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ORIGINAL ARTICLE

Nasopharyngeal sialoceles in 11 brachycephalic dogs

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Abstract

Objective: To report clinical features, diagnosis, surgical treatment, and outcomes of brachycephalic dogs with nasopharyngeal sialoceles (NPS).

Study Design: Retrospective case series.

Animals: Eleven brachycephalic dogs with NPS.

Methods: Diagnosis of NPS was based on identification of cystic nasopharyngeal masses containing saliva-like mucoid material on retroflexed endoscopy. Biopsies were obtained to support the diagnosis. Surgical deroofing was performed under endoscopic guidance, and endoscopic examination was repeated at least 3 months after surgery. The owners rated the outcome of surgery via a follow-up telephone inquiry.

Results: Pugs (n = 8), French bulldogs (n = 2), and English bulldogs (n = 1) were included in this study. Eight of the NPS were ventral, 6 were obstructive, and 2 were multiple; both the obstructive and the multiple sialoceles were ventral. Surgical deroofing of the sialoceles was performed under endoscopic guidance with biopsy forceps in 5 cases and by diode laser in 6 cases. No recurrence was diagnosed. Ten of 11 owners reported that they were satisfied with the outcome.

Conclusions and Clinical Significance: NPS may be an uncommon consequence of chronic nonphysiologic mechanical stress causing changes in minor nasopharyngeal salivary glands. Direct visualization by nasopharyngoscopy is the best technique for detection of NPS, and we advise routine examination of the nasopharynx by endoscope in all brachycephalic dogs before performing surgery. Either endoscopic guided biopsy forceps or diode laser surgery can be used for noninvasive, rapid, and effective resolution of NPS in all of the dogs.

1 | INTRODUCTION

A sialocele is an accumulation of saliva within the fascial planes of the throat, oral cavity, or orbit.[1] The cranial submandibular and sublingual regions are the most common sites,[1,2] but the pharyngeal,[3,4] parotid,[5] and zygomatic[6] areas can also be involved, depending on the affected gland and the site of the disruption. Although trauma has been reported as the primary cause of sialocele,[7] evidence is lacking to support this theory. Attempts to reproduce sialoceles by ligating the mandibular and sublingual ducts rostrally or caudally or by damaging them directly were consistently unsuccessful in creating sialoceles.[8]

In addition to the major salivary glands, there are numerous minor salivary glands located throughout the oral cavity and the pharynx. These lay within the submucosa of the buccal, labial, pharyngeal, and lingual mucosa[9] and are designated as *labial, buccal, molar, lingual,* or *palatine,* depending on their location.[10] Rarely, trauma or obstruction in the minor salivary glands gives rise to blister-like cystic lesions, particularly around the soft palate.[11]

Although major salivary gland sialoceles have been thoroughly described in dogs,[1-6] a review of the literature failed to yield any previous report of nasopharyngeal sialocele (NPS) of the minor palatine salivary glands (*glandulae*

