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Neck pain and Headache Complicated by Persistent Syringomyelia After Foramen Magnum Decompression for Chiari I Malformation: Improvement with Multimodal Chiropractic Therapies

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		Patient: agnosis: aptoms: ication: cedure: ecialty:	Female, 62-year-old Cervical spondylosis • syringomyelia Headache • neck pain • upper extremity numbness and weakness — Exercises • soft tissue manipulation • spinal mobilization • therapeutic ultrasound Chiropractic			
Objective: Background: Case Report: Conclusions:			 Rare disease Patients with Arnold-Chiari Malformation I (CM-I) treated with foramen magnum decompression (FMD) can have ongoing neck pain, headaches, and other symptoms complicated by persistent syringomyelia, yet there is little research regarding treatment of these symptoms. A 62-year-old woman with a history of residual syringomyelia following FMD and ventriculoperitoneal shunt for CM-I presented to a chiropractor with progressively worsening neck pain, occipital headache, upper extremity numbness and weakness, and gait abnormality, with a World Health Organization Quality of Life score (WHO-QOL) of 52%. Symptoms were improved by FMD 16 years prior, then progressively worsened, and had resisted other forms of treatment, including exercises, acupuncture, and medications. Examination by the chiropractor revealed upper extremity neurologic deficits, including grip strength. The chiropractor ordered whole spine magnetic resonance imaging, which demonstrated a persistent cervico-thoracic syrinx and findings of cervical spondylosis, and treated the patient using a multimodal approach, with gentle cervical spine mobilization, soft tissue manipulation, and core and finger muscle rehabilitative exercises. The patient responded positively, and at the 6-month follow-up her WHO-QOL score was 80%, her grip strength and forward head position had improved, and she was now able to eat using chopsticks. This case highlights a patient with neck pain, headaches, and persistent syringomyelia after FMD for CM-I who 			
	Ke	ywords:	Arnold-Chiari Malformation • Chiropractic • Exe Therapy, Soft Tissue	rcise Therapy • Manipulation, Spinal • Neck Pain •		
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Background

Arnold-Chiari malformation type I (CM-I), the most common type of Arnold-Chiari malformation, is defined as a downward descent of the cerebellar tonsils at least 5 mm inferior to the level of the foramen magnum [1]. Between 30% and 80% of patients with Arnold-Chiari malformation also have syringomyelia [2], an abnormal fluid-filled cavity within the spinal cord [3]. While symptoms of CM-I are varied, patients with syringomyelia can present with sensory symptoms, hand weakness, scoliosis, neck and back pain, and headaches [2]. While these patients often undergo foramen magnum decompression (FMD) surgery, there has been little research on treatment of neck pain and other symptoms that can persist along with syringomyelia after surgery.

About 93% of patients with a syrinx undergoing FMD have a reduction in the size of the syrinx [4]. However, among those treated surgically for arachnoid cyst with syrinx, symptoms can improve even with minimal or no change in the syrinx size [5]. Persistence of the syrinx and associated symptoms are thought to relate to ongoing impairment of cerebrospinal fluid (CSF) flow [6,7]. In those patients without improvement in CSF flow following surgery, symptoms can persist, including weakness in 100% of patients, vision symptoms in 84%, dizziness in 78%, neck pain in 66%, and headache in 50% [7].

Half of patients with persistent or recurrent syringomyelia after FMD are treated conservatively and can experience improvement 1 to 2 years following their original surgery [5]. However, the remainder of patients undergo another surgical procedure, at a mean 15 months after surgery [5]. CSF diversion is the most commonly utilized procedure among these patients and involves a syringo-subarachnoid shunt [5]. Another method of CSF diversion is ventriculoperitoneal shunting [4,5]. Revision surgery is another option which is only rarely considered, when cerebellar tonsillar herniation persists after surgery [5].

