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SPECIAL ANNUAL ISSUE

Decompressive surgery for Chiari I malformation in children without dural repair: a still effective and safe procedure?

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Abstract

Purpose There are numerous publications about the technical aspects of decompressive surgery for Chiari I malformation highlighting many variations of this procedure. Each approach has its followers. Bony decompression of the foramen magnum alone or with the removal of a portion of the posterior arch of C1, dural splitting with keeping arachnoid intact, and durotomy are described. Dural closure is done with various materials. We retrospectively reviewed foramen magnum decompression without dural repair (FMDWDR) following the technique used by Gardener and Williams as an option in pediatric patients with Chiari I malformation in terms of complication rate and clinical outcome.

Methods The surgical database of our unit identified 65 consecutive children who underwent FMDWDR surgery for Chiari I malformation between 2009 and 2016. The retrospective assessment included patient demographics, clinical data, surgical technique, revision rate, complications, and clinical outcome.

Results Durotomy without repair was performed in 65 patients. Complications included aseptic meningitis and subdural hematoma respectively in three cases, intradiploic CSF collections in three patients, and CSF leaks in six children. The CSF leak rate has reduced dramatically after introducing a technical modification. Revision surgery was performed in seven cases. None of the patients was identified with postoperative hydrocephalus or infection. There was no mortality and no long-term surgical morbidity. In terms of clinical outcome, 52 patients reported postoperative improvement, 10 were clinically unchanged, and three noticed worsening of symptoms.

Conclusion Applying a "T"-shaped fascial incision which allows a watertight closure of the fascia FMDWDR is still a safe and effective treatment option for Chiari I malformation in children.

Keywords Chiari I malformation · Foramen magnum decompression · Durotomy · Dural repair · Cerebrospinal fluid · Complications

Introduction

In symptomatic adult and pediatric patients with Chiari I malformation, decompressive surgery [31] is performed to improve the flow of cerebrospinal fluid. In the literature, there is no class I evidence of how the best decompressive surgery should be performed to minimize complications and provide the best clinical outcome. The extent of the decompression of the foramen magnum [16] and the necessity to resect a portion of the posterior arch of C1 [19] and to perform durotomy [25] or dural splitting [13, 21] or peeling [4] and dural closure [35] are still a matter of debate [3, 6, 8, 12, 14, 20, 22, 23, 26, 36, 37]. Since 2009, in our pediatric neurosurgical unit, decompressive surgery for Chiari I malformation has been performed by two consultants with a very similar technique that is characterized by a limited decompression of the foramen magnum and to leave the dura open. The renunciation of dural repair is not a new concept: 70 years ago, Gardener [9] operated on 17 adult and adolescent Chiari I patients and left the dura intentionally open in all cases. In 1990, Williams [34] argued for leaving the dura open in the surgical treatment of syringomyelia.

The idea behind the renunciation of dural closure [35] is to create a bigger cisterna magna without provoking the downward herniation of the cerebellum. Leaving the dural defect

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open merges the intra- and extradural compartments. It provides a pseudomeningocele that serves as an enlarged cisterna magna and permits improved CSF flow. Reclosure of the dura with or without a graft, however, neutralizes at least partially the maximal space-gaining effect of the decompressive surgery. Autologous and artificial grafts may fail, favor fibrosis, or create a valvular mechanism. In addition, dural patching is costly in terms of operation time and in case of artificial patches also from a financial point of view.

We performed a retrospective review of the database of our pediatric neurosurgical unit between 2009 and 2016 to identify the children who underwent surgery for Chiari I malformation to evaluate the complication rate and clinical outcome.

Methods

Institutional data

The surgical database of our Department of Pediatric Neurosurgery of the John Radcliffe Hospital in Oxford revealed 65 children who underwent FMDWDR surgery for Chiari I malformation between 2009 and 2016. All patients were included in this study irrespective of concomitant pathologies. The assessment of cerebellar tonsil position was performed on sagittal T2-weighted magnetic resonance images (MRI). Because of the common asymmetry of tonsillar ectopia [32], a line drawn perpendicular from the lowest tonsillar tip to the foramen magnum line was measured. The first clinical follow-up appointment was scheduled 6 weeks after surgery. Further appointments were scheduled on a case-by-case basis. Postoperative MRI was only performed in the event of complications or the persistence of preoperative symptoms depicted at the follow-up exam. The retrospective assessment of the records included patient demographics (Table 1), preoperative symptoms and signs (Table 2), surgical technique (Table 3), revision rate and complications (Table 4), and final clinical outcome (Table 5).

