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제1형 아놀드 키아리 기형을 가진 33세 남자 환자에서 소 심장막 유래 인공뇌경막을 이용한 두개성형술 후 발생한 호산구성 수막염

Eosinophilic Meningitis after Bovine Graft Duraplasty for Arnold-Chiari Malformation Type 1 in a 33-year-old Man

이환태 · 박필환 · 서일혜 · 김경희 · 서자영 · 정지훈 · 김문진 · 안정열
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Cases of pediatric eosinophilic meningitis following duraplasty with a bovine graft have been reported. These patients recovered following the surgical removal of the dural graft or steroid therapy. Decompression for Chiari malformation is a common procedure in both pediatric and adult neurosurgery. We describe the case of a 33-yr-old male patient with eosinophilic meningitis following Chiari decompression via bovine graft duraplasty. Cerebrospinal fluid (CSF) study showed 49 red blood cells/μL and 129 leukocytes/μL with 17% eosinophils. There was no evidence of infectious disease. To our knowledge, this is the first report of adult eosinophilic meningitis after bovine graft duraplasty in Korea.

Key Words: Eosinophilic meningitis, Bovine graft, Arnold-Chiari malformation

INTRODUCTION

Eosinophilic meningitis is defined as the presence of more than 10 eosinophils/μL in the cerebrospinal fluid (CSF) and/or eosinophils accounting for more than 10 percent of CSF leukocytes. Parasitic infection is one of the most common causes of eosinophilic meningitis, but other infectious conditions, neoplastic diseases, and foreign bodies are also associated [1]. Collagenous dural patches derived from bovine tissues are routinely used for dural closure in cranial and spinal surgery because they provide a watertight dural closure and should have low antigenicity [2-4]. Pediatric cases of eosinophilic meningitis following duraplasty with a bovine graft have been previously published [3, 5-10]. Here, we describe an adult patient with eosinophilic meningitis following Chiari decompression surgery via bovine graft duraplasty.

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CASE

The 33-yr-old man presented with 1-yr history of having difficulty walking, and MRI showed a type 1 Arnold-Chiari malformation. He underwent decompression with suboccipital craniectomy, and duraplasty was performed using a Lyoplant® matrix (B.Braun Aesculap, Tuttlingen, Germany). One month later, he complained of fever, headache, and dizziness. CSF analysis following admission revealed 49 red blood cells/μL and 129 leukocytes/μL with 17% eosinophils, 73% lymphocytes, and 10% monocytes without malignant cells (Fig. 1A). His CSF protein level was

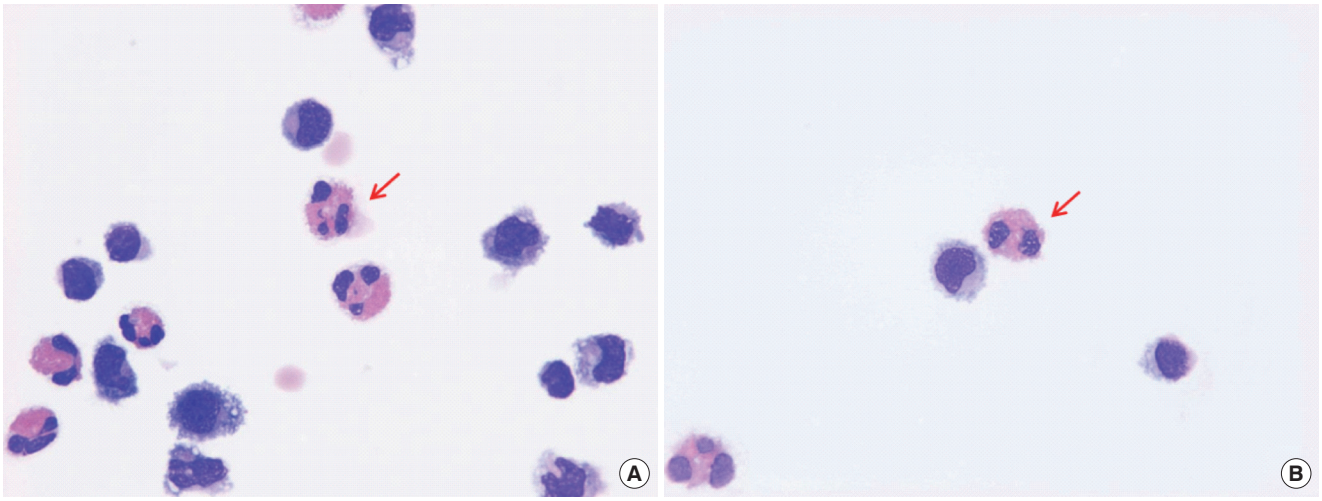

Fig. 1. Wright-Giemsa-stained cytospin smear (×1,000) of cerebrospinal fluid demonstrated increased eosinophils (arrow) at diagnosis (A) and follow up after 2 months (B).

