Technical Note

Fibrin sealant augmentation with autologous pericranium for duraplasty after suboccipital decompression in Chiari 1 patients: A case series

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Abstract

Background: The Chiari 1 malformation (CM1) involves decent of the tonsils of the cerebellum through the foramen magnum. Symptomatic disease requires a posterior fossa decompression with or without an expansile duraplasty. To date, the optimal surgical treatment for CM1 has not been delineated. The extent of bony removal, size of the dural opening, necessity for expansion of the dural space, choice of materials for the duraplasty, and possible need for augmentation with dural sealant are all factors that continue to be debated amongst neurological surgeons worldwide. We herein evaluate the use of fibrin sealant augmentation in combination with locally harvested autologous pericranium for duraplasty in adult CM1 decompression.

Methods: Retrospective data collected from January 2006 to December 2011. Data were reviewed for surgical site infection or meningitis, cerebrospinal fluid leak, symptomatic pseudomeningocele, radiographic improvement of hindbrain compression, and postoperative recurrence of symptoms at a minimum of 1 year of follow-up. Outcomes were studied clinically, radiographically, as well as by using a patient-specific questionnaire.

Results: Twenty-two consecutive patients were included. One patient required a revision for a delayed graft dehiscence in the setting of a rare form of aseptic meningitis with cerebrospinal fluid (CSF) pleocytosis due to a nonsteroidal anti-inflammatory drug (NSAID) allergy. All remaining patients had successful decompressions with full resolution of their symptoms except for one patient who had persistent headaches.

Conclusion: Autologous pericranium with dural sealant augmentation is an effective technique for expansile duraplasty in CM1 decompressions.
INTRODUCTION

The Chiari 1 malformation (CM1) is defined as protrusion of the cerebellar tonsils below the foramen magnum of greater than 5 mm [Figure 1a] with a prevalence of approximately 1 in 1,280 individuals, the majority of which are asymptomatic. A shallow posterior fossa (p-fossa) along with a congenitally smaller foramen magnum leads to hindbrain compression with descent of the tonsils, often causing symptoms such as tussive headaches, drop attacks, neck-, arm-, or back pain, swallowing difficulties, upper extremity dissociated sensory loss, or lower cranial nerve findings.

Surgical treatment of CM1 remains one of the most debated issues in neurosurgery today. CM1 with or without syringomyelia is managed with a suboccipital craniectomy with or without removal of at least the posterior arch of the C1-vertebra, depending on the extent of tonsillar descent and option of opening the dura to increase the craniospinal CSF space. Controversy persists around the optimal amount of bony removal, necessity for dissection of subarachnoid webs, need for duraplasty and choice of employed material, augmentation of dural closure with sealants, and need for tonsillar shrinkage. A survey by the AANS of pediatric surgeons revealed that 9% performed prophylactic surgery in asymptomatic patients, with treatment for symptomatic patients varying widely with 20% performing bony decompression alone, 30% augmenting with a dural graft, another 25% including intradural dissection of adhesions along with dural grafting, and 30% performing tonsillar resection on top of everything else. Recent meta-analysis comparing decompression with or without duraplasty found that added duraplasty was associated with a lower risk of reoperation than suboccipital decompression alone but had a greater risk of CSF-related complications. Specific to treatment of CM1 associated with a syrinx, Matsumoto and Symon did not notice any difference in reduction of syrinx size following duraplasty, but Munshi and colleagues found that patients had greater improvement of symptoms following duraplasty.

Expansion of the dura with graft material following bony decompression is intended to create a capacious p-fossa to prevent recurrence of symptoms. A watertight dural repair is ideal for preventing risks of infection, aseptic meningitis, pseudomeningocele, and inflow of blood. There are currently a host of autologous and foreign dural graft materials available for use. The ideal graft should not cause inflammatory reactions or adhesions, is watertight, readily available, inexpensive, and easily sterilized. Synthetic materials, such as polyester mesh, silastic, Teflon or GoreTex, bovine or porcine patches, are expensive, can be difficult to shape or suture, and can cause toxic or inflammatory reactions or even hemorrhage. Allogenic patches can induce immunoallergic reactions and have also been reported to be a conduit for transmittable diseases such as Creutzfeldt-Jakob Disease (CJD), hence they are considered suboptimal. The best material is one that will disappear when replaced by dura mater over time.

