Severe cervical stenosis due to ossification of the posterior longitudinal ligament without neurological manifestations ("silent OPLL")

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We report a case of severe cervical spinal stenosis due to ossification of the posterior longitudinal ligament (OPLL) whose clinical examination showed no symptoms or signs of neurological damage ("silent OPLL"). Questions about the diagnosis and the treatment of silent OPLL are discussed in the light of published data.

Key-Words: Cervical stenosis — ossification of posterior longitudinal ligament

Introduction

It is widely accepted that cervical spinal canal dimensions are determinants of neurological compromises and the risk of myelopathy is very high with a sagittal diameter of 10 mm or less. Ossification of the posterior longitudinal ligament (OPLL) is a not uncommon cause of spinal stenosis and myelopathy particularly in Japan [11], but also in caucasians [5]. This report describes the unique case of a patient with a cervical OPLL causing a severe progressive spinal canal stenosis without clinical signs of neurological damage ("silent OPLL").

Case report

This 64 year old woman had been suffering from back pain for over 20 years and cervical pain for 10 years with morning stiffness in the shoulder and pelvic girdles. Coming for an examination at the Istituto Ortopedico Rizzoli, she brought with her spinal x-rays taken 6 years before (Fig. 1). There was hyperlordosis of the cervical spine and exaggerated thoracic kyphosis with a marked reduction in forward flexion of the spine particularly at the cervical level. The right scapulo-humeral joint was rigid and painful. Active and passive range of motion was normal but painful in the other joints. The neurological examination was normal. Laboratory data were within normal limits except alkaline phosphatase, which was 190 (60-170) and sedimentation rate which was 35 (NV < 15).

The new radiological study of the cervical spine (Fig. 2A and B) showed an increase in the ossification of the anterior longitudinal ligament (OALL) and of the posterior longitudinal ligament (OPLL) that had initially been overlooked. Besides an increase in the OALL, the radiological study of the thoracic spine suggested a possible
ossification of the anterior longitudinal ligament with normal height of the disc spaces. A continuous type of OPLL from C2 to C5 had been overlooked (black arrows).

ossification of the ligamenta flava. The radiological study of the lumbar spine showed a remarkable increase in the OALL, an ossification of inter- and supra-spinous ligaments, and a possible localized OPLL at L2-L3. There was no significant intervertebral disc space loss along the entire spine. Cervical CT scans helped to evaluate (Fig. 2C) the residual dimensions of the vertebral canal and the presence of a segmental OPLL at C6 and to reveal it at C7. In spite of the marked ossification of the posterior longitudinal ligament causing severe reduction in the sagittal diameter of the cervical canal this patient with radiological signs of diffuse idiopathic skeletal hyperostosis (DISH) showed no symptoms nor signs of neurological damage ("silent" OPLL). Corticosteroid treatment was useful to control symptoms of polymyalgia.

Discussion

It has been widely accepted that a decreased diameter of the spinal canal is crucial to the development of cervical myelopathy, which may be expected when the sagittal spinal canal measures 10 mm or less [4]. Degenerative changes involving vertebral bodies and intervertebral discs are the well known and most frequent factors in developing acquired spinal stenosis. Stenotic myelopathy can also occur as a result of spinal ligament ossifications. OPLL, although initially described by Coste [3], began to be fully recognized only after the papers of Terayama [10] and Tsuyama [11], and from then on it was regarded mostly as a "Japanese disease" [2]. A roentgenological review study recently done in Bologna at the Istituto Ortopedico Rizzoli [1] on 1258 cervical X-rays showed 22 cases of OPLL with a frequency of 1.75%, a figure close to that found in Japan [11]. The reported case gives rise to a few comments and at least one unresolved question. First of all, in this case as in others OPLL is not an isolated entity but an aspect of a more diffuse tendency towards ossification of the ligaments called diffuse idiopathic skeletal hyperostosis (DISH) after Resnick [9]. In the reported case, however there were no signs of spinal cord involvement apart...