

Easily misdiagnosed intramuscular hemangioma: a case report

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Abstract

An intramuscular hemangioma is a benign vascular tumor that often occurs in the lower extremities. We herein report a rare case of an intramuscular hemangioma that occurred in the gluteus medius muscle and was misdiagnosed as lumbar disc herniation. A 36-year-old woman presented with incidental and infrequent pain of the left buttock. She was diagnosed with lumbar disc herniation and underwent treatment. Although her pain was slightly relieved, relapse soon occurred. X-ray examination and magnetic resonance imaging revealed a mass in the gluteus medius muscle. The mass was suspected to be a malignant tumor and was therefore resected. The final diagnosis was an intramuscular hemangioma. Her pain completely disappeared thereafter and did not recur. Patients with intramuscular hemangiomas usually have no specific symptoms; therefore, this tumor is often misdiagnosed. When a satisfactory treatment effect is not obtained, the diagnosis should be reassessed in a timely manner.

Keywords

Intramuscular hemangioma, gluteus medius muscle, misdiagnosis, vascular tumor, benign, case report

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Introduction

A hemangioma is a tumor that originates from vascular tissue and may develop in almost any part of the body, including the skin, subcutaneous tissue, muscle, splanchnic tissue, and bone. Intramuscular hemangiomas represent <0.8% of all hemangiomas.¹ However, they are the most common type of deep intramuscular

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tumor^{2,3} and are more common in people aged <30 years. Intramuscular hemangiomas are usually asymptomatic at the initial stage. When the tumor becomes enlarged, however, it may compress and push the adjacent muscles and nerves, resulting in various symptoms. Clinical lesions can involve any skeletal muscles but are most common in the lower limbs. Intramuscular hemangiomas become more difficult to treat as they enlarge. The early symptoms of intramuscular hemangiomas are nonspecific and not obvious, making them difficult to detect; thus, intramuscular hemangiomas are often misdiagnosed. We herein describe a patient who presented with incidental and infrequent pain of the left buttock accompanied by radiating pain in the back of the thigh. She was diagnosed with lumbar disc herniation and began long-term treatment. However, her pain persisted and frequently recurred.

Case report

A 36-year-old woman presented with a 5-year history of incidental and infrequent pain of the left buttock accompanied by radiating pain in the back of the thigh. She had previously been a saleswoman and now had mild obesity. Five years before presentation, she had begun to feel pain in the left buttock. The pain was tolerable and occurred six to eight times per year. One year before presentation, she sought medical advice. No abnormalities were found in a physical examination of the spine and left hip, the range of motion of the spine and hip was normal, and her gait was normal. However, the straight-leg raise test result was suspicious: during the test, she felt some discomfort in the left buttock, but she was unable to clearly describe the pain and did not feel obvious radiating pain or numbness in the lower limbs. Considering these findings, the doctor suggested that she reduce daily movement, rest,

lie down, and undergo physiotherapy including traction, magnetic thermotherapy, and application of a waistband-like device to help stabilize the lumbar spine. Each time she underwent these treatments, her pain was slightly relieved. However, when she returned to work, especially when she was required to walk or stand for a long period of time, the pain recurred. As long as she continued the above-mentioned physiotherapy along with medication, her pain was improved but did not disappear.

Six months before the writing of the present report, she was referred to an orthopedic surgeon, who requested a pelvic X-ray examination (Figure 1(a)) and magnetic resonance imaging (MRI) without contrast (Figure 1(b)–(d)). These examinations showed that the density of the left ilium was not uniform, the edges were fuzzy, high-density shadows were present in the soft tissue, lumpy mixed-intensity abnormal signals were present in the gluteus medius and gluteus minimus muscles with unclear boundaries, and bone destruction was present in the ilium. These findings were similar to the imaging features of a malignant tumor. Moreover, single-photon emission computed tomography also revealed findings suspicious for bone metastasis of a malignant tumor.