Meticulous closure of the dura is required following bony decompression for preventing CSF-related complications including meningitis, encephalitis, low-pressure headaches, arachnoiditis, and wound dehiscence. As true watertight dural closures are often impossible to achieve, dural sealants have been developed to be applied to the sutured dural perimeter to help prevent CSF-related complications. Several dural sealants are currently available for use, including trisylamine solution and polyethylene glycol hydrogels (Dura SealTM, Covidien LLC, Mansfield, MA, USA) and fibrin sealant (TISSEEL, Baxter, Deerfield, IL, USA). Adjuvant use of such sealants may be prudent particularly in p-fossa surgeries as the incidence of CSF leaks has been quoted to be as high as approximately 15%-28% with such surgeries, with an increased odds ratio of 5.84 when compared to supratentorial procedures. Given these potentially higher complication rates, we believe that diligent adjuvant use of a dural sealant along with autologous materials such as pericranium should decrease the rates of CSF-related or graft-related complications in CM1 decompression. We herein report our results with this technique.

MATERIALS AND METHODS

Study design

An institutional review board approved retrospective analysis was performed of clinical, and radiographic data collected in adult cases of symptomatic CM1. All patients underwent a standardized bilateral suboccipital decompression including at least a C1 laminectomy and a dural repair using locally harvested autologous pericranium augmented with dural sealant.

Operative procedure

Patients are positioned prone on the operative table with their head secured in Mayfield pins. Following clipping of the posterior hairline using an electric razor, a midline incision is fashioned one inch above the inion extending inferiorly to the level of the mid cervical spine [Figure 1b]. By extending the incision superiorly, we are able to perform blunt dissection of the subgaleal connective tissue to harvest adequate amounts of pericranium for dural grafting, as recently described by Stevens and colleagues. The graft is kept moist separately in Bacitracin enriched sterile saline until time for duraplasty [Figure 1c]. A suboccipital craniectomy is then performed using an Anspach highspeed side cutting drill bit by first removing occipital bone on either side.
of the midline keel, and then removing that keel using the drill as well as Kerrison rongeurs [Figure 1d]. The posterior C1 arch is removed in all cases (and possibly more inferior levels too) depending on the degree of tonsillar decent. Adequate bone removal exposes the p-fossa dura overlying the cerebellar hemispheres and the cervicomedullary junction. Dural bands constricting the craniocervical junction are released [Figure 1e] and the dura is then opened sharply in a Y-shaped fashion exposing the inferior aspect of the cerebellar hemispheres and the dorsal surface of the hindbrain [Figure 1f]. The arachnoid layer is opened and may be dissected in cases of arachnoid webs, but we do not routinely resect the cerebellar tonsils. Expansile duraplasty is then performed using the harvested pericranium [Figure 1g]. An induced Valsalva maneuver is then performed to ensure a watertight closure and a thin layer of dural sealant is then applied over the suture line and also the entire graft [Figure 1h], which is furthermore covered with a piece of Surgicel followed by another layer of dural sealant and finally GelFoam. A second induced Valsalva maneuver is then performed to document a watertight closure. The surgical wound is then closed in multiple layers beginning with muscle, ligamentum nuchae, subcutaneous tissue, and then, skin.

Postoperative follow-up
Patients were discharged from hospital and were seen in our clinic at 10-14 days, 3 months, and at 1 year following surgery for evidence of surgical site infection or meningitis, CSF leak, pseudomeningocele, and radiographic improvement of hindbrain compression as detected with a MRI scan [Figure 1i]. A questionnaire survey was filled out during interviewing of patients and recorded “yes” or “no” as well as quantified responses to the presence of the following symptoms prior to and at three months following, surgical decompression: tussive headaches, vertigo, visual disturbance, tinnitus, drop attacks, neck pain, gait and balance disturbances, dysmetria, bladder or bowel symptoms, dysphagia, and sensory deficits in the extremities.

RESULTS
Twenty-two CM1 patients were identified. Twenty-one had tussive headaches, five had vertigo, three had visual changes, four had neck pain, three had gait disturbances, and four had dissociated sensory deficits, prior to surgery [Table 1]. At 3 months and at final follow-up, all patients had full resolution of their clinical symptoms except one patient, who had persistent headaches. This

| Table 1: Patient demographics and summary of postoperative complications |
|--------------------------------------------------|
| Total cases                                      | 22                        |
| Male                                             | 4                         |
| Female                                           | 18                        |
| Average age (years±SD)                           | 37.3±12.7                 |
| Min                                              | 21                        |
| Max                                              | 61                        |
| Symptoms                                         |                           |
| Bladder/bowel dysfunction                        | 0                         |
| Drop attacks                                     | 0                         |
| Dysmetria                                       | 0                         |
| Dysphagia                                       | 0                         |
| Gait/balance difficulties                       | 3                         |
| Neck pain                                       | 4                         |
| Sensory deficits in the extremities              | 4                         |
| Tussive headaches                               | 21                        |
| Tinnitus                                        | 0                         |
| Vertigo                                         | 5                         |
| Visual disturbances                             | 3                         |
| Complications                                   |                           |
| Surgical site infection                         | 0                         |
| Meningitis                                      | 1*                        |
| CSF leak                                        | 1*                        |
| Pseudomeningocele                               | 0                         |

