Extradural spinal hydatid cyst causing hindlimb ataxia in a horse

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Summary

This case report describes a 13-year-old cob-cross gelding presented for evaluation of recent onset hindlimb ataxia. The gelding had undergone general anaesthesia and tenoscopy of the right hindlimb digital flexor tendon sheath at a nearby clinic three months earlier and had appeared normal at routine post-operative assessments until the sudden onset of neurological deficits. Spinal trauma was suspected initially but radiography and scintigraphy were unremarkable. Due to the severity and progressive nature of the clinical signs the gelding was subjected to euthanasia. Post mortem examinations (computed tomography, dissection and histopathology) revealed spinal cord compression caused by a single extradural hydatid cyst (*Echinococcus equinus*), confirmed with PCR, at the level of the 15th thoracic vertebra. This is the first report of a spinal hydatid cyst causing hindlimb ataxia and should therefore be considered a potential differential diagnosis for ataxia in the equine patient.

Keywords: Horse, hydatid cyst, hindlimb ataxia, spinal cord, imaging

Ataxie der Hintergliedmasse beim Pferd verursacht durch eine zystische Echinokokkose

Dieser Fallbericht beschreibt einen 13-jährigen Tinker Wallach, der zur Untersuchung einer kürzlich aufgetretenen Hinterbeinataxie vorgestellt wurde. Vor drei Monate wurde bei dem Wallach unter Allgemeinanästhesie eine Tenoskopie der Beugesehnenscheide der rechten Hintergliedmasse durchgeführt. Bei den routinemäßigen postoperativen Untersuchungen zeigte das Tier keine abnormalen Befunde bis zum plötzlichen Auftreten von neurologischen Defiziten. Bei der initialen Untersuchung wurde ein spinales Trauma vermutet, jedoch waren die Radiologischen Untersuchungen und Szintigraphie unauffällig. Aufgrund der Schwere und Progressivität der klinischen Symptome wurde der Wallach euthanasiert. Post-mortem-Untersuchungen (Computertomographie, Sektion und Histopathologie) zeigten eine Rückenmarkkompression auf Höhe des 15. Brustwirbels, die durch eine einzige extradurale Echinokokkuszyste (*Echinococcus equinus*) verursacht wurde. Dies ist der erste Bericht einer eine spinalen Hydatidenzyste die eine Hinterbeinataxie verursachte. Daher sollte eine Echinokokkuszyste als potentielle Differentialdiagnose für eine Ataxie beim Pferd betrachtet gezogen werden.

Schlüsselwörter: Pferd, Hydatidenzyste, Ataxie, Hintergliedmasse, Rückenmark, Bildgebende Diagnostik

https://doi.org/10.17236/sat00183

Received: 22.05.2018
Accepted: 13.08.2018

Introduction

Hindlimb ataxia is encountered in equine practice and is most frequently the result of cervical spinal cord compression caused by cervical vertebral malformation, trauma or enlargement of the intervertebral articulations due to osteoarthritis[12,14]. Thoracic lesions are much less common and are typically traumatic, often resulting in severe neurological deficits. Due to the large forces required to damage the spinal cord in this region and the associated soft tissue and bone damage, lesions in this area are often untreatable and necessitate euthanasia.

Hydatid cysts result from infection with a metacestode of the genus *Echinococcus* and have been reported in the liver and lungs of horses after natural and experimental infection[4,13,20]. Hydatid disease can affect great apes and

humans where ataxia caused by hydatid cysts have been reported[1,3,15,18]. This is the first case of a spinal hydatid cyst (*Echinococcus equinus*) reported in the horse.

Case report

History

A 13-year-old cob-cross gelding was diagnosed with right hindlimb lameness caused by a longitudinal tear of the lateral aspect of the deep digital flexor tendon within the digital sheath. The gelding was treated at a nearby clinic where the horse underwent general anaesthesia and tenoscopy while in dorsal recumbency with the right hindlimb extended. The damaged tendon was debrided and the plantar annular ligament transsected before the gelding recovered uneventfully from anaesthesia. Post- operative management included bandaging and suture removal after 14 days. Mesenchymal stem cells, cultured from a sternal bone marrow aspirate obtained at the time of surgery, were injected into the digital sheath 13 and 22 days post operatively. At 80 days after surgery, the horse displayed severe hindlimb ataxia, which the owner described as having deteriorated steadily over the previous two months. The gelding was monitored for a further few weeks before being referred to Rossdales Diagnostic Centre for further evaluation. In the interim the horse had been turned out with a stable companion without active exercise. The worming history of the animal was unknown. The horses stabled at the same location did not show any abnormal clinical signs.

