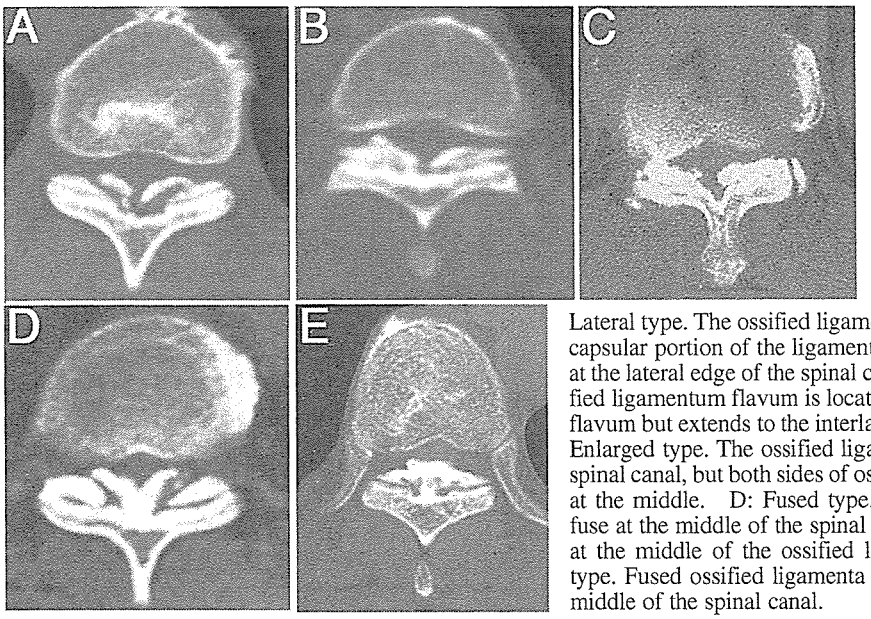

FIG. 2. The CT classification of OLF, with scans obtained at the middle of the facet joint. A: Lateral type. The ossified ligamentum flavum is located only in the capsular portion of the ligamentum flavum, which can be detected at the lateral edge of the spinal canal. B: Extended type. The ossified ligamentum flavum is located at the surface of the ligamentum flavum but extends to the interlaminar portion of the ligament. C: Enlarged type. The ossified ligamentum flavum protrudes into the spinal canal, but both sides of ossified ligamenta flava are not fused at the middle. D: Fused type. Bilateral ossified ligamenta flava fuse at the middle of the spinal canal, but an incision can be found at the middle of the ossified ligamentum flavum. E: Tuberous type. Fused ossified ligamenta flava make a tuberous mass at the middle of the spinal canal.

Several previous studies on OLF myelopathy involved fewer than 50 patients, and thus OLF myelopathy's clinical features remain unclear.[1,3,8,9,12,14,16,18,22,24] In the present study, we collected data in 72 cases, which represents the largest population reported to date. Most reports of OLF myelopathy have originated from Japan, and this fact may suggest that the number of patients might be fewer outside Japan.[3,15–18,24] Recently, however, several investigators have reported on Caucasian, Indian, North African, and Chinese patients.[1,3,9,12,14,22]

The reason for the high incidence of OLF in the Japanese population is not clear. Authors of several recent studies have indicated that the development of OPLL is associated with certain genetic factors.[6,10,20,23] These factors may also play a specific role in the origin of OLF.

In the present study we found that OLF-induced myelopathy frequently developed in the lower thoracic region in elderly males. The authors of previous studies have also indicated that OLF-related myelopathy most commonly occurred in the lower one third of the thoracic spine.[13,19] The symptoms of OLF-induced myelopathy mimic those of lumbar disorders, resulting in misdiagnosis.[12,16,24] Half of our patients first noticed lower-extremity tingling and numbness or pain; these symptoms can be the chief complaints among patients with lumbar disorders. Interestingly, 11% of the patients complained of back pain, which is in contrast to patients with cervical myelopathy who rarely experienced neck pain first.[2,4,7] To establish a correct diagnosis of OLF-related myelopathy, a detailed neurological examination should be performed. In addition, a lower thoracic region magnetic resonance imaging study should be conducted once OLF-related myelopathy is suspected due to spasticity or multisegmental neurological deficits in the lower extremities.

Because OLF-related myelopathy affects the posterior part of the spinal canal, a laminectomy is indicated. Some technical modifications, however, have been developed during the 15 years of the study, based on the conditions of OLF.[15,17] The ligamentum flavum bilaterally has two portions: medially, the interlaminar portion and, laterally, the capsular portion.[22] Ossification usually begins in the capsular portion and spreads to the laminar portion. Ossification enlarges anteriorly toward the spinal cord. Bilateral ossifications then fuse in the middle of the lamina and thicken to form a central tuberous mass.[15,22] These fused or

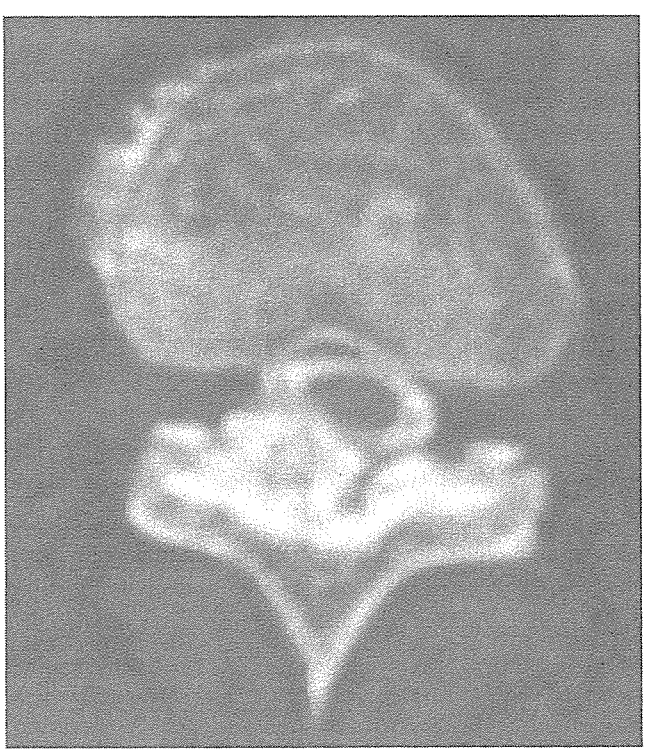

FIG. 3. Preoperative CT myelogram obtained in a patient with OLF and ossified dura mater at the T10–11 level. The ossified ligamentum flavum adheres to the ossified dura on the right side of the spinal cord, and little contrast medium between the cord and ossified ligamentum flavum is detected.

TABLE 3

Relationship of postoperative JOA score and recovery rate to various patient factors

Factor	Postop Variable (range)	
	JOA Score	Recovery Rate (%)
sex		
male	7.7 (0–11)	46 (−38 to 100)
female	8.3 (4–10)	54 (0 to 88)
age (yrs)		
≤65	8.0 (0-11)	46 (−38 to 100)
>65	7.7 (2-11)	48 (0 to 100)
sex & age		
male ≤65	8.0 (0–11)	47 (−38 to 100)
male >65	7.3 (2–11)	42 (0 to 100)
female ≤65	8.0 (6–9)	43 (0 to 60)
female >65	8.4 (4–10)	60 (22 to 88)
preop duration of symptoms (mos)		
≤6	8.0 (4–11)	54 (−25 to 100)
6–12	8.3 (5–11)	47 (0 to 100)
12–24	7.6 (0–11)	43 (−38 to 100)
≥24	7.7 (4–10)	41 (−25 to 75)
ossification of dura mater		
present	6.5 (0–11)	30 (−38 to 100)
absent	8.0 (4–11)	49 (−25 to 100)
preop severity of myelopathy*		
mild (JOA score ≥7)	9.0 (6–11)	45 (−38 to 100)
moderate (JOA score 4–6)	8.2 (4–11)	51 (0 to 100)
severe (JOA score ≤3)	5.9 (0–11)	41 (−25 to 100)

* A statistically significant difference ($p < 0.05$) was detected only in the relationship between the JOA score and the preoperative severity of myelopathy.

