

EXTENSIVE CERVICOTHORACIC SYRINGOMYELIA ASSOCIATED WITH BORDERLINE TONSILLAR DESCENT: A CASE REPORT OF POSTERIOR FOSSA DECOMPRESSION

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ABSTRACT

Chiari malformation type I is a structural disorder characterised by herniation of the cerebellar tonsils through the foramen magnum, frequently associated with syringomyelia. Although a descent of at least 5 mm is conventionally required for diagnosis, lesser degrees may be clinically significant when cerebrospinal fluid dynamics at the craniocervical junction are disturbed. A 40-year-old female patient presented with cervical and left shoulder pain persisting for two months, without motor or sensory deficit. Cervical magnetic resonance imaging revealed a crowded posterior fossa, tonsillar herniation of only 4.5 mm below the basion-opisthion line, reduced subarachnoid spaces at the craniocervical junction, and a 130 mm hydrosyringomyelia cavity extending from the odontoid process to T2, 9×12 mm at its widest, with cord expansion and perisyrinx T2-weighted hyperintensity suggestive of chronic spinal cord injury. Surgery was indicated by the extent of the syrinx and the risk of irreversible cord injury, rather than pain alone. The patient underwent posterior fossa decompression consisting of suboccipital craniectomy, C1 posterior arch excision, and duraplasty. The early postoperative course was uneventful; a minor superficial wound problem was managed conservatively during follow-up. At 18 months her pain had improved, the neurological examination was unchanged, and serial imaging showed sustained decompression with syrinx regression. This case suggests that craniocervical morphology and cerebrospinal fluid flow, rather than the millimetric extent of tonsillar descent alone, should guide assessment and the decision to operate.

Keywords: Case report, Chiari malformation type I, posterior cranial fossa, surgical decompression, syringomyelia

INTRODUCTION

Chiari malformation type I (CM-I) is a structural disorder of the posterior cranial fossa characterised by the herniation of the cerebellar tonsils through the foramen magnum (1, 2). Hans von Chiari first described and classified these malformations according to the degree of tonsillar herniation at the end of the 19th century (2, 3). CM-I is the most prevalent subtype, with a reported incidence ranging from 1 in 1,000 to 1 in

5,000 and is relatively uncommon in the adult population (2, 4). Reported prevalence, age at presentation and access to specialist care also vary across sociodemographic groups (5). Several pathophysiological mechanisms have been proposed: the predominant hypothesis attributes CM-I to an aberrantly small posterior cranial fossa in which the neural contents are crowded and herniate into the spinal canal (3); others suggest that excessive cerebellar growth during the embryonic period



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displaces the tonsils caudally (3); and a further theory implicates mechanical downward traction of the brainstem and cerebellum by the filum terminale (3). Clinically, the condition spans a wide spectrum extending from entirely asymptomatic individuals to patients with serious neurological impairment (2, 6). Although nausea, dysphagia, apnoea, clonus, cerebellar signs, drop attacks, muscle atrophy, and dysphonia have all been reported, cervical pain and headache remain the most frequently observed presenting complaints (1, 2, 6, 7).

Diagnosis relies on a combination of clinical history, neurological examination, and magnetic resonance imaging (MRI) of the craniocervical junction (3, 8). A tonsillar descent of at least 5 mm below the basion-opisthion line is conventionally applied as the radiological criterion. However, this threshold is arbitrary and does not by itself determine clinical relevance: herniations of 3-5 mm may be symptomatic when accompanied by syringomyelia, obliteration of the retrocerebellar cerebrospinal fluid (CSF) spaces, or an unfavourable posterior fossa morphology (7-9). Surgical intervention is the primary treatment, and accurate patient selection combined with an appropriate operative strategy is associated with a high rate of symptomatic improvement which is consistent with the potentially reversible nature of the condition (3). Nevertheless, the natural history of CM-I remains incompletely understood, and ongoing clinical and imaging research continues to refine current knowledge of the disease (2). We present an adult patient in whom an extensive cervicothoracic syrinx was associated with a tonsillar descent of only 4.5 mm, in order to illustrate that the clinical significance of Chiari-related pathology cannot be judged by the millimetric extent of tonsillar herniation alone, and that management should follow an individualised assessment integrating the clinical findings, posterior fossa crowding, craniocervical junction anatomy, syrinx burden and the probable disturbance of CSF dynamics.

