

Scoliosis as the first sign of a cystic spinal cord lesion

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Summary. We reviewed the cases of ten patients with scoliosis as the first sign of a cystic spinal cord lesion with the aim of identifying and studying early patient and curve characteristics. All patients were examined with magnetic resonance (MR) imaging of the brain and spinal cord and quantitative thermal testing (QTT). The mean Cobb angle was 22° and the curves were right thoracic in seven patients, left thoracic in two, and left lumbar in one, when first seen for scoliosis. In six patients the cystic lesion was found in routine screening for syringomyelia, which is performed in all cases of congenital and juvenile scoliosis and in adolescent scoliosis before bracing. In four patients, it took up to 17 years following the initial diagnosis of scoliosis before neurologic deterioration warranted MR imaging, disclosing two Chiari I associated syrinxes and two cystic spinal cord tumors. QTT revealed a subclinically decreased sensation in two of the patients with no findings other than scoliosis. It also verified the decreased sensation in all patients in whom neurologic deterioration had complicated the clinical course of their scoliosis. More frequent spinal MR screening of patients with supposed juvenile or adolescent idiopathic scoliosis is indicated, regardless of curve type, to exclude a neurogenic cause. QTT documents subclinical as well as overt decreased sensation, and is valuable in the serial follow-up of these patients to monitor the progress or the response to treatment of the cystic lesion.

Key words: Chiari malformation – Magnetic resonance imaging – Scoliosis – Syringomyelia – Sensory testing

Cystic spinal cord lesions have an insidious and varied symptomatology which may remain unrecognized or misunderstood, causing pain and unnecessary neurologic de-

terioration before proper treatment is started [5, 10]. The possibility of recovery is small once a severe neurologic deficit has become established [7]. Scoliosis may be an early sign of a syrinx, regardless of any association with a Chiari malformation [39] or tumor [7, 21]. Some of these patients are referred to orthopedic surgeons for supposed idiopathic scoliosis long before sensory disturbances or other localizing neurologic signs appear [1, 25, 31, 32, 35, 38]. Many questions remain to be answered about initial signs and symptoms both of the syrinx and the associated scoliosis. Decreased sensitivity may be a presenting sign, but a slight sensory impairment is often difficult to assess on clinical examination. Quantitative estimation of thermal sensitivity (QTT), which is particularly applicable to patients with spinal cord lesions, may disclose subclinical thermal sensory loss and document obvious sensory deficits for cold, warmth, and heat-pain stimuli [4, 32].

The aim of this report was to study the early signs and symptoms in patients in whom scoliosis was the first sign of a cystic spinal cord lesion. We also wanted to assess the diagnostic value of QTT in patients with supposed or evident decreased sensation, by correlating the findings with the extent and type of syringomyelia.

Patients and methods

The group in our study consisted of ten patients initially diagnosed with idiopathic scoliosis in whom subsequent MR imaging had disclosed syringomyelia of an idiopathic type, associated with structural scoliosis, Chiari I malformation, or a spinal cord tumor. There were eight girls and two boys, with a mean age at diagnosis of scoliosis of 11 (range 7–14) years and a mean age at diagnosis of syringomyelia of 16 (range 7–26) years. All of them were clinically examined and diagnosed between August 1989 and February 1993, a period when children diagnosed with juvenile or congenital scoliosis routinely underwent MR imaging of the spine in a screening program, and before bracing in the case of an adolescent scoliosis. Four of the patients were initially assessed and/or treated for supposed juvenile or adolescent scoliosis and were followed up for up to 17 years before the clinical diagnosis of myelopathy was made and MR imaging disclosed the syrinx. A clinical examination was performed in all patients before MR of the spine and brain and QTT was performed. MR images were obtained with a 1.0-T superconducting magnet (Magnetom, Siemens, Erlangen) using a spin echo technique with T1-weighted images (TR 600/TE15 ms). The brain stem and cervical spinal cord were examined with 15 sagittal scans of 4-mm thickness. The dorsal and lumbar spinal cord

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Table 1. Clinical data for ten patients with scoliosis and a cystic spinal cord lesion

