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Original Article

Pseudomeningocele formation following chiari decompression: 19-year retrospective review of predisposing and prognostic factors

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Abstract

Background: Pseudomeningocele is a known operative complication of Chiari decompression with significant morbidity.

Methods: A retrospective analysis of 150 consecutive patients from November 1991 to June 2011 was conducted. Symptomatic pseudomeningocele was defined clinically; to meet definition it must have required operative intervention. Variables evaluated included sex, age, use of graft, and use of operative sealant. The Chi-square, Fisher test, and the two-sample *t*-test were used as appropriate to determine significance. Multiple logistic regression was used to determine independent risk factors for complication.

Results: A total of 67.3% of patients were female, with average age being 39.7 years. A total of 67.3% of patients had a graft placed with the most common being fascia lata. Only nine patients (6%) presented with pseudomeningocele. Factors observed to be significantly associated with pseudomeningocele development were age and use of sealant. Age and sealant use were also independent risk factors for complication. Adjusted for the significant effect of age, odds for complication among patients with sealant usage were 6.67 times those for patients without sealant. Adjusted for the significance of sealant usage, there is a 6% increase in odds for complication for every year increase in patient's age.

Conclusions: A statistically significant relationship exists between age and sealant use and the risk of developing a postoperative pseudomeningocele. Emphasis and attention must be placed on meticulous closure technique. This information can aide in preoperative planning and patient selection.

Key Words: Arnold-Chiari malformation, operative complications, operative sealant, pseudomeningocele

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INTRODUCTION

Chiari malformations represent a broad group of craniovertebral anomalies in which patients may present in a spectrum of different clinical scenarios.[17,21,22] Chiari is traditionally defined as an extension of the cerebellar tonsils through the foramen magnum, with distance required for radiographic diagnosis variable with age.[4,5]

Sub-occipital decompression for Chiari malformation represents a procedure with a wide range of techniques. Since the beginning of surgical approaches to this pathology, reported morbidity has been profound. In 1932, Van Houweninge Graftdijk described the first surgical treatment of Chiari malformation in his doctorate of medicine thesis entitled *Over Hydrocephalus*.[34] There, he advocated resection of the occipital bone, incision of the dura mater, and reduction of the tonsils in an attempt to improve cerebrospinal fluid (CSF) circulation at the level of the foramen magnum. The surgical mortality in this series was 100%. In 1935, Russell and Donald extended decompression to the spinal cord at the level of the foramen magnum to facilitate CSF circulation.[34] Complication rates currently published in the literature range from 4% to 30%.[24,28,35] Those most commonly reported include aseptic meningitis, CSF leak, wound infection, failure of procedure, and pseudomeningocele.[1,11]

The most clinically volatile complication of Chiari decompression is the pseudomeningocele.[25] Treatment management algorithms, although varying from institution to institution, adhere to the same clinical principles. The focus is on diversion of flow from the weakened construct to allow for further healing processes to strengthen the closure. The first standard approach is tightly wrapping the head. Fluid, following the path of least resistance, is less likely to test the posterior fossa closure. This reduces the initial burden on the assumed watertight closure. This is often combined with a large volume lumbar puncture. Again, the focus is on flow diversion removing large volumes, often 40–50 cc. This will reduce the initial volume of pseudomeningocele, and combining this procedure with wrapping the head in a tight ace bandage maximizes its effectiveness of flow diversion.[30] Failure of these initial management protocols will usually result in the placement of a lumbar drain.[33] This more invasive technique maximizes the same physiological principles. Generally, this is left for a period of 3–5 days and is often combined with wrapping the head. Those cases failing in conservative management ultimately require surgical intervention with re-establishment of a watertight closure.[34]

Primary closure techniques vary significantly from center to center and can involve many possible permutations of auto- or allograft and sealant.[7,24,41] Duraplasty is supplemented by a variety of synthetic and autologous substitutes.[7] Pericranium can be harvested during the dissection.[36] Fascia lata can be harvested by use of an additional incision. Bovine pericardium can be substituted as well as a variety of synthetic materials.[19] This can be supplemented with synthetic or fibrin-based sealant.[16] However, the primary protection and burden is placed on surgical technique. Watertight closure must be ensured through meticulous closure.

