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CASE REPORT

Atypical compartment syndrome of the forearm due to mixed infection with *Proteus mirabilis* and *Morganella morganii* after a penetrating injury: A limb-saving approach

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ABSTRACT

Compartment syndrome is a well-described clinical condition and is considered an orthopedic emergency affecting individuals of all ages. A typical scenario for acute compartment syndrome involves lower limb fractures or crush injuries. However, physicians may occasionally encounter atypical presentations, defined as atypical compartment syndrome (ACS). A 38-year-old, left-handed male patient without any comorbidities developed ACS of the forearm and clinical presentation of sepsis after a small penetrating injury to his right forearm. He developed ACS secondary to infected hematoma and subsequent soft tissue infection caused by *Proteus mirabilis* and *Morganella morganii*. Both bacteria infected the patient by direct contamination after injury with a knife, resulting in multifloral contamination. The patient was successfully treated with reconstructive surgery. In conclusion, ACS secondary to this type of penetrating injury shows a subtle clinical course at the time of hospital admission and can insidiously progress from an infected hematoma, posing a serious threat to the limb or even cause mortality. Good extremity function without any disability can be achieved with an accurate diagnosis during the initial evaluation of the patient in the emergency department and prompt surgical intervention followed by appropriate reconstructive methods.

Keywords: Atypical compartment syndrome, coinfection, hematoma, *Morganella morganii*, *Proteus mirabilis*.

Compartment syndrome requires early surgical intervention and close follow-up with proper medical treatment, as it has a progressive course over time, and irreversible tissue necrosis may occur if ischemia is not treated for >8 h.[1,2] The gold-standard approach in the diagnosis of compartment syndrome is to measure the difference between the diastolic blood pressure and compartment pressure.[3,4] However, this method is not preferred in routine practice, and appropriate equipment is not available in most centers. Therefore, the diagnosis of compartment syndrome relies heavily on clinical suspicion.[5]

Compartment syndrome can sometimes have an atypical presentation with an insidious onset, and this type of condition is referred to as "atypical compartment syndrome" (ACS).[6] Hematoma and infection are two rare causes that may play a role in the development of ACS, and a case of ACS secondary to *Proteus mirabilis* (*P. mirabilis*)-infected hematoma has been reported in the literature.[7] However, to the best of our knowledge, our case is the first report of ACS caused by a subcutaneous hematoma coinfected with *P. mirabilis* and *Morganella morganii* (*M. morganii*).

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FIGURE 1. A 2 cm penetrating injury in the volar compartment of the right forearm at the time of second admission to the emergency room is indicated by the arrow.

In this article, we emphasize that ACS can be easily misdiagnosed due to its atypical presentation or insidious onset, and may also lead to catastrophic outcomes, including both morbidity and mortality. Nevertheless, this case report describes satisfactory results after a combination of timely surgical intervention and appropriate medical treatment as part of a limb-salvage approach. It is of utmost importance to suspect ACS in similar scenarios where penetrating injury insidiously progresses from an infected hematoma and poses a serious threat to the limb.

CASE REPORT

A 38-year-old left-handed male patient was admitted to the emergency department with a penetrating injury 1.5-cm long and approximately 2-cm deep in the midline volar region of the forearm. The patient accidentally injured himself with a knife that was used to clean the river fish and separate the cattle offal. The patient had no comorbidities, immunosuppression, or any other preexisting conditions. Physical examination revealed no signs of vascular impairment at the time of initial admission. All peripheral pulses were palpable, and all sensory and motor functions were normal during the examination. No active bleeding from the wound was observed. Diagnostic arterial computed tomography angiography (CTA) or Doppler ultrasound examinations were not performed, as there were no soft or hard signs of vascular impairment, as described previously.[8]

The plain radiographs were interpreted as normal. Tetanus prophylaxis and antibiotics (cefazolin 2 g, gentamicin 1.5 g/kg intravenous [IV]) were administered to the patient following bolus wound irrigation with standard 0.9% isotonic saline solution, superficial debridement, and primary suturing of the wound (Figure 1). A conservative observational strategy was preferred; therefore, the patient was discharged with oral antibiotic treatment and the Rest, Ice, Compression, and Elevation (RICE) instructions after 6 h of observation. There was no pulsatile or expanding hematoma at the injury site or any other pathological findings during the observation period.

