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Case report
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THE IMPORTANCE OF EARLY DIAGNOSIS AND TREATMENT IN PATIENTS WITH ANKYLOSING SPONDYLITIS – CASE REPORT

ZNAČAJ RANOG OTKRIVANJA I LEČENJA BOLESNIKA SA ANKILOZIRAJUĆIM SPONDILITISOM − PRIKAZ SLUČAJA

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Summary

Introduction. Ankylosing spondylitis is a common, chronic musculoskeletal condition associated with substantial functional limitations. Inflammation in later phases of the disease may lead to fibrosis and calcification of the spine, causing poor quality of life. This case report emphasizes the importance of early diagnosis of ankylosing spondylitis as one of the major factors in further course of the disease. **Case report.** A 27-year-old man was diagnosed with ankylosing spondylitis in 2019. He was first time examined in 2005 because of joint pain which was then characterized as growing pains. Symptoms of joint pain reappeared approximately 10 years later, and were present in lumbar and thoracic spine accompanied by morning stiffness. New pain spots also appeared, as well as positive laboratory results regarding inflammation. Despite the lasting diarrhea, additional tests such as anti-smooth muscle and anti-mitochondrial antibody, Hepatitis B surface Antigen and anti-Hepatitis C virus antibody test as well as stool test results all turned out to be negative. The magnetic resonance imaging showed edema of lower edges of the lumbosacral corpus, most likely as part of spondylitis, changes to the sacroiliac joints in terms of chronic phase of sacroiliitis with discrete activity, and right shoulder active synovitis. The human lymphocyte antigen B27 testing showed positive results. Despite the prescribed medical therapy, the disease activity remained high with positive clinical presentation. This patient may be a candidate for biological therapy. **Conclusion.** Early diagnosis and effective treatment provide reduction of pain, fatigue and disease activity and also prevent functional limitations, therefore improving the quality of life.

Key words: Spondylitis, Ankylosing; Early Diagnosis; Risk Factors; Treatment Outcome; Pain; Quality of Life

Sažetak

Uvod. Ankilozirajući spondilitis predstavlja hronično muskuloskeletno oboljenje praćeno postepenom funkcionalnom onesposobljenošću. U kasnijim fazama inflamacija u sklopu ovog oboljenja može dovesti do fibroze i kalcifikacije kičme sa posledičnim smanjenjem kvaliteta života. Ovaj prikaz slučaja naglašava značaj ranog dijagnostikovanja ankilozirajućeg spondilitisa kao jednog od bitnih faktora u daljem toku same bolesti. **Prikaz slučaja.** Dvadesetsedmogodišnjem bolesniku je ankilozirajući spondilitis dijagnostikovan 2019. godine. Bolesnik je prvi put pregledan zbog bolova u zglobovima 2005. godine, kada je bol povezan sa rastom i razvojem. Međutim nakon deset godina javili su se bolovi u području slabinskog i grudnog dela kičme praćeni jutarnjom ukočenošću. Takođe, javila su se i nova bolna mesta uz pozitivne laboratorijske nalaze u pravcu inflamacije. Uprkos perzistentnim dijarejama dodatna ispitivanja koprokulture, antiglatkomišićna i antimitohondrijalna antitela kao i antihepatitis B površni antigen i antihepatitis C virus antitela su pokazala negativan rezultat. Magnetnorezonantnom tehnikom prikazan je otok donjih delova tela slabinsko-trtičnih pršljenova najpre u sklopu spondilitisa, promene u sakroilijačnom zglobu u pravcu hroničnog sakroiliitisa sa diskretnom aktivnosti kao i sinovitis desnog ramena. Nalaz humanog leukocitnog antigena B27 pokazao je pozitivan rezultat. Uprkos prepisanoj medikamentnoj terapiji registruje se visoka aktivnost bolesti praćena pozitivnim kliničkim nalazom. Prikazani bolesnik je potencijalno kandidat za biološku terapiju. **Zaključak.** Rano postavljena dijagnoza ankilozirajućeg spondilitisa kao i efektivna terapija omogućavaju smanjenje intenziteta bola, napora, aktivnosti bolesti i preveniraju funkcionalna ograničenja, a time poboljšavaju kvalitet života pacijenata obolelih od ankilozirajućeg spondilitisa.

Ključne reči: ankilozirajući spondilitis; rana dijagnoza; faktori rizika; ishod lečenja; bol; kvalitet života

Introduction

Ankylosing spondylitis (AS) is an autoimmune disease that mainly involves spine joints, sacroiliac joints (SIJs) and their adjacent soft tissues, such as tendons and ligaments [1].

In more advanced cases, it can cause immobile position due to fibrosis and calcification. AS primarily manifests with back pain, spinal rigidity, and inflammation in the joints, fingers or toes. However, extra-articular manifestations such as acute anterior uveitis and inflammatory bowel disease (IBD) can

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Abbreviations

AS – ankylosing spondylitis
SIJs – sacroiliac joints
IBD – inflammatory bowel disease
HLA B27 – human lymphocyte antigen B27
NSAIDs – anti-inflammatory drugs
MRI – magnetic resonance imaging
BASMI – Bath Ankylosing Spondylitis Metrology Index
BASDAI – Bath Ankylosing Spondylitis Disease Activity Index
BASFI – Bath Ankylosing Spondylitis Functional Index
ASDAS – Ankylosing Spondylitis Disease Activity Score
CD – Crohn's disease
SpA – axial spondyloarthritis

also occur [2]. Crohn's disease (CD) and ulcerative colitis occur in 5-10% of patients with AS due to genetic, clinical, immunological, and microbial connections between AS and CD [3, 4].

