Thoracic Myelopathy in Japan: Epidemiological Retrospective Study in Miyagi Prefecture during 15 Years

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Aizawa, T., Sato T., Tanaka, Y., Ozawa, H., Hoshikawa, T., Ishii, Y., Morozumi, N., Ishibashi, K., Kasama F., Hyodo, H., Murakami, E., Nishihira T. and Kokubun, S. Thoracic Myelopathy in Japan: Epidemiological Retrospective Study in Miyagi Prefecture during 15 Years. Tohoku J. Exp. Med., 210 (3), 199-208 — Thoracic myelopathy is defined as spinal cord compression in the thoracic region, leading to sensory and motor dysfunctions in the trunk and lower extremities, and can be caused by various degenerative processes of the spine. Thoracic myelopathy is rare, and there are many unsolved problems including its epidemiological and clinical features. We have established a registration system of spinal surgeries, which covered almost all surgeries in Miyagi Prefecture, and enrolled the data of 265 patients with thoracic myelopathy from 1988 to 2002. The annual rate of surgery gradually increased and averaged 0.9 per 100,000 inhabitants, which was less than 1/10 of that for cervical myelopathy. About 20 patients with thoracic myelopathy are operated on in Miyagi Prefecture each year. It frequently develops in middle-aged males. About half of the cases were caused by ossification of the ligamentum flavum, followed by ossification of the posterior longitudinal ligament, intervertebral disc herniation and posterior spur. Patients usually noticed numbness or pain in the legs and the preoperative duration was long, averaging 2 years. Its symptomatic similarities to lumbar disorders might cause difficulty in making a correct diagnosis. Since thoracic myelopathy can markedly restrict the activities of daily life, even general physicians should recognize this entity. —— thoracic myelopathy; epidemiological study; ossification of the ligamentum flavum; ossification of the posterior longitudinal ligament

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Thoracic myelopathy is defined as spinal cord compression in the thoracic region, leading to sensory and motor dysfunctions in the trunk and lower extremities and urinary disturbance, and can be caused by various degenerative processes of the spine such as intervertebral disc herniation (HE), ossification of the posterior longitudinal ligament (OPLL), and posterior spur (SP) (Smith and Godersky 1987; Yonenobu et al. 1987; Otani et al. 1988; Mitra et al. 1996; Sato et al. 1997a; Fong and Wong 2004). The posterior longitudinal ligament connects the posterior aspects of the vertebral body while the ligamentum flavum is located in the interlaminar space. OPLL, HE and SP can compress the spinal cord from the anterior and OLF from the posterior.

Unlike cervical myelopathy, the symptomatology of thoracic myelopathy is not well recognized by orthopedists or even by spine surgeons or neurosurgeons. It has often been overlooked or misdiagnosed as lumbar spinal disorders as the symptoms involve mainly the lower extremities (Mitra et al. 1996; Sato et al. 1997a). The number of the patients with thoracic myelopathy is much smaller than that of those with cervical or lumbar disorders (Mitra et al. 1996; Sato et al. 1997a). Most previous studies reviewed fewer than 100 patients, usually 30 patients or less (Yonenobu et al. 1987; Shiokawa et al. 2001; Hamouda et al. 2003). Thus, few epidemiological studies have assessed the true incidence of thoracic myelopathy necessitating surgery compared with cervical or lumbar disorders. In addition, the clinical features of thoracic myelopathy including the prevalence, age distribution, initial symptoms and the rate of corresponding spinal factors remain unclear.

Since 1988, all spine surgeries at the orthopedic departments in Miyagi Prefecture, a province in northeastern Japan with a population of about 2.3 million, have been enrolled in the registration system of the Department of Orthopaedic Surgery, Tohoku University School of Medicine (Kokubun et al. 1996; Sato et al. 1997a). Historically in Japan, patients with compressive myelopathy have been usually treated by orthopedic surgeons rather than by neurosurgeons. Therefore, the data from this registration system should be reliable and, based on these data, we have reported several epidemiological studies (Kokubun et al. 1996; Sato et al. 1997a; Tanaka et al. 2003). Sato et al. (1997a), reported the epidemiological data on 81 patients with thoracic myelopathy based on this registration system for the 7 years between 1988 and 1994. To our knowledge, no other epidemiological studies on it have been reported from Japan or other countries. As the registry continued, more than 250 patients were surgically treated for thoracic myelopathy during the 15 years to 2002. Using these data, the purpose of this study was to define the epidemiological and clinical features of thoracic myelopathy in the Japanese. This paper focuses on the epidemiological findings of this myelopathy in Miyagi Prefecture and not on the surgical outcomes nor on an analysis of the factors affecting the postoperative improvement.