The current investigation reviews the long-term trends in complications following Chiari decompression at a large, academic teaching hospital.

MATERIALS AND METHODS

Review Protocol was approved by the Institutional Review Board. A retrospective analysis of patient charts, operative records, and radiology reports was performed upon consecutive patients undergoing operative intervention for Chiari malformation at our institution from April 1991 to June 2011. This was performed using the historical database available for all surgical patients created by the Department of Neurosurgery. Patient characteristics collected and recorded included age, sex, race, diagnosis, surgical treatment, including use of graft or operative sealant, date of admission, and imaging characteristics.

The information was extracted by the same chart reviewer using an electronic database as well as the electronic chart system. Inclusion criteria included patients undergoing a first time decompression via sub-occipital craniectomy for Chiari malformation. Furthermore, this was limited to type I Chiari malformations as linked with the admission or surgical diagnosis. Focus was on pseudomeningocele that was both symptomatic and operative. Again, this implies that the pseudomeningocele had to be identified clinically and radiographically; it also had to result in an operative intervention. Other complications were not considered in the aim of this study.

Data on 150 patients were analyzed to determine factors significantly associated with complication. The Chi-square or Fisher test was used to determine significant categorical factors, and the two-sample *t*-test was used to determine association of age with complications. Multiple logistic regression was used to determine independent risk factors for complication.

Operative procedure

Surgical goals for Chiari decompression include decompression of the inferior cerebellum, enlargement of the posterior fossa, and the establishment of CSF flow.[9] Following induction of general anesthesia, patients are positioned in the prone position with cranial immobilization utilizing a Mayfield 3-point rigid cranial fixation system. Overlying hair is clipped to accommodate a skin incision from the inion to the spinous process of C2, and the region is prepped and draped in the usual sterile fashion. Suboccipital musculature is dissected with the aid of electrocautery in a subperiosteal fashion so as to expose the occiput approximately 3 cm superior to the foramen magnum and 1.5 cm from the midline bilaterally as well as the entirety of the posterior arch of the C1 vertebra. Depending upon attending preference, pericranium may be harvested prior to bony removal,

or alternately following the durotomy. Sub-occipital craniectomy is then performed approximately 3 × 3 cm from the foramen magnum.[12] In cases involving tonsillar descent below the posterior arch of C1, a laminectomy is usually performed from 10 to 12 mm from the midline, paying particular attention to the location of the posterior loop of the vertebral artery.[26] The posterior fossa dura is opened in the midline in the region of C1 and extended in a Y-shaped fashion over the convexities of the cerebellar hemispheres, bilaterally. Duraplasty is then performed with either previously harvested pericranium, bovine pericardium, autologous fascia lata or synthetic graft, depending upon primary surgeon preference.[23] This is closed in a continuous fashion beginning at opposing ends of the graft utilizing 4-0 braided, monofilament sutures, and a water-tight closure is ensured with a Valsalva maneuver. The decision to utilize an operative sealant is made on a case by case basis and includes either an absorbable polyethlelene glycol ester (DuraSeal, Covidien, Mansfield, MA) or a human fibrin sealant (TISSEEL, Baxter Healthcare, Deerfield, IL) The incision is then closed in multiple layers using interrupted absorbable sutures in muscle, fascia, and subcutaneous layers and a continuous monofilament suture at the skin.

RESULTS

A total of 150 patient met inclusion criteria. Demographic information, operative variables, and incidence of complications are included in [Table 1]. Mean age at presentation was 39.7 years, with a range of 1–76 years and a standard deviation of 17.6 years. The majority of patients (101/150) were female (67.3%). Only 49 (32.7%) were male. Forty-six patients (30.7%) had sealant utilized prior to wound closure. Nineteen (12.7%) cases utilized absorbable polyethlelene glycol ester, 9 (6.0%) cases utilized fibrin sealant, and 17 (11.3%) cases utilized a sealant without specific mention of type. A majority of patents (67.35%) had a graft placed at the time of surgery. The most common was fascia lata, in 52 patients (34.7%). Five cases, or 3.3%, utilized bovine pericardium. Pericranium was harvested and used in 18 (12%) patients. Twenty-six cases (17.3%) utilized a synthetic graft.

