



Case Report

Headshaking associated with a unique presentation of guttural pouches infection and otitis media in a Quarter Horse stallion: diagnosis, medical treatment and outcome

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ABSTRACT

A 3-year-old Quarter Horse stallion was referred for headshaking (HS) syndrome of one month duration. The horse underwent complete HS workup. Physical examination revealed up-and-down head movements associated with nose snorting during lunging and riding, in the absence of other abnormalities at rest or in motion. Lameness, neurologic and behavioural evaluations were unremarkable. Endoscopic examination of the upper respiratory tract showed bilateral guttural pouches (GPs) inflammation. The mucosal surface was altered by the presence of areas of microbullous appearance coalescing into some bullae of larger diameter. A bacterial culture obtained by flushing, brushing and biopsy of the GP mucosa was positive for *Serratia marcescens*. Computed tomographic evaluation was indicative of: moderate bilateral GP inflammation, bilateral otitis media, and nuchal ligament enthesopathy, with uncertain significance. The horse was treated with antimicrobials according to the sensitivity test, systemic NSAIDs and rest. At the one-month follow-up, HS condition was improved and the endoscopic examination showed a limited number of bullous lesions remaining. Bacterial culture re-obtained from the GPs lavages was positive for *Streptococcus equi zooepidemicus*. Based on the residual evidence of GP infection, the horse was re-treated with antimicrobials according to the sensitivity test, systemic NSAIDs and rest. At the two-month follow-up, HS condition was considered resolved based on clinical evaluation, the horse resumed gradually training with satisfactory results. Endoscopy and bacteriology showed resolution of GP infection. At one-year follow-up no relapses were reported.

1. Introduction

Sporadic shaking of the head can be normal in horses as an avoidance manifestation, a poor rider technique or a behavioural issue [1,2]. On the other hand, headshaking (HS) syndrome is defined as a repeated, often involuntary, movement of the head, and sometimes neck, occurring predominantly in the vertical plane, but also horizontally or rotationally [3]. This display is often associated with signs of nasal irritation, such as twitching and rubbing, and can occur both during exercise and at rest [4,2]. HS can originate from multiple abnormalities: functional causes such as trigeminal nerve-mediated pain (previously referred as idiopathic HS), and anatomical ones, which include temporohyoid osteoarthropathy (THO), fracture of the paracondylar process of the occipital bone, sinusitis, tooth root infections, affections of the ear canal,

ocular diseases, cervical injury [1,5,6,2,7]. Due to the contiguity of the guttural pouches (GPs) with the cranial nerves, also diseases affecting these anatomical structures, especially mycosis, have also been reported as possible cause of HS in horses [8]. In addition, given the vicinity of the various structures of the head that can be affected by pathology that causes HS, simultaneous involvement of more than one is not uncommon. For example, chronic otitis media with bony inflammation of the bulla often leads to secondary THO, making it tricky to discriminate the source of the clinical sign [5].

Because of the wide variety of conditions that can cause HS, the diagnostic work-up is particularly challenging and often requires advanced imaging techniques [9,3].

The present report describes an atypical presentation of guttural pouches infection and otitis media in a Quarter Horse stallion as the

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putative causes of HS.

2. Case description

The owner provided informed consent, as part of the veterinary hospital admission consent form, for the use of the horse's data in the study.

2.1. History

A 3-year-old Quarter Horse stallion was referred to the Veterinary Teaching Hospital (VTH) of the Department of Veterinary Medical Sciences of the University of Bologna, Italy due to subacute onset of HS, about a month before referral. The horse was in active training and performed in its first reining competition a month prior to the onset of clinical signs. The owner reported a progressive onset of up-and-down head movements which manifested both during lunging and riding, emphasized during collection request, and associated with snorting. No other abnormalities were described and no treatment was attempted prior to hospitalization. Nevertheless, the owner reported the occurrence of a strangles outbreak at the stable about three months before, however the horse remained fully asymptomatic.

