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Surgical outcomes of six bulldogs with spinal lumbosacral meningomyelocele or meningocele

This is the final peer-reviewed author's accepted manuscript (postprint) of the following publication:

Published Version:
Martin Muniz, L., Del Magno, S., Gandini, G., Pisoni, L., Menchetti, M., Foglia, A., et al. (2020). Surgical outcomes of six bulldogs with spinal lumbosacral meningomyelocele or meningocele. VETERINARY SURGERY, 49(1), 200-206 [10.1111/vsu.13342].

Availability:
This version is available at: https://hdl.handle.net/11585/733297 since: 2020-02-24

Published:

DOI: http://doi.org/10.1111/vsu.13342

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13 September 2026

Surgical treatment of six meningo(myelo)celes

SURGICAL OUTCOME IN SIX BULLDOGS WITH SPINAL LUMBOSACRAL MENINGOMYELOCELE OR MENINGOCELE.

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ABSTRACT

Objectives: To report the surgical treatment and outcome in 6 bulldogs with spina bifida (SB) and meningocele (MC) or meningomyelocele (MMC).

Study design: Case series.

Animals: Six client-owned dogs (5 French bulldogs and 1 English bulldog) with MC or MMC.

Methods: The surgical treatment and outcome of spinal MC or MMC diagnosed by magnetic resonance imaging in dogs at two institutions between 2013 and 2016 were retrospectively reviewed. Surgical treatment included dissection of the meningeal sac to the vertebral column defect. In dogs with MMC, nerves were repositioned and protruded meninges removed, prior to suturing excised meninges.

Results: Two dogs were diagnosed with MC and 4 with MMC. A lumbosacral dimple was noted in all dogs, along with neurological deficits most commonly consisting of urinary and fecal incontinence (n=6) and mild/moderate paraparesis (n=3). Dorsal laminectomy was performed in all dogs. Resection of adhesions and filum terminale was performed in 2 dogs with suspected tethered cord syndrome (TCS). Urinary and fecal incontinence improved in 2 cases and remained unchanged in four. Paraparesis improved in 2 dogs.

Conclusions: Surgical treatment resulted in partial improvement of the urinary and fecal incontinence (2/6 dogs) and paraparesis (2/3 dogs) or stable neurological condition (3/6 dogs) with only minor temporary complications.

Clinical significance: In the absence of published data comparing surgical and conservative treatment of puppies affected by SB and MC or MMC, early surgical treatment can be considered in order to prevent future deterioration of neurological signs and, eventually, facilitate improvement of the neurological condition.

INTRODUCTION

Meningocele (MC) and meningomyelocele (MMC) are rare and probably underestimated congenital neural tube malformations,[1] which are responsible for various degrees of neurological deficits in dogs.[2,3] The associated spina bifida (SB) is characterized by incomplete dorsal fusion of the vertebral arches, classified as open (*aperta*) or closed (*occulta*) in the case of communication or not with the external enviroment.[1] MC defines a protrusion of the meninges associated with an accumulation of cerebrospinal fluid (CSF) outside the vertebral canal through the bone defect.[1-4] MMC differs from MC because the meningeal protrusion includes nervous tissue.[3,5-9] Tethered cord syndrome (TCS), a rare condition in dogs, is characterized by an abnormal caudal traction of the conus medullaris, potentially associated with SB, MC, and MMC,[6,8,10] usually causing progressive neurologic deterioration.[6,8,10]

In contrast to companion animals, the human literature defines MMC as an open lesion, characterized by leakage of CSF and exposure of the neural tissue to the environment. MMC is frequently associated with other serious central nervous system (CNS) anomalies, such as Chiari type II malformation and hydrocephalus.[11] In human medicine, surgical treatment is clearly advised during fetal life for spina bifida aperta[12-14] and recommended as soon as possible in the case of spina bifida occulta with clinical signs, especially in the case of TCS.[11] The outcome regarding urinary continence is variable; however, ambulatory function and mental status improve with early surgery.[15]

