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Case report Κλινικό περιστατικό

Nasal Epidermal Inclusion Cyst in a horse: A Case Report

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ABSTRACT: Nasal epidermal inclusion cysts, previously known as "atheroma" or "sebaceous cyst", are cutaneous lesions that occur in the caudodorsal aspect of the nasal diverticulum, between the epithelium of the diverticulum and the skin of the nostril. They are mostly benign, asymptomatic, non-painful, and are primarily of cosmetic significance. Nasal epidermal inclusion cysts are rather rare in horses in contrast with other species or humans. According to our knowledge, nasal epidermal inclusion cysts have not previously been reported across the Mediterranean basin. A 12-year-old, Warmblood gelding was presented to the Equine unit, Aristotle University of Thessaloniki for evaluation of a facial swelling on the right side of the nose. The swelling was first observed 2 years before presentation and had progressively increased in size. On presentation, the horse was bright, alert, responsive and all parameters of the clinical examination were within normal limits. The presence of a large, fluctuating cyst, of approximately 6cm in diameter was noticed on the caudodorsal aspect of the right nasal diverticulum. Based on the location and clinical characteristics, a diagnosis of nasal epidermal inclusion cyst was made, and surgical removal was elected. Following anesthesia, a linear skin incision up approximately 8cm long, directly over the epidermal inclusion cyst was made through the skin and subcutaneous tissues. The cyst was dissected from the surrounding tissues, drainage of the wound into the false nostril was done by making a 2-cm incision from the defect into the false nostril and the operating site was thoroughly lavaged. The horse was re-examined 2 months later and showed complete resolution of clinical signs with no complications. The surgical wound had closed completely leaving no obvious scar. The horse was still blemish - free two years after surgery.

Keywords: horse; nasal epidermal inclusion cyst; atheroma; cyst removal

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INTRODUCTION

n the horse epidermoid or dermoid cysts are found L in the nostrils, the ventral thorax, the withers (Boles and Charles, 1979; Pascoe and Summers, 1981; Wellington and Scott, 1991), the central nervous system, the gastrointestinal system, the musculoskeletal system (Boles and Charles, 1979; Gaughan and DeBowes, 1993; Gaugham, 1995; Dixon, 1991), the reproductive system, the esophagus, the nasal and paranasal cavities, and the retrobulbar space (McGavin and Zachary, 2012; Scott et al., 1977; Gunnarsdottir et al., 2014; Muñoz et al., 2007; Visser et al., 2019). These cysts most likely represent embryologic remnants that they can cause severe damage followed by remarking clinical symptoms (Fraseret al., 2006; Headley et al., 2009; Kelly and Watson, 1976; Peters et al., 2003; Sanz et al., 2006; Stenberget al., 2004).

Nasal epidermal inclusion cysts, previously known as "atheroma" (Hamlat et al., 2005)or "sebaceous cyst" (Boles and Charles, 1979), are cutaneous lesions that occur in the caudodorsal aspect of the nasal diverticulum (Gaughan and DeBowes, 1993), between the epithelium of the diverticulum and the skin of the nostril (Gaugham, 1995). These nasal cysts can be bilateral but are usually unilateral and vary in size (1.5 - 4 cm). Nasal inclusion cysts can be seen in all age groups. Most commonly, they occur in younger horses (1-3 years old) but, less frequently, they can be present at birth or develop by the age of 6 months (Boles and Charles, 1979; Gaughan and DeBowes, 1993; Gaugham, 1995; Dixon, 1991). Still, it is not unusual to be found in older horses (Boles and Charles, 1979; Gaughan and DeBowes, 1993; Gaugham, 1995; Dixon, 1991). They are mostly benign, asymptomatic, non-painful, and are primarily of cosmetic significance. On the contrary, in humans there are several reports of epidermal inclusion cysts that can undergo malignant transformation to squamous cell carcinoma (Hamlat, 2005;), a scenario that is extremely rare in horses (Peters, 2003). If untreated, due to continuous exfoliation of squamous cells from its lining (Tremaine andDixon,2001),this cyst can gradually or rapidly expand and even become large enough to obstruct airflow or cause exercise intolerance (Boles and Charles, 1979).