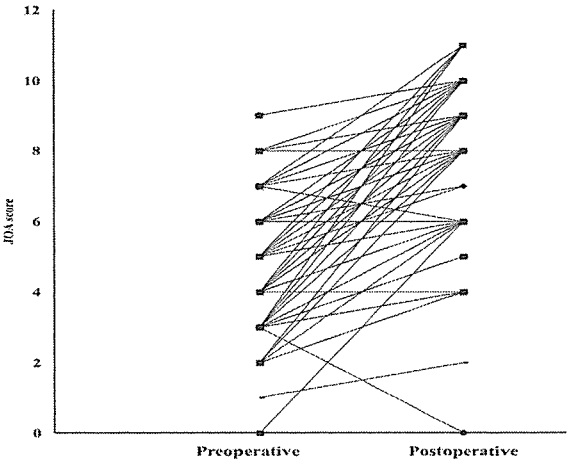

FIG. 4. Line graph showing the pre- and postoperative JOA scores obtained in 72 patients. The score improved in 66 patients during a mean follow-up period of 46 months.

central tuberous types of OLF frequently adhere to the dura mater or fuse with its ossification.[3,8,15,17,22] The current procedure of choice involves fenestration involving all ligamenta flava for uni- or bilateral OLF at a single level without fusion in the middle. Fenestration, or French-door laminectomy, is performed for nonfused-type OLF at two levels or more. En bloc laminectomy is chosen to treat the fused or central tuberous types of OLF.[15,17]

Several peri- and postoperative complications have been reported in patients with OLF-induced myelopathy.[3,9,22] Cerebrospinal fluid leakage followed by the disruption of dura is one of the major intraoperative complications. In this series, dural tears occurred in nine patients and most of them showed the ossified dura. In such cases, the ossified ligamentum flavum needs to be excised together with the ossified dura, keeping the arachnoid intact to avoid iatrogenic spinal cord damage.[3,15,17,22] Subsequently, duraplasty is required, usually with the placement of artificial dura.[3,15,17,22] Neurological deterioration can occur immediately after surgery because of unintended intraoperative spinal cord manipulation.[9] A postoperative epidural hematoma can also cause dense paralysis, which was seen in one of our cases. Increased kyphotic spinal deformity after laminectomy can cause late-onset neurological deterioration or localized back pain.[3]

Myelopathy caused by OLF is generally believed to progress slowly.[18,24] In the present study, the mean preoperative duration of initial symptoms was nearly 2 years. Forty percent of the patients in whom the preoperative duration of symptoms was shorter than 6 months, however, had severe myelopathy, which suggests that in some cases, myelopathy progresses rapidly. Surgical decompression has been the treatment of choice for compressive myelopathy, and the results of this study indicate that outcomes after decompression for OLF-induced myelopathy are stable.[11] The postoperative neurological condition depended on the preoperative severity of myelopathy; patients with a shorter preoperative duration of symptoms tended to fare better than those in whom the duration of myelopathy was longer. Thus, patients who present early in the course of OLF with fewer disabilities should undergo surgery quickly to avoid deterioration of myelopathy and poorer results, which are highly possible if surgery is delayed.

Conclusions

Myelopathy caused by OLF is uncommon, particularly outside Japan. We have presented the clinical features and surgical results obtained in 72 patients with this disorder; this is the largest population reported to date. The surgical outcome was relatively good and depended on the severity of myelopathy; thus, early and correct diagnosis is required to avoid poorer results. Further education and study of OLF-induced myelopathy are necessary, not only for spine surgeons in Japan but also for neurosurgeons all over the world.

References

1. Ben Hamouda K, Jemel H, Haouet S, Khaldi M: Thoracic myelopathy caused by ossification of the ligamentum flavum: a report of 18 cases. **J Neurosurg 99 (2 Suppl):**157–161, 2003
2. Bernhardt M, Hynes RA, Blume HW, White AA III: Cervical spondylotic myelopathy. **J Bone Joint Surg Am 75:**119–128, 1993
3. Fong SY, Wong HK: Thoracic myelopathy secondary to ligamentum flavum ossification. **Ann Acad Med Singapore 33:** 340–346, 2004
4. Hattori S: [Cervical myelopathy.] **J Jpn Orthop Assoc 52:** 581–593, 1978 (Jpn)
5. Japanese Orthopaedic Association: [Scoring system for cervical myelopathy.] **J Jpn Orthop Assoc 68:**490–503, 1994 (Jpn)

6. Koga H, Sakou T, Taketomi E, Hayashi K, Numasawa T, Harata S, et al: Genetic mapping of ossification of the posterior longitudinal ligament of the spine. **Am J Hum Genet 62:** 1460–1467, 1998
7. Kokubun S, Sato T, Ishii Y, Tanaka Y: Cervical myelopathy in the Japanese. **Clin Orthop Relat Res 323:**129–138, 1996
8. Li KK, Chung OM, Chang YP, So YC: Myelopathy caused by ossification of ligamentum flavum. **Spine 27:**E308–E312, 2002
9. Liao CC, Chen TY, Jung SM, Chen LR: Surgical experience with symptomatic thoracic ossification of the ligamentum flavum. **J Neurosurg Spine 2:**34–39, 2005
10. Maeda S, Koga H, Matsunaga S, Numasawa T, Ikari K, Furushima K, et al: Gender-specific haplotype association of collagen α2 gene in ossification of the posterior longitudinal ligament of the spine. **J Hum Genet 46:**1–4, 2001
11. Matsumoto M, Chiba K, Ishikawa M, Maruiwa H, Fujimura Y, Toyama Y: Relationships between outcomes of conservative treatment and magnetic resonance imaging findings in patients with mild cervical myelopathy caused by soft disc herniations. **Spine 26:**1592–1598, 2001
12. Mitra SR, Gurjar SG, Mitra KR: Degenerative disease of the thoracic spine in central India. **Spinal Cord 34:**333–337, 1996
13. Okada K, Oka S, Tohge K, Ono K, Yonenobu K, Hosoya T: Thoracic myelopathy caused by ossification of the ligamentum flavum. Clinicopathologic study and surgical treatment. **Spine 16:**280–287, 1991
14. Payer M, Bruder E, Fischer JA, Benini A: Thoracic myelopathy due to enlarged ossified yellow ligaments. **J Neurosurg 92 (1 Suppl):**105–108, 2000
15. Sato T, Kokubun S, Ishii Y: [Choice of operative method for ossification of ligamentum flavum based on CT findings.] **Rinsho Seikeigeka 31:**541–545, 1996 (Jpn)
16. Sato T, Kokubun S, Tanaka Y, Ishii Y: Thoracic myelopathy in the Japanese: epidemiological and clinical observations on the cases in Miyagi Prefecture. **Tohoku J Exp Med 184:**1–11, 1998
17. Sato T, Tanaka Y, Aizawa T, Koizumi Y, Kokubun S: [Surgical treatment for ossification of ligamentum flavum in the thoracic spine and its complications.] **Spine Spinal Cord 11:**505–510, 1998 (Jpn)
18. Shiokawa K, Hanakita J, Suwa H, Saiki M, Oda M, Kajiwara M: Clinical analysis and prognostic study of ossified ligamentum flavum of the thoracic spine. **J Neurosurg 94 (2 Suppl):** 221–226, 2001
19. Smith DE, Godersky JC: Thoracic spondylosis: an unusual cause of myelopathy. **Neurosurgery 20:**589–593, 1987
20. Tahara M, Aiba A, Yamazaki M, Ikeda Y, Goto S, Moriya H, et al: The extent of ossification of the posterior longitudinal ligament of the spine associated with nucleotide pyrophosphatase gene and leptin receptor gene polymorphisms. **Spine 30:** 877–881, 2005
21. Tanaka Y, Kokubun S, Sato T, Ishii Y: [Changes on spine and spinal cord lesions in frequencies of their surgeries; an observation based on the registered cases for 14 years.] **Orthop Surg Traumatol 46:**391–398, 2003 (Jpn)
22. Trivedi P, Behari S, Paul L, Banerji D, Jain VK, Chhabra DK: Thoracic myelopathy secondary to ossified ligamentum flavum. **Acta Neurochir (Wien) 143:**775–782, 2001
23. Yamamoto Y, Furukawa K, Ueyama K, Nakanishi T, Takigawa M, Harata S: Possible roles of CTGF/Hcs24 in the initiation and development of ossification of the posterior longitudinal ligament. **Spine 27:**1852–1857, 2002
24. Yonenobu K, Ebara S, Fujiwara K, Yamashita K, Ono K, Yamamoto T, et al: Thoracic myelopathy secondary to ossification of the spinal ligament. **J Neurosurg 66:**511–518, 1987

Manuscript received April 7, 2006.
Accepted in final form August 28, 2006.
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Tohoku J. Exp. Med., 2006, **210**, 199-208

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

Received July 27, 2006; revision accepted for publication August 30, 2006.
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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 the ligamentum flavum (OLF), 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