Only nine patients (6.0%) experienced an operative pseudomeningocele. The factors observed to be significantly associated with having an operative pseudomeningocele were age and utilization of operative sealant during closure. Comparisons between patients with and without complications are shown in [Table 2]. The mean age of patients with a complication was noted to be 52.0 years (range 17–76 years). Only 6/150 (4%) were pediatric cases (age < 14) and 8.7% (13/150) of cases were over the age of 65. The average age of those patients without complication was 38.9 (range 1–76 years) (P = 0.03). Six (66.7%) patients suffering an operative pseudomeningocele had operative sealant placed at the time of their surgery, while only 28.4% (40/150) of those patients without a complication had sealant utilized during their operation (P = 0.02). A total of 7/9 (77.8%) with pseudomeningoceles were in patients with a documented duroplasty. A total of 7/102 (6.9%) patients with documented duroplasty had a pseudomeningocele as opposed to 4.1% (2/48) of those who did not (P = 0.71).

Following multiple logistic regression analysis, age and usage of operative sealant were determined to be independent risk factors for complication [Table 3]. Adjusted for the significant effect of age, odds of experiencing an operative pseudomeningocele among patients with were 6.67 times higher with the usage of operative sealant than without (P = 0.01). Adjusted for the significant of operative sealant, there is a 6% increase in odds for complication for every year increase in patient's age (P = 0.02).

Table 1: Summary statistics on patient characteristics and outcomes (*N*=150)

Characteristics/outcome	Number (%) or mean±SD, range
Gender	
Female	101 (67.3)
Male	49 (32.7)
Age (years)	39.7±17.6 (1-76)
Graft	
None	49 (32.7)
Bovine pericardium	5 (3.3)
Fascia lata	52 (34.7)
Pericranium	18 (12.0)
Synthetic	26 (17.3)
Sealant usage	46 (30.7)
Had complication (s)	9 (6.0)

Table 2: Significant comparisons between patients with and without operative pseudomeningoceles

	Mean±SD, range or number (%)		
Factor	Had complications (*N*=9)	No complications (*N*=141)	*P* value
Age (years)	52.0±19.3, 17-76	38.9±17.3, 1-76	0.03*
sealant usage	6 (66.7)	40 (28.4)	0.02*

*Significant at 5% level (0.01< *P* value <0.05)

Table 3: Independent significant risk factors for operative pseudomeningoceles

Factor	Adjusted odds ratio (OR)	95% CI for OR	*P* value
Sealant	6.67	1.49-29.41	0.01*
Age	1.06	1.01-1.10	0.02*

DISCUSSION

The pathophysiology of Chiari malformation, as well as the most appropriate management of its myriad symptoms has vexed physicians for centuries. Among the first documented observations were made by Realdo Colombo in 1572, when he described the syndrome of atlas assimilation.[19] Ackermann would go on to describe basilar impression (also known as basilar assimilation) in 1790.[2] In 1883, John Cleland described the case of a child presenting with spina bifida, hydrocephalus, as well as anatomical abnormalities of the brainstem and cerebellum.[29,34] The following year, Julius Arnold published a similar case of spina bifida associated with elongation and descent of the inferior cerebellum into the spinal canal. In 1891, a Viennese pathologist named Hans Chiari reported the case of a 17-year-old female with elongation of the tonsils and medial divisions of the inferior lobules of the cerebellum into cone shaped projections, which accompanied the medulla oblongata into the spinal canal.[2,8] At the time of discovery, Chiari thought these anatomical malformations must have been due to hydrocephalus; although he acknowledged the discoveries of Cleland and Arnold in his series of 14 additional cases, he also expanded his description to include descent of the inferior vermis, pons, medulla, and fourth ventricle into the spinal canal. In postulating the pathogenesis of this series of malformations, he believed that the most important factor was related to insufficient skull growth resulting in increased intracranial pressure. Chiari also believed that the degree of tonsillar descent could be secondary to the age of onset, as well as the length and severity of hydrocephalus.[34]