Clinical Findings

At presentation the horse was bright and responsive with normal mentation. The gelding was eating, drinking, defecating and urinating normally. There were no cranial nerve deficits and there was a normal range of neck movement. Normal tail tone and anal reflex were present. A pronounced left more than right sided panniculus reflex was noted with hypoalgesia mid thorax. There was asymmetry of the thoracic epaxial musculature with notable reduction in muscle mass on the right side. Moderate distension of the right hind digital flexor tendon sheath was appreciated. At the walk, there was unilateral (right-sided) pacing with moderate to severe bilateral hindlimb weakness, the gelding displayed great difficulty co-ordinating the movements of the hind legs (ataxia grade 3/4 according to Mayhew's grading system)[10]. Forelimb proprioception appeared unaffected. These clinical findings strongly suggested a lesion in the thoracic spinal cord.

Diagnostic Imaging

Diagnostic imaging was challenging due to the marked ataxia of the patient. Radiographs were acquired using a computed radiography system (AGFA DX-G System, Needle-based detector plates, wall mounted camera). Lateral radiographs of the cervical, thoracic and lumbar spine in the standing patient were unremarkable.

Subsequently the horse underwent delayed phase bone scintigraphy examination of the neck and back three hours following intravenous injection of Tc^{99m}-labelled methylene diphosphonate using a MiE High Resolution Scintron Equine Scanner. There were no clinically significant findings identified.

Although there was no specific diagnosis, the gelding was subjected to euthanasia due to the severe and progressive clinical signs indicating a lesion of the thoracic spinal cord.

Post mortem examination and Imaging

A *post mortem* examination included computed tomographic (CT) imaging, detailed dissection of the thoracic spine, histopathology and microbiology (PCR).

CT data were acquired using a 16 slice helical mode scanner (16 slice GE Lightspeed Scanner). A slice thickness of 0.625mm, an X-ray tube current of 125 kV and an exposure time of 64.8 seconds was utilised and the data was reconstructed using soft tissue and bone algorithm in 1.25mm and 0.625mm slice thickness. The images revealed deviation and compression of the spinal cord to the left at the level of the $15^{th}/16^{th}$ thoracic intervertebral disc caused by a right sided, extradural, homogenous, ellipsoid soft tissue mass, with a length of 26 millimetres and width of 20 millimetres. A mild concavity of the adjacent bone appreciated, likely a result of the mass effect. There was no evidence of sclerosis of irregular lysis (Figures 1 a and b).

Dissection of the thoracic spinal cord confirmed a cystic extradural mass expanding the right side of the spinal canal displacing the cord to the left and causing moderate spinal compression (Figures 2 a and b). The cyst had a thin, partially mineralized wall and clear watery fluid content. No brood capsules or hydatid sand were detected macroscopically. Meningeal blood vessels overlying and adjacent to the lesion were congested. The cyst was closely adhered to the dura and lateral wall of the spinal canal, with a sunken region of bony remodeling in the adjacent bone. No other lesions were detected in the brain, remainder of the vertebral column, liver, kidneys, lungs, or any other internal organs.

Histopathology

The cyst had a thin inner germinal membrane, supported on a thick, laminated membrane and an outer fibrous pericyst. In the latter, there were localized areas of stromal mineralization and moderate inflammatory infiltrates, predominantly lymphocytes, with some eosino-