83.8 mg/dL (reference range, 8-32 mg/dL), and his glucose level was 17 mg/dL. His hsCRP level was 0.38 mg/dL (reference range, 0-0.5 mg/dL) and his procalcitonin level was less than 0.05 ng/mL (reference range, <0.05 ng/mL). The complete blood count (CBC) results were as follows: hemoglobin, 13.1 g/dL; white blood cell (WBC) 3.92×10^9/L (differential count: segment neutrophil, 63.3%; lymphocyte, 24.5%; monocyte 9.9%; eosinophil, 2%; basophil, 0.3%), and platelets 248×10^9/L. Although evaluations for infectious disease including bacterial and fungal were negative for blood, CSF, and sputum, vancomycin and cefepime were started empirically. Brain computed tomography (CT) revealed fluid collection in the suboccipital region, and he underwent repair surgery for CSF leaking. The Lyoplant® matrix was not removed. Intraoperative CSF culture and post-operative drain culture did not show evidence of infection. He was regularly administered only antibiotics without intravenous or oral steroids after the repair surgery. Two months after admission, repeat CSF analysis revealed 41 red blood cells/μL and 52 leukocytes/μL with 16% eosinophils, 5% segment neutrophil, 36% lymphocytes, 41% monocytes, and 2% macrophages (Fig. 1B). His CSF protein level was 83.3 mg/dL, and his glucose level was 23 mg/dL. There was no evidence of infection on CSF culture, but his WBC count was 3.14×10^9/L (differential count: segment neutrophil, 54.1%; lymphocyte, 25.2%; monocyte, 17.5%; eosinophil, 2.9%; basophil, 0.3%). At his 6-month follow-up, the gait disturbance and operative wound were improved, and he was without fever. His WBC count was 2.40×10^9/L.

DISCUSSION

To the best of our knowledge, this is the first report of adult eosinophilic meningitis after bovine graft duraplasty in Korea. Although several pediatric cases of eosinophilic meningitis following duraplasty with a bovine graft have been reported [3, 5-10], the case of noninfectious eosinophilic meningitis in adult patients have been associated with malignancies such as Hodgkin's disease or eosinophilic leukemia, medications including vancomycin and ibuprofen, or ventriculoperitoneal shunts [11-14]. The cases of pediatric eosinophilic meningitis following a bovine tissue dural graft were successfully treated after the surgical removal of the dural graft [5]. Another patient suffered an intense allergic reaction including a rash, swelling, intermittent fever, and eosinophilia to the bovine dural graft, and she recovered after replacement of the dural graft. Her explanted dural graft showed evidence of eosinophilc and chronic lymphocytic infiltration on pathological examination [3]. There have been recent reports of patients with eosinophlic meningitis following duraplasty with a bovine dural graft responding to intravenous or oral steroid treatment [6, 9, 10]. Ostendorft and Connolly reported a case of 7-yr-old girl who was diagnosed with eosinophilic meningitis following bovine graft duraplasty [9]. Eosinophlic meningitis did not improve after initially administering cefepime, vancomycin, and doxycycline. However, she was symptom free after starting intravenous dexamethasone.

Our patient had no evidence of infectious or neoplastic disease, and he was not taking any medication previously associated with

eosinophilic meningitis. He underwent decompression with Lyoplant® matrix 1 month prior to the onset of symptoms. We believe that a reaction to the Lyoplant® matrix was the most likely cause of the eosinophilic meningitis, although the possibility that the Lyoplant® matrix actually caused eosinophilic meningitis cannot be ruled out.

Lyoplant® is an acellular and avascular bovine derived extracellular matrix and is manufactured with a controlled lyophilization process. The avascular characteristic prevents hyperacute rejections. This xenogeneic extracellular matrix graft activates a T-helper cell type 2 immunogenic response, which is consistent with a remodeling reaction rather than rejection [2]. However, eosinophilic meningitis after bovine dural graft placement has been reported in pediatric patients. The cause of eosinophilic meningitis is not entirely clear.

Eosinophilc meningitis after bovine graft duraplasty for Arnold-Chiari malformation has not been reported in adults, although decompression for Chiari malformations is commonly performed in both pediatric and adult neurosurgery [15]. Perhaps differences in the immune response between pediatric and adult patients are related, and further studies are needed. Based on this case, we also suggest that careful review of the medical history and evaluations for infectious disease are necessary to prevent use of improper antibiotics and surgical management in adult eosinophilic meningitis patients.

요 약

소 심장막 유래 인공뇌경막을 이용한 두개성형술을 받은 소아 환자에서 호산구성 수막염이 발생한 보고가 있었으며, 소아 환자들의 경우 이식된 인공뇌경막을 제거하거나 스테로이드 치료를 통해서 증상이 회복되었다. 소아와 성인 아놀드 키아리 기형 환자 모두에서 감압두개술은 흔하게 시행되며, 본 환자는 33세 성인으로 소 심장막 유래 인공뇌경막을 이용한 키아리 감압술을 받은 후 호산구성 수막염이 발생하였다. 환자의 뇌척수액 검사에서 적혈구 49개/μL, 백혈구 129개/μL였고, 이 중 호산구가 17%였다. 감염성 질환의 증거는 없었다. 이는 한국에서 소 심장막 유래 인공뇌경막을 이용한 두개성형술을 받은 성인에서 호산구성 수막염을 보인 첫 번째 사례이다.

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