The etiology is likely multifactorial, including genetic mutations, breed predisposition,[16,17] exposure to drugs that interfere with mitosis,[18,19] and nutritional deficiencies.[20] Manx cats

70 are genetically predisposed to these spinal disorders,[17] while an inherited etiology was
71 supposed in English bulldogs.[16]
72 In dogs, clinical presentation of SB and MMC or MC depends on the severity and location
73 of the malformation. The most frequently affected site is the lumbosacral area and the
74 clinical signs reflect the involvement of the caudal lumbosacral intumescence or adjacent
75 nerve roots. The most common signs include fecal and urinary incontinence,[3,5-7]
76 reduced/absent anal tone and perineal sensation, mild flexor paresis of the hind limbs,[3,5,6,21]
77 and dimpling of the skin.[3] The clinical suspicion is usually confirmed by magnetic
78 resonance imaging (MRI).[3]
79 Few case reports are present in the veterinary literature, and no guidelines are available
80 about conservative or surgical management of symptomatic dogs with MC or MMC not
81 communicating with the environment.[3] Surgical treatment has been suggested, especially
82 in cases of communication of the MC/MMC with the external environment or in cases of
83 TCS with a variable outcome, ranging from an unchanged neurological condition to
84 (rarely) return to normal function.[3,5,6,11,21,22] To the best of the authors' knowledge, only 2
85 dogs with spina bifida occulta that were surgically treated have been reported in the
86 literature,[6,17] and another surgical treatment of a dog with myelomeningocele and dermoid
87 sinus has been described.[18]
88 The aim of the present study was to increase the information available in the veterinary
89 literature on closed SB and MC or MMC in dogs, considering the hypothesis that dogs
90 treated surgically at a young age could improve their neurological condition after surgical
91 treatment. For this reason, the study reports retrospectively the clinical signs, surgical

92 treatment, outcome, and long-term follow-up results in a case series of dogs affected by
93 closed SB and MC or MMC.

MATERIALS AND METHODS

Medical records (2013–2016) of dogs with SB and MMC or MC surgically treated from 2 different establishments were identified retrospectively. The owners of the dogs were informed about the risks of surgery and the outcomes reported in the literature, including the lack of information about conservative therapy. They all chose surgery in an attempt to help to improve the continence and gait function of their dogs and to prevent the anticipate progression of the neurological signs. Dogs less than one year of age were included in the study if they had a complete physical and neurological examination, MRI of the lumbosacral spine, surgical treatment, and outcome information.

Plain radiographs of the lumbosacral spine were performed, including latero–lateral and ventro–dorsal views. MRI of the lumbosacral spine was performed using a 0.2T permanent magnet (Esaote Vet-MR unit, Esaote Biomedica, Genova, Italy) or a 0.22T MRI unit (Mr J 2200, Paramed, Italy). In all patients, MRI examination included sagittal, dorsal, and transverse T1- and T2-weighted images. Follow-up information was collected by re-examining the dogs at 2 weeks and 2 years after surgery or by telephone interviews with the owners or referring veterinarians at the same time period.

Anesthesia, analgesia, and perioperative period

Intraoperative analgesia, consisting of continuous intravenous infusion (CRI) of a cocktail of morphine, lidocaine, and ketamine (MLK)[23] was also maintained for approximately 24 hours after surgery to ensure good pain control. Intra- and postoperatively, a broad-spectrum antibiotic therapy was administered (cephalexin 30 mg/kg twice daily, intravenously or orally), and gastric protection (omeprazole 0.7 mg/kg once daily, orally) was maintained for 8–10 days after surgery.

Postoperative analgesia was adapted to each patient. After the first 24 hours on MLK CRI, opioids (methadone and/or buprenorphine) were administered for 2 to 3 days and then lowered with tramadol for 5 days. Owners were advised to restrict dogs to a crate for 4 weeks after surgery.

