Acute spontaneous compartment syndrome in recent onset type 1 diabetes

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Acute spontaneous compartment syndrome is a rare orthopaedic emergency that usually presents to general physicians as an acute medical admission. Most cases reported to date, in patients with diabetes, are in those with long-standing disease or with evidence of diabetic complications. An acute spontaneous compartment syndrome in a girl with recent diagnosis of type 1 diabetes is reported here. Awareness of the condition allows early recognition and diagnosis, thereby preventing more severe muscle necrosis and disability.

A 17-year-old Asian girl was referred to our admission unit by her general practitioner with a suspected deep vein thrombosis (DVT). She presented with a 4-day history of severe pain in the lateral aspect of the right leg, which was associated with swelling and difficulty in walking. There was no history of previous injury, injection of drugs or excessive exercise. The patient had been diagnosed as having type 1 diabetes mellitus 2 months previously, and had been given Novomix 30 (Biospart insulin) subcutaneously, twice daily. She was not taking any other drugs, and had no evidence of any diabetic complications. There was no history of illicit drug use. On examination, she was afebrile. There was swelling of the right leg with lateral tenderness, with a localised increase in temperature, but no erythema. Arterial pulses were palpable in both feet. A mild weakness of eversion of the right ankle was found. DVT was excluded on the basis of a normal D dimer and negative Doppler ultrasound of the leg. Routine blood results showed mildly raised inflammatory markers, including a C reactive protein level of 67 mg/l, white cell count of $13.8 \times 10^9/l$ and erythrocyte sedimentation rate of 40 mm/h. Urea and creatinine levels were within the normal range. After the finding of an increased creatine kinase level at 6167 U/l, an MRI scan of the leg was performed, which showed swelling and an abnormal signal of the whole peroneus muscle with no bony involvement. An urgent orthopaedic opinion was obtained, and isolated lateral compartment syndrome was confirmed by the measurement of compartment pressure by a Stryker needle unit, providing pressures of 120 mm Hg in the lateral compartment and 19 mm Hg in the superficial posterior compartment (fasciotomy indicated if pressure was >30 mm Hg).¹ Emergency decompression of the lateral compartment was

carried out, which confirmed a tense lateral compartment and necrotic peroneus muscles. There was no evidence of purulent discharge, haematoma or active bleeding. Fasciotomy and excision of the peroneus muscles were performed, and subsequent histological examination of muscle biopsy confirmed acute ischaemic changes. The patient made a good recovery and was

discharged home with mild weakness of ankle eversion. She remained asymptomatic, and a follow-up MRI at 3 months showed a generalised high signal within the anterior compartment of both tibialis anterior and extensor longus muscles, but no swelling, suggesting an early stage of the disease.

DISCUSSION

Non-traumatic leg pain is a common and important medical emergency. The differential diagnoses usually include DVT, cellulitis, abscess, thrombophlebitis, ruptured Baker's cyst, muscle tear/strain, osteomyelitis, haematoma and, rarely, acute spontaneous compartment syndrome.

Compartment syndrome is an orthopaedic emergency requiring urgent fasciotomy to prevent irreversible damage. It is defined as an increase in interstitial pressure in a closed osteofacial compartment, leading to microvascular compromise. The most common causes

Figure 1 MRI scan showing abnormal high signals from the peroneus muscles (lateral compartment).

of compartment syndrome are trauma, burns, compression and arterial injury. Spontaneous compartment syndrome is very rare. The few reported cases have been associated with a number of medical conditions, including hypothyroidism, nephrotic syndrome, leukaemic infiltration, influenza-virus-induced myositis and bleeding.²

However, the most frequently reported single association, with spontaneous compartment syndrome is diabetes mellitus. Muscle infarction in patients with diabetes is a well-reported, but rare, complication of long-standing diabetes. The mean duration of diabetes to the first episode of diabetes-associated muscle infarction (DMI) is 14.3 years.¹ However, <200 episodes of DMI have been reported in the literature worldwide.¹ The development of an acute compartment syndrome after DMI is an even rarer occurrence. Only six cases of acute spontaneous compartment syndrome associated with diabetes have been reported in the literature.² Unfortunately, the diagnosis is usually delayed because of lack of awareness of the condition. To our knowledge, ours is the first report of spontaneous compartment syndrome in a patient

with recent-onset type 1 diabetes with no evidence of diabetic complications. This is also the youngest patient in the literature with DMI (the reported age range is 19–81 years).^{3,4}

The mechanism of muscle infarction in DMI is unknown, although it is thought to be the result of microvascular disease associated with long-standing diabetes leading to ischaemia and cellular oedema, and to an increase in interstitial pressure within the closed facial compartment. This leads to a reduced capillary blood flow, and further, muscle ischaemia. The relative young age and lack of diabetic complications in our patient are, therefore, unusual, and suggest an alternative mechanism.

Unfortunately, our patient presented relatively late to the admission unit, and we were unable to save the lateral compartment muscles from necrosis. The awareness of spontaneous compartment syndrome, particularly in patients with diabetes mellitus, should be considered in the differential diagnosis of severe leg pain, with prompt diagnosis leading to an urgent referral to the orthopaedic surgeons.

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