The patient was readmitted to the emergency room at 12 h after discharge with a complaint of confusion. The patient had a Glasgow Coma Scale (GCS) score of 13, and other vital signs were as follows: blood pressure 80/50 mmHg, pulse rate 115 bpm and body temperature 38.4°C. Laboratory findings revealed leukocytosis (white blood cell [WBC] count: 19.67/m^3) and elevated C-reactive protein values of 48.9 mg/L.

TABLE I
Diagnostic algorithm of sepsis

Diagnosis	Criteria	Definition
SIRS	• Fever ≥38°C or ≤36°C • Heart rate ≥90 beats/min • Respiratory rate ≥20/min • Leukocyte count ≥12,000 cells/mm^3 or ≤4,000 cells/mm^3 or >10% immature neutrophils	Clinical response defined by determining ≥2 of the findings
Sepsis	• Presence of known or suspected infection + ≥2 SIRS criteria	
Severe sepsis	• Sepsis + development of dysfunction involving one or multiple organs (acute encephalopathy, hypoxemia, hypotension, oliguria)	
Septic shock	• Organ dysfunction + • Refractory hypotension after adequate fluid resuscitation	

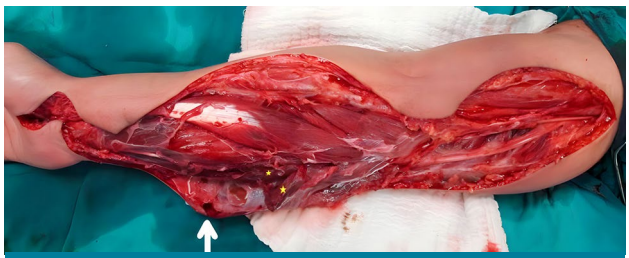

FIGURE 2. Forearm after fasciotomy. Arrow: penetrating injury site and the entry hole. Asterisk: areas of myonecrosis.

FIGURE 3. Image of the forearm after serial debridement and antibiotherapy. Please note the worsened condition of the wound.

FIGURE 4. Early postoperative image after split thickness skin graft application.

Physical examination revealed diffuse swelling and excessive tension in the volar forearm and elbow. The distal pulses were nonpalpable and there was intense pain with passive movement.

The gold-standard approach in the diagnosis of compartment syndrome is to measure the difference between the diastolic blood pressure and compartment pressure. A compartment pressure above 35 mmHg or a value within 30 mmHg of diastolic pressure is considered significantly indicative of compartment syndrome.[3,4] However, this method is not preferred in routine practice, and appropriate equipment was not available. Previous studies have suggested that the diagnosis of compartment syndrome highly relies on suspicion, and the diagnostic criteria include pain out of proportion to the initial injury, palpably tense extremity, pain on passive stretching, paresthesia, and paralysis of the involved muscles.[3] Therefore, this patient was diagnosed with compartment syndrome concurrent with a clinical condition of pre-sepsis (Table I) and underwent aggressive debridement and emergent fasciotomy.

Fasciotomy was performed with a single volar incision starting from the volar region of the arm and extending toward the cubital region. Remarkable myonecrosis was observed, particularly in the forearm volar region (Figure 2). An antibiotic regimen of piperacillin-tazobactam (4×4.5 g IV) and daptomycin (1×500 mg IV) was initiated in consultation with the Department of Infectious Diseases and Clinical Microbiology. Following the procedure, the patient underwent daily superficial debridement, wound irrigation, and open wound dressing.

FIGURE 5. Functional outcome at the eighth month follow-up visit.

TABLE II
Range of motion (ROM) in upper extremity joints

	Flexion	Extension	Total ROM
Elbow joint	130	0	130
Wrist	40	30	70
MCP	90	-10	100
PIP	90	0	90
DIP	70	0	70

MCP: Metacarpophalangeal joint; PIP: Proximal interphalangeal joint; DIP: Distal interphalangeal joint.

