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## Case Report

# Spontaneous acute forearm compartment syndrome: Case report of a clinical diagnosis with limited imaging options <sup>☆</sup>

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## ABSTRACT

Diagnostic radiology is essential for providing targeted management of different diseases. Thus, there has been a dramatic increase in the demand for medical imaging. However, acute compartment syndrome (ACS) is one of the clinical scenarios in which radiology has limited value. The authors report a nontraumatic spontaneous ACS in the forearm of a 56-year-old female. The roles of Ultrasound and MRI, if available, are also illustrated. Limited reports of spontaneous ACS are published in the literature; we hope this case adds to the limited data. Our goal in reporting this case is to improve clinical practice with favorable outcomes for the patients involved globally by alert to the onset of ACS to promote early detection and timely fasciotomy. Also, we aim to increase awareness among physicians and radiologists of the limitations of radiology in specific clinical scenarios. Finally, it may aid in illuminating a possible link between malignant hypertension, spontaneous bleeding/hematoma, and ACS.

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## Introduction

Acute compartment syndrome (ACS) is a surgical emergency. It can theoretically occur in all muscular compartments, but it is most often seen in the forearm and lower

leg. If not appropriately treated, the elevated pressure results in ischemia and eventually irreversible necrosis to muscles and the associated nerves [1,2]. Therefore, as little time as possible should be spent confirming the diagnosis [1]. Idiopathic spontaneous ACS is an uncommon occurrence [3,4].

Abbreviations: BP, blood pressure; BAT-score, The ISTH-SSC Bleeding Assessment Tool; ECG, electrocardiogram; HB, hemoglobin; O<sub>2</sub> Sat, O<sub>2</sub> Saturation; P, pulse; RR, respiration rate; Temp, temperature; WBC, white blood cells; RBC, red blood cell.

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The rarity of spontaneous ACS in the upper limbs makes it challenging to recognize and can lead to adverse outcomes. Only 17 nontraumatic ACS of the forearm from 1993 to 2016 have been reported [5], of which only one had an idiopathic cause that was later attributed to nephrotic syndrome and transient bacteremia [6]. The search of Google Scholar from 1980 to 2022 for similar publications to the presented publication did not turn up any. We report this case to add to the scarce data. Thus, it might aid in providing the best evidence-based care to colleagues and patients globally. Clinical, radiological, and surgical findings are illustrated accordingly.

## Case report

A 56-year-old Caucasian woman presented to the emergency department (ER) around 03:00 PM with acute spontaneous left-sided anterior forearm pain and swelling. The pain had started around 09:30 AM and worsened through the day. Her general practitioner referred her to the ER with a blood pressure of 210/99. Her medical history included a probability of micronodular pulmonary sarcoidosis, stable since 2007 with no treatment. The patient used acetylsalicylic acid 75 mg daily, Terbutaline (Bricanyl) and paracetamol occasionally, and no use of anticoagulants. She had no diagnosis of hypertension or bleeding disorder nor any history of past trauma, surgical intervention, insect bites or exertional exercise.

At the time of presentation, the pain was diffuse, worst 3–5 cm above the wrist joint on the volar aspect. The pain worsened with movement and showed gradual aggravation despite paracetamol and morphine. Decreased sensory perception in 3 ulnar fingers. Ulnar and radial pulses were present, and capillary refill time was < 2 seconds. BP. 217/104, P.77, Temp. 37.6, O<sub>2</sub> Sat. 99%, and RR. 16. Various blood tests were carried out. There was a normal leucocyte count, including WBC differentiation cells, HB, RBC, thrombocytes, renal function, arterial blood gas analysis, lipid profile, B12-Vitamin, and ECG. Also, there was a normal coagulation profile, except for a near-to-normal antithrombin level:  $1.28 \times 10^3$  International Units (IU)/L (> 16 years:  $0.80\text{--}1.20 \times 10^3$  IU/L). D-dimer was slightly elevated 0.81 mg/L (Normal is < 0.7 mg/L). Normal fibrinogen. Myoglobin was normal at the presentation time, but slightly increased to 81  $\mu$ g/L (Normal is < 75  $\mu$ g/L) when measured again after 5 hours from presentation to the ER. Creatinine kinase, measured after 5 hours from presentation to the ER, also showed mild elevation: 247 U/L (Normal for females > 18 years: 50–150 U/L).

The acute ultrasound examination was done between 05:15 and 06:00 PM. During the ultrasound examination, the patient was in severe pain. The patient exhibited tenderness in the forearm when pressed by the probe. Acute ultrasound (US) of the left-sided upper limb showed a well-defined inhomogeneous, mainly hypoechoic, space-occupying lesion 10 cm above the left wrist in the anterior compartment of the forearm. It measured 2.7 cm X 1.2 cm X 1.2 cm. It was interpreted as most likely a hematoma. Normal flow in the ulnar and radial arteries. Ultrasound excluded a thrombosis in the upper limb and showed no active bleeding (Fig. 1). Acute magnetic resonance imaging (MRI) services are unavail-

able in the hospital after 03:00 PM when the evening shift starts.

