The Association of Acromegaly and Ankylosing Spondylitis

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SUMMARY

Coexistence of acromegaly and anklosing spondylitis had been rarely reported. Only two case reports were described coexistence of two diseases in literature. These two diseases have some similar clinical and radiographic features. Calcaneal epin formation, enteshopathy and caude equina syndrome can be seen both acromegaly and ankylosing spondylitis. Our case had clinical and radiological features of both acromegaly and ankylosing spondylitis. Acral enlargement, coarsening of feature, malocclusion, non-suppressed growth hormone levels with oral glucose tolerance test and evidence of pituitary adenoma were support diagnose of acromegaly. Morning stiffness, positive Schober and Moll test, elevated erythrocyte sedimentation rate and grade 4 sacroiletis of the patient lead us to diagnose ankylosing spondylitis at the same time. In this case report, we aim to discuss interesting coexistence of two disease

Key words: *acromegaly, ankylosing spondylitis.*

Ankilozan ve Akromegali Birlikteliği

ÖZET

Akromegali ve ankilozan spondilitin nadiren rapor edilmiştir. Bu iki hastalığın birlikteliği literatürde iki olgu sunumu ile gösterilmiştir. Akromegali ve ankilozan spondilit benzer klinik ve radiografik bulgulara sahiptir. Kalkaneal epin varlığı, entezopati, cauda equina sendromu gibi bulgular hem akromegalide hemde ankilozan spondilit de gözlenebilir. Bu olgu hem ankilozan spondilitin hemde akromegalinin klinik ve radyolojik bulgularına sahipti. Uçlarda büyüme, yüz yapısının bozulması, malokluzyon, glikoz tolerans testinden sonra baskılanmayan büyüme hormon düzeyinin olmasına ek olarak da bir hipofizer adenom varlığı akromegali tanısını destekleyen bulgulardı. Sabah tutukluğu, Schober ve Moll testinin pozitif olması, sedimentasyon yüksekliği ile birlikte dördüncü dereceden sakroileitin varlığı hastada akromegaliye ek olarak ankilozan spondilitin de varlığını göstermekteydi. Bu olgu sunumunda ankilozan spondilit ve akromegali tanısı konulan bir hasta tartışılmıştır.

Anahtar kelimeler: *akromegali, ankilozan spondilit*

INTRODUCTION

Acromegaly is a rare disabling disorder that resulting in premature death. It caused by growth hormone hypersecretion. The definitive test for the diagnosis of acromegaly is failure of serum growth hormone levels to decrease to less than 2μg/L after oral glucose test. IGF-1 levels are increased in acromegalic patients. Magnetic resonance imaging of pituitary can be used for definition of tumour mass. Ankylosing spondylitis is considered the

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prototype of the spondyloarthropathies. Ankylosing spondylitis primarily affects the axial skeleton; peripheral joints and extraarticular structures.The initial symptom is usually dull pain insidious in onset, felt deep in the lower lumbar or gluteal region, accompanied by low-back morning stiffness of up to a few hours.

Early in the course of the acromegaly, joint spaces are increased secondary to cartilage proliferation. Synovial and periarticular swelling produces joint swelling without effusion. Weight bearing and proliferating cartilage in joint lead to ulceration and osteoarthritis. Ankylosing spondiliytis is a chronic inflammatory rheumatoid disease and characterised by inflammation of sacroiliac joints, the enthuses, and the spine (1) Spinal mobility is normal or increased in acromegaly but, decreased in ankylosing spondylitis both frontal and sagital plane (2). Joint lesion of ankylosing spondylitis and acromegaly are similar, but coexisting of the two diseases is very rare in the literature.

CASE

The patient is a 38 years old man; he has been suffering headache, neck pain, low back pain, which was increasing with inactivity and decreasing with activity, and morning stiffness lasting at least an hour, since 1993. In physical examination**:** the patient cervical mobility was restricted; occiput-wall and jaw-sternum distances were 4.5 and 4 cm, respectively. Chest expansion distance was 1 cm. Schober test and Moll lateral flexion were measured as 1cm and 0.7 cm, respectively. Sacroiliac compression test and Mennel test were bilaterally positive, and Fabere test was bilaterally restricted**.**

Laboratory evaluation: Erythrocyte sedimentation rate was 57mm/h and C-reactive protein was 43u/l(Normal range 0-6 u/l). HLA-B27 was found to be positive Prolactin levels were 53ng/ml (normal range: 4.1-18.4 ng/ml), twenty-four hour GH levels did not reach below 6µg/L. IGF-1 level was 783 ng /ml (N 130-354 ng /ml). Bone density measurements of the lumbar spine was normal range by dual energy x-ray absorptiometry (T score was 0.4 and Z score was 0.2) other biochemical findings were normal.

