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Postnatal Management and Outcomes of Hydrocephalus in Patients with Myelomeningocele: a Retrospective Study from a Single Institution

Miyelomeningoselli Hastalarda Hidrosefalinin Doğum Sonrası Yönetimi ve Sonuçları: Tek Bir Kurumdan Retrospektif Bir Çalışma

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ABSTRACT

Background: This study aims to analyze retrospectively the incidence and management of hydrocephalus in patients with myelomeningocele (MMC) treated at the Gülhane Training and Research Hospital between 2017 and 2022. The study focuses on patients diagnosed with both MMC and hydrocephalus, either prenatally or postnatally.

Materials and Methods: We retrospectively reviewed the medical records of 30 patients diagnosed with MMC and hydrocephalus. Variables such as gender, birth weight, localization of the MMC sac, timing of MMC repair surgery, and the onset and management of hydrocephalus were documented. Exclusion criteria included patients operated on alone for either MMC or hydrocephalus, patients with diastematomyelia, myelocele, or lipomyelomeningocele, as well as cases with incomplete records.

Results: Of the 30 patients included in the study, 13 (43.3%) were male, and 17 (56.7%) were female. Hydrocephalus was detected at birth in 10 patients (33.3%), while it developed postoperatively in 20 patients (66.7%). MMC lesions were observed in the thoracic region (4 patients, 13.3%); the lumbar region (6 patients, 20%); the sacral region (5 patients, 16.7%); the thoracolumbar region (6 patients, 20%); and the lumbosacral region (9 patients, 30%). Ventriculoperitoneal shunt (VPS) placement was performed in 12 (60%) of the 20 patients who developed hydrocephalus postoperatively; while endoscopic third ventriculostomy (ETV) was performed in 8 patients (40%). Among the 10 patients who presented with hydrocephalus at birth, VPS was performed after initial placement of an Ommaya reservoir.

Conclusion: Hydrocephalus remains a significant and challenging complication in MMC patients, often requiring surgical intervention. The timing and approach to the treatment of hydrocephalus, particularly in the context of MMC, demand careful consideration to minimize the risk of infection and surgical complications. Our findings suggest that while ETV combined with VPS can be effective, close monitoring and timely intervention are crucial to managing hydrocephalus in these patients.

Keywords: Hydrocephalus, myelomeningocele, ETV, Ommaya reservoir

ÖZ

Amaç: Bu çalışma, myelomeningosel (MMC) hastalarında hidrosefalinin insidansını ve yönetimini retrospektif olarak analiz etmeyi amaçlamaktadır. Çalışma, 2017 ve 2023 yılları arasında Gülhane Eğitim ve Araştırma Hastanesi'nde tedavi edilen MMC tanılı hastaları kapsamaktadır. Çalışma, hem MMC hem de hidrosefalisi olan hastalara odaklanmaktadır ve bu hastaların hidrosefalisi ya doğuştan ya da doğum sonrası gelişmiştir.

Gereç ve Yöntemler: MMC tanısı konmuş ve hidrosefalisi olan 30 hastanın tıbbi kayıtları retrospektif olarak incelenmiştir. Cinsiyet, doğum ağırlığı, MMC'nin yeri ve hidrosefalinin yönetimi gibi değişkenler analiz edilmiştir. Çalışmaya dahil edilen hastalar arasında diğer nöral tüp defekti türleriyle (ensefalosel, lipomiyelomeningosel, myelosel, lipomyelosel) ameliyat edilenler yer almamaktadır.

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öz

Bulgular: Çalışmaya dahil edilen 30 hastanın 13'ü (%43,3) erkek, 17'si (%56,7) kız idi. Hidrosefali doğumda 10 hastada (%33,3) saptanırken, 20 hastada (%66,7) doğumdan sonra gelişti. MMC lezyonları, torasik bölgede 4 hastada (%13,3), lomber bölgede 6 hastada (%20), sakral bölgede 5 hastada (%16,7), torakolomber bölgede 6 hastada (%20) ve lumbosakral bölgede 9 hastada (%30) gözlendi. Ventriküloperitoneal şant (VPS) yerleştirme işlemi, hidrosefalisi olan 20 hastanın 12'sinde (%60) uygulandı. Endoskopik üçüncü ventrikülostomi (ETV) 8 hastada (%40) uygulandı. Hidrosefali ile başvuran ve ameliyat edilen 10 hasta arasında, VPS yerleştirilmeden önce Ommaya rezervuarı takıldı.

