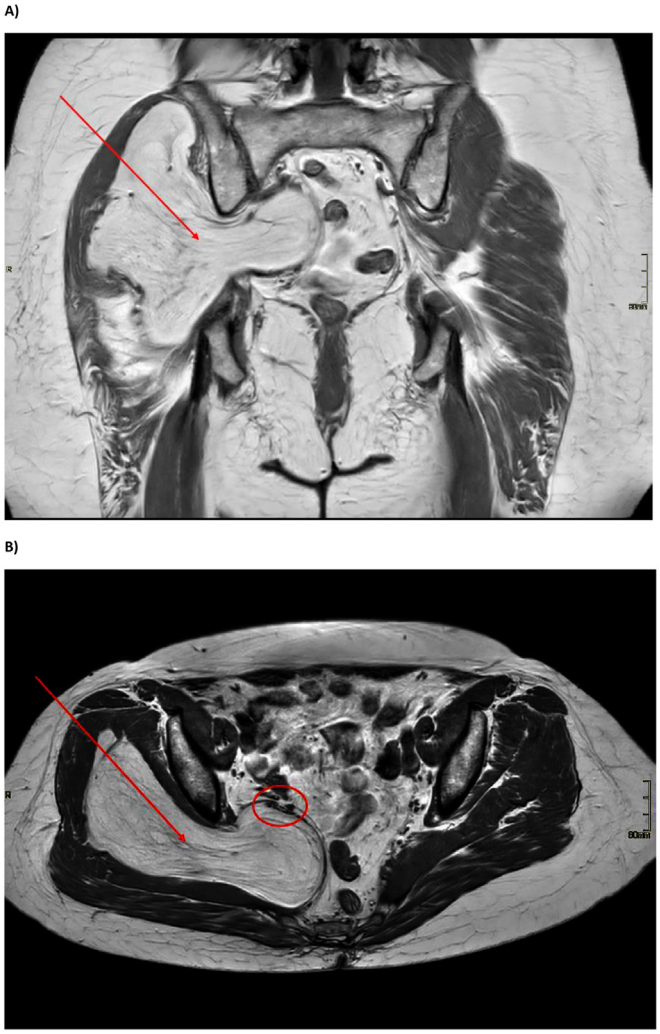

Fig. 2 – MRI scan of the pelvis (T1 - coronal view (A), axial view (B)) showed a large intramuscular mass within the gluteus medius and gluteus minimus muscles of 7.3 x 15.5 x 16.0 cm (antero-posterior x latero-lateral x cranio-caudal)(arrow), with significant extension of the lesion into the pelvis, causing compression of the sacral plexus and sciatic nerve (circle).

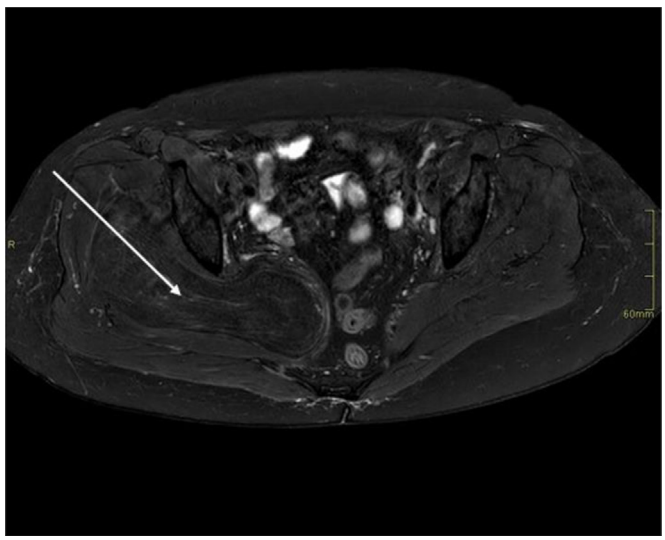

Fig. 3 – MRI scan of the pelvis (T2 with fat saturation – axial view) showing a hypo-intense gluteal mass (arrow).

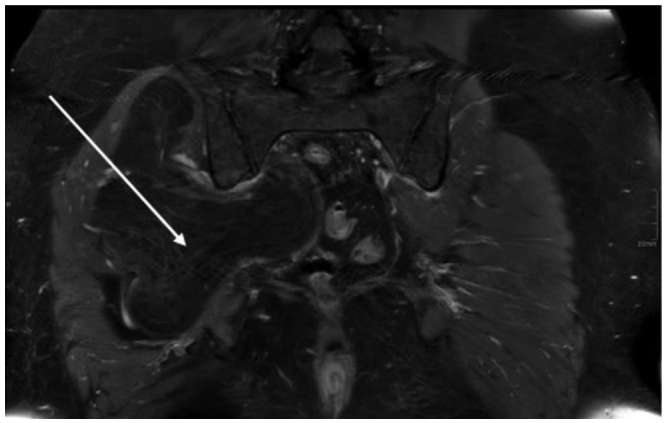

Fig. 4 – MRI scan of the pelvis with gadolinium contrast (T1 with fat saturation – coronal view) showed no clear contrast enhancement of the mass, nor any thickened septations or nodular zones (arrow).

