Erosive cervical spine involvement in primary Sjögren’s syndrome

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A 69-year-old woman, diagnosed with primary Sjögren’s syndrome that was characterized by the presence of sicca symptoms and anti-nuclear and anti-Ro antibodies and histologically confirmed with a minor salivary gland biopsy 11 years ago (Figure 1a), consulted our rheumatologic department for a recent onset of inflammatory neck pain. Physical examination revealed pronounced cervical stiffness, but no signs of synovitis in the peripheral joints. Magnetic resonance imaging of the cervical spine in sagittal T2-weighted scan showed the presence of synovitis of the atlantoaxial joint, with synovial pannus surrounding the odontoid process of the second cervical vertebra and compressing the cord (Figure 1b). Bone marrow edema and erosions were also detected in the odontoid process. Subsequent coronal (Figure 1c) and sagittal computed tomography scans (Figure 1d) confirmed the presence of well-defined multiple erosions at the base and apex of the odontoid process. Rheumatoid factor and anti-citrullinated protein antibodies were absent. X-rays of the hands, wrists, and knees revealed only mild signs of osteoarthritis.

Figure 1. a-d. Histological examination (a) of a minor salivary gland biopsy revealing a lymphocytic infiltrate suggestive of Sjögren’s syndrome, with a focus score of 4. Magnetic resonance imaging (b) (T2-weighted sagittal scan) shows synovial hypertrophy of the atlantoaxial joint with compression on the spinal cord and bone marrow edema of the odontoid process, whereas the coronal (c) and sagittal (d) computed tomography scans detail the presence of multiple erosions of the odontoid process.
The atlantoaxial joint could be involved in different articular inflammatory diseases. Calcium pyrophosphate deposition disease could appear as calcifications in the ligaments surrounding the odontoid process (“crowned dens syndrome”) (1). However, the atlantoaxial joint is one of the chief target in rheumatoid arthritis (2). To our knowledge, this is the first depiction of inflammatory and erosive cervical spine involvement in primary Sjögren’s syndrome.

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References