Idiopathic hypoparathyroidism mimicking ankylosing spondylitis: a case report

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ABSTRACT

Idiopathic hypoparathyroidism, due to an inadequate secretion of parathyroid hormone of unknown etiology, may mimic ankylosing spondylitis both clinically and radiologically. Spinal complaints may be the first sign of this endocrinological disorder.

Keywords: Hypoparathyroidism; Ankylosing spondylitis.

INTRODUCTION

Inherited or acquired, idiopathic hypoparathyroidism (IH) is the absence or atrophy of parathyroid glands of unknown etiology that results in inadequate secretion of parathyroid hormone (PTH), which in turn produces hypocalcemia and hyperphosphatemia[1,2]. Skeletal manifestations of IH are caused by ligaments calcification, which give rise to morning stiffness, spinal pain and change in posture, resembling ankylosing spondylitis (AS)[2-4]. We report a case of undiagnosed IH, who was referred to us for a rehabilitation programme for the spinal pain and postural changes, with the diagnosis of AS.

CASE REPORT

A 58-year-old female patient, previously diagnosed as AS by the family physician, was referred to our clinic for the spinal pain unresponsive to nonsteroidal anti-inflammatory drugs (NSAID) and postural rehabilitation. She had been suffering from neck, thoracic spine and low back pain for 35 years. Pain was in both mechanical and inflammatory in character, occurring at day and night, worsening with movement but not completely resolving by rest. She also had morning stiffness lasting at most 30 minutes. She complained about lassitude, fatigue, headache and anorexia for the last year. Her past medical history revealed she had been suffering for 20 years of idiopathic hypertension and type 2 diabetes mellitus, had a coronary by-pass 6 years before and bilateral cataracts with surgery on the left eye. She was using a hearing device due to hearing loss on the left ear. She denied any paresthesias, muscle cramps or previous episodes of convulsion or confusion.

The physical examination of the patient revealed typical AS posture due to limitation of spinal range of motion, with increased thoracic kyphosis (Figure1). The neck was forwardly projected and completely ankylosed, shoulders were limited in all directions: fle-

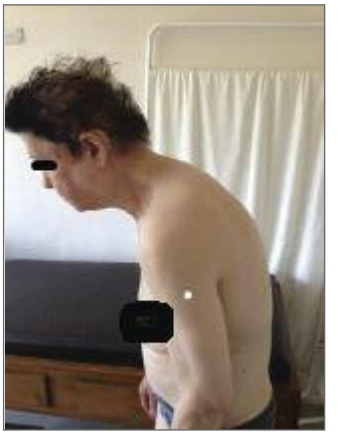

FIGURE 1. Lateral view of the patient

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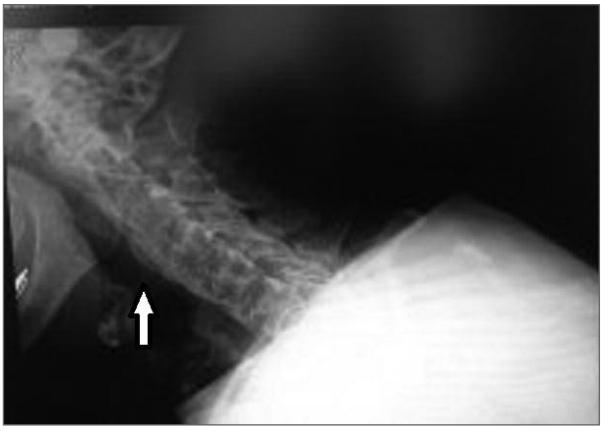

FIGURE 2. Roentgenogram of the cervical spine showing ligament calcifications

xion 80º (0-180), abduction 60º (0-180), external rotation 50º (0-90) and internal rotation 40º (0-70) bilaterally. Hips and lumbar spine were painful on active and passive mobilization and limited in all planes. She had no alopecia, only dry skin without nail or dental changes. Neurological examination was normal with normoactive deep tendon reflex. The lumbar Schober test was 0 cm, the lumbar lateral flexion was 3cm and the finger - floor distance was 27cm, jaw-manubrium distance of 9.5 cm, tragus-wall distance of 23 cm and chest expansion of 1.5 cm. No peripheral arthritis was detected.

