ESETISMERTETÉS

CASE REPORT

Cervical syringomyelia associated with cervical disc disease

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Background and purpose – Syringomyelia is a neurological condition in which a longitudinal fluid-filled cavity is formed within the spinal cord. It usually occurs in the cervical region and is associated with Chiari malformation, infections, trauma, and tumors of the spinal cord. However, syringomyelia associated with cervical disc disease (SCD) is very rare and only a few cases have been reported so far. This case report presents the clinical and radiological findings of 13 cases of SCD to describe the properties of SCD and explore the nature of the relationship between syringomyelia and cervical disc disease.
Methods – SCD was diagnosed in 13 using MRI findings, including coexistence of syringomyelia and cervical disc disease, presence of narrowed cervical subarachnoid space secondary to the cervical disc herniation or cervical local kyphosis associated with cervical disc degeneration or herniation, and the cervical disc herniation or segmental kyphosis and syrinx should be located within the same levels. The MRI findings were used to grade the syrinx and determine whether the cervical disc herniation or local kyphosis was located at the proximal or distal end of the syrinx.
Results – All patients had single-level disc herniation or kyphosis, the most common level being C5–6 (n = 6), followed by C6–7 (n = 4) and C4–5 (n = 3). Eight patients had a distal type (disc disease located in the proximal end of the syrinx) SCD while five had the proximal variety (cervical disc disease located in the distal end of the syrinx). The average

Nyaki porckorong-betegséghez társuló nyaki syringomyelia

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Háttér és cél – A syringomyelia olyan neurológiai állapot, amelyben a gerincvelőben egy folyadékkal teli, hosszanti üreg keletkezik. Általában a nyaki régióban fordul elő, és Chiari-malformációval, fertőzésekkel, traumával és a gerincvelő daganataival hozható összefüggésbe. Mindazonáltal, a nyaki porckorongbetegséggel társuló syringomyelia (syringomyelia with cervical disc disease, SCD) nagyon ritka, és eddig csak néhány esetről számoltak be. Ez az esetismertetés 13 SCD-s eset klinikai és radiológiai leleteit mutatja be, hogy leírja az SCD tulajdonságait, és feltárja a syringomyelia és a nyaki porckorongbetegség közötti kapcsolat természetét.
Módszerek – Tizenhárom, MRI-leletek alapján SCD diagnózist kapott esetről számolunk be; az esetek között van olyan, amikor a syringomyelia és a nyaki porckorongbetegség együttesen fordult elő, van olyan, amikor a nyaki porckorongsérvhez vagy a nyaki porckorong-degenerációhoz társuló nyaki kyphosis miatt másodlagosan alakult ki a cervicalis subarachnoidealis tér szűkülete, valamint olyan, amikor a nyaki porckorongsérv vagy a szegmentális kyphosis és a syrinx ugyanazon a szinten belül helyezkedik el. Az MRI-leletek alapján osztályoztuk a syrinxet, és meghatároztuk, hogy a nyaki porckorongsérv vagy a helyi kyphosis a syrinx proximális vagy distalis végén található.
Eredmények – Minden betegnél egyszintű porckorongsérv vagy kyphosis volt jelen. A leggyakoribb szint a C5–6 (n = 6), majd a C6–7 (n = 4) és a C4–5 (n = 3) volt. Nyolc betegnél az SCD distalis típusú volt (a syrinx

length of the syrinx was two vertebral segments. Surgery was performed in five cases and some degree of syrinx resolution was observed in all of them.

Discussion – The main cause of syringomyelia is obstruction of cerebrospinal fluid (CSF) pathways; total obstruction could cause distal syrinx, whereas partial obstruction could cause proximal or distal syrinxes. Restoration of CSF pathways may result in some degree of resolution of syringomyelia. A causal association may exist between cervical disc disease and cervical syringomyelia but needs further exploration.

Conclusion – SCD is a mild form of syringomyelia with symptoms primarily arising due to disc herniation or local kyphosis. The surgical treatment of the cervical disc disease is sufficient and results in a syringomyelia resolution of some degree.

Keywords: cervical, disc herniation, mri, syringomyelia

proximális végén elhelyezkedő porckorongbetegség), míg ötnél a proximális változat fordult elő (a syrinx distalis végén elhelyezkedő nyaki porckorongbetegség). A syrinx átlagos hossza két csigolyaszegmens volt. Öt esetben végeztek műtétet, és mindegyiküknél megfigyelhető volt a syrinx bizonyos fokú feloldódása.