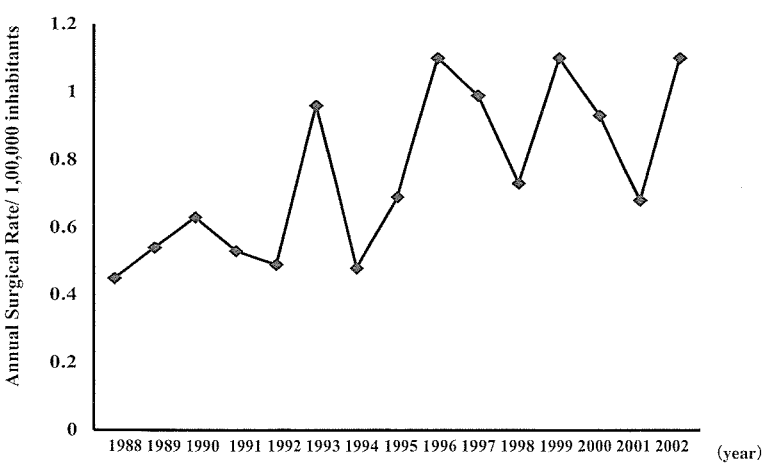


Fig. 1. Yearly changes in the surgical rate for patients with thoracic myelopathy in Miyagi Prefecture from 1988 to 2002.

TABLE 1. Summary of the patients with thoracic myelopathy.

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

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. $^{*}p < 0.05$.

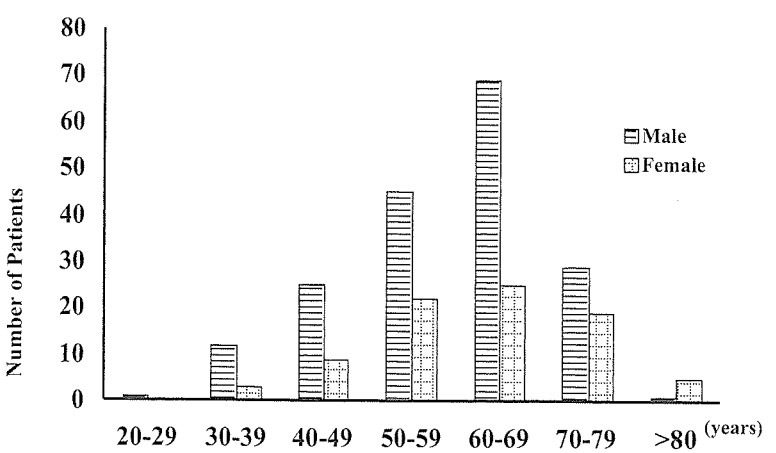


Fig. 2. Age distribution of the patients with thoracic myelopathy in Miyagi Prefecture. Among males, the patients in their sixties show the highest prevalence, and among females, those in the fifties, sixties and seventies show almost equal prevalences.

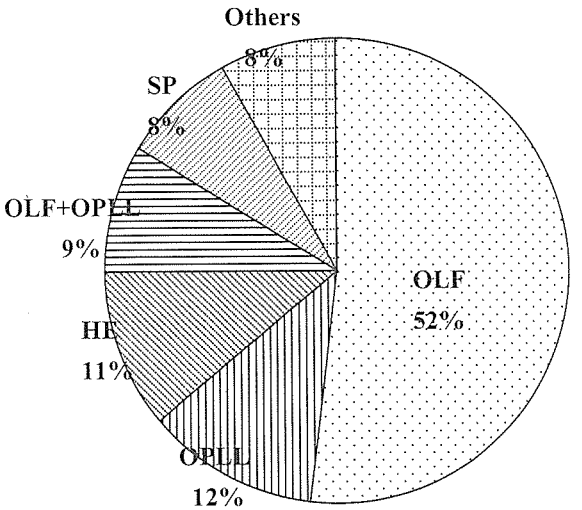


Fig. 3. Spinal factors compressing the spinal cord leading to thoracic myelopathy.
OLF, ossification of the ligamentum flavum; OPLL, ossification of the posterior longitudinal ligament; HE, intervertebral disc herniation; SP, posterior spur.

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

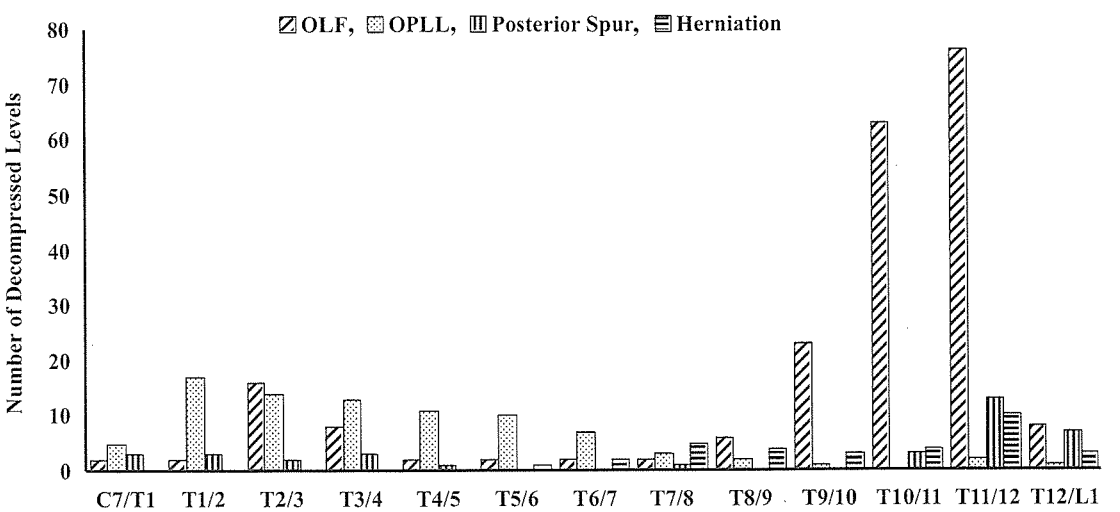


Fig. 4. Distribution of the four major compressive factors in the spinal cord in relation to the intervertebral disc level.
OLF is most common in the lower thoracic spine while OPLL is frequently distributed in the upper to middle thoracic. Intervertebral disc herniation is mainly detected in the middle and lower thoracic spine and posterior spur in the lower thoracic spine.

TABLE 2. Surgical procedures for the common compressing spinal factors in thoracic myelopathy.

OLF	139 patients
Laminectomy	78
Fenestration	45
Laminectomy + fenestration	9
Hemilaminectomy	3
Others	4
OPLL	33 patients
Laminectomy	24
Anterior decompression through posterior approach "Otsuka"	2
Anterior decompression & spinal fusion	2
Anterior decompression through a diagonal anterior and posterior approach	2
Others	3
OLF + OPLL	25 patients
Laminectomy	18
Laminoplasty + laminectomy	3
Fenestration	2
Anterior decompression through posterior approach "Otsuka"	2
Disc herniation	30 patients
Anterior decompression & spinal fusion	13
Hemilaminectomy	6
Transverso-arthro-pediculectomy	6
Others	5
Posterior spur	22 patients
Anterior decompression & spinal fusion	12
Laminectomy	6
Others	4

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

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).

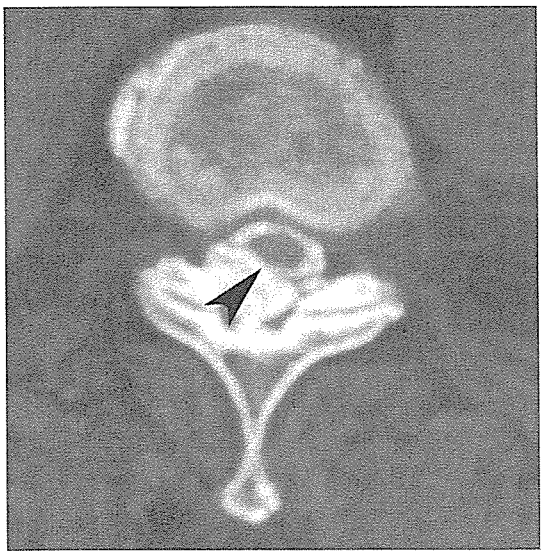
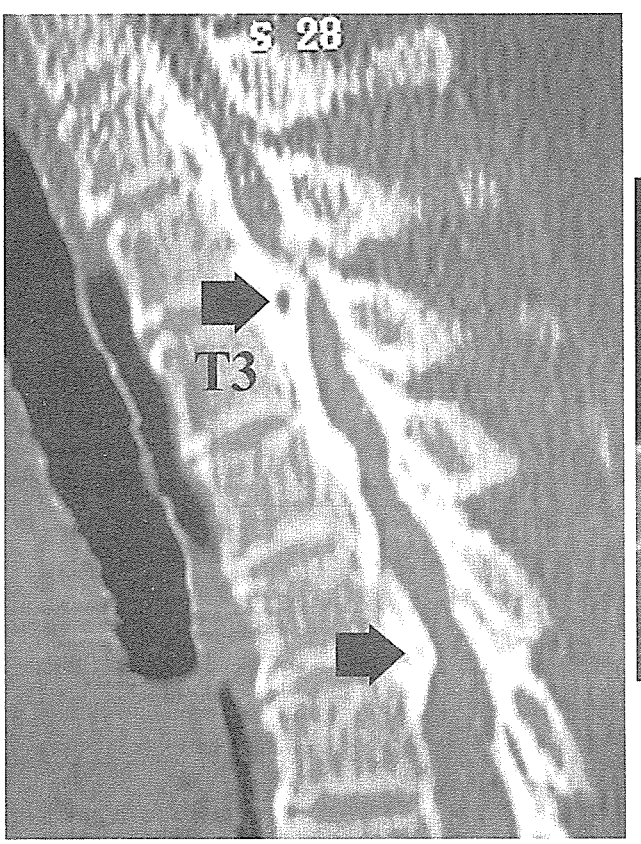