CASE REPORT

A 40-year-old female patient presented to Trakya University Hospital Department of Neurosurgery with complaints of neck pain and left shoulder pain persisting for two months. Her medical history included hypothyroidism managed with levothyroxine (Euthyrox; Merck, Darmstadt, Germany) at 25 µg daily, increased to twice daily on weekends; two prior caesarean sections; and a previous right wrist surgery. Her family history was unremarkable.

On neurological examination the patient was alert and fully orientated, and the cranial nerve examination was intact. Motor examination revealed no focal weakness, with muscle strength graded 5/5 in all extremity muscle groups. Sensory examination, including light touch, pinprick, and vibration, was unremarkable, and there was no suspended or dissociated sensory loss. Mild hypertonia was noted in the extremities. Deep tendon reflexes were normoactive and symmetrical (2+/2+ in both upper and lower extremities), plantar responses were flexor bilaterally, and neither the Hoffmann's sign nor clonus could be elicited.

Cerebellar examination demonstrated no dysmetria or dysidiadochokinesia, and the Romberg test was negative. Gait was normal. Spurling's test was negative bilaterally. The patient described occasional stress urinary incontinence triggered by coughing and sneezing; this symptom was not investigated further with urological, gynaecological or urodynamic assessment.

Laboratory investigations revealed no abnormalities. A cervical MRI study, obtained at Trakya University Faculty of Medicine (14.05.2024), demonstrated a crowded posterior fossa with the cerebellar tonsils herniated 4.5 mm below the basion-opisthion line. A decrease in the anterior and posterior subarachnoid spaces with cord expansion was observed at the C2 vertebral level. In addition, a hydrosyringomyelia cavity extending from the level of the odontoid process to T2, spanning a 130 mm segment and reaching a maximum diameter of 9×12 mm at C2, was identified, causing significant cord expansion and exhibiting T2-weighted hyperintensity in the spinal cord surrounding the syrinx, suggestive of chronic spinal cord injury (Figure 1). The tonsillar descent was therefore below the conventional 5 mm threshold, and the diagnosis was not based on this single measurement. CM-I with extensive cervicothoracic hydrosyringomyelia was diagnosed on the combined basis of posterior fossa crowding, effacement of the subarachnoid spaces at the craniocervical junction suggestive of impaired CSF flow, the craniocaudal extent of the syrinx, the associated cord expansion, the perisyrinx signal change, and the clinical presentation.

The principal indication for surgery was not the pain itself. The patient had neither motor nor sensory deficit, and her symptoms in isolation would not have mandated operative treatment. Decompression was recommended because of the craniocaudal extent of the syrinx, the degree of cord expansion, the perisyrinx signal change indicating established cord injury, and the consequent risk of progressive and potentially irreversible myelopathy should the underlying obstruction to CSF flow be left untreated. Posterior fossa decompression was recommended, and the patient was counselled regarding the procedure and its associated risks.

The patient underwent surgery on 26 August 2024 under general anaesthesia. She was positioned prone with the head secured in a Mayfield clamp in flexion. Although a horseshoe headrest is an equally acceptable and less invasive alternative for this procedure, the Mayfield clamp was selected to provide rigid fixation and a reproducible degree of flexion during microsurgical dural work. A midline incision of approximately 10 cm was made, extending from 1 cm above theinion to the C2 spinous process. The nuchal fascia was opened along the midline using monopolar cautery, and the nuchal muscles were subperiosteally stripped from the occipital bone. The occipital bone, the posterior rim of the foramen magnum, and the C1 posterior arch were exposed. The operating microscope was introduced. Using a high-speed drill and Kerrison rongeurs, a suboccipital craniectomy measuring approximately 2×4 cm