Megbeszélés – A syringomyelia fő oka a cerebrospinalis folyadék (CSF) járatainak elzáródása; a teljes elzáródás distalis syrinx, míg a részleges elzáródás proximális vagy distalis syrinx kialakulását okozhatja. A liquorközlekedés helyreállítása a syringomyelia bizonyos fokú megszűnését eredményezheti. A nyaki porckorongbetegség és a nyaki syringomyelia között ok-okozati összefüggés állhat fenn, de ennek igazolása további vizsgálatokat igényel.

Következtetés – Az SCD a syringomyelia enyhe formája, aminek a tünetei elsősorban porckorongsérv vagy helyi kyphosis miatt jelentkeznek. A nyaki porckorongbetegség műtéti kezelése elegendő, és a syringomyelia bizonyos fokú megszűnését eredményezi.

Kulcsszavak: cervicalis, porckorongsérv, MRI, syringomyelia

Syringomyelia is a condition in which a fluid-filled longitudinal cavity is formed within the spinal cord. It usually occurs in the cervical region and is associated with Chiari malformations[1], with arachnoiditis secondary to infection or trauma and spinal tumors being other common causes[1, 2]. Notably, syringomyelia associated with cervical spondylosis is extremely rare[3, 4]. So far, only four cases of syringomyelia with cervical disc disease (SCD) have been reported in the literature[5–8]. In this case report, we describe 13 cases of SCD.

Methods

Between 2008 and 2021, 13 patients (10 female, 3 male; mean age = 42.1 years, range = 21–59 years) were evaluated and diagnosed with SCD by the primary investigator (ACI) at two different neurosurgical clinics. All patients were diagnosed based on the magnetic resonance imaging (MRI) findings. The clinical and radiological data is summarized in **Table 1**.

The study was reviewed and approved by the local ethics committee and written informed consent was obtained from all patients.

SCD cases were included using the following criteria: 1. Radiological absence of the major causes of cervical syringomyelia, such as Chiari malformation, tumor, trauma, and infection; 2. Presence of narrowed cervical subarachnoid space secondary to cervical disc herniation or cervical local kyphosis without the obliteration of the posterior subarachnoid space due to spondylotic changes. Multilevel cervical disc disease was not an exclusion criterion; 3. Localization of the cervical syrinx and disc disease level should be in accordance, i.e., the disc pathology was located either at the upper or lower end of the syrinx.

The syrinx was graded based on MRI appearance: Grade 1: visible only on T2-weighted MRI; Grade 2: visible on both T1-weighted and T2-weighted MRI without spinal cord expansion; Grade 3: with spinal cord expansion; Grade 4: massive spinal cord expansion.

Results

All 13 patients had single-level disc herniation or local kyphosis, the most common level being C5–6 (n = 6), followed by C6–7 (n = 4) and C4–5 (n = 3). The most

Table 1. *Clinical and radiological data of 13 patients with syringomyelia associated with cervical disc disease*

Case	Age	Sex	Symptoms	Neurological findings	Herniated disc level or segmental kyphosis level	Grade of syrinx	Length of syrinx	Type of syrinx	Surgery	Syrinx resolution
1	56	F	Neck and arm pain	Hypoesthesia pain	C6-7	1	2 segments	Proximal	Microdiscectomy + fusion with cage	+
2	42	F	Weakness in the left arm	Motor weakness in the left arm	C5-6	2	1 segment	Distal	–	
3	33	F	Neck pain	Hypoesthesia	C4-5	3	3 segments	Distal	–	
4	32	F	Neck and arm pain	Motor weakness in the left arm	C5-6	1	1 segment	Distal	Microdiscectomy + fusion with cage	+
5	59	M	Neck pain	Hypoesthesia	C6-7	2	2 segment	Proximal	–	
6	47	F	Neck pain	Hypoesthesia pain	C4-5	3	2 segment	Distal	–	
7	32	F	Neck pain	Hypoesthesia	C5-6	2	2 segment	Distal	–	
8	54	F	Neck and arm pain	Motor weakness in the right arm	C5-6	2	2 segment	Distal	Microdiscectomy + fusion with cage	+
9	50	M	Pain in the left arm	Motor weakness and decreased reflex in the left arm	C6-7	1	2 segment	Proximal	Microdiscectomy + fusion with cage	+
10	38	F	Arm pain and paresthesia	Motor weakness and decreased reflex in the right arm	C4-5	3	3 segment	Distal	Microdiscectomy + fusion with plate	+
11	29	F	Neck pain	Hypoesthesia	C5-6	2	4 segment	Proximal	–	
12	21	M	Pain in the right arm	Hypoesthesia pain	C5-6	1	1 segment	Distal	–	
13	55	F	Paresthesia in the left arm	Hypoesthesia	C6-7	1	1 segment	Proximal	–	