Case No.	Age at diagnosis (years)		Sex	Scoliosis		Clinical presentation	Neurologic findings	MR findings
	Scoliosis	Syrinx		Apex	Cobb angle			
1	14	16	F	T8 (R)	32°	Scoliosis	Normal	Syrinx, C6–C8
2	16	17	F	L2 (L)	27°	Scoliosis	Normal	Syrinx, T6–T8
3	10	13	M	T4 (L) T9 (R)	24° 27°	Scoliosis	Normal	Syrinx, T6–T8
4	11	13	F	T3 (R) T3–T9	37°	Congenital scoliosis	Normal	Syrinx, T5
5	13	21	F	T4 (R) T12 (L)	7° 7°	Scoliosis. Left hip pain 5 years later	Painful dysesthesia T5–L2 left side	Chiari I with syrinx, C2–T5
6	7	12	F	T7 (R) L3 (L)	20° 25°	Scoliosis, Painless wound on thumb	Decreased sensitivity C6–C8 right hand	Chiari I with syrinx, C1–T12
7	7	8	F	T7 (R) L2 (L)	13° 19°	Scoliosis. Torticollis 1 year later	Subclinical sensory deficit on QTT	Chiari I with syrinx, C1–T9
8	7	7	M	T9 (L)	20°	Scoliosis. abdominal pain and torticollis	Subclinical sensory loss on QTT	Spinal cord tumor with syrinx, C1–T10
9	9	26	F	T10 (R)	28°	Scoliosis. Back pain 13 years later	Arm and leg weakness, painful dysesthesia	Spinal cord tumor with syrinx, C1–T12
10	14	25	F	T8 (R)	13°	Scoliosis. Back pain 3 years later	Painful abdominal and leg dysesthesia, wasting right thigh	Spinal cord tumor with syrinx, T2–T5

were examined with 34 continuous axial scans of 10-mm thickness. In patients with neurologic deficits at clinical examination, enhanced MR images were obtained by administering dimeglumine gadopentate (Magnevist, Schering, Berlin) intravenously at a standard dose of 0.1 mmol/kg, followed by postcontrast T1-weighted images.

The scoliosis curve was measured by Cobb's method [8] on frontal images obtained in the standing position. In cases with delayed diagnosis of the cystic spinal cord lesion, radiographs obtained after the first clinical presentation of scoliosis were reevaluated.

Sensory loss was quantified by determining discrimination thresholds for cold, warmth and heat pain with a modified Marstock thermostimulator [13] operating on the Peltier principle (Thermotest, Somedic, Stockholm, Sweden) delivering controlled focal thermal stimulation of the skin. The thermode, with a stimulating surface of 2.5 × 5 cm, can be heated or cooled at a rate of 1–2°C per second and the stimulating temperature continuously measured by a thermocouple and recorded by a pen recorder. When the stimulator is in good contact with the skin, patients are asked to press a switch as soon as they perceive the sensation of cold, warmth, and heat pain. All examinations were performed by the same person (L.S.). Each test session started with a series of presentations of thermal stimuli on the abdominal skin (T10 dermatome) to introduce the patient to the test technique.

Thresholds for cold, warmth, and heat pain were recorded from the cheek (n. maxillaris) and hand (thenar or hypothenar eminence). Additional abdominal or extremity recordings were obtained from areas with suspected decreased sensation. A predetermined adaptive temperature of 32°C was used at the start of each test. The outer temperature limits were set at 10°C for cold and 50°C for heat to avoid tissue damage. The registration from the same location on the contralateral side was used as a control, and in cases with bilateral sensory deficit, we used historical controls.

Results

Demographic data, clinical symptoms and signs, and radiologic and MR findings in the ten patients are detailed in

Table 1. Screening for cystic spinal cord lesions revealed three idiopathic syrinxes, one syrinx associated with multiple vertebral anomalies of the thoracic spine, one Chiari I associated syringomyelia, and one tumor-associated syrinx. In the remaining four patients, diagnosis was delayed by 5 to 17 years until neurologic complications were evident and further investigation with MR imaging disclosed their cystic lesions. Two of them had Chiari I associated syringomyelia and two had cystic tumors of the spinal cord. The curve types were double major with the proximal curve right thoracic in three of four patients, right thoracic in four, left thoracic in one and left lumbar in one. The mean Cobb angle of the proximal curve at presentation was 22°.

