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**OLGU SUNUMU / CASE REPORT**

# Delayed post-surgical hypoparathyroidism mimicking ankylosing spondylitis: a case report

## Ankilozan spondiliti taklit eden gecikmiş cerrahi sonrası hipoparatiroidizm: bir olgu sunumu

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**Abstract**

Hypoparathyroidism is a rare clinical condition characterized by parathyroid hormone insufficiency, decreased serum calcium, and increased serum phosphorus levels. Neck surgery is the most common cause of acquired hypoparathyroidism in adults. Post-surgical hypoparathyroidism is reported as one of the important complications in thyroid gland surgeries and if not diagnosed timely, may be associated with severe morbidity. The clinical manifestations of hypoparathyroidism are variable and may affect almost all organ systems. It is usually manifested by symptoms associated with hypocalcemia. The most common symptom is neuromuscular irritability, which includes paresthesias and muscle spasms in hands, feet, and around mouth. Rarely, some cases may present with inflammatory low back and neck pain, widespread pain, posture disorders, muscle weakness or, limited vertebral range of motion in the long term. Some patients may be misdiagnosed as spondyloarthropathy and may receive unnecessary treatments. In this case report, we present the clinical findings of a 55-year-old male patient with delayed post-surgical hypoparathyroidism who had been previously diagnosed ankylosing spondylitis and referred to our clinics for further treatment.

**Keywords:** Ankylosing spondylitis, hyperphosphatemia, hypocalcemia, hypoparathyroidism, spondylarthropathies

**Öz**

Hipoparatiroidizm, paratiroid hormonu yetersizliği, azalmış serum kalsiyum ve artmış serum fosfor seviyeleri ile karakterize nadir görülen bir klinik durumdur. Boyun cerrahisi erişkinlerde edinilmiş hipoparatiroidizmin en yaygın nedenidir. Ameliyat sonrası hipoparatiroidizm tiroid bezi ameliyatlarında önemli komplikasyonlardan biri olarak bildirilir ve zamanında teşhis edilmezse ciddi morbidite ile ilişkili olabilir. Hipoparatiroidizmin klinik belirtileri değişkendir ve hemen hemen tüm organ sistemlerini etkileyebilir. Genellikle hipokalsemi ile ilişkili semptomlarla kendini gösterir. En yaygın semptom, el, ayak ve ağız çevresindeki paresteziler ve kas spazmlarını içeren nöromüsküler irritabilitedir. Nadiren, bazı vakalarda uzun vadede inflamatuvar bel ve boyun ağrısı, yaygın ağrı, duruş bozuklukları, kas güçsüzlüğü veya vertebral hareket açıklığında kısıtlanma görülebilir. Bazı hastalara spondiloartropati tanısı konabilir ve gereksiz tedaviler uygulanabilir. Bu olgu sunumunda, daha önce ankilozan spondilit tanısı konan ve daha ileri tedavi için kliniklerimize sevk edilen, gecikmiş cerrahi sonrası hipoparatiroidizmi olan 55 yaşında bir erkek hastanın klinik bulguları sunulmuştur. .

**Anahtar kelimeler**: Ankilozan spondilit, hipoparatiroidi, hipokalsemi, hiperfosfatemi, spondiloartropati

## INTRODUCTION

Hypoparathyroidism (HypoPT) is a rare clinical condition characterized by parathyroid hormone (PTH) insufficiency, decreased serum calcium, and increased serum phosphorus levels. This endocrinological disease may occur due to inadequate hormone synthesis from the parathyroid glands or, more rarely, due to PTH receptor resistance[1]. While neck surgery is the most common cause of acquired HypoPT in adults, it may also develop due to

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autoimmune diseases, irradiation to the neck, parathyroid metastases, or heavy metal accumulation in the gland. Besides, HypoPT can be observed in severe deficiencies of magnesium, which is a co-factor for PTH secretion[1,2].

