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CASE REPORT – OPEN ACCESS

A Case of Primary Hyperparathyroidism Coincident with Advanced Iliac Artery Aneurism

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ABSTRACT

Background: Primary hyperparathyroidism (PHPT) is a clinical entity characterized by clinical symptoms specific to high parathyroid hormone (PTH) and hypercalcemia. Patients are generally asymptomatic, and high calcium levels are detected by biochemical tests. We present a case of a patient with a rare vascular complication of hyperparathyroidism.

Case Report: In our 58-year-old male patient, a high calcium level was attributed to PHPT. The iliac artery aneurysm detected in our patient was evaluated as a complication of an elevated PTH level. No risk factors for IAA were detected in the patient's history. Surgery was performed for the iliac artery, followed by parathyroidectomy.

Conclusion: In this case report, a PHPT case without a risk factor for IAA is presented. This case demonstrates the relation between higher PTH levels and vascular damage. This case presented with the absence of advanced-stage iliac artery aneurism with PHPT coexistence.

Keywords: Primary hyperparathyroidism, iliac artery aneurism, vascular complications

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INTRODUCTION

Primary hyperparathyroidism (PHPT) is a clinical condition characterized by elevated serum parathyroid hormone (PTH) and calcium levels. Patients often remain asymptomatic, and usually, hypercalcemia is detected during routine biochemical examination. Typical symptoms include bone pain, osteopenia, osteoporosis, and kidney stones, and patients may also present with atypical symptoms. In this report, we present a case of a patient with PHPT who presented with bilateral iliac artery aneurysm.

CASE REPORT

A 58-year-old male patient presented to another health center with swelling and weakness in his legs and abdominal pain and then was referred to our clinic after the detection of hypercalcemia. On examination, the mean arterial pressure and heart rate were 100/70 mmHg and 95 beats/min, respectively, and the cardiac and pulmonary sounds were normal. There was no defense or rebound on abdominal examination. Bilateral (++/++) pretibial edema was observed. On electrocardiography (ECG), a normal sinus rhythm was noted. In biochemical analysis, the calcium level was 12.9 mg/dL (the corrected calcium level was 13.7 mg/dL), and the PTH level was 1,027.4 pg/mL (the baseline laboratory parameters are shown in Table 1). Abdominal ultrasonography (USG) showed a 14-mm kidney stone in the proximal one-third of the right kidney and dilatation of the renal collecting system proximal to the kidney stone. An arterial and venous Doppler USG showed an impression of bilateral iliac artery aneurysm. Subsequently, computed tomography (CT) showed thrombotic fusiform aneurysm in bilateral main iliac arteries, which had a length of 11 cm, outer diameter of 53 mm, and lumen diameter of 25 mm in the right iliac artery and a length of 14 cm, outer diameter of 106 mm, and lumen diameter of 85 mm in the right iliac artery. Additionally, CT showed thrombotic fusiform aneurysm in the left internal iliac artery, which had a length of approximately 7 cm, outer diameter of 63 mm, and lumen diameter of 11 mm (Fig. 1). On ECG examination, the diameter of the ascending aorta was 47 mm; no dissection was observed; the ejection fraction (EF) was 60%; the pulmonary artery pressure was 40–45 mmHg; and mild aortic, mitral, and tricuspid insufficiencies were detected. The patient was consulted to the cardiovascular surgery department, which recommended the normalization of the serum calcium level prior to the surgery. Neck USG showed an impression of a 40 × 22-mm mass suggestive of parathyroid adenoma in the left parathyroid gland. Parathyroid scintigraphy visualized a mass suggestive of parathyroid adenoma that covered two-thirds of the thyroid gland in late images and showed diffuse MIBI involvement (Fig. 2). Neck magnetic resonance imaging showed a 24 × 33-mm heterogeneous hyperintense nodular lesion suggestive of parathyroid adenoma at a location posterior to the left thyroid lobe in T2A images (Fig. 3). All three imaging modalities showed the mass at a location inferior to the left thyroid lobe. Since surgery was the primary form of treatment due to the

