Acute instability of the nuchal ligament following cervical neuromuscular dysfunction in a dressage horse

Acute instabiliteit van het ligamentum nuchae ten gevolge van cervicale neuromusculaire disfunctie bij een dressuurpaard

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ABSTRACT

A ten-year-old Warmblood dressage gelding was presented with acute instability of the nuchal ligament after paddock turnout. Based on the clinical signs, orthopedic and neurologic examination, diagnostic imaging and electromyography, cervical neuromuscular dysfunction of the M. obliquus capitis caudalis on the right side of the neck was diagnosed. Conservative treatment including steroidal anti-inflammatory medication in combination with oral supplementation with vitamin B1 and box rest resulted in complete recovery of the horse within six months.

SAMENVATTING

Een warmbloed-dressuurpaard (ruin) van tien jaar oud werd aangeboden met de klacht van acute instabiliteit van het ligamentum nuchae na vrije beweging in een paddock. Op basis van de klinische presentatie, orthopedisch en neurologisch onderzoek, medische beeldvorming en elektromyografie werd cervicale neuromusculaire disfunctie van de M. obliquus capitis caudalis aan de rechterzijde van de hals vastgesteld. Conservatieve therapie met corticosteroïden in combinatie met orale supplementatie van vitamine B1 en boxrust resulteerden na zes maanden in volledig herstel.

INTRODUCTION

The vertebrate nuchal ligament is a large elastic structure in the dorsal cervical midline between the occiput, the cervical vertebrae and the cranial thoracic spinous processes, which helps to support the head and neck of the horse (Gellman and Bertram, 2002a). The nuchal ligament consists of funicular (cord-like) and lamellar (sheet-like) parts (Dyson, 2003; Gellman and Bertram, 2002a). The funicular part is broad and flat at its origin on the cranial thoracic spines, and becomes more cord-like and narrow towards its insertion on the skull. The fibre bundles of the lamellar part are closely interwoven with those of the funicular part, with the lamellar bands coursing cranioventrally to insert on the dorsal spines of cervical vertebrae two through six (Gellman and Bertram, 2002a). The lamellar part separates the bilateral muscle groups of the neck (Dyson, 2003). The nuchal ligament primarily consists of the highly extensible biological polymer elastin (Gellman and Bertram, 2002b; Minns et al., 1973; Wainwright et al., 1982). When the head is lowered, the nuchal ligament is stretched and elastic strain energy is stored, whereas when the head is raised, the ligament returns to its previous length (Gellman and Bertram, 2001b). During locomotion, the caudal funicular and cranial lamellar regions contribute most to elastic strain energy storage, and therefore, the line of function is from the withers to the second cervical vertebra and not from the withers to the skull. Because of the capacities for energy storage in the nuchal ligament, this structure is substituting passive work for active muscular work, which allows the horse to preserve its metabolic energy resources (Gellman and Bertram, 2001b).

In the scientific literature, pathology of the equine

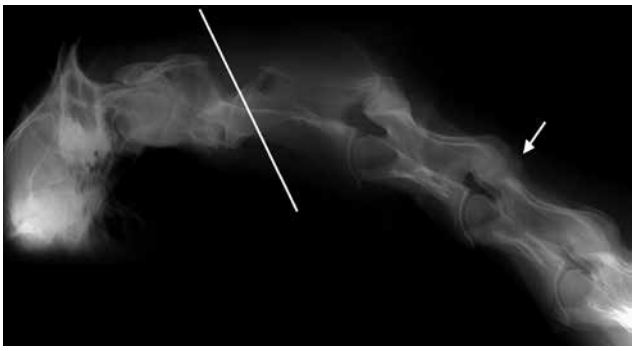

Figure 1. Left lateral view of the cervical vertebrae, illustrating the irregularly outlined bony fragment between the articular processes of the third and fourth cervical vertebrae (arrow). The white line is shown as a reference for the anatomical cross-section of the cadaver specimen presented in Figure 3.