Discussion

Lower back pain may manifest with radicular symptoms resulting from lumbosacral nerve root pathology [4]. Lumbar disc herniation is a predominant etiology of lower back pain, sciatica and radicular leg pain, and is typically the first diagnosis considered [4]. In this case, lumbar spine MRI did not provide a definitive explanation for the radiating pain in the right deep gluteal area and right posterior leg. Consequently, additional testing was conducted, including ENMG and pelvic MRI, which revealed a significant mass in the gluteal muscles compressing the sciatic nerve. This case underscores the importance of a thorough diagnostic evaluation when initial imaging does not correlate with clinical symptoms.

In 2010, Mentzel et al. introduced the term "atypical spindle cell lipoma" for the first time, suggesting that these neoplasms likely represent a distinct entity closely related to spindle cell lipomas, rather than a morphological variant of an atypical lipomatous tumor or well-differentiated liposarcoma [3]. In 2020, the World Health Organization (WHO) recognized atypical spindle cell lipomatous tumor as a novel entity within the classification of benign or low-grade adipocytic neoplasms (Table 1) [2]. This entity primarily affects middle-aged adults, with a peak incidence in the sixth decade of life, and exhibits a slight male predominance [1,5]. Unlike classic spindle cell or pleomorphic lipomas, atypical spindle cell lipomatous tumors have a wider anatomical distribution, with the extremities being the most common sites of involvement, while the abdominal wall and groin are less frequently affected [5]. Lesions in the posterior neck or shoulder regions are more likely to be diagnosed as classic spindle cell or pleomorphic lipomas [6].

For palpable lesions, ultrasound can serve as an initial imaging modality; however, magnetic resonance imaging (MRI) is regarded as the gold standard for further assessment [7]. In this case, the MRI findings including the absence of thick septa, nodular or globular mass-like areas, contrast enhancement, along with a fat composition exceeding 75%, suggested a benign soft-tissue tumor. However, the lesion's substantial size and the patient's age raised concerns regarding its potential malignancy. Bird et al. identified tumor size exceeding 10 cm, thigh location, and patient age over 60 years as clinical risk factors for an atypical lipomatous tumor or well-differentiated liposarcoma [8]. MRI features indicative of malignancy include large lesion size, thick septa (typically >2 mm), nodular or globular or nonadipose mass-like areas, contrast enhancement, and a reduced fat composition (<75%) [9,10].

When feasible and clinically indicated, core needle biopsy can be considered for histological assessment, particularly when a sarcoma is suspected [11]. In the present case, given the lesion's compression of the sacral plexus and sciatic nerve and the initial suspicion of a benign tumor, we opted for direct excisional biopsy. The lesion was completely removed with clear margins, minimizing aesthetic and functional impact.

Histopathologically, atypical spindle cell lipomatous tumors exhibit a morphological spectrum of benign adipocytic

Table 1 – The 2020 WHO classification of soft tissue tumors, entities characterized by adipocytic differentiation (2).

Adipocytic tumors
Benign
Lipoma and lipomatosis
Lipomatosis of nerve
Lipoblastoma and lipoblastomatosis
Angiolipoma
Myolipoma of soft parts
Chrondroid lipoma
Spindle cell/pleomorphic lipoma
Atypical spindle cell/pleomorphic atypical lipomatous tumor
Hibernoma
Intermediate (locally aggressive)
Atypical lipomatous tumor
Malignant adipocytic tumours
Well differentiated liposarcoma: lipoma-like, sclerosing, inflammatory
Dedifferentiated liposarcoma
Myxoid liposarcoma
Pleomorphic liposarcoma
Myxoid pleomorphic liposarcoma