The clinical manifestations of HypoPT are variable and may affect almost all organ systems. It is usually manifested by symptoms associated with hypocalcemia. Although hypocalcemia is often asymptomatic, there may be various symptoms associated with the levels of blood calcium, duration of hypocalcemia, and the rate of the development of hypocalcemia[3]. The most common symptom is neuromuscular irritability, which includes paresthesias and muscle spasms in hands, feet, and around mouth. Besides, fatigue, anxiety, memory problems, depression, frontal lobe and basal ganglia calcifications, pseudotumor cerebri, Chvostek sign, and Trousseau sign may be seen[3,4]. Although hypocalcemia usually occurs immediately after surgery in the postoperative cases, progressive atrophy of the parathyroid glands may result in late clinical presentation. Rarely, some cases may present with inflammatory low back and neck pain, widespread pain, posture disorders, muscle weakness, or limited vertebral range of motion in the long term[5,6]. Radiological findings such as ossification of spinal ligaments, bridging osteophytes, ossification of iliolumbar, sacrospinous and sacrotuberous ligaments and ossification of acetabular margins may also be detected[7,8]. As a result, some patients may be misdiagnosed as spondyloarthropathy (SpA) and may receive unnecessary treatments. Few cases of idiopathic HypoPT mimicking ankylosing spondylitis (AS) or diffuse idiopathic skeletal hyperostosis (DISH) have been previously reported in the literature, but post-surgical delayed diagnoses are very rare[6,7,9-11].

In this case report, we aimed to present the clinical findings of a patient with delayed post-surgical HypoPT who had been previously diagnosed AS and referred to our clinics for further treatment and also to briefly discuss the current literature.

## CASE

A 55-year-old male foundry worker admitted to our outpatient clinic with chronic low back pain and morning stiffness. His past medical history revealed that he had been receiving indomethacin 50mg/day and sulfasalazine 1000mg/day with the diagnosis of AS for the last 3 years. He also stated that he had a thyroid surgery 30 years ago. He could not remember any more surgical details. There was no history of major trauma or lumbar operation. His family history was unremarkable. He did not report any rash, aphthous ulcer, diarrhea, abdominal pain, a recent infection, fever, or weight loss. Bath Ankylosing Spondylitis Disease Activity Index (BASDAI) score was 6.5 and Visual Analog Scale (VAS) score was 8/10.

On his physical examination, anteflexed posture with increased thoracic kyphosis and decreased lumbar lordosis was seen. Cervical and lumbar vertebral range of motions (ROM) were limited and painful on the sagittal and frontal planes (Figure 1). FABER and FADIR tests were positive bilaterally. Sacroiliac compression test was positive on the right, but Mennel test was negative on both sides. Fingertip-to-floor distance was 45 cm, occiput-to-wall distance was 24 cm, tragus-to-wall test was 24 cm, Modified Schober's Test was 1.5 cm, chest expansion was 2 cm and intermalleolar distance was 75 cm. Lateral flexion of the lumbar spine was 4 cm on the right and 3.5 cm on the left side. There was no sign of peripheral arthritis or enthesitis. Neurological exam was normal. The Trousseau and Chvostek's signs were both negative.

**Figure 1. Lateral view of the patient. Anteflexed posture with increased thoracic kyphosis and decreased lumbar flexion was seen.**

On laboratory examination; creatinine 1.41 mg/dL (0.7-1.3), erythrocyte sedimentation rate 21mm/hour, C-reactive protein (CRP) 1.56 mg/L (0-

5), calcium 7.4 mg/dL (8.5-10.5), phosphorus 5.1 mg/dL (2.3-4.7), albumin 45 g/L (35-50), parathyroid hormone 11 mg/dL (18.5-88), 25-OH vitamin D 42 µg/L (20-70), calcium in spot urine 10.8 mg/dL, and calcium in 24 hour urine 325 mg/day (100-300). Complete blood count, creatinine kinase, alkaline phosphatase and thyroid stimulating hormone (TSH) level were in normal ranges. HLA-B27, rheumatoid factor, antinuclear antibody (ANA) tests were negative.