There is a limited understanding of the optimal conservative treatment options for patients with neck pain or other symptoms and persistent syringomyelia after FMD [4,8]. One article described that pain medications and rest were commonly prescribed for these patients [9]. Most research regarding manual therapies has been devoted to CM-I prior to or instead of surgical decompression, mostly in the form of case reports. These have shown a variety of responses to care, including negative [10-13], equivocal [14,15], or positive outcomes [16-22]. As of July 4, 2022, we are aware of only 2 cases which reported the use of manual therapies for a patient following FMD, both yielding a positive outcome [8,23].

The literature regarding rehabilitative and manual therapies for syringomyelia in general is also limited [24]. However, according

to one survey, patients reported that physical therapy was helpful with symptom management [24]. Other case reports have noted improvement in syringomyelia-related symptoms with various conservative treatments, including cervical traction, joint mobilization, soft tissue manual therapies, posture correction, and acupuncture [25-28].

Given the limited research on treatments for neck pain, headache, and other symptoms persistent in conjunction with syringomyelia after FMD for CM-I, we present a patient who responded positively to a multimodal chiropractic treatment regimen.

Case Report

Patient Information

A 62-year-old woman presented to a chiropractor in a multidisciplinary office with frequent severe neck pain and occipital headache and intermittent numbness and weakness of the bilateral upper limbs, more prominently affecting the right side. The pattern of numbness chiefly affected her right deltoid region to the lateral arm, forearm, and the hand, diffusely. The patient's self-reported pain intensity was a 9 out of 10 on the numeric pain rating scale. She noted being unable to use chopsticks with her right hand due to numbness, weakness, and hand deformity. Her World Health Organization Quality of Life Score (WHO-QOL) was 52%.

Her medical history was significant for CM-I with FMD, persistent syringomyelia, and epilepsy. She denied having any difficulty swallowing or speaking, hoarseness, sleep apnea, and gastrointestinal or cardiovascular symptoms. She was a homemaker, nonsmoker, and did not drink alcohol. Her family history was negative for scoliosis, cancer, connective tissue disease, and any neurological disorders.

Thirty years prior to her presentation to the chiropractor, at age 32, she had episodic nausea and dizziness that occurred about once per week depending on activity and lasted about 5 min. She was initially diagnosed with benign paroxysmal positional vertigo and treated with physiotherapy.

Sixteen years prior to presentation, at age 46, she began to have sudden loss of balance and unprovoked episodes of epilepsy. She then visited a neurosurgeon and underwent magnetic resonance imaging (MRI), which revealed herniation of the cerebellar tonsils through the foramen magnum greater than 5 mm and syringomyelia, and CM-I was accordingly diagnosed. Her primary care provider and neurosurgeon recommended surgery, and she underwent FMD that year. The neurosurgeon performed a suboccipital craniectomy and laminectomy of C1 and C2 and simultaneously implanted a ventriculoperitoneal shunt. Following surgery, her neck pain, headache, and dizziness improved but were not completely alleviated. At this time, she still had occasional epileptic seizures, and these were managed with oral sodium valproate (200 mg) and levetiracetam (500 mg). At the time of presentation to the chiropractor, she had no recent seizures.

Six years prior to presentation to the chiropractor, at age 56, she began to experience bilateral upper limb weakness, worse on the right side, and revisited her neurosurgeon, who ordered a brain and cervical spine MRI, and computed tomography (CT) angiogram of the head without and with contrast. The CT angiogram exhibited small focal calcified plaque causing mild (20%) stenosis at the left carotid bulb, which was deemed to not be a source of her symptoms. The MRI revealed persistent syringomyelia; however, the surgeon reportedly determined that the patient was not a candidate for further surgeries and instead referred the patient to physical therapy.

At physical therapy, the patient recalled performing neck exercises and cervical traction; other specific details, however, were unclear. The patient also received acupuncture treatment. None of these therapies improved her symptoms, which slowly became worse.

