Title of Paper

Atlantooccipital subluxation in an adult Thoroughbred gelding

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Summary

A thirteen-year-old gelding was referred to the University of Liverpool Equine Hospital for further investigation of ataxia and neck pain following a suspected traumatic incident in the field five days prior. The following case report documents the clinical presentation, ultrasonographic, radiographic and computed tomographic (CT) findings of a right lateral atlantooccipital (AO) subluxation. In brief, clinical presentation included abnormal head carriage, ataxia and cranial cervical swelling with associated neck pain. Radiography showed lateral deviation of the poll and subluxation of the right AO joint with significant widening of the left AO joint. CT was undertaken standing which confirmed lateral luxation of the right occipital condyle in relation to the right articular process of the AO joint such that the right articular process of the atlas was located medial to the right occipital condyle. The gelding was euthanised and post-mortem the subluxation was resolved with a closed traction procedure. This case initiates discussion of diagnosis, management and outcome for this uncommon injury. The use of CT in this case gives previously undocumented detail on the nature of the subluxation and assisted in the management and post-mortem closed reduction procedure.

Introduction

The equine atlantooccipital (AO) articulation is a paired ellipsoid joint, formed by the articular surface of the two convex condyles of the occipital bone and the corresponding two oval concave foveae of the atlas. The AO joint is stabilised by the dorsal and ventral AO membranes and the lateral AO ligaments (Gutiérrez-Crespo *et al.* 2014). The dorsal AO membrane extends from the dorsal border of the foramen magnum and occipital condyles to the cranial border of the dorsal arch of the atlas (Gutiérrez-Crespo *et al.* 2014) and is fused with the joint membrane. The dorsal AO membrane has two re-enforcing symmetric oblique long bands of fibres that cross, forming an X-shape on the sagittal plane. The ventral AO membrane extends from the ventral arc of the atlas to the ventral border of the foramen magnum and is fused with the joint capsule (Gutiérrez-Crespo *et al.* 2014). The lateral AO ligaments are two short bands that are partially blended with the joint capsules (Gutiérrez-Crespo *et al.* 2014). The lateral AO ligaments attach cranially to the base of the jugular processes and part of the paracondylar processes of the occipital bone and caudally to the craniolateral border of the dorsal arch of the atlas; these fibres are also fused with the joint membrane (Wright et al. 2018). The medial AO joint margin lies adjacent to the lateral aspect of the dura mater and spinal cord meaning distention of the AO joint can result in spinal cord compression (Wright *et al.* 2018).

Subluxation occurs when a bone is partially displaced from its articulation, resulting in a portion of its articular surface remaining in the natural cavity or upon its edge. Subluxation can be congenital or acquired. Cranial cervical congenital subluxations include occipitoatlantoaxial malformation (OAAM), atlantoaxial subluxation (AAS) and atlantoaxial instability (AI), but these conditions are uncommon and AAS and AI specifically relate to subluxation of the atlas and axis. Acquired subluxation in the cranial cervical region is also rare, traumatic in origin and usually occurs at the atlanto-axial articulation (Gerlach *et al.* 2012). AO subluxation case reports are limited and include three neonates (Griffin *et al.* 2007) and a foal with a concurrent atlantoaxial luxation (Licka 2002).

Conventional two-dimensional imaging modalities such as radiography and ultrasonography are limited in the AO region due to the complexity of the anatomy, superimposition of osseous and soft tissue structures and difficulty in obtaining orthogonal views (Gough *et al.* 2020). Ultrasonography is further limited due to acoustic shadowing making assessment of deeper structures impossible. Multi-planar reconstruction, possible with computed tomography

(CT) and magnetic resonance imaging (MRI), provides a more detailed assessment of the cervical region (Gough *et al.* 2020). CT has become the imaging modality of choice for the diagnosis of cervical vertebral pathology in the equine patient (Lindgren *et al.* 2021). In canine and human orthopaedic trauma, CT is commonly used in the diagnosis of traumatic spinal cord injury and is considered the gold standard for the investigation of acute spinal trauma (Steffen *et al*. 2003).

