—Case Reports—

Chronic Expanding Hematoma Extending over Multiple Gluteal Muscles Associated with Piriformis Syndrome

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Abstract

We report on a patient with an unusual, slowly enlarging hematoma of the left buttock. A 62-year-old man presented with a 6-year history of an enlarging mass of the left buttock. He had first noted the mass 6 years earlier and had had sciatica of the left lower limb for the last 2 months. He denied any history of antecedent trauma. The lesion extended over 3 gluteal muscles (the gluteus medius, gluteus minimus, and piriformis). On microscopic examination, the lesion showed typical signs of chronic expanding hematoma. The sciatica was relieved after surgical removal of the lesion. The lesion had not recurred at the last follow-up 4 years after the operation. The present case suggested that chronic expanding hematoma can extend into multiple muscles due, perhaps, to long-term growth and the anatomical and functional conditions of the affected muscles. Our case also suggests that chronic expanding hematoma can be a cause of piriformis syndrome.
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Key words: hematoma, gluteal, piriformis syndrome

Introduction

Hematoma is usually associated with trauma or surgery and will gradually decrease in size and disappear naturally over time. Chronic expanding hematoma (CEH) is a rare type of hematoma that slowly and continuously enlarges without an impairment of coagulation[1]. It is essentially identical to subdural hematoma[1,2]. The specific mechanism underlying this enlargement is not completely understood. A typical CEH is a slowly growing, simple, oval hematoma caused by trauma or surgery adjacent to the fascia of a lower extremity or buttock[1,3]. However, CEHs are occasionally extensive and difficult to differentiate from sarcoma. Treatment of CEH requires excision, but outcomes are usually good[3]. We report an unusual case CEH extending over multiple muscles of the gluteal region which was associated with piriformis syndrome, without a history of trauma.

Case Report

A 62-year-old man presented with an enlarging mass on the left buttock. He had first noted the

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mass about 6 years earlier and had had sciatica of the left lower limb for the last 2 months. The patient had no history of antecedent trauma or ecchymosis of the left buttock. He had mild hypertension but no other diseases, such as hyperlipidemia, diabetes mellitus, paralysis, or addictions, such as alcoholism. He had worked in a bed factory for more than 20 years. His family history was unremarkable.

On physical examination, the patient was 160 cm tall and weighed 61 kg. A large mass of the left buttock, measuring 20 cm × 20 cm, was elastic and soft on palpation with mild tenderness and fluctuation but without redness or heat. The circumference of the left thigh was 2 cm less than that of the right thigh. The abduction power of the left hip was graded as 4 on manual muscle testing. Results of laboratory tests, including bleeding time and coagulation, were unremarkable.

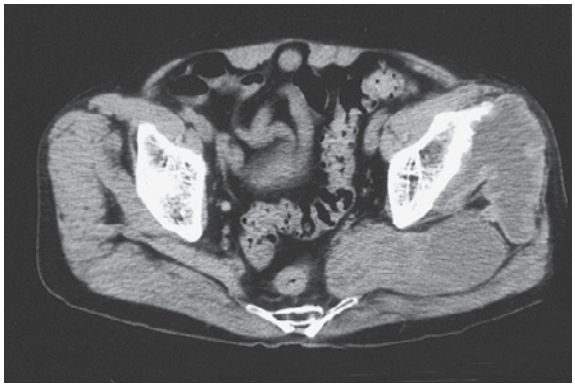

Fig. 1 Computed tomography scan of the pelvis showing swelling of the left gluteus medius, gluteus minimus, and piriformis muscles and the sclerotic, irregular contour of the outer cortex of the ilium.

Radiographs of the left pelvis revealed a sclerotic, irregular contour of the outer cortex of the left ilium. Computed tomography revealed 3 swollen muscles, the gluteus medius, gluteus minimus, and piriformis muscles, showing internal low-density areas without calcification **(Fig. 1)**. Magnetic resonance (MR) imaging revealed a large cystic lesion involving these muscles **(Fig. 2)**. The lesion exhibited a slightly increased signal intensity with a low signal intensity rim and small peripheral nodules on both T1- and T2-weighted images. The intrapelvic portion of the lesion (the proximal part of the piriformis muscle) was enhanced after gadolinium administration **(Fig. 3)**. The gluteus maximus muscle appeared atrophied. Signal intensity of the tensor fascia lata muscle was similar to that of fat. Gallium scintigraphy revealed abnormal uptake in the ilium and the adjacent lateral soft tissues. Bone scintigraphy revealed abnormal uptake in the ilium. The differential diagnosis, based on clinical and radiological findings, included CEH, abscess, and soft-tissue sarcoma.

