Upper Oesophageal Sphincter Aplasia in Equids: Observations in Preoperative Patients and a Possible Association with some Cases of Dorsal Pharyngeal Collapse and Paradoxical Sleep Deprivation

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Abstract: Upper oesophageal sphincter aplasia and subsequent incompetence had been reported as one element of the fourth branchial arch defect syndrome and also as a possible sequelae to the placement of a laryngeal abductor prosthesis. Fourth branchial arch defects were thought to be uncommon and when diagnosed it was more often the cartilaginous abnormalities with associated arytenoid abductor incompetence that were discussed. As a consequence of introducing manual assessments of upper oesophageal sphincter tone to the oropharyngeal examination performed prior to palatoplasty procedures, it was determined that this presentation was more common than previously reported. In addition and as a result of pre and post operative examinations and history collection there appeared to be a possible association in some instances with cases of dorsal pharyngeal collapse and paradoxical sleep deprivation.

Keywords: Horse, sleep disorders, upper airway, aerophagia, hiccup.

INTRODUCTION

The upper oesophageal sphincter (UOS) is formed by elements of the crico and thyro-pharyngeus muscles [1,2]. Its purpose is to prevent food and water being returned to the nasopharynx during the process of deglutition. UOS aplasia and incompetence had been reported in five horses following prosthetic laryngoplasty [3]. The suspected etiology in these cases was intraoperative trauma to either the musculature or to the neural supply [3]. Chronic coughing and upper tracheal contamination were common presenting complaints.

In cases of aplasia, which were one element of fourth brachial arch defects (4BAD), associated reflux could precipitate airways contamination with horses presenting with histories of repeated airways infections [4-7]. Aerophagia, as a consequence of UOS aplasia in 4BAD horses, could in some cases culminate in instances of tympanitic colic [2,5].

Most cases of 4BAD were diagnosed when either rostral displacement of the palatopharyngeal arch (RDPA) [8,9] was evident on endoscopy or when laryngeal abductor dysfunction along with palpable abnormalities of the cricothyroid articulation were detected [2,4,5]. Cases of right sided abductor dysfunction were frequently diagnosed as being part of 4BAD condition [4,5].

It was uncommon to see reports where the only recorded abnormality was that of UOS hypoplasia or aplasia. This segment of the 4BAD complex was normally diagnosed by either utilising radiography to detect the presence of air in the proximal oesophagus [4] or with ultrasonography of the larynx which included the cricopharynx [10]. There were no reports in the literature of manual assessments of UOS integrity.

Dorsal pharyngeal wall collapse, which could occur in association with lateral pharyngeal wall collapse or independently, was regarded as part of the condition referred to as dynamic pharyngeal collapse (PC) [11-13]. Causative theories centered around the function and thence potential dysfunction of the stylopharyngeus muscle [14,15].

Paradoxical sleep deprivation and with this a number of possible sequelae including sleep crashing had drawn interest from researchers in recent times [16-18]. Many cases of recumbent sleep deprivation were successfully managed with changes in the horses environment, choice of guard or companion or with pain management [16,17]. There were also two case in which the horses returned to normal recumbent sleep following upper airways surgery to reduce the impact of palatal instability on nasopharyngeal airway competence [19,20].

MATERIALS AND METHOD

A total number of 300 horses which presented for surgery in an attempt to reduce the incidence of palatal instability (PI) [21,22] with or without progression to dorsal displacement of the soft palate (DDSP) [23-25] were used in this study. All horses underwent a modified oral palato-pharyngoplasty (OPP) [26,27].

