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Oligo-secretory myeloma in a patient with ankylosing spondylitis

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Abstract Non-secretory myeloma is a plasma cell dyscrasia characterized by the absence of serum and urinary monoclonal immunoglobulins on electrophoretic tests. Because of the lack of monoclonal protein, the identification of the disease is more difficult than for secretory myelomas. The coexistence of ankylosing spondylitis and multiple myeloma has been reported occasionally. We report a rare case of oligo-secretory myeloma coexistent with ankylosing spondylitis.

Keywords Ankylosing spondylitis · Multiple myeloma · Non-secretory myeloma · Immunoglobulin light chains

Introduction

The association of ankylosing spondylitis (AS) and multiple myeloma (MM) has been reported occasionally [1, 2]. Moreover, the coexistence of AS and oligo-secretory myeloma (OSM) is exceptional [3]. Here, we report a case of OSM in a patient with long-standing AS.

Case report

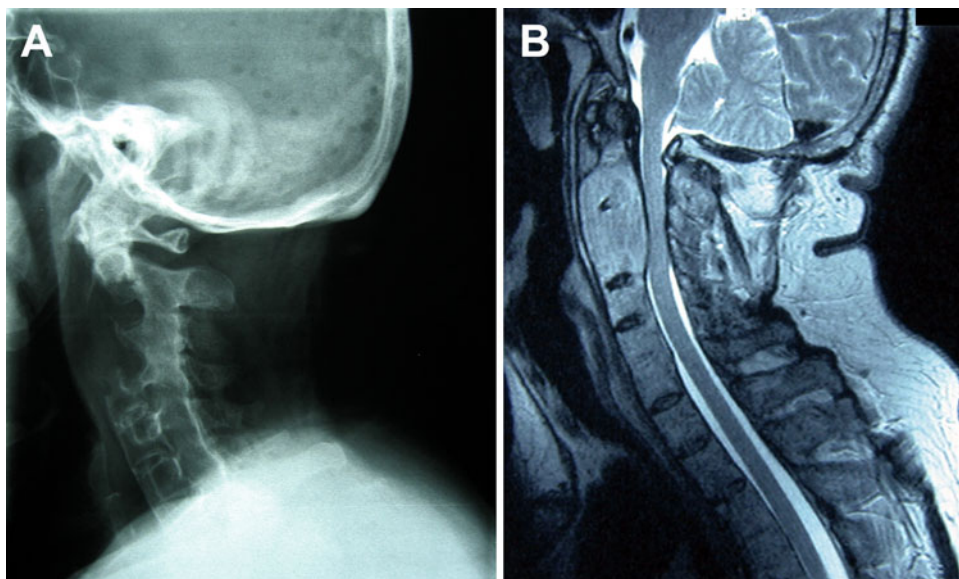
A 55-year-old man with a 35-year history of AS (HLA B27 positive) and schizophrenia was admitted to the hospital because of intractable cervical pain. The patient was maintained on stable medication with fluphenazine decanoate, haloperidol, ziprasidone, paroxetine, biperiden and indomethacin at standard doses and pain was alleviated with a combination of tramadol and acetaminophen. Also, he received a short course of infliximab but was discontinued due to inefficacy. He had no history of therapeutic irradiation. He presented with a 3-month history of cervical pain, right upper extremity numbness and progressive weakness of all four extremities. On examination, he showed right upper limb paresis and dysesthesia, hyporeflexia in right biceps with hyperactive reflexes in lower limbs and normal bilateral plantar stimulation responses but no sphincter dysfunction. The rest of the physical examination was normal. The routine laboratory data showed a normal complete blood count and coagulation parameters; also, serum creatinine, alkaline phosphatase, calcium and ESR levels were normal. Quantification of serum immunoglobulins detected hypogammaglobulinemia with a serum IgG value of 4.4 g/l (normal range, 7.0–16.0), IgA 0.9 g/l (normal range, 0.5–3.0) and IgM 0.2 g/l (normal range, 0.8–4.7). A chest radiograph revealed no pathologic findings, but cervical spine radiograph demonstrated a partial resorption of the vertebral bodies of C2, C3 and C4 (Fig. 1a). X-ray study of the rest of the spinal cord showed typical skeletal findings of advanced AS. A CT-scan of the cervical spine revealed a lytic lesion of C2, C3 and C4 vertebral bodies with canal obliteration. MRI of the cervical spine confirmed the lesion of the vertebral bodies and revealed a soft tissue mass determining spinal cord compression (Fig. 1b). A surgical approach was planned in order to ensure spinal decompression and