In the clinical context, the near-normal biochemical parameters, acute ultrasound imaging and the above-mentioned symptoms led to the tentative diagnose of ACS in the anterior compartment of the left forearm despite the absence of an obvious etiology other than the possible intramuscular hematoma (Fig. 1). The patient was started on labetalol intravenously to treat hypertension. At approximately 11.15 PM, the intracompartmental pressure was 40 mmHg in the flexor digitorum superficialis supporting the diagnosis. Pressure was measured using a Centurion Compass Universal Hg Device. Surgery was planned, and the patient was put under general anesthesia at 00.05 AM.

Intraoperatively, the patient's muscles in the volar compartment bulged out when incising the fascia, and the proximal part of the flexor digitorum superficialis was slightly discolored. A hematoma, corresponding to what was demonstrated by ultrasound, was removed. No ongoing bleeding was observed (Fig. 2).

## Discussion

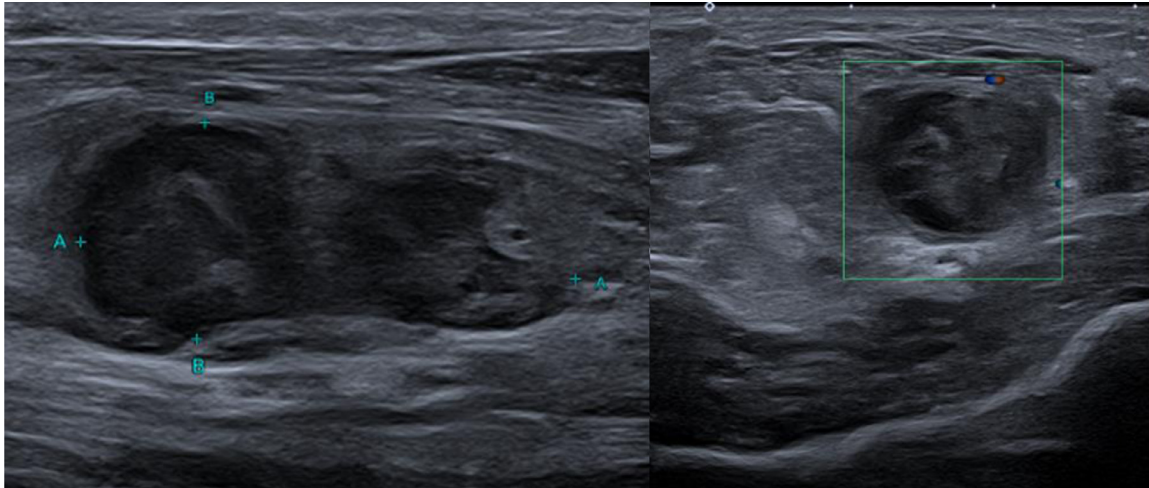
ACS is a surgical emergency. The accepted normal pressure within a compartment is < 10 mmHg. To aid the diagnosis of ACS, intracompartmental pressure (ICP) > 30 mmHg can be used as a threshold. However, a single normal ICP reading does not exclude ACS [1].

Fractures represent 75% of the predisposing factor to ACS. ACS can also be seen in penetrating traumas, burns, vascular injuries, bleeding disorders including hematological malignancies, tumor infiltration, reperfusion injuries, thrombosis, and intense athletic activity syndrome, as seen in the case of 'march gangrene' etc. [1,7]. ACS can occur several months or even years after the trauma [8]. Idiopathic spontaneous ACS is an uncommon occurrence [3,4]. The term 'spontaneous' compartment syndrome has been used to suggest a non-traumatic cause such as diabetes mellitus, nephrotic syndrome, hypothyroidism, and secondary medication [8].

The \*Gold standard\* of diagnosing ACS is based on the clinical assessment of symptomatology [9]. Imaging options are generally limited [10] and are rarely used because imaging delays appropriate intervention [11]. Furthermore, nonclinical diagnostic means should have no bearing on the urgent "get out of bed and take the patient to the [operating room]" [12].

Ultrasound (US) is more readily available and allows dynamic imaging of muscle trauma. US in ACS can show edema [10], which appears as an increase in echogenicity [7]. However, this is not specific to CS. Also, edema starts later in the case of CS. US can exclude other differential diagnoses that overlap with CS [1]. For example, US can exclude thrombus, muscle tear, and occlusion [1,13]. It is also used to evaluate arterial flow [13].

Magnetic Resonance Imaging (MRI) is superior to US in diagnosing soft tissue pathologies [11]. MRI service complements US. However, obtaining an MRI exam can cause a delay in the diagnosis and time for surgical treatment, thus resulting in a poor outcome. Therefore, only when rapidly available



**Fig. 1 – (A & B) Ultrasound image of the space-occupying lesion in 2 planes. 10 cm above the left wrist in the anterior compartment of the forearm. A well-defined mixed echogenic mass measured 2.7 cm x 2 cm x 1.2 cm. Color-Doppler ultrasound revealed no blood flow neither inside the mass nor outside. Radiologically, it was compatible with the features of a hematoma. The scanner is a Toshiba, Aplio 500.**



**Fig. 2 – Intraoperative photo showing the corresponding hematoma as seen by the acute diagnostic ultrasound. No bleeding was identified. It was evaluated intraoperatively that the hematoma was located between the flexor digitorum superficialis, flexor pollicis longus and the flexor digitorum profundus.**

can an MRI assist in the diagnostic workup of ACS, but it is ineffective in diagnosing early ACS [12].