Oral glucose tolerance test: 0. 30^{th}, 60^{th}, 90^{th}, 120^{th} minutes glucose were 90mg/dl, 155mg/dl, 120mg/dl, 117mg/dl, 113mg/dl, respectively, and growth hormone levels measured at the same time were 12.8µg/L, 6.98µg/L 10.5µg/L,10.7µg/L, 10.0 µg/L, respectively

Radiographic findings: Arrowhead finding, grade 4 sacroileitis were detected in plain films (figure-1).

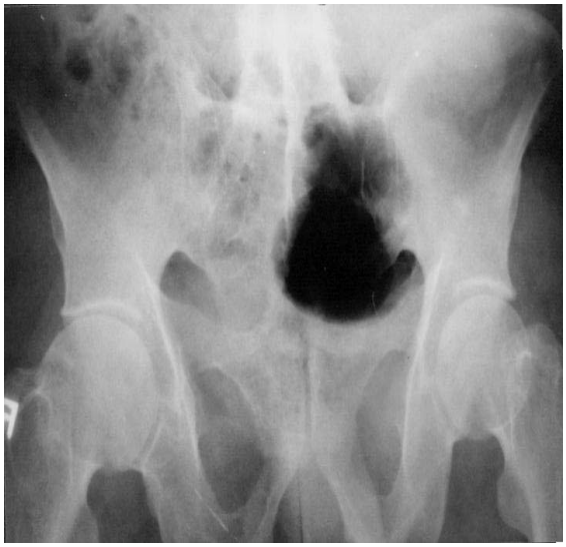

Figure-1: Grade 4 sacroileitis of the patient were detected on sacroiliac plain films

There was ‘bamboo spine’ imagine on anterior-posterior lumbosacral graphy (figure-2).

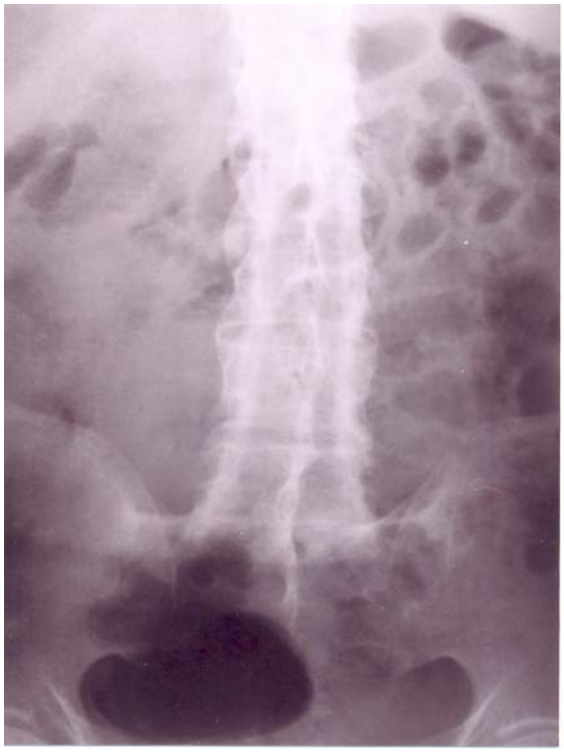

Figure-2: Bamboo spine imagine on anterior-posterior lumbosacral graphy

Calceneal epin and enthesopathy of achille tendon was confirmed on lateral foot films. Hell pad distance was 28mm. Ossification of anterior and posterior longitudinal ligaments in lateral cervical radiography. Evidence of a microadenoma of pituitary was seen by magnetic resonance imaging after godolinium –DTPA injection. (Figure-3)

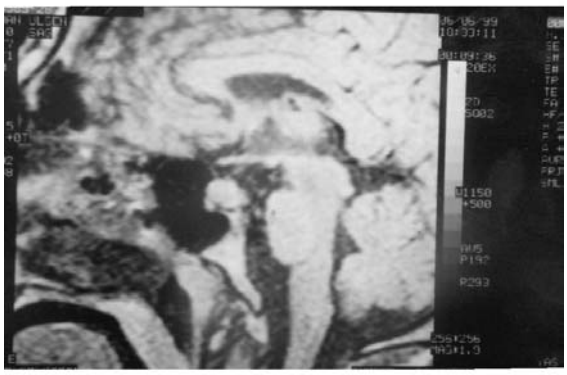

Figure-3: Magnetic resonance imaging of pituitary microadenoma

DISCUSSION

Ankylosing spondylitis and acromegaly have some similar radiographic findings however, these two diseases have different etiology. Coexistence of acromegaly and ankylosing spondylitis is interesting. Clinical features of our patient such as acral enlargement, malocclusion and headache lead us to think acromegaly. The patient's GH levels did not decrease below 2 μg/L after the glucose tolerance test. This finding was diagnostic for acromegaly. Evidence of micro adenoma by magnetic resonance imaging of pituitary supported our diagnosis of acromegaly.

