CASE REPORT

Fulminant crural compartment syndrome preceded by psychogenic polydipsia

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SUMMARY

We report a case of bilateral anterolateral crural compartment syndrome elicited by hyponatraemia and psychogenic polydipsia. The unusual constellation of clinical findings and diminished pain expression made initial diagnostic procedures challenging. The possible pathogenesis and treatment options are discussed. Impairment of lower extremity function at follow-up was serious and permanent.

BACKGROUND

Compartment syndrome (CS) is characterised by localised muscle damage causing inflammation, swelling and increased pressure within an osteofascial compartment when fascial compartment pressures exceed perfusion pressure. This leads to muscular pain and potentially to muscle necrosis, and nerve and vessel injury. CS is most commonly related to musculoskeletal trauma and to reperfusion after acute peripheral ischaemia,[1] yet non-traumatic causes of muscle swelling and necrosis might also lead to CS.[2–7]

The cellular manifestation of CS is rhabdomyolysis. This presents serologically with elevated serum creatine kinase (CK) and myoglobin.[1 8] Besides trauma, aetiologies of rhabdomyolysis are legio, examples being hypothyroidism, hypokalaemia, hypophosphataemia, hyponatraemia/hypernatraemia, hypocalcaemia, hyperglycaemia, G6PD deficiency and familial rhabdomyolysis.[2] Elevated levels of myoglobin may severely compromise kidney function by clogging the renal tubuli with myoglobin casts. Accompanying lipid peroxidation may also contribute to acute renal failure.[1]

Psychogenic polydipsia is a mental disorder most often described in schizophrenics.[9–11] After drinking large amounts of water, acute severe hypotonic hyponatraemia may develop due to diminished renal water excretion. Severe hyponatraemia might be related to convulsions and cerebral oedema, concomitant rhabdomyolysis and, very rarely, CS.[1] Another cause of polydipsia is diabetes insipidus caused by deficient secretion of antidiuretic hormone or a diminished kidney response.

We present a clinical case of bilateral CS preceded by water intoxication, and discuss the possible pathogenesis and treatment options. Several factors may have contributed to the development of CS in this case, and the diagnosis and treatment was relatively complex and surgically radical.

CASE PRESENTATION, INVESTIGATIONS AND TREATMENT

A 30-year-old man with a schizophrenic disorder was admitted to the emergency department (ED) from a psychiatric hospital where he had been found on a bathroom floor in a comatose state. Convulsions had not been witnessed but were suspected.

The patient had previously been somatically healthy. However, 3 days prior to admission, he had been admitted voluntarily to the psychiatric hospital due to anxiety and delusions. The second generation antipsychotic drug, aripiprazole, was prescribed but compliance was questionable as the patient only ingested the drug on one occasion. In a psychotic state, he was compulsively eating, and drinking excessive amounts of fluids, resulting in multiple vomiting episodes. The large water intake was part of a psychosis and the patient later explained on one occasion that he intended to purge bacteria from his body, but on other occasions he stated that his excessive water intake was due to unspecified mental stress and an attempt to soothe anxiety. There was no previous record of polydipsia, alcohol or drug abuse.

On arriving at the ED on day 1 he was conscious (Glasgow Coma Score of 14) with a high spontaneous respiratory rate (30–40/min). Blood pressure was 170/90 mm Hg, oxygen saturation 92% and ear temperature 35.1°C. On examination, there was no sign of trauma or focal neurological lesions. There was a state of global myoclonic fasciculations, explosive watery vomiting and a declining level of consciousness. Selected laboratory data were Na^+ 115 mmol/L, white cell count 25 *$\times 10^9$/L, haemoglobin 8.0 mmol/L (table 1) and lactate 8.0 mmol/L. A normal sodium level of 144 mmol/L 3 days before was noted indicating acutely developed hyponatraemia due to psychogenic polydipsia. A chest X-ray showed bilateral infiltrates indicating aspiration pneumonia. Treatment with hypertonic saline and antibiotics (piperacillin/tazobactam and metronidazole) was instituted and the patient was transferred to the intensive care unit for further treatment of water intoxication. Sodium level was corrected from 115 to 129 mmol/L during the first 18 h and total diuresis over the first 36 h was 16 000 mL. Despite rapid correction of the sodium level there were no clinical signs of central pontine myelinolysis. On day 2, he was transferred to the medical department for further observation and treatment of pneumonia.

