Interlayer Dural Split Technique for Chiari I Malformation Treatment in Adult – Technical Note

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ABSTRACT

Objective: To present an alternative surgical technique in treating cases of Chiari I Malformation with mild-to-moderate syringomyelia after decompressive suboccipital craniectomy: incising only the outer layer of the dura mater, then dissecting it from the inner layer without opening the latter.
Methods and Results: We utilized this technique in a short series of three cases who were admitted in our department for mild symptoms such as intermittent headache and dissociated sensory loss in the upper limbs, caused by a Chiari Malformation Type I. The patients were placed in the sitting position. We performed a reduced median suboccipital craniectomy and resection of the posterior arch of C1 adapted to the level of tonsil descent, from a limited superior half to a complete resection. Afterwards, we incised the outer dural layer, while sparing the inner one. Using a fine dissector, we then split apart the outer and inner layers to the margin of the craniectomy. Through the transparency of the inner layer and the arachnoid, the cerebellum and the medulla were visible and pulsating. An autologous fascia duraplasty was then performed. The postoperative course was favorable in all cases, patients being discharged without any deficits and with complete symptom resolution.
Conclusions: Interlayer dural split technique can be used effectively in treating symptomatic cases of type I Chiari malformation in adults, with mild-to-moderate syringomyelia. It is less invasive than opening the dura and possibly more effective than decompressive craniectomy and C1 laminectomy alone. This technique must be validated in a larger case-control series.

Key words: Chiari I Malformation, Suboccipital Craniectomy, Partial C1 Laminectomy, Interlayer Dural Split Technique, Posterior Fossa Decompression, Duraplasty

- Arnold Chiari 1 treatment involves the decompression of the foramen magnum via craniectomy, duraplasty, and resection of cerebellar tonsils.
- These methods have varying success rates and there is much debate whether more or less invasive methods are required.
- Our method aims to combine the invasive and less-invasive methods by only incising the outer layer of the dura mater, without entering into the subdural spaces, and performing a duraplasty to further prevent any CSF leakage.

Introduction

In his two monographs published in 1891 and 1895, professor Hans Chiari defined the Chiari malformations (CMs) as varying degrees of herniation through the foramen magnum. Based on the findings of these malformations in 40 autopsies that were performed in Prague, he devised a four-tier classification system (type I, II, III, IV) for these entities [1-4].

Over time, new classifications appeared for the variants not fitting Chiari's original descriptions new classifications (e.g. Chiari 0, Chiari 1.5 and Chiari 3.5 malformations) [5]. Chiari malformation type I (CMI) constitutes a syndrome wherein the cerebellar tonsils descend through the foramen magnum for at least 3–5 mm. This results in alterations of cerebrospinal fluid (CSF) flow [6]. According to morphometric data, a hypoplastic occipital bone produces posterior fossa overcrowding, wherein normally developed neural structures are contained [7]. Occipital bone hypoplasia is probably the result of a paraxial mesodermal defect of the parachordal plate. However, CMI associated with a thick occipital bone has also been described [8]. Another theory states that CMI is the result of a premature closure of the spheno-occipital synchondrosis [9, 10]. According to this theory, the structure of the posterior cranial fossa gradually shifts towards the shape of a narrow funnel.

The average age of detection of CMI is 24.9 + 15.8 years [11]. The most common presenting symptom is headache in the occipital and cervical region, being short of duration and induced by Valsalva activities. Other presenting symptoms may include weakness and hyporeflexia in the upper extremities, spasticity, hyperreflexia and fasciculation in the lower extremities, lower cranial nerve deficits resulting in central sleep apnea and/or dysphagia, and/or difficulty with balance or coordination. In up to 70% of the cases of CMI, scoliosis and syringomyelia are associated [12-15]. In association with syringomyelia the craniospinal pressure gradients are higher in patients with CMI [16].

MRI is the best imaging modality for determining cerebellar tonsillar ectopia and concern for CMI and syringomyelia that previously with other techniques remained unrecognized or was misdiagnosed. The following findings may be observed: hindbrain abnormalities, obstructive hydrocephalus, herniation of the cerebellar tonsils through the foramen magnum, narrow posterior cranial fossa, syringomyelia and skeletal abnormalities. Phase-contrast MRI or cine MRI can be useful for evaluating CMI patients and demonstrate the disturbance of CSF velocity and flow at the foramen magnum [17-20].