Laboratory analysis parameters were as follows: calcium: 5.3 mg/dl (8.4-10.2 mg/dl), phosphorus: 6.3 mg/dl (2.5-4.6 mg/dl), albumin: 3.7g/dl (3.5-5.5 g/dl), PTH: 4.7 pg/ml (22.2-108.5 pg/ml), vitamin D: 27.2 ng/ml (>20), c-reactive protein: 0.4 mg/dl (0-0.5 mg/dl), erythyrocyte sedimentation rate:42 mm/h, hemoglobin:11.8 gr/dl (12-18 g/dl). She had mild iron deficiency anemia with low mean corpuscular volume and ferritin values. HLA-B27 test was negative.

Radiologic evaluation revealed increased cervical lordosis, calcification of the posterior longitudinal ligament and enthesopatic changes at posterior elements of cervical spine (Figure 2), syndesmophyte formations at the lumbar spine producing bamboo spine image (Figure 3) and periarticular calcifications at the shoulder and hip joints bilaterally.

Calcium and vitamin D replacement therapy was initiated as a treatment regimen of calcitriol 1mcg/day and calcium carbonate 2g/day. Pain was significantly reduced (VAS decreased from 9 to 5) with no change in the posture at the second month of follow-up.

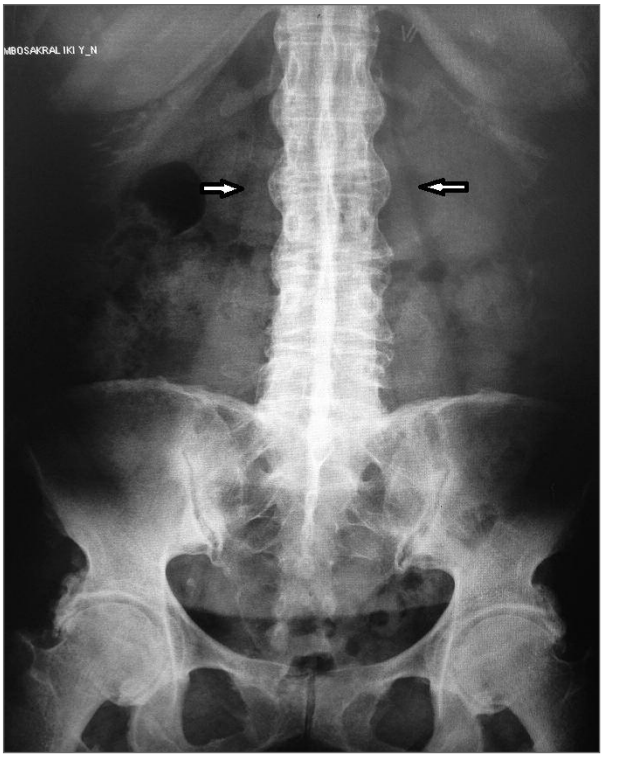

FIGURE 3. Roentgenogram of lumbar spine showing bamboo spine features

DISCUSSION

We diagnosed the patient with IH based on the hypocalcemia, hyperphosphatemia and low serum levels of PTH. As no genetical analysis was performed it was impossible to exclude rare causes (e.g DiGeorge Syndrome, Kenny-Caffey syndrome) of IH, however our diagnosis is supported by no previous history of parathyroid surgery, radiation exposure or familial autoimmune polyendocrinopathies.