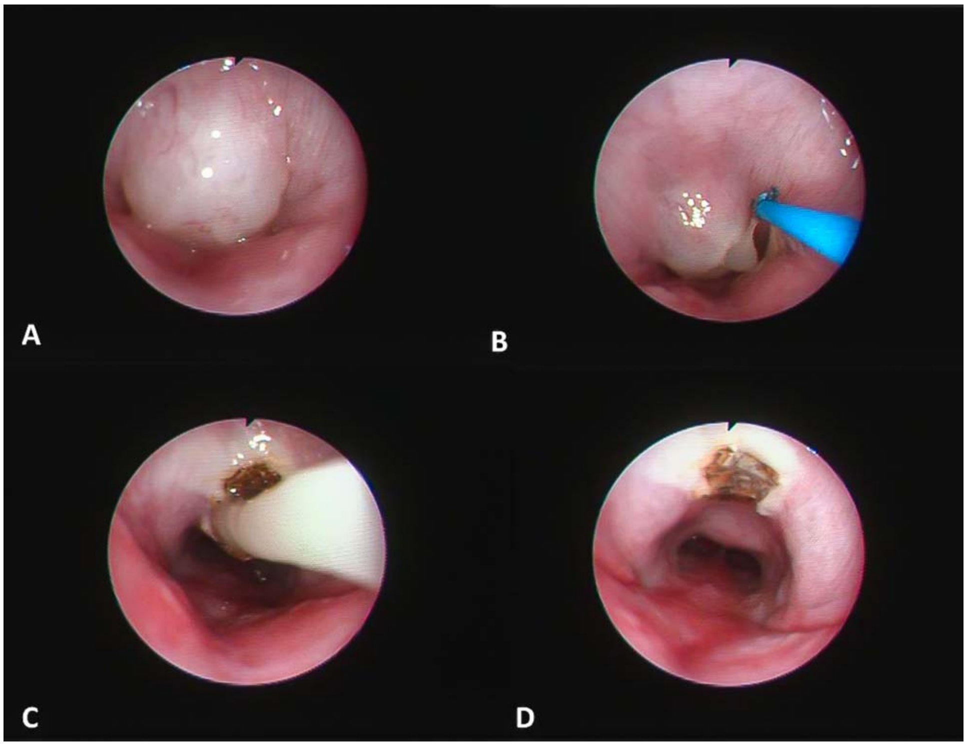

FIGURE 1 Ventral obstructive nasopharyngeal sialocele (case 8). A, Large sessile NPS completely obstructs the nasopharyngeal passage. Image in the retroflexed endoscope is inverted: the ventral nasopharyngeal wall is in view. B, Diode laser quartz fiber begins vaporizing the outer surface of the sialocele, allowing the mucoid content to escape. C, Outer surface is fully removed, and mucoid fluid is aspirated. D, A crater is now evident in place of the NPS. Both choanae are fully visible, and the nasopharyngeal diameter is restored. NPS, nasopharyngeal sialocele

palatinae) in dogs, and specifically in brachycephalic breeds. This retrospective study sought (1) to characterize the signalment, history, clinical signs, imaging, and endoscopic features of 11 brachycephalic dogs with NPS; (2) to describe the treatments used; and (3) to evaluate the follow-up.

2 | MATERIALS AND METHODS

Medical records of the Veterinary Hospital "I Portoni Rossi" were searched for cases of nasopharyngeal masses with a final diagnosis of NPS between January 2013 and January 2016. For each case, the recorded information included signalment, history, clinical signs, cervical radiographs (and, when available, computed tomography [CT] scan of the same area), upper and lower airway endoscopic findings, cytological and histological findings, surgical procedures, and outcome. Patients were excluded from this study if their digital files were incomplete or if they were lost to follow-up. In all dogs, a lateral radiograph of the larynx and pharynx was made after induction of general anesthesia, with the endotracheal tube removed if possible.

Diagnosis was based on identification of a nasopharyngeal cyst-like mass filled with thick and mucoid material on retroflexed endoscopy (Fibroscope 60001VL; Karl Storz, Tuttlingen, Germany; Figure 1A). The wall of the cyst was penetrated with biopsy forceps, and a sample of the fluid was collected via the endoscope. Examination of a drop of the fluid smeared on a glass slide suggested that it was saliva (Figure 2). A sialocele was confirmed when occasional foamy macrophages along with a small number of neutrophils and erythrocytes were found on a pinkish-blue fibrillar background on a May–Grüwald–Giemsa-stained smear.

NPS were classified as "dorsal" (Figure 3) or "ventral" (Figures 1A, 4), according to the location affected. They were further classified as "obstructive" if they occupied 70%-100% of the nasopharyngeal opening (Figures 1A, 4) and

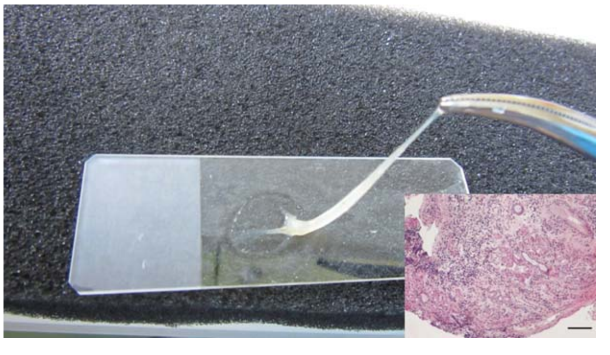

FIGURE 2 Appearance of the fluid contained in the nasopharyngeal sialocele; a sticky, viscous fluid is consistent with saliva. Inset, Histopathological appearance of a nasopharyngeal sialocele (case 6). Moderate salivary gland hyperplasia, duct dilation, and mucus stasis mixed with inflammatory cells can be seen. Hematoxylin and eosin stain ×100. Scale bar = 100 μm

