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Surgical stabilisation of hiatal hernia and gastroesophageal reflux associated with idiopathic inflammatory polymyopathy in a Wire Fox Terrier

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Abstract: A one-and-a-half-year-old male Wire Fox Terrier weighing 3 kg presented with continuous drooling and vomiting. Its body condition score was 3/9, and severe atrophy of the temporal/masticatory muscle, trismus, and enophthalmos was observed on physical examination. The radiographic examination and fluoroscopic oesophagography revealed a type 1 hiatal hernia with gastroesophageal reflux. The serology tests revealed increased muscle enzyme activities. The antibody tests for acetylcholine receptor, type 2M fibre, and infectious agents were negative. A conventional surgical treatment was performed, and a thickened, stretched, and flaccid diaphragmatic muscle and an extended inelastic oesophageal hiatus were observed. On the histological examination of the diaphragmatic muscle, a diffuse histiocytic myositis was confirmed. Although the postoperative gastroesophageal reflux totally disappeared, the oesophageal motility and clinical signs did not improve significantly. Medical treatment with immunosuppressive agents was attempted and was effective in alleviating the clinical signs and abnormal oesophageal motility. The health condition of the dog was adequately maintained in the 12-month monitoring period. These findings suggest that, although the medical and surgical treatment have different therapeutic effects, they should be considered simultaneously for the management of a hiatal hernia associated with polymyopathy in dogs.

Keywords: muscular dystrophy; myositis; oesophageal hiatus; oesophageal motility

A hiatal hernia describes the condition in which the abdominal contents bulge through the oesophageal hiatus of the diaphragm into the thorax. Four types of hiatal hernia have been described in dogs and cats (Bright et al. 1990; Callan et al. 1993; Katsianou et al. 2014). Type 1, also known as a sliding hiatal hernia, is the most commonly reported type and is defined as the axial displacement of the distal oesophagus, gastroesophageal junction, and stomach into the thorax (Sivacolundhu et al. 2002; Katsianou et al. 2014). Other less frequent types include type 2, known as paraoesophageal hiatal hernia, in which the gastroesophageal junction remains in its normal position, but a portion of the fundus bulges through the hiatus into the thorax; type 3, combines elements of both type 1 and type 2; and type 4, which is a type 3 hernia complicated with a herniation of the organs other than the stomach

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(Sivacolundhu et al. 2002; Katsianou et al. 2014). It is essential to differentiate among the hiatal hernia types due to the differences in the pathophysiology, which necessitate different treatment strategies (Prymak et al. 1989; Sivacolundhu et al. 2002; Katsianou et al. 2014). Type 1 hiatal hernias are usually congenital and, thus, observed mainly in young dogs (Keeley et al. 2008). The acquired form of a type 1 hiatal hernia occasionally occurs as a result of a traumatic event or in combination with severe respiratory disease (Sivacolundhu et al. 2002). Clinical signs of a type 1 hiatal hernia include hypersalivation, regurgitation, vomiting, and dysphagia, which are usually secondary to reflux oesophagitis or megaoesophagus (Sivacolundhu et al. 2002; Tauro et al. 2015). Occasionally, these clinical signs may cause aspiration pneumonia (Ellison et al. 1987; Lorinson and Bright 1998). The two main treatment options for type 1 hiatal hernias are medical therapy and surgery. Medical treatment aimed at resolving the reflux oesophagitis and associated megaoesophagus has been recommended as a first-line treatment by some authors (Prymak et al. 1989; Lorinson and Bright 1998; Sivacolundhu et al. 2002). The most common surgical treatment for type 1 hiatal hernias is a combination of a diaphragmatic hiatal plication, an oesophagopexy, and a left-sided gastropexy (Prymak et al. 1989; Callan et al. 1993; Lorinson and Bright 1998; Guiot et al. 2008). The use of fundoplication techniques in dogs has resulted in poor outcomes, despite modification of the original surgery (Ellison et al. 1987; Prymak et al. 1989; Callan et al. 1993; Lorinson and Bright 1998).