Surgical technique

All patients had been operated by one of two consultants who applied a very similar technique with the only difference in terms of the handling of the cerebellar tonsils. After a single shot of prophylactic antibiotics, the patient was placed in the Concorde position with the head fixed with a three-pin head holder. In the majority of our cases, however, pin fixation was not feasible due to thin skull, so that the Mayfield head rest was used. Disinfection and sterile draping were conducted. From the external occipital protuberance down to C2, the skin and the subcutaneous tissue were incised in the midline to expose the cervical fascia (Fig. 1a). Lateral dissection from the top of incision to C1 was done in between the avascular plane of fascia and subcutaneous fat. During our series, we introduced a new technical feature: a "T"-shaped myofascial cuff [7,28] was created with midline dissection in the avascular plane and subperiostal exposure of the suboccipital bone and the posterior arch of C1 (Fig. 1b). With a craniotome and a high-speed drill, a limited craniectomy of 2.5 × 1.5 cm to enlarge the foramen magnum was performed and a part of the posterior arch of the C1 resected (Fig. 1c). The dura was opened with a straight incision following a paramedian line from C1 to the enlarged foramen magnum. Lateral dural releasing incisions on both sides were performed at the level of C0–C1 and C1–C2 (Figs. 1d and 2). The two arisen dural flaps were hitched laterally to the muscles (Fig. 1e). The arachnoid was opened, and all visible adhesions were resected. The opening of the fourth ventricle was checked to see if there is good egress of cerebrospinal fluid (CSF). One surgeon performed subpial cerebellar tonsil resection if judged necessary. Strict meticulous hemostasis was done in all cases, and copious irrigation was used to flush out all residual blood mixed with CSF. A piece of thin sterile and water-insoluble porcine gelatin–absorbable sponge (Spongostan™) [11,27] was loosely placed without sutures on the dural defect which acts as a temporary barrier between the intradural and extradural compartments. The latter contains in the postoperative phase blood and exuded fluid from traumatized muscles. Tight wound closure with absorbable stitches was done respecting the four anatomical planes, muscles, fascia, subcutaneous tissue, and skin. There was no postoperative treatment with antibiotics or corticosteroids.

Results

Patient characteristics

Sixty-five consecutive symptomatic pediatric patients underwent FMDWDR surgery for Chiari I malformation in our unit from 2009 to 2016 (Table 1). The mean age was 10.4 years with a range from 2 to 19 years. All patients had radiologically confirmed Chiari I malformation with the cerebellar tonsils extending more than 5 mm inferior to the foramen magnum [1]: The mean position of the cerebellar tonsils below the foramen magnum was 13.24 mm with a range between 6 and 25 mm. Thirty-two patients had syringomyelia and Chiari I malformation. Nineteen patients had concomitant scoliosis, and 13 children presented with syringomyelia and scoliosis. The median hospital stay was 3 days. The follow-up time was between 6 and 48 months. The most frequent specified symptom was headache. The patients' signs and

Table 1 Patient demographics

	Present study, 2019	Krieger et al., 1999 [17]	Tubbs et al., 2011 [33]
Number of patients	65	31	500
Mean age	10.4 years	12 years	11 years
Range age	2 years to 19 years	6 months to 18 years	2 months to 20 years
Mean position of cerebellar tonsils inferior to the foramen magnum	13.24 mm	N/A	Inferior border of the lowest cerebellar tonsil between the foramen magnum and C-1 in 110 patients (22%), at C-1 in 187 (37.4%), at C-2 in 197 (39.4%), and at C-3 in 6 (1.2%)
Range of mean position of cerebellar tonsils inferior to the foramen magnum	6–25 mm	N/A	
Length of hospital stay	median: 3 days	median: 2.7 days	2 data mentioned: 2 to 7 days (mean: 3 days) and 1 to 8 days (mean: 3.1 days)
Follow-up	4 to 48 months	15 to 93 months	2 to 180 months

symptoms at time of presentation in outpatients are listed in Table 2.

Complications

CSF leaks occurred in 6 children. These were patients that had been operated early in our series without the creation of a T-shaped myofascial cuff. Aseptic meningitis and subdural hematoma respectively happened in 3 cases, and intradiploic CSF collections in 3 patients. Revision surgery was performed in 7 children. None of the patients was identified with postoperative hydrocephalus or infection. There was no mortality and no long-term surgical morbidity.