Sonuç: Hidrosefali, MMC'de önemli ve zorlu bir komplikasyon olarak kalmaya devam etmektedir ve çoğu zaman cerrahi müdahale gerektirmektedir. MMC bağlamında tedaviye yaklaşımda zamanlama ve yöntem, enfeksiyon riskini ve cerrahi komplikasyonları minimize etmek için büyük önem taşır. Bulgularımız, ETV ve VPS'nin her ikisinin de etkili olduğunu, ancak yakından izleme ve zamanında müdahalenin tedavi başarısı için kritik olduğunu göstermektedir.

Anahtar Kelimeler: Hidrosefali, miyelomeningosel, ETV, Ommaya rezervuarı

Introduction

Cranial defects, encompassing a spectrum of congenital anomalies known as neural tube defects (NTDs) that include open or closed spinal dysraphism, are among the most common severe central nervous system anomalies and rank second only to cardiovascular anomalies in causing congenital morbidity and mortality (1). Myelomeningocele (MMC), which occurs due to the incomplete closure of the spinal neural tube during the first month of embryonic development, is the most severe form of spinal dysraphism and is characterized by the protrusion of the spinal cord and meninges through a defect in the vertebral column (2). The incidence of MMC varies between 0.2 and 2 per 1000 live births, and depending on the affected vertebral region, it can result in devastating clinical manifestations such as Chiari II malformation, genitourinary dysfunction, sensory and motor deficits, and hydrocephalus. Therefore, if not diagnosed early or left untreated, the prognosis generally worsens (3). Historically, the relationship between MMC and hydrocephalus was first suspected by Morgagni as early as 1769, with the hypothesis that excess fluid was responsible for both the spinal cyst and hydrocephalus (4). The exact pathophysiology of hydrocephalus associated with MMC remains unclear; however, one theory suggests that inappropriate intrauterine cerebrospinal fluid (CSF) leakage through the MMC defect leads to the underdevelopment of normal CSF drainage pathways, while another theory posits that meningeal irritation caused by amniotic fluid during the intrauterine period contributes to this condition (5,6). These theories provided the scientific basis for early animal studies and pilot human studies evaluating prenatal MMC closure, with the hope that early defect closure would lead to milder neurological deficits and reduced postnatal hydrocephalus (7).

Symptomatic hydrocephalus usually occurs in the first month after MMC repair, and more than half of the patients require surgical treatment (8). Initially, a CSF shunt was the only treatment for MMC-associated hydrocephalus, but nowadays endoscopic third ventriculostomy (ETV) and choroid plexus cauterization (CPC) may be preferred (9,10). Additionally, there is still no consensus in the literature regarding the timing of treatment for these patients; some are treated for hydrocephalus simultaneously with the initial closure, while others are monitored post-closure to determine whether they will require intervention for hydrocephalus (11).

Although managing hydrocephalus associated with MMC is challenging, if patients are well-managed, particularly with the prevention of infections, a favorable cognitive outcome can be achieved in these children. In this study, we retrospectively reviewed the postnatal management of patients with hydrocephalus associated with MMC who were operated on in our clinic.

Materials and Methods

Ethical approval of the project was obtained from the Gülhane Faculty of Medicine Clinical Research Ethics Committee (approval number: 2023-159, dated: 05.07.2023). Patient consent was not required as the study was retrospective, based on computer records, and did not include figures showing the patient's face. In this study, patients with hydrocephalus who underwent surgery between 2017 and 2022 were retrospectively analyzed, with a specific focus on those with hydrocephalus associated with MMC. Patients included in the study were those who had intrauterine detection of MMC and hydrocephalus and were born alive, those who presented with both MMC and hydrocephalus postnatally, and those who developed hydrocephalus during follow-up after MMC surgery, and required treatment. Patients who were operated on solely for MMC or hydrocephalus, those with diastematomyelia, myelocele, and lipomyelomeningocele, and those whose medical records were incomplete, were excluded from the study. A total of 30 patients met these criteria and were included in the study.

The patients' files were retrospectively reviewed. The recorded data included gender, birth weight, location of the MMC sac, timing of MMC repair surgery, timing of the first detection and treatment of hydrocephalus, type of treatment, and the need for and timing of revision surgery in patients treated for hydrocephalus.