We document the clinical examination, radiographic, ultrasonographic and CT findings of a lateral AO subluxation in a mature Thoroughbred gelding. To our knowledge, this injury has not been reported in a mature equine before and thus, advanced diagnostic imaging of this injury is unreported in equine veterinary literature.

Case Description

Case history

A thirteen-year-old Thoroughbred gelding presented to the University of Liverpool Equine Hospital for investigation of dullness, ataxia, abnormal head carriage, neck swelling and neck pain following a suspected traumatic incident in the field five days prior. The referring veterinarian had identified low head carriage, bilateral soft tissue swelling in the poll region and a reduced lateral and dorsoventral range of motion of the neck. The gelding was treated with phenylbutazone (Equipalazone[1] 4.4mg/kg bwt IV) and dexamethasone (Duphacort Q[2], 0.1mg/kg bwt IV) initially and prescribed phenylbutazone (Equipalazone[1], 4.4mg/kg bwt PO) twice daily and prednisolone (Equipred[3], 1mg/kg bwt PO) once daily. An initial improvement in the neck swelling and comfort of the horse was noted but after four days, when the prednisolone dose was tapered (Equipred[3], 0.5mg/kg bwt PO), the gelding appeared more painful and was unable to elevate his head, prompting referral.

Clinical Findings

On presentation, the gelding was quiet, but alert and responsive and all vital parameters were within normal limits. At rest, the horse stood with a low head carriage equally weight bearing on all four limbs. A left-sided soft swelling (~5cm x 8cm) was palpable dorsal to the vertebral column in the poll region with no associated heat, and an associated asymmetry of the cranial cervical region was observed when viewing the neck from dorsal (Figure 1a). A mild pain response was elicited on palpation of the cervical vertebral column. The left wing of the atlas was more prominent than the right wing with deviation of the head to the left of midline. This resulted in a palpable concavity on the right-hand side of the cranial cervical spine and a convexity of the left-hand side of the cranial cervical spine (Figure 1b).

A targeted cranial nerve exam was considered largely unremarkable with pupillary light, dazzle and facial sensation reflexes all within normal limits. Menace and palpebral reflexes on the left side elicited a mild hyperreactive and myokymia response of the eyelid. Voluntary movement, range of motion and flexion of the neck was assessed by tempting the horse to prehend carrots in different directions. Cervical range of motion and left lateroflexion was good, while range of motion and right lateroflexion was poor; this could be improved with gentle pressure. Dorsoventral flexion and extension were markedly reduced.

The horse was comfortable at walk and able to walk in a serpentine pattern without any lameness. Only very subtle intermittent proprioceptive deficits of the fore and hind limbs were apparent when the horse was walked in a straight line. When walked in tight circles to the right, the gelding pivoted on the front feet and demonstrated a reluctance to cross the hindlimbs but displayed no evidence of interference. When walked in a tight circle to the left, the same signs of ataxia were noted but more pronounced. The horse backed up normally and was able to walk up a gradual incline with a normal gait. When the head was held in an elevated position at rest the horse showed discomfort including a wide-base stance and reluctance to go forward; at walk the horse showed an increased ataxic and hypermetric gait. The horse's ataxia was graded two out of five on the Modified Mayhew System.

Initial Management

An intravenous catheter was placed in the left jugular vein and phenylbutazone (Equipalazone[1], 4.4 mg/kg bwt IV) and paracetamol (Paracetamol[4], 20mg/kg bwt PO) were administered twice daily. The gelding was confined to a stable and carefully monitored.