Clinical outcome

At the follow-up appointment, the patients or their parents were asked to evaluate the evolution of the preoperative symptoms. We limited possible answers to improvement, unchanged, and worsening. Fifty-two patients reported postoperative improvement, 10 were clinically unchanged, and 3 noticed worsening of symptoms.

Table 2 Signs and symptoms before first decompressive surgery for Chiari I malformation

Symptoms and signs	Present study, 2019, number of patients 65 *n* (%)	Krieger et al., 1999 [17], number of patients 31 *n* (%)	Tubbs et al., 2011 [33], number of patients 500 *n* (%)
Headache	43 (66.2)	16 (51.6)	
Occipital pain	13 (20.0)	N/A	200 (40.0)
Neck pain	11 (16.9)	N/A	
Sensory disturbances	14 (21.5)	9 (29)	54 (10.8)
Gait disturbances	12 (18.5)	7 (22.6)	N/A
Motor power deficit	12 (18.5)	12 (38.7) upper extremities	53 (10.6)
Sphincter function	12 (18.5)	N/A	2 (0.4)
Swallow function	5 (7.7)	N/A	20 (4.0)
Preoperative hydrocephalus	1 (1.5)	N/A	48 (9.6)
Syringomyelia	32 (49.2)	26 (83.9)	285 (57)
Scoliosis	19 (29.2)	20 (64.5)	90 (18)
Syringomyelia and scoliosis	13 (20.0)	N/A	74 (14.8)

Table 3 Technical details of decompressive surgery for Chiari I malformation

	Present study, 2019	Krieger et al., 1999 [17]	Tubbs et al., 2011 [33]
Number of patients	65	31	500
Size of suboccipital craniectomy	2.5 × 1.5 cm	2.5 × 2.5 cm	width 4.0 to 5.0 cm
Inclusion of C1 in decompressive surgery	Removal of the mid-15-mm part of the posterior arch of C1	Removal of the mid-15-mm part of the posterior arch of C1	Removal of posterior arch of C1, width 4.0 to 5.0 cm
Durotomy	65	31	499
Arachnoid opened and adhesion lysed	40	31	499
Cerebellar tonsils shrunk	15	N/A	49
Dural repair	0	0	499

Discussion

To the best of our knowledge, the present study with 65 patients is the biggest series of children with Chiari I malformation who have undergone FMDWDR. Krieger et al. [18] reported in 2011 a study of 79 patients who had CM-I decompression but where the dura was loosely approximated.

The intercomparability of our results with those of other studies is limited. Most studies included pediatric and adult [24, 30] or only adult patients [29] or different scoring systems for outcome assessment had been applied [2]. Moreover, pre- and postoperative imaging, the recording of pre- and postoperative signs and symptoms and the applied surgical techniques vary considerably.

In 1999, Krieger et al. [17] published a retrospective study with 31 operated pediatric patients that had been operated without dural closure and where a piece of oxidized surgical cellulose was placed over the area of the opened dura. The rate of CSF leaks and infection is similar in both studies (Table 4).

In terms of clinical outcome, the comparison of these two studies is limited because the prevalence of certain preoperative symptoms is not mentioned in both studies (Table 2) and we did not systematically follow up the evolution of scoliosis and syringomyelia with postoperative MRI. Krieger et al. [17] mentioned that 94% of the total signs and symptoms significantly resolved by the 6-month follow-up exam (Table 5), whereas we categorized the preoperative symptoms as "improved," "unchanged," or "worsened" (Table 5).

The comparison of the outcome assessment of our cohort with pediatric patients who underwent decompressive surgery with duraplasty also turns out to be difficult. Tubbs et al. [33] reported 500 surgically treated pediatric patients with Chiari I malformation. Duraplasty was performed in 499 cases. In 83% of the patients, relief of preoperative symptoms or signs was seen [33] without further specification (Table 5).

Tubbs et al. [33] mentioned revision surgery in 15 patients (3.0%). Of our cohort, 7 (10.8%) patients underwent revision surgery. One of these patients had been operated on before in another hospital. Another patient had surgery early in our series before we introduced the T-shaped fascial incision and needed revision surgery due to CSF leak.