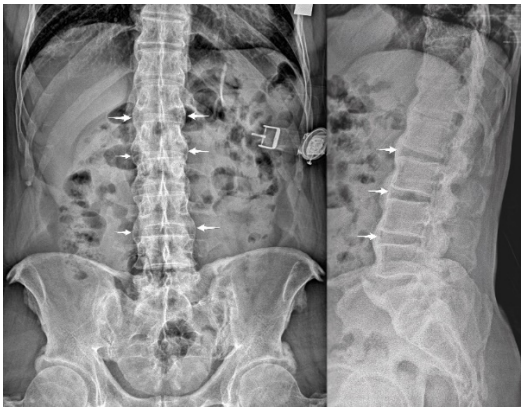

**Figure 2. Anteroposterior and lateral radiograph of the thoracolumbar spine shows syndesmophytes (white arrows).**

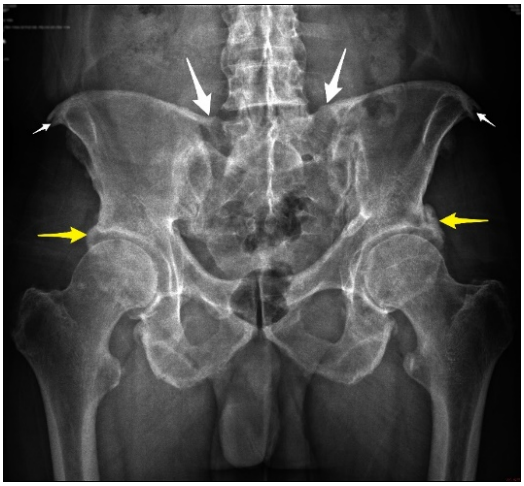

**Figure 3. Anteroposterior (AP) radiograph of the pelvis shows calcification of iliolumbar ligament (big white arrows), osseous proliferation, and irregular bony excrescences (small white arrows), calcification of the superior acetabular margins (yellow arrows).**

On radiological examination, anteroposterior and lateral cervical-thoracolumbar spinal x-ray series revealed cervical-thoracolumbar syndesmophytes, calcifications of interspinous ligaments and ankylosis of apophyseal joints. Vertebral spaces were also preserved and there was no excessive disk disease. Calcification of acetabular lateral margins were observed bilaterally on anteroposterior pelvic x-ray. There was mild periarticular sclerosis of sacroiliac joints, but the joint space was normal.Irregular bony excrescences were seen involving the right and left iliac bones and also involving ischial tuberosity and superior pubic ramus. Calcifications of interosseous membrane were examined on bilateral anteroposterior forearm radiographs (Figure 2-5). On magnetic resonance imaging of the sacroiliac joints, there was no evidence of active or chronic sacroiliitis.

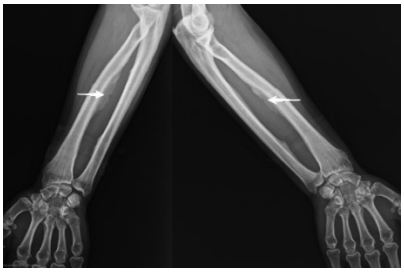

**Figure 4. Radiograph of the bilateral forearm showing interosseous membrane calcification (white arrows).**

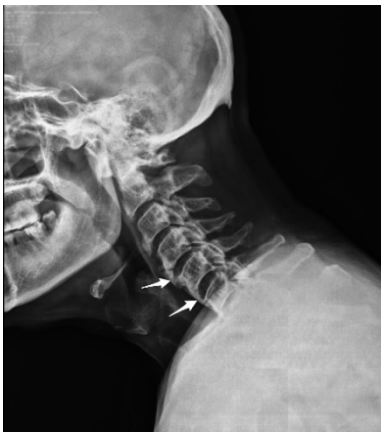

**Figure 5. Lateral radiograph of the cervical spine shows prominent and bridging osteophytes (white arrows) and calcification of interspinous ligaments.**

Patient was diagnosed as delayed post-surgical HypoPT mimicking AS. Patient's medications including indomethacin and sulphasalazine were stopped due to decreased renal function. Paracetamol 1000 mg/day was prescribed for analgesia. Patient was consulted with Department of Endocrinology for HypoPT and calcitriol 0.50 micrograms/day and

calcium carbonate 2000mg+vitamin D3 1760 IU/day treatments were prescribed. Physical therapy including hot pack, transcutaneous electrical nerve stimulation (TENS), and therapeutic ultrasound (US) were started and core stabilization exercises were given. After15 sessions physical therapy, VAS score was 4. Laboratory tests at the end of two weeks revealed improved renal function and serum calcium and phosphorus levels returned to normal ranges.

## DISCUSSION

Transient (absence of PTH/low PTH levels lasting <6 months) or permanent (HypoPT for >6 months) post-surgical HypoPT is reported as one of the important complications in thyroid gland surgeries and may be associated with severe morbidity[1,12]. Recently, a study revealed an almost 5-fold increased risk of renal insufficiency and a doubled risk of a diagnosis of any malignancy in patients with permanent HypoPT after thyroidectomy[12]. In cases of long-term untreated HypoPT, clinical signs of SpA may be observed and typical radiological findings may be detected. There are a few reports of SpA described in post-surgical HypoPT and detailed clinical information is lacking since the disease is rarely seen in endocrine practice[5].