Fig. 5. Preoperative CT-myelogram of OLF with ossified dura mater at T10/11.
OLF protrudes right-ventrally and the contrast medium between the spinal cord and OLF can not be detected (arrowhead).

A



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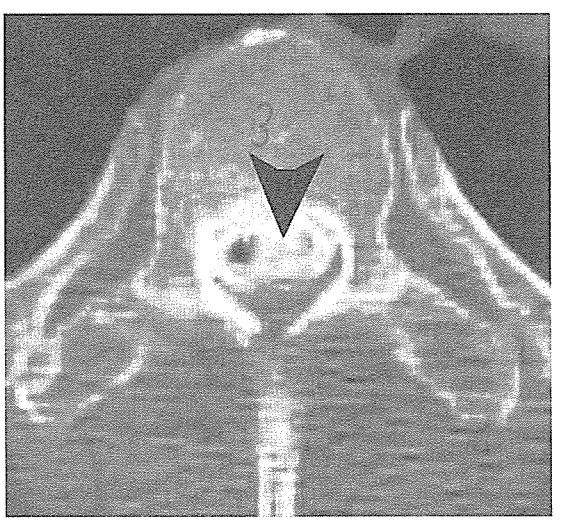

Fig. 6. Preoperative CT of a patient with OPLL.
A: Sagittal plane. Continuous OPLL can be found in the upper thoracic spine including beak-like regions (arrows).
B: Axial plane. Thick OPLL occupies the spinal canal at T3 level (arrowhead).

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 (Ohtsuka 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

Aizawa, T., Sato, T., Tanaka, Y., Kotajima, S., Sekiya, M. & Kokubun, S. (2001) Idiopathic herniation of the thoracic spinal cord. *Spine,* **26**, E488-E491.

Bernhardt, M., Hynes, R.A., Blume, H.W. & White, A.A., III (1993) Current concepts review: cervical spondylotic myelopathy. *J. Bone Joint Surg. Am.,* **75**, 119-128.

Fong, S.Y. & Wong, H.K. (2004) Thoracic myelopathy secondary to ligamentum flavum ossification. *Annals. Acad. Med.,* **33**, 340-346.

Hamouda, K.B., Jemel, H. & Khaldi, M. (2003) Thoracic myelopathy caused by ossification of the ligamentum flavum: a report of 18 cases. *J. Neurosurg. (Spine 2),* **99**, 157-161.

Koga, H., Sakou, T., Taketomi, E., Hayashi, K., Numasawa, T., Harata, S., Yone, K., Matsunaga, S., Otterud, B., Inoue, I. & Leppert, M. (1998) Genetic mapping of ossification of the posterior longitudinal ligament of the spine. *Am. J. Hum. Genet.,* **62**, 1460-1467.

Kokubun, S., Kashimoto, O., Ozawa, H., Hashimoto, Y., Kikawa, T., Gomibuchi, S. & Sakurai, M. (1991) Anterior decompression in the thoracic spine through a diagonal antero-posterior approach. *Tohoku Archives Orthop. Surg. Traumatol.,* **35**, 71-74. (in Japanese)

Kokubun, S., Sato, S., Ishii, Y. & Tanaka, Y. (1996) Cervical myelopathy in the Japanese. *Clin. Orthop.,* **323**, 129-138.

Mitra, S.R., Gurjar, S.G. & Mitra, K.R. (1996) Degenerative disease of the thoracic spine in central India. *Spinal Cord.,* **34**, 333-337.

Otani, K., Yoshida, M., Fujii, E., Nakai, S. & Shibasaki, K. (1988) Thoracic disc herniation-surgical treatment in 23 patients-. *Spine,* **13**, 1262-1267.

Otsuka, K., Terayama, K., Tsuchiya, T., Wada, K., Furukawa, K. & Okubo, M. (1983) A surgical procedure of the anterior decompression of the thoracic spinal cord through the posterior approach. *Orthop. Surg. Traumatol.,* **26**, 1083-1090. (in Japanese)

Payer, M., Bruder, E., Fischer, J.A. & Benini, A. (2000) Thoracic myelopathy due to enlarged ossified yellow ligaments. *J. Neurosurg. (Spine 1),* **92**, 105-108.

Smith, D.E. & Godersky, J.C. (1987) Thoracic spondylosis: an unusual cause of myelopathy. *Neurosurgery,* **20**, 589-593.

Sato, T., Kokubun, S., Tanaka, Y. & Ishii, Y. (1997a) Thoracic myelopathy in the Japanese: epidemiological and clinical observations on the cases in Miyagi Prefecture. *Tohoku J. Exp. Med.,* **184**, 1-11.

Sato, T., Kokubun, S., Tanaka, Y. & Aizawa, T. (1997b) Paraparesis associated with mild congenital kyphoscoliosis in an adult. *Tohoku J. Exp. Med.,* **183**, 303-308.

Sato, T., Tanaka, Y., Aizawa, T., Koizumi, Y.& Kokubun, S. (1998) Surgical treatment for ossification of ligamentum flavum in the thoracic spine and its complications. *Spine Spinal Cord,* **11**, 505-510. (in Japanese)

Sato, T. (2003) Operative method for thoracic myelopathy based on each spinal pathological factor. *Orthop. Surg. Traumatol.,* **46**, 523-531. (in Japanese)

Shiokawa, K., Hanakita, J., Suwa, H., Saiki, M., Oda, M. &

Kajiwara, M. (2001) Clinical analysis and prognostic study of ossified ligamentum flavum of the thoracic spine. *J. Neurosurg. (Spine 2)*, **94**, 221-226.

Stillerman, C.B. & Weiss, M.H. (1991) Management of thoracic disc disease. In: *Proceeding of the Congress of Neurological Surgeons. Los Angeles, California 1990. Clin. Neurosurg. vol 38*, edited by W. Selman, Williams & Wilkins, Baltimore, pp. 325-352.

Tanaka, Y., Kokubun, S., Sato, T. & Ishii, Y. (2003) Changes on spine and spinal cord lesions in frequencies of their surgeries; an observation based on the registered cases for 14 years. *Orthop. Surg. Traumatol.*, **46**, 391-398. (in Japanese)

Yonenobu, K., Ebara, S., Fujiwara, K., Yamashita, K., Ono, K., Yamamoto, T., Harada, N., Ogino, H. & Ojima, S. (1987) Thoracic myelopathy secondary to ossification of the spinal ligament. *J. Neurosurg.*, **66**, 511-518.

Yamamoto, Y., Furukawa, K., Ueyama, K., Nakanishi, T., Takigawa, M. & Harata, S. (2002) Possible roles of CTGF/Hcs24 in the initiation and development of ossification of the posterior longitudinal ligament. *Spine*, **27**, 1852-1857.

J Neurosurg Spine 5:398–403, 2006

Facet cyst in the lumbar spine: radiological and histopathological findings and possible pathogenesis

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Object. The authors define facet cyst as a cyst located beside the facet joint and exhibiting a communication with the joint, as demonstrated on arthrography and subsequent computed tomography (CT) of the joint space. The purpose of this study was to determine the pathogenesis of facet cysts based on their radiological and histological features.