After embolization of the tumor's main blood supply, debulking surgery was conducted. The operation was performed with the patient under general anesthesia in the lateral position. A curved incision was made in the buttock, extending along the direction of gluteus maximus muscle fibers, and the gluteal muscle fibers were separated to expose the tumor. The tumor was present within the gluteus medius muscle and was very large, measuring about $15 \times 10 \times 5 \text{ cm}^3$. Calcified lesions were present in the gluteus maximus and gluteus medius muscles above the sciatic nerve but were not in direct contact with

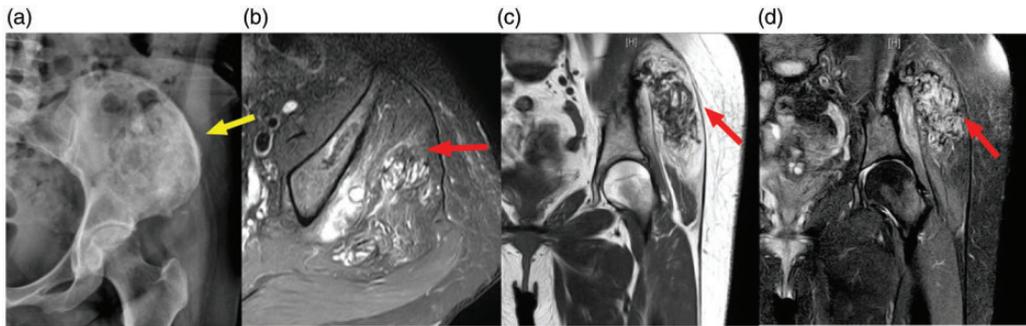


Figure 1. (a) X-ray shows that the density of the left ilium is not uniform, the edge is fuzzy, high-density shadows are present in the soft tissue, and calcified lesions are present near the left ilium (yellow arrow). (b) Magnetic resonance imaging shows a soft tissue mass in the left buttock (red arrow). (c, d) Lumpy mixed-intensity abnormal signal in the gluteus medius and gluteus minimus muscles with an unclear boundary and bone destruction of the ilium (red arrow).

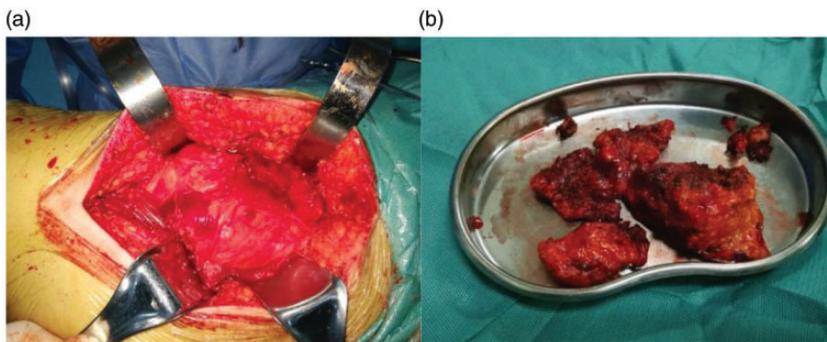


Figure 2. (a) Gross appearance of the tumor, with no obvious capsule or boundary. (b) Excised tumor tissue.

the sciatic nerve. After careful separation and exposure, the tumor was removed in its entirety (Figure 2). The gluteus medius muscle exhibited degeneration and atrophy due to compression and erosion by the tumor. The amount of intraoperative blood loss was much less than that predicted preoperatively. No postoperative complications or gluteal weakness occurred. Immunohistochemical staining of pathological sections led to a definitive diagnosis of an intramuscular hemangioma with no evidence of malignancy (Figure 3). After the operation, the patient was referred for rehabilitation exercise and

gradually returned to work. During the first week postoperatively, she was allowed to perform only isometric contraction of the lower limb muscles each day without hip movement. The next week, she was encouraged to move the hip, first passively and then actively and first with flexion and then abduction; aquatic therapy was then added and she was thus able to gradually recover her muscle strength. About 11 months after the operation, the manual muscle testing score of her gluteus medius muscle was 5, and she resumed her normal work and exercise levels with no recurrence of pain.