Patients with HypoPT may present with musculoskeletal system symptoms like back and hip pain, stiffness, limited vertebral range of motion, posture problems, and myopathy[3,6]. Patients may often fulfill the classification criteria for AS[9]. Hypocalcemia is often asymptomatic and low serum albumin-corrected or ionized calcium levels and low PTH levels are detected incidentally on laboratory examination. HLAB27 positivity rate is not more common than the general population[5,7]. Our patient also had inflammatory low back pain without neurological deficits and laboratory findings were consistent with literature data. To the best of our knowledge, this is a very rare case presented with an AS clinic and diagnosed as HypoPT 30 years after thyroid surgery.

PTH increases renal and intestinal calcium absorption and renal phosphate excretion. Deficiency of PTH causes hyperphosphatemia due to increased renal tubular reabsorption of phosphate and excessive calcifications in soft tissues and bones may develop[1,5]. On radiological examination, frontal and basal ganglion calcifications, soft tissue-ligament calcifications, enthesopathies, vertebral syndesmophytes, calcifications on the acetabular edges of the hips, and increased sclerosis in sacroiliac joints without erosions may be seen[8,13]. These radiological findings are different from those seen in SpA and DISH. In SpA, usually, the sacroiliac joints are retained in the early period and erosions can be detected. While calcifications often develop in anterior spinous ligaments, posterior ligament calcifications may also be observed in patients with HypoPT. In DISH, commonly anterior spinal ligament calcification is detected while sacroiliac and facet joints are spared. Lower thoracic spine involvement is typical of DISH, but the lumbar and cervical spine can also be affected. The left side of the spine is typically spared or less involved, which is probably attributable to the pulsating aorta. Also, intramembranous calcification of the forearm is not an expected finding. Generally, there is an underlying disease such as diabetes and hypothyroidism[5,9,11]. Consistent with previous case reports, calcifications of vertebral interspinous and iliolumbar ligaments, forearm interosseous membranes and superior acetabular margins, bridging osteophytes, increased sclerosis in thoracolumbar vertebrae and bilateral sacroiliac joints were detected in conventional x-rays of our patient.

While many cases of idiopathic HypoPT have been described in the literature, postoperative cases are rare. Recently, Türkoğu et al. presented a case of iatrogenic HypoPT manifesting without hypocalcemia symptoms but accompanied by SpA clinic[11]. Zabihiyeganeh et al. reported a case of HypoPT that was misdiagnosed as refractory SpA and therefore, had been treated with anti-tumor necrosis factor (anti-TNF)[14]. Goswami et al. radiologically examined 40 patients with HypoPT and found signs suggestive of AS such as sacroiliitis, spondylitis, acetabular calcification in 14 patients[5]. Jiang et al. reported a case with systemic lupus erythematosus, HypoPT, and AS. This relationship has been reported in relation to autoimmunity and it has been concluded that HypoPT develops due to autoantibodies against calcium-sensing receptors[15]. In our case, we believe that post-surgical HypoPT, which develops due to the patient's irregular follow-up, causes proliferative spinal involvement misdiagnosed.

As a result, the diagnosis of patients with post-surgical HypoPT may be delayed and these patients may be treated with the diagnosis of AS and DISH. This situation causes a serious financial burden and

side effects profile. Calcium, phosphorus, alkaline phosphatase, PTH should be checked in patients with SpA, and HypoPT should be considered in the differential diagnosis, and the diagnosis should be revised in patients who do not respond to anti-inflammatory and immunosuppressive therapy or whose symptoms worsen.

**Yazar Katkıları:** Çalışma konsepti/Tasarımı: AA, İB, İŞ, AA, AT; Veri toplama: İB, AA; Veri analizi ve yorumlama: İB, AA; Yazı taslağı: :AA, İŞ, AT; İçeriğin eleştirel incelenmesi: AA, İB, İŞ, AA, AT; Son onay ve sorumluluk: AA, İB, İŞ, AA, AT; Teknik ve malzeme desteği: AA, İB, İŞ, AA, AT; Süpervizyon: AA, İB, İŞ, AA, AT; Fon sağlama (mevcut ise): yok.
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**Author Contributions:** Concept/Design : AA, İB, İŞ, AA, AT; Data acquisition: İB, AA; Data analysis and interpretation: İB, AA; Drafting manuscript: AA, İŞ, AT; Critical revision of manuscript: AA, İB, İŞ, AA, AT; Final approval and accountability: AA, İB, İŞ, AA, AT; Technical or material support: AA, İB, İŞ, AA, AT; Supervision: AA, İB, İŞ, AA, AT; Securing funding (if available): n/a.
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