Inflammatory myopathies are characterised by immunological responses in the skeletal muscles (Tauro et al. 2015). The underlying aetiology of these diseases in dogs includes an immune malfunction and infection by protozoa, bacteria, rickettsiae, and/or parasites (Evans et al. 2004; Tauro et al. 2015). In human medicine, the most common inflammatory myopathies include polymyositis, dermatomyositis, necrotising autoimmune myositis, and inclusion body myositis (Dalakas 2012). Polymyositis has been reported in various breeds, such as Boxers, German Shepherds, Retrievers, and the Hungarian Vizsla. Clinical, serological, electromyographic, and histological criteria are required for the diagnosis of inflammatory polymyopathy (Podell 2002; Evans et al. 2004; Platt et al. 2006; Tauro et al. 2015). There have been a few reports describing hiatal hernias associated with polymyopathy; however, type 1 hiatal hernias related to inflammatory polymyopathy have rarely been reported. In particular, no report has been published distinguishing the roles of the medical and surgical treatment for this condition.

The present report describes the successful surgical stabilisation of a type 1 hiatal hernia and gastroesophageal reflux presumably related to the idiopathic inflammatory polymyopathy in a Wire Fox Terrier, and the ensuing discussion addresses the respective roles of the medical and surgical treatment.

Case description

A one-and-a-half-year-old male Wire Fox Terrier was presented to the Veterinary Medical Teaching Hospital of Konkuk University (Seoul, Republic of Korea) for evaluation of a hiatal hernia. The referring veterinarian reported perpetual vomiting, drooling, and acute weight loss on the initial observation. On presentation, the patient was depressed, with a body condition score of 3/9. The patient exhibited continuous vomiting and drooling, with severe atrophy of the temporal/masticatory muscle, trismus, and enophthalmos. The serum biochemical abnormalities included elevated activities of aspartate aminotransferase, 147 IU/l (reference range, 0–50 IU/l); alanine aminotransferase, 441 IU/l (reference range, 10–100 IU/l); and creatine kinase, 1 661 IU/l (reference range, 10–200 IU/l). A plain thoracic radiography revealed findings compatible with a hiatal hernia with a proximal stomach herniation into the thorax through the oesophageal hiatus (Figure 1). On the fluoroscopic oesophagography, the esophagogastric junction and a portion of the stomach were displaced cranially into the thoracic cavity, along with a severely decreased oesophageal motility and gastroesophageal reflux (Figure 2). The gastroesophageal junction sphincter did not appear to contract properly (Figure 2). The contrast medium refluxed from the stomach to the oesophagus, and the diameter of the gastroesophageal junction was almost equal to that of the influx. The serum anti-acetylcholine receptor antibody, type 2M fibre antibody, and infectious agent antibody tests were negative. A diagnosis of a type 1 hiatal hernia was made. The dog was fed liquefied food in an elevated position to prevent food aspiration. An empirical medical management was first

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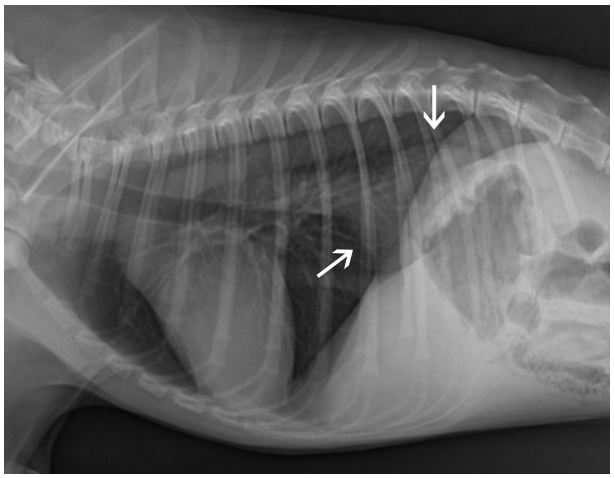


Figure 1. Right lateral thorax radiograph with a soft tissue opacity (white arrows), which represents the herniated cardia and the proximal part of the stomach