**MATERIALS AND METHODS**

This study was approved by the Ethical Committee of Tohoku University School of Medicine. Between 1988 and 2002, 15,714 surgical operations at 30 hospitals in Miyagi Prefecture were enrolled by the registration system of the Department of Orthopaedic Surgery, Tohoku University School of Medicine (Kokubun et al. 1996; Sato et al. 1997a). Two hundreds and sixty five patients with thoracic myelopathy required surgical intervention among the 14,458 patients in total who were the residents of this prefecture and underwent spinal surgeries within the prefecture. These 265 patients were the subjects of this study. Ten patients required 2 to 3 revisions, secondary posterior fusion, dura mater repair, addition decompression, and therefore, totally 278 surgical operations were performed, which accounted for 2% of all the spinal surgeries.

Neurological deficits attributed to thoracic myelopathy mostly included lower-extremity hyperreflexia, plank paraparesis, and/or sphincter dysfunction. Neurodiagnostic studies confirming this disorder included abnormal myelograms, computed tomograms (CT) and/or magnetic resonance imaging (MRI) studies. The diagnoses and subsequent surgical operations were performed by highly experienced spinal surgeons at 15 of the 30 hospitals in Miyagi prefecture. Cases of thoracic
myelopathy caused by spinal cord tumor, primary or metastatic bone tumor, infection, spinal cord herniation (Aizawa et al. 2001), and fracture or fracture dislocation, were excluded from the current study.

The number of operations in each year was counted and the annual rate per 100,000 inhabitants in Miyagi Prefecture was calculated using the annual population of this prefecture. Variables contributing to the clinical features of thoracic myelopathy were assessed for the 265 patients: the gender and age, the initial symptoms, the preoperative duration from the onset of the initial symptoms, the compressive factors for the spinal cord and the locations in relation to the intervertebral disc levels, and the types of surgical procedures. In addition, operative findings on ossification of the dura mater that could not be dissected from OLF, which is closely related to the difficulty of the surgery, were investigated in the OLF patients. The preoperative disease period was divided into four: shorter than 6 months, from 6 months to 1 year, from 1 to 2 years, and 2 years or longer.

**RESULTS**

The annual rate of surgery for thoracic myelopathy in Miyagi Prefecture gradually increased and the average rate per 100,000 inhabitants for 5-year periods was 0.5 between 1988 and 1992, 0.8 between 1993 and 1997 and 0.9 between 1998 and 2002 (Fig. 1). The last rate was thus almost double that of the earliest period.

Of the 265 patients undergoing thoracic decompressions, males significantly outnumbered females (2.2 ratio), and were younger on average than their female counterparts (Table 1). The highest prevalence was for male patients in their sixties, followed by those in their fifties and seventies. On the other hand, the prevalence was almost similar for female patients in their fifties, sixties and seventies and they together accounted for about 80% of all patients (Fig. 2).

OLF, OPLL, HE, and SP were most consistently contributing spinal factors to thoracic myelopathy, with half showing OLF, followed by OPLL, HE, and OLF with OPLL and SP (Fig. 3). Three patients had a combination of two of OLF, HE or SP. The remaining 13 patients had rare factors such as kyphoscoliosis (Sato et al. 1997b), spondylolisthesis, or spinal canal stenosis in association with achondroplasia.

The most common initial symptoms included numbness and tingling or pain in the lower extremities, followed by spastic gait and/or weakness. A handful (5%) of patients complained of back pain. A few patients noted atrophy/cramping of the lower extremities first. The preoperative duration from the onset of the initial symptoms averaged 2 years and about half of the patients showed symptoms longer than 1 year while one third were less than 6 months. HE patients showed the shortest preoperative durations while those of the SP patients were relatively long. The mean age at surgery of the SP and HE patients

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**Fig. 1.** Yearly changes in the surgical rate for patients with thoracic myelopathy in Miyagi Prefecture from 1988 to 2002.
Table 1. Summary of the patients with thoracic myelopathy.

