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NOTE

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Congenital syringohydromyelia in a crossbred (Holstein-Friesian × Japanese Black) beef calf

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ABSTRACT. A 5-day-old male crossbred beef calf presented with a well-coordinated bilateral hopping gait of the hind limbs. Postmortem CT showed a poorly defined oval-shaped region at the L3–L4 spinal segments, which had high signal intensity on T2 weighted postmortem MRI images. On pathological examination, we identified a large cystic cavity filled with a large amount of cerebrospinal fluid on the cut surface of the spinal region. Histopathological examination revealed that the spinal cord parenchyma was compressed by the cystic structure, and the cystic cavity was lined with a thin layer of discrete ependymal cells, indicating syringohydromyelia. This is the first reported case of a Holstein-Friesian × Japanese Black crossbred calf with solitary syringohydromyelia. Our findings suggest that myelodysplasia with cystic cavities can be suspected by CT, without the need for MRI.

KEY WORDS: congenital spinal disease, crossbred calf, myelodysplasia, postmortem examination, syringohydromyelia

Syringohydromyelia is a condition in which the central canal is abnormally dilated and the cavity is partly lined by ependymal cells [1, 19]. In previous reports, myelodysplasia including syringohydromyelia was suspected based on ultrasonography, computed tomography (CT), magnetic resonance imaging (MRI), and autopsy findings in calves, and eventually diagnosed by histopathological inspection [4, 16, 21]. Clinical signs of congenital myelodysplasia in cattle include recumbency, inability to stand, and ataxia, which are usually non-progressive. For rare cases in which animals are capable of walking, they have been reported to exhibit a well-coordinated hopping action, called a "bunny hopping" gait [1–4, 8, 17, 23]. Here we report a case of a solitary syringohydromyelia in a crossbred beef calf with a well-coordinated bilateral hopping gait of the hind limbs, which was suspected by postmortem CT and MRI, and confirmed by pathological examination.

A 5-day-old male crossbred beef calf (Holstein-Friesian × Japanese Black) presented with an abnormal gait at a loose-barn dairy farm in Chiba, Japan. The dam of the calf was vaccinated every year to prevent bovine viral diarrhea virus infection. Delivery of the calf required some assistance by a farmer. The calf was separated from the dam immediately after birth, kept in an individual calf pen for 5 days, and moved to a sawdust-bedded group pen with automated milk feeding systems, where a farmer noticed the calf's hopping gait and inability to return backward from the milk feeding space by itself. On clinical examination, pulse and body temperature were normal (120 beats/min and 39.0°C, respectively), and intestinal movements on auscultation and the appearance of feces were normal. A well-coordinated bilateral hopping gait of the hind limbs was observed (Fig. 1). There was no dislocation found in the hip joint by palpation. The calf was moved to an individual pen again to limit walking motion for a week.

Hematological, neurological, and ultrasonographic examinations were conducted. Dislocation of the hip joint was initially suspected, and ultrasonographic examination of the hip joints was performed using a 9.0 MHz linear transducer (B-mode, CTS-800, SIUI, Guangdong, China) at age 13 days [20]. The calf was sedated during the examination with intravenous xylazine (0.2 mg/kg) administration. The results were normal, excluding the possibility of hip joint dislocation. At age 25 days, comprehensive blood work revealed normal range values for white blood cell count (8.2×10^3 / μl), red blood cell count (7.33×10^6 /μl), hematocrit (25.3%), hemoglobin concentration (9.0 g/dl), protein concentration (6.0 g/dl), albumin concentration (3.4 g/dl), aspartate aminotransferase (45 U/l), lactate dehydrogenase (666 U/l), and creatine phosphokinase (85 U/l), suggesting the absence of inflammatory disease. Results of ultrasonography of the spinal cord from the L6–S1 intervertebral space using a 9.0 MHz linear transducer at age 30 days were

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Fig. 1. Well-coordinated bilateral hopping gait in the patient calf (serial photographs from left to right).

normal. Cranial tibial reflex (reflex center: L6 spinal segment) of the right hind limb was decreased and that of the left was increased; patellar tendon reflex (L4–L6 spinal segments), gastrocnemius reflex (S1 spinal segment), and flexion reflex of the hind limbs (L6–S2 spinal segments) were normal at age 48 days. The well-coordinated bilateral hopping gait of the hind limbs persisted to age 48 days. The calf was euthanized following the owner's request.

