Reumatismo, 2011; 63 (1): 38-43

Spondylodiscitis as the only clinical manifestation of the onset of psoriatic spondyloarthritis

Spondilodiscite come unica manifestazione clinica di esordio di spondiloartrite psoriasica

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RIASSUNTO

Viene descritto il caso di un uomo di 47 anni, con esordio subdolo e progressivamente invalidante di rachialgia dorsale. Il paziente presentava minime lesioni dermatitiche ai gomiti e retro auricolari che venivano attribuite a psoriasi minima. Un' iniziale RMN del rachide dorsale, effettuata un mese dopo l'inizio dei sintomi, mostrava un'alterazione del segnale a livello delle vertebre D7 –D8, come da edema osseo. Una successiva TAC del rachide, circa 6 mesi dopo, evidenziava un processo osteolitico importante di D7 e D8 e vasta tumefazione dei tessuti circostanti. Ad una contemporanea TAC polmonare si evidenziava un'opacità polmonare. Una prima ipotesi di neoplasia polmonare con metastasi vertebrali veniva scartata dalla broncoscopia negativa e la successiva scomparsa dell'opacità polmonare dopo terapia antibiotica. Un'agobiopsia vertebrale TAC guidata dava esito negativo per patologia tumorale, granulomatosa ed infettiva. La comparsa successiva anche di poliartrite periferica e l'iniziale edema osseo giustificava la diagnosi di spondilodiscite psoriasica. Veniva iniziata terapia con Anti TNF-alpha (Etanercept) con la quale si risolveva rapidamente sia la sintomatologia dolorosa che il danno radiologico.

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INTRODUCTION

Psoriatic arthritis falls within the family of seronegative spondyloarthritis given that the involvement of the spine, whether in combination with peripheral arthritis or not, is one of the most common clinical findings of the disease.

Symptomatological polymorphism of the disease, however, still includes several clinical subsets ranging from peripheral arthritis, mono-, oligo- and polyarticular, to enthesitis and dactylitis, all the way to crippling arthritis.

Despite the frequent and characteristic axial involvement of psoriatic arthritis, an evident spondyloscitis, either at the onset or in the course of the disease, is an extremely rare event and in literature it has only been described in SAPHO syndrome (1, 2). Here we describe the case report of a man who came under our observation for severe disabling pain in the spine which, after several investigations, was attributed to psoriatic spondylodiscitis.

CASE REPORT

PL, a 47-year-old man, came under our observation in March 2009 for a symptomatology characterized by violent, excruciating and continuous pain in the dorsal region. The pain had arose in October 2008 and progressively worsened. At the same time he was going through sporadic episodes of evening fever. He had no other painful symptomatology in the joints.

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Previous history showed arterial hypertension treated with beta-blockers, diabetes treated with Metformin and a pulmonary thromboembolism for posttraumatic deep vein thrombosis in the right leg. Never in the past had he experienced back pain, peripheral arthritis, dactylitis or enthesitis.
No trauma to the vertebral column, neither surgical nor instrumental diagnostic maneuvers were recorded.
He had reported to have minimal dermatitis on the elbows and behind the ears for some years, to which he had never given much importance.
In November 2008 he had an MRI of the spine which showed a partial hyperintensity, a sign compatible with bone marrow edema. This had appeared in the transverse TFE sequences and in the sagittal STIR sequence, both in the frontal view and at the level of the D7-D8 vertebral bodies (Fig. 1). The patient was treated with an increasing dose of NSAIDs and analgesics for several months in order to relieve the strong back pain, but he was still unable to perform his normal daily and work activities.
In March 2009, under our observation, the patient appeared to be severely suffering from a nagging pain located in the spine. The physical examination showed a shortness of breath in the right lung with no relevance to other organs or systems. The skin inspection showed the slightest dermatitis on the elbows and behind the ears, similar to psoriasis.
Our first tests showed an elevated ESR (48 mm/h), a high level of CRP and fibrinogen and a small increase in alpha-2 and gamma globulins. Normal immunoglobulins. CBC, liver and kidney function tests normal. The dermatological examination confirmed the diagnosis of minimal psoriasis.
A chest x-ray showed a right posterior lung consolidation similar to atelectasis and a spine x-ray showed a structural alteration between the vertebral bodies of D7 and D8. The x-ray of the sacro-iliac joints was normal.
A CT scan of the lungs was then requested which confirmed a coarse right lung consolidation. Furthermore, a CT scan of the spine showed a severe osteolytic lesion of

