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Case Report

Rapidly Growing Leiomyoma Mimicking Schwannoma of the Saphenous Nerve in the Lower Extremity: An Unusual Case Report

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Abstract

Leiomyomas and schwannomas are both types of rare benign soft tissue tumours. Leiomyomas are more commonly found in the lower limbs than in the upper extremities, while schwannomas are rare peripheral nerve sheath tumours that can occur in different anatomical regions. However, they rarely occur in the saphenous nerve. This case study presents a 41-year-old female patient with a solitary mass lesion located deep in the soft tissue of the anteromedial lower extremity. The physical examination revealed a palpable, elastic-hard, mobile and non-tender mass. Magnetic resonance imaging (MRI) showed an oval-shaped subcutaneous mass on contrast-enhanced T1-weighted sections. The initial MRI images suggested a schwannoma, but the tumour was later confirmed to be a leiomyoma after total enucleation. An immunohistochemical study was performed for differential diagnosis. Solitary mass lesions in the lower extremities can be mistaken for various types of tumours and misdiagnosed and require histopathological examination and good radiological imaging for differential diagnosis. Complete surgical excision is usually a safe and effective treatment for leiomyomas.

Keywords: Leiomyoma, Lower Extremity Tumour, Neoplasm, Schwannoma, Soft Tissue Tumour

Introduction

Leiomyomas are benign tumours of smooth muscle origin and account for 4.4% of all benign soft tissue neoplasms[1]. They are classified as tumours of the skin, vascular system and deep soft tissues. Leiomyomas rarely occur in the extremities and are more common in the lower extremities than in the upper extremities[2]. The number of cases of deep soft tissue leiomyomas reported in the literature is extremely low. Leiomyomas that occur outside the uterus and gastrointestinal tract are also extremely rare. Leiomyomas most commonly occur in women in their third and fourth decades of life[3]. They were first described by Virchow in 1854[4,5].

Extremity leiomyomas are classified into two types: superficial and deep tumours[6]. Deep extravascular soft tissue leiomyomas are rare, with only a few cases reported in the literature. Leiomyomas in soft tissue are usually small and are found in cutaneous or subcutaneous locations. However, lower extremity leiomyomas can be mistaken for benign peripheral nerve tumours known as schwannomas.

The peripheral nervous system consists of peripheral nerves and ganglia that originate from the brainstem or spinal cord[7]. Schwannomas originate from the Schwann cells that normally form the bundles of myelinated axons in the peripheral nerves and these tumours grow along the longitudinal axis of the nerve[8]. Schwannomas account for 1–3% of peripheral nerve tumours and often occur in the head, neck and upper extremities[9,10]. In the lower extremities, schwannomas occur most commonly in the sciatic and tibial nerves[11,12].

Schwannomas have the highest incidence between the fourth and sixth decades of life and affect both genders equally[13]. Schwannomas that cause a mass effect may elicit compressive neuropathy. Patients with schwannomas usually present with a slowly growing mass on the flexor surfaces of

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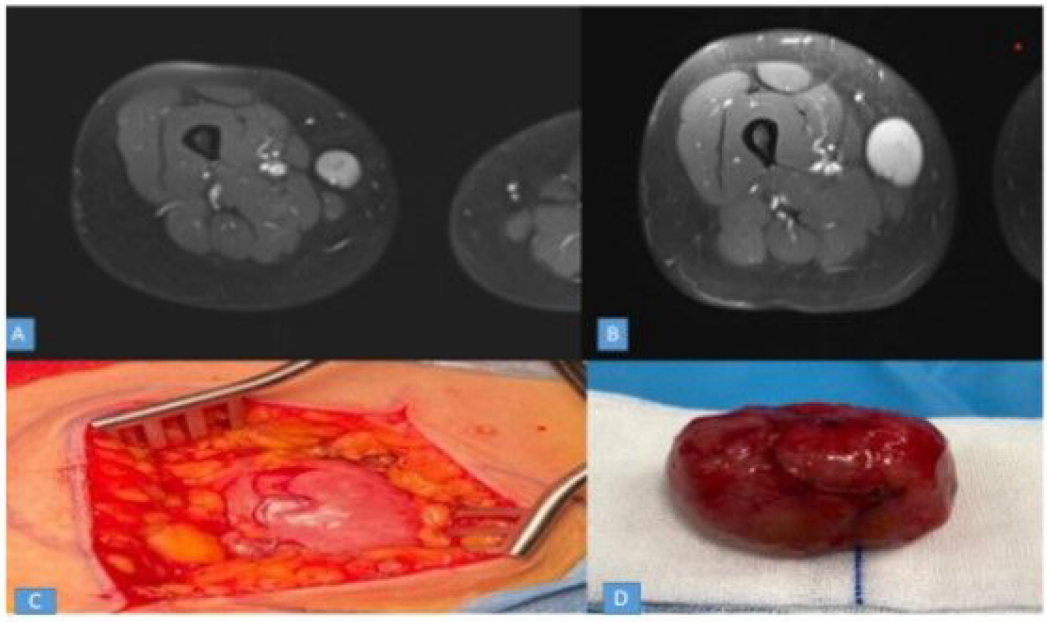

Figure 1. **A.** Second quarter of 2023, **B.** Last quarter of 2023; Axial magnetic resonance imaging showed that the mass had higher signal intensity on contrast-enhanced T1-weighted sequence. **C.** Intraoperative photograph of the tumor and the saphenous nerve. **D.** Gross appearance of the glistening tan-white tumor.

their extremities. However, solitary schwannomas and lower extremity leiomyomas can both present as asymptomatic masses, palpable growing masses, or pain. Both tumours can also present different clinical manifestations, depending on the anatomical site at which the tumour appears.