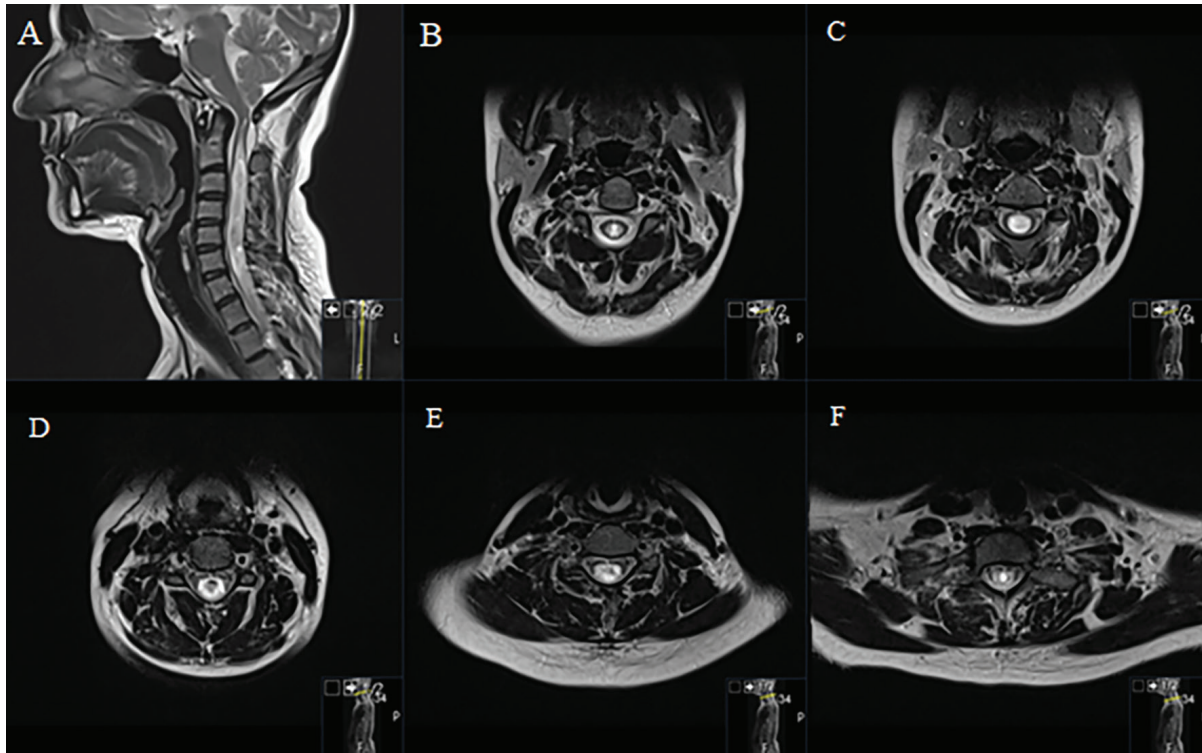


Figure 1: Preoperative cervical MRI. (A) Sagittal T2-weighted image demonstrating cerebellar tonsillar herniation below the foramen magnum and extensive hydrosyringomyelia extending from the odontoid process to T2. (B-F) Axial T2-weighted images between C2-C7 levels showing the syrinx cavity (9×12 mm) with surrounding T2-weighted hyperintensity.

MRI: Magnetic resonance imaging.

and extending roughly 2 cm lateral to the midline bilaterally was performed, along with excision of the C1 posterior arch. The posterior borders of the foramen magnum were further widened with Kerrison rongeurs.

The occipital sinus at the posterior rim of the foramen magnum was coagulated with bipolar diathermy and divided under microscopic vision before the dural leaves were reflected. The dura and overlying fibrotic bands were opened down to the C2 level using a blunt-tipped hook and a no. 11 blade. The subarachnoid membranes appeared fibrotic; CSF exit was confirmed from several millimetric openings, and the cisterna magna was adequately decompressed. A synthetic dura graft, approximately 3.5×1 cm and fashioned in a fish-mouth configuration, was sutured to the dural edges with 4-0 Vicryl in a watertight fashion. An autologous fascial or aponeurotic graft harvested from the operative field would have been an equally appropriate, and in several respects preferable, alternative; a synthetic substitute was used in this patient according to the preference of the operating surgeon and the graft material available in theatre at the time of operation. A small CSF leak at the uppermost suture line was reinforced with a muscle graft secured by a separate suture, followed by application of tissue adhesive and a thin layer of Spongostan. The nuchal muscles, fascia, and periosteum were closed with 2-0 Vicryl, and the remaining layers were approximated anatomically. The operative duration was 2 hours and 45 minutes, and blood loss

was minimal (<50 mL). No intraoperative complications were encountered. The patient was extubated uneventfully and transferred to the ward in stable condition.