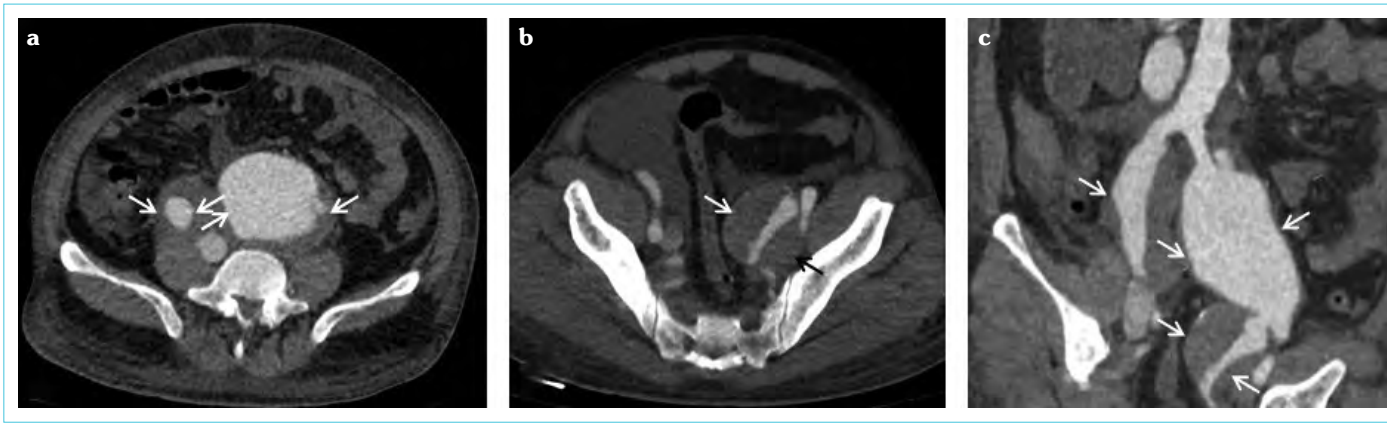

Figure 1. **Axial (a, b) and curved MPR (c) CT angiography images show bilateral; partially thrombosed common iliac and left internal iliac artey aneurysms (arrows)**

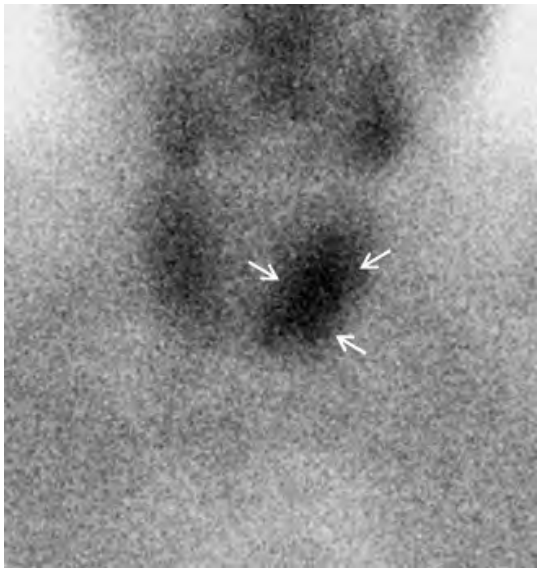

Figure 2. **Anterior view of late-phase parathyroid scintigraphy with 99mTc- sestamibi shows intense activity located on inferior 2/3 of the lobe of the thyroid supporting the diagnosis of parathyroid adenoma (arrows)**

presence of aneurysm, the patient was initiated on a pamidronate 60-mg therapy. Following this therapy, the serum calcium level decreased to 10.5 mg/dL. Subsequently, the serum calcium level was maintained within the normal range with a cinacalcet (3 × 30 mg) therapy, and the patient was transferred to the cardiovascular surgery department. The patient was followed up with the cinacalcet therapy for the following 2 months, and parathyroidectomy was performed when the patient's general condition improved. The postoperative PTH levels were normal, and no complication was observed intraoperatively and postoperatively. Table 1 presents the comparison of preoperative and postoperative laboratory parameters. Histological analysis showed two mitoses per 10 high power fields. Moreover, no clear capsule formation and no lymphovascular or perineural invasion were observed. In immunohistochemical analysis, the cells were positive for PTH and negative for Thyroid transcription factor-1 and calcitonin, and the Ki-67 proliferation index was 1%. Based on histological and immunophenotypic analyses, the patient was considered as having atypical parathyroid adenoma since the findings did not meet the malignancy criteria. The patient is still followed up by our department at regular intervals.