This study presents a rare case of a soft tissue leiomyoma mimicking a solitary schwannoma in a middle-aged female patient. Written permission was obtained from the patient for publication of the case.

Case Presentation

A female patient, aged 41, was admitted to our clinic with a history of a slow-growing, painful mass in the anteromedial part of her right upper and lower leg. The patient experienced electric shock-like pain on palpation and percussion of the mass. Magnetic resonance imaging (MRI) revealed a distinct mass on the saphenous nerve, located approximately 18 mm deep in the medial thigh. The mass exhibited diffuse enhancement and smooth contours. Comparison to an examination conducted in June 2023 revealed an increase in the lesion volume, with previous dimensions of 40×35×25 mm and current dimensions of 50×38×32 mm. The lesion had doubled in size from approximately 15 cc to 30 cc (Figure 1).

Surgical treatment was decided based on the clinical findings of the mass, and the patient underwent surgery under general anaesthesia. The tumour was macroscopically exposed, and the entire mass was removed en bloc. It was initially evaluated as a schwannoma due to its smooth surface and white colour, but was sent for confirmation by histopathological examination.

Histopathological examination

A tissue sample, measuring 4×4×3.5 cm in its largest diameter, was divided into five pieces and placed in five cassettes (Figure 1). The immunohistochemical study was conducted in our laboratory using a fully automated Ventana Benchmark XT instrument (Arizona, USA). The tissues were fixed in buffered formaldehyde and an internal control for antibodies was employed. Antigen retrieval was performed using an automatic system. Ten different immunohistochemical examinations were conducted for differential diagnosis. Negative (-) staining was observed for CD34, S100, Sox10, CD117 and DOG1. Actin was positive (+), desmin was palely positive (+) and H-caldesmon showed diffuse staining (+) (Figure 2). Evaluation of INI-1 and Ki67 indicated artefactual staining at 2–3%. A diagnosis of leiomyoma was made based on histopathological examination of the specimen by a double-blinded pathologist with at least 10 years of experience. The surgical margins were clear.

Discussion

The histopathological examination of our case led to the conclusion that her tumour was a benign mesenchymal

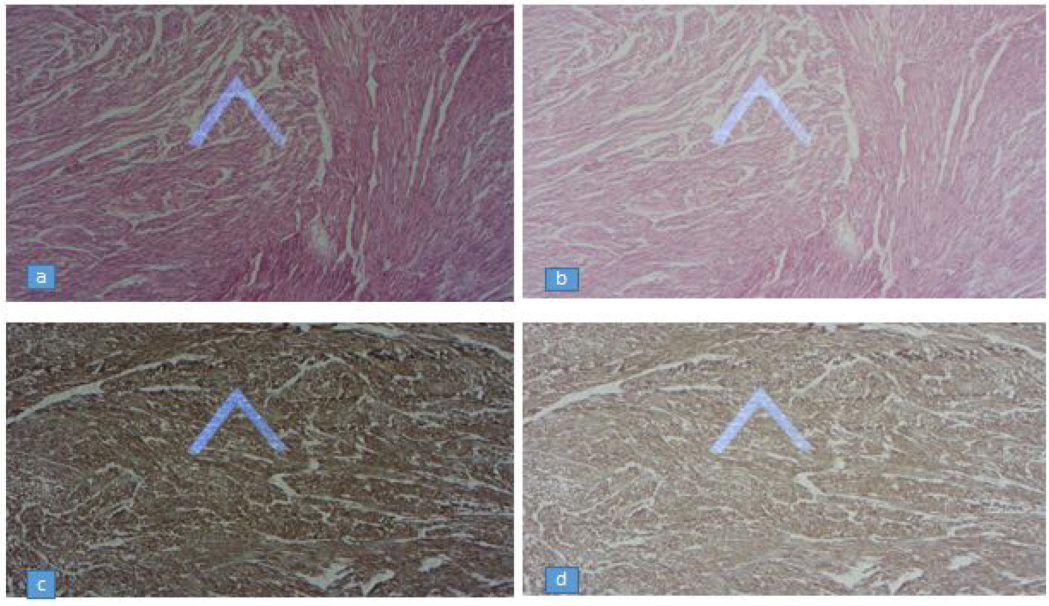

Figure 2. A-B: (1x100) Diffuse nodular tumor with spindle nuclei and cytoplasm borders not visible in Hematoxylin&Eosin histochemical study. **C-D:** Diffuse positive staining with H-caldesmon immunohistochemical marker (1x100).

neoplasm, specifically a leiomyoma. Deep extravascular soft tissue leiomyomas like this one are rare, particularly in the lower extremities. They are typically single and well-circumscribed and can be mistaken for other solitary mass lesions in the extremities, with differential diagnoses that can include lipoma, leiomyosarcoma, schwannoma, neurofibroma, haemangioma and soft tissue giant cell tumour of the tendon sheath. The pathogenesis of leiomyomas remains unclear. Deep leiomyomas can originate from undifferentiated mesenchymal cells or smooth muscle remnants[14]. These tumours may also possibly arise from the smooth muscles in the walls of blood vessels[15].