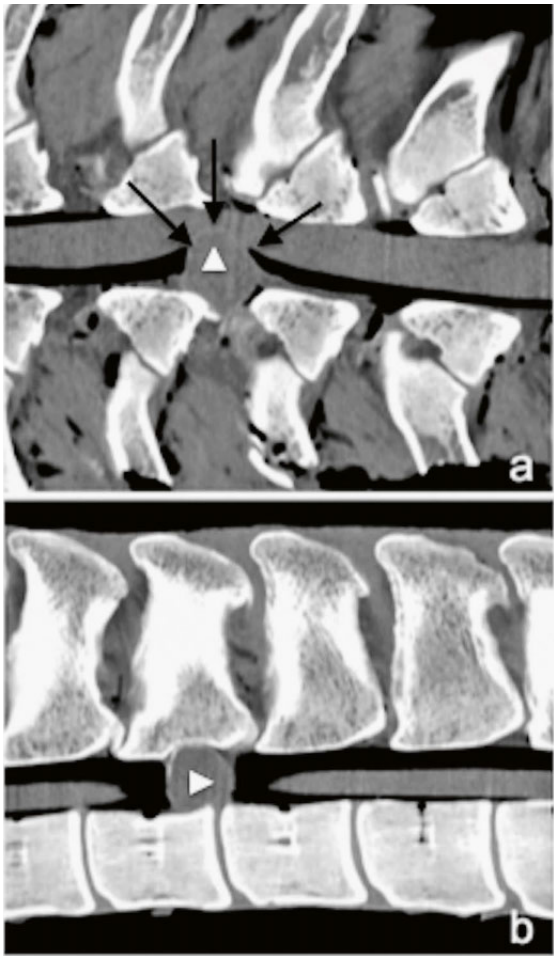

Figure 1: Dorsal (a) and sagittal (b) plane reformatted post mortem images of the thoracic spine at the level of T14-17 from a gelding presenting with hind limb ataxia and hypoalgesia. A right sided, extradural, homogenous, ellipsoid soft tissue mass (white triangle) displacing and compressing the spinal cord (black arrows) can be appreciated.

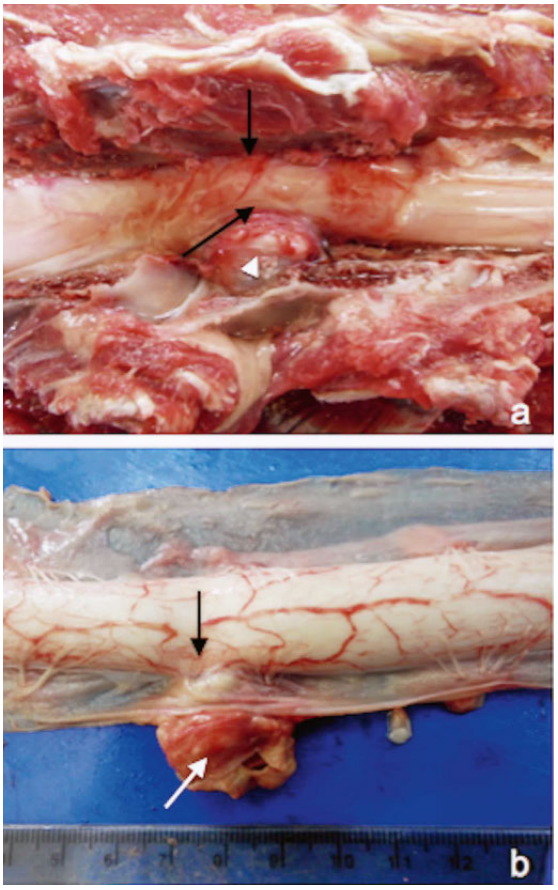

Figure 2: Post mortem dissection of the thoracic spine at the level of T14-16 from a gelding presenting with hind limb ataxia and hypoalgesia. The extradural hydatid cyst (white triangle) causing spinal cord displacement and compression (black arrow) is clearly visible from the dorsal aspect (a). Opening of the dura mater after removal of the spinal cord from the vertebral canal (b) highlights the extradural location of the hydatid cyst (white arrow), with an indentation of the spinal cord at the site of compression (black arrow).

phils and plasma cells. A single protoscolex was detected adhered to the germinal layer, with approximately 23 visible rostellar hooks (Figure 3). Sections of the spinal cord confirmed significant compression and malacia of the white matter, with frequent axonal spheroids and moderate gliosis.

Polymerase chain reaction (PCR)

E. equinus (formerly *E. granulosus*; strain G4) was identified using PCR. *E. equinus* specific primers G4F168 (5′-ATTAGTTATAGTGGTCTTTCTGTATTA-3′) and G4R426 (5′-CCATATATTAACGGAAATAACAACCAA-3′) were used to amplify a 260 base-pair long fragment of the partial region of the 471 base pair segment of the mitochondrial ND1 gene[5]. The PCR was performed in a PTC-200 gradient thermocycler (MJ Research Inc, Waltham, MA) with an initial denaturation step of 95 °C for 10 minutes, followed by 45 cycles at 95 °C for 30 s, 58 °C for 45 s and 72 °C for 60 s. The PCR