She reported her episodes of neck pain and dizziness in the month prior to presenting to the chiropractor would occur 4 to 5 days per week, 3 to 4 times per day, depending on activity. She visited an orthopedic surgeon, who ordered full spine radiographs, which revealed a reversed cervical lordosis and degenerative changes of the cervical zygapophyseal and uncovertebral joints, suggestive of cervical spondylosis, as well as 3 mild scoliotic curvatures (Figure 1). The ventriculoperitoneal shunt appeared intact. The patient was prescribed a nonsteroidal anti-inflammatory drug and tramadol, 100 to 200 mg per day, and again was referred for physical therapy with cervical traction.

Because the patient had only temporary relief with these therapies previously and had progressively worsened over the past 6 years, she presented to a chiropractor for a second opinion.

Clinical Findings

The patient was brought by her husband to the chiropractor. Upon examination by the chiropractor, she was in distress and crying. She demonstrated a slow gait and held her husband's arm to help maintain her balance while walking. She was slow to rise from a seated position and required the use of her arms to help push herself into a standing position. She had a kyphotic posture with her head and neck positioned anteriorly deviated to the right side. Muscle atrophy was evident, affecting the right upper extremity and hand and bilateral calf muscles. The patient also had a right claw hand deformity. Her active cervical range of motion was limited by pain to 10° of extension (normal >70°) and 40° of cervical rotation bilaterally (normal >80°), which appeared to trigger a spasm of the neck muscles. Although her lumbar range of motion was full, flexion caused a dull ache in the lower back. Palpation identified hypertonicity of the sternocleidomastoid, trapezius, rhomboid, and levator scapulae muscles bilaterally and tenderness at C2. A cranial nerve examination was normal, no overt cerebellar signs were noted (eg, intention tremor, nystagmus), and muscle stretch reflexes were normal bilaterally. Motion palpation identified restriction of the C4/5, C6/7, T2/3, and T7/8 segments.

The patient had diminished sensation to light touch and pain in the right lateral arm, forearm, and hand; however, temperature sense was not assessed. Her motor strengths for right shoulder abduction, arm flexion, extension, and wrist flexion and extension were 4 out of 5 (Medical Research Council scale) for each action. The patient also demonstrated lower extremity weakness with 4 out of 5 strength for hip flexion and extension, quadriceps extension, and plantar flexion bilaterally. Her grip strength was diminished bilaterally, as measured using a digital dynamometer (Jamar Plus, JLW Instruments, USA), which yielded 4.3 kg for the left hand (normal 20.7 ± 4.6) and 8.7 kg for the right (dominant) hand (normal 25.0 ± 4.6).

The chiropractor's differential diagnosis included persistence of the patient's syringomyelia, concurrent degenerative spondylosis causing cervical and lumbar radiculopathy, and cervical facet arthropathy related to scoliosis. The chiropractor also considered the possibility that the syringomyelia extended further inferiorly into the thoracic spinal cord. Given the patient's complex medical history, including scoliosis and history of CM-I, syringomyelia, and previous FMD, which are precautions to certain manual therapies used by chiropractors, as well as her upper and lower extremity neurologic deficits, gait abnormality, and progressive worsening despite conservative care, the chiropractor ordered whole spine MRI, which was conducted that week.

The MRI revealed the presence of post-surgical FMD changes and syringomyelia extending from C1 to T9 (Figure 2). There was also cervical spondylosis with degenerative disc changes from C3 to C7. Most notably, at C4/5, there was moderate narrowing of the central canal and right neuroforamen, caused by disc displacement (Figures 3, 4). At C5/6, there was mild narrowing of the left neural foramen due to uncovertebral joint hypertrophy. At C6/7, there was a small disc osteophyte complex with an annular fissure causing mild canal stenosis and moderate right neuroforamen stenosis (Figure 4).



Figure 1. Full spine radiographs taken at an outside facility prior to presentation to the chiropractor. In the lateral view (A), the patient is noted to have an anterior head position with loss of the cervical lordosis. There is evidence of suboccipital craniectomy and laminectomy of C1 and C2. In the anteroposterior view (B), 3 scoliotic curvatures are evident, including a 20° levoconvex cervicothoracic curvature (upper Θ), a 20° dextroconvex lumbar curvature (lower Θ), and 13° levoconvex thoracic curvature (not shown). The visible span of the ventriculoperitoneal shunt appears intact in both views (arrows).