Table 1. Comparison of laboratory parameters preoperatively and postoperatively

	Pre.	Post.	RR
Blood urea nitrogen (BUN) (mg/dL)	17	8	6–20
Creatinine (mg/dL)	0.94	1.27	0.67–1.17
Calcium (mg/dL)	12.9	8.6	8.8–10.6
Phosphor (mg/dL)	1.42	2.42	2.5–4.5
Albumin (g/dL)	3	3.4	3.5–5.2
Magnesium (mg/dL)	1.98	1.39	1.7–2.55
Alkaline phosphatase (U/L)	132	100	30–120
Parathyroid hormone (PTH) (pg/mL)	1027.4	4.8	12–88
24-h urine calcium (mg/day)	271.5	100	<300
25-OH vitamin D3 (µg/L)	8.8	15.8	20–120

Pre: Preoperative; Post: Postoperative; RR: Reference range

DISCUSSION

Patients with PHPT have an increased mortality risk before and after parathyroidectomy due to cardiovascular reasons (1). In epidemiological controlled studies, increased calcium and PTH levels have been associated with increased cardiovascular mortality in patients with PHPT (2).

Isolated iliac artery aneurysm (IIAA) is relatively uncommon, which has been shown to have an incidence of 0.03% in the general population (3). Moreover, IIAA accounts for 0.4%–1.9% of all aneurysms (4). Risk factors for degenerative IAA are similar to those of other degenerative large vessel aneurysms (abdominal aortic aneurysm, popliteal aneurysm) and include male sex, White race, advancing age, history of smoking, hypertension, and atherosclerotic disease (5). In the case presented, ECG showed an increased pulmonary artery pressure (40–45 mmHg); an EF of 60%; an ascending aortic diameter of 46.5 mm; a normal aortic valve; and mild mitral, tricuspid, and pulmonary insufficiencies. Due to the absence

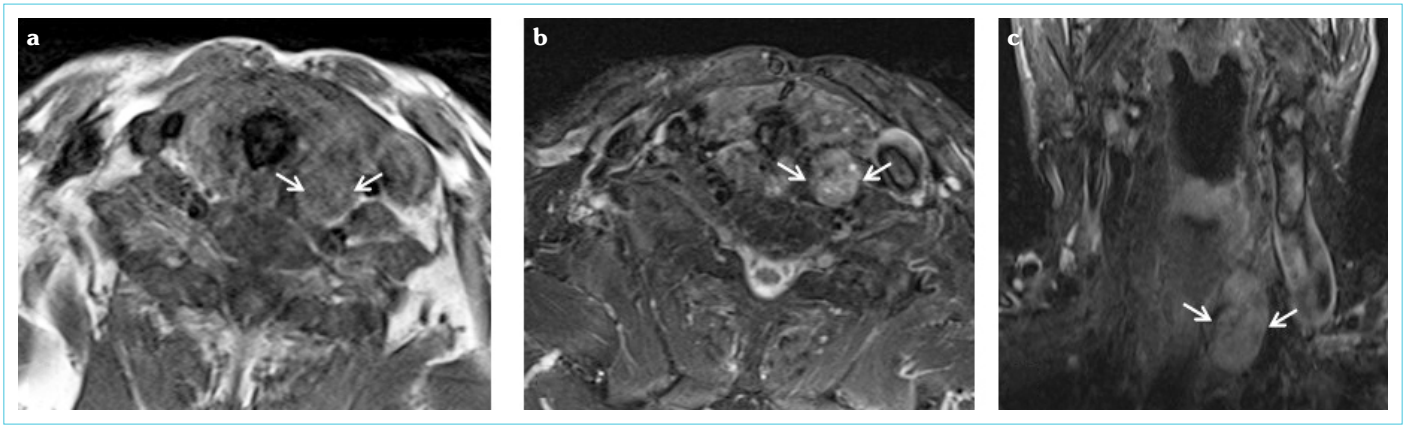

Figure 3. Axiay (a, b) and coronal (c) MRI images show T1 iso-hypointense, T2 heterogeneous, hyperintense nodular lesion posterior to the left lobe of the thyroid consistend with parathyroid adenoma (arrows)

of ascending and abdominal aortic aneurysms, the IIAA detected in our patient could not be explained by standard etiologies.