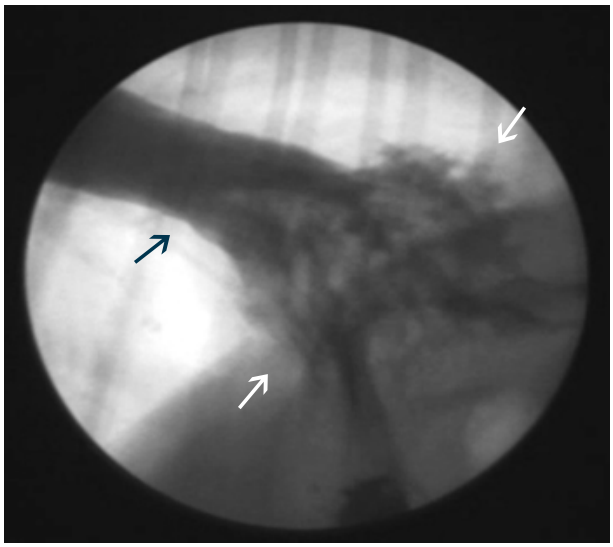
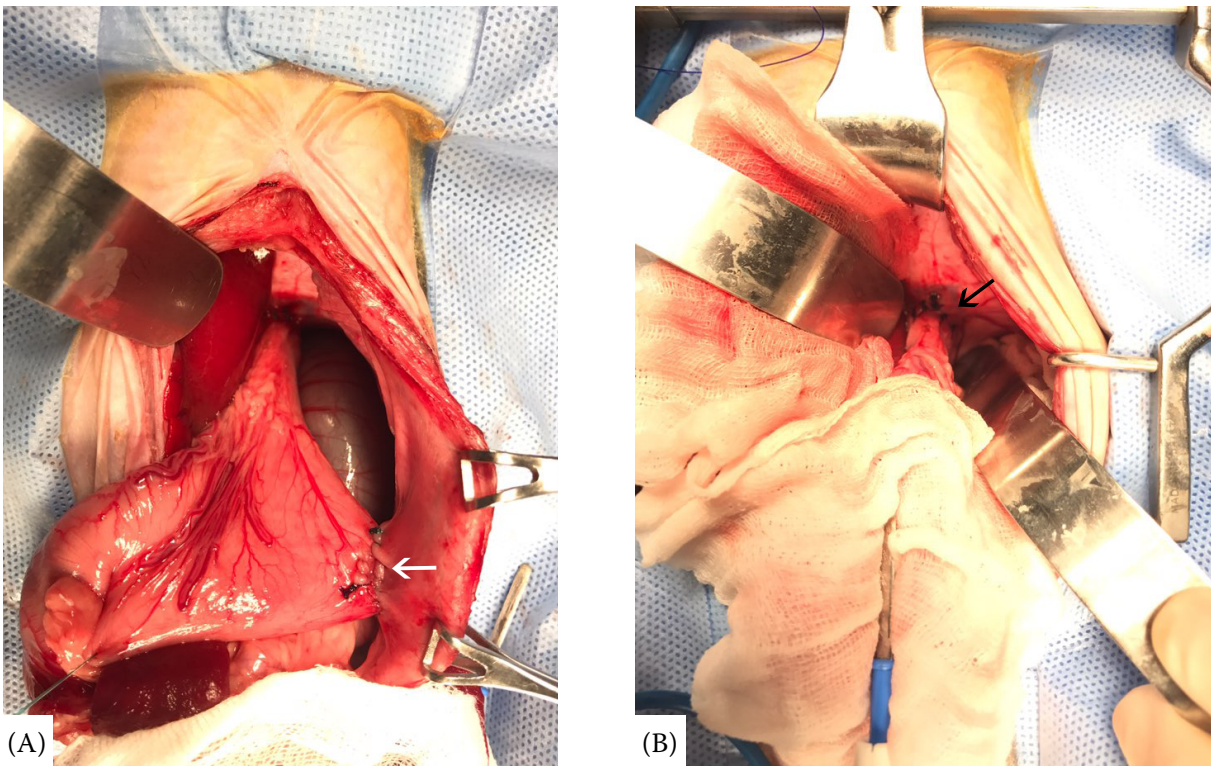