Incidence of operative pseudomeningocele

From the beginning of operative intervention for Chiari malformation, surgical morbidity has been high. Contemporary rates have significantly diminished from historical rates as high as 100%. Overall, the 6% rate of symptomatic, operative pseudomeningocele demonstrated in this series compares favorably with published rates. In 2012, Klekamp *et al.* reported a 5.9% rate of CSF fistula in 371 consecutive decompressions with duraplasty,[15] while several well-cited pediatric series have demonstrated rates as low as 2.4%.[13,38] Similarly, Alfieri *et al.* reported a 2.7% rate of CSF leak in a mixed retro- and prospective series of 109 consecutive cases, with only 1 case (0.9%) requiring surgical intervention.

Patient age

Increasing patient age was found to be significantly associated with increasing rates of complication. Intuitively, age is seen as risk factor for developing a pseudomeningocele, as it has been well established as a predictor of operative morbidity and mortality in multiple series.[6,10,40] However, several large series have found no role for patient age in the incidence of postoperative complications following posterior fossa surgery.[18,32] This makes our series, to the best of our knowledge, the first to associate patient age with incidence of complication following posterior fossa decompression.

Operative sealant

Additionally, the use of any sealant, without respect to type, increased the chances of operative pseudomeningocele in this series. It is likely surgeons are more likely to use sealant in the face of a possible complicated closure. Than *et al.* performed a direct comparison between fibrin-based and polyethylene glycol (PEG) sealant augmented dural closure in 100 consecutive posterior fossa surgeries and found a significantly higher rate of incisional CSF leak in the former group.[37] Parker *et al.* found exactly the opposite, with 50% of the cases utilizing PEG sealant experiencing a complication with a rate of only 18.7% with fibrin glue in their series of 114 pediatric cases.[27] Laboratory work has recently identified a pathophysiological difference between these two choices of operative sealant. In 2013, Ito *et al.* compared the clinical and pathological effects of duraplasty in Japanese white rabbits by creating bilateral dural defects in the same animal and treating each with opposing sealants. Statistically significant increases in dural regeneration were seen in rabbits treated with fibrin sealant versus synthetic sealant. This also corresponded with higher rates of abscess and granulation tissue formation in the PEG group.[14] While the current investigation was not sufficiently powered to demonstrate a difference among the difference sealants used, we believe our data corroborates a growing body of literature implicating these agents in wound healing complications. This represents a correlation of spinal fluid leaks with the use of sealant. Safe assumption in our series is that cases without duroplasty would not receive operative sealant.

Graft selection

While the current series did not find any correlation or statistically significant difference in outcome based upon the selection of dural graft, the literature is replete with case series purporting the superiority of autologous, synthetic, or xenographic materials.[9,20,31,39] In 2011, Abla *et al.* undertook a critical review of the existing literature, including 108 publications referencing nonrevision Chiari decompression, including duraplasty.[1] After narrowing these results to three that directly compared different types of dural grafts, they concluded that no difference in outcome existed between cases utilizing autologous versus nonautologus grafts. Interestingly, in the subgroup analysis, a nonsignificant increased rate of incisional CSF leaks and symptomatic pseudomeningocele in the group utilizing pericranium as compared with the expanded polytetrafluroethylene (ePTFE) group; however, the rate of asymptomatic pseudomeningocele was found to be higher in the nonautologous group (22% vs. 10%). Despite these findings, the authors continued to

recommend the utilization of pericranium as dural graft secondary to its nonimmunogenicity and capability of "creating a watertight closure."