2.2. Clinical findings and diagnosis

At presentation, clinical examination was normal, except for a mild reaction to palpation of the poll region. Both neurological and lameness examinations were performed, both at rest and during lunging, which showed no signs of obvious lameness nor other neurologic signs, but the presence of a vertical HS and snorting consistent with a grade 1/3 according to the 3-point scale proposed by Roberts and colleagues [10]. Ophthalmic, oral and behavioral examinations were unremarkable. Hematochemistry evaluation showed a leucocyte count at the upper normal limits for the horse's age with normal inflammatory markers fibrinogen and Serum Amyloid A. Radiographs of skull and neck resulted inconclusive for a temporohyoid osteoarthropathy. Otoscopic and endoscopic examinations of the ears showed a moderate amount of keratin scales around the entrance to the bony part of both external ear canals, which did not make the tympanic membrane clearly visible.

Endoscopy of the upper respiratory tract was normal, whereas the GPs presented several abnormalities: the mucous membrane was thickened and presented multiple bullous-like lesions covering the temporohyoid joint (THJ) and the stylohyoid in both pouches (Fig. 1), and also extending to the surface of medial and lateral compartments in the left GP (Fig. 2). The content of the intramural bullae was aspirated, and consisted of air and mucus. Due to the mucosal abnormality, a slight thickening of the styloid could not be ruled out. Flushing, brushing and biopsy samples were collected from both GPs for bacterial culture, which resulted positive for a pure growth of *Serratia marcescens*, whereas fungal culture was negative. A quantitative PCR for *Streptococcus equi equi* yielded a positive result.

A CT evaluation of the head performed under general anesthesia revealed: moderate bilateral pouch mucosal thickening and fluid accumulation, consistent with the clinically reported GP infection; bilateral soft tissue accumulation within the tympanic bulla, consistent with otitis media; and nuchal ligament enthesopathy, with uncertain significance.

Based on the findings, a diagnosis of bullous GP infection, otitis media and nuchal ligament enthesopathy was reached, while a temporohyoid osteoarthropathy was ruled out.

2.3. Treatment and outcome

Once clinical investigations were concluded, the horse was discharged from the VTH and therapies were administered at the stable. Based on the isolation of *Serratia marcescens* and sensitivity test, the horse was treated with oral trimethoprim potentiated sulfadiazine at a dose of 30 mg/kg BWT twice daily (Equibactin, Dechra Veterinary Products SRL) and oral firocoxib at a dose of 0.1 mg/kg BWT once daily (Previcox, Boehringer Ingelheim Animal Health Italia S.p.A.) for seven days, and rest.

At the one-month follow-up, the HS condition was gradually improving and the endoscopic examination showed only bullous lesions of a larger diameter and limited number (3 to 5) remained over the temporohyoid joint of the left GP, and scarce traces of mucus were present on both GPs. Bacterial culture re-obtained from the GPs lavages was positive for a pure growth of *Streptococcus equi zooepidemicus*, whereas PCR for *Streptococcus equi equi* was negative. Based on the residual evidence of GP infection, the horse was treated again with

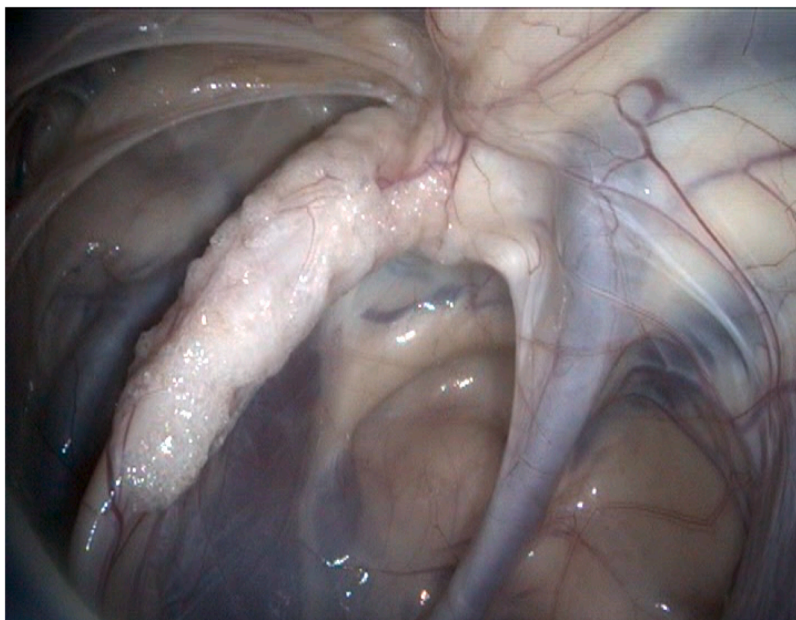


Fig. 1. Endoscopic image from the right guttural pouch. Intramural bullous appearance of the mucosal surface extending from the temporohyoid joint to the stylohyoid bone.