On fine-needle aspiration obtained a brown, turbid, serous fluid with fine suspended granules. The cytological analysis of the fluid showed an amorphous substance, histiocytes, and peripheral blood cells without any tumor cells. Bacterial and

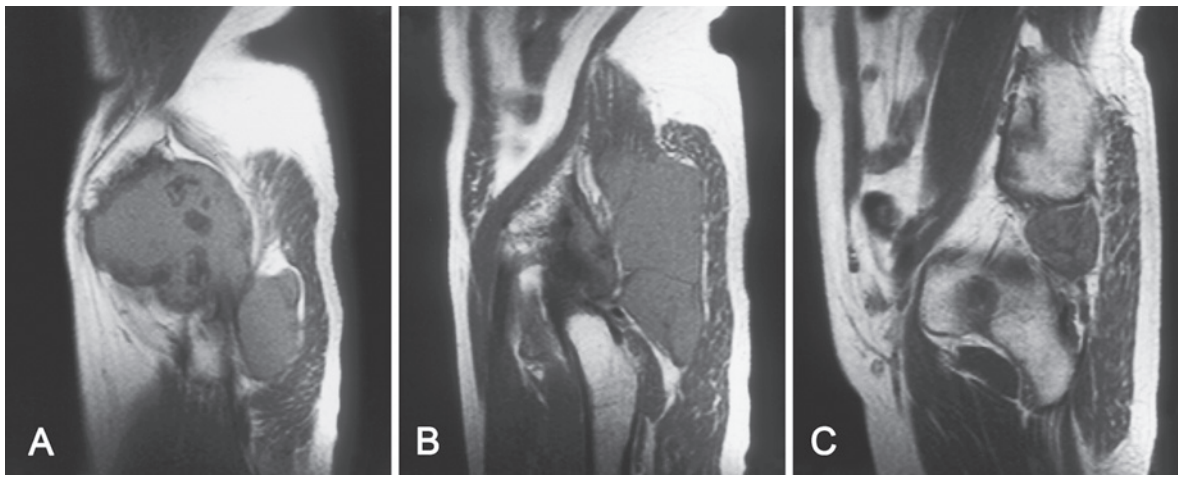

Fig. 2 **A–C**: T1-weighted sagittal magnetic resonance images demonstrating a cystic lesion involving the gluteus medius, gluteus minimus, and piriformis muscles. (A) Plane including the greater trochanter of the femur. (B) Plane including the lesser trochanter of the femur. (C) Plane including the greater sciatic foramen.

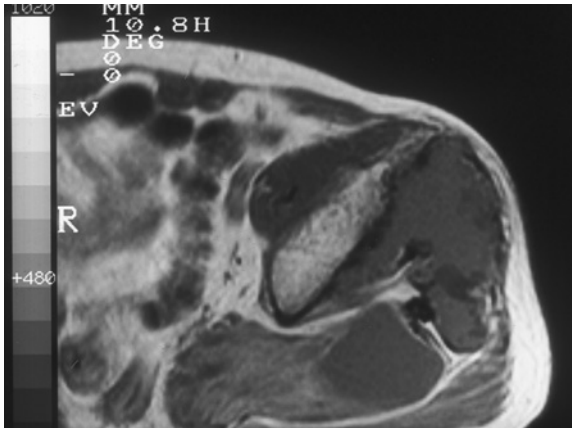

Fig. 3 T1-weighted axial magnetic resonance image demonstrating that the part of the cystic lesion over the piriformis muscle is enhanced after gadolinium administration, whereas almost all parts of the cystic lesions over the gluteus medius and gluteus minimus muscles are not enhanced.

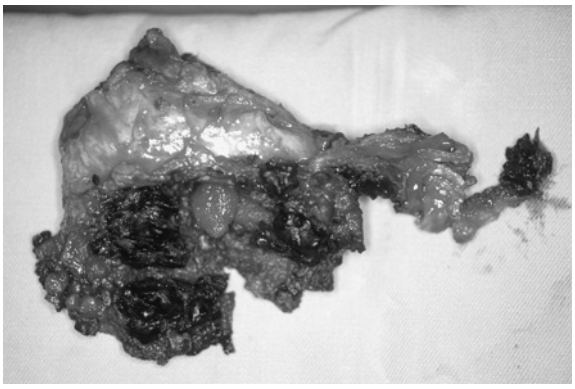

Fig. 4 Gross specimen of the cystic lesion over the gluteus medius and gluteus minimus muscles showing granulationlike tissues and blood clots on its inner surface and blended muscle fibers on its outer surface.

fungal cultures and tuberculosis polymerase chain reaction analysis of the fluid were negative for pathogenic microorganisms.