common symptoms were neck pain and paresthesia in the upper extremities. In terms of neurological status, five patients had motor weakness in the upper extremities, while three patients demonstrated diminished deep tendon reflexes. Pathological reflexes, *Babinski* and *Hoffman*, or muscle atrophy and dissociated sensory loss in the upper extremities were not observed in any of the patients.

MRI findings

The average syrinx length in the study patients was two vertebral segments. Grade 1 and 2 syrinxes were observed in five patients each, while the remaining three patients had a Grade 3 syrinx. None of the patients had a massive cord expansion.

Figure 1. *Case 2 C5-C6 segmental kyphosis and small disc herniation Grade 1syrinx* **A** *T2W and* **B** *T1W MRI appearances. Kyphosis is 7° on standing X-Ray*

Based on the location of the syrinx and cervical disc disease, SCD was classified into two types: proximal and distal. In the proximal variety, cervical disc disease was located in the distal end of the syrinx; this type of localization was observed in five patients. In the distal type, disc disease was located in the proximal end of the syrinx, and was observed in eight patients **(Figures 1–4)**. Seven patients had disc herniation, whereas the remaining six had cervical angulation (local or segmental kyphosis) associated with disc herniation or disc degeneration, which was observed in MRI and confirmed on standing cervical radiographs. Hyperextension and hyperflexion radiographs were also evaluated. The average segmental kyphosis was 6.5° (range: 3–14°). Cervical instability, defined as horizontal vertebral displacement of >3 mm and/or angular displacement of >11°, was not observed in any case.

Surgical intervention

The indications for surgery were motor weakness in the upper extremities and intractable radicular or axial neck pain. Four patients underwent an anterior microdiscectomy and fusion with an intervertebral cage; in one patient, fusion was done using a plate following microdiscectomy. Postoperative syrinx resolution was observed in all these patients **(Figures 3, 4)**; the postoperative longitudinal diameter of the syrinx decreased by an average of 1 vertebral segment in all cases, while the MRI grade decreased by 1 grade in three cases. No procedure was done for the syringomyelia.

The primary reason for the small number of operated cases was the presence of relatively mild symptoms and findings in the non-operated cases. Among the non-operated cases, only four patients were followed up for an average duration of 6 months; however, no changes in syrinx, kyphotic angulation, and disc herniation were observed in the follow-up examinations.

Discussion

Cervical syringomyelia associated with cervical spondylosis is an uncommon occurrence[3, 4] and SCD is a further rare presentation[5–8]. *Ball* and *Little* reported a case of cervical syringomyelia along with Chiari malformation in which the syrinx resolution occurred following the cervical disc surgery[6]. Furthermore, the case reported by *Athanasiou* and *Magras* had multilevel cervical spondylosis[5]. In this study, we prefer the term "cervical disc disease" instead of "disc herniation" because nearly half of the patients had segmental kyphosis associated with cervical disc degeneration or herniation. In these cases, the kyphotic angulation was more prominent than the disc herniation. Kyphotic angulation usually develops after a disc herniation and disc space collapse, followed by subsequent vertebral subluxation[9–13]. If the segmental