Surgical management

Surgical treatment consisted of the correction of MC or MMC. Prior to surgery, the hair was shaved, followed by aseptic skin preparation over the lumbosacral area, from the third or fourth lumbar vertebra to the tail. The dogs were positioned in sternal recumbency with their pelvic limbs cranially placed. The surgery was performed as previously described.[3,5,22] Briefly, blunt and sharp dissection until the opening of the lamina was performed to isolate the protrusion of the meninges (Figure 1). A laminectomy was performed to improve visualization of the anomalous protrusion of the meninges. With the help of magnification (ocular loops 2.5 x), durotomy and consequent opening of the meningeal sac were performed in all cases with iris scissors or scalpel, until CSF flowed out. After placement of stay sutures, *cauda equina* nerve roots and *filum terminale* were identified in cases of MMC, and meningeal adhesions were broken down to allow careful repositioning of the neural tissue. The excessive meninges were removed (Figure 2) and the dural defect sutured with absorbable or non-absorbable suture material (Monosyn 6/0, Braun, Aesculap AG, Germany, and Prolene 6-0, Ethicon, Johnson & Johnson, USA) in a simple continuous or interrupted pattern to restore the linearity of the dural sac (Figure 3). Standard closure of epaxial muscles, subcutaneous tissue, and skin was performed.

Complications were classified as major or minor, and intraoperative or postoperative. Major complications were defined as those life-threatening circumstances requiring urgent

surgical or medical management. Minor complications were defined as self-limiting or medically managed conditions.[3]

RESULTS

Six dogs fulfilled the inclusion criteria and were included in the study. Affected breeds included 5 French bulldogs (two from the same litter) and one English bulldog (Table 1).

History and clinical signs

All cases were referred for fecal and urinary incontinence since birth. Gait abnormalities were observed in 3 dogs (dogs 2, 3, and 4) (Table 1).

On general physical examination, an inflamed perianal region was observed in 3 dogs. Localized skin depression was confirmed on palpation of the lumbosacral region in all dogs (Figure 4). In dog 2, left quadriceps contracture and ipsilateral hip luxation were noted. The rest of the physical examination was within normal limits.

Neurological examination was consistent with a lesion affecting the S1–S3 spinal cord segments in 3 cases (dogs 1, 5, and 6), and L6–S3 in the other 3 (dogs 2, 3, and 4) (Table 1). Decreased or absent perineal sensation and reflex were observed in all dogs. Based on the age, history, and clinical and neurological signs, congenital anomalies including SB associated with MMC or MC in the lumbosacral region were suspected.

Preoperative evaluation

Routine blood works were within normal limits. Plain radiographs of the lumbosacral region showed the incomplete dorsal lamina and abnormal spinous process in all patients at the level of the sixth or seventh lumbar vertebra.

MRI findings consisted of a lack of fusion of the dorsal lamina (in L7 or L6 according to the dog) and absence of the spinous process (Figure 5). Moreover, a dorsal displacement of the meninges and subarachnoid space was observed through the bony defect extending dorsally or caudo-dorsally to the level of the subcutaneous tissue in all dogs. MC was

diagnosed in dogs 1 and 2, whereas in dogs 3, 4, 5, and 6 MRI confirmed a MMC. A midline depression in the skin corresponding to the area of the defect was present. Concomitant mild syringomyelia and subarachnoid diverticulum were observed in cases 3 and 2, respectively (Table 1). TCS was suspected in dogs 3 and 4 due to the middle dorsal displacement of the conus medullaris (Figure 5A, 5B).