Figure 2. Fluoroscopic oesophagography during the gastroesophageal reflux revealing the cranial displacement of the oesophagogastric junction and part of the stomach into the thorax (white arrows), with a concurrent gastroesophageal junction dilation (blue arrow)

initiated with metoclopramide [0.5 mg/kg, subcutaneous (s.c.), twice per day (b.i.d.)], metronidazole [15 mg/kg, intravenous (i.v.), b.i.d.], cefazolin (30 mg/kg, s.c., b.i.d.), tramadol (4 mg/kg, s.c., b.i.d.), famotidine (1 mg/kg, i.v., b.i.d.), maropitant [1 mg/kg, s.c., once per day (s.i.d.)], and aluminium sucrose sulfate [Ulcerlmin, 5 ml, per os (p.o.), three times per day (t.i.d.)]. After failure of the empirical medical treatment, surgical stabilisation involving a phrenoplasty, an oesophagopexy, and a left-sided gastropexy was performed (Figure 3). The dog was premedicated with cefazoline (30 mg/kg, i.v.), at-



(A) (B)

Figure 3. Intraoperative images demonstrating the left-sided gastropexy (white arrow) (A) and phrenoplasty/oesophagopexy (black arrow) (B)

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ropine sulfate (0.02 mg/kg, s.c.), and butorphanol (0.1 mg/kg, i.v.), followed by anaesthesia induction with propofol (6 mg/kg, i.v.). The dog was intubated, and anaesthesia was maintained with isoflurane in oxygen. Intravenous fluids were administered at a rate of 5 ml/kg/h intraoperatively until complete recovery from anaesthesia. A ventral midline celiotomy incision was performed, and the sliding hiatal hernia was reduced by manual traction on the gastric fundus. The diameter of the oesophageal hiatus was approximately 4 cm. The phrenoplasty was performed by approximating the left and right crura of the diaphragm using two simple interrupted 3-0 polypropylene sutures. The oesophageal hiatus was reduced to a diameter through which only the index finger could be inserted. The oesophagus was then sutured to the diaphragm by placing two simple interrupted 3-0 polypropylene sutures on each side of the oesophageal hiatus between the tunica muscularis along the surface of the oesophagus and the diaphragm. Finally, a left-sided gastropexy was performed by incising the seromuscular layer of the gastric fundus and left ventrolateral abdominal wall. The abdominal wall incision was extended through the peritoneum and transversus abdominis muscle. The gastric incision edges were sutured to the corresponding abdominal wall incision edges using 3-0 polydioxanone in a simple continuous suture pattern. Copious lavage with sterile saline and a routine closure completed the procedure. During the surgery, there was evidence of a thickened, stretched, and flaccid diaphragmatic muscle and an extended inelastic oesophageal hiatus. The postoperative management included intravenous fluid therapy and maintenance of the preoperative medications. The postoperative analgesia was provided by a continuous infusion of butorphanol (0.1 mg/kg/h, i.v.) for 24 h and intermittent doses of butorphanol (0.1 mg/kg, i.v.) as needed for pain. Diffuse histiocytic myositis of the diaphragmatic muscle was diagnosed based on the histopathologic examination showing an inflammatory cell infiltration and fragmented/degenerated myocytes (Figure 4). On the fluoroscopic oesophagography, 5 days after surgery, the gastroesophageal reflux virtually disappeared, and the gastroesophageal junction appeared to be in the normal position. In contrast, the oesophageal motility remained decreased, and vomiting was frequently observed. An acetylcholinesterase inhibitor (pyridostigmine, 0.5 mg/kg, p.o., b.i.d.) was additionally administered; however,