In our case, the initial diagnosis was schwannoma, based on the patient's symptoms of pain and the presence of a palpable mass lesion, as well as the MRI report. However, the histopathological examination revealed a diagnosis of benign mesenchymal neoplasia, specifically leiomyoma. A point worth noting is that schwannomas are relatively common in individuals aged 20 to 50, with no significant gender differences[8]. By contrast, our patient was a 41-year-old female.

Histopathological examinations of schwannomas often show that they are encapsulated by fibrous tissue. Pathology slides reveal that classic schwannomas exhibit two distinct cell patterns: a cellular, compact Antoni A pattern and a loose, cobweb-like Antoni B pattern. Another typical histological feature of Antoni A schwannomas is the presence of 'Verocay bodies', which are cellular clusters with eosinophilic cytoplasmic cell extensions in the centre and a palisadic nuclear arrangement around them. In our case, we performed 10 different immunohistochemical examinations for differential diagnosis. The diagnosis of leiomyoma was confirmed based on the observed negative staining for CD34, S100, Sox10, CD117 and DOG1. Additionally, actin was positive, desmin was pale positive, and H-caldesmon showed diffuse staining. INI-1 and artefactual staining, as well as Ki67 staining, were evaluated at 2-3%. All these immunohistochemical results confirmed the diagnosis of *leiomyoma*.

An examination of the literature indicates that leiomyomas can appear in various locations, including the uterus, stomach, heart and extremities[16-18]. However, deep extravascular soft tissue leiomyomas are rare, with only a few cases reported in the literature. One case presentation by Bommireddy described a 25-year-old female with a slowly enlarging mass in the right forearm that had been present for 6 months[1]. The patient reported occasional dull pain over the lesion. Similarly, Cigna et al. reported a 69-year-old woman with a painful, slowly growing mass in the medial malleolus of her right leg, which they diagnosed as a large subcutaneous vascular leiomyoma[19].

According to Cigna et al., vascular leiomyomas or angioleiomyomas are benign, solitary tumours that consist of smooth muscle cells originating from the muscle layer of the vascular walls[19]. Pain is the most common symptom, occurring in 60–75% of cases. Diagnosis is made by identifying histopathological features using various staining techniques. Treatment usually involves surgical excision. Due

to the biological characteristics of leiomyoma, the recurrence rate is low, ranging from 19–21%.

Leiomyomas and schwannomas can both grow along the nerve sheath and cause similar local symptoms, such as swelling, pain and paraesthesia, due to the mass effect and mechanical compression of the nerve and surrounding structures. Our patient also complained of local pain and a palpable mass lesion. According to Stout et al., contraction of the smooth muscles in these tumours may cause spontaneous paroxysmal pain[15]. The decision for surgical treatment of our patient, who was under clinical follow-up, was based on the progressive growth of the mass within the same year. Imaging techniques for the diagnosis of leiomyoma are limited, and although ultrasonography is frequently used, we imaged the lesion with MRI. The volume of the lesion was approximately 15 cc in the second quarter of 2023, but had increased to approximately 30 cc in the last quarter.

Complete surgical excision is generally a reliable and safe treatment option for leiomyomas, with positive outcomes observed in the majority of published case reports[19]. In our case, we performed a linear surgical incision, dissection and gross total excision of the tumour. The patient's symptoms improved acutely, and no local recurrence was observed during the short-term (first 3 months) follow-up. The patient's intermittent outpatient clinic checks continue.

The main limitation of this study is the relatively short follow-up period of three months, which is not sufficient to obtain a comprehensive evaluation of the results and effectiveness of the therapeutic interventions.

Conclusion

Solitary leiomyomas, which are rare, especially in the lower extremities, can be misdiagnosed as different tumour types. Schwannoma of the saphenous nerve is a rare tumour in the lower extremity. However, it may be the first pathology that comes to mind when encountering a lower extremity tumour near this location in neurosurgery practice.

Authors' contributions

ŞG and YG contributed equally to the development of the manuscript. ŞG and KK contributed equally and provided expert oversight for the completion of the manuscript. ŞG, KK and FT provided images and modified them. ŞG and YG provided input on the preparation of the manuscript and later modified the article. FT contributed to the histopathological examination All of the authors accepted responsibility for the integrity of the data analysis. Also, all authors read and approved the final version of the manuscript.

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