Idiopathic syringomyelia

There were two patients with syringomyelia at T6–T8 and one patient with a syrinx at C6–C8 (Fig. 1). All were centrally located and none were associated with any other intraspinal abnormality or neurologic deficit on neurologic examination. There were no findings of decreased sensitivity on QTT either on the standard test sites or on the additional sites chosen with respect to syrinx location. The magnitude of Cobb angle ranged from 24 to 32°, and curve types were right thoracic, left thoracic, and left thoracic/right lumbar.

Congenital scoliosis with syringomyelia

One patient with a right thoracic structural curve of 37° had a small, centrally located syrinx at T5 and multiple vertebral anomalies between T3 and T9. The neurologic examination and QTT showed no abnormalities.

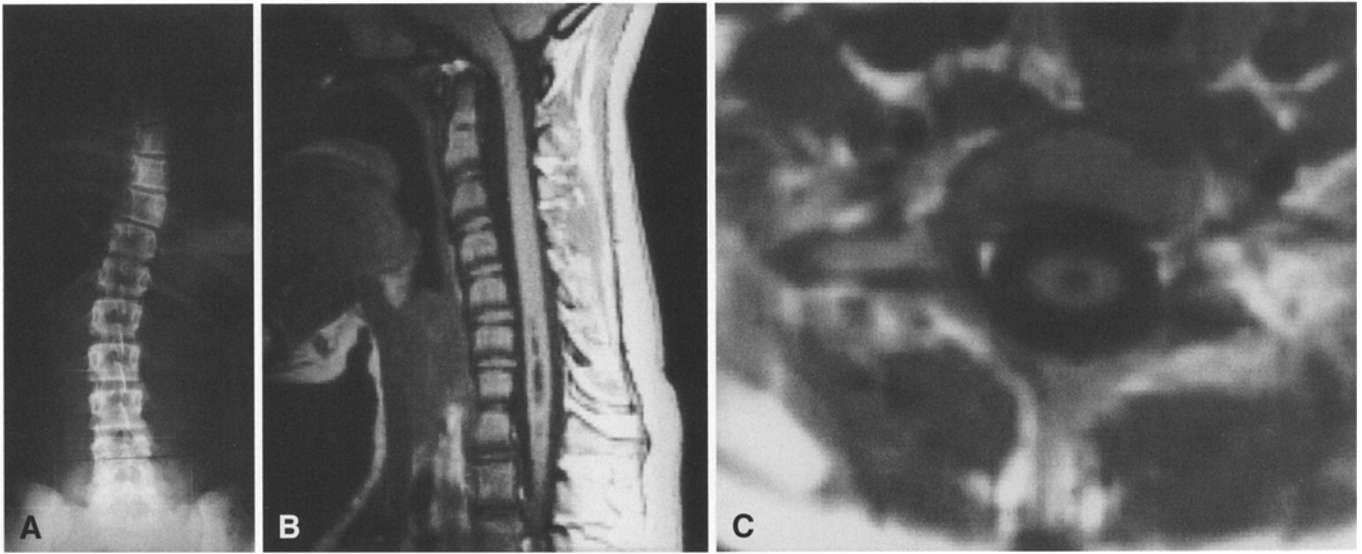


Fig. 1 A–C. *Case 1.* **A** Plain anteroposterior (AP) radiograph of 14-year-old girl with right thoracic scoliosis. **B** Midline sagittal and **C** axial MRI of idiopathic syrinx located at C6–C8 at the age of 16 years

Chiari I malformation with syringomyelia

All three patients in this group had cysts extending from the upper cervical to the middle or lower thoracic spinal cord. Two of these patients deteriorated neurologically and presented with decreased sensation or painful dysesthesia 5 and 8 years respectively after diagnosis of their scoliosis. Both had decreased sensation documented with QTT (Fig. 2). The third patient was diagnosed after routine screening for intraspinal pathology (Fig. 3). She later experienced a period of neck pain and torticollis. On QTT a subclinically decreased sensation was shown in her right hand. All three patients had double primary scoliosis, the proximal curve being right thoracic and the distal curve left lumbar. Two of the patients had an early onset scoliosis at 7 years of age.