Diagnostic Imaging

Radiography

Radiographs were obtained the day after presentation. The gelding was sedated with detomidine hydrochloride (Detonervin[5], 0.1 mg/kg bwt IV) and butorphanol (Torphasol[5], 0.1 mg/kg bwt IV). Standing laterolateral, lateral-oblique and ventrodorsal radiographs of the head and cervical spine were obtained using settings of 88kV and 20mAs. The laterolateral radiograph showed asymmetry of the left and right side of the atlas with one side being dorsally displaced relatively to the occipital condyle. Additionally, there was widening of the AO joint space on one side suggestive of subluxation (Figure 2a). The ventrodorsal radiograph showed left lateral deviation of the atlas with asymmetric AO joint spaces (left versus right) and marked widening of the left AO joint consistent with subluxation of the left and right AO joints. The right articular process of the atlas was displaced medially to the right occipital condyle and the left articular process of the atlas was positioned craniolaterally to the left occipital condyle (Figure 2b).

CT

CT examination of the cranial cervical spine and head was performed under standing sedation, with further sedation of acepromazine (Tranquinervin[1], 0.03mg/kg bwt) and morphine sulphate (Morphine Sulphate[7], 0.1mg/kg). CT images were obtained using a 16 slice, 90 cm bore CT scanner **(**Canon Aquillion Prime 160[8]), mounted on a sliding gantry system. Images were acquired using 16 row × 1.0 mm detector width, 1.0mm slice thickness, 550 mm FOV, tube rotation time 0.75s, collimator pitch 0.688, 120 KVp and 300 mAs. Bone and soft tissue reconstructions were performed. Images were viewed on a computer monitor, using proprietary DICOM software (HOROSTM, GNU Lesser General Public License, Version 3.0, LGPL 3.0) in single and multiplanar views using multiplanar reconstruction.

CT identified left lateral deviation of the atlas with asymmetric AO joint spaces, marked widening of the left AO joint and the right cranial articular process of the atlas was located medial to the right occipital condyle, within the right side of the foramen magnum (Figure 3a and 3b). The left cranial articular process of the atlas was craniolaterally displaced resulting in a significant widening of the left AO joint (Figure 3b and 4a). Despite the abnormal bone placement there was minimal compression of the dura at the foramen magnum, however at the level of the atlas there was significant extra-dural compression resulting in right sided displacement of the spinal cord. There was marked soft tissue swelling, dorsal to the left occipital condyle. The soft tissue swelling in the left side of the vertebral canal at the level of the AO joint created a mass effect on the spinal cord resulting in deviation of the spinal cord to the right (Figure 4b). Heterogeneous soft tissue attenuation filled the gap between the left articular process of the atlas and the left occipital condyle, consistent with organising haematoma and fluid. There was heterogeneous soft tissue attenuating material within the expected area of the AO joint capsule, consistent with haemorrhage and likely rupture of the joint capsule (Figure 4a). In addition to this, several osseous fragments, approximately 3 x 4 mm were identified ventral and medial to the right occipital condyle and a smaller 2 x 3 mm osseous fragment was located extra-durally within the vertebral canal at the level of the AO joint (Figure 3b). The CT confirmed AO subluxation with fragmentation and extradural compression.

Ultrasonography

Ultrasonographic examination (Logic S7 Expert[9]) was performed following CT to assess this diagnostic imaging modality as a tool for ongoing assessment of subluxation and soft tissue trauma. This identified marked soft tissue enlargement at the level of the left AO joint and a loss of the normal relationship between the cranial fovea of the atlas and caudal surface of the occipital condyles on both sides of the neck. From a right dorsal ultrasonographic window the bone contour of the right atlas could be appreciated located in an abnormally medial position to the bone surface of the right occipital condyle. This procedure was well tolerated with sedation alone.

Outcome

These findings were discussed with the owners; conservative management was considered inappropriate in this case and so closed reduction was offered. The owners opted for euthanasia with no gross post-mortem performed. The horse was humanely euthanised with intravenous injection of secobarbital sodium (400mg/ml) and cinchocaine hydrochloride (25mg/ml) (Somulose[1]). Immediately after euthanasia, the horse was positioned in right lateral recumbency, and a 30 x 30 x 4 cm thick wooden block was placed under the horse's head to elevate the head, with the caudal margin of the block aligned to the caudal ramus of the mandible. A head collar was fitted tightly to the horse's head and the head was fully extended. A single person applied manual traction to the head in a cranial direction whilst a board-certified surgeon simultaneously placed a hand on the lateral aspect of the left atlantal wing and applied downward pressure in a short pulsing movement. A firm clunking noise was audible as the AO joint was successfully reduced. A ventrodorsal radiograph confirmed reduction.