Nasal epidermal inclusion cysts are rather rare in horses in contrast with other species or humans. The differential diagnosis should include all the mass-like structures; however,an easy tentative diagnosis is supported by its typical location and physical appearance. A definite diagnosis can be made with further histopathological evaluation.

There are many therapeutic options, ranging from a less invasive chemical ablation with formalin or corticosteroids, to a complete surgical excision. The efficacy of chemical ablation is debatable due to a low success rate and considerable side effects (Schumacher et al.,1998; Williamsand Nickels, 2020; Schumacher and Dixon, 2007). Surgical removal is the treatment of choice, especially before the cyst increases dramatically in size or shows signs of malignancy. Unfortunately, reports on successful surgical management of equine nasal epidermal inclusion cysts are limited (Tremaine and Dixon, 2002). According to our knowledge, nasal epidermal inclusion cysts have not previously been reported across the Mediterranean basin.

CASE DESCRIPTION

A 12-year-old, Warmblood gelding was presented to the Equine unit, Aristotle University of Thessaloniki for evaluation of a facial swelling on the right side of the nose. The swelling was first observed 2 years before presentation and had progressively increased in size. There was no history of known trauma. Intralesional injection of 6mg betamethasone (Celestone Chondrose[®], Schering-Plough)that was performed twice by the referring veterinarian in order to decrease inflammation and inhibit fluid production, led to a partial, temporary improvement of the condition of the blemish, but the lesion reoccurred.

On presentation, the horse was bright, alert, responsive and all parameters of the clinical examination were within normal limits. The presence of a large, fluctuating cyst, of approximately 6cm in diameter was noticed on the caudodorsal aspect of the right nasal diverticulum (false nostril) (Fig.1). There was no respiratory distress, and the horse was performing and eating normally, showing no signs of pain or discomfort. No nasal discharge or regional lymphadenopathy was seen at presentation. The examination of the head indicated a unilateral, soft, fluctuant, nonpainful, cool, smooth, round, immobile mass with a diameter of approximately 6cm, attached to the dorsolateral wall of the right nasal diverticulum. The overlying skin was normal. No signs of head shaking or head bumping were noted during clinical examination or reported by the owner. Palpation with one finger in the nasal diverticulum and another over the skin indicated that the mass was located between the skin and



Figure 1. Initial presentation. A large cyst on the right nasal diverticulum



Figure 2. A linear skin incision directly over the cyst was performed under standing sedation

epithelium of the nasal diverticulum. Radiographic examination showed no abnormalities or connection to the nasal bones. Following fine needle aspiration, the fluid within the mass was viscous and grey, resembling sebum. Although histopathology was suggested, the owner declined that option because of financial constraints. Based on the location and clinical characteristics, a tentative diagnosis of nasal epidermal inclusion cyst was made, and surgical removal was elected with the owner's consent.

The horse was pre-medicated with penicillin/ streptomycin (20,000IU and 15mg/kg respectively, intramuscularly, SID) (Ilcocillin PS®, Norbrook) and phenylbutazone (4mg/kg intravenously, SID) (Stoplac®, Provet). A standing en-bloc removal of the cyst was decided. For that purpose, the horse was sedated with 0,1mg/kg romifidine (Sedivet[®], Boehringer Ingelheim), the hair over the nostril was clipped and the skin and nasal diverticulum was aseptically prepared to minimize surgical contamination. The site was desensitized with the subcutaneous injection of lidocaine 2% (Xylozane®, Demo) in a longitudinal line 12cm long, centered directly over the cyst and the tissues around it. A linear skin incision up approximately8cm long, directly over the epidermal inclusion cyst was made through the skin and subcutaneous tissues (Fig.2). The cyst was dissected from the surrounding tissues using a Metzenbaum scissors around the margin of the cyst (Adams and Fessler, 2000). Despite careful dissection the cyst ruptured. Following complete removal, drainage of the wound into the false nostril was done by making a2-cm incision from the defect into the false nostril (Adams and Fessler, 2000). After complete removal, the operating site was thoroughly lavaged with saline to discard any cyst debris. The incision was sutured in two layers to minimize the dead space. For the subcutaneous tissue, a 2/0 polyglactin suture material (Vicryl[®], Ethicon) was used, and the dermis was carefully apposed with a 2/0 polypropylene suture material (Prolene[®], Ethicon), using the intradermal suturing technique to avoid complication with a noticeable scar post-operatively.