If acute MRI services are available, it can show edema in subcutaneous tissue and muscles [13]. Also, it can demonstrate necrosis [7]. Necrotic muscle tissue is depicted as a non-enhancing intramuscular region on SE T1-weighted sequences following IV gadolinium administration. MR angiography, in some cases, can be used to detect abnormal peripheral vessels [8]. MRI has a role in chronic exertional compartment syndrome [12].

Delay in treatment results from delay in diagnosis. Delayed diagnoses occur due to an unclear presentation, a slow-evolving presentation, unclear etiology, or delays in seeking medical care [14]. At the presentation time of ACS, only one of the classic symptoms can be present, which can often be underestimated or confused with other injuries. Therefore, waiting for the onset of all the typical symptoms of ACS appears

censurable [15]. ACS is currently one of the malpractice claims [15].

Surgical decompression through fasciotomy is advised as soon as a diagnosis of CS is suspected [13]. There is still controversy about the right time that fasciotomy should be done to avoid irreversible ischemic changes [2]. But it is recommended within 6 hours of injury, and fasciotomy is not recommended after 36 hours because it may not be beneficial in this situation [1]. Delayed fasciotomy is controversial and associated with increased rates of late amputation and infection [2,14]. ACS is associated with potential systemic risks such as rhabdomyolysis, myonecrosis, and death. Also, a functional loss is possible [1,14]. Therefore, conducting a prophylactic fasciotomy may be unnecessary but is generally recommended better than doing it too late or ignoring a true ACS [2]. Regarding our patient, it took about 9 hours to execute a fasciotomy from the presentation time to the ER.

During the hospital's admission, our patient's malignant hypertension appears to be the predisposing factor to ACS. The elevated BP, a 210/99, measured at the patient's General practitioner appears to have led the patient to have spontaneous bleeding from small ruptured vessels and/or a rupture of an undiagnosed underlying congenital aneurysm/AV malformation. Subsequently, bleeding led to the formation of the intramuscular hematoma, which increased the intracompartmental pressure leading to ACS.

Reference [16] reported a case of ACS following a trivial injury in a hypertensive 69-year-old man with systemic hypertension for 20 years, who had discontinued acetylsalicylic acid and antihypertensive medicines 13 days before this injury. BP at his time of presentation was 200/120 mmHg. Fasciotomy of the anterior compartment revealed a hematoma of about 750 ml anteromedially in the subcutaneous plane, but no bleeding or ruptured vessel was found. Hypertension appeared to be the predisposing factor, producing continuous bleeding from small ruptured vessels allowing a large hematoma to develop [16].

Despite no history of bleeding disorders and having a normal coagulation profile during the acute admission, during the later follow-up, the patient was referred to the hematology department to exclude an undiagnosed bleeding disorder. The hematology department repeated relevant blood tests. The testing of thrombin time, thrombocytes, hemoglobin levels, WBC, fibrinogen, and clotting factors II, VII, VIII, XIII were all normal. The Von Willebrand Ristocetin Cofactor assay measured at 0.64 (normal:  $0.38\text{--}1.52 \times 10^3$  int.enh./L) with a normal Von Willebrand factor (VWF) antigen at 0.90 (normal:  $0.47\text{--}2.07 \times 10^3$  int.enh./L. Because the patient's BAT-score was 8–10 based on a postpartum bleeding that required endometrial scraping, and having the function of VWF in the low range, the patient was offered flow cytometry of the thrombocytes and repetition of VWF assays. The flow cytometry showed no absence of glycoproteins (Ia, IIIa, Ib, IIb, IX) on the surface of the thrombocytes. Also, adding the agonists (Collagen, ADP, and TRAP) revealed normal fibrinogen-binding with a normal expression of P-selectin and CD63-antigen. Thus, this concludes that the patient has no undiagnosed hematological disease.

The local multidisciplinary team (MDT) meetings, including vascular surgeons and vascular radiologists at the Regional Hospital Viborg, sent the patient for a CT angiography including both arterial and venous phases of the left upper limb 6 months after the operation. In addition, the proximal part of the aorta was also to be scanned in the CT angiography. The results were normal, with no vascular malformations. Therefore, the local MDT concluded that there was no need for further follow-up or extra investigations.

The authors conclude retrospectively that malignant hypertension was the predisposing cause for ACS. However, it is necessary to highlight that at the time of writing the manuscript, the patient was stable and improving. Therefore, neither further follow-up nor investigations are warranted.

Finally, this case also emphasizes the importance of having clinical expertise and prowess to consider an obvious diagnosis even in its rarer form of presentation. Furthermore, it highlights the importance of radiologists understanding the limitation of imaging in specific clinical scenarios.

## Patient consent

Oral and written informed consent to publish this case and use anonymized radiologic and surgical photos was obtained from the patient. Obtaining the consent has also been registered in the electronic journal of the patient.

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