*The same patient experienced a cerebrospinal fluid leak and aseptic meningitis

Figure 1: Intraoperative images outlining the technique used for decompressing a symptomatic Chiari 1 malformation. (a) Representative sagittal T2-weighted MRI of a patient with a symptomatic CM1. (b) Midline incision from one inch above the inion extending inferiorly to the mid-cervical spine. Extension of the incision above the inion allows for harvesting of the occipital pericranium using Bovie cautery. (c) Harvested pericranium kept moist in saline. (d) Suboccipital craniectomy showing initial preservation of the midline bony keel, which is later removed using Kerrison rongeurs. (e) Release of dural bands exposing the cervicomedullary junction. (f) Y-shaped dural opening revealing contents of the hindbrain. (g) Expansile duraplasty with autologous pericranium. (h) Application of dural sealant over the dural graft and suture line. (i) Postoperative MRI showing full resolution of tonsillar herniation and a patent foramen magnum.
particular patient had a long history of frequent tussive headaches presenting as the only symptom of her CM1. One patient presented with a delayed CSF leak at 8 weeks due to a dehiscence of the dural graft requiring surgical revision and also subsequently developed aseptic meningitis and CSF pleocytosis due to an allergy to NSAID medications. None of the other patients had any CSF-related complications or surgical morbidities at the time of follow-up.

**DISCUSSION**

Symptomatic progression of CM1 is secondary to crowding of the cerebellar tonsils as they sit in a shallow p-fossa and smaller than normal foramen magnum. A suboccipital craniectomy releases compression at the cervicomedullary junction and aims to restore normal CSF dynamics between the spinal and cortical subarachnoid spaces, indirectly treating CM1 that is associated with syringomyelia. Whether dural expansion is required in CM1 decompression has remained a debatable topic. Studies have shown improvements in brain stem auditory evoked potentials after bony decompression alone with little additional gains associated with duraplasty. Matsumoto and Symon showed no difference in reduction of the size of the syrinx following duraplasty, but Munshi and colleagues demonstrated that patients had improvement in their symptoms following duraplasty. A meta-analysis of five retrospective and two prospective cohort studies totaling 582 patients demonstrated a significantly lower reoperation rate but higher rates of CSF-related complications in patients who received a duraplasty. Patients without duraplasty did not have significant improvements in their clinical exam or syringomyelia. Given existing evidence that supports the rationale of enlarging the craniospinal CSF spaces, we regularly include an expansile duraplasty in all of our CM1 surgeries. A recent study using a cerebellar tonsillar descent grading scale to determine when duraplasty is required during CM1 surgery found that duraplasty improves functional outcomes in patients with grade 3 tonsillar descent (descent below the C1 arch), while a suboccipital craniectomy alone may be adequate in grades 1 and 2 descent (descent not extending below the C1 arch). All of our study patients had an improvement in their neurological function follow expansile duraplasty, supporting the need for a generous enlargement of the p-fossa in selected patients.

We believe that, when available, autologous grafts should be used to prevent known adverse reactions to nonautologous materials including graft dissolution, encapsulation, immunoallergic reactions, and adhesions. In particular, autologous pericranium can be locally harvested using the same incision, is nonimmunogenic, inexpensive, and effective at creating a watertight closure, especially when combined with available modern dural sealants. This is in contrast to fascia lata grafts which require a second incision, ligamentum nuchae which may compromise fascial closure, or posterior atlantooccipital membrane grafts which have an increased risk of vertebral artery injury during harvesting. A survey of pediatric neurosurgeons by the AANS showed that most of the surgeons preferred to use autologous pericranium (52%). Vanaclocha and Saiz-Sapena compared freeze-dried cadaveric dura with fibrin sealant augmentation to autologous occipital pericranium alone and reported two CSF leaks and five pseudomeningoceles in the cadaveric dura group, while no complications were reported using autologous pericranium. We did not have any CSF-related complications in our study, thus further supporting the reported benefits of autologous pericranium when used in combination with fibrin- or hydrogel-based sealants. We have also expanded and validated our technique in a case series of 100 p-fossa surgeries in which none of the patients, other than the one Chiari patient reported in this study, required revision of their dural closure. However, a recent literature review did not find any superiority when comparing autologous to nonautologous grafts, which we feel is most likely due to the heterogeneity amongst studies. The authors of that study also state that their institutional experience dictates that autologous pericranium should be utilized when available and of good quality.