<table>
<thead>
<tr>
<th></th>
<th>Male: Female</th>
<th>182: 83</th>
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<tbody>
<tr>
<td><strong>Mean age at surgery</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male ($n = 182$)</td>
<td>59 yrs (range; 29-84)</td>
<td></td>
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<tr>
<td>Female ($n = 83$)</td>
<td>63 yrs (range; 32-85)</td>
<td></td>
</tr>
<tr>
<td>OLF ($n = 139$)</td>
<td>64 yrs (range; 38-85)</td>
<td></td>
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<tr>
<td>OPLL ($n = 33$)</td>
<td>58 yrs (range; 39-84)</td>
<td></td>
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<tr>
<td>OLF + OPLL ($n = 25$)</td>
<td>58 yrs (range; 36-73)</td>
<td></td>
</tr>
<tr>
<td>HE ($n = 30$)</td>
<td>56 yrs (range; 38-79)</td>
<td></td>
</tr>
<tr>
<td>SP ($n = 22$)</td>
<td>49 yrs (range; 29-72)</td>
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<tr>
<td><strong>Initial symptoms ($n = 178$)</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tingling, numbness or pain in legs</td>
<td>56% ($n = 100$)</td>
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<tr>
<td>Gait disturbance</td>
<td>35% ($n = 62$)</td>
<td></td>
</tr>
<tr>
<td>Back pain</td>
<td>5% ($n = 9$)</td>
<td></td>
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<tr>
<td>Others</td>
<td>4% ($n = 7$)</td>
<td></td>
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<tr>
<td><strong>Averaged preoperative duration of symptoms</strong></td>
<td>2 yrs (range; 1 month-18 yrs)</td>
<td></td>
</tr>
<tr>
<td>$\leq$ 6 month</td>
<td>49 patients</td>
<td></td>
</tr>
<tr>
<td>6 months &lt; $\leq$ 1yr</td>
<td>34 patients</td>
<td></td>
</tr>
<tr>
<td>1 yr $\leq$ &lt; 2 yrs</td>
<td>29 patients</td>
<td></td>
</tr>
<tr>
<td>$\geq$ 2 yrs</td>
<td>43 patients</td>
<td></td>
</tr>
<tr>
<td>Unknown or not decidable</td>
<td>110 patients</td>
<td></td>
</tr>
<tr>
<td>OLF ($n = 139$)</td>
<td>1.8 yrs (range; 1 month-11 yrs)</td>
<td></td>
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<tr>
<td>OPLL ($n = 33$)</td>
<td>2.1 yrs (range; 1 month-17 yrs)</td>
<td></td>
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<tr>
<td>OLF + OPLL ($n = 25$)</td>
<td>2.4 yrs (range; 2 month-14 yrs)</td>
<td></td>
</tr>
<tr>
<td>HE ($n = 30$)</td>
<td>1.0 yrs (range; 1 month-6 yrs)</td>
<td></td>
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<tr>
<td>SP ($n = 22$)</td>
<td>3.0 yrs (range; 2 month-18 yrs)</td>
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</table>

OLF, ossification of the ligamentum flavum; OPLL, ossification of the posterior longitudinal ligament; HE, intervertebral disc herniation; SP, posterior spur.

Statistically significant differences can be detected only in the male/female ratio. *$p < 0.05$.

Fig. 2. Age distribution of the patients with thoracic myelopathy in Miyagi Prefecture. Among males, the patients in their sixties show the highest prevalence, and among females, those in the fifties, sixties and seventies show almost equal prevalences.
was lower than that of those with the other compressive spinal factors (Table 1).

The decompression levels for the four major factors, OLF, OPLL, HE and SP, considered to be responsible for the thoracic myelopathy are shown in Fig. 4. The patients having a combination of two of those factors were excluded as it was uncertain which factor was responsible for the myelopathy. The surgical levels for OLF were between T10/11 and T11/12 in 65%, for OPLL they were between T1/2 and T6/7 in 84%, for HE at T7/8 or lower in 90%, and for SP between T10/11 and T12/L1 in 64%.