Cystic spinal cord tumor

Two patients presenting with juvenile scoliosis had extensive syrinxes engaging the whole cervical and dorsal spinal cord associated with mid-thoracic spinal cord tumors. One of them underwent spinal fusion at the age of 14 years owing to severe progression of her scoliosis (Fig. 4). She developed back pain 16 years after the presentation of scoliosis, and sensory loss and lower extremity weakness 1 year later. QTT showed decreased thermosensitivity and, before a decompressive operation was performed, she developed an acute tetraparesis. The other patient, initially diagnosed from screening for syringomyelia, presented at the age of 7 years with a left thoracic scoliosis and torticollis after surgery for appendicitis. QTT revealed a subclinically decreased abdominal sensation. After decompressive operations both patients improved their thermal sensitivity.

The third patient in this group presented with a right thoracic adolescent curve at the age of 14 years. She complained of back pain 3 years later but there was a further delay of 8 years before decreased left-side abdominal and leg sensation and wasting of the right thigh and painful dysesthesia were noticed. MR imaging showed a cystic lesion from T2 to T5 associated with a small tumor at T5. Her thermosensitivity was decreased according to the clinical findings, but there was an extended area of subclinical sensory loss beyond the clinically documented one.

The scoliotic curves in these three patients had apexes between T8 and T10 and were all single. Two were right thoracic and one left thoracic.

Discussion

The variable clinical picture and unpredictable natural history of syringomyelia [14, 34] demands methods capable of detecting subtle neurologic deterioration due to syrinx progression, sometimes after years of spontaneous arrest of the disease. In a report on preoperative sensory examination before surgery in progressive scoliosis, Chopin et al. [6] claim that beside thermal disturbances, absent or asymmetrical cutaneous abdominal reflexes are also associated with syringomyelia and may be an early diagnostic indication. However, an absent abdominal reflex is an unspecific sign indicating a corticospinal tract lesion, and has to be evaluated together with other neurologic findings such as spasticity or paresis. Thermal disturbance is expected to appear earlier than corticospinal symptoms since the spinothalamic tract passes the midline, which is the most common location of an early cystic lesion.

A systematic quantification of sensory impairment has not previously been performed in patients with scoliosis as the first sign of a cystic lesion. QTT has mostly been used in the assessment of metabolic and traumatic neuropathies in the leg or arm [13, 15, 36], but the technique is also applicable to the study of central thermal pathways

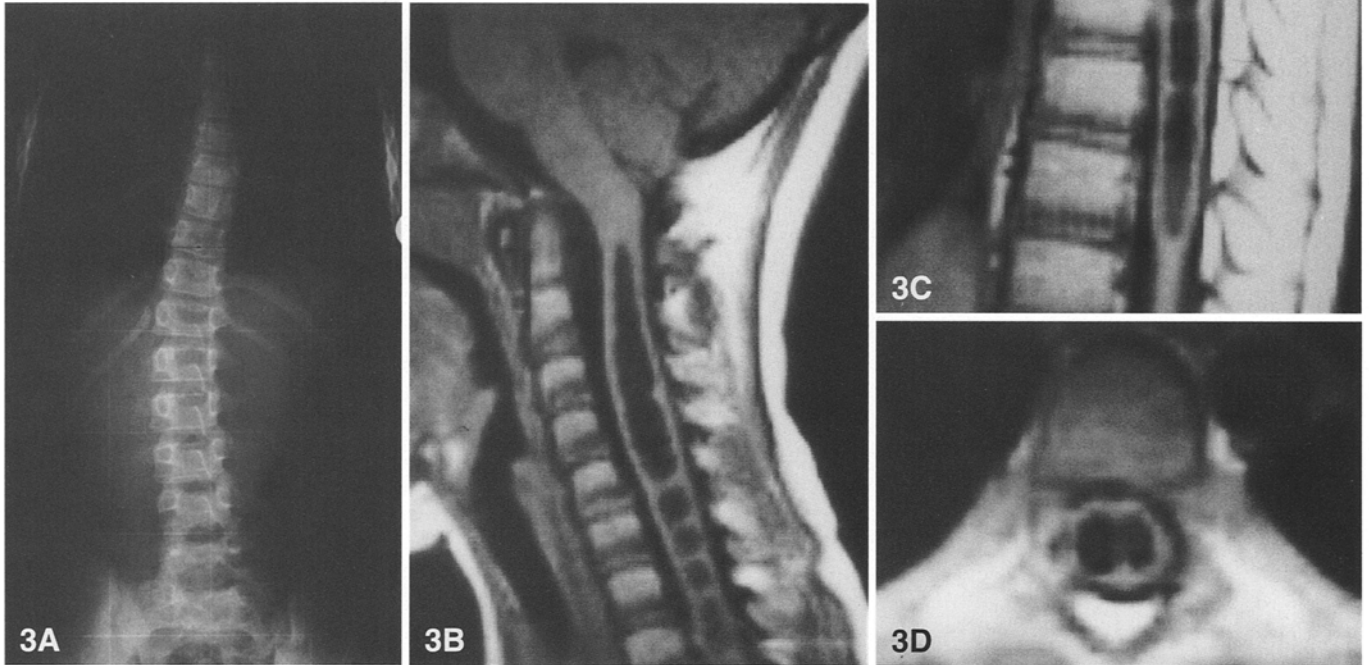
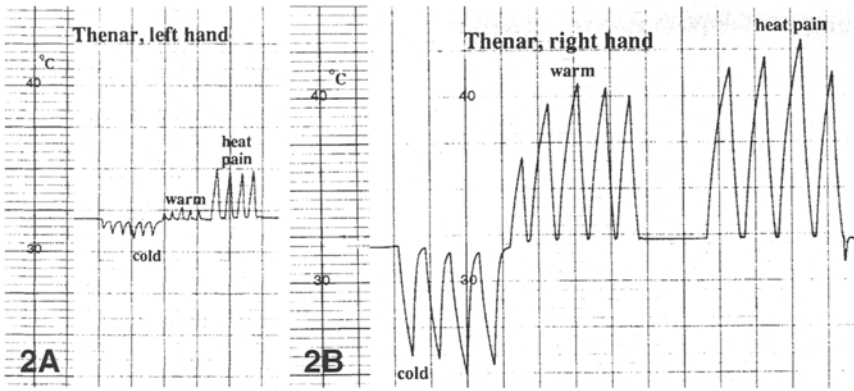