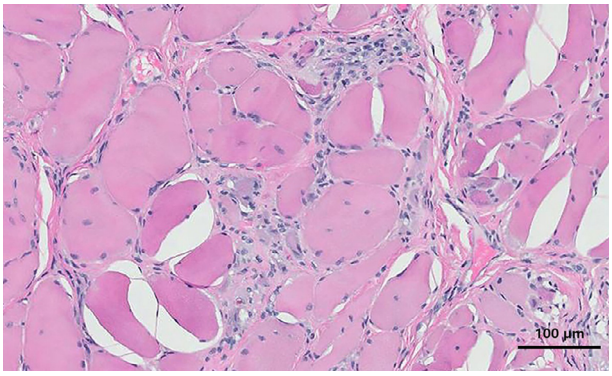

Figure 4. Histopathological examination of the diaphragmatic muscle showing an inflammatory cell infiltration and fragmented/degenerated myocytes

even after increasing the dose to 1 mg/kg, the clinical signs were not alleviated. After administration of azathioprine (2 mg/kg, p.o., s.i.d.), the patient started to take food voluntarily and gained weight. Azathioprine was slowly tapered off and replaced by cyclosporin microemulsion (25 mg/dog, p.o., s.i.d.) over a 1-month period. Although intermittent vomiting (once per week) was observed by the owner, the dog was reported to be in good health during the 12-month monitoring period.

DISCUSSION AND CONCLUSIONS

Since the first report of a hiatal hernia in dogs by Gaskell, numerous additional cases have been reported in the veterinary literature (Gaskell et al. 1974; Ellis 1980; Kirkby et al. 2005; Guiot et al. 2008). Although the exact aetiology and pathogenesis remain uncertain, several interrelated factors have been postulated to be responsible for hiatal hernias, including: anatomical deformities of the hiatal canal and phrenoesophageal ligament; a neurological or muscular disorder; a severe respiratory disease; trauma; and an increasing gradient between the intraabdominal and intrathoracic pressure (Kahrilas et al. 1999; Sivacolundhu et al. 2002). These factors compromise the function of the lower oesophageal sphincter or separate the extrinsic anatomical structures (diaphragmatic crus, phrenoesophageal ligament, fixation of the oesophagus to the liver) from the lower oesophageal sphincter, causing the dynamic axial motion of the gastroesophageal junction into the thorax (Sivacolundhu et al. 2002; Mayhew et al. 2017). In the majority of the type 1 hiatal hernia cases reported to date, the clinical

https://doi.org/10.17221/47/2020-VETMED

signs had been caused by gastroesophageal reflux (Katsianou et al. 2014). Mayhew et al. (2017) suggested that the clinical signs are mediated by the axial movement of the gastroesophageal junction rather than the dysfunction of the lower oesophageal sphincter. However, many dogs and cats have asymptomatic type 1 hiatal hernias (Bright et al. 1990). Why and how the gastroesophageal reflux occurs in some individuals and not others remains unclear. In human studies, a gastroesophageal reflux disease by itself may further worsen the function of the lower oesophageal sphincter, thus initiating a self-perpetuating cycle (Ellis 1980; Dalakas 2012).

Muscular dysfunction caused by tetanus, myasthenia gravis, masticatory muscle myositis, idiopathic polymyopathy, and/or muscular dystrophy has been reported to be associated with hiatal hernias (Ham and Bree 1992; Acke et al. 2004; Dalakas 2012; Tauro et al. 2015). However, the exact mechanism of a hiatal hernia and gastroesophageal reflux secondary to the muscular dysfunction remains unclear. Tetanus appears to cause muscular diaphragmatic spasms, which in turn cause the stretching of the central tendon of the diaphragm (Ham and Bree 1992; Acke et al. 2004). Concurrent with this spasm, shortening of the oesophagus may result in a hiatal hernia. Merieux et al. hypothesised a possible relationship between the polymyositis/dermatomyositis and oesophageal abnormalities causing food reflux (Merieux et al. 1983). Muscular dystrophy associated with oesophageal and diaphragmatic muscular dysfunction has been studied in various canine breeds (Merieux et al. 1983; Evans et al. 2004; Brumitt et al. 2006; Dalakas 2012). In a report describing idiopathic inflammatory polymyopathy in the Hungarian Vizsla, the relationship between idiopathic inflammatory polymyopathy and other idiopathic immune-mediated diseases, including atopic dermatitis, immune-mediated polyarthritis, inflammatory bowel disease, keratoconjunctivitis sicca, sebaceous adenitis, and steroid responsive meningitis arteritis, have been suggested (Tauro et al. 2015).