The surgical procedures for the four major compressing factors are summarized in Table 2. Laminectomy was most frequently performed for OLF followed by fenestration, partial resection of the lamina with the spinous process and upper part of the lamina kept intact. Before 1993, laminectomy was performed in 24 of 27 patients and hemilaminectomy in the others. Thereafter, OLF at a single level without fusion in the middle of the spinal canal was usually removed by fenestration and laminectomy, and fenestration was performed with equal frequency (Sato et al. 1998). Ossified dura mater that could not be dissected

Fig. 3. Spinal factors compressing the spinal cord leading to thoracic myelopathy.
OLF, ossification of the ligamentum flavum; OPLL, ossification of the posterior longitudinal ligament; HE, intervertebral disc herniation; SP, posterior spur.

Fig. 4. Distribution of the four major compressive factors in the spinal cord in relation to the intervertebral disc level.
OLF is most common in the lower thoracic spine while OPLL is frequently distributed in the upper to middle thoracic. Intervertebral disc herniation is mainly detected in the middle and lower thoracic spine and posterior spur in the lower thoracic spine.
from the OLF was observed in 12 (9%) of 139 patients who had OLF alone (Fig. 5). Thick or beak-like OPLL combined with or without OLF (Fig. 6) usually compressed the spinal cord very severely and laminectomy alone could not achieve sufficient decompression. In such cases, anterior decompression through a diagonal anterior and posterior approach was done for two patients before 1994 (Kokubun et al. 1991), and anterior decompression through posterior approach described by Otsuka et al. (1983) was adopted for four patients after 1997. HE was treated by anterior decompression and spinal fusion (ASF) through an extrapleural or thoracotomy approach before 1996 and mainly by discectomy through a transverso-arthro-pediculectomy approach from the posterior afterwards (Sato 2003).
Fig. 5. Preoperative CT-myelogram of OLF with ossified dura mater at T10/11. OLF protrudes right-ventrally and the contrast medium between the spinal cord and OLF can not be detected (arrowhead).

Fig. 6. Preoperative CT of a patient with OPLL.
A: Sagittal plane. Continuous OPLL can be found in the upper thoracic spine including beak-like regions (arrows).
B: Axial plane. Thick OPLL occupies the spinal canal at T3 level (arrowhead).
DISCUSSION

Thoracic spinal disorders including tumors and fractures warranting surgical intervention constituted only 7% of all spinal procedures performed in the registration system of the Department of Orthopaedic Surgery, Tohoku University School of Medicine (Tanaka et al. 2003). Cases of thoracic myelopathy caused by degenerative processes of spine were even fewer, representing only 2% of all the spinal surgeries in this study. The annual rate of surgery was 0.9 per 100,000 inhabitants from 1998 to 2002 in Miyagi Prefecture, which was less than 1/10 of that for cervical myelopathy (10.9) in the same prefecture in the same period, but nearly doubled during those 15 years (Tanaka et al. 2003). This increase might be attributed to advances in neurodiagnostic imaging including CT and MRI and in the training of spinal surgeons.

The present study showed that thoracic myelopathy more frequently develops in middle-aged males. Previous studies also indicated that male cases were more common than female cases (Sato et al. 1997a; Shiokawa et al. 2001; Hamouda et al. 2003). Thoracic myelopathy patients were younger at operation than those with cervical myelopathy. The patients in the latter group were more frequently in their sixties to seventies (Tanaka et al. 2003). It is unclear why thoracic myelopathy more often develops in middle-aged people compared to cervical myelopathy patients. OLF and OPLL, the major causes of thoracic myelopathy, might be associated with some genetic factors (Koga et al. 1998; Yamamoto et al. 2002). The thoracic spine is naturally kyphotic and the spinal cord runs anterior of the spinal canal, which suggests the cord is more easily damaged from the anterior side. Additionally, the spinal cord in the thoracic spine has a particularly vulnerable region called the “watershed zone” due to poor blood supply and the ratio of the cord to the canal is larger than in other parts of the spine (Stillerman and Weiss 1991). Since the cord is debilitated more easily by compressive spinal factors, thoracic myelopathy might develop earlier than in the cases of cervical myelopathy.

