

Bilateral recurrent external obturator muscle hematoma: An unusual cause of pelvic pain in hemophilia

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Abstract. Following joint hemorrhages, intramuscular hemorrhages are the second most prevalent bleeding pattern in hemophiliac patients. Hematomas of the iliopsoas muscle are a well-known complication of hemophilia; however, obturator muscle hematomas are rare. We herein report a case of spontaneous bleeding of the bilateral external obturator muscles, which occurred three times within a period of 9 months in a hemophilia patient with factor VIII inhibitors. To the best of our knowledge, this is the first published case of an obturator externus muscle hematoma in hemophilia. In addition to hip hemarthrosis, iliopsoas hematomas and acute appendicitis, obturator muscle hematoma should be considered as one of the diagnostic alternatives for pelvic pain in hemophiliac patients. Magnetic resonance imaging enables rapid diagnosis of obturator muscle hematoma.

Introduction

Intramuscular hematomas represent 10-25% of all hemorrhagic complications in patients with hemophilia (1). Factor VIII (FVIII) replacement therapy is crucial to the therapeutic and conservative treatment of hemorrhagic complications. However, the emergence of FVIII inhibitors is a major complication in haemophilia. Upon the occurrence of a hemorrhagic event, subsequent events may become unpredictable and their management becomes more demanding in hemophiliac patients with factor VIII inhibitors.

The diagnosis of pelvic pain may also be challenging in patients with hemophilia. Under such conditions, evaluation should be based on three main pathological findings: hip hemarthrosis, iliopsoas hematoma and acute digestive tract

illness, such as appendicitis or diverticulitis. However, rare diagnoses must be considered. Obturator muscle hematomas are very rarely encountered. Aouba *et al* reported two cases of spontaneous obturator internus muscle hematoma in hemophilia, which is the only publication regarding obturator muscle hematomas in the literature (2). To the best of our knowledge, cases of obturator externus muscle hematoma in the context of hemophilia have not been published to date. We herein report a case of spontaneous bleeding of the bilateral obturator externus muscles, which occurred three times within a period of 9 months in a hemophilia patient with factor VIII inhibitors. The most frequently published cases involving the obturator muscles are primary pyomyositis or intramuscular abscess (3-6). We also underline the possibility of underdiagnosing obturator muscle hematoma with ultrasonography, which requires more advanced imaging modalities, such as magnetic resonance imaging (MRI) or computed tomography (CT).

Case report

A 22-year-old male patient diagnosed with haemophilia A (FVIII <1%) with high responding inhibitors (6.4 BU) for 2 years, was admitted to our emergency department with left hip, groin and thigh pain, extending to the pubic area. At rest, the patient maintained his hip in mild flexion, whereas movement resulted in pain and muscular spasm. The psoas test was within normal limits. The findings on examination of other joints, cardiovascular and respiratory systems were normal. Routine radiographs and ultrasonography of the pelvis and hips revealed no abnormalities. A pelvic MRI revealed extensive bleeding and edema in the left obturator externus muscle (Fig. 1A and B). The clinical findings subsided following initiation of treatment with recombinant activated FVII (rFVIIa). The treatment included 90 mcg kg⁻¹ every 3 h for 3 days. The control MRI performed 3 weeks after the first hemorrhagic event revealed regression of the intramuscular hematoma and edema (Fig. 1C and D).

The patient was admitted to the emergency department with right hip, groin and thigh pain 5 months after the first event. The musculoskeletal findings were nearly identical to the first presentation, when the patient was admitted to the hospital. An MRI revealed a focal hematoma and mild peripheral edema in

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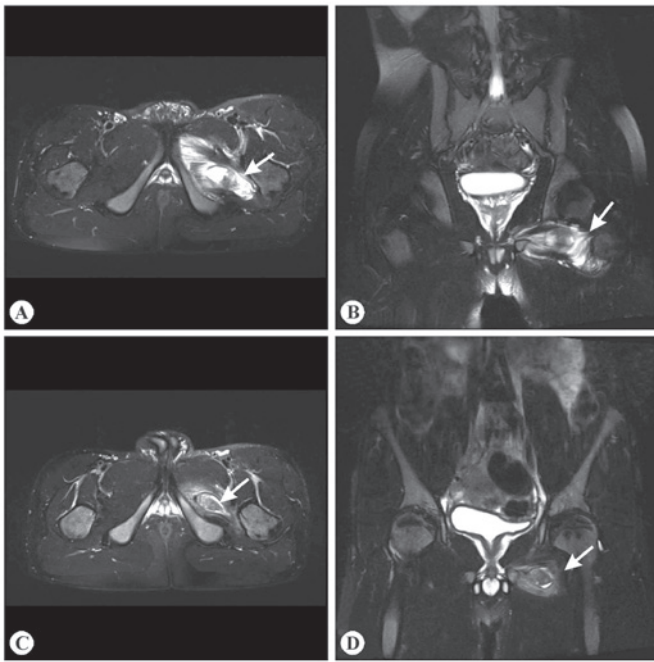


Figure 1. Fat-suppressed T2-weighted (A) axial and (B) coronal magnetic resonance images of the pelvis, demonstrating a hematoma and peripheral edema in the left obturator externus muscle. Fat-suppressed T2-weighted (C) axial and (D) coronal magnetic resonance images obtained 3 weeks after the first bleeding event revealed regression of the intramuscular hematoma and edema.