Surgery

In all cases, a dorsal approach was used to detach the meningeal protrusion from the surrounding tissues. After the incision of the meningeal sac, in dogs 3, 4, 5, and 6 some nerve roots were dorsally displaced outside the vertebral canal, inside the protruded meninges. In MMC, after careful detachment of adhesions between the nerve roots and the meninges, difficulties were encountered to arrange the redundant nerves in their normal anatomical position. They tended to regain the previous position within the defect (dogs 2 and 3). In 2 cases (3 and 4), a dorsally displaced and tight *conus medullaris* was observed and tethered spinal cord was suspected, and, according to the literature,[3,6,10] resection of the *filum terminale* and adhesion resolution were performed. No intraoperative complications were recorded.

Follow-up

Minor postoperative complications were recorded in 4 out of the 6 dogs. In the immediate postoperative period, dogs 2 and 3 showed temporary worsening of paraparesis, recovering at the pre-surgical condition within 24 hours. Dog 3 exhibited moderate swelling of the wound that did not require treatment. Dog 5 showed lameness in the left hind limb during the first 3 days after surgery, which spontaneously improved. In dogs 5 and 6, diarrhea was observed during the first three days, spontaneously resolving without specific treatment.

Dogs were discharged from the hospital between 3 and 13 days after surgery. Gabapentin (10 mg/kg every eight hours, PO) was used in dogs 3 and 5. At 15 days after surgery, gait improvement was observed in dog 3, and complete continence was obtained in dog 6.

In the long-term follow-up, 3 dogs (3, 4, and 6) showed improvement compared with their preoperative status, while the other 3 dogs (1, 2, and 5) presented an unchanged neurological condition (Table 1). In the postoperative period, although an increase of the anal tone was detectable in all cases, only dog 6 was urinary and fecal continent 2 weeks after surgery, and dog 3 presented fecal and urinary incontinence selectively during physical exercise. All the other dogs remained incontinent at the last control after surgical intervention (for dogs still alive, at 2 years after surgery).

DISCUSSION

The present case series provides some information on the early surgical management of dogs affected by spina bifida occulta and MC or MMC. The long-term follow-up only partially support the hypothesis that dogs affected by MC or MMC treated surgically at a young age could improve their neurological condition after surgical treatment.

The clinical signs recorded in our case series reflected the most frequently reported signs in the literature, including fecal and urinary incontinence and gait abnormalities, depending on the area of the spinal cord involved.[3,5,6,22,24] The presence of a dimple in the lumbo-sacral region was a constant sign in the dogs included in the study. This external characteristic can be easily found, and together with a radiograph, it can help breeders and first-opinion practitioners to quickly identify possibly affected puppies.

After surgery, on the long-term follow-up, the clinical signs remained unchanged in 3 dogs, partial resolution of the neurological abnormalities was observed in 2 dogs, and complete continence was noticed in only 1 dog. Unfortunately, this latter dog was followed only until 1 month after surgery, when he died after parvovirus infection.

In the dogs included in the study, MC did not show different clinical signs or a better outcome after surgery in comparison to MMC. The 2 dogs affected by MC remained stable after surgery, while 3 out of the 4 dogs with MMC improved after surgery, and the remaining dog maintained the pre-surgical neurological status. In addition, 2 dogs with MMC presented a suspected TCS, and both dogs improved in continence and/or gait after surgery. These results suggest that surgical release of displaced nerve roots and adhesions could be potentially beneficial in affected dogs. However, the low number of cases does not permit the drawing of definitive conclusions, and further studies are warranted. The 2

MC-affected dogs had other spinal anomalies, which could have contributed to the lack of significant improvement (Table 1). In our case series, syringomyelia and arachnoid diverticulum were found in 2 dogs, but other associated neural anomalies could have been missed because the MRI, according to the neurological localization, was performed only on the lumbosacral region.

The lack of improvement could be explained by malformations of the *cauda equina* itself,[9] myelodisplasia,[6] or the acquired damage of the nerve roots during chronic displacement/traction or during surgery. Indeed, the abnormal position that the nerve roots tend to maintain after detachment of adhesions in some dogs[21] may support the hypothesis of severe chronic changes. Other causes include the inability of the surgery to regain a normal anatomy in the lumbosacral region or retethering of the spinal cord, as reported in humans and probably due to scar tissue formation.[25] Unfortunately, a control MRI, useful for confirming or excluding the above-mentioned hypotheses, was not performed in any dog included in the study.