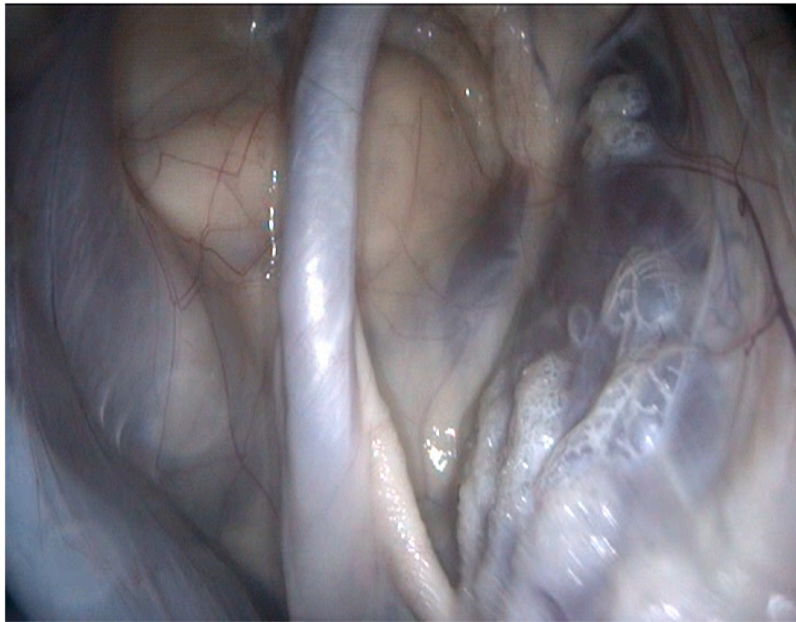


Fig. 2. Endoscopic image from the left guttural pouch. Intramural bullous appearance of the mucosal surface extending to the lateral and medial walls of the pouch.

antimicrobials for 4 weeks according to the sensitivity test: intramuscular procaine penicillin at a dose of 22 mg/kg BWT twice daily (Depocillina, MSD Animal Health SRL) for seven days and oral trimethoprim potentiated sulfadiazine at a dose of 30 mg/kg BWT twice daily for the remaining three weeks, oral firocoxib at a dose of 0.1 mg/kg BWT once daily was administered for 15 days and light exercise recommended until the next check-up.

At the two-month follow-up, HS condition was considered resolved based on clinical evaluation, the horse resumed gradually training with satisfactory results. Endoscopy and bacteriology showed no residual signs of GP inflammation and culture was negative.

The horse engaged in regular competitive activity, and no relapse was reported at one year follow-up.

3. Discussion

GPs pathology has been previously reported as a cause of HS. While mycosis cases have been mainly mentioned, bacterial infections have been rarely reported, and treatment did not result in resolution of HS [11,12]. This differs from our case, in which the signs encountered complete regression with antimicrobial therapy, despite the bacterial positivity dynamic renders difficult to identify a primary or single cause. The initial infection with *Serratia marcescens*, a gram-negative facultative anaerobic Enterobacteriaceae, may have induced the production and accumulation of gas and mucus in the thickness of the mucosa, that produced the atypical microbullous aspect of its surface [13]. This bacterium is widely present in water and soils, and is usually considered opportunistic, causing infections in debilitated or immunosuppressed patients, both in human and veterinary medicine [14].