Postmortem CT and MRI of the lumbar spinal cord, and pathological examination were performed after euthanasia. CT scan was performed using a 80-row multi-slice CT scanner (Aquilion Prime, Canon Medical Systems Corp., Otawara, Japan) with the following parameters: tube voltage, 120 kV; tube current, 50–300 mA; tube rotation time, 0.5 sec/rotation; slice thickness, 0.5 mm; field of view, 500 mm; and a matrix size, 512 × 512. Digital Imaging and Communications in Medicine (DICOM) data were sent to a viewer (Newton OsiriX, Newton-Graphics, Sapporo, Japan) to measure CT values. An MRI scan was performed using a 3.0 T MR unit (Vantage Galan 3T, Canon Medical Systems Corp.). T2-weighted images (TR=4,000 msec and TE=60 msec) and T1-weighted images (TR=2,900 msec and TE=10 msec) of the lumbar spinal cord were obtained.

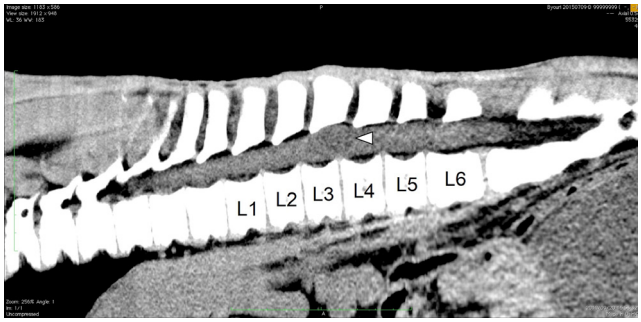

Fig. 2. Two-dimensional reconstruction computed tomographic image of the lumbar spinal cord. The L3–L4 spinal segments show focal enlargement and a poorly defined oval-shaped region with reduced Hounsfield units (arrowhead).

CT showed focal enlargement of the lumbar spinal cord, and a poorly defined oval-shaped region at the L3–L4 spinal segments (Fig. 2). The CT value of the region (15–20 HU; Hounsfield Unit) was reduced compared with that of other spinal regions (30–40 HU). There was no evidence of morphological abnormality in the vertebrae, and the numbers of cervical, thoracic, lumbar, and sacral vertebrae were normal on CT scans. MRI revealed a cavity filled with fluid in the cord at the L3–L4 spinal segments, which showed T2-weighted hyperintensity and T1-weighted hypointensity (Fig. 3A and 3B). The cranial portion of the cavity was connected with the central canal of the spinal cord.

At necropsy, there were no significant lesions in the vertebrae, the skeletal muscles of the four limbs, cerebrum, and cerebellum. The spinal cord at the L3–L4 lumbar vertebrae was markedly swollen (Fig. 4). On the cut surface of the spinal region, there was a large cystic space filled with a large amount of cerebrospinal fluid. Histopathological examination revealed the spinal cord parenchyma to be compressed by the enlarged cystic structure that was connected to the central canal (Fig. 5A), and the surface of the cystic cavity was lined with a thin layer of discrete ependymal cells (Fig. 5C and 5D). The caudal part of the cavity was disconnected to the central canal (Fig. 5C). These postmortem findings confirmed the diagnosis of solitary syringohydromyelia at the third and fourth spinal cord segments. In visceral organs other than the spinal cord, mild bronchopneumonia was observed.