Aortic stiffness has become a significant independent risk factor in the determination of cardiovascular morbidity and mortality (6). Studies investigating the effect of PHPT on large arteries have reported a significant association between an elevated PTH level and aortic stiffness (7). A previous study evaluated 24 patients with mild PHPT and 48 control subjects individually matched with cases by age, sex, and blood pressure and found higher aortic peak wave velocity (PWV) values in patients with PHPT (11.4±2 vs. 9.6±2 m/s, $p<0.001$). The authors also noted that the aortic PWV values of patients with PHPT significantly decreased at 4 weeks after surgery (8). Another study compared 17 hypercalcemic and 16 normocalcemic patients with PHPT with 15 control subjects and found higher aortic PWV values in patients with PHPT compared with those in the control subjects (9). In the present study, it was revealed that the serum PTH level may be effective on arterial stiffness irrespective of serum calcium level. A recent study compared 30 healthy controls (group I), 34 patients with PHPT who had no surgical indications and were followed up with medical treatment (group II), and 31 patients with PHPT with surgical indications (group III) and reported that the aortic intima-media thickness values significantly increased in groups II and III compared with those in group I. Moreover, these values also significantly increased in group III compared with those in group II (10).

Taken together, the aforementioned notions suggest that an elevated PTH level is an independent risk factor particularly for the stiffness of large arteries and the severity of this vascular injury increases with the severity of the disease. On the other hand, we consider that the high PTH level in our patient (>1,000 pg/mL) could have further aggravated the vascular injury and, thus, led to aneurysm formation. Moreover, the absence of a history of smoking and hypertension and a family history of aneurysm in our patient implicates the presence of severe PHPT.

In conclusion, the case presented in this study demonstrated a relationship between an elevated PTH level and vascular injury. To our knowledge, there has been no report of a PHPT case accompanied by advanced-stage bilateral iliac artery aneurysm in the literature.

Informed Consent: Written informed consent was obtained from patient who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – SVK; Design – SVK; Supervision – SVK; Data Collection and/or Processing – YEĞ, SA, İN; Analysis and/or Interpretation – SVK; Literature Search – YEĞ, SVK; Writing – YEĞ, SVK; Critical Reviews – SVK, İN.

Conflict of Interest: The authors have no conflict of interest to declare.

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REFERENCES

1. Hedbäck G, Odén A. Increased risk of death from primary hyperparathyroidism--an update. Eur J Clin Invest 1998; 28(4): 271–6. [CrossRef]
2. Lundgren E, Lind L, Palmér M, Jakobsson S, Ljunghall S, Rastad J. Increased cardiovascular mortality and normalized serum calcium in patients with mild hypercalcemia followed up for 25 years. Surgery 2001; 130(6): 978–85. [CrossRef]
3. Brunkwall J, Hauksson H, Bengtsson H, Bergqvist D, Takolander R, Bergentz SE. Solitary aneurysms of the iliac arterial system: an estimate of their frequency of occurrence. J Vasc Surg 1989; 10(4): 381–4.
4. Levi N, Schroeder TV. Isolated iliac artery aneurysms. Eur J Vasc Endovasc Surg 1998; 16(4): 342–4. [CrossRef]
5. Kent KC, Zwolak RM, Egorova NN, Riles TS, Manganaro A, Moskowitz AJ, et al. Analysis of risk factors for abdominal aortic aneurysm in a cohort of more than 3 million individuals. J Vasc Surg 2010; 52(3): 539–48. [CrossRef]
6. Mitchell GF, Hwang SJ, Vasan RS, Larson MG, Pencina MJ, Hamburg NM, et al. Arterial stiffness and cardiovascular events: the Framingham Heart Study. Circulation 2010; 121(4): 505–11. [CrossRef]
7. Bitigen A, Tanalp AC, Kaynak E, KAravelioğlu Y, Kırma C, Adaş A, et al. Elastic properties of aorta in patients with primary hyperparathyroidism. Int J Clin Pract 2006; 60(12): 1572–5. [CrossRef]
8. Schillaci G, Pucci G, Pirro M, Monacelli M, Scarponi AM, Manfredelli MR, et al. Large-artery stiffness: a reversible marker of cardiovascular risk in primary hyperparathyroidism. Atherosclerosis 2011; 218(1): 96–101. [CrossRef]
9. Cansu GB, Yılmaz N, Özdem S, Balcı MK, Süleymanlar G, Arıcı C, et al. Parathyroidectomy in asymptomatic primary hyperparathyroidism reduces carotid intima-media thickness and arterial stiffness. Clin Endocrinol (Oxf) 2016; 84(1): 39–47. [CrossRef]
10. Sumbul HE, Koc AS. The abdominal aortic intima-media thickness increases in patients with primary hyperparathyroidism. Exp Clin Endocrinol Diabetes 2019; 127(6): 387–95. [CrossRef]