The early postoperative course was uneventful. Neurological examination findings remained stable. The patient was mobilised on the first postoperative day and discharged in good condition. An early postoperative cervical MRI, obtained on the second postoperative day, confirmed adequate decompression of the posterior fossa (Figure 2).

At the seven-month follow-up visit in March 2025, the patient reported improvement in her cervical symptoms. Examination of the surgical wound revealed a millimetric crust, which was excised; a small wound opening was managed with local wound care and topical mupirocin ointment (Thiocilline; Abdi İbrahim, İstanbul, Türkiye). This minor superficial wound problem resolved completely with conservative treatment and required neither surgical revision nor systemic antibiotic therapy. A follow-up cervical MRI at six months postoperatively demonstrated satisfactory decompression of the craniocervical junction and interval reduction of the syrinx cavity (Figure 3). Measured on the sagittal T2-weighted sequence and on the axial section at C2, corresponding to the levels used preoperatively, the syrinx measured 95 mm in length and 1.5×2.5 mm at C2, compared with 130 mm and 9×12 mm before surgery. The patient was advised to undergo a repeat cervical MRI at one year postoperatively.

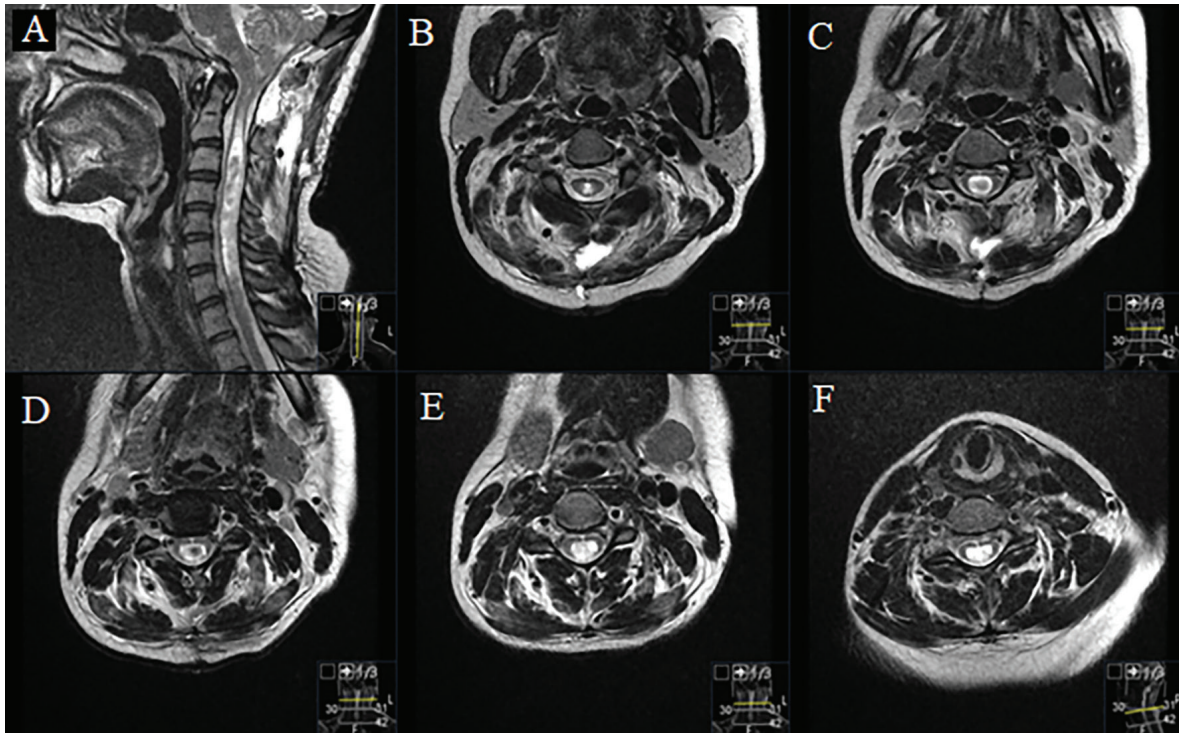


Figure 2: Early postoperative cervical MRI (postoperative day 2). Sagittal (A) and axial (B-F) T2-weighted images demonstrating adequate posterior fossa decompression following suboccipital craniectomy and C1 posterior arch excision.

MRI: Magnetic resonance imaging.

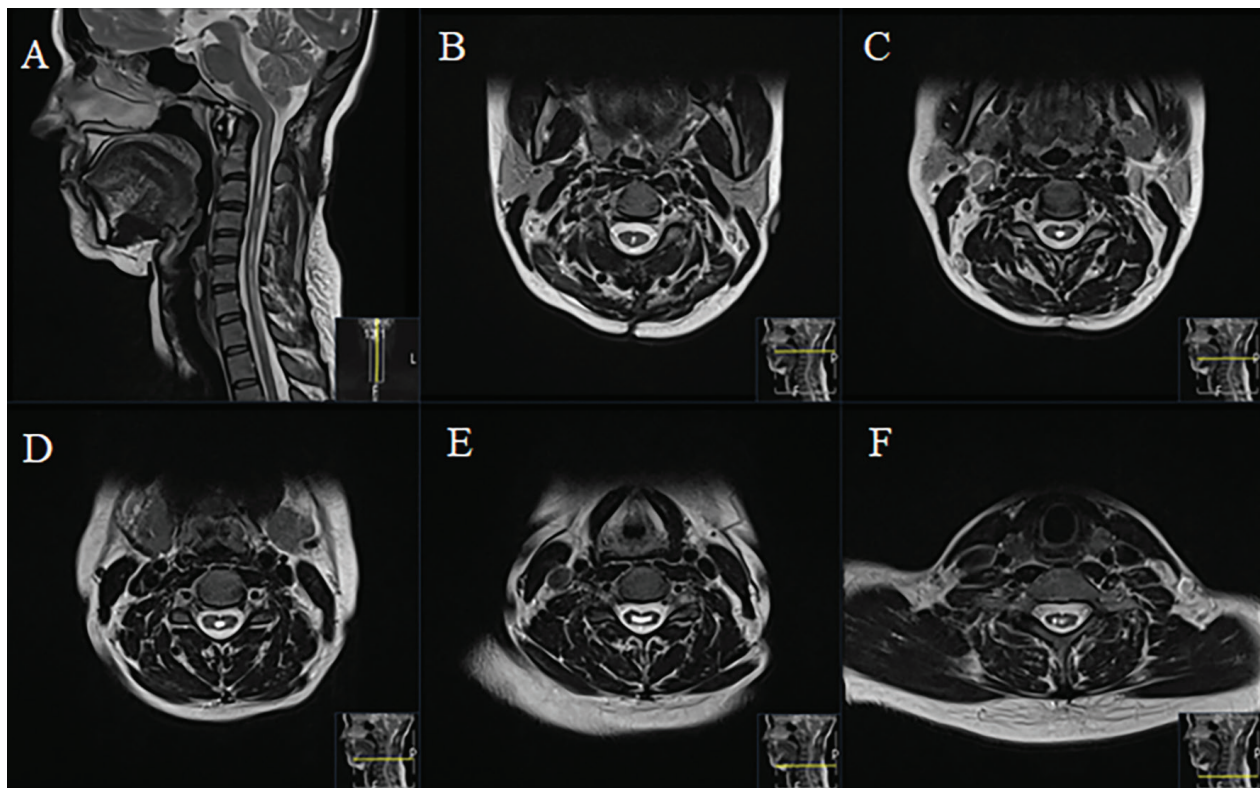


Figure 3: Six-month postoperative cervical MRI. Sagittal (A) and axial (B-F) T2-weighted images showing maintained posterior fossa decompression and interval reduction of the syrinx cavity.