The present case was a solitary syringohydromyelia of a crossbreed beef calf with a well-coordinated bilateral hopping gait of the hind limbs. Hydromyelia is a condition in which the central canal is abnormally dilated and the cavity is lined by ependymal cells [21]. In contrast, syringomyelia is an abnormal condition in which paracentral cavities located within the neuroparenchyma [21]. In some cases, hydromyelia and syringomyelia may coexist, and the cystic cavity is lined by discrete ependymal cells. It makes difficult to differentiate between the two conditions. Therefore, syringohydromyelia is used as an umbrella term of hydromyelia and syringomyelia [1, 19]. Hydromyelia, syringomyelia, and syringohydromyelia frequently occur together with skeletal deformities [6, 13], arthrogryposis [9], Arnold-Chiari malformations [3], and other myelodysplasias such as diplomyelia, diastematomyelia, hypoplasia, and aplasia [4, 7, 8, 10, 11, 13, 15–17, 21–23]. Hydromyelia, syringomyelia, and syringohydromyelia have previously been reported in Holstein, Japanese Black, Charolais, Brown Swiss, Tudanca, Simmental, and Brahman-crossed calves [1, 2, 4, 7–11, 15–17, 21]. To date, one case of a Brahman-crossed calf and two cases of Holstein calves with solitary hydromyelia or syringohydromyelia have been reported to exhibit a well-coordinated bilateral hopping action [2, 4, 8].

In addition to MRI scans [7, 13, 16, 23], ultrasonographic diagnosis of syringohydromyelia, diplomyelia, and hypoplasia from intervertebral spaces have been reported [21, 22]. We attempted to initially diagnose the calf by ultrasonography prior to CT and MRI scans. However, we failed to make a diagnosis because we examined the L6–S1 intervertebral spaces, rather than the affected L3–L4 spinal segments. MRI scans are the gold standard for diagnosing cerebral and spinal diseases and are reportedly useful

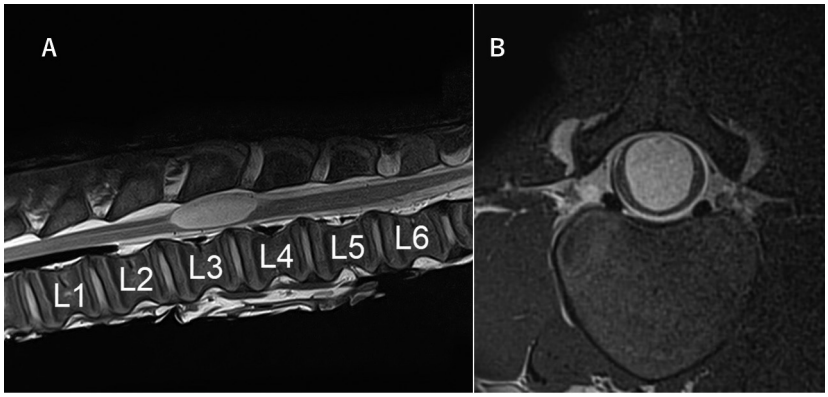

Fig. 3. Magnetic resonance images of the lumbar spinal cord. Sagittal (**A**) and transverse (**B**) T2-weighted images of the cavity in the cord at the L3–L4 spinal segments.