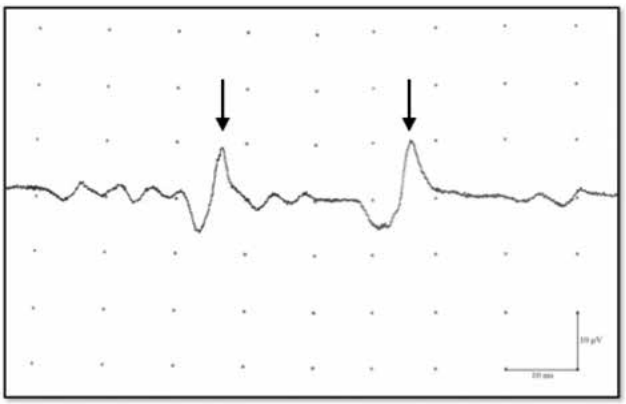

Figure 2. Electromyographic recording (filter 20Hz) of the right M. obliquus capitis caudalis presenting positive sharp waves (arrows), indicative of muscle denervation.

nuchal ligament is mainly limited to focal mineralization (Collobert et al., 1995; van Volkenberg, 1922) and insertional desmopathy (Nowak and Huskamp, 1989; Nowak, 2001), but these are often incidental findings. However, acute instability of the nuchal ligament has not been described before.

CASE REPORT

A ten-year-old Warmblood dressage gelding was referred with acute pain in the cranial cervical region after paddock turnout the day before. Before paddock turnout, the horse did not present any abnormalities and competed at an (inter-)national level. No specific traumatic events were noted but the horse was not under supervision the whole time. When retrieving the horse from the paddock, the owner was alerted by the very stiff and uncoordinated gait. Upon closer inspection, the horse was standing with an extended neck and was unable to raise the head. These symptoms did not disappear after one night of box rest. Therefore, the horse was admitted to the clinic for further examination. On clinical examination, instability of the nuchal ligament at the level of the second cervical vertebra was observed during lateroflexion of the head and neck, without any local deformation. The nuchal ligament intermittently luxated from its median position into a left or right paramedian position, depending on left or right cervical lateroflexion, respectively. Subjectively, more pronounced luxation was observed to the right than to the left side. The horse presented generalized neck stiffness and moderate to severe ataxia seen as lack of hind limb propulsion, toe-dragging and difficulties in gait transitions.

Lateral radiographic imaging of the cervical vertebrae revealed a rounded bony fragment in the dorsocaudal region of the intervertebral joint between the third and fourth cervical vertebrae and a concurrent mild dorsal angulation of the vertebral canal (Figure 1). The inter- and intravertebral ratios were within normal limits. No concurrent abnormalities of the cervical vertebrae were visible on the radiographic projections.

Ultrasonography (7.5-12 Mhz lineair probe) under sedation in a neutral position, confirmed a left-right shifting of the nuchal ligament into a paramedian position at the level of the second cervical vertebra, depending on left or right cervical lateroflexion, respectively. The ultrasonographic examination could not reveal relevant muscle atrophy on admission.

Neurological examination using transcranial magnetic stimulation was performed to assess the integrity of the motor tracts (Nollet et al., 2002). Electromyographic responses were recorded bilaterally from needle electrodes in the M. extensor carpi radialis and the M. tibialis cranialis. The onset latency in the left forelimb was 21.75±0.07 ms, right forelimb 22.00±0.42 ms, left hind limb 40.05±0.31 ms and right hind limb 41.25±0.19 ms. The amplitude of the left forelimb was 5.75 mV, right forelimb 5.33 mV, left hind limb 5.64 mV and right hind limb 3.39 mV. Comparing these data with reference values published by Nollet et al. (2002), mildly increased onset latency in the four limbs but no decrease in peak-to-peak amplitude in the descending motor tracts was recorded.

Electromyography (EMG) was used to identify insertional activity and pathological spontaneous electrical activity in the left and right M. rectus capitis major, the M. spinalis, the M. semispinalis and the M. obliquus capitis caudalis, which were all examined bilaterally at the day of admission. In contrast with the left side, the M. obliquus capitis caudalis on the right side of the neck presented positive sharp waves and fibrillation potentials persisting after needle insertion (Figure 2).