Statistical Analysis

IBM SPSS Statistics software version 29.0.2.0 (20) (IBM, SPSS, Chicago, Illinois, USA) was used for the statistical analysis of the data for this study. Categorized variables were reported using descriptive statistics as the number of patients (n) and percentage (%). The distribution properties of numerical variables were evaluated with the Shapiro-Wilk test. The homogeneity of the variances was analyzed by Levene's test. Differences at the $p<0.05$ level were considered statistically significant.

Results

Of the 30 patients who met the inclusion criteria, 13 (43.3%) were male, and 17 (56.7%) were female. The average birth weight of the patients was 3305 grams. MMC surgery was performed on 15 patients (50%) on the first day after birth, on 10 patients (33.3%) on the second day, on 2 patients (6.7%) on the third day, on 2 patients (6.7%) on the fourth day, and on 1 patient (3.3%) in the first month due to late presentation by the family (Table 1).

The MMC lesion was located in the thoracic region in 4 patients (13.3%), the lumbar region in 6 patients (20%), the sacral region in 5 patients (16.7%), the thoracolumbar region in 6 patients (20%), and the lumbosacral region in 9 patients (30%) (Figure 1 and Table 2).

Of the 30 patients included in the study, 20 (66.7%) initially underwent MMC surgery and were monitored for hydrocephalus. Among the 20 patients who developed hydrocephalus during follow-up, ventriculoperitoneal shunt (VPS) placement was performed in 12 patients (60%) between one week and five months after MMC surgery. ETV was performed in 8 patients (40%) with hydrocephalus, and simultaneous VPS placement was performed in 6 of these patients, which is 75%. An Ommaya reservoir were placed in 2 patients (25%) who weighed less than 3000 grams, and VPS placement surgery was performed once they reached an appropriate weight within 2 months. VPS dysfunction

Table 1. Demographic and surgical timing data

Variable	Number of patients (n=30)	Percentage (%)
Gender		
Male	13	43.3%
Female	17	56.7%
Average birth weight	3305 grams	
Timing of MMC surgery		
First day	15	50%
Second day	10	33.3%
Third day	2	6.7%
Fourth day	2	6.7%
Within 1 month (late presentation)	1	3.3%