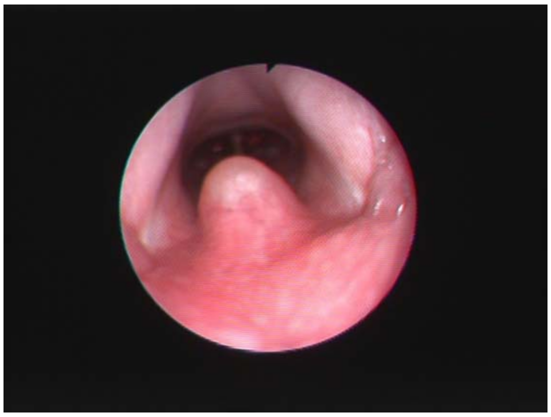

FIGURE 3 Dorsal nonobstructive nasopharyngeal sialocele (case 5)

"multiple" if more than 1 cyst-like mass was identified by imaging (Figure 5) or endoscopy. Only cases in which biopsies from both the outer layer and the floor of the nasopharyngeal mass excluded an alternative pathology were included in this study. Specimens for histological examination obtained during endoscopy were fixed in 10% neutral buffered formalin, and paraffin-embedded sections were cut at 4 µm and stained with hematoxylin and eosin. They were evaluated by a pathologist who was blind to the cytological diagnosis.

Surgical deroofing of the sialocele was performed at the same time as correction of any other component of the brachycephalic syndrome. The surgery sought to remove as much as possible of the external wall of the cyst and to prevent further accumulation of saliva and recurrence of a sialocele. We used 2 techniques, depending on availability of instrumentation. In the first method, flexible endoscopic biopsy forceps with alligator jaws (FB211D.A; Olympus, Tokyo, Japan) via the working channel of the endoscope were used to remove the external mucosal surface piece by piece, in so far as possible. The second method sought to remove the outer wall with a 400-µm-diameter, 4-W-power, 980-mµ-wavelength diode laser (Quanta Sistem, Milan, Italy) in contact, continuous wave mode (Figure 1A-D).

FIGURE 4 Ventral obstructive nasopharyngeal sialocele producing complete nasopharyngeal obstruction (case 6)

Postsurgical endoscopic nasopharyngeal examination was performed in all dogs at least 3 months after surgery. After surgical correction of NPS and any other component of the brachycephalic syndrome, telephone interviews with the owners of the dogs were conducted 6 months or more after surgery. The owners were asked to rate the outcome of surgery as poor (if the dog's condition was unchanged or worse), moderate (if there was good improvement but some limitation of physical activity), or adequate (if improvement was fully satisfactory).

3 | RESULTS

3.1 | Descriptive data

One hundred fourteen brachycephalic dogs underwent thorough endoscopic evaluation of the respiratory system between January 1, 2013 and January 30, 2016. This population included 29 English bulldogs (25.4%), 46 French bulldogs (40.3%), 35 Pugs (30.7%), and 4 Cavalier King Charles spaniels (3.5%). Eleven of 19 cases with a nasopharyngeal cyst-like mass on endoscopy fulfilled the inclusion criteria (10.8%). Among the remaining 8 cases, 4 were excluded because of lack of postsurgical endoscopic control, 3 were excluded because of lack of a histological diagnosis, and 1 was lost to follow-up. The prevalence of NPS in our general brachycephalic breed dog population was 9.6%; the prevalence in the different breeds was 3.4% in English bulldogs, 4.3% in French bulldogs, and 22.8% in Pugs.

The population meeting our criteria for inclusion consisted of 6 males and 5 females, aged 1-6 years (mean 4.5, median 5.1) including 8 Pugs, 2 French bulldogs, and 1 English bulldog (Table 1). Five of the 11 dogs had been treated surgically for 1 or more components of the brachycephalic syndrome, without substantial improvement. Eight of the NPS were ventral, 6 were obstructive, and 2 were multiple; both obstructive and multiple NPS were ventral (Table 1). All of the NPS were located dorsal to the soft palate.

3.2 | Clinical signs and imaging findings

The most frequent clinical signs were snoring, extreme discomfort during sleeping, hypersomnolence, nasal discharge, and air swallowing (Table 2). Extreme discomfort during sleeping and hypersomnolence were present in all dogs with obstructive NPS.

