Unusual presentation of more common disease/injury

Vanishing polyuria and respiratory failure

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Summary

A 44-year-old man with headache, sweating, subfebrile temperature and profound fatigue was found to have hypercalcaemic crisis with renal failure. Despite standard therapy, calcium levels remained high, he became anuric and developed multi-organ failure with acute respiratory distress syndrome requiring high ventilatory support, norepinephrine, dobutamine and continuous veno-venous haemodiafiltration. Ectopic calcification was found in the lungs, in the thyroid, kidneys, heart and stomach. A large parathyroid adenoma was then removed. When last seen, 11 months after surgery, the patient no longer required oxygen, and total lung capacity had returned to normal.

BACKGROUND

Hypercalcaemia preceding and presenting later with kidney failure is an unusual cause of ectopic calcification. Primary hyperparathyroidism is a common condition, whereas parathyroid crisis with circulatory shock and multiple organ failure is rare. Aggressive fluid replacement is crucial for the emergency management of hypercalcaemia. In case of severely restricted diuresis and multi-organ failure, continuous renal replacement therapy (preferably, veno-venous haemofiltration with citrate) is the treatment of choice and should be considered at the onset.

CASE PRESENTATION

A 44-year-old man presented to the emergency with a 2-day history of headache, sweating, subfebrile temperature and profound fatigue. The day before, his physician suspected urinary tract infection and prescribed ciprofloxacin. At presentation, his medical history was unremarkable and he was taking no regular medication. However, for the past 7 months, according to his wife, he had suffered from increasing fatigue, muscle weakness, constipation, polydipsia, polyuria and nocturia; the latter, disappearing 2 weeks previously. On examination, the patient was in distress. Body temperature was 37.1°C, blood pressure 90/40 mm Hg, heart rate 80/min and respiration rate 47/min.

INVESTIGATIONS

Urine analysis showed erythrocytes, leucocytes and protein (6 g/l), but no bacteria. Blood tests showed haemoglobin of 143 g/l, leucocytes, $25.7 \times 10^9/l$; C-reactive protein, 275 mg/l; creatinine, 420 $\mu\text{mol/l}$; glucose, 6.2 mmol/l; sodium, 127 mmol/l; potassium, 7.6 mmol/l; CK, 1051 U/l (<190); phosphate, 1.88 mmol/l (0.87–1.45); total calcium, 5.56 mmol/l (2.09–2.54); albumin, 38 g/l; and parathyroid hormone, 3323 ng/l (15–65). ECG showed diffuse repolarisation abnormalities, troponin-T was 8.08 $\mu\text{g/l}$ (<0.03) and echocardiography revealed diffuse ventricular hypokinesis with an ejection fraction of 25%.

A chest radiograph showed a wide, upper mediastinum and the following CT scan showed a large cystic mass at the lower pole of the thyroid gland extending into the mediastinum (figure 1). Because of clinical evolution with high ventilatory demand, acute respiratory distress syndrome (ARDS), severe hypercalcaemia and ectopic calcification were suspected: $^{99\text{m}}\text{Tc-DPD}$ bone scintigraphy^{1 2} revealed marked isotope uptake in the thyroid, lungs, kidneys, heart and stomach (figure 2).

TREATMENT

The next day, despite fluid replacement and zoledronate treatment, calcium levels remained high (4.12 mmol/l); urine output was not normalised. Piperacillin–tazobactam was given for possible urosepsis. However, blood cultures remained negative. The patient developed multi-organ

Figure 1 CT of the upper mediastinum with parathyroid mass (arrows) and bilateral pleural effusion.

Figure 2 Bone scintigraphy (a=anterior, b=posterior view) showing increased isotope uptake in the thyroid, in the lungs, kidneys, heart and stomach.

failure with ARDS requiring ventilatory support, dobutamine and norepinephrine. Continuous veno-venous haemodiafiltration with citrate anticoagulation³ was started. Removal of the 6.2×4.7×7.7 cm tumour (histology: parathyroid adenoma) on day 16 after admission required both trans-sternal and cervical access and double lung ventilation; open lung biopsy showed ectopic calcification.

OUTCOME AND FOLLOW-UP

By day 4, his clinical condition improved; calcium levels decreased to 2.35 mmol/l and creatinine to 163 µmol/l. On further questioning, the patient stated that his physician

had found hypercalcaemia 2 years ago; however, there was no follow-up. Pulmonary function tests the day before hospital discharge showed severely reduced total lung capacity (TLC, 44% of predicted) and CO diffusion capacity (DLCO) (28% of predicted). He was discharged on oxygen 6 weeks after surgery. When last seen, 11 months after surgery, he no longer required oxygen, TLC returned to normal and DLCO improved to 51%.

DISCUSSION

Aggressive fluid replacement is essential for emergency management of hypercalcaemia, and bisphosphonates are an established cornerstone to block bone resorption. In case of severely restricted diuresis and multi-organ failure, continuous renal replacement therapy (preferably, veno-venous haemofiltration with citrate) is the treatment of choice and should be considered at the onset. Systemic calcinosis is a potential complication of severe hyperphosphataemia. In patients on dialysis it may present as painful skin necrosis (calciophylaxis), while kidney and lung calcinosis may remain asymptomatic. Hypercalcaemia preceding and presenting later with kidney failure and hyperphosphataemia is an unusual cause of ectopic calcification and markedly impaired lung diffusion capacity. Marked hypercalcaemia and metastatic calcification have been reported in a patient with parathyroid carcinoma who died 24 h after admission.⁴ We are not aware of similar reports of parathyroid adenomas leading to circulatory shock with multiple organ failure.

Learning points

- ▶ Primary hyperparathyroidism is a common condition, mostly of relatively benign course,⁵ whereas parathyroid crisis with multiple organ failure is rare.
- ▶ Aggressive fluid replacement is crucial for emergency management of hypercalcaemia.
- ▶ By restricted diuresis, continuous renal replacement therapy is the treatment of choice and should be considered at the onset.

Competing interests None.

Patient consent Obtained.

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