Fig. 2A, B. Case 6. Recordings of thermal thresholds for cold, warmth, and heat pain from **A** left hand and **B** right hand thenar regions in a 16-year-old girl with scoliosis and Chiari I associated syringomyelia. Readings show normal sensation at left thenar and decreased sensation for all modalities at right thenar

Fig. 3A–D. Case 7. **A** Plain AP radiograph of 7-year-old girl with right thoracic/left lumbar scoliosis. **B, C** Midline sagittal, and **D** axial MRI of Chiari I associated syringomyelia at C1–T9 at the age of 8 years

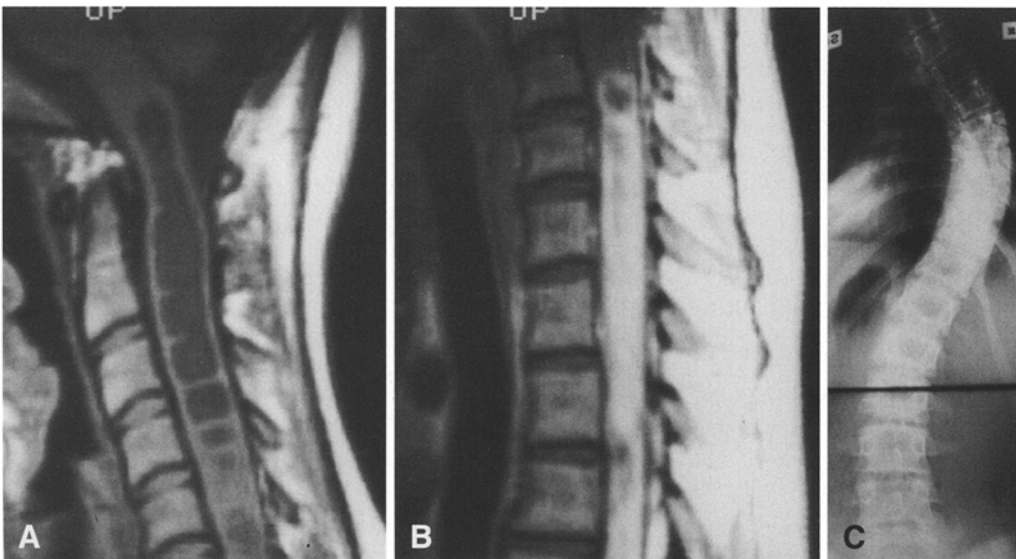


Fig. 4A–C. Case 9. **A, B** Sagittal MRI of 25-year-old woman with right thoracic scoliosis and a cystic spinal cord tumour at C1–T12. **C** Plain AP radiograph of her scoliosis before spinal fusion at the age of 16 years