Ramón et al. stated that for the patients with mild or asymptomatic symptoms there is little evidence to suggest that surgery should be performed on the basis of radiological findings alone. Patients with progressive posterior fossa or spinal cord signs are more likely to undergo surgical treatment. The target of surgery is to restore normal CSF dynamics at the foramen magnum [21-23]. Conventional treatment is the posterior fossa bony decompression and upper cervical laminectomy with or without duraplasty. Despite the persistent debate, there is little evidence to support either posterior fossa decompression without dural opening or posterior fossa decompression with duraplasty is more suitable for a given patient [24-27].

Material and Methods

Patients. Three patients (one male, two female) diagnosed with Chiari Malformation type I were admitted in our department between June and December 2019 and, after acquiring their informed consent, were subjected to surgical decompression of the posterior fossa using our original technique. Their symptoms were mild, ranging from recurrent and intermittent headache, to dissociated sensory loss in the upper limbs. One of them (Case 1) had a mild cervical syrinx, the other two had a moderate syringomyelia. Considering their symptomology, lack of response to conservative management, and findings on imaging studies, the operative board decided for the surgical decompression of the posterior fossa. As their symptoms were mild and the degrees of tonsil descent were low, we opted for a technique that would imply minimal risk of CSF fistula, as well as a lower chance of recurrence than simple bony decompression.

Surgical technique. All of the patients were placed in the sitting position. After exposing the bone, we plugged the foramina of the emissary veins to avoid air embolism. Incision and dissection of superior insertion of the atlantooccipital ligament allowed for a radial foramen magnum decompression starting from its inferior border up to 1,5-2 cm using either a Kerrison punch, or a diamond drill. The atlantooccipital ligament was then incised vertically in the midline, dissected from atlas and anchored laterally. Adjusted to the degree of cerebellar tonsil descent, the superior half or entire posterior arch of C1 was resected. The dura mater, which is usually thick and stenotic at this level, was carefully incised only at the external layer after occlusion of transverse venous channel via bipolar coagulation. The outer and inner layers are then split apart from each other all the way to the border of the craniectomy using a fine double-ended dissector. Through the transparency of the inner dural layer and the arachnoid, the pulsating cerebellar tonsils and the medulla are clearly visible, suggesting adequate flow of CSF through the cisterna magna and the foramen magnum (Video 1). This was also later confirmed by the delayed postoperative MRI controls and clinical amelioration. No CSF leakage was noticed, neither was there any pneumocephalus on postoperative imaging studies; however, as a precautionary measure to prevent a later extension of the inner layer, a duraplasty was performed using autologous fascia and a 5/0 polypropylene monofilament suture in either a separate, or a continuous fashion (Figure 1). No unforeseen incidents such as air embolism occurred during surgery in any of the patients.

Case Description

Case 1. A 31-year-old female patient was admitted to our neurosurgical department complaining of recurrent episodes of numbness, alternating between her upper and lower limbs, as well as headache and upper back pain. The symptoms began approximately a year prior to presentation and were refractory to mild analgesics.
On admission, the neurological examination revealed dissociated sensory loss in her upper limbs, without discernible motor deficit. The non-contrasted MRI disclosed the inferiorly herniating cerebellar tonsils pressing against the posterior medulla, a “crowded foramen magnum,” and cervical syrinx. A complete MRI evaluation of the spine ruled out any other visible causes of syrinx. Therefore, the diagnosis was consistent with mild CMI (Figure 2A).
Postoperative course. The control cranio-cervical MRI performed at 72 hours after the procedure showed a slight increase in distance between the medulla and the cerebellar tonsils, as well as a shrinkage of the syringomyelia, with no signs of intracranial hemorrhage

or pneumocephalus (Figure 2B). Her postoperative evolution was favorable with swift amelioration of the dissociated sensory loss and cervicalgia. She was discharged on the fifth postoperative day without any neurological complaints and fully capable to resume normal daily life. The control MRI at 3 months showed a marked reduction of the medullary compression and an enlargement in the space between the medulla and the cerebellar tonsils, as well as a slight shrinkage of the syrinx (Figure 2C).

