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Parathyroid Adenoma in a Patient with Left Hip Pain: A Case Report

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Authors' contributions

This work was carried out in collaboration between all authors. Author MK wrote the draft of the manuscript. Author MK managed the literature searches. Authors ZM and MRH designed the figures, managed literature searches and contributed to the correction of the draft. Authors ZM, MRH and BM provided the case, the figures and supervised the work. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Diagnosis of primary hyperparathyroidism (PHPT) is a challenging issue in some cases because of the uncommon and various presentations. Primary hyperparathyroidism is a relatively common condition, originating from parathyroid adenomas, with a prevalence of 25 per 100,000 people. In this paper we reported two cases of PHPT. First one was a 37-year-old female with a 3-month history of pain in the left hip and morning stiffness lasted for 1-2 hours. The second case was a 73-year-old woman with one month history of swelling and pain in the right wrist and fever, chills,

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erythema on the dorsum of hands, and limited range of motion in the right hip, from five days before admission; she had also experienced weight loss (3 kg) and bone pain over the past month. Diagnostic processes including laboratory, imaging, and clinical interventions were compared and reported in this paper.

Keywords: Hip pain; hyperparathyroidism; parathyroid adenoma.

1. INTRODUCTION

Primary hyperparathyroidism (PHPT) is a relatively common condition, originating from parathyroid adenomas, with a prevalence of 25 per 100,000 [1,2]. PHPT is usually diagnosed during the fifth or sixth decade of life [1-6]. Although this disease can be occurred at any age, the majority of cases are postmenopausal women. In fact, PHPT affects women three times more than men [7].

Manifestations of PHPT are usually non-specific such as renal stones, bone related symptoms, peptic ulcers, and hypertension. Other symptoms of this condition include weakness, nausea, vomiting, anorexia, cerebral symptoms, and reduced muscle tone [8].

In some studies, large adenomas have been reported which were weighing more than 3.5 grams [6] or 5 grams [2]. However, in some cases, parathyroid adenomas have been estimated to weigh more than 90 g [5]. Compared to the normal weight of the gland (0.05 g), these values are actually 100 times higher.

Hypercalcemia in PHPT results from the elevated level of parathyroid hormone (PTH) and activates osteoclasts [9]. Most cases of PHPT originate from adenomas (80-85%), while the majority of remaining cases are hyperplasia [10,11]. Parathyroid cancer is a rare cause of hyperparathyroidism [12].

2. CASE PRESENTATION

2.1 Case 1

A 37-year-old woman with a three-month history of left hip pain was referred to our clinic. Her pain worsened with activity. Although pain subsided with rest, its severity awake her at night. She also complained of prolonged morning stiffness (lasted for 1-2 hours) and weight loss (6 kg) over the past 3 months. And she was suffered from anorexia and polyuria. She did not mention fever, sweating or excessive thirst. She had a history of

cholecystectomy 3 years before. Sacroiliac joint tenderness was detected in physical examination. Hip examination and FABER test showed normal results. A palpable mass (3x4 cm) was found in the inferior part of neck. In laboratory analysis, Wright, 2ME, and HLA-B27 results were negative.

Various radiologic assessments were performed for this patient. Two lobulated, hypoechoic, heterogeneous masses with well-defined borders were confirmed in the left posterior-inferior pole of the thyroid lobe by ultrasound investigation. Its diameters were 50 mm×14 mm×18 mm, and no cervical adenopathy was detected, while abdominal ultrasound results were normal. These findings proposed parathyroid adenoma.

Parathyroid technetium-99m methoxy-isobutylisonitrile (99 mTc-MIBI) scan revealed a large parathyroid adenoma in the inferior thyroid lobe extended to the thorax. Left thyroid lobe and parathyroid mass pathologic assessment showed tumoral changes which were confined to the capsule (Fig. 1). The tumour was solid and trabecular, with a round, monomorphic nucleus, acidophilic cytoplasm in the stroma, and fine vessels. With these findings, parathyroid adenoma was confirmed. Pain and joint inflammation were subsided after treatment, she underwent surgery.

2.2 Case 2

A 73-year-old woman with one-month history of right wrist swelling and pain, along with movement limitations of the right hip, was referred to our clinic. Fever, chills, and erythema on the dorsum of the hands had started 5 days before admission. She had also experienced weight loss (3 kg) and bone pain over the past month. She had no positive findings in head and neck physical examination.

The only positive point in her family history was breast cancer in her sister. She had also undergone brain hemangioma and right knee surgeries 18 and 10 years ago, respectively.

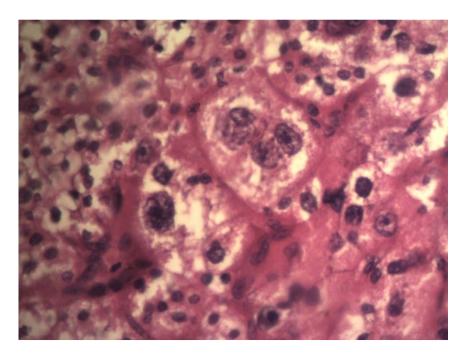


Fig. 1. Pathology findings of removed mass. Variations in nuclear size and shape (H&E, ×400); no vascular or capsular invasion was reported

The patient mentioned developing a urinary tract infection two months before admission. In physical examinations, swelling, warmth, erythema, and movement limitations were detected in the right wrist and second and third metacarpophalangeal (MCP) joints. Right FABER test was positive, and internal hip rotation was restricted. Laboratory results of both patients are summarized in Table 1.