No radiographic evidence of nasopharyngeal mass was detected in this study. CT was obtained in 3 of 11 dogs. A single soft tissue opacity arising from the ventral side of the nasopharynx and obstructing completely the lumen was clearly seen in 2 dogs (Figure 6). Two fluid-filled masses were identified in the nasopharynx of the third dog; 1 lesion

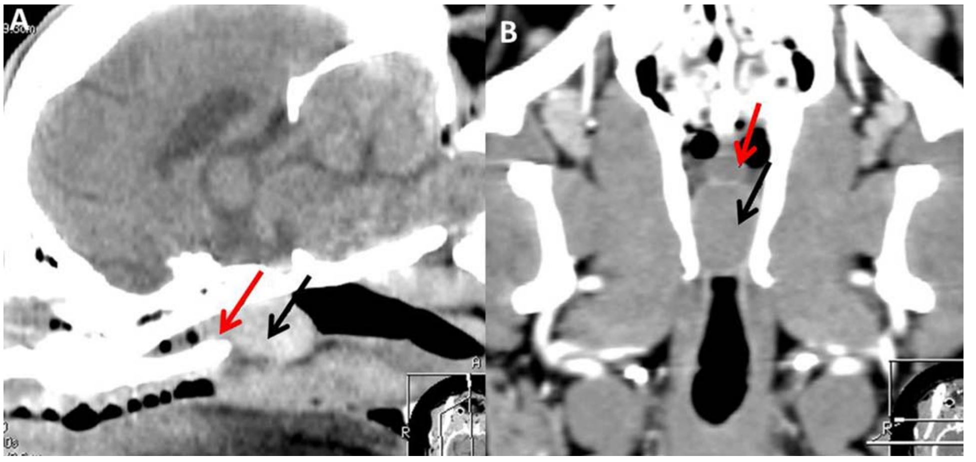

FIGURE 5 Midsagittal (A) and dorsal (B) computed tomograms from a Pug with multiple ventral obstructive nasopharyngeal sialoceles (case 9). A small cyst-like lesion was present at the end of the hard palate (red arrow), and a large lesion was located dorsal to the soft palate (black arrow)

measured 3 mm in width, and the other completely obstructed the nasopharynx (Figure 5).

3.3 | Pathology

Cytological examination of fluid from all of the cysts revealed features consistent with a sialocele[12]; an amorphous basophilic background, highly vacuolated macrophages, and nondegenerate neutrophils were the most common cells. Erythrocytes were uncommon, and there were no hematoidin crystals.

The histological findings included salivary gland hyperplasia (n = 8), moderate duct dilation (n = 8), mucus stasis and exfoliated epithelial cells (n = 6), focal mucous spilling (n = 5), and moderate lymphocytic and neutrophilic inflammation (n = 5). The external capsule was inflammatory and fibrous tissue, with no epithelial demarcation.

3.4 | Endoscopy, therapy, and outcome

A nasopharyngeal mass resembling a cyst, having a smooth surface and covered with well-vascularized pinkish mucosa, was identified on endoscopy in all dogs (Figures 1A, 3–4). The ventral or dorsal origin of the NPS was easily identified as was the degree of nasopharyngeal obstruction. In 2 cases of multiple NPS, the second lesion was detected after the first had been removed. In all cases, the NPS was located in the nasopharynx dorsal to the soft palate (Figures 5, 6); in 1

TABLE 1 Signalment, previous treatments, endoscopic findings, and surgical technique used in 11 brachycephalic breed dogs with a nasopharyngeal sialocele

Case	Breed (sex/age, y)	History	Endoscopy	Surgery
1	Pug (M/4)	REP, SNC, ESR	Obstructive, ventral, single	Forceps
2	Pug (M/5)	NPT	Nonobstructive, ventral, single	Forceps
3	Pug (FN/4)	NPT	Obstructive, ventral, multiple	Forceps
4	Pug (M/6)	REP, SNC	Obstructive, ventral, single	Laser
5	Pug (M/3)	NPT	Nonobstructive, dorsal, single	Forceps
6	Pug (FN/7)	REP, SNC, ESR	Obstructive, ventral, single	Laser
7	Pug (FN/5)	NPT	Nonobstructive, ventral, single	Laser
8	FBD (FN/6)	REP, SNC	Obstructive, ventral, single	Laser
9	Pug (M/2)	NPT	Obstructive, ventral, multiple	Laser
10	FBD (M/2)	NPT	Nonobstructive, dorsal, single	Laser
11	EB (FN/6)	REP, SNC, ESR	Nonobstructive, dorsal, single	Laser

EB, English bulldog; ESR, everted saccules resection; F, female; FBD, French bulldog; FN, neutered female; Forceps, surgery performed with forceps; Laser, surgery performed with diode laser; M, male; NPT, no previous treatment; REP, resection of elongated soft palate; SNC, stenotic nares correction.