Postoperatively, penicillin streptomycin, -(20,000IU and 15mg/kg respectively, intramuscularly SID) (Ilcocillin PS®, Norbrook) and phenylbutazone (4mg/kg intravenously SID) (Stop-lac®, Provet)were administered for 5 days. The horse was monitored daily by the owner and it was re-evaluated 14 days after surgery, when the sutures were removed. At that time, there was no visible, external swelling of the nose. The horse was re-examined 2 months later and showed complete resolution of clinical signs with no complications. The surgical wound had closed completely leaving no obvious scar. The horse was still blemish - free two years after surgery.

DISCUSSION

Cysts are non-neoplastic, simple sac-like struc-

tures with an epithelial wall and keratinous to amorphous content (Ginel et al., 2007). Cutaneous cysts are rare in the horse and have generally been described as epidermal or dermoid cyst. The terms most frequently used are epidermal, epidermoid, or epidermal inclusion cysts and they have recently been categorized as infundibular, follicular, or keratinizing cysts (Hillyer et al., 2003).In the past, the terms "atheroma" and "sebaceous cyst" were also used, as it was falsely believed that the nasal diverticulum is lined by thin skin and has an abundance of sebaceous glands (Boles and Charles, 1979). Lately, it has been proven that these terms are inappropriate as sebaceous glands are not usually identified in their lining (Tremaine, and Dixon, 2002).

The pathogenesis of these cysts is perplexing. They are thought to be congenital, originating from defective embryological tissue development during embryogenesis. Epidemiologically, there is no breed or gender predisposition (Barnes et al., 2021), and a large age variation has been reported (2-18 years old) (Tremaine, and Dixon,2001). In our case the gelding was 12 years old, and the cyst had been gradually increasing in size for 2 years before referral. Therefore, the congenital theory seems unlikely in this case. Another possible explanation would be for the cyst to be post-traumatic. It has been postulated that they can occur after traumatic displacement of the epithelial tissue (Hillyer et al., 2003; Grant, 2016;), suggesting that environmental factors are responsible for the cyst occurrence. This can explain the late age presentation despite the congenital basis. Although, the gelding in our case had no history or evidence of trauma, it is common for small wound to escape the owner's attention.

As mentioned before, epidermoid cysts in horses are rather rare. In their majority, they do not have any clinical significance causing mostly a cosmetic defect (Frankeny, 2003; Camus et al., 1996; Hillyer et al., 2003), so they can be left untreated. However, there are cases where the cyst interferes with the nose band or is large enough to obstruct the airflow; thus, it is advisable to be addressed as soon as possible before they grow substantially in size. Extreme clinical signs caused by the mechanical pressure of the cysts are to be expected when they occur in other organs, such as the colon, tendons, or brain (Peters et al., 2003). Therefore, they should not be generally considered as asymptomatic and should be included in the differential diagnosis.

The differential diagnosis of inclusion cysts can include any mass-like formation on the site, such as abscess, hematoma, neoplastic tissue, other cyst, foreign body, or fistula. Due to the clinical characteristics and the typical location of the epidermal inclusion cyst - unilateral, fluctuating, on the nasal diverticulum without obstructing air flow - a tentative diagnosis is easy to make. In this case, diagnosis was based on history, clinical presentation and fine needle aspiration of the mass, and it was confirmed on surgery. Ideally, a histopathologic examination would lead to a definite diagnosis of the lesion. Additionally, although there is only one report of a cyst with histological signs of malignancy (Peters 2003), it would differentiate it from neoplasia. The diagnostic procedure can also include the noninvasive approach of ultrasonographic examination, useful for the differentiation of abscess and hematoma due to their characteristic ultrasonographic appearance (Ali et al., 2012). Unfortunately, the owner declined further diagnostic evaluation due to financial constraints. Still, diagnosis can be based solely on the characteristic clinical signs (Tremaine and Dixon, 2002).