Ankylosing spondylitis is a chronic systemic disease of unknown etiology. Low back pain, morning stiffness and loss of spinal mobility are main complaints of the disease (3). According to modified New York criteria, presence of radiographic sacroileitis plus one of the other criteria is accepted to be sufficient for diagnosis of definite ankylosing spondylitis (4). Our patient had bilateral grade-4 sacroileitis on suprapubic pelvis plain films. Spinal arthritis usually develops after the presence of radiographic sacroileitis. This patient had sclerosis and ankylosing of symmetric facet articulation, ligament ossification and syndesmophytes on cervical, thoracal and lumbosacral graphies. These radiographic findings are also supported the diagnosis of ankylosing spondylitis. European Spondyloarthropathy Study Group has accepted Enthesopathy as a criterion of ankylosing spondylitis since 1991. Sensitivity and speciality of enthesopathy was determined to be 56%, 76%, respectively (5). Enthesopathy is also one of the radiographic findings of acromegalic joint disease (4). Podgorski (6) have reported spinal and peripheral abnormalities in 47% and 74% of acromegalic patients, respectively. Chronically elevated growth hormon in acromegaly leads to increased IGF-1 production within the joints and articular tissues. Then, collogenous overgrowth occurs, followed by misalignment and destabilisation of joint architecture, subsequently, joint spaces widen and osteoarthritis occurs as the affected joint

gradually becomes damaged. In acromegalic patients GH and IGF-1 stimulate collagen production within tendon, causing laxity and leading to further joint destabilization. Excessive GH and IGF-1 stimulate osteoblast activity lead to osteophytes formation and osteoarthritis, calcification of tendons and ligaments insertion (7).

Our patient showed limitation of motion of the lumbar spine in both sagittal and frontal plane. Syndesmophytes formation and ossification of ligament of spine are responsible for limitation of motion of the spine. In the lumbar spine, progression of the disease leads to straightening caused by loss of lordosis and reactive sclerosis caused by osteoitis of the anterior corners of the vertebral bodies with subsequent erosion, thereby leading to 'squaring' of the vertebral bodies(8) Squaring of vertebral bodies was detected on the thoracal and lumbosacral graphies of our patient. Costovertebral, costosternal, manibrosternal, sternoclavicular enthesopathy areas and ankylosing of the joints cause chest pain and limiting of chest expansion. Chest expansion of our patients was severely limited and he had also chest pain. Encondral bone formation leads to enlargement of costocondral joint, and then, acromegalic rosary occurs. These findings also cause chest pain, but our patient had no acromegalic rosary (6).

Calcaneal epin can be seen either in acromegalic or ankylosing spondylitis by radiographic evaluation of feet. (4). Our patient have been suffering from ankylosing spondylitis for 9 years, therefore, we could suggest that calcaneal epin of our patient was due to ankylosinig spondylitis. Morning stiffness is a significant diagnostic criterion of ankylosinig spondylitis, although it is not observed in acromegalic patients (4). Cauda equina syndrome is one of the neurologic complications of either acromegaly or ankylosing spondylitis. Our patients had no cauda equina syndrome, but we will follow him about this finding.

Ankylosing spondylitis shows a striking correlation with the histocompatibility antigen HLA-B27 (9). The frequency of HLA-antigens in acromegalic patients is not clear, but, a study showed that the frequency of HLA-DRW9 was found slightly higher in acromegalic patients than the controls (10). We did not evaluate HLA-DRW9 because of technical failure, but, the patient was HLA-B27 positive which was found higher positivity in ankylosing spondylitis. Coexistence of acromegaly and ankylosinig spondylitis is rare. Prier N revealed in 1983 a patient coexisting of ankylosinig spondylitis and acromegaly with cauda equina syndrome(11). Ionescu N reported an acromegalic patient with ankylosing spondylitis in 1968(12). Owing to the low incidence of acromegaly in the general population, coexistence of two diseases may unlikely to be fortuitous.

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