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Foramen Magnum Decompression with Adipose Tissue Grafting for Caudal Occipital Malformation Syndrome in a Maltese Dog

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Abstract : A 7-year-old intact female Maltese dog was referred to the hospital with a history of paresis in the hind limbs, left head turn, and a loss of balance that persisted for 2 weeks. Her condition was initially managed with steroids, prescribed by the referring veterinarian, but her neurological symptoms were not alleviated. Physical and neurological examinations, radiography, computed tomography, and magnetic resonance imaging were performed. Based on the findings on these examinations, caudal occipital malformation syndrome (COMS) with syringohydromyelia was diagnosed. Medical treatment was not effective in the previous trial; therefore, foramen magnum decompression, durotomy, and free autogenous adipose tissue grafting were performed. After 3 days, an improvement was observed in the clinical symptoms and was maintained for 8 months postoperatively. Based on the results, it is suggested that the decompression method with a fat graft may be considered an effective surgical treatment for the management of COMS that did not respond well to previous medical treatment.

Key words **:** caudal occipital malformation syndrome, foramen magnum decompression, fat graft, dog.

Introduction

Caudal occipital malformation syndrome (COMS), which is analogous to Chiari Type I malformation in humans, has been recognized in small breed dogs and commonly affects Cavalier King Charles Spaniels (CKCS) (4,10,12). The cause of this disorder is unknown, but is considered a developmental disorder of the occipital bone mesoderm (16). Comparable to Chiari type 1 malformation in humans, dogs with COMS may have a variety of neurologic manifestations, including myelopathy (generally cervical), central vestibular dysfunction, and seizures (4,8,15,16). The treatment method of choice for humans with Chiari type 1 malformation is the surgical decompression of the foramen magnum (16); however, 8-30% of cases require reoperation. Similar to humans, the recurrence rate of foramen magnum decompression (FMD) in cases of canine COMS was 47% (4). To reduce the relapse rate, various techniques have been studied, for example, FMD with a titanium-mesh cranioplasty (5). According to the report, the use of this technique demonstrated the absence of recurrence in 21 cases, with a 1-year follow-up period (5).

In this case, the occipital bone was too fragile to perform a titanium-mesh cranioplasty. Therefore, the application of durotomy with a free adipose tissue autograft implanted over the durotomy (9) was performed as an alternative method of surgical treatment. The aim of this case report is to describe the radiographic findings and clinical prognosis in a dog treated surgically using FMD, durotomy, and adipose tissue graft.

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Case

A 7-year-old, intact female Maltese weighting 2.5 kg was referred to the veterinary medical teaching hospital at the Gyeongsang national university animal medical center for evaluation of hind limb paresis that persisted for 2 weeks. The owner reported that the dog had received steroids for 10 days but her clinical condition did not improve.

Upon presentation, the dog was unwilling to walk and was supporting its weight with abducted forelimbs. Observations made during the physical examination included both paresis and the absence of conscious proprioception in the hind limbs. Hopping and tactile placement of the hind limbs were absent on the left side and decreased on the right. The head was turned to the left side and a loss of balance was observed during the neurological examination. According to the owner, paresis of both hind limbs had recently worsened. Based on the results of physical and neurological examination, the cerebellovestibular system was localized.

The complete blood count, biochemical assay, and radiography showed no significant findings and were within the reference ranges. Brain and spinal CT and MRI were performed under general anesthesia. The MRI revealed dilation of the ventricular system, compression of the cerebellum, and protrusion of the cerebellum through the foramen magnum (Fig 1A). The midsagittal MRI of the cervical spinal cord

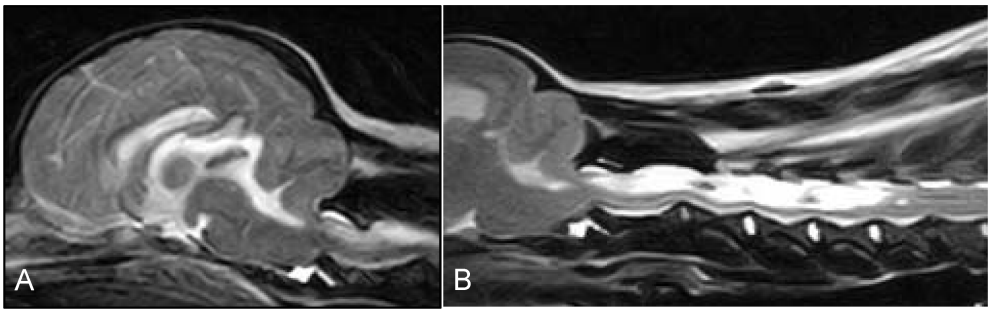

Fig 1. (A) A T2W-sagittal magnetic resonance (MR) image showing herniation of cerebellum, ventriculomegaly, and rostral displacement of the cerebellum (B) T2W-sagittal MR image showing SM of the spinal cord.