In a retrospective review of their pediatric experience, Attenello *et al.* published a radiographic and clinical outcome comparison between autologous pericranial and an "antiadhesive" ePTFE dural substitute graft.[3] Their findings suggested an improved maintenance of posterior fossa decompression without scarring, as evidenced by cine-flow studies as well as significant improvement in radiographic evidence of syrinx when compared with pericranium grafting. Revision decompression was required in 10% in the pericranium group and in none of those utilizing synthetic graft ($P = 0.090$). The authors, however, reported no significant differences in the incidence of CSF leak or symptomatic pseudomeningocele. Historical preference was for extradural decompression.

Most recently, in 2013, Williams *et al.* published a prospective, randomized comparison of sutureless synthetic duraplasty with a watertight closure with bovine pericardium grafting, without the use of operative sealant in either case.[41] Their results indicate a short-term improvement in quality of life measures at 2 months postoperatively ($P < 0.05$); however, no differences existed between groups at long-term follow up. Radiographic pseudomeningocele occurred more commonly in the patients undergoing sutureless closure (31.2% vs. 22.2%) and CSF leak occurred in 12.5% of this group, as compared with 0% of the sutured bovine pericardium group. Neither of these comparisons, however, reached statistical significance, likely secondary to the somewhat small sample size ($n = 34$).

The preponderance of contradictory data regarding choice of dural graft and its effect on postoperative complications seems to suggest that the factors likely responsible for poor outcomes are more surgeon and technique-driven. The creation of a watertight duraplasty and proper, layered closure of the operative incision probably contribute significantly to the overall outcome more so than the specific choice of material for implantation. Of course the corollary of this argument also holds true: no amount of technological advancement will even overcome the effects of a nonmeticulous surgical closure.

Limitations

This investigation is limited by several factors inherent to its retrospective design. Information regarding specific operative technique not listed in the operative note was unavailable. This included but was not limited to type or technique of stitch used, level of training for surgeon closing the wound, and the reason regarding the type of graft used. Specific information regarding the opening of the arachnoid membrane, a known risk factor for pseudeomeningocele formation, in duroplasty cases was also not available. The decision-making process regarding when to use sealant was also unavailable. While standard operating technique at our facility would dictate a Valsalva maneuver following completion of the duraplasty, if these results were not noted in the operative report, they were unavailable for analysis. This includes whether or not the dura was fully opened. Additionally, data regarding the selection of graft material selection was not available in 49 patients. As a result, we were unable to accurately compare this variable over the 20-year study period. The effect of graft selection on outcome following posterior fossa decompression needs to be further evaluated and investigated in future study.

Furthermore, this study defined a complication as a symptomatic and operative pseudomeningocele. Size, presence of a transcutaneous fistula, or attending's treatment algorithm for pseudomeningocele treatment was not available. While this complication has been shown to significantly reduce the immediate efficacy of decompression and diminish overall improvement in health at one year, the data presented here does not capture the incidence and outcome of symptomatic pseudomeningoceles treated conservatively with either placement of a lumbar drain or needle decompression followed by a tight head wrap.[25]

CONCLUSION

This investigation is the first to establish a statistically significant relationship between the risk of developing of a postoperative pseudomeningocele following posterior fossa decompression and increasing patient age as well as use of operative sealant. This data can aide surgeons in risk stratification and preoperative planning prior to performing these procedures. Focus is on operative technique. We believe that these results emphasize the importance, not only of meticulous attention to detail when performing the duraplasty, but also flexibility in technique allowing for alternate choices in dural substitute or operative sealant as the individual case may dictate.

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Dr. Anil Nanda is Professor and Chairman, Department of Neurosurgery at LSU Health Sciences Center in Shreveport, LA.

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Commentary

The modern era of Chiari surgery was ushered in by Bernard Williams, the first neurosurgeon to intensively deal with the pathology of Chiari I malformation.

At the end of his prematurely interrupted life, he wrote an editorial advocating for leaving the dura open at the end of Chiari decompressions.[6]

In his opinion, leaving behind an iatrogenic pseudomeningocele was more acceptable than dealing with the mass effect of a pseudomeningocele caused by the ball valve mechanisms of an imperfectly closed duraplasty.