The cystic lesion was excised following an incisional biopsy. During the operation, the gluteus medius, gluteus minimus, and piriformis muscles were noted to have been extensively affected by a large, irregularly shaped cystic lesion with a thick wall. The inner surface of the cystic lesion was irregular with granulationlike tissues and old blood clots **(Fig. 4)**. The cystic lesion was removed with adherent muscle fibers except for the intrapelvic

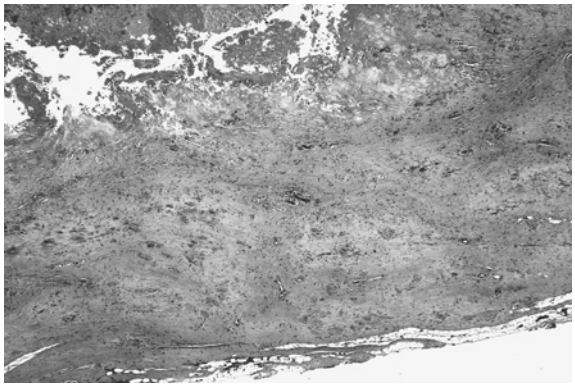

Fig. 5 Histological section of the wall of the cystic lesion over the gluteus medius and gluteus minimus muscles showing hyalinized, dense, fibrous connective tissues with a small amount of dystrophic calcified material (stain, hematoxylin and eosin; original magnification, ×40).

part of the cystic lesion (the proximal part of the piriformis muscle). Curettage of the intrapelvic part of the cystic lesion (the proximal part of the piriformis muscle) and the affected part of the ilium was performed. The sciatic nerve, which was compressed by the swollen piriformis muscle, was carefully released.

Microscopic examination revealed typical diagnostic findings of CEH in the gluteus medius and gluteus minimus muscle parts of the cystic lesion, whereas the part involving the piriformis muscle exhibited a more pronounced inflammation. The wall of the cystic lesion consisted of hyalinized, densely fibrous connective tissues, but the wall of the part of the lesion over the gluteus medius and gluteus minimus muscles showed greater hyalinization and lesser cellularity than did the part over the piriformis muscle **(Fig. 5)**. Deposition of a small amount of a dystrophic calcified material was noted in the wall of the part of the lesion over the gluteus medius and gluteus minimus muscles. The inner surface of the wall of the cystic lesion showed blood clots, granulation tissues with histiocytic proliferation containing ingested eosinophilic amorphous substance, and capillary ingrowth. A more pronounced proliferation of such histiocytes that had infiltrated atrophied or necrotic skeletal muscles was seen in the piriformis muscle part of the cystic lesion **(Fig. 6)** along with capillary

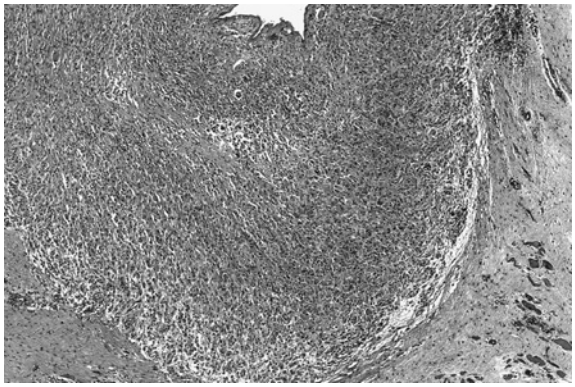

Fig. 6 Histological section of the wall of the cystic lesion involving the piriformis muscle showing pronounced proliferation of histiocytes (stain, hematoxylin and eosin; original magnification, ×100).

ingrowth, fresh bleeding, cholesterol clefts, and multinucleated giant cells. Histiocytic infiltration of the bone marrow was also noted. The tensor fascia lata muscle exhibited extensive fatty degeneration.

The patient obtained relief from the sciatica postoperatively. Four years after the operation, the patient had neither recurrence nor sciatica but complained of the left lower limb being easily fatigued, probably owing to weakness of the hip abductors. The hip abduction power was graded as 3 on manual muscle testing.

