

Cervical spine involvement as initial manifestation of rheumatoid arthritis: a case report

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ABSTRACT

Rheumatoid arthritis' synovitis affects mostly small hand and feet joints, although it may compromise any joint with a synovial lining. Cervical involvement occurs usually in longstanding disease in over half of these patients. We report the case of a 35-year old male patient who was referred to our outpatient clinic for a 2-year severe and disabling inflammatory neck pain, with incomplete response to intramuscular non-steroidal anti-inflammatory drugs and unremarkable cervical imaging studies. He also mentioned self-limited episodes of symmetric polyarthralgia involving hands, wrists, elbows, knees and feet, which started after his cervical complaints. On laboratorial workup, positive rheumatoid factor and anti-citrullinated peptide antibody and negative HLA-B27 were found. Cervical spine magnetic resonance imaging revealed atlantoaxial subluxation and odontoid process inflammatory pannus and erosions. Rheumatoid arthritis with cervical spine involvement as initial manifestation of disease was the definite diagnosis. The patient was started on methotrexate and prednisone and he was referred to neurosurgery outpatient clinic for cervical spine fixation.

Keywords: Rheumatoid arthritis; Cervical spine; Inflammatory pannus; Atlantoaxial subluxation

INTRODUCTION

Rheumatoid arthritis (RA) is the most important and frequent chronic inflammatory arthropathy. Although it is known to cause progressive joint destruction and severe disability, the current available disease-modifying antirheumatic drugs (DMARD's) have changed RA's natural history and reduced its personal, social and economic burden.

In addition to RA poorly understood etiology and pathogenesis, there is also heterogeneity in its clinical presentation. The main feature is synovitis, which can affect potentially any synovial membrane in the body (articular and periarticular). The classic presentation is symmetric, additive and persistent inflammatory polyarthritis involving the proximal interphalangeal and metacarpophalangeal joints, wrists, elbows, shoulders, hips, knees, ankles and metatarsophalangeal joints. There can be also prolonged morning stiffness and functional impairment. The development of these symptoms is usually gradual but it can also be abrupt. Less frequently, initial RA involvement may be monoarticular or oligoarticular¹.

Spine involvement is a common late finding and it's mostly limited to the cervical spine. It occurs in over half the patients after an average of 10-year of disease duration².

The authors report an average of case of cervical spine involvement as initial manifestation of RA.

CLINICAL CASE

A 35 year-old caucasian male patient was referred to our rheumatology outpatient clinic from his primary care physician for severe and disabling neck pain. The pain started 2 years earlier without a recognizable triggering event and got progressively worse. No pain radiation or neurologic symptoms were reported. The

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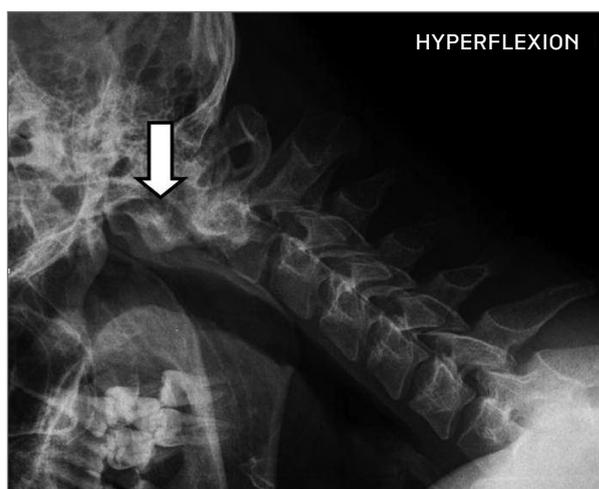


FIGURE 1. Full flexion lateral view radiography. Notice the increased space between the anterior arch of the atlas and the odontoid process, also known as atlantoaxial subluxation (white arrow)

