

Case Report

Primary repair of open neural tube defect in adulthood: case example and review of management strategies

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Received 24 March 2015; revised 10 June 2015; accepted 23 July 2015

Abstract

BACKGROUND CONTEXT: Neural tube defects are congenital malformations that develop when the neural plate fails to close during embryogenesis. The most common open neural tube defect, myelomeningocele (MMC), is declining in frequency in North America. If identified, an MMC must be closed in the perinatal period to prevent lethal complications. Lesions presenting in older adults are, thus, very uncommon.

PURPOSE: The purpose of this study was to describe the surgical management of an adult with an unrepaired ulcerated lumbosacral MMC who presented with persistent cerebrospinal fluid (CSF) leakage and to review the management strategies for adult patients with unrepaired MMC.

STUDY DESIGN: A case report was used for this study.

METHODS: The patient was a 62-year-old woman with an unrepaired ulcerated lumbosacral MMC and associated lower extremity weakness. She sought medical care for persistent lumbar tenderness and ulceration after sustaining a fall 4 months before admission. Physical and radiological assessment revealed a lumbosacral MMC at the L5 and S1 levels and a tethered cord. Surgical resection of the placode and de-tethering were performed.

RESULTS: One week after repair of the lumbosacral MMC, the patient was readmitted for management of continued CSF leakage and hydrocephalus, requiring external ventricular drainage, wound revision, and placement of lumboperitoneal shunt. The patient experienced complete resolution of back pain without additional episodes of CSF leakage.

CONCLUSIONS: This rare case and review of management strategies suggests that proper surgical management of open MMC in adulthood can successfully be performed and improve patient symptoms and prevent further complications. © 2015 Elsevier Inc. All rights reserved.

Keywords:

Cerebrospinal fluid; Hydrocephalus; Lumboperitoneal shunt; Myelomeningocele; Spinal dysraphism; Tethered cord

Abbreviations: MMC, myelomeningocele; CSF, cerebrospinal fluid.

FDA device/drug status: Not applicable.

Author disclosures: **JG:** Nothing to disclose. **VMR:** Nothing to disclose. **WZR:** Nothing to disclose. **RE:** Nothing to disclose. **ATD:** Dr Dailey reports grants and personal fees from Biomet (C), personal fees from AONA (B), outside the submitted work.

The authors report no conflict of interest concerning the materials or methods used in this study or findings specified in this paper.

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Introduction

Neural tube defects are a family of congenital malformations that are thought to develop when the neural plate fails to close during embryogenesis [1]. Myelomeningocele (MMC), the most common type of open neural tube defect, is characterized by herniation of meningeal membranes and spinal cord tissue through a bony defect in the vertebral column, frequently in the lumbosacral region [2]. According to Centers for Disease Control and Prevention estimates, MMCs occur in 3 out of every 10,000 live births, accounting for the chronic disability of nearly 100,000 individuals

in the United States (<http://www.cdc.gov/ncbddd/spinabifida/data.html>) [1].

As a result of increased prenatal screening, the vast majority of neural tube defects are diagnosed early in pregnancy, and if pregnancy is carried to term, the defect is surgically repaired within 72 hours of birth. Although the benefits of prenatal and early postnatal repair are well established [3], primary MMC repair in adolescent and adult patients has been sporadically reported [4–6]. The risks of not treating MMC include progressive neurologic dysfunction, chronic meningitis, and death [7]. Here, we report a unique case of a 62-year-old patient with open lumbosacral MMC who underwent primary surgical repair, and we review management strategies for similar published cases. This is one of a few cases of delayed MMC repair in an adult requiring surgical intervention for persistent cerebrospinal fluid (CSF) leakage following an injury.

Case report

History and examination

A 62-year-old woman from a rural area had a lifelong history of an open lumbosacral MMC that had been judged non-operable at the time of birth. Minimal CSF leakage from the ulcerated lesion had begun 8 years before her presentation and had been managed with daily wound dressing and intermittent oral antibiotic treatment. The patient sought medical attention 4 months after suffering a fall that led to focal tenderness and copious CSF drainage; it was unclear whether pain was related to muscular strain or irritation from CSF leakage. At admission, the MMC measured 10×10 cm and was noted to be primarily epithelialized with small ulcerations and CSF leak in multiple areas (Fig. 1).