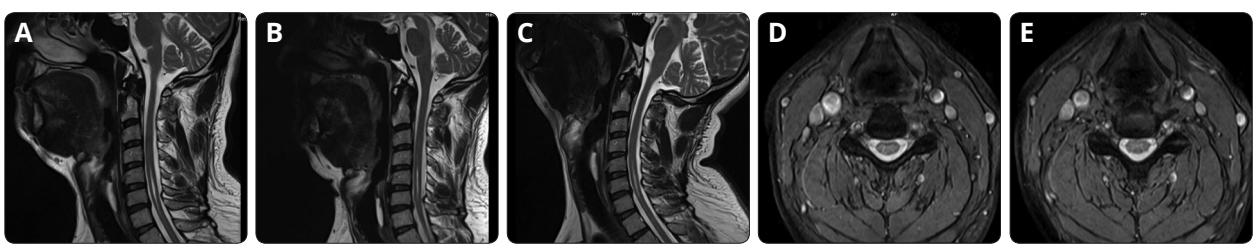

Figure 2. *Case 5 proximal type, Grade 2 syrinx and C6-C7 disc herniation.* **A** *Neutral* **B** *Flexion and* **C** *Extension sagittal MRI slices, and axial slices* **D** *Neutral and* **E** *Flexion. Obliteration of anterior subarachnoid space increases in both flexion and extension*

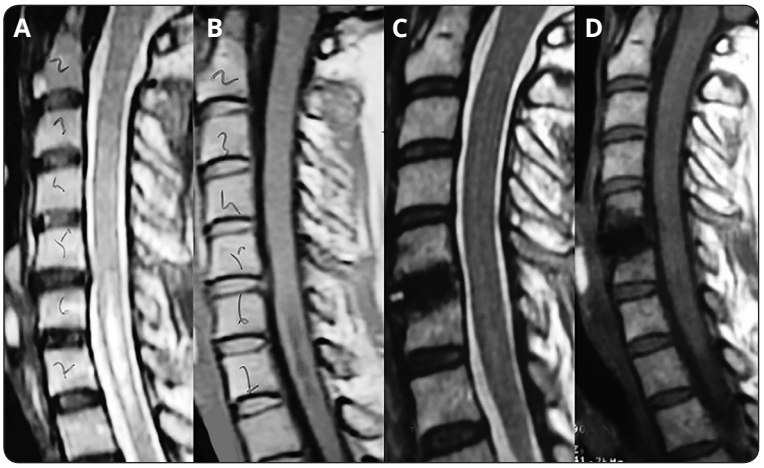

Figure 3. *Case 8 preoperative T2 weighted* **A** *and T1 weighted* **B** *MRI of distal type Grade 2 syrinx associated with C5-C6 disc herniation. Postoperative T2 weighted* **C** *and T1 weighted* **D** *MRI shows resolution of syrinx*

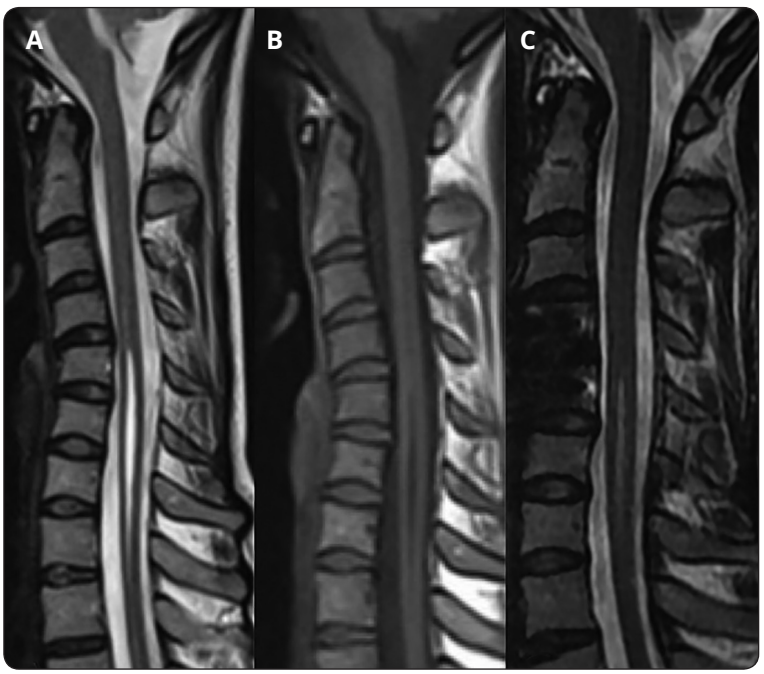

Figure 4. *Case 10 T2 weighted* **A** *and* **B** *T2 weighted sagittal MRI of distal type Grade 2 syrinx and C4-C5 segmental kyphosis. T2 Weighted* **C** *MRI following C4-C5 microdiscectomy and anterior cervical plating shows resolution of syrinx*

kyphosis is corrected (becomes straight or lordotic) on extension radiographs, it is considered a reversible kyphosis. Reversible kyphosis is usually related to disc degeneration and herniation and is treated surgically in the same way as cervical disc herniation.