Discussion

This case adds detail to the clinical presentation and diagnostic imaging of the rare condition of traumatic AO subluxation in an adult equid. Whilst there are no reports of traumatic AO subluxation in the adult horse, neonate and foal luxations have been recorded; Licka (2002) reports a case of traumatic AO luxation and atlantoaxial subluxation in a three-month-old Warmblood colt and Griffin *et al.* (2007) reports a short case series of three neonates with congenital AO luxation. Traumatic luxation elsewhere in the cervical region in the adult horse has been reported, including a traumatic atlantoaxial luxation in a mature 500kg Warmblood mare (Gerlach *et al.* 2012). Based on these cases (Gerlach *et al*. 2012; Licka 2002), closed reduction was offered as a viable treatment option in this case and performed post mortem to determine the feasibility of the procedure in a mature equid. The procedure was conducted as similarly described by Gerlach *et al.* (2012) with the horse in lateral recumbency and traction applied to the head by an assistant. Reduction was first attempted with the head in a flexed position but this was not possible. Manual traction with additional laterolateral force resulted in reduction of the subluxation with far less traction force required than if machine traction were to have been utilised. This is an important consideration as the strain exerted upon the spinal cord during manipulation of the foal was described as considerable and causative of the ataxia seen immediately after reduction (Licka 2002).

Although closed reduction is considered a non-invasive or conservative treatment for AO subluxation it should be strongly emphasised that any manipulation of the AO joint can result in deteriorating neurological status and even death (Griffin *et al.* 2007). The dorsal AO membrane is interwoven with large collagen bundles, and this forms the middle contact of the muscle-membrane-spinal dura mater connection (myodural bridging) linking the suboccipital musculature to the dura mater. We hypothesise that stretching of this membrane and myodural bridge either from the initial trauma or during traction applied in a close reduction procedure may result in dura mater and even spinal cord trauma. As the closed reduction in the present case was attempted, and achieved, after euthanasia it is not possible to predict what effect the closed reduction procedure would have had on this patient.

In this case the initial radiographs identified the AO subluxation, but CT provided useful additional information including spinal cord compression, soft tissue swelling within and outside the vertebral canal, deviation of the spinal cord at the level of the atlas as a result of the mass effect of soft tissue swelling consistent with haemorrhage and several separate osseous fragments within the vertebral canal at the level of the AO joint. Advanced cross-sectional imaging techniques, such as MRI and CT, have been shown to improve ante-mortem diagnosis of cervical pathology in equine patients (Griffin *et al.* 2007; Gutiérrez-Crespo *et al.* 2014; Lindgren *et al.* 2021). Additionally, advanced cross-sectional imaging techniques has been shown to improve ante-mortem diagnosis of AO joint pathology and treatment selection (Steffen *et al.* 2003).

Although myelography would have provided further information relating to the spinal cord, the procedure is contra-indicated in this case due to potential increased intracranial pressure (ICP). AO subluxation has the potential to obstruct cerebrospinal fluid (CSF) drainage from the cranium resulting in increased ICP. Whilst determining increased ICP in horses is challenging the altered mental status of our patient and the type of injury was enough that we considered a myelogram potentially fatal. If a myelogram is performed in a patient with ICP it is possible during the aspiration of CSF for brain tissue to herniate through the foramen magnum and for a Cushin-type reflex to be inducted which can lead to asystole (Bennell and Bardell 2021).