Early blood culture results were (-), but *P. mirabilis* and *M. morganii* were isolated in tissue culture. Therefore, a new treatment regimen consisting of meropenem (3×2 g IV) and metronidazole (4×500 mg IV) was initiated on the sixth day after fasciotomy. Daily serial debridement and mesh gauze dressing with rifamycin sodium (Rifocin®; Sanofi Sağlık Ürünleri Ltd. Şti, İstanbul Türkiye) and nitrofurazone (Furacin®; Sanofi Sanayi ve Tic. A.Ş, İstanbul Türkiye) were applied for another two weeks (Figure 3). Vacuum-assisted closure was not preferred for the wound due to the anaerobic infection and the gradual necrotic tissue requiring serial debridement.[9,10] Subsequently, reconstructive surgery was planned for this patient, whose clinical condition gradually improved, with negative infective markers. A second surgery included a local skin flap to the exposed median nerve in the midline of the forearm and application of a split-thickness skin graft from both thighs for large skin defects (Figure 4).

Two weeks after reconstructive surgery, the patient was discharged using a standard postoperative rehabilitation protocol. At the eighth month of follow-up, the skin integrity was intact, no fistula mouth or discharge was found in any region (Figure 5), and the range of motion of the elbow and wrist joints was normal (Table II).

DISCUSSION

This case report describes penetrating upper injury trauma leading to an infected hematoma and eventual ACS. Current literature suggests that the accuracy of physical examination for detecting vascular injury is very high in patients after penetrating trauma.[11,12] Previous studies have reported that hard signs of vascular injury (i.e., active hemorrhage, absent distal pulses, ischemia, expanding or pulsatile hematoma, bruit or thrill) mandate surgical exploration, and diagnostic CTA is indicated in patients with hard or soft signs (subjective reduced or unequal pulses, large non-pulsatile hematoma, neural injury, history of bleeding).[8] Considering that our patient had neither of these signs indicating vascular impairment, a conservative observational strategy was chosen at the initial admission.

Compartment syndrome is described as increased pressure in a contained fibro-osseous compartment, leading to impaired perfusion pressure and subsequent neuromuscular hypoxia or ischemia.[13] The typical scenario for acute compartment syndrome involves lower limb fractures or crush injuries. However, physicians may occasionally encounter insidious and atypical presentations, defined as ACS.[13] Current literature suggests that surgical fasciotomy of the involved compartments remains the gold-standard treatment approach, irrespective of the cause, condition, or location.[14] Stull et al.[7] performed dual-incision fasciotomy instead of a standard single-incision to avoid contamination of the uninvolved compartments in a patient with ACS secondary to a Proteus-infected hematoma. However, we decided to use a standard single-incision approach for our patient. Nonetheless, there were no signs of contamination in the uninvolved compartments or any persistent wound infections during the postoperative follow-up. In this context, we emphasize the importance of combining the correct antibiotic regimen with a timely surgical procedure to prevent contamination after a single-incision technique.

Papachristos et al.[13] reported that atypical manifestations of ACS could lead to delays in treatment and unfavorable clinical outcomes. The literature review shows that unusual anatomical locations, rare conditions, drug interactions, and their side effects as well as surgical procedures and rare fractures can be associated with ACS.[7,13,14] There is limited knowledge about an infected hematoma associated with ACS,[7] and the authors of this case report describe a coinfection of forearm hematoma leading to ACS for the first time in the literature.[15] *Proteus mirabilis* and *M. morganii* were isolated as coinfectious agents in this case.

Proteus mirabilis is a Gram-negative *bacillus* species found in the gastrointestinal tract of both humans and animals. It is most commonly associated with open sores and osteomyelitis in immunosuppressed individuals.[16] On the other hand, *M. morganii* is a Gram-negative, anaerobic, facultative *bacillus*. *Morganella morganii* can be found in the oral cavities of animals and infect individuals; moreover, some studies have suggested that it has evolved as a zoonotic pathogenic bacterium.[17]

Proteus mirabilis and *M. morganii* spp. are known to cause opportunistic infections and belong to different flora. We believe that the coinfection in this patient possibly originated from the knife, which was contaminated with different flora (river fish and cattle offal). The *P. mirabilis* and *M. morganii* both possess *peritrichous flagella*, which make them motile with superior swarming ability, thus allowing the infection to spread easily at an early stage.

This patient achieved satisfactory clinical outcomes, including good functionality and significant relief of symptoms, owing to a limb-salvage approach consisting of an accurate diagnosis and prompt surgical.

In conclusion, this case report contributes to the understanding of the diagnosis of ACS after a minor injury and emphasizes the potential for catastrophic results for both the surgeon and patient, in cases of ACS with delayed intervention or misdiagnosis.

Patient Consent for Publication: A written informed consent was obtained from patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

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