CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 51 (2018) 335–339

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Treatment of hypocalcemia in hungry bone syndrome: A case report

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ARTICLE INFO

Article history:
Received 1 March 2018
Received in revised form 29 July 2018
Accepted 6 August 2018
Available online 13 August 2018

Keywords:
Hungry bone syndrome case report
Parathyroid hormone
Primary/secondary hyperparathyroidism
Hypocalcemia
Parathyroidectomy
Calcium gluconate/citrate
Calcitriol
Elemental calcium

ABSTRACT

INTRODUCTION: Hungry bone syndrome (HBS) is rapid, intense and prolonged hypocalcemia that follows parathyroidectomy. The focus of this paper is HBS in patients with secondary hyperparathyroidism (SHPT) due to end stage renal disease (ESRD). Various risk factors are correlated with developing HBS post-parathyroidectomy due to SHPT which include: old age (>60 years); the preoperative level of parathyroid hormone (PTH); increased osteoclasts; and evidence of bone disease before surgery.
PRESENTATION OF CASE: A 25-year-old woman, who underwent parathyroidectomy of all four parathyroid glands due to SHPT caused by ESRD. Her calcium deficit was prolonged, as expected in patients who undergo parathyroidectomy, however her calcium levels remained low despite unprecedented supplementation of elemental calcium and calcitriol.
DISCUSSION: Unfortunately, there is not enough data-based evidence to help prevent or minimize severe complications of hypocalcemia prior to parathyroidectomy. The main goal of treatment is replenishing the calcium deficiency through supplementation with calcium salts, high doses of active metabolites of vitamin D, and electrolytes.
CONCLUSION: The ultimate goal of reviewing and analyzing this particular case is to obtain a better understanding for the treatment of Hungry bone syndrome. Although, there are very few cases as severe as this patient, hopefully this case study will result in greater insight and lead to improvement in the overall treatment of hypocalcemia.

1. Introduction

HBS is considered in patients following parathyroidectomy when the hypocalcemia is prolonged and severe, specifically total serum calcium less than 8.4 mg/dL (2.1 mmol/L) or ionized calcium less than 4.48 mg/dL (1.12 mmol/L) for more than four days post-surgery. It is believed to be caused due to the sudden drop in PTH levels and the impact that the hormone had on osteoclastic resorption [1]. The decrease in PTH is a result of unopposed activation of osteoblasts and increased influx of calcium into "calcium-starved" bone. HBS is reported most often after surgery for parathyroidectomy or thyroidectomy because of primary or secondary hyperparathyroidism. The longer the duration of elevated levels of PTH in the system, the greater the severity of hypocalcemia post parathyroidectomy.

There are few reported cases of HBS in literature, and data regarding incidence is sparse and varied. Patients who undergo parathyroidectomy due to primary hyperparathyroidism are believed to experience a milder form of the syndrome in comparison to patients with SHPT who develop a more severe and persist HBS. The incidence of the syndrome due to primary hyperparathyroidism (PHPT) ranges from 13 to 20%, while it can range from 27 to 51% for patients with SHPT [2]. Another review done in 2012 reported HBS to be found in 4 to 87% of patients who underwent parathyroidectomy [3].

The sudden decline of PTH causes an increase in bone density, and the longer the syndrome persists, the greater the increase in bone density. Some risk factors for developing HBS include: increased age; elevated levels of PTH preoperatively; increased number of osteoclasts on bone biopsy; as well as radiological evidence of bone disease prior to surgery [4].

Some clinical manifestations of severe hypocalcemia include perioral paresthesia, positive Chvostek and Trousseau signs, convulsions, carpopedal spasms, laryngospasm, and cardiac arrhythmias. Generally, six to 12 g per day of elemental calcium is considered safe to administer to patients, along with 2–4 μg/day of calcitriol [5]. The case discussed below is HBS with severe hypocalcemia in a 25-year-old patient following parathyroidectomy due to secondary hyperparathyroidism, that required unprecedented amounts of calcium during her post-operative treatment. This case study is constructed in order to satisfy the scare criteria [6].

2. Case report

A 25-year-old woman presented to the ENT clinic on April 2017 with complaints of generalized weakness and difficulty walking

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https://doi.org/10.1016/j.ijscr.2018.08.011

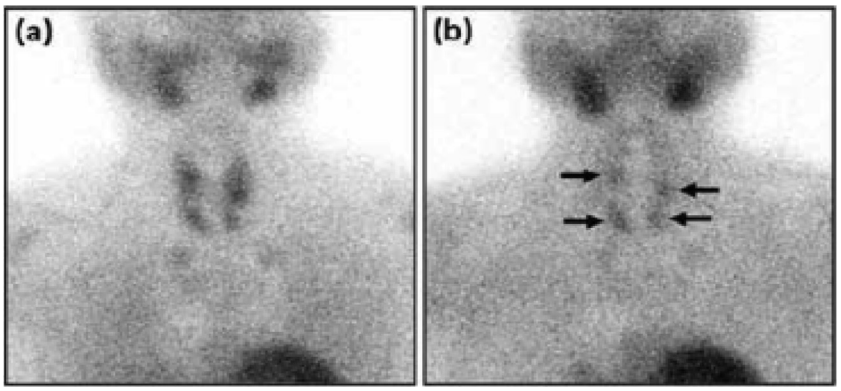

Fig. 1. 99mTc-sestamibi dual phase images showing a 4-gland parathyroid hyperplasia.
(a) Tracer uptake by the thyroid and parathyroid glands in the early phase image.
(b) Delated tracer washout from all the hyperplastic parathyroid glands (the arrows), in the late phase image.

due to progressive worsening of leg pain. The leg pain had started several months before and was initially mild at onset, however, now caused the patient significant pain. Her past medical history included Bartter's syndrome (diagnosed at age two), ESRD (2014), secondary hyperparathyroidism, previous pulmonary embolism (2016) and anemia of chronic disease. She was currently taking iron, thiamine, zinc, vitamin c, mag-ox, aspirin, lovenox and KCl. On physical examination- height was 158 cm, weight was 80.5 kg, vital signs were within normal limits. No other significant findings were noted.