TABLE 2 Preoperative clinical signs in 11 brachycephalic dogs with a nasopharyngeal sialocele

Clinical signs	No. of dogs
Snoring	11
Discomfort during sleeping	11
Hypersomnolence	10
Nasal discharge	7
Air swallowing	7
Exercise intolerance	8
Cyanosis	2

case of multiple NPS, 1 lesion was located at the end of the hard palate, and the other was dorsal to the soft palate.

Five dogs were treated by removing as much of the mass as possible with a biopsy forceps. This required 20-30 minutes and resulted in a crater with negligible hemorrhage. In the other 6 cases, the cyst was deroofed by diode laser. This required an average of 12 minutes, seemed easier than with biopsy forceps (Figure 1), and was also without significant hemorrhage. There were no complications during surgery in any of the patients.

Postsurgical evaluation and endoscopic examination were performed in all patients at 3-18 months. There was no difference in the outcome of the 2 procedures, and no recurrence of NPS was observed. The owners of 10 of the dogs reported that the outcome was satisfactory and that there were no residual signs other than occasional snoring during exercise and sleeping. However, in 1 dog, nasal discharge ceased after removal of the sialocele but severe exercise intolerance and mild discomfort during sleeping persisted despite previous correction of stenotic nares and resection of an elongated soft palate and laryngeal saccules. Laryngeal collapse, laryngeal granuloma, and epiglottal retroversion were diagnosed on re-examination, but the owner declined further surgery.