Table 4 Complications for decompressive surgery in Chiari I malformation

Complication type	Present study, 2019, number of patients 65 *n* (%)	Krieger et al., 1999 [17], number of patients 31 *n* (%)	Tubbs et al., 2011 [33], number of patients 500 *n* (%)
CSF leak	6 (9.2)	3 (9.7)	1 (0.2)
Intradiploic CSF collections	3 (4.6)	N/A	N/A
Aseptic meningitis	3 (4.6)	N/A	0 (0.0)
Subdural hematoma	3 (4.6)	N/A	4 (0.8)
Acute postoperative hydrocephalus	0 (0.0)	1 (3.2)	4 (0.8)
Infection	0 (0.0)	0 (0.0)	1 (0.2)
Revision surgery	7 (10.8)	N/A	15 (3.0)
Long-term morbidity	0 (0.0)	0 (0.0)	1 (0.2)
Mortality	0 (0.0)	0 (0.0)	0 (0.0)

Table 5 Evaluation of postoperative symptoms after first decompressive surgery

Patients clinically	Present study, 2019, number of patients 65 *n* (%)	Krieger et al., 1999 [17], number of patients 31 *n* (%)	Tubbs et al., 2011 [33], number of patients 500 *n* (%)
Improved	52 (80.0)	94% of signs and symptoms significantly resolved by 6-month postoperative exam	Relief of preoperative symptoms or signs was seen in 83% of patients
Unchanged	10 (15.4)		
Worse	3 (4.6)		4 (0.8)

The CSF leak rate has reduced dramatically after introducing the T-shaped fascial incision which allows a watertight closure of the fascia. We noticed also the reduction of the length of hospital stay after the introduction of this technical modification. Meticulous hemostasis and copious irrigation to wash the blood-stained CSF reduces the stimulus for production of excess CSF and possibly the incidence of CSF leak.