In our case series, several clinical and radiological properties of SCD were observed. The clinical symptoms and findings were primarily due to disc herniation or segmental kyphosis. Neck pain and mild radicular pain were common, whereas muscle atrophy, dissociative sensory loss, and pathological reflexes, which are typically attributed to syringomyelia, were not observed. Cervical disc herniation usually occurs in the midline and obliterates the anterior cervical subarachnoid space totally or partially. In six of the cases, an increased kyphotic angulation of the cervical spine was detected in the MRI and standing radiographs, suggesting greater subarachnoid space obliteration in the standing and hyperflexed positions.

The most common location of disc disease was C5–6, followed by C6–7 and C4–5 levels. These levels are also known to have the narrowest subarachnoid space due to cervical spinal cord enlargement. The average syrinx length was two segments, which was less than that reported for syrinxes associated with Chiari malformation. We also graded the syrinx according to its visibility on MRI and the spinal cord expansion independent of the syrinx length. In our study, most of the syrinxes were classified as Grades 1 or 2. Therefore, a syrinx associated with disc herniation may manifest as a mild form of cervical syringomyelia; consequently, it may be easily overlooked or interpreted as idiopathic syringomyelia. Notably, the localization of the syrinx corresponded to the level of disc disease in our patients; in all cases, the level of disc disease was either on the upper or the lower end of the syrinx. Presumably, this situation is associated with the pathophysiology of syrinx development; the distal type syrinxes typically resemble syringomyelia associated with Chiari malformation, whereas the proximal type resembles syringobulbia or hydrocephaly associated with Chiari malformation.

The pathogenesis of syringomyelia is still unclear probably due to the multiple mechanisms responsible for syrinx development[14–16]. Many theories have been proposed to explain the development of syringomyelia associated with Chiari malformation[15, 16]. Some authors also believe that syringomyelia associated with spinal pathology should be classified as primary spinal syringomyelia[17, 18]. Recent studies have shown that cerebrospinal fluid (CSF) flow impairment is responsible for the development of primary spinal syringomyelia. The obliter-

ation of the spinal subarachnoid space reduces its compliance and increases the subarachnoid pulse pressure proximal to the obstruction during the cardiac systole[16]. The subarachnoid pressure waves occur as a result of brain expansion during cardiac systole and elevation of CSF pressure; this pulse pressure drives the CSF through the Virchow–Robin and the extracellular spaces, and eventually into the syrinx[14, 16, 19]. Furthermore, the observation of spinal cord edema and swelling in the pre-syrinx state confirms the transparenchymal passing of CSF[20–22]. This theory satisfactorily explains the development of syringomyelia associated with traumatic injury and inflammatory conditions in which the CSF pathway is totally obstructed[4]. However, studies on CSF flow and CSF pulse wave revealed two properties of CSF flow dynamics. First, anterior and posterior spinal subarachnoid spaces have different flow patterns[23–25]. *Mauer* et al. showed that in cardiac-gated phase-contrast MRI of CSF flow, the flow signal was located ventrally in the cervical region and shifted dorsally at the craniocervical junction[24]. It was also clearly located dorsally in the midthoracic spine and shifted ventrally again at the level of the medullary cone. This finding correlates well with the assumption that the spinal cord traverses closer to the concave side of the spinal curvatures, just as in a bow and string, i.e., in the cervical region, the anterior subarachnoid space is more prominent than the posterior subarachnoid space. Second, partial obstruction of subarachnoid space is sufficient for the development of syringomyelia[23]. Partial obstruction of CSF causes delays in the CSF waveform[25]; the change in the timing of the CSF waveform with respect to the cardiac cycle drives more fluid along the perivascular space and into the spinal cord. Indeed, partial obstruction of subarachnoid space was observed in most of the reported cases, as well as in our study. This obstruction could be increased by flexion, extension, and standing **(Figure 2)**. Also, a segmental kyphosis of >7.5° could increase intramedullary spinal pressure[18].