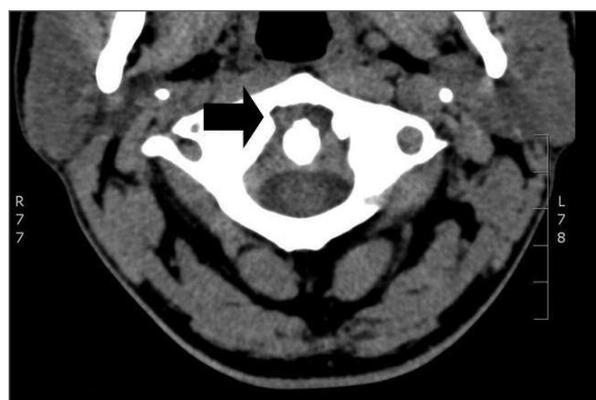


FIGURE 2. Cervical spine computerized axial tomography showing the increased space between the anterior arch of the atlas and the odontoid process (atlantoaxial subluxation, black arrow) as well as the irregularities of the odontoid process.

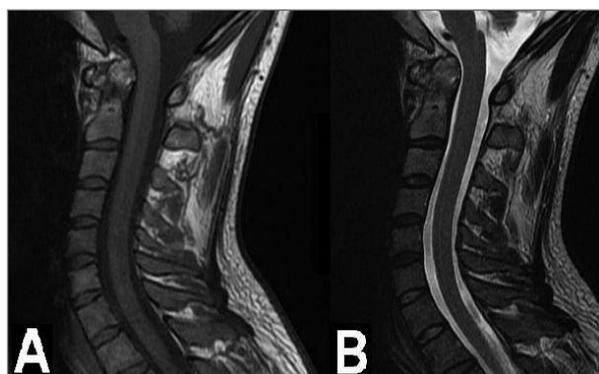


FIGURE 3. T1 (A) and T2 (B) weighted sagittal magnetic resonance of the cervical spine, showing the atlantoaxial subluxation, the inflammatory pannus heterogeneity and the erosions of the odontoid process

pain had an inflammatory rhythm and was associated with limited range of motion and prolonged morning stiffness. Activities of daily living were gradually compromised, including the performance of personal hygiene, bathing and dressing. Work attendance was also reduced. He went to his family physician and he was submitted to cervical spine plain radiography and computerized tomography, which showed no abnormalities. During this period, the patient had to resort repeatedly to the emergency department (2-3 times per week) because of his neck complaints; he was always treated with intramuscular non-steroidal anti-inflammatory drugs (NSAIDs) with only temporary relief.

The patient had no relevant past medical or surgical history and no tobacco, alcohol or illicit drug abuse was reported.

Physical examination revealed normal blood pressure, pulse and respiratory rate. He was afebrile. Chest, abdomen and limb examination showed no abnormalities. There was tenderness in his cervical spinous processes as well as pain and serious limitation of active and passive cervical movement (maximum of 30 degrees of rotation and 10 degrees of lateral flexion). The remainder musculoskeletal examination and the neurologic examination were unremarkable.

At this point, an axial seronegative spondyloarthritis was considered as the most likely diagnosis and the patient was started on aceclofenac 100 mg twice a day.

Posterior laboratory workup revealed a normal complete blood count, as well as normal kidney and liver function tests. Serum protein electrophoresis revealed no abnormalities. C-reactive protein level was 0.79 mg/dL and erythrocyte sedimentation rate was 4 mm/1st. Negative HIV and HCV but positive HBs and HBc antibodies were found. Immunology tests revealed a positive rheumatoid factor (93 U/mL) and positive low titer anti-citrullinated peptide antibody (30 U/mL). Antinuclear antibody titer was 1/160 and antibodies against extractable nuclear antigens were negative. C3 and C4 complement levels were within normal range. HLA-B27 was negative. Chest and pelvic radiographies were unremarkable.

In the first follow-up consultation, the patient reported little improvement of the cervical complaints with aceclofenac and referred concomitant left wrist

pain. Left wrist arthritis as well as right hand flexor tenosynovitis were objectified. He then recalled having self-limited episodes of symmetric inflammatory polyarthralgia involving wrists, elbows, shoulders, knees and ankles, sometimes associated with swelling, starting a few months after his neck pain. In light of all the clinical and laboratorial findings, RA was considered and urgent cervical imaging studies were performed, including spine gadolinium-based magnetic resonance imaging (MRI). This latter imaging study showed atlantoaxial subluxation (AAS) and odontoid process inflammatory pannus with multiple small erosions. Inflammation of the atlas' articular processes was also present. There was no evidence of medullary compromise (Figures 1, 2, 3).