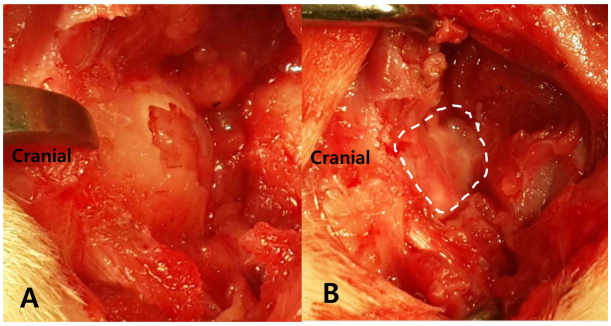

Fig 2. (A) FMD defect on the occipital bone and C1 (B) Satisfactory positioning of free adipose tissue before the wound was closed.

revealed a syrinx formation in C2-5 (Fig 1B). Based on previous tests and the MRI findings, the clinical signs were attributed to COMS and subsequent SM. Medical treatment was not effective in the previous trial, surgical treatment was recommended.

The patient received mannitol (0.5 g/kg, IV, over 15 minutes), cefazolin (25 mg/kg, IV), and methyl prednisolone sodium succinate (MPSS) (30 mg/kg, IV) preoperatively and was premedicated with atropine (0.04 mg/kg, SC), butorphanol (0.2 mg/kg, SC) and diazepam (0.2 mg/kg, IV). Anesthesia was induced with propofol (6 mg/kg, IV) and maintained with isoflurane inhalation anesthesia with an oxygen flow rate of 2 L/min. The patient was placed in sternal recumbency, with the neck ventroflexed. The surgical site was clipped from the bregma to the level of the C3-4, with a width equal to that of the atlas. A towel was placed under the neck to extend the surgical site. The skin was incised from the occipital protuberance cranially to the level of C3 caudally to expose the median raphe and underlying biventer cervicis muscles. The paired biventer cervicis muscles were divided along the midline by using blunt dissection, exposing the rectus capitis dorsalis muscles. The rectus capitis dorsalis muscles were dissected to expose the caudal portion of the occiput, arch of the atlas, and dorsal lamina of the C1. A high-speed air drill and Lempert rongeur were used to remove a portion of the occiput and dorsal aspect of C1 (Fig 2A). After creating the FMD defect, the durotomy was performed with a 23-G syringe needle to further decompression of the cerebellum and to aid the cerebrospinal fluid (CSF) flow.

In order to reduce scar tissue formation and the recurrence rate, the cranioplasty procedure using titanium mesh and polymethylmethacrylate was prepared. However, in this case, the occipital bone was too fragile to perform screw insertion; instead, a free adipose tissue autograft (obtained from the neck subcutaneous fat) was implanted over the site that FMD was performed (Fig 2B). The wound was closed in routine fashion.

Butorphanol (0.2 mg/kg SC), cefazolin (25 mg/kg, IV), famotidine (0.5 mg/kg, SC) and MPSS (30 mg/kg, IV) were administered postoperatively. A soft, padded cervical bandage was applied to prevent excessive cervical movement after the surgery. The patient received prednisolone (0.5 mg/kg, PO), tramadol (5 mg/kg, PO), misoprostol (5 μg/kg, PO), famotidine (0.5 mg/kg, PO), and cefadroxil (25 mg/kg, PO) for 14 days.

The patient was able to stand without any support after 1 day postoperatively. After 3 days the patient was ambulatory. Although her neurological signs such as head turn and the lack of balance and ambulation improved, the symptoms were not completely diminished. A reevaluation was performed monthly thereafter to examine ambulation, mentation, and the surgical site. After 8 months, there was no observation of recurrence or deterioration. The owner reported that the gait was well maintained was satisfied with the results of surgery.

