A rare cause of acute compartment syndrome after gluteal cyst rupture

Moaaz Baghal, Viral Amrutiya, Husam Ali, Siddhant Mehta, Adam Atoot and Abraham Lo

Department of Internal Medicine and Department of Family Medicine Hackensack Meridian Health Palisades Medical CenterNorth Bergen NJ USA

ABSTRACT
Acute Compartment Syndrome (ACS) is a serious medical condition that often results in high morbidity. ACS more commonly presents after substantial trauma in the extremities where compartment pressures can be measured. Here, we present an unusual case of a healthy man who presented with ACS of the thigh secondary to rupture of the gluteal cyst which was formed after multiple unsupervised testosterone injections. Our goal is to encourage a broad differential diagnosis while keeping limb threatening conditions and emergencies on top in patients complaining of leg pain and also to educate the patient population on safety concerns of unsupervised intramuscular injections.

1. Introduction
Acute compartment syndrome (ACS) is a limb threatening surgical emergency, and delay in diagnosis and intervention can result in significant morbidities from limb loss and systemic morbidities and even death in some cases [1]. The most common etiology of ACS in extremities is trauma-related fracture or massive soft tissue injury, yet when it is present in non-trauma setting, ACS could be missed or delayed to recognize and intervene upon. Especially in the atypical presentation when classic six Ps: pain out of proportion, palsy, paresthesia, poikilothermia, pulsation, paralysis are absent or partially present [2]. Common causes of ACS of the thigh include extreme exercise, DVT, anticoagulation therapy, surgical complications and trauma, with and without fractures [3–5]. The literature review showed a high incidence of ACS in young male patients who have experienced significant trauma. We present an unusual case in which we believe the ACS was secondary to gluteal cyst rupture formed from long-term self-injections of testosterone.

2. Case presentation
A healthy 48 year-old Caucasian American man with no significant past medical history presented to the emergency department with severe left anterior thigh pain. Initially, he noticed a small tender mass in the left buttocks approximately 4 weeks ago. He also described that during his exercise in the gym 4 days prior to arrival he felt like mass was ruptured and reduced in size giving him slight relief in pain. Yet over the period of 4 days, he has worsening of pain and gradually he has decreased in the range of motion at the left hip joint. The patient’s home medication includes self-administered intramuscular testosterone injection usually to his either of buttocks for at least the past 3 years. Complete review of the system was negative for pain at another site, loss of sensation in left extremity, knee pain or ankle pain, skin rash or color change, fever, swelling of left hip or knee joint. Vitals and labs were within normal limits. On physical examination, he was in moderate distress, left thigh was tender to touch, and the patient experienced pain out of proportion on passive extension of the left thigh, with tense anterior thigh muscles, raising the suspicion of compartment syndrome. On admission, a Computed Tomography scan of the left hip showed a left gluteal fluid collection, and a $2 \times 2.7$ cm cystic mass (Figure 1). Left knee synovial fluid was collected and results ruled out infectious processes.

Orthopedic surgery had high suspicion for ACS; therefore, the patient underwent urgent fasciectomy of the left anterior thigh compartment with excision debridement of the left lower extremity including skin, subcutaneous tissues, fascia, and muscle with the placement of wound vacuum-assisted therapy. After 6 weeks, when the patient was seen in the clinic, he had no symptoms and a full range of motion was achieved. Interim he also went through rehabilitation and physical therapy. The patient was educated to refrain from further self-intramuscular testosterone injection.

CONTACT Moaz Baghal moaz.baghal@gmail.com Department of Internal Medicine and Department of Family Medicine, Moaz Baghal Hackensack Meridian Health Palisades Medical Center North Bergen NJ USA

© 2021 The Author(s). Published by Informa UK Limited, trading as Taylor & Francis Group on behalf of Greater Baltimore Medical Center. This is an Open Access article distributed under the terms of the Creative Commons Attribution-NonCommercial License (http://creativecommons.org/licenses/by-nc/4.0/), which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.
3. Discussion

Herein, we present a rare case of ACS of the thigh due to rupture of a gluteal cyst in the setting of long-term self-intramuscular injection of steroids. Generally, ACS presents in the forearm and leg following significant trauma [6,7]. Chronic intramuscular injections have been associated with hematoma, nerve damage, abscess and necrosis, but cystic formation with intramuscular injection is yet to be described in the literature [8].