Spinal pain, morning stiffness, postural changes and radiological findings such as thoracic and/or lumbar spinal syndesmophytes, mild sacroiliitis, enthesopathy of hips and shoulders may lead the clinician to misdiagnose IH as AS[4-6]. This patient was referred us for the rehabilitation of her postural deformity and related complaints, which were typical for, and previously interpreted, as AS. Sacroiliitis, the most common and even the first sign of AS[3,8], is very infrequent or mild in IH[8]. In our case, despite the presence of severe spinal involvement, sacroiliac joint space was unaffected, except for mild sclerosis. There was sclerosis on both sacral and iliac margins of the sacroiliac joint

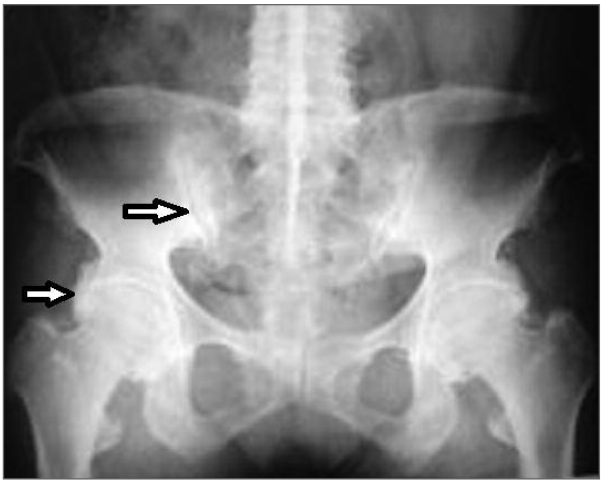

FIGURE 4. Roentgenogram of the pelvis showing sclerosis of the sacroiliac joints and ischiatic enthesopathy

however the joint space was preserved and regular (Figure 4). Since the patient's posture is unfavorable to MRI, it was not performed.

Korkmaz *et al.* reported the case of a 45-year-old male who was misdiagnosed as AS due to typical clinical and radiological findings. Persistent pain despite 2 months of treatment with sulfasalazine, methotrexate and indomethacin lead to further evaluation of the patient. Detailed laboratory investigations revealed hypocalcemia, hyperphosphatemia and low serum levels of PTH with the eventual diagnosis of IH. They reported an important decrease in pain levels after 2 months of treatment with calcitriol and calcium carbonate[7].

A study by Goswami *et al.* stated that 3 of 40 patients with hypoparathyroidism had clinical and radiological findings suggestive of AS. Preserved bone mineral density, negative HLA-B27 test and equal distribution among both sexes which may be helpful to distinguish IH from AS[8]. The mechanism underlying these skeletal changes in IH is not well defined. Paravertebral ligamentous ossification is shown to be related with decreased intestinal calcium absorption via 1,25 dihydroxyvitamin D^2. Hypocalcemia, the ultimate result of IH may be the causative factor in the skeletal and ligamentous changes resembling AS.

Tetany, the acute manifestation of hypocalcemia in IH was not present in our case. Chvostek's and Trousseau's signs were negative, due to the chronicity of the hipocalcemia. She had bilateral cataracts due to discrete layers of opacity in the peripheral cortex of the lens which is the typical lesion of hypoparathyroidism.

Early onset of symptoms (approximately 25 years of age), typical posture, spinal deformity and vertical syndesmophyte formations at the thoracolumbar region are AS-like features in our case. Despite the severe spinal involvement the lack of typical changes at the SI joint is striking and makes AS less likely. Laboratory findings indicating the diagnosis of IH are hypocalcemia, hyperphosphatemia and a low serum level of PTH. The negative acute phase reactants and HLA-B27 are the other features in our case that make the diagnosis of AS less likely.

Additionally, periarticular calcifications at the hips and shoulders, cataract formation - a long-term complication of hypoparathyroidism - and pain unresponsive to NSAIDs, all support the diagnosis of IH.

Nakamura reported the prevalence of IH as 7.2 (5.5--8.8) per million persons in Japan[11]. IH is also rare in Turkey but the exact prevalence is not known. Despite its rare occurance, hypoparathyroidism may be the underlying pathology in patients with spinal complaints. Evaluation of serum calcium and phosphate levels may clarify the diagnosis and prevent further disease progression since the management of IH and AS differ significantly.

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