As a matter of fact, the present horse was a carrier of *Streptococcus equi equi* confirmed by PCR at the time of examination, and this may have negatively influenced local and systemic immunity [15]. In equines, *Serratia marcescens* has been reported to be a frequent inhabitant of the conjunctiva of healthy horses [16], but also to cause endocarditis [17], abortion [18] and septicemia [19], with no distinctive pathological features. Nevertheless, in human medicine, infections from this bacterium have manifested in several cases as a particular bullous cellulitis, affecting either the site of entrance of the infection, or the lower extremities when spread via hematogenous route [20,21]. The microbiological picture is further complicated by the streptococcal

infection diagnosed at the first follow-up, that in turn was probably secondary to a functional loss of the upper airway immunity [15].

In the present case, the diagnosis of bacterial GP infection was reached by means of endoscopy and bacteriology. CT examination showed that the THJ had a normal appearance and therefore allowed to rule out a THO. In fact, CT has been demonstrated to be a highly sensitive diagnostic technique for diseases of the head, with a 100 % sensitivity and 96.7 % specificity, while radiology has shown a sensitivity between 16.7 % and 43.5 % depending on the region of the skull being investigated [22]. Nevertheless, the most common cause of HS syndrome in horses is trigeminal-mediated HS, which diagnosis is made by exclusion of other anatomical sources, therefore CT becomes an important diagnostic aid in these cases [3].

Furthermore, CT showed the presence of bilateral otitis media, which could not be confirmed during otoscopy given the absence of exudate that could be sampled non-invasively and the poor visualization, due to the amount of debris present, which, nevertheless, does not represent a pathological finding [23]. Otitis media is a well-known possible cause of HS in horses [24] and the simultaneous involvement of the GP is not uncommon, due to their anatomical vicinity [23]. Nevertheless, the mechanism that leads to the infection of both these structures is still not fully understood. Blythe [5] reported the hematogenous route to be the most common way of entry of infectious agents into the middle ear, whereas an extension from the GP was defined as less common, and the infection never sustained by the same microorganism, as reported by Shusser [25]. Moreover, the *ostium tympanicum tubae auditivae*, which is the opening between the middle ear and the GP, is too small to allow exudate from the GP to drain into the middle ear [25]. In fact, otitis media was described to be an initiating cause for THO [5,23], which was excluded in the present case by CT, but not for GP infection. Therefore, the pathogenesis of bilateral otitis media and GP infection in this case is still uncertain. Given the horse's response to medical treatment and the relative invasiveness, it was not elected to sample the contents of the tympanic bullae by the trans-membranous method [5,26].

The CT examination revealed also the presence of nuchal ligament enthesopathy. Voight and colleagues [27] reported two cases of horses referred for HS, which was caused by nuchal crest avulsion. In addition, new bone proliferation at the site of insertion of the nuchal ligament may result in a tendency of the affected horse to shake their head, especially when the flexion of the poll region is requested [28], as in the

present case. Nevertheless, these calcifications may be observed also as an incidental finding [28], as demonstrated in the present case, since the horse got a full recovery after resolution of the infectious process.

A limitation in the present report is represented by the fact that a transtympanocentesis was not performed, therefore it was not possible to assess if the infectious agent was the same both in the GPs and in the tympanic bullae. Another limitation in the present report is that a second CT examination was not performed, due to obvious financial constraint in face of a clinical resolution. Therefore, we were not able to assess whether otitis media was also resolved by the administration of antimicrobials. Since it is reported that a minimum of 30 days therapy is usually necessary for otitis [5,29,30], the similar antimicrobial course in this case may have been effective on both the GP infection and the otitis media.

To the authors' knowledge, this is the first reported case of a bilateral guttural pouch microbullous infection caused by *Serratia marcescens* and otitis media in a horse presented with HS, that underwent complete resolution with medical therapy.

CRedit authorship contribution statement

G. Forni: Writing – review & editing, Writing – original draft, Visualization, Investigation, Data curation, Conceptualization. **N. Ellero:** Writing – review & editing, Visualization, Investigation, Data curation, Conceptualization. **A. Mannini:** Writing – review & editing, Visualization, Investigation, Data curation, Conceptualization. **L. Scacco:** Writing – review & editing, Visualization, Investigation, Data curation, Conceptualization. **F. Freccero:** Writing – review & editing, Visualization, Supervision, Project administration, Investigation, Data curation, Conceptualization.

Declaration of competing interest

None of the authors has any financial or personal relationships that could inappropriately influence or bias the content of the paper.

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