The PTH level was 1849 pg/mL during the initial visit, therefore a Sestamibi scan was performed, however the scan did not show a localized adenoma. Fig. 1 shows a Sestamibi scan for a patient with secondary hyperparathyroidism.

The patient was immediately scheduled for resection of all four parathyroid glands based on several factors; including her PTH level, complaints of generalized weakness and worsening bone pain. The procedure was performed under general anesthesia, with no complications. Sections of the parathyroid glands were taken and sent for pathology. The PTH level intraoperatively was noted to be 71 pg/mL. The patient was then admitted to the ICU in stable condition following surgery. Goals that were set for the patient following surgery- 8.0–8.5 mg/dL in serum calcium and 4.5 mg/dL for ionized calcium.

On postoperative day (POD) one, the nephrologist was put on consult and became part of the patient's care team till her discharge. They helped to determine daily amount of calcium administered and whether any adjustments were needed. Other recommendations made by the nephrologists: daily weights, hemodialysis twice per week, vitamin D supplements, and close observation of magnesium and phosphorus levels.

The development of hypocalcemia in the patient was rapid and progressive. On POD 1, the patient had a PTH level of 12 units/l, serum calcium level of 7.3 mg/dL and an ionized calcium of 2.9 mg/dL. However, she was asymptomatic. POD 2, the patient was stable and still in ICU, but complained of pain at the intravenous (IV) site on her right hand.

She was scheduled for hemodialysis with high-calcium bath twice a week. The patient's weight decreased 25 kg (from 80.5 kg to 55.4 kg) within a span of four days post-surgery. The baseline weight of the patient being unknown it can be assumed that the 80.5 kg was substantially higher than her norm and could potentially be attributed to the patient's missed dialysis sessions as well as the bilateral lower extremity edema. Once the patient was able to tolerate oral calcium supplements she was transferred from the ICU to the internal floor. The patient received 11.37 g of elemental calcium, consisting of 27.5 g of calcium carbonate and 4 g of calcium gluconate, on her first day of oral calcium replacement. The concentrations of elemental calcium increased substantially thereafter, from 11.37 g to 17 g → 30 g → 35.9 g. The highest amount of calcium was provided on POD 11 where 28 500 (x6) g of calcium citrate was given to the patient, resulting in 35.9 g of elemental calcium. During the 11 days of treatment her total serum calcium maintained at an average of 7.8 mg/dL and ionized calcium at 4 mg/dL, while these levels are considered low they did not indicate the indicate to start the patient on intravenous calcium. Fig. 2 illustrates the patient's changes in serum/ ionized calcium and PTH levels. The amount of calcium which this patient received has never been reported or recommended for any patient with HBS. The current recommendations are between 6–12 g/day [3]. The patients POD 7 laboratory values are presented in Table 1.

Table 1
Laboratory Results for Postoperative Day 7[a].

Hematologic Labs		Routine Chemistry	
WBC	6.4 × 10(3)/mcL	Glucose	96 mg/dL
RBC	2.5 × 10(6)/mcL	Calcium level	7.4 mg/dL
Hemoglobin	6.4 gm/dL	Sodium	140 mmol/L
Hematocrit	20.30%	Potassium	4.3 mmol/L
MCV	81 fL	Chloride	102 mmol/L
Platelet	162 × 10(3) mcL	CO2	27 mmol/L
Iron	37 mcg/dL	Alk Phos	696.0 unit/L
TIBC	195 mcg/dL	BUN	21 mg/dL
Ferritin	535 ng/mL	Creatinine	4.51 mg/dL
Transferrin	130 mg/dL	BUN/Creatinine Ratio	5
		Albumin Level	2.7 gm/dL
		Total Protein	5.0 gm/dL

TIBC: Total Iron Binding Capacity, WBC: white blood cell count, BUN: blood urea nitrogen, Alk Phos: Alkaline phosphatase, RBC: Red blood cell count, MCV: Mean corpuscular volume.
[a] POD 7 is when the patient was transferred from the ICU to the internal team for care.

The treatment was stopped abruptly due to the patient's request. She was discharged with 2 days of medication and given prescription for another 14 days. The importance of maintaining her bi-weekly dialysis appointments was discussed with the patient, where her serum calcium, ionized Ca, BMP and Alkaline phosphatase will be closely monitored. Calcium levels on discharge were - serum calcium of 9.5 mg/dL and ionized calcium of 4.9 mg/dL. Had the patient not requested discharge, the medical team wanted to continue the ongoing treatment plan. That plan involved providing the necessary amount of calcium, closely monitoring her serum levels as well as her weight for an additional few days (Figs. 3 and 4).

The discharge medications included: calcium citrate 30 g/QID; calcitriol 5 mcg/daily; calcium citrate 200 mg/QID; epoetin alfa 10