Case 2. A 73-year old male patient presented with vertigo, numbness on the left hemiface and upper extremity, as well as mild headache. The symptoms started two months before admission and did not respond to mild analgesics. Personal history lacked any heart-related comorbidities or symptoms signaling ischemic events.
On admission, the objective neurological examination revealed mild dissociated sensory loss in the left upper limb, without additional motor deficit. The non-contrasted MRI showed the crowding of the posterior fossa and foramen magnum, as well as a moderate cervical syringomyelia (Figure 3A). Additionally, imaging studies failed to reveal any ischemic areas, whether chronic or acute, therefore, cerebral ischemia was ruled out as a cause of his symptoms.
Postoperative course. The control cranio-cervical MRI performed on the second day after the intervention showed no signs of hemorrhage or pneumocephalus. His symptoms attenuated and he was discharged one week after surgery without any additional complaints. The control MRI performed at 3 months displayed a significant reduction of the syringomyelia, as well as an enlarged space between the medulla and the cerebellar tonsils (Figure 3B).

Case 3. A 38-year old female patient, known with hypothyroidism, was admitted in our department complaining of cervical pain, numbness in the upper limbs and headache. Her symptoms appeared 6 months prior to presentation, had a progressive evolution, and did not ameliorate with analgesic medication.
On admission, the neurological exam showed moderate and symmetrical dissociated sensory loss in her upper limbs, without apparent motor deficit. The non-contrasted MRI was consistent with CMI and moderate syringomyelia in the cervical and dorsal regions (Figure 4A).
Postoperative course. The control cranio-cervical MRI performed 48 hours after surgery showed no signs of hemorrhage or pneumocephalus (Figure 4B). She had a discernible reduction of her complaints and was discharged one week after surgery having no symptoms. The control MRI at 3 months showed a substantial shrinkage of the syringomyelic cavity, as well as a larger distance between the medulla and the cerebellar tonsils (Figure 4C).

The clinical and radiological follow-up duration was two years for each patient, with repeated controls at 3, 6, 12 and 24 months. None of the three patients had any complications or recurrence of symptoms during this time.

Discussion

The standard surgical treatment for CMI is represented by a sufficiently broad decompression of the cranio-cervical junction, involving a suboccipital craniotomy and C1 posterior arch resection [28]. This is then occasionally supplemented by a C2 laminectomy, followed by incision of the dura and duraplasty [29, 30]. Nevertheless, the optimal treatment method remains elusive. There has been a lot of debate regarding the extent of craniectomy, the necessity of dural incision, opening the arachnoid, the removal of the cerebellar tonsils, even the placement of duraplasty and its source, whether autologous or synthetic [31-35]. Surgery can be performed in either prone, concorde, semi-sitting or even sitting position [29, 36-42]. The majority of authors prefer the prone position because this offers comfort of the surgical team, and the risk of air embolism is minimal. We are generally in favor of sitting position for cranio-cervical junction interventions, as it offers a wide workspace, heightened spatial orientation, optimal venous drainage, decreased intracranial pressure, and adequate gravitational cerebellar traction [43]. However, since there is no need for opening the CSF spaces in our described technique, one might argue that the sitting position was not warranted. Nevertheless, we opted for this position more for the exposure, drainage and personal preference.

Asymptomatic patients with CMI are generally managed conservatively, as there is little chance of them developing symptoms during their lifetime [44, 45]. This also applies to individuals presenting with mild symptoms, with significant evidence suggesting that their symptoms remain stable or might alleviate after a period of time. Studies indicate a higher frequency of symptomatic presentation in the adult population, as well as poorer improvement rates following non-operative management [46]. Presentation with cough headache was repeatedly associated with a decreased chance of associated symptom improvement, whereas headaches of other types were identified as positive predictors for recovery in the absence of surgery [46, 47]. Additionally, surgical intervention possesses the highest likelihood of improving headache and ataxia, however nausea or non-specific symptoms may persist after intervention [44, 48]. A large syrinx may also be a reason for intervention, although the absence of clinical manifestation or the lack of syrinx enlargement on imaging studies may justify conservative management. Spontaneous resolution of CMI in adults has also been documented on serial MRI imaging, although this event usually occurs in children [49, 50]. As our cases presented with headache and varying degrees of dissociated sensory loss that did not respond to conservative management, as well as the mild-to-moderate syringomyelia in the latter two, we argue that surgical intervention was mandated.

