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CASE REPORT

Companion or pet animals

Clinical and imaging features of a feline lumbar myelopathy resembling ossification of the posterior longitudinal ligament in humans

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Abstract

A 9-year-old, male, neutered Russian blue cat was presented for a 2-week history of pelvic limb weakness and reluctance to jump and climb stairs. Neurological examination was consistent with an L4–L6 myelopathy. Ectopic calcification of soft tissues within the vertebral canal involving the dorsal longitudinal ligament over the L5–L6 vertebrae was diagnosed based on radiography, magnetic resonance imaging and computed tomography. The lesion was surgically excised, and histopathological examination demonstrated fibrous connective tissue with calcification. The patient was neurologically normal 2 months after surgery, and no relapses have occurred in a follow-up period of 2 years. The findings in this case had not been previously reported in cats, and share some similarities with a clinical entity in humans named ossification of the posterior longitudinal ligament.

KEYWORDS

cats, dorsal longitudinal ligament, ectopic calcification, spinal cord

BACKGROUND

Ossification of the posterior longitudinal ligament (OPLL) is a clinical entity described in human medicine caused by ectopic calcification within this ligament. This hyperostotic condition causes spinal cord compression more commonly in the cervical spine, but it can also affect the thoracic and lumbar spine. Although 5% of human cases are asymptomatic, most show signs of myelopathy or radiculopathy that are variable depending on the affected area and the severity of vertebral canal stenosis.[1] The pathogenesis of OPLL remains obscure, and a multifactorial aetiology including genetic and environmental factors has been suggested. Some authors have also suggested that the condition in humans is a variant of diffuse idiopathic skeletal hyperostosis (DISH).[1,2]

In veterinary medicine, the most common hyperostotic disorders that affect the vertebral column are spondylosis deformans and DISH.[3] DISH is a systemic disease characterised by ossification of soft tissues including the ventral longitudinal ligament and has been reported in one cat.[4] Although DISH in dogs often remains clinically silent, the only case reported in domestic cats showed signs consistent with a T3–L3 myelopathy. The dorsal longitudinal ligament in dogs and cats is the analogue of the posterior longitudinal ligament in human beings. Neither spondylosis deformans nor DISH in dogs and cats are reported to cause calcification of the dorsal longitudinal ligament.

The following case report describes the clinical features, multimodal imaging findings, surgical treatment and outcome of a feline patient with an L4–L6 myelopathy that shares striking similarities with OPLL in humans. To the authors' knowledge, this entity has not been previously described in veterinary medicine.

CASE PRESENTATION

A 9-year-old, male, neutered Russian blue cat was presented for a 2-week history of progressive pelvic limb weakness and reluctance to jump and climb stairs. According to the owner, the patient showed clinical signs of pain/discomfort.

On presentation, the physical examination was considered normal. Neurological examination revealed a mild ambulatory paraparesis, proprioceptive deficits and reduced patellar reflexes in both pelvic limbs. Pain was elicited upon palpation of the caudal lumbar spine. The remaining neurological examination was considered normal. Neuroanatomic localisation was consistent with an L4–L6 myelopathy, and main differential diagnoses included intervertebral disc disease, neoplasia and inflammatory/infectious diseases.

INVESTIGATIONS

Complete blood cell count and serum biochemistry (including ionised calcium) did not reveal any abnormalities. Serological testing for feline immunodeficiency virus and feline leukaemia virus were negative. Thoracic radiographs and abdominal ultrasound were unremarkable.

Lateral radiographs of the lumbar spine were performed under general anaesthesia. The L5–L6 disc space was slightly narrowed, and a thin double radiopaque line was observed within the vertebral canal overlying the same intervertebral disc space (Figure 1).

Magnetic resonance imaging (MRI) of the lumbar vertebral column was performed with a low-field MRI (Airis Light 0.25T, Hitachi). Sequences included sagittal and transverse T2-weighted (T2W; TR = 3200 ms, TE = 80 ms), sagittal short tau inversion recovery (STIR; TR = 2800 ms, TE = 16 ms, TI = 90 ms), sagittal 3D heavily T2W sequence with myelographic effect (3DFSEMyelo; TE = 8000 ms, TE = 525 ms) and sagittal and transverse T1-weighted images (T1W; TR = 512 ms, TE = 18 ms) before and after intravenous administration of paramagnetic contrast (gadoteric acid, 0.1 mmol/kg bodyweight; Dotarem, Guerbet). On T2W images, all the nuclei pulposi of the lumbar intervertebral discs had normal hyperintense appearance except for the L5–L6 and L6–L7 discs that had a partial and a total loss of the T2 hyperintense signal, respectively. There was an extradural mass lesion overlying the L5–L6 intervertebral disc space causing severe compression of the spinal cord. On the sagittal plane, the lesion had a peripheral thin rim that was hypointense on T2W, T1W and STIR sequences, consistent with calcification. The centre of the lesion was hyperintense on T2W/STIR images and isointense on T1W images (Figure 2). On the transverse plane, the hypointense peripheral thin rim had a reverse C-shaped morphology (Figure 3). Moderate contrast enhancement of the centre of the lesion was detected on post-contrast T1W images.