[4, 32, 33]. As the neurologic deficit is related to the location of the spinal cord lesion, reports on thermal thresholds for each of our test sites in healthy controls are sparse [3, 11–13]. In order to minimize variations in the registrations due to use of different testing procedures, we used historical controls from earlier studies performed by the same examiner using the same technique as the present study. However, in cases with strictly unilaterally decreased sensation, we used the unaffected contralateral side as a control.

There are few reports on neurophysiologic quantification of abnormalities in association with syringomyelia. Boivie [4], using QTT, showed that decreased temperature sensation in syringomyelia was an indication of an internal dorsal horn spinal cord lesion. Jabbari [19] found pathological somatosensory evoked potentials (SEPs) in 16 out of 22 patients with syringomyelia (72%). In patients with clinical loss of temperature and superficial pain, SEPs were abnormal in the same percentage, and in some cases, pathologic SEPs were registered in asymptomatic extremities. Pathologic SEPs are not reliable indicators of the syringomyelia-Chiari I complex, as only 25% were pathologic in this study. In our study, QTT confirmed the clinically observed decreased sensation as well as showing a subclinical sensory loss in some cases. MR imaging has proved to be the method of choice in evaluation of spinal cord and brain stem lesions [14, 24, 30, 40]. This is reflected in an increase in the reported prevalence of scoliosis associated with syringomyelia in children and adolescents, to between 32 and 61% compared with earlier reports from when MR units were not in routine use of between 3 and 15%. The increase shows that a progressive syrinx may be present for decades before it is diagnosed, and the first sign of syringomyelia may vary from scoliosis wrongly supposed to be of an idiopathic type [1, 2, 17, 25, 35] to acute paraplegia [41].

Early diagnosis affords the best opportunity for operative arrest of syrinx progression, which is why recognition of early clinical features is important [22, 28, 37]. In cases of scoliosis, the patient may be both braced and/or have spinal fusion before neurologic deterioration warrants further investigation and a syrinx is diagnosed. Operative correction of such a scoliosis has had a high incidence of neurologic complications [16, 25, 35, 38]. Furthermore, Muhonen et al. [22] reported that scoliosis corrected itself in all patients below 10 years of age with Chiari I associated syringomyelia after operative restoration of normal cerebrospinal fluid dynamics. In contrast to this and according to Phillips et al. and Nohria and Oakes [23, 26], the effect of Chiari and syrinx surgery on scoliosis in immature patients is inconclusive. We could not confirm the observation of Barnes et al. [2] that left thoracic curves were more common in syringomyelia [9]. However, our screening for syringomyelia patients was only to select those with early onset scoliosis, and not unusual curve types. According to Williams [38], more recent reports show no preponderance for left or right convex curves in scoliosis associated with syringomyelia [18, 20, 26, 35]. Neurogenic scoliosis is probably right convex as often as left convex, even if the latter is more conspicuous and, up to now, studied more extensively in search of a neurogenic etiology of the

scoliosis. When a syrinx is found, this gives a false impression that left convex curves are more often associated with syringomyelia. There were, however, four double primary curves in our material, three of which were associated with Chiari I malformations and syringomyelia extending over at least ten segments, which is in accordance with the reports of Riseborough and Herndon [29] and Isu et al. [18].

Besides scoliosis, decreased temperature and pain sensation are probably some of the earliest findings, which may appear long before muscle weakness, atrophy, or gait disturbances are evident [6, 34, 38]. Isu et al. [18] reported on 17 patients with Chiari I and syringomyelia, all of whom had sensory deficits on admission, while scoliosis was present in 85%. Six of our patients eventually developed decreased sensation, but only two of them had muscular weakness or wasting, in both cases caused by cystic cord tumors. These two patients presented with back pain 3 and 13 years respectively after their scoliosis was diagnosed, and with muscle weakness 11 and 17 years after diagnosis. In cases of neurologic deficit at levels above the cystic tumor, it seems reasonable to assume that the cystic lesion contributes to the development of both scoliosis and sensorimotor deficits.