The prevalence of AS is generally believed to be between 0.1% and 1.4% globally [5].

It is important to note that the human lymphocyte antigen B27 (HLA B27) gene is present in approximately 90% of individuals with Ankylosing Spondylitis (AS), while it is found in less than 8% of the general population. However, the exact mechanism by which this gene contributes to the development of AS remains uncertain [6]. HLA B27 also plays a crucial role in the diagnosis, classification, and severity of axial spondyloarthritis (SpA) [7].

This case report emphasizes the importance of early diagnosis of ankylosing spondylitis as one of the major factors in further course of the disease.

Case report

A 27-year-old man, was diagnosed with ankylosing spondylitis in 2019. The patient did not have any underlaying comorbidities. He was first examined in 2005 because of joint pain that was then characterized as growth pains. The symptoms of joint pain reoccurred approximately 10 years later and were present in lumbar and thoracic spine accompanied by morning stiffness. He was treated by a physiatrist only with nonsteroidal anti-inflammatory drugs (NSAIDs) with transient improving effect. Looking at the chronicity of his problem, especially with minimal improvement and new pain spots, he was suggested to an X-ray of the right shoulder in regard to the newly onset symptoms. Meanwhile he had episodes of one-month lasting diarrheas without increased body temperature. Laboratory findings showed a confirmed inflammatory syndrome (sedimentation-SE 90 mm/h and C reactive protein - CRP 150 mg/l) followed by the increased level of transaminases. He underwent additional testing such as anti-smooth muscle antibody (ASMA) and anti-mitochondrial antibody (AMA), Hepatitis B surface Antigen (HBsAg) and anti-hepatitis C virus antibody (HCV), as well as a stool test, which all turned out to be negative. As the right shoulder X-ray showed deformity of the humerus head and reverse cervical lordosis, he was referred for further testing. The magnetic resonance imaging (MRI) showed edema of lower edges of the lumbosacral corpus, most likely as part of spondylitis, changes in the sacroiliac joints (SIJs) in terms of chronic phase of sacroiliitis with discrete activity and right shoulder active synovitis. HLA testing showed HLAB27 positivity. The patient was diagnosed with ankylosing spondylitis and started sulfasalazine and methylprednisolone therapy. Methylprednisolone therapy was soon discontinued. Furthermore, the patient was eligible for biological therapy, but was rejected as his condition got complicated by a urogenital infection. Since his condition did not improve (Bath Ankylosing Spondylitis Metrology Index- BASMI 6.8, Bath Ankylosing Spondylitis Disease Activity Index – BASDAI 6.1, Bath Ankylosing Spondylitis Functional Index – BASFI 5.8, Ankylosing Spondylitis Disease Activity Score – ASDAS 5.06) the sulfasalazine doses accompanied with NSAIDs were increased and he was advised to do a new MRI scan of spine and shoulder in the next three months. His latest results of the MRI scans showed the already seen signs of spondylitis and synovitis followed with high disease activity (BASDAI 4.95, BASFI 7.0, ASDAS 5.48) and positive inflammatory results. Bone density scan (DEXA) was referent. Clinical presentation showed positive Mennell's sign, thoracic kyphosis, Schober test 1 cm, tragus to wall test 20 cm, Thomayer distance 40 cm, respiratory index 4 cm, lateral flexion 4 cm and cervical rotation 10 cm. Finally, after the urogenital infection healed, he was approved to take the biological drugs.

Discussion

The prognosis of patients with AS varies depending on the presence of extraspinal manifestations (i.e. uveitis, psoriasis, and IBD), age at diagnosis and the treatment received [8, 9]. AS usually initially appears during the third decade of life, and rarely after the age of 45 [5]. However, diagnosis of AS is often delayed. Since AS is a rare condition, both medical professionals and patients are less familiar with it compared to the more common back disorders [10–12]. Compared to other rheumatic diseases, patients usually experience a delay between symptoms and diagnosis [13]. European data indicate an average time from symptom onset to diagnosis of between 8.5 and 11 years [14, 15]. A key reason for the delay in AS diagnosis is the requirement for radiographic evidence of sacroiliitis [16]. Radiographic changes develop slowly, with only 70% of AS patients fulfilling these diagnostic criteria after 5 years of symptoms [17]. Even though it has been reported that onset before the age of 10 or after the age of 45 is rare, approximately 15% of patients have onset of their disease in childhood (before the age of 16), but this percentage may be as high as 40% in some developing countries [18, 19].

Various studies conducted in China, Taiwan, and India have revealed that juvenile-onset AS patients are more prone to experiencing hip involvement than those with adult-onset AS. Due to substantial individual differences in clinical manifestations and the developmental stage of sacroiliac joints in children,

imaging diagnoses are limited, potentially leading to delayed diagnosis. Within 10 to 15 years, approximately 40% of juvenile spondyloarthropathy patients progress to functional disability, and hip involvement has been closely linked to a poor prognosis [20]. Considering all these facts, there is a need to develop a distinct strategy for children and adolescents with SpA.

Early diagnosis of spondylitis of any cause can significantly affect the patients' quality of life, but also reduce treatment costs before any accompanying complications occur [21].

Conclusion

Early diagnosis and effective treatment provide reduction of pain, fatigue and disease activity and also prevent functional limitations, therefore improving the quality of life.

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