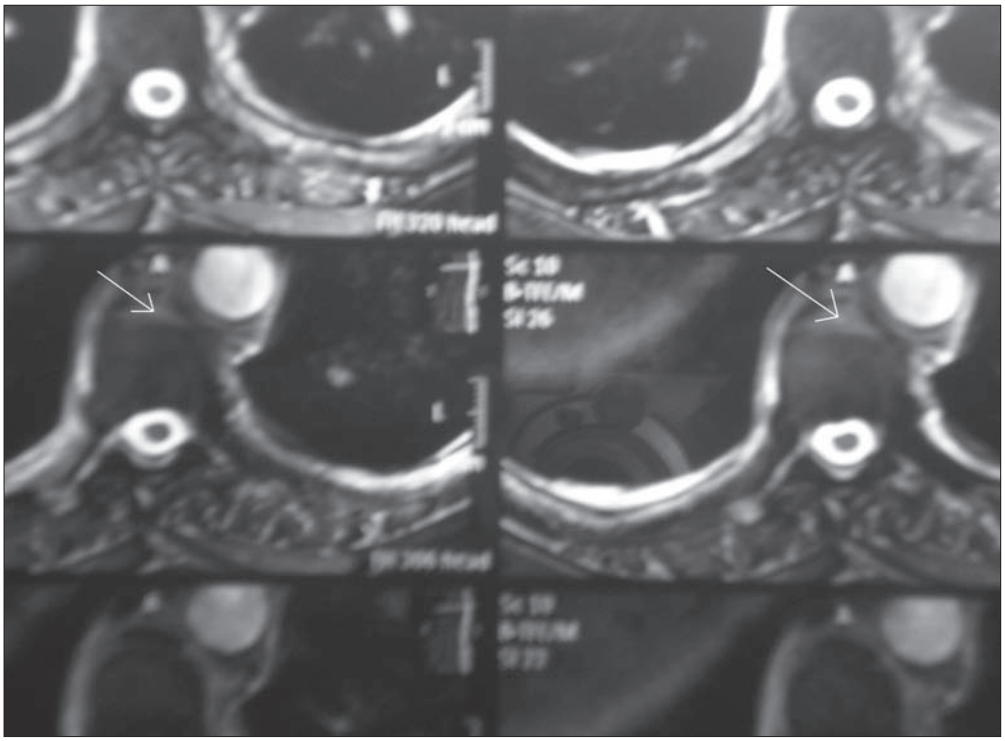

Figure 1 - Bone marrow oedema.

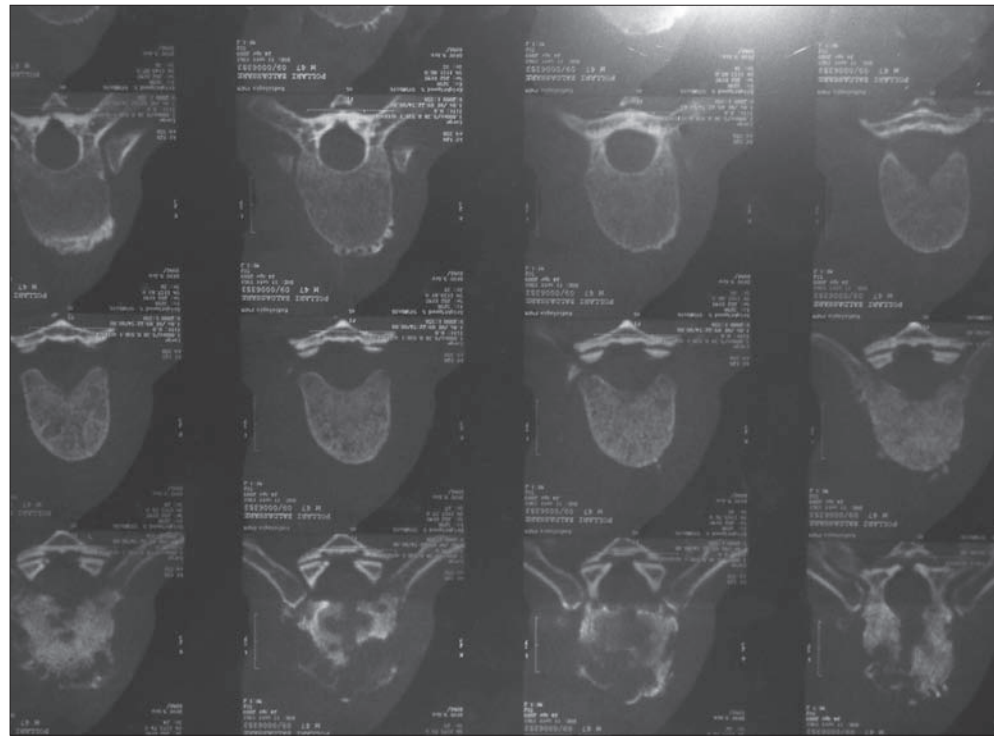

Figure 2 - Severe osteolytic lesion.

the lower border of the D7 vertebral body and a lesion on the top border of the D8 vertebra with significant thickening of surrounding soft tissues (Fig. 2). The hypothesis of metastatic lung cancer caused us to make a bone scan which showed an increased signal intensity in correspondence of the seventh and eighth dorsal vertebrae. Moreover, a bronchoscopy showed only a slight hypoventilation in the middle and lower right lobe bronchi, due to extrinsic compression.