Four out of the five patients with small, centrally located syrinxes were among the patients screened for syringomyelia owing to early onset or prior to bracing. None were found to have any neurologic deficit, whether examined clinically or by QTT. The nature of these idiopathic syrinxes is unknown, and they may just have been found by chance when the spinal cord was examined with MR imaging. On the other hand, there are, to our knowledge, no reports on the prevalence of syringomyelia in patients having MR imaging of their spines for other reason. Bradford et al. [5] reported a 10% prevalence of syringomyelia in patients with congenital scoliosis [27] and recommended MR imaging in patients with lumbosacral kyphosis, pain, neurologic findings, or a cutaneous hairy patch. However, this procedure would not have resulted in correct diagnosis of our one asymptomatic patient with a syrinx associated with a congenital scoliosis. Torticollis is reported to be associated with syringomyelia according to Dure et al. [10], who found 1 out of 11 patients with this symptom. In our study, we had two patients out of ten with torticollis associated with early onset scoliosis. One of them had a Chiari I associated syrinx, while the other one had a spinal cord tumor.

This review provides evidence for the conclusion that scoliosis as the first sign of cystic spinal cord lesions is not rare. A large curve in a patient below 10 years of age should always raise suspicion, not only when it is associated with back pain or other atypical signs such as torticollis. Left convex curves are more conspicuous than right convex ones, but are probably not more frequently associated with a syrinx. Double primary curves may be associated with extensive syringomyelia, especially in Chiari I patients. The natural history of cystic spinal cord lesions is poorly documented, but our study shows that progression may be very slow, extending over decades before neurologic complications lead to diagnosis. Quantitative measurements of thermal sensation can demonstrate subclini-

cal sensory disturbances as well as known sensory deficits. It is also useful in the serial follow-up of patients to monitor the progress or the response to treatment of a cystic spinal cord lesion, as a complement to serial MR investigations, which may be less sensitive to subtle changes of syrinx size. The relevance of the small idiopathic syrinxes will be established by long-term clinical follow-up supplemented by serial MR imaging and QTT. Active screening for early onset scoliosis in school populations and more frequent spinal MR screening of patients with supposed idiopathic scoliosis, even without atypical features, is recommended until further knowledge scoliosis with cystic spinal cord lesions is obtained. However, MR imaging is expensive and can not become the single screening tool for syringomyelia in otherwise asymptomatic scoliotic patients. A meticulous clinical neurologic examination, including sensory modalities is mandatory to ensure early diagnosis of scoliosis caused by a cystic spinal cord lesion.

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Anterior Column Support of the Thoracolumbar Spine 8-9 December 1995, New York, NY, USA

This course is designed for orthopedic and neurosurgeons who want to explore both theoretical and practical approaches to this timely topic. Lectures will survey biomechanics, grafts and implants as related to intradiscal fusions, and reconstruction of vertebral body defects. New techniques, including structural alternatives to allograft as well as percutaneous and laparoscopic techniques, will be presented. Case presentations from the faculty will include tumor, trauma, infection, and post-traumatic deformity cases.

For further information, please contact: Gary L. Lowery, MD, PhD, c/o Medical Education Resources, 1500 West Canal Court, Suite 500, Littleton, CO 80120-4528, USA; Tel. 1-800-421-3756 (in USA), outside USA: +1-303-798-9682, Fax +1-303-798-5731. AMA Category 1 credits will be given.

1st International R. Roy-Camille Meeting on Spine - Failures and Complications of Spine Surgery/ Revision Surgery 18-20 January 1996, Paris, France

For further information, please contact: Dr. Claude Laville, 206, avenue de Versailles, F-75016 Paris, France; Fax +33-1-4071-0984.

23rd Annual Meeting of the International Society for the Study of the Lumbar Spine 25-29 June 1996, Burlington, VT, USA

If you are interested in submitting an abstract for that meeting please contact Dr. Bjorn Rydevik, Secretary, ISSLS, Sunnybrook Medical Center, Room A309, 2075 Bayview Avenue, Toronto, Canada, M4N 3M5. *Deadline for abstracts* is November 15, 1995. In order to attend the meeting as a non-member, you must have a paper or poster on the program or be invited by a member as his guest. Tel. 416-480-4833, Fax 416-480-6055.