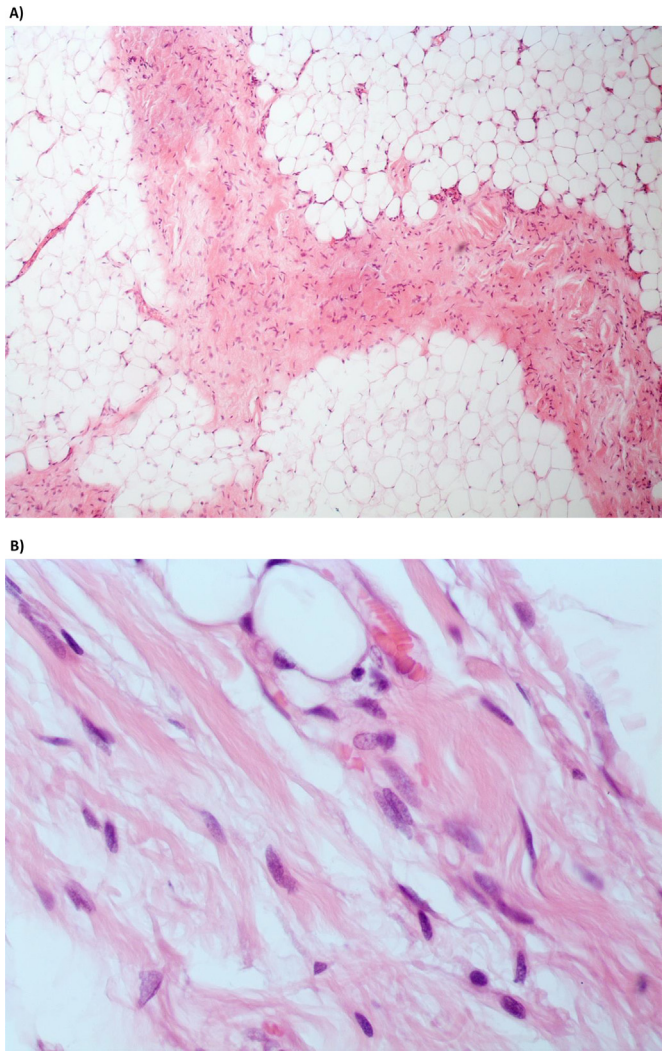

Fig. 5 – Histopathological images. (A) Low-power view showing a well-differentiated lipomatous tumor, characterized by fibrous septa containing spindle cells with mild nuclear atypia. (B) High-power view highlighting slightly atypical spindle cells, predominantly located within the fibrous septa.

neoplasms, reflecting varying proportions of spindle cells, adipocytes, lipoblasts, and extracellular matrix components [1,2]. Notably, RB1 expression is lost in approximately 60% of cases [2,12]. In this case, the lesion comprised mature adipocytes with slight size variability and no significant nuclear atypia. Lipoblasts were absent. Both fine and broad septa were observed, accompanied by an increased number of spindle cells, with mild atypia, occasionally displaying a plumper nucleus (Fig. 5). There was no presence of ropy collagen. FISH-analysis showed RB1 deletion (monosomy 13q). Amplification of MDM2 was absent.

The histological differential diagnosis of atypical spindle cell lipomatous tumors includes several entities:

- Spindle Cell Lipoma: Characterized by a limited anatomical distribution, typically affecting the upper back, shoulders, head, and neck, with the presence of distinctive ropy collagen bundles.
- Atypical Lipomatous Tumors/Well-Differentiated Liposarcoma: These tumors exhibit unusual hyperchromatic stromal cells within a densely collagenous stroma and demonstrate nuclear expression of MDM2 and CDK4.
- Other Entities: These include diffuse neurofibroma, myofibroblastoma, cellular angiofibroma, fat-forming solitary fibrous tumor, pleomorphic liposarcoma, and dedifferentiated liposarcoma [2,6,13].

Complete surgical excision with clear margins is the recommended treatment for atypical spindle cell lipomatous tumors. Incomplete excision may result in local recurrence in approximately 10%-15% of cases [1,2]. In case of incomplete removal, options include re-excision and close clinical and radiological follow-up for potential local recurrence. Regular imaging surveillance is not required after complete resection [1]. There is no associated risk of dedifferentiating or metastasis, and therefore, adjuvant therapy is not indicated [14].

Conclusion

Despite the significant challenges in diagnosing and differentiating radicular pain, clinicians should remain vigilant in considering this condition and exclude space-occupying lesions in the pelvis when evaluating patients with radicular pain. This case highlights the atypical presentation of an intramuscular atypical spindle cell lipomatous tumor in the pelvis, which caused sciatic nerve compression. Complete surgical excision is the recommended treatment for atypical spindle cell lipomatous tumors. There is no associated risk of dedifferentiation or metastasis, and thus, adjuvant therapy is not required.

Patient consent

Complete written informed consent was obtained from the patient for the publication of this study and accompanying images.

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