Methods. Forty-six juxtafacet cysts in 45 patients (26 men and 19 women, age range 41–82 years) were surgically treated after evaluation by arthrography and subsequent CT scanning. A communication channel between the cyst and the joint was confirmed in all cases and thus the lesions were designated facet cysts. In almost all patients the involved facet joint showed moderate to severe degeneration. After a thorough preoperative radiological evaluation, these cysts were excised en bloc by medial facetectomy with the entire ligamentum flavum. The specimens were cut axially at the maximum diameter and were histologically investigated.

Morphologically, the cysts exhibited three shapes, appearing as: 1) a small protrusion, 2) a semicircular cyst, or 3) a round cyst. The cyst walls consisted of elastic and collagen fibers undergoing fibrinoid degeneration, but no synovial lining cells were detected. The cystic cavities were regularly filled with fibrinoids, and myxoid degeneration was found particularly in the larger cysts.

Conclusions. Facet cysts are closely related to the degeneration of the neighboring facet joint. Analysis of the findings in this histological study suggests that there are several shapes of facet cysts. The authors propose that the shape could depend on the stage of the cyst's development.

KEY WORDS • **synovial cyst** • **ganglion cyst** • **facet joint** • **lumbar spine** • **histopathological study**

ADVANCES in MR imaging have made it possible to easily diagnose an intraspinal cystic lesion beside the facet joint of the lumbar spine. Many hospitals have MR imaging units and the numbers of reports in which such cysts have been described is increasing.[3,5,13,14,21,25] Several terms for these cysts, including synovial cyst,[1,4,7,8,11,12,14,15] ganglion cyst,[6,10,12] and pseudocystic lesion,[25] have been used. Synovial cysts and ganglion cysts can be distinguished only by their histological findings, which indicate whether they have synovial lining cells.[9–12] In 1974, Kao, et al.,[11] proposed the term "juxtafacet cyst" to represent both synovial and ganglion cysts. Even thereafter, however, the term "intraspinal synovial cyst" was more frequently used in the literature, on occasion despite the histological findings.[3,8,14,15,21] In 1995, Hsu, et al.,[9] first used the term "intraspinal facet cyst" to designate cysts associated with the facet joints regardless of whether synovial lining cells were present, because there were cysts that exhibited the histological features of both synovial and ganglion cysts. Different terms may indicate the same disorders, and several authors have suggested that synovial and nonsynovial cysts are not entirely separate entities.[9,11,12,13] Such confusion in the terminology is a fundamental cause of the difficulty in understanding this entity.

Many researchers have suggested that the formation of the juxtafacet cyst is closely related to ligamentum flavum degeneration and also to facet joint degeneration.[3,5,9,12,14,25] A communicating channel between the cyst and the facet joint could exist and play a developmental role. In several studies, investigators have confirmed the communication between the cyst and the joint using facet joint arthrography,[4,7,9] but this modality has not been applied in all cases.[9,12,14] In addition, there have been only few reports in which this communication was confirmed intraoperatively or histopathologically.[6,12,22] The cyst–joint communication must be proved radiologically or histologically to clarify the pathogenesis of juxtafacet cysts. Therefore, in this study, we have defined facet cysts as those communicating with the neighboring facet joint, as demonstrated on arthrography and subsequent CT scanning, although there might be other types of cysts in addition to those in the facet joints.

Abbreviations used in this paper: CT = computed tomography; MR = magnetic resonance.

In the present study, we performed facet joint arthrography and CT in all patients with juxtafacet cysts. In addition, we performed a medial facetectomy, removed the cysts en bloc, and investigated tissue samples microscopically. Here, we present the radiological and histological findings of these cysts in detail and discuss their pathogenesis.

Clinical Material and Methods

Patient Population

Between January 1992 and December 2005, we established MR imaging–based diagnoses of 75 lumbar juxtafacet cysts in a consecutive series of 72 patients at the Tohoku Rosai Hospital, Sendai, Japan. All patients underwent facet joint arthrography and subsequent CT scanning, and communication with the neighboring facet joint was confirmed in all cases (Fig. 1). Thus, all the cysts in the present study are described as "facet cysts." There were 43 male and 29 female patients, whose mean age at the initial visit to the hospital was 65 years (range 15–86 years). Lumbar radiculopathy was demonstrated in 50 patients and cauda equina syndrome in 22. Initially, the patients received conservative treatment involving the injection of a steroid agent into the affected facet joints as well as medical therapy,[8,13,15] and approximately one third of the patients experienced symptomatic improvement.

The remaining 46 lumbar facet cysts in 45 patients were surgically treated. These 45 cases form the basis of this study. There were 26 men and 19 women whose mean age at surgery was 66 years (range 41–82 years). Of the 46 cysts, three (6%) were located at L2–3, 10 (22%) at L3–4, 29 (63%) at L4–5, and four (9%) at L5–S1. We first determined the duration between the onset of symptoms and treatment. Thereafter, patients were evaluated radiologically.

Radiological Examination

Plain lateral radiographs were reviewed to assess whether lumbar degenerative spondylolisthesis was present, and if so it was divided into four quarters according to the Meyerding classification.[16] The degenerative changes of the involved facet joints were evaluated on CT scans and graded as follows: Grade 0, normal; Grade 1, joint space narrowing; Grade 2, joint space narrowing with osseous sclerosis or hypertrophy; and Grade 3, severe degeneration with joint space narrowing, sclerosis, and osteophytes.[20]

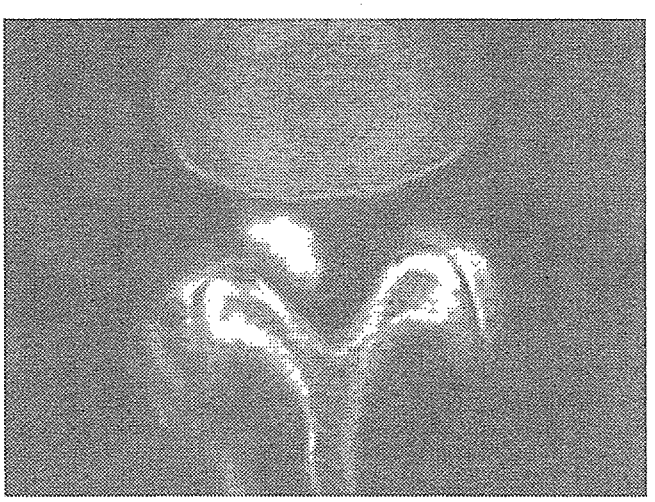

FIG. 1. Axial CT scan obtained after facet joint arthrography. The contrast medium clearly helps to delineate the communication between the juxtafacet cyst and neighboring facet joint.

Surgical Procedure

At surgery, the facet cyst, usually adherent to the dura mater, was resected en bloc with all of the ligamentum flavum through medial facetectomy. This technique is considered not to affect lumbar stability.[2] The specimens were immediately fixed in 10% neutral formalin, decalcified using ethylenediaminetetraacetic acid, cut axially at the cyst's maximum diameter, and embedded in paraffin. Both H & E and elastica–Masson trichrome staining techniques were performed.

Histopathological Investigation

Morphological characteristics of the cysts were investigated using a low-magnification microscope. Histopathological features were examined under a high-magnification microscope to characterize the walls and contents of the cysts. The findings of fibrinoid and myxoid degeneration, granulation, and hemorrhage were graded as follows: severe (findings were abundant); moderate (some present); mild (minimal); and negative (absent). All findings were reviewed by two of the authors (T.K., F.K.). The interobserver reliability of the findings was tested using the Cohen kappa coefficient and found to be excellent—93% agreement between the two observers (= 0.907).

Study Approval

This study was approved by the ethics committee of Tohoku Rosai Hospital, and all patients provided detailed informed consent before being treated.

Statistical Analysis

The relationships between the histopathological and clinical and radiological findings were analyzed. For statistical analysis, the Tukey–Kramer and Kruskal–Wallis tests were conducted. Probability values less than 0.05 were considered statistically significant.

Results

The mean duration between onset of symptoms and surgical treatment was 13 months (range 4 weeks–7 years). After surgery, symptoms and neurological deficits improved in all patients. Nineteen of 45 patients had Grade 1 degenerative spondylolisthesis at the involved level.[16] Computed tomography demonstrated degenerative changes in all the involved facet joints: joint space narrowing, sclerosis of the subchondral bone, osseous cyst formation, and bone spur formation. Based on the CT findings,[20] we determined that one joint (2%) exhibited Grade 1 features, 12 joints (26%) Grade 2 features, and 33 joints (72%) Grade 3 features. In almost all patients we observed moderate to severe facet joint degeneration.