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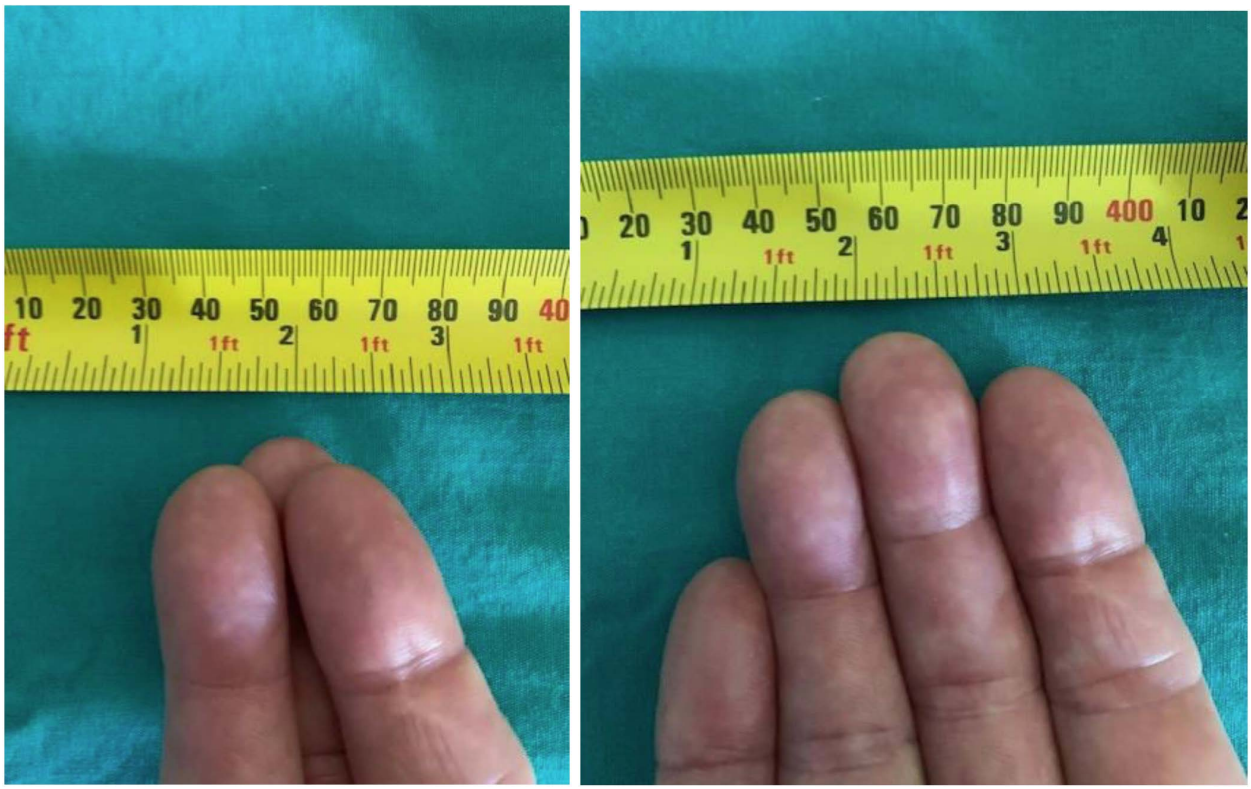
E-ISSN: 2310-0796/21



A **B**

Figure 1: A. Three digit test for UOS tone. **B**. Grade 4 - four digit linear.

Assessments of UOS constrictor tone were made manually by the same operator following the induction of anaesthesia. The same induction protocol was used in all cases and examinations were made within 3 to 4 minutes of the horse attaining recumbency. Initially three fingers (except thumb and little finger) were inserted into the oesophagus in a triangular formation (see Figure 1). The most common, and what was then regarded as a normal response, was when the fingers were firstly compressed and then held tight. This was graded as 0. Grades from 1 to 5 reflected progressive lessening in this response (see Figure **2A**). For grades 2 to 5 the fingers were held in a linear arrangement (see Figure **1A**). Grade 5 had been previously recorded but not in this study. In these cases the operators five fingers were inserted in a linear arrangement with next to no contractile response. These horses presented with chronic reflux, nasal return of mucus and green feed and with repeated airways infections.

The incidence and degree of UOS dysfunction along with age, gender and breed were recorded (see Figures **2** and **3**). Only grades 2-3 and above were included in the study in an attempt to remove any possible variation in contractile response to anaesthesia. There were however a number of grade 1-2 cases where clinical histories strongly suggested the presence of UOS incompetence.

A.

Grade:
1 - reduced tone, three fingers triangular
2 - reduced tone, three fingers linear
3 - reduced tone, four fingers linear
4 - poor tone, four fingers linear
5 - very poor tone , five fingers

B.

Grade:	No. horses:
1-2	(not included)
2-3	27 (58%)
3	10 (22%)
3-4	6 (13%)
4	3 (7%)
5	0

Figure 2: A. Grading protocol: Degrees of reduced contractile tone (palpation).