At that time a transbronchial biopsy was performed with cyto-histological and bacteriological examinations, including one for mycobacteria. All of these had negative results.

A subsequent CT scan of the abdomen and pelvis was negative, just as were all tumor markers.

In light of a clinical history which lasted for over six months, in the complete absence of respiratory symptoms and in the negativity of the lung biopsy, the hypothesis of lung cancer was abandoned. This clinical direction was also confirmed by a subsequent lung CT scan that showed the complete resolution of the consolidation after antibiotic therapy.

Subsequent tests carried out (HIV, lymphocyte populations, serodiagnosis, Bence-Jones urine and serum tests, tuberculin test, calcitonin) were all normal. The gastroscopy and colonoscopy were also negative.

The patient still continued to suffer violent, persistent pain in the dorsal region, which had irradiated in recent weeks to the chest band and was not responsive to treatment with opioid analgesics.

In May 2009 we decided to make a CT guided transthoracic needle biopsy of the spine. The CT scan proved severe osteolytic vertebral damage and the thickening of soft tissue, comparable to the previous examination.

Adequate bone frustule was taken for cyto-histological and bacteriological examinations which all came out negative for both neoplastic or granulomatous processes and for infectious processes.

We decided to start a broad-spectrum of

antibiotic therapy (ciprofloxacin, clarithromycin and amoxicillin) associated with ibandronate.
Therapy was carried out for three months without getting any benefit.
In July 2009, the patient also began to experience pain with functional limitations and slight swelling in the wrists and in the MCP joints of both hands, including pain in the feet and ankles.
At this point, also in light of the early vertebral bone marrow edema, the first suspicion of psoriatic arthritis was reconsidered, evaluating an unusual onset with spondylodiscitis. In August, all antibiotics were suspended and treatment started with anti-TNF-alpha (Etanercept 25 mg/2 times a week).
After the first month of treatment the patient already showed a significant improvement in pain symptomatology which allowed to stop all analgesics. Within two months the complete cessation of any symptoms was reported, both in the spine and in the peripheral joints; even the skin psoriasis disappeared.
In October 2009, a CT scan of the spine showed a recovery in osteosclerosis of the D7 and D8 vertebrae with a net reduction in osteolytic phenomena and in soft tissue swelling (Fig. 3).
The analysis showed normalization of ESR and CRP levels.
After 6 months of therapy with Etanercept, the patient, still in therapy, enjoys excellent health and has resumed all of his work activities.

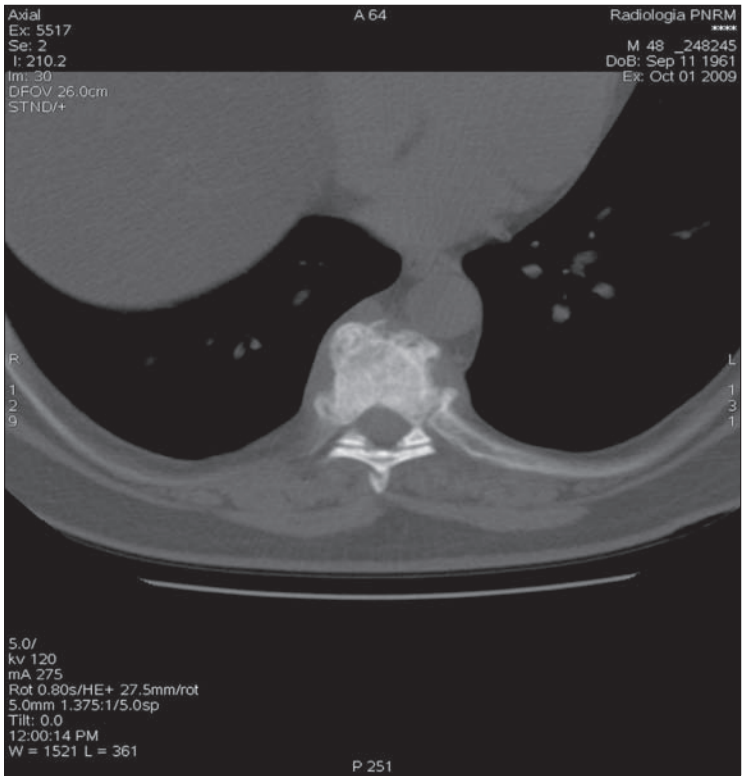

Figure 3 - Recovery in osteosclerosis.