At low magnification, a communicating channel be-

tween the cyst and the neighboring facet joint was clearly detected in one section in three cases. Based on their shapes, the facet cysts were divided into three types: those with a small protrusion from the ligamentum flavum, those exhibiting a semicircular shape, and those showing a round shape like a papilla. Examination of the small protrusions revealed collagen rather than elastic fibers inside the cyst walls, with either a fissure or an obvious cavity. Inspection of the semicircular and round cysts also showed two types, with thick or thin cyst walls. According to these findings, we classified the facet cysts into four types: Type 1 was characterized by a small protrusion with a fissure (Fig. 2A); Type 2, by a small protrusion with an obvious cavity (Fig. 2B); Type 3, by a semicircular cyst; and Type 4, by a round cyst. Types 3 and 4 showed two subtypes, with thick cyst walls (Type 3a or 4a) or thin cyst walls (Type 3b or 4b) (Fig. 2C–F). Six cysts were classified as Type 1, eight as Type 2, five as Type 3a, 13 as Type 3b, and seven each as Types 4a and 4b.

The ligamentum flavum showed degeneration with irregularly arranged and disrupted elastic fibers, irregular collagenous scarring, an increasing number of fibroblasts, small-vessel proliferation, chondroid metaplasia, and occasional calcium deposition and hemorrhage. Synovial lining cells were not noted in any of the cases (Fig. 3). The histological findings of the cyst walls and the contents of the cysts are summarized in Table 1. The cyst walls consisted of elastic and collagen fibers with fibroblasts. The fibrinoid degeneration of Type 1 was mild. As the cysts became larger, more frequent and severe fibrinoid degeneration was observed, and the ratio of the involved elastic fibers became smaller. Types 2, 3, and 4a cysts also had granulation and vessel proliferation in the cyst walls, particularly in the base of the cysts. Cavities were usually filled with fibrinoids. Hemorrhage was occasionally found in all types of the facet cysts but was most common in Types 3 and 4 cysts. Many Types 3b and 4b cysts exhibited myxoid degeneration in the cavity.

The relationships between the type of cyst and the preoperative duration of symptoms, and lumbar degeneration are presented in Table 2. Twelve patients underwent surgery within 3 months of symptom onset. The preoperative symptom onset–treatment interval of patients with Type 3b cysts was the smallest, but there was no statistically significant difference between the types. No relationship was found between the type of cyst and the severity of the spondylolisthesis at the involved level. Larger cysts, however, tended to be associated with more severely degenerated facet joints. More than three fourths of Types 3 and 4 cysts showed Grade 3 facet joint degeneration.

Discussion

The varying terms ascribed to intraspinal cysts that lie near the facet joints have created confusion in the past. The phrases synovial cyst, ganglion cyst, and pseudocyst have been used;[9–12,25] however, certain cysts exhibit characteristics of each type.[9] Whether a cyst has synovial lining cells might not be clinically important.[3,9,12,13,22] In the present report, we defined facet cyst as a cyst located beside the facet joint and in communication with the neighboring fac-

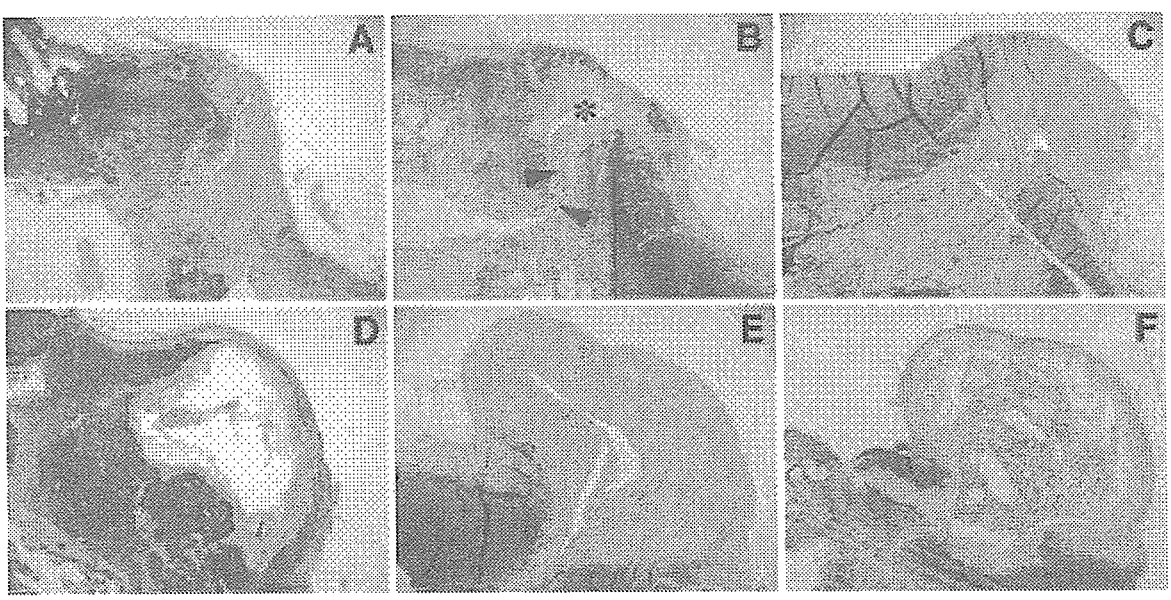

FIG. 2. Photomicrographs of the facet cysts resected en bloc with the neighboring facet joint. A: Type 1: small protrusion with a fissure. Only a fissure can be found inside the protruding ligamentum flavum. B: Type 2: small protrusion with a cavity. The cystic cavity is formed with fibrinoid degeneration. A communicating channel is clearly detected *(arrowheads)* between the cyst *(asterisk)* and the facet joint. C: Type 3a: semicircular cyst with a thickened cyst wall. Granulation and small-vessel proliferations are noted in the wall of the cyst. D: Type 3b: semicircular cyst with a thin cyst wall. Fibrinoid degeneration and partial myxoid degeneration are noted in the cystic cavity. E: Type 4a: round cyst with a thickened cyst wall. Increased collagen fibers and fibrinoid degeneration are observed in the wall of the cyst. F: Type 4b: round cyst with a thin cyst wall. Marked myxoid degeneration is detected in the cystic cavity. Elastica–Masson stain, original magnification 12.5. Bars = 1 mm.

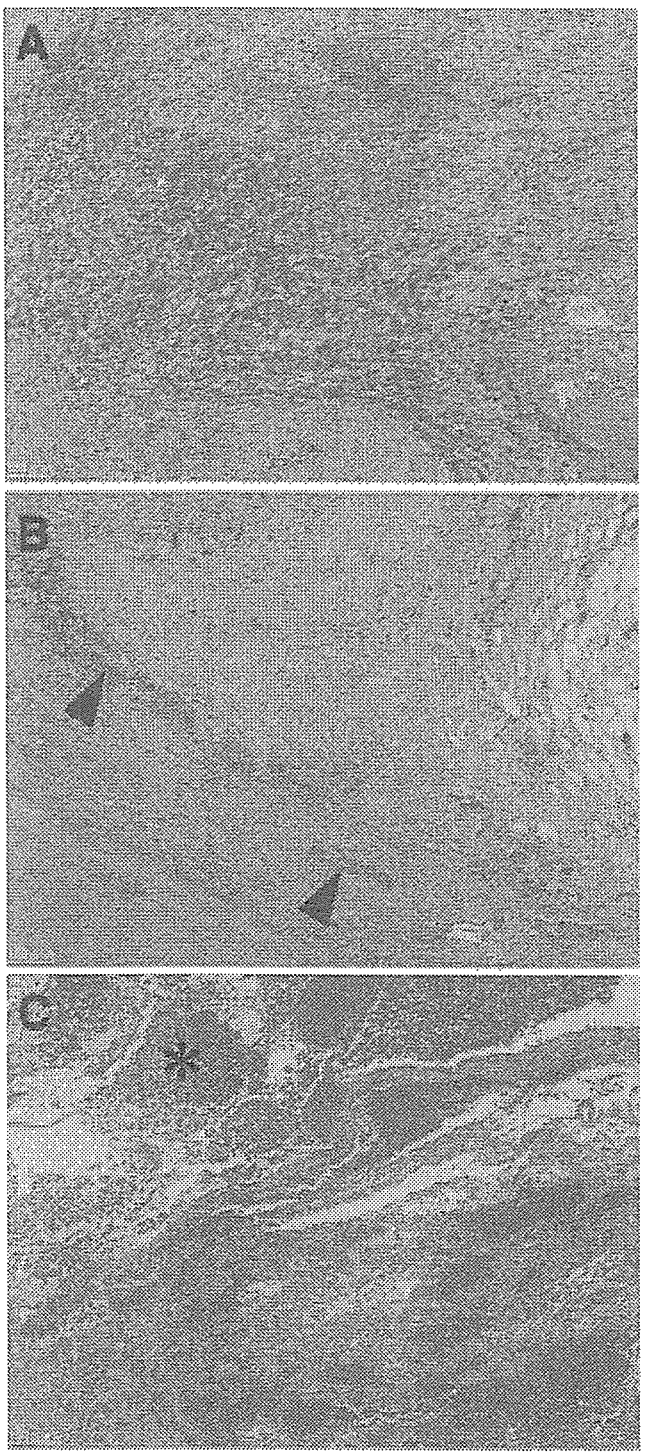

FIG. 3. Photomicrographs of the wall and contents of the facet cyst. A: In the base of the cyst, small inflammatory cells are detected in the degenerative elastic and collagen fibers, whereas synovial lining cells are not noted. B: Small-vessel proliferations *(arrowheads)* are found at the base of the cyst. C: Fibrinoids and hemorrhage *(asterisk)* are noted in the cystic cavity. H & E, original magnification 100. Bars = 100 m.