More recently, Goel proposed that Chiari malformation was a potential secondary manifestation of atlantoaxial instability and could be with only atlantoaxial fixation[26]. Later, his group reported nine cases of idiopathic cervical syringomyelia treated by atlantoaxial fixation[27]. It is well known that atlantoaxial instability may cause cervical syringomyelia. Segmental kyphosis aggravates the compression of the anterior subarachnoid space in flexion and standing positions. This is similar to the mechanisms described for instability observed with syringomyelia[26, 27]. We also observed decreased anterior subarachnoid space in both extension and flexion positions in dynamic MRI scans, although cervical instability was not detected in any patient.

There are reports describing a relationship between syrinx and obstruction level in arachnoiditis-related syringomyelia. *Klekamp* et al. reported that in all cases, arachnoiditis was at a level close to the syrinx[28]. *Holly* and *Batzdorf* also found that the cause of syrinx was located in all cases but one[29]. Mauer et al. noted that in 30% of the cases, the CSF blockage level was at either the upper or lower end of the syrinx, while in 50% of the cases, it was within the syrinx levels[24]. Most of our patients had syringomyelia distal to the obstruction, whereas five patients had syrinxes proximal to the herniated disc level. Josephson et al. conducted an experimental study involving the dural sac construction model and observed that the syrinxes developed proximally and distally in the partial obstruction; in contrast, proximal cysts were developed in the total obstruction group[30]. Therefore, in the case of distal syringomyelia associated with cervical disc disease, CSF pressure should be higher in the region distal to the obstruction because it is presumed that cerebellar tonsils prevent the CSF movement from the spinal to the cranial space during coughing, sneezing, or straining; consequently, the CSF pressure increases[14–16]. Similarly, cervical disc herniation obliterating the subarachnoid space partially may act as a one-way valve, increasing the CSF pressure distally and causing a distal type syrinx. We believe that SCD is a mild form of syringomyelia. However, different mechanisms may be responsible for the development of the different types of syrinx.

In this report, only five patients underwent surgical treatment, and some degree of syrinx resolution was observed in the follow-up examinations of all operated cases. Furthermore, some non-operated cases were followed up clinically and radiologically and no progression of syringomyelia was observed. If a patient presents with cervical disc disease and cervical syringomyelia, it is worth pondering whether this coexistence is coincidental or causal. Our inclusion criteria described above supported the idea of a causative relationship between cervical disc disease and syringomyelia. First, the presence of subarachnoid obstruction due to disc herniation or kyphotic angulation may cause syringomyelia. Second, the obstruction was located in the upper or lower end of the syrinx cavity, a situation that is often observed in spinal syringomyelia. Third, some resolution of syringomyelia occurred in all operated cases, as seen in previously reported cases[5-8]. However, some aspects must be considered while interpreting our results. We did not perform CSF flow studies in our patients, and cervical disc disease could be a factor that secondarily contributed to the syringomyelia which was already present.

Conversely, in many cases, cervical disc herniation and segmental kyphosis may be associated with cervical cord compression and myelopathy but without syringomyelia. There are several explanations for this situation. First, in some cases, the syrinx is small or mild with an average length of two segments, so these cases are often not reported[8]. Second, in experimental studies using the extradural construction model, syrinx cavities take 8–11

weeks to develop[23]. If in human beings, syrinx cavities developed within a similar duration, patients who are treated within this period and whose CSF pathways are restored, would not suffer from syringomyelia. However, our suggestions about the causative relationship between cervical disc disease and syringomyelia are speculative, and more observations of similar or incidental syringomyelic cases are needed for confirmation.

Conclusion

SCD is a very rare presentation and syringomyelia in these cases is in a very mild form. In such cases, clinical findings are more associated with cervical disc disease. Cervical disc herniation usually occurs in the midline and herniation or kyphotic angulation cause an obliteration of the anterior subarachnoid space, which is presumably the cause of syrinx development. Based on the MRI findings and the disc disease level, SCD has two main types (proximal and distal types), and probably different mechanisms are responsible for the development of the different types. SCD can be adequately managed surgically using the standard treatment for cervical disc herniation or segmental kyphosis and decompression of obliterated anterior cervical subarachnoid space, resulting in syrinx resolution; no additional treatment for the syringomyelia is necessary. However, a causal relationship between cervical disc disease and syringomyelia cannot be established.

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