Discussion

In 1980, Reid et al. described CEH as a rare type of hematoma persisting and increasing in size for more than 1 month after the initial hemorrhage[1]. Thereafter, this term has been widely used. Characteristic MR findings of CEH is a mass exhibiting heterogeneous signal intensities on both T1- and T2-weighted images with a peripheral rim of low signal intensity[4]. Histological features of CEH consist of 3 components: a peripheral wall of dense fibrous tissues, a central space containing fresh and altered blood, and a midzone of loose connective tissues[1]. Involvement of an adjacent bone is not uncommon[4]. Treatment of CEH requires excision in many cases. Okada et al. have reported that 7 of 9 patients with CEH had an uneventful clinical course after marginal excision, and 2 patients had recurrent lesions[3]. The clinical, radiological, and histological findings of the present case were consistent with CEH.

To the best of our knowledge, 10 cases of intramuscular CEH have been reported in the English-language literature. According to these reports, the affected muscles and patients were as follows: the tensor fascia lata muscles in 2 patients, the gastrocnemius muscles in 2, the psoas muscles in 2, the quadriceps femoris muscle in 1, the rectus abdominis muscle in 1, and unspecified muscles in 2[1–3,5–8]. Half of these cases had no distinct history of trauma, as in the present case.

The specific mechanism underlying the enlargement of a CEH is not completely understood. Labadie et al. have stated that irritant effects of blood and breakdown products could cause repeated exudation or bleeding from capillaries in the granulation tissue[9]. In their report on a CEH in the tensor fascia lata muscle, Mentzel et al. have suggested that muscle compression within a fascial envelope and constant stretching of the compressed muscle are important factors in the development of repeated intramuscular hemorrhages leading to such hematomas[5]. Our patient had long worked in a bed factory, where he had frequently lifted heavy items. Therefore, strong contractions of the left gluteus medius and gluteus minimus muscles during the lifting of heavy items could have caused repeated hemorrhages in these muscles.

The lesion of the present case showed extensive growth over 3 muscles. Although many lesions described in the literature showed a simple oval shape[10–12], Aoki et al. have reported 3 extensive lesions of irregular shape like that of the present lesion[4]. All the patients with extensive, irregularly shaped lesions had long histories; a 62-year-old man with a lesion in the leg of 2 years' duration, an 87-year-old man with a lesion in the thigh of 8 years' duration, and a 70-year-old man with a lesion in the axilla of 3 years' duration[4]. The extensiveness of the present lesion was possibly due to the long duration of lesion's progress and the close anatomical and functional relationships among the affected muscles. We speculate the process of the lesion's growth over the 3 muscles was as follows. The initial bleeding

occurred in the gluteus medius or minimus and involved both muscles because of their close anatomical and functional relationships. Then the hematoma extended into the piriformis muscle through its point of contact with the gluteus minimus muscle, because the sciatica occurred very late as piriformis syndrome and the histopathological analysis revealed more active inflammation in the piriformis muscle. Moreover, the hematoma of the gluteal medius and minimus muscles must have extended more easily toward the piriformis muscle, because the gluteus medius and minimus muscles are firmly surrounded by the wing of the ilium and the combined layers of the fascia lata in all directions except in the direction of the piriformis muscle[13].

To the best of our knowledge, this is the first report of piriformis syndrome associated with CEH. Piriformis syndrome is an entrapment neuropathy of the sciatic nerve by the piriformis muscle at the greater sciatic notch. Causes of entrapment can include hypertrophy of the piriformis muscle, trauma, and myositis ossificans. Diagnosis of piriformis syndrome can often be missed or delayed because of the rarity, nonspecific clinical symptoms, and absence of definitive diagnostic tests[14]. MR imaging can be useful for diagnosing this syndrome[14]. Treatment of piriformis syndrome includes conservative approaches, such as administration of anti-inflammatory agents, and surgical approaches, such as release of the piriformis muscle[14]. In the present case, the surgical removal of the CEH was effective in relieving the sciatica.

CEH may be misdiagnosed as a malignant tumor owing to its deep location, large size, progressive enlargement, and occasional absence of a history of trauma[4]. In general, MR imaging can be used to distinguish a simple hematoma from a soft-tissue sarcoma, because a soft-tissue sarcoma usually has a large solid portion, even if it contains an intratumoral hematoma. However, according to the literature, most portions of a soft tissue sarcoma can be occupied by hematoma, and a solid mass can be hard to recognize in a soft-tissue sarcoma, whereas a CEH can demonstrate thick areas of nodular internal enhancement on MR imaging[14,15]. Accordingly, a CEH can be difficult to distinguish from a soft-tissue sarcoma clinically and radiologically: thus, a biopsy is often necessary to diagnose CEH[16].

The present case suggests that CEH can extend into multiple muscles due, perhaps, to long-term growth and to the anatomical and functional conditions of the affected muscles. Our case also suggests that CEH can be a cause of piriformis syndrome.

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