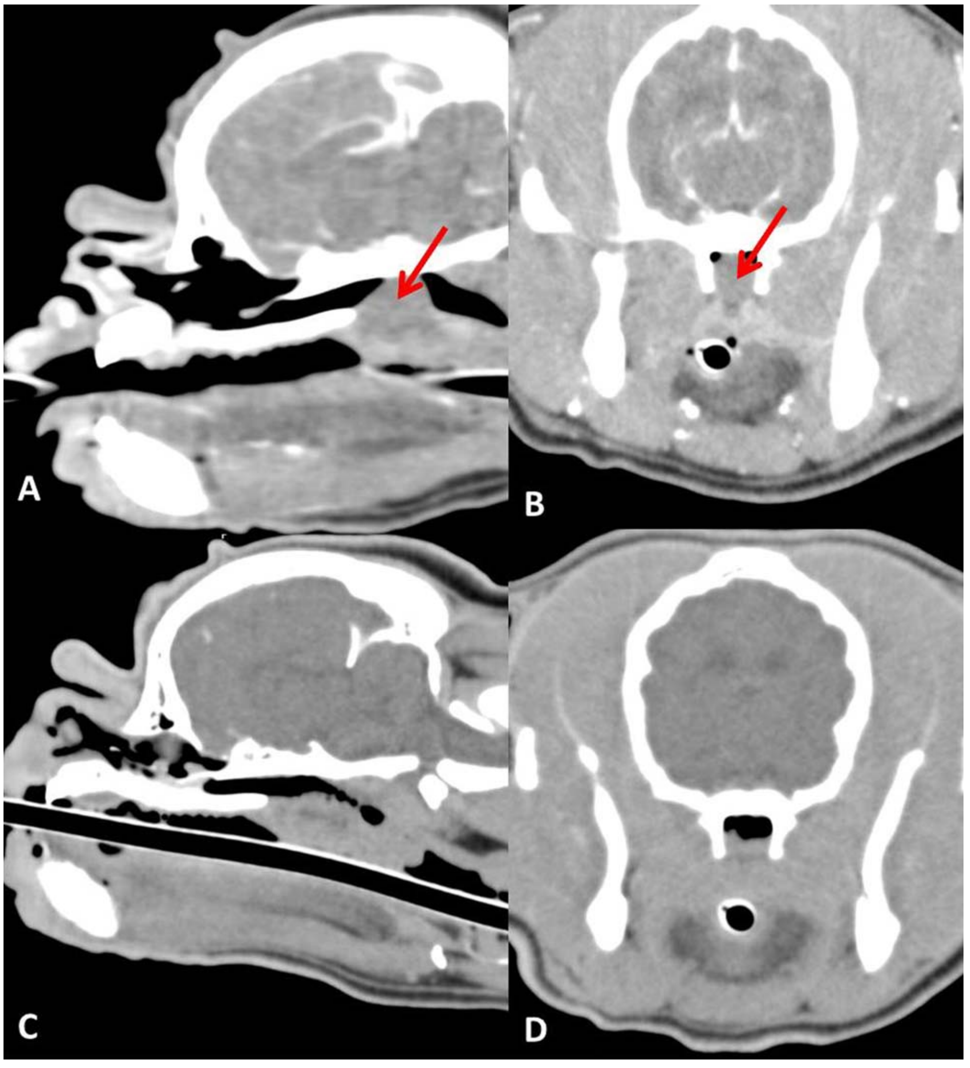

FIGURE 6 Midsagittal (A) and transverse (B) computed tomograms from a Pug with a single ventral obstructive nasopharyngeal sialocele dorsal to the soft palate (case 1). C, D, The same dog after removal of the external surface of the sialocele with biopsy forceps

4 | DISCUSSION

Masses causing nasopharyngeal obstruction include abscesses, tumors,[13] polyps,[14] and, rarely, cysts derived from Rathke's pouch[15,16] or the nasopharyngeal mucosa.[17] A review of the literature revealed no previous report of obstructive nasopharyngeal cysts derived from minor salivary gland in dogs. This study is therefore the first to describe NPS in a series of brachycephalic dogs. The pathogenesis of this condition remains unclear; several authors have suggested that trauma may be involved, but the great majority of published cases have no history of preceding trauma.[1,18] In addition, attempts to induce progressive sialoceles experimentally in dogs, by ligating or incising the mandibular salivary ducts or by traumatizing the mandibular gland, have failed.[8-19] We propose that friction and vibrations of nasopharyngeal mucosa during inspiratory effort in brachycephalic dogs could contribute to nasopharyngeal gland hyperplasia and trauma. These changes may lead to leakage of saliva, formation of a sialocele, and associated nasopharyngeal obstruction.

Thickening and elongation of the soft palate have been suggested to cause a narrowing of the oropharyngeal and nasopharyngeal passages, affecting respiratory activity in all brachycephalic dogs with brachycephalic airway obstructive syndrome (BOAS). The thick, redundant, and hyperplastic appearance of the soft palate in brachycephalic breeds has been described both macroscopically[20,21] and microanatomically.[22] Microscopic features of the soft palate in adult brachycephalic dogs include mucous gland hyperplasia on the nasopharyngeal or the oral side of the palate. The most important microscopic findings reported in a study of 11 adult brachycephalic dogs with grade I BOAS[22] were severely dilated glandular ducts, glandular hyperplasia, and mucocele. In a more recent study by the same authors,[23] none of the layers constituting the soft palate of neonate brachycephalic dogs had signs of palatine glands hyperplasia or mucous spilling. The authors therefore concluded that palatine glandular abnormalities develop after birth and may result from airway turbulence and consequent chronic barotraumas, as previously suggested.[24] Although this evidence supports the concept of chronic exposure to mechanical stress as a cause for NPS in brachycephalic breeds, the low incidence of the condition reported here leads us to suspect the contribution of other causative factors, such as the number and topography of minor salivary glands, individual variation in nasopharyngeal and soft palate dimensions and shape, turbinate topography, and the number of mucosal contact points.

In humans, mucocele of minor salivary glands occurs in both the oral and the nasopharyngeal cavities,[25-27] and 2 types of mucocele are recognized, extravasation mucocele and retention mucocele, the former being more frequent. Experimental and clinical studies of extravasation mucocele have shown that rupture of a minor salivary gland duct leads to the release of mucus into the surrounding connective tissues. The resulting cavity is well circumscribed and fills with mucus containing neutrophils, histiocytes, and foamy macrophages. The cavity is surrounded by nonsecretory, inflammatory, and connective tissue. Conversely, retention mucoceles arise from the dilation of minor salivary gland ducts that become filled with mucinous material and are lined by secretory, cuboidal ductal epithelium. All of the NPS reported here can be defined as extravasation cysts, supporting the contribution of chronic trauma as a cause for NPS in our case series (Figure 2B).