Proponents of cranio-cervical decompression with duraplasty claim that it offers a superior relief of symptoms, an acceptable rate of morbidity, and has a lower rate of reintervention [31, 37, 51]. Even more so, duraplasty leads to a very high rate of resolution of the syringomyelia, although recrudescence of the syrinx has been noted in a small number of cases [52]. In the retrospective series of 105 patients treated via duraplasty, De Vlieger *et al*. concluded that the majority of patients describe an amelioration or stabilization of the symptoms, especially if there were no signs of syringomyelia [53]. According to Lam *et al*., autologous pericranium augmented with dural sealant (such as

DuraSeal™, Covidien LLC, Mansfield, MA, USA) is both safe and effective for CMI, removing the need to perform watertight dural sutures [54]. This is especially useful considering that a watertight closure of the dura using only sutures is virtually impossible, as the needles cause small holes in the dura, and a tightly placed patch may rip from the tension in the suture. Takeshima *et al*. demonstrated that the posterior fossa becomes progressively enlarged over time following duraplasty with local fascia, in a manner depending on the size of the patch itself [55]. Moreover, the degree of posterior fossa enlargement was significantly correlated with the likelihood of a positive postoperative outcome. As such, duraplasty is a more reliable treatment choice in patients with syringomyelia or a higher grade of cerebellar tonsil herniation, whereas simple extradural decompression is commonly not advised in children with syringomyelia [56, 57].

Arachnoid manipulation may be needed in case of significant adhesions, although Vidal et al. reported a higher incidence of complications following subarachnoid exploration [58]. Foramen magnum decompression without dural repair, in which the fascia in opened with a T-shaped incision and then sutured in a watertight fashion is an available alternative, despite also bearing high risks of CSF fistulas [42]. This maneuver increases space, leading to the development of a larger neo cisterna magna and permitting the CSF to flow behind the tonsils. According to Tonkins *et al*., dural hitching, where the dura is sutured to the covering tissues and the incision is then closed in anatomical layers, is superior to standard dural closure and considerably more likely to cause syrinx resolution [30]. The reasoning behind our decision not to open the dura entirely was to avoid any CSF leaks, the low grade of tonsil descent in our patients, and the fact that we enlarged the outer layer of the dura using autologous fascia, offering a widening of the posterior fossa and a sufficient space for adequate CSF flow.

Outer membranectomy, or dural peeling, is a technique similar to the one we have described, wherein the outer layer of the dura is carefully peeled off so as not to open the subarachnoid spaces [26, 59, 60]. It has a shorter operation time and leads to lower risks of CSF-related complications than duraplasty, yet with the drawback of an inferior success rate and a higher chance of reintervention. In the case series reported by Del Gaudio et al, all ten patients who underwent duraplasty achieved clinical improvement, whereas only twelve out of eighteen (66.7%) subjected to dural peeling did the same [59]. Nevertheless, dural peeling led to no postoperative, as compared to duraplasty (4 patients, 40%), but no predictive factors of clinical amelioration following dural peeling were discovered. Certain authors support a minimally invasive surgical (MIS) decompression of the foramen magnum, in which a MIS tube is inserted after performing a midline incision no larger than 3 cm [61-63]. After performing a midline incision of 2 or 3 cm centered over the atlanto-occipital junction, a MIS tube or a Gelpi self-retaining retractor is inserted. The MIS tube limits the extent of the subperiosteal dissection, bone resection, dural peeling or delamination and, if required, durotomy. MIS decompression apparently offers higher quality-of-life improvements, lower rates of surgery-related complications, as well as an overall shorter intervention compared to the classical open surgery. Furthermore, it is associated with minimal damage to the connective and muscular tissues, shorter recovery time, earlier

mobilization and discharge, and an adequate widening of the posterior aspect of the foramen magnum [40].