The horse was initially treated with dexamethasone (Rapidexon, Eurovet, the Netherlands) IV for four days using a gradually reducing dosage (0.1mg/kg – 0.025 mg/kg), followed by oral prednisolone (Equisolon, Boehringer Ingelheim, Germany), 1.5mg/kg for three days, 1 mg/kg for another seven days, 0.5 mg/kg for the next seven days and 0.5 mg/kg every other day for seven days. The horse was box-rested

and hand-walked for three months, and vitamin B1 (Thiamine, ABC Chemicals, Belgium) was supplemented (0.03g/kg/day) daily. Radiographic follow-up after eight weeks did not reveal any changes, whereas the stability of the nuchal ligament at its normal position had already returned. At that moment, no more signs of pain or restriction in motion of the cervical region were noted, no morphological abnormalities were detected and the ataxia was fully resolved. Six months after initial presentation, the horse had successfully returned to its previous athletic level.

DISCUSSION

In this case report, an unusual case of acute instability of the nuchal ligament is described in a ten-year-old dressage horse. The etiology of this condition is unknown. Chronic insertional desmopathy and dystrophic mineralization of the nuchal ligament and injury of the M. semispinalis have been described in 85% of Warmbloods suffering cervical trauma (e.g. pulling back when tied up) or undergoing an excessive amount of lunging exercise while restricted with side or draw reins (Dyson, 2003). However, none of these conditions leads to instability of the nuchal ligament.

In the literature regarding the equine nuchal ligament, predominantly, the craniocaudal attachments of the funicular part and the cranioventral insertion of the lamellar part on the dorsal spines of the cervical vertebrae have been described (Gellman, 2001). However, the lateromedial stability of this dynamic structure has not been investigated. The nuchal ligament is surrounded by the left and right M. semispinalis and by the M. rectus capitis dorsalis major ventrolaterally (Dyson, 2003; Mülling et al., 2014) (Figure 3). The latter is known to be relatively weak and occasionally atrophied without clinical significance (Mülling et al., 2014). Post-hoc anatomical dissections were performed to evaluate the surrounding musculature, and revealed the large dimensions of the M. obliquus capitis caudalis at the cranial aspect of the cervical spine. Therefore, the authors conclude that the latter muscle is of major importance for stabilizing the nuchal ligament besides its stabilizing function of the atlantoaxial joint and its role in the rotation of the head and atlas around the dens of the axis (Mülling et al., 2014; Nickel et al., 1986). This muscle, which lies cranially to the Mm. multifidi and connects the spinous process of the axis to the wing of the atlas, is very large at the level of the second vertebra. The unilateral neurogenic damage of this muscle may have caused instability of the overlying nuchal ligament (Figure 3). Therefore, trauma to the dorsal branch of the second cervical spinal nerve was presumed, as this nerve supplies the motor innervations of that muscle (Mülling et al., 2014). Although the exact pathogenesis in the present case remains unknown, chronic trauma and/or predisposing injuries are considered highly unlikely because the horse had not presented any abnormalities before and had successfully performed on (inter-) national level. Therefore, in the authors' opinion, an acute traumatic event causing instability in the cranial cervical region and subsequent ataxia and neurogenic damage to the M. obliquus capitis caudalis are most likely.

Although it could be argued that unilateral neuromuscular damage to the M. obliquus capitis caudalis would result in luxation of the nuchal ligament only to the ipsilateral side, the present case has clearly demonstrated instability to both sides albeit more pronounced to the right. In the authors' opinion, this may be explained by the position of the nuchal ligament dorsal to the M. obliquus capitis caudalis.

