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Surgical Neurology International

Editor-in-Chief: Nancy E. Epstein, MD, NYU Winthrop Hospital, Mineola, NY, USA.

SNI: Unique Case Observations

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Case Report

Repair of encephalocele and cerebrospinal fluid leak with the use of bone morphogenetic protein: A case report

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Received : 13 December 18
Accepted : 20 February 19
Published : 24 April 19

DOI
10.25259/SNI-137-2019

Quick Response Code:

ABSTRACT

Background: Encephaloceles are rare phenomena which occur when brain parenchyma herniates through a skull defect which, if left untreated, may lead to significant issues such as cerebrospinal fluid (CSF) fistulas, meningitis, and intractable seizures. Due to the rarity and variety in size and location of encephaloceles, no standard technique has been established for the resultant defect. Herein, we demonstrate the safe and effective use of bone morphogenetic protein (BMP) in the repair of CSF leak caused by encephalocele.

Case Description: A retrospective chart review was conducted on a 50-year-old female who presented with sudden onset spontaneous right nostril CSF leak due to the right lateral sphenoid sinus recess encephalocele, for which she underwent surgical repair. After resecting the encephalocele, cadaver crushed bone was used to fill the skull base defect. Following, an absorbable sponge from the extra-small BMP kit was cut in half and soaked with recombinant human BMP-2 (rhBMP-2) before being laid over the bony defect. On postoperative clinic visits at 2 weeks and at 3 months, the patient demonstrated good recovery without evidence of recurrent CSF leak. On follow-up computed tomography imaging at 9 months' postsurgery, there was no evidence of recurrent CSF leak or encephalocele, infection, ectopic bone formation, excessive inflammation, or neoplasm.

Conclusion: In this case, we demonstrate the successful use of BMP for the repair of CSF leak due to encephalocele. It is our extrapolation that the pro-inflammatory properties of rhBMP-2 lead to the prevention of recurrent CSF leak.

Keywords: Bone morphogenetic protein, cerebrospinal fluid leak, encephalocele, encephalocele repair

INTRODUCTION

A herniation of brain parenchyma through a skull defect is considered to be heterotopic brain tissue known as encephalocele [Figure 1].[12] They are rare phenomena that carry significant morbidity and mortality if not treated, including cerebrospinal fluid (CSF) fistulas, meningitis, and intractable seizures.[12,13] Due to the rarity of the condition and variable size and location of encephaloceles, there is no standard technique for the treatment of the resultant defect. Techniques used for the resection and repair include endonasal endoscopic and open craniotomy approaches. The endoscopic approach requires a well-experienced rhinologist and poses

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a challenge if the defect is located in the lateral sphenoid recess.[6] The open craniotomy approach is more common and familiar to neurosurgeons; however, it can lead to neurologic complications due to brain retraction.[9] In both approaches, the recurrent CSF leak is one of the most fraught after complications.[4,8]

We present a case of spontaneous right lateral sphenoid sinus encephalocele with CSF leak in a 50-year-old female who underwent a right middle fossa craniotomy for resection of the encephalocele. In our institution, we have developed a technique for the repair of dural defect for recurrent CSF leak with the use of recombinant human bone morphogenetic protein-2 (rhBMP-2) after transsphenoidal pituitary resection. BMPs are signaling molecules and belong to a superfamily of proteins known as transforming growth factor-β that are mostly involved in osteogenesis.[10] BMP use has been associated with complications such as ectopic bone growth, osteolysis, and systemic neoplasm.[2] We sought to demonstrate that BMP could be used in encephalocele repair to prevent recurrent CSF leak without long-term complications.

CASE REPORT

A 50-year-old female without a significant history of infection, tumor, or trauma presented to the neurosurgery clinic with profuse watery drainage from her right nostril. The drainage was found to be consistent with CSF. The computed tomography (CT) and magnetic resonance imaging demonstrated a defect in the middle cranial fossa on the right with the encephalocele extending into the sphenoid sinus [Figure 2]. The endoscopic endonasal approach was not feasible due to the lateral sphenoid recess location of the encephalocele.

A lumbar drain was inserted before surgery. A right temporal craniotomy was performed in a standard fashion. Using the microscope, the dura was separated from the middle cranial fossa until the dural defect and encephalocele were identified. The encephalocele was pulled through the bony defect. At this point, the dura was incised. The temporal lobe was identified and gently lifted to expose the encephalocele which was protruding through the dural defect. Following, the encephalocele was resected and was measuring 2 cm × 2 cm × 0.4 cm. The bony defect was then repaired by tamping crushed cancellous cadaver bone into the opening. The absorbable sponge from the extra-small BMP kit was soaked with rhBMP-2, cut in half, and laid over the bony defect. A piece of collagen matrix (DuraGen®) was placed above the BMP sponge to completely cover the BMP. Another piece of DuraGen® was laid inside the dural defect due to the lack of access to the dural defect for primary repair [Figure 3]. The remaining part of the surgery was completed in the standard manner.

The lumbar drain remained in place and was open to drain for 7 days. The patient was discharged home on the postoperative day 8. She was followed up clinically at 2 weeks, 3 months, and 9 months after the surgery. CT scan at 9 months' postsurgery showed no findings to indicate recurrent CSF leak, encephalocele, infection, extraneous bony growth, excessive inflammation, or neoplasm [Figure 4].