In our case series, the rationale behind the early surgical treatment was to try to restore a normal anatomy of the meninges and the *cauda equina*, eliminating abnormal CSF accumulation and, in case, to prevent possible further deterioration of the nervous tissue. With the same aim, the adhesions between the meninges and nerves, when present, were also carefully detached, and, in the case of suspected TCS, the *filum terminale* was resected to release the nervous tissue from abnormal tension. Unlike Shamir et al.,[6] who reported the use of artificial dura for closing the meningeal defect created by excising the protruded meninges in dogs, in the present study the primary dural closure was considered satisfactory in all cases.

In dogs it is unknown whether clinical signs linked to MC, MMC and concurrent anomalies[3] will progress with conservative management and treatment recommendations are extrapolated from the human literature.[11,12] The surgical outcomes previously described for MC and MMC in dogs are limited to a few cases in the literature focusing on surgical treatment of spina bifida aperta[3,5] or with concomitant anomalies like dermoid sinus[26] or TCS.[6,8] Comparison between surgical and conservative treatment is lacking in the veterinary literature. Unfortunately, due to the retrospective nature of this study, no comparison with dogs treated conservatively was available, preventing the acquisition of useful data. Only one successful treatment, with complete remission of urinary and fecal incontinence and gait abnormalities, was reported in a seven-week-old Yorkshire Terrier with a diagnosis of closed MMC.[21] Other case reports have documented no regain of urinary continence and improvement of the mild gait abnormalities after surgical treatment.[5,6,22]

The present case series confirms the variable success of surgery in improving the clinical signs, especially urinary incontinence. It is worth noting that none of the dogs showed worsening of the neurological condition in the long-term follow-up.

As for humans, the time of surgery is claimed to potentially play an important role in terms of enhanced neurological improvement as in dogs.[3] Unfortunately, this statement is not demonstrated in the veterinary literature, and further studies are necessary. In human medicine, early diagnosis and treatment of MMC can be performed using sequential ultrasonographic evaluation during fetal life.[13,14] In the case of spina bifida occulta, treatment is suggested as soon as possible in the case of neurological signs.[11] In dogs, the intrauterine approach is not currently available, and only post-natal advanced imaging

techniques can support the diagnosis and, consequently, the treatment.[3,8] In our case series, the dog showing the worse neurological condition was treated at 2 months of age and had a remarkable improvement from a non-ambulatory paraparesis and complete urinary and fecal incontinence to ambulatory paraparesis and incontinence only during vigorous physical activity.

In our population, French bulldogs accounted for 83% of the dogs, and two of them were from the same litter. The overrepresentation of French bulldogs in our case series reinforces the suspicion of an inherited etiology, as already observed in Manx cats,[17] or the presence of a breed predisposition as in English bulldogs.[16]

The present study has several limitations mainly related to its retrospective nature, which prevented more objective monitoring of outcome. Limitations include the low number of cases, due to the low incidence of the disease; the lack of control MRI; and the lack of a control group of dogs treated conservatively. The authors decided not to use an objective scale to measure gait abnormalities, mainly consisting in flexor muscles weakness, because the lumbosacral localization prevented efficient use of the published scales for thoracolumbar spinal disorders.[26]

In conclusion, the present study showed that the early surgical management of dogs affected by spina bifida occulta and MC or MMC in puppies did not produce any major complication or deterioration of the neurological condition in the long term. On the contrary, a stable or improved clinical condition was observed. In the absence of clear guidelines on the management of this disease, early surgery could be considered as a treatment option. Information about these congenital anomalies should be promoted to

290 first-line practitioners and breeders, especially of bulldogs, to allow early diagnosis and
291 future studies. Investigations comparing the medical and surgical outcome in dogs with
292 MC and MMC are warranted to detail the effective value of surgical intervention and
293 provide precise treatment guidelines.