B. Incidence of varying grades of reduced contractile tone.

Any elements of the presenting histories that were suggestive of UOS incompetence and or the presence

	Case No. (Population %)		UOS aplasia No. (% 0f same breed/age/gender)	
Breed:				
Thoroughbred -	284	(94.7%)	37	(13.0%)
Standardbred -	11	(3.6%)	6	(54.5 %)
Warmblood -	5	(1.7%)	3	(60.0 %)
TOTAL	300		46	(15.3%)
Age:				
2yr	84	(28.0%)	10	(12.0 %)
3yr	116	(38.5%)	17	(14.5%)
4yr	50	(16.6%)	7	(14.0 %)
5yr	27	(9.0%)	5	(18.5%)
6yr	9	(3.0%)	4	(44.5%)
7-11yrs	14	(4.6%)	3	(21.5%)
Gender:				
Fillies	113	(37.6%)	18	(15.9%)
Geldings	179	(59.6%)	27	(15.0%)
Colts	8	(2.8%)	1	(12.5%)

Figure 3: Horses presented for surgery from August 2015 to December 2020. Breed and age distribution

of 4BAD were recorded along with any relevant findings from endoscopic examinations.

RESULTS

Of the 300 horses included in this study, 284 (94.7%) were Thoroughbreds (TB), 11(3.7%) Standardbreds (STB) and 5 (1.6%) Warmbloods (WB).

The grading system used in assessing UOS tone or contractility was developed by the author (see Figure **1A**). Forty-six horses (15.3%) were graded greater than 1-2 (see Figure **3**).

The incidence in TB's at 13% was probably a truer reflection of the incidence of this condition in this surgical population. The total population figure of 15.3% was heavily influenced by a small number of STB and WB cases. There appeared to be no gender bias and little age influence except where case numbers were limited (5 -11yrs).

Twenty-seven (58%) horses were assessed with moderate loss of tone (grades 2 -3) , whilst 19 (42%) were more severe. The numbers of horses declined with increasing severity of UOS incompetence.

Of the 46 cases graded 2-3 and above, none were recorded as having (RDPA) [7-9] and only one had significant laryngeal abductor dysfunction which was determined to be due to an atrophic thyro-cricoid articulation (left side) [4-7]. Bilateral dorsal pharyngeal collapse was evident in three horses, whilst mucus with feed material in the upper trachea was also present. 1 horse had unilateral collapse.Ten horses had inflammation of the trachea and bronchi with heavy mucus loads and feed material present within the mucus.

In the process of procuring preoperative 'histories' a number of potentially significant symptoms were recorded. Some of these had been previously reported as occurring in horses subsequently diagnosed with 4BAD's whilst others related more to the horses general behaviour or demure and also their recumbent sleep habits. As not all horses were investigated preoperatively with a view to assessing these factors, their frequencies are therefore unlikely to be a reflection of their true incidence.

Stable or Yard

Apparent absence of recumbent sleep -7, Cribbing or crib biting - 5 horses, Mucus/saliva film on drinking water -2. Green mucus in nostrils -2. Chronic cough -5, Sleeping in sternal recumbency only -2, Long periods of recumbency (2-3 hours) often late AM - 2, Excitement followed by colic (one stallion when calling to a mare would ingest enough air to initiate colic) - 2, History of repeated episodes of tympanitic colic - 2, and noises similar to hiccups - 1.

Behaviour

Somnolent - 5, Somnolent to hyper-reactive - 6 [19,20],

Race/Event and Training Related

Extremely loud inspiratory noises - 8, Extended recovery (up to 30 minutes) - 8, Flatulence following exercise - 6, Nasal return of water during mouth wash -

4. Chronic cough in work - 7. Frequent airways infections - 4. Curious noises described as trumpet - 3 or flute like - 1. White foamy discharge after work - 3.

There were also peri-operative observations that may have been related to this condition. On induction, as the horses head moved ventrally with sedation, a continuous white mucoid discharge became apparent in one nostril - 3. Long periods of recumbency/recovery post anaesthesia (1 to 2 hours) - 7. Saliva from laryngotomy -2. Copious flatulence on recovery - 5. Chronic cough (normally only occasional coughing post operatively) - 3.