Computed tomography (CT) of the lumbar spine using a two-slice scanner (Somaton Spirit, Siemens) was performed immediately after MRI for further characterisation of the lesion. Bone and soft tissue window settings were used. An extradural lesion with a hyperattenuating peripheral rim and a central hypoattenuating core was detected. On sagittal reconstruction, a double-layer hyperattenuating rim was clearly seen overlying the L5–L6 intervertebral disc space (Figure 4a,b). No other ectopic calcification/ossification sites were detected.

A cerebrospinal fluid (CSF) sample was collected from the cisterna magna, and CSF analysis was unremarkable (three nucleated cells/μL, total protein concentration 19 mg/dL; reference ranges: <5 nucleated cells/μL and <25 mg/dL).

DIFFERENTIAL DIAGNOSIS

Based on the clinical and imaging findings, the presumptive diagnosis was compressive myelopathy caused by ectopic calcification/ossification of soft tissue within vertebral canal involving the dorsal longitudinal ligament. Intervertebral disc protrusion with atypical imaging findings, a calcified inflammatory/infectious granuloma or, less likely, a calcified neoplasia could not be completely ruled-out.

LEARNING POINTS/TAKE-HOME MESSAGES

- Calcification of the dorsal longitudinal ligament should be considered when calcified lesions are seen within the vertebral canal.
- Combined magnetic resonance imaging and computed tomography is a useful diagnostic imaging approach to assess uncommon spinal disorders, as they provide complementary information.
- Surgical treatment in this case resulted in a long-lasting good outcome.

TREATMENT

A right-sided hemilaminectomy was performed over the L5–L6 vertebrae. Upon opening the vertebral canal, the spinal cord was observed to be severely deviated dorsally, but no obvious mass or extruded disc material were visible. Careful inspection of the ventral epidural space was performed using Rothon microdissectors, and a very hard epidural mass was detected in the midline overlying the intervertebral disc space. The lesion was firmly attached to the spinal cord, and it was not possible to remove it without causing excessive traction over the nervous tissue. A no. 11 scalpel blade was used to gently separate the mass from the dura mater and from the floor of the vertebral canal. A plaque of white calcified material attached to fibrous tissue debris was retrieved and submitted for histopathological analysis. CSF leakage was observed just after spinal cord decompression, and absorbable dura substitute (Lyoplant; B. Braun Surgical) was used to repair the dural tear. The patient recovered uneventfully from the surgical procedure.

OUTCOME AND FOLLOW-UP

Microscopically, the lesion was a proliferation of fibrous connective tissue with low cellularity associated with a granular basophilic material (calcified tissue). There were also small fragments of cancellous bone and haemorrhagic foci (Figure 4c). There were no intervertebral disc material, neoplastic cells or inflammatory reaction. These findings were suggestive of a diagnosis of calcification/ossification of the dorsal longitudinal ligament.

The patient was rechecked periodically by the neurology personnel, and 2 months after surgery, the neurological examination was normal. No relapses were observed in a follow-up period of 2 years.

DISCUSSION

The findings described in this case report are consistent with an ectopic calcification of the dorsal longitudinal ligament, causing a lumbar myelopathy in a feline patient. The imaging findings are very similar, if not identical, to those described in OPLL in humans.[1] According to the human classification system, which relies on lateral radiography, there are four OPLL

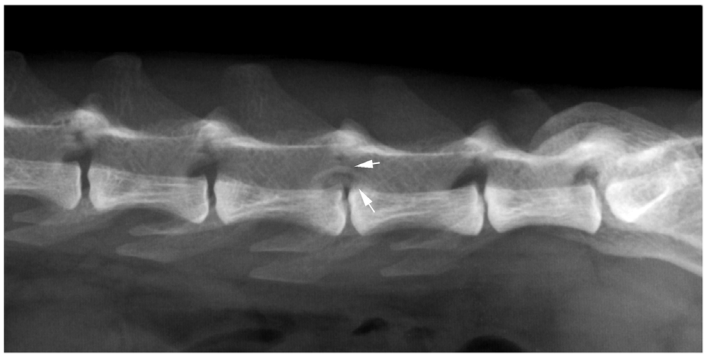

FIGURE 1 Lateral radiography of the lumbar vertebral column, showing a double radiopaque line overlying the L5–L6 intervertebral disc space within the vertebral canal (white arrows).