MMC: Myelomeningocele

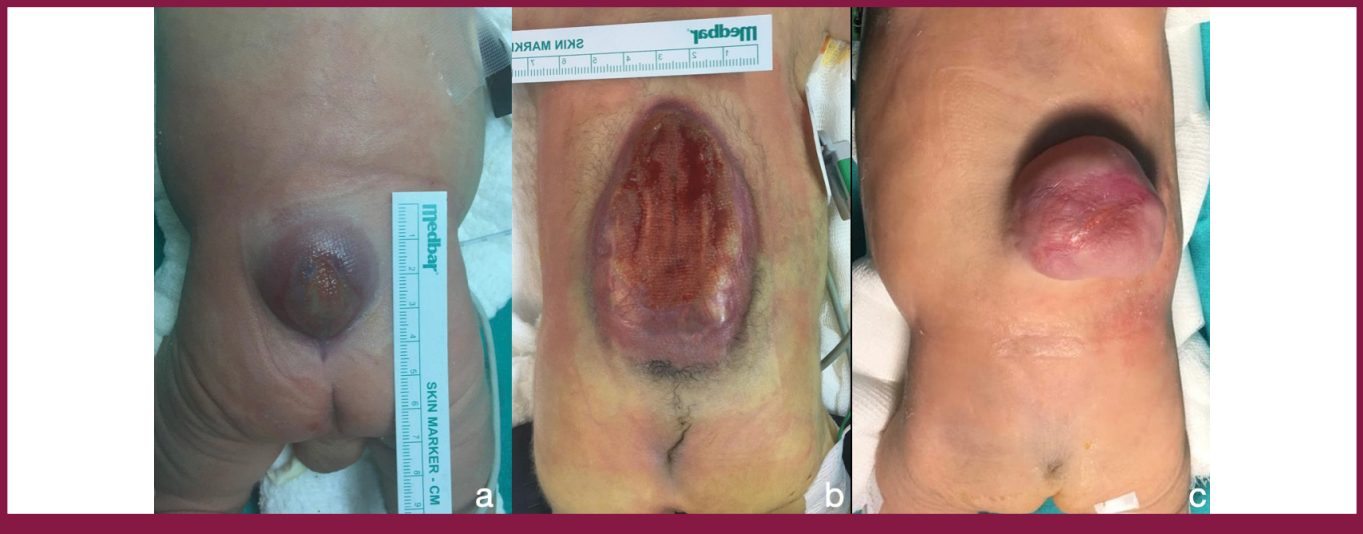

Figure 1. Preoperative images of (a) sacral, (b) thoracolumbar, and (c) thoracic myemomeningocele patients

requiring revision occurred in 17 patients (85%) between 3 and 18 months postoperatively. Three patients (15%) did not need revision surgery (Table 3).

Hydrocephalus was observed at birth in 10 patients alongside MMC. An Ommaya reservoir was placed simultaneously with MMC closure surgery in 6 of these 10 patients (60%), and a VPS was placed once they reached an appropriate weight within 2 months. In 4 patients (40%), ETV was performed simultaneously with MMC closure surgery, followed by the placement of an Ommaya reservoir, and VPS placement was performed within 1 month. VPS dysfunction requiring revision occurred in 8 patients (80%), between 1 and 6 months during follow-up. Two patients (20%) did not need revision surgery (Table 3).

Table 2. Location of MMC lesion

Location	Number of patients (n=30)	Percentage (%)
Thoracic region	4	13.3%
Lumbar region	6	20%
Sacral region	5	16.7%
Thoracolumbar region	6	20%
Lumbosacral region	9	30%

MMC: Myelomeningocele

Table 3. Hydrocephalus management and outcomes

Variable	Number of patients	Percentage (%)
Hydrocephalus following MMC surgery (n=20)		
VPS placement	12	60%
ETV	8	40%
Simultaneous VPS placement	6	75%
• Ommaya reservoir placement	2	25%
• VPS dysfunction (requiring revision)	17	85%
No revision required	3	15%
Hydrocephalus present at birth (n=10)		
Ommaya reservoir placement simultaneous with MMC closure	6	60%
ETV	4	40%
VPS dysfunction (requiring revision)	8	80%
No revision required	2	20%

MMC: Myelomeningocele, VPS: Ventriculoperitoneal shunt, ETV: Endoscopic third ventriculostomy

Discussion

Hydrocephalus is a clinical and neuroradiographic diagnosis characterized by abnormal CSF accumulation within the brain ventricles, leading to ventricular enlargement, and is often associated with increased intracranial pressure (12). Hydrocephalus complicates 35-91% of MMCs, and while it may be evident at birth, it can also develop following MMC repair (13). Among the 30 MMC patients included in our study, 10 (33.3%) had evident hydrocephalus at birth, while 20 (66.7%) developed hydrocephalus after MMC repair (Table 3).

It has been observed that the likelihood of developing hydrocephalus requiring treatment in patients with MMC varied according to the anatomical level of the lesion, and hydrocephalus developed in 60.7% in sacral lesions, 82.4% in lumbar lesions, and 92.2% in thoracic lesions (14). Among the MMC patients requiring surgery for hydrocephalus in our study, the lesion levels were distributed as follows: thoracic in 4 patients (13.3%), lumbar in 6 patients (20%), sacral in 5 patients (16.7%), thoracolumbar in 6 patients (20%), and lumbosacral in 9 patients (30%) (Table 2).

The primary principle of MMC surgical repair is to identify and carefully handle neural tissues to minimize the risk of damage to remaining functional neural tissue. A secondary principle involves preserving and reconstructing the covering structures to re-establish CSF circulation and prevent CSF leakage. Lastly, correct timing of surgical repair and the prevention of perioperative complications ensure proper MMC management (15). Surgery within the first 48 hours of life is crucial to reduce the risk of infection (e.g., reducing the incidence of ventriculitis from 37% to 7% if surgery is performed within 48 hours) and to decrease the likelihood of neurological deterioration due to dehydration or stretching of the placode (16). In our center, we aim to perform MMC repair within 24 hours of birth for all newborns with MMC. However, surgeries were delayed for patients referred from other hospitals. On the first day, in 15 patients (50%), MMC surgery was performed; on the first day in 15 patients (50%), on the second day, in 10 patients (33.3%); on the second day in 10 patients (33.3%), on the third day in 2 patients (6.7%), on the fourth day, in 2 patients (6.7%); on the fourth day in 2 patients (6.7%), and, in 1 patient (3.3%), surgery was performed at 1 month due to delayed referral by the family (Table 1).