MRI: Magnetic resonance imaging.

At the eighteen-month follow-up in March 2026, the patient remained clinically stable with no new neurological deficits. A repeat cervical MRI demonstrated sustained decompression at the craniocervical junction and interval reduction in the hydrosyringomyelia cavity (Figure 4). At the same reference levels, the syrinx measured 65 mm in length and 1.5×2 mm at C2. Because the follow-up examinations were not acquired with sequence parameters identical to those of the preoperative study, these values are presented as a comparison at matched anatomical levels rather than as an exact volumetric assessment. Clinically, the patient described her neck and left shoulder pain as substantially reduced and no longer limiting her daily activities, and she had returned to her usual occupational and domestic routine. No validated outcome instrument, such as a visual analogue scale or the Chicago Chiari Outcome Scale, was applied prospectively; therefore, the reported clinical improvement is descriptive.

DISCUSSION

The present case illustrates the diagnostic evaluation and surgical management of a 40-year-old female patient with CM-I associated with extensive cervicothoracic hydrosyringomyelia who was treated by posterior fossa decompression with duraplasty. Its instructive feature is the discrepancy between a tonsillar descent of only 4.5 mm and a syrinx spanning 130 mm of the cervicothoracic cord with associated cord

expansion and perisyrinx signal change. The case therefore argues against assessing Chiari-related pathology by a fixed numerical threshold of tonsillar herniation, and in favour of an individualised evaluation that integrates the clinical findings, posterior fossa crowding, craniocervical junction anatomy, syrinx burden, and the probable disturbance of CSF dynamics.

CM-I is characterised by the caudal displacement of the cerebellar tonsils through the foramen magnum, leading to crowding of the posterior fossa and disruption of normal CSF dynamics at the craniocervical junction (8). Obstruction of CSF flow at the level of the foramen magnum is considered the principal mechanism underlying the development of hydrosyringomyelia, which has been reported in 23-80% of patients with CM-I (4, 7, 8). Although the classical radiological threshold for the diagnosis of CM-I is ≥ 5 mm of tonsillar descent, Milhorat et al. (7) demonstrated in a cohort of 364 symptomatic patients that herniations as small as 3-5 mm may be clinically significant when accompanied by syringomyelia and other radiological features such as obliteration of the retrocerebellar CSF spaces and a steepened tentorial angle. Studies of posterior fossa morphology support this position. Sekula et al. (9) examined 22 patients who were symptomatic for CM-I but did not meet the accepted radiological criterion for tonsillar descent and found the dimensions of the posterior fossa to be reduced in this group, indicating that a crowded posterior fossa may be clinically relevant even when the descent falls below