There is a variety of therapeutic options for epidermal inclusion cysts. Aspiration of the cysts usually results in reoccurrence or occasionally in local abscessation (Tremaine and Dixon, 2002). Intralesional corticosteroid injections are an integral part of the dermatological therapeutic armamentarium for human medicine since 1951 (Nickels and O'Neill, 2019). They have been indicated for various epidermal lesion, including mass-like, nodular, and cystic lesions, with promising results (Cramp et al., 2014). The number of injections depends on the disease, site, size and severity of lesions, age of the patient and response to previous injections. The interval between injections is dictated by the improvement produced by the first injection and varies with the type of the lesion being treated (Deshmukh et al., 2014). In our case, the cyst has been previously treated with two intralesional injections of 6mg betamethasone (Celestone Chondrose[®], Schering-Plough) and led to a partial, temporary improvement but the lesion reoccurred.

To avoid surgery, a conservative therapeutic approach has been described (Frankeny 2003), where a single intralesional formalin injection resulted in successful total resolution of epidermal inclusion cysts in 5 horses. According to that report, the adverse effects seen were cyst enlargement after injection and nose rubbing, signs that resolved completely within

few days (Frankeny, 2003). This therapeutic method was not elected in our case, as formalin injection can cause soft tissue damage and necrosis and has been linked with laminitis (Schumacher et al., 1998), long-lasting local inflammation and hyperalgesia or even abscessation of the injection site (Williams and Nickels, 2020). A newer report highlights the formation of a large nasocutaneous fistula as a sequel to intralesional formalin administration into a nasal inclusion cyst which warranted further and rather complicated and costly surgical intervention (Schumacher and Dixon,2007). That is why intralesional injection of formalin was not elected in this case.

The treatment of choice is complete surgical removal and various techniques have been described. The procedure presented in this paper can be easily performed in the standing sedated horse with the use of local anesthesia, either infiltrating the skin over the cyst or with perineural ipsilateral infraorbital anesthesia. The standard approach is the en-block excision of the cyst with a longitudinal incision directly over it and the entire cyst lining removal with a Metzenbaum scissors (Barnes et al., 2021), as it was performed in our case. Another technique is the approach through the nasal diverticulum to prevent leaving a noticeable scar on the skin after surgery. Proposed advantages of surgical resection via the standard approach versus an incision through the diverticulum include the overall easier technique, and the substantial elimination of the possibility of leaving part of the cyst lining unremoved thus resulting in reoccurrence (Cramp et al., 2014). Moreover, the operating site through the diverticulum is rather narrow and may require general anesthesia to have a clear look inside it. An alternative technique on the standing sedated horse with perineural infraorbital anesthesia has also been described (Schumacher et al., 1997). According to

this method, the cyst is lanced with a stab incision through the nasal diverticulum and a roaring bur is used to completely remove the lining of the cyst. Subsequently, the wound is left open to heal by second intention. The last surgical option would be to create a nasal flap over the cyst to dissect it; this technique is problematic and unnecessary considering the hemorrhage, the bigger scar, and the superficial location of the cyst. In this case, the complete surgical excision over the cyst was chosen as this technique provides better visualization of the entire cyst and provides the benefit of avoiding the risks of general anesthesia, as the operation was performed with the horse standing, under sedation and local anesthesia. The possible disadvantage of scar formation was addressed with the intradermal suturing pattern of the skin, leading to an excellent cosmetic result.

During dissection, the cyst ruptured and that made the removal challenging as the cyst lining was not easily retractable, and the operating site was contaminated with the cyst sebum-like material. This is a common intraoperative complication because the cyst has a very thin membrane. In that case drainage of the wound into the false nostril should be done by making a 2-cm incision from the defect into the false nostril (Ffrooz et al., 1995) and thorough lavage of the site with saline following complete removal of the cyst lining.

Post-operation complications have not been reported and after complete surgical removal, recurrence is uncommon. The horse of our study was blemish free 2 years after the procedure with no other complains from the owner.

CONFLICT OF INTEREST

None declared.

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