Although this case series is limited to 11 brachycephalic dogs and was biased toward dogs with clinical signs of other upper airway obstruction, Pugs seemed overrepresented (breed prevalence of 22.8%). Several factors can influence the severity of changes secondary to BOAS and sialocele formation. Recent studies have shown breed-specific anatomical and pathological differences between Pugs and French bulldogs.[28-30] Among those, the nasopharyngeal cross-sectional area was found on CT to be smaller in Pugs than in bulldogs, despite their smaller soft palate.[29] Based on these findings, Pugs may be predisposed to dynamic nasopharyngeal collapse secondary to greater negative luminal pressure generated on inspiration, increasing mechanical stress.

Radiographic examination can be important in the diagnosis of pharyngeal respiratory problems because most of these are associated with a reduction in pharyngeal air spaces; nevertheless, relevant radiographic abnormalities were detected in only 4 of 67 dogs with pharyngeal disorders in a previous study.[31] Radiography failed to detect a nasopharyngeal mass in all of the dogs described here and is not considered useful for the diagnosis of NPS. Latero-lateral radiographs of the pharynx in brachycephalic dogs can be misleading, because a thick soft palate and redundant nasopharyngeal mucosa may fill most or all of the normal radiolucent pharyngeal air space, with lack of soft tissue detail.[32] In spite of these limitations, thoracic and cervical radiographs are recommended in all dyspneic brachycephalic dogs to rule out other causes of dyspnea. Endoscopy also assists in the diagnosis of all types of pharyngeal disorders, but a flexible endoscope is essential for adequate investigation of the nasopharyngeal area.[31] Detection and adequate biopsy sampling of nasopharyngeal masses were achieved by retroflexed endoscopy in all dogs in the present study. For these reasons, we recommend preoperative endoscopic examination of the nasopharynx in all brachycephalic dogs.

Because of the sparse and inconstant distribution of minor salivary glands in the nasopharynx and soft palate, complete removal of the affected glands is practically impossible. Treatment of minor gland mucous extravasation in humans includes excision of the lesion with or without removal of the involved gland, marsupialization, cryosurgery, argon laser, and micromarsupialization.[33-36] Treatment of oral mucocele by high intensity diode laser has also recently been described as a viable option in humans.[37-39] Laser management is considered preferable over conventional surgery for its good hemostasis, reduced postoperative swelling, reduction in the bacterial population at the surgical site, limited placement of sutures, faster healing, and decreased postoperative pain. In 5 of the dogs described here, the entire portion of the sialocele protruding into the pharynx was resected with biopsy forceps under direct endoscopic visualization. The procedure was accomplished without complications and with negligible bleeding, although the smooth penetration of the hard cyst surface was sometimes difficult because of the small jaws of the biopsy forceps. In the other 6 dogs, laser vaporization of the sialocele was accomplished in an average surgical time of 12 minutes and seemed easier. No complication was encountered in any patient, and we consider both treatment methods suitable for NPS.

Five dogs had previously undergone upper airway surgery, without substantial improvement. Four of these dogs had undiagnosed obstructive NPS and improved after surgical deroofing with forceps or diode laser. These findings prompt us to suggest that nasopharyngeal obstruction by NPS should be considered in some cases as an important contributor of BOAS. Six dogs in our series were also treated with procedures traditionally recommended to treat brachycephalic syndrome, such as alaplasty, staphylectomy, and, when required, excision of everted laryngeal saccules. These adjunct procedures limit our ability to evaluate the actual contribution of nasopharyngeal obstruction to clinical signs as well as the efficacy of NPS surgery in this group of dogs.

In conclusion, to the best of our knowledge, this is the first report of NPS in dogs, and it is noteworthy that all dogs were brachycephalic. Although we cannot ascertain the pathogenesis of this condition, exposure of minor nasopharyngeal glands to chronic supraphysiologic mechanical stress should be considered as a potential causative factor for upper airway obstruction in BOAS. Direct visualization by retroflexed nasopharyngoscopy is the best means to detect NPS and grade the severity of BOAS.

Thorough resection of the external surface of the cyst-like lesion with forceps or diode laser under endoscopic guidance was successful in all of these patients. Although the number of brachycephalic dogs in this study was small, Pugs appeared to be overrepresented. Larger prospective studies are warranted to further explore the epidemiology and pathogenesis of NPS.

CONFLICT OF INTEREST

The authors declare no conflicts of interest related to this report.

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