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Table 1 Serological data in chronological order

	Day 1	Day 2	Day 3	Day 4	Day 5	Day 6	Day 7
Na^+	115	129	137	135	133	137	137
K^+	3.1		3.8	4.1	4.2	4.3	4.3
Creatinine	53	63	78	72	64	52	56
CK		2830	29 900	15 000	6120	3520	159
Leucocytes	25.0	14.0	21.7	19.1	16.9	11.5	14.0
CRP	<3	82	145	230	207	196	150

Normal values: sodium 137–144 mmol/L, potassium 3.5–4.6 mmol/L, creatinine 60–105 μmol/L, CK 50–400 U/L, leucocytes 3.5–8.8×10^9/L, CRP<10 mg/L.
CK, creatine kinase; CRP, C reactive protein.

However, on day 3, infection parameters and CK increased (table 1), and on day 4 the patient developed fever and tachycardia, and a symmetrical, diffuse erythema was noted bilaterally on his calves. He was still able to walk and did not have significant clinical signs of pneumonia. Furthermore, cultures from blood and tracheal secretion were negative. The CK level had risen to 29 900 U/L. The possibility of necrotising fasciitis or CS was considered and an orthopaedic evaluation was required.

An orthopaedic surgeon found moderate pain anteriorly in both crurae (visual analogue scale score 5/10). There was bilateral anterolateral crural erythema with no other skin changes on the rest of the body. There was some aggravation of bilateral pain in the anterior and peroneal muscle compartments on palpation and stretching of the corresponding fasciae. The muscle compartments were hard and the patient was unable to dorsiflex his feet. The posterior calf compartments were virtually pain free and soft on palpation. There was a pulse in both dorsal pedal arteries. Laboratory parameters (table 1) corresponded to a Laboratory Risk Indicator for Necrotizing Fasciitis (LRINEC) score of 6 points.[12] Necrotising fasciitis, or CS, was suspected and the patient was taken to the operating theatre for emergency anterolateral release of those muscle compartments. However, the decision was challenged at the time, as pain was considerably less than in a classical clinical presentation of CS. The antibiotic therapy was changed to intravenous ciprofloxacin, meropenem and clindamycin on an empirical basis. An intracompartmental pressure measurement was not performed.

One and a half hours later, the operation was performed on both crurae. The anterior and peroneal compartments were released through a single crural skin incision. The fasciae were intact and displayed no signs of infection or necrosis. The musculature was hard and grey, had sparse bleeding and displayed no certain signs of contractility. Musculofascial biopsies from each leg were sent for microbiological analysis but no bacteria were identified on microscopic examination or in cultures. Preoperative X-rays showed no bone anomalies. Postoperatively, pain levels subsided significantly. Three more revisions were required for full resection of all necrotic tissue in the compartments.

Leucocytes, C reactive protein and CK diminished considerably during days 5–10. The patient had orthoses applied to keep his feet plantigrade and was referred to an orthopaedic shoemaker for manufacturing of permanent drop foot orthoses. He was referred to physiotherapy.

OUTCOME AND FOLLOW-UP

During the postoperative period, the patient refused the aripiprazole medication. On day 11 he was transferred to a psychiatric hospital. There were no other incidents of polydipsia, no further pathological thirst drive and no further delusional incidents associated with water intake. The patient was confirmed to have a schizophrenic mental illness and generalised anxiety. He rejected medication with aripiprazole but did take olanzapine, which was later changed to risperidone. He experienced a beneficial effect and reverted from his severe delusional state. He was discharged after some months to a protected supervised environment. Four months postoperatively, the patient had permanent drop foot orthoses in his shoes. He was able to walk with a symmetric, discretely waddling gait and experienced no limitations on distance of ambulation.

DISCUSSION

We present a fulminant course of bilateral anterolateral crural CS preceded by water intoxication in a young man with no previous polydipsia behaviour and no history of hyponatraemia. The patient presented as a diagnostic challenge because of the rare constellation of findings and diminished pain expression possibly due to underlying schizophrenic disease.