Diagnostic examination of the neck is challenging and can be done by radiography, ultrasonography, nuclear scintigraphy, computer tomography (CT) and myelography. Transcranial magnetic stimulation or electromyography can be used as ancillary tests. In the present case, a combination of radiography, ultrasonography, transcranial magnetic stimulation and

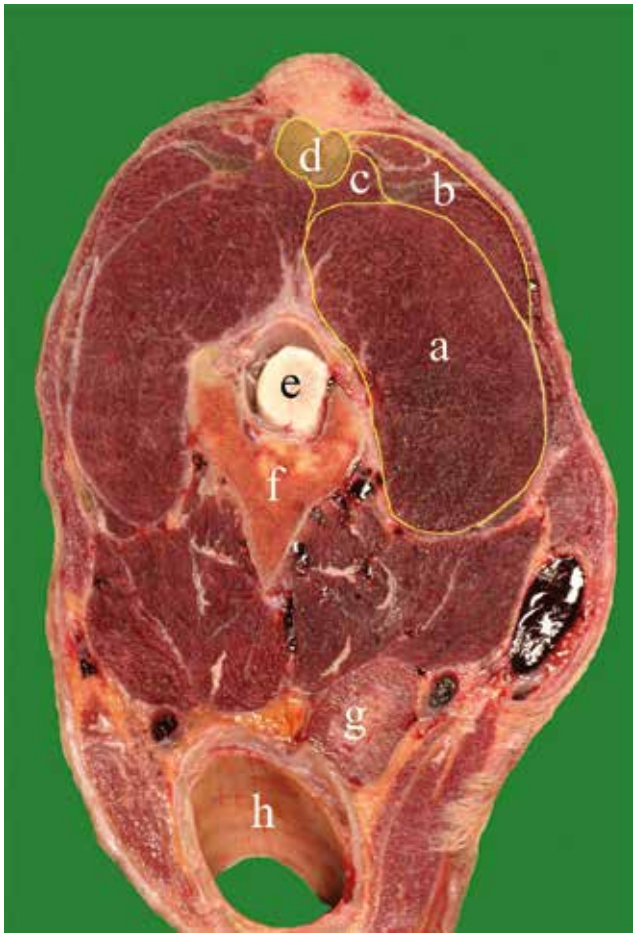

Figure 3. Anatomical cross-section of a cadaver specimen at the level of the second cervical vertebra (cranial view), illustrating the stabilizing function of the large muscle bodies of the bilateral M. obliquus capitis caudalis (a: left side); M. semispinalis (b: left side); M. rectus capitis dorsalis major (c: left side); nuchal ligament (d); spinal cord within the vertebral canal (e); second cervical vertebra (f); esophagus (g); trachea (h).

electromyography was required to fine-tune the diagnosis. From a diagnostic perspective, CT and myelography could have been of additional value. However, the horse in this case report presented with moderate to severe ataxia and therefore, general anesthesia and the significant risk of injury associated with anesthetic recovery (Young and Taylor, 1993) were considered inacceptable. In horses, myelography is routinely performed under general anesthesia, although it can be performed in the standing horse, albeit with increased risks, like generalized seizure (Foley et al., 1986). Under general anesthesia, computed tomography of the full equine neck can be performed using a minimal gantry opening of 85 cm and scanning field of 70 cm diameter (Kristofferson et al., 2014). Recently, a full body CT scanner (Equimagine, Four Dimensional Digital Imaging (4DDI), New York, USA) for the standing horse has been developed. Unfortunately, this equipment was not available in Belgium at the time of this case.

In the present case, only lateral radiographic projections were obtained and therefore, the presence of the rounded bony fragment between the third and fourth cervical vertebrae could not be assigned to the left or right side. Additional oblique radiographic projections would have been required to answer this question. However, the clinical importance of the rounded bony fragment is unclear, as similar fragments can be detected incidentally in normal horses (Stewart et al., 1991). Based on the absence of any local sensitivity or other structural abnormalities at this level and based on its distance to the spinal cord, the detected fragment is probably clinically irrelevant.