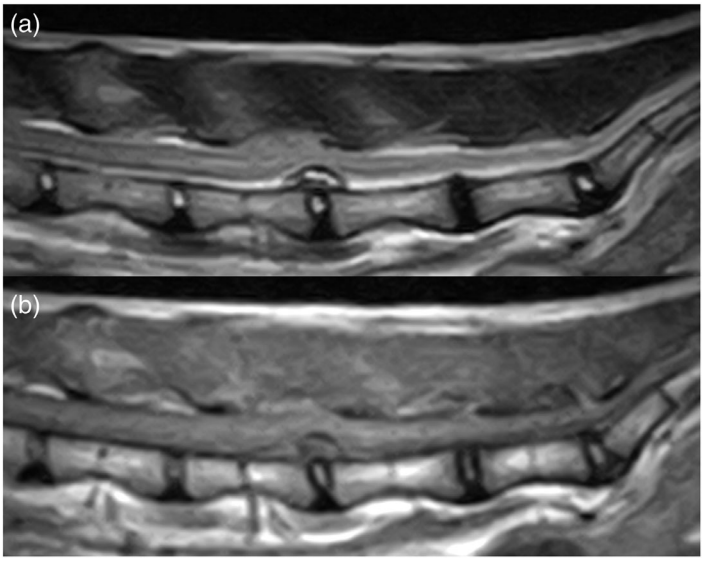

FIGURE 2 Magnetic resonance imaging of the lumbar spine: T2-weighted (T2W) sagittal (a) and T1-weighted (T1W) sagittal (b) image. Extradural mass lesion dorsal to the L5–L6 intervertebral disc space causing a severe compression of spinal cord. The lesion had a peripheral thin rim hypointense in T2W and T1W. The centre of the lesion is hyperintense in T2W and isointense in T1W images.

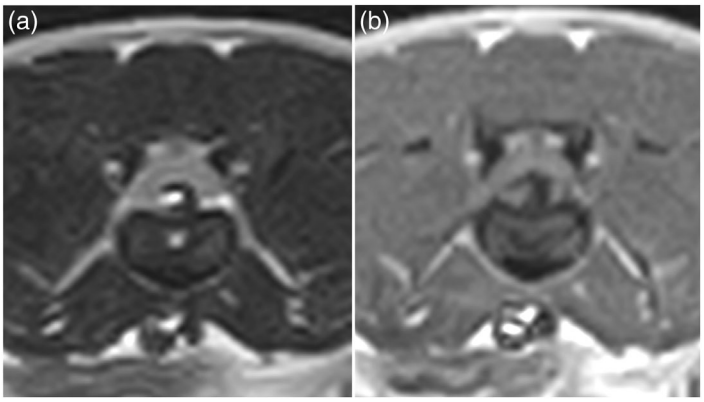

FIGURE 3 Magnetic resonance imaging of the lumbar spine: T2-weighted (T2W) transverse (a) and T1-weighted (T1W) transverse (b) image. The extradural mass lesion has a peripheral thin rim hypointense in T2W and T1W images and a reverse C-shaped morphology.

variants: continuous, segmental, localised and mixed.[1] As the dorsal longitudinal ligament in dogs and cats is the analogue of the posterior longitudinal ligament in humans, the case reported here would be similar to the localised form of the disease in which ligament ossification is distributed over the intervertebral disc space without involvement of the vertebral body.

Furthermore, OPLL can be subclassified into single-layer and double-layer type depending on the morphology assessed by CT scan.[5,6] The single-layer sign consists of a single homogeneous ossified posterior longitudinal ligament mass and the double-layer sign describes anterior and posterior hyperattenuating rims (corresponding to ossification), separated by a central hypoattenuating area (corresponding to hypertrophied non-ossified ligament).[6] Surgical treatment of OPLL may result in a dural defect that often causes CSF fistulas, because the ossified ligament is firmly attached to or can even penetrate the dura mater. The presence of a double-layer sign is important, because it is associated more often with the presence of a dural defect than the single-layer sign.[1,5–7]

In the case reported here, the double-layer sign was clearly identified on CT images, and the lesion was confirmed to be tightly adherent to dura mater during surgery.