RA with cervical spine involvement as initial manifestation of disease was the definite diagnosis. Prednisone 10 mg qid, methotrexate 15 mg per week and folic acid 10 mg per week were added and the patient was referred to neurosurgery for cervical spine fixation surgery.

DISCUSSION

The authors report this case because, although cervical spine involvement is a common feature in longstanding disease, it is a very unusual form of presentation of RA. Axial skeleton involvement is a common feature of RA. It has a variable prevalence ranging from 25 to 80% (3), with the cervical spine being almost exclusively affected. Although typical of longstanding disease, it may develop rather early with a reported prevalence of 10% after 2 years of disease duration and being increasingly frequent with ongoing inflammatory activity⁴. In the literature, there are dozens of reports of cervical involvement of RA that occur with longstanding disease but we only found two of cervical involvement as a presenting feature of the disease, both reported in 2010 (one in Germany⁵ and the other in Spain⁶). This case represented a diagnostic challenge because the inflammatory pattern of the patient's axial symptoms resembled a spondyloarthritis and his serum inflammatory markers were negative. However, the symmetric involvement of peripheral joints as well as the incomplete response to NSAIDs, the positive rheumatoid factor and the positive anti-citrullinated peptide antibody raised the suspicion of RA.

There are three types of cervical spine involvement in rheumatoid arthritis (RA). The most common is AAS (49%), followed by superior migration of the odontoid

(SMO, 38%) and subaxial subluxation (SAS, 10-20%)⁷. In AAS, synovitis of the transverse ligament causes laxity and erosions of the odontoid process in which case the anterior arch of the atlas may move forward on neck flexion and upper cervical cord compression may occur¹. SMO is due to the erosion and bone loss in the occipitoatlantal and atlantoaxial joints, which causes the cranial migration of the odontoid process and compression of vital structures of the brainstem. In SAS there is unstable movement of vertebrae below the axis due to destruction of facets, intervertebral discs and interspinous ligaments¹.

The earliest and most frequent symptom of cervical involvement is neck pain radiating to the occiput⁸. Other symptoms may include a sensation of head falling forward upon flexion, dysphagia, vertigo, dysarthria, nystagmus, peripheral paresthesias, quadriplegia, loss of sphincter control, respiratory dysfunction and consciousness changes⁹. Transient ischemic attacks may occur due to vertebral artery compromise. Suggestive physical findings include loss of physiological lordosis, painful and limited active and passive cervical movements.

Diagnostic confirmation is radiological and should combine the use of cervical lateral view plain radiography in full flexion (to assess the AAS) and cervical MRI (to assess synovitis and neural structures compression)⁴. In lateral plain radiography, the distance between the posterior margin of the anterior ring of the atlas and the anterior surface of the odontoid process (anterior atlantodental distance) is considered abnormal when > 3 mm in the adult patient with RA¹⁰. Although an anterior atlantodental distance of ≥ 9 mm was associated with an increased incidence of paralysis, a distance of ≤ 14 mm between the posterior surface of the odontoid and the anterior margin of the posterior ring of the atlas showed a much higher sensitivity and negative predictive value for poor neurologic outcomes¹¹.

Treatment may be surgical or conservative, depending on the presence or absence of signs of spinal cord compression, respectively. DMARDs are the cornerstone of RA therapy and they are essential for controlling inflammation and preventing joint damage in both peripheral and axial joints. There is some evidence that DMARD combination therapy may offer benefit over monotherapy in preventing cervical spine involvement¹². Conservative treatment included the use of stiff collars for stabilization but their benefit in preventing serious injury is still controversial; its use should be

decided on an individual basis^{12,13}. The role and safety of cervical physical exercises is also uncertain. Patients with refractory pain or evidence of spinal cord compromise must be submitted to surgery in order to prevent irreversible neurological damage and death. Surgical management includes reduction and stabilization of the injured spine segment and decompression of neural structures¹⁴.

Cervical involvement is a rather common late finding in RA and should always be considered when patients with long-standing disease present with new-onset cervical pain or neurologic symptoms. The reported case of cervical arthritis, as a presenting feature of RA, corroborates that this is a heterogeneous disease and may have atypical presentations for which the physician must be aware.

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