Case Report

Trauma International | 2020 January-June; 6(1):26-29

A Rare Case of Atraumatic Unilateral Isolated Compartment Syndrome of the Peroneal Compartment Leading to Common Peroneal Nerve Palsy

Jun Guang Kendric Tan [1], Rajitha Gunaratne[1], Geoff Cooper[1]

Abstract

Acute Compartment syndrome (ACS) is an orthopaedic surgical emergency with very poor prognosis when left untreated. The majority of cases are preceded by trauma, but importantly there is a small percentage of patients presenting with non-traumatic causes. Although rare with only two other case reports describing non-traumatic, spontaneous compartment syndrome of the peroneal compartment, the complications are equally devastating.

We report a 67-year-old female with a history of Factor V Leiden, pulmonary embolism (PE) and deep vein thrombosis (DVT). She was seen by orthopaedic surgery six weeks after acute onset right leg pain with associated foot-drop. Her symptoms were initially managed by a medical team as she had no history of trauma and no fractures were evident on plain radiographs. This was on a background of severe bilateral knee osteoarthritis, chronic bilateral lower leg pain and neurogenic claudication for the preceding six months. Given her atypical presentation with confounding comorbidities, she was admitted under the medical team for further investigations instead of urgent fasciotomy.

As orthopaedics was not involved in the acute phase of her symptoms, a non-surgical approach was taken. MRI right knee and calf six weeks after onset of symptoms were suggestive of sequela of compartment syndrome isolated to the peroneal compartment. This scan showed myonecrosis in this compartment as well as denervation myopathy of the anterior compartment. Over the next twelve months, there was resolution of leg pain and gradual progression of a positive Tinel's sign from the fibular head to anterior shin with associated return of some ankle range of movement and power.

Given the atraumatic presentation and patient risk factors, we believe she suffered a venous thrombotic event at the popliteal trifurcation leading to a subacute compartment syndrome of the peroneal compartment. Swelling of the compartment led to compression of the deep peroneal nerve, causing denervation of the anterior compartment. This is of interest given the rarity of such presentations, and will serve as a timely reminder for non-traumatic causes of ACS.

Keywords: Compartment syndrome; Non-traumatic; Factor V Leiden; Common peroneal nerve palsy.

Introduction

Acute Compartment syndrome (ACS) most commonly occurs in the setting of trauma, with tibial shaft fractures shown to be the most common cause at up to nine percent, but can be found in cases of compression rhabdomyolysis as well [1]. Other documented non-traumatic causes of ACS include medical comorbidities such as diabetes, end stage liver disease, hypothyroidism, localised soft tissue infection, bleeding diathesis/anticoagulation or thrombophilia [2].

Factor V Leiden is the most common inherited form of thrombophilia, with up to 1 in 5, 000 people with European ancestry having two copies of the mutation. Factor V is a coagulation factor normally controlled by activated protein C (APC), which regulates the clotting process. Individuals with Factor V Leiden have Factor V that cannot be inactivated by APC, resulting in overproduction and unopposed function of Factor V, leading to hypercoagulability [3]. Association studies have shown Factor V Leiden to substantially increase the risk of deep vein thrombosis, especially in large venous blood vessels [4]. If found in an extremity, the thrombus can compromise venous outflow, increasing hydrostatic pressure, causing fluid extravasation to the interstitium and subsequent swelling of the affected compartment.

[1]Department of Orthopaedics, Joondalup Health Campus, Grand Blvd & Shenton Ave, Joondalup, WA 6027.

Address of Correspondence
Dr. Jun Guang Kendric Tan,
Resident Medical Officer, MBBS, Joondalup Health Campus, Grand Blvd & Shenton Ave, Joondalup, WA 6027.
E-mail: junguangt@hotmail.com

Case presentation

A 67-year-old lady presented to the emergency department for sudden onset right anterior lower leg pain extending onto the dorsum of her foot, followed by development of a right foot-drop. She denies any trauma or new onset back pain. This was on a background of bilateral lower leg pain over the past six months, limiting her walking to about ten metres before needing to sit.

On examination, she had profound weakness of right ankle dorsiflexion, ankle eversion, toe extension and decreased light touch at the mid-aspect of shin and dorsum of foot. Significant swelling of her right calf was noted. She otherwise had strong dorsalis pedis and popliteal pulses and a warm, well perfused limb with no infective signs or significant calf tenderness.