Several factors contribute to hydrocephalus in MMC patients. McLone and Knepper (5) reported that permanent CSF leakage due to NTDs disrupts the development of the brain and CSF pathways, leading to crowding of the posterior fossa, which results in downward displacement of the brain stem and hydrocephalus (17).

Prenatal diagnosis of hydrocephalus in MMC can be achieved using ultrasound and magnetic resonance imaging (MRI). High-resolution fetal ultrasound is a non-invasive, sensitive, affordable, and widely available examination, but it is dependent on the observer. Fetal MRI is more accurate and non-invasive but is expensive and more difficult to access (18). In our study, of the 10 patients with concurrent MMC and hydrocephalus at birth, the condition was detected by fetal ultrasound during obstetric follow-ups in 7 patients. The families were informed about the process.

In addition to ultrasound and MRI, computed tomography (CT) can also be used in the postnatal diagnosis of hydrocephalus. Ultrasonography, which has the advantages of being cost-effective and non-invasive, is one of the most commonly used methods for the diagnosis of hydrocephalus in newborns (Figure 2a). CT remains one of the most sensitive and widely used modalities to confirm hydrocephalus and aid in treatment planning, although there are increasing concerns about the radiation effects of CT on the developing brain, particularly regarding cognitive development and tumor induction (Figure 2b) (19). In our clinical practice, patients undergoing surgery for MMC are closely monitored for head circumference and fontanel tension, and in cases of suspicious increases, they are evaluated for hydrocephalus with ultrasonography and MRI (Figure 2c). In cases of acute neurological deterioration, urgent CT imaging is performed.

The two main accepted procedures for hydrocephalus treatment in MMC patients are CSF shunt placement and ETV (20,21). Although VPS surgery is the gold standard and most commonly used CSF shunt procedure in hydrocephalus treatment, other alternatives include ventriculopleural and ventriculoatrial shunts (22). In our routine clinical practice, VPS surgery is the first choice, provided there are no contraindications.

While many authors believe that placement of VPS at the same time as MMC surgery, increases the risk of shunt infection, others believe that there is no association, which shows that there is still controversy about the timing of surgery (23-26). MMC may increase the risk of shunt infection by causing CSF contamination through the defect (27,28). Therefore, it is recommended that VPS placement be performed meticulously, with particular attention to preventing infection. At our center, we adopt a cautious approach to VPS placement while closely monitoring for hydrocephalus development. Ventricular tapping, external ventricular drainage, and Ommaya reservoir placement are preferred treatment alternatives until VPS surgery, especially if there is evidence or suspicion of MMC repair breakdown or infection. Of the 30 patients included in this study, we initially performed MMC repair surgery without prophylactic hydrocephalus surgery in 20 (66.7%) patients who did not have evident hydrocephalus at birth, while closely monitoring them for hydrocephalus. In the 10 (33.3%) patients with hydrocephalus at birth, we opted for simultaneous placement of an Ommaya reservoir during MMC surgery and delayed VPS surgery.

In recent years, ETV has become an important treatment option, especially for obstructive hydrocephalus because of its lower risk of infection and the fact that it does not rely on extracranial mechanical drainage (29). Although the success rate of ETV in patients with MMC-associated hydrocephalus is known to be as low as 35%, the success rate is 76%, in combination with CPC (30). We prefer not to perform ETV in newborns with hydrocephalus associated with MMC due to the low success rate in this age group. In this study, among the 10 patients diagnosed with MMC and hydrocephalus at birth and operated on during the neonatal period, 6 received an Ommaya reservoir, while 4 underwent ETV along with the placement of an Ommaya reservoir. Of the 20 patients