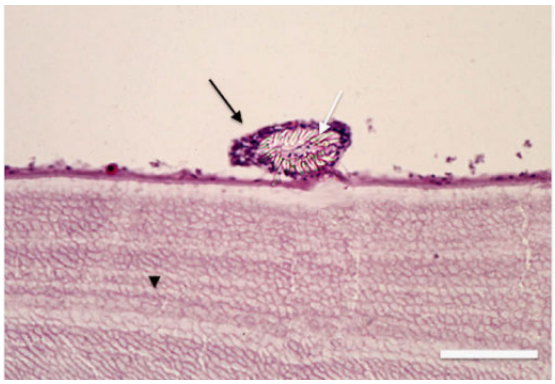

Figure 3: Histology of the wall of the hydatid cyst: a single protoscolex (black arrow) identified by PCR as *E. Equinus* was detected adhered to the germinal layer, with approximately 23 visible rostellar hooks (white arrow). The underlying acellular laminated layer of the cyst wall is visible (black triangle). Sections of the spinal cord confirmed significant compression and malacia of the white matter, with frequent axonal spheroids and moderate gliosis (not shown). Scale bar = 100µm

was terminated with a final extension step at 72 °C for 7 minutes. After amplification 10 µl of the amplification products were resolved on a 1.5% ethidium bromide stained agarose gel and the amplified DNA fragments of specific sizes were visualized by ultraviolet fluorescence. Their sizes were verified by a standard DNA ladder (Bio-RAD) run simultaneously. The expected size of the PCR products was 260 base pairs. Control DNA from the *E. equinus* reference strain (AJ508084) and a negative control were included in each reaction. To prove the specificity of our new PCR, the 260 base pair product was sequenced by a commercial company (Eurofins Genomics, Ebersberg, Germany) in both directions using the corresponding PCR primers as sequencing primers. Nucleotide sequence analysis was undertaken using the National Center for Biotechnology Information BLAST programs and database. Nucleotide sequence identity with the *E. equinus* reference strain was 100%.

Discussion

Neurological examination, diagnostic imaging pre and post mortem and pathological examination identified an extradural spinal hydatid cyst at the level of the 15th/16th intervertebral disc. Subsequent PCR identified the aetiological agent was *Echinococcus equinus*. *E. equinus* has not been reported as a cause of hindlimb ataxia in the equine patient. The basic life cycle of *Echinococcus* includes carnivores (*canidae*) as the definitive host, in which the tapeworm matures in the small intestine[16]. Domestic dogs and foxes, depending on *Echinococcus* species, are the main source of infection, providing eggs for infection of intermediate hosts, which are typically sheep, goats and cattle or rodents[16]. Eggs eaten by the suitable intermediate hosts develop into hydatid cysts, usually within the viscera[16]. Within the cyst numerous protoscolices develop, each capable of producing an adult in the final host[16]. The taxonomy of the genus *Echinococcus* is presently undergoing changes[10,19]. Currently, four species are recognized in Europe, one of which, *E. equinus*, uses dogs as a definitive host and *equidae* (horses, donkeys, mules, zebras) as an intermediate host, making *E.equinus* a highly specific parasite of Equidae[16]. More commonly known are *E. granulosus* (sheep and cattle as intermediate hosts) and *E. multilocularis* (small mammals, predominantly rodents as intermediate host). *E. granulosus* is responsible for cystic echinococcosis and is affecting approximately 2-3 million humans living in endemic areas[7]. *E. multilocularis* can result in alveolar echinococcosis, a zoonosis of public health significance due to almost 100% lethality rates in humans[7]. The level of risk for development of echinococcosis in humans caused by equine strains of the parasite is unclear, epidemiologic data suggests that the equine strain may not be infective to humans[17]. Hydatid disease in humans occurs on every continent except Antarctica[9]. Skeletal involvement occurs in 0.5-2% of all human cases, half of which are in the spine[1]. The initial treatment of choice in humans is surgical excision for neural decompression and to establish a diagnosis[1,18]. After excision local scolicidal agents like hypertonic saline or certimide is used for irrigation1. Pre-surgical use of Albendazole in *Echinococcus* infestations reduces risk of recurrence and/or facilitates surgery by reducing intra-cystic pressure[18]. The overall recurrence rate is 30-40%[18]. In Europe, echinococcosis affecting horses and donkeys has been described in Great Britain, Ireland, Belgium, Switzerland, Italy and Spain[7,8,13]. The disease is also prevalent in *equidae* in the Middle East and Africa[4]. In Ireland the prevalence in horses at slaughterhouses varies between 10% and 62%[4]. In *equidae*, cysts most commonly form in the liver and lungs[9]. Cysts typically range in diameter from 10 to 70 millimetres, in rare cases up to 200 millimetres. Clinical signs are often related to pressure on adjacent organs and tissues from the expanding cyst[4,19,20]. There are no definitive ante mortem tests for echinococcosis in horses. If the liver is affected, ultrasonographic examination may provide evidence of cystic lesions[20]. Unfortunately, due to the size of the average horse, imaging examinations of the entire spinal canal in the live animal are limited.