Since birth, the patient's clinical condition had been characterized by lower extremity weakness, limited ambulation

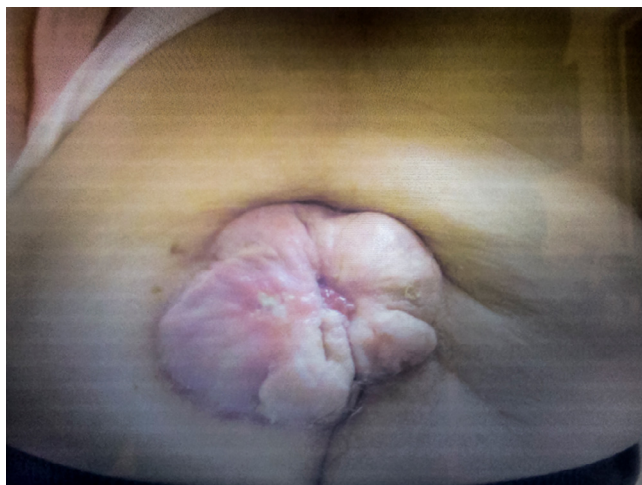


Fig. 1. Photograph demonstrating MMC measuring 10×10 cm. There were several areas noted to be ulcerating, with CSF leaking from multiple areas along the defect.

with use of a front-wheeled walker, as well as bowel and overflow incontinence due to urinary retention. On examination, the patient demonstrated reduced distal lower extremity strength bilaterally: dorsiflexion (3/5), plantarflexion (3/5), and extensor hallucis longus (3/5). Sensation of the ankles and feet was diminished, with normal reflexes and no hyperreflexia. Her general health was otherwise unremarkable and unchanged after her fall. She had no previous history or treatment of severe meningitis requiring hospitalization, and during the last 8 years she had experienced minimal CSF leakage.

Radiographic studies revealed a large complex MMC with associated tethered spinal cord and posterior defect involving nearly the entire sacrum (Fig. 2). Computed tomography imaging demonstrated a Chiari II malformation but no ventriculomegaly.

Initial operative intervention

Because of the persistent CSF leakage after her fall, surgery was pursued. Surgical treatment consisted of primary closure of the MMC and debulking of the surrounding dysplastic scar tissue. The subperiosteal dissection was carried down to the level of the lamina at the L5, identifying the normal anatomy surrounding the defect. The epithelial tissue planes were carefully dissected free from the scar tissue and adjoining placode (Fig. 3A and B). The subdural space was dissected and the tethered structures were released. The dura was inadequate to fully cover the placode; therefore, a 4×6-cm AlloDerm patch (Lifecell, Branchburg, New Jersey, USA) was used to achieve a watertight closure. Fig. 4 illustrates the layers of closure of an MMC repair and neural placode.

Postoperative course and secondary operative intervention

Postoperatively, the patient's neurologic condition was unchanged, but she developed leakage of CSF from her wound on postoperative day 5 as well as increased ventricular size on cranial imaging. On postoperative day eight, an external ventricular drain was inserted, but subsequent attempts to wean the drain were unsuccessful. Thirteen days after the initial repair, the patient underwent a second surgical procedure for wound revision and placement of lumbar drain because of wound dehiscence and continued CSF leakage. Before the patient's discharge, the drain was converted to a slit valve lumboperitoneal shunt system.

After shunt placement, the patient experienced complete resolution of back pain without additional episodes of CSF leakage. She complained of recurrent occipital headaches that were responsive to over-the-counter medications. Eleven months after surgery, her Oswestry Disability Index score was 26%, and magnetic resonance imaging showed an intact repair without evidence of pseudomeningocele (Fig. 5). There was no change in lower extremity neurologic function, and she was free of headaches with a well-healed wound 2 years after surgery.

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