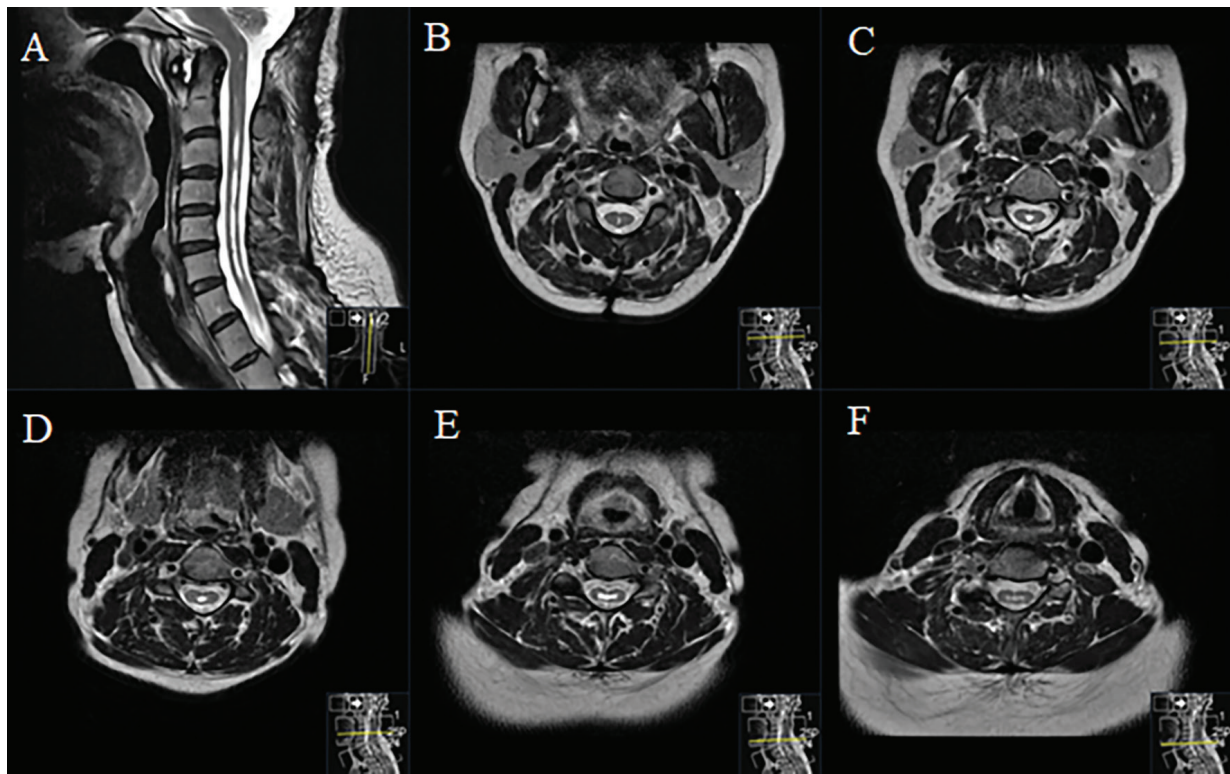


Figure 4: Eighteen-month postoperative cervical MRI. Sagittal (A) and axial (B-F) T2-weighted images demonstrating sustained posterior fossa decompression and reduction in the hydrosyringomyelia cavity compared to preoperative imaging.

MRI: Magnetic resonance imaging.

the conventional threshold. The disturbance of CSF flow and the overall craniocervical morphology therefore appear more clinically relevant than the absolute extent of tonsillar descent. In the present patient the descent was borderline at 4.5 mm, yet the subarachnoid spaces at C2 were effaced and the syrinx was extensive, indicating a functionally significant obstruction at the foramen magnum despite a sub-threshold measurement.

The clinical presentation of CM-I spans a broad spectrum, ranging from asymptomatic to severe neurological impairment (2, 6). The most frequently reported complaints are cervical pain and headache, and particularly suboccipital headache exacerbated by Valsalva manoeuvres. These are attributed to the stretching of pain-sensitive structures at the foramen magnum and to the elevated posterior fossa pressure (1, 2, 6). Less familiar presentations, including sleep-disordered breathing and orthostatic intolerance, are increasingly recognised but may be overlooked when the degree of tonsillar descent is borderline (10, 11). Our patient presented primarily with neck and left shoulder pain persisting for two months, consistent with the predominantly pain-related symptom profile described in the literature. The occasional stress urinary incontinence was considered nonspecific and could not be confidently attributed to the syrinx. In the absence of urological, gynaecological, or urodynamic assessment, neither a myelopathic origin nor pelvic floor dysfunction related to previous pregnancies can be established, and referral for formal evaluation would have been the appropriate course.

The development of a syrinx and the progression of neurological symptoms are widely regarded as indications for surgical intervention (1, 8, 12). In the present case the indication was not pain in isolation but the combination of an extensive syrinx, cord expansion and perisyrinx signal change, which together carried a risk of progressive and irreversible cord injury; this distinction is important because pain alone, in the absence of a syrinx or neurological deficit, would not ordinarily justify decompression. The objectives of surgery are to relieve compression at the craniocervical junction and to restore normal CSF circulation, with the aim of arresting or reversing syrinx progression (12, 13). Individualised selection of the extent of decompression according to the pattern of obstruction and the characteristics of the syrinx has been advocated, and dedicated scales have been proposed to grade syrinx resolution after surgery (13).