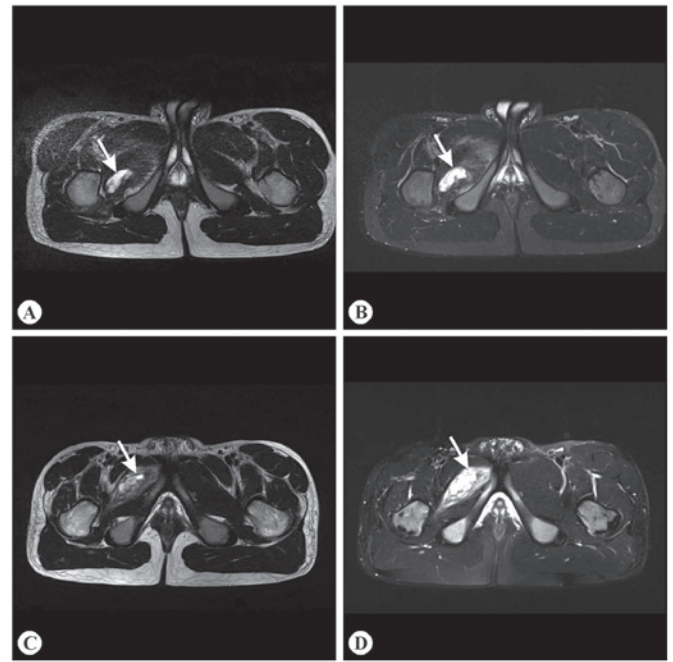


Figure 2. (A) T2-weighted axial and (B) fat-suppressed T2-weighted axial magnetic resonance (MR) images obtained 5 months after the first bleeding event revealed a focal hematoma and mild peripheral edema in the right obturator externus muscle. (C) T2-weighted axial and (D) fat-suppressed T2-weighted axial MR images obtained 4 months after the second bleeding event demonstrated a larger hematoma and prominent peripheral edema in the same muscle.

the right obturator externus muscle (Fig. 2A-B). The symptoms were relieved following treatment with rVIIa (90 mcg kg^{-1} every 3 h for 4 days).

The patient was again admitted to the emergency department with right iliopelvic pain and identical musculoskeletal findings 4 months after the second event. An MRI revealed a larger hematoma and prominent edema in the right obturator externus muscle (Fig. 2C and D). The clinical findings were alleviated following treatment with rFVIIa (90 mcg kg^{-1} every 3 h for 4 days).

The patient provided written informed consent for the publication of the details of this case.

Discussion

The majority of the hemorrhagic complications in hemophilic patients occur within the musculoskeletal system. Following joint hemorrhages, intramuscular hemorrhages are the second most prevalent bleeding pattern in haemophilia. Hemorrhaging generally occurs following trauma, although it may also occur spontaneously (7). The clinical findings are based on the affected muscle, its dimensions and the extent of the coagulation defect. Bleeding causes a soft swelling in the muscle, quickly progressing into defensive spasm and leading to flexion of the adjacent joint, with limitation of movement and intense pain. A limited number of studies have been conducted on the pathology of the mentioned muscle hematomas. It is considered that muscle necrosis happens quickly due to ischemia of the fibres as the developing hematoma damages the neurovascular bundles (8).

A significant location of hematoma occurrence is the iliopsoas muscle; this is a well-known complication of hemophilia

and is considered as a possibly life-threatening complication, significantly associated with morbidity. Iliopsoas haematoma leads to compression of the femoral nerve due to the developing lesion in the muscle; there is generally no traumatic etiology and the first occurrence is sudden, with intense lower abdominal or groin pain (9).

The obturators are the pelvi-trochanteric muscles originating from the posterior (obturator internus) and the anterior (obturator externus) walls of the obturator foramen. These two muscles attach to the greater trochanter of the femur and they are involved in thigh external rotation and abduction, or external rotation and adduction respectively. The differential diagnosis of the obturator externus muscle hematoma should also include primary pyomyositis or abscess. However, only two cases of isolated obturator externus muscle abscess have been reported in the literature to date (5-6). In intramuscular abscesses, fever and elevation of the erythrocyte sedimentation rate or C-reactive protein levels are likely to occur. An MRI with intravenous gadolinium administration achieves a rapid diagnosis of muscle inflammation and rim-enhancing abscess formation; it also enables the detection of additional local pathologies (5).

Ultrasonography is generally of little value in the diagnosis of obturator muscle hematomas, contrary to pelvic MRI, as mentioned in the present case. The CT findings of the intramuscular hematoma are non-specific asymmetric muscular hypertrophy and possible blood attenuation in the muscle (2). MRI demonstrates hemorrhagic signal abnormality and edema in the affected muscle (10).

As a result, in addition to hip hemarthrosis, iliopsoas hematomas and acute appendicitis, obturator muscle hematoma

must be considered as one of the diagnostic alternatives for pelvic pain in patients with hemophilia.

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