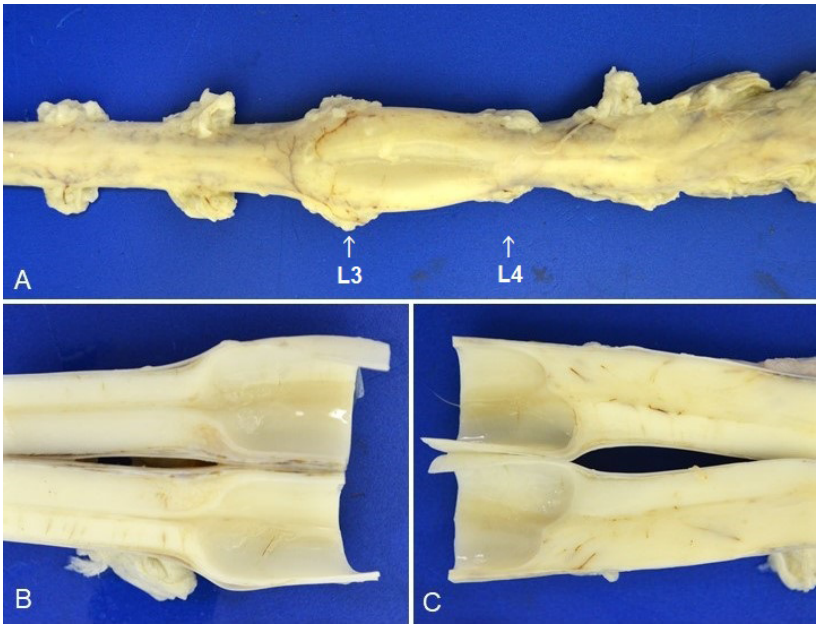

Fig. 4. Macroscopic findings of the spinal cord (**A**). Arrows indicate the L3 and L4 nerves. Sagittal plane of the cranial portion of the cavity (**B**). Sagittal plane of the caudal portion of the cavity (**C**). The spinal cord at the area of the L3–L4 vertebrae is clearly swollen.

for diagnosing syringohydromyelia and diplomyelia while calves are still alive [7, 16, 24]. Accessibility to MRI scans in large animal veterinary practices, however, is more limited than CT scans due to costs associated with MRI scanners and the difficulty and ancillary costs of accommodating patients at facilities equipped with CT or MRI scanners [18]. Since diagnosis of bovine myelodysplasia by CT scans was previously reported but rarely discussed in detail [4, 13], we compared MRI with CT images after the calf was euthanized in the present study. Focal enlargement of the spinal cord, and a poorly defined oval-shaped region with a reduced CT value, which was confirmed to be a distinct cavity on MRI, were observed at the L3–L4 spinal segments on CT. This suggests that myelodysplasia with cystic cavities can be suspected by CT, without the need for MRI.

Generally, a bilateral hopping gait indicates a lower motor neuronal or orthopedic disorder [5]. The affected spinal area may alter spinal nerve reflexes and can be identified by neurological examinations [5]. Clinical signs of spinal cord malformation vary based on the affected area. To date, calves exhibiting a hopping gait with hydromyelia, or syringohydromyelia, have shown involvement of the lumbar or lumbosacral spinal cord [2–4, 8]. Hopping gait, however, has also been reported in calves with diplomyelia, syringomyelia, myelodysplasia, hypoplasia, or aplasia, with involvement of the L1–S2 spinal segments [1, 4, 8, 23]; this is considered to occur due to a malfunction of the central pattern generator network of the pelvic limbs within the lumbar intumescence [4]. The most focally affected area in calves with a hopping gait is the L5 spinal segment [8]. The features of our case with syringohydromyelia in the L3–L4 spinal segments were consistent with those reported in previous studies. Our findings suggest that dysfunction of the L3–L4 spinal segments may also induce a hopping gait, similar to findings from a study by Wunderink *et al.* [23].