Posterior fossa decompression, which comprises suboccipital craniectomy, excision of the C1 posterior arch, and duraplasty, is the most commonly performed and widely accepted surgical procedure for CM-I (14, 15). Whether the dura should be opened remains debated. Bone-only decompression and dura-splitting techniques avoid entry into the subarachnoid space and are associated with shorter operative times, whereas duraplasty achieves a more reliable enlargement of the cisterna magna. A systematic review and meta-analysis found decompression with duraplasty and arachnoid preservation to be beneficial and to reduce the rate of reoperation, and comparative series have examined dura-splitting against combined techniques in

patients with syringomyelia (15, 16). Duraplasty was therefore preferred in this patient, in whom the syrinx was the dominant radiological abnormality. Regarding graft material, autologous fascia, aponeurosis or pericranium can be harvested from within the operative field, avoid the cost of a commercial product and are regarded by many surgeons as more physiological than synthetic substitutes; a synthetic graft was used in this case according to the operating surgeon's preference and the material available at the time of operation, and an autologous graft would have been an equally appropriate alternative. Our patient underwent a two-hour and 45-minute posterior fossa decompression. Blood loss was minimal. No intraoperative complications were encountered.

Following foramen magnum decompression, improvement in symptoms such as cervical pain, headache, and ataxia has been reported in the majority of patients. Klekamp reported that 73.6% of 371 patients demonstrated improvement at three months following decompression, whilst Alfieri and Pinna observed sustained clinical improvement in over 90% of their cohort with CM-I-related syringomyelia over a median follow-up of 12.7 years (17, 18). Han et al. (19) similarly documented clinical and radiological improvement one year after posterior fossa decompression in adults with CM-I and syringomyelia. However, established neurological deficits, such as motor weakness and sensory loss, are less likely to recover, underscoring the importance of timely intervention before irreversible spinal cord damage occurs (1, 3, 18). Despite a generally favourable safety profile, complications including CSF leakage, pseudomeningocele formation, wound infection, and, rarely, postoperative hydrocephalus have been reported in a proportion of patients (3, 15, 17, 20). In the present case the early postoperative course was uneventful, and the only adverse event during follow-up was a minor superficial wound problem at seven months that resolved with local care and topical antibiotic treatment. The patient described a substantial reduction in her neck and shoulder pain, the neurological examination remained unchanged throughout the 18-month follow-up period, and serial MRI demonstrated sustained decompression of the craniocervical junction with regression of the syrinx cavity, consistent with the favourable radiological outcomes reported in the literature (18, 19).

This case has certain limitations. First, as a single case report the findings are not generalisable. Second, the reported symptomatic improvement remains subjective. The clinical outcome was assessed descriptively: no validated instrument such as a visual analogue scale or the Chicago Chiari Outcome Scale was applied prospectively, and analgesic requirement and functional status were not recorded systematically. Third, the follow-up MRI studies were not acquired with sequence parameters identical to those of the preoperative examination, so the comparison of syrinx dimensions should be interpreted as an approximation at matched anatomical levels. Fourth, the urinary symptom was not evaluated by a urologist, a gynaecologist, or by urodynamic study, and its origin therefore remains undetermined. Finally,

the follow-up period, although 18 months, may not be sufficient to capture long-term complications or late recurrence, as neurological deterioration has been reported in up to 15% of patients within ten years of surgery (17). Continued clinical and radiological surveillance is therefore warranted.

CONCLUSION

This case suggests that posterior fossa decompression may provide clinical and radiological benefit in selected patients with extensive syringomyelia and borderline tonsillar descent when the overall findings are suggestive of impaired CSF dynamics at the craniocervical junction. It also illustrates that the clinical significance of Chiari-related pathology cannot be determined by the millimetric extent of tonsillar descent alone, and that the decision to operate should follow an individualised assessment of craniocervical anatomy, syrinx burden, and CSF flow rather than a single radiological cut-off. Larger series with longer follow-up and standardised outcome measures are needed to define which patients with borderline herniation derive the greatest benefit from decompression.

Ethics

Informed Consent: Written and verbal informed consent was obtained from the patient for this article.

Footnotes

Conflict of Interest: The authors declared no conflict of interest.

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