Water intoxication is related to schizophrenia and 20% of patients with schizophrenia are estimated to suffer from psychogenic polydipsia.[13] The mechanism is unclear but an altered set point for vasopressin has been proposed.[14] However, in this case, the sodium and creatinine levels were normal 3 days before admission indicating a normal level of vasopressin and normal kidney function. Our patient's dysfunctional thirst drive seems to have been elicited by a delusional desire to soothe severe anxiety.

Acute hyponatraemia is a life-threatening condition resulting in cerebral oedema and risk of generalised convulsions. In this case, convulsions were not witnessed but considered highly probable and these could have elicited rhabdomyolysis and CS. However, rhabdomyolysis and CS developed after correction of hyponatraemia. Certain antipsychotic medications are also associated with hyponatraemia and rhabdomyolysis.[8 15] These include aripiprazole, which was prescribed at the psychiatric department; but only one dose was administered. Furthermore, previous treatment with aripiprazole was not associated with related side effects and an element of drug-induced rhabdomyolysis was considered unlikely. Lastly, it is important to consider the possibility of exertional rhabdomyolysis. The patient had been out for a 30 km walk and bicycle ride 1 day before admission to the psychiatric ward, but rhabdomyolysis and CS developed considerably later. Hence, a causal relationship seems improbable.

Why this patient developed rhabdomyolysis and CS is unknown, and possibly multifactorial. One hypothesis is that hyponatraemia decreases the ion exchange of calcium and sodium ions across the cell membrane. Thus, calcium levels would rise intracellularly, activating proteases and lipases, and lead to cell death. Such local muscle damage would release free radicals and intracellular toxins leading to further myolysis.[15–17] Others speculate that by ingesting vast amounts of water, the extracellular osmolarity would decrease. The muscle cells would swell, and this swelling would subside by the extrusion of potassium ions. This again would change the transmembrane potential and lead to myolysis.[15–17] Because such cascades possibly develop within hours, the proposed mechanisms may explain why myolysis was delayed, and why CK levels rose after hyponatraemia was identified and corrected, as observed in this case.[15] Furthermore, it has been speculated that expedient correction of hyponatraemia might lead to rhabdomyolysis.[18] However, a causal relationship could not be demonstrated in rats.[19] The sodium level in this case was intended to be corrected to 10 mmol/L/day. However, the sodium level was

unintentionally corrected faster despite repeated measurements. The rapid correction might be one of several concurrent reasons for CS to develop.

CS could develop due to myolysis and inflammation, causing the intracompartmental pressure to rise and compromise perfusion. This would expedite further muscle cell death and, hence, a continued building up of pressure.[8 17] Why only the anterolateral crural compartments were affected is unknown, but it might be due to an inert relatively high muscle to compartment volume in an unaffected extremity, or perhaps the fasciae in these compartments are simply less expandable than in others.

Having signs of CS and with some suspicion of musculofascial infection, an operative decompression of both lower limbs was performed. The decision was challenged by a surprisingly modest pain reaction, which directly contradicted the fulminant pattern reflected by the operative findings with ischaemic muscle in both anterolateral compartments. Retrospectively considered, this pattern of pain insensitivity is likely related to the underlying schizophrenic disease, as diminished pain expression can be a pronounced schizophrenic trait. The reason for this has been discussed in observational and experimental studies. It seems to be related to neurochemical mechanisms involved in pain perception, especially the excitatory system, but it may also depend on cognitive factors related to the recognition of pain. The impact of antipsychotic medication on pain perception is disputed. In the present case, however, the patient ingested aripiprazole on only one occasion. Thus, we find it unlikely that antipsychotic medication solely has changed the pain perception of pain.[11 20 21]

CONCLUSION

A combination of disposing factors seems to have elicited massive rhabdomyolysis and CS in this patient. Diagnosis and treatment were hindered because of possibly altered pain expression and infection. We find it important to direct attention to the seemingly uncharacteristic moderate pain level, which might, however, not be an unusual expression in a patient with schizophrenia. Care providers should be alerted to the possibility of rare and multifactorial causes of these conditions in particularly problematic cases.

Learning points

- Psychogenic polydipsia may elicit severe hyponatraemia and rhabdomyolysis. Under certain circumstances, this may lead to compartment syndrome.
- Patients with schizophrenia may have diminished pain expression leading to difficulty in interpreting normally painful conditions.
- Compartment syndrome may develop under certain circumstances with no trauma or ischaemia.

Competing interests None.

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