Exophthalmus secondary to a sinonasal cyst in a horse

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Case Description—A 13-year-old female Miniature Horse was evaluated for progressive unilateral exophthalmia of the left globe of 3 weeks’ duration.

Clinical Findings—Results of a physical examination were unremarkable. Ophthalmic examination identified exophthalmus of the left globe with complete resistance to retropulsion and mild blepharocconjunctivitis. Computed tomography revealed a large, space-occupying mass within the left caudal maxillary and left conchofrontal sinuses. The mass extended into the left retrolbulbar space and contacted the cribriform plate. Trephination yielded copious amounts of turbid yellow fluid. The diagnosis was a sinonasal cyst.

Treatment and Outcome—Subtotal surgical excision of the cyst via a frontonasal osteoplastic flap was curative, with complete resolution of the exophthalmus. Histologic examination confirmed diagnosis of a sinonasal cyst. There was no evidence of cyst recurrence by 4 months after surgery.

Clinical Relevance—Sinonasal cyst should be a differential diagnosis for retrobulbar disease in horses. Exophthalmia may be the only clinical finding in horses with a sinonasal cyst. (J Am Vet Med Assoc 2008;233:285–288)

A 13-year-old 130-kg (286-lb) female Miniature Horse with a 3-week history of progressive unilateral exophthalmia was evaluated at the veterinary medical teaching hospital of The Ohio State University. The horse had been treated by the referring veterinarian with an ophthalmic corticosteroid preparation applied topically every 8 hours, but there had been no improvement in the condition. The horse was considered to be current on vaccinations, and the medical history was otherwise unremarkable.

Results of an initial physical examination at our facility were unremarkable, except for exophthalmus of the left eye. To facilitate ocular examination, xylazine hydrochloride (0.30 mg/kg [0.14 mg/lb], IV) was administered. Examination of the left eye identified exophthalmia with rostral and lateral displacement of the globe, mild conjunctival hyperemia, and blepharopharyngeal edema. There was complete resistance to retropulsion, and evaluation of globe size and ocular motility revealed no abnormalities. A positive result for the menace response confirmed diagnosis of a sinonasal cyst. There was no evidence of cyst recurrence by 4 months after surgery.

Initial diagnostic evaluation included hematologic and serum biochemical analyses, thoracic radiography for evaluation of evidence of metastatic disease, and CT imaging of the head. Results of a CBC were unremarkable, except for hyperfibrinogenemia (439 mg/dL; reference range, 193 to 422 mg/dL). Serum biochemical analysis revealed increases in activity of alkaline phosphatase (276 U/L; reference range, 80 to 187 U/L), γ-glutamyltransferase (41 U/L; reference range, 7 to 20 U/L), sorbitol dehydrogenase (15.1 U/L; reference range, 4 to 13 U/L), creatine kinase (464 U/L; reference range, 150 to 360 U/L), and hypertriglyceridemia (71 mg/dL; reference range, 10 to 60 mg/dL). Results of thoracic radiography were unremarkable, and there was no evidence of pulmonary metastasis.

The horse was anesthetized for CT imaging of the head. Preanesthetic medications administered included potassium penicillin (22,000 U/kg [10,000 U/lb], IV), gentamicin (7 mg/kg [3.2 mg/lb], IV), and flunixin meglumine (0.6 mg/kg [0.27 mg/lb], IV). Tetanus toxoid was administered IM. Anesthesia was induced by IV administration of xylazine (0.75 mg/kg [0.34 mg/lb]),
A 13-year-old Miniature Horse with unilateral exophthalmia of the left globe (asterisk) was positioned in dorsal recumbency. Anesthesia was maintained, and the horse was positioned in right lateral recumbency. Rhinoscopy was performed with a flexible endoscope. Hyperemia of the nasal turbinates with mild distortion of the nasal cavity was visible, but no other abnormalities could be identified. The lining of the left caudal maxillary sinus was performed by use of a Steinmann pin and Jacobs chuck, as described elsewhere.

Copious amounts of turbid, yellow fluid immediately flowed from the trephination site, and a sample was aspirated. Anesthesia was discontinued, and the horse recovered without incident. Treatment was initiated with sulfamethoxazole-trimethoprim (15 mg/kg [6.8 mg/lb], PO, q 12 h) and flunixin meglumine (0.6 mg/kg, IV, q 12 h).

