Asian Journal of Surgery (2016) 39, 255–259

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CASE REPORT

# Parathyroid carcinoma in tertiary hyperparathyroidism

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Received 6 August 2012; received in revised form 6 November 2012; accepted 9 January 2013
Available online 26 March 2013

**KEYWORDS**
kidney transplantation;
parathyroid carcinoma

**Summary** Parathyroid carcinoma is a rare disease of unknown etiology. This study presents a case of parathyroid carcinoma in a patient with tertiary hyperparathyroidism. Despite a successful kidney transplantation, the intact parathyroid hormone (iPTH) level of the patient was elevated consistently and could not be controlled by medical therapy. Due to the development of tertiary hyperparathyroidism with bone pain and osteoporosis, subtotal parathyroidectomy was performed 4 months after the kidney transplantation. Histological evaluation revealed that one of four parathyroid lesions was a parathyroid carcinoma, while the others were diffuse hyperplasia. Postoperative laboratory studies indicated a decreased level of iPTH. A positron emission tomography–computed tomography performed 6 months after the operation revealed no evidence of local recurrence or distant metastasis.


## 1. Introduction

Parathyroid carcinoma is a rare disease of unknown etiology.[1] Here, we report a case of parathyroid carcinoma in a patient with tertiary hyperparathyroidism.

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http://dx.doi.org/10.1016/j.asjsur.2013.01.007

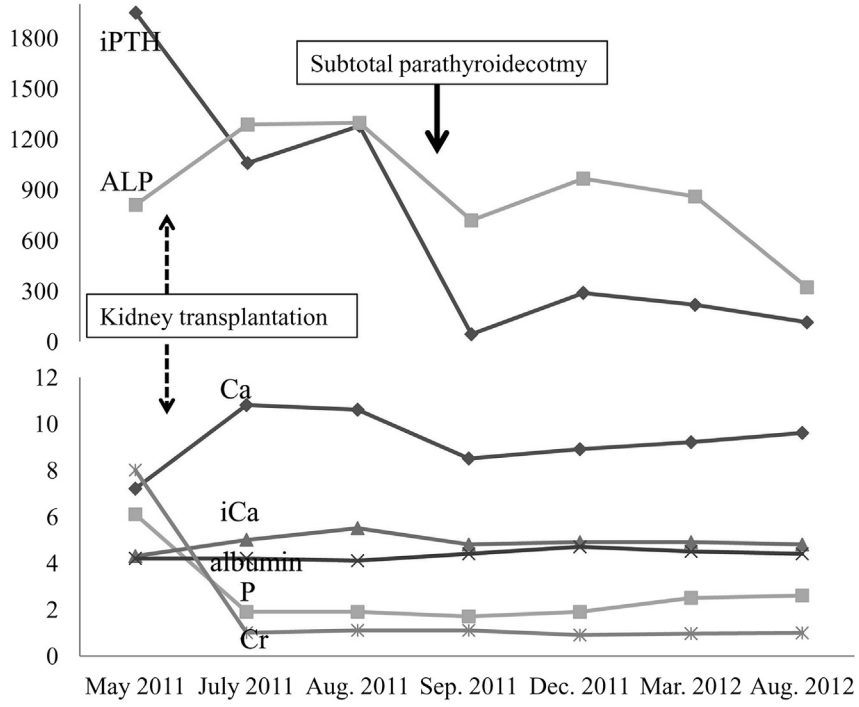
## 2. Case report

A 57-year-old man had received long-term hemodialysis (for 11 years) to manage his chronic renal failure caused by polycystic kidney disease. In 2011, he underwent a kidney transplant; since then he has never experienced any complications or rejection reactions.

Blood tests performed prior to the kidney transplantation indicated that serum total calcium (Ca) and ionized Ca levels were 7.2 mg/dL (normal, 8.2—10.4 mg/dL) and 4.3 mg/dL (normal, 4.5—5.3 mg/dL), respectively. Serum phosphorus, alkaline phosphatase (ALP), and intact parathyroid hormone (iPTH) levels were found to be 6.1 mg/dL (normal, 1.9—4.4 mg/dL), 810 IU/L (normal, 104—338 IU/L), and above 1900 pg/mL (normal, 14—72 pg/mL), respectively. Postoperative laboratory tests performed 3 months following a kidney transplant surgery showed an elevated level of serum Ca and ALP, which were increased to 10.6 mg/dL and 1297 IU/L, respectively. Blood tests also confirmed a slightly decreased level of iPTH, which was measured to be 1278.4 pg/mL (Fig. 1). The patient took the medication cinacalcet to manage his markedly elevated serum Ca and iPTH levels, but the treatment was ineffective. He complained consistently of knee and ankle pain.

To treat symptomatic tertiary hyperparathyroidism with osteoporosis, the patient was referred to endocrine surgeon. The mean T-score of the lumbar spine (L1—L4), measured by dual-energy X-ray absorptiometry, was −3.1. Computed tomography of the neck showed three enlarged parathyroid glands (Fig. 2). Fig. 3 represents a 2-hour delayed 99mTc-sestamibi scintigraphy (MIBI scan) showing an increased uptake in the right inferior parathyroid gland. During the neck exploration, four enlarged, soft, oval, and yellowish parathyroid glands were observed (right superior: 1.8 × 1.5 $cm^2$; right inferior: 1 × 1 $cm^2$; left superior: 1 × 0.7 $cm^2$; left inferior: 2.5 × 1.8 $cm^2$). Frozen biopsy performed intraoperatively on the left superior parathyroid revealed parathyroid hyperplasia. Left superior parathyroid gland was preserved partially, while other lesions were removed completely.

Histological findings revealed a parathyroid carcinoma on a 1.7 × 1.5 × 1.0 $cm^3$ mass of the right superior parathyroid gland; its capsular penetration was present and vascular or perineural invasion was absent. Its surgical resection margin was clear. Histological evaluation

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|---|---|---|---|---|---|---|---|
| iPTH (pg/mL) | 1900 | 1058.4 | 1278.4 | 43.6 | 288.3 | 217.8 | 114 |
| ALP (IU/L) | 810 | 1287 | 1297 | 718 | 966 | 860 | 322 |
| Ca (mg/dL) | 7.2 | 10.8 | 10.6 | 8.5 | 8.9 | 9.2 | 9.6 |
| iCa (mg/dL) | 4.3 | 5 | 5.5 | 4.8 | 4.9 | 4.9 | 4.8 |
| albumin(g/dL) | 4.2 | 4.2 | 4.1 | 4.4 | 4.7 | 4.5 | 4.4 |
| P (mg/dL) | 6.1 | 1.9 | 1.9 | 1.7 | 1.9 | 2.5 | 2.6 |
| Cr (mg/dL) | 8 | 1 | 1.1 | 1.1 | 0.9 | 0.96 | 0.99 |

**Figure 1** Serum iPTH, ALP, Ca, iCa, albumin, P, and Cr levels from kidney transplantation to subtotal parathyroidectomy. ALP = alkaline phosphatase; Ca = calcium; Cr = creatinine; iCa = ionized calcium; iPTH = intact parathyroid hormone; P = phosphorus.