The symptomatology of thoracic myelopathy is similar to that of lumbar disorders (Mitra et al. 1996; Sato et al. 1997a). It usually appears first in the lower extremities (Sato et al. 1997a). In the current study, more than one half of the patients initially noticed tingling, numbness or pain in the lower legs. Interestingly enough, 5% of the patients complained of back pain, which was in contrast to patients with cervical myelopathy who rarely presented with neck pain (Smith and Godersky 1987; Bernhardt et al. 1993; Kokubun et al. 1996; Mitra et al. 1996; Sato et al. 1997a). Thoracic myelopathy usually progresses slowly (Shiokawa et al. 2001; Fong and Wong 2004). In the present study, the preoperative duration from the initial onset of symptoms until surgery was also relatively long, 2 years on average, which might result from by this slow progression and the difficulty of the diagnosis because of similarities with lumbar disorders. However, the fact that about one third of the patients showed preoperative durations of less than 6 months suggests that thoracic myelopathy sometimes progresses rapidly (Otani 1988; Shiokawa et al. 2001; Fong and Wong 2004). Careful observation is necessary in order to avoid deterioration of the myelopathy.

OLF is the most common compressive factor contributing to thoracic myelopathy in the Japanese (Yonenobu et al. 1987; Sato et al. 1997a), and was responsible for 60% of all thoracic cord compression diseases in this series, either alone or in combination with OPLL. As for the location of the ossification of spinal ligaments, OLF and OPLL showed contrasting appearances. OLF is hardly found in the cervical spine (Kokubun et al. 1996). On the other hand, OPLL is one of the most frequent compressive factors of cervical myelopathy and was found in 20% of such patients (Kokubun et al. 1996). OPLL was found mostly in the upper to middle thoracic spine, whereas OLF was mostly in the lower thoracic in the current study as previously described (Yonenobu et al. 1987; Sato et al. 1997a). For the development of OLF, mechanical stress also plays an important role. Higher mechanical forces cause more pronounced degenerative changes of the facet joints and intervertebral discs at the
thoracolumbar junction, which might lead to further degeneration of the ligamentum flavum at the lower thoracic spine (Payer et al. 2000).

Laminectomy is the standard procedure for OLF. From our experience, however, some technical modifications have been required based on the conditions of OLF, depending on whether bilateral OLFS are fused at the middle of the spinal canal (Sato et al. 1998; Sato 2003). Fenestration or French-door laminectomy was performed for the non-fused OLF as longitudinal resection of the lamina and ligamentum flavum at the middle of the spinal canal is easy. Since it is difficult to cut the fused OLF at the middle, en bloc laminectomy was selected in such cases (Sato et al. 1998; Sato 2003). Based on this surgical strategy, the number of surgical procedures for OLF myelopathy changed. About 10% of the OLF patients in this study had the ossification of the dura mater. The OLF needs to be removed together with the ossified dura while keeping the arachnoid intact, and subsequent dural repair is required in order to avoid cerebrospinal fluid leak (Sato 2003; Fong and Wong 2004).

There are still many unsolved problems in the treatment of thoracic OPLL. In some patients, laminectomy alone does not sufficiently decompress the spinal cord impinged from the front by thick or beak-like OPLL in the thoracic spine since the spinal curvature is kyphotic and the spinal cord does not shift backward enough by it (Yonenobu et al. 1987). Several surgical procedures have been developed in order to ensure sufficient decompression for OPLL, and these had also been performed in this series (Ohetsuka et al. 1983; Kokubun et al. 1991). But the indications for each procedure, the range of decompression and the necessity for combined spinal fusion have not been clarified completely.

Thoracic myelopathy is not common and not well recognized by orthopedists or even by spinal surgeons, particularly outside Japan (Yonenobu et al. 1987). Not only orthopaedists but also general physicians should know this entity since a delay in diagnosis and subsequent treatment might result in severe gait and urinary disturbance, which can markedly restrict the activities of daily life. In addition, spinal surgeons should better recognize the clinical features and neurodiagnostic findings of thoracic myelopathy that indicate the need for surgical intervention.

References


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