Proper physical examination and measurement of intra-compartment pressures are required for the diagnosis of ACS. On presentation, the ‘6 P’s’ oftentimes initially manifest as pain which is described as pain out of proportion due to injury [9]. If ACS involves one of the patient’s extremities, the pain would also be increased on passive flexion of the extremity. Skin pallor, paresthesia manifesting as burning or tingling sensation, loss of pulsations distal to the affected compartment, and increased intracompartamental pressure are also seen [10,11].

Konstantontos et al. reported that compartment pressure compared to diastolic blood pressure can be used to aid in the diagnosis of ACS with a cutoff of 30 mmHg; however, no established values were set to diagnose compartment syndrome of the thigh [12]. In this case, pressure measurement of the anterior thigh compartment was not performed due to the lack of established criteria present to use such a method for diagnosis of acute anterior thigh compartment. Management of ACS would require prompt fasciotomy <12 hours after diagnosis to avoid long-term complications [13].

Given a high mortality of up to 47% related to thigh ACS, a high index of clinical suspicion must be used to arrive at a diagnosis to prevent limb amputation and even mortality [14]. Long-term unsupervised self-intramuscular injection complicated by cystic formation should be considered as a possible cause of ACS, as a missed diagnosis of ACS and delay of care in these patients could lead to gravid irrevocable complications.

Disclosure statement

No potential conflict of interest was reported by the authors.

References

[1] Verwiebe EG, Kanlic EM, Saller J, et al. Thigh compartment syndrome, presentation and complications. Bosn J Basic Med Sci. 2009;9(Suppl 1):S28.
[2] Burns B, Sproule J, Smyth H. Acute compartment syndrome of the anterior thigh following quadriceps strain in a footballer. Br J Sports Med. 2004;38:218–220.

[3] Rahm M, Probe R. Extensive deep venous thrombosis resulting in compartment syndrome of the thigh and leg. A case report. J Bone Joint Surg Am. 1994;76:1854–1857.

[4] Ebraheim NA, Hoeflinger MJ, Savolaine ER, et al. Anterior compartment syndrome of the thigh as a complication of blunt trauma in a patient on prolonged anticoagulation therapy. Clin Orthop Relat Res. 1991;263:180–184.

[5] Sancineto C, Godoy Monzon D. Compartment syndrome of the thigh: an unusual complication after spinal surgery. J Spinal Disord Tech. 2004;17:336–338.

[6] Viegas SF, Rimoldi R, Scarborough M, et al. Acute compartment syndrome in the thigh: a case report and a review of the literature. Clin Orthop Relat Res. 1988;234:232–234.

[7] Mithoefer K, Lhowe DW, Vrahas MS, et al. Functional outcome after acute compartment syndrome of the thigh. J Bone Joint Surg Am. 2006;88:729–737.

[8] McQueen MM, Gaston P, Court-Brown CM. Acute compartment syndrome. Who is at risk? J Bone Joint Surg Br. 2000;82:200–203.

[9] Lie C, Leung F, Chow SP. Nicolau syndrome following intramuscular diclofenac administration: a case report. J Orthop Surg (Hong Kong). 2006 Apr;14(1):104–107.

[10] Whitesides TE, Heckman MM. Acute compartment syndrome: update on diagnosis and treatment. J Am Acad Orthop Surg. 1996;4(4):209–218.

[11] Schwartz JT Jr, Brumback RJ, Lakatos R, et al. Acute compartment syndrome of the thigh. A spectrum of injury. J Bone Joint Surg Am. 1989 Mar;71(3):392–400.

[12] Konstantakos EK, Dalstrom DJ, Nelles ME, Laughlin RT, Prayson MJ. Diagnosis and management of extremity compartment syndromes: an orthopaedic perspective. Am Surg. 2007 Dec;73(12):1199–209. PMID: 18186372

[13] Sheridan GW, Matsen FA 3rd. Fasciotomy in the treatment of the acute compartment syndrome. J Bone Joint Surg Am. 1976 Jan;58(1):112–5. PMID: 1249096