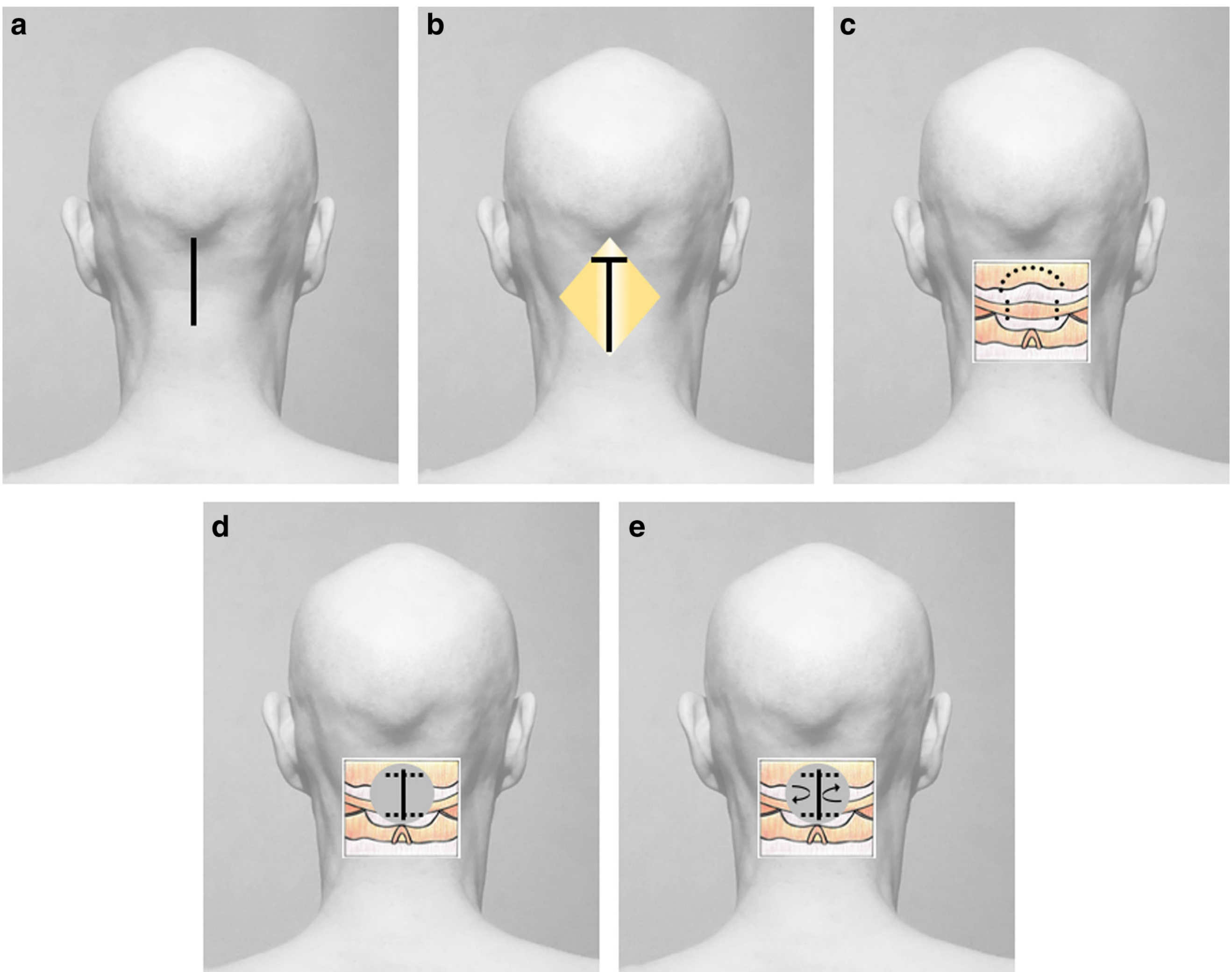

Fig. 1 Illustrations of the suboccipital region reflect the steps of decompressive surgery for Chiari I malformation without dural repair. **a** Cutaneous incision in the midline from the external occipital protuberance down to C2 (solid line). **b** "T"-shaped incision to create a myofascial cuff (solid lines). **c** Limited craniectomy of 2.5 × 1.5 cm to enlarge the foramen magnum and removal of a part of the posterior arch of the C1 (dotted lines). **d** Straight paramedian durotomy from C1 up to the enlarged foramen magnum (solid line) with lateral dural releasing incisions on both sides at the level of C0–C1 and C1–C2 (dotted lines). **e** After durotomy, the two dural flaps are hitched laterally to the muscles (curved arrows)

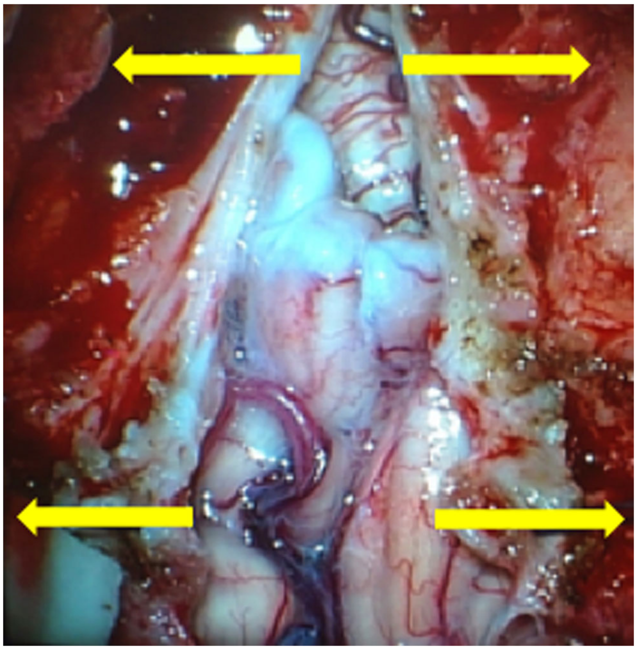

Fig. 2 Intraoperative photograph showing the straight paramedian dural incision from C1 up to the enlarged foramen magnum. Lateral dural releasing incisions on both sides at the level of C0–C1 and C1–C2 are suggested (arrows)

In few cases where the dura was not opened or duraplasty had been performed, there is anecdotal experience about the reformation of a tight foramen magnum resulting in the necessity to redo its decompression. This could be due to the osteogenic effect of the dura in children and the capability of their skull to grow so that our rationale is to keep the dura open especially in children.

FMDWDR is a space-gaining maneuver leading to the formation of a bigger neo cisterna magna and allowing for CSF flow behind the tonsils. Figure 3 shows exemplarily the pre- and postoperative MRI of a 4-year-old patient with Chiari I malformation who had FMDWDR resulting in the formation of a pseudomeningocele that creates an enlarged cisterna magna.

Applying a T-shaped fascial incision which allows a watertight closure of the fascia, we feel that on children FMDWDR is safe and effective as our study has concluded. In children, due to ongoing growth potential, it is essential to get good flow through the foramen magnum; our technique which is a slight modification of the original one is safe and effective.