DISCUSSION

Spondylodiscitis usually represents a complication of sepsis. The most involved pathogens are Staphylococcus aureus in approximately 60% of cases and Enterobacter in 30% (3).
The least common infection is from Streptococcus pneumoniae in the course of pleuropneumonia (4).
Tuberculous spondylodiscitis is usually an expression of systemic tuberculosis in an immunocompromised patient (5).
Salmonella and Brucella are rarely involved. Spondylodiscitis may also occur in the course of Hodgkin's lymphoma and especially as a complication of instrumental spinal maneuvers.
Seronegative spondyloarthritis and inflammatory autoimmune diseases are not the common cause of spondylodiscitis. In these cases the manifestation of spondylodiscitis is also an expression of a long-term illness, aggressive in its course.
Ankylosing spondylitis, among these, is the disease that is most commonly complicated by spondylodiscitis (6).
Only in the SAPHO variant of psoriatic spondyloarthritis have other cases of spondylodiscitis been reported, but never as a clinical expression of an onset of a disease (1, 2). Other cases of spondylodiscitis have also been described in rare variants of spondyloarthritis, such as in acne fulminans or in patients on hemodialysis therapy for a long time (7).
Our case therefore presents unique clinical

features. Spondylodiscitis had manifested as the only clinical symptom of an onset and as the only axial lesion of an unrecognizable psoriatic arthritis. In fact, the patient was not even aware that he suffered from skin psoriasis and had never suffered from joint disease in the past.
Only after several months since the beginning of axial symptoms due to spondylodiscitis, the patient experienced peripheral arthritis.
Spondylodiscitis during psoriatic arthritis is rare and, to the best of our knowledge, has never been reported in literature.
Just as those which complicate ankylosing spondylitis, it should be an expression of a severe chronic illness in an immunocompromised patient.
Our patient had not done immunosuppressive therapy.
He was not a carrier of HIV or other infectious diseases. Furthermore, the needle biopsy excluded any infectious pathogenesis, just as cancer and granulomatous diseases were excluded.
The other peculiarity of our case is represented by the complete and rapid healing response to anti-TNF-alpha therapy. After only two months of therapy with Etanercept, spinal pain symptomatology, which had tormented the patient for about a year, had completely disappeared.
He also resumed full work activity without needing analgesic therapy. The CT scan showed the healing in sclerosis of the spondylodiscitis lesion.
This confirms that the TNF-alpha cytokine was strongly implicated in the genesis of spondylodiscitis, as it is for skin lesions and joints of psoriatic spondyloarthritis.
After the failure of non-steroidal anti-inflammatory drugs, the new guidelines for psoriatic arthritis, developed by the "Group for Research and Assessment of Psoriasis and Psoriatic Arthritis (GRAPPA)", in cases of axial involvement, identify the second therapeutic step directly in anti-TNF-alpha drugs (8).
In several clinical trials, these drugs have also amply demonstrated their effectiveness in clinical control and in radiological progression of the disease (9-11).
Our patient is being treated with Eternacept for over six months. He enjoys excellent health and has not been met with any infectious complications. A complication of this type was the biggest fear that had constrained us the most when undertaking therapy with biologicals. As a result of the rarity of the case, a remote doubt of an infectious cause had still remained.
Our case demonstrates how a severe spondylodiscitis can be the first clinical manifestation of psoriatic spondyloarthritis. Although it may be unrecognizable, it confirms that this disease may have clinical symptoms that are most varied and often unpredictable.

SUMMARY

We report the case of a 47-year-old man with insidious onset of progressively disabling back pain in the dorsal region. The patient had minimal dermatitic lesions to the elbows and behind the ears, which were attributed to minimal psoriasis. An initial MRI of the spine, one month after the onset of symptoms, showed an alteration in the D7-D8 vertebrae as from bone marrow edema. The successive CT scan of the spine, after about six months, showed a significant osteolytic process of the D7 and D8 vertebrae and extensive swelling of surrounding tissues. A contemporary lung CT scan showed opacity in the right lung. A first hypothesis of lung cancer with vertebral metastases was ruled out by the negative bronchoscopy and the subsequent disappearance of lung opacity after antibiotic therapy. A CT-guided needle biopsy of the spine gave negative results for granulomatous and infectious tumor pathology. The later appearance of peripheral polyarthritis and the presence of initial bone marrow edema justified the diagnosis of psoriatic spondylodiscitis. Therapy with anti-TNF-alpha (Eternacept) was initiated, with which both the painful symptomatology and the radiological damage were quickly resolved. This is the first case in literature about spondylodiscitis as the manifestation of the onset of psoriatic spondyloarthritis.

Parole chiave: spondilodiscite, psoriasi minima, agobiopsia vertebrale, anti-TNF- alpha
Key words: *spondylodiscitis, minimal psoriasis, needle biopsy of the spine, anti-TNF-alpha.*

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