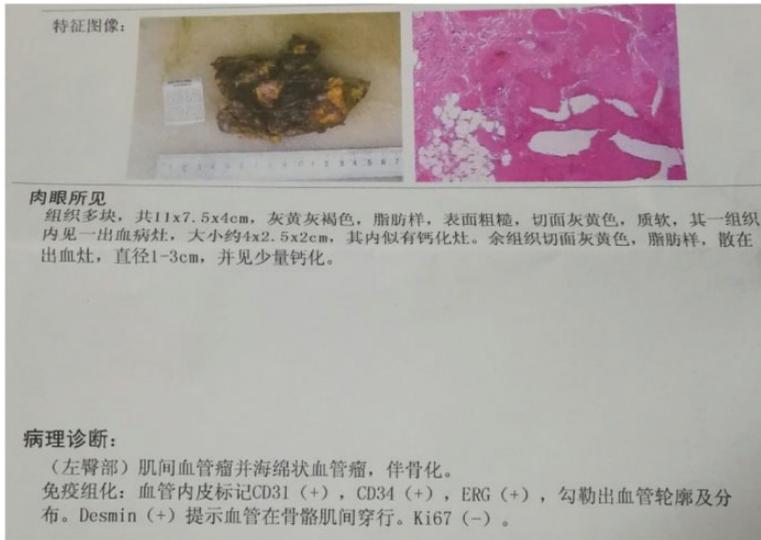


Figure 3. Immunohistochemical results of pathological sections prove that the specimen is an intramuscular hemangioma with no evidence of a malignant tumor.

We retrospectively reviewed the patient's medical history. Five years ago, she began to feel pain in the left buttock while working or exercising, and this was accompanied by radiating pain in the back of the thigh. This symptom is very similar to lumbar disc herniation. The pain was not severe, and the patient did not initially seek medical attention. At this point, the mass might not have been very large, and its compression of the sciatic nerve was likely minimal. Furthermore, the patient had mild obesity and her job required her to move quickly. Therefore, she was diagnosed with lumbar disc herniation, which delayed the correct diagnosis and treatment to some extent. As the mass gradually enlarged during the next few years, the patient's pain became more severe, and the treatment effect was unsatisfactory. The patient therefore sought medical attention again, leading to consideration of other diagnoses. Although a definite diagnosis had not been reached before the operation, lumbar disc herniation had been excluded. X-ray examination

and MRI are often used in the clinical diagnosis of intramuscular hemangioma. The X-ray image shows a soft tissue block shadow or reticular high-density shadow, and about half of cases show a vein stone (phlebolith) shadow. MRI can provide more diagnostic clues. In the present case, T1-weighted imaging of the tumor showed a mixed-density shadow with an isointense or slightly hyperintense signal, and T2-weighted imaging showed a mixed-density shadow with a hyperintense signal. The patient's X-ray demonstrated nonuniform density of the left ilium with large calcified lesions in the gluteus medius muscle. MRI showed high-density shadows in the soft tissue as well as lumpy mixed-intensity abnormal signals in the gluteus medius and gluteus minimus muscles with unclear boundaries. We were unable to obtain a clear diagnosis based on these findings; furthermore, the single-photon emission computed tomography results were suspicious for a malignant tumor. We therefore performed angiography to identify the

main blood supply. Generally, if a definite bruit is present and arteriography reveals the feeding vessels before resection of the tumor, embolization of the vessels followed by meticulous extirpation of the tumor is recommended. Embolization decreases profuse bleeding and shrinks the tumor, allowing for safer and more complete resection. Resection should follow embolization within hours because collaterals rapidly reestablish themselves. Therefore, in such cases, we embolize the main blood supply of the tumor during angiography to control the bleeding and increase the safety of surgery.