An important feature of decompressing the posterior fossa is represented by a sufficient cephalocaudal extension of the suboccipital craniectomy [10]. Regarding specifics of bone removal, it is possible to perform an expansive cranioplasty, implying an osteoplastic suboccipital craniotomy, with the bone flap attached to the periosteum [36]. Fixation of the flap with resorbable plates ensures a faster and sturdier result. This method would, in theory, enhance the tightness of the dural closure, as well as prevent complications such as pseudomeningocele or meningitis. Pijpker *et al*. have described a novel method of reconstructing the occipital bone after posterior fossa craniectomy by using a 3D-printed polymethylmethacrylate casting mold [64]. This method might help reduce the frequency of complications following posterior fossa decompression. Implant casting is highly precise and can be performed at the same time with the operation, as opposed to cranioplasty with autologous bone or titanium mesh that need to be shaped intraoperatively by the surgeon himself and with a decreased accuracy. On the other hand, performing a titanium mesh cranioplasty and tenting the dura ensures an expansive posterior fossa and may prove useful in precluding cerebellar ptosis and dural prolapse [65]. For this to be effective, the dura must be augmented in a watertight manner, and the cranioplasty should cover only the superior portion of the craniectomy to avoid re-stenosis of the foramen magnum. Although some authors prefer a partial superior C2 laminectomy so as to avoid kyphotic deformity [65], to the best of our knowledge, no study was conducted on the advantages of a limited C1 posterior arch removal. Following our own surgical experience, we are in favor of restricting the extent of posterior atlas resection to its upper half, as it maintains a stable arch, does not affect the mechanical integrity of the craniocervical junction and is, at least in our opinion, sufficient for adequately decompressing the foramen magnum.

Based on long-term follow-up, high rates of recurrence have been reported in patients undergoing cranio-cervical bony decompression without dural opening, regardless of age [66-69]. Nonetheless, the low risk of complications makes it a veritable treatment option for select patients [70, 71]. Although some surgeons reserve it solely for patients complaining of headache alone [69]. Intraoperative ultrasonography is helpful in verifying CSF flow and establish whether dural opening is warranted [33, 72]. A more recent current of thought is that neither surgical option for CMI is superior, and that the decision of treatment should be tailored to each individual [33, 34, 73].

Our technique is a hybrid between simple bony decompression and duraplasty, in the sense that the inner layer of the dura is not opened, so, in theory, there is minimal risk of CSF-related postoperative morbidity. Also, by dissecting the inner layer from the outer one and placing an autologous dural patch, we enlarge the foramen magnum and the posterior fossa, while further strengthening the tightness of the dura. By this rationale, our technique is hypothetically superior to extradural decompression alone. We are confident that, in the near future, this method will prove a safe and effective variant for CMI treatment, and plan on validating its efficiency in a prospective study. In that regard, to

compare treatment outcomes between conventional strategies and the interlayer dural split technique, scores such as the Chicago Chiari Outcome Scale (CCOS) may prove useful [74].

Conclusion

Our cases demonstrate that Type I Chiari Malformation in adults can be effectively treated via this innovative method of incising the outer dural layer and carefully separating the inner and outer dural layers from each other with a fine dissector. It is highly possible that our method will prove more effective than simple craniectomy without dural incision, while being less invasive than opening the inner dura, the arachnoid, or resecting the cerebellar tonsils. The “Interlayer dural split” technique must be validated by future a larger case series and prospectively compared to the other surgical methods.

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Conflict of Interest
The authors have no conflicts of interest to disclose.

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Figure 1. Intraoperative aspect. The outer dura is incised medially, with the inner layer kept intact and separated from the outer one with a dissector (1A). After ensuring proper CSF flow, seen as pulsating cerebellar tonsils through the transparency of the inner layer, we performed duraplasty with autologous fascia (1B).

Figure 2. Patient 1; A – preoperative MRI scan. Descending cerebellar tonsil herniation and cervical syrinx can be observed, consistent with mild Chiari Malformation type I. B – postoperative MRI scan at 72 hours. A slightly enlarged posterior fossa and a narrower ependymal canal can be noticed. C – control MRI at 3 months. A complete resolution of the foramen magnum crowding is visible.

Figure 3. Patient 2; A – preoperative MRI scan. The descending cerebellar tonsil herniation can be noticed, as well as the significant cervical syrinx. B – control MRI at 3 months. The cervical syrinx is markedly reduced, as is the foramen magnum crowding.

Figure 4. Patient 3; A – preoperative MRI scan. Descending cerebellar tonsil herniation and moderate syringomyelia can be noticed, consistent with Chiari Malformation type I. B –

postoperative MRI scan at 48 hours. A slightly enlarged posterior fossa is observed. C – control MRI at 3 months. The resolution of the foramen magnum crowding is noticeable, as well as a significant decrease of the syringomyelic cavity.