Analysis of fluid obtained from the mass revealed a specific gravity of 1.035, total protein concentration of 10.5 g/dL, and WBC count of 1,320 cells/μL. Cytologic examination revealed a proteinaceous background with nondegenerate neutrophils and vacuolated macrophages, some of which contained RBCs and hemosiderin. Findings were compatible with a diagnosis of sinonasal cyst. Less likely differential diagnoses were a necrotic, fluid-containing neoplastic lesion or the possibility that fluid samples had been obtained from the obstructed sinus.

Surgical excision of the mass was recommended and was performed 4 days after the initial examination at our facility. Anesthesia was as previously described, and the anesthetized horse was positioned in right lateral recumbency. An oscillating saw was used to create a large frontonasal osteoplastic flap in the left frontal bone, as described in another report. A thick lining, consistent with a sinonasal cyst, was identified within the maxillary and conchofrontal sinuses. The lining of the cyst was removed by digital manipulation combined with use of a rongeur and curettage. Excision was incomplete because of the extensive nature of the lesion, involvement of the cribiform plate, and moderate intraoperative hemorrhage. The sinus cavity was packed with gauze and the osteoplastic flap was pressed back into place. Skin margins were apposed with 2-0 polydioxanone. Because of intraoperative blood loss, 1 L of plasma was administered IV during the procedure. The horse recovered from anesthesia without complication; however, mild hemorrhage persisted after surgery. Postoperative hemorrhage was controlled by the application of ice, administration of epsilon-aminocaproic acid (30 mg/kg [13.6 mg/lb], IV), and insertion of a suture to close the left naris. Two days after surgery, the suture was removed from the naris and the gauze packing was extracted from the sinus cavity without incident.

Histologic examination of the excised tissue revealed a ciliated, pseudostratified columnar epithelium with mucus-producing goblet cells. A thick fibrous capsule was evident, and spicules of trabecular bone were detected (Figure 2). Extensive hemorrhage and edema were evident within the subepithelial tissues, with evidence of a mild inflammatory response. Aerobic and anaerobic culture of the fluid sample obtained from the sinus yielded no growth after 7 days. These findings confirmed the diagnosis of a sinonasal cyst.

Forty-eight hours after surgery, there was a reduction in the degree of exophthalmia and slight improvement in retropulsion of the left globe. Conjunctival hyperemia of the left eye was persistent but diminished. Complete physical and ophthalmic examination findings were otherwise unremarkable. The horse was discharged from the hospital 3 days after surgery. Medications prescribed at the time of discharge included phenylbutasone (2 mg/kg [0.9 mg/lb], PO, q 24 h for 2 days) and sulfamethoxazole-trimethoprim (15 mg/kg, PO, q 12 h for 10 days).

Four months after surgery, evaluation by the referring veterinarian revealed complete resolution of the exophthalmus and blepharedema, and the left eye appeared normal. There was no evidence of recurrence of the sinonasal cyst, and no postsurgical complications were identified.
Humans has been associated with obstruction of the sinus mucosa, which can be congenital or may be caused by persistent inflammation, trauma, scarring, repeated infection, or previous surgery. A higher incidence of exophthalmia has been described in primary sinus mucoceles in humans, relative to mucoceles secondary to trauma or maxillofacial surgery. Whether the specific cause of sinonasal cysts in horses similarly influences their potential for orbital extension remains to be determined, but certainly the horse reported here had no known history of trauma or previous surgical intervention.

Computed tomography was chosen for imaging the head of the horse reported here. In a study of orbital disease in dogs, investigators found diagnostic value to be similar for CT and ultrasonography; however, CT more frequently revealed extraorbital involvement. Magnetic resonance imaging is an alternative imaging modality, and the superior soft tissue detail of MRI may be more sensitive for distinguishing a sinonasal cyst from neoplasia. However, MRI has limited value in assessing bone erosion and sinus configuration.