An unusual aspect of the present case was that the hiatal hernia with the gastroesophageal reflux occurred concurrently with the regional specific severe atrophy of the temporal/masseter muscle, trismus, and enophthalmos. Enophthalmos has been reported to be secondary to pterygoid muscle atrophy (Tauro et al. 2015). Given that the muscular atrophy and increased serum levels of creatine kinase were observed together, serological testing for the determination of antibodies associated with myopathies, including an acetylcholine receptor antibody (myasthenia gravis), a type 2M fibre antibody (anti-masticatory muscle myositis), and an infectious agent antibody (ehrlichiosis, toxoplasma, neosporosis, leishmaniosis), was performed. However, all of these test results were negative. A marked elevation of creatine kinase levels is an indication of skeletal muscle damage and, in the case reported herein, was considered to be associated with the temporal/masseter/pterygoid muscle atrophy and diaphragm muscle myositis. However, other skeletal muscles, such as the pharyngeal and oesophageal muscles, may also be associated with serum creatine kinase activity, given that the myopathy of these muscles could contribute to the dysphagia and gastroesophageal reflux. An association between the magnetic resonance imaging (MRI) and the histopathologically identified muscle inflammation has been reported (Platt et al. 2006). As such, an MRI may be a useful diagnostic method for differentiating abnormal from unaffected muscles and, therefore, may facilitate a more accurate biopsy.

The compromised function due to the diaphragmatic myositis in the present case was regarded as the major cause of the hiatal hernia. A diaphragmatic muscle myopathy may cause the stretching of the oesophageal hiatus, which may induce separation of the gastroesophageal junction from the hiatus. Conventional surgical techniques, including phrenoplasty, oesophagopexy, and left-sided gastropexy, to correct the type 1 hiatal hernia were performed, and the postoperative fluoroscopic oesophagography demonstrated that these surgical techniques prevented the relapse of the hiatal hernia and gastroesophageal reflux. However, considering that the clinical signs and oesophageal motility abnormalities were not appreciably alleviated, the surgical treatment did not appear to improve the muscular function of the oesophagus or other musculature. Alleviation of the clinical signs after administration of the immunosuppressive agents (azathioprine and cyclosporine) used to relieve the idiopathic inflammation of the associated muscular structures indirectly suggests that the abnormally decreased oesophageal motility was responsive to the immunosuppressive agents. Consequently, the gastroesophageal reflux occurred not only

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because of the muscular dysfunction of the lower oesophageal sphincter and diaphragm, but also because of the malpositioning of the gastroesophageal junction. Similar to other reports, abdominal pressure on the oesophagus and the angle of the oesophagus into the cardia may act as part of the antireflux mechanism.

The results of this study demonstrated that medical management alone may not be sufficient for a hiatal hernia caused by idiopathic inflammatory polymyopathy. Moreover, the concurrent surgical repositioning of the displaced gastroesophageal junction into the normal position could effectively alleviate the clinical signs. The surgical techniques used for this patient were not aimed at the functional improvement of the respective muscular structures, but for repositioning of the gastroesophageal junction and extrinsic anatomical structures in a harmonised manner.

In conclusion, the results of this study support the suspicion that other types of inflammatory polymyopathy could be involved in a type 1 hiatal hernia and gastroesophageal reflux disease. However, the histological or immunohistochemical evaluation of the affected muscle tissues would be necessary to confirm this hypothesis.

Our data, along with those from other reports, demonstrate that a hiatal hernia with gastroesophageal reflux caused by idiopathic inflammatory polymyopathy should be treated both surgically and medically.

Conflict of interest

The authors declare no conflict of interest.

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