A posterior nodule was detected in the thyroid ultrasound. Computed tomography (CT) of the lung, abdominal and pelvic ultrasounds, mammography, chest X-ray, and echocardiography showed normal results.

No fluid was detected in the right hip ultrasound; on the other hand, increased echogenicity and edema were reported in the wrist ultrasound. In bone mineral density (BMD) test, osteopenia in the right wrist bones and generalized osteoporosis (with a T-score <3.5) were reported. Moreover, Tc 99m-MIBI scan of parathyroid glands revealed a parathyroid adenoma. Symptoms relief was observed after surgical removal of adenoma.

3. DISCUSSION

PHPT is one of the most frequent endocrine disorders, affecting women more than men [1], most patients aged between 50 and 60 years [2].

It rarely manifested with major neuromuscular signs and symptoms. In this study we reported two cases in which muscular and joint inflammation were the predominant manifestations (Fig. 2). To best of our knowledge sacroiliitis is a rare musculoskeletal sign of hyperparathyroidism. One of our patients has a large adenoma (3x4 cm mass), while thyroid physical examination was normal in other case. Previous studies reported bone pain and muscle weakness in younger patients Pathological fractures also had been reported in some cases [14].

Our patients have non-specific symptoms of hyperparathyroidism, and combination of clinical and physical findings lead us to the accurate diagnosis. Facial swelling and pain in the left maxillary area are the other non-specific symptoms of PHPT [14]. It can be concluded that hyperparathyroidism in the context of parathyroid adenoma can present by a variety of manifestations. PTH level becomes normal right after surgery in both patients.

Both of our mentioned cases suffered from hypercalcemia. Confusion and renal disease and gastro-intestinal manifestations are associated with hypercalcemia. Diplopia or blurred vision, depression and renal stones might be occurred in these patients [15,13].

Table 1. Laboratory results

Case 2	Case 1	Case 2	Normal range
Alkaline phosphatase (Alp)	502	320	35-100 U/L
Phosphor (Ph)	1.4	2.3	2.5-4.5 mEq/L
C-reactive protein (CRP)	10	14	1-2 mg/L
Rheumatoid factor (RF)	-	Negative	<40 IŬ/mL
Wright, 2ME, and Cooms-Wright test results	-	Negative	-
PTH	1736	137	11-54 pg/ml
25-hydroxy vitamin D test	14	15	10-55 ng/ml
Calcium (Ča)	11.7 mg/dl	11.3	8.5-10 mg/dl





Fig. 2. Radiologic manifestations of PHPT in reported cases. A: Left sacroilitis with pseudowidening, sclerosis, and erosion. B: Bilateral anterior-posterior radiographic view of hands shows subperiosteal bone resorption along the radial aspects of middle phalanges

Neck examinations are very important in PHPT cases and revealed mild to severe enlargement of the thyroid gland or mass. The mass might be non-tender and mobile, with no thrills or bruits [13]. Although one of our patients had thyroid mass, neck examinations were near normal in the other one. So, radiologic and laboratory tests should be considered in cases like this.

Radiologic investigations might be helpful in PHPT diagnosis; most patients have a mottled appearance of the calvarium, presenting the characteristic salt-and-pepper pattern of hyperparathyroidism. Ultrasonography of the neck and parathyroid scan are the most helpful assessment for PHPT diagnosis. Although

imaging is useful to localize the PHPT, it is not sufficient to confirm the diagnosis [9].

Both cases had hip pain, which might have a wide range of deferential diagnosis, particularly in elderly. Femoroacetabular impingement, gluteal tendon injury, greater trochanteric bursitis, capsular laxity and avascular necrosis of the hip are some probable causes. Physical and radiologic examinations did not confirm any of the following diagnosis in our cases, on the other hand, their pain relief after treatment.

Considering the non-specific signs and symptoms of PHPT, it is suggested that serum Ca and P levels be evaluated in every patient

with suspicious clinical manifestations and medical history. In cases with elevated serum Calcium level and reduced Phosphor level, assessment of PTH level can help with the diagnosis. Serum Ca and PTH levels should both be evaluated to make an accurate diagnosis, since a patient with PHPT may have a normal blood Ca level due to a variety of factors such as vitamin D deficiency [14,15].

PHPT diagnosis should be considered in patients with persistent hypercalcemia and inappropriately normal or elevated PTH level. The normal feedback disturbs between PTH and extracellular calcium, so the set point changes in adenomas. In parathyroid hyperplasia, this feedback affects by the increase in cell numbers [10,11].

4. CONCLUSION

PHPT diagnosis might be complicated in elderly patients because of various manifestations and laboratory impairment (due to underlying diseases). So, clinical doubt of this condition and careful physical examination is crucial. Our case was a novel one due to rare manifestation (sacroiliitis) of PHPT.

CONSENT

All authors declare that 'written informed consent was obtained from both patient for publication of this case report and accompanying images.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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