Fourth branchial arch defects in full siblings had been previously reported [28]. In this study two horses had two half siblings, that had previously been recorded as having significant UOS incompetence. Another had two full siblings.

DISCUSSION

It had been reported that approximately two in a thousand Thoroughbreds were born with a 4BAD [2]. Aides to diagnosis included palpation [5,2], ultrasonography [10,29], radiography [29], magnetic resonance imaging (MRI) [30-32], computed tomography (CT) [31,33] and endoscopy. The more common indications that would trigger an investigation into the possibility of a 4BAD being present were endoscopic evidence of RDPA [7-9], oesophageal exudate [3], right sided laryngeal abductor dysfunction [2,6] or a history of tympanitic colic. Others often only came to light when surgical attempts to abduct the corniculate process, using a laryngoplasty, failed.

In this study approximately one in seven horses that presented for palatoplasty surgery had palpable reductions in UOS contractile response. Given that DDSP has a reported incidence of 10-20% in TB's, the results of this study would suggest a higher incidence of 4BAD in this breed [2]. Interestingly none of the 46 horses presented with RDPA and only one with a left sided abductor deficit that was a consequence of an atrophic crico-thyroid articulation. In addition only two horses had a history of tympanitic colic whilst five others had a white foamy or green tinged nasal discharge suggestive of oesophageal reflux [4,5].

There were four horses with dorsal pharyngeal collapse (one unilateral and three bilateral) evident on endoscopy. A previously reported 4BAD case in a pony presented with gross enlargement of the right crico-thyroid notch, a resting right sided abductor dysfunction and dorsal pharyngeal collapse during exercise [37]. Interestingly the right corniculate cartilage was seen to fully abduct during an exercise test whilst the dorsal pharynx collapsed. This suggested that a reported adventitious noise was likely being created by the pharyngeal collapse rather than the 4BAD [37].

It was postulated that if dilation of the proximal oesophagus was to occur during exercise then the dorsal excursion of the roof of the oesophagus would apply a positive pressure to the caudal floor of the guttural pouch. This compression would then create a positive pressure within the pouch which could then result in the dorsal wall of the nasopharynx moving ventrally. Indeed, ventral collapse of the nasopharynx along with dorsal movement of the soft palate and epiglottis along with dilation of the proximal oesophagus were already recognised as being integral contributors to the pharyngeal 'stripping wave' [2]. As much as the action of the stylo-pharyngeus muscle and any possible interference to its neural supply had been postulated as an aetiology for this form of pharyngeal collapse, to date there appeared to be no evidence to support this theory [11,13-15].

In two previously published case studies of horses that had issues with achieving adequate paradoxical sleep and that subsequently underwent palatoplastys, UOS constrictor grades of 2-3 had been recorded [19,20]. At the time these findings were not thought to be relevant and thus were not mentioned in the publications.

In this study 11 (24%) of the 46 (Graded 2-3 and above) cases presented with histories of somnolence [16]. In 9 of these cases owners reported that they had never observed the animals in lateral recumbency. The other 2 horses were often seen in extended (several hours) periods of recumbency. However in no cases was 24 hour monitoring carried out so paradoxical sleep deprivation could not be confirmed.Theoretically during periods of recumbent sleep reduced muscle tone along with UOS weakness could trigger proximal oesophagus dilation. As previously suggested a resultant positive pressure created within the guttural pouch could then contribute to the ventral movement of the dorsal pharyngeal wall. If PI with its dorsal movement of the soft palate or DDSP were to occur along with this ventral excursion of the roof of the nasopharynx, an airway obstruction similar to that described in human cases of obstructive sleep apnea (OSA) could ensue [38].

In the present study many of the theories presented lack sufficient evidence other than that of an anecdotal nature to support them. Further rigorous investigations would be required to either support or refute these theories. However from a practical viewpoint it would be useful if practitioners when encountering cases of dorsal pharyngeal collapse, other than those associated with pole flexion, or recumbent sleep deprivation where aetiologies were unknown, could include an assessment of UOS structure and competence in their clinical workups.

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Received on 15-01-2021 Accepted on 27-02-2021 Published on 03-03-2021

https://doi.org/10.12970/2310-0796.2021.09.03