Disclosure

The authors declare no conflict of interest related to this report.

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Figure legends

Figure 1: Dog 3: Dissected meningomyelocele (asterisk) protrusion anchored by a stay suture from the bifid arch of L6 and L7 (cranial part of the patient corresponds with the left side of the picture).

Figure 2: Dog 2: After resection of the meningocele, CSF and neural tissue (arrowhead) are visible through the resected meninges (arrow), anchored by stay sutures.

Figure 3: Dog 1: Intraoperative image of a meningocele. Dural sac after durotomy and closure by simple suture pattern (arrowhead).

Figure 4: Dorsal view of lumbosacral area in dog 4 (A) and dog 5 (B). The hair on the dorsal midline has an abnormal appearance, and a dimpling of the skin can be noticed (A, B; white arrowheads).

Figures 5 A, B, C, and D: Transverse and sagittal (T2W) views in dog 4 (Figures 5A and 5B) and dog 5 (Figures 5C and 5D). Note the middle dorsal displacement in Figure 5A and 5B (black arrowhead, suspected tethered cord syndrome) compared with Figure 5D (black arrowhead). Displacement of meninges with or without nervous tissue through the bone defect in Figures 5A and 5C (white arrows), respectively.

381 Table 1

382 The data regarding signalment, neurological signs, magnetic resonance imaging, surgery,
383 and outcome are reported for each dog.

384

Signalment	Neurological signs	MRI	Surgery	Outcome
Dog 1: English bulldog, 4 months old, M	Perianal reflex absent, urinary and fecal incontinence	Multiples vertebral malformations from T8 to L1 and SB with MC L7-S1	Resection of MC	Neurologically unchanged after 2 years
Dog 2: French bulldog, 4 months old, M	Paraparesis postural deficits HL, flexor reflexes decreased HL, perianal reflex absent, urinary and fecal incontinence	SB in L6-L7 with accompanying MC in L7 Arachnoid diverticulum in L6-L7	Resection of MC	Neurologically unchanged after 2 years
Dog 3: French bulldog, 2 months old, F	Severe non-ambulatory paraparesis, spontaneous proprioceptive deficits HL, flexor reflexes decreased HL, perianal reflex absent, urinary and fecal incontinence	SB in L6-L7 with MMC Presence of syringohydromyelia of L5-L6 spinal cord segments Suspected TCS	Resection of MC and resolution of neural tissue adhesions. Filum terminale resection	Improved: able to walk with moderate paraparesis. Fecal and urinary incontinence improved, only during exercise 2 years post-op
Dog 4: French bulldog, 5 months old, M	Paraparesis, bunny hopping, minimal postural deficits in HL, perianal reflex absent, urinary and fecal incontinence	SB in L7-S1 with MMC Suspected TCS	Resection of MC and resolution of neural tissue adhesions. Filum terminale resection	Improved at 8 months post-op. Bunny hopping disappeared. Urinary and fecal incontinence persisted

Dog 5: French bulldog, 4 months old, F	Perianal reflex absent, urinary and fecal incontinence	SB in L7-S1 with MMC	Resection of MC and resolution of neural tissue adhesions	Neurologically stable after 3 months post-op and euthanized.
Dog 6: French bulldog, 4 months old, F	Perianal reflex absent, urinary and fecal incontinence, episodic voluntary urination	SB in L7-L6 with MMC	Resection of MC and resolution of neural tissue adhesions	Improved, complete continence 15 days post-op. Dead at 1 month post-op for parvovirosis infection.

385 SB= spina bifida

386 MC= meningocele

387 MMC= meningomyelocele

388 TCS= tethered cord syndrome

389 HL= hind limbs

390 M= male

391 F= female

392