et joint, as confirmed on arthrography and CT scanning of the facet joint. All patients in our consecutive series were placed in this category. In addition, we could confirm the communication histologically. Such a definition would be

TABLE 1
*Summary of histological findings in facet cysts**

Cyst Feature	Type 1				Type 2				Type 3a				Type 3b				Type 4a				Type 4b			
walls																								
fibrinoid degeneration	3	3	—	—	—	8	—	—	—	2	3	—	9	4	—	—	—	—	7	—	—	6	1	—
granulation	6	—	—	—	2	—	6	—	—	—	1	4	8	3	2	—	—	4	3	—	5	1	1	—
vessel proliferation	6	—	—	—	2	6	—	—	—	1	4	—	5	8	—	—	—	3	4	—	5	1	1	—
calcification	6	—	—	—	7	1	—	—	4	1	—	—	13	—	—	—	7	—	—	—	5	1	—	1
contents																								
fibrinoid	2	4	—	—	—	—	8	—	—	—	4	1	—	—	4	9	—	—	1	6	—	—	—	7
myxoid degeneration	6	—	—	—	8	—	—	—	4	—	1	—	3	—	8	2	4	2	1	—	—	—	6	1
hemorrhage	3	1	2	—	6	—	2	—	1	4	—	—	3	5	4	1	—	4	3	—	1	4	2	—

* There were six Type 1, eight Type 2, five Type 3a, 13 Type 3b, seven Type 4a, and seven Type 4b cysts. = negative; = mild; = moderate; = severe.

TABLE 2

Relationships between the type of facet cyst and the preoperative symptom duration and spinal degeneration

Variable	Type 1	Type 2	Type 3a	Type 3b	Type 4a	Type 4b
mean preop duration (mos)*	13 (3–36)	12 (3–26)	22 (1–72)	6 (2–14)	13 (1–48)	19 (1–84)
3 mos (no. of cases)	1	1	1	5	2	2
spondylolisthesis (no. of cases)	3	5	2	4	2	3
Grade 3 facet degeneration (no. of cases)	2	5	4	10	6	6

* Parenthetical values reflect the range of preoperative symptoms in months.

useful when considering the pathogenesis of this type of cyst.

Several hypotheses concerning the origin and development of juxtafacet cysts have been reported. These lesions have been described as originating from a synovium herniation through a defective joint capsule,[9,11,12,15] from mucinous degeneration within the periarticular fibrous tissue,[12,22] from proliferation of pluripotential mesenchymal cells in the ligamentum flavum,[1,12,15,22] from repetitive microtrauma with focal hemorrhage,[12,21,22] and from a part of the articular capsule of the facet joint or some distensible tissue penetrating into a ruptured degenerative ligamentum flavum.[22] Those hypotheses were based on the findings of a small number of juxtafacet cysts, which were not always removed en bloc with the ligamentum flavum and the involved facet joint.[9,12,22,25] In addition to the radiological examinations of 46 facet cysts, we investigated the cysts grossly and histologically after their complete en bloc resection by medial facetectomy with all of the ligamentum flavum. A medial facetectomy should not affect lumbar spinal stability,[2] and complete excision of the cyst might be necessary to prevent its postoperative recurrence. Using these specimens, we could clearly evaluate not only the cysts themselves but also the joints including the ligaments.

In many patients with facet cysts we observed chronic symptoms during a mean preoperative interval of more than 12 months. The mean duration of the symptoms prior to surgical treatment in patients with Type 3b cysts was shorter than that in patients with other types. Type 3b cysts were not as large as Type 4 lesions, and most of them had hemorrhage in their cavities. The size of the cyst and bleeding into its cavity might be related to acute onset or sudden worsening of symptoms.

There are two portions of the ligamentum flavum, the interlaminar and the capsular parts.[17,23,27] The normal ligamentum flavum consists of 80% elastic fibers and 20% collagen fibers, but in spondylotic specimens the contents vary from 50 to 80% elastic and 20 to 50% collagen fibers.[26] Mechanical and biochemical factors are closely related to this process.[19,23] An increase of collagen fibers has been noted more markedly in the capsular portion,[27] which suggests that this part might be broken more easily. As in previous reports,[3,9,12,14,25] in this study there was instability of the involved spinal region and degenerative facet joints in many of the patients. Types 3 and 4 cysts were associated with more severe facet degeneration, one reason for which could be that these lesions required more time to form than the other cyst types. Spinal instability can cause the ligamentum flavum to rupture, particularly in the capsular portion. In all of our cases, facet joint arthrography and CT scanning showed communication between the cyst and the facet joint. However, synovial lining cells, described as synovial cysts in the literature, were absent in all cases.[1,4,5,9,11,12,14] This fact suggested that synovial cells had not yet invaded into the cysts or that the origin of the lesions was not from a herniation of synovium into the ligamentum flavum.[9,11,12,15] Synovial fluid from synovial cells could infiltrate through the communicating channel, which might enlarge the cysts.

Based on the radiological and histopathological findings in this study, we propose that the following pathogenesis occurs during the formation of the facet cysts. Our classification is related to this proposed pathogenesis (Fig. 4). 1) Degenerative arthritic changes and increased joint motion in the lumbar spine cause ligamentum flavum degeneration; collagen fibers substitute for elastic fibers in areas where they become loose, and these then protrude into the spinal canal. Spinal instability also causes rupture and fissure in the ligamentum flavum, particularly in the capsular portion. The fissure develops into a cavity, mainly because of joint fluid from the connected facet joint and possibly because of secretion from fibroblasts.[18,24] Occasionally, hemorrhage can occur. These stages of the cyst would correspond to Types 1 and 2 cysts in this study. 2) Growth of the cyst wall leads to more damage in the ligamentum flavum so that an inflammatory reaction with a fibroblastic healing response occurs around the cyst wall. This healing response thickens the cyst wall. These stages are represented in Types 2 and 3a cysts. 3) The shape and size of the cyst depend on the elasticity and intensity of the cyst wall as well as the local anatomy of the spinal canal—Type 3a cysts can transform into Type 3b or develop

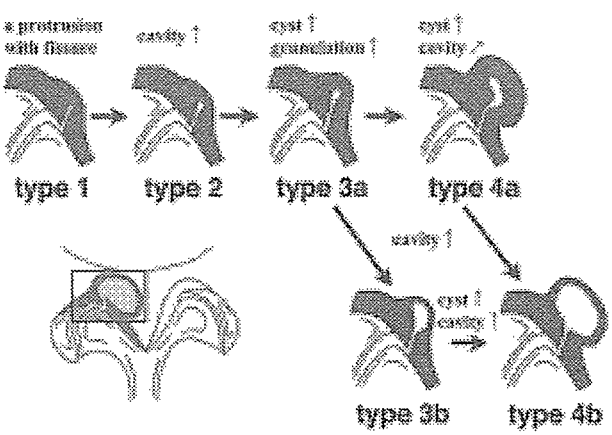

FIG. 4. Schematic drawings showing the development of facet cysts in the lumbar spine, illustrating the proposed classification.

into Type 4 lesions. As the cystic cavity becomes larger, the cyst wall becomes thinner with fibrinoid degeneration as seen in Types 3b and 4b. The contents of the cavity show myxoid degeneration during the resorption of hemorrhage and fibrinoid debris.