At present, there is no class I evidence of how best surgical treatment of Chiari I malformation should be designed to deliver best clinical outcome and to minimize complications. A main reason is that the underlying disease has not been unraveled so far. Chiari I malformation seems not to be a

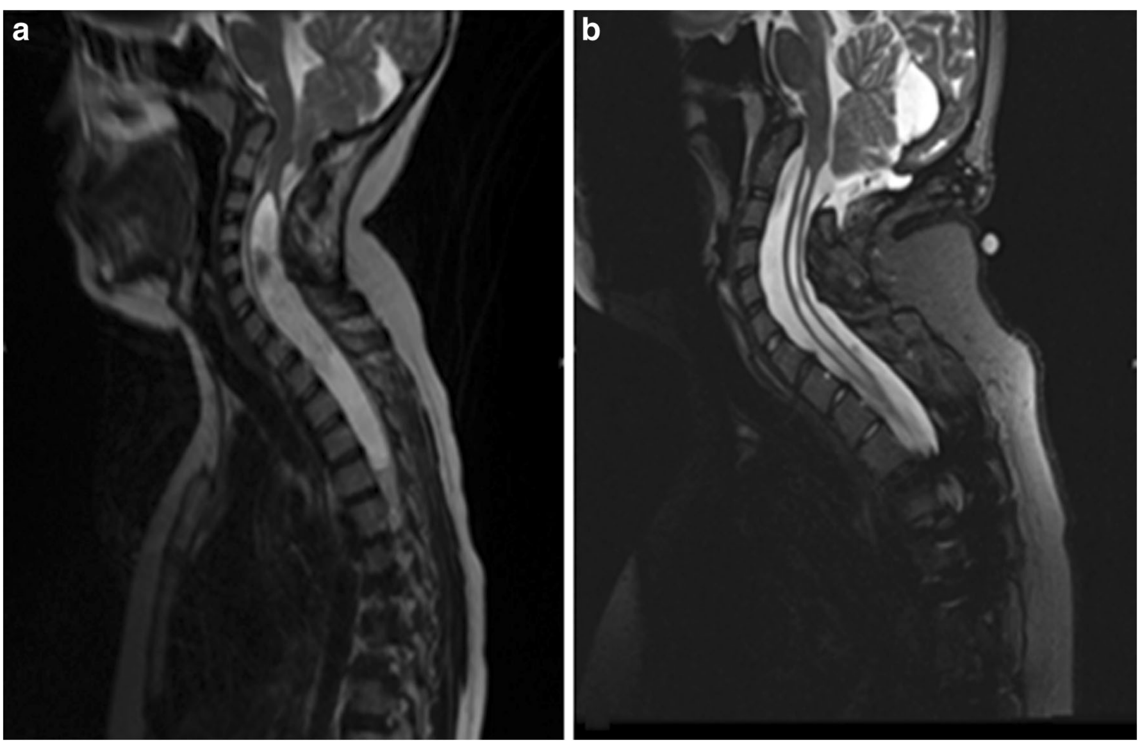

Fig. 3 **a** Preoperative sagittal T2-weighted magnetic resonance image demonstrating the patient's Chiari I malformation with the cerebellar tonsils lying at the C2 level and a concomitant syrinx extending from C2–C3 to T5. **b** Sagittal T2-weighted magnetic resonance image performed 7 years postoperatively showing the pseudomeningocele that creates an enlarged cisterna magna with the cerebellar tonsils lying at the C2 level. Sustained postoperative decrease of the syrinx suggests a suitable decompression of the posterior fossa and sufficient CFS flow at the foramen magnum

single entity; it is probably a collective name that comprises several entities with still unknown interactions. In this context, the contribution of occipital-cervical instability [10, 15], raised intracranial pressure [5], the pathophysiology of syringomyelia, and scoliosis need to be further examined. National or international registers might help to elucidate the pathophysiology of Chiari I malformation and offer directions for further studies. A unified scoring system for outcome assessment may facilitate the intercomparability of studies.

Limitations

Our study had no control group which made it difficult to perform appropriate correlation and predictive statistical analyses. We did not perform routinely postoperative imaging to monitor the evolution of syringomyelia and scoliosis.

Conclusion

Decompressive surgery without dural repair with a T-shaped fascial incision which allows a watertight closure of the fascia is a safe and effective treatment option for Chiari I malformation in children.

Compliance with ethical standards

Conflict of interest The authors report no conflict of interest.

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