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Figure 2. Sagittal cervical spine magnetic resonance imaging. Most evident on the T1-weighted image (A), bone defects are noted around the foramen magnum and posterior elements of C1 and C2 (bracket; }), suggestive of suboccipital craniectomy and laminectomy of C1 and C2. Also evident on the T1-weighted and T2-weighted image (B) is a spinal cord syrinx (arrowheads). At the lowermost extent of the image, the upper thoracic spinal canal is not visible, given the patient's scoliotic curvature at this region.

There was no evidence of dens retroflexion, pseudomeningocele, or other pathology in the vicinity of the prior FMD. The thoracic spine did not demonstrate any canal stenosis, with the only abnormality being the syrinx, which extended to the level of T9. The lumbar spine was relatively normal, showing only mild posterior disc bulges at L3/4, L4/5, and L5/S1, without nerve root compression.

The chiropractor considered the whole spine MRI findings to be most consistent with symptoms related to the patient's cervicothoracic cord syrinx. In addition, the cervical spondylosis was considered to potentially account for some of the patient's neck pain and upper extremity symptoms. In contrast, the lumbar spine findings were deemed inconsistent with a clinical diagnosis of lumbar radiculopathy. Accordingly, treatment focused on the cervical and thoracic regions rather than the lumbar spine. The patient consented to a chiropractic treatment plan which began at a frequency of 3 visits per week and included a variety of therapies (Table 1). Gentle manual spinal mobilization was administered to the mid to lower cervical and upper thoracic segments to alleviate muscle hypertonicity and restore spinal mobility (Figure 5). This treatment was well tolerated by the patient. Therapeutic ultrasound and instrument-assisted soft tissue manipulation (also called *gua sha*) using a massage tool (Strig, Korea) was applied to further alleviate muscular hypertonicity and reduce pain (Figure 6).

After the first week of care, the patient reported she was able to turn her head better without an increase in pain, yet her baseline pain was still severe at 7 to 8 out of 10. She also reported improvement in her grip strength, which improved to 4.9 kg on her left hand; however, her right hand strength remained similar at 8.8 kg.



Figure 3. Axial T2-weighted cervical magnetic resonance image at C4/5. There is moderate narrowing of the central canal due to an eccentric disc osteophyte complex on the right (arrow). There is also moderate narrowing of the right neural foramen due to disc herniation (arrowhead).

After the first month of treatment, the patient's reported neck pain severity decreased to 4 out of 10. Given the patient's improvement, her visit frequency was reduced to once per week. After the second month of treatment, all numbness symptoms were resolved. The patient had stopped taking tramadol and was now able to use chopsticks. Low-impact isometric core muscle stabilization training was added, with the goal of strengthening the paraspinal muscles and spine and improving posture. This was accomplished by having the patient sit in a robotic device that rotated slowly at different angles of inclination while the patient maintained a neutral spine position; this was performed for 10 min per visit (AllCore360° [29], USA; Figure 7)



Figure 4. Right oblique sagittal proton density turbo spin echo cervical magnetic resonance image. Moderate narrowing of the right C4/5 and C6/7 neural foramina due to disc herniation is evident (arrows).

Hand exercises were also added, which were conducted using robotic gloves (Ober, China). These were rented to the patient for home use for 15 to 30 min per day (Figure 8). This type of robotic glove is used to augment hand motion for patients with weakness or functional deficits of the hand and fingers and has been previously studied for use with patients with spinal cord injury [30]. In the setting used, the device matched the patient's own finger movement and provided mechanical assistance. The degree of assistance was reduced as the patient

Table 1. Timing and details of multimodal chiropractic and rehabilitative therapies.