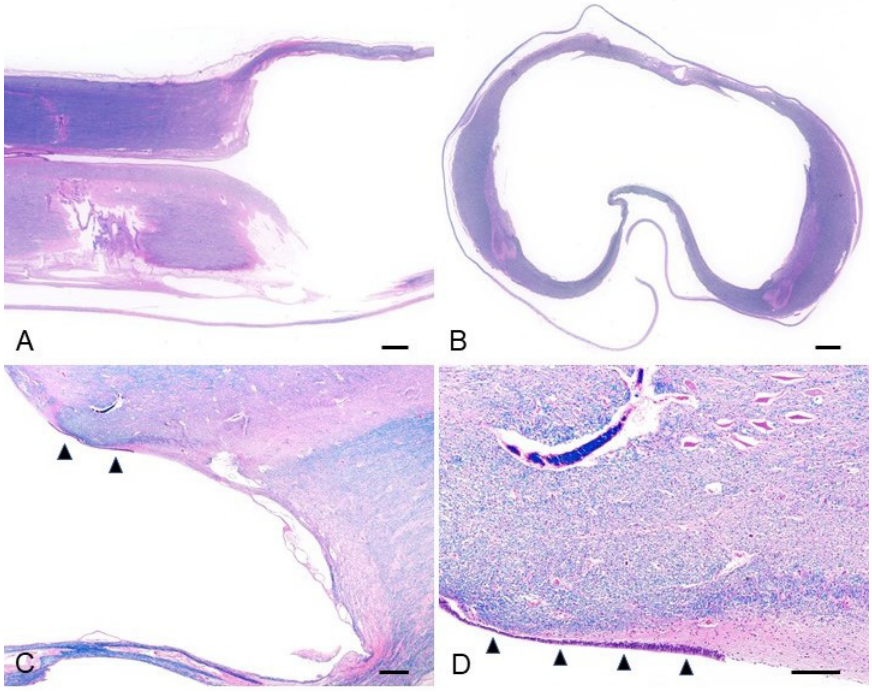

Fig. 5. Histological findings of the spinal cord. Cranial and sagittal plane (**A**). Transverse plane (**B**). Magnified images of caudal (**C**) and sagittal (**D**) planes. The central canal is connected to the cavity (A), the cavity is centrally located and compresses the surrounding parenchyma (B), and some parts of the inner surface are lined by ependymal cells (C and D). The caudal part of the cavity was disconnected to the central canal (C). Arrowheads indicate discrete ependymal cells. Luxol fast blue-hematoxylin-eosin staining (A–D). Bars: 1 mm (A and B), 500 µm (C), and 200 µm (D).

The cause of solitary hydromyelia or syringohydromyelia remains unclear. Since our calf was kept in an individual calf pen after birth for 5 days, we speculate that the gait was not clearly observed until the calf was moved to the wide group pen. Based on this, the bilateral hopping gait observed at age 5 days seemed congenital. The cystic cavity located at the L3–L4 spinal segments was connected at the cranial portion but disconnected at the caudal portion to the central canal; no other abnormalities related to the bilateral hopping gait were discovered from blood tests or histopathological examination. These findings indicate that due to focal dysplasia or obstruction of the central canal at the L4 spinal segment cerebrospinal fluid in the disconnected central canal of the calf gradually compressed the parenchyma before birth and formed the cystic cavity. Myelodysplasia in cattle, including hydromyelia, syringomyelia, and syringohydromyelia, is reported to occur particularly in the lumbar area [3, 4], indicating a common pathogenic mechanism. Recurrent outbreaks of myelodysplasia in Spain are thought to be caused by neuroteratogenic plants [17]. On the other hand, involvement of genetic factors in myelodysplasia has been proposed for Charolais, Hereford, and Holstein calves [12, 14]. The potential cause of vertebral and spinal dysplasia in Holstein calves, which have several lesions in the cervical to coccygeal vertebrae including hydromyelia, syringomyelia, and syringohydromyelia, has been reported to be a missense mutation in *T-box transcription factor T* (*TBXT*) [12, 13]. Further studies will be needed to elucidate the pathogenesis of bovine spinal malformations.

We report the first case of a Holstein-Friesian × Japanese Black crossbred calf with solitary syringohydromyelia. Our findings suggest that myelodysplasia with cystic cavities can be suspected by CT, without the need for MRI. This clinical case adds to our growing knowledge of the relationship between a bilateral hopping gait of the hind limbs and myelodysplasia.

CONFLICT OF INTEREST. The authors declare that there are no conflicts of interest.

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