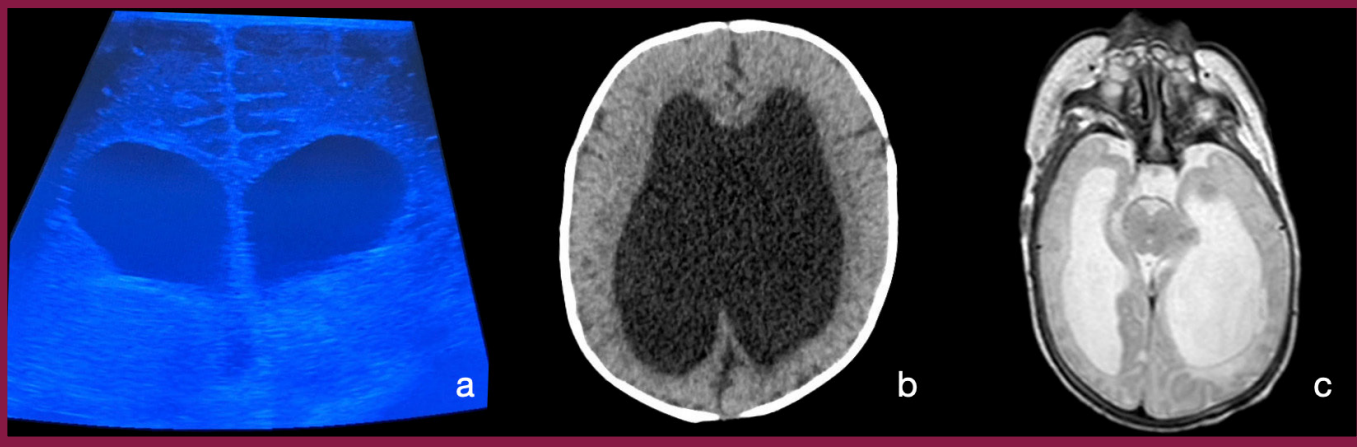

Figure 2. (a) Ultrasonography, (b) computed tomography and (c) magnetic resonance imaging in the diagnosis of hydrocephalus in patients with myelomeningocele

who developed hydrocephalus during follow-up, 8 (40%) underwent ETV, but 6 of these also received concurrent VPS placement, and 2 received an Ommaya reservoir. None of our patients underwent the CPC procedure.

In these patients, it is very important to remain vigilant regardless of the treatment modality, which makes close follow-up necessary. Initially, a follow-up every three months is recommended. Once patients have stabilized, annual follow-up is preferred.

Conclusion

This study provides valuable insights into the management of hydrocephalus in patients with MMC and highlights the importance of individualized treatment strategies. Our findings indicate that while the incidence of hydrocephalus remains significant, the timing and choice of surgical interventions, particularly the use of VPS and ETV, play a critical role in patient outcomes. The decision to delay VPS placement in favor of vigilant monitoring and the selective use of Ommaya reservoirs or ETV has been shown to be effective in reducing the risk of infection and other complications associated with premature shunt insertion.

Furthermore, the data underscore the need for careful consideration of anatomical factors, such as an abnormal base of the third ventricle and the presence of communicating hydrocephalus, when determining the suitability of ETV in neonates. The success rate of ETV in neonates with MMC remains limited, and alternative or adjunctive approaches are required.

The study also highlights the necessity for close postoperative follow-up to promptly detect and address potential shunt failures or other complications, thereby ensuring better long-term neurological outcomes. As the field of pediatric neurosurgery continues to evolve, our experience suggests that a cautious, tailored approach to the management of MMC-associated hydrocephalus, informed by ongoing clinical assessment and the latest surgical techniques, will likely yield the best results for this vulnerable patient population.

Future research should focus on further refining these treatment protocols, exploring the potential of newer technologies and minimally invasive procedures, and developing long-term follow-up strategies to enhance the quality of life for patients with MMC and hydrocephalus.

Ethics

Ethics Committee Approval: Ethical approval of the project was obtained from the Gülhane Faculty of Medicine Clinical Research Ethics Committee (approval number: 2023-159, dated: 05.07.2023).

Informed Consent: Retrospective study.

Footnotes

Authorship Contributions

Surgical and Medical Practices: G.K., D.E.K., M.C.E., Concept: M.B.K., D.E.K., Design: G.K., M.B.K., D.E.K., M.C.E., Data Collection or Processing: G.K., M.C.E., Analysis or Interpretation: M.B.K., D.E.K., Literature Search: G.K., M.C.E., Writing: G.K., D.E.K., M.C.E.

Conflict of Interest: No conflict of interest was declared by the authors.

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