

# Idiopathic hypoparathyroidism, reversible cardiomyopathy and nephrogenic diabetes insipidus - case report

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## ABSTRACT

*We are presenting a case of a 36-year-old patient with idiopathic hypoparathyroidism and reversible dilated cardiomyopathy as a result of hypocalcaemia. Twelve years later, the patient presented a picture of nephrogenic diabetes insipidus, which according to available literature has so far not yet been described.*

**Key words:** hypoparathyroidism, cardiomyopathy, nephrogenic diabetes insipidus

## Introduction

Idiopathic hypoparathyroidism manifests with signs of hypocalcaemia. Dilated cardiomyopathy caused by hypocalcaemia is rare. Association between idiopathic hypoparathyroidism and nephrogenic diabetes insipidus is also rare.

## Case report

A thirty-six-year-old patient was transferred to the intensive care unit from the department of cardiology because of deterioration of cardiac decompensation. This was a patient who was treated for alcoholism seven years ago and has been abstinent since. Four years ago he was hospitalized for acute psychosis. He was already treated at the Department of Cardiology 2.5 months earlier for recurrent pulmonary oedema and dilated cardiomyopathy.

The patient was of medium height, conscious, afebrile and eupneic. Lung auscultation showed rales on both sides of the lungs. The action of the heart was

rhythmic with quiet tones, and blood pressure was 135/80 mm Hg. The extremities were without oedema. A phenomenon of gear and "gynecological hand" were found in neurological status with the tongue and left hand tremor and increased muscle tone. The psychiatrist stated that the patient had signs of psychotic anxiety that occasionally had the proportions of panic and fear of immediate death. Attacks were mostly present at night, lasting about five minutes, accompanied by hyperventilation with the worsening of extrapyramidal side effects. He was treated with biperiden, clozapine, fluvoxamine, furosemide 1 tablet at 40 mg every other day with 500 mg of potassium chloride.

Arterial blood gas measurements and acid-base status showed metabolic alkalosis and hypoxemia. Corrected calcium was 0.1 mmol/l (normal values 2.25-2.75 mmol/l), phosphorus 3.5 mmol/l (normal values 0.65-1.62 mmol/l), potassium 3.0 mmol/l (normal values 4.1-5.5 mmol/l), parathyroid hormone (PTH) 4.4 pg/ml (normal values 20 - 90 pg/ml). Magnesium, as well as T3, T4 and TSH, were within limits. The

X-ray of the heart and the lungs showed an enlarged cardiac shadow with severe intrapulmonary stasis. The electrocardiogram revealed sinus rhythm, frequency of 108/min and prolonged corrected QT interval (QTc) of 517 ms.

An ultrasound of the heart showed dilated cardiomyopathy (left ventricular-diastolic diameter-LVDd) with reduced ejection fraction (EF) of 48% and severe interventricular septum (IVS) and apex hypocontractility, as well as increased atria - left atrium (LA) 50 mm with severe degree tricuspid and mitral regurgitation (both 3-4 +). IVS thickness was 12 mm, and the posterior LV wall (LVPW) 10 mm. A brain CT scan pointed out almost symmetrical calcifications in the area of the basal ganglia (in the heads nn. caudati, nn. lentiform as both thalamus pulvinars). An elongated calx shadow was visible in the left hemisphere of the cerebellum. The ocular fundus examination was normal.

Considering the test results, a diagnosis of primary idiopathic hypoparathyroidism was made. The therapy involved the preparation of calcium and calcitriol with furosemide, spironolactone, biperiden,

sulpiride, nitrazepam. The therapy led to an improvement of the general condition of the patient, and the patient's heart compensated. Control tests showed the following values: corrected calcium 1.7 mmol/l, phosphorus 2.36 mmol/l, potassium 4.5 mmol/l, the electrocardiogram monitored the reduction in QTc - 395 ms. A neurological examination was made after a few days and it showed no disturbances other than the patient's slow thought process and incomplete space orientation. The patient was discharged after 32 days in the hospital with the recommendation of further outpatient follow-ups with a cardiologist, an endocrinologist and a psychiatrist.