The use of magnetic motor evoked potentials is a quantitative and valuable diagnostic tool in cervical spinal cord disease in horses (Nollet et al., 2002). In the present case, this technique revealed mildly increased onset latency in the four limbs but no decrease in peak-to-peak amplitude, thereby providing objective and quantitative data and confirming the deficit in the descending motor tracts. In the absence of myelography, it is impossible to conclude about the exact site of spinal cord compression and about its static and/or dynamic nature. The final diagnosis was based on EMG, which presented fibrillation potentials and positive sharp waves on the right M. obliquus capitis caudalis. Fibrillation potentials are the most commonly observed abnormal spontaneous electropotential in EMG and strongly suggest denervation. Fibrillation potentials are thought to be spontaneous discharges from acethylcholine-hypersensitive denervated muscle fibers, or may result from muscle necrosis, muscle inflammation and focal muscle degeneration (Chrisman et al., 1972; Ettinger, 2005). The presence of fibrillation potentials is important in diagnosing denervation before clinical discernible muscle atrophy (Andrews and Lacombe, 2010). Positive sharp waves are potentials, in which the primary deflection is downward, followed by a lower-amplitude, longer-duration negative deflection, resembling a sawtooth. They occur with muscle denervation and muscular diseases (Andrews and Lacombe, 2010). In the present case, the M. obliquus capitis caudalis was affected, leading to the presumptive etiological diagnosis of trauma of to the dorsal branch of the second cervical spinal nerve innervating this muscle (Mülling et al., 2014). The acute stage, in which the present case was examined, did not allow detecting any morphological abnormalities in this muscle. At the eight-week check-up, no atrophy could be observed, but unfortunately, the EMG was not repeated. It would have been interesting to compare the previously detected positive sharp waves and fibrillation potentials with the muscle activity after resolution of the clinical signs.

In the case presented here, conservative treatment using steroidal anti-inflammatory medication and oral supplementation with vitamin B1 (thiamine) was successful. Steroidal anti-inflammatory drugs are the most commonly used drugs for acute traumatic injuries to the central nervous system (Nout, 2010). The neuroprotective effect of corticosteroids is thought primarily to be mediated by free radical scavenging but may include decreased catecholamines and glutamate levels, as well as decreased apoptosis-related cell death (Zurita et al., 2002). Other potential beneficial effects of corticosteroids include reduction in the spread of morphologic damage, preservation of vascular membrane integrity, prevention of the loss of axonal conduction and reflex activity and stabilization of white matter neuronal cell membranes in the presence of central hemorrhagic lesions. Furthermore, their anti-inflammatory properties are useful in reducing edema (Nout, 2010; de la Torre, 1981). Vitamin B1, also known as thiamine, may have a supporting role in the regeneration of nerve fibers and specifically of axons (Karachalias et al., 2010); however, evidence-based information for its efficacy is lacking.

The duration of rehabilitation was determined empirically based on clinical experience with cases presenting acute neurological deficits. Nerve injury has been described in different degrees, with neurapraxia being the first. Recovery occurs normally after several weeks up to three months following injury. However, with more severe injury like axonotmesis, the nerve will only regenerate at a rate of 2.54 cm/month (Lee and Wolfe, 2000). Therefore, a total rehabilitation protocol of three months was advised for this case. Box rest and handwalking were preferred over paddock turnout based on the temperament of the horse and the earlier traumatic event in the paddock. The follow-up at eight weeks and six months did not allow determining if the instability disappeared prior to the ataxia or vice versa.

Electrical muscle and/or nerve stimulation techniques may be useful in the management of neuromuscular damage (Sheffler and Chae, 2007) and were considered in the present case. However, for practical

reasons, the owner preferred a purely conservative approach. Theoretically, a surgical treatment option would have been bilateral imbrication and/or mesh implantation at the level of the nuchal ligament. However, this has not been described yet and therefore, its efficacy and potential complications remain unknown.

In conclusion, this report is the first to describe a case of acute instability of the nuchal ligament following traumatic cervical neuromuscular dysfunction. A combination of examination techniques was required to fine-tune the diagnosis. This case and the subsequent anatomical dissections performed by the authors suggest a stabilizing role of the M. obliquus capitis caudalis on the nuchal ligament in the cranial cervical area, which has not been described before. In this case, conservative management resulted in full recovery and the horse successfully returned to its previous athletic function.

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