Conclusion

AO subluxation is a rare condition and CT imaging provided clinically relevant and useful information not ascertainable from conventional two-dimensional imaging. Extrapolating from the veterinary literature and the successful post mortem reduction achieved in this case, closed reduction may be considered as treatment for AO luxation in the mature equine population but with all risks considered and communicated. The method of closed reduction described in this report shows a practical and feasible means of achieving reduction which could be trialled after appropriate case selection.

Manufacturers' addresses

[1]Dechra Pharmaceuticals plc, Shrewsbury, UK
[2]Zoetis UK Ltd, Surrey, UK
[3]Virbac Ltd, Suffolk, UK
[4]Milpharm Ltd, Ruislip, UK
[5]Animalcare Ltd, York, UK
[6]DMS Imaging, Gallargues-le-Monteux, France
[7]Martindale Pharmaceuticals, Romford, UK

[8]Canon Medical Systems Ltd, West Sussex, UK
[9]GE Ultrasound Korea Ltd, Gyeonggi-do, South Korea

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Figure Legends

Previously 1

Figure 1a) Image taken from craniodorsal showing a left-sided swelling in the cranial cervical region (arrowed) and an asymmetry of the cranial cervical region

Previously 2

Figure 1b) Photograph of the cranial cervical region taken from dorsal to demonstrate the asymmetry in the region, the bottom of the image is the caudal aspect of the neck, and the convexity on the left side of the cranial cervical spine can be seen (arrowed).

Previously 3

Figure 2a) Laterolateral radiograph of the cranial cervical region showing mild obliquity of the atlas with asymmetric atlantooccipital joint spaces (arrowed). Cranial is to the left of the image.

Previously 4

Figure 2b) Ventrodorsal radiograph of the cranial cervical region showing left lateral deviation of the atlas with asymmetric atlantooccipital (AO) joint spaces (left versus right) and marked widening of the left AO joint (double headed arrow) The right cranial articular process of the atlas (arrow head) is displaced medial to right occipital condyle (starred), consistent with subluxation of the left and right AO joint. The patients left is displayed to the right of the image.

Previously 5

Figure 3a; 3D volumetric reconstruction of a computed tomographic study viewed from dorsal, demonstrating the left cranial articular process of the atlas (arrowed) displaced craniolaterally and the right cranial articular process of the atlas (arrow head) located medial to the right occipital condyle (starred), within the foramen magnum (circled), consistent with a lateral atlantooccipital joint subluxation. The patients left is displayed to the left of the image.

Previously 6

Figure 3b; Dorsal multiplanar computed tomographic image reconstructed with a standard bone algorithm, displayed in a bone window (window level 350; window width 1500) at the level of the atlantooccipital joint demonstrating the right cranial articular process of the atlas (arrow head) abnormally positioned within the right side of the foramen magnum (circled). A displaced separate bone fragment (arrowed) is evident medial to the left occipital condyle. The patients left is displayed to the right of the image.

Previously 7

Figure 4a) Transverse multiplanar computed tomographic image reconstructed with a smooth soft tissue algorithm and displayed in soft tissue window (window length 350; window width 35) at the level of the atlantooccipital (AO) joint. Only the left AO joint is evident due to the marked asymmetry and rotation of the atlas and the left AO joint is markedly enlarged with loss of congruency (arrowed). There is marked heterogeneously attenuating material within the area of the joint capsule, consistent with haemorrhage and suspected rupture of the joint capsule (circled). The patients left is displayed to the right of the image.

Figure 4b) Transverse multiplanar computed tomography image reconstructed with a smooth soft tissue algorithm and displayed in soft tissue window (window length 350; window width 35) at the level of the atlas. The left occipital condyle (blue circle) is abnormally positioned and evident caudal to its expected position and dorsal to the body of the atlas in the vertebral canal. There is marked soft tissue swelling consistent with haemorrhage, dorsal to the abnormally positioned left occipital condyle (red arrows). The abnormal soft tissue swelling in *the left side of the vertebral canal is creating a mass effect on the spinal cord (white star), which it deviated to the right. The patients left is displayed to the right of the image.*