Her past medical history includes bilateral severe knee osteoarthritis with valgus alignment of the right knee, chronic back pain, factor V Leiden and pulmonary embolism in 2013. Bloods were unremarkable other than a slight neutrophilia on presentation. Given her atypical

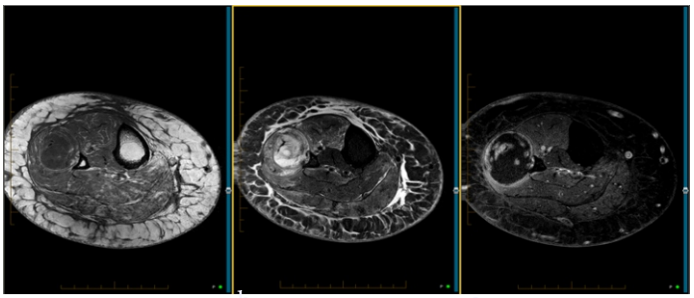

Figure 1: a: T1 – Hypointense peroneal compartment; b: T2 with SPAIR – Hyperintense signal in peroneal compartment (Acute ACS); c: Post-contrast – Areas of non-enhancement

presentation with confounding comorbidities, she was admitted under the medical team for further investigations instead of urgent fasciotomy.

Given her history of neurogenic claudication and sudden onset right foot-drop, an urgent inpatient MRI lumbosacral spine was performed which revealed grade one degenerative spondylolisthesis at L4/5, moderate spinal canal and subarticular recess stenosis at L4/5 with no evidence of right L5 or S1 nerve compression. There were also moderate to moderately severe foraminal stenosis on the right at L2/3, mild to moderate stenosis at L2/3 and moderate spinal canal stenosis at L3/4. In view of the multilevel canal stenosis, a provisional diagnosis of compressive common peroneal nerve palsy was made. There was gradual improvement of pain over the next week and our patient was discharged with analgesia and outpatient follow-ups with orthopaedic and physiotherapy.

On orthopaedic review six weeks after her onset of symptoms, the pain had largely resolved but there was still ongoing foot drop. Further scans were ordered to identify any evidence of local compression. CT right knee and leg showed extensive swelling of the peroneal compartment. It appeared of low attenuation and to contain a few locules of gas, suggesting necrosis and developing gangrene.

MRI right knee and calf on the same day revealed gross oedema as well as signal change involving the entire peroneal compartment of the leg. Post-contrast, there were large central areas of non-enhancement suggestive of necrosis. The common peroneal nerve and its branches also appeared thickened just proximal to peroneal compartment, possibly compressed between the swollen peroneal compartment and the lateral aspect of the fibular shaft. In addition, patchy muscle belly oedema was noted in the anterior compartment. The appearances were suggestive of compartment syndrome isolated to the peroneal compartment with extensive myonecrosis, and compression of the deep peroneal nerve leading to the subacute changes in the anterior compartment likely secondary to denervation myopathy (Figure 1).

CT aorto-bifemoral angiogram was performed to investigate the cause of peroneal compartment necrosis. No stenotic or occlusive lesion of the lower limb arteries was found, and there was no dissection or focus of arterial occlusion.

Differential diagnosis

On initial review by neurology, a presumptive diagnosis of common peroneal nerve palsy was made after her MRI scan reported as showing no evidence of a right L5 or S1 nerve root compression.

When the patient sought orthopaedic input, an ongoing lack of active dorsiflexion and eversion at the ankle was noted. Subsequent CT and MRI revealed evidence of isolated compartment syndrome of the peroneal compartment, with likely denervation myopathy of anterior compartment.

Given the atraumatic presentation and our patient's history of Factor V Leiden, PE and DVT, differentials considered included a venous thrombotic event at the popliteal trifurcation leading to a sub-acute compartment syndrome. Arterial dissection or embolus, although rare, were ruled out on CT aorto-bifemoral angiogram. Double crush phenomena was also considered given the spinal canal and subarticular recess stenosis at L4/5 [5].

Treatment

Following extensive discussion between orthopaedic surgery and neurology, it was decided that given the lack of convincing anatomy to surgically decompress, management would instead revolve around regular follow-ups and conservative measures including intensive physiotherapy and orthotics for her foot drop. Pain medicine specialist input was also obtained for symptom management.

Outcome and follow-up

Over the next twelve months, gradual progression of a positive Tinel's sign was noted. There was also an associated return of motor function at the ankle joint from a plantar position to approximately neutral. We note our patient to be actively participating in physiotherapy and managing her foot drop with a "toe-up" orthosis. At week twenty-three, there was some return of movement in her toes. At twenty-eight weeks, progression of Tinel's sign was noted further along her anterior shin. This together with ongoing functional muscle recovery was encouraging for spontaneous recovery.