This article caused a schism in the Chiari surgical technique, which still endures today, between the American lore of closing the dura with a duraplasty and the British tradition of deference to the parting words of Mr. Williams.

In the following years, Ulrich Batzdorf M.D., Arnold Menezes M.D., and Thomas Milhorat M.D., demonstrated that through repetition, improvement, and impeccable technique, the incidence of pseudomeningocele could be contained to values below 5%.[1]

Years later, the pseudomeningocele is still the defining complication in the field of Chiari surgery.[2,5]

Pseudomeningoceles can negatively affect the patient via a number of different mechanisms: (i) mass effect on the duraplasty and the dural contents; (ii) aseptic meningitis; (iii) negative effects on cerebrospinal fluid (CSF) flow and pressure; and (iv) transcutaneous fistulas (with consequent bacterial meningitis).

Patients with a history of large postoperative pseudomeningoceles are often the recipients of subsequent ventriculo-peritoneal shunts, in the face of elevated CSF pressures.

The real overall incidence of pseudomeningoceles is probably underestimated, since many Neurosurgeons spring to corrective surgical action only in the case of transcutaneous CSF fistulae. Moreover, a number of Neurosurgeons tend to "accept" the chronic presence of nonexpanding pseudomeningoceles, as a part of the normal spectrum of postoperative surgical results.

The concepts of "failed Chiari surgery" and of "posterior fossa revision" are quite recent and have gained traction thanks to the birth of Centers dedicated to the diagnosis and management of Chiari I malformation. Persistent pseudomeningoceles are increasingly recognized as one of the causes for "failed Chiari surgery" in quite a large number of patients.

Running locked, running unlocked, and interrupted stitching configurations have been used in Chiari surgery, with similar results in the best of hands. Flawless execution is the only determining factor for successfully avoiding pseudomeningoceles, in the face of different needles and stitching materials used.

The dura of Chiari patients has several unique, challenging features. It is extremely thin over the cerebellar hemispheres (at the very top of the Y-shaped dural incision), because of the combined effects of a small posterior fossa and a pulsatile, crowded cerebellum. It is also very adherent to the periosteum at the level of the foramen magnum. Dural rents or shredding of the outer dural layer during the craniectomy can increase the chance of a postoperative pseudomeningocele.

The mismatch between a thin, fragile dura and a stiff, thick duraplasty (i.e., bovine pericardium, GoreTex) can create undue tension at the dural edge of the durarraphy, with leaking stitch-holes.

Watertight closure is to be tested with Valsalva maneuvers. As a rule, one or two Valsalva maneuvers are not enough. Attention should be paid to the air–fluid level (often visible through the thinned and transparent dura), being sure that the entire height of the durarraphy is below such level, to guarantee reliable Valsalva testing. We routinely fill the subdural space with 20–40 cc of injectable saline before passing the last dural stitch, to improve the odds.

Dural sealants have been engineered to prevent pseudomeningoceles.[4] While these compounds have demonstrated themselves quite valuable in the supratentorial compartment, their record in the posterior fossa (and especially with Chiari surgeries) is not as impressive.

In the posterior fossa, gravity tends to bring the CSF through the dural stitching (and not away from it like in the supratentorial compartment).

Dural sealants cannot and should not be regarded as a magical remedy or a compensation to less than meticulous dural suturing.

Extradural techniques of Chiari decompression have been advocated to try to avoid pseudomeningoceles altogether.[3] Since the intrinsic limitations of the extradural Chiari techniques are not the focus of this Editorial, we will only say that a downside of this strategy has been the decreased number of dural openings and duraplasties.

Our preoccupation is that the new generations of Residents, tempted by the availability of sealants and extradural techniques, could end up becoming less proficient in the fine art of a watertight dural closure of the posterior fossa.

Dr. Liu and his colleagues have recently combined a subpial tonsillar resection along with a primary durarraphy (i.e., without a duraplasty). Using a variant of this technique, we were able to perform more than 250 cases without a single instance of pseudomeningocele, exploiting the finding that the Chiari dura is at its thickest along the midline.

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