The locaion of the sinonasal cyst in this Miniature Horse was typical (i.e., within the conchofrontal and caudal maxillary sinuses). Extension into the orbit reflected the potential of sinonasal cysts for bone destruction. This has been correlated with direct pressure of the cyst wall on bone. Increased concentrations of prostaglandin E2, interleukins, and tumor necrosis factor within the mucosa of sinus mucoceles in humans may also contribute to the invasive nature of these conditions.

Results of rhinoscopy are typically not specific for sinonasal cyst disease, which was true for the horse reported here. Abnormal respiratory tract secretions and nasal cavity distortion are commonly reported and support imaging findings of sinus disease. Direct endoscopic examination of the paranasal sinuses may be of greater diagnostic value. For the horse reported here, evaluation of a biopsy specimen of the mass yielded similar results without the benefit of gross examination. Fluid obtained from the cyst in this horse had negative results for bacterial culture and had cytologic evidence of chronic inflammation and erythrophagocytosis, which are findings consistent with previous reports. Bacterial colonization of sinonasal cysts has been described, but this appears to be atypical.

Discussion

Sinonasal cysts have been described with some frequency in horses, accounting for approximately 13% of all paranasal sinus disease in horses. However only 1 report describing exophthalmus associated with sinonasal cyst disease was identified by the authors. In that report, exophthalmus was evident in addition to more typical signs of sinonasal cyst disease, such as distortion of the facial bone. Other common reported findings include unilateral nasal discharge, obstruction of nasal airflow, obstructive respiratory noise, epiphora, and submandibular lymphadenopathy. In the horse described here, exophthalmia was the only clinical finding identified.

Similarities between sinonasal cysts in horses and sinus mucoceles in humans have been described. Both are expansile, mucus-filled cavities within the paranasal sinuses and are capable of bone destruction. Histologically, both possess a mucus-secreting epithelial lining, which is typically accompanied by a mild inflammatory response. Orbital extension of sinus mucoceles in humans has been verified. Exophthalmus is most commonly reported in these patients, although exophthalmus related to bone destruction has also been described. The apparent lower incidence of orbital extension in sinonasal cysts in horses may reflect a paucity of reports in the literature. It is also possible that differences in comparative anatomy, specifically the physical proximity of the sinus ostia to the orbit, may make sinonasal cysts in horses less prone to orbital extension.

The cause of these conditions has not been elucidated. However, the development of sinus mucoceles in humans has been associated with obstruction of the sinus ostium, which can be congenital or may be caused by persistent inflammation, trauma, scarring, repeated infection, or previous surgery. A higher incidence of exophthalmia has been described in primary sinus mucoceles in humans, relative to mucoceles secondary to trauma or maxillofacial surgery. Whether the specific cause of sinonasal cysts in horses similarly influences their potential for orbital extension remains to be determined, but certainly the horse reported here had no known history of trauma or previous surgical intervention.

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Sinonasal cysts typically require surgical excision because of their tendency for bone destruction and continued expansion. Although complete extirpation is advocated, this is rarely achieved because of the sensitivity of associated anatomic structures (such as the cribriform plate) and the potential for hemorrhage. In the horse reported here, excision of the sinonasal cyst was performed through a large frontal osteoplastic flap, as described elsewhere. This technique provided access to the conchofrontal and maxillary sinuses and enabled maximal debridement of the extensive lesion. Endoscopic surgery is an alternative and has been advocated for treatment of humans that have sinus mucoceles with orbital involvement. Endoscopic debridement and lavage performed in 3 standing, sedated horses with sinonasal cysts indicated that this is a technique best reserved for selected horses with small cysts.

In the horse reported here, surgical excision was curative, with complete resolution of the exophthalmus.
without complication or recurrence by 4 months after surgery. This outcome was consistent with that in other reports of horses with sinonasal cyst disease. Cyst recurrence appears to be a rare event; however, a persistent mucoid nasal discharge has been described in a small percentage of patients. Transient postoperative nasal discharge is a more commonly reported sequela but is of minimal importance.

The information reported here should make equine practitioners aware of sinonasal cysts as a cause of retrobulbar disease in horses. Additionally, exophthalmia may be the only clinical finding in horses with sinonasal cyst disease. Diagnosis is aided by use of CT, and the prognosis for resolution after subtotal surgical excision via a frontonasal osteoplastic flap is good.

References