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Fig. 1 Plain lateral radiograph of the cervical spine showing resorption of C2, C3 and C4 vertebral bodies (a) and sagittal T2-weighted MRI slices demonstrating a soft tissue mass (b)



stabilization. The biopsy of the soft tissue mass showed atypical plasma cells (kappa type). No paraprotein was detected on serum or urinary protein electrophoresis and immunofixation, but the serum-free light chain (FLC) assay (FreeLite™, The Binding Site, Birmingham, UK) showed a kappa–lambda ratio of 55:1 (reference range, 0.26–1.65). The bone marrow biopsy also showed atypical plasma cell infiltration accounting for 16% of the cells. These plasma cells were positive for CD56, CD38, BB4 and kappa light chains. The karyotype of the bone marrow showed no abnormalities. In addition to cervical radiotherapy, an alternating VBCMP/VBAD (vincristine, carmustine, melphalan, cyclophosphamide and prednisone/vincristine, carmustine, doxorubicin and high-dose dexamethasone) chemotherapy scheme was initiated without objective response. Bortezomib plus dexamethasone was started but after six courses disease progression (renal failure and hypercalcemia) was observed. A bone marrow biopsy revealed 35% atypical plasma cells. Fluorescence in situ hybridization (FISH) was performed showing 13q deletion and IgH rearrangement. DHAP (cytosine arabinoside–cisplatin–dexamethasone) was initiated but the patient developed nosocomial pneumonia with respiratory distress requiring intubation and mechanical ventilation. Finally, he presented refractory respiratory failure and died.

Discussion

The risk of monoclonal gammopathy and MM is increased in patients with some autoimmune and inflammatory diseases and specifically in AS [4]. However, other studies did not show increased risk of hematological malignancies,

except in patients with AS subjected to radiation treatment [5–7]. There are few studies in the literature which supported an association between AS and MM and most of the reported cases involved the IgA class [1, 2]. Predisposing factors for the development of MM included preceding monoclonal gammopathy, severe activity of AS, prolonged plasma cell stimulation and proliferation in response to elevated levels of IgA, and high number of X-ray investigations. Others, as dysregulation in the mechanisms leading to apoptosis can only be speculated.

Non-secretory myeloma (NSM) represents approximately 1–5% of all myeloma cases and is defined by the absence of detectable monoclonal protein in both the serum and urine in patients who otherwise manifest the typical clinical picture of MM (lytic bone lesions, renal failure, hypercalcemia) [8]. The FLC assay is a nephelometric measurement of light chains that are not bound to immunoglobulin heavy chain. The introduction of serum FLC assays reveals that approximately three-fourths of the so-called NSM cases do have evidence of clonal immunoglobulin production and could be classified as OSM, as in our case [9]. In the rest, named as true NSM, the absence of a monoclonal protein may be a result of the inability of plasmatic cells to synthesize immunoglobulin, or the failure of such components to be exported from the cells. A frameshift mutation in the light-chain constant region has been demonstrated in a patient with a true NSM [10].

In summary, the present case is of interest because of the rare association between AS and OSM. In our knowledge, there is only one previous report about a patient with NSM and AS [3].

Conflict of interest statement The authors declare that they have no conflict of interest.

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