Intervention and description	Timing and frequency	Duration
Gentle manual spinal mobilization applied to the mid-to-lower cervical spine and upper thoracic spine	Month 1: 3×/week Months 2-4: 1×/week Months 4-6: 1×/month	2-5 min
Therapeutic ultrasound applied to the cervical and upper thoracic spine	Month 1: 3×/week Months 2-4: 1×/week Months 4-6: 1×/month	8 min
Instrument-assisted soft tissue manipulation applied to the cervico-thoracic musculature	Month 1: 3×/week Months 2-4: 1×/week Months 4-6: 1×/month	5-10 min
Isometric core muscle stabilization training (AllCore360°, USA)	Months 2-4: 1×/week Months 4-6: 1×/month	10 min
Hand exercises using robotic gloves (Ober, China)	Months 2-6: daily (at home)	15-30 min

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Figure 5. Cervical mobilization demonstration. The provider applies gentle posterior to anterior pressure to the left mid-cervical spine while the opposite hand ipsilaterally laterally flexes and slightly rotates the head and stabilizes it against the head rest. This procedure is repeated on the contralateral side as well as in the lower cervical and upper thoracic spine (not shown).



Figure 7. Core strengthening exercise demonstration. The patient sits in this device (Allcore360°, USA) that rotates slowly at different angles of inclination, while the patient isometrically contracts the core muscles to remain in the neutral, seated posture while holding a large ring in the hands and ball between the knees.



Figure 6. Instrument-assisted soft tissue manipulation. The provider applies emollient to the skin surface then gently strokes a massage tool (Strig, Korea) across the skin surface (arrows). This treatment was applied in the region of the upper trapezius (shown), as well as the rhomboids and levator scapulae muscles bilaterally.



Figure 8. Hand strength rehabilitation demonstration. The patient is wearing robotic gloves (Ober, China) and conducts fine motor activities for up to 30 min per day. Also shown is the control console that allows the level of mechanical assistance to be altered. Image from EC.

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regained strength. The patient was recommended to perform tasks such as opening and closing the hands and squeezing a ball with the aid of the gloves.

At the 4-month follow-up, the patient no longer had neck pain, and her other symptoms were now mild or absent. Her active cervical spine range of motion improved to 40° of extension and 60° of cervical rotation bilaterally. Owing to her continued improvement, the patient's visit frequency was reduced to once per month.

At the 6-month follow-up, the patient was pain free, with no neck pain or headaches. Her WHO-QOL score improved to 80%. Measurement of grip strength showed improvements to 5.8 kg (left hand) and 10.0 kg (right hand), and upon visual inspection, her anterior head posture had improved. The patient still had residual slowness of gait and limitations of strength, and although she could eat more easily, she still had difficulty cooking due to an inability to hold pots and pans. Because of these subjective and objective measures of improvement, the patient chose to remain in chiropractic care with monthly appointments.

The patient provided written consent for the publication of this case report and accompanying images. There were no adverse events in relation to the multimodal chiropractic interventions provided.

Discussion

This case illustrates an older woman with persistent syringomyelia and persistent and progressively worsening neck pain, headaches, and neurological deficits after FMD for CM-I years prior. The chiropractor played a pivotal role in ordering imaging and implementing a cautious but effective conservative treatment regimen which led to significant improvements in the patient's symptoms and quality of life.

The patient's symptoms, including occipital headache, neck pain, and upper extremity sensory and motor deficits, were typical for CM-I and persistent syringomyelia [2,7,31,32]. However, it is possible that overlapping degenerative cervical spine changes contributed to her neck pain and upper extremity neurologic deficits. While many individuals in their 60s will have some degree of degenerative changes [33], recent research has shown that the severity of cervical foraminal stenosis correlates with the severity of upper extremity symptoms [34]. In the current case, the patient had predominantly right-sided cervical foraminal stenosis at C4/5 and C6/7, which could be expected to correspond to a C5 and C7 radiculopathy, respectively. However, even if these degenerative changes were clinically relevant, they would account for only some, but not all, of the motor and sensory deficits in the patient's upper extremities [35].