Putting together our patient's history of positive Factor V Leiden, PE and DVT, her acute presentation and spontaneous but prolonged recovery, we believe our patient is presenting with an atypical case of compartmental syndrome involving the peroneal compartment. It is likely a venous thrombotic vascular event led to a reduction in venous drainage, causing a reduced arteriovenous pressure gradient. This in turn reduces local tissue perfusion, causing a spontaneous atraumatic compartment syndrome of her peroneal compartment and her presentation of sudden onset of severe pain of with calf swelling. Swelling of her peroneal compartment led to compression of her deep peroneal nerve against the fibular shaft, leading to denervation of the anterior compartment and associated foot drop. As the initial vascular event resolved, swelling of the peroneal compartment subsided, leading to rapid recovery of the deep peroneal nerve and associated return of tinel's sign and motor function.

MRI right knee and calf was repeated at twelve weeks post presentation. Resolution of the previously evident swelling at the peroneal compartment with no extrinsic compressive abnormality was noted. No common peroneal nerve abnormality down to the level of the fibular head was observed. No fatty infiltration, muscle atrophy, rim enhancement or stippled enhancement was noted.

Discussion

A literature review on PubMed with keywords 'compartmental syndrome' and 'non-traumatic' revealed 42 case reports. Of them, two are of particular relevance. L. Andrew reports an otherwise fit and healthy rugby league player who experienced a dull ache of his leg while warming up [6]. He subsequently developing peroneal compartment syndrome requiring decompressive fasciotomy. K. Pentz reports a 48

year old female with protein S deficiency on Warfarin [7]. Unfortunately, her Warfarin was ceased and she subsequently developed non-traumatic compartment syndrome of the hand and thigh requiring decompressive fasciotomies. We note that in both cases, their subjects either had no confounding comorbidities or had a clear inciting cause for thrombosis.

In presentations to the emergency department post-trauma, clinicians should always be vigilant not to miss a diagnosis of ACS. In high risk patients, there is little debate on the need for an urgent fasciotomy. However, diagnosis is often difficult in patients who are unable to give a clear history or participate in a thorough clinical examination such as children, the critically ill or those with ongoing neurological illness [8]. In view of this, compartmental pressures are a good adjunct to assist diagnosis. Whitesides et al. suggests when the difference between diastolic blood pressure and compartmental pressure reaches less than 30mmHg that there is a high likelihood of inadequate perfusion and relative ischemia, aiding early diagnosis and appropriate intervention [9]. Unless presented with challenges that impedes the traditional bedside examination, it is important to remember ACS is a clinical diagnosis and imaging should not delay surgery. If ACS fails to be managed timely, myonecrosis will inevitably occur.

In situations where the diagnosis is not as clear, it is important to consider ACS mimics such as muscular denervation leading to myopathy. As an adjunct to the clinical examination, this diagnosis is frequently made with electromyography (EMG) findings showing absence or reduction of motor unit recruitment during voluntary movement and fibrillation potentials at rest [10].

Factor V Leiden is the most common inherited form of thrombophilia, with strong evidence showing association with venous thrombotic events [11]. Several well designed studies have demonstrated Factor V Leiden's tendency to increase the relative risk of deep vein thrombosis up to eight times for heterozygotes and eighty times for homozygotes [12]. A common classification system for DVTs is based on thrombus location, with proximal DVTs defined as thrombosis involving the iliac, femoral and popliteal vein and distal DVTs if involving only the calf veins. Numerous ultrasound series and contrast venography has shown that up to 50-80% of all DVTs are proximal, [14, 15] but there is little literature categorising the location of thrombosis in terms of exact vein involvement. It is very possible our patient had a venous thrombotic event around the peroneal vein to popliteal trifurcation, causing venous outflow obstruction and a sub-acute compartment syndrome.

Conclusion

ACS can present in atypical ways, with non-traumatic causes uncommon but equally severe. It is essential for clinicians to consider the possibility even in patients with confounding factors and multiple differentials. Individuals with thrombophilia are at particularly high risk and should serve as a red flag for early orthopaedic input. In retrospect, it is understandably difficult to opt for invasive procedures such as decompressive fasciectomy when the diagnosis is unclear.

Our case also highlighted the potential for spontaneous recovery in selected patients with compartmental syndrome. Awareness of ACS mimics and ability to identify complications such as denervation myopathy will enable us to appreciate the potential for spontaneous recovery, reducing unnecessary retrospective surgical interventions and improve patient outcomes.

Clinical message

Non-traumatic causes of ACS are rare but equally debilitating. Thrombophilia is a documented risk factor and should serve as a red flag. In patients presenting with acute limb pain with multiple confounding comorbidities but no clear cause, early surgical input should be sought.

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Conflict of Interest: NIL
Source of Support: NIL

How to Cite this Article

Tan JGK, Gunaratne R, Cooper G | A Rare Case of Atraumatic Unilateral Isolated Compartment Syndrome of the Peroneal Compartment Leading to Common Peroneal Nerve Palsy | Trauma International | January-June 2020; 6(1): 26-29.