The main imaging differential diagnosis for a calcified extradural mass overlying the intervertebral disc space is intervertebral disc extrusion. The presence of a double-layer sign on the CT images, as well as the presence of a calcified

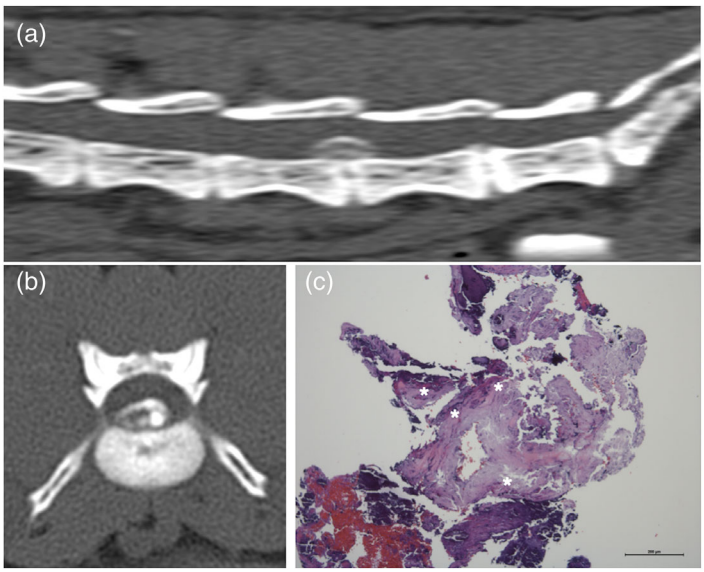

FIGURE 4 Computed tomography (CT) of the lumbar spine: sagittal reconstruction (a) and transverse CT image at the level of the L5–L6 vertebrae (b). An extradural lesion with hyperattenuating peripheral rim and central hypoattenuation (double layer sign). (c) Microscopic appearance of fragments of the dorsal longitudinal ligament. Note the presence of several ossified focal areas between fascicles of collagen fibres (white asterisks). Haematoxylin–eosin stain; bar: 200 μm.

thin rim extending over the vertebral bodies beyond the vertebral epiphyses, could help to differentiate between calcification/ossification of the dorsal longitudinal ligament and calcified intervertebral disc material.

The multimodal imaging diagnosis approach used in this case is also used in human medicine. MRI is helpful in detecting presence of myelomalacia secondary to compression, the extent of spinal cord compression and the presence of foraminal stenosis.[1]

In human medicine, the pathogenesis of OPLL is poorly understood, and some authors have suggested that OPLL is a variant of DISH.[1,2] There is only one case of feline DISH reported, and the imaging findings are the same as those described in dogs.[4] The case reported here does not fulfil the diagnostic criteria for DISH, and to date, involvement of the dorsal longitudinal ligament has not been associated with DISH in veterinary medicine.[3,4]

The cat was not hypercalcaemic and no other sites of ectopic calcification were detected on thoracic and abdominal images, thus an underlying metabolic disorder was not found for the spinal calcification. Hypervitaminosis A can cause a metabolic deforming osteopathy in cats.[8] Although serum vitamin A concentration was not determined, hypervitaminosis A seems unlikely because the cat was on a well-balanced diet, the lesion was focal and there has been a long follow-up period without signs of relapse or other clinical signs.

To the authors' knowledge, this case, supported by the imaging and histological findings, is the first description of a feline lumbar compressive myelopathy resembling OPLL in humans.

AUTHOR CONTRIBUTIONS

Mar Reus-Coll wrote the manuscript. Ignacio Montes de Oca, Mar Reus and Cristian de la Fuente acquired and interpreted the imaging part. Marti Pumarola performed histopathological study. Sonia Añor and Cristian de la Fuente conceived, designed and supervised the project. All authors read and approved the final version of the manuscript.

CONFLICT OF INTEREST STATEMENT

The authors declare they have no conflicts of interest.

FUNDING INFORMATION

The authors received no specific funding for this work.

ETHICS STATEMENT

The presented case report fulfils the high ethical standards concerning animal welfare. All investigations and treatments were performed with the client's consent.

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