Discussion

COMS is a developmental abnormality, which results in overcrowding of the caudal fossa and compression of the cervicomedullary junction (2,4). The pathogenesis of COMS and SM are not fully understood (12). A major factor is an inadequately small caudal fossa volume that is caused by a relatively short occipital bone. However, studies comparing intracranial dimension did not demonstrate a difference between the size of the cranial fossa in CKCSs with or without COMS (3).

COMS is predominantly found in small breed dogs; the disorder is overwhelmingly represented in CKCSs (2,14). A total of 92% of CKCSs have at least one craniocervical morphologic abnormality while 50% have concurrent SM (3). To date the syndrome has been reported in breeds such as the CKCS, Brussels Griffon, Yorkshire Terrier, Maltese Terrier, Chihuahua, Miniature Dachshund, Miniature Toy Poodle, Bichon Frise, Pug, Shih Tzu, Pomeranian, Staffordshire Bull

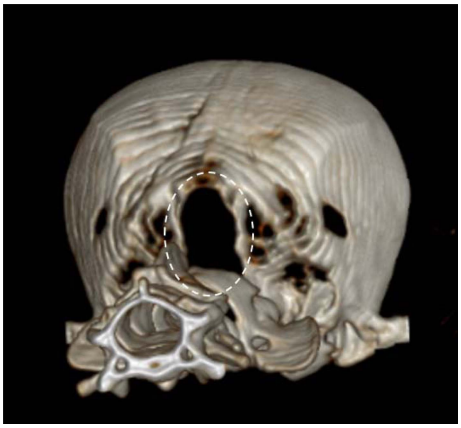

Fig 3. Three-dimensional reconstruction image of the surgically decompressed region using the computed tomography images. The FMD defect in the occiput was identified.

Terrier, Boston Terrier, French Bulldog, Pekingese, and Miniature Pinscher (16). Diverse neurological presentations are observed in these dogs. In addition, most COMS cases have a multitude of clinical presentations, reflecting the multifocal nature of the disease (5,8,11,16).

Clinical features of COMS and SM are extremely variable, but the most important and consistent clinical presentation is pain (8); however, this may be difficult to localize because it is often intermittent. Other clinical signs include cerebellovestibular dysfunction, cervical myelopathy, scratching activity, seizure, scoliosis, and facial nerve paralysis (1,2,12). In this report, in addition to hind limb paresis and head turn to the left side, the dog showed cerebellovestibular dysfunction such as imbalance, wide-based stance, falling to the left side, and abnormal postural reaction.

MRI is the most useful method of diagnosing SM. MRI findings in humans with Chiari type 1 malformation involve obliteration or attenuation of the subarachnoid space at the level of the foramen magnum, cerebellar tonsillar herniation, anterior displacement of the cerebellum, and "kinked" image of the medulla (7,17). Similar to the disease in humans, MRI findings in 30 dogs with COMS included attenuation or obliteration of the dorsal subarachnoid space at the cervicomedullary junction (all dogs); SM was observed in 26 (87%) cases. Cerebellar herniation was noted in 18 (60%) cases. A kinked image of the caudal medulla was observed in 12 (40%) dogs (4). In this case, all of the aforementioned MRI abnormalities were observed. At 3 months, postoperative MRI images showed that these abnormalities were not resolved; however, the surgically decompressed site was well maintained (Fig 3).

The reported medical treatment for dogs with COMS generally consists of three categories: analgesic medication, drugs that reduce CSF production, and anti-inflammatory doses of oral glucocorticoids (e.g., prednisone [0.5 mg/kg q12h]) (1,13). Medical treatment will initiate a favorable response, but clinical signs of dysfunction are not resolved. In one study of 10 dogs with COMS that were treated medically, 5 dogs were euthanized within 2 years. In another study, 36% of COMS dogs were euthanized at a mean of 1.7 years from the day of diagnosis (1,4). In this case, medical therapy was administered for 10 days, but an improvement of clinical symptoms was not observed. Therefore, surgical intervention was recommended.