Twelve years later, the patient was hospitalised in the nephrology department for a diagnostic treatment of hypertension, polyuria and polydipsia that had been present for two years. The patient was monitored for psychosis by a psychiatrist. A cardiologist monitored the reversibility of dilated cardiomyopathy. The patient was on fluphenazine, biperiden, risperidone, zolpidem, atorvastatin, lisinopril, atenolol, CaCO<sub>3</sub> and calcitriol therapy. A cardiac ultrasound was done and showed left ventricular concentric hypertrophy, incomplete diastolic relaxation without regional contractility disturbances, functional valves, moderate tricuspid regurgitation with no indirect signs of pulmonary hypertension (LVDd 3.9 cm, right ventricle 1.0 cm, LA 4.8 cm, IVS 1.7 cm, LVPW 1.3 cm, EF 73 %). The values of urea, creatinine, calcium, phosphorus, potassium, magnesium, TSH, T3 and T4 were within normal limits. PTH was < 6 pg/ml, the creatinine clearance was 65.9 ml/min and

the protein in 24 h urine 0.36g. QTc was 400 ms. The kidneys were examined by ultrasound and were of normal size. Holter blood pressure recorded an average value of 135.2/97 mm Hg and the brain CT scan showed no significant changes compared to the scan made 12 years ago. Aldosterone and plasma renin values were within normal limits as was the value of antidiuretic hormone (ADH) in the urine. Urine concentration test indicated the isosthenuria (plasma osmolality 290 mOsm/kg, urine osmolality 150 mOsm/kg) with diuresis 3000-5500 ml for 24 h. Renal biopsy found a 20% altered glomeruli (connective tissue) without significant interstitial fibrosis, tubular atrophy and without significant changes in the blood vessels. Based on these results, we concluded that the patient suffered from nephrogenic diabetes insipidus, which required further monitoring. The patient was discharged in good general condition with the recommendation for further follow-ups.

## Discussion

Idiopathic hypoparathyroidism manifests with signs of hypocalcaemia such as tetany, epileptic seizures, hypotension (in acute hypocalcaemia), prolongation of the QT interval in the electrocardiogram, papilloedema and psychiatric manifestations including psychosis. A few cases of congestive heart failure and dilated cardiomyopathy are also described. Although the mechanism remains undefined, calcium plays a critical role in excitation-contraction coupling and is required for epinephrine-induced glycolysis in the heart. (1) As a result of

long-term hypoparathyroidism, calcifications of the basal ganglia may occur, which may or may not be associated with extrapyramidal symptoms. (1)

According to available data to date, the literature has described 29 cases of hypocalcemic cardiomyopathy in patients of all ages. (2-4)

Our case concerned hypoparathyroidism with a distinct clinical picture of hypocalcaemia, including dilated cardiomyopathy that withdrew after treatment with preparations of calcium and vitamin D. The psychiatric and neurological disorders that were present at hospital admission were also significantly improved due to calcium correction.

Twelve years later, the patient was diagnosed with idiopathic nephrogenic diabetes insipidus. Theoretically, transient hypercalcaemia could have been the cause, possibly as a consequence of hypoparathyroidism treatment. (5) To our knowledge, one case of autosomal dominant hypoparathyroidism and nephrogenic diabetes insipidus has been described so far. (6) There are no data so far indicating that the drugs that the patient took could be associated with the development of nephrogenic diabetes insipidus.

## Conclusion

We have presented a case of a 36-year-old patient with idiopathic hypoparathyroidism and reversible dilated cardiomyopathy as a result of hypocalcaemia. Twelve years later, the patient presented a picture of nephrogenic diabetes insipidus, which according to available literature has so far not yet been described.

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