Facet cysts occasionally regress spontaneously or through conservative treatment.[8,13,15] Symptoms in approximately one third of our patients improved with nonoperative management.[13] Rupture or shrinkage of cysts could facilitate regression, but when this would occur is unpredictable.[8,13] If the patients' symptoms do not improve in response to conservative treatment, excision of the cyst should be performed.

Conclusions

Juxtafacet cysts that communicate with the neighboring facet joints were defined as facet cysts in the present study. We found that facet cysts have several shapes. Their shape probably depends on the stage of their development. The pathogenesis of facet cysts is closely related to degenerative instability of the lumbar spine and degenerative changes in the ligamentum flavum and the facet joints. A classification of these cysts and a pathogenesis, based on our intensive study of 46 of them, is proposed.

References

1. Abdullah AF, Chambers RW, Daut DP: Lumbar nerve root compression by synovial cysts of the ligamentum flavum. Report of four cases. **J Neurosurg 60:**617–620, 1984
2. Abumi K, Panjabi MM, Kramer KM, Duranceau J, Oxland T, Crisco JJ: Biomechanical evaluation of lumbar spinal stability after graded facetectomies. **Spine 15:**1142–1147, 1990
3. Banning CS, Thorell WE, Leibrock LG: Patient outcome after resection of lumbar juxtafacet cysts. **Spine 26:**969–972, 2001
4. Casselman ES: Radiologic recognition of symptomatic spinal synovial cysts. **AJNR Am J Neuroradiol 6:**971–973, 1985
5. Epstein NE: Lumbar synovial cysts: a review of diagnosis, surgical management, and outcome assessment. **J Spinal Disord Tech 17:**321–325, 2004
6. Gritzka TL, Taylor TK: A ganglion arising from a lumbar articular facet associated with low back pain and sciatica. Report of a case. **J Bone Joint Surg Br 52:**528–531, 1970
7. Hemminghytt S, Daniels DL, Williams AL, Haughton VM: Intraspinal synovial cysts: natural history and diagnosis by CT. **Radiology 145:**375–376, 1982
8. Houten JK, Sanderson SP, Cooper PR: Spontaneous regression of symptomatic lumbar synovial cysts. Report of three cases. **J Neurosurg 99 (2 Suppl):**235–238, 2003
9. Hsu KY, Zucherman JF, Shea WJ, Jeffrey RA: Lumbar intraspinal synovial and ganglion cysts (facet cysts). Ten-year experience in evaluation and treatment. **Spine 20:**80–89, 1995
10. Kao CC, Uihlein A, Bickel WH, Soule EH: Lumbar intraspinal extradural ganglion cyst. **J Neurosurg 29:**168–172, 1968
11. Kao CC, Winkler SS, Turner JH: Synovial cyst of spinal facet. Case report. **J Neurosurg 41:**372–376, 1974
12. Kjerulf TD, Terry DW Jr, Boubelik RJ: Lumbar synovial or ganglion cysts. **Neurosurgery 19:**415–420, 1986
13. Kusakabe T, Kasama F, Sato K, Sato T: [Conservative treatment for facet cyst in the lumbar spine.] **Seikei Saigai Geka 46:**259–265, 2003 (Jpn)
14. Lyons MK, Atkinson JL, Wharen RE, Deen HG, Zimmerman RS, Lemens SM: Surgical evaluation and management of lumbar synovial cysts: the Mayo Clinic experience. **J Neurosurg 93 (1 Suppl):**53–57, 2000
15. Maezawa Y, Baba H, Uchida K, Furusawa N, Kubota C, Yoshizawa K: Spontaneous remission of a solitary intraspinal synovial cyst of the lumbar spine. **Eur Spine J 9:**85–87, 2000
16. Meyerding HW: Spondylolisthesis. **Surg Gynecol Obstet 54:** 371–377, 1932
17. Naffziger HC, Inman V, Saunders JB: Lesions of the intervertebral disc and ligamenta flava. Clinical and anatomical studies. **Surg Gynecol Obstet 66:**288–299, 1938
18. Ohshima M, Ogoshi T, Ogawa H, Muto A, Suzuki K, Otsuka K: Effect of dental cyst epithelium-conditioned medium on collagenase production by periodontal ligament fibroblasts. **J Nihon Univ Sch Dent 39:**31–33, 1997
19. Park JB, Chang H, Lee JK: Quantitative analysis of transforming growth factor-beta 1 in ligamentum flavum of lumbar spinal stenosis and disc herniation. **Spine 26:**E492–E495, 2001
20. Pathria M, Sartoris DJ, Resnick D: Osteoarthritis of the facet joints: accuracy of oblique radiographic assessment. **Radiology 164:**227–230, 1987
21. Pirotte B, Gabrovsky N, Massager N, Levivier M, David P, Brotchi J: Synovial cysts of the lumbar spine: surgery-related results and outcome. **J Neurosurg 99 (1 Suppl):**14–19, 2003
22. Prestar FJ: Juxta facet cysts of the lumbar spine. **Minim Invasive Neurosurg 39:**45–49, 1996
23. Ramsey RH: The anatomy of the ligamenta flava. **Clin Orthop Relat Res 44:**129–140, 1966
24. Ricard J, Pelloux H, Pathak S, Pipy B, Ambroise-Thomas P: TNF- enhances Toxoplasma gondii cyst formation in human fibroblasts through the sphingomyelinase pathway. **Cell Signal 8:**439–442, 1996
25. Wildi LM, Kurrer MO, Benini A, Weishaupt D, Michel BA, Bruhlmann P: Pseudocystic degeneration of the lumbar ligamentum flavum: a little known entity. **J Spinal Disord Tech 17:**395–400, 2004
26. Yong-Hing K, Reilly J, Kirkaldy-Willis WH: The ligamentum flavum. **Spine 1:**226–234, 1976
27. Yoshida M, Shima K, Taniguchi Y, Tamaki T, Tanaka T: Hypertrophied ligamentum flavum in lumbar spinal canal stenosis. Pathogenesis and morphologic and immunohistochemical observation. **Spine 17:**1353–1360, 1992

Manuscript received April 10, 2006.
Accepted in final form July 27, 2006.
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Geriat. Med. 44(2)：143～147, 2006

特集 高齢者の転倒骨折予防

2

わが国における大腿骨近位部骨折の発生率とその経年推移

萩野　浩*

KEY WORD

大腿骨近位部骨折
大腿骨頸部骨折
疫学
発生率
経年推移

POINT

- 日本人を含めたアジア人の大腿骨近位部骨折発生率は，欧米白人より低値である．
- わが国での性・年齢階級別発生率は経年的に上昇している．
- わが国における年間新規患者数は2010年に約15万例，2030年には約25万例に達すると推計される．

0387-1088/06/¥500/論文/JCLS

はじめに

大腿骨近位部骨折は，80歳以上の後期高齢者あるいは超高齢者と呼ばれる年齢群で発生率が高い．わが国では今後も人口の高齢化が急速に進むと予測されていて，大腿骨近位部骨折の患者数も増加すると見込まれている．しかしながら，最近に行われた大腿骨近位部骨折の経年的な疫学調査結果によれば，患者数の増加は単に高齢者人口の増加が原因ではなく，年齢ごとの骨折発生率が近年上昇傾向にあることも原因となっている．すなわち，高齢者数が増加しているのは，「長生き」をする方々が増えていることを意味するが，「骨が折れやすい」高齢者の割合もまた増加していることになる．

大腿骨頸部(近位部)骨折とはどこの骨折を指すのか？

わが国では一般に，大腿骨近位部骨折をいわゆる「大腿骨頸部骨折」と称し，内側骨折(関節包内)，外側骨折(関節包外)の2つの骨折型に分けていた(**図1**)[1]．しかしながら最近では，大腿骨近位部骨折を頸部骨折(neck fracture)，転子部骨折(trochanteric fracture)に分類することが多くなり，この分類の頸部骨折が「内側骨折」に当たり，転子部骨折が「外側骨折」に当たる．したがって「頸部」という名称を用いる際には注意が必要である．本稿では，頸部骨折(内側骨折)および転子部骨折(外側骨折)を合わせたものを大腿骨近位部骨折とする．なお英語論文では，この「大腿骨近位部骨折」はhip fractureと記述される．

日本人は発生率が低い

大腿骨近位部骨折の発生率を**図2**に示す(1994年の鳥取県での調査結果)[2]．性・年齢階級別の発生率は男女とも70歳以降に指数関数的に上昇し，75～79歳では女性で約480(年間人口10万人当たり)，80～84歳では約800，85歳以上では約1,900に達する．このほかに国内各地で行われた調査結果や全国規模での調査を比べ

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