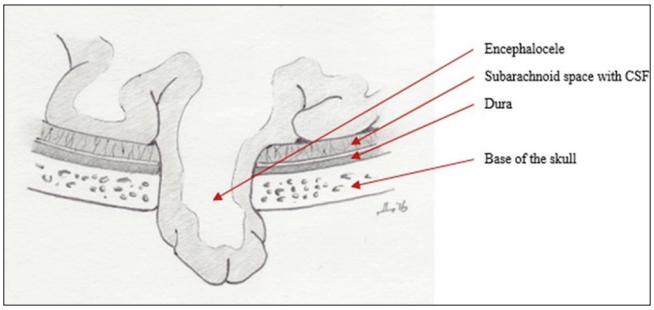

Figure 1: Brain parenchyma extrusion through a skull defect creating an encephalocele (Original material).

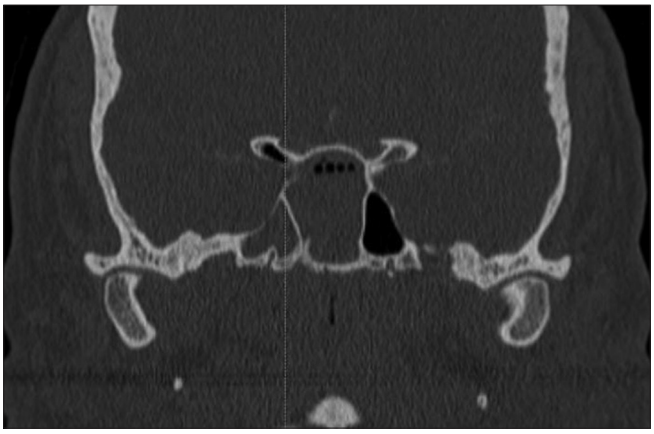

Figure 2: Postoperative CT w/o intrathecal contrast shows the satisfactory appearance of the paranasal sinuses indicating no recurrent cerebrospinal fluid leak.

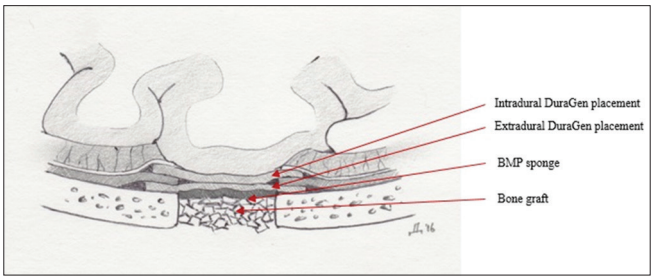

Figure 3: Preoperative CT with intrathecal contrast depicts the presence of the contrast dye in the right sphenoid sinus which is indicative of encephalocele that is not clearly identifiable on the image.

DISCUSSION

Encephaloceles are uncommon abnormalities of brain tissue herniations through a skull defect that can be of congenital, iatrogenic, posttraumatic, or spontaneous origin.[1,12] A spontaneous skull base encephalocele in an adult carries significant morbidity and mortality associated with the potential development of CSF fistulas, meningitis, and intractable seizures.[12,13] Surgical

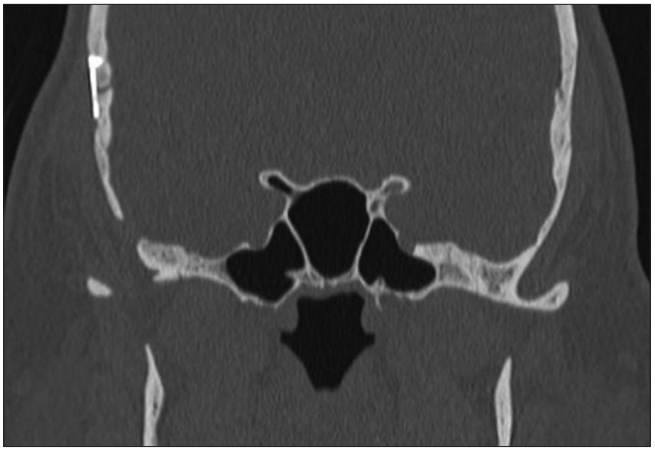

Figure 4: Repair of encephalocele with the use of bone morphogenetic protein, DuraGen®, and bone graft (Original material).

techniques either endoscopic endonasal or open craniotomy can be utilized to repair the defect. One of the more common complications of either approach is the development of recurrent CSF leak quoted anywhere between 6.6% and 9%.[6]

In our case, we were able to show a safe and effective technique for the repair of CSF leak secondary to the right lateral sphenoid sinus recess encephalocele with the use of BMP. In 2002, rhBMP-2 was approved by the United States Food and Drug Administration for use in lumbar fusions due to its osteoinductive properties.[7] A recent article by Huang *et al.* demonstrated that rhBMP-2 induced an inflammatory state as evident on histologic tissue sections and systemic blood samples of rats with subcutaneously implanted BMP.[5] We believe that this pro-inflammatory BMP-induced response leads to faster scarring and healing of the skull base defect helping prevent a recurrent CSF leak. In our institution, we successfully treated recurrent CSF leaks with BMP in a series of four patients after a transsphenoidal pituitary tumor resection.[11]

In recent years, articles have been published on complications associated with the use of BMP that includes ectopic bone formation, osteolysis, and malignancies with higher BMP dose formulations.[2,3] In our study, none of the complications associated with BMP use was observed. Although BMPs are meticulously investigated signaling molecules, more research is required to completely elucidate their function and use in surgery, wound healing, and tissue repair. Future studies should focus on further demonstrating the safety of BMP in the treatment of CSF leak through stronger study designs, such as a randomized controlled trial.

CONCLUSIONS

Encephaloceles are rare brain parenchymal herniations through a skull defect, that if left untreated may lead to CSF fistula formations, seizures, and meningitis. Here we demonstrate a safe application of BMP in the repair of encephalocele and persistent CSF leak.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Slavnic D, Tong D, Barrett RP, Soo TP. Repair of encephalocele and cerebrospinal fluid leak with the use of bone morphogenetic protein: A case report. Surg Neurol Int 2019:10:55.