A watertight dural closure is often difficult to achieve when using dural grafts because of holes produced by surgical needles. Resultant CSF leaks and its associated complications, including low-pressure headaches and pseudomeningoceles, have led to the development of several types of dural sealants to reduce the risks of such complications. In particular, the incidence of CSF leak has been reported to be as high as 28% in p-fossa procedures due to its location being at the dependent portion of the skull base. To date, two prospective studies have been published using dural sealant alone or in combination with autologous duraplasty materials in cranial surgeries. The DuraSeal Pivotal Trial reported an infection rate of 8.1%, deep surgical infection rate of 7.2%, and 4.5% incidence of CSF leak. The second trial by Cosgrove and colleagues used a polyethylene glycol (PEG) hydrogel sealant and did not report any adverse events after three months of follow-up. Similarly, Boogaarts et al. prospectively treated 46 patients with a dural sealant in combination with different autologous dural substitutes and reported one overt CSF leak and one pseudomeningocele. There were no CSF leaks, pseudomeningoceles, or wound infections in our patients at any point during the follow-up period, further lending support for the use of a dural sealant in combination with autologous pericranium for dural...
 closure. This is in sharp contrast to increased rates of CSF leak, pseudomeningoecele formation, and infections when nonautologous materials are used in combination with a dural sealant.\cite{11}

A recent study compared retrospective data of patients who underwent cranial operations using nonautologous materials augmented with PEG hydrogel sealant for dural closure to patients from the Pivotal Trial who received autologous materials and found that there were no significant differences in the rates of CSF leaks, but that Pivotal Trial patients had a higher rate of meningitis.\cite{14} As this study included all patients who received craniotomies not specific to the p-fossa location alone, it is difficult to say whether the difference may be more apparent if a subgroup analysis had been performed. Of caution, there has been one case report in the literature of mass effect requiring reoperation due to in vivo postoperative swelling of the hydrogel sealant, highlighting the need for appropriate thickness of less than two millimeters when applying the sealant.\cite{1}

We attempted to further quantify the outcomes of our surgical technique in suitable CM1 patients using a questionnaire survey asking for the absence or presence of tussive headaches, vertigo, visual disturbance, tinnitus, drop attacks, neck pain, gait and balance disturbances, dysmetria, bladder or bowel symptoms, dysphagia, and sensory deficits in the extremities, before and after the decompression. Our survey is similar to the Chicago Chiari Outcome Scale (CCOS) published recently by Aliaga and colleagues in which they used four categories: pain-related symptoms (tussive headaches, neck and shoulder pain, dyesthesias in the upper extremities), nonpain-related symptoms (dysphagia, ataxia, vertigo, muscle weakness, sensory loss, tinnitus, paresthesias, and drop attacks), functionality, and surgical complications to assess their Chiari outcomes.\cite{5} They stratified 141 patients into two categories of either improved or unchanged outcomes. In the category of patients with improved outcomes (n = 101), approximately 45% of their patients had full resolution of their pain-related and nonpain-related symptoms, while approximately 65% had no complications and 30% had complications which resolved after a single lumbar puncture and lowering of CSF pressure. We did not include complications as part of our survey and kept it under a separate category of analysis. Our results differ to their study in that 90% of our patients (19 of 21 patients presenting with pain-related symptoms) had full resolution of their pain-related symptoms, and 95% of patients had no complications following the decompression (21 of 22 patients). The patients in their study also used autologous pericranium as a dural graft, but there is no mention of augmentation with a dural sealant, and they also included children in their study (mean age of diagnosis 20 ± 16.7 years, range 1-55 years), who are more prone to CSF-related complications following surgery. These factors may have contributed to their lower rates of resolution of symptoms and higher rates of complications compared to our study, further suggesting that augmentation with dural sealant enhances surgical results. Limitations of our study include a relatively small pool of patients with the lack of a control arm due to an absence of a gold standard protocol and a relatively short period of follow-up.

In summary, locally harvested autologous pericranium with dural sealant augmentation is a safe and effective surgical technique for CM1 decompression. Further validation with a larger patient pool, inclusion of a comparison arm, and longer follow-up period are required to establish superiority compared to other techniques.

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