Discussion

An intramuscular hemangioma is a benign vascular tumor that contains neoplastic proliferations of blood vessels and often occurs in skeletal muscle. It is characterized by the proliferation of endothelial cells that originate from the vascular tissue of the mesoderm; pathological changes that become more obvious after trauma; and infiltrative growth, invasive growth, and no obvious capsule or boundary. The cause of intramuscular hemangiomas remains uncertain, but both congenital and post-traumatic theories have been considered.⁴ Traumatic and hormonal influences have been suggested and may contribute to the etiology or growth spurts of the tumor.^{5,6} Most intramuscular hemangiomas are also accompanied by various types of mature adipocyte tissues; these tumors are also known as intramuscular angiolipomas. In some cases, chronic anoxia of myocytes and accumulation of metabolites may occur because of poor local blood circulation, which may manifest as muscle swelling, pain, infiltrative scleroderma involving the whole muscle, fibrosis, calcification, ossification, and other bone damage. Most patients with intramuscular hemangioma have no specific symptoms in

the early stage (i.e., usually no mass, pain, or swelling). In the later stage, patients may develop myosclerosis, contracture, and muscle and joint deformities and dysfunction. The most common symptom of intramuscular hemangioma is a slowly enlarging mass that is usually not associated with cutaneous changes, and patients may experience pain without any bruit, pulsation, or thrill,⁷ especially after exercise, as in the present case. Because of its deep location, variable size and shape, lack of specific clinical symptoms, and absence of obvious biological characteristics, the preoperative diagnostic rate of intramuscular hemangioma is only 8% to 19%.⁸ Therefore, it is easily misdiagnosed clinically and must be differentiated from soft tissue malignant tumors and other conditions. In the present case, the patient demonstrated the above-mentioned typical but nonspecific symptoms and was initially misdiagnosed. When similar cases are encountered in the clinical setting, the patient should be carefully examined and an intramuscular hemangioma should be considered. Ultrasound is a convenient and inexpensive examination technique that may be able to provide some clues. However, MRI may be the most important imaging method. The definitive diagnosis depends on the patient's symptoms, physical signs, plain radiographs, and MRI results. Once the diagnosis is confirmed, complete surgical excision of the tumor and sometimes the entire muscle remains the best treatment solution and is recommended by most authors. In the present case, the tumor and surrounding normal muscle tissue were completely removed, and the tumor was resected in its entirety.

Many reports have stated that intramuscular hemangiomas can occur in various sites within the muscles of the trunk and the upper and lower limbs, with predominance in the thigh muscles.⁹⁻¹¹ About 45% of intramuscular hemangioma are found in

the lower extremities, 27% are found in the upper extremities, and the remaining are equally distributed between the head and neck area and the trunk. Because intramuscular hemangiomas are rare vascular tumors and are not usually suspected based on clinical findings, they are of interest to surgeons as a cause of diagnostic problems.¹² In one report, an intramuscular hemangioma was misdiagnosed as an osteoid osteoma.¹³ The symptoms of intramuscular hemangioma are sometimes characterized as nerve compression.¹⁴ With respect to the imaging features, the most similar disease is myositis ossificans because it often occurs in the muscle and is characterized by calcification lesions and edema.¹⁵ In the present case, the intramuscular hemangioma occurred in the gluteus medius muscle and produced symptoms similar to those of lumbar disc herniation. Intramuscular hemangiomas are very rare and easy to misdiagnose because they often lack of specific symptoms. Importantly, when the treatment effect is not satisfactory, further inspection is required.

This is the first known case of an intramuscular hemangioma within the gluteal muscle. The patient's symptoms were similar to those of disc herniation, and it was therefore easily misdiagnosed. This case illustrates that intramuscular hemangiomas often lack specific symptoms and that they should be considered in suspicious cases.

This case has two main limitations. First, this was a rare case and does not represent the more common clinical situations. Second, we did not achieve a definite diagnosis before the operation and even suspected a malignant tumor; this affected the operative plan. However, the tumor was completely removed during the operation; the patient's pain disappeared thereafter, and she was satisfied with the result.

Authors' contributions

YL wrote, edited, and reviewed the manuscript and agreed to be accountable for all aspects of the revision in ensuring the accuracy and integrity of all parts of the work. KC and MY performed the operation. MY conducted the follow-up and data collection. All authors read and approved the final manuscript.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Ethics approval and consent to participate

This is a case report; therefore, ethics approval was not required. We treated the patient according to an established regimen with no complications. The patient provided informed consent for the operation and treatment and for publication of this case report.

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