This case is similar to the 2 previously published cases describing conservative, rehabilitative, and manual therapies for patients with persistent symptoms after FMD [8,23]. In one case similar to ours, with persistent symptoms and syringomyelia after FMD, a physical therapist utilized a suboccipital manual release to alleviate neck pain, in conjunction with several spine stabilizing and strengthening exercises, which led to improvements in pain, gait, and balance [8]. In a similar case of headaches after FMD, but without syringomyelia, spinal manipulation was utilized with success; however, this treatment was modified to avoid any cervical rotation [23].

There are several mechanisms whereby the treatment provided in the present case could have led to a reduction in symptoms. We suggest that the cervical spine mobilizations may have alleviated radicular components of the patient's neck pain, in accordance with a recent systematic review that found manual therapies were effective in reducing cervical radiculopathy [36]. However, this would only partially explain the improvement from her spectrum of symptoms (eg, not the broader pattern of neurologic deficits). We therefore also suspect that rehabilitative core exercises had a beneficial effect on the patient's posture, while hand exercises were responsible for her improvement in grip strength and functional use of the hand.

It is also possible that improved postural changes had a beneficial impact on CSF flow and subsequent syrinx-related symptoms. However, this hypothesis could not be confirmed as it would require serial cine phase-contrast MRI to examine the CSF velocity and flow [6], which would not be clinically justified or practical. Research on this topic is limited; however, a recent study found an association between a loss of cervical lordosis and CM-I and suggested that anteflexion of the cervical cord could impair CSF flow, thus promoting syrinx formation [37]. In the present study, anterior head posture was improved at the 6-month follow-up.

Clinicians considering a rehabilitative or manual therapy approach to patients with persistent symptoms and syringomyelia after FMD should do so with caution. In the present case and those previously published, providers took a measured, gentle approach to therapy, avoiding any excessive cervical rotation [8,23]. While there is limited research regarding the safety or harm of spinal manipulation in the presence of cervical syringomyelia (a cause of myelopathy), previous case reports have described exacerbation of degenerative cervical myelopathy with high velocity spinal manipulative therapy [38,39]. Accordingly, any rehabilitative or manual therapy approach for such patients should also be done in with consideration of the patient's neurological status, imaging findings,

their preferences toward treatment, and informed consent of other available options. In the present case, high-velocity lowamplitude manipulation, a common chiropractic treatment, was altogether avoided in preference for more gentle mobilization techniques, soft tissue manipulation, and exercises.

Future research should examine the safety and effectiveness of multimodal chiropractic and rehabilitative treatments for persistent symptoms and syringomyelia after FMD. Given that this patient presentation may be uncommon, this type of research may be more feasible in the form of case series or retrospective chart review studies. Clinicians should consider serial measurements to track patient progress, in addition to those obtained in the present study, such as patient-reported outcome assessments/questionnaires, grip strength, and markers of posture and balance.

Limitations

First, as a single case, the results shown may not be generalizable to all patients following FMD. It is possible that the patient's cervical spondylosis played a role in her response to manual therapies and exercise. In contrast, other patients with persistent symptoms after FMD may not have concomitant spondylosis and thus may not respond to a similar treatment program. Second, several tests were unavailable in the present case that could have aided in objective measures of improvement, such as balance testing and functional gait assessment. Further, images from before the patient's surgery were unavailable upon request and only imaging after surgery could be identified. Finally, our literature search for similar cases describing the use of manual therapies for patients with

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persistent symptoms after FMD included PubMed, the Index to Chiropractic Literature, and Google Scholar. This search identified only a conference abstract and case report, which was not indexed in PubMed [8,23]. Accordingly, these references have limited detail and/or reliability.

Conclusions

This case demonstrates an adult woman with recalcitrant neck pain and headache complicated by a chronic, persistent cervicothoracic cord syrinx after FMD for CM-I who improved after a combination of conservative chiropractic and rehabilitative therapies. Despite the success in this case, there remains a limited, low level of evidence supporting the therapies used in this case for patients with neck pain, headache, or other symptoms and syringomyelia after FMD for CM-I. These treatments should not be broadly applied to other similar patients and should be approached with caution on a case-by-case basis. Further research should examine the safety and efficacy of such therapies in this patient population.

Department and Institution Where Work Was Done

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Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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