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CASE REPORT

Ossification of the posterior longitudinal ligament in dizygotic twins with schizophrenia: a case report

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Abstract The pathogenesis of ossification of the posterior longitudinal ligaments (OPLL) has not been clarified. We here report dizygotic twin sisters with OPLL of the cervical spine and propose a new pathogenesis of OPLL. This is the first report of dizygotic twins with OPLL. The twins suffered from schizophrenia, which might be related to the pathogenesis of OPLL. In addition, we investigated the occurrence of OPLL in 30 patients with schizophrenia who had been admitted to a mental hospital. OPLL of the cervical spine was found in six (20%) of them, with an incidence almost five times higher than the incidence of OPLL among the general population in Japan. Schizophrenia may have a increased susceptibility to OPLL.

Keywords Ossification of the posterior longitudinal ligament (OPLL) · Schizophrenia · Dizygotic twins · Calcineurin · Myelopathy

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Introduction

Ossification of the posterior longitudinal ligament (OPLL) is a hypertrophic condition of the spine associated with severe neurological deficit [1–5]. The disease was first reported by Key [6] in 1838, and Tsukimoto [7] reported a postmortem examination of a Japanese patient. The pathogenesis of OPLL has been studied since 1975 by members of the Investigation Committee of The Japanese Ministry of Public Health and Welfare. However, the pathogenesis of OPLL has not been clarified. Genetic background is thought to play a major role in OPLL from results of studies involving families [8] and twins [9]. However, DNA analysis has not shown specific genes conferring susceptibility to OPLL. The Japan-wide study of twins with OPLL revealed six of eight monozygotic twins had OPLL of the cervical spine. However, no dizygotic twins with OPLL were found, and the mode of inheritance of OPLL could not been determined. In this study, dizygotic twin sisters with OPLL of the cervical spine are reported, and a new pathogenesis is proposed.

Case

Dizygotic twin sisters (Fig. 1), aged 69 years, suffered from schizophrenia for 40 years. The younger of the two exhibited OPLL of the cervical spine (Fig. 2), with a maximum spinal canal stenosis rate of 45%, but she did not exhibit myelopathy. In contrast, the older twin had almost the same size and degree of OPLL of the cervical spine (Fig. 3), but she exhibited severe myelopathy. Both sisters were short in stature, but laboratory data did not correspond

Fig. 1 **a** The younger sister of the dizygotic twins. **b** The elder sister of the dizygotic twins

Fig. 2 Plain X-ray film of the cervical spine and computed tomography of the younger sister of the dizygotic twins; *white arrows* show ossification of the posterior longitudinal ligament

to vitamin-D-resistant rickets, which is frequently associated with OPLL.

Supplementary study

With the agreement of the families, we investigated the occurrence of OPLL in 30 patients with schizophrenia who had been admitted to a mental hospital. The study comprised 25 male patients and five female patients. OPLL of the cervical spine was present in six (20%) (five men and one woman) of the 30 patients with schizophrenia (Fig. 4).

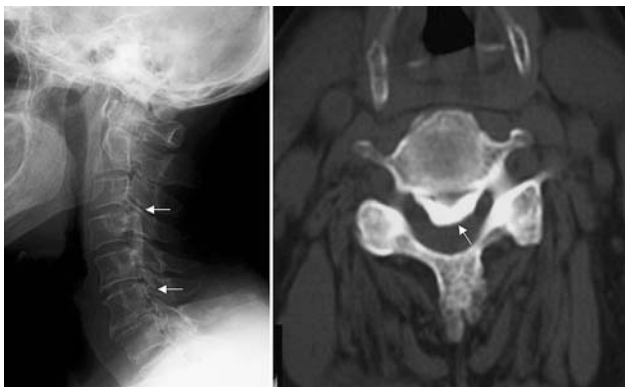

Fig. 3 Plain X-ray film of the cervical spine and computed tomography of the elder sister of the dizugotic twins, *white arrows* show ossification of the posterior longitudinal ligament

The maximum spinal canal stenosis caused by the ossified ligament ranged from 35% to 68% with an average of 56.7%. Two of the six patients who had OPLL in the cervical spine exhibited myelopathy, but the remaining four did not.

Discussion

Genetic background is thought to play a major role in OPLL, as shown by the results of Japan-wide studies of families and twins. This study is the first to report dizygotic twins with OPLL. The case of dizygotic twins presented here is one of schizophrenia associated with OPLL. Schizophrenia might be related to the pathogenesis of OPLL. Recently, calcineurin has been reported to be significantly associated with susceptibility to schizophrenia [10, 11], and it has also shown osteogenesis by the activation of osteoblasts [12, 13]. The association between OPLL and schizophrenia may not be coincidental. In our study, 20% of patients with schizophrenia exhibited OPLL, an incidence almost five times higher than among the general population in Japan (2.0∼4.3%) [14]. The drugs used to treat schizophrenia are muscle relaxant, a feature that may induce malalignment of the spine. The biomechanical changes induced by the malalignment are also related to the development of OPLL. However, patients with OPLL in our series did not have apparent spinal deformities. Some diseases, such as vitamin-D-resistant rickets [15] or polycystic ovary syndrome [16], have been reported to be associated with OPLL. Schizophrenia might also be in the category.

Fig. 4 Ossification of the posterior longitudinal ligament (OPLL) of the six patients with schizophrenia. **a** A 78-year-old man, **b** a 75-year-old man, **c** a 59-year-old man, **d** a 79-year-old woman, **e** a 67-year-old man, and **f** a 51-year-old man. *White arrows* show the OPLL

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