Reported surgical techniques for COMS include FMD, FMD with titanium mesh/PMMA cranioplasty, FMD with graft, and FMD with duroplasty. The surgical success rate of FMD in dogs with COMS is about 80%; the relapse rate is approximately 25% to 47% (8,11). According to a study on FMD with titanium/PMMA cranioplasty, the short-term success rate is 81% and the recurrence rate is 0%-8% (11). Additionally, a recent analysis of more than 100 cases involving this surgical technique determined the re-operative rate to be 7% (6). A surgical technique that performed FMD with duroplasty using lyophilized swine sub-mucosa and the placement of a free autogenous adipose tissue graft has been studied. Of all the patients that underwent this procedure, none required additional surgery; 94% (16/17) showed improvement after surgery with no observation of deterioration beyond their pre-surgical status (9). In this case, the occipital bone was too fragile to perform FMD with titanium/PMMA cranioplasty. Therefore, FMD with a free adipose tissue autograft implanted over the durotomy was performed. This surgical technique was tolerated well in the patient and resulted in sustained clinical improvement.

Further studies and follow-up evaluations are needed before this method is determined to be an effective alternative to other surgical methods. Small breed dogs, like the patient in this study, may benefit from cranioplasty and duroplasty using titanium/PMMA and lyophilized swine sub-mucosa, respectively. This case report describes the successful treatment of COMS and SM with FMD, durotomy, and adipose tissue graft.

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References

1. Bagley RS, Gavin PR, Silver GM, Moore MP, Kippenes H, Connors R. Syringomyelia and hydromyelia in dogs and cats. Comp Cont Educ Pract 2000; 22: 471-479.
2. Cagle L. Concurrent occipital hypoplasia, occipital dysplasia, syringohydromyelia, and hydrocephalus in a Yorkshire terrier. Can Vet J 2010; 51: 904.
3. Carruthers H, Rusbridge C, Dubé MP, Holmes M, Jeffery N. Association between cervical and intracranial dimensions and syringomyelia in the cavalier King Charles spaniel. J Small Anim Pract 2009; 50: 394-398.
4. Dewey CW, Berg JM, Stefanacci JD, Barone G, Marino DJ. Caudal occipital malformation syndrome in dogs. Comp Cont Educ Pract 2004; 26: 886-896.
5. Dewey CW, Marino DJ, Bailey KS, Loughin CA, Barone G, Bolognese P, Milhorat T, Poppe D. Foramen magnum decompression with cranioplasty for treatment of caudal occipital malformation syndrome in dogs. Vet Surg 2007; 36: 406-415.
6. Fossum TW. Small animal surgery textbook: Elsevier Health Sciences: 2013.

7. Karagöz F, Izgi N, Sencer SK. Morphometric measurements of the cranium in patients with Chiari type I malformation and comparison with the normal population. Acta Neurochir 2002; 144: 165-171.
8. Lu D, Lamb C, Pfeiffer D, Targett M. Neurological signs and results of magnetic resonance imaging in 40 cavalier King Charles spaniels with Chiari type 1-like malformations. Vet Rec 2003; 153: 260-263.
9. Ortinau N, Vitale S, Akin EY, Beasley M, Shores A. Foramen magnum decompression surgery in 23 Chiari-like malformation patients 2007-2010: outcomes and owner survey results. Can Vet J 2015; 56: 288-291.
10. Rusbridge C. Chiari-Like Malformation with Syringomyelia in the Cavalier King Charles Spaniel: Long-Term Outcome After Surgical Management. Vet Surg 2007; 36: 396-405.
11. Rusbridge C. Neurological diseases of the Cavalier King Charles spaniel. J Small Anim Pract 2005; 46: 265-272.
12. Rusbridge C, Greitz D, Iskandar BJ. Syringomyelia: current concepts in pathogenesis, diagnosis, and treatment. J Vet Intern Med 2006; 20: 469-479.
13. Rusbridge C, Jeffery ND. Pathophysiology and treatment of neuropathic pain associated with syringomyelia. Vet J 2008; 175: 164-172.
14. Rusbridge C, Knowler P, Rouleau GA, Minassian BA, Rothuizen J. Inherited occipital hypoplasia/syringomyelia in the cavalier King Charles spaniel: experiences in setting up a worldwide DNA collection. J hered 2005; 96: 745-749.
15. Rusbridge C, Knowler S. Hereditary aspects of occipital bone hypoplasia and syringomyelia (Chiari type I malformation) in cavalier King Charles spaniels. Vet Rec 2003; 153: 107-112.
16. Rusbridge C, MacSweeny J, Davies J, Chandler K, Fitzmaurice S, Dennis R, Cappello R, Wheeler S. Syringohydromyelia in cavalier King Charles spaniels. J Am Anim Hosp Assoc 2000; 36: 34-41.
17. Strayer A. Chiari I malformation: clinical presentation and management. J Neurosci Nurs 2001; 33: 90-104.