In this reported case, neurological signs were found in the trunk and hindlimbs with normal mentation, which lead to the conclusion that the location of the lesion must be between the second thoracic vertebra and the second sacral vertebra. The hypoalgesia of the skin noted in the mid thoracic area supported the localization of the lesion in the thoracic spinal cord as degrees of hypoalgesia and analgesia have been described caudal to the sites of severe thoracolumbar spinal cord lesions[2]. We considered trauma or space occupying lesions as possible differential diagnosis and excluded equine post anaesthetic myelopathy due to the time lapsed between the surgery and the onset of neurological signs. The lack of a history of trauma and the progressive clinical signs led us to believe that a space-occupying lesion was most likely. CT examination allowed the confirmation of a mass within the spinal canal. Unfortunately the definitive diagnosis of spinal hydatid disease in the horse is to date only possible post mortem[20].

This case demonstrates that spinal hydatid disease should be considered a possible differential diagnosis in horses with neurological deficits and no history of recent trauma. Although not currently readily available, the development of imaging modalities, which allow evaluation of the entire equine spine, will increase the likelihood that this, and other lesions of the thoracic spine can more readily be diagnosed in the future.

Acknowledgements

Sarah E. Powell for the assistance with the cross-sectional imaging acquisition and interpretation.

Kyste hydatique rachidien extradural provoquant une ataxie des membres postérieurs chez un cheval

Ce rapport décrit le cas d'un hongre croisé cob de 13 ans présenté pour l'évaluation d'une ataxie des membres postérieurs d'apparition récente. Le hongre avait subi une anesthésie générale et une ténoscopie de la gaine du tendon du fléchisseur digital du membre postérieur droit dans une clinique voisine trois mois auparavant et avait semblé normal lors des évaluations postopératoires de routine jusqu'à l'apparition soudaine de déficits neurologiques. Un traumatisme rachidien était suspecté au départ, mais la radiographie et la scintigraphie étaient sans particularité. En raison de la gravité et de la nature progressive des signes cliniques, le hongre a été euthanasié. Les examens post mortem (tomodensitométrie, dissection et histopathologie) ont révélé une compression de la moelle épinière provoquée par un unique kyste hydatique extradural (*Echinococcus equinus*), confirmé par PCR, au niveau de la 15e vertèbre thoracique. Il s'agit du premier cas rapporté d'un kyste hydatique au niveau de la colonne vertébrale causant une ataxie des membres postérieurs et doit donc être considéré comme un diagnostic différentiel potentiel de l'ataxie chez le patient équin.

Mots-clés: cheval, kyste hydatique, ataxie des membres postérieurs, moelle épinière, imagerie

Cisti idatidea extradurale spinale che ha causato atassia nell'arto posteriore di un cavallo

Questo caso clinico descrive un castrone di cob-cross di 13 anni presentato per valutare la recente atassia dell'arto posteriore. Tre mesi prima, il castrone aveva subito un'anestesia generale e una tenoscopia nell'arto posteriore destro della guaina tendinea del flessore digitale posteriore in una clinica vicina. Il decorso post-operatorio era apparso normale fino all'improvviso insorgere di deficit neurologici. Inizialmente si è sospettato un trauma spinale ma i risultati della radiografia e della scintigrafia erano irrilevanti. A causa della gravità e della natura progressiva dei segni clinici, il castrone è stato eutanasiato. Gli esami post mortem (tomografia computerizzata, dissezione e istopatologia) hanno rivelato la compressione della spina dorsale causata da un'unica cisti idatidea extradurale (*Echinococcus equinus*), confermata via PCR a livello della 15esima vertebra toracica. Questo è il primo caso di una cisti idatidea spinale che causa atassia dell'arto posteriore e quindi dovrebbe essere preso in considerazione per una potenziale diagnosi differenziale nei casi di atassia negli equini.